National Collaborating Centre for Mental Health

Draft for consultation

Challenging Behaviour and Learning Disabilities:

Prevention and intervention for people with learning disabilities whose behaviour challenges

Clinical Guideline <…>

Methods, evidence and recommendations

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Draft for Consultation

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The British Psychological Society
NATIONAL COLLABORATING CENTRE FOR MENTAL HEALTH

Disclaimer
Healthcare professionals are expected to take NICE clinical guidelines fully into account when exercising their clinical judgement. However, the guidance does not override the responsibility of healthcare professionals to make decisions appropriate to the circumstances of each patient, in consultation with the patient and/or their guardian or carer.

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Contents

Guideline Development Group members and National Collaborating Centre for Mental Health (NCCMH) review team

Glynis Murphy (Chair)

Professor of Clinical Psychology & Disability, Co-Director of the Tizard Centre, University of Kent

Steve Pilling (Facilitator)

Director, NCCMH

David Allen

Clinical Director, Positive Response Training & Consultancy and Professor, Tizard Centre, University of Kent

Katherine Andrea

Senior Project Manager, NCCMH

David Branford

Chief Pharmacist Derbyshire Healthcare Foundation NHS Trust (retired)

Alick Bush

Lead Psychologist, St Andrews Healthcare

Carole Buckley

General Practitioner, The Old School Surgery

Vivien Cooper

Carer representative/CEO, The Challenging Behaviour Foundation

Jo Dwyer

Clinical Specialist Occupational Therapist, Lewisham Team for Adults With Learning Disabilities, Guys and St Thomas's NHS Foundation Trust

David Glynn

Health Economist, NCCMH (September 2014 onwards)

Bronwyn Harrison

Systematic Reviewer, NCCMH

Angela Hassiotis

Professor, UCL

Honorary Consultant Psychiatrist, Camden & Islington Foundation Trust

Phil Howell

PIAS Manager and PBS Consultant, British Institute of Learning Disabilities

Simon Jones

Head of Behavioural Support, Care UK

Elena Marcus

Research Assistant, NCCMH (August 2014 onwards)

Ifigeneia Mavranezouli

Senior Health Economist, NCCMH

Richard Mills

Research Director, Research Autism

David Newton

Team Manager, Adult Safeguarding Quality Assurance Team, Adult Social Care Directorate, Nottingham City Council

Steve Noone

Consultant Clinical Psychologist, Northumberland, Tyne and Wear Foundation NHS Trust

Cheryl Palmer

Research Assistant, NCCMH

Phil Perkins

Senior Community Learning Disability Nurse for Children and Young People, Surrey and Borders Partnership NHS Foundation Trust

Victoria Slonims

Senior Consultant Speech and Language Therapist; Honorary Senior Lecturer, Evelina Children’s Hospital, Guy's and St Thomas' NHS Foundation Trust

Clare Taylor

Senior Editor, NCCMH

Craig Whittington

Associate Director (Clinical Effectiveness)/Lead Systematic Reviewer, NCCMH (September 2014 onwards)

Keith Wyncoll

Carer representative

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Gemma Griffiths, Bangor University

Mieke Heyvaert, Katholieke Universiteit Leuven

1 Preface

This guideline has been developed to advise on the management and support of people with a learning disability and behaviour that challenges, and prevention of behaviour and challenges. This guideline covers children (aged 12 years or younger), young people (aged 13 to 17 years) and adults (aged 18 years or older).

The guideline recommendations have been developed by a multidisciplinary team of healthcare professionals, people who care for those with a learning disability and behaviour that challenges and guideline methodologists after careful consideration of the best available evidence. It is intended that the guideline will be useful to clinicians and service commissioners in providing and planning high-quality care for people with a learning disability and behaviour that challenges while also emphasising the importance of the experience of care for people with a learning disability and behaviour that challenges and their families and carers (see Appendix A for more details on the scope of the guideline).

Although the evidence base is rapidly expanding, there are a number of major gaps. The guideline makes a number of research recommendations specifically to address gaps in the evidence base. In the meantime, it is hoped that the guideline will assist clinicians, and people with a learning disability and behaviour that challenges and their families and carers, by identifying the merits of particular treatment approaches where the evidence from research and clinical experience exists.

1.1 National clinical guidelines

1.1.1 What are clinical guidelines?

Clinical guidelines are ‘systematically developed statements that assist clinicians and service users in making decisions about appropriate treatment for specific conditions’ (Mann, 1996). They are derived from the best available research evidence, using predetermined and systematic methods to identify and evaluate the evidence relating to the specific condition in question. Where evidence is lacking, the guidelines include statements and recommendations based upon the consensus statements developed by the Guideline Development Group (GDG).

Clinical guidelines are intended to improve the process and outcomes of healthcare in a number of different ways. They can:

- provide up-to-date evidence-based recommendations for the management of conditions and disorders by healthcare professionals
- be used as the basis to set standards to assess the practice of healthcare professionals
- form the basis for education and training of healthcare professionals
- assist service users and their families and carers in making informed decisions about their treatment and care
- improve communication between healthcare professionals, service users and their families and carers
- help identify priority areas for further research.

1.1.2 Uses and limitations of clinical guidelines

Guidelines are not a substitute for professional knowledge and clinical judgement. They can be limited in their usefulness and applicability by a number of different factors: the availability of high-quality research evidence, the quality of the methodology used in the development of the guideline, the generalisability of research findings and the uniqueness of individuals.

Although the quality of research in this field is variable, the methodology used here reflects current international understanding on the appropriate practice for guideline development (Appraisal of Guidelines for Research and Evaluation Instrument [AGREE]; www.agreetrust.org; (AGREE Collaboration, 2003)), ensuring the collection and selection of the best research evidence available and the systematic generation of treatment recommendations applicable to the majority of people with a learning disability and behaviour that challenges. However, there will always be some people and situations where clinical guideline recommendations are not readily applicable. This guideline does not, therefore, override the individual responsibility of healthcare professionals to make appropriate decisions in the circumstances of the individual, in consultation with the person with a learning disability and behaviour that challenges or their families and carers.

In addition to the clinical evidence, cost-effectiveness information, where available, is taken into account in the generation of statements and recommendations in clinical guidelines. While national guidelines are concerned with clinical and cost effectiveness, issues of affordability and implementation costs are to be determined by the National Health Service (NHS).

In using guidelines, it is important to remember that the absence of empirical evidence for the effectiveness of a particular intervention is not the same as evidence for ineffectiveness. In addition, and of particular relevance in mental health, evidence-based treatments are often delivered within the context of an overall treatment programme including a range of activities, the purpose of which may be to help engage the person and provide an appropriate context for the delivery of specific interventions. It is important to maintain and enhance the service context in which these interventions are delivered, otherwise the specific benefits of effective interventions will be lost. Indeed, the importance of organising care in order to support and encourage a good therapeutic relationship is at times as important as the specific treatments offered.

1.1.3 Why develop national guidelines?

The National Institute for Health and Care Excellence (NICE) was established as a Special Health Authority for England and Wales in 1999, with a remit to provide a single source of authoritative and reliable guidance for service users, professionals and the public. NICE guidance aims to improve standards of care, diminish unacceptable variations in the provision and quality of care across the NHS, and ensure that the health service is person-centred. All guidance is developed in a transparent and collaborative manner, using the best available evidence and involving all relevant stakeholders.

NICE generates guidance in a number of different ways, 4 of which are relevant here. First, national guidance is produced by the Technology Appraisal Committee to give robust advice about a particular treatment, intervention, procedure or other health technology. Second, NICE commissions public health intervention guidance focused on types of activity (interventions) that help to reduce people's risk of developing a disease or condition, or help to promote or maintain a healthy lifestyle. Third, NICE commissions social care guidance which makes recommendations that span across health, public health and social care, allowing a more integrated approach to supporting people and ensuring their needs are met. Forth, NICE commissions the production of national clinical guidelines focused upon the overall treatment and management of a specific condition. To enable this latter development, NICE has established 4 National Collaborating Centres in conjunction with a range of professional organisations involved in healthcare.

1.1.4 From national clinical guidelines to local protocols

Once a national guideline has been published and disseminated, local healthcare groups will be expected to produce a plan and identify resources for implementation, along with appropriate timetables. Subsequently, a multidisciplinary group involving commissioners of

healthcare, primary care and specialist mental health professionals, service users and carers should undertake the translation of the implementation plan into local protocols, taking into account both the recommendations set out in this guideline and the priorities in the National Service Framework for Mental Health (Department of Health, 1999) and related documentation. The nature and pace of the local plan will reflect local healthcare needs and the nature of existing services; full implementation may take a considerable time, especially where substantial training needs are identified.

1.1.5 Auditing the implementation of clinical guidelines

This guideline identifies key areas of clinical practice and service delivery for local and national audit. Although the generation of audit standards is an important and necessary step in the implementation of this guidance, a more broadly-based implementation strategy will be developed. Nevertheless, it should be noted that the Care Quality Commission in England, and the Healthcare Inspectorate Wales, will monitor the extent to which commissioners and providers of health and social care and Health Authorities have implemented these guidelines.

1.2 The national Challenging Behaviour and Learning Disabilities guideline

1.2.1 Who has developed this guideline?

This guideline has been commissioned by NICE and developed within the National Collaborating Centre for Mental Health (NCCMH). The NCCMH is a collaboration of the professional organisations involved in the field of mental health, national service user and carer organisations, a number of academic institutions and NICE. The NCCMH is funded by NICE and is led by a partnership between the Royal College of Psychiatrists and the British Psychological Society's Centre for Outcomes Research and Effectiveness, based at University College London.

The GDG was convened by the NCCMH and supported by funding from NICE. The GDG included people with a learning disability and behaviour that challenges and carers, and professionals from psychiatry, clinical psychology, nursing, social work, speech and language therapy, and general practice; academic experts in psychiatry and psychology; commissioning managers; and carers and representatives from service user and carer organisations.

Staff from the NCCMH provided leadership and support throughout the process of guideline development, undertaking systematic searches, information retrieval, appraisal and systematic review of the evidence. Members of the GDG received training in the process of guideline development from NCCMH staff, and the service users and carers received training and support from the NICE Patient and Public Involvement Programme. The NICE Guidelines Technical Adviser provided advice and assistance regarding aspects of the guideline development process.

All GDG members made formal declarations of interest at the outset, which were updated at every GDG meeting. The GDG met a total of 11 times throughout the process of guideline development. The group oversaw the production and synthesis of research evidence before presentation. All statements and recommendations in this guideline have been generated and agreed by the whole GDG.

1.2.2 For whom is this guideline intended?

This guideline will be relevant for children, young people and adults with a learning disability and behaviour that challenges and covers the care provided by primary, community,

secondary, tertiary and other healthcare professionals who have direct contact with, and make decisions concerning the care of, children, young people and adults with a learning disability and behaviour that challenges.

The guideline will also be relevant to the work, but will not cover the practice, of those in:

- occupational health services
- social services
- the independent sector.

1.2.3 Specific aims of this guideline

The guideline makes recommendations for the management and support of children, young people and adults with a learning disability and behaviour that challenges. It aims to:

- improve access and engagement with treatment and services for people with a learning disability and behaviour that challenges
- improve the methods of assessment and identification of those at risk of developing challenging behaviour
- evaluate the role of specific psychological, psychosocial, environmental and pharmacological interventions
- integrate the above to provide best-practice advice on the care of individuals
- promote the implementation of best clinical practice through the development of recommendations tailored to the requirements of the NHS in England and Wales.

1.2.4 The structure of this guideline

The guideline is divided into chapters, each covering a set of related topics. The first 3 chapters provide a general introduction to guidelines, an introduction to the topic of learning disabilities and behaviour that challenges, and to the methods used to develop guidelines. Chapter 4 to Chapter 13 provide the evidence that underpins the recommendations about the support and management of people with a learning disability and behaviour that challenges.

Each evidence chapter begins with a general introduction to the topic that sets the recommendations in context. Depending on the nature of the evidence, narrative reviews or meta-analyses were conducted, and the structure of the chapters varies accordingly. Where appropriate, details about current practice, the evidence base and any research limitations are provided. Where meta-analyses were conducted, information is given about both the interventions included and the studies considered for review. Clinical summaries are then used to summarise the evidence presented. Finally, recommendations related to each topic are presented at the end of each chapter. Full details about the included studies can be found in Appendix L, Appendix M and Appendix N . Where meta-analyses were conducted, the data are presented using forest plots in Appendix P (see Table 1 for details).

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2 Introduction

Some people with a learning disability display behaviour that challenges. 'Behaviour that challenges' is not a diagnosis and is used in this guideline to indicate that such behaviour is a challenge to services, family members and carers, but may be functional for the person with a learning disability. The behaviour may appear in only certain environments, and the same behaviour may be considered challenging in some settings or cultures but not in others. It may be used by the person for reasons such as creating sensory stimulation. Some care environments increase the likelihood of behaviour that challenges. This includes those with limited social interaction and meaningful occupation, lack of choice and sensory input, excessive noise, those that are crowded, unresponsive or unpredictable, and those characterised by neglect and abuse.

When children, young people or adults with a learning disability engage in behaviour that challenges, they may experience a series of escalating reductions in their quality of life, such as restrictive practices (restraint and locked doors), physical abuse, placement breakdown and out of area placements (Department of Health, 2007; Emerson & Einfeld, 2011; Royal College of Psychiatrists, 2007)). Families, carers and staff also experience a reduction in quality of life, often reporting frustration, fatigue, exhaustion, burnout and feeling unable to continue in their caring role (Hastings, 2002a; Lecavalier et al., 2006). Meanwhile, when families, carers or staff are unable to cope, service commissioners are often uncertain about what to do. At times, they fund the person's care in poor quality services that are out of area, that may be very expensive, and that may increase the risk of behaviour that challenges even further (Allen et al., 2007; Barron et al., 2011; McGill & Poynter, 2012). Such placements are often a long distance from families, meaning that their quality of life, and that of their family member, may be even more compromised (Bonell et al., 2011; Chinn et al., 2011). This guideline addresses these important issues for people with a learning disability, their families and carers, staff and service providers and commissioners.

2.1 Definitions and terminology

2.1.1 Learning disabilities

In the UK, the term 'learning disabilities' was first used formally in 1991 in a speech by the then Health Minister, Stephen Dorrell, to refer to what had previously been termed 'mental handicap' or 'mental retardation' (which people with a learning disability and their families found unacceptable). Since then 'learning disabilities' has been the accepted term in government documents. Recently, in the White Paper *Valuing People*, the Department of Health (2001) defined a learning disability as:

- a significantly reduced ability to understand complex information or learn new skills (impaired intelligence)
- a reduced ability to cope independently (impaired social functioning)
- a condition which started before adulthood (18 years of age), and has a lasting effect.

It is important to be clear that the term 'learning disabilities' employed in this guideline implies *pervasive* or *global* learning disabilities, affecting most aspects of cognitive functioning, and not *specific* learning disabilities, such as dyslexia.

Services for adults with a learning disability are familiar with the above definition in the UK. In children's services, however, rather different terms are used, because the education authorities prefer the term 'learning difficulties' which covers a broader group of children.

Internationally, the term 'learning disability' is often confused with dyslexia and so in international contexts the preferred phrase is 'intellectual disability'. This is becoming the accepted term in Australia, New Zealand, Canada, USA, Europe and Scandinavia. In the UK,

the term ‘learning disability’ is still the most widely used and accepted - only the British Psychological Society and the Royal College of Psychiatrists have adopted the phrase ‘intellectual disability’ (December 2013). Therefore in this guideline the term ‘learning disability’ is used.

Whatever the term used, it is widely recognised that learning disability is largely a socially constructed phenomenon (Finlay & Lyons, 2005), which has had varying different definitions over time and across countries. Currently most developed countries accept a 3-part definition:

1. Significant impairments in cognitive functioning
2. Significant impairments in adaptive behaviours
3. Occurring in the developmental period.

The disabilities are thus seen as being located in the individual, and a major challenge to this so-called ‘medical’ model has come from those who espouse a social model of disability and who argue that disability arises from the inability of social environments to adapt to fit a person’s needs. With a responsive environment, they argue, impairments would not become disabilities (Shakespeare, 2006; Thomas, 2007).

People with a learning disability may have a very wide range of impairments and there have been numerous attempts to sub-divide the population on the basis of cognitive ability. For example, the WHO ICD-10 sub-division is into:

- Mild learning disability - IQ between 50 and 69
- Moderate learning disability - IQ between 35 and 49
- Severe learning disability - IQ between 20 and 34
- Profound learning disability - IQ less than 20.

Such classifications have been heavily criticised however, not least because they rely on IQ. It is important to be aware that IQ cannot be measured with much accuracy below 50, and certainly the accuracy is highly compromised below 35. Moreover a person’s IQ can vary depending on the test and when the test is conducted, and it may change over longer periods of time. In addition, people’s everyday skills are not only dependent on IQ: some people with relatively high IQ can seem very disabled if they are very socially impaired (for example, people with Asperger syndrome) and/or if they have major difficulties with communication, while conversely others with good social skills and expressive language can appear more able than their IQ might suggest. Consequently, taking all of this into account, the sub-divisions above are not very useful. The picture becomes even more complicated when considering children: education authorities in the UK refer to children with moderate and severe learning difficulties, and these terms do not map well onto the WHO sub-divisions above. Thus a child with ‘moderate learning difficulties’ in school becomes an adult with a ‘mild learning disability’, and a child with ‘severe learning difficulties’ in school becomes an adult with a ‘moderate learning disability’ in adult services.

Nevertheless, the GDG recognises that there is a very large range of abilities among people with a learning disability: some people have good mobility, considerable language skills, adequate self-care skills, and may only need help with more complex tasks, while others may have far more extreme degrees of disability, with very poor mobility, little or no language skills and need a great deal of assistance with self-care and other tasks. Consequently it will sometimes be necessary in this guideline to distinguish people with more skills from those with fewer skills, for example when recommending assessments or treatments that will not all be suitable for everyone.

2.1.21 Behaviour that challenges

It is widely recognised that people with a learning disability are at increased risk of various mental and physical health problems. In addition, some engage in behaviour that has been called challenging. Emerson's definition of 'challenging behaviour' is:

> *Culturally abnormal behaviour(s) of such an intensity, frequency or duration that the physical safety of the person or others is likely to be placed in serious jeopardy, or behaviour which is likely to seriously limit use of, or result in the person being denied access to, ordinary community facilities (Emerson, 1995).*

The Royal College of Psychiatrists (2007) defined 'challenging behaviour' very similarly as:

> *Behaviour of such an intensity, frequency or duration as to threaten the quality of life and/or the physical safety of the individual or others and is likely to lead to responses that are restrictive, aversive or result in exclusion.*

Historically, such behaviour had been described as 'inappropriate', 'abnormal', 'disordered', 'dysfunctional', 'problem' or 'maladaptive'. However, research has shown that the behaviour in question is actually quite adaptive and functional in some ways, and not disordered. The newer term, 'challenging behaviour', was thought to have some advantages over these earlier terms, in that it suffers from fewer semantic contradictions, and it was also intended to remind professionals, staff and policy makers that such behaviour was a challenge to services.

The intention of the term 'challenging behaviour' was to prevent the phrase being used as a diagnosis and to stop people feeling that we needed to 'fix' the person, so that they would instead concentrate on 'fixing' the environment. However, since the introduction of the term many professionals and carers have felt that the reason for the change in terminology has been lost sight of. The frequent use of personal pronouns and verbs (such as 'his challenging behaviour' or 'she has challenging behaviour'), imply that the problem is within the person. It is important to recognise that 'challenging behaviour' is rather the result of an interaction between the person and their environment, and as such is largely socially constructed. The term 'behaviour that challenges' is preferred as an alternative, and this phrase will be used in this guideline.

The kinds of behaviour referred to include: aggressive behaviour (such as verbal abuse, threats and physical violence), destructive behaviour (such as breaking or destroying furniture and other objects and setting fires), disruptive behaviour (such as repetitive screaming, smearing faeces, setting off fire alarms when there is no fire, calling the emergency services when there is no emergency), self-injurious behaviour (including self-biting, head banging), sexually harmful behaviour (including sexual assaults, rape and stalking). Some of these behaviours may fall under the purview of the criminal justice system, but by no means all those with a learning disability who engage in illegal behaviour are arrested, as the criminal justice system requires not just *actus reus* but also *mens rea*, so that most people with severe disabilities who engage in potentially illegal behaviour are not involved in the criminal justice system.

The setting in which behaviours occur can influence whether the behaviour is considered challenging. For example, behaviours such as shouting and jumping are acceptable at a rock concert but not in a library, and physical aggression is acceptable in a boxing ring but not outside of the ring. Similarly, some behaviours, such as running away from home, may be seen as challenging in some circumstances, such as when the person lives with supervision at home and is unsafe out alone, but they may not be challenging in other circumstances, such as if the person is on a fitness programme involving daily running, and is safe out on their own. Likewise, for many carers, sleep difficulties in the person they care for may feel very challenging. For example, if someone with severe disabilities who is not safe to be up alone, frequently wakes for large parts of the night, wanders about the house, falls down the

stairs, destroys household objects and exhausts his or her carers, it is likely that such acts would be seen by them as behaviour that challenges. In circumstances such as these, it is important to be clear that it is not the poor sleep per se that is challenging, but the behaviour that occurs when the person would normally be asleep. If this person lived in a staffed house with waking night staff, the poor sleep might not be seen as challenging, and likewise if they woke at night and were lying quietly in bed, poor sleep might not be seen as challenging.

2.1.37 Carers

In this guideline the word 'carer' is used to refer to a person who provides unpaid support to a partner, family member, friend or neighbour with a learning disability and behaviour that challenges. It does not refer to paid carers or care workers, who are defined as 'staff' in this guideline (see below), unless otherwise specified.

2.1.42 Staff

In this guideline, the term 'staff' includes health and social care professionals, including those working in community teams for adults or children (such as psychologists, psychiatrists, social workers, speech therapists, nurses, occupational therapists, physiotherapists), care workers in a variety of settings (including residential homes, supported living settings and day services) and teachers.

2.28 Prevalence

The prevalence of behaviour that challenges has been the subject of numerous studies, which have produced a range of figures. The reason there is such a range is that the prevalence depends on a variety of methodological issues. For example:

a) Studies in hospital/institutional environments always produce much higher figures. This may be partly because people have been admitted there as a result of behaviour that challenges, and partly because aspects of the hospital/institutional environment (such as low engagement levels) may cause an increase in behaviour that challenges. For example, Oliver et al (1987) in a well-known study of self-injurious behaviour in a total population of people with a learning disability in touch with services, reported a prevalence rate for self-injury of 12% in hospitals for people with a learning disability, but only 3% for adults with a learning disability in the community. Borthwick Duffy (1994) showed an even bigger discrepancy between institutional and community-based prevalence rates for behaviour that challenges: 49% versus 3% respectively.

b) Studies may use different definitions of the behaviour to be counted. For example, they may count only 1 type of behaviour. Oliver and colleagues (1987), for instance, asked whether anyone had shown self-injurious behaviour of the following kind: 'repeated, self-inflicted, non-accidental injury, producing bruising, bleeding or other temporary or permanent tissue damage' within the previous 4 months. Had they used a definition that did not require the behaviour to have caused 'tissue damage', they would have probably found higher figures. Likewise, had they employed a longer period of time, for example 'in the last year', they may well have found higher figures. Moreover had they also counted other behaviour that challenges, such as aggression, they would have found even higher figures.

c) Most studies count prevalence by asking staff or carers for their opinions. It is likely that the staff and carers vary in their observational powers and their memory so that some may recall some behaviours that others do not. Likewise, behaviour that challenges varies with the environment, including the social environment, such that the behaviour might be far more problematic for some staff or carers than others, so that different people will report different rates.

With these provisos in mind, the accepted range for prevalence of behaviour that challenges, is approximately 6 to 14% of people with a learning disability who are known to services (Borthwick-Duffy, 1994; Emerson, 2001; Emerson & Bromley, 1995; Kiernan & Qureshi, 1993). These figures derive from surveys of total populations of people with a learning disability (administratively defined) and including all types of behaviour that challenges. According to Emerson and Einfeld (2011) this translates to a prevalence of between 2 and 5 per 10,000 of general population (using administrative prevalence rates for learning disabilities in the general population), in other words between 12,000 and 30,000 people across the UK (assuming a general population of 60 million people).

Typically, in these surveys, researchers interview staff and carers about people with a learning disability, and use a specific definition of behaviour that challenges. As an example, that of Kiernan and Qureshi (1993), which defines quite a serious level of behaviour, is given below:

a) Showed behaviour that 'at some time caused more than minor injury to themselves or others, or destroyed their immediate living or working environment'.

b) Showed behaviour 'at least once a week that required the intervention of more than one member of staff to control, or placed them in danger, or caused damage that could not be rectified by care staff or caused more than 1 hour's disruption'.

c) Showed behaviour 'at least daily that caused more than a few minutes disruption'.

It is relatively rare for studies to use a particular questionnaire, with a specified cut-off point, to establish prevalence, as would be common in medical or other diagnostic studies, based on a widespread view that this is an inappropriate approach for the topic of learning disabilities and behaviour that challenges, partly because of the great variations seen for the same person in different environments.

Few prevalence studies have asked about behaviour that has come to the attention of the criminal justice system. One exception to this is McBrien and colleagues (2003) who surveyed all adults known to learning disabilities services in an area with a general population of about 200,000. They reported that 3% of the adults with a learning disability known to services had a current or previous conviction and a further 7% had had previous contact with the criminal justice system but no conviction. As Murphy and Mason (2014) point out, this is likely to be an overestimate of the true proportion of people with a mild learning disability involved with the criminal justice system, as most people with a mild learning disability do not receive services (between one and two thirds disappear from services on leaving school) and therefore they were probably not included in the survey.

This fact, that most studies of the prevalence of behaviour that challenges consider only the people with a learning disability who are known to services (so-called administrative prevalence), together with the fact that many people with a mild learning disability disappear from services after school age, means that the prevalence of behaviour that challenges in people with a severe learning disability, who almost all receive services, is fairly well established. The prevalence of behaviour that challenges among people with a mild learning disability is more difficult to know. As already noted, people with a mild learning disability are more likely to lose touch with services if they have no special needs when they leave school, but to remain in touch with services if they have behaviour that challenges. Nevertheless, the uncertainties of this administrative prevalence approach has brought some researchers to examine total cohort studies of a general population of children. These studies, however, while they may solve the problem of ensuring a total population is captured, encounter other problems, such as how learning disabilities and behaviour that challenges are defined within the survey. Emerson and Einfeld (2011) describe 3 surveys of this type, 1 giving the prevalence of 'conduct disorder' among children aged 5 to 16 years with 'intellectual disabilities' as 12% (while that of non-disabled children was 4%), 1 giving a figure of 'behavioural difficulties' for children aged 6 to 7 years with 'intellectual disabilities' of 24%

(compared with 8% for non-disabled children), and the third giving a figure for 'behavioural difficulties' for British children aged 3 years with 'early cognitive delay' of 30% (compared with 10% for children without delays). Clearly the fact that these surveys often use a variety of definitions of intellectual or learning disabilities and/or cognitive delay, as well as a variety of definitions of the behaviour to be counted, make them difficult to compare with the more common studies of administrative prevalence of behaviour that challenges. Nevertheless, they all broadly agree that behaviour that challenges is about 3 times more common in children with disabilities than in typically developing children.

2.39 Co-occurrence and persistence

It is known that behaviour that challenges can co-occur, such that between a half and two thirds of people who show behaviour that challenges, engage in more than 1 form (where 'form' is classified as 'aggression', 'self-injury', 'property destruction' and 'other', Emerson, 2001). Matson and colleagues (2008), for example, found that people who showed self-injury were more likely to have other behaviour that challenges such as aggression, when compared with those without self-injury, matched for age, gender and degree of disability. In a recent study, in which Oliver and colleagues (2012) also found considerable co-occurrence between self-injury, aggression and repetitive behaviours in children with a severe learning disability, Oliver and colleagues (2012) argued that high-frequency repetitive behaviours could be a risk marker for other behaviour that challenges.

Even with 1 'form' of behaviour that challenges, such as self-injury, it is common for people to show more than 1 topography: for example, Oliver and colleagues (1987) in their survey found 54% of those who showed self-injury had more than 1 topography, 3% showed more than 5 topographies, and, among those who wore protective devices, 7% had 5 or more topographies.

It has been repeatedly found that the prevalence rates of behaviour that challenges varies considerably with age, peaking in people with a learning disability in their late teens and early twenties and gradually reducing thereafter (Borthwick-Duffy, 1994; Davies & Oliver, 2013; Kiernan & Kiernan, 1994; Oliver et al., 1987). Some behaviours that challenge are persistent, however, and it appears that when such behaviour is very severe, it can be long-lasting. For example, Murphy and colleagues (1993) reported in their study of those whose self-injury was so severe as to require protective devices, that the average age of onset of self-injury was 7 years and the duration (so far) was 14 years. In a follow-up of this Murphy and Oliver cohort, Taylor and colleagues (2011), found that 84% of those who showed self-injury in the 1987 study, continued to show self-injurious behaviour 18 years later. Similarly, Murphy and colleagues (2005) found that, in a total population of South London children with a learning disability or autism who were known to services, the presence of 'behaviour problems' at mean age of 8.9 years predicted the presence of 'behaviour problems' in the same individuals as adults (mean age 20.9 years). Likewise, Emerson and colleagues (1988) reported that when local authority agencies were asked who their 2 or 3 'most challenging' individuals were, the people they named had been showing that same behaviour for over 20 years.

Nevertheless, while some people show behaviour that has a lengthy and serious trajectory, behaviour that challenges that emerges in some young children disappears over time (Oliver et al., 2005). Cooper and colleagues (2009a; 2009b) have also reported considerable change in aggressive and self-injurious behaviours over a 2-year period in adults with a learning disability, when all such behaviours are counted and not just the most serious levels of such behaviours.

2.41 Associated characteristics

A number of characteristics are known to be associated with behaviour that challenges, including gender, degree of disability, communication skills, sensory impairments, various historical factors, and the presence of some genetic and other disorders:

a) Gender: males are somewhat more likely than females to show certain types of behaviour that challenges, especially aggressive behaviour (Borthwick-Duffy, 1994; McClintock et al., 2003). Males and females are about equally likely to show self-injury (Oliver et al., 1987).

b) Degree of disability: there is very broad agreement across numerous studies (Borthwick-Duffy, 1994; Cooper et al., 2009a; Cooper et al., 2009b; Kiernan & Qureshi, 1993; Oliver et al., 1987) that behaviour that challenges is more prevalent among people with severe and profound disabilities, and this is especially so for self-injurious behaviour (McClintock et al., 2003). This does not mean that people with a mild disability are never challenging: some may be very challenging, but most will not be. The lower prevalence in less disabled people may not be obvious to professionals working in adult services because many people with a mild disability (the most numerous group) 'disappear' from adult services after they leave school, and those who remain in touch with adult services may well be there because they are the ones whose behaviour is challenging.

c) Communication skills: children and adults with poorer communication skills tend to have higher rates of behaviour that challenges (Emerson, 2001; Kiernan & Kiernan, 1994; Murphy et al., 2005), especially self-injury (McClintock et al., 2003). This may be the important variable (or one of them) underlying the relationship between the degree of learning disability and behaviour that challenges.

d) Sensory impairments: sensory impairments, such as hearing and/or visual impairments put people at increased risk of behaviour that challenges (Cooper et al., 2009a; Kiernan & Kiernan, 1994).

e) Low mood: there are very few studies that examine the relationship between mood and behaviour that challenges. One reason for this is the difficulty of measuring mood in people with a severe disability. However, Hayes and colleagues (2011) demonstrated that low mood, reliably rated on the Mood Interest and Pleasure Questionnaire, was associated with the presence of behaviour that challenges in people with a severe learning disability.

f) Attachment: attachment towards carers and staff, and the associated behaviours, have been considered to have the function of promoting carers' and staff support of children, assisting them in regulating their own emotions at times of stress. There are very few studies of attachment and behaviour that challenges in children or adults with a learning disability (Schuengel et al., 2013). However, in 1 study of young people with a learning disability in a day care setting, it was shown that young people with poor attachment had higher levels of behaviour that challenges, and this was not explained by factors such as the presence of autism (De Schipper & Schuengel, 2010).

g) Traumatic events: it has been supposed for many years that traumatic experiences may lead to behaviour that challenges. It is only recently that this has been reliably established by 2 different studies. In 1, a group of adults with a learning disability who had been abused were matched for age, gender, communication skills and degree of disability to a non-abused group (Sequeira et al., 2003). The abused group had significantly more mental health needs, PTSD symptoms and behaviours that challenge. In the other study, carers of people with a severe learning disability were asked about their family members' behaviours before and after abusive events, using standardised measures (Murphy et al., 2007). A very consistent pattern emerged of significantly fewer behaviours that challenge before the traumatic event, significantly raised levels just after

the traumatic event, and some improvement years later. Adaptive behaviours changed in the opposite direction: they were significantly higher before the traumatic event, fell significantly immediately afterwards, and recovered somewhat years later.

h) Mental health needs: some researchers have argued that the presence of mental health needs raises the risk of behaviour that challenges (Cooper et al., 2009a; Cooper et al., 2009b; Hemmings et al., 2006; Moss et al., 2000). This has been much disputed, mainly because the presence of mental health needs is usually based on self-report of distress in the general population, and yet the people with most severe behaviour that challenges often have the least verbal skills, making diagnosis of mental health needs difficult. This is further complicated by arguments about whether behaviour (including behaviour that challenges) can be seen as a 'symptom' of mental health needs, and, if this premise is accepted, then the co-occurrence of the 2 becomes tautological.

i) Behavioural phenotypes: a number of specific syndromes associated with learning disabilities have raised risks of particular types of behaviour associated with them (this is discussed further in 2.5.1). Occasionally the links between syndromes and behaviour are very specific, to the extent that almost everyone with that specific diagnosis shows that specific behaviour. One example of this is Lesch-Nyhan syndrome, an X-linked metabolic disorder resulting in mild or moderate learning disabilities but severe physical disabilities, in which a characteristic form of self-injury appears in the first few years of life, specifically severe self-biting, in most affected children (Hall et al., 2001). Such a close link between syndrome and behaviour, however, is rare – typically syndromes simply raise the risk of specific behaviours, such that they are only somewhat more common than in other disorders (see Table 2 for some examples of these).

2.5 Causes

There is very broad agreement that behaviour that challenges results from a multiplicity of causes. These include biological, psychological, social and environmental causes. They can be conceptualised through diagrams such as Oliver's biopsychosocial model of self-injury (Oliver, 1993), Murphy's biopsychosocial model of aggression (Murphy, 1997) and Langthorne and colleagues' (2007) integrative model for behaviour that challenges. Individualised formulation diagrams, such as Murphy and Clare's case examples (2012), also show similar factors at play, for particular individuals. The contributions of the various factors are summarised below.

2.5.1 Biological causes

In the past, biological causes were thought to be the most prominent reason for behaviour that challenges and it was partly this idea that led to the belief that the behaviour in some sense 'sat inside' the person with a disability. There were a number of pieces of evidence that were thought to support this view:

a) The higher prevalence of behaviour that challenges in people with a more severe disability and therefore, some have argued, more extensive brain damage or dysfunction (see section 2.2).

b) The co-occurrence of behaviour that challenges with genetic syndromes and other diagnoses (see below & Table 2).

c) The discovery that some very specific biochemical substances were associated with particular types of behaviour that challenges (for example, high endogenous opioids associated with severe self-injury).

There are, of course, reasons why more severe disability is associated with the presence of behaviour that challenges, which might be unrelated to degree of brain damage or

dysfunction. For example, more severe degrees of disability are usually associated with poorer communication skills and there are very clear psychological reasons why poor communication skills may underlie the causes of behaviour that challenges (see section 2.5.2).

Table 2: Behavioural phenotypes in some common syndromes

Diagnosis/syndrome	Behaviour that challenges	Reference
Autism	Raised risk of a variety of behaviours that challenge, compared with children with a learning disability and no autism, especially for self-injury, stereotypy and aggression	(McClintock et al., 2003; Murphy et al., 2005)
Fragile X	Raised risk of hyperactivity, stereotypy, self-injury and autistic-like behaviours, fewer compulsions	(Hagerman, 2002; Langthorne & McGill, 2012)
Cornelia de Lange	Raised risk of hyperactivity, stereotypy, self-injury and autistic-like behaviours, including compulsions	(Basile et al., 2007; Oliver et al., 2008)
Lesch-Nyhan	Very high risk of developing self-injury, starting with self-biting and progressing to other forms of self-injury	(Jinnah et al., 2010; Jinnah & Friedmann, 2001; Lesch & Nyhan, 1964)
Prader Willi	Raised risk of behaviour that challenges, particularly repetitive questions and temper tantrums that are often food-related	(Holland et al., 2003; Oliver et al., 2009)
Rett	Typical development followed by regression, with raised risk of breathing difficulties, self-injury and stereotypies, particularly in centre line, and including hand wringing, plus autistic–like behaviours	(Hagberg et al., 1983; Mount et al., 2001)
Smith Magenis	Raised risk of self-injury, aggression, and sleep disorders	(Dykens & Smith, 1998; Finucane et al., 2001; Taylor & Oliver, 2008)

Nevertheless, it is difficult to explain why *specific* syndromes would produce raised risks of *specific* behaviour that challenges, without some biological component (see Table 2). In Lesch-Nyhan syndrome, for example, it used to be thought that all those with the syndrome showed a very specific behaviour, early self-biting, which frequently was so distinctive, and severe, that it led to the diagnosis, and which often then extended into other forms of serious self-injury. It is now known that in some Lesch-Nyhan variants self-injury does not appear (Jinnah et al., 2010) and so it may be that this will help in finding the exact link between the disorder and the self-injury. Of course, in many syndromes the links between the syndrome and the behaviour are nothing like so specific, and even when there are apparent links, environmental effects are still often present (Bergen et al., 2002; Hall et al., 2001; Langthorne & McGill, 2012; Taylor & Oliver, 2008).

Finally, as regards 'biological causes', there are also a number of conditions that would broadly fall into the 'biological' category that are known to worsen behaviour that challenges, and these include pain and physical illnesses or discomfort. People with a learning disability have more health problems than those without a disability because of a variety of comorbidities, and these health needs are difficult to diagnose, partly because people with a learning disability have associated communication problems. As a result, there have been a number of high-profile reports on the poor health outcomes of people with a disability in the

UK, that have been likened to those of non-disabled people in the developing world (Mencap, 2007); (Michael, 2008); (Heslop et al., 2013).

The relationship between behaviour that challenges, and the person's health needs is complex, and has been studied both in large-scale cross-sectional surveys, often relating to annual health checks (Cooper et al., 2006), and in small-scale single case series (Bosch et al., 1997; Kennedy & O'Reilly, 2006; Peebles & Price, 2012). De Winter and colleagues (2011), in a systematic review of physical health issues and behaviour that challenges, found 45 relevant studies, covering issues as diverse as motor disorders, sensory impairment, epilepsy, gastrointestinal disease, sleep disorders and dementia. They noted the absence of evidence related to infectious diseases, cancer, pulmonary and cardiac disease. They concluded that strong evidence existed for a relationship between visual impairment and self-injurious behaviour, pain in cerebral palsy and problem behaviour, and some evidence for a relationship between both gastrointestinal reflux and poor sleep, and behaviour that challenges. They concluded there was no evidence that epilepsy was related to behaviour that challenges.

2.5.2 Psychosocial causes

Psychosocial causes have probably been investigated more frequently than any other causes and it seems that psychosocial factors have a very widespread influence on behaviour that challenges. Children, young people and adults with a learning disability are among the most stigmatised individuals in society, especially when they show behaviour that challenges. They tend to have very little power and struggle to obtain what they need to make a success of life. The psychosocial factors relevant to behaviour that challenges have been studied in very different ways for different sub-populations, and these are briefly described below. Generally it has been agreed that behaviours are mostly learnt, and the psychosocial environment is crucial to their appearance, escalation, elicitation and extinction.

For people with a severe disability, it appears that behaviour that is challenging for others, is often functional for them, allowing them to control their lives in particular ways, such as gaining sensory stimulation, attracting the attention of carers or staff members, removing demands or gaining tangible items. Essentially, behaviour that challenges, may produce the desired effect by itself, through self-stimulation, or it may 'teach' carers and staff to respond in particular ways through social positive or social negative reinforcement: for instance, if someone is aggressive or self-injurious, carers and staff may well try to meet their needs by taking some action contingent on the behaviour. They may go and speak with the person (a form of social positive reinforcement), offer them food, drink or their favourite toy, activity or tangible item (if made available through social means, this is also a type of social positive reinforcement). Carers and staff may stop asking the person to do a task (the removal of the task negatively reinforces the behaviour) or they may move away to leave the person alone (social negative reinforcement). Essentially, these actions may 'teach' the person with a disability to repeat those behaviours in similar circumstances, in the presence of discriminative stimuli, and at the same time, any cessation in the behaviour may in turn 'teach' carers and staff to use the same strategy next time to stop the behaviour. Stimuli that signal that reinforcers are available act as discriminative stimuli and deprivation states produce motivating operations (Vollmer & Iwata, 1991), accounting for some of the variability of behaviour in different circumstances. Many children, young people and adults who show behaviour that challenges have no speech or very little speech, and it seems that much behaviour that challenges can be seen as functioning like communication for those with very poor language skills, even though they may lack intent.

The discovery of the variety of possible psychosocial functions of behaviour that challenges, in the 1980s and 1990s, led to attempts to match a number of specific behavioural strategies (such as extinction) to the putative functions of behaviour that challenges, in attempts to reduce it. The likelihood of the behaviour serving communicative functions, in turn, led to the development of interventions teaching specific communicative acts (so-called functional

communication training originated by Carr and Durand (1985)), which, it was hypothesised, could replace the function of the behaviour that challenges. In both cases, one of the necessary first steps was to develop a way of analysing the behavioural function of an individual's behaviour, in order to match intervention strategies to the function, and a number of methods of functional behaviour assessment were developed (Lloyd & Kennedy, in press). Very simple analyses could be conducted through the use of ABC charts and scatter plots but these gave a limited amount of information. Functional behaviour assessments began to be developed which involved interviews or questionnaires, conducted with staff or carers, such as the Functional Analysis Interview (O'Neill et al., 1997) and the Behavior Assessment Guide (Willis et al., 1993), the Motivation Assessment Scale (Durand & Crimmins, 1992), the Questions about Behavioral Functioning (Vollmer & Matson, 1995), and the Functional Analysis Screening Tool (Iwata et al., 2013)(FAST, Iwata et al, 2013).

More direct methods of analysing the function of behaviour were also developed: in some cases this involved conducting direct observations of the person in their naturalistic environment, with subsequent sophisticated analysis of data, such as by conditional probabilities (Oliver et al., 2005). In other cases, this was undertaken by experimental functional analysis, involving the use of analogue conditions in which the behaviour of the person was directly assessed, while providing brief periods in which discriminative stimuli and specific reinforcers were deliberately presented, in order to examine which ones set off the behaviour (Iwata et al., 1994). These experimental functional analyses could be lengthy, however, and sometimes inconclusive, such that various adapted methods were developed (Hagopian et al., 2013), including brief versions that could be done at out-patient settings (Northup et al., 1991).

For people with a mild learning disability, these methods of functional behavioural assessment were sometimes more difficult to use, partly because the behaviours occurred less frequently, despite being extremely serious when they did occur (such as, arson or sexually harmful behaviour). According to Didden and colleagues(2006), functional analyses still led to more effective behavioural treatments, though increasingly since then assessments have been adapted for people with a mild learning disability that use self-report rather than carer report (Murphy & Clare, 1995; Novaco & Taylor, 2004); (SOTSEC-ID collaborative, 2010) and intervention methods have increasingly become cognitive-behavioural rather than simply behavioural (Lindsay, 2005)(Lindsay, 2005; SOTSEC-ID, 2010; Willner et al,2013). The influence of psychosocial variables has also broadened to include psychological distress (assessed directly with the person with a learning disability) and cognitive distortions, including those arising from causes such as perceived stigma (Dagnan & Waring, 2004), as well as those arising from abusive experiences (Lindsay, 2005).

2.5.38 Environmental causes

The reliable appearance of much higher rates of behaviour that challenges in certain environments (see section 2.2) led to the proposal that some environments have *such* a major role in causing behaviour that challenges, that we should be intervening with environments and social systems, rather than with individuals, in order to reduce behaviour that challenges. Very high rates of behaviour that challenges have been reported in institutions, which typically entail a relative lack of activities, poorer social support, higher rates of physical interventions and restrictive practices (such as locked doors), and more frequent reports of abusive practices. Very high rates of behaviour that challenge are also associated with poor parenting, particularly with abusive practices. Such practices, of course, do not only occur in institutions and in particular families but may occur in all types of environments at times. McGill (in press) has termed these 'challenging environments' and has developed the concept of the opposite kind of environment: the 'capable' environment, in which good quality care reduces the risk of behaviour that challenges. This approach is inextricably linked with the Positive Behaviour Support (PBS) approach, which developed from applied behavioural approaches, amalgamating these with person-centred planning,

non-aversive methods and quality of life interventions. According to one of the founding fathers of PBS, Ted Carr, PBS is 'an applied science that uses educational and systems change methods to enhance quality of life and minimise problem behavior' (Carr et al., 2002a). According to McGill (in press), the characteristics of the 'capable' environment include positive social interactions, support for communication, support for meaningful activity, provision of predictable and consistent environments, support to establish and maintain relationships with family and friends, provision of choice, encouragement of more independent functioning, support for personal healthcare, an acceptable physical environment, mindful and skilled carers, effective management and staff support, and effective organisational context.

2.61 Current care in the UK

Every area of the country has designated services, intended to provide assessments and interventions for children, young people and adults with a learning disability and behaviour that challenges. However, in the past, especially for children, these services have been fragmented and at times ineffective and unresponsive to family needs, to the point sometimes of being abusive (Mencap, 2013). Typically, for children and young people with behaviour that challenges, services have been provided within education (through their school and the educational psychology service), as well as through generic child and adolescent mental health services (CAMHS). CAMHS are run by the NHS and consist of a variety of professionals (such as nurses, psychologists, psychiatrists, occupational therapists and speech and language therapists), seeing any local children and young people with mental health needs (considered to include behaviour that challenges), not just those with disabilities. In some CAMHS teams, there have been professionals (usually clinical psychologists) who specialise in seeing children and young people with a learning disability; occasionally, in some part s of the country, there are completely separate teams with a full range of allied health professionals for children and young people with a learning disability. Social workers meanwhile have operated in yet other teams: the Child in Need teams for any child with a disability, and children and families (including child protection) teams for those children at risk. Families find the number of unrelated services bewildering and report that it is all too easy to find that none of them will offer help. Moreover there are very few early intervention services routinely available for children with a learning disability. The government's *Joint Improvement Programme* following the *Winterbourne View* scandal and the new *Children and Families Bill* aim to improve this fragmented situation by requiring improved commissioning of better services at all levels, and by legislating that all children with disabilities must have an Education, Health & Care plan and ensuring that local authorities (education and social care) and health work together.

In the past, referral pathways for children with a learning disability, who were showing behaviour that challenges, have been very complex. At school, when behaviour that challenges began to emerge, the schools provided individual educational plans and they sometimes sought the advice of an educational psychologist. Where the behaviour also occurred at home, schools provided support for families through a family-liaison worker, but this was unlikely to involve more than 1 visit per term. Many families would therefore seek help elsewhere, such as from their local general practitioner (GP). The GP could refer them either to their local paediatrician (usually for younger children) or to their local CAMHS team. The professional most likely to provide assessment and treatment for behaviour that challenges, in either case, would be the psychologist, who would typically visit and assess the child at school and at home, and construct an intervention that would aim to be effective across home and school. Other professionals likely to be involved included speech and language therapists, occupational therapist and nurses, each of whom may contribute to part of the assessment and intervention. In practice, however, families of the children with severe behaviour that challenges frequently found generic CAMHS teams workers insufficiently expert, and even unhelpful, and if the school placement also broke down, the families often

ended up being told that their son or daughter had to be placed in a residential placement many miles from the family home (McGill et al., 2006b).

Meanwhile for adults, in all areas, there are community learning disability teams (CLDTs), again consisting of a variety of professionals, typically learning disability nurses, psychologists, psychiatrists, occupational therapists, physiotherapists and speech and language therapists, all working as a team. In many areas, social workers are co-located and integrated into the CLDTs. However, in some areas they are located at separate social services offices, so that there is effectively an NHS-based and social services-based CLDT, which is unhelpful. For adults with a learning disability, their day services, or their residential/supported living service (if they are no longer living with families), may first try to deal with behaviour that challenges themselves (many independent day/residential services now employ their own 'challenging behaviour workers'). These services may refer them to the CLDT if they continue to show behaviour that challenges and/or their families may also access the CLDT through the local GP or other agencies. Again, the most likely professional to work with them is the psychologist but speech and language therapists and occupational therapists may be involved, and many teams also have behaviourally trained nurses and 'challenging behaviour support workers' (who would typically work under the supervision of psychologists).

For both children and adults, the CAMHS or CLDT team psychiatrists may also provide assessments and interventions, when the person with a learning disability is thought to have underlying mental health needs. Good practice would involve joint working by psychologist, psychiatrists, speech and language therapists and others, as described in the RCP/BPS document *A Unified Approach* (2007). However, for adults, as for children, with behaviour that challenges, the experience of carers has too often been that there is insufficient support from professionals, who are often not expert enough, providing help that arrives too late (or even never), that is poorly coordinated (Griffith & Hastings, 2013), and that where services and /or families cannot cope, the likely outcomes may include over-medication of the individual with a learning disability, disengagement by professionals, and eventually 'out of area' placements, often very far removed from families, some with restrictive practices and very high costs (many 'assessment and treatment' units cost in the region of £250,000 per person per year). As a result of such experiences the *Challenging Behaviour Foundation* drew up a charter of *Rights and Values* and *Actions to be Taken*, to better support families and people with a learning disability whose behaviour is said to be challenging (see www.challengingbehaviour.org.uk/learning-disability-files/CBF-Charter-2013).

The events at *Winterbourne View* reflect the kinds of dislocation and poor quality of services that occur all too often for children, young people and adults with a learning disability whose behaviour challenges services, with restrictive practices replacing any kind of positive assessment or intervention. As part of the Government's response to Winterbourne View (*Transforming Care: A national response to Winterbourne View Hospital)*(Department of Health, 2012) there was a resolve to improve commissioning and the Joint Implementation Team has now produced a draft of *Core Principles Commissioning Tool* to be used for the development of local specifications for services supporting children, young people, adults and older people with a learning disability and / or autism who display or are at risk of displaying behaviour that challenges. This, alongside the proposed '*Education, Health and Care Plans*' for all people younger than 25 years identified with Special Educational Needs (specified in the *Children and Families Bill*), better transition to adult services, which is the focus of the *Preparing for Adulthood Programme*, personal health budgets which will be available to those in receipt of continuing healthcare, and better integration of services, are intended by the Government to improve services for all people with a learning disability and behaviour that challenges.

2.71 Economic costs

Behaviour that challenges exhibited by people with learning difficulties can place an additional strain on resources across a range of budgets. Given the diverse sectors of society in which care and support is provided for people with learning difficulties, additional financial costs may be borne by families, charities, local or national governments. Though the link between behaviour that challenges and resource use makes strong intuitive sense little data exists to explore and quantify the association in the UK.

In an attempt to quantify the financial impact of psychiatric and neurological issues in the UK, Fineberg and colleagues (2013) found learning disabilities to be the tenth most costly issue costing €5975 million (2010 prices). The study took into account productivity losses and direct non-medical costs though it did not link the costs associated with learning disabilities to behaviour that challenges.

A number of studies have assessed the predictors and costs of out-of-area placements for people with a learning disability and behaviour that challenges in the UK, as out-of-area placements are often perceived as one of the most substantial cost elements of care provided to this population. Predictors of out-of-area placements include young age, behaviours resulting in physical injury to self, staff or others and exclusion from service settings, a history of formal detention under the mental health act, the presence of mental health problems, a diagnosis of autism, a higher total score on the Adaptive Behaviour Scale and multiple health problems (Allen et al., 2007; Hassiotis et al., 2008). In contrast to the perception that out-of-area placements impose considerable costs to the public purse, research shows that out-of-area placements have in fact similar or lower costs compared with within-area placements for people with a learning disability and behaviour that challenges (Allen et al., 2007; Hassiotis et al., 2008; Perry et al., 2013).

In order to investigate the relationship between service costs and the severity of behaviour that challenges, Knapp and colleagues (2005)analysed data on characteristics and service receipt from 1,120 people with a learning disability and behaviour that challenges living in residential accommodation, and found a complex relationship between cost, severity of learning disabilities and levels of behaviour that challenges. At moderate levels of learning disability a linear relationship with service costs was observed. At higher levels of learning disability this relationship appeared to decrease but costs remained higher for people who exhibited more severe behaviour that challenges. The largest component of service costs was, as anticipated, accommodation, accounting for 85% of the total cost. Service costs tended to be higher in NHS settings (including long-stay hospital settings, hostels and NHS-provided residential care in ordinary housing) compared with private and voluntary settings. However, people living in NHS settings scored more highly on both learning disability and behaviour that challenges indicators, which may partly explain the higher costs in NHS settings

Doran (2012) used self-completed questionnaires to estimate the cost of learning disabilities to both families and the government in Australia. This was reported to reach $14,720 billion annually (AUS$, 2006 prices). Though the independent impact of behaviour that challenges on resource use was not estimated in the study, components of financial cost such as replacing broken toys/furniture and respite care were highlighted as associated with the occurrence of behaviour that challenges. The study reported that families carry the majority of the financial burden and are insufficiently compensated by the government, with an annual net loss per family of approximately $37,000 and $58,000 for mild and severe/profound learning disabilities, respectively.

Using the same Australian data set Einfeld and colleagues (2010)investigated the relationship between patient characteristics as measured by the demographic behavioural checklist and the costs associated with behaviour that challenges in people with a learning disability. The aggregate outcome of total behavioural problem score was significantly related

to both direct costs (replacing damaged toys, expenses for care) and opportunity costs (reduced time in employment to provide care) to families. Disruptive and self-absorbed behaviour (which includes self-injury) subscales were statistically related to out of pocket and opportunity costs respectively.

Though measurement of the independent financial effect of behaviour that challenges could not be carried out, these studies illustrate the link between behaviour that challenges and the distribution of these costs in society.

In addition to the measured financial impacts, it is acknowledged that intangible costs represent a significant component of burden that is not possible to capture (Doran et al., 2012). Among others these costs include loss of both role performance and social participation.

Although it is difficult to quantify the contribution of behaviour that challenges to the costs associated with learning disabilities this is likely to be substantial. As these financial costs are borne by a variety of stakeholders, public policy must be devised and applied sensitively to responsibly provide value for service users, families and society in general.

3 Methods used to develop this guideline

3.1 Overview

The development of this guideline followed *The Guidelines Manual* (NICE, 2012). A team of health and social care professionals, lay representatives and technical experts known as the Guideline Development Group (GDG), with support from the NCCMH staff, undertook the development of a person-centred, evidence-based guideline. There are 7 basic steps in the process of developing a guideline:

1. Define the scope, which lays out exactly what will be included (and excluded) in the guidance.
2. Define review questions that cover all areas specified in the scope.
3. Develop a review protocol for each systematic review, specifying the search strategy and method of evidence synthesis for each review question.
4. Synthesise data retrieved, guided by the review protocols.
5. Produce evidence profiles and summaries using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) system.
6. Consider the implications of the research findings for clinical practice and reach consensus decisions on areas where evidence is not found.
7. Answer review questions with evidence-based recommendations for clinical practice.

The clinical practice recommendations made by the GDG are therefore derived from the most up-to-date and robust evidence for the clinical and cost effectiveness of the interventions and services covered in the scope. Where evidence was not found or was inconclusive, the GDG adopted both formal and informal methods to reach consensus on what should be recommended, factoring in any relevant issues. In addition, to ensure a service user and carer focus, the concerns of service users and carers regarding health and social care have been highlighted and addressed by recommendations agreed by the whole GDG.

3.2 The scope

Topics are referred by the Secretary of State and the letter of referral defines the remit, which defines the main areas to be covered (see *The Guidelines Manual* (NICE, 2012) for further information). The NCCMH developed a scope for the guideline based on the remit (see Appendix A). The purpose of the scope is to:

- provide an overview of what the guideline will include and exclude
- identify the key aspects of care that must be included
- set the boundaries of the development work and provide a clear framework to enable work to stay within the priorities agreed by NICE and the National Collaborating Centre, and the remit from the Department of Health/Welsh Assembly Government
- inform the development of the review questions and search strategy
- inform professionals and the public about expected content of the guideline
- keep the guideline to a reasonable size to ensure that its development can be carried out within the allocated period.

An initial draft of the scope was sent to registered stakeholders who had agreed to attend a scoping workshop. The workshop was used to:

- obtain feedback on the selected key clinical issues
- identify which population subgroups should be specified (if any)
- seek views on the composition of the GDG

- encourage applications for GDG membership.

The draft scope was subject to consultation with registered stakeholders over a 4-week period. During the consultation period, the scope was posted on the NICE website (www.nice.org.uk). Comments were invited from stakeholder organisations The NCCMH and NICE reviewed the scope in light of comments received, and the revised scope was signed off by NICE.

3.3 The Guideline Development Group

During the consultation phase, members of the GDG were appointed by an open recruitment process. GDG membership consisted of: professionals in psychiatry, clinical psychology, nursing, social work, speech and language therapy, and general practice; academic experts in psychiatry and psychology; commissioning managers; and carers and representatives from service user and carer organisations. The guideline development process was supported by staff from the NCCMH, who undertook the clinical and health economic literature searches, reviewed and presented the evidence to the GDG, managed the process, and contributed to drafting the guideline.

3.3.1 Guideline Development Group meetings

Eleven GDG meetings were held between July 2013 and February 2015. During each day-long GDG meeting, in a plenary session, review questions and clinical and economic evidence were reviewed and assessed, and recommendations formulated. At each meeting, all GDG members declared any potential conflicts of interest (see Appendix B), and service user and carer concerns were routinely discussed as a standing agenda item.

3.3.2 Service users and carers

Individuals with direct experience of services gave an integral service-user focus to the GDG and the guideline. The GDG included carers and a representative of a national service user group. They contributed as full GDG members to writing the review questions, providing advice on outcomes most relevant to service users and carers, helping to ensure that the evidence addressed their views and preferences, highlighting sensitive issues and terminology relevant to the guideline, and bringing service user research to the attention of the GDG. In drafting the guideline, they met with the NCCMH team on several occasions to develop the chapter on experience of care and they contributed to writing the guideline's introduction and identified recommendations from the service user and carer perspective.

3.3.3 Expert advisers

Expert advisers, who had specific expertise in one or more aspects of treatment and management relevant to the guideline, assisted the GDG, commenting on specific aspects of the developing guideline and making presentations to the GDG. Appendix C lists those who agreed to act as expert advisers.

3.3.4 National and international experts

National and international experts in the area under review were identified through the literature search and through the experience of the GDG members. These experts were contacted to identify unpublished or soon-to-be published studies, to ensure that up-to-date evidence was included in the development of the guideline. They informed the GDG about completed trials at the pre-publication stage, systematic reviews in the process of being published, studies relating to the cost effectiveness of treatment and trial data if the GDG could be provided with full access to the complete trial report. Appendix E lists researchers who were contacted.

3.41 Review protocols

2 Review questions drafted during the scoping phase were discussed by the GDG at the first
3 few meetings and amended as necessary. The review questions were used as the starting
4 point for developing review protocols for each systematic review (described in more detail
5 below). Where appropriate, the review questions were refined once the evidence had been
6 searched and, where necessary, sub-questions were generated. The final list of review
7 questions can be found in Appendix F.

8 For questions about interventions, the PICO (Population, Intervention, Comparison and
9 Outcome) framework was used to structure each question (see Table 3).

10 **Table 3: Features of a well-formulated question on the effectiveness of an intervention**
11 **– PICO**

Population:	Which population of service users are we interested in? How can they be best described? Are there subgroups that need to be considered?
Intervention:	Which intervention, treatment or approach should be used?
Comparison:	What is/are the main alternative/s to compare with the intervention?
Outcome:	What is really important for the service user? Which outcomes should be considered: intermediate or short-term measures; mortality; morbidity and treatment complications; rates of relapse; late morbidity and readmission; return to work, physical and social functioning and other measures such as quality of life; general health status?

12 Questions relating to case identification and assessment tools and methods do not involve
13 an intervention designed to treat a particular condition, and therefore the PICO framework
14 was not used. Rather, the questions were designed to pick up key issues specifically relevant
15 to clinical utility, for example their accuracy, reliability, safety and acceptability to the service
16 user.

17 In some situations, the prognosis of a particular condition is of fundamental importance, over
18 and above its general significance in relation to specific interventions. Areas where this is
19 particularly likely to occur relate to assessment of risk, for example in terms of behaviour
20 modification or screening and early intervention. In addition, review questions related to
21 issues of service delivery are occasionally specified in the remit from the Department of
22 Health/Welsh Assembly Government. In these cases, appropriate review questions were
23 developed to be clear and concise.

24 Where review questions about service user experience were specified in the scope, the
25 SPICE format was used to structure the questions (Table 4).

26 **Table 4: Features of a well-formulated question about the experience of care**
27 **(qualitative evidence) – SPICE**

Setting	Where? In what context?
Perspective	For who?
Intervention (phenomenon of interest):	Which intervention/interest should be included?
Comparison:	What?
Evaluation:	How well? What result?
Adapted from (Booth, 2003)	

For each topic, addressed by one or more review questions, a review protocol was drafted by the technical team using a standardised template (based on PROSPERO[a]), review and agreed by the GDG (all protocols are included in Appendix F).

To help facilitate the literature review, a note was made of the best study design type to answer each question. There are 4 main types of review question of relevance to NICE guidelines. These are listed in Table 5. For each type of question, the best primary study design varies, where 'best' is interpreted as 'least likely to give misleading answers to the question'. For questions about the effectiveness of interventions, where randomised controlled trials (RCTs) were not available, the review of other types of evidence was pursued only if there was reason to believe that it would help the GDG to formulate a recommendation.

However, in all cases, a well-conducted systematic review (of the appropriate type of study) is likely to always yield a better answer than a single study.

Table 5: Best study design to answer each type of question

Type of question	Best primary study design
Effectiveness or other impact of an intervention	Randomised controlled trial (RCT); other studies that may be considered in the absence of RCTs are the following: internally/externally controlled before and after trial, interrupted time-series
Accuracy of information (for example, risk factor, test, prediction rule)	Comparing the information against a valid gold standard in an RCT or inception cohort study
Rates (of disease, service user experience, rare side effects)	Prospective cohort, registry, cross-sectional study
Experience of care	Qualitative research (for example, grounded theory, ethnographic research)

3.5 Clinical review methods

The aim of the clinical literature review was to systematically identify and synthesise relevant evidence from the literature in order to answer the specific review questions developed by the GDG. Thus, clinical practice recommendations are evidence-based, where possible, and, if evidence is not available, informal consensus methods are used to try and reach general agreement between GDG members (see Section 3.3.1) and the need for future research is specified.

3.5.1 The search process

3.5.1.1 Scoping searches

A broad preliminary search of the literature was undertaken in April 2013 to obtain an overview of the issues likely to be covered by the scope, and to help define key areas. The searches were restricted to clinical guidelines, Health Technology Assessment (HTA) reports, key systematic reviews and RCTs. A list of databases and websites searched can be found in Appendix H.

3.5.1.2 Systematic literature searches

After the scope was finalised, a systematic search strategy was developed to locate as much relevant evidence as possible. The balance between sensitivity (the power to identify all studies on a particular topic) and specificity (the ability to exclude irrelevant studies from the

[a] http://www.crd.york.ac.uk/prospero/

results) was carefully considered, and a decision made to utilise a broad approach to searching to maximise retrieval of evidence to all parts of the guideline. Searches were restricted to certain study designs if specified in the review protocol, and conducted in the following databases:

- Applied Social Sciences Index and Abstracts (ASSIA)
- Australian Education Index (AEI)
- British Education Index
- CINAHL
- Cochrane Database of Abstracts of Reviews of Effects (DARE)
- Cochrane Database of Systematic Reviews (CDSR)
- CENTRAL
- Education Resources Information Center (ERIC)
- Embase
- HTA database (technology assessments)
- International Bibliography of the Social Sciences (IBSS)
- MEDLINE/MEDLINE In-Process
- Psychological Information Database (PsycINFO)
- Sociological Abstracts
- Social Services Abstracts
- Social Sciences Citation Index (SSCI)

The search strategies were initially developed for MEDLINE before being translated for use in other databases/interfaces. Strategies were built up through a number of trial searches and discussions of the results of the searches with the review team and GDG to ensure that all possible relevant search terms were covered. In order to assure comprehensive coverage, search terms for CBLD were kept purposely broad to help counter dissimilarities in database indexing practices and thesaurus terms, and imprecise reporting of study populations by authors in the titles and abstracts of records. The search terms for each search are set out in full in Appendix H.

3.5.1.3 Reference Management

Citations from each search were downloaded into reference management software and duplicates removed. Records were then screened against the eligibility criteria of the reviews before being appraised for methodological quality (see below). The unfiltered search results were saved and retained for future potential re-analysis to help keep the process both replicable and transparent.

3.5.1.4 Search filters

To aid retrieval of relevant and sound studies, filters were used to limit a number of searches to systematic reviews, randomised controlled trials and qualitative studies. The search filters for systematic reviews and randomised controlled trials are adaptations of validated filters designed by the Health Information Research Unit (HIRU) at McMaster University. The qualitative research filter was developed in-house. Each filter comprises index terms relating to the study type(s) and associated text words for the methodological description of the design(s).

3.5.1.5 Date and language restrictions

Systematic database searches were initially conducted in August 2013 up to the most recent searchable date. Search updates were generated on a 6-monthly basis, with the final re-runs

carried out in October 2014 ahead of the guideline consultation. After this point, studies were only included if they were judged by the GDG to be exceptional (for example, if the evidence was likely to change a recommendation).

Although no language restrictions were applied at the searching stage, foreign language papers were not requested or reviewed, unless they were of particular importance to a review question.

Date restrictions were not applied, except for searches of systematic reviews which were limited to research published from 1999. The search for systematic reviews was restricted to the last 15 years as older reviews were thought to be less useful.

3.5.1.6 Other search methods

Other search methods involved: (a) scanning the reference lists of all eligible publications (systematic reviews, stakeholder evidence and included studies) for more published reports and citations of unpublished research; (b) sending lists of studies meeting the inclusion criteria to subject experts (identified through searches and the GDG) and asking them to check the lists for completeness, and to provide information of any published or unpublished research for consideration (see Appendix E); (c) checking the tables of contents of key journals for studies that might have been missed by the database and reference list searches; (d) tracking key papers in the Science Citation Index (prospectively) over time for further useful references; (e) conducting searches in ClinicalTrials.gov for unpublished trial reports; (f) contacting included study authors for unpublished or incomplete datasets. Searches conducted for existing NICE guidelines were updated where necessary. Other relevant guidelines were assessed for quality using the AGREE instrument (AGREE Collaboration, 2003). The evidence base underlying high-quality existing guidelines was utilised and updated as appropriate.

Full details of the search strategies and filters used for the systematic review of clinical evidence are provided in Appendix H.

3.5.1.7 Study selection and assessment of methodological quality

All primary-level studies included after the first scan of citations were acquired in full and re-evaluated for eligibility at the time they were being entered into the study information database. More specific eligibility criteria were developed for each review question and are described in the relevant clinical evidence chapters. Eligible systematic reviews and primary-level studies were critically appraised for methodological quality (risk of bias) using a checklist (see *The Guidelines Manual* (NICE, 2012) for templates). The eligibility of each study was confirmed by at least 1 member of the GDG.

For some review questions, it was necessary to prioritise the evidence with respect to the UK context (that is, external validity). To make this process explicit, the GDG took into account the following factors when assessing the evidence:

- participant factors (for example, gender, age and ethnicity)
- provider factors (for example, model fidelity, the conditions under which the intervention was performed and the availability of experienced staff to undertake the procedure)
- cultural factors (for example, differences in standard care and differences in the welfare system).

It was the responsibility of the GDG to decide which prioritisation factors were relevant to each review question in light of the UK context.

Unpublished evidence

Stakeholders were invited to submit any relevant unpublished data using the call for evidence process set out in the NICE manual (NICE, 2012). Additionally, authors and principal investigators were approached for unpublished evidence. The GDG used a number of criteria when deciding whether or not to accept unpublished data. First, the evidence must have been accompanied by a trial report containing sufficient detail to properly assess risk of bias. Second, the evidence must have been submitted with the understanding that data from the study and a summary of the study's characteristics would be published in the full guideline. Therefore, in most circumstances the GDG did not accept evidence submitted 'in confidence'. However, the GDG recognised that unpublished evidence submitted by investigators might later be retracted by those investigators if the inclusion of such data would jeopardise publication of their research.

Experience of care

Reviews were sought of qualitative studies that used relevant first-hand experiences of service users and their families, partners or carers. A particular outcome was not specified by the GDG. Instead, the review was concerned with narrative data that highlighted the experience of care.

Data extraction

Quantitative analysis

Study characteristics, aspects of methodological quality, and outcome data were extracted from all eligible studies, using Review Manager Version 5.3 (Cochrane Collaboration, 2014) and an Excel-based form (see Appendix K).

In most circumstances, for a given outcome (continuous and dichotomous), where more than 50% of the number randomised to any group were missing or incomplete, the study results were excluded from the analysis (except for the outcome 'leaving the study early', in which case, the denominator was the number randomised). Where there were limited data for a particular review, the 50% rule was not applied. In these circumstances the evidence was downgraded (see section 3.5.5).

Where possible, outcome data from an intention-to-treat analysis (ITT) (that is, a 'once-randomised-always-analyse' basis) were used. Where ITT had not been used or there were missing data, the effect size for dichotomous outcomes were recalculated using worse-case scenarios. Where conclusions varied between scenarios, the evidence was downgraded (see section 3.5.5).

Where some of the studies failed to report standard deviations (for a continuous outcome), and where an estimate of the variance could not be computed from other reported data or obtained from the study author, the following approach was taken.[b] When the number of studies with missing standard deviations was less than one-third and when the total number of studies was at least 10, the pooled standard deviation was imputed (calculated from all the other studies in the same meta-analysis that used the same version of the outcome measure). In this case, the appropriateness of the imputation was made by comparing the standardised mean differences (SMDs) of those trials that had reported standard deviations against the hypothetical SMDs of the same trials based on the imputed standard deviations. If they converged, the meta-analytical results were considered to be reliable.

[b] Based on the approach suggested by Furukawa and colleagues (2006).

When the conditions above could not be met, standard deviations were taken from another related systematic review (if available). In this case, the results were considered to be less reliable.

The meta-analysis of survival data, such as time to any mood episode, was based on log hazard ratios and standard errors. Since individual participant data were not available in included studies, hazard ratios and standard errors calculated from a Cox proportional hazard model were extracted. Where necessary, standard errors were calculated from confidence intervals (CIs) or *p* value according to standard formulae (see the Cochrane Reviewers' Handbook 5.1.0 (Higgins & Green, 2011)). Data were summarised using the generic inverse variance method using Review Manager.

Consultation with another reviewer or members of the GDG was used to overcome difficulties with coding. Data from studies included in existing systematic reviews were extracted independently by 1 reviewer and cross-checked with the existing dataset. Where possible, 2 independent reviewers extracted data from new studies. Where double data extraction was not possible, data extracted by 1 reviewer was checked by the second reviewer. Disagreements were resolved through discussion. Where consensus could not be reached, a third reviewer or GDG members resolved the disagreement. Masked assessment (that is, blind to the journal from which the article comes, the authors, the institution and the magnitude of the effect) was not used since it is unclear that doing so reduces bias (Berlin, 2001; Jadad et al., 1996).

3.5.3 Single-case and small-n studies

Single-case and small-n (SCSn) studies (also known as N of 1 trials) make up a substantial part of the empirical evidence that is published in the field of learning disabilities. Unlike group-studies that present aggregated data for a group of participants that received either treatment or control, SCSn studies report effectiveness data for each participant separately. The approach uses a process of repeated observation during a certain period of time which allows for the assessment of change in targeted behaviours under different treatments of at least 1 independent variable (Onghena, 2005). Experimental designs typically follow an ABA withdrawal format whilst quasi-experimental designs follow an AB format. The primary strengths of the SCSn design are the analysis of behaviour of a single case, the assessment of the both the process and product of change and the allowance of complex analysis in to the particular characteristics of 'responders' and 'non responders' (Horner et al., 2005). Limitations of the SCSn design include publication bias, carry-over and order effects, irreversibility and the generalisability of results. However, by aggregating the results from several SCSn studies in a meta-analysis generalisability becomes more feasible (Van den Noortgate & Onghena, 2007; Van den Noortgate & Onghena, 2008).

The frequent use of SCSn designs in the field of learning disabilities contrasts with the limited use of the RCT to evaluate treatment effects. Recruitment, ethical considerations and obtaining consent to randomisation have all contributed to a limitation of RCTs and other group comparison methods.

3.5.4 Evidence synthesis

The method used to synthesise evidence depended on the review question and availability and type of evidence (see Appendix F for full details). Briefly, for questions about the psychometric properties of instruments, reliability, validity and clinical utility were synthesised narratively based on accepted criteria. For questions about test accuracy, bivariate test accuracy meta-analysis was conducted where appropriate. For questions about the effectiveness of interventions, standard meta-analysis or network meta-analysis was used where appropriate, otherwise narrative methods were used with clinical advice from the GDG. In the absence of high-quality research, formal and informal consensus process were used (see 3.5.8).

3.5.5 Grading the quality of evidence

For questions about the effectiveness of interventions, the GRADE approach[c] was used to grade the quality of evidence from group comparisons for each outcome (Guyatt et al., 2011). Evidence from systematic reviews of SCSn designs was graded as 'low' or 'very low' quality without using the formal GRADE approach because specific methodology has not been developed to grade this type of evidence (see section 3.5.3 for limitations, which account for the low or very low quality grade). For questions about the experience of care and the organisation and delivery of care, methodology checklists (see section 3.5.1) were used to assess the risk of bias, and this information was taken into account when interpreting the evidence. The technical team produced GRADE evidence profiles (see below) using GRADEprofiler (GRADEpro) software (Version 3.6), following advice set out in the GRADE handbook (Schünemann et al., 2009). All staff doing GRADE ratings were trained, and calibration exercises were used to improve reliability (Mustafa et al., 2013).

3.5.5.1 Evidence profiles

A GRADE evidence profile was used to summarise both the quality of the evidence and the results of the evidence synthesis for each 'critical' and 'important' outcome (see Appendix O for completed evidence profiles). The GRADE approach is based on a sequential assessment of the quality of evidence, followed by judgment about the balance between desirable and undesirable effects, and subsequent decision about the strength of a recommendation.

Within the GRADE approach to grading the quality of evidence, the following is used as a starting point:

- RCTs without important limitations provide high quality evidence
- observational studies without special strengths or important limitations provide low quality evidence.

For each outcome, quality may be reduced depending on 5 factors: limitations, inconsistency, indirectness, imprecision and publication bias. For the purposes of the guideline, each factor was evaluated using criteria provided in Table 6.

For observational studies without any reasons for down-grading, the quality may be up-graded if there is a large effect, all plausible confounding would reduce the demonstrated effect (or increase the effect if no effect was observed), or there is evidence of a dose-response gradient (details would be provided under the 'other' column).

Each evidence profile includes a summary of findings: number of participants included in each group, an estimate of the magnitude of the effect, and the overall quality of the evidence for each outcome. Under the GRADE approach, the overall quality for each outcome is categorised into 1 of 4 groups (high, moderate, low, very low).

Table 6: Factors that decrease quality of evidence

Factor	Description	Criteria
Limitations	Methodological quality/ risk of bias.	Serious risks across most studies (that reported a particular outcome). The evaluation of risk of bias was made for each study using NICE methodology checklists (see Section 3.5.1).
Inconsistency	Unexplained heterogeneity of results.	Moderate or greater heterogeneity (see Appendix O for further information about how this was evaluated)
Indirectness	How closely the outcome	If the comparison was indirect, or if the question

[c] For further information about GRADE, see www.gradeworkinggroup.org

Factor	Description	Criteria
	measures, interventions and participants match those of interest.	being addressed by the GDG was substantially different from the available evidence regarding the population, intervention, comparator, or an outcome.
Imprecision	Results are imprecise when studies include relatively few patients and few events and thus have wide confidence intervals around the estimate of the effect.	If either of the following 2 situations were met: • the optimal information size (for dichotomous outcomes, OIS = 300 events; for continuous outcomes, OIS = 400 participants) was not achieved • the 95% confidence interval around the pooled or best estimate of effect included both 1) no effect and 2) appreciable benefit or appreciable harm
Publication bias	Systematic underestimate or an overestimate of the underlying beneficial or harmful effect due to the selective publication of studies.	Evidence of selective publication. This may be detected during the search for evidence, or through statistical analysis of the available evidence.

3.5.61 Presenting evidence to the Guideline Development Group

Study characteristics tables and, where appropriate, forest plots generated with Review Manager Version 5.2 and GRADE summary of findings tables (see below) were presented to the GDG.

Where meta-analysis was not appropriate and/ or possible, the reported results from each primary-level study were reported in the study characteristics table and presented to the GDG. The range of effect estimates were included in the GRADE profile, and where appropriate, described narratively.

3.5.6.19 Summary of findings tables

Summary of findings tables generated from GRADEpro were used to summarise the evidence for each outcome and the quality of that evidence (Table 7). The tables provide illustrative comparative risks, especially useful when the baseline risk varies for different groups within the population.

Table 7: Example of a GRADE summary of findings table

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Any control**	Corresponding risk **Cognitive behavioural intervention**			
Carer health and wellbeing (depression) - post-treatment		The mean carer health and wellbeing (depression) - post-treatment in the intervention groups was **0.35 standard deviations lower** (0.54 to 0.15 lower)		428 (5 studies)	**Moderate**[1]
Carer health and wellbeing (depression) - follow-up Follow-up: 46 to 104 weeks		The mean carer health and wellbeing (depression) - follow-up in the intervention groups was **0.41 standard deviations lower** (0.79 to 0.04 lower)		130 (2 studies)	**low**[1,2]
Carer health and wellbeing (clinically depressed) - post-treatment	**224 per 1000**	**56 per 1000** (18 to 188)	**RR 0.25** (0.08 to 0.84)	111 (1 study)	**very low**[1,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The

corresponding risk (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

Note. CI = Confidence interval; RR = Risk ratio.

[1] Most information is from studies at moderate risk of bias
[2] Optimal information size not met
[3] Optimal information size not met; small, single study.

3.5.72 Extrapolation

When answering review questions, if there is no direct evidence from a primary dataset,[d] based on the initial search for evidence, it may be appropriate to extrapolate from another data set. In this situation, the following principles were used to determine when to extrapolate:

- a primary dataset is absent, of low quality or is judged to be not relevant to the review question under consideration, and
- a review question is deemed by the GDG to be important, such that in the absence of direct evidence, other data sources should be considered, and
- non-primary data source(s) is in the view of the GDG available, which may inform the review question.

When the decision to extrapolate was made, the following principles were used to inform the choice of the non-primary dataset:

- the populations (usually in relation to the specified diagnosis or problem which characterises the population) under consideration share some common characteristic but differ in other ways, such as age, gender or in the nature of the disorder (for example, a common behavioural problem; acute versus chronic presentations of the same disorder) , and
- the interventions under consideration in the view of the GDG have 1 or more of the following characteristics:
  - share a common mode of action (for example, the pharmacodynamics of drug; a common psychological model of change - operant conditioning)
  - be feasible to deliver in both populations (for example, in terms of the required skills or the demands of the health care system)
  - share common side effects/harms in both populations, and
- the context or comparator involved in the evaluation of the different datasets shares some common elements which support extrapolation, and
- the outcomes involved in the evaluation of the different datasets shares some common elements which support extrapolation (for example, improved mood or a reduction in behaviour that challenges).

When the choice of the non-primary dataset was made, the following principles were used to guide the application of extrapolation:

- the GDG should first consider the need for extrapolation through a review of the relevant primary dataset and be guided in these decisions by the principles for the use of extrapolation
- in all areas of extrapolation datasets should be assessed against the principles for determining the choice of datasets. In general the criteria in the 4 principles set out above for determining the choice should be met

[d] A primary data set is defined as a data set which contains evidence on the population and intervention under review

- in deciding on the use of extrapolation, the GDG will have to determine if the extrapolation can be held to be reasonable, including ensuring that:
  - the reasoning behind the decision can be justified by the clinical need for a recommendation to be made
  - the absence of other more direct evidence, and by the relevance of the potential dataset to the review question can be established
  - the reasoning and the method adopted is clearly set out in the relevant section of the guideline.

3.5.8 Method used to answer a review question in the absence of appropriately designed, high-quality research

In the absence of appropriately designed, high-quality research (including indirect evidence where it would be appropriate to use extrapolation), both formal and informal consensus processes were adopted.

3.5.8.1 Formal method of consensus

The modified nominal group technique (Bernstein et al., 1992)was chosen due to its suitability within the guideline development process. The method is concerned with deriving a group decision from a set of expert individuals and has been identified as the method most commonly used for the development of consensus in health care (Murphy et al., 1998).

In round 1, members were presented with an overview of the modified nominal group technique, a short summary of the available evidence, a consensus questionnaire and a covering letter giving instructions and definitions. Members were asked to rate their agreement with the statements taking into account the available evidence and their clinical expertise. Ratings were made using a 9-point scale, when 1 represented least agreement (that is, the strategy was not appropriate) and 9 most agreement (that is, the strategy was appropriate).

At the subsequent GDG (round 2), anonymised distributions of responses to each statement were given to all members, together with members additional comments and the ranking of statements based on consensus percentage. Those statements in the top half of the ranking table were discussed and recommendations developed from them.

Table 8: Definition of agreement within the consensus panel

Agreement	Definition
100% consensus	Ratings of all 16 members fall within a single 3-point region, i.e. 1–3 (inappropriate strategy), 4–6 (equivocal) or 7–9 (appropriate strategy)
Less than 100% consensus but greater than 75% consensus	For the GDG group of 16 members, the ratings of at least 12 members must lie within the 3-point region of consensus (1–3 or 7–9).
No consensus	Any distribution of ratings outside the limits described above was regarded as no consensus

3.5.8.2 Informal method of consensus

The informal consensus process involved a group discussion of what is known about the issues. The views of GDG were synthesised narratively by a member of the review team, and circulated after the meeting. Feedback was used to revise the text, which was then included in the appropriate evidence review chapter.

3.6 Health economics methods

The aim of the health economics was to contribute to the guideline's development by providing evidence on the cost effectiveness of interventions for people with a learning disability and behaviour that challenges covered in the guideline. This was achieved by:

- systematic literature review of existing economic evidence
- decision-analytic economic modelling.

Systematic reviews of economic literature were conducted in all areas covered in the guideline. Economic modelling was undertaken in areas with likely major resource implications, where the current extent of uncertainty over cost effectiveness was significant and economic analysis was expected to reduce this uncertainty, in accordance with *The Guidelines Manual* (NICE, 2012). Prioritisation of areas for economic modelling was a joint decision between the Health Economist and the GDG. The rationale for prioritising review questions for economic modelling was set out in an economic plan agreed between NICE, the GDG, the Health Economist and the other members of the technical team. The following economic questions were selected as key issues that were addressed by economic modelling:

- parent training for the management of behaviour that challenges in children and young people with a learning disability
- psychological and pharmacological interventions for the management of sleep problems in children and young people with a learning disability
- the use of antipsychotics for the management of behaviour that challenges in children and young people with a learning disability

In addition, literature on the health-related quality of life of people with a learning disability and behaviour that challenges was systematically searched to identify studies reporting appropriate utility scores that could be utilised in a cost-utility analysis.

The rest of this section describes the methods adopted in the systematic literature review of economic studies. Methods employed in economic modelling are described in the relevant economic sections of the evidence chapters.

3.6.1 Search strategy for economic evidence

3.6.1.1 Scoping searches

A broad preliminary search of the literature was undertaken in April 2013 to obtain an overview of the issues likely to be covered by the scope, and help define key areas. Searches were restricted to economic studies and HTA reports, and conducted in the following databases:

- Embase
- MEDLINE/MEDLINE In-Process
- HTA database (technology assessments)
- NHS Economic Evaluation Database (NHS EED).

Any relevant economic evidence arising from the clinical scoping searches was also made available to the health economist during the same period.

3.6.1.2 Systematic literature searches

After the scope was finalised, a systematic search strategy was developed to locate all the relevant evidence. The balance between sensitivity (the power to identify all studies on a particular topic) and specificity (the ability to exclude irrelevant studies from the results) was

carefully considered, and a decision made to utilise a broad approach to searching to maximise retrieval of evidence to all parts of the guideline. Searches were restricted to economic studies and health technology assessment reports, and conducted in the following databases:

- Embase
- HTA database (technology assessments)
- MEDLINE/MEDLINE In-Process
- NHS EED
- PsycINFO.

Any relevant economic evidence arising from the clinical searches was also made available to the health economist during the same period.

The search strategies were initially developed for MEDLINE before being translated for use in other databases/interfaces. Strategies were built up through a number of trial searches, and discussions of the results of the searches with the review team and GDG to ensure that all possible relevant search terms were covered. In order to assure comprehensive coverage, search terms for the guideline topic were kept purposely broad to help counter dissimilarities in database indexing practices and thesaurus terms, and imprecise reporting of study interventions by authors in the titles and abstracts of records.

For standard mainstream bibliographic databases (Embase, MEDLINE and PsycINFO) search terms for the guideline topic combined with a search filter for health economic studies. For searches generated in topic-specific databases (HTA, NHS EED) search terms for the guideline topic were used without a filter. The sensitivity of this approach was aimed at minimising the risk of overlooking relevant publications, due to potential weaknesses resulting from more focused search strategies. The search terms are set out in full in Appendix H.

3.6.1.3 Reference Management

Citations from each search were downloaded into reference management software and duplicates removed. Records were then screened against the inclusion criteria of the reviews before being quality appraised. The unfiltered search results were saved and retained for future potential re-analysis to help keep the process both replicable and transparent.

3.6.1.4 Search filters

The search filter for health economics is an adaptation of a pre-tested strategy designed by CRD (2007). The search filter is designed to retrieve records of economic evidence (including full and partial economic evaluations) from the vast amount of literature indexed to major medical databases such as MEDLINE. The filter, which comprises a combination of controlled vocabulary and free-text retrieval methods, maximises sensitivity (or recall) to ensure that as many potentially relevant records as possible are retrieved from a search. A full description of the filter is provided in Appendix H.

3.6.1.5 Date and language restrictions

Systematic database searches were initially conducted in August 2013 up to the most recent searchable date. Search updates were generated on a 6-monthly basis, with the final re-runs carried out in October 2014. After this point, studies were included only if they were judged by the GDG to be exceptional (for example, the evidence was likely to change a recommendation).

Although no language restrictions were applied at the searching stage, foreign language papers were not requested or reviewed, unless they were of particular importance to an area

under review. All the searches were restricted to research published from 1998 onwards in order to obtain data relevant to current healthcare settings and costs.

3.6.1.6 Other search methods

Other search methods involved scanning the reference lists of all eligible publications (systematic reviews, stakeholder evidence and included studies from the economic and clinical reviews) to identify further studies for consideration.

Full details of the search strategies and filter used for the systematic review of health economic evidence are provided in Appendix I.

3.6.2 Inclusion criteria for economic studies

The following inclusion criteria were applied to select studies identified by the economic searches for further consideration:

1. Only studies from Organisation for Economic Co-operation and Development countries were included, as the aim of the review was to identify economic information transferable to the UK context.
2. Selection criteria based on types of clinical conditions and service users as well as interventions assessed were identical to the clinical literature review.
3. Studies were included provided that sufficient details regarding methods and results were available to enable the methodological quality of the study to be assessed, and provided that the study's data and results were extractable. Poster presentations of abstracts were excluded.
4. Full economic evaluations that compared 2 or more relevant options and considered both costs and consequences as well as costing analyses that compared only costs between 2 or more interventions were included in the review.
5. Studies that adopted a very narrow perspective, ignoring major categories of costs to the NHS, were excluded; for example studies that estimated exclusively drug acquisition costs were considered non-informative to the guideline development process.

3.6.3 Applicability and quality criteria for economic studies

All economic papers eligible for inclusion were appraised for their applicability and quality using the methodology checklist for economic evaluations recommended by NICE (NICE, 2012). The methodology checklist for economic evaluations was also applied to the economic models developed specifically for this guideline. All studies that fully or partially met the applicability and quality criteria described in the methodology checklist were considered during the guideline development process, along with the results of the economic modelling conducted specifically for this guideline. The completed methodology checklists for all economic evaluations considered in the guideline are provided in Appendix R.

3.6.4 Presentation of economic evidence

The economic evidence considered in the guideline is provided in the respective evidence chapters, following presentation of the relevant clinical evidence. The references to included studies and the respective evidence tables with the study characteristics and results are provided in Appendix S. Methods and results of economic modelling undertaken alongside the guideline development process are presented in the relevant evidence chapters. Characteristics and results of all economic studies considered during the guideline development process (including modelling studies conducted for this guideline) are summarised in economic evidence profiles accompanying respective GRADE clinical evidence profiles in Appendix T.

3.6.5 Results of the systematic search of economic literature

The titles of all studies identified by the systematic search of the literature were screened for their relevance to the topic (that is, economic issues and information on health-related quality of life). References that were clearly not relevant were excluded first. The abstracts of all potentially relevant studies (60 references) were then assessed against the inclusion criteria for economic evaluations by the health economist. Full texts of the studies potentially meeting the inclusion criteria (including those for which eligibility was not clear from the abstract) were obtained. Studies that did not meet the inclusion criteria, were duplicates, were secondary publications of 1 study, or had been updated in more recent publications were subsequently excluded. Economic evaluations eligible for inclusion (8 studies) were then appraised for their applicability and quality using the methodology checklist for economic evaluations. Finally, those studies that fully or partially met the applicability and quality criteria set by NICE were considered at formulation of the guideline recommendations.

3.7 Using NICE evidence reviews and recommendations from existing NICE clinical guidelines

When review questions overlap and evidence from another guideline applies to a question in the current guideline, it might be desirable and practical to incorporate or adapt recommendations published in NICE guidelines. Adaptation refers to the process by which an existing recommendation is modified in order to facilitate its placement in a new guideline. Incorporation refers to the placement of a recommendation that was developed for another guideline into a new guideline, with no material changes to wording or structure. Incorporation would be used in relatively rare circumstances, as cross-referring to the other guideline will often be all that is necessary.

Incorporation or adaptation is likely to be substantially more complex where health economics were a major part of the decision making. In these circumstances, these methods are only used rarely after full and detailed consideration.

3.7.1 Incorporation

In the current guideline, the following criteria were used to determine when a recommendation could be incorporated:

- a review question in the current guideline was addressed in another NICE guideline
- evidence for the review question and related recommendation(s) has not changed in important ways
- evidence for the previous question is judged by the GDG to support the existing recommendation(s), and be relevant to the current question
- the relevant recommendation can ‘stand alone’ and does not need other recommendations from the original guideline to be relevant or understood within the current guideline.

3.7.2 Adaptation

The following criteria were used to determine when a recommendation could be adapted:

- a review question in the current guideline is similar to a question addressed in another NICE guideline
- evidence for the review question and related recommendations has not changed in important ways
- evidence for the previous question is judged by the GDG to support the existing recommendation(s), and be relevant to the current question

- the relevant recommendation can 'stand alone' and does not need other recommendations from the original guideline to be relevant
- contextual evidence, such as background information about how an intervention is provided in the healthcare settings that are the focus of the guideline, informs the re-drafting or re-structuring of the recommendation but does not alter its meaning or intent (if meaning or intent were altered, a new recommendation should be developed).

In deciding whether to choose between incorporation or adaption of existing guideline recommendations, the GDG considered whether the direct evidence obtained from the current guideline dataset was of sufficient quality to allow development of recommendations. It was only where (a) such evidence was not available or insufficient to draw robust conclusions and (b) where methods used in other NICE guidelines were sufficiently robust that the 'incorporate and adapt' method could be used. Recommendations were only incorporated or adapted after the GDG had reviewed evidence supporting previous recommendations and confirmed that they agreed with the original recommendations.

When adaptation is used, the meaning and intent of the original recommendation is preserved but the wording and structure of the recommendation may change. Preservation of the original meaning (that is, that the recommendation faithfully represents the assessment and interpretation of the evidence contained in the original guideline evidence reviews) and intent (that is, the intended action[s] specified in the original recommendation will be achieved) is an essential element of the process of adaptation.

3.7.3 Roles and responsibilities

The guideline review team, in consultation with the guideline Facilitator and Chair, were responsible for identifying overlapping questions and deciding if it would be appropriate to incorporate or to adapt following the principles above. For adapted recommendations, at least 2 members of the GDG for the original guideline were consulted to ensure the meaning and intent of the original recommendation was preserved. The GDG confirmed the process had been followed, that there was insufficient evidence to make new recommendations, and agreed all adaptations to existing recommendations.

In evidence chapters where incorporation and adaptation have been used, the original review questions are listed with the rationale for the judgement on the similarity of questions. Tables are then provided that set out the original recommendation, a brief summary of the original evidence, the new recommendation, and the reasons for adaptation. For an adapted recommendation, details of any contextual information are provided, along with information about how the GDG ensured that the meaning and intent of the adapted recommendation was preserved.

3.7.4 Drafting of adapted recommendations

The drafting of adapted recommendations conformed to standard NICE procedures for the drafting of guideline recommendations, preserved the original meaning and intent, and aimed to minimise the degree or re-writing and re-structuring.

3.8 From evidence to recommendations

Once the clinical and health economic evidence was summarised, the GDG drafted the recommendations. In making recommendations, the GDG took into account the trade-off between the benefits and harms of the intervention/instrument, as well as other important factors, such as the trade-off between net health benefits and resource use, values of the

GDG and society, the requirements to prevent discrimination and to promote equality[e], and the GDG's awareness of practical issues (Eccles et al., 1998; NICE, 2012).

Finally, to show clearly how the GDG moved from the evidence to the recommendations, each chapter (or sub-section) has a section called 'recommendations and link to evidence'. Underpinning this section is the concept of the 'strength' of a recommendation (Schünemann et al., 2003). This takes into account the quality of the evidence but is conceptually different. Some recommendations are 'strong' in that the GDG believes that the vast majority of healthcare professionals and service users would choose a particular intervention if they considered the evidence in the same way that the GDG has. This is generally the case if the benefits clearly outweigh the harms for most people and the intervention is likely to be cost effective. However, there is often a closer balance between benefits and harms, and some service users would not choose an intervention whereas others would. This may happen, for example, if some service users are particularly averse to some side effect and others are not. In these circumstances the recommendation is generally weaker, although it may be possible to make stronger recommendations about specific groups of service users. The strength of each recommendation is reflected in the wording of the recommendation, rather than by using ratings, labels or symbols.

Where the GDG identified areas in which there are uncertainties or where robust evidence was lacking, they developed research recommendations. Those that were identified as 'high priority' were developed further in the NICE version of the guideline, and presented in Appendix G.

3.9 Stakeholder contributions

Professionals, service users, and companies have contributed to and commented on the guideline at key stages in its development. Stakeholders for this guideline include:

- service user and carer stakeholders: national service user and carer organisations that represent the interests of people whose care will be covered by the guideline
- local service user and carer organisations: but only if there is no relevant national organisation
- professional stakeholders' national organisations: that represent the healthcare professionals who provide the services described in the guideline
- commercial stakeholders: companies that manufacture drugs or devices used in treatment of the condition covered by the guideline and whose interests may be significantly affected by the guideline
- providers and commissioners of health services in England and Wales
- statutory organisations: including the Department of Health, the Welsh Assembly
- Government, NHS Quality Improvement Scotland, the Care Quality Commission and the National Patient Safety Agency
- research organisations: that have carried out nationally recognised research in the area.

NICE clinical guidelines are produced for the NHS in England and Wales, so a 'national' organisation is defined as 1 that represents England and/or Wales, or has a commercial interest in England and/or Wales.

Stakeholders have been involved in the guideline's development at the following points:

- commenting on the initial scope of the guideline and attending a scoping workshop held by NICE
- commenting on the draft of the guideline.

[e]See NICE's equality scheme: www.nice.org.uk/aboutnice/howwework/NICEEqualityScheme.jsp

3.10 Validation of the guideline

Registered stakeholders had an opportunity to comment on the draft guideline, which was posted on the NICE website during the consultation period. Following the consultation, all comments from stakeholders and experts (see Appendix D) were responded to, and the guideline updated as appropriate. NICE also reviewed the guideline and checked that stakeholders' comments had been addressed.

Following the consultation period, the GDG finalised the recommendations and the NCCMH produced the final documents. These were then submitted to NICE for a quality assurance check. Any errors were corrected by the NCCMH, then the guideline was formally approved by NICE and issued as guidance to the NHS in England and Wales.

4 Experience of care for service users, families and carers

4.1 Introduction

Most, if not all learning disabilities are identified very early in life and many families will have a central caring role. For many people this care will be lifelong. Similarly, most behaviour that challenges is also first identified in the home and the burden of care that stems from this usually falls on the family; 20% or more of people who live at home(Joyce et al., 2001) may have behaviour that challenges and the numbers are similar for those attending day schools (Kiernan & Kiernan, 1994). Even when behaviour that challenges emerges in another setting, families are almost always involved in the care of a person.

Families, therefore, are key providers of support, and it is important that they are acknowledged as valued partners in the care of people with a learning disability and behaviour that challenges and are provided with information and support that is practical, tailored to their needs and evidence based, as set out in the charter of The Challenging Behaviour Foundation (http://www.challengingbehaviour.org.uk/strategy-group/charter.html). However, the experience of families is commonly that information is sparse, support inadequate and collaboration often also very limited. Families describe a lack of practical information, and struggling to access any training in understanding behaviour that challenges and supporting behaviour change. Family members may be excluded from support and services for learning disabilities because of the behaviour that challenges, which means that those families who are most in need of short breaks, for example, are not able to access them. Despite being well placed to spot the early warning signs of support breaking down, or additional support needs developing, these are often ignored or not recognised until a crisis develops. Families also regularly describe navigating and engaging with the systems and processes to access support services as confusing and difficult.

Families also report a lack of training in understanding and responding to their child's behaviour that challenges. While most families will describe the many positive characteristics of their relative, the day to day challenges are wide-ranging, and have a cumulative effect on the whole family, having an impact on relationships, the home environment, social, leisure and employment opportunities and finances, as well as taking a toll on emotional and physical health and wellbeing, including sleep. All of this can lead some families to feel isolated and excluded, and as a result of their experiences, they can develop low expectations of services.

While for some people with a learning disability, the opportunities of personalisation, and the associated financial support, have enabled them to have a good quality of life in their local community, and successive government and other documents have aimed to place people who use services at the heart of policy (Hatton & Taylor, 2008; Moss et al., 1993; Moss et al., 1998; Sturmey et al., 2005), many people with a learning disability and behaviour that challenges continue to be marginalised. They are at risk of living in segregated settings far from their families and local communities and of being subjected to a range of restrictive practices and abuse.

Investigations into the abuse at Winterbourne View Hospital *(Aman et al., 1986)* have highlighted the ease with which inappropriate and excessive use of restrictive and abusive practices can be utilised and can inflict pain and cause distress. Unfortunately Winterbourne is just the most recent in a long list of scandals going back many years. Martin and Evans (Kazdin et al., 1983) reviewed the findings of 16 inquiries between 1969 and 1981, identifying many of the now familiar lessons about the abuses inflicted upon the most vulnerable members of our society. Since then, there has continued to be a steady stream of examples

of abuse in which the needs of the person with a learning disability have been overlooked by both individual members of staff and services as a whole.

The Learning Disabilities Census across England (Linaker, 1991) provides an audit of current service provision, numbers of out of area placements and lengths of stay. The data for the census were collected on the 30 September 2013, providing a snapshot of the treatment and care people with a learning disability, autism and/or behaviour that challenges received from the NHS and independent learning disability service providers on that day. The subsequent report contains information relating to the experience of care including drug administration, incidents, ward accommodation, uses of the Mental Health Act 1983, and information on the commissioning and provision of learning disability services including costs and care planning. The report found that:

- Over half of the service users (56.6 per cent or 1,841) had been the subject of at least 1 incident involving self-harm, an accident, physical assault on the service user, hands-on restraint or seclusion during the 3 months preceding the census. Proportionally, more females experienced every type of incident than males. There appears to be an association between hands-on restraint and the administration of drugs; 40.4% (889) of the 2,220 given these drugs had experienced at least 1 instance of hands-on restraint compared with 21.4% (221) of the 1,030 who were not given any medication.
- Almost half of service users (46.4% or 1,508 people) were in receipt of an active care plan without a discharge plan in place. Around 1 in 20 service users (4% or 152 people) were experiencing a delayed transfer of care.
- Almost four fifths of service users (78.0% or 2,536) were subject to the Mental Health Act 1983 on census day, compared with 22% (714 people) who were classed as 'informal patients'. Of those subject to the Mental Health Act, the majority (99.5% or 2,524) were subject to 'longer term hospital orders' (of a duration of greater than 72 hours).

The need to gain the perspective of people with a learning disability whose behaviour is challenging is self-evident if services are to provide support that is based upon an understanding of the function of their behaviour. Understanding this perspective and that of their families and carers is the primary focus of this chapter.

4.2 Review question: In people with a learning disability and behaviour that challenges, what are their experiences of having a learning disability and behaviour that challenges, of access to services, and of treatment?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 9. A systematic search for published reviews of relevant qualitative studies of people with a learning disability and behaviour that challenges was undertaken using standard NCCMH procedures as described in Chapter 3. Reviews were sought of qualitative studies that used relevant first-hand experiences of adults witha learning disability and behaviour that challenges and their families, partners and carers. The GDG did not specify a particular outcome. Instead the review was concerned with any narrative data that highlighted the experience of care.

A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 9: Clinical review protocol summary for the review of service user experience of care

Component	Description

Component	Description
Review question	In people with a learning disability and behaviour that challenges, what are their experiences of having a learning disability and behaviour that challenges, of access to services, and of treatment? (RQ8.1)
Perspective	People with a learning disability and behaviour that challenges
Phenomenon of interest	The individuals experiences of: • having a learning disability and behaviour that challenges • access to services • treatment.
Primary outcome/ Evaluation	Experience of care
Study design	Systematic reviews and qualitative research

4.2.1 Evidence

One systematic review providing relevant qualitative evidence met the eligibility criteria and was selected as the basis for this section of the guideline: Griffith 2013a (Griffith et al., 2013). The systematic review carried out a narrative thematic synthesis of qualitative studies using the methods described by (Thomas & Harden, 2008). A quality evaluation was completed for all included studies based on guidelines developed by (Cesario et al., 2002). A summary of the included review can be found in Table 10.

Table 10: Study information table for the systematic review included in the review of service user experience of care

	Griffith 2013a
Review question/ Aim	Examine qualitative research on the experiences of people with a learning disability and behaviour that challenges in relation to received service supports and interventions.
Method used to synthesise evidence	Thematic synthesis
Design of included studies	Qualitative studies
Dates searched	No restriction to January 2013
Electronic databases	PsycINFO, Web of Science, PUBMED, and the Cochrane Library.
No. of included studies (N[1])	17 (163)
Participant characteristics	People with a learning disability, or a learning disability and a co-diagnosis of ASD, who were reported to engage in behaviour that challenges.
Intervention	N/A
Comparison	N/A
Outcome	Service user experience of care.
Review Quality	High
Notes. ASD = autism spectrum disorder. [1]Number of participants.	

The systematic review included 17 studies (N = 163) evaluating service users' experience, or a researcher observation, of care: Brown 2009 (Brown & Beail, 2009), Clare 1993 (Clare & Murphy, 1993), Clarkson 2009 (Clarkson et al., 2009), Duperouzel 2010 (Duperouzel & Fish, 2010), Fish 2005 (Fish & Culshaw, 2005), Hall 2008 (Hall & Deb, 2008), Harker-longton 2002 (Harker-Longton & Fish, 2002), Hawkins 2005 (Hawkins et al., 2005), Hubert 2006 (Hubert & Hollins, 2006), Hubert 2010 (Hubert & Hollins, 2010), Jones 2006 (Jones & Kroese, 2006), Lunsky 2009 (Lunsky & Gracey, 2009), MacDonald 2011 (MacDonald et al., 2011), Murphy

1996 (Murphy et al., 1996), Ruef 1999 (Ruef et al., 1999), Ruef 2002 (Ruef & Turnbull, 2002), Sequeira 2001 (Sequeira & Halstead, 2001).

Of the included studies, 14 were conducted in the UK, 2 in the USA and 1 in Canada. Of the included participants, 30% were female and the age ranged from 18 to 76 years. The vast majority (97%) were currently residing in a residential placement, with 33% in secure or forensic placements. Of those studies that provided information on the severity of participants' learning disability (k = 8; N = 94), 48% had a mild learning disability, 15% had a mild-to-moderate learning disability, 12% had a moderate learning disability, 21% had a severe learning disability, and 4% had a diagnosis of autism with no clear information about learning disabilities, although they had reported difficulties with verbal expressive communication and received state services for people with developmental disabilities. The type of behaviour that challenges, when specified, included aggressive behaviour, criminal behaviour and self-injurious behaviour.

The quality of the included studies as a whole was rated good. Of the 17 included studies, 12 were rated as high quality (75% to 100% of the total quality criteria being met), and 3 were rated as medium quality (50% to 74% of the total quality criteria being met). The quality of the remaining 2 studies could not be evaluated because they did not present data in a format suitable for quality rating.

Although the original focus of the systematic review was on service users' experience of all support services for behaviour that challenges, the majority of the included studies concern the experience of residential settings.

Further information about included and excluded studies can be found in Griffith 2013a.

A summary of the findings from Griffith 2013a is presented below for each theme.

4.2.1.1 Theme 1: Imbalance of power

Service users reported not feeling in control of their immediate living environment, nor of the direction of their own lives. Apparent throughout all studies was the imbalance of power between staff and service users. Service users in residential care were dependant on staff for most of their daily needs. However, some service users felt that the quality and consistency of the care they received was dependent on staff moods, behaviour, and attitudes:

> *I was really annoyed 'cos they said I can go home and then they changed their mind.* (Brown & Beail, 2009, p. 507)

The casual denial of service users' requests by support staff highlights how little power and control service users sometimes had:

> *[During a meal the service user] said 'drink' and was told he could have some when he was finished.* (Hubert and Hollins, 2010, p. 193)

Many service users spoke of their frustration at the authoritarian attitude of staff and of the limited influence they had over the decisions about their own lives:

> *I don't like people comin' into my room and tellin' me what to do, saying 'Well, you should do this, and you should do that' [mimics authoritarian voice].* (Ruef et al., 1999, p. 49)

> *They are drawing up my guidelines, they'll tell me though, not ask me.* (Harker-Longton & Fish, 2010, p. 147)

The 'imbalance of power' was apparent across all aspects of service users' experience of care, but most explicitly in direct relation to support staff. Service users regarded some support staff as indifferent to their individual attributes and 1 researcher noted:

> *All of the men, even those without any speech, spent a considerable amount of time trying to communicate their feelings and needs […] There was often little recognition of or response to these attempts to communicate [by staff], and thus there was a rejection of these men as interactive, social beings.* (Hubert & Hollins, 2006, p. 71)

It was clear that some service users felt the need to emphasise their individuality and personhood as a means of overcoming the indifference and highlighting the imbalance of power that endured:

> *I'm not a patient, I'm a person.* (Brown & Beail, 2009, p. 507)

4.2.1.2 Theme 2: Participants' causal attributions about behaviour that challenges

There were numerous reports of participants having to endure institutional residential placements that were experienced as depersonalised and constraining. In the case of forensic placements, many also reported living with violent and unpredictable peers. Many spoke of their feelings of frustration, injustice, helplessness, and anger, provoked by living in an environment in which they had little control. The very residential placements that were supposed to support people in improving their behaviour that challenges were perceived by many participants as *causes* of their behaviour that challenges.

4.2.1.2.1 Atmosphere in residential placement.

The majority of service users described the atmosphere in their residential placements extremely negatively, and this was also the case in researchers' observations:

> *We observed again a generally rather cold atmosphere, under another of a series of managers, where staff seemed to have lost control of one resident, whose behaviour caused others to become nervous and demanding, giving the house a palatable sense of instability and unease.* (Hubert and Hollins, 2010, p. 193)

The auditory stimulation in residential placements was found to be particularly annoying and stressful. Examples included the radio being on loudly; the constant ringing of telephones, and the other service users making noise (Brown & Beail, 2009; Ruef & Turnbull, 2002; Ruef et al., 1999).

Some service users reported sometimes violent living environments. Clare and Murphy (1993) found that 4 of 6 service users described times when they were frightened by the violence of other service users, and MacDonald et al. (2011) reported that 3 of 8 participants spoke of being punched, being hit, or having items thrown at them by other service users:

> *Violence was a part of everyday life.* (MacDonald, 2011, p. 49)

Service users felt as though they had limited autonomy, lacking control over both their environment and their choice of activities:

> *They wouldn't even leave me alone. They wouldn't let me read, they wouldn't let me do anything. And that kind of made me mad…I don't like it when people like say that I can't do what I want to do. You ain't my mother, I'm a grown man.* (Ruef & Turnbull, 2002, p. 132)

They also reported felt infringements of their liberty (Ruef & Turnbull, 2002; Ruef et al., 1999) and recounted instances such as the front door being kept locked (Clare & Murphy, 1993; Ruef & Turnbull, 2002) and personal belongings being removed from their bedroom (Brown & Beail, 2009; Harker-Longton & Fish, 2002).

> *I can't go out of the apartment, we get in trouble*. (Ruef & Turnbull, 2002, p. 131)

Conversely, participants valued being in charge of their day-to-day routines and recreational activities (Murphy, Estien, & Clare, 1996; Ruef & Turnbull, 2002). Common responses for

preferring some residential placements over others included being '*more independent*' and having '*more freedom*' (Murphy et al., 1996, pp. 273–274).

Despite the consistently negative descriptions of their living environments, few service users with aggressive behaviour identified this as a causal factor for their behaviour that challenges; they would largely talk about specific situational factors as triggering a particular episode. Only a minority made the link between the negative environment and their aggressive behaviour:

> *But people get pissed off living here. That's why a lot of people kick off.* (Fish & Culshaw, 2005, p. 99)

However, in the case of service users who self-harmed, the majority recognised their residential placement as a causal factor in their self-injurious behaviour:

> *I'm not a kid or a baby, I'm not an animal either but I'm in this cage.* (Harker-Longton & Fish, 2002, p. 146)

4.2.1.2.2 Staff Attitudes: A Trigger

The poor attitude of support staff was highlighted by service users as a primary 'trigger' to their aggressive behaviour:

> *If we want a drink and they tell us 'no' then we kick off. Staff wind people up.* (Jones & Kroese, 2006, p. 52)

Service users felt that support staff made little effort to hide negative feelings toward them and found staff to be rude, authoritarian, and '*not bothered*' (Clarkson et al., 2009, p. 286):

> *They should be more honest shouldn't they? They should get it right. There wouldn't be half the aggro on the ward would it?* (Clarkson et al., 2009, p. 287)

The most common reported reason for engaging in behaviour that challenges was frustration as a result of not being listened to, or feeling misunderstood by staff (Brown & Beail, 2009; Fish & Culshaw, 2005; Jones & Kroese, 2006):

> *You've got something on your mind and staff's like not listening, you like play up and they don't listen.* (Fish & Culshaw, 2005, p. 99)

4.2.1.2.3 Self-injurious behaviour as a form of coping

Self-harm was consistently reported as an intensely emotional experience. Service users spoke of short and long-term, environmental and internal factors that they felt contributed to their behaviour. The most common reason given for engaging in self-injurious behaviour was as a means of relief from overwhelming mental distress relating to feelings of sadness, hopelessness and shame, or anger and frustration:

> *Whatever I'm sad about its steam coming out.* (Harker-Longton & Fish, 2002, p. 143)

> *It were 'cos of anger, 'cos I felt angry, and I used to cut.* (Brown & Beail, 2009, p. 508)

Other reasons given for engaging in self-injurious behaviour included past events such as abuse or a close bereavement (Brown & Beail, 2009), as a means of self-punishment (Duperouzel & Fish, 2010; Harker-Longton & Fish, 2002), or as an alternative to hurting others:

> *I just lose my temper so much and I don't want to hurt the staff, so I take it out on myself.* (Brown & Beail, 2009, p. 507)

All these reasons suggest that self-injurious behaviour was regarded by service users as a coping mechanism and 1 that was beyond their control:

Your body gets addicted [...] when you get angry, your body expects to be cut. (Brown & Beail, 2009, p. 508)

4.2.1.3 Theme 3: Experiences of restrictive interventions

Of the included studies, 6 focused explicitly on how service users perceived restrictive practices. Throughout these studies, all physical interventions were reported to be stressful and painful, and some service users demonstrated a limited understanding about why or when physical restraint procedures would be used. It was therefore difficult from the reports to ascertain if they were reporting properly conducted restrictive practices, or unethical practice, although some situations that some participants recalled were clearly unethical. In a similar vein, 1 study examined participants' understanding of chemical restraint (Hall & Deb, 2008) and found a lack of knowledge of the drugs taken for their behaviour that challenges.

Standard restrictive interventions after an episode of self-harm were found to be hugely disliked by service users, who reported that they were not just ineffective but also stressful.

4.2.1.3.1 Understanding of restrictive interventions

Service users' understanding about why restrictive interventions are used varied widely across studies.

The majority felt that restrictive interventions served a purpose:

Stop me from getting hurt. (Jones & Kroese, 2006, p. 52)

To make sure I didn't hit or kick. (MacDonald et al., 2011, p. 50)

However, some service users felt that interventions were used for purposes of punishment and as a means of gaining control by staff:

I reckon some of the staff here might seclude people just to prove they are in charge. (Sequeira & Halstead, 2001, p. 468)

Some service users differentiated between restrictive procedures that seemed justifiable and those that were not:

Sometimes it's necessary and sometimes it isn't, it's stupid things for someone to be restrained about, I mean if you were going to attack someone well that's alright, but restraining you just for the hell of it. (Fish & Culshaw, 2005, p. 104)

Service users generally perceived staff to be reluctant to physically intervene:

They probably feel upset because they don't like doing it. (Jones & Kroese, 2006, p. 52)

However, some service users thought staff were angry when delivering physical interventions (MacDonald et al., 2011; Sequeira & Halstead, 2001).

4.2.1.3.2 Unethical practice

Some of the reports by service users were indicative of unethical and abusive practice:

I've seen staff hitting clients, after clients have hit them. A bit frightening, lot of staff on top of him. (Jones & Kroese, 2006, p. 52)

They just hold you down and hit you. Sometimes they put you in a dirty bath. (MacDonald et al., 2011, p. 48)

'We're going to the pub' they tell you when you're in seclusion. (Jones & Kroese, 2006, p. 52)

> *Laughing and joking and punching me at the same time.* (MacDonald et al., 2011, p. 50)

However, because of the service group, it can be difficult to ascertain whether service users are describing instances of abuse by staff or whether there is a lack of understanding of sanctioned restrictive procedures. For example, Hawkins, Allen, and Jenkins (2005) noted that very few service users understood that physical restraint would stop if their behaviour that challenges stopped. Nonetheless, due to reports of abusive practices appearing across multiple research studies, and the specific details in each report, dismissing them as simply lack of understanding becomes very difficult.

4.2.1.3.3 Physical and emotional discomfort

Of the 5 studies that examined services users' experience of physical interventions, all consistently reported physical pain as a consequence:

> *People sitting on my legs and it hurts my legs.* (Hawkins et al., 2005, p. 26)

> *Oh aye, it's painful. You squeal and squeal but they just hold you down.* (MacDonald et al., 2011, p. 48)

Numerous accounts of emotional discomfort caused by restraining practices were also reported, including fear, anger, desperation, anxiety, and sadness:

> *It's awful, when they restraint you it's awful. Nurses and doctors say you're awful and they give you one of these (mimics giving self an injection).* (Sequeira & Halstead, 2001, p. 467)

Several service users spoke of becoming angrier when restrained:

> *When you have got people holding you, you kick off more than you have done.* (Sequeira & Halstead, 2001, p. 468)

One service user found restraint and treatment at the service so distressing that they thought about suicide as a means of escape:

> *I wished I was dead, I tried anything to get out. I used to lie in bed at night and try and do that to myself (demonstrates strangling self). I was trying to kill myself…I wanted out of it. (MacDonald et al., 2011, p. 49)*

One service user said she had nightmares about restraint (Sequeira & Halstead, 2001); another reported physical restraint brought back memories of previous abuse, particularly if male staff were involved (Fish & Culshaw, 2005). Other service users were thought to be so traumatised by their experience of restraint that they avoided talking about it at all (MacDonald et al., 2011).

Not one service user reported a restrictive practice as anything other than physically or emotionally painful, and some felt the use of restrictive practices such as restraint was unfair to themselves and to other service users:

> *I thought they [staff] were terrible doing that to us. It was pretty bad*. (MacDonald et al., 2011, p. 50)

4.2.1.3.4 Self-injurious behaviour: Effects of special observation

A common procedure following a service user engaging in self-injurious behaviour is to place him or her under 24- hour observation. Service users reported a strong dislike for the procedure, finding them both degrading and invasive:

> *They check your pockets, check your socks, totally degrading, things like that, open your mouth.* (Duperouzel & Fish, 2010, p. 611)

The emotional distress caused by the procedure could in turn lead to repeated self-injurious behaviour; this process was described by 1 service user as a *'vicious circle'* (Duperouzel & Fish, 2010, p. 612).

Some service users talked about special observation being ineffective, as they could still find ways to self-injure:

> *Don't they know after all this time it's not who's with me, it's whether I want to or not.* (Harker-Longton & Fish, 2002, p. 145)

In addition, some staff members did not hide their annoyance or animosity toward service users when having to observe them after an episode of self-injurious behaviour:

> *They've said 'we want you off a level 3 [special observation] immediately because we're not happy following you round the flat' (Duperouzel & Fish, 2010, p. 612)*

This perceived animosity created a tense situation for service users during a time of immense vulnerability (Duperouzel & Fish, 2010).

4.2.1.3.5 Medication

Service users had large gaps in their knowledge about the medication taken for their behaviour that challenges (Hall and Deb, 2008). From 20 service users who were receiving prescribed medication for their behaviour that challenges, only 5 could recall the name of their medication and the majority (N = 13) were unable to accurately say why they took the medication. The responses of the 7 service users who did give an accurate reason as to why they were on prescribed medication included '*my temper*' and '*to help my nerves*' (Hall & Deb, 2008, p. 31).

Rather than being actively involved in decisions surrounding their medication, the majority of service users deferred to the doctors' advice:

> *You're my doctor, it's not up to me.* (Hall & Deb, 2008, p. 32)

In contrast, women who received emergency psychiatric services were steadfast in not wanting to be sedated and reported feeling disempowered when forced to do so:

> *I don't want it, they force me to take meds—strap me down.* (Lunsky & Gracey, 2009, p. 92)

4.2.1.4 Theme 4: Opportunities for improvement and proactive interventions

Across some studies, a positive view of practice within 'challenging behaviour' services was described.

Service users reported beneficial and helpful relationships with staff. '*Good*' staff members were those that showed good interpersonal skills with service users, that displayed a respectful attitude, and that treated service users as individuals.

Similarly, service users wanted fewer restrictive interventions and felt that these could be prevented if staff helped calm the situation by talking to them to.

Some service users spoke of finding their own behaviour that challenges aversive but still could not control it and wanted help to control their behaviour that challenges.

4.2.1.4.1 Beneficial relationship with staff members

Some service users talked about the positive impact that a good relationship with support staff had on their emotional wellbeing and behaviour that challenges. However, good relationships with staff members did not come easily for service users, and many said it took a long time to get to a stage where they trusted a staff member:

> *I have difficulty in trusting people […] so I have to build trust up with someone, build it up.* (Fish & Culshaw, 2005, p. 103)

Establishing a trusting relationship with a staff member was further compounded by high staff turnover:

> *It feels strange them leaving and then some other new staff come in and you have to get used to them.* (Clarkson et al., 2009, p. 286)

Service users provided various suggestions about how the staff of psychiatric hospitals could be improved:

> *Be more nicer to people and don't judge them for their issues—everyone has issues.* (Lunsky & Gracey, 2009, p. 93)

> *Treat us like we are people, not babies, don't tell us 'Sit and don't move.'* (Lunsky & Gracey, 2009, p. 93)

Service users spoke about the qualities possessed by '*good*' staff members which included: patience, helpfulness, being able to laugh together, mutual respect, having a calm and consistent approach, and explaining information clearly. A balance of power between service user and staff member was also highly valued:

> *He just like, asks me very politely…and me and him both work together*. (Ruef & Turnbull, 2002, p. 135)

Positive relationships gave service users the confidence to progress towards valued goals:

> *The people I work with now really believe in what I'm doing and believe in me. So I'm starting to believe in myself.* (Ruef & Turnbull, 2002, p. 134)

Service users reported responding best to staff members who were genuinely interested in their wellbeing and cared for them:

> *I can tell when they like me […] everyone wants to be liked don't they? Make it easier when they like you.* (Harker-Longton & Fish, 2005, p. 146)

4.2.1.4.26 Strategies for calming down

Many service users found their own behaviour that challenges aversive and described feeling guilty and regretful about their behaviour after the event (Brown & Beail, 2009; Duperouzel & Fish, 2010; Ruef et al., 1999).

Service users across studies wanted less restrictive staff responses when dealing with a situation that could escalate into an episode of behaviour that challenges (Duperouzel & Fish, 2010; Hall & Deb, 2008):

> *Talk to you, ask you why you are worked up, talk to you.* (Fish & Culshaw, 2005, p. 102)

When asked what could have been done to prevent his aggressive behaviour, 1 service user replied:

> *They could take me to my room and speak to me. That's what they could have done, it would have helped me and could have helped them as well.* (MacDonald et al., 2011, p. 50)

A history of a good relationship with a staff member could prevent or reduce behaviour that challenges for some service users:

> *It were Stella's shift, so when she came down I settled dead easy.* (Fish & Culshaw, 2005, p. 103)

Other strategies for calming down included deep breathing (Hawkins et al., 2005), spending time away from the setting, counting to 10 (Hall & Deb, 2008), or going to their bedroom to calm down (Fish & Culshaw, 2005; Hall & Deb, 2008).

4.2.1.4.3 A need for better strategies

Throughout the studies, service users reported being keen to learn strategies to better manage their behaviour that challenges:

> *I know I have a hard time being polite, but I'm tryin', tryin' my best to be polite to everybody.* (Ruef & Turnbull, 2002, p. 135)

Few service users were reported as receiving proactive interventions for their behaviour. No studies focused on the effects of any psychological interventions for behaviour that challenges in any detail, although there were a few broad comments by some service users (Ruef et al.1999).

Three service users from a study by Clare and Murphy (1993) continued to practice self-help strategies learned from a psychological program and were successful in reducing their behaviour that challenges. However, in another study, anger management was not regarded as useful for a service user with self-harm:

> *I thought that [anger management] would work but it never…I don't know who to go to, I do want to get out of it.* (Duperouzel & Fish, 2010, p. 610)

Some service users felt that support services would be more helpful if they offered structured and regular support, such as better outpatient facilities and regular group therapy. Such support was considered by service users to prevent behaviour that challenges and the subsequent restrictive interventions or admission (Hall & Deb, 2008; Lunsky & Gracey, 2009):

> *Seeing a doctor once a week works fine.* (Lunsky & Gracey, 2009, p. 94)

4.2.2 Evidence statements concerning service user experience

Evidence from 17 (163 participants) qualitative studies was synthesised by 1 systematic review using thematic analysis. The review was judged to be of high quality and the authors assessed the quality of the included studies as primarily high.

Four main themes were identified:

(1) Imbalance of power,

(2) Participants' causal attributions about behaviour that challenges,

(3) Experiences of restrictive interventions,

(4) Opportunities for improvement: proactive interventions. The recommendations which were developed from this section and the link to the evidence are at the end of the chapter where they are brought together with the reviews of the carer's experience and the validation exercise with service users and carers undertaken for this guideline.

4.3 Review question: For families and carers of people with a learning disability and behaviour that challenges, what are their experiences of caring for people with a learning

disability and behaviour that challenges, and what support is available for families, partners and carers?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 11. A systematic search for published reviews of relevant qualitative studies of people with a learning disability and behaviour that challenges was undertaken using standard NCCMH procedures as described in Chapter 3. Reviews were sought of qualitative studies that used relevant first-hand experiences of adults with autism and their families, partners and carers. The GDG did not specify a particular outcome. Instead the review was concerned with any narrative data that highlighted the experience of care.

A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 11: Clinical review protocol summary for the review of service user experience of care

Component	Description
Review question	For the families and carers of people with a learning disability and behaviour that challenges, what are their experiences of caring for people with a learning disability and behaviour that challenges, and what support is available for families, partners and carers? (RQ8.2)
Perspective	Families and carers of people with a learning disability and behaviour that challenges.
Phenomenon of interest	Families' and carers' experiences of: • caring for people with a learning disability and behaviour that challenges • the support available.
Primary outcome/ Evaluation	Experience of the family/carer
Study design	Systematic reviews and qualitative research

4.3.1 Evidence

One systematic review providing relevant qualitative evidence met the eligibility criteria and was selected as the basis for this section of the guideline: Griffith 2013b (Griffith & Hastings, 2013). The systematic review carried out a meta-synthesis of qualitative studies using Noblit and Hare's (1988) meta-ethnography. A summary of the included review can be found in Table 12.

Table 12: Study information table for the systematic review included in the review of carers' experience of care

	Griffith 2013b
Review question/ Aim	Synthesise the qualitative literature on the perspectives of those caring for a family member with a learning disability and behaviour that challenges, with a focus on their experiences of support services
Method used to synthesise evidence	Meta-ethnography
Design of included studies	Qualitative studies
Dates searched	No restriction to December 2012
Electronic databases	PsycINFO, Web of Science, PUBMED, and the Cochrane Library.

	Griffith 2013b
No. of included studies (N[1])	17 (391)
Participant characteristics	Carers of people with a learning disability and behaviour that challenges who have received support services or interventions.
Intervention	N/A
Comparison	N/A
Outcome	Carers' experience of care
Review Quality	Adequate[2]
[1]Number of participants. [2]No quality assessment of included studies was carried out.	

The systematic review included 17 studies (N = 391) evaluating perspectives of those caring for a family member with a learning disability and behaviour that challenges: Allen 2006 (Allen et al., 2006), Brown 2011 (Brown et al., 2011), Elford 2010 (Elford et al., 2010), Fox 1997 (Fox et al., 1997), Fox 2002 (Fox et al., 2002), Fredheim 2011 (Fredheim et al., 2011), Hubert 2010 (Hubert, 2010), McConkey 2011 (McConkey et al., 2011), McGill 2006a (McGill et al., 2006a), McGill 2006b (McGill et al., 2006b), Qureshi 1992 (Qureshi, 1992), Robertson 1996 (Robertson et al., 1996), Ruef 1999 (Ruef et al., 1999), Turnbull & Reuf 1996 (Turnbull & Reuf, 1996), Turnbull & Reuf 1997 (Turnbull & Reuf, 1997), Weiss 2009 (Weiss et al., 2009), Wodehouse & McGill 2009 (Wodehouse & McGill, 2009).

Of the included studies, 11 were conducted in the UK, 4 in the USA, 1 in Canada and 1 in Norway. Participant characteristics were poorly reported by the included studies. The relationships between the carer and family member with a learning disability were not specified for 55% of carers (N = 217). Of the remaining participants, 36% were mothers, 7% fathers and 2% 'others' (siblings, grandparents, and so on). Only 6 studies gave information about the carer's age, which ranged from 27 to 78 years.

The focus of the 17 studies was varied: 11 focused broadly on carers' experiences of caring for a family member with behaviour that challenges, and receipt of support services/ interventions; 3 studies interviewed parents whose child attended residential schools; and 3 studies addressed other specific aspects of carers' experience such as admissions to an emergency psychiatric service, experiences of using restraint procedures with their adult offspring, and support received from GPs.

Further information about included and excluded studies can be found in Griffith 2013b. A summary of the findings from Griffith 2013b is presented below for each theme.

4.3.1.1 Theme 1: Love

The love carers had for their family member with a learning disability was a constant presence throughout the interviews, although was only explored directly in 1 study (Hubert 2010) in which the author described:

> *A… love. (…) mothers often admitted to quite explicitly.* (Hubert 2010; p. 219)

Despite love being fundamental to the experience of being a carer, the theme was only addressed directly by 1 study (Hubert 2010). For many mothers in this study, their family member with behaviour that challenges had become the centre of their lives:

> *My heart is always where he is… I feel closer to him than to anybody*. (Hubert 2010, p. 219)

Getting good support services for their family member with behaviour that challenges goes to the heart of their role as carers. Carers wanted to maintain their family member's dignity,

safety and to ensure that they were genuinely cared for as an individual and included in the community around them:

> *At home we try to give Andrew a little bit of independence and privacy.* (Elford et al. 2010, p. 79)

Carers holistic concerns about their family members' intellectual, social and emotional development were often beyond the boundaries of what support services were reported to deliver (see Theme 4).

Frustration was evident when support services did not provide appropriate care or when they failed to understand the needs of their family member (Qureshi 1992; Robertson et al. 1996; McGill et al. 2006a):

> *It's having mental tick boxes in their [service providers'] heads of autistic traits that don't actually have any bearing, or fit in at all with what your son's like.* (Wodehouse & McGill 2009, p. 649)

The theme of love was also apparent in reports of putting their family member's safety before their own:

> *Rather than [...] both of us getting hurt [...] I'd sooner, rather he didn't get [...] seriously hurt, I'd sooner [...] put myself […] in that position, I'm his mother.'* (Elford et al. 2010; p. 80)

Carers expressed motivation for wanting excellent support, and also the resultant frustration whenever support services did not meet expectations, further highlights their love for their family member:

> *Very little of the time did they ever speak to her [family member]. They would just talk to me about what she needed, but she is fairly high functioning…I felt it was a respect thing; they would ignore her and talk to me.* (Weiss et al. 2009, p. 358)

Love for their family member helps carry some parents through many of the difficulties of raising and supporting a family member with a learning disability and behaviour that challenges:

> *He's a good wee soul. He's hard work, but he's worth it, you know. I wouldn't part with him. (Hubert 2010, p. 219)*

4.3.1.2 Theme 2: Altered identity

Whilst caring deeply for their family member, carers reported a loss of a wider self-identity:

> *I'm not allowed to be a person, I'm just Penny's mum that cares for her 24 hours a day.* (Qureshi 1992; p. 113)

> *I am so stressed, I'm just living without a life.* (Allen et al. 2006, p. 359)

For many, the role of a 'carer' becomes the predominant identity, which has an insular effect on themselves and their immediate family. Conversely, the minority of carers wholly identified with and valued their all-consuming caring role:

> *I'm not worried…about what I'm missing out because none of it, if I didn't have him [son], none of it is worth anything anyway (…) that's why it's no big deal to look after him, I'm doing what I want really*. (Hubert 2010; p. 219-20)

For carers who had their family member living at home with them, the home was reported to be a place of hard work, where carers were 'on-duty' at all times:

> *It's a 24 hour, 7-day involvement. It's always Matthew. It gets kind of hard for me and my kids. Everyday we're affected.* (Fox et al. 2002, p. 444-45)

Carers also spoke of having little spare time:

> *Everything suffers because you haven't got time for yourselves, any quality time because everything centres on time for the child.* (Brown, et al., 2011, p. 913)

Many carers spoke of themselves and their family becoming socially isolated. This was explicitly linked to behaviour that challenges, which meant that they could rarely take their family member out of the family home, for fear of an episode:

> *She [mother] was in prison virtually because of his behaviour, she couldn't even go out in the garden without him misbehaving. We didn't get any visitors, as they were too scared of him to come round. It was a lonely life*. (Robertson et al. 1996, p. 86)

As their family member gets older, carer isolation increases as behaviour that challenges become progressively more difficult and embarrassing to manage in public

> *It's growing up that has separated me with the outside world with Arturo, because you are limited to where you can go with him, because of his behaviour problems*. (Fox et al. 2002; p. 447)

Although underpinned by deep love for their family members, the caring role was often described as a chronic strain for carers and the whole family. While on the surface, these seemed like 2 disparate emotions, the dual occurrence of love and strain ran throughout reports: the strain arising from the all-consuming role of providing good and loving care to their family member all day, every day.

4.3.1.3 Theme 3. Crisis management

An episode of behaviour that challenges was always reported to have a significant emotional and/or physical impact. Carers recounted some of the most difficult instances of behaviour that challenges:

> *I was attacked by my son – punched, kicked, hair pulled – then, in the same incident, pushed against a wall. Whilst I lost consciousness and was on the ground, I was repeatedly kicked*. (Allen 2006, p.358-59)

Other, low-intensity but high-frequency behaviours that challenge were also reported to be very stressful for parents:

> *When I am around him it is constant noise. He talks or squawks. By afternoon I am frazzled.* (Turnbull & Reuf 1996, p. 283)

As well as dealing with the immediate physical effects of an episode of behaviour that challenges, the emotional strain of self-harming and aggressive behaviours was described as equally difficult:

> *It's the most distressing thing possible to watch your child self harming. As a mother, it kills you*. (Allen et al. 2006; p. 359)

> *I was bruised all over, but the emotional pain was far more to cope with.* (Allen et al. 2006, p. 359)

In some instances, behaviour that challenges became so severe that carers needed to utilise crisis management, such as restrictive interventions (such as direct physical contact, use of barriers [such as bed rails or padding] or equipment [such as splints and straps]) or admission to a hospital emergency department. These options were fraught with difficulties

for carers and were reported to be used only as a last resort (Weiss et al. 2009; Elford et al. 2010).

As well as being a very stressful crisis management situation, the ethical dilemma faced by carers when using restrictive interventions themselves was also reported to be a significant emotional strain:

> *It's a very fine line between whether it's right to restrain or wrong, and I'm not qualified to say.* (Elford et al. 2010, p. 78)

In Canada, families in crisis as the result of their family members' severe behaviour that challenges turned to the hospital emergency department, but did not always receive helpful support. Families were asked to wait in noisy waiting rooms, causing additional agitation to their family member, and staff lacked experience and skill:

> *They do not have psychiatrists trained to deal with this population.* (Weiss et al. 2009, p. 357)

In no paper did carers attribute blame to their family member for engaging in behaviour that challenges or resent them for causing them strain. Instead, causal attributions focused on the lack of support services for their family member or on their family member's inability to communicate:

> *He would bite his thumb almost in half, he can't communicate.* (Brown et al. 2011, p. 912)

Carers felt that access to proactive and consistent support for their family member's behaviour that challenges, rather than a reactive crisis management support, would reduce the frequency of severe episodes of behaviour that challenges.

4.3.1.4 Theme 4: Support is not just 'challenging behaviour services'

Despite the strain of caring being evident throughout the reviewed studies, carers rarely spoke of the need for emotional support for themselves. Instead, their talk focused on the support needed for their family member with a learning disability.

Across all studies, carers did not differentiate between specific 'challenging behaviour' support and more general support issues. Carers had a holistic view of the support their family member needed, in which behaviour that challenges issues and more general support were clearly intertwined. Carers felt that all support services (from schools, to respite care, to day centres) needed to have an understanding of their family members' behaviour that challenges to support them adequately. Thus, all services needed to have an element of being a 'challenging behaviour' service. Themes 4.1– 4.3 reflect carers' relationships with support services, the difficulties caused by bureaucratic processes, the impact of poorly trained professionals and support staff, and the positive impact of receiving reliable and proactive support services for their family member.

4.3.1.4.1 'Us' versus 'them:' Relationships with support services

Cares' most frequent description of professionals and support services were negative in tone, and phases such as '*battle*' and '*banging your head against a brick wall*' (Elford et al. 2010; p. 80) were frequently used. In addition, there was talk about being overwhelmed and stressed by bureaucratic processes (Qureshi 1992; Ruef et al. 1999; McGill et al. 2006b):

> *It just seems overwhelming, and after years and years of fighting the bureaucracy, and looking for services, and trying to get someone to listen, that we run out of energy after a while.* (Ruef et al. 1999; p. 50)

This was particularly evident when bureaucracy got in the way of meeting the needs of carers:

> *I don't want to know about that [explanations of joint planning or interagency relationships], I just wanted to know about a night's sleep and a break.* (Qureshi 1992, p.109)

There was little evidence of collaboration and partnership with services and professionals in the majority of studies. Many carers found that receiving a support service was typically only a result of huge effort of their part:

> *Find[ing] out what provision was available on our own, no-one offered direction or advice*. (McGill et al. 2006b; p. 606)

> *I feel that unless…make a nuisance…pester people to death, nothing is done.* (McGill et al. 2006a; p.162)

Some reported that respite care – a highly valued break – was very difficult to obtain:

> *The pot-luck aspect of respite care… most effective tool for coping in my view-is a national disgrace.* (McGill et al. 2006a; p. 162)

Such valued services were reported to be either unavailable or very difficult to obtain:

> *A joke, the only time you could get it was at times you didn't really need it like a Wednesday evening. We needed it at weekends really.* (Robertson et al. 1996; p. 85)

Support services were regarded as complex and cumbersome systems, and parents were often overwhelmed; 1 parent described arranging services for her son as '*a full-time job in itself*' (Ruef et al. 1999, p. 50).

In addition, carers sometimes felt that their opinions were marginalised or ignored by services:

> *Nobody listens, I found out that professionals actually hold another meeting after I have attended an arranged meeting.* (McGill et al. 2006b; p. 606)

> *You've got all that experience of dealing with Jenny and your views aren't, you know, as if it doesn't matter.* (Elford et al. 2010, p.80).

A few carers recognised that some professionals tried their best to help but, like carers themselves, they had little individual power within their support services:

> *I think she [social worker] does her best to within what limits she can go.* (Qureshi 1992, p. 118)

Carers could see that professionals were bound by the same bureaucracy as they were, and overall found the structure of service systems as unhelpful to collaborative working, cumbersome, time-consuming and tiring.

4.3.1.4.25 Level of need exceeds level of service

A primary complaint of carers was that professionals did not have the expertise to be able to understand the complex needs of their family member and thus could not provide a service that met their needs:

> *I'm just thoroughly and continually amazed and appalled at the lack of information that the professionals have on autism.* (Ruef et al. 1999; p. 49)

> *I am aware of his behaviour triggers but I cannot…get the support or understanding outside of my care to ensure my child's behaviour is managed.* (McGill et al. 2006a; p.162)

Carers deemed the advice of professionals that lacked the expertise to deal with complex behaviour that challenges as ineffective:

> *They were sort of saying (...) 'just keep doing what you are doing,' they sort of didn't really come up with any [strategies].* (Wodehouse & McGill 2009; p. 649)

Lack of expertise meant that some professionals were not flexible enough to take individual circumstances into account. After explaining the advice she had received about implementing a behavioural intervention at home, 1 carer said:

> *You come and live my life for a day and see how you would put that intervention in, if it's actually applicable and appropriate.* (Wodehouse & McGill 2009, p. 649)

Lack of skilled support/teaching staff and the resultant inability to deal with behaviour that challenges could lead to the family member being excluded from school or other support services (Ruef et al. 1999; McGill et al. 2006b; Wodehouse & McGill 2009; Hubert 2010). Exclusion, a common experience throughout the reviewed studies, leaves carers to cope at home for more hours with no additional support:

> *School were 'phoning saying 'Can you come and pick him up? We can't cope.' I just think 'Yeah it's me on my own here, you've got a whole team of people.* (Wodehouse & McGill 2009; p. 650)

Some respite services asked carers to be '*on call*' in case they couldn't cope with the family member's behaviour that challenges. This meant that carers were unable to relax and prevented them from having a '*true*' break:

> *They say 'We'll take her a night as long as you are at the other end of the 'phone in case we can't cope'. And I thought 'Well that's no good to me.' You know I couldn't send her there with piece of mind.* (Qureshi 1992, p. 133)

Apparent throughout the studies was carer's general frustration and distrust of support services as a consequence of the limited expertise among their staff. Some parents reported instances when their family member came back from a support service with increased behaviour that challenges, indicative of it not being well managed, or with unexplained physical injuries:

> *It must be three or four times he's come back like that [with physical injuries] – one day all his head was cut open. And they don't let you know how it's happened.* (Qureshi 1992, p.116)

Some carers reported ceasing to use much-needed services because of concerns for their family member's wellbeing, or because the efforts involved in organising access to the service far outweighed any benefit gained from a break.

4.3.1.4.3 Appreciation of good support services

The majority of included papers reported very few positive comments about services. Of the positive comments that were reported, carers were deeply appreciative of '*good*' professionals, who were pro-active, genuinely interested in the wellbeing of their family member, and who communicated openly and honestly (Ruef et al. 1999):

> *Because our children are very challenging, you've got to have respect and honesty and be family-orientated. It's got to be, because we are all quite vulnerable; parents at times are at their lowest points.* (McConkey et al. 2011, p. 259)

In 5 studies, carers generally reported high levels of satisfaction with a particular service their family member received. These services were praised by carers for having professionals with high levels of expertise, collaborative working between carers and professionals, their family members' behaviour improving and having confidence in services being able to cope with behaviour that challenges. However, all of the 5 studies were conducted in close collaboration with the service providers themselves.

These points almost exactly mirror areas carers felt were lacking in most received support (Themes 4.2.2.4.1 and 4.2.2.4.2). Thus, these features seem to be core to carers' experiences of services – whether good or bad.

Three studies were conducted in collaboration with residential schools (Brown et al. 2011; McGill et al. 2006b; Robertson et al. 1996), 2 of which used a behaviourally orientated approach. Most carers in these studies reported a dramatic improvement in their family members' behaviour after attending the school:

> *He used to be very violent and wreaked the house but while at Beech Tree his behaviour improved drastically. You could take him out to pubs and out for meals.* (Robertson et al. 1996, p. 86)

Some carers reported that the improvement in their family members' behaviour affected the whole family:

> *We've seen a noticeable improvement in his behaviour, so much so that home life for everyone, myself, my wife, and the other two children, has improved dramatically.* (Brown et al. 2011, p. 913)

In 2 studies (Fox et al. 1997; McConkey et al. 2011), community support services were praised for a collaborative approach and their honest and open communication with carers:

> *Look [s] at how best to serve the child and the family (…) It's always about problem solving and how to make it work.* (McConkey et al. 2011; p. 259)

Services most appreciated by carers were those that were proactive and able to work with parents when problems arose. Some carers reported learning techniques from staff at respite placements that they began to use at home:

> *I have learned from the staff what they were doing and I took it home and extended it, so now he does sleep.* (McConkey et al. 2011; p. 263)

In contrast to the previous subtheme (Level of need exceeds level of service), papers did report that high quality respite care can help the entire family:

> *Although the short break was to provide us with a break (…) I realised it was providing my son with a break as well (…) I am happy that he is happy there.* (McConkey et al. 2011, p. 261)

Finally, although carers rarely spoke of their own needs as a priority for support services, they did appreciate having their own needs addressed:

> *And every time I talk to him [Dr] he'll give me word of encouragement. He'll say something like (…) 'the best thing you can do for him [child] is to love him' (…) I want to cry every time I come out of there.* (Fox et al. 2002, p. 444)

4.3.1.5 Theme 5: The future: Low expectations, high hopes

The majority of carers looked towards the future care of their family member with anxiety and fear:

> *His future is such a big, dark thing…so many things could go horribly wrong*. (McGill et al. 2006b; p. 610)

The main concern centred on the care of the family member when carers are no longer around to look after them. A primary fear was that their family member would not be loved and cared for like they are in the family home, would not have a genuine close relationship with anyone and would not be treated like an individual:

> *I worry that he [would not be] well cared for, that's what bothers me, who would care for him?* (Hubert 2010, p. 222)

Due to the lack of demographic information provided, it is difficult to ascertain patterns in the data, such as what services specific age groups received, although Hubert (2010) reported that carers rated support services for adults as being of poorer quality, and less reliable than when their family member was a child.

Some carers struggled to get support services to prepare for the transition to adulthood support services:

> *We have tried to get them on board since he's been 16 and a half asking why we had no input from the young adult team…he is 19 soon and we have heard nothing.* (McGill et al. 2006b; p. 610)

Others spoke of lack of funding, limited options for residential care and confusion about the process. A general feeling of helplessness about the future was often reported:

> *We are looking, but like we said there is nowhere for our Mary to go. We can't really, they haven't told us, like when she's 40 or 30, where she's supposed to go.* (Qureshi 1992, p. 117)

Some carers who had family members with a severe/ profound learning disability were so fearful for the wellbeing of their family member at the hands of support services that they hoped that their family member would not outlive them:

> *I'd rather give him an overdose, then see him go in there [residential service]…he'd be better off dead. What sort of life would he have? …They're [other service users] suffering in there because they can't say any different…you've got to think about the content of life, haven't you?* (Hubert 2010; p. 222)

> *I'd like to have the guts to do her in, rather than let her go there (…) she's not going to have any life in there so she might as well be done in.* (Qureshi 1992; p. 117)

Carers feared that if they were no longer able to oversee the care, their family member may be an easy target for sexual assault, or might be heavily drugged to control their behaviour that challenges (McGill et al. 2006b; Hubert 2010).

Despite low expectations, some carers still possessed high hopes for their family member's future care:

> *Ideally I would like him to be half an hour from home…in a very small home…looked after by familiar people where he is loved.* (McGill et al. 2006b, p. 611)

However, past experiences of support services for their family member meant that few carers felt this situation was likely to be a reality and for many, future was a place of both anxiety and uncertainty.

4.3.2 Evidence statements carer experience

Evidence from 17 (392 participants) qualitative studies was synthesised by 1 systematic review using meta-ethnography. The review was judged to be of adequate quality although

the authors did not assess the quality of the included studies. Five main themes were identified: (1) Love, (2) Altered identify, (3) Crisis management, (4) Support is not just 'challenging behaviour services,' and (5) The future. From theme (4), 3 further subthemes were identified: a) 'us' versus 'them' relationships, b) level of need exceeds level of service, and c) appreciation of good support services.

The recommendations which were developed from this section and the link to the evidence are at the end of the chapter where they are brought together with the reviews of the service user experience and the validation exercise with service users and carers undertaken for this guideline.

4.4 Expert advisory group validation

4.4.1 Introduction

Individuals with direct experience of services – that is, experts by experience – are integral to developing a service user and carer focus to the GDG and the guideline. The GDG included 3 parents of people with a learning disability and behaviour that challenges, who contributed as full GDG members to develop review questions, highlight sensitive issues and terminology and to bring the experiences of carers and families to the attention of the GDG. Unfortunately, it was not possible to recruit a service user to the GDG, due in part to the time demands of the GDG member role and format of the GDG meetings. However, it was considered crucial that the experiences of people with a learning disability were incorporated into the guideline. In order to achieve this, the GDG sought the views of people with a learning disability to inform the development of the guideline via the following organisations: The Elfrida Society and the Camden Speaking Up Rights Group whose aim is to improve the lives of people with a learning disability by educating health and council services and providing support. The GDG also sought the views of 2 groups of carers of people with a learning disability who display behaviour that challenges through The Challenging Behaviour Foundation, which provides information and support to families, carers and professionals caring for people with a learning disability and behaviour that challenges. The intention of this validation exercise was to test out emerging themes which related both to the themes in this chapter and also others that emerged during the course of the development of the guideline.

4.4.2 Service user focus group

4.4.2.1 Method

To recruit members of the group, staff at the Power and Control Group at The Elfrida Society (http://www.elfrida.com/) and the coordinator of the Camden Speaking Up Rights Group (http://www.advocacyproject.org.uk/service/surge/) were contacted. The Power and Control group is a group of people with a learning disability who represent the views of people with a learning disability in Islington, London. The group are consulted on local services and issues and hold larger forum meetings, which anyone with a learning difficulty in Islington can attend. The Camden Speaking Up Rights Group is a group of people with a learning disability who give advice to health and council services on what people with a learning disability need in London. Members of each group were asked if they were interested in taking part in the service user focus group. In total 4 members of the Power and Control Group and 5 members of the Camden Speaking Up Rights Group agreed to take part. The group were given a presentation on key emerging themes of the guideline and specifically their views and experiences on the following areas were covered: (1) the causes of behaviour that challenges, (2) staff training, (3) medication for behaviour that challenges, (4) other therapies for behaviour that challenges. Responses were recorded on a flip chart and have been summarised below. For a full report of the focus group see Appendix U.

4.4.2.2 Summary of findings

What are the causes of behaviour that challenges in people with a learning disability?

One of the main causes of behaviour that challenges the group described was an underlying physical or mental health problem which had not been addressed. The group described personal experiences of difficulties communicating physical or emotional problems to carers and family members. The general view was that professionals or family member's had often not taken the time to try and understand the person's underlying problem:

> *I had difficult behaviour as a child because it was hard to say how I was feeling.*

People did not find out early what was upsetting me, they did not do a proper assessment.

Some members of the group said that their own physical health problems had also been ignored by healthcare professionals in the past:

I had a lot of health needs in my life, but my needs were not being met.

Late diagnosis of health problems.

Within the group there was an overall sense that service users were rarely included in decisions about their care as their views were deemed unimportant. They also felt that there were too many healthcare professionals involved in their care. Being undermined in such situations was perceived as a potential contributor to behaviour which may challenge:

What the person themselves wants can get left out. Services are not person centred, not including the person in everything about their lives.

There are too many people involved in your life – staff, friends, family.

The group felt very strongly that a lack of support could lead to behaviour that challenges. They stressed the importance of having good quality relationships with staff and other people who supported them:

You need someone to talk to who you can trust.

What should staff training involve?

There was a strong feeling from the group that people with a learning disability should be involved in the interview process for recruiting members of staff and in delivering training. This was seen as a good way to empower service users and to make sure potential candidates were suitable for the role:

Staff should be interviewed by people with learning disabilities.

They need training from people with learning disabilities before they start, about what their job is about.

In light of the Winterbourne View report, some members of the group felt that there was an extra need to monitor staff and to check they did not have a history of abusive behaviour. They also stressed that staff members should have more support from managers as the role was likely to be stressful:

Staff need good back up support and expert advice from their managers and others.

What are your views on medication for behaviour that challenges?

The general view among the group was that medication should only be used in the short term or in addition to other approaches. They also felt that it was important to take the time to understand the cause of the behaviour before resorting to medication:

A balance of both can work – medication can help the person to be calm so problems can be sorted out.

It is important to talk to the person and try to solve the problem at its root cause.

What are your views on psychological therapies for behaviour that challenges?

The group did not have any experience of psychological therapies for behaviour that challenges so instead they talked about therapies, other than drug treatment, which may help

in preventing or reducing behaviour that challenges in this population. These included art, music and dance therapies, relaxation therapies but also simple interventions, *'someone there to listen would be helpful', 'giving the person the chance for a break, respite, change of scenery'.*

4.4.3 Carer focus group

4.4.3.1 Method

The Challenging Behaviour Foundation invited 18 family members to 1 of 2 focus groups, 1 in London and 1 in Birmingham. Of these, 17 attended and contributed. The carers were divided into 2 groups: (1) carers of family members aged 18 to 37 years, and (2) carers of family members aged 7 to 21 years. The families worked in small groups and addressed each question in turn recording their discussion on flip chart paper. They then came together as a larger group to discuss their key issues and concerns and this information was also recorded. The same method was used to generate and record the 'Any Other Issues' concerns. Finally, each participant was asked to write out on a piece of paper his or her individual key priority statement for the GDG. Findings are summarised below, for a full report of the focus group see Appendix V.

4.4.3.2 Summary of findings

Access to assessments: what are the experiences of families accessing services for children, young people and adults with a learning disability and behaviour that challenges?

The carers thought that assessment should start early and be seen as part of a preventative strategy. It was viewed as a dynamic ongoing process that needs to be regularly reviewed and updated:

> *We need to be proactively planning for life to prevent problems developing. Everything is so short term and narrow in focus.*

The overarching message of the carers taking part in both the workshops was that assessment should always lead to something- an outcome, and too frequently this does not happen:

> *Assessments do not produce action plans or guidance. The behaviour specialist came in and did an assessment, discussed it with the staff team but never followed it up to see if it had been implemented and it wasn't! What a waste of time that was!*

There was also a real concern that assessments are not person centred and individualised. One carer pointed out that often:

> *The tools they use are not person centred. I don't think they see Peter as a person in the round he is just a cluster of labels to them.*

A factor that families felt contributed to the lack of person centred assessment and the ability of people to really 'see' their child/ adult was caused by 'diagnostic overshadowing':

> *Their label means other things about them get missed, (such as health needs), there are so many assumptions.*

The families told us that they often feet 'under the spotlight' when meeting professionals, and that they are being assessed themselves, but this is never explicitly stated. They often feel that they are not listened to and judged to be part of the problem rather than partners in working to find the best solution for their family member.

What is the experience of the use of medication for children, young people and adults with a learning disability and behaviour that challenges and their families?

The families that participated in both workshops shared many of the same concerns about medication. They were concerned that medication is frequently the only sort of intervention offered to their family member:

> *My daughter was offered Risperidone at 15 years old. On reading the research I questioned why it was being offered when there were no positive results for females. I asked for therapy and not medication. I was told there is not enough money so it was medication or nothing. I chose nothing.*

The families said they are not being offered enough information about the medications that are being prescribed for their family member. This includes issues like:

- Potential side effects
- Interaction (poly-pharmacy) with any other drugs being prescribed
- Interaction with any home based remedies the person might take for a cold or a headache.

There was also a very strong view that:

> *[A]ntipsychotics should never be used for challenging behaviour unless there is an underlying mental health problem.*

CAMHS were specifically singled out for criticism in the children and young people workshop. The feeling was that Ritalin has some very bad side effects so assessment about whether to use it had to be extensive and thorough. There was a concern that local CAMHS services lacked the sort of expertise that is needed to do this properly. This was also felt to be true in relation to the prescribing of melatonin:

> *CAMHS need to be more than just drug pushers.*

There was a consensus that there should be a minimum of a mandatory annual review of medication and this should involve a blood test to review medication levels and physical functioning. This consensus links to a strong feeling that there should be more information provided to GP's and a better link between primary care and specialist prescribers should be developed.

Behavioural interventions: what support is given to families when involved in behavioural programmes and do they help children, young people and adults with a learning disability and behaviour that challenges in the long term?

After medication, behavioural interventions were identified as the second most widely used approach for supporting and managing the needs of children, young people and adults with a learning disability and behaviour that challenges. The families participating in the workshops were unanimously positive about this approach. However, they were concerned that there is not enough Positive Behavioural Support (PBS) (or ABA) on offer and available in all areas.

All the families were concerned over the issue of equity of access to positive behavioural interventions both in terms of information and availability in their local area. The families of the children's' group also feel strongly that access to PBS (and ABA) should be part of a proactive early preventative strategy:

> *I cannot imagine what our life would be like now if we hadn't found out about ABA early on. It has made such a difference to all our lives!*

This same mother also said that she felt lucky to have been told about ABA from another parent, and when services refused to pay for the assessment, that they were fortunate to have the money to pay for her son's assessment.

There were also concerns that some services think they are offering PBS (CAMHS and other providers were mentioned) but were not providing the 'real deal':

> *Behavioural interventions are only as good as the people delivering them.*

Staff development and workforce issues were a big concern for families:

> *Consistency and expertise are needed.*

Yet the families' experience is often the opposite:

> *We don't pay them enough. They can get more working stacking shelves in a supermarket. If we don't value them how can we expect them to value our children.*

Transition between services: what are the experiences of transitioning or moving between services? (for example, child to adult services)

Families were clear that all good transitions involve preparation, planning and execution of an action plan that everyone has signed up to, whatever the transition is. Preparation and planning always need to involve the person, (even if they lack capacity), and their family. Even if the person with a learning disability who displays behaviour that challenges cannot communicate using verbal communication, it is essential to find other ways of finding what their preferences would be as they make a change in their life. The families said they thought that people with a learning disability and behaviour that challenges are particularly vulnerable to experiencing chaotic transitions. They attribute this to the lack of expertise in local services to enable the needs of people with more complex needs to be met:

> *There is a lot of great information out there now to help you prepare and plan for the time your child moves into adulthood. The sad thing is that where we lived it was all left to the last minute and we were told that when he left school his only choice was the local college but when we talked to the college they made it clear that they couldn't cope with Josh and he ended up sitting at home with me! He got bored and things went from bad to worse and he ended up being placed in a home miles away.*

Families shared their good and bad experiences of transition but it has to be acknowledged that the bad experiences heavily outnumbered the good. The good practice examples demonstrated that when an investment was made in giving time to preparing and planning the transition, it worked well.

> *The new staff team worked with Kay in her old environment for four months before supporting her to move to her new home. We (my daughter and myself) were involved in recruiting the new staff team. Videos of the interview questions were sent to Kay.*

Any other issues: not covered explicitly in relation to the other questions

Carers expressed other issues which were not explicitly elicited from the questions asked. These included: not feeling valued by professionals, the importance of having good information about the disorder and services, the lack of integrated care, the need for a more flexible approach to evidence, personal budgets and having access to family advocates.

4.5 Recommendations and link to evidence

Recommendations	
	1. Work in partnership with people who have a learning disability

	and behaviour that challenges, and their family members or carers, and: • involve them in decisions about care • support self-management and encourage the person to be independent • build and maintain a continuing, trusting and non-judgemental relationship • provide information about the nature of the person's needs, and the range of interventions (environmental, psychosocial, psychological and pharmacological) and services available to them, in an appropriate language or format (including spoken and picture formats, and written versions in Easy Read style and different colours and fonts) • develop a shared understanding about the function of the behaviour and what maintains it • help family members and carers to provide the level of support they feel able to. 2. When providing support and interventions for people with a learning disability and behaviour that challenges, and their family members or carers: • take into account the severity of the person's learning disability and their developmental stage • aim to provide support and interventions in the person's home, or as close to their home as possible, in the least restrictive setting • aim to prevent the development of future episodes of behaviour that challenges • offer support and interventions respectfully, and ensure that the focus is on improving the person's support rather than changing the person • ensure that they know who to contact if they are concerned about care or interventions, including the right to a second opinion • offer independent advocacy to the person and to their family members or carers.
Relative values of different outcomes	The GDG agreed that experience and satisfaction of service users and carers was the most important outcome. Involvement in the planning of care provided and adequate information that allowed for proper participation in decision making was also important.
Trade-off between clinical benefits and harms	The GDG agreed that lack of involvement in care planning and inadequate information were a serious impediment to the provision of effective care. Harms were likely very limited but attention should be paid to the right to confidentiality of both service users and carers.

Trade-off between net health benefits and resource use	The GDG took into account that providing information and support to service users and carers, as well as promoting their involvement in care planning, might entail modest resource implications, which would, however, be offset by provision of more effective care and of improved outcomes resulting from service users' and carers' involvement in decision making. Improved outcomes for people with learning disabilities and behaviour that challenges are also expected to lead to a reduction in costs associated with behaviour that challenges, which can be substantial, for example costs incurred by inpatient placements.
Quality of evidence	Published systematic reviews judged to be of high quality was used, and overall the included studies were rated as good quality.
Other considerations	The experience of care for service users, families and carers demonstrated than many people had experienced significant shortfalls in access to services and the quality of care provided. It was striking that although many service users, families and carers had clear views on what might help them, they felt that often their voices were not heard. Families felt that the support that they provided was not recognised and lack of support often undermined them in their attempts to support their relative. A number of specific concerns were also identified including the over use of medication, limited access to psychological interventions, avoidable and costly out of home placements and assessments often not being followed through. Considering all this information, the GDG judged that it was important to set out some general principles underpinning good care. These focused on the proactive involvement of services users, families and carers in the planning and delivery of their care and the setting in which it is delivered. In addition to the development of the recommendations in this chapter the reviews of service user and carer experience also contributed to the development of recommendations in other chapters in this guideline, in particular the chapters on assessment, interventions for carers and the organisation and delivery of care.

1
2

5 Interventions for carers

5.1 Introduction

The economic value of unpaid carers in the UK has been estimated at £119 billion per year (Buckner & Yeandle, 2011) with approximately 15% of all carers in the UK caring for someone with a learning disability (The Princess Royal Trust for Carers, 2004). It is estimated that more than 65% of people with a learning disability in England are living with their parents or another relative (Emerson & Hatton, 2008). A large number of carers are therefore faced with meeting the needs of their family member, partner or friend often with minimum support from statutory services (see Section 4.1).

Family members who care for adults with a learning disability and behaviour that challenges are a vulnerable group. This group has been shown to be at increased risk for a variety of negative outcomes including poorer mental and physical health and reduced socio-economic resources compared with the general population (Gallagher et al., 2008; Hastings, 2002b; Most et al., 2006).

A recent systematic review of carers of family members with a learning disability and behaviour that challenges (Griffith & Hastings, 2013) revealed that carers performed a complex juggling act, managing day-to-day general care demands and the particular stresses associated with behaviour that challenges (for example, physical injury and fear), battling with services or the general lack of suitable support from services, and preparing for a future when they would no longer be able to provide care and support to their relative. It was also clear from this review that these considerable demands were managed in the context of a strong commitment to the person with a learning disability.

Providing adequate support and appropriate interventions to carers first requires that they are identified. At present there is no clear service that has been tasked with this role, although some improvements have been made in recent years. Social services have a statutory duty to offer carer assessments but this only benefits a number of families and resources may be limited to implement the outcome of the assessment.

GPs are now encouraged to identify patients who have a role as a carer. They can offer additional support in the form of carer packs and seasonal flu jabs, but records can be patchy and often do not have sufficient information. GPs may not always recognise the burden of caring for someone with a learning disability and behaviour that challenges. There will also be families who no longer offer direct care (because their child has grown and left home) who may still have significant additional needs but are unlikely to be identified in the records.

Families often report fears for the future care of their child and worry that services might fail them because previous experiences may not always have been adequate. Current services can appear to have a bias to crisis management with fewer resources being made available for early intervention or prevention. Without a commitment to reduce the risk of behaviour that challenges, problems have to escalate before additional support is offered. Response to crisis can be inadequate and too late and result in placement breakdown. This can result in people moving to inappropriate placements, often at some distance from the family home, for an unnecessarily long time.

Systematic reviews (Griffith & Hastings, 2013) have suggested a need for trusted partnership between professionals/services and family members, increased skills for family members, and the need for support in coping with the emotional demands of caring for an adult with a learning disability and behaviour that challenges. Parents, in particular, reported being socially isolated, with almost their whole existence focused on supporting their son or daughter.

Intervention and support for parents of children (rather than adults) with a learning disability and behaviour that challenges have been subject to some research attention. In particular, behavioural parenting training methods have been applied to parents of children and subjected to evaluations in RCTs (McIntyre & Brown, 2013). As yet, no RCT has been undertaken with families with children who are now adults.

5.2 Review question: In families and carers of people with a learning disability and behaviour that challenges, what are the benefits and potential harms of interventions aimed at improving their health and wellbeing?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 13. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 13: Clinical review protocol summary for the review of interventions aimed at improving carers' health and wellbeing

Component	Description
Review question	In family and carers of people with a learning disability and behaviour that challenges, what are the benefits and potential harms of interventions aimed at improving their health and wellbeing? (RQ5.1)
Population	Family and carers of children, young people or adults with mild, moderate, severe or profound a learning disability and behaviour that challenges. The term 'carers' encompasses both family carers and paid carers.
Intervention(s)	Included interventions: All interventions targeted at improving the health and wellbeing of family and carers. Excluded Interventions: Interventions targeted at improving the health and wellbeing of people with a learning disability and behaviour that challenges Studies evaluating the process of interventions rather than outcomes (for example, uptake of programme).
Comparison	• Any control • Treatment as usual, no treatment, waitlist control, attention control or any alternative management strategy.
Critical outcomes	• Family and carer quality of life • Family and carer mental and psychological health outcomes • Family and carer stress and resilience • Family and carer satisfaction.
Study design	RCTs and systematic reviews.
Note. RCT = Randomised controlled trial.	

5.2.1 Clinical evidence

5.2.1.1 Cognitive behavioural interventions versus any control for family and carers

Ten RCTs (N = 837) met the eligibility criteria for this review: Feinberg 2014 (Feinberg et al., 2014), Gammon 1991 (Gammon, 1991), Greaves 1997 (Greaves, 1997), Kirkham 1990 (Kirkham, 1990), Neece 2014 (Neece, 2014), Nixon 1993 (Nixon, 1993), Schultz 1993 (Schultz C.L., 1993), Singer 1988 (Singer, 1988), Singer 1989 (Singer, 1989), Wong 2010

(Wong, 2010). Of the 10 eligible studies, 7 (N = 610) included sufficient data to be included in the evidence syntheses and 3 (N = 147) included critical outcome data that could not be included in the meta-analyses because of the way the data had been reported (Gammon 1991; Greaves 1997; Neece 2014); a brief narrative synthesis is therefore given to assess whether the findings support or refute the meta-analyses. Greaves 1997 was a 3-armed trial (N = 54); for the purposes of this review comparison only the experimental and no treatment control group will be utilised (N = 37). An overview of the trials included in the meta-analysis can be found in Table 14.

Summary of findings can be found in Table 15. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

No data were available for the critical outcomes of family or carer satisfaction.

The study flow diagram and evidence tables can be found in Appendix N, and exclusion list in Appendix Q.

Table 14: Study information table for trials included in the meta-analysis of cognitive behavioural therapy (CBT) for family and carers versus any control

	CBT versus any control
Total no. of studies (N[1])	10 (820)
Study ID	(1) Gammon 1991[2] (2) Greaves 1997[2,3] (3) Feinberg 2014 (4) Kirkham 1990 (5) Neece 2014[2] (6) Nixon 1993 (7) Schultz 1993 (8) Singer 1988 (9) Singer 1989 (10) Wong 2010
Country	(1, 3 to 6, 8 to 9) USA (2, 7, 10) Australia
Diagnosis	(1, 4 to 5, 8 to 10) DD (2) Down Syndrome (3) Autism (6 to 7) LD
Carer age (mean)	(1, 3 to 5, 7, 10) 34-47 (2, 6, 8, 9) Not reported
Carer sex (% Female)	(1 to 4, 6, 10) 95-100 (5, 8) Not reported (7, 9) 50-65
Carer ethnicity (% White)	(1, 2, 5 to 9) Not reported (3) 44 (4) 92 (10) 0
Treatment length (weeks)	(1 to 5, 8, 10) 8-10 (6, 7) 5-6

	CBT versus any control
	(9) 16
Intervention	(1, 9) Coping Skills Training Program (2) Rational-Emotive Parent Education Program (3) Problem-solving education (4) Life skills intervention training (5) Mindfulness-based stress reduction (6) Cognitive restructuring treatment program (7) Caring for Parent Caregivers (8) Stress management training (10) CBT
Comparison	(1, 2, 7) No treatment (3, 4, 8, 9) TAU (5, 6, 10) Wait list

Notes: N = total number of participants; DD = developmental disabilities; LD = learning disability; TAU = treatment as usual.
[1] Number randomised.
[2] Data not reported in a meta-analysable format; findings are described narratively.
[3] 3-armed trial; only intervention and no treatment control arms utilised.

Table 15: Clinical evidence profile: cognitive behavioural interventions versus any control for family and carers of people with a learning disability and behaviour that challenges

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Any control**	Corresponding risk **Cognitive behavioural intervention**			
Carer health and wellbeing (depression) - post-treatment		The mean carer health and wellbeing (depression) - post-treatment in the intervention groups was **0.35 standard deviations lower** (0.54 to 0.15 lower)		428 (5 studies)	**Moderate**[1]
Carer health and wellbeing (depression) - follow-up Follow-up: 46 to 104 weeks		The mean carer health and wellbeing (depression) - follow-up in the intervention groups was **0.41 standard deviations lower** (0.79 to 0.04 lower)		130 (2 studies)	**low**[1,2]
Carer health and wellbeing (clinically depressed) - post-treatment	**224 per 1000**	**56 per 1000** (18 to 188)	**RR 0.25** (0.08 to 0.84)	111 (1 study)	**very low**[1,3]
Carer health and wellbeing (anxiety, trait) - post-treatment		The mean carer health and wellbeing (anxiety, trait) - post-treatment in the intervention groups was **0.5 standard deviations lower** (1.03 lower to 0.03 higher)		68 (2 studies)	**low**[1,2]
Carer health and wellbeing (anxiety, state) - post-treatment		The mean carer health and wellbeing (anxiety, state) - post-treatment in the intervention groups was **0.46 standard deviations lower** (1.12 lower to 0.2 higher)		36 (1 study)	**very low**[3,4]
Carer health and wellbeing (mental ill health) - post-treatment		The mean carer health and wellbeing (mental ill health) - post-treatment in the intervention groups was **2.19 standard deviations lower**		58 (1 study)	**very low**[3,4]

		(2.85 to 1.53 lower)			
Carer health and wellbeing (quality of life) - post-treatment		The mean carer health and wellbeing (quality of life) - post-treatment in the intervention groups was **0.87 standard deviations higher** (0.33 to 1.41 higher)		58 (1 study)	**very low**[3,4]
Carer health and wellbeing (stress) - post-treatment		The mean carer health and wellbeing (stress) - post-treatment in the intervention groups was **0.45 standard deviations lower** (0.78 to 0.12 lower)		384 (3 studies)	**very low**[1,2,5]
Carer health and wellbeing (stress) - follow-up Follow-up: mean 104 weeks		The mean carer health and wellbeing (stress) - follow-up in the intervention groups was **0.43 standard deviations lower** (0.9 lower to 0.05 higher)		76 (1 study)	**very low**[3,4]
Carer health and wellbeing (clinically stressed) - post-treatment	**293 per 1000**	**38 per 1000** (9 to 155)	**RR 0.13** (0.03 to 0.53)	111 (1 study)	**very low**[3,4]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

Note. CI = Confidence interval; RR = Risk ratio.

[1] Most information is from studies at moderate risk of bias
[2] Optimal information size not met
[3] Optimal information size not met; small, single study
[4] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[5] $I^2 > 40\%$

5.2.1.21 Support versus any control for family and carers

One RCT (N = 80) met the eligibility criteria for this review and was included in the evidence synthesis: Davis 1991 (Davis, 1991). An overview of the single trial included in the meta-analysis can be found in Table 16.

Summary of findings can be found in Table 17. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No data were available for the critical outcomes of family and carer quality of life, mental and psychological health, and satisfaction.

The study flow diagram and evidence tables can be found in Appendix N, and exclusion list in Appendix Q.

Table 16: Study information table for trials included in the meta-analysis of support and psychoeducation for family and carers versus any control

	Support versus any control	Psychoeducation versus any control
Total no. of studies (N[1])	1 (80)	2 (180)
Study ID	Davis 1991	(1) Bilgin 2009 (2) Yildrim 2013
Country	UK	(1, 2) Turkey
Diagnosis	LD	(1, 2) LD
Carer age (mean)	33	(1) 34 (2) 42
Carer sex (% Female)	100	(1, 2) 100
Carer	65	(1, 2) Not reported

ethnicity (% White)		
Treatment length (weeks)	66	(1) 1 (2) 4
Intervention	Parent Advisor Scheme	(1) Interactive education sessions (2) Psychosocial education program
Comparison	TAU	(1) Waitlist (2) TAU

Notes: N = total number of participants; DD = developmental disabilities; LD = learning disability; TAU = treatment as usual.
[1] Number randomised.

Table 17: Clinical evidence profile: support versus any control for family and carers of people with a learning disability and behaviour that challenges

Outcomes	Comparative risks (95% CI)		No of Participants (studies)	Quality of the evidence (GRADE)
	Any control	Support interventions		
Carer health and wellbeing (stress) - post-treatment		The mean carer health and wellbeing (stress) - post-treatment in the intervention groups was **1.21 standard deviations lower** (2.04 to 0.39 lower)	28 (1 study)	**very low**[1,2]

Note. CI = Confidence interval.

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

5.2.1.3 Psychoeducation versus any control for family and carers

Two RCTs (N = 180) met the eligibility criteria for this review and were included in the evidence synthesis: Bilgin 2009 (Bilgin, 2009), Yildrim 2013 (Yildirim et al., 2013). An overview of the trials included in the meta-analysis can be found in Table 16.

Summary of findings can be found in Table 18. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No data were available for the critical outcomes of family and carer quality of life, stress and resilience, and satisfaction.

The study flow diagram and evidence tables can be found in Appendix N, and exclusion list in Appendix Q.

Table 18: Clinical evidence profile: psychoeducation versus any control for family and carers of people with a learning disability and behaviour that challenges

Outcomes	Comparative risks (95% CI)		No of Participants (studies)	Quality of the evidence (GRADE)
		Corresponding risk		
	Any control	Psychoeducation		
Carer health and wellbeing (depression) - follow-up		The mean carer health and wellbeing (depression) - follow-up in the intervention groups was **0.84 standard deviations lower** (1.31 to 0.36 lower)	75 (1 study)	**very low**[1,2]

Follow-up: mean 4 weeks				
Carer health and wellbeing (burnout) - follow-up Follow-up: mean 8 weeks		The mean carer health and wellbeing (burnout) - follow-up in the intervention groups was **0.35 standard deviations lower** (0.77 lower to 0.06 higher)	90 (1 study)	**very low**[1,2]

Note. CI = Confidence interval.

[1] Crucial limitation for one criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

1

5.2.1.42 Mindfulness versus any control for paid carers

3 Two RCTs (N = 194) met the eligibility criteria for this review and were included in the
4 evidence synthesis: Bethay 2013 (Bethay et al., 2013), McConachie 2014 (McConachie et
5 al., 2014). An overview of the trials included in the meta-analysis can be found in Table 19.

6 Summary of findings can be found in Table 20. The full GRADE evidence profiles and
7 associated forest plots can be found in Appendix O.

8 No evidence was identified in relation to the specific subgroups identified in the review
9 protocol.

10 No data were available for the critical outcomes of family and carer quality of life, and
11 satisfaction.

12 The study flow diagram and evidence tables can be found in Appendix N, and exclusion list
13 in Appendix Q.

14 **Table 19: Study information table for trials included in the meta-analysis of**
15 **mindfulness interventions for paid carers versus any control**

	Mindfulness versus any control
Total no. of studies (N[1])	2 (194)
Study ID	(1) Bethay 2013 (2) McConachie 2014
Country	(1) USA (2) UK
Diagnosis	(1, 2) LD
Carer age (mean)	(1) 38 (2) 43
Carer sex (% Female)	(1) 77 (2) 26
Carer ethnicity (% White)	(1) 50 (2) Not reported
Treatment length (weeks)	(1) 6 (2) 3
Intervention	(1) Mindfulness and acceptance-based work stress reduction intervention + Applied Behaviour Analysis (2) Acceptance and Mindfulness Workshop
Comparison	(1) TAU/ Applied Behaviour Analysis (2) Wait list
Notes: N = total number of participants; DD = developmental disabilities; LD = learning disability;	

	Mindfulness versus any control
TAU = treatment as usual [1] Number randomised.	

Table 20: Clinical evidence profile: mindfulness versus any control for paid carers of people with a learning disability and behaviour that challenges

Outcomes	Comparative risks (95% CI)		No of Participants (studies)	Quality of the evidence (GRADE)
	Corresponding risk			
	Any control	Psychoeducation		
Carer health and wellbeing (mental wellbeing) - post-treatment		The mean carer health and wellbeing (mental wellbeing) - post-treatment in the intervention groups was **0.17 standard deviations higher** (0.19 lower to 0.53 higher)	120 (1 study)	**very low**[1,2]
Carer health and wellbeing (mental wellbeing) - follow-up Follow-up: mean 6 weeks		The mean carer health and wellbeing (mental wellbeing) - follow-up in the intervention groups was **0.28 standard deviations higher** (0.08 lower to 0.64 higher)	120 (1 study)	**very low**[1,2]
Carer health and wellbeing (mental ill health) - post-treatment		The mean carer health and wellbeing (mental ill health) - post-treatment in the intervention groups was **0.54 standard deviations lower** (1.06 to 0.02 lower)	154 (2 studies)	**very low**[3,4,5]
Carer health and wellbeing (mental ill health) - follow-up Follow-up: 6-13 weeks		The mean carer health and wellbeing (mental ill health) - follow-up in the intervention groups was **0.24 standard deviations lower** (0.72 lower to 0.24 higher)	154 (2 studies)	**very low**[3,4,5]
Carer health and wellbeing (stress) - post-treatment		The mean carer health and wellbeing (stress) - post-treatment in the intervention groups was **0.17 standard deviations higher** (0.19 lower to 0.53 higher)	120 (1 study)	**very low**[1,2]
Carer health and wellbeing (stress) - follow-up Follow-up: mean 6 weeks		The mean carer health and wellbeing (stress) - follow-up in the intervention groups was **0.05 standard deviations lower** (0.41 lower to 0.31 higher)	120 (1 study)	**very low**[1,2]
Carer health and wellbeing (burnout) - post-treatment		The mean carer health and wellbeing (burnout) - post-treatment in the intervention groups was **0.18 standard deviations lower** (0.86 lower to 0.49 higher)	34 (1 study)	**very low**[1,2]
Carer health and wellbeing (burnout) - follow-up Follow-up: mean 13 weeks		The mean carer health and wellbeing (burnout) - follow-up in the intervention groups was **0.08 standard deviations lower** (0.76 lower to 0.59 higher)	34 (1 study)	**very low**[1,2]

Note. CI = Confidence interval.

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study
[3] Most information is from studies at moderate risk of bias
[4] $I^2 > 40\%$
[5] Optimal information size not met

5.2.12 Economic evidence

No studies assessing the cost effectiveness of interventions for family and carers of people with a learning disability and behaviour that challenges were identified by the systematic

search of the economic literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

5.2.3 Clinical evidence statements

5.2.3.1 Cognitive behavioural interventions versus any control for family and carers

- Moderate quality evidence from 5 studies (N = 428), suggested that the cognitive behavioural intervention was more effective than the control in reducing depression in family and carers at the end of the intervention. At up to 2 years follow up, the intervention was similarly effective, but the evidence was from 2 studies (N = 130) and graded as low quality.
- Low to very low quality evidence from single studies with at most 111 participants, suggested that the cognitive behavioural intervention had a positive impact on other mental and psychological outcomes, quality of life and stress when compared with a control.
- 3 trials could not be included in the meta-analysis (N = 130). The authors of both Greaves 1997 (N = 37) and Neece 2014 (N = 51) reported that the cognitive behavioural intervention was more effective than no treatment control in reducing stress. Neece 2014 also reported that the mindfulness intervention was more effective than waitlist control in reducing depression. Conversely, Gammon 1991 (n = 42) reported no overall effect of the cognitive behavioural intervention, when compared with a control, on dimensions of parental stress at the end of the intervention.

5.2.3.2 Support versus any control for family and carers

- Very low quality evidence from a single study (N = 28), suggested that support was more effective than a control in reducing stress at end of the intervention.

5.2.3.3 Psychoeducation versus any control for family and carers

- Very low quality evidence from single studies (N = 75-90), suggested that psychoeducation was more effective than a control in reducing depression and burnout at 4 to 8 weeks follow-up.

5.2.3.4 Mindfulness versus any control for paid carers

- Very low quality evidence from up to 2 studies (N = 154) demonstrated some benefit in improving mental ill health of a mindfulness intervention when compared with a control at the end of the intervention, but was inconclusive with regard to mental wellbeing, stress and burnout.

5.2.4 Economic evidence statements

No economic evidence on interventions for family and carers of people with a learning disability and behaviour that challenges is available.

5.2.5 Recommendations and link to evidence

See section 5.4 for the recommendations and link to evidence relating to this section.

5.3 Review question: What are the benefits and potential harms of strategies aimed at engaging the family and carers of people with a learning disability and behaviour that challenges as a resource in the design, implementation and monitoring of interventions for the person with a learning disability and behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 21. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 21: Clinical review protocol summary for the review of strategies to engage family and carers as a resource in the design, implementation and monitoring of interventions

Component	Description
Review question	What are the benefits and potential harms of strategies aimed at engaging the family and carers of people with a learning disability and behaviour that challenges as a resource in the design, implementation and monitoring of interventions for the person with a learning disability and behaviour that challenges? (RQ5.2)
Population	Family and carers of children, young people or adults with mild, moderate, severe or profound a learning disability and behaviour that challenges. The term 'carers' encompasses both family carers and paid carers.
Intervention(s)	Strategies aimed at engaging the family and carers of people with a learning disability and behaviour that challenges as a resource in the design, implementation and monitoring of interventions.
Comparison	• Any control • Treatment as usual, no treatment, waitlist control, attention control or any alternative management strategy.
Critical outcomes	• Severity, frequency and duration of the targeted behaviour that challenges • Quality of life • Family and carer stress and resilience • Use of inpatient placements • Service user and carer satisfaction.
Study design	RCTs and systematic review of RCTs.
Note. RCTs = Randomised controlled trials	

5.3.1 Clinical evidence

The evidence base available for this section of the guideline was both anticipated to be and found to be extremely poor. No randomised controlled trials or systematic reviews were identified in the search. Consequently the GDG decided to adopt a more formal method of consensus (the modified nominal group technique) to identify areas of agreement on which to base guidance (see Chapter 3 for further details about the method).

A recent literature review on the area was used to develop the consensus questionnaire (see Appendix N): McIntyre 2013 (McIntyre & Brown, 2013). The literature review concerned recommended strategies for engaging family and carers as a resource in the design, implementation and monitoring of interventions for individuals with learning disability and behaviour that challenges. These strategies were adapted into 15 separate statements. In

order to address the various stages of behaviour that challenges in people with a learning disability, statements were split to address 3 levels: 1) universal prevention (all family and carers of people with a learning disability), 2) selective prevention (family and carers of people with a learning disability whose risk for developing behaviour that challenges is above average), and 3) indicated prevention/ intervention strategies (family and carers of people with a learning disability who have, or have specific risk factors for, behaviour that challenges).

The 16 GDG members' ratings of each of the 15 statements were compiled and ranked 1 to 15. The results of the consensus are presented in Table 22.

Table 22: Consensus results for statements concerning proposed strategies to engage family and carers as a resource in the design, implementation and monitoring of interventions

Statement	1st Round Consensus (%)	Rank
Universal prevention strategies		
1. Informal social support: Identify network of family and friends to provide emotional support and encouragement	75	12th
2. Formal social support: Identify formal resources available in the community	75	12th
3. Stress management: Practice self-care and healthy lifestyle	68.75	15th
4. Assessment: Developmental and behavioural screening surveillance, and monitoring	87.5	*6th
5. Parent education/ family behavioural supports: Widely available materials aimed at promoting positive parenting practices and behaviour management	100	*1st
Selective prevention strategies		
6. Informal social support: Identify network of family and friends to provide emotional support, encouragement, and instrumental support.	81.25	9th
7. Formal social support: Use of formal supports, including disability-specific services and specialty care.	100	*1st
8. Stress management: Practice self-care and healthy lifestyle	87.5	*6th
9. Assessment: Use behaviour-specific assessments (for example, direct observations, rating scales)	100	*1st
10. Parent education/ family behavioural supports: Group based parent management training	87.5	*6th
Indicated prevention/ intervention strategies		
11. Informal social support: Regularly utilise network of family and friends for emotional and instrumental support.	81.25	9th
12. Formal social support: Use of formal supports, including disability-specific services and specialty care.	100	*1st
13. Stress management: Practice self-care and healthy lifestyle, engage in individual or family counselling specially targeting stress management.	75	14th
14. Assessment: Use functional behavioural assessments or experimental functional analyses developed to inform behavioural treatment.	93.75	*5th
15. Parent education/ family behavioural supports: Group based parent management training	81.25	9th
*Ranked in the top half of the ranking table and will form the basis of evidence statements.		

Those consensus statements ranked in the upper half of the ranking table (rank 1st to 6th) were used to form the basis for the clinical evidence statements.

5.3.2 Clinical evidence statements

- At the level of universal prevention (that is all parents of a child with a learning disability), the GDG supported the use of: a) parent education/ family behavioural supports (materials aimed at promoting positive parenting practices and behaviour management); and b) assessment (developmental and behavioural screening surveillance, and monitoring).
- At the level of selective prevention, the GDG supported the use of: a) formal social support (including disability-specific services and specialty care); b) behaviour-specific assessments (for example, direct observations, rating scales); c) stress management (self-care and healthy lifestyle).
- At the level of indicated prevention/ intervention strategies, the GDG supported the use of: a) formal social support (including disability-specific services and specialty care); and b) assessment (functional behavioural assessments or experimental functional analyses developed to inform behavioural treatment).

5.3.3 Economic evidence

No economic evidence strategies aimed at engaging the family and carers as a resource in the design, implementation and monitoring of interventions for the person with a learning disability and behaviour that challenges was identified by the systematic search of the economic literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

5.3.4 Economic evidence statements

No economic evidence on strategies aimed at engaging the family and carers as a resource in the design, implementation and monitoring of interventions for the person with a learning disability and behaviour that challenges is available.

5.4 Recommendations and link to evidence

5.4.1 Support and interventions for family members or carers

Recommendations	3. Advise family members or carers about their right to a formal carer's assessment of their own needs (including their physical and mental health) and explain how to obtain it. 4. When providing support to family members or carers: • recognise the impact of caring for a person with a learning disability and behaviour that challenges • explain how to access family advocacy • consider family support and information groups if there is a risk of behaviour that challenges, or it is emerging • consider formal support through disability-specific support groups for family members or carers and regular assessment of the extent and severity of the behaviour that challenges. 5. If a family member or carer has an identified mental health

	problem, consider: • interventions in line with existing NICE guidelines **or** • referral to a mental health professional who can provide interventions in line with existing NICE guidelines.
Relative values of different outcomes	The GDG agreed that the following 4 outcomes for family and carers were critical: quality of life, mental and psychological health, stress and resilience, and satisfaction.
Trade-off between clinical benefits and harms	The GDG agreed that based on the available data there was reasonable evidence that some interventions for families and carers can have important benefits. The GDG also agreed by informal consensus to make a recommendation that all parents and carers should be made aware of and offered a carer's assessment. Although there was evidence for the treatment of depression only, the GDG was of the view that for those with identified mental health problems, healthcare professionals should consider providing, or referring for, interventions in line with existing NICE guidelines.
Trade-off between net health benefits and resource use	No economic evidence is available. Provision of interventions for families and carers has some resource implications. However, the GDG expressed the opinion that effective interventions for families and carers are likely value for money since they improve outcomes for families and carers and may consequently reduce healthcare resource utilisation associated with mental and psychological health problems experienced by carers, including depression and anxiety.
Quality of evidence	Although evidence came from RCTs, it was generally downgraded to low or very low quality due to risk of bias and small sample sizes. The notable exception to this was for the review of CBT (5 RCTs with over 400 participants). Nevertheless, this evidence was downgraded to moderate quality due to some concerns about risk of bias.
Other considerations	Although carers' assessments and NICE-recommended interventions should be readily accessible for all carers, the GDG noted from the review of carer experience that these options were often not available to carers of people with a learning disability and therefore considered that recommendations in this area were needed to improve carers' experience.

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5.4.22 Involving families and carers

Recommendations	6. Involve family members or carers in developing and delivering the support and intervention plan for the person with a learning disability and behaviour that challenges. Give them information about support and interventions in an appropriate language and format, including NICE's 'Information for the public'.
Relative values of different outcomes	The GDG agreed that the following were critical outcomes: severity, frequency and duration of the targeted behaviour that challenges, quality of life, family and carer stress and resilience, use of inpatient placements, and service user and carer satisfaction.
Trade-off between clinical benefits and harms	Due to the paucity of evidence, the GDG used a formal consensus approach to determining strategies to engage family and carers as a resource in the design, implementation and monitoring of interventions. These strategies were grouped in terms of universal prevention, selective prevention and indicated prevention/ intervention strategies. The consensus process clearly identified a number of strategies with strong support by the GDG. Assessment was seen as important across all levels of prevention/

	intervention. In addition, at the universal level, parent education/family behavioural supports were seen as important. At the selective level, stress management was seen as important and at the selective and indicated/ intervention level, formal social support was seen as important.
Trade-off between net health benefits and resource use	No economic evidence is available. The GDG expressed the view that implementation of strategies aimed at engaging the family and carers as a resource in the design, implementation and monitoring of interventions for the person with a learning disability and behaviour that challenges is likely to be cost-effective if it enhances improvement of outcomes for the person with a learning disability and behaviour that challenges, which, in turn, is expected to reduce associated costs which can be substantial, for example costs incurred by inpatient placements.
Quality of evidence	The review was not based on empirical evidence and therefore there was no quality assessment. The formal consensus process involved the use of the modified nominal group technique, which was chosen due to its suitability within the guideline development process. The method is concerned with deriving a group decision from a set of expert individuals and is commonly used for the development of consensus in health care.
Other considerations	N/A

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6 Organisation and delivery of care (including training)

6.1 Introduction

The overall organisation of services for people with behaviour that challenge has been briefly described in Chapter 2. This chapter is specifically concerned with 2 aspects of the organisation and delivery of care. The first concerns transition between settings (care, health and educational settings), which has been identified as a major problem by staff working in the field and in a number of recent reports (for example, (Sloper et al., 2010)). The second is concerned with the training of staff across a range of care settings, which, again, is a long-standing concern in the field and has been the subject of a number of recent reports (Department of Health, 2012)

6.1.1 Transition

Most people with a learning disability rely on others, including families, friends, formal and informal carers and a range of professionals to provide care throughout their lives, especially at times of substantial change. Some transitions, for example moving to a new school or to more independent living, can be a very positive experience but my nonetheless present a significant challenge. Where moves are not desired by the person, or are brought a sudden change in personal circumstances, for example a change in health status (of the person themselves or a carer), the challenge can be even greater. Transitions may occur in a planned way, as a result of the natural aging process (such as an individual moving from children's services into adult services), or may happen in a reactive, unplanned way (for example when an established placement breaks down and a new one is sought). Finding the right services and support for a person with a learning disability and behaviour that challenges can be a difficult process. Often a large number of assessments will be undertaken to inform the decision making as well as knowledge and views from both the person concerned and their immediate family. Opinions of those involved may differ, making the choice of services and support, and the development of a support plan, a delicate and complex process.

Whatever the reason for a transition across or between services, the challenge for commissioners and service providers is to manage the period of change in such a way as to minimise anxiety and uncertainty for those involved. Arguably a period of transition is one of the most testing times both for services and for the people who use those services. In addition to identifying the needs of the person, other important considerations include the allocation to, and use of, particular funding streams, availability and suitability of any given placement, the training and experience of staff members, the resources of carers and the continuity of care across the transition. Often what has sustained the person previously cannot be replicated, leading to a period of significant change, with all of the challenges commensurate with that.

Staff involved in transition, and care delivery in general, can make a significant contribution to the success of a given placement and help maintain an element of stability in a period of transition. The established skills, experience and training of staff and carers will have a great impact.

6.1.21 Training

There is growing evidence that when there is an understanding of the person with behaviour that challenges, the function of their behaviour and also how particular approaches and techniques may be applied, this correlates with better outcomes. In general such approaches relate to the development of whole service approaches that may then be personalised to the needs of the individual. Herein lies a problem, in that many approaches to behaviour that challenges to date have relied on what can be called 'reductionist' behavioural techniques, involving the teaching of specific methods designed to decrease the unwanted behaviours rather than understand their purpose. Fidelity is usually weak and the approach ineffective because it ignores critical information about the person or their circumstances.

However, the majority of staff (59%) involved in the care of people with a learning disability, have no formal professional training and this, along with the relatively high turnover in staff, represents a source of considerable concern in the provision of high quality services for people with a learning disability and behaviour that challenges as such people are often in receipt of support from staff in residential settings where levels of training may be lower than those of staff working in community teams and other specialist services (Bamford, 2007).

Training of staff is highly dependent on the circumstances of the individual service user's support setting. Some support organisations place great emphases on ensuring staff have regular and relevant accredited and professional training. However, at the other end of the spectrum some support services rely on 'on-the- job' staff coaching, often by individuals who themselves may have received little formal training.

Many families and carers report being left to acquire knowledge and information entirely unsupported and often learning lessons 'the hard way'. Learning 'the hard way' can mean unwittingly reinforcing behaviour that challenges, which can lead to inappropriate and costly interventions.

Past scandals involving the abuse of people who display behaviour that challenges invariably cite training as a key issue and recommend investment in it. This does not appear to be sustained in any meaningful way, at least so far as front line staff and carers are concerned. In the light of the enquiry into Winterbourne View Hospital, there is recognition of improving services through training both as a way of improving people's quality of life and reducing the risk that inexperienced or uninformed staff will accept abusive and dehumanising treatment as acceptable**.**

6.2 Review question: In people with a learning disability and behaviour that challenges, what are the effective models for transition between services?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 9. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 23: Clinical review protocol summary for the review of effective models for transition between services

Component	Description
Review question	In people with a learning disability and behaviour that challenges, what are the effective models for transition between services (for example child-adult, adult-older adult, NHS-social care/residential)? (RQ7.1)

Component	Description
	To answer this question, consideration should be given to: • The structure, design and delivery of care pathways • The nature and duration of support provided during transition.
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability and behaviour that challenges
Intervention(s)	All models aimed at effective transition between services
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Targeted behaviour that challenges • Quality of life • Rates of placement breakdown • Use of inpatient placements (including out-of-area placements) • Effects on carer stress and resilience • Service user and carer satisfaction
Study design	RCTs and systematic reviews
Note. RCTs = Randomised controlled trials	

6.2.1 Clinical evidence

No RCTs or systematic reviews met the eligibility criteria for this review. Further information about excluded studies can be found in Appendix Q.

The GDG noted the lack of high quality evidence in this area and the limitations of existing studies (see Appendix Q) which were almost entirely descriptive in nature and tended to be focused on transition from child and adolescent health, education or social care services to adult services. The relevance of this literature was further limited by the fact that much of the current descriptive data were concerned with children with a range of disabilities and was often not specifically concerned with learning disabilities or with behaviour that challenges. Even less relevant literature on adults was identified.

In the absence of high quality evidence the GDG considered whether to make any recommendations at all in this area. They drew on their expert knowledge in the area and the very considerable concerns that they had about the nature of transition between services (which they believed were shared by many professionals in the field). The GDG took the view that the current transitions were poorly planned, lacked proper oversight and often led to inappropriate and costly placements. The GDG took the view that recommendations elsewhere in this guideline, for example on assessment, could make a significant contribution to addressing these problems but that recommendations that set out the key principles which should underpin the proper organisation of transitions between and within services could have real value in improving the care and support of people with a learning disability and behaviour that challenges.

The GDG also noted that a similar problem had arisen in the development of another guideline: *Autism: Recognition, referral, diagnosis and management of adults on the autism spectrum* (NICE, 2012). The autism guideline was concerned with the development of care pathways for adults with autism, including but going beyond issues concerned with transition between services. In developing the recommendations in that area the GDG for the autism guideline had drawn on the evidence and recommendations in the *Common Mental Health Disorders* guideline (NICE, 2011). The GDG for this guideline on behaviour that challenges in people with a learning disability decided to adopt the same method (outlined in Chapter 3) but with a somewhat narrower focus (that is, on the development of recommendations which

would support more effective transition between services). In order to do this, the GDG first compiled a list of recommendations from the *Common Mental Health Disorders* guideline that could potentially be included in this current guideline – 23 in total (see Table 2). The underlying evidence is described fully in Chapter 7 of *Common Mental Health Disorders* (NCCMH, 2011). The GDG also considered the review of the evidence in Chapter 4 on the experience of care of people with a learning disability and their families and carers. The GDG then identified a number of recommendations (see 6.2.6) that they judged were important for the transition between services of people with a learning disability and behaviour that challenges. The GDG reviewed these recommendations and some minor adaptations to them to ensure that they were relevant to the current context. The detail of the adaptations and the rationale for them are given below in Table 25.

Table 24: Initial list of potential recommendations from the *Common Mental Health Disorders* guideline for inclusion

Recommendations
1. Primary and secondary care clinicians, managers and commissioners should collaborate to develop local care pathways that promote access to services for people with common mental health disorders by: • supporting the integrated delivery of services across primary and secondary care • having clear and explicit criteria for entry to the service • focusing on entry and not exclusion criteria • having multiple means (including self-referral) to access the service • providing multiple points of access that facilitate links with the wider healthcare system and community in which the service is located.
2. Provide information about the services and interventions that constitute the local care pathway, including the: • range and nature of the interventions provided • settings in which services are delivered • processes by which a person moves through the pathway • means by which progress and outcomes are assessed • delivery of care in related health and social care services.
3. When providing information about local care pathways to people with common mental health disorders and their families and carers, all healthcare professionals should: • take into account the person's knowledge and understanding of mental health disorders and their treatment • ensure that such information is appropriate to the communities using the pathway.
4. Provide all information about services in a range of languages and formats (visual, verbal and aural) and ensure that it is available from a range of settings throughout the whole community to which the service is responsible.
5. Primary and secondary care clinicians, managers and commissioners should collaborate to develop care pathways that promote access to services for people with common mental health disorders by: • supporting the integrated delivery of services across primary and secondary care • having clear and explicit criteria for entry to the service • focusing on entry and not exclusion criteria

Recommendations

- having multiple means (including self-referral) to access the service
- providing multiple points of access that facilitate links with the wider healthcare system and community in which the service is located.

6. Primary and secondary care clinicians, managers and commissioners should collaborate to develop local care pathways that promote access to services for people with common mental health disorders from a range of socially excluded groups including:

- black and minority ethnic groups
- older people
- those in prison or in contact with the criminal justice system
- ex-service personnel.

7. Support access to services and increase the uptake of interventions by:

- ensuring systems are in place to provide for the overall coordination and continuity of care of people with common mental health disorders
- designating a healthcare professional to oversee the whole period of care (usually a GP in primary care settings).

8. Support access to services and increase the uptake of interventions by providing services for people with common mental health disorders in a variety of settings. Use an assessment of local needs as a basis for the structure and distribution of services, which should typically include delivery of:

- assessment and interventions outside normal working hours
- interventions in the person's home or other residential settings
- specialist assessment and interventions in non-traditional community-based settings (for example, community centres and social centres) and where appropriate, in conjunction with staff from those settings
- both generalist and specialist assessment and intervention services in primary care settings.

9. Primary and secondary care clinicians, managers and commissioners should consider a range of support services to facilitate access and uptake of services. These may include providing:

- crèche facilities
- assistance with travel
- advocacy services.

10. When discussing treatment options with a person with a common mental health disorder, consider:

- their past experience of the disorder
- their experience of, and response to, previous treatment
- the trajectory of symptoms
- the diagnosis or problem specification, severity and duration of the problem
- the extent of any associated functional impairment arising from the disorder itself or any chronic physical health problem
- the presence of any social or personal factors that may have a role in the development or maintenance of the disorder

Recommendations

- the presence of any comorbid disorders.

- 11. When discussing treatment options with a person with a common mental health disorder, provide information about:
- the nature, content and duration of any proposed intervention
- the acceptability and tolerability of any proposed intervention
- possible interactions with any current interventions
- the implications for the continuing provision of any current interventions.

12. When making a referral for the treatment of a common mental health disorder, take account of patient preference when choosing from a range of evidence-based treatments.

13. When offering treatment for a common mental health disorder or making a referral, follow the stepped-care approach, usually offering or referring for the least intrusive, most effective intervention first.

14. Local care pathways should be developed to promote implementation of key principles of good care. Pathways should be:

- negotiable, workable and understandable for people with common mental health disorders, their families and carers, and professionals
- accessible and acceptable to all people in need of the services served by the pathway
- responsive to the needs of people with common mental health disorders and their families and carers
- integrated so that there are no barriers to movement between different levels of the pathway
- outcomes focused (including measures of quality, service-user experience and harm).

15. Responsibility for the development, management and evaluation of local care pathways should lie with a designated leadership team, which should include primary and secondary care clinicians, managers and commissioners. The leadership team should have particular responsibility for:

- developing clear policy and protocols for the operation of the pathway
- providing training and support on the operation of the pathway
- auditing and reviewing the performance of the pathway.

16. Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that promote a stepped-care model of service delivery that:

- provides the least intrusive, most effective intervention first
- has clear and explicit criteria for the thresholds determining access to and movement between the different levels of the pathway
- does not use single criteria such as symptom severity to determine movement between steps
- monitors progress and outcomes to ensure the most effective interventions are delivered and the person moves to a higher step if needed.

17. Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that promote a range of evidence-based interventions at each step in the pathway and support people with common mental health disorders in their choice of interventions.

Recommendations

18. All staff should ensure effective engagement with families and carers, where appropriate, to:

- inform and improve the care of the person with a common mental health disorder
- meet the identified needs of the families and carers.

19. Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that promote the active engagement of all populations served by the pathway. Pathways should:

- offer prompt assessments and interventions that are appropriately adapted to the cultural, gender, age and communication needs of people with common mental health disorders
- keep to a minimum the number of assessments needed to access interventions.

21. Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that provide an integrated programme of care across both primary and secondary care services. Pathways should:

- minimise the need for transition between different services or providers
- allow services to be built around the pathway and not the pathway around the services
- establish clear links (including access and entry points) to other care pathways (including those for physical healthcare needs)
- have designated staff who are responsible for the coordination of people's engagement with the pathway.

22. Primary and secondary care clinicians, managers and commissioners should work together to ensure effective communication about the functioning of the local care pathway. There should be protocols for:

- sharing and communicating information with people with common mental health disorders, and where appropriate families and carers, about their care
- sharing and communicating information about the care of services users with other professionals (including GPs)
- communicating information between the services provided within the pathway
- communicating information to services outside the pathway.

23. Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that have robust systems for outcome measurement in place, which should be used to inform all involved in a pathway about its effectiveness. This should include providing:

- individual routine outcome measurement systems
- effective electronic systems for the routine reporting and aggregation of outcome measures
- effective systems for the audit and review of the overall clinical and cost-effectiveness of the pathway.

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Table 25: Revised list of recommendations from the *Common Mental Health Disorders* guideline to be included

Recommendations
1. (14). Local care pathways should be developed to promote implementation of key principles of good care. Pathways should be: • negotiable, workable and understandable for people with common mental health disorders, their families and carers, and professionals • accessible and acceptable to all people in need of the services served by the pathway • responsive to the needs of people with common mental health disorders and their families and carers • integrated so that there are no barriers to movement between different levels of the pathway • outcomes focused (including measures of quality, service-user experience and harm).
2. (15). Responsibility for the development, management and evaluation of local care pathways should lie with a designated leadership team, which should include primary and secondary care clinicians, managers and commissioners. The leadership team should have particular responsibility for: • developing clear policy and protocols for the operation of the pathway • providing training and support on the operation of the pathway • auditing and reviewing the performance of the pathway.
3. (17). Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that promote a range of evidence-based interventions at each step in the pathway and support people with common mental health disorders in their choice of interventions.
4. (20). Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that respond promptly and effectively to the changing needs of all populations served by the pathways. Pathways should have in place: • clear and agreed goals for the services offered to a person with a common mental health disorder • robust and effective means for measuring and evaluating the outcomes associated with the agreed goals • clear and agreed mechanisms for responding promptly to identified changes to the person's needs.
5. (21). Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that provide an integrated programme of care across both primary and secondary care services. Pathways should: • minimise the need for transition between different services or providers • allow services to be built around the pathway and not the pathway around the services • establish clear links (including access and entry points) to other care pathways (including those for physical healthcare needs) • have designated staff who are responsible for the coordination of people's engagement with the pathway.
6. (22). Primary and secondary care clinicians, managers and commissioners should work together to ensure effective communication about the functioning of the local care pathway. There should be protocols for:

Recommendations
• sharing and communicating information with people with common mental health disorders, and where appropriate families and carers, about their care • sharing and communicating information about the care of services users with other professionals (including GPs) • communicating information between the services provided within the pathway • communicating information to services outside the pathway.

Table 26: Final list of recommendations from the *Common Mental Health Disorders* guideline after adaptation

Original recommendation from Common Mental Health Disorders	Review question and evidence base of existing recommendation	Recommendation following adaptation/incorporation for this guideline (numbering is from the NICE guideline recommendations)	Reasons for adaptation/incorporation
1.5.1.1 Local care pathways should be developed to promote implementation of key principles of good care. Pathways should be: negotiable, workable and understandable for people with common mental health disorders, their families and carers, and professionals accessible and acceptable to all people in need of the services served by the pathway responsive to the needs of people with common mental health disorders and their families and carers integrated so that there are no barriers to movement between different levels of the pathway outcomes focused (including measures of quality, service-user experience and harm).	Review question: In adults (18 years and older) with depression (including subthreshold disorders) or an anxiety disorder, what are the aspects of a clinical care pathway that are associated with better individual or organisational outcomes? Evidence base: 21 systematic reviews of clinical care pathways, the majority of which were of the treatment of depression. See Chapter 7 of Common Mental Health Disorders.	Develop care pathways for people with a learning disability and behaviour that challenges for the effective delivery of care and the transition between and within services that are: • negotiable, workable and understandable for people with a learning disability and behaviour that challenges, their family members or carers, and professionals •accessible and acceptable to people using the services, and responsive to their needs •integrated (to avoid barriers to movement between different levels of the care pathways) • focused on outcomes (including measures of quality, service-user experience and harm).	The GDG considered this recommendation relevant to the organisation of care of people with a learning disability and behaviour that challenges, including children and young people. Minor changes were made to the wording of the recommendations according to current NICE style for recommendations (direct instructions in plain English) and also to indicate the current context of the recommendation (the delivery of care and the transition between and within services for people with a learning disability and behaviour that challenges).
1.5.1.2 Responsibility for the development, management and evaluation of local care pathways should lie with a designated leadership team, which should include	Review question: In adults (18 years and older) with depression (including subthreshold	A designated leadership team of primary and secondary care professionals, managers and commissioners should be responsible for	The GDG considered this recommendation relevant to the organisation of care of people with a learning disability and behaviour that challenges

Original recommendation from Common Mental Health Disorders	Review question and evidence base of existing recommendation	Recommendation following adaptation/incorporation for this guideline (numbering is from the NICE guideline recommendations)	Reasons for adaptation/incorporation
primary and secondary care clinicians, managers and commissioners. The leadership team should have particular responsibility for: developing clear policy and protocols for the operation of the pathway providing training and support on the operation of the pathway auditing and reviewing the performance of the pathway.	disorders) or an anxiety disorder, what are the aspects of a clinical care pathway that are associated with better individual or organisational outcomes? Evidence base: 21 systematic reviews of clinical care pathways, the majority of which were of the treatment of depression. See Chapter 7 of Common Mental Health Disorders.	developing, managing and evaluating care pathways, including: • developing clear policies and protocols for care pathway operation • providing training and support on care pathway operation • auditing and reviewing care pathway performance.	including children and young people. Minor changes were made to the wording of the recommendations according to current NICE style for recommendations (direct instructions in plain English).
1.5.1.4 Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that promote a range of evidence-based interventions at each step in the pathway and support people with common mental health disorders in their choice of interventions.	Review question: In adults (18 years and older) with depression (including subthreshold disorders) or an anxiety disorder, what are the aspects of a clinical care pathway that are associated with better individual or organisational outcomes? Evidence base: 21 systematic reviews of clinical care pathways, the majority of which were of the treatment of depression. See	Primary and secondary care professionals, managers and commissioners should work together to design care pathways that promote a range of evidence-based interventions at each step and support people in their choice of interventions.	The GDG considered this recommendation relevant to the organisation of care of people with a learning disability and behaviour that challenges, including children and young people, and adapted it accordingly (removing ‘people with common mental health disorders’).

Original recommendation from Common Mental Health Disorders	Review question and evidence base of existing recommendation	Recommendation following adaptation/incorporation for this guideline (numbering is from the NICE guideline recommendations)	Reasons for adaptation/incorporation
	Chapter 7 of Common Mental Health Disorders.		
1.5.1.7 Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that respond promptly and effectively to the changing needs of all populations served by the pathways. Pathways should have in place: • clear and agreed goals for the services offered to a person with a common mental health disorder • robust and effective means for measuring and evaluating the outcomes associated with the agreed goals • clear and agreed mechanisms for responding promptly to identified changes to the person's needs.	Review question: In adults (18 years and older) with depression (including subthreshold disorders) or an anxiety disorder, what are the aspects of a clinical care pathway that are associated with better individual or organisational outcomes? Evidence base: 21 systematic reviews of clinical care pathways, the majority of which were of the treatment of depression. See Chapter 7 of Common Mental Health Disorders.	Primary and secondary care professionals, managers and commissioners should work together to design care pathways that respond promptly and effectively to the changing needs of the people they serve and have: • clear and agreed goals for the services offered • robust and effective ways to measure and evaluate the outcomes associated with the agreed goals • clear and agreed mechanisms for responding promptly to identified changes to the person's needs.	The GDG considered this recommendation relevant to the organisation of care of people with a learning disability and behaviour that challenges. including children and young people. Minor changes were made to the wording of the recommendations to indicate the current context of the recommendation (the delivery of care and the transition between and within services for people with a learning disability and behaviour that challenges). The last bullet point was omitted because it was covered sufficiently in the main body of the recommendation.
1.5.1.8 Primary and secondary care clinicians, managers and commissioners should work together to design local care pathways that provide an integrated programme of care across both primary and secondary care services. Pathways should: • minimise the need for transition between different services or providers • allow services to be built around the pathway and not the pathway around the services	Review question: In adults (18 years and older) with depression (including subthreshold disorders) or an anxiety disorder, what are the aspects of a clinical care pathway that are associated with better individual or organisational	Primary and secondary care professionals, managers and commissioners should work together to design care pathways that provide an integrated programme of care across both primary and secondary care services and: • minimise the need for transition between different services or providers • provide the least restrictive alternatives	The GDG considered this recommendation relevant to the organisation of care of people with a learning disability and behaviour that challenges, including children and young people. Minor changes were made to the wording of the recommendations according to current NICE style for recommendations (direct instructions in

Original recommendation from Common Mental Health Disorders	Review question and evidence base of existing recommendation	Recommendation following adaptation/incorporation for this guideline (numbering is from the NICE guideline recommendations)	Reasons for adaptation/incorporation
• establish clear links (including access and entry points) to other care pathways (including those for physical healthcare needs) • have designated staff who are responsible for the coordination of people's engagement with the pathway.	outcomes? Evidence base: 21 systematic reviews of clinical care pathways, the majority of which were of the treatment of depression. See Chapter 7 of Common Mental Health Disorders.	for people with behaviour that challenges • allow services to be built around the care pathway (and not the other way around) • establish clear links (including access and entry points) to other care pathways (including those for physical healthcare needs) • have designated staff who are responsible for coordinating people's engagement with a care pathway and transition between services within and between care pathways.	plain English) and also to indicate the current context of the recommendation (the delivery of care and the transition between and within services for people with a learning disability and behaviour that challenges). The GDG considered that a bullet point should be added to this recommendation about the use of restrictive practices in people with a learning disability and behaviour that challenges, given the concerns about over-use of restriction. In the final bullet point, the GDG added 'transition between services within and between care pathways, given their concerns about transitions for people with a learning disability and behaviour that challenges.
1.5.1.9 Primary and secondary care clinicians, managers and commissioners should work together to ensure effective communication about the functioning of the local care pathway. There should be protocols for: • sharing and communicating information with people with common mental health disorders, and where appropriate families and carers, about their care • sharing and communicating information about the care of services users with other	Review question: In adults (18 years and older) with depression (including subthreshold disorders) or an anxiety disorder, what are the aspects of a clinical care pathway that are associated with better individual or organisational outcomes? Evidence base:	Primary and secondary care professionals, managers and commissioners should work together to ensure effective communication about the functioning of care pathways. There should be protocols for sharing information: • with people with a learning disability and behaviour that challenges, and their family members or carers (if appropriate), about their care • about a person's care with other professionals	The GDG considered this recommendation relevant to the organisation of care of people with a learning disability and behaviour that challenges, including children and young people. Minor changes were made to the wording of the recommendations according to current NICE style for recommendations (direct instructions in plain English) and also to indicate the current context of the

Original recommendation from Common Mental Health Disorders	Review question and evidence base of existing recommendation	Recommendation following adaptation/incorporation for this guideline (numbering is from the NICE guideline recommendations)	Reasons for adaptation/incorporation
professionals (including GPs) • communicating information between the services provided within the pathway • communicating information to services outside the pathway.	21 systematic reviews of clinical care pathways, the majority of which were of the treatment of depression. See Chapter 7 of Common Mental Health Disorders.	(including GPs) • with all the services provided in the care pathway • with services outside the care pathway.	recommendation (the delivery of care and the transition between and within services for people with a learning disability and behaviour that challenges).

6.2.2 Clinical summary of evidence

The GDG drew from 2 evidence sources in developing the recommendations in this section: the *Common Mental Health Disorders* guideline (because this guideline developed a set or principles for the development of care pathways in the field of mental health) and the review of the evidence in Chapter 4 on experience of care. The GDG considered these 2 evidence sources and identified a number of recommendations that in their view were important in improving transitions for people with a learning disability and behaviour that challenges. The GDG then adapted the recommendations based on the method outlined in Chapter 3.

6.2.3 Economic evidence

No studies assessing the cost effectiveness of models for transition between services for people with a learning disability and behaviour that challenges were identified by the systematic search of the economic literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3. Nevertheless, 2 UK studies were identified that provided information on costs associated with transition to adult services for young people with a learning disability and behaviour that challenges (Barron et al., 2013) and for young people with disabilities and complex health needs (Sloper et al., 2010). Although these studies do not meet inclusion criteria for this review as none of them assess the cost effectiveness of models of transition, they do offer an insight into the types of costs associated with the period of transition of young people with a learning disability and behaviour that challenges to adult services and thus are briefly described in this section.

Barron and colleagues (2013) conducted a survey of all young people aged 16-18 years with a learning disability and behaviour that challenges that were in transition to adult services between 2006 and 2008 in one London borough. The survey identified 59 young people that were suitable for adult community learning disability services, of which 36 were identified as having behaviour that challenges; 27 of them agreed to take part in the study. At the time of the interview, the participants' mean Challenging Behaviour Checklist (CBC) score was 16.8 (sd 11.1; range 0-36); 3 individuals scored zero and 15 had a CBC score ≥ 17. Eighteen individuals showed 2 or more types of behaviour that challenges. The types of behaviour that were recorded included self-injury, harm to others and destruction to property. The cost elements measured in the survey included day time activities (day centre, social club, drop-in centre, adult education), education (special needs & mainstream day school, residential

school), hospital-based services (inpatient, outpatient, accident and emergency), community-based services (for example, GP, psychiatrist, psychologist, community psychiatric nurse, social worker, speech & language therapist, occupational therapist, art therapist, home care), police and informal care. The mean weekly cost per young person in transition was estimated at £2,543 (2009 prices), attributed mainly to informal care (65% of total cost) and education (22% of total cost). The authors reported that individuals' access to services showed wide variation in terms of number and type of services used, with lack of access to community specialist nursing and employment services being notable. Individuals with higher levels of behaviour that challenges (as measured by the CBC score) or more complex needs (indicated total number of coexisting mental and physical health diagnoses) were not found to be in receipt of higher cost care packages; the only clinical parameter linked to the cost of care was the level of learning disability.

Sloper and colleagues (2010) conducted a national survey of multi-agency co-ordinated transition services for disabled young people and their families. The aim of the study was to investigate arrangements across local authority areas in England for multiagency assessment for, planning of and actual transfer from child to adult services for young people with disabilities or complex health needs, compare the implementation and operation of different models of transition services, assess outcomes for parents and young people, and also investigate sources of funding and costs of different models of transition services. Of the 34 transition services participating in the survey, 16 provided sufficient data on whole-time equivalent composition of their teams, their professions and employing organisations that allowed estimation of staffing costs (i.e. salary costs of transition workers and managers). Based on this information, the mean annual cost per young person supported by a transition team was estimated at £1,483 (2007/8 prices), ranging from £490 (at a service supporting 220 people) to £3,190 (at a service supporting 34 people). These figures do not include costs of clerical and administrative support, office-related costs, travel costs, client-related service costs, building costs and overheads.

In addition, a detailed study on 5 multi-agency co-ordinated transition services for disabled young people and their families was undertaken, focusing on young people in special schools with a severe learning disability. The 5 services encompassed different models of working and had key differences in terms of co-ordinating services and transition teams. The mean annual cost per person supported ranged from £395 (at a service covering 2 urban centres and surrounding villages and supporting 72 people at the time of the study) to £3,545 (at a service covering an outer London borough and supporting 76 people at the time of the study). Costs were driven by the professional mix in the transition team and the costs of employing those professionals.

The study also reported service costs for young people who were in the process of transition planning but had not yet transferred to adult services (pre-transition sample, N=105), and those who had transferred within the last 2 years and had received the transition service (post-transition sample, N=23). The 3-month service cost per person pre- and post-transition was £6,259 and £5,047, respectively; residential services (including both education and accommodation) accounted for 84% of this cost, with remaining costs incurred by hospital and community health services (10%) and other social care services (6%).

6.2.4 Clinical evidence statements

No clinical evidence was identified for this review.

6.2.5 Economic evidence statements

There is evidence that young people with a learning disability and behaviour that challenges in transition to adult services incur considerable costs associated mainly with informal care and residential service use, and in a lesser degree with health and other social service use. There is wide variation in the cost of transition services per supported person across the UK,

which is driven by the professional mix in the transition team and the co-ordination of services. However, there is no evidence on the cost effectiveness of different models of transition for people with a learning disability and behaviour that challenges.

6.2.6 Recommendations and link to evidence

See section 6.4 for recommendations and link to evidence relating to this section.

6.3 Review question: What are the benefits and potential harms of training and education programmes to allow health and social care professionals and carers to provide good-quality services and carry out evidence based interventions designed to reduce or manage behaviour that challenges in people with a learning disability?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 27. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 27: Clinical review protocol summary for the review of training and education programmes

Component	Description
Review question	What are the benefits and potential harms of training and education programmes to allow health and social care professionals and carers to provide good-quality services and carry out evidence based interventions designed to reduce or manage behaviour that challenges in people with a learning disability? (RQ6.1)
Population	Health and social care professionals, and carers of children, young people or adults with a mild, moderate, severe or profound learning disability and behaviour that challenges. The term 'carers' encompasses both family carers and paid carers.
Intervention(s)	Training and education programs to allow health and social care professionals and carers provide good-quality services and carry out evidence based interventions targeted at the reduction or management of behaviour that challenges.
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Targeted behaviour that challenges • Effects on carer stress and resilience • Quality of life • Fidelity • Service user and carer satisfaction
Study design	RCTs and systematic reviews
Note. RCTs = Randomised controlled trials	

6.3.1 Clinical evidence

No RCTs met the eligibility criteria for this review. The GDG therefore selected an existing systematic review of non-randomised studies as the basis for this section of the guideline: Macdonald 2013 (MacDonald & McGill, 2013). The systematic review included 14 studies:

Baker 1998 (Baker, 1998), Browning-Wright 2007 (Browning-Wright et al., 2007), Crates 2012 (Crates & Spicer, 2012), Dench 2005 (Dench, 2005), Freeman 2005 (Freeman et al., 2005), Gore 2011 (Gore & Umizawa, 2011), Grey 2007 (Grey & McClean, 2007), Kraemer 2008 (Kraemer et al., 2008), Lowe 2007 (Lowe et al., 2007b), McClean 2005 (McClean et al., 2005), McClean 2012 (McClean & Grey, 2012), McGill 2007 (McGill et al., 2007), Reid 2003 (Reid et al., 2003), Reynolds 2011 (Reynolds et al., 2011). Although the systematic review allowed for any type of study design, all included studies were repeated measures. A summary of the included review can be found in Table 28.

All included studies were published in peer-reviewed journals between 1998 and 2012 and specifically involved training in Positive Behaviour Support. Of the 14 included studies, 4 were from Ireland, 5 from the USA, 3 from the UK, 1 from Canada, and 1 from Australia.

Six of the included studies focused on staff outcomes, 4 focused on service user outcomes and 4 focused on both staff and service user outcomes. Studies that focused only on outcomes for families and carers were excluded, although some studies that focused on staff and family/carer outcomes, as well as the other outcomes of interest, were included.

Further information about both included and excluded studies can be found in Macdonald 2013.

As a result of considerable differences between the studies, including the length of training and outcome measures used, no meta-analysis was possible. A narrative synthesis of the evidence was, therefore, applied.

Table 28: Study information table for the systematic review included in the review of training and education programmes

	Macdonald 2013
Review question/ Aim	To evaluate the research on the outcomes of Positive Behaviour Support training in relation to either children or adults with a learning disability and behaviour that challenges.
Method used to synthesise evidence	Narrative Synthesis
Design of included studies	Repeated measures
Dates searched	1990 to 2012
Electronic databases	1) Google Scholar; 2) Web of Science; 3) Pub Med; 4) PsycINFO
No. of included studies (N[1])	14 (1,466)
Participant characteristics	Children, young people and adults with a learning disability, and/or the staff that provide their support. Excluded studies relating to families and carers only.
Intervention	Positive behavioural support staff training
Comparison	N/A
Outcome	• Staff outcomes (including changes in skills, confidence, knowledge, attributions and emotional responses) • Service user outcomes (including frequency, severity and management of behaviour that challenges and quality of life)
Review Quality	Poor[3]

[1]Number of participants.

[2]The included studies randomised 57 participants; however 7 participants were excluded from the review as they did not have SIB.

[3]The design of included studies was deemed inappropriate for the guideline review and the quality of them was not assessed or reported.

6.3.12 Economic evidence

No studies assessing the cost effectiveness of training and education programmes for health and social care professionals and carers of people with a learning disability and behaviour that challenges were identified by the systematic search of the economic literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

6.3.37 Clinical evidence statements

6.3.3.18 Service user outcomes

- In 1 poor quality systematic review of 14 studies, there was evidence from 8 of these studies that training staff in positive behavioural support may reduce behaviour that challenges, but it was unclear whether this also improves quality of life.

6.3.3.22 Staff outcomes

- In 1 poor quality systematic review of 14 studies, there was evidence from 7 of these studies that training staff in positive behavioural support may improve staff skills.

6.3.45 Economic evidence statements

There is no evidence on the cost effectiveness of training and education programmes for health and social care professionals and carers of people with a learning disability and behaviour that challenges.

6.49 Recommendations and link to evidence

6.4.10 Delivering effective care

Recommendations	
	7. Develop care pathways for people with a learning disability and behaviour that challenges for the effective delivery of care and the transition between and within services that are: • negotiable, workable and understandable for people with a learning disability and behaviour that challenges, their family members or carers, and staff • accessible and acceptable to people using the services, and responsive to their needs • integrated (to avoid barriers to movement between different levels of the care pathways) • focused on outcomes (including measures of quality, service-user experience and harm). 8. A designated leadership team of primary and secondary care professionals, managers and commissioners should be responsible for developing, managing and evaluating care pathways, including: • developing clear policies and protocols for care pathway operation • providing training and support on care pathway operation

	• auditing and reviewing care pathway performance. 9. Primary and secondary care professionals, managers and commissioners should work together to design care pathways that promote a range of evidence-based interventions at each step and support people in their choice of interventions. 10. Primary and secondary care professionals, managers and commissioners should work together to design care pathways that respond promptly and effectively to the changing needs of the people they serve and have: • clear and agreed goals for the services offered • robust and effective ways to measure and evaluate the outcomes associated with the agreed goals. 11. Primary and secondary care professionals, managers and commissioners should work together to design care pathways that provide an integrated programme of care across both primary and secondary care services and: • minimise the need for transition between different services or providers • provide the least restrictive alternatives for people with behaviour that challenges • allow services to be built around the care pathway (and not the other way around) • establish clear links (including access and entry points) to other care pathways (including those for physical healthcare needs) • have designated staff who are responsible for coordinating people's engagement with a care pathway and transition between services within and between care pathways. 12. Primary and secondary care professionals, managers and commissioners should work together to ensure effective communication about the functioning of care pathways. There should be protocols for sharing information: • with people with a learning disability and behaviour that challenges, and their family members or carers (if appropriate), about their care • about a person's care with other professionals (including GPs) • with all the services provided in the care pathway • with services outside the care pathway.
Relative values of different outcomes	There was a clear view from the GDG that many services failed to achieve successful transitions for people with a learning disability and behaviour that challenges, with poor outcomes a clear consequence of this. Reduction in

	behaviour that challenges, quality of life and service user and carer satisfaction were agreed to be critical outcomes.
Trade-off between clinical benefits and harms	The current situation is unsatisfactory with poor coordination of care and poor resource allocation. Formalising pathways through care should improve this situation but the absence of empirical evidence means that there is a risk this will not be the case.
Trade-off between net health benefits and resource use	Young people with a learning disability and behaviour that challenges in transition to adult services incur considerable costs associated mainly with informal care and residential service use, and in a lesser degree with health and other social service use. Currently, there is wide variation in costs of transition services across the UK. The GDG expressed the opinion that formalising care pathways for people with a learning disability and behaviour that challenges, including transition between and within services, would enable more effective delivery of care and better outcomes for service users, reducing, at the same time, the high variation in care costs resulting from provision of ineffective and poorly coordinated care.
Quality of evidence	The very limited evidence available was of low quality.
Other considerations	In the absence of high quality evidence in this area, the GDG drew on a review of the recommendations on care pathways in the *Common Mental Health Disorders* guideline and the review of experience of care (Chapter 4 of the current guideline). The GDG judged that adapting recommendations from *Common Mental Health Disorders* would add value to the overall guideline in order to improve transitions for people with a learning disability and behaviour that challenges. Adaptations to the wording of the recommendations from *Common Mental Health Disorders* were considered necessary in order to reflect the different organisational context in which services for learning disabilities are provided.

6.4.21 Understanding learning disabilities and behaviour that challenges

Recommendations	13. Everyone involved in delivering support and interventions for people with a learning disability and behaviour that challenges (including family members and carers) should understand: • the nature, development and course of learning disabilities • individual and environmental factors related to the development and maintenance of behaviour that challenges • that behaviour that challenges is communicating an unmet need • the effect of learning disabilities and behaviour that challenges on the person's personal, social, educational and occupational functioning • the effect of the social and physical environment on learning disabilities and behaviour that challenges (and vice versa), including how staff and carer responses to the behaviour may maintain it.

6.4.2.11 Team working

Recommendations	
	14. Health and social care provider organisations should ensure that the assessment and management of behaviour that challenges in people with a learning disability are undertaken by teams that have skills and competencies in routine assessment and intervention methods. 15. If initial assessment (see section 8.5) and management have not been effective, or the person has more complex needs, health and social care provider organisations should ensure that teams providing routine assessment and interventions have access to: • specialist assessment • specialist support and intervention services • advice, supervision and training to support the implementation of any care or intervention. Specialist support and intervention services should include nurses, psychologists, psychiatrists, social workers, and speech and language therapists. Occupational therapists, physiotherapists, physicians**,** paediatricians and pharmacists may also be involved.

6.4.2.22 Staff training and supervision

Recommendations	
	16. All staff working with people with a learning disability and behaviour that challenges should be trained to deliver proactive strategies to reduce the risk of behaviour that challenges, including: • developing personalised daily activities • adapting a person's environment and routine • developing strategies to help the person develop an alternative behaviour to achieve the same purpose by developing a new skill (for example, improved communication, emotional regulation or social interaction) • the importance of including people, and their family members or carers, in planning support and interventions • strategies designed to calm and divert the person if they show early signs of distress. Training should also include delivering reactive strategies to manage behaviour that is not preventable. 17. All interventions for people with learning disabilities and behaviour that challenges should be delivered by competent staff. Staff should: • receive regular high-quality supervision that takes into account the impact of individual, social and environmental factors

	• deliver interventions based on the relevant manuals • use routine sessional outcome measures (for example, the Adaptive Behaviour Scale and the Aberrant Behaviour Checklist) • take part in monitoring and evaluating adherence to interventions and practitioner competence (for example, by using Periodic Service Review methods, video and audio recording, and external audit and scrutiny).

6.4.2.31 Link to evidence across all topics

Relative values of different outcomes	The GDG agreed that the following outcomes were critical to decision making: targeted behaviour that challenges, effects on carer stress and resilience, quality of life, fidelity and service user and carer satisfaction.
Trade-off between clinical benefits and harms	The evidence suggested that training staff may have benefits in terms of reduced behaviour that challenges and improved fidelity of treatment through improved staff skills. There was insufficient or no evidence to determine the impact on quality of life, satisfaction or carer stress and resilience.
Trade-off between net health benefits and resource use	Training health and social care professionals who care for people with a learning disability and behaviour that challenges is likely to incur considerable costs. Nevertheless, the GDG considered that the benefits from effective programmes may potentially outweigh costs, if these programmes lead to a reduction in, or more effective management of, behaviour that challenges in this population.
Quality of evidence	The evidence came from a poor quality systematic review that had not appraised the quality of the individual studies.
Other considerations	The GDG also drew on its expert knowledge in developing the recommendations in this section and in doing sought to emphasise the following; (a) that all staff working in the area should have a full understanding of learning disabilities and people's needs, (b) that interventions should always be provided in a team whose knowledge and expertise might need to be supplemented by external experts, (c) that training should emphasise positive proactive approaches to care as well as reactive approaches and that this should be central to any training, and (d) training will only be effective if it is supported by proper supervision and audit of outcomes.

6.4.3.2 Research recommendations

1. **Does providing care where people live compared with out-of-area placement lead to improvements in both the clinical and cost effectiveness of care for people with a learning disability and behaviour that challenges?**

2. **What factors (including service management, staff composition, training and supervision, and the content of care and support) are associated with sustained high-quality residential care for people with a learning disability and behaviour that challenges?**

7 Identification of behaviour that challenges

7.1 Introduction

The appearance of behaviour that challenges in people with a learning disability is not usually a random event. It has been thought for some time that some people are more at risk of developing behaviour that challenges than others (McClintock et al., 2003) (see Section 2.4); possible risk factors include the degree of disability, gender, presence of certain comorbid conditions (such as autism and epilepsy), levels of communication skills, and sensory and other impairments.

The knowledge that some of these factors are associated with a greater risk of behaviour that challenges provides 2 kinds of opportunities. First, the influence of a particular factor on the emergence of behaviour that challenges should inform theories about why the behaviour has appeared and what is maintaining it. At the very least such theories need to be able to account for the factors that turn out to be of influence in the appearance of behaviour that challenges. Second, and more importantly in many ways, this knowledge should be seen as an opportunity for early interventions to be put in place, given the presence of relevant characteristics, to reduce the likelihood of behaviour that challenges arising or persisting.

In services currently, such knowledge is rarely utilised. In general, services are reactive rather than proactive in intervening with behaviour that challenges, even in circumstances where such behaviour is highly likely to appear. At the very least such interventions could include psychoeducation for carers, regular monitoring and early interventions if and when the behaviour first begins to appear. The improved knowledge provided by the evidence reviewed below gives services an opportunity to use that knowledge in providing improved and more proactive support for people with a learning disability and behaviour that challenges, and their families and carers.

7.2 Review question: In people with a learning disability, what are the circumstances, risk factors and antecedents associated with the development of behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 29. A complete list of review questions and full review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 29: Clinical review protocol summary for the review of circumstances, risk factors and antecedents associated with the development of behaviour that challenges

Component	Description
Review question	In people with a learning disability, what are the circumstances, risk factors and antecedents associated with the development of behaviour that challenges? (RQ1.1)
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability
Intervention(s)	Circumstances, risk factors and antecedents for behaviour that challenges: • Circumstance = A factor or condition connected with or relevant to an event or action • Risk factor = a variable associated with an increased risk of

Component	Description
	disease/disorder • Antecedent = anything that precedes another thing, especially the cause of the second thing.
Comparison	Not applicable
Critical outcomes	Risk of behaviour that challenges (event or odds ratio for risk of behaviour that challenges)
Study design	Any

7.2.1 Clinical evidence

The GDG selected an existing systematic review (McClintock et al., 2003) as the basis for this section of the guideline, with a new search conducted to update the existing review. The existing review identified 86 potentially relevant studies. Of these, 20 studies provided sufficient data to be included in the evidence synthesis: Ando 1979 (Ando & Yoshimura, 1979a; Ando & Yoshimura, 1979b), Ballinger 1971 (Ballinger, 1971), Berkson 1985 (Berkson et al., 1985), Bhaumick 1997 (Bhaumik et al., 1997), Bott 1997 (Bott et al., 1997), Davidson 1994 (Davidson et al., 1994), Eyman 1977 (Eyman & Call, 1977), Griffin 1986 (Griffin et al., 1986), Hardan 1997 (Hardan & Sahl, 1997), Jacobson 1982 (Jacobson, 1982), Kebbon 1986 (Kebbon & Windahl, 1986), Kieman 1996 (Kieman & Alborz, 1996), Maisto 1978 (Maisto et al., 1978), Maurice 1982 (Maurice & Trudel, 1982), McLean 1996 (McLean et al., 1996), Quine 1986 (Quine, 1986), Rojahn 1986 (Rojahn, 1986) , Ross 1972 (Ross, 1972), Schroeder 1978 (Schroeder et al., 1978), Shodell 1968 (Shodell & Reiter, 1968).

An additional 52 potentially relevant studies were identified by the update search conducted for the guideline, of which 12 provided sufficient data to be included in the evidence synthesis: Baghdadli 2003 (Baghdadli et al., 2003), Bradley 2004 (Bradley et al., 2004), Cooper 2009 (Cooper et al., 2009a), Crocker 2006 (Crocker et al., 2006), Crocker 2013 (Crocker et al., 2013), Hill 2006 (Hill & Furniss, 2006), Holden 2006 (Holden & Gitlesen, 2006), Lundqvist 2013 (Lundqvist, 2013), Myrbakk 2008 (Myrbakk & Von Tetzcnner, 2008), Richards 2012 (Richards et al., 2012), Tenneij 2009 (Tenneij et al., 2009b), Tyrer 2006 (Tyrer et al., 2006). Ando 1979 reported findings for different risk factors among the same group of participants across 2 separate papers, which will be referred to herein as Ando 1979a and Ando 1979b.

In total, 138 observational studies therefore met the eligibility criteria for this review. Of these, 32 (N = 127,298) reported sufficient data to be included in the evidence synthesis. All were published in peer-reviewed journals between 1968 and 2013. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

7.2.1.1 Autism diagnosis

Seven studies examined a comorbid diagnosis of autism as a potential risk factor for behaviour that challenges in people with a learning disability (N = 7,662): Ando 1979 (Ando & Yoshimura, 1979a), Bhaumick 1997 (Bhaumik et al., 1997), Bradley 2004 (Bradley et al., 2004), Cooper 2009 (Cooper et al., 2009a), Davidson 1994 (Davidson et al., 1994), Lundqvist 2013 (Lundqvist, 2013), Tyrer 2006 (Tyrer et al., 2006). Of the included studies, 2 focused on combined physical, verbal and destructive aggression (Cooper 2009, Lundqvist 2013), 2 on destruction of property (Ando & Yoshimura 1979a, Bhaumick 1997), 4 on physical aggression (Ando & Yoshimura 1979a, Bhaumick 1997, Davidson 1994, Tyrer 2006) and 5 on self-injury (Ando & Yoshimura 1979a, Bhaumick 1997, Bradley 2004, Cooper 2009, Lundqvist 2013). An overview of the trials included in the meta-analysis can be found in Table 30. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

Subgroup analysis was carried out to compare the effect of a comorbid autism diagnosis on behaviour that challenges across different settings (mixed and educational) and different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 31. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 30: Study information table for trials included in the meta-analysis of autism as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Destruction of property	Physical aggression	Self-injury
Total no. of studies (N)	2 (1,938)	2 (2,436)	4 (5,700)	5 (4,398)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013	(1) Ando 1979a (2) Bhaumick 1997	(1) Ando 1979a (2) Bhaumick 1997 (3) Davidson 1994 (4) Tyrer 2006	(1) Ando 1979a (2) Bhaumick 1997 (3) Bradley 2004 (4) Cooper 2009 (5) Lundqvist 2013
Country	(1) UK (2) Sweden	(1) Japan (2) UK	(1) Japan (2, 4) UK (3) USA	(1) Japan (2, 4) UK (3) Canada (5) Sweden
Diagnosis	(1, 2) LD	(1) Autism + LD (2) LD	(1) Autism + LD (2, 4) LD (3) DD	(1) Autism + LD (2 - 5) LD
Population	(1, 2) Adults	(1) C & YP (2) Adults	(1) C & YP (2, 4) Adults (3) Mixed	(1, 3) C & YP (2, 4, 5) Adults
Setting	(1, 2) Mixed	(1) Education (2) Mixed	(1) Education (2 to 4) Mixed	(1) Education (2 to 5) Mixed
Age (mean)	(1, 2) 43	(1, 2) Not reported	Not reported (3) 28	(1, 2) Not reported (3) 16 (4, 5) 43
Sex (% Female)	(1, 2) 45	(1) 35 (2) Not reported	35-43 (2) Not reported	33-45 (2) Not reported
IQ (mean)	(1, 2) Not reported	(1) 43 (2) Not reported	(1, 3) 43-44 (2, 4) Not reported	(1) 43 (2 to 5) Not reported

Notes: N = total number of participants; LD = learning disability; DD = developmental disabilities; C & YP = children and young people

Table 31: Summary of findings table for the review of autism as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks*	Relative	No of	Quality of the

	(95% CI)		effect (95% CI)	Participants (studies)	evidence (GRADE)
	Assumed risk **No autism diagnosis**	Corresponding risk **Autism diagnosis**			
All aggression (physical, verbal and destructive) Validated questionnaires, interviews and medical records	**196 per 1000**	**300 per 1000** (222 to 393)	**OR 1.76** (1.17 to 2.65)	1938 (2 studies)	**very low**[1]
Destruction of property Questionnaire and interviews with both service user and carer	**94 per 1000**	**368 per 1000** (126 to 701)	**OR 5.6** (1.39 to 22.56)	2376 (2 studies)	**very low**[2, 3]
Physical aggression Validated questionnaires, interviews and medical records	**159 per 1000**	**446 per 1000** (316 to 634)	**RR 2.80** (1.98 to 3.98)	5637 (4 studies)	**moderate**[3]
Self-injury Validated questionnaires and interviews with both service user and carer	**138 per 1000**	**332 per 1000** (225 to 461)	**OR 3.11** (1.81 to 5.35)	4338 (5 studies)	**very low**[2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio; **OR:** Odds ratio;

[1] I2 > 40%
[2] I2 > 75%
[3] RR >2

7.2.1.21 Gender

Seventeen studies examined gender as a potential risk factor for behaviour that challenges in people with a learning disability (N = 43,281): Ballinger 1971 (Ballinger, 1971), Cooper 2009 (Cooper et al., 2009a), Crocker 2006 (Crocker et al., 2006), Crocker 2013 (Crocker et al., 2013), Davidson 1994 (Davidson et al., 1994), Griffin 1986 (Griffin et al., 1986), Holden 2006 (Holden & Gitlesen, 2006), Lundqvist 2013 (Lundqvist, 2013), Maisto 1978 (Maisto et al., 1978), Maurice 1982 (Maurice & Trudel, 1982), Quine 1986 (Quine, 1986), Myrbakk 2008 (Myrbakk & Von Tetzcnner, 2008), Richards 2012 (Richards et al., 2012), Rojahn 1986 (Rojahn, 1986), Schroeder 1978 (Schroeder et al., 1978), Tenneij 2009 (Tenneij et al., 2009b), Tyrer 2006 (Tyrer et al., 2006). Of the included studies, 3 focused on all aggression (physical, verbal and destructive) (Cooper 2009, Lundqvist 2013, Tenneij 2009), 2 on destruction of property (Crocker 2006, Crocker 2013), 5 on physical aggression (Crocker 2006, Crocker 2013, Davidson 1994, Quine 1986, Tyrer 2006) and 2 on verbal aggression (Crocker 2006, Crocker 2013). Eleven of the 17 included studies focused on self-injury (Ballinger 1971, Cooper 2009, Crocker 2006, Griffin 1986, Lundqvist 2013, Maisto 1978, Maurice 1982, Quine 1986, Richards 2012, Rojahn 1986, Schroeder 1978), 1 each focused on inappropriate sexual behaviour (Crocker 2006) and stereotypy (Lundqvist 2013) and 2 focused on global behaviour that challenges (Holden 2006, Myrbakk 2008). An overview of the trials included in the meta-analysis can be found in Table 32 and Table 33. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

One study concerned a mixed population of adults with a learning disability and psychotic disorders (Maurice 1982). Because less than 50% of the combined population was diagnosed with a learning disability, a sensitivity analysis excluding this study was conducted to explore the robustness of the findings. In the sensitivity analysis, all effects remained consistent with the main analysis.

Subgroup analysis was carried out to compare the effect of a comorbid autism diagnosis on behaviour that challenges across different settings (mixed and inpatient) and different

populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 34. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 32: Study information table for trials included in the meta-analysis of gender as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Destruction of property	Physical aggression	Verbal aggression
Total no. of studies (N)	3 (2,046)	2 (3,461)	5 (6,925)	2 (3,461)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013 (3) Tenneij 2009	(1) Crocker 2006 (2) Crocker 2013	(1) Crocker 2006 (2) Crocker 2013 (3) Davidson 1994 (4) Quine 1986 (5) Tyrer 2006	(1) Crocker 2006 (2) Crocker 2013
Country	(1) UK (2) Sweden (3) Netherlands	(1, 2) Canada	(1, 2) Canada (3) USA (4, 5) UK	(1, 2) Canada
Diagnosis	(1, 2) LD (3) Mild LD	(1) LD (2) Moderate LD	(1, 5) LD (2) Moderate LD (3) DD (4) Severe LD	(1) LD (2) Moderate LD
Population	(1 to 3) Adults	(1, 2) Adults	(1, 2, 5) Adults (3) Mixed (4) C & YP	(1, 2) Adults
Setting	(1, 2) Mixed (3) Inpatient	(1, 2) Mixed	(1 to 5) Mixed	(1, 2) Mixed
Age (mean)	(1, 2) 43 (3) 26	(1, 2) 41	(1, 2) 41 (3) 28 (4, 5) Not reported	(1, 2) 41
Sex (% Female)	(1, 2) 45 (3) 24	(1) 48 (2) 45	37-48	(1) 48 (2) 45
IQ (mean)	(1, 2) Not reported (3) 66	(1, 2) Not reported	(1, 2, 4, 5) Not reported (3) 44	(1, 2) Not reported

Note. N = total number of participants; LD = learning disability; DD = developmental disabilities; C & YP = children and young people

Table 33: Study information table for trials included in the meta-analysis of gender as a risk factor for behaviour that challenges in people with a learning disability

	Inappropriate sexual behaviour	Self-injury	Stereotypy	Behaviour that challenges (global)

Total no. of studies (N)	1 (3,165)	11 (38,569)	1 (222)	2 (1044)
Study ID	Crocker 2006	(1) Ballinger 1971 (2) Cooper 2009 (3) Crocker 2006 (4) Griffin 1986 (5) Lundqvist 2013 (6) Maisto 1978 (7) Maurice 1982 (8) Quine 1986 (9) Richards 2012 (10) Rojahn 1986 (11) Schroeder 1978	Lundqvist 2013	(1) Holden 2006 (2) Myrbakk 2008
Country	Canada	(1, 2, 8, 9) UK (3, 7) Canada (4, 6, 11) USA (5) Sweden (10) Germany	Sweden	(1, 2) Norway
Diagnosis	LD	(1 to 6, 10, 11) LD (7) Mixed[1] (8) Severe LD (9) Autism	LD	(1, 2) LD
Population	Adults	(1 to 3, 5, 7) Adults (4, 6, 9 to 11) Mixed (8) C & YP	Adults	(1, 2) Mixed
Setting	Mixed	(1, 4, 6, 7, 11) Inpatient (2, 3, 5, 8 to 10) Mixed	Mixed	(1, 2) Mixed
Age (mean)	41	(1, 8, 10, 11) Not reported (2) 30-46 (3) 10	43	(1) Not reported (2) 40
Sex (% Female)	48	37-55 (9) 11	45	(1) 45 (2) 48
IQ (mean)	Not reported	(1 to 11) Not reported	Not reported	(1, 2) Not reported

Notes: N = total number of participants; LD = learning disability; C & YP = children and young people
[1] Participants diagnosed as having learning disability (43.7%) or psychotic or related diagnoses (48.5%); study excluded in sensitivity analysis.

Table 34: Summary of findings table for the review of gender as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk	Corresponding risk			
	Female gender	**Male gender**			

All aggression (physical, verbal and destructive) Validated questionnaire and observation	**264 per 1000**	**184 per 1000** (155 to 221)	**OR 0.63** (0.51 to 0.79)	2046 (3 studies)	**low**
Behaviour that challenges (global) Validated survey	**92 per 1000**	**126 per 1000** (83 to 184)	**OR 1.42** (0.9 to 2.23)	816 (1 study)	**very low**[1]
Destruction of property Validated questionnaire Follow-up: 0 to 12 months	See comment[2]	See comment[2]	Not estimable	3461 (2 studies)	**low**
Inappropriate sexual behaviour Questionnaire Follow-up: mean 12 months	**76 per 1000**	**119 per 1000** (96 to 147)	**OR 1.64** (1.29 to 2.09)	3160 (1 study)	**very low**[1]
Physical aggression Validated questionnaires, interviews, observations and medical records Follow-up: 0 to 12 months	See comment[2]	See comment[2]	Not estimable	6925 (5 studies)	**very low**[3]
Self-injury - mixed settings Questionnaire and survey Follow-up: 0 to 12 months	**293 per 1000**	**252 per 1000** (223 to 285)	**OR 0.81** (0.69 to 0.96)	6174 (6 studies)	**low**
Self-injury- inpatient setting Non-validated questionnaire, survey and interview Follow-up: 0 to 3 years	**122 per 1000**	**119 per 1000** (96 to 146)	**OR 0.97** (0.76 to 1.23)	18227 (5 studies)	**very low**[4]
Stereotypy Validated questionnaire	**411 per 1000**	**415 per 1000** (354 to 485)	**RR 1.01** (0.86 to 1.18)	915 (1 study)	**very low**[1]
Verbal aggression Validated questionnaire Follow-up: 0 to 12 months	See comment[2]	See comment[2]	Not estimable	3461 (2 studies)	Not estimable

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio; **OR:** Odds ratio;

[1] Optimal information size not met; single study
[2] N/A; Generic inverse variance
[3] I2 > 40%
[4] I2 > 75%

7.2.1.31 Severity of learning disability

Seventeen studies examined severity of learning disability as a potential risk factor for behaviour that challenges in people with a learning disability (N = 115,647): Ballinger 1971 (Ballinger, 1971), Berkson 1985 (Berkson et al., 1985), Cooper 2009 (Cooper et al., 2009a), Crocker 2006 (Crocker et al., 2006), Davidson 1994 (Davidson et al., 1994), Eyman 1977 (Eyman & Call, 1977), Hardan 1997 (Hardan & Sahl, 1997), Holden 2006 (Holden & Gitlesen, 2006), Jacobson 1982 (Jacobson, 1982) , Kebbon 1986 (Kebbon & Windahl, 1986), Lundqvist 2013 (Lundqvist, 2013), Maisto 1978 (Maisto et al., 1978), Myrbakk 2008 (Myrbakk & Von Tetzcnner, 2008), Rojahn 1986 (Rojahn, 1986), Ross 1972 (Ross, 1972), Schroeder 1978 (Schroeder et al., 1978), Tyrer 2006 (Tyrer et al., 2006). Of the included studies, 2 focused on all aggression (physical, verbal and destructive) (Cooper 2009, Lundqvist 2013), 1 focused on destruction of property (Crocker 2006), 7 focused on physical aggression (Crocker 2006, Davidson 1994, Eyman & Call 1977, Hardan & Sahl 1997, Jacobson 1982, Ross 1972, Tyrer 2006) and 1 focused on verbal aggression. Twelve of the 17 included studies focused on self-injury (Ballinger 1971, Cooper 2009, Crocker 2006, Eyman 1977, Hardan 1997, Jacobson 1982, Kebbon 1986, Lundqvist 2013, Maisto 1978, Rojahn 1986, Ross 1972, Schroeder 1978), 6 on stereotypy (Berkson 1985, Eyman 1977, Holden 2006, Jacobson 1982, Lundqvist 2013, Myrbakk 2008), 2 on global behaviour that challenges (Holden 2006, Myrbakk 2008) and a single study focused on inappropriate sexual behaviour (Crocker 2006).

An overview of the trials included in the meta-analysis can be found in Table 35 and

Table **36**. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

Subgroup analysis was carried out to compare the effect of severity of learning disability on behaviour that challenges across different settings (mixed and inpatient) and different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 37. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 35: Study information table for trials included in the meta-analysis of severity of learning disability as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Destruction of property	Physical aggression	Verbal aggression
Total no. of studies (N)	2 (1,938)	1 (3,165)	7 (55,249)	1 (3,165)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013	Crocker 2006	(1) Crocker 2006 (2) Davidson 1994 (3) Eyman 1977 (4) Hardan 1997 (5) Jacobson 1982 (6) Ross 1972 (7) Tyrer 2006	Crocker 2006
Country	(1) UK (2) Sweden	Canada	(1) Canada (2 to 6) USA (7) UK	Canada
Diagnosis	(1, 2) LD	LD	(1, 3 to 7) LD (2) DD	LD
Population	(1, 2) Adults	Adults	(1, 7) Adults (2, 3, 5, 6) Mixed (4) C & YP	Adults
Setting	(1, 2) Mixed	Mixed	(1 to 5) Mixed (6) Inpatient (7) Mixed	Mixed
Age (mean)	(1, 2) 43	41	(1) 41 (2, 6) 23-28 (3, 4, 7) Not reported (4) 9	41
Sex (% Female)	(1, 2) 45	48	(1 to 7) 41-48 (4) 28	48
IQ (mean)	(1, 2) Not reported	Not reported	(1, 3 to 7) Not reported (2) 44	Not reported

Notes: N = total number of participants; LD = learning disability; DD = developmental disabilities; C & YP = children and young people				

Table 36: Study information table for trials included in the meta-analysis of severity of learning disability as a risk factor for behaviour that challenges in people with a learning disability

	Inappropriate sexual behaviour	Self-injury	Stereotypy	Behaviour that challenges (global)
Total no. of studies (N)	1 (3,165)	12 (111,086)	6 (39,660)	2 (1,044)
Study ID	Crocker 2006	(1) Ballinger 1971 (2) Cooper 2009 (3) Crocker 2006 (4) Eyman 1977 (5) Hardan 1997 (6) Jacobson 1982 (7) Kebbon 1986 (8) Lundqvist 2013 (9) Maisto 1978 (10) Rojahn 1986 (11) Ross 1972 (12) Schroeder 1978	(1) Berkson 1985 (2) Eyman 1977 (3) Holden 2006 (4) Jacobson 1982 (5) Lundqvist 2013 (6) Myrbakk 2008	(1) Holden 2006 (2) Myrbakk 2008
Country	Canada	(1, 2) UK (3) Canada (4 to 6, 9, 11, 12) USA (7, 8) Sweden (10) Germany	(1, 2, 4) USA (3, 6) Norway (5) Sweden	(1, 2) Norway
Diagnosis	LD	(1 to 12) LD	(1 to 6) LD	(1, 2) LD
Population	Adults	(1 to 3, 8) Adults (4, 6, 7, 9 to 12) Mixed (5) C & YP	(1) C & YP (2 to 4, 6) Mixed (5) Adults	(1, 2) Mixed
Setting	Mixed	(1) Inpatient (2 to 8, 10) Mixed (9, 11, 12) Inpatient	(1 to 6) Mixed	(1, 2) Mixed
Age (mean)	41	(1, 4, 6, 7, 10, 12) Not reported (2, 3, 8) 41-43 (5) 9 (9) 34 (11) 23	(1 to 4) Not reported (5) 43 (6) 40	(1) Not reported (2) 40
Sex (% Female)	48	(42-55 (5) 28	44-48 (1)Not reported	(1) 45 (2) 48
IQ (mean)	Not reported	(1 to 12) Not reported	(1 to 6) Not	(1, 2) Not reported

			reported	

Notes: N = total number of participants; LD = learning disability; C & YP = children and young people

Table 37: Summary of findings table for the review of the severity of learning disability as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Mild/ Moderate LD**	Corresponding risk **Severe/ Profound LD**			
All aggression (physical, verbal and destructive) Validated questionnaires	**215 per 1000**	**317 per 1000** (181 to 494)	**OR 1.70** (0.81 to 3.57)	1918 (2 studies)	**very low**[1]
Behaviour that challenges (global) Survey	**66 per 1000**	**234 per 1000** (163 to 323)	**OR 4.31** (2.75 to 6.74)	822 (1 study)	**low**[2,3]
Destruction of property Validated questionnaire Follow-up: 12 months	**229 per 1000**	**260 per 1000** (229 to 295)	**OR 1.18** (1 to 1.41)	3160 (1 study)	**very low**[2]
Inappropriate sexual behaviour Validated questionnaire Follow-up: 12 months	**97 per 1000**	**99 per 1000** (80 to 125)	**OR 1.02** (0.8 to 1.32)	3160 (1 study)	**very low**[2]
Physical aggression - inpatient setting Survey	**294 per 1000**	**218 per 1000** (200 to 236)	**OR 0.67** (0.6 to 0.74)	11139 (1 study)	**very low**[2,4]
Physical aggression - mixed setting Validated questionnaires, interviews, observations and medical records	**136 per 1000**	**217 per 1000** (181 to 257)	**OR 1.76** (1.4 to 2.2)	43864 (6 studies)	**very low**[1]
Self-injury Validated questionnaires, surveys and medical records Follow-up: 0 to 36 months	**53 per 1000**	**172 per 1000** (127 to 230)	**OR 3.75** (2.62 to 5.38)	85888 (12 studies)	**very low**[1,3]
Stereotypy Validated questionnaires and surveys	**65 per 1000**	**306 per 1000** (89 to 664)	**OR 6.38** (1.42 to 28.65)	23946 (4 studies)	**very low**[1,3]
Verbal aggression Validated questionnaire	**414 per 1000**	**294 per 1000** (261 to 328)	**OR 0.59** (0.5 to 0.69)	3160 (1 study)	**very low**[2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] I2 > 75%
[2] Optimal information size not met; single study
[3] RR > 2
[4] Partial applicability to review population- high risk inpatient

Notes LD = Learning disability

7.2.1.44 Epilepsy diagnosis

Three studies examined a comorbid diagnosis of epilepsy as a potential risk factor for behaviour that challenges in people with a learning disability (N = 2,160): Baghdadli 2003 (Baghdadli et al., 2003), Cooper 2009 (Cooper et al., 2009a), Lundqvist 2013 (Lundqvist, 2013). Of the included studies, all focused on self-injury, 2 focused on combined physical, verbal and destructive aggression (Cooper 2009, Lundqvist 2013) and 1 on stereotypy (Lundqvist 2013). An overview of the trials included in the meta-analysis can be found in Table 38. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

Subgroup analysis was carried out to compare the effect of a comorbid epilepsy diagnosis on behaviour that challenges across different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 39. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 38: Study information table for trials included in the meta-analysis of epilepsy as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Self-injury	Stereotypy
Total no. of studies (N)	2 (1,938)	3 (2,160)	1 (915)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013	(1) Baghdadli 2003 (2) Cooper 3009 (3) Lundqvist 2013	Lundqvist 2013
Country	(1) UK (2) Sweden	(1) France (2) UK (3) Sweden	Sweden
Diagnosis	(1, 2) LD	(1) Autism + LD (2, 3) LD	LD
Population	(1, 2) Adults	(1) C & YP (2, 3) Adults	Adults
Setting	(1, 2) Mixed	(1 to 3) Mixed	Mixed
Age (mean)	(1, 2) 43	(1) 5 (2, 3) 43	43
Sex (% Female)	(1, 2) 45	(1) 21 (2, 3) 45	45
IQ (mean)	(1, 2) Not reported	(1 to 3) Not reported	Not reported
Notes: N = total number of participants; LD = learning disability; C & YP = children and young people			

Table 39: Summary of findings table for the review of epilepsy as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk No diagnosis of epilepsy	Corresponding risk Diagnosis of epilepsy	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
All aggression (physical, verbal and destructive) Validated questionnaire	**224 per 1000**	**271 per 1000** (218 to 331)	**OR 1.29** (0.97 to 1.72)	1927 (2 studies)	**low**
Self-injury- adults Validated questionnaire	**172 per 1000**	**302 per 1000** (239 to 373)	**OR 2.08** (1.51 to 2.86)	1927 (2 studies)	**low**
Self-injury- children and young people Questionnaire	**536 per 1000**	**429 per 1000** (203 to 692)	**OR 0.65** (0.22 to 1.94)	206 (1 study)	**very low**[1, 2]
Stereotypy Validated questionnaire	**399 per 1000**	**499 per 1000** (407 to 594)	**OR 1.5** (1.03 to 2.2)	915 (1 study)	**very low**[2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the

intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] Unclear if outcome assessment was validated
[2] Optimal information size not met; Single study

7.2.1.51 Mental health needs

2 Four studies examined the presence of mental health needs as a potential risk factor for
3 behaviour that challenges in people with a learning disability (N = 32,812): Jacobson 1982
4 (Jacobson, 1982), Cooper 2009 (Cooper et al., 2009a), Crocker 2013 (Crocker et al., 2013),
5 Lundqvist 2013 (Lundqvist, 2013). Of the included studies, 2 focused on combined physical,
6 verbal and destructive aggression (Cooper 2009, Lundqvist 2013), 2 on physical aggression,
7 verbal aggression and destruction of property (Crocker 2013, Jacobson 1982), 2 on
8 stereotypy (Lundqvist 2013, Jacobson 1982) and 3 on self-injury (Cooper 2009, Lundqvist
9 2013, Jacobson 1982). An overview of the trials included in the meta-analysis can be found
10 in Table 40 and Table 41. Further information about both included and excluded studies can
11 be found in Appendix L and Appendix Q.

12 Subgroup analysis was carried out to compare the effect of an expressive communication
13 deficit on behaviour that challenges across different populations (children and young people
14 and adults). The results for each subgroup will only be reported if findings between groups
15 were conflicting.

16 Summary of findings can be found in Table 42. The full GRADE evidence profiles and
17 associated forest plots can be found in Appendix O and Appendix P.

18 The methodology checklists can be found in Appendix J, study evidence tables in Appendix
19 L, and exclusion list in Appendix Q.

20 **Table 40: Study information table for trials included in the meta-analysis of mental**
21 **health needs as a risk factor for behaviour that challenges in people with a learning**
22 **disability**

	All aggression (physical, verbal, destructive)	Destruction of property	Physical aggression	Verbal aggression
Total no. of studies (N)	2 (1,938)	2 (33,743)	2 (33,743)	2 (33,743)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013	(1) Crocker 2006 (2) Jacobson 1982	(1) Crocker 2006 (2) Jacobson 1982	(1) Crocker 2006 (2) Jacobson 1982
Country	(1) UK (2) Sweden	(1) Canada (2) USA	(1) Canada (2) USA	(1) Canada (2) USA
Diagnosis	(1, 2) LD	(1, 2) LD	(1, 2) LD	(1, 2) LD
Population	(1, 2) Adults	(1) Adults (2) Mixed	(1) Adults (2) Mixed	(1) Adults (2) Mixed
Setting	(1, 2) Mixed	(1, 2) Mixed	(1, 2) Mixed	(1, 2) Mixed
Age (mean)	(1, 2) 43	(1) 41 (2) Not reported	(1) 41 (2) Not reported	(1) 41 (2) Not reported
Sex (% Female)	(1, 2) 45	(1) 48 (2) 44	(1) 48 (2) 44	(1) 48 (2) 44
IQ (mean)	(1, 2) Not reported	(1, 2) Not reported	(1, 2) Not reported	(1, 2) Not

				reported
Notes: N = total number of participants; LD = learning disability.				

Table 41: Study information table for trials included in the meta-analysis of mental health needs as a risk factor for behaviour that challenges in people with a learning disability

	Self-injury	Stereotypy
Total no. of studies (N)	3 (32,516)	2 (31,493)
Study ID	(1) Cooper 2009 (2) Jacobson 1982 (3) Lundqvist 2013	(1) Jacobson 1982 (2) Lundqvist 2013
Country	(1) UK (2) USA (3) Sweden	(1) USA (2) Sweden
Diagnosis	(1 to 3) LD	(1, 2) LD
Population	Adults (2) Mixed	(1) Mixed (2) Adults
Setting	(1 to 3) Mixed	(1, 2) Mixed
Age (mean)	43 (2) Not reported	(1) Not reported (2) 43
Sex (% Female)	44-45	(1) 44 (2) 45
IQ (mean)	(1 to 3) Not reported	(1, 2) Not reported
Notes: N = total number of participants; LD = learning disability.		

Table 42: Summary of findings table for the review of mental health needs as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk **No mental health needs**	 Corresponding risk **Mental health needs**	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
All aggression (physical, verbal and destructive) Validated questionnaire	**205 per 1000**	**344 per 1000** (251 to 449)	**OR 2.03** (1.3 to 3.15)	1938 (2 studies)	**low**
Destruction of property Validated questionnaire and survey	See comment[1]	See comment[1]	Not estimable	30874 (2 studies)	**very low**[2]
Physical aggression Validated questionnaire and survey	See comment[1]	See comment[1]	Not estimable	30874 (2 studies)	**very low**[2]
Self-injury Validated questionnaires and survey	**93 per 1000**	**126 per 1000** (115 to 138)	**OR 1.4** (1.26 to 1.56)	32516 (3 studies)	**low**
Stereotypy Validated questionnaire and survey	**71 per 1000**	**87 per 1000** (77 to 98)	**OR 1.26** (1.1 to 1.43)	31493 (2 studies)	**low**
Verbal aggression Validated questionnaire and survey	See comment[1]	See comment[1]	Not estimable	30874 (2 studies)	⊕⊕⊕⊖ **moderate**[3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] N/A; Generic inverse variance
[2] I2 > 75%
[3] RR > 2

7.2.1.6 Expressive communication

Nine studies examined the presence of an expressive communication deficit as a potential risk factor for behaviour that challenges in people with a learning disability (N = 7,565): Ando 1979 (Ando & Yoshimura, 1979b), Baghdadli 2003 (Baghdadli et al., 2003), Bott 1997 (Bott et al., 1997), Cooper 2009 (Cooper et al., 2009a), Lundqvist 2013 (Lundqvist, 2013), McLean 1996 (McLean et al., 1996), Richards 2012 (Richards et al., 2012), Schroeder 1978 (Schroeder et al., 1978), Shodell 1968 (Shodell & Reiter, 1968). Of the included studies, all focused on self-injury, 2 focused on combined physical, verbal and destructive aggression (Cooper 2009, Lundqvist 2013), 2 on physical aggression (Bott 1997, McLean 1996) and 1 on stereotypy (Lundqvist 2013). An overview of the trials included in the meta-analysis can be found in Table 43. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

One study concerned a mixed population of verbal and non-verbal children with schizophrenia (Shodell 1968). Because it could not be verified whether the sample also had a diagnosis of learning disability, a sensitivity analysis excluding this study was conducted to explore the robustness of the findings. In the sensitivity analysis, all effects remained consistent with the main analysis.

Subgroup analysis was carried out to compare the effect of an expressive communication deficit on behaviour that challenges across different settings (mixed, education and inpatient) and different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 44. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 43: Study information table for trials included in the meta-analysis of expressive communication deficit as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Physical aggression	Self-injury	Stereotypy
Total no. of studies (N)	2 (1,938)	2 (3,873)	9 (7,565)	1 (915)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013	(1) Bott 1997 (2) McLean 1996	(1) Ando 1979b (2) Baghdadli 2003 (3) Bott 1997 (4) Cooper 2009 (5) Lundqvist 2013 (6) McLean 1996 (7) Richards 2012 (8) Schroeder 1978 (9) Shodell 1968	Lundqvist 2013
Country	(1) UK (2) Sweden	(1) UK (2) USA	(1) Japan (2) France	Sweden

			(3 to 4, 7) UK (5) Sweden (6, 8-9) USA	
Diagnosis	(1, 2) LD	(1) LD (2) Severe LD	(1) Autism + LD (2 to 5, 8) LD (6) Severe LD (7) Autism (9) LD + Schizophrenia[1]	LD
Population	(1, 2) Adults	(1) Adults (2) Mixed	(1, 2, 9) C & YP (3 to 5) Adults (6 to 8) Mixed	Adults
Setting	(1, 2) Mixed	(1, 2) Mixed	(1, 9) Education (2 to 7) Mixed (8) Inpatient	Mixed
Age (mean)	(1, 2) 43	(1, 2) Not reported	(1, 3, 6, 8, 9) Not reported (2) 5 (4 to 5) 43 (7) 10	43
Sex (% Female)	(1, 2) 45	(1) Not reported (2) 34	(1, 4, 6, 8) 34-55 (2) 21 (3, 9) Not reported (7) 11	45
IQ (mean)	(1, 2) Not reported	(1, 2) Not reported	(1) 43 (2 to 9) Not reported	Not reported
Notes: N = total number of participants; LD = learning disability; C & YP = children and young people [1] Not a verified LD sample; study removed in sensitivity analysis				

Table 44: Summary of findings table for the review of expressive communication deficit as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **No deficit**	Corresponding risk **Expressive communication deficit**			
All aggression (physical, verbal and destructive) Validated questionnaire	**229 per 1000**	**295 per 1000** (243 to 356)	**OR 1.41** (1.08 to 1.86)	1936 (2 studies)	**low**
Physical aggression- adult population Questionnaire	**262 per 1000**	**375 per 1000** (333 to 416)	**OR 1.69** (1.41 to 2.01)	3662 (1 study)	**very low**[1,2]
Physical aggression- mixed population Non-validated questionnaire	**313 per 1000**	**44 per 1000** (9 to 167)	**OR 0.10** (0.02 to 0.44)	211 (1 study)	**low**[2,3,4]
Self-injury Questionnaires, interviews and formal assessments	**146 per 1000**	**333 per 1000** (235 to 449)	**OR 2.93** (1.8 to 4.78)	7502 (9 studies)	**very low**[5,6]

Follow-up: 0 to 3 years					
Stereotypy Validated questionnaire	**377 per 1000**	**603 per 1000** (513 to 685)	**OR 2.51** (1.74 to 3.6)	915 (1 study)	**very low**[2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] Non validated checklist for risk and outcome assessment
[2] Optimal information size not met; single study
[3] Questionnaire for risk and outcome assessment was not validated
[4] RR < 0.2
[5] I^2 > 75%
[6] RR > 2

7.2.1.71 Receptive communication

Three studies examined the presence of a receptive communication deficit as a potential risk factor for behaviour that challenges in people with a learning disability (N = 1,359): Ando 1979 (Ando & Yoshimura, 1979b), Kieman 1996 (Kieman & Alborz, 1996), Schroeder 1978 (Schroeder et al., 1978). All of the included studies focused on self-injury. An overview of the trials included in the meta-analysis can be found in Table 45. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

Subgroup analysis was carried out to compare the effect of an expressive communication deficit on behaviour that challenges across different settings (education, inpatient and mixed) and different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 46. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 45: Study information table for trials included in the meta-analysis of receptive communication deficit as a risk factor for behaviour that challenges in people with a learning disability

	Self-injury
Total no. of studies (N)	3 (1,359)
Study ID	(1) Ando 1979b (2) Kieman 1996 (3) Schroeder 1978
Country	(1) Japan (2) UK (3) USA
Diagnosis	(1) Autism + LD (2, 3) LD
Population	(1) C & YP (2, 3) Adults
Setting	(1) Education (2) Community (3) Inpatient
Age (mean)	Not reported
Sex (% Female)	35-55
IQ (mean)	(1) 43

	(2, 3) Not reported
Notes: N = total number of participants; LD = learning disability; C & YP = children and young people	

Table 46: Summary of findings table for the review of expressive communication deficit as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk	Corresponding risk	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	No deficit	**Receptive communication deficit**			
Self-injury Questionnaire and interview Follow-up: 0 to 3 years	**135 per 1000**	**350 per 1000** (280 to 427)	**OR 3.46** (2.5 to 4.79)	1321 (3 studies)	⊕⊕⊕⊖ **moderate**[1]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] RR > 2

7.2.1.85 Hearing impairment

Three studies examined the presence of an hearing impairment as a potential risk factor for behaviour that challenges in people with a learning disability (N = 2,087): Cooper 2009 (Cooper et al., 2009a), Lundqvist 2013 (Lundqvist, 2013), Richards 2012 (Richards et al., 2012). Of the included studies, all focused on self-injury, 2 focused on combined physical, verbal and destructive aggression (Cooper 2009, Lundqvist 2013) and 1 on stereotypy (Lundqvist 2013). An overview of the trials included in the meta-analysis can be found in Table 47. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

Subgroup analysis was carried out to compare the effect of an auditory impairment on behaviour that challenges across different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 48. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 47: Study information table for trials included in the meta-analysis of auditory impairment as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Self-injury	Stereotypy
Total no. of studies (N)	2 (1,938)	3 (2,087)	1 (915)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013	(1) Cooper 3009 (2) Lundqvist 2013 (3) Richards 2012	Lundqvist 2013
Country	(1) UK	(1, 3) UK	Sweden

	(2) Sweden	(2) Sweden	
Diagnosis	(1, 2) LD	(1, 2) LD (3) Autism	LD
Population	(1, 2) Adults	(1, 2) Adults (3) Mixed	Adults
Setting	(1, 2) Mixed	(1 to 3) Mixed	Mixed
Age (mean)	(1, 2) 43	(1, 2) 43 (3) 10	43
Sex (% Female)	(1, 2) 45	(1, 2) 45 (3) 11	45
IQ (mean)	(1, 2) Not reported	(1 to 3) Not reported	Not reported
Notes: N = total number of participants; LD = learning disability; C & YP = children and young people			

Table 48: Summary of findings table for the review of auditory impairment as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **No impairment**	Corresponding risk **Auditory impairment**			
All aggression (physical, verbal and destructive) Validated questionnaire	**233 per 1000**	**228 per 1000** (113 to 404)	**OR 0.97** (0.42 to 2.23)	1938 (2 studies)	**very low**[1]
Self-injury Validated questionnaire	**237 per 1000**	**246 per 1000** (132 to 415)	**OR 1.05** (0.49 to 2.29)	2086 (3 studies)	**very low**[1]
Stereotypy Validated questionnaire	**411 per 1000**	**470 per 1000** (309 to 638)	**OR 1.27** (0.64 to 2.53)	915 (1 study)	**very low**[2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] $I^2 > 40\%$
[2] Optimal information size not met; single study

7.2.1.93 Mobility impairment

Two studies examined the presence of a mobility impairment as a potential risk factor for behaviour that challenges in people with a learning disability (N = 1,172): Cooper 2009 (Cooper et al., 2009a), Richards 2012 (Richards et al., 2012). Of the included studies, all focused on self-injury and 1 focused on combined physical, verbal and destructive aggression (Cooper 2009). An overview of the trials included in the meta-analysis can be found in Table 49. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

Subgroup analysis was carried out to compare the effect of mobility impairment on behaviour that challenges across different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 50. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 49: Study information table for trials included in the meta-analysis of mobility impairment as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Self-injury
Total no. of studies (N)	1 (1,023)	2 (1,172)
Study ID	Cooper 2009	(1) Cooper 2009 (2) Richards 2012
Country	UK	UK
Diagnosis	LD	(1) LD (2) Autism
Population	Adults	(1) Adults (2) Mixed
Setting	Mixed	Mixed
Age (mean)	43	(1) 43 (2) 10
Sex (% Female)	45	(1) 45 (2) 11
IQ (mean)	Not reported	Not reported
Notes: N = total number of participants; LD = learning disability.		

Table 50: Summary of findings table for the review of mobility impairment as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk No impairment	 Corresponding risk Mobility impairment	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
All aggression (physical, verbal and destructive) Validated questionnaire	**101 per 1000**	**89 per 1000** (56 to 138)	**OR 0.87** (0.53 to 1.43)	1023 (1 study)	**very low**[1]
Self-injury- adult population Validated questionnaire	**101 per 1000**	**89 per 1000** (56 to 138)	**OR 0.87** (0.53 to 1.43)	1023 (1 study)	**very low**[1]
Self-injury- children and young people population Validated questionnaire	**478 per 1000**	**692 per 1000** (397 to 885)	**OR 2.46** (0.72 to 8.38)	147 (1 study)	**very low**[1]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] Optimal information size not met; single study

7.2.1.106 Visual impairment

Three studies examined the presence of a visual impairment as a potential risk factor for behaviour that challenges in people with a learning disability (N = 2,087): Cooper 2009 (Cooper et al., 2009a), Lundqvist 2013 (Lundqvist, 2013), Richards 2012 (Richards et al., 2012). Of the included studies, all focused on self-injury, 2 focused on combined physical, verbal and destructive aggression (Cooper 2009, Lundqvist 2013) and 1 on stereotypy (Lundqvist 2013). An overview of the trials included in the meta-analysis can be found in Table 51. Further information about both included and excluded studies can be found in Appendix L and Appendix Q.

Subgroup analysis was carried out to compare the effect of a visual impairment on behaviour that challenges across different populations (children and young people and adults). The results for each subgroup will only be reported if findings between groups were conflicting.

Summary of findings can be found in Table 52. The full GRADE evidence profiles and associated forest plots can be found in Appendix O and Appendix P.

The methodology checklists can be found in Appendix J, study evidence tables in Appendix L, and exclusion list in Appendix Q.

Table 51: Study information table for trials included in the meta-analysis of visual impairment as a risk factor for behaviour that challenges in people with a learning disability

	All aggression (physical, verbal, destructive)	Self-injury	Stereotypy
Total no. of studies (N)	2 (1,938)	3 (2,087)	1 (915)
Study ID	(1) Cooper 2009 (2) Lundqvist 2013	(1) Cooper 3009 (2) Lundqvist 2013 (3) Richards 2012	Lundqvist 2013
Country	(1) UK (2) Sweden	UK (2) Sweden	Sweden
Diagnosis	LD	LD (3) Autism	LD
Population	Adults	Adults (3) Mixed	Adults
Setting	Mixed	(1 to 3) Mixed	Mixed
Age (mean)	43	43 (3) 10	43
Sex (% Female)	45	45 (3) 11	45
IQ (mean)	Not reported	Not reported	Not reported
Notes: C & YP = children and young people; LD = learning disability; N = total number of participants.			

Table 52: Summary of findings table for the review of visual impairment as a risk factor for behaviour that challenges in people with a learning disability

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)	Comments
	Assumed risk **No impairment**	Corresponding risk **Visual impairment**				
All aggression (physical, verbal and destructive) Validated questionnaire	**245 per 1000**	**284 per 1000** (202 to 384)	**OR 1.22** (0.78 to 1.92)	1938 (2 studies)	**low**	
Self-injury Validated questionnaire	**246 per 1000**	**321 per 1000** (249 to 401)	**OR 1.45** (1.02 to 2.06)	2086 (3 studies)	**low**	
Stereotypy Validated questionnaire	**405 per 1000**	**628 per 1000** (457 to 773)	**OR 2.49** (1.24 to 5.01)	915 (1 study)	**very low**[1]	

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **OR:** Odds ratio;

[1] Optimal information size; single study

7.2.2 Health economic evidence

Identification of circumstances, risk factors and antecedents associated with the development of behaviour that challenges in people with a learning disability may lead to better prediction (and thus more timely management) and possibly prevention of incidents of behaviour that challenges and has therefore potentially important resource implications. However, this review question is not relevant for economic analysis.

7.2.3 Clinical evidence statements

7.2.3.1 Autism diagnosis

- Very low quality evidence from up to 5 studies (N = 4,338) suggested that a comorbid diagnosis of autism was associated with increased risk of all aggression, destruction of property and self-injury.
- Moderate quality evidence from 4 studies (N = 5,637) suggested that a comorbid diagnosis of autism was associated with increased risk of physical aggression.

7.2.3.2 Gender

- Low quality evidence from 3 studies (N = 2,046) suggested that male gender was associated with reduced risk of combined physical, verbal and destructive aggression (in mixed or inpatient settings).
- Very low quality evidence from a single study (N = 816) suggested that male gender was associated with increased risk of global behaviour that challenges (in mixed settings). However, precision of the estimate is poor.
- Very low quality evidence from up to 2 studies (N = 3,461) suggested that male gender was associated with increased risk of property destruction, inappropriate sexual behaviour and physical aggression (in mixed settings).
- Low quality evidence from 6 studies (N = 6,174) suggested that male gender was associated with reduced risk of self-injury in mixed settings. However, evidence was inconclusive for inpatient settings (k = 5; N = 18,227).
- Very low quality evidence from a single study (N = 915) was inconclusive as to whether male gender was associated with the increased risk of verbal aggression or stereotypy (in a mixed setting).

7.2.3.3 Severity of learning disability

- Very low quality evidence from 2 studies (N = 1,918) suggested that severe/ profound learning disability was associated with increased risk of combined physical, verbal and destructive aggression although the precision of the estimate was poor.
- Low quality evidence from a single study (N = 822) suggested that severe/ profound learning disability was associated with increased risk of global behaviour that challenges and destruction of property.
- Very low quality evidence from a single study (N = 3,160) was inconclusive as to whether severe/ profound learning disability was associated with the increased risk of inappropriate sexual behaviour.
- Very low quality evidence from a single study (N = 11,139) suggested that severe/ profound learning disability was associated with reduced risk of physical aggression in an inpatient setting. However, very low quality evidence from 6 studies (N = 43, 864) suggested that in a mixed setting, severe/ profound learning disability was associated with increased risk of physical aggression.
- Very low quality evidence from up to 12 studies (N = 85,888) suggested that severe/ profound learning disability was associated with increased risk of self-injury and stereotypy.

- Very low quality evidence from a single study (N = 3,160) suggested that severe/ profound learning disability was associated with reduced risk of verbal aggression.

7.2.3.4 Epilepsy diagnosis

- Low quality evidence from up to 2 studies (N = 1,927) suggested that a comorbid diagnosis of epilepsy was associated with increased risk of all aggression and stereotypy.
- Very low quality evidence from up to 2 studies (N = 1,927) suggested that a comorbid diagnosis of epilepsy was associated with increased risk of self-injury in adults. However, evidence was inconclusive for children and young people (k = 1; N = 206).

7.2.3.5 Mental health needs

- Low quality evidence from up to 3 studies (N = 32,516) suggested that the presence of mental health needs was associated with increased risk of all aggression, self-injury and stereotypy.
- Very low quality evidence from 2 studies (N = 30,874) suggested that the presence of mental health needs was associated with increased risk of property destruction although the precision of the effect was poor.
- Very low quality evidence from 2 studies (N = 30,874) suggested that the presence of mental health needs was associated with increased risk of physical aggression.
- Moderate quality evidence from 2 studies (N = 30,874) suggested that the presence of mental health needs was associated with increased risk of verbal aggression.

7.2.3.6 Expressive communication

- Very low quality evidence from up to 9 studies (N = 7,502) suggested that the presence of an expressive communication deficit was associated with increased risk of all aggression, self-injury and stereotypy.
- Very low quality evidence from a single study (N = 3,662) suggested that the presence of an expressive communication deficit was associated with increased physical aggression in an adult population. However, the opposite effect was found for a mixed population of children, young people and adults (k = 1; N = 211).

7.2.3.7 Receptive communication

- Moderate quality evidence from 3 studies (N = 1,321) suggested that the presence of a receptive communication deficit was associated with increased risk of self-injury.

7.2.3.8 Auditory impairment

- Very low quality evidence from up to 3 studies (N = 2,086) was inconclusive as to whether an auditory impairment was associated with the risk of all aggression, self-injury or stereotypy.

7.2.3.9 Mobility impairment

- Very low quality evidence from a single study (N = 1,023) was inconclusive as to whether a mobility impairment was associated with the risk of combined physical, verbal and destructive aggression.
- Very low quality evidence from a single study (N = 147) suggested that a mobility impairment was associated with increased risk of self-injury in children and young people although precision of the estimate is poor. Evidence from the adult population was inconclusive (k = 1; N = 1023).

7.2.3.101 Visual impairment

- Low quality evidence from 2 studies (N = 1,938) was inconclusive as to whether a visual impairment was associated with the risk of combined physical, verbal and destructive aggression.
- Low quality evidence from 3 studies (N = 2,086) suggested that visual impairment was associated with increased risk of self-injury and stereotypy.

7.2.47 Economic evidence statements

This review question was not relevant for economic analysis.

7.3 Review question: In people with a learning disability, what is the utility of methods and tools used to assess the circumstances, risk factors and antecedents associated with the development of behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 53. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 53: Clinical review protocol summary for the review of the utility of methods and tools used to assess the circumstances, risk factors and antecedents associated with the development of behaviour that challenges

Component	Description
Review question(s)	In people with a learning disability, what is the utility of methods and tools used to assess the circumstances, risk factors and antecedents associated with the development of behaviour that challenges?
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability
Intervention(s)	Methods and tools used to assess the circumstances, risk factors and antecedents associated with the development of behaviour that challenges: • Methods and tools for personal assessment including assessment of sensory deficits, sensory processing disorders, physical health status, communication needs, emotional needs and mental health needs • assessment of environmental factors including the physical environment, the social environment, parent, carers and staff attitudes, skills and staff competence
Comparison	Not applicable
Critical outcomes	Sensitivity, specificity, reliability, validity
Study design	Any

7.3.1 Studies considered

The search for evidence identified 47 studies that met the eligibility criteria for this review: Atchinson 1998 (Atchison et al., 1998), Bamburg 2001 (Bamburg et al., 2001), Barratt 2012 (Barratt et al., 2012), Breau 2000 (Breau et al., 2000), Breau 2002 (Breau et al., 2002), Carr 2008 (Carr et al., 2008), Clifford 2010 (Clifford et al., 2010), Fisher 2000 (Fisher et al., 2000), Gleason 2012 (Gleason & Coster, 2012), Hatton 2008 (Hatton & Taylor, 2008), Hillier 2010 (Hillier et al., 2010), Iacono 2009 (Iacono et al., 2009), Kottorp 2008 (Kottorp, 2008), LeBlanc 1999 (LeBlanc et al., 1999), Linaker 1991 (Linaker, 1991), Lotan 2009a (Lotan et al., 2009a), Lotan 2009b (Lotan et al., 2009b), Lotan 2010 (Lotan et al., 2010), Lotan 2013 (Lotan et al., 2013), Mailloux 1990 (Mailloux, 1990), Manohari 2013 (Manohari et al., 2013), Masi 2002 (Masi et al., 2002), Matson 1984 (Matson et al., 1984), Matson 1991 (Matson et al., 1991), Matson 1997a (Matson & Smiroldo, 1997a), Matson 1997b (Matson et al., 1997b), Matson 1998a (Matson et al., 1998a), Matson 1998b (Matson et al., 1998b), Matson 1999 (Matson et al., 1999), McAtee 2004 (McAtee et al., 2004), McGill 2005 (McGill et al., 2005), Moss 1993 (Moss et al., 1993), Moss 1998 (Moss et al., 1998), Paclawskyj 1997 (Paclawskyj et al., 1997), Prosser 1998 (Prosser et al., 1998), Roy 2002a (Roy et al., 2002a), Sevin 1995 (Sevin et al., 1995), Stinnett 1999 (Stinnett et al., 1999), Sturmey 1990 (Sturmey & Ley, 1990), Sturmey 2004 (Sturmey et al., 2004), Sturmey 2005 (Sturmey et al., 2005), Swiezy 1995 (Swiezy et al., 1995), Tenneij 2009 (Tenneij et al., 2009a), Van der Gaag 1988 (Van

der Gaag, 1988), Van der Gaag 1990 (van der Gaag & Lawler, 1990), Walsh 1999 (Walsh & Shenouda, 1999), Watkins 2002 (Watkins et al., 2002).

Only 2 studies provided data for the critical outcomes of sensitivity and specificity. Data for reliability and validity were reported for the following assessment instruments:

- American Association on Mental Retardation (AAMR) Adaptive Behaviour Scale-School, Second Edition (AAMR ABS-S2)
- American Association on Mental Retardation Adaptive Behaviour Scale - Residential and Community (AAMR ABS)
- Assessment of Motor and Process Skills (AMPS)
- Checklist of Communicative Competencies Revised (Triple-C Revised)
- Communication Assessment Profile (CASP)
- Contextual Assessment Inventory (CAI)
- Diagnostic Assessment for the Severely Handicapped-II (DASH-II)
- Ecological Interview (EI)
- Health of the Nation Outcome Scales for People with Learning Disabilities (HoNOS-LD)
- Matson Evaluation of Social Skills for Individuals with Severe Retardation (MESSIER)
- Mini Psychiatric Assessment Schedule for Adults with a Developmental Disability (Mini PAS-ADD)
- Modified Classroom Observation Schedule to Measure Intentional Communication (M-COSMIC)
- Non communicating adults pain checklist (NCAPC)
- Non Communicating Children's Pain Checklist - Postoperative version (NCCPC-PV)
- Non Communicating Children's Pain Checklist (NCCPC)
- Psychiatric Assessment Schedule for Adults with a Developmental Disability (PAS-ADD)
- Psychiatric Assessment Schedule for Adults with a Developmental Disability Checklist (PAS-ADD Checklist)
- Psychopathology Instrument for Mentally Retarded Adults (PIMRA)
- School Assessment of Motor and Process Skills (School AMPS)
- Sensory Integration and Praxis Test (SIPT)
- Vineland Adaptive Behaviour Scales II (VABS II)

For ease of presentation, the evidence is organised by instrument and grouped within the following domains: communication needs, environmental factors, health status, mental health needs, pain assessment, sensory deficit, and severity of learning disability. Further details about the characteristics and psychometric properties of each instrument can be found in Appendix L.

7.3.2 Clinical evidence for assessment instruments

7.3.2.1 Communication needs

7.3.2.1.1 Communication Assessment Profile (CASP)

The CASP is a questionnaire and observation instrument which assesses the communicative competence of adults with a learning disability, including the form, function and context of language. There are 2 parts, plus an appendix. Part 1 is a staff questionnaire with 48 items, to be filled in by someone who works closely with the individual being assessed (such as a keyworker). Part 2 is completed by the speech therapist and has 8 sections which assess communication, for example, in one section photographs are presented to assess auditory

discrimination. The instrument takes 20-45 minutes to administer and costs £199.20. It is the only UK standardised assessment tool for adults with a severe to moderate learning disability.

The CASP was found to have high inter-rater reliability for therapist-to-therapist agreement (81%-99%) whereas therapist to key worker agreement has been found to be good for all subscales (70% - 82%), with the exception of the talking to self sub-scale which was moderate (56%) (Van der Gaag 1988; Van der Gaag 1990). Significant correlations have been found between CASP and the Adaptive Behaviour Scale (ABS) and Communicative Ground Scale (CGS), offering evidence of convergent validity (Van der Gaag1990).

7.3.2.1.2 Modified Classroom Observation Schedule to Measure Intentional Communication (M-COSMIC)

The M-COSMIC is an observation instrument for use in children with a learning disability. It was developed as an ecologically valid measure of social-communication behaviour, delineating forms, functions, and intended partners of children's spontaneous communication acts. It evaluates social-communication in children with autism with more varied levels of functioning and language ability than intended with the original measure which focused on low functioning individuals. It is completed by a researcher and takes approximately 25 minutes to administer. In Clifford 2010, researchers received approximately 25 hours of training of administration of the instrument.

The M-COSMIC was found to have good inter-rater reliability with the majority of intra-class correlations above 0.84. Good convergent validity has been found between the M-COSMIC and the Autism Diagnostic Observation Schedule – Generic algorithm total scores (ADOS-G), but not for specific items. Significant associations were also found between the M-COSMIC and several subscales of the Preschool Language Scales, the MacArthur-Bates Communicative Development Inventory and the VABS.

7.3.2.1.3 Matson Evaluation of Social Skills for Individuals with Severe Retardation (MESSIER)

The MESSIER is an 85-item instrument completed by a staff member. It is designed to assess social skills in adults with severe and profound learning disability.

The MESSIER has been found to have excellent internal consistency for the entire scale (0.94). Positive subscales have shown good to excellent internal consistency, ranging from 0.87-0.96, whereas negative subscales show acceptable internal consistency ranging from 0.73-0.81. Spearman rank-order correlation coefficients ranged from 0.14 to 0.89, suggesting inadequate to high inter-rater consistency on individual items. There was good inter-rater reliability for the scale as a whole (r = 0.73). Good convergent validity has been found between the MESSIER and relative measures including sociometric ranking and the Vineland.

7.3.2.1.4 Checklist of Communicative Competencies Revised (Triple C – Revised)

The Triple C – Revised is an 81-item observation instrument, completed by a staff member, which assesses communication among adolescents and adults with little to no speech. The revised checklist comprises 5 stages that reflect the continuum from unintentional to symbolic communication. The instrument takes 1 to 2 weeks to complete and the cost of the manual and checklists is £65.55.

The Triple C – Revised has been found to have excellent internal consistency (Kuder–Richardson Formula 20 ranged from 0.83-0.93 for individual stages). Cohen's kappa has been found to yield a moderate to high coefficient (k=0.63) indicating good inter-rater reliability. Factor analysis has confirmed a 1-factor solution indicating good structural validity.

7.3.2.2 Environmental factors

7.3.2.2.1 Contextual Assessment Inventory (CAI)

The CAI is an 80-item questionnaire completed by a staff member. It rapidly identifies generic classes of contextual variables associated with problem behaviour in adults with developmental disabilities. Subcategories include social/cultural contexts, task/activity contexts, physical contexts, and biological contexts. The instrument takes 25 minutes to administer and is available for free.

The CAI has shown good test-retest reliability across studies. Inter-rater reliability has ranged from good (mean percentage agreement 94.8%) to poor (intra-class correlation = 0.28). Internal consistency has been found to be excellent (α=0.95). Significantly more behaviour log entries corresponded to items rated as frequently associated with problem behaviour on the CAI than corresponded to items rated as rarely associated with problem behaviour (effect size = 0.76). Problem behaviour was significantly more likely to occur in the contexts rated on the CAI as frequently associated with problem behaviour in contrast to those rated as rarely associated with problem behaviour (effect size 0.85).

7.3.2.2.2 Ecological Interview (EI)

The EI is a 76-item interview completed by a staff member for use in children, young people and adults with a learning disability. It investigates the relationship between environmental events and variability in behaviour that challenges. The instrument is available for free.

The EI has shown good test-retest reliability (weighted kappa =0.64). McGill (2005) demonstrated 100% agreement between staff ratings of frequency and 98.7% agreement for ratings of likelihood of behaviour that challenges using the EI. Barratt (2012) found that some items of the EI showed significant correlation with the CAI but this was not consistent.

7.3.2.3 Health status

7.3.2.3.1 Health of the Nation Outcome Scales for People with Learning Disabilities (HoNOS-LD)

The HoNOS-LD is an 18-item questionnaire completed by a staff member. It was developed to measure health and social functioning among adults with learning disability. Scales cover a wide range of health and social domains: psychiatric symptoms, physical health, functioning, relationships and housing. One-day training and a half-day re-training every 2 years for clinical staff is required. The course can be delivered with up to 25 delegates for £3,000.00. The measure itself is free to use in NHS funded care.

The HoNOS-LD has been found to have acceptable to good internal consistency (α=0.74-0.89) (Tenneij 2009). Inter-rater reliability has been found to be good (kappa = 0.58-0.86; Pearson's r=0.82) (Roy 2002a; Tenneij 2009).The HoNOS-LD has been found to be a useful tool in measuring clinical outcomes. Hillier 2010 demonstrated significant improvements in mental state, behaviour and social functioning following in-patient treatment and Roy 2002a found a significant difference in ratings over time for individuals engaged in treatment, suggesting sensitivity to change. Nurses' ratings on the HoNOS-LD have been found to distinguish between individuals placed on closed wards and outpatients, although psychiatrist/psychologists ratings have not been found to do so (Tenneij 2009). The HoNOS-LD has been found to be positively correlated with the ABC, Social Functioning Scale for the Mentally Retarded and Adult Behavior Checklist indicating good convergent validity (Roy 2002a; Tenneij 2009).

7.3.2.4 Mental health needs

7.3.2.4.1 Diagnostic Assessment for the Severely Handicapped-II (DASH-II)

The DASH-II is an 84-item questionnaire completed by a staff member or family member or carer for use in people with a severe and profound learning disability. It is a measure of comorbid psychopathology and consists of 13 subscales: anxiety, depression, mania, PDD/autism, schizophrenia, stereotypies, self-injury, elimination, eating, sleeping, sexual, organic, and impulse control. The instrument costs £192 including the manual, 50 protocols, 50 score sheets and shipping from the USA.

Sevin 1995 found the mean percentage agreement (MPA) across all items to be 0.86 for frequency, 0.85 for duration, and 0.95 for severity of the disorder. Intra-class correlation coefficients were greater than 0.5 for 10 of the subscales, indicating adequate agreement. However, they were less than 0.5 for the anxiety, schizophrenia and sexual disorders subscales indicating poor agreement. Sevin 1995 calculated percentage agreement and kappa coefficients. MPA across all items was 0.84 for frequency, 0.84 for duration, and 0.91 for severity. Good inter-rater reliability was also reported by Matson 1991. Internal consistency has been found to vary from unacceptable to good across subscales, with good internal consistency for the total scale (0.87; Paclawski 1997). Numerous studies have evaluated the subscales of the DASH-II and have found them to be valid for the diagnosis of depression (Matson et al, 1997), mania (Matson & Smiroldo, 1997), schizophrenia (Bamburg 2001), and autism/pervasive developmental disorder (Matson et al, 1998). However, caution has been reported in terms of the validity of the anxiety subscale due to high rates of false positive diagnoses (Matson et al, 1997). Sturmey 2004 found 5 factors that were named emotional lability/antisocial, language disorder, dementia/anxiety, sleep disorder, and psychosis. Scales derived from this factor analysis were internally consistent. The DASH-II demonstrates good convergent and discriminant validity with the Aberrant Behavior Checklist (ABC), MESSIER, and Vineland Adaptive Behavior Scales (VABS). (Paclawski 1997; Sturmey 2004).

7.3.2.4.2 Mini Psychiatric Assessment Schedule for Adults with a Developmental Disability (Mini PAS-ADD)

The Mini PAS-ADD is an 86-item instrument for use in adults with a learning disability. Rather than being an interview, the mini version of the PAS-ADD provides a framework for an individual, or team to collect together relevant information on psychiatric symptomatology which is available without the need for interviewing. Secondly, the Mini PAS-ADD is aimed at case identification, rather than full ICD-10 diagnostic evaluation. The Mini PAS-ADD is a more elaborate instrument that requires some training in its administration, and that provides information that is more detailed, and more rigorously coded, than the PAS-ADD Checklist.

Prosser 1998 found alpha coefficients to range from questionable to excellent (α=0.60-0.95). Inter-rater reliability for case identification has been found to be moderate (kappa=0.44, Prosser 1998). There was no available data on validity.

7.3.2.4.3 Psychiatric Assessment Schedule for Adults with a Developmental Disability (PAS-ADD)

The PAS-ADD is a 66-item interview primarily designed for adults with a level of language that enables them to give some verbal contribution to the interview. It provides full diagnoses under both ICD-10 and DSM-IV (TR).

The PAS-ADD has been found to have good inter-rater reliability across all items (Moss 1993). There was no available data on validity.

7.3.2.4.4 *Psychiatric Assessment Schedule for Adults with a Developmental Disability Checklist (PAS-ADD Checklist)*

The PAS-ADD checklist is a screening instrument specifically designed to help staff recognise mental health problems in the adults with learning disability for whom they care, and to make informed referral decisions. It consists of a life-events checklist, and 29 symptom items scored on a 4-point scale. It covers: appetite and sleep, tension and worry, phobias and panics, depression and hypomania, obsessions and compulsions, psychoses, and autism. The cost of a pack of 20 checklists is £60.

Two studies assessed the sensitivity and specificity of the measure in adults with a learning disability (Moss 1998, Sturmey 2005). Both studies showed that the sensitivity and specificity of the measure was moderate. In Moss 1998 (N = 59) sensitivity was 0.7 and specificity was 0.69. In Sturmey 2005 (N = 226) sensitivity was 0.66 and specificity was 0.7.

Inter-rater reliability has been found to be good when the PAS-ADD Checklist is used for case identification purposes (Moss 1998). Internal consistency has been found to be acceptable for the total checklist but variable for subscales (0.51-0.87; Moss 1998, Sturmey 2008). Moss (1998) found that although the checklist showed broadly satisfactory validity, 2 individuals had been judged by the psychiatrist as having a severe condition, but were not detected by instrument. Hatton 2008 concluded that given the inconsistency of empirically derived subscales, the PAS-ADD Checklist should not be used to identify specific types of psychopathology. The checklist may have more utility as a screening tool for general psychopathology and subsequent referral for more detailed assessment.

7.3.2.4.5 *Psychopathology Instrument for Mentally Retarded Adults (PIMRA)*

The PIMRA is a 56-item diagnostic instrument for psychiatric diagnoses in adolescents and adults with different degrees of learning disability. It is completed by a staff member, family member or carer or is self-completed. Items are grouped in 8 subscales: schizophrenia, affective disorders, adjustment disorders, anxiety disorders, somatoform disorders, personality disorders and poor adjustment which correspond to DSM-III classifications. The cost of the instrument kit and shipping is £194.

Inter-rater reliability for case identification has been found to be good (86% agreement, Linkaker 1990; kappa 0.64, Linaker 1991). Internal consistency has been found to be variable, ranging from unacceptable to good for informant and self-report measures across studies (α=0.40-0.85, Matson 1984; Sturmey 1990; Watson 1988). The stability of scores over time has been found to be variable. Small to large correlations have been found for PIMRA subscale scores taken at 5 month intervals (Watson 1988), although total PIMRA scores have been found to be highly correlated over time (Matson 1984; Watson 1988). A good level of correspondence has been found between PIMRA and DSM diagnosis classifications in general, although may not be satisfactory when a high level of diagnostic precision is required (Linaker 1991; Linaker 1994). Authors have pointed out that the PIMRA may not be satisfactory as the only basis for diagnosis. Total PIMRA scores have been found to be significantly correlated with the ABC, Child Behaviour Checklist (CBCL), DSM-III and the Zung Anxiety Scale, but not with CBCL and Zung depression subscales (Masi 2002; Sturmey 1990; Swiezy 1995). Matson 1984 found inconsistency between the factors identified for the self-report and informant versions of the PIMRA. The authors suggested that this may demonstrate difficulty on the part of mentally retarded patients to discriminate on the particular type of psychopathology that they are experiencing.

7.3.2.5 Pain assessment

7.3.2.5.1 *Non Communicating Adults Pain Checklist (NCAPC)*

The NCAPC is an 18-item observation instrument which measures pain behaviour among adults with a learning disability. It includes 6 sub-categories of pain behaviour: vocal reaction,

emotional reaction, facial expression, body language, protective reaction, and physiological reaction. The instrument is completed by a staff member or a researcher and is available for free.

Internal consistency of the NCAPC has been shown to be acceptable to good (α=0.72-0.85) (Lotan, 2009b; Lotan 2010; Lotan 2013). Inter-rater reliability has been found to vary from low (0.40-0.49 within groups of nurses and case managers) to high (0.77-0.92 within groups of paid carers and therapists) (ICC(1,1) = 0.40–0.88). Reliability between paid carer and therapists has been found to be moderate (0.71-0.75) (Lotan 2009a). Lotan (2013) found high inter-rater reliability between 2 observers (role unspecified). Relative intra-rater reliability has been found to be high (ICC 0.93 - 0.94) (Lotan 2009a). The NCAPC has shown moderate sensitivity to detect pain: a standardised response means (SRM) of 0.57 was found in Lotan 2013. Lotan 2009b and Lotan 2010 found that SRM values were high for the whole sample as well as for all levels of learning disability. The mean NCAPC sum scores monitored across different situations have shown significantly lower values ($p < 0.05$) during no pain situations (dormitory and dental clinic waiting room), than during pain situations (influenza injection and dental hygiene treatment) (Lotan, 2010). Significant correlations have been found between the NCAPC and the Pain and Discomfort Scale (PADS) indication good convergent validity (Lotan, 2013).

7.3.2.5.2 Non Communicating Children's Pain Checklist (NCCPC)

The NCCPC is a 26-item observation instrument completed by a staff member and researchers, which measures pain behaviour among children with a learning disability. It takes 10 minutes to administer and is available to use for free.

The NCCPC has shown acceptable internal consistency (Breau 2000).The number of items reported by carers during pain has been found to be consistent over time. This indicates that the Checklist was reliable when used by the same observer for 2 discrete pain events. It also provides evidence that the pain behaviour of those with cognitive impairments may be consistent over time (Breau 2000). NCCPC scores have been found to be significantly correlated with carers' numerical pain ratings which indicates how helpful the specific behaviour is for deciding on the presence of pain, however this comparison scale was not validated (Breau, 2000).

7.3.2.5.3 Non Communicating Children's Pain Checklist - Postoperative version (NCCPC-PV)

The NCCPC-PV is a 27-item observation instrument completed by a staff member, researcher, family member or carer, which assesses postoperative pain among children with a learning disability. It takes 10 minutes to administer and is available to use for free.

The NCCPC-PV has been found to be internally reliable (α=0.71-0.91; Breau 2002). Intra-class correlations for total scores have been found to be 0.82 before surgery and 0.78 after surgery. Thus, total scores showed good inter-rater reliability (Breau 2002). Postoperative NCCPC-PV scores have been found to be correlated with visual analogue scale ratings provided by carers and researchers, but not with those of nurses (Breau 2002).

7.3.2.6 Sensory deficits

7.3.2.6.1 Sensory Integration and Praxis Test (SIPT)

The SIPT is an observation instrument completed by a psychologist (or related discipline) which is designed to measure the sensory integration processes that underlie learning and behaviour in children. It consists of 17 subtests requiring children to perform visual, tactile, kinesthetic, and motor tasks. It takes 120 minutes to administer and 30-45 minutes to score. The cost of the instrument is £634 which includes 10 copies of all test materials.

Test-retest coefficients for the major test scores on the 17 subtests of the SIPT have been found to range from 0.48 - 0.93 indicating poor to excellent reliability (Mailloux 1990).The inter-rater reliability coefficients have been found to range between 0.94 and 0.99 indicating excellent reliability (Mailloux, 1990). Factor analyses of the SIPT generally demonstrate the emergence of factors that can be seen as logically related to past groupings of scores, with the addition of new factors specifically reflecting the inclusion of additional measures of praxis (Mailloux 1990). The SIPT has been found to discriminate between children without dysfunction and those with dysfunction at a statistically significant level (Mailloux 1990).

7.3.2.7 Severity of learning disability

7.3.2.7.1 American Association on Mental Retardation Adaptive Behaviour Scale - Residential and Community (ABS)

The ABS is a questionnaire with 612 items which measures adaptive behaviour among adults in community and residential settings. Part 1 evaluates adaptive behaviours considered important to personal responsibility and independent living. Part 2 assesses social adaptations and maladaptive behaviour. The measure takes 30 minutes to administer.

There was no available data on the reliability of this measure however the previous version of this measure (AAMD ABS) was found to have good internal consistency and variable inter-rater reliability (Bean & Roszkowski, 1982; Roszkowski, 1982). Significant correlations have been found between the ABS Part II and Reiss Screen, ABC Irritability and Hyperactivity subscales, indicating good convergent validity (Walsh 1999). Discriminant validity was not reported for this measure however the previous version of this measure was found to successfully discriminate between children placed at different levels of special education and between children with different levels of learning disability (Malone & Christian, 1974).

7.3.2.7.2 American Association on Mental Retardation Adaptive Behaviour Scale-School, Second Edition (ABS-S2)

The AMS-S2 is a 2-part instrument with 437 items designed to evaluate adaptive behaviour in children aged 3 to 18 who are being evaluated for learning disability, autism, and/or behaviour disorders. Part 1 features 9 behaviour domains and evaluates adaptive behaviours considered important to personal responsibility and independent living. Part 2 features 4 behaviour domains that assess social adaptations and maladaptive behaviour. The instrument is completed by clinicians and takes 15-30 minutes to administer. To administer the measure there is a requirement to complete a graduate-level course in tests and measurement at a university or equivalent documented training. The cost of 2 exam booklets is £44.36 and 25 forms cost £21.60.

There was no available data on the reliability of this measure. Watkins 2002 and Stinnett 1999 found that a 2-factor solution provided the best dimensional model. These results suggest that interpretation of the AAMR ABS-S2 should focus on its 2 major conceptual components (personal independence and social behaviour) rather than the 5 factors and 16 domains endorsed by its authors.

7.3.2.7.3 Assessment of Motor and Process Skills (AMPS)

The AMPS is a 36-item observation instrument completed by an occupational therapist. It is designed to evaluate how well adults with a learning disability are able to perform personal or instrumental daily living activities. Participants receive a score based on the quality of 16 motor and 20 process performance skills. The measure takes 60 minutes to administer and score. The training course to administer the instrument costs £592 and the manual and scoring guide costs £57.

There was no available data on the reliability of this measure. Kottorp 2008 found that a difference of 1.0 logit on the AMPS process scale increases the likelihood of needing minimal

or no assistance by more than 3 times (odds ratio = 3.11), although the motor ability measure did not add significantly to the predictive value of the model.

7.3.2.7.4 School Assessment of Motor and Process Skills (School AMPS)

The School AMPS is a 36-item observation-based instrument completed by an occupational therapist and designed to measure students' ability to perform functional school tasks. The School AMPS is similar to the original AMPS in design, with several important modifications: (a) the tasks are related to school work instead of activities of daily living; (b) the scoring manual includes examples applicable to classroom tasks; and (c) the occupational therapist interviews a student's educational team members to determine a student's problem tasks (instead of choosing assessment tasks on the basis of a student interview) and matches these problem tasks with School AMPS tasks. The measure takes 60 minutes to administer and score. The training course to administer the instrument costs £586 and the manual costs £39.

The School AMPS has been found to have strong intra-rater reliability and goodness-of-fit demonstrating consistency of scoring (Atchinson, 1998; Fisher, 2000). Studies have used Rasch analysis to assess structural validity. Four facets were Motor skill items have been found to show acceptable goodness-of-fit, although Atchison 1998 found that findings for process items are more mixed (Atchison 1998; Fisher 2000). The School AMPS has suggested that the person response validity is acceptable for the motor scale but not for the process scale (Fisher 2000). Good convergent validity has been found between the Peabody Developmental Motor Scale–Fine Motor (PDMS-FM) and Motor scale of the AMPS (Atchinson 1998).

7.3.2.7.5 Vineland Adaptive Behaviour Scales II (VABS II)

The VABS-II is a 297-item interview completed by a researcher, family member or carer for children and young people with a learning disability. It is designed to support the diagnosis of learning and developmental disabilities, autism and ADHD by assessing adaptive functioning in 5 domains: communication (receptive, expressive and written), socialisation (interpersonal relationships, play and leisure time and coping skills), daily living skills (personal, domestic and community) and motor skills (gross and fine, only applicable for children under 6); maladaptive behaviour (optional for children 5 years and over). The instrument takes 20-60 minutes to administer and 15-30 minutes to score. Examiners and scorers should have graduate training in test administration and interpretation. The cost of an interview starter set is £118 and the manual costs £56.

There was no available data on the reliability of this measure however the previous version of this measure showed good internal consistency, inter-rater reliability and test-retest reliability. Gleason 2012 used content analysis to demonstrate that the items of the Vineland II map well onto the International Classification of Functioning, Disability and Health (ICF), demonstrating good convergent validity. Manohari 2013 suggested that the Vineland may not be readily generalisable to Indian participants due to differences in gender roles and self-care activities between the West and India.

7.3.3 Health economic evidence

No studies assessing the cost effectiveness of methods and tools used to assess the circumstances, risk factors and antecedents associated with the development of behaviour that challenges in people with a learning disability were identified by the systematic search of the economic literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

7.3.14 Clinical evidence statements

- For the CASP instrument, there was evidence from 2 studies demonstrating adequate reliability and validity, although evidence for test-retest reliability, internal consistency and criterion validity were not available.
- For the M-COSMIC instrument, there was evidence from 1 study demonstrating good reliability and validity, although evidence for test-retest reliability, internal consistency and criterion validity were not available.
- For the MESSIER instrument, there was evidence from 5 studies demonstrating adequate reliability and validity, although evidence for test-retest reliability and criterion validity was not available and inter-rater reliability for subscales was mixed.
- For the Triple-C revised instrument, there was evidence from 1 study demonstrating adequate reliability and validity, although evidence for test-retest reliability and criterion validity were not available.
- For the CAI instrument, there was evidence from 2 studies demonstrating adequate reliability and validity, however for inter-rater reliability the evidence was mixed.
- For the EI instrument, there was evidence from 2 studies demonstrating adequate reliability, however the evidence for construct validity was unclear and there was no evidence for internal consistency or criterion validity.
- For the HoNOS-LD instrument, there was evidence from 3 studies demonstrating good reliability and validity, although there was no evidence for re-retest reliability and evidence for criterion validity was mixed.
- For the DASH-II instrument, there was evidence from 9 studies demonstrating adequate reliability and validity, however inter-rater reliability was mixed and criterion validity was not available.
- For the Mini PAS-ADD instrument there was evidence from 1 study demonstrating adequate internal consistency, however inter-rater reliability was poor and there was no evidence for test-retest reliability, construct or criterion validity.
- For the PAS-ADD instrument, there was evidence from 1 study demonstrating good inter-rater reliability, however there was no evidence for test-retest reliability, internal consistency or validity.
- For the PAS-ADD checklist, there was evidence from 2 studies demonstrating moderate sensitivity and specificity. Evidence from 3 studies demonstrated good inter-rater reliability and internal consistency for the total checklist, however evidence for construct validity was poor and there was no evidence for test-retest reliability and criterion validity.
- For the PIMRA instrument, there was evidence from 5 studies demonstrating adequate reliability, however evidence for internal consistency and structural validity was mixed and there was no evidence for criterion validity.
- For the NCAPC instrument, there was evidence from 4 studies demonstrating adequate reliability and validity, although evidence for criterion validity was not available and inter-rater reliability was mixed.
- For the NCCPC instrument, there was evidence from 1 study demonstrating adequate reliability and validity, although evidence for inter-rater reliability and criterion validity was not available.
- For the NCCPC-PV instrument, there was evidence from 1 study demonstrating adequate reliability and validity, although evidence for test-retest reliability and criterion validity was not available.
- For the SIPT instrument, there was evidence from 1 study demonstrating adequate reliability and validity, although evidence for internal consistency and criterion validity was not available and evidence for test-retest reliability varied for each subscale.
- For the ABS instrument, there was evidence from 1 study demonstrating good construct validity, however evidence for reliability and criterion validity was not available.

- For the ABS-S2 instrument, there was evidence from 2 studies demonstrating good construct validity, however evidence for reliability and criterion validity was not available.
- For the AMPS instrument, there was evidence from 1 study indicating adequate validity, however evidence for reliability and construct validity was not available.
- For the School AMPS instrument, there was evidence from 2 studies indicating adequate reliability and validity, although evidence for test-retest reliability, internal consistency, and criterion validity was not available.
- For the VABS II instrument, there was evidence from 2 studies indicating adequate validity, however evidence for reliability and criterion validity was not available.

7.3.5 Economic evidence statements

No evidence on the cost effectiveness of methods and tools used to assess the circumstances, risk factors and antecedents associated with the development of behaviour that challenges in people with a learning disability is available.

7.4.4 Recommendations and link to evidence

Recommendations	
	18. Be aware of the risk of behaviour that challenges when working with people with a learning disability and their family members or carers, and that it often develops gradually. Pay attention to factors that may increase this risk, including: • personal factors, such as o a severe learning disability o autism o communication difficulties (expressive or receptive) o visual impairment (which may lead to increased self-injury and stereotypy) o physical health problems o variations with age (peaking in the teens and twenties) • environmental factors, such as: o abusive or restrictive social environments o environments with little sensory stimulation and those with low engagement levels o developmentally inappropriate environments (for example, a curriculum that makes too many demands on a child or young person) o environments where disrespectful social relationships and poor communication are typical. 19. Consider using direct observation and recording or formal rating scales (for example, the Adaptive Behaviour Scale or

	Aberrant Behaviour Checklist) to monitor the development of behaviour that challenges.
Relative values of different outcomes	The GDG specified that all of the following outcomes were of critical importance: determining the factors associated the risk of developing behaviour that challenges and identifying tools that support the recognition of those factors associated with increased risk of developing behaviour that challenges.
Trade-off between clinical benefits and harms	A number of personal factors (for example, autism) may be associated with an increased risk of developing behaviour that challenges. Some findings did not accord with GDG experience (that is, male gender reducing risk of any aggression), but this may be explained by selection bias. Less evidence was identified for environmental factors, for example, impoverished social environments. A number of tools were also identified which also had evidence to support their use in recognising risk factors (largely personal factors). The GDG considered that such tools could support early intervention or careful monitoring to reduce the likelihood of behaviour that challenges developing. However, there are a number of limitations with this evidence. The importance of the various risk factors may vary with the setting in which they present, for example, gender may vary in importance as a risk factor, being less important in inpatient settings, where risk of behaviour that challenges may be the major consideration in determining admission. In addition, some factors may rely on information obtained from previous diagnostic or other form of assessment which may have limited reliability. These and other factors raise the possibility of harm arising from unnecessary concern or actions, such as increased monitoring, which might negatively impact on the person with a learning disability or their family
Trade-off between net health benefits and resource use	Identification of circumstances, risk factors and antecedents associated with the development of behaviour that challenges in people with a learning disability has important resource implications. Some methods and tools come with cost associated with examiner manuals, licences and testing materials. However, better assessment is likely to lead to potential cost savings if it allows better prediction (and thus more timely and effective management) and potentially prevention of incidents of behaviour that challenges.
Quality of evidence	The evidence across nearly all studies on the identification of risk factors was of low or very low quality. For the majority of the tools assessed the quality of the evidence was also low with considerable inconsistency in the reporting of sensitivity, specificity, reliability and validity of the tools.
Other considerations	In developing recommendations in this area the GDG were concerned to balance the potential advantages of early intervention with the potential harms of unnecessary anxiety or intervention. The GDG also drew on their expert knowledge as the potential risks factors associated with certain characteristics of the care environment had not been identified in the reviews undertaken. The GDG therefore identified a limited number of factors that both the evidence review and their own expert knowledge suggested are associated with the development of behaviour that challenges. They also drew on their knowledge to identify a number of characteristics of the care environment that could themselves precipitate behaviour that challenges, but which might also interactive negatively with personal risk factors. Finally the GDG saw the benefit of recommending the use of formal rating scales (such as the ABS and the Aberrant Behaviour Checklist) for monitoring behaviour. Behaviour that challenges often develops gradually and the GDG considered that not using formal and reliable rating scales might delay the deployment of effective interventions.

1

8 Assessment

8.1 Introduction

The assessment of behaviour that challenges because assessing the nature of the behaviour alone is rarely, if ever, sufficient to allow for the development of a support and intervention plan. Assessment needs to be able to adequately characterise the behaviour, its antecedents and its consequences, which may require a consideration of a person's developmental history, their mental and physical health, the social and physical quality of their environment, the nature of any care provided and the skills and capacities of those caring for them. It follows from this that the methods of assessment will need to be able to properly and reliably capture important dimensions of all these factors and that a range of assessment methods and skills will need to be available and may be best undertaken in a team context where teams members can draw on the skills and knowledge of each other and those of expert staff when needed. Central to assessment in this area is a consideration of the function of the behaviour, attempts to understand which are central to gaining an understanding of why the behaviour has emerged and what is maintaining it. Although potentially a complex and protracted process, assessment can also be relatively straightforward, for example, understanding that an increase in aggressive behaviour resulting from a painful and treatable tooth abscess, which a person with a learning disability was otherwise unable to communicate other than by changing their behaviour.

To be effective, assessment has to be able to more than simply set out an understanding of the function of the behaviour. It has to ensure the most appropriate means to involve service users, families and carers in the process so that not only is the assessment comprehensive and accurate but also that all involved can play an active part in the development of any support and intervention plan. In addition, if an assessment is to be comprehensive it means that skills of particular professionals may be needed; these could include a GP, psychiatrist, neurologist, paediatrician, speech and language therapist or psychologist. The presence of neurodevelopmental disorders such as autism or ADHD may complicate assessment, for example, because of communication problems arising from the disorder or associated behavioural problems if the neurodevelopmental disorder is not recognised. As noted above, unrecognised or untreated physical health problems may underlie the problem—sometimes it may be a simple problem such as toothache but it may be a more complex and life threatening disorder. Both neurodevelopmental and physical disorders can also complicate the identification of emerging mental disorders. Although the link between behaviour that challenges and mental illness is not well understood, new presentations of behaviour that challenges may be a manifestation of a new mental disorder or the relapse of a previously diagnosed one. However, the diagnosis of mental disorder in people with a learning disability poses difficulties resulting from communication problems, the developmental trajectory of a person with a learning disability and the presentation of the symptoms of mental disorders per se given the existing cognitive limitations.

Furthermore, behaviour that challenge may have an adverse impact on the person but also on those in caring roles. Therefore, it is acknowledged that the wellbeing of families and carers needs to be assured and an assessment of their ability to cope with the behaviour that challenges of the person they support is paramount. As part of the management of complex needs and behaviour that challenges in the community by secondary care mental health services the care programme approach (Department of Health, 2008) may be implemented. A formal carer's assessment carried out by social care is part of such a coordinated approach to management.

Before provision of any interventions for behaviour that challenges, it is recognised that an assessment of carers' capacity and resources ought to be made and clear objectives set in order to not only manage expectations but also to monitor the implementation of the support and intervention plan (Ali et al., 2014)

8.2 Review question: In people with a learning disability, what are the key components of, and the most effective structure for, an assessment of the behaviour that challenges across a range of settings?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 54. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 54: Clinical review protocol summary for the review of the key components of, and the most effective structure for, an assessment of the behaviour that challenges across a range of settings

Component	Description
Review question	In people with a learning disability, what are the key components of, and the most effective structure for, an assessment of the behaviour that challenges across a range of settings? (RQ2.1) To answer this question, consideration should be given to: • methods of assessment (including functional analysis) • formal assessment tools/ psychological instruments (including risk assessment) • biological and physical health measures
Population	Children, young people and adults with mild, moderate, severe or profound learning
Intervention(s)	Assessment of the behaviour that challenges (across a range of settings)
Comparison	• any control • another alternative assessment strategy
Critical outcomes	Clinical utility (including key components of, and the most effective structure for, an assessment of the behaviour that challenges)
Study design	N/A; GDG consensus-based

8.2.1 Clinical evidence

No studies assessing the methods and structure of instruments for the assessment of behaviour that challenges in people with a learning disability were identified by the systematic search of the literature undertaken for this guideline.

8.2.2 Clinical evidence statement

No evidence on the methods and structure of instruments for the assessment of behaviour that challenges in people with a learning disability is available.

8.3 Review question: In people with a learning disability and behaviour that challenges, what is the utility of methods and tools for assessment?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 55. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 55: Clinical review protocol summary for the review of the utility of methods and tools used to assess behaviour that challenges

Component	Description
Review question	In people with a learning disability and behaviour that challenges, what is the utility of methods and tools for assessment? (RQ2.2)
Population	Children, young people and adults with mild, moderate, severe or profound a learning disability
Intervention(s)	• Methods and tools for assessment (including assessment of sensory deficits, sensory processing disorders, physical health status, communication needs, emotional needs, individual, environmental risk factors and mental health needs) • Assessment of environmental factors (including the physical environment, the social environment, parent, carers and staff attitudes, skills and staff competence)
Comparison	N/A
Critical outcomes	**Sensitivity**: the proportion of true positives of all cases with behaviour that challenges **Specificity**: the proportion of true negatives of all cases without behaviour that challenges **Reliability**: inter-rater, test-retest, internal consistency **Validity**: criterion, construct
Study design	Any

8.3.1 Clinical evidence

The search for evidence (supplemented by GDG advice) identified 56 studies that met the eligibility criteria for this review: Akande 1998 (Akande, 1998), Aman 1985a (Aman et al., 1985a), Aman 1985b (Aman et al., 1985b), Aman 1987a (Aman et al., 1987a), Aman 1987b (Aman et al., 1987b), Aman 1995 (Aman et al., 1995), Aman 1996 (Aman et al., 1996), Barnard-Brak 2013 (Barnard-Brak et al., 2013), Bihm 1991 (Bihm & Poindexter, 1991), Brinkley 2007 (Brinkley et al., 2007), Brown 2002 (Brown et al., 2002), Clarke 2003 (Clarke et al., 2003), Crawford 1992 (Crawford et al., 1992), Dekker 2002 (Dekker et al., 2002), Duker 1998 (Duker & Sigafoos, 1998), Durand 1988 (Durand & Crimmins, 1988), Einfeld 1995 (Einfeld & Tonge, 1995), Gonzalez 2009 (Gonzalez et al., 2009), Haynes 2013 (Haynes et al., 2013), Hill 2008 (Hill et al., 2008), Joosten 2008 (Joosten & Bundy, 2008), Kearney 1994 (Kearney, 1994), Kearney 2006 (Kearney et al., 2006), Koritsas 2013 (Koritsas & Iacono, 2013), Lecavalier 2004 (Lecavalier et al., 2004), Marshburn 1992 (Marshburn & Aman, 1992), Matson 1999b (Matson et al., 1999b), Matson 2007c (Matson & Boisjoli, 2007c), Matson 2009 (Matson & Wilkins, 2009), Mohr 2005 (Mohr et al., 2005), Mohr 2011 (Mohr et al., 2011), Newton 1988 (Newton & Sturmey, 1988), Newton 1991 (Newton & Sturmey, 1991), Nicholson 2006 (Nicholson et al., 2006), Norris 2011 (Norris & Lecavalier, 2011), Oliver 2003 (Oliver et al., 2003), Oliver 2007 (Oliver et al., 2007), Paclawskyj 2000 (Paclawskyj et al., 2000), Paclawskyj 2001 (Paclawskyj et al., 2001), Rojahn 2001 (Rojahn et al., 2001), Rojahn 2003 (Rojahn et al., 2003), Rojahn 2010a (Rojahn et al., 2010a), Rojahn

2010b (Rojahn et al., 2010b), Rojahn 2012b (Rojahn et al., 2012b), Rojahn 2013 (Rojahn et al., 2013), Roy 2002a (Roy et al., 2002a), Sansone 2012 (Sansone et al., 2012), Shogren 2003 (Shogren & Rojahn, 2003), Sigafoos 1994 (Sigafoos et al., 1994), Singh 1993 (Singh et al., 1993), Spreat 1996 (Spreat & Connelly, 1996), Thompson 1995 (Thompson & Emerson, 1995), Walsh 1999 (Walsh & Shenouda, 1999), Watkins 2013 (Watkins & Rapp, 2013), Zaja 2011 (Zaja et al., 2011), Zarcone 1991 (Zarcone et al., 1991).

No studies provided data for the critical outcomes of sensitivity and specificity. Data for reliability and validity were reported for the following assessment instruments:

- Aberrant behaviour checklist (ABC)
- Behaviour Problem Inventory - Short Form (BPI-S)
- Behaviour Problem Inventory (BPI-01)
- Challenging Behaviour Interview (CBI)
- Developmental Behaviour Checklist (DBC-P)
- Developmental Behaviour Checklist for adults (DBC-A)
- Functional Analysis Screening Tool (FAST)
- Modified Overt Aggression Scale (MOAS)
- Motivation Assessment Scale (MAS)
- Nisonger Child Behaviour Rating Form (NCBRF)
- Questions About behavioural Function (QABF)
- Strengths and Difficulties Questionnaire (SDQ)

For ease of presentation, the evidence is organised by instrument and grouped within the following domains: behaviour that challenges (any), behaviour that challenges (aggression) and functional analysis. Further details about the characteristics and psychometric properties of each instrument can be found in Appendix L.

8.3.1.1 Behaviour that challenges (any)

8.3.1.1.1 Aberrant behaviour checklist (ABC)

The ABC is a 58-item questionnaire completed by unpaid carers, paid carers or teachers. It was designed as a problem behaviour rating scale to assess treatment effects in people with a learning disability. There are 5 subscales including: irritability, lethargy/social withdrawal; stereotypic behaviour; hyperactivity/noncompliance; and inappropriate speech.

In a sample of participants with any learning disability the internal consistency of the ABC ranged from good to excellent for subscales: irritability subscale, α=0.92-0.93; lethargy/social withdrawal subscale, α=0.90-0.91; stereotypic behaviour, α=0.84-0.90; hyperactivity, α=0.93-0.96; inappropriate speech α=0.76-0.86 (Aman 1995; Aman 1985b; Marshburn 1992). Test-retest reliability ranged from moderate to good. In Aman 1987a, inter-rater and test-retest reliability correlations varied markedly both across subscales and raters but were comparable to levels derived with other symptom checklists and were deemed to be adequate.

In a sample of participants with fragile X syndrome, internal consistency ranged from good to excellent (based on modified 6-factor solution): irritability subscale, α=0.94; hyperactivity, α=0.92, lethargy/social withdrawal α=0.86, social avoidance α=0.92 (newly derived factor), stereotypic behaviour, α=0.87, inappropriate speech, α=0.80 (Sansone 2012).

The 5-factor solution of the ABC has been replicated with learning disability and autism samples (Aman 1987b; Aman 1995; Bihm 1991; Brinkley 2007; Newton 1988). Brown 2002 and Marshburn 1992 found a 4-factor solution to be most appropriate with a learning disability sample, as the inappropriate speech factor was not replicated. Moderate to

excellent congruence has been found between the original ABC factor structure and that found with learning disability samples (0.62-0.97) (Aman 1987b; Aman 1995; Brown 2002; Marshburn 1992). Good convergent and divergent validity has been demonstrated by significant relationships between the ABC, Health of the Nation Outcome Scales for People with Learning Disabilities (HoNOS-LD), Vineland Adaptive Behaviour Scales II, Reiss Screen, Challenging Behaviour Inventory (CBI), Diagnostic Assessment for the Severely Handicapped-II (DASH-II) and Adaptive Behaviour Scale (ABS) (Aman 1985b; Hill 2008; Oliver 2003; Paclawski 1997; Rojahn 2003; Roy 2002a; Walsh 1999).

A 6-factor solution, which adds a 'social avoidance' factor to the original ABC factors has been found in a sample of participants with fragile X syndrome (Sansone 2012).

8.3.1.1.21 Behaviour Problem Inventory (BPI-01)

The BPI-01 is a 52-item respondent-based behaviour rating instrument. It is suitable for both children and adults with a learning disability and completed by unpaid carers, paid carers or teachers. It reports the frequency and severity of behaviour on 3 subscales: self-injurious; stereotypic; and aggressive/destructive.

In Rojahn 2010b the BPI-01 showed good reliability between teacher informants, but it was poor between parent and teacher informants. Gonzalez 2009 found that the inter-rater and re-test reliability coefficients of the self-injurious behaviour items and subscale were generally good, whereas the overall inter-rater and test–retest reliability coefficients of the aggression/destruction items and subscale were good to excellent. The stereotypy items and subscale had fair to low inter-rater and test-retest reliability coefficients (Gonzalez 2009). Internal consistency values range from poor to acceptable for the self-injurious behaviour subscale, poor to excellent for the stereotypy items and acceptable to good for aggressive/destructive behaviour (Gonzalez 2009; Rojahn 2001; Rojahn 2010b; Rojahn 2012b). Good convergent and divergent validity has been demonstrated by significant correlations in predicted directions between the BPI-01 and measures including the ABC, Nisonger Child Behaviour Rating Form (NCBRF), Inventory for Client and Agency Planning (ICAP), Autism Spectrum Disorders-Behaviour Problems for Intellectually Disabled Adults (ASD-BPA) and DASH-II (Hill 2008; Rojahn 2003; Rojahn 2010a; Rojahn 2010b; Rojahn 2012b).There have been mixed findings regarding structural validity. Rojahn 2001 and Gonzalez 2009 replicated a 3-factor solution and Hill 2008 found a 6-factor solution which mapped onto the 3-subscale structure. However, Rojahn 2010 failed to replicate a 3-factor solution. Barnard-Brak 2013 used confirmatory factor analysis to indicate acceptable model fit for each latent construct suggesting support for the one-dimensional nature of each trait. Individuals with a diagnosis of PDD had higher scores on the self-injurious behaviour and stereotyped behaviour subscales than those without; in addition, they also had elevated aggression/destruction scores. Higher stereotyped behaviour scores among people with a diagnosis of stereotyped behaviour disorder, compared with residents without, can be considered as another sign of validity of the BPI-01.

Rojahn 2013 included a sample of participants with Cornelia de Lange Syndrome only. In this study internal consistency values ranged from questionable to excellent (α=0.66-0.90) and there was evidence of a sufficient factor structure for each of the subscales identified by the BPI-01.

8.3.1.1.24 Behaviour Problem Inventory - Short Form (BPI-S)

The BPI-S is a shortened 30-item version of the BPI-01 completed by unpaid carers, paid carers or teachers. It is used for children and adults with a learning disability and contains the same 3 subscales as the BPI-01: self-injurious behaviour; stereotyped behaviour; and aggressive/destructive behaviour.

Internal consistency was found to be acceptable for the aggressive/destructive and stereotyped behaviour subscales of the BPI-S. For the self-injurious behaviour subscale,

values ranged from unacceptable to acceptable (Rojahn 2012b). Confirmatory factor analysis results indicated an acceptable model fit for each latent construct suggesting support for the one-dimensional nature of each trait (Barnard-Brak 2013). Good convergent and divergent validity has been demonstrated by significant correlations in predicted directions between the BPI and measures including the ABC, NCBRF, Inventory for Client and Agency Planning (ICAP) and DASH-II (Rojahn 2012b).

8.3.1.1.4 Challenging Behaviour Interview (CBI)

The CBI is a 19-item instrument completed by paid carers or teachers which measures the severity of behaviour that challenges in children and adults with a learning disability. It is divided into 2 parts. Part I of the interview identifies the occurrence of 5 clearly operationalised forms of behaviour that challenges that have occurred in the previous month. Part II of the interview assesses the severity of the behaviours identified on 14 scales measuring the frequency and duration of episodes, effects on the individual and others and the management strategies used by carers.

The CBI has been found to demonstrate good inter-rater reliability (kappa=0.50-0.80) and test-retest reliability (kappa=0.70-0.91). The CBI has also been found to be significantly correlated with the ABC showing good convergent validity (Oliver 2003).

8.3.1.1.5 Developmental Behaviour Checklist for adults (DBC-A)

The DBC-A is a 107-item questionnaire completed by unpaid or paid carers. It assesses a comprehensive range of emotional, behavioural and mental health problems in adults with mild, moderate and more severe levels of learning disability. The manual and supplement cost £64.92 and a pack of 10 checklists cost £5.90.

The DBC-A has shown substantial agreement between family members (ICC=0.72; Mohr 2005) and acceptable agreement between paid carers (ICC 0.69; Mohr 2011). Test-retest reliability has been found to be good, ranging from 0.75-0.85 (ICC; Mohr 2005). A strong positive correlation has been demonstrated between the DBC-A and both the PAS-ADD and ABC, providing evidence of good convergent validity (Mohr 2005).

8.3.1.1.6 Developmental Behaviour Checklist (DBC-P)

The DBC-P is a suite of instruments for the assessment of behavioural and emotional problems of children and young people with developmental and learning disabilities completed by unpaid and paid carers. It has 96 items and takes 10 to 15 minutes to administer. The starter kit, which consists of a manual and a packet of checklists and score sheets, costs £77.46.

Internal consistency has been found to be questionable for the antisocial subscale (α=0.67) and acceptable to excellent for the remaining subscales (α=0.73-0.91) based on the original 6-factor solution (Einfeld 1995). Internal consistencies for a revised 5-factor solution have been found to range from questionable for the anxiety subscale (α=0.66) to excellent for the disruptive/antisocial and self-absorbed subscales (α=0.91) (Dekker 2002). Inter-rater reliability for parent ratings was moderate to substantial (ICC=0.75-0.80) and poor to substantial for teacher ratings (ICC=0.30 - antisocial subscale; ICC=0.74 - self-absorbed subscale) (Einfeld 1995). Test-retest reliability was found to be moderate to substantial (ICC=0.75-0.80) (Einfeld, 1995).

Post-treatment change as measured by the DBC has been found to be strongly correlated with change as rated by an experienced clinician (Clarke 2003). Einfeld 1995 produced 6 clinically meaningful and factorially valid subscales using principle components analysis: disruptive, self-absorbed, communication disturbance, anxiety, social relating, and antisocial. However, Dekker 2002 suggested that a 5-factor solution was more appropriate, which included the following subscales: disruptive/antisocial, self-absorbed, communication disturbance, anxiety, and social relating. Dekker 2002 suggested that this revised scale

structure constitutes an improvement over the original structure given that it is based on a larger sample and one that better represents all levels of learning disability. Strong positive correlations have been found between the DBC and the Adaptive Behaviour Scale (0.72) and the Scales of Independent Behaviour (0.72 $p < .001$ in each case). Pearson product-moment correlations between the DBC total score and psychiatrist ratings has been found to be significant (0.81, $p < .001$) (Einfeld 1995).

8.3.1.1.7 Nisonger Child Behaviour Rating Form (NCBRF)

The NCBRF is a standardised instrument for assessing child and adolescent behaviour completed by families, carers or teachers. It has 76 items and a scoring time of 8 minutes. The instrument is available for free.

Poor inter-rater reliability for the NCBRF prosocial scales has been found between teacher and parent-teacher ratings. For the problem behaviour scales teacher-teacher agreement was fair, but parent-teacher agreement ranged from poor to moderate (Aman 1996; Rojahn 2010b). Rojahn 2010b found fair reliability for prosocial and problem behaviour subscales. Internal consistency has been found to be fair to good for the prosocial scales and good for the problem behaviour scales, based on a learning disabilities sample (Aman 1996; Norris 1999; Rojahn 2010b). Based on a sample of participants with autism, Lecavalier 2004 found questionable to good consistency for the adaptive social subscale (α=0.63-0.79), acceptable to good consistency for the compliant/calm (α=0.79) based on parent and teacher ratings, respectively. Studies indicated strong convergent and divergent validity between the NCBRF and BPI-01, ABC and DBC (Aman 1996; Norris 1999; Rojahn 2010b). There have been mixed findings regarding the factor structure of the NCBRF. Lecavalier 2004 and Norris 1999 replicated a 2-factor structure for social competence items based on autism and learning disabilities samples. But Rojahn 2010b found the fit for a 2-factor solution to be poor. Lecavalier 2004 found a 5-factor solution to be more appropriate than the original 6-factor solution for problem behaviour items. Other studies have demonstrated poor fit for both 5- and 6-factor solutions for this scale (Norris 1999; Rojahn 2010b).

8.3.1.1.8 Strengths and Difficulties Questionnaire (SDQ)

The SDQ is one of the most widely used brief questionnaires for assessing mental health problems in children and adolescents. It has 25 items and is divided into 5 domains: emotional symptoms, conduct problems, hyperactivity, peer problems and pro-social behaviour. It can be self-completed or administered by families, carers and teachers, and is available for free.

The SDQ has been found to show acceptable internal consistency overall (α=0.71) with subscales ranging from unacceptable (α=0.30 for peer problems) to good (α=0.87 for total impact) (Emerson 2005). Inter-rater reliability has been found to be modest for child ratings when compared with parent and teacher ratings (0.11 for peer problems subscale - 0.49 for hyperactivity) (Emerson 2005). Self-reported difficulties have been found to be significantly correlated with ICD-10 diagnoses (Emerson 2005). In a population of children with a learning disability Haynes 2013 found that a 3-factor model was a better measure than the original 5-factor model.

8.3.1.2 Behaviour that challenges (aggression)

8.3.1.2.1 Modified Overt Aggression Scale (MOAS)

The MOAS is designed to measure aggressive behaviours in adults and children. It is a 20-item instrument which is divided into 5 categories: verbal aggression towards others, verbal aggression towards self, physical aggression against objects, physical aggression against self and physical aggression against others. The MOAS differs from the original Overt Aggression Scale by modifications to wording and the addition of items measuring verbal aggression toward self. It is completed by unpaid or paid carers and is available for free.

The MOAS has been found to have a high level of agreement between raters for verbal aggression (ICC =0.90), physical aggression against others (ICC=0.90) and for total MOAS score (ICC=0.93). Levels of agreement on the other 2 subscales have been found to be lower but still in the moderate range (ICC=0.49-0.56) (Oliver 2007). There were no data available for the validity of the measure.

8.3.1.3 Functional analysis

8.3.1.3.1 Functional Analysis Screening Tool (FAST)

The FAST is a functional assessment tool designed to assess 4 functional properties of problem behaviour in adults with a learning disability. The 4 subscales are labelled: social (attention/preferred items), social (escape from tasks/activities), automatic (sensory stimulation) and automatic (pain attenuation).It has 16 items and is completed by a paid carer, family carer or teacher. It takes approximately 10 minutes to score and is available for free.

The FAST has been found to have unacceptably low internal consistency (α=0.05-0.77 for each subscale with a mean of 0.39) especially for the social attention and social escape subscales (Zaja 2011). Correlations for inter-rater agreement have been found to range from poor to good (ICC=0.48–0.71) (Zaja 2011). Test-retest correlation coefficients have been found to range from fair to excellent for total FAST scores (0.55-0.82) (Zaja 2011). Convergent and discriminant validity (Spearman p) has been found to be better between the FACT and the QABF (0.80) than between the FAST and the FACT (0.50) or the FAST and the QABF (0.51) (Zaja 2011).

8.3.1.3.2 Motivation Assessment Scale (MAS)

The MAS is a 16-item instrument completed by unpaid and paid carers or teachers. It is designed to provide information about the function of the target behaviour of children and adults with a learning disability. Each item refers to one of 4 potential functions, with each item rated on a 7-point Likert scale. The MAS is supposed to reveal whether the target behaviour is related to sensory, escape, attention, or tangible variables. The instrument takes approximately 10 minutes to score and is free.

Internal consistency has been found to range from questionable to good for the sensory subscale (α=0.67-0.83), questionable to good for escape (α=0.68-0.88), questionable to excellent for attention items (α=0.69-0.96) and good to excellent for tangible items (α=0.80-0.91) (Bihm 1991; Duker 1998; Koritsas 2013; Newton 1991; Shogren 2003; Spreat 1996). There have been mixed findings concerning inter-rater reliability with levels of agreement ranging from poor to almost perfect. However, the majority of studies report poor agreement (Akande 1998; Crawford 1992; Duker 1998; Durand 1988; Kearney 1994; Koritsas 2013; Newton 1991; Shogren 2003; Sigafoos 1994; Spreat 1996; Thompson 1995; Zarcone 1991). The MAS correlates with functionally analogous scales of the QABF, offering evidence of convergent validity (Koritsas 2013; Paclawskyj 2001; Shogren 2003). There have been mixed findings about the factor structure of the MAS. Several studies have failed to replicate the original factor structure of the MAS (Duker 1998; Kearney 2006; Joosten 2008; Koritsas 2013) and others have offered support for the structure in institutional but not school samples (Bihm 1991; Singh 1993). Durand 1988 found that teacher's ratings on the MAS predicted their student's behaviour in experimental conditions.

8.3.1.3.3 Questions About behavioural Function (QABF)

The QABF is a 25-item report completed by unpaid and paid carers. It is designed to identify behavioural functions which are important in maintaining aberrant behaviour in children and adults. The 5 subscales of the assessment relate to 5 possible variables influencing problem behaviour: attention, escape from task demands or social contact, non-social reinforcement, physical discomfort, and tangible reinforcement. The instrument is available to use for free.

Internal consistency has been found to be generally acceptable to excellent for all subscales (Koritsas 2013; Nicholson 2006; Paclawskyj 2000; Shogren 2003; Zaja 2011). Although Paclawskyj 2000 found that it was questionable for the test as a whole (α=0.60). Inter-rater reliability for subscales has been found to range from poor to almost perfect (kappa=0.21-0.95) (Koritsas 2013; Matson 2007c; Matson 2009; Nicholson 2006; Paclawskyj 2000; Shogren 2003; Zaja 2011). Scores have been found to be stable over time indicating good test-retest reliability (Paclawskyj 2000; Zaja 2011). The Motivation Assessment Scale (MAS) and Functional Assessment for Multiple Causality (FACT) have been found to correlate with functionally analogous scales of the QABF, offering evidence of convergent validity (Koritsas 2013; Paclawskyj 2001; Shogren 2003; Zaja 2011). Watkins 2013 also demonstrated that the QABF identified the same behavioural functions in participants when compared with a brief functional analysis. Participants with treatments developed from functional assessment (QABF results) have been found to improve significantly when compared with controls receiving standard treatments not based on functional analysis (Matson 1999b). Paclawskyj 2000 replicated the original 5-factor solution. Nicholson 2006 also found 5 factors that corresponded to the 5 subscales of the QABF, however their analysis suggested the existence of a sixth factor with a high loading from only a single item, concerning the repetitive nature of the behaviour. The proposed explanation for this was that respondents differentiated repetitiveness of behaviour from aspects suggesting sensory or other automatic reinforcement.

8.2.2 Health economic evidence

No studies assessing the cost effectiveness of methods and tools for the assessment of behaviour that challenges in people with a learning disability were identified by the systematic search of the literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

8.2.3 Clinical evidence statements

- For the ABC instrument, there was evidence from 16 studies demonstrating adequate reliability and validity, although evidence for inter-rater and criterion validity were not available.
- For the BPI-01 instrument, there was evidence from 8 studies demonstrating adequate reliability and validity, although evidence for criterion validity was not available.
- For the BPI-S, there was evidence from 2 studies demonstrating adequate internal consistency and validity, although evidence for inter-rater reliability, test-retest reliability and criterion validity was not available.
- For the CBI, there was evidence from 1 study demonstrating adequate reliability and validity, although evidence for internal consistency and criterion validity was not available.
- For the DBC-A there was evidence from 2 studies demonstrating adequate reliability and validity, although evidence for internal consistency and criterion validity was not available.
- For the DBC-P there was evidence from 3 studies demonstrating adequate reliability and validity.
- For the NCBRF there was evidence from 4 studies demonstrating adequate test-rest reliability, internal consistency and convergent validity, however inter-rater reliability was poor, structural validity was unclear and criterion validity was not available.
- For the SDQ there was evidence from 1 study demonstrating adequate internal consistency and criterion validity, however inter-rater reliability was poor and test-retest reliability and structural validity were not available.

- For the MOAS there was evidence from 1 study indicating adequate reliability, although evidence for test-retest reliability, internal consistency and validity was not available.
- For the FAST there was evidence from 1 study demonstrating adequate reliability, however internal consistency was poor and construct validity was mixed. Criterion validity was not available.
- For the MAS there was evidence from 17 studies demonstrating adequate internal consistency and convergent validity, however test-rest reliability was mixed and there was no evidence for inter-rater reliability and criterion validity.
- For the QABF there was evidence from 10 studies demonstrating adequate reliability and construct validity, however inter-rater reliability was mixed and criterion validity was not available.

8.3.4 Economic evidence statements

No evidence on the cost effectiveness of methods and tools for the assessment of behaviour that challenges in people with a learning disability is available.

The recommendations which were developed from this section and the link to the evidence are at the end of the chapter where they are brought together with the reviews of other instruments. This was because GDG consider it most appropriate to develop and integrated approach to assessment.

8.4 Review question: In carers of people with a learning disability and behaviour that challenges, what is the utility of methods used to assess and monitor their capacity to support the person?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 56. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 56: Clinical review protocol summary for the review of the utility of methods and tools used to assess and monitor carers' capacity to support the person

Component	Description
Review question	In carers of people with a learning disability and behaviour that challenges, what is the utility of methods used to assess and monitor their capacity to support the person? (RQ2.3) To answer this question, consideration should be given to the: • identification of appropriate carers • assessment of carers skills and capacity
Population	Carers of people (children, young people and adults) with a learning disability and behaviour that challenges. The term 'carers' encompasses both family carers and paid carers.
Intervention(s)	Methods used to assess and monitor family carers and paid carers capacity to support the person with a learning disability and behaviour that challenges
Comparison	N/A
Critical outcomes	Clinical utility (including sensitivity and specificity, reliability and reliability)
Study design	Any

8.4.1 Clinical evidence

The search for evidence (supplemented by GDG advice) identified 8 studies that met the eligibility criteria for this review: Chao 2011 (Chao et al., 2011), Friedrich 1983 (Friedrich et al., 1983), Hastings 2004 (Hastings et al., 2004), Hatton 1995a (Hatton et al., 1995a), Hatton 1995b (Hatton & Emerson, 1995b), Honey 2005 (Honey et al., 2005), Knussen 1992 (Knussen et al., 1992), Scott 1989 (Scott et al., 1989).

No studies provided data for the critical outcomes of sensitivity and specificity. Data for reliability and validity were reported for the following assessment instruments:

- Maslach Burnout Inventory (MBI)
- Shortened Ways of Coping (Revised) Questionnaire
- Ways of Coping Questionnaire - Revised
- Questionnaire on Resources and Stress (QRS-F)

For ease of presentation, the evidence is organised by instrument and grouped within the following domains: carer burnout, carer needs and carer stress. Further details about the characteristics and psychometric properties of each instrument can be found in Appendix L.

8.4.1.1 Carer burnout

8.4.1.1.1 Maslach Burnout Inventory (MBI)

The MBI is a self-report instrument with 22 items which has been developed to assess burnout in professional paid carer's. The licence to conduct 50 and 500 paper and pencil administrations costs £59.59 and £214.51 respectively. The licence to use the online version for 50 and 500 administrations costs £71.50 and 257.42 respectively. The manual for the MBI costs £23.83.

The MBI has been found to have acceptable to good internal consistency for the emotional exhaustion subscale (α=0.87-0.90) and the personal accomplishment subscale (α=0.76). Internal consistency for the depersonalisation subscale has varied from unacceptable to acceptable (α=0.68-0.71) (Chao 2011, Hastings 2004).

Chao 2001 found that while a 3-factor solution suggested an acceptable fit for the data, a 4-factor solution provided a better fit than the original 3-factor solution. Items on the 3 subscale all had positive loadings greater than 0.40 on the anticipated factors. Of the 22 items, 19 loaded above 0.40 on the appropriate factor and less than 0.40 on the other factors.

8.4.1.2 Carer Needs

8.4.1.2.1 Shortened Ways of Coping Questionnaire - Revised (SWC-R)

The SWC-R a 14-item self-report questionnaire for adults to represent thoughts and actions used to deal with the demands of a stressful encounter. The measure is scored on 2 subscales which represent distinct ways of coping: practical coping and wishful thinking.

Internal consistency for the SWC-R has been found to range from poor to good for the wishful thinking subscale (α=0.52-0.82), and acceptable to good for the practical coping subscale (α=0.70 - 0.80) (Hatton 1995b). Subscale scores were stable over time demonstrating good test-retest reliability: paired t-tests showing no significant differences between measurements over a 16 month period (Hatton 1995b).

A significant association has been found between 1991 Wishful Thinking scores and 1993 distress scores (Hatton 1995b).

8.4.1.2.2 *Ways of Coping Questionnaire – Revised (WC-R)*

The WC-R is a full length version of the SWC-R. It has 66 items and takes approximately 10 minutes to complete. As in the SWC-R, it is used to represent thoughts and actions which can be used to deal with the demands of a stressful encounter. The licence to conduct 50 and 500 paper and pencil administrations costs £59.59 and £214.51 respectively. The licence to use the online version for 50 and 500 administrations costs £71.50 and £257.42 respectively. The WC-R manual costs £23.83.

In a study which included participants with Down's syndrome only, internal consistency was found to be poor for the passive acceptance subscale (α=0.53), questionable for the stoicism subscale (α=0.65), and acceptable for the practical coping, wishful thinking and seeking social support subscales (α=0.77 - 0.90) (Knussen 1992). In Hatton 1995a, 4 out of 5 subscales showed adequate levels of test-retest reliability for mothers (α> 0.6), with only the passive acceptance subscale failing to reach an adequate level. For fathers, all the coping subscales except stoicism showed adequate levels.

In a study which included participants with Down's syndrome only subscales resulting from factor analysis were found to be similar to those reported in earlier studies, with differences attributable to variations of personal and situational variables (Knussen 1992).

8.4.1.3 **Carer stress**

8.4.1.3.1 *Questionnaire on Resources and Stress (QRS-F)*

The QRS-F is a 52-item self-report questionnaire for families and carers, used widely with parents of children with disabilities. It assesses 4 subcomponents of parental perceptions: parent and family problems (stressful aspects of the impact of the child with disability on parents and the wider family), pessimism (parents' pessimistic beliefs about the child's future), child characteristics (features of the child that are associated with increased demands on parents), and physical incapacity (the extent to which the child is able to perform a range of typical activities). The QRS-F is a free instrument.

The 52-item version of the QRS-F has been found to have excellent internal consistency (Kuder-Richardson coefficient=0.89-0.93) (Friedrich 1983, Scott 1989). In Honey 2005, a good level of internal consistency has been found for mothers (KD-20= 0.85) and for both mothers and fathers (KD-20=0.93) of young children with autism, using a 31-item version of the QRS-F derived from factor analysis. Honey and colleagues (2005) also found no significant difference between mothers' (mean = 10.67, SD = 7.08) and fathers' (mean = 9.91, SD =5.95) scores ($t(42)=1.34$, $p=0.19$), suggesting good inter-rater reliability with the 31-item version.

The QRS-F shows significant correlations in the expected direction with the Beck Depression Inventory, Marlowe-Crowne Social Desirability Scale, suggesting good convergent validity (Friedrich 1983). Scott 1989 successfully replicated the 4-factor solution found by Friedrich 1983. Scores have been found to vary reliably with handicapping condition, offering support for criterion validity (Scott 1989).

In a sample of participants with autism only, Honey 2005 did not find a 2- or 3-factor structure that had any resemblance to the existing QRS–F scales. Rather, the majority of the items loaded significantly onto the first factor extracted in most analyses. Adaptation (Judson scale) has been found to be significantly correlated with maternal stress ($r(54) = –0.70$, $p <0.001$) and paternal stress ($r(43) = –0.46$,$p < 0.01$), offering evidence of convergent validity (Honey 2005).

8.4.2 Health economic evidence

No studies assessing the cost effectiveness of methods used to assess and monitor the capacity of carers to support a person with a learning disability and behaviour that challenges were identified by the systematic search of the literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

8.4.3 Clinical evidence statements

- For the MBI there was evidence from 2 studies demonstrating adequate internal consistency and construct validity, however there was no evidence for criterion validity, inter-rater and test-retest reliability.
- For the SWC-R there was evidence from 1 study demonstrating adequate reliability and criterion validity, however there was no evidence for inter-rater reliability construct validity.
- For the WC-R there was evidence from 2 studies demonstrating adequate structural validity, however reliability varied and there was no available evidence for inter-rater reliability and criterion validity.
- For the QRS-F there was evidence from 3 studies demonstrating good reliability and construct validity, although there was no evidence for test-retest reliability and criterion validity.

8.4.4 Economic evidence statements

No evidence on the cost effectiveness of methods used to assess and monitor the capacity of carers to support a person with a learning disability and behaviour that challenges is available.

8.51 Recommendations and link to evidence

8.5.21 The assessment process

Recommendations	
	20. When assessing behaviour that challenges in people with a learning disability, follow a graduated approach (see recommendations 23–**Error! Reference source not found.**). Aim to gain a functional understanding of why the behaviour occurs and develop a behaviour support plan (see recommendation 32) as soon as possible. 21. When assessing behaviour that challenges ensure that: • the person and their family members or carers are engaged in the assessment process • the complexity and duration of the assessment is proportionate to the severity, impact, frequency and duration of the behaviour • everyone involved in delivering an assessment understands the criteria for moving to more complex and intensive assessment • the person being assessed remains at the centre of concern and is supported throughout the process • all individual and environmental factors that may lead to behaviour that challenges are taken into account • assessment is a flexible rather than fixed process, because factors that trigger and maintain behaviour may change over time • assessments are repeated after any change in behaviour • assessment is outcome focused • the resilience and resources of family members and carers are assessed • the capacity, sustainability and commitment of the staff delivering the behaviour support plan (see recommendation 32) are assessed. 22. Explain how the person and their family members or carers will be told about the outcome of any assessment of behaviour that challenges. Ensure that feedback is personalised and involves a family member, carer or advocate to support the person and help them to understand the feedback if needed.

8.5.23 Initial assessment of behaviour that challenges

Recommendations	23. If behaviour that challenges is emerging or apparent, or a family member, carer or member of staff, including a teacher, has concerns about behaviour, carry out an initial assessment that includes: • a description of the behaviour (including its severity, frequency, duration and impact on the person and others) from the person (if possible) and a family member, carer or a member of staff, including a teacher • an explanation of the individual and environmental factors involved in developing or maintaining the behaviour from the person (if possible) and a family member, carer or a member of staff, including a teacher • the role of the service, staff or family in developing or maintaining the behaviour. Consider using a formal rating scale (for example, the Aberrant Behaviour Checklist) to provide baseline levels for the behaviour and a scale (such as the Functional Analysis Screening Tool) to understand its function. 24. As part of initial assessment of behaviour that challenges, take into account: • developmental history • any previous interventions for behaviour that challenges • social and interpersonal history, including relationships with family members, carers or staff, including teachers • the person's abilities and needs (in particular, their expressive and receptive communication) • recent life events • any physical or mental health problems, and the effect of prescribed and other medication • the person's sensory sensitivities, preferences and needs • the physical environment, including heat, light, noise and smell • the care environment, including the range of activities available, how it engages people and promotes choice, and how well organised it is. 25. After initial assessment, develop a written statement (formulation) that sets out an understanding of what has led to the behaviour that challenges, the function of the behaviour and what maintains it. Use this to develop a behaviour support plan (see recommendation 32).

8.5.31 Risk assessment

Recommendations	
	26. Assess the following risks during any assessment of behaviour that challenges: • self-harm (in particular in people with depression) and self-injury • harm to others • self-neglect • breakdown of family or residential support • exploitation or abuse by others • rapid escalation of the behaviour that challenges or level of risk. Ensure that the behaviour support plan includes risk management (see recommendation 32).

8.5.41 Further assessment of behaviour that challenges

Recommendations	
	27. If the behaviour that challenges is severe or complex, or does not respond to the behaviour support plan, review the plan and carry out a further assessment, integrated with an assessment of need. Carry out a functional assessment (see recommendations 29–31) and identify and evaluate any factors that may provoke or maintain the behaviour. Consider including the following in the further assessment: • any physical health problems • the social environment (including contact and relationships with friends, family members, carers and staff, including teachers) • the physical environment, including sensory needs and any restrictions imposed by the environment • any coexisting mental health problems • response to previous or current treatment for a mental or physical health problem or intervention for behaviour that challenges, including side effects of medication • receptive and expressive communication problems • life history, including any history of trauma or abuse • current functioning at home, in education or in the care environment • neurodevelopmental problems (including the severity of the learning disability and the presence of autism or other behavioural phenotypes) • sensory abnormalities or sensitivities (for example, to heat, light, noise, smell or touch) • changes to routine or personal circumstances. Consider using formal (for example, the Adaptive Behaviour Scale or the Aberrant Behaviour Checklist) and idiographic (personalised) measures to assess the severity of the behaviour and the progress of any intervention. 28. After further assessment, develop a written statement (formulation) that sets out an understanding of what has led to the behaviour that challenges and what maintains it. Use this with the functional assessment of behaviour to develop a behaviour support plan (see recommendation 32).

8.5.52 Functional assessment of behaviour

Recommendations	29. Carry out a functional assessment of the behaviour that challenges to help inform decisions about interventions. This should include: • a clear description of the behaviour, including classes or sequences of behaviours that typically occur together • identifying the events, times and situations that predict when the behaviour will and will not occur across the full range of the person's daily routines and usual environments • identifying the consequences (or reinforcers) that maintain the behaviour (that is, the function or purpose that the behaviour serves) • developing summary statements or hypotheses that describe the relationships between personal and environmental triggers, the behaviour and its reinforcers • collecting direct observational data to inform the summary statements or hypotheses. 30. Include the following in all functional assessments: • a baseline measure of current behaviour, and its frequency and intensity, and repeated measurements in order to evaluate change • measures taken using direct observations and scales such as the Aberrant Behaviour Checklist and self-reporting • a baseline measure of quality of life (such as the Life Experiences Checklist and the Quality of Life Questionnaire) • assessment of the impact of current or past interventions, including reactive strategies.

Recommendations	
	31. Vary the complexity and intensity of the functional assessment according to the complexity and intensity of behaviour that challenges, following a graduated approach as set out below. • For recent-onset behaviour that challenges, consider brief structured assessments such as the Functional Analysis Screening Tool or Motivation Assessment Scale to identify relationships between the behaviour and what triggers and reinforces it. • Carry out pre-assessment data gathering to help shape the focus and level of the assessment. • For recent-onset behaviour that challenges, or marked changes in patterns of existing behaviours, take into account whether any significant alterations to the person's environment and physical or psychological health are associated with the development or maintenance of the behaviour. • Consider in-depth assessment involving interviews with family members, carers and others, direct observations, structured record keeping, questionnaires and reviews of case records. • If a mental health problem may underlie behaviour that challenges, consider initial screening using assessment scales such as the Diagnostic Assessment Schedule for the Severely Handicapped-II, Psychiatric Assessment Schedule for Adults with a Developmental Disability or the Psychopathology Instrument for Mentally Retarded Adults and seek expert opinion. • If the behaviour poses a risk to the person or others, carry out a risk assessment (see recommendation 26).

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8.5.61 Behaviour support plan

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Recommendations	
	32. If the behaviour that challenges continues after assessment, develop a behaviour support plan based on a shared understanding about the function of the behaviour and what maintains it. This should: • identify proactive strategies designed to stop the conditions likely to promote behaviour that challenges, including changing the environment (for example, reducing noise, increasing predictability) and promoting active engagement through structured and personalised daily activities, including the school curriculum for children and young people • identify adaptations to a person's environment and routine, and strategies to help them develop an alternative behaviour to achieve the function of the behaviour that challenges by developing a new skill (for example, improved communication, emotional regulation or social interaction) • identify secondary prevention strategies to calm the person when they begin to show early signs of distress, including: o individual relaxation techniques o distraction and diversion onto activities they find enjoyable and rewarding • identify reactive strategies to manage any behaviours that are not preventable (see section 13.3), including how family members, carers or staff should respond if a person's agitation escalates and there is a significant risk of harm to them or others • incorporate risk management and take into account the effect of the behaviour support plan on the level of risk • be compatible with the abilities and resources of the person's family members, carers or staff, including managing risk, and can be implemented within these resources • be monitored using data collection and reviewed regularly • identify any training for family members, carers or staff to improve their understanding of behaviour that challenges in people with a learning disability.

8.5.73 Interventions for coexisting health problems

Recommendations	33. Offer people with a learning disability and behaviour that challenges interventions for any coexisting mental or physical health problems in line with the relevant NICE guideline for that condition. Adjust the nature, content and delivery of the interventions to take into account the impact of the person's learning disability and behaviour that challenges.

8.5.81 Link to evidence across all topics

2

Relative values of different outcomes	The GDG decided that clinical utility (including the key components of assessment, sensitivity and specificity, reliability and reliability) was the critical outcome.
Trade-off between clinical benefits and harms	The GDG decided to adopt a graduated approach to assessment. This was because, in their expert opinion and experience, in a number of circumstances only limited assessment was necessary. The GDG recognised that while this is less intrusive and less consuming of resources, it does increase the risk that more complex factors contributing to the behavioural problem may not be identified.
Trade-off between net health benefits and resource use	Effective assessment and monitoring of carers' capacity in supporting people with a learning disability and behaviour that challenges has important clinical and resource implications for the carers, in terms of intervention costs and the carers' coping and HRQoL; it has also important clinical and resource implications for people with a learning disability, as it enables carers to assess and monitor them most effectively, which, in turn, contributes to the effective and cost-effective anticipation and management of behaviour that challenges. It is therefore likely that costs of assessment and monitoring may be offset, at least partially, by savings associated with earlier and more effective management of behaviour that challenges.
Quality of evidence	There was very limited evidence on the structure and content of assessment. There was moderate to low quality evidence on the psychometric properties of a number of measures reviewed.
Other considerations	In absence of evidence on the structure, content and validity of the assessment process, the GDG used informal consensus methods to arrive at the recommendations related to this topic in this chapter. The GDG also drew on the evidence in the chapter on experience of care (which provided evidence of service users' and carers' experience of the assessment process) and the chapter on psychosocial interventions, which identified functional assessment as a moderator of treatment effectiveness. The GDG decided first that a graduated approach to assessment was needed to balance the burden of assessment with the need to understand the drivers behind any behavioural problem. They judged that this should start with an initial assessment, including a risk assessment, followed by further assessment if the behaviour is severe or complex, or has not responded to the behaviour support plan. To ensure that the assessment is fully informed and that any plan that emerged has full service user and carer involvement, the GDG judged that both service users and carers should be fully involved in all stages of the assessment. The evidence drawn from the chapter on psychosocial interventions that functional assessment is an important moderator of a good outcome led the GDG to recommend this as an integral part of a further assessment. Formal rating scales (for which there was evidence for their reliability and validity – including behaviour that challenges, mental state and quality of life) were also considered to be of use in informing the assessment and providing reliable data on the impact of any interventions. The GDG were aware that any assessment or intervention

	that focused on behaviour that challenges could increase risk and so recommended that a risk assessment be an integral part of any assessment. The GDG were also aware of the reactive nature of many interventions for behaviour that challenges and decided that wherever possible all interventions should be contained within a behavioural support plan, which emphasises proactive as well as reactive strategies. Finally, where the assessment indicated a coexisting mental or physical health problem, the GDG agreed that it would be good practice to offer an appropriate intervention in line with relevant NICE guidance, but the nature, content and delivery should be adjusted to take account of the impact the person's learning disability and behaviour that challenges.

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9 Interventions aimed at preventing behaviour that challenges

9.1 Introduction

Behaviour that challenges has serious implications for people with a learning disability and their family and carers. For the former, these include social exclusion, institutionalisation, deprivation, physical harm, abuse, misdiagnosis, exposure to ineffective or aversive interventions, and failure to access evidence-based interventions (Baker & Allen, 2001; Emerson, 2001; Guess et al., 1987; Lowe et al., 2005; Rusch et al., 1986; White et al., 1995). Children with severe behaviour that challenges are at risk of placement in 52-week residential schools (Pilling et al., 2007) and adults in out-of-area assessment and treatment facilities (Health and Social Care Information Centre, 2014). For families and carers, these implications may include elevated risks of physical and mental ill health, physical injury, increased financial burdens, and reduced quality of life (Allen et al., 2006; Qureshi, 1994). Given that behaviour that challenges may first appear in childhood (Einfeld et al., 2007; Murphy et al., 1999) and, in the absence of appropriate intervention, often seems to be enduring (Einfeld et al., 2006; Kiernan & Alborz, 1996), significant care costs may be incurred over protracted periods of time for some people. For example, in the early 1990s, the National Institutes for Health (1991) estimated that 200,000 individuals with developmental disabilities in the United States displayed significant degrees of destructive behaviour at an annual cost to care services exceeding US $3 billion. Annual individual service costs of between £100-450,000 have recently been identified in the UK (Emerson & Robertson, 2008; Lowe et al., 2007a).

Conditions that have a similar impact within the general population (for example, coronary heart disease and smoking-related illnesses) are typically subject to high-profile public health interventions whose focus is prevention. In contrast, behavioural and emotional difficulties in people with a learning disability are often only addressed when they have become fully established in a person's behavioural repertoire, present for many years, and therefore likely to be more resistant to effective intervention.

People with a learning disability will, in general, experience high levels of exposure to many of the known risk factors for emotional and behavioural difficulties. For example, Emerson and Hatton (2007) showed that cumulative risk of exposure to a variety of indicators of social disadvantage (lone parent family, income poverty, exposure to 2 or more negative life events, poor family functioning, primary carer with no educational qualifications, potential maternal mental health issues, and poor maternal self-rating of health) were associated with increased prevalence of emotional disorders, conduct disorders and hyperactivity in children. While this was true for those with and without a learning disability, the former were at significantly greater risk of exposure to all the variables studied. People with a learning disability are also at significant risk of experiencing social isolation (McVilly et al., 2006; Stancliffe et al., 2007), being unemployed (Martorelli et al., 2008) and being supported in settings where there are low levels of activity and stimulation (Mansell et al., 2003). While they are at increased risk of experiencing a wide variety of general health problems, the treatment that they receive for these problems often falls below optimal levels (Scheepers et al., 2005). Some service settings will themselves have characteristics that serve to promote and encourage the development and maintenance of behaviour that challenges (McGill et al., 2003) and fail to offer or provide evidence-based interventions for behaviour that challenges when it develops.

9.2 Review question: In people with a learning disability, what are the benefits and potential harms of interventions aimed

at preventing the development of behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 57. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 57: Clinical review protocol summary for the review of interventions (including early intervention) aimed at preventing the development of behaviour that challenges

Component	Description
Review question	In people with a learning disability, what are the benefits and potential harms of interventions (including early intervention) aimed at preventing the development of behaviour that challenges? (RQ3.1)
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability.
Intervention(s)	Psychosocial, pharmacological, environmental and complex interventions (for example, combined psychological and pharmacological interventions)
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Behaviour that challenges • Adaptive functioning, including communication skills • Quality of life • Service user and carer satisfaction
Study design	RCTs and systematic reviews.
Note. RCTs = Randomised controlled trials	

9.2.1 Clinical evidence

9.2.1.1 Educational intervention versus any control

One RCT (N = 294) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Strain 2011 (Strain & Bovey, 2011). An overview of the included trial can be found in Table 58.

Summary of findings can be found in the Table 59. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 58: Study information table for trials included in the meta-analysis of preventative interventions versus any control

	Educational intervention versus any control	Home-based versus centre-based early behavioural intervention
Total no. of studies (N[1])	1 (294)	1 (67)
Study ID	Strain 2011	Roberts 2011
Country	USA	Australia
Diagnosis	ASD	ASD
Age (mean)	4	4
Sex (% Female)	Not reported	10
Ethnicity (% White)	Not reported	Not reported
IQ (mean)	Not reported	62
Treatment length (weeks)	104	40
Intervention	Learning Experiences and Alternative Program for Pre-schoolers and their Parents (LEAP) - Full replication.	Home-based EBI 'Building Blocks' programme.
Comparison	Attention control/LEAP intervention manual-only control	Centre-based EBI 'Building Blocks' programme.

Notes: N = total number of participants; ASD = autism spectrum disorder; EBI = early behavioural intervention

[1] Number randomised

Table 59: Summary of findings table for educational intervention compared with any control

Outcomes	Comparative risks (95% CI) Control	Educational intervention	No of Participants (studies)	Quality of the evidence (GRADE)
Behaviour that challenges (severity) - post-treatment Change score[1]	–	The mean behaviour that challenges (severity) - post-treatment in the intervention groups was **0.19 standard deviations lower** (0.42 lower to 0.04 higher)	294 (1 study)	**very low**[2,3,4]
Adaptive functioning (social) - post-treatment	–	The mean adaptive functioning (social) - post-treatment in the intervention groups was **0.76 standard deviations higher** (0.52 to 1 higher)	294 (1 study)	**very low**[2,3,4]
Adaptive functioning (communication) - post-treatment	–	The mean adaptive functioning (communication) - post-treatment in the intervention groups was **0.94 standard deviations higher** (0.7 to 1.19 higher)	294 (1 study)	**very low**[2,3,4]

[1] Due to significant baseline differences, standard deviation of change and estimates of mean change were derived using initial and final mean values and utilising r = 0.5. Sensitivity analyses were used to explore the impact of altering assumptions about the calculation of the effect size, but this resulted in no change to conclusions.
[2] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[3] Applicability concerns: autism population; no information reported concerning learning disability
[4] Optimal information size not met

9.2.1.21 Home-based early behavioural intervention versus centre-based early behavioural intervention

One RCT (N = 67) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Roberts 2011 (Roberts et al., 2011). An overview of the included trial can be found in Table 58.

Summary of findings can be found in the Table 60. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 60: Summary of findings table for home-based early behavioural intervention compared with centre-based early behavioural intervention

Outcomes	Comparative risks (95% CI)		No of Participants (studies)	Quality of the evidence (GRADE)
	Centre-based early behavioural intervention	Home-based early behavioural intervention		
Behaviour that challenges (severity) - post-treatment	–	The mean behaviour that challenges (severity) - post-treatment in the intervention groups was **0.11 standard deviations lower** (0.7 lower to 0.48 higher)	44 (1 study)	**very low**[1,2]
Adaptive functioning (social) - post-treatment	–	The mean adaptive functioning (social) - post-treatment in the intervention groups was **0.63 standard deviations lower** (1.17 to 0.09 lower)	56 (1 study)	**very low**[1,2]
Adaptive functioning (communication) - post-treatment	–	The mean adaptive functioning (communication) - post-treatment in the intervention groups was **0.46 standard deviations lower** (1 lower to 0.07 higher)	55 (1 study)	**very low**[1,2]

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

9.2.1.37 Early intensive behavioural intervention (EIBI) versus parent delivered Lovas intervention

One RCT (N = 28) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Smith 2000 (Smith et al., 2000). An overview of the included trial can be found in Table 61.

Summary of findings can be found in the Table 62. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 61: Study information table for trials included in the meta-analysis of preventative interventions versus any control

	EIBI versus parent delivered Lovas intervention	High supervision EIBI (clinic-directed) versus low supervision EIBI (parent-directed)
Total no. of studies (N[1])	1 (28)	1 (24)
Study ID	Smith 2000	Sallows 2005
Country	USA	USA
Diagnosis	ASD	ASD
Age (mean)	3	3
Sex (% Female)	18	21
Ethnicity (% White)	50	Not reported
IQ (mean)	51	51
Treatment length (weeks)	Early intensive behavioural intervention = 145 Parent delivered Lovas interventions = 13 to 39	209
Intervention	Early intensive behavioural intervention	Clinic-directed early intensive behavioural treatment
Comparison	Parent delivered Lovas interventions	Parent-directed early intensive behavioural treatment

Notes: N = total number of participants; ASD = autism spectrum disorder
[1] Number randomised

Table 62: Summary of findings tables for EIBI versus parent delivered Lovas intervention

Outcomes	Comparative risks* (95% CI)		No of Participants (studies)	Quality of the evidence (GRADE)
	Parent intervention	Early intensive behavioural intervention		
Behaviour that challenges (severity) - post-treatment Parent-rated	–	The mean behaviour that challenges (severity) - post-treatment in the intervention groups was **0.36 standard deviations lower** (1.1 lower to 0.39 higher)	28 (1 study)	**very low**[1,2]
Behaviour that challenges (severity) - post-treatment Teacher-report	–	The mean behaviour that challenges (severity) - post-treatment in the intervention groups was **0.47 standard deviations higher** (0.28 lower to 1.23 higher)	28 (1 study)	**very low**[1,2]
Adaptive functioning (communication) - post-treatment	–	The mean adaptive functioning (communication) - post-treatment in the intervention groups was **0.63 standard deviations higher** (0.13 lower to 1.39 higher)	28 (1 study)	**very low**[1,2]
Adaptive functioning (global) - post-treatment	–	The mean adaptive functioning (global) - post-treatment in the intervention groups was **0.11 standard deviations higher** (0.64 lower to 0.85 higher)	28 (1 study)	**very low**[1,2]

[1] Applicability concerns: autism population; no information reported concerning learning disability

[2] Optimal information size not met; small, single study

9.2.1.41 High supervision EIBI (clinic-directed) versus low-supervision EIBI (parent-directed)

2 One RCT (N = 24) met the eligibility criteria for this review and included sufficient data to be
3 included in the evidence syntheses: Sallows 2005 (Sallows & Graupner, 2005). An overview
4 of the included trial can be found in Table 61.

5 Summary of findings can be found in the Table 63. The full GRADE evidence profiles and
6 associated forest plots can be found in Appendices O and P.

7 No evidence was identified in relation to the specific subgroups identified in the review
8 protocol.

9 No data were available for the critical outcomes of behaviour that challenges, quality of life or
10 service user and carer satisfaction.

11 The study flow diagram and evidence tables (including methodology checklists) can be found
12 in Appendix N, and exclusion list in Appendix Q.

13 **Table 63: Summary of findings table for clinic-directed EIBI versus parent-directed**
14 **EIBI**

Outcomes	Comparative risks* (95% CI)		No of Participants (studies)	Quality of the evidence (GRADE)
	Low supervision EIBI (parent-directed)	High supervision EIBI (clinic-directed)		
Adaptive functioning (communication) -post-treatment	–	The mean adaptive functioning - communication; post-treatment in the intervention groups was **0.25 standard deviations lower** (1.08 lower to 0.57 higher)	23 (1 study)	**very low**[1,2,3]

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability concerns: autism population; no information reported concerning learning disability
[3] Optimal information size not met; small, single study

9.2.1.5 15 Parent education, support and skills training versus any control

16 Two RCTs (N = 170) met the eligibility criteria for this review and included sufficient data to
17 be included in the evidence syntheses: Rickards 2007 (Rickards et al., 2007), Tonge 2006
18 (Tonge et al., 2006). Tonge 2006 was a 3-arm study; for the purposes of this review, the
19 parent education and behaviour management intervention (PEBM) arm was compared with
20 the parent education and counselling (PEC) arm (N = 70). An overview of the trials included
21 in the meta-analysis can be found in Table 66. Unlike the parent training interventions
22 reviewed in Chapter 9, which focused specifically on reducing child's targeted behaviour that
23 challenges, these interventions focused on parental mental health and on the global needs
24 the child (in both populations all children had autism and a learning disability).

25 Summary of findings can be found in Table 65. The full GRADE evidence profiles and
26 associated forest plots can be found in Appendix O.

27 No evidence was identified in relation to the specific subgroups identified in the review
28 protocol.

29 No data were available for the critical outcomes of quality of life or service user and carer
30 satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 64: Study information table for trials included in the meta-analysis of parent education, support and skills training versus any control

	Parent training versus any control
Total no. of studies (N[1])	2 (135)
Study ID	(1) Rickards 2007[2] (2) Tonge 2006
Country	Australia
Diagnosis	ASD
Age (mean)	4
Sex (% Female)	(1) 20 (2) 16
Ethnicity (% White)	Not reported
IQ (mean)	(1) 60 (2) 59
Treatment length (weeks)	(1) 40 (2) 20
Intervention	(1) Parent education, support and skills training (+ early intervention centre programme) (2) Parent education and behaviour management training
Comparison	(1) TAU/early intervention centre programme only (2) Attention control/parent education and counselling
Notes: N = total number of participants; TAU = Treatment as usual; ASD = autism spectrum disorder [1]Number randomised. [2]Three armed trial; parent education and behaviour management intervention (PEBM) and parent education and counselling (PEC) utilised.	

Table 65: Summary of findings table for parent education, support and skills training versus any control

Outcomes	Comparative risks* (95% CI) Control	 Parent education, support and skills training	No of Participants (studies)	Quality of the evidence (GRADE)
Behaviour that challenges (severity) - post-treatment		The mean behaviour that challenges (severity) - post-treatment in the intervention groups was **0.4 standard deviations lower** (0.93 lower to 0.12 higher)	57 (1 study)	**low**[1]
Behaviour that challenges (severity) - follow up Follow-up: 26 to 52 weeks		The mean behaviour that challenges (severity) - follow up in the intervention groups was **0.37 standard deviations lower** (0.79 lower to 0.05 higher)	117 (2 studies)	**low**[2,3]
Adaptive functioning (global) - post-treatment		The mean adaptive functioning (global) - post-treatment in the intervention groups was **0.25 standard deviations higher** (0.27 lower to 0.77 higher)	58 (1 study)	**low**[1]
Adaptive functioning (global) - follow-up Follow-up: 26 to 52 weeks		The mean adaptive functioning (global) - follow-up in the intervention groups was **0.52 standard deviations higher** (0.15 to 0.88 higher)	119 (2 studies)	**low**[2,3]
Adaptive functioning (communication) - follow-up Follow-up: mean 26 weeks		The mean adaptive functioning (communication) - follow-up in the intervention groups was **0.75 standard deviations higher** (0.26 to 1.25 higher)	68 (1 study)	**low**[1]

[1] Optimal information size not met; small, single study
[2] Most information is from studies at moderate risk of bias

[3] Optimal information size not met

9.2.2 Economic evidence

The systematic search of the economic literature did not identify any evidence on the cost effectiveness of interventions exclusively aimed at the prevention of behaviour that challenged in people with a learning disability. However, 4 studies were identified that assessed the cost effectiveness of early intensive behavioural intervention (EIBI) focusing on impairments in adaptive behaviour in children and young people with autism (Chasson et al., 2007; Jacobson, 1998; Motiwala et al., 2006; Peters-Scheffer et al., 2012). Three studies were conducted in the US (Chasson et al., 2007; Jacobson, 1998; Motiwala et al., 2006) and the other one was carried out in the Netherlands (Peters-Scheffer et al., 2012). All studies were based on decision-economic modelling. Details on the methods used for the systematic review of the economic literature are described in Chapter 3; full references to the included studies and evidence tables for all economic evaluations included in the systematic literature review are provided in Appendix S. Completed methodology checklists of the studies are provided in Appendix R. Economic evidence profiles of studies considered during guideline development (that is, studies that fully or partly met the applicability and quality criteria) are presented in Appendix T.

Chasson and colleagues (2007) estimated the net cost-savings associated with provision of EIBI to children with autism aged 4 years, resulting exclusively from improvement in children's functioning and subsequent reduction in need for special education. The study was conducted in the US (Texas) and considered only intervention costs and costs of special education (including state-budgeted, local, federal, and private); regular education costs were omitted from the analysis, as these are standard baseline costs. The time horizon of the analysis was 18 years (from 4 to 22 years of age). Resource use and cost data were based on local (state) data, personal communication and further assumptions. Estimates of clinical effectiveness were based on a non-systematic review of published studies and further assumptions made by the authors. According to these estimates, without EIBI provision all children with autism require special education for 18 years, while when they receive 3 years of EIBI only 28% of the children require special education and the remaining children can attend exclusively mainstream, regular education. The total special education cost per child with autism not receiving EIBI was $360,000 (without EIBI 100% of children receive special education), while the mean total cost per child with autism following provision of EIBI was $151,500, consisting of the intervention cost of EIBI and the special education cost for 28% of children still requiring special education. EIBI was therefore associated with a total net cost-saving of $208,500 per child (cost year not reported but it was likely 2004; no discounting was undertaken). When this figure was applied to a conservative estimate of 10,000 children with autism in Texas, it was estimated that provision of EIBI would result in a total net saving to the State of $2.09 billion.

The study is characterised by potentially serious limitations, mainly relating to the selective use of clinical effectiveness data associated with the provision of EIBI which were further modified by authors' assumptions; moreover, the study was carried out in the US and its findings are therefore only partially applicable to the UK context.

Jacobson (1998) reported the wider total net savings associated with provision of EIBI in preschool children with autism or pervasive developmental disorder. The study was conducted in the US (Pennsylvania) and adopted a societal perspective. The authors estimated the net incremental cost of EIBI per person with autism from the age of 3 years (mean age of provision of EIBI) and up to 55 years of age. Costs were estimated for children with normal functioning following EIBI, children experiencing a partial effect of EIBI, and children where EIBI had a minimal effect. Clinical efficacy parameters were based on data derived from a non-systematic review of published literature. The authors reported overall net

savings assuming different levels of EIBI effectiveness, which was expressed as the percentage of children achieving normal functioning. Net savings ranged from $656,385 for levels of normal functioning reaching 20% to $1,081,984 for levels of normal functioning reaching 50% (1996 prices). These figures were estimated assuming marginal effects, that is, children with normal range effects improved from partial effects, and those with partial effects improved from minimal effects. However, estimation of cost-savings using this methodology is underlined by the unrealistic implicit assumption that the marginal effect of normal functioning is achieved only after provision of EIBI, and that without EIBI no children achieve normal functioning. This assumption, which led to overestimation of cost-savings associated with EIBI, was considered a very serious methodological limitation, and therefore, although the study met inclusion criteria, it was not considered at guideline development.

Motiwala and colleagues (2006) conducted a modelling study to estimate the cost effectiveness of a programme of expansion of 3 years of EIBI to all eligible children with autism, aged 2-5 years, in Ontario, Canada, compared with the standard service in Ontario at the time of the analysis, which consisted of EIBI for 37% of eligible children with autism aged 2-5 years and no intervention for 63% of eligible children with autism aged 2-5 years. Expansion of EIBI was also compared with no intervention. The study adopted a public sector perspective and estimated costs starting from the preschool age and up to the age of 65 years. Costs included the cost of providing EIBI (consisting of therapists' training costs; contractual payments to service providers; salaries, benefits & overheads incurred by provincial civil servants), educational and respite service costs, costs of adult day programmes, accommodation and supported employment. Costs were estimated separately for children with autism and normal functioning, semi-dependent children with autism and very dependent children with autism. The total cost of the 3 alternative strategies was subsequently estimated based on the proportion of children with normal functioning, semi-dependent children and heavily dependent children in each strategy. The measure of outcome was the number of dependency-free years per person. Resource use and unit costs were based on provincial government data; clinical data were based on a non-systematic literature review and further assumptions.

Expansion of EIBI led to a higher number of dependency-free years per child with autism over the time horizon of the analysis (14.0), compared with standard service (11.2) and no intervention (9.6). The overall cost of expansion of EIBI, standard service, and no intervention per child with autism was $960,595, $995,074 and $1,014,315, respectively (2003 Canadian dollars, discounted at an annual rate of 3%), meaning that expansion of EIBI would produce an overall saving of $34,479 per child with autism, compared with standard service, and $53,720 per child with autism, compared with no intervention. By applying this cost-saving to the estimated population of 1,309 children with autism, aged 2-5 years, in Ontario, who at the time of the study received the standard service, the total net saving that would be accrued by expanding EIBI to all eligible children would reach $45,133,011. Results were sensitive to the EIBI efficacy (expressed as the proportion of children that achieved normal functioning following EIBI) and the discount rate used.

The study is characterised by potentially serious limitations relating to the assumptions made at the estimation of the clinical parameters of the economic model; furthermore, as it was conducted from a Canadian public sector perspective, it is only partially applicable to the UK setting.

Peters-Scheffer and colleagues (2012)(conducted a cost analysis to estimate the cost savings associated with provision of EIBI - in addition to treatment as usual (TAU) - to children with autism of preschool age in the Netherlands. The comparator of the analysis was TAU alone. The study adopted a public service perspective and estimated costs starting from the preschool age and up to the age of 65 years. Cost elements included implementation of EIBI (personnel, capital assets, transportation, materials and supplies), speech therapy & physiotherapy, educational services, daytime services, daytime activities and care, social benefits for parents, payments for future adult living expenses, day programs or supported

work and sheltered environment services. Like Motiwala and colleagues (2006), the study estimated costs for children with autism and normal functioning, semi-dependent children with autism and very dependent children with autism, and subsequently estimated costs for EIBI and TAU based on the proportion of children achieving normal functioning, semi-dependent children and heavily dependent children following EIBI and TAU, respectively. Resource use and unit costs were based on national data and further assumptions; clinical data were based on a review of meta-analyses, selection of the reported data according to their applicability to the Dutch setting, and further assumptions.

EIBI and TAU were associated with an overall cost per child with autism up to the age of 65 years of €2,578,746 and €3,681,813, respectively, meaning that EIBI resulted in an overall cost-saving of €1,103,067 (cost year not reported but it was likely 2011; discounting was not applied). The authors reported that if these cost-savings per child were extended to the total number of children with autism born every year in the Netherlands (approximately 1092 to 1820 children), the estimated cost savings would reach €109.2–€182 billion, excluding costs associated with inflation.

The study is characterised by potentially serious limitations relating to the assumptions made at the selection of the data used to populate the economic model, and is only partially applicable to the UK setting since it was undertaken in the Netherlands.

Overall, although the studies included in the systematic literature review suggested that provision of EIBI focusing on impairments in adaptive behaviour in pre-school children with autism may result in important cost-savings, all studies suffered from potentially serious methodological limitations, especially regarding the identification and selective use of clinical effectiveness data, which may have significantly affected the study results and conclusions. Moreover, none of the studies identified in the review were conducted in the UK, and therefore their applicability to the NICE context is limited.

In addition to the economic evidence described above, one of the RCTs included in the guideline systematic review (Roberts 2011) reported the intervention cost per child receiving either home-based or centre-based EIBI, comprising exclusively staff costs as monitored for the trial (Roberts et al., 2011). This cost was estimated at $6383AU (likely in 2007 prices) per child, regardless of which treatment the child received. This corresponds to approximately £3,337 per child in 2013 prices. The authors expressed the view that this is a small cost compared with a range of other interventions currently available to children and families with autism. It needs to be noted that the intervention cost may be different in the UK, due to differences in service organisation and delivery as well as staff unit costs.

9.2.3 Clinical evidence statements

9.2.3.1 Educational intervention versus any control

- Very low quality evidence from a single study (N = 294) suggested that the educational intervention was more effective than control at reducing the severity of behaviour that challenges at end of treatment. However, the precision of this estimate was poor.
- Very low quality evidence from a single study (N = 294) suggested that the educational intervention was more effective than control at increasing both social and communicative adaptive functioning at end of treatment.

9.2.3.2 Home-based early behavioural intervention versus centre-based early behavioural intervention

- Very low quality evidence from a single study (N = 44) was inconclusive as to the effectiveness of home-based when compared with centre-based early behavioural intervention in reducing the severity of behaviour that challenges at the end of treatment.

- Very low quality evidence from a single study (N = 56) suggested that the home-based early behavioural intervention was less effective than the centre-based early behavioural intervention at increasing social and communicative adaptive functioning. However, the precision of the estimate for communicative adaptive functioning was poor.

9.2.3.3 Early intensive behavioural intervention (EIBI) versus parent delivered Lovas intervention

- Very low quality evidence from a single study (N = 28) was inconclusive as to the effectiveness of the early intensive behavioural intervention when compared with parent delivered Lovas interventions in reducing the severity of parent-rated behaviour that challenges at end of treatment.
- Very low quality evidence from a single study (N = 28) suggested that the early intensive behavioural intervention was less effective than parent delivered Lovas intervention reducing the severity of behaviour that challenges at the end of treatment. However, the precision of the estimate was poor.
- Very low quality evidence from a single study (N = 28) suggested that the early intensive behavioural intervention was more effective than parent delivered Lovas intervention in increasing communicative adaptive functioning at the end of treatment. However, the precision of the estimate was poor.
- Very low quality evidence from a single study (N = 28) was inconclusive as to the effectiveness of the early intensive behavioural intervention when compared with delivered Lovas intervention in increasing global adaptive functioning at the end of treatment.

9.2.3.4 High supervision EIBI (clinic-directed) versus low supervision EIBI (parent-directed)

- Very low quality evidence from a single study (N = 23) was inconclusive as to the effectiveness of the clinic-directed when compared with parent-directed early intensive behavioural intervention at increasing communicative adaptive functioning at end of treatment.

9.2.3.5 Parent education, support and skills training versus any control

- Low quality evidence from up to 2 studies (N = 117) suggested that parent education, support and skills training was more effective than control in reducing the severity of behaviour that challenges at the end of treatment and up to 52 weeks follow-up. However, the precision of the estimate was poor.
- Low quality evidence from a single study (N = 58) was inconclusive as to the effectiveness of the parent education, support and skills training when compared with control in improving adaptive functioning at the end of treatment. However, at up to 52-week follow-up, 2 studies (N = 119) suggested that parent education, support and skills training was more effective than control.
- Low quality evidence from a single study (N = 68) suggested that parent education, support and skills training was more effective than control in improving communicative adaptive functioning at 26-week follow-up.

9.2.4 Economic evidence statements

- Low quality evidence from 4 model-based studies suggested that provision of EIBI in pre-school children with autism may result in important cost-savings. However, this evidence is coming from children with autism and thus is not directly relevant to the study population of this guideline. Furthermore, the evidence is characterised by potentially serious methodological limitations. Finally, this evidence is based on US studies and therefore its applicability to the NICE context is limited.

9.2.15 Recommendations and link to evidence

2 See section 9.5 for recommendations and link to evidence relating to this section.

9.3 3 Health awareness interventions

9.3.1 4 Introduction

5 There is an increasing recognition that behaviour that challenges are sometimes the only
6 apparent means of communication available to those with a learning difficulty. This form of
7 communication may represent significant distress about either a physical or a mental health
8 problem.

9 There is ample evidence that people with a learning difficulty have poorer health than their
10 non-disabled peers. It is believed that this represents both difficulty identifying important
11 symptoms and difficulty accessing care (Disability Rights Commission, 2006; Mencap, 2007).

12 There is robust evidence that offering health checks in primary care is effective at identifying
13 previously unidentified morbidity in those with a learning disability (Robertson et al., 2010;
14 Robertson et al., 2011).

15 Extrapolating from this would lead us to believe that an annual health check in primary care
16 can reduce the risk of behaviour that challenges. These checks were introduced into the
17 NHS in the form of a Directed Enhanced Service (DES) in 2009(Michael, 2008) .This
18 incentivises GP practices to offer an annual health check to all adults with a learning
19 difficulty. In 2014 this was extended to include young people from age 14 to 18.

20 The checks are comprehensive and include:

21 • Assessment of feeding, bowel and bladder function
22 • Assessment of behavioural disturbance
23 • Assessment of vision and hearing
24 • Along with a general health review, medication review, and syndrome specific health issue
25 review

26 The Public Health Observatory for learning disability has produced 5 years of reports
27 showing steady progression in the uptake of the annual health check and in health
28 outcomes. However uptake around the country varies considerably with an average of 52%
29 of eligible adults receiving the checks. The range of uptake is from 20% - 80% in different
30 parts of England (Glover et al., 2012).

31 Clearly there remains room for improvement. There is no evidence of harm from the checks,
32 and reports from areas of high uptake indicate considerable benefits in detection of
33 previously unrecognised health need.

34 Additionally there has been interest in facilitating access to both primary and secondary care
35 for those with a learning disability by offering Personal health profiles and Health Action
36 plans that can give important information to care givers. In July 2014 Baroness Angela
37 Browning launched an autism-specific ‘health passport’ in an attempt to improve access for
38 people with autism who are more likely to demonstrate behaviour that challenges in a health
39 environment. The behaviour can be a significant barrier to accessing health care but may
40 represent an unmet health need. Reasonable adjustments to enable access to health care
41 are a requirement of the Equality Act but may not be recognised for those with a learning
42 difficulty.

43 The family and carers of those with a learning difficulty have their own burdens with an
44 increase in mental health problems reported. Carer interventions have been shown to
45 improve depression significantly and to help with anxiety, stress or burnout. The available

evidence only concerns the parents of children with a learning disability but the experience of health professionals working in this field would imply that the needs of carers across the spectrum are significant and that behaviour that challenges are very disruptive to the carers' lives. It causes increased isolation, poor economic status and often physical pain from injuries caused by their dependent. This groups needs are not well met. General Practice is being encouraged to identify patients who also act as carers, but the support then available is patchy and their additional heath needs are often not met. As has already been stated behaviour that challenges often starts in childhood and may become an ingrained form of behaviour and communication. More needs to be done to encourage carers to identify early signs of behaviour that challenges and then offer practical help to enable them both to deal with it and manage their own distress.

9.4 Review question: In people with a learning disability, and their carers, what are the benefits and potential harms of interventions aimed at reducing health risks and increasing understanding of physical illness or mental health problems in relation to the prevention or management of the behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 66. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 66: Clinical review protocol summary for the review of interventions aimed at reducing health risks and increasing understanding of physical illness or mental health problems

Component	Description
Review question	In people with a learning disability, and their carers, what are the benefits and potential harms of interventions aimed at reducing health risks and increasing understanding of physical illness or mental health problems in relation to the prevention or management of the behaviour that challenges? (RQ3.2)
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability and behaviour that challenges
Intervention(s)	Any intervention that aims to reduce health risks and increase understanding of health problems in relation to the prevention or management of behaviour that challenges, such as annual health checks or hand held health records.
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Adaptive functioning, including communication skills • Behaviour that challenges • Mental and psychological health outcomes • Physical health outcomes • Premature death • Quality of life • Service user and carer understanding of health problems
Study design	RCTs and systematic reviews.

Component	Description
Note. RCTs = Randomised controlled trials	

9.4.1 Clinical evidence

9.4.1.1 Hand-held health record versus treatment as usual

Two RCTs (N = 473) met the eligibility criteria for this review: Lennox 2010 (Lennox et al., 2010), Turk 2010 (Turk et al., 2010). Both of the eligible studies included sufficient data to be included in the evidence syntheses. Lennox 2010 had 4 study arms; for the purposes of this review, only the arm that received the hand held health record and the no treatment arm were utilised (N = 134). An overview of the trials included in the meta-analysis can be found in Table 67.

Summary of findings can be found in the Table 68. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of mental and psychological health outcomes, adaptive functioning, behaviour that challenges or quality of life.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 67: Study information table for trials included in the meta-analysis of interventions aimed at reducing health risks and increasing understanding of physical illness or mental health problems versus treatment as usual

	Hand-held health record versus treatment as usual	Annual health check versus treatment as usual
Total no. of studies (N[1])	2 (335)	2 (592)
Study ID	(1) Lennox 2010[2] (2) Turk 2010	(1) Lennox 2007 (2) Lennox 2010[3]
Country	(1) Australia (2) UK	Australia
Diagnosis	LD	LD
Age (mean)	(1) 36 (2) 40	(1) 39 (2) 36
Sex (% Female)	(1) 43 (2) 39	(1) 44 (2) 43
Ethnicity (% White)	(1) Not reported (2) 92	Not reported
IQ (mean)	Not reported	Not reported
Treatment length (weeks)	52	One-off check; 52-week follow-up
Intervention	(1) Advocacy Skills Kit Diary (2) Personal Health Profile	(1 – 2) Comprehensive Health Assessment Program
Comparison	TAU	TAU

Notes: N = total number of participants; LD = learning disability; TAU = treatment as usual
[1]Number randomised.
[2]Four armed trial; hand-held health record arm and no treatment arm utilised.

	Hand-held health record versus treatment as usual	Annual health check versus treatment as usual

[3]Four armed trial; health check arm and no treatment arm utilised.

Table 68: Summary of findings table for hand-held health record versus treatment as usual

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Treatment as usual**	Corresponding risk **Hand-held health record**			
Health promotion (blood pressure checked) Follow-up: mean 52 weeks	**471 per 1000**	**551 per 1000** (386 to 781)	**RR 1.17** (0.82 to 1.66)	119 (1 study)	**low**[1]
Health promotion (constipation investigation) Follow-up: mean 52 weeks	**15 per 1000**	**98 per 1000** (12 to 814)	**RR 6.67** (0.8 to 55.33)	119 (1 study)	**low**[1]
Health promotion (hearing test) Follow-up: mean 52 weeks	**29 per 1000**	**59 per 1000** (10 to 339)	**RR 2** (0.35 to 11.53)	119 (1 study)	**low**[1]
Health promotion (vision test) Follow-up: mean 52 weeks	**59 per 1000**	**137 per 1000** (42 to 444)	**RR 2.33** (0.72 to 7.55)	119 (1 study)	**low**[1]
Health promotion (weight measured) Follow-up: mean 52 weeks	**250 per 1000**	**352 per 1000** (203 to 615)	**RR 1.41** (0.81 to 2.46)	119 (1 study)	**low**[1]
Health promotion (weight management plan) Follow-up: mean 52 weeks	**176 per 1000**	**99 per 1000** (37 to 261)	**RR 0.56** (0.21 to 1.48)	119 (1 study)	**low**[1]
Health promotion (epilepsy review) Follow-up: mean 52 weeks	**118 per 1000**	**215 per 1000** (94 to 498)	**RR 1.83** (0.8 to 4.23)	119 (1 study)	**low**[1]
Service user knowledge of health problems Knowledge of Health Problems and Terminology Checklist (unvalidated measure)		The mean service user knowledge of health problems in the intervention groups was **0.32 standard deviations lower** (0.81 lower to 0.16 higher)		66 (1 study)	**very low**[1,2]
Carer knowledge of health problems Knowledge of Health Problems and Terminology Checklist (unvalidated measure)		The mean carer knowledge of health problems in the intervention groups was **0 standard deviations higher** (0.33 lower to 0.33 higher)		144 (1 study)	**very low**[1,2]
Carer satisfaction		The mean carer satisfaction in the intervention groups was **0 standard deviations higher** (0.39 lower to 0.39 higher)		101 (1 study)	**very low**[1,2]
Service user satisfaction		The mean service user satisfaction in the intervention groups was **0.6 standard deviations higher** (0.08 lower to 1.27 higher)		36 (1 study)	**very low**[1,2]
Premature death	**23 per 1000**	**62 per 1000** (12 to 309)	**RR 2.72** (0.54 to 13.61)	169 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Optimal information size not met; small, single study
[2] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect

9.4.1.21 Annual health check versus treatment as usual

Two RCTs (N = 730) met the eligibility criteria for this review and provided sufficient data to be included in the evidence syntheses: Lennox 2007 (Lennox et al., 2007), Lennox 2010 (Lennox et al., 2010). Lennox 2010 had 4 study arms but for the purposes of this review, only the arm that received the annual health check and the no treatment arm were utilised (N = 138). An overview of the trials included in the meta-analyses can be found in Table 67.

Summary of findings can be found in the Table 69. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of mental and psychological health outcomes, behaviour that challenges, adaptive functioning, quality of life or service user and carer understanding of health problems.

The study flow diagram and evidence tables (including methodological checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 69: Summary of findings table for annual health check versus treatment as usual

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Treatment as usual**	Corresponding risk **Annual health check**			
Health promotion (blood pressure checked) Follow-up: mean 52 weeks	**456 per 1000**	**498 per 1000** (420 to 593)	**RR 1.09** (0.92 to 1.30)	574 (2 studies)	**very low**[1,2]
Health promotion (constipation investigation) Follow-up: mean 52 weeks	**15 per 1000**	**75 per 1000** (9 to 656)	**RR 5.13** (0.59 to 44.58)	121 (1 study)	**low**[3]
Health promotion (hearing test) Follow-up: mean 52 weeks	**10 per 1000**	**128 per 1000** (25 to 643)	**RR 12.22** (2.43 to 61.49)	574 (2 studies)	**low**[2,4]
Health promotion (vision test) Follow-up: mean 52 weeks	**56 per 1000**	**209 per 1000** (123 to 355)	**RR 3.75** (2.21 to 6.36)	574 (2 studies)	**moderate**[2]
Health promotion (acuity corrected by glasses) Follow-up: mean 52 weeks	**0 per 1000**	**0 per 1000** (0 to 0)	**RR 6.55** (0.34 to 126.14)	453 (1 study)	**low**[3]
Health promotion (otoscopic examination) Follow-up: mean 52 weeks	**228 per 1000**	**393 per 1000** (295 to 525)	**RR 1.72** (1.29 to 2.3)	453 (1 study)	**low**[3]
Health promotion (weight measurement) Follow-up: mean 52 weeks	**185 per 1000**	**454 per 1000** (345 to 596)	**RR 2.46** (1.87 to 3.23)	574 (2 studies)	**moderate**[2]
Health promotion (weight management plan) Follow-up: mean 52 weeks	**45 per 1000**	**105 per 1000** (30 to 369)	**RR 2.32** (0.66 to 8.14)	574 (2 studies)	**low**[2,4]
Health promotion (epilepsy review) Follow-up: mean 52 weeks	**118 per 1000**	**169 per 1000** (71 to 411)	**RR 1.44** (0.6 to 3.49)	121 (1 study)	**low**[3]
Identification of physical health problem (hearing loss) Follow-up: mean 52 weeks	**0 per 1000**	**0 per 1000** (0 to 0)	**RR 29.02** (1.75 to 482.11)	453 (1 study)	**low**[3]
Identification of physical health problem (visual impairment) Follow-up: mean 52 weeks	**5 per 1000**	**30 per 1000** (4 to 241)	**RR 6.55** (0.81 to 52.82)	453 (1 study)	**low**[3]
Identification of physical health problem (obesity) Follow-up: mean 52 weeks	**18 per 1000**	**73 per 1000** (25 to 213)	**RR 3.98** (1.36 to 11.64)	453 (1 study)	**low**[3]

Premature death Follow-up: mean 52 weeks	**5 per 1000**	**4 per 1000** (0 to 68)	**RR 0.94** (0.06 to 14.87)	453 (1 study)	**low**[3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] $I^2 > 75\%$
[2] Optimal information size not met
[3] Optimal information size not met; small, single study
[4] $I^2 > 40\%$

9.4.1.3 Annual health check versus hand-held health record

One RCT (N = 272) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Lennox 2010 (Lennox et al., 2010). Lennox 2010 had 4 study arms; for the purposes of this review, only the arm that received the annual health check and the arm that received the hand-held health record were utilised (N = 118). An overview of the trial included in the meta-analysis can be found in Table 70.

Summary of findings can be found in the Table 71. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of mental and psychological health outcomes, behaviour that challenges, adaptive functioning, premature death, quality of life or service user and carer understanding of health problems.

The study flow diagram and evidence tables (including methodological checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 70: Study information table for trials included in the meta-analysis of interventions aimed at reducing health risks and increasing understanding of physical illness or mental health problems

	Annual health check versus held health record	Annual health check + held health record versus treatment as usual	Opportunistic health check versus treatment as usual
Total no. of studies (N[1])	1 (118)	1 (154)	1 (111)
Study ID	Lennox 2010[2]	Lennox 2010[3]	Jones 1997
Country	Australia	Australia	UK
Diagnosis	LD	LD	LD
Age (mean)	36	36	41
Sex (% Female)	43	43	50
Ethnicity (% White)	Not reported	Not reported	Not reported
IQ (mean)	Not reported	Not reported	Not reported
Treatment length (weeks)	One-off check; 52 week FU	52	One-off check; 26 week FU
Intervention	Comprehensive Health Assessment Program	Comprehensive Health Assessment Program +	Opportunistic health check

	Annual health check versus held health record	Annual health check + held health record versus treatment as usual	Opportunistic health check versus treatment as usual
		Advocacy Skills Kit Diary	
Comparison	Advocacy Skills Kit Diary	TAU	TAU

Notes: FU = follow-up; LD = learning disability; N = total number of participants; TAU = treatment as usual
[1]Number randomised.
[2]Four armed trial; annual health check arm and hand-held health record arm utilised.
[3]Four armed trial; annual health check + hand-held health check arm and no treatment arm utilised.

Table 71: Summary of findings table for annual health check versus hand-held health record

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Hand-held health record**	Corresponding risk **Annual health check**			
Health promotion (blood pressure checked) Follow-up: mean 52 weeks	**549 per 1000**	**489 per 1000** (340 to 708)	**RR 0.89** (0.62 to 1.29)	104 (1 study)	**low**[1]
Health promotion (constipation investigation) Follow-up: mean 52 weeks	**98 per 1000**	**75 per 1000** (22 to 266)	**RR 0.77** (0.22 to 2.71)	104 (1 study)	**low**[1]
Health promotion (hearing test) Follow-up: mean 52 weeks	**59 per 1000**	**189 per 1000** (55 to 646)	**RR 3.21** (0.94 to 10.99)	104 (1 study)	**low**[1]
Health promotion (vision test) Follow-up: mean 52 weeks	**137 per 1000**	**207 per 1000** (88 to 494)	**RR 1.51** (0.64 to 3.60)	104 (1 study)	**low**[1]
Health promotion (weight measured) Follow-up: mean 52 weeks	**353 per 1000**	**547 per 1000** (349 to 854)	**RR 1.55** (0.99 to 2.42)	104 (1 study)	**low**[1]
Health promotion (weight management plan) Follow-up: mean 52 weeks	**98 per 1000**	**283 per 1000** (111 to 722)	**RR 2.89** (1.13 to 7.36)	104 (1 study)	**low**[1]
Health promotion (epilepsy review) Follow-up: mean 52 weeks	**216 per 1000**	**170 per 1000** (78 to 375)	**RR 0.79** (0.36 to 1.74)	104 (1 study)	**low**[1]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Optimal information size not met; small, single study

9.4.1.43 Annual health check plus hand-held health record versus treatment as usual

One RCT (N = 272) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Lennox 2010 (Lennox et al., 2010). Lennox 2010 had 4 study arms; for the purposes of this review, only the arm that received the annual health check plus the hand-held health record and the no treatment arm were utilised (N = 154). An overview of the trial included in the meta-analysis can be found in Table 70.

Summary of findings can be found in the Table 72. The full GRADE evidence profiles and associated forest plots can be found in Appendix O.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of mental and psychological health outcomes, behaviour that challenges, adaptive functioning, premature death, quality of life or service user and carer understanding of health problems.

The study flow diagram and evidence tables (including methodological checklists) can be found in Appendix N, and exclusion list in Appendix Q.

9.4.1.5 Table 72: Summary of findings table for annual health check plus hand-held health record versus treatment as usual

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Treatment as usual**	Corresponding risk **Annual health check + hand-held health record**			
Health promotion (blood pressure checked) Follow-up: mean 52 weeks	**471 per 1000**	**659 per 1000** (485 to 889)	**RR 1.4** (1.03 to 1.89)	138 (1 study)	**low**[1]
Health promotion (constipation investigation) Follow-up: mean 52 weeks	**15 per 1000**	**57 per 1000** (7 to 498)	**RR 3.89** (0.45 to 33.89)	138 (1 study)	**low**[1]
Health promotion (hearing test) Follow-up: mean 52 weeks	**29 per 1000**	**143 per 1000** (32 to 628)	**RR 4.86** (1.1 to 21.36)	138 (1 study)	**low**[1]
Health promotion (vision test) Follow-up: mean 52 weeks	**59 per 1000**	**286 per 1000** (103 to 792)	**RR 4.86** (1.75 to 13.47)	138 (1 study)	**low**[1]
Health promotion (weight measured) Follow-up: mean 52 weeks	**250 per 1000**	**585 per 1000** (370 to 925)	**RR 2.34** (1.48 to 3.7)	138 (1 study)	**low**[1]
Health promotion (weight management plan) Follow-up: mean 52 weeks	**176 per 1000**	**101 per 1000** (42 to 238)	**RR 0.57** (0.24 to 1.35)	138 (1 study)	**low**[1]
Health promotion (epilepsy review) Follow-up: mean 52 weeks	**118 per 1000**	**100 per 1000** (39 to 261)	**RR 0.85** (0.33 to 2.22)	138 (1 study)	**low**[1]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Optimal information size not met; small, single study

9.4.1.6 Opportunistic health check versus any control

One RCT (N = 111) met the eligibility criteria for this review: Jones 1997 (Jones & Kerr, 1997). However, the trial reported critical outcomes that could not be included in the meta-analyses due to the way the data had been reported; a brief narrative synthesis is therefore given. An overview of the included trial can be found in Table 70.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of behaviour that challenges, adaptive functioning, premature death, quality of life or service user and carer understanding of health problems.

The study flow diagram and evidence tables (including methodological checklists) can be found in Appendix N, and exclusion list in Appendix Q.

9.4.2 Economic evidence

The systematic search of the economic literature identified 1 study that assessed the cost effectiveness of health checks aimed at reducing health risks in people with a learning disability (Romeo et al., 2009). Details on the methods used for the systematic review of the economic literature are described in Chapter 3; full references to the included studies and evidence tables for all economic evaluations included in the systematic literature review are provided in Appendix S. Completed methodology checklists of the studies are provided in Appendix R. Economic evidence profiles of studies considered during guideline development (that is, studies that fully or partly met the applicability and quality criteria) are presented in Appendix T.

Romeo and colleagues (2009) evaluated the costs and outcomes of a health-check intervention versus standard care offered to adults with a learning disability registered with primary care services in the UK. The health-check intervention comprised a review of participants' GP records by an experienced nurse; assessment of participants' general physical & mental health, development & problem behaviours, selected physical examination and blood tests; discussion of the results with a GP; preparing a report of findings and recommendations to the participants' GP; and referral algorithms to learning disabilities services. The economic analysis was based on a cohort study with matched controls that followed 100 people for a period of 12 months (Cooper et al., 2006) Participants were matched with controls for age, gender and level of learning disability. The analysis adopted a societal perspective; costs consisted of intervention costs (equipment & staff time), primary, inpatient, outpatient & specialist learning disability service costs, costs of other healthcare services, daytime activity costs comprising unsupported & supported paid employment, voluntary work, adult education classes, day centres and additional support, costs of respite care, costs of aids and adaptations, as well as costs associated with paid and unpaid care. Costs were collected prospectively for the intervention group and retrospectively for the control group. Unit costs were based on national sources & further estimates. The effectiveness of the intervention was measured by the levels of health need detection, met new health needs, met health promotion and monitoring needs.

According to the study findings, the mean total cost of intervention was £82 per person. Total mean service costs were similar for the intervention and standard care. However, the total costs per person were significantly lower for the intervention compared with control (bootstrapped cost difference -£22,772 per person in 2003 prices, 95%CI -£37,569 to -£6,400), resulting from lower mean carer support costs per person associated with the intervention. The intervention resulted in a higher number of newly identified health needs and new health needs that were met per person, and a higher level of met health promotion and health monitoring needs per person; all differences in outcomes between the health-check intervention and standard care were statistically significant. Therefore, the intervention was shown to be dominant over standard care, as it resulted in better outcomes, similar service costs and lower carer support and total costs compared with standard care. The study is directly applicable to the guideline context as it was undertaken in the UK, but it is characterised by potentially serious limitations, mainly relating to the study design (retrospective measurement of control costs) and the small number of people participating in the study.

9.4.3 Clinical evidence statements

9.4.3.1 Hand-held health record versus treatment as usual

- Low quality evidence from a single study (N = 121) was inconclusive as to the effectiveness of the hand-held health record when compared with treatment as usual in increasing the probability of receiving a blood pressure check, a hearing test or a weight management plan by 52-week follow-up.

- Low quality evidence from a single study (N = 119), suggested that the hand-held health record increased the probability of receiving a constipation investigation, a vision test and a weight measurement by 52-week follow-up when compared with treatment as usual. However, the precision of this estimate is poor.
- Low quality evidence from a single study (N = 119), suggested that the hand-held health record increased the probability of receiving an epilepsy review by 52-week follow-up when compared with treatment as usual. However, the precision of this estimate is poor.
- Very low quality evidence from a single study (N = 144) was inconclusive as to the effectiveness of the hand-held health record when compared with treatment as usual in increasing carer knowledge of health problems at 52-week follow-up.
- Low quality evidence from a single study (N = 66) suggested that the hand-held health record was less effective than treatment as usual in increasing service user knowledge of health problems at 52-week follow-up, but the precision of this estimate is poor.
- Very low quality evidence from a single study (N = 101) was inconclusive as to the effectiveness of the hand-held health record when compared with treatment as usual in increasing carer satisfaction at the end of intervention.
- Very low quality evidence from a single study (N = 36) suggested that the hand-held health record was more effective than treatment as usual in increasing service user satisfaction at the end of intervention. However, the precision of this estimate is poor.
- Very low quality evidence from a single study (N = 169) suggested that the hand-held health record was less effective than treatment as usual in reducing premature deaths at the end of intervention. However, the precision of this estimate is poor.

9.4.3.2 Annual health checks versus treatment as usual

- Very low quality evidence from 2 studies (N = 576) was inconclusive as to the effectiveness of the annual health check when compared with treatment as usual in increasing the probability of receiving a blood pressure check by 52-week follow-up.
- Low quality evidence from up to 2 studies (N = 574), suggested that the annual health check increased the probability of receiving a constipation investigation, having acuity corrected by glasses and receiving a weight management plan by 52-week follow-up when compared with treatment as usual. However, the precision of all of these estimates is poor.
- Moderate quality evidence from up to 2 studies (N = 574), suggested that the annual health check increased the probability of having a hearing test, vision test, otoscopic examination and weight measurement by 52-week follow-up when compared with treatment as usual.
- Low quality evidence from a single study (N = 121) was inconclusive as to the effectiveness of the annual health check when compared with treatment as usual in increasing the probability of receiving an epilepsy review at 52-week follow-up.
- Low quality evidence from a single study (N = 453) suggested that the annual health check increased the probability of identifying hearing loss, visual impairment and obesity at 52-week follow-up when compared with treatment as usual.
- Low quality evidence from a single study (N = 453) was inconclusive as to the effectiveness of the annual health check when compared with treatment as usual in reducing the probability of premature death at 52-week follow-up.

9.4.3.3 Annual health check versus hand-held health record

- Low quality evidence from a single study (N = 104) was inconclusive as to the effectiveness of the annual health check when compared with hand-held health records in increasing the probability of receiving a blood pressure check or a constipation investigation by 52-week follow-up.

- Low quality evidence from a single study (N = 104) suggested that the annual health check increased the probability of receiving a hearing test and a vision test by 52-week follow-up when compared with a hand-held health record. However, the precision of both of these estimates is poor.
- Low quality evidence from a single study (N = 104) suggested that the annual health check increased the probability of having weight measured and receiving a weight management plan by 52-week follow-up when compared with a hand-held health record.
- Low quality evidence from a single study (N = 104) was inconclusive as to the effectiveness of the annual health check when compared with hand-held health records in increasing the probability of receiving an epilepsy review by 52-week follow-up.

9.4.3.4 Annual health check plus hand-held health record versus treatment as usual

- Low quality evidence from a single study (N = 138) suggested that the annual health check plus a hand-held health record increased the probability of receiving a blood pressure check, a constipation investigation, a hearing test, a vision test and a weight measurement by 52-week follow-up when compared with treatment as usual. However, the precision of the estimate for the blood pressure check was poor.
- Low quality evidence from a single study (N = 138) suggested that the annual health check plus a hand-held health record reduced the probability of receiving a weight management plan at 52-week follow-up when compared with treatment as usual, although the precision of the estimate is poor.
- Low quality evidence from a single study (N = 138) was inconclusive as to the effectiveness of the annual health check plus a hand-held health record when compared with treatment as usual in increasing the probability of receiving an epilepsy review by 52-week follow-up.

9.4.3.5 Opportunistic health check versus any control

- One trial could not be included in the meta-analysis (N = 111). The authors reported no significant differences in consultation patterns between the 2 groups at 26-week follow-up, either in the total number of consultations, or in the outcome (advice, prescription, intervention or referral) of the consultations. Moreover, the authors reported no significant difference across a range of health promotion issues.

9.4.4 Economic evidence statements

- Low quality evidence from a cohort study with matched controls (N = 100) suggested that regular health checks aiming to identify and manage health needs of people with a learning disability are cost-effective as they result in a higher number of new health needs identified and met, and similar service costs. The evidence is directly relevant to the UK but is characterised by potentially serious limitations.

9.5 Recommendations and link to evidence

9.5.1 Psychosocial interventions aimed at prevention of behaviour that challenges

Recommendations	
	34. Consider preschool classroom-based interventions for children aged 3–5 years. 35. Preschool classroom-based interventions should have multiple components, including: • curriculum design and development

	• social and communication skills training for the children • skills training in behavioural strategies for parents or carers • training on how to mediate the intervention for teachers.
Relative values of different outcomes	The GDG agreed that the following outcomes were critical: behaviour that challenges, adaptive functioning (including integration into mainstream education and social and communication skills), quality of life, and service user and carer satisfaction. There were limited data available on these outcomes and the study populations were diagnosed with autism and so did not represent the full range of learning disabilities covered by this guideline.
Trade-off between clinical benefits and harms	The evidence suggested that educational interventions in pre-school children have benefits in terms of behaviour that challenges and adaptive functioning. The GDG was of the view that these interventions with young children at risk of developing behaviour that challenges may also have long-term benefits in supporting their integration into mainstream education. There was no evidence regarding quality of life, satisfaction, or specific harms. There was insufficient evidence to make a distinction between: (1) home- and centre-based early behavioural interventions, (2) EIBI and parent training, and (3) high and low supervision EIBI, or to support a recommendation for various parent-delivered interventions.
Trade-off between net health benefits and resource use	Existing economic evidence on EIBI is limited, flawed, and only partially applicable to the UK context. The GDG considered that the benefits of educational interventions in pre-school children in terms of behaviour that challenges and adaptive functioning may lead to substantial future cost-savings, primarily associated with integration of children into mainstream education and thus reduced need for high cost special education. Improvements in behaviour that challenges may also lead to cost-savings due to reduction in the need for assessment and management of such behaviour.
Quality of evidence Other considerations	All evidence was graded low to very low quality because it was based on 1 or 2 studies with fewer than 300 participants in total, and there were concerns about risk of bias and applicability. In developing the recommendations the GDG was mindful of: (a) the very considerable burden experienced both by those who have behaviour that challenges and by their families and carers, and (b) the evidence reviewed in the chapter on the experience of care and on the evidence of effectiveness for parent training and psychosocial interventions to support carers and the considerable problems that many carers experienced in accessing care for family members. A consideration of all these factors led the GDG to make recommendations that would offer increased opportunities through pre-school interventions to children with a learning disability, many of whom have an increased risk of developing behaviour that challenges.

1

9.5.22 Health care interventions aimed at prevention of behaviour that challenges

Recommendations	
	36. Offer an annual physical health check to people with a learning disability in all settings. Carry out the physical health check

	together with a family member, carer or healthcare professional or social care practitioner who knows the person. Ensure that it takes into account any known or emerging behaviour that challenges and how it may be linked to any physical health problems, and contains: • a physical health review • a review of all current health interventions, including medication and any side effects • an agreed and shared care plan for managing any physical health problems.
Relative values of different outcomes	The GDG agreed that the following outcomes were critical: behaviour that challenges, adaptive functioning (including communication skills), mental and psychological health outcomes, physical health outcomes, premature death, quality of life, and service user and carer understanding of health problems.
Trade-off between clinical benefits and harms	For people with a learning disability, the evidence was inconclusive in determining which of the following interventions were effective in supporting improved health outcomes: 1) hand-held health records when compared with treatment as usual, 2) combining an annual health check with hand-held health records, and 3) undertaking opportunistic health checks. The evidence for the overall benefits on health outcomes for annual health checks compared with treatment as usual was limited, although there was some evidence of improved probability of having various tests (that is, a hearing test, vision test, otoscopic examination and weight measurement) and identifying hearing loss, visual impairment and obesity. When annual health checks were compared with hand-held health records, the evidence was generally inconclusive, although the former may increase the probability of having weight measured and receiving a weight management plan.
Trade-off between net health benefits and resource use	Regular health checks offered to people with a learning disability appear to be cost effective because they improve health outcomes in terms of health needs identified and met, at a similar service cost to standard care. The GDG considered that annual health checks in this population were likely to lead to identification and management of underlying physical health problems at an earlier, milder stage, before they become severe and require more resource intensive management, thus leading to improved health outcomes in the longer term and potential future cost-savings. Moreover, the GDG took into consideration that unrecognised physical illness in people with a learning disability may lead to pain and discomfort, which, in turn, may be an important precipitant of behaviour that challenges in this population. Therefore, early identification of physical health problems in people with a learning disability may prevent or reduce the levels of behaviour that challenges, thus leading to a reduction in costs associated with the assessment and management of such behaviour.
Quality of evidence	Most evidence was graded low to very low quality because it was based on 1 or 2 studies with relatively few participants, and there were concerns about risk of bias or inconsistency. The only moderate quality evidence was for annual health checks compared with treatment as usual, and this was downgraded for imprecision.
Other considerations	In developing recommendations in this area, the GDG took into consideration 2 factors about the physical health of people with a learning disability: (1) many types of physical disease go unrecognised in people with a learning disability, in part because of the communication difficulties some people experience and in part because of healthcare professionals' lack of

	knowledge and awareness about how to communicate with and assess people with a learning disability who may be physically unwell, and (2) that unrecognised physical illness and the associated pain and discomfort can be an important precipitant of behaviour that challenges in people with a learning disability. Regular proactive monitoring of physical health problems was therefore supported by the GDG as a means both to reduce the likelihood of behaviour that challenges developing and understanding possible causal mechanisms where it already exists.

9.5.3 Research recommendations

3. Can positive behaviour support provided for children aged under 5 years with a learning disability reduce the risk of developing behaviour that challenges?

10 Environmental interventions

10.1 Introduction

The context in which behaviour that challenges occurs is an essential component in attempting to understand and hence change the frequency and/or intensity of the behaviour. In order to provide successful interventions it is necessary to understand the function of that behaviour for the person. The environment is one element of a functional analysis that needs to be considered when assessing the reason for that behaviour occurring. There may be features of a particular environment that contribute to the occurrence of particular behaviour. It is therefore possible, that by changing the environment (sometimes referred to as 'ecological manipulation'), the likelihood of the behaviour occurring can be reduced.

Behaviour that challenges is known to increase in institutional type settings or impoverished environments where there is a lack of engagement, poor social support, higher rates of restrictive practices and often higher reports of abusive practices (Department of Health, 2007). Poor parenting experiences can also increase the rate of behaviour that challenges, and may too be abusive. Over recent years there has been a shift from providing support to people with behaviour that challenges in institutional settings, to community-focused models of support that advocate person-centred planning and individualised care (Lowe et al., 2007a).

The environment is not just the physical space that a person occupies, but also the people, culture, social factors and opportunities that surround and influence the person. These factors are not mutually exclusive and will need to be considered as a whole when thinking about the right environment for a person. It has been recognised that the physical environment will need to be capable of meeting the person's needs and be tolerant of unintended use (Brand, 2010) and that the people within the environment will need to be provided with the tools to deliver person centred care and support effectively.

McGill and colleagues (McGill et al., in press) use the terms 'challenging' and 'capable' environments. Challenging environments would include the practices often associated with institutional-style care and support or poor parenting practices. Capable environments are those that support a person effectively and provide the optimal setting to support positive interactions and opportunities. It is an holistic approach to align the multiple factors that form part of a person's environment including building design, an appropriate physical environment, consistency of support for communication, opportunities to engage in meaningful activities and develop independent skills, opportunities to make positive social interactions and to maintain relationships, provision of real choice, support to maintain good health, and a skilled staff team, supported through management and organisational values that promote personal preference and aspirations.

In order to ensure the right environmental fit for a person with a learning disability, it is necessary to understand their individual needs. Alongside understanding the function of their behaviour, this will often also include understanding their communication, sensory, health and support needs, preferences for activities, skill level, and engagement style. This will tend to require support from health and social care professionals to undertake assessments and provide a clear understanding of the person's needs. This work may be undertaken directly with the person with a learning disability and behaviour that challenges, or with their support networks to equip them to meet that person's needs.

There are approaches that seek to provide such understanding. Positive behavioural support (Allen et al., 2005) seeks to better understand and so reduce the behaviour that challenges through use of a multi-element format to consider changing the environment, developing skills, providing focused support and developing reactive strategies. In this way environmental adaptations are not solely aimed at reducing the behaviour that challenges,

but also at improving the person's quality of life (Mackenzie-Davies & Hardy, 2010). Person-centred active support (Mansell, 2007) seeks to provide an understanding of how to effectively engage people within their environments. Both models seek to enable people with a learning disability and behaviour that challenges to increase their confidence and self-esteem through exploration of their 'capable' environment, providing opportunity for developing interests and skills, and ultimately supporting mastery of the environment.

10.2 Review question: In people with a learning disability and behaviour that challenges, what are the benefits and potential harms associated with environmental changes aimed at reducing and managing behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 73. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 73: Clinical review protocol summary for the review of environmental interventions aimed at reducing and managing behaviour that challenges

Component	Description
Review question(s)	In people with a learning disability and behaviour that challenges, what are the benefits and potential harms associated with environmental changes aimed at reducing and managing behaviour that challenges? (RQ4.1)
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability and behaviour that challenges.
Intervention(s)	All environmental changes, including the physical and social environments.
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Targeted behaviour that challenges • Adaptive functioning, including communication skills. • Quality of life. • Service user and carer satisfaction.
Study design	RCTs and systematic reviews.
Note. RCT = Randomised controlled trial.	

10.2.1 Clinical evidence

The GDG considered the RCT evidence for this section of the guideline to be limited in terms of quality, directness and quantity. The range of included studies was therefore expanded to systematic reviews of non-randomised studies (see Table 74).

10.2.1.1 Sensory intervention versus any control

Three RCTs (N = 137) met the eligibility criteria for this review: Chan 2005 (Chan et al., 2005), Lundqvist 2009 (Lundqvist et al., 2009), Martin 1998 (Martin et al., 1998). Of the eligible studies, only 2 (N = 109) included sufficient data to be included in the evidence syntheses (Chan 2005; Lundqvist 2009). One trial (Martin 1998; N = 27) included critical outcomes that could not be included in the meta-analyses because of the way the data had

been reported; a brief narrative synthesis is therefore given to assess whether the findings support or refute the meta-analyses. An overview of the trials included in the meta-analysis can be found in Table 74.

Summary of findings can be found in Table 75. The full GRADE evidence profiles and associated forest plots can be found in Appendices P and O.

No data were available for the critical outcomes of quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables can be found in Appendix N, and exclusion list in Appendix Q.

Table 74: Study information table for trials included in the meta-analysis of environmental interventions versus any control

	Sensory intervention versus any control	Structured versus unstructured activity
Total no. of studies (N[1])	3 (136)	1 (26)
Study ID	(1) Chan 2005 (2) Lundqvist 2009 (3) Martin 1998[2]	Gencoz 1997
Country	(1) Hong Kong (2) Sweden (3) UK	Turkey
Diagnosis	(1, 2) LD (3) Severe to profound LD	LD
Age (mean)	(2, 3) 37-38 (1) Not reported	12
Sex (% Female)	(1) 60 (2, 3) 33-35	Not reported
Ethnicity (% White)	(1, 3) Not reported (2) 100	Not reported
IQ (mean)	Not reported	Not reported
Targeted behaviour that challenges	(1) Aggressive and maladaptive behaviour (2, 3) Not specified	Maladaptive behaviours
Treatment length (weeks)	(1, 3) 12-16 (2) 5	7
Intervention	(1, 3) Multisensory environment (2) Vibroacoustic chair	Special Olympics Sports Skill Instructional Program
Comparison	(1, 3) Attention control (2) Waiting list control	Attention control

Note. LD = learning disability; N = total number of participants; RCT = randomised controlled trial; TAU = treatment as usual.
[1] Number randomised.
[2] Data not reported in a meta-analysable format; findings are described narratively.

Table 75: Summary of findings table for sensory interventions compared with any control

Outcomes	Sensory intervention versus any control	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (global) - post-treatment Change score[1]	The mean targeted behaviour that challenges (global) - post-treatment in the intervention groups was **1.69 standard deviations higher** (1.2 to 2.18 higher)	89 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (global) - follow-up Change score[1] Follow-up: mean 12 weeks	The mean targeted behaviour that challenges (global) - follow-up in the intervention groups was **0.00 standard deviations higher** (0.42 lower to 0.42 higher)	89 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (self-injurious behaviour, severity) - post-treatment	The mean targeted behaviour that challenges (self-injurious behaviour, severity) - post-treatment in the intervention groups was **0.2 standard deviations lower** (1.08 lower to 0.68 higher)	20 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (self-injurious behaviour, frequency) - post-treatment	The mean targeted behaviour that challenges (self-injurious behaviour, frequency) - post-treatment in the intervention groups was **0.25 standard deviations lower** (1.14 lower to 0.63 higher)	20 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (stereotypical behaviour, severity) - post-treatment	The mean targeted behaviour that challenges (stereotypical behaviour, severity) - post-treatment in the intervention groups was **0.33 standard deviations higher** (0.55 lower to 1.21 higher)	20 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (stereotypical behaviour, frequency) - post-treatment	The mean targeted behaviour that challenges (stereotypical behaviour, frequency) - post-treatment in the intervention groups was **0.22 standard deviations lower** (1.1 lower to 0.66 higher)	20 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (aggressive/ destructive behaviour, severity) - post-treatment	The mean targeted behaviour that challenges (aggressive/ destructive behaviour, severity) - post-treatment in the intervention groups was **0.15 standard deviations lower** (1.03 lower to 0.72 higher)	20 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (aggressive/ destructive behaviour, frequency) - post-treatment	The mean targeted behaviour that challenges (aggressive/ destructive behaviour, frequency) - post-treatment in the intervention groups was **0.22 standard deviations lower** (1.1 lower to 0.66 higher)	20 (1 study)	**very low**[2,3]
Adaptive functioning - post-treatment Change score[1]	The mean adaptive functioning - post-treatment in the intervention groups was **1.12 standard deviations lower** (1.57 to 0.67 lower)	89 (1 study)	**very low**[2,3]
Adaptive functioning - follow-up Change score[1] Follow-up: mean 12 weeks	The mean adaptive functioning - follow-up in the intervention groups was **0.48 standard deviations lower** (0.9 to 0.05 lower)	89 (1 study)	**very low**[2,3]

Note. CI = confidence interval.

[1] Due to significant baseline differences, standard deviation of change and estimates of mean change were derived using initial and final mean values and utilising r = 0.5. Sensitivity analyses were used to explore the impact of altering assumptions about the calculation of the effect size, but this resulted in no change to conclusions.
[2] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[3] Optimal information size not met

10.2.1.23 Structured activity versus unstructured activity

One RCT (N = 26) met the eligibility criteria for this review and provided sufficient data to be included in the evidence syntheses: Gencoz 1997 (Gencoz, 1997). An overview of the included trial can be found in Table 74.

Summary of findings can be found in Table 76. The full GRADE evidence profiles and associated forest plots can be found in Appendix P and O.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 76: Summary of findings table for structured compared with unstructured activity

Outcomes	Structured activity versus unstructured activity	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (severity) - post-treatment Change score[1]	The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.87 standard deviations lower** (1.68 to 0.06 lower)	26 (1 study)	**very low**[2,3]
Targeted behaviour that challenges (severity) - follow-up Change score[1] Follow-up: mean 6 weeks	The mean targeted behaviour that challenges (severity) - follow-up in the intervention groups was **0.95 standard deviations lower** (1.77 to 0.13 lower)	26 (1 study)	**very low**[2,3]

Note. CI = confidence interval.

[1] Due to significant baseline differences, standard deviation of change and estimates of mean change were derived using initial and final mean values and utilising r = 0.5. Sensitivity analyses were used to explore the impact of altering assumptions about the calculation of the effect size, but this resulted in no change to conclusions.
[2] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[3] Optimal information size not met

10.2.1.37 Motivating operations

For the purposes of this review, motivating operations are defined as those variables that alter both the effectiveness of reinforcement or punishment (the value-altering effect) and the frequency of operant response classes related to those consequences (the behaviour-altering effect).

No RCTs or systematic review of RCTs met eligibility criteria for this review. The search for additional systematic reviews identified only 1 that the GDG considered to be relevant: Simo-Pinatella 2013 (Simo-Pinatella et al., 2013). This systematic review included 31 single-n or small-n studies (N = 55): Ahearn 2003 (Ahearn, 2003), Buckley 2006 (Buckley & Newchok, 2006), Butler 2007 (Butler & Luiselli, 2007), Carey 2002 (Carey & Halle, 2002), Carter 2007 (Carter & Wheeler, 2007), Cautilli 2004 (Cautilli & Dziewolska, 2004), Chung 2010 (Chung & Cannella-Malone, 2010b), Kuhn 2009 (Kuhn et al., 2009), Lang 2009 (Lang et al., 2009), Lang 2010 (Lang et al., 2010), Lanovaz 2009 (Lanovaz et al., 2009), LeBlanc 2001 (LeBlanc et al., 2001), Levin & Carr 2011 (Levin & Carr, 2001), Lomas 2010 (Lomas et al., 2010), McComas 2000 (McComas et al., 2000), McComas 2003 (McComas et al., 2003), McGinnis 2010 (McGinnis et al., 2010), O'Reilly 2007 (O'Reilly et al., 2007), O'Reilly 2000 (O'Reilly & Lancioni, 2000), O'Reilly 2009 (O'Reilly et al., 2009), O'Reilly 2006 (O'Reilly et al., 2006) , O'Reilly 2008 (O'Reilly et al., 2008), Pace 2000 (Pace & Toyer, 2000), Piazza 2000 (Piazza et al., 2000), Rapp 2004 (Rapp, 2004), Rapp 2005 (Rapp, 2005), Reed 2005 (Reed et al., 2005), Ringdahl 2002 (Ringdahl et al., 2002), Roantree 2006 (Roantree & Kennedy, 2006), Thiele 2001 (Thiele et al., 2001), van Camp 2000 (Van Camp et al., 2000). Of the included studies, 15 were single-n studies and 16 were small-n studies. A summary of the included review can be found in Table 77.

All included studies were published in peer-reviewed journals between 2000 and 2010 and involved a process of functional assessment plus an intervention focused on the modification of a motivating operation. The mean age of included participants was 9 years (range 4-17 years) and 20% were females. All participants were diagnosed with a learning disability.

Fourteen of the included studies were conducted at the participants' school. Other settings in which studies were conducted included an inpatient unit or facility (k = 4), family home (k = 2), short-term residential facility (k = 2), an outpatient setting (k = 1), day service (k = 1), intensive day-treatment programme (k = 1), community-based group home (k = 1) and Centre Behaviour Analysis Clinic (k = 1).

Among the included participants, the most common behaviour that challenges was aggression (N = 22), stereotypic behaviour (N = 17), destructive behaviour (N = 17), self-injurious behaviour (N = 14) and tantrums (N = 11). Other behaviour that challenges included feeding problems (N = 5), disruptive behaviour (N = 2), pica (N = 1) and property destruction (N = 1). Behaviour that challenges was maintained by automatic reinforcement (N = 19), escape (N = 12), attention (N = 9) and tangible reinforcement (N = 6). Behaviour that challenges was maintained by multiple functions for 6 participants, and the behavioural function was not specified for 3 participants.

Motivating operations were classified as follows:

- Social context variables, involving attention from others and factors related to others' characteristics
- Activity or nature of the task, involving instructional requests, presentation of work and the method of instruction
- Characteristics of the environment, involving factors related to objects or activities and environmental enrichment
- Personal context, involving physiological states.

Appendix N provides the review characteristic table and methodology checklist; the review was judged to be of poor quality (that is, it met only 3 of the 5 criteria), and the quality of evidence for each outcome was graded as very low quality because of limitations inherent in single-case and small-n studies (see section 3.5.3) and the risk of bias associated with individual studies had not been assessed by Simo-Pinatella 2013. The authors did not include unpublished research, arguing that they are 'usually incomplete and their accuracy may be difficult to assess.' However, they did supplement the electronic search by manually searching the reference lists of included studies and the table of content of journals that publish this type of research. In addition, a search was done of authors who commonly publish in this area.

Further information about both included and excluded studies can be found in Simo-Pinatella 2013.

Table 77: Study information table for the systematic review included in the review of antecedent modification

	Simo-Pinatella 2013
Review question/ Aim	To conduct a systematic review of studies that have conducted a functional assessment and a subsequent motivating operation based intervention with school-aged children with a learning disability and behaviour that challenges.
Method used to synthesise evidence	Narrative synthesis
Design of included studies	Small-n and single-n studies[1] • Reversal design (k = 17) • Multi-element (k = 16) • Multiple baseline (k = 3) • Alternating treatments (k = 3) • Multi-probe design (k = 2)
Dates searched	January 2000 to December 2010

	Simo-Pinatella 2013
Electronic databases	PsycINFO, Education Resources Information Center (ERIC), Science Direct, Blackwell, SAGE, and Medline (Ebsco and PubMed).
No. of included studies (N^2)	31 (55)
Participant characteristics	Children and young people (under 18 years old) with a learning disability and behaviour that challenges
Intervention	Process of functional assessment plus an intervention focused on the modification of a motivating operation.
Comparison	N/A
Outcome	Behaviour that challenges
Review Quality	Poor[3]

Note. k = number of studies.
[1] 9 studies used more than one design.
[2] Number of participants.
[3] No quality assessment of included studies was carried out; only published studies searched for.

Evidence from each participant was summarised by the review authors graphically and is reproduced in Table 78.

Table 78: Effect of different types of motivating operations (MOs) on participants' behaviour that challenges in relation to its function (reproduced with permission of the copyright owner)

	Behavioral function				
Type of MO	Automatic reinforcement	Escape	Attention	Access to tangible	Not specified
Social context					
Therapist gender (female)			↓[a]		
Preferred staff (noncontingent social reinforcement)			↓		
Type of attention (verbal and physical attention)			↓		
PSC deprivation (no attention)	↓ ↓ ↑[b] ↑	=* [c] =[d] =	↑* ↑ ↑ ↑ ↑ ↑		
PSC attention	↓ ↓ ↑ ↑	=* = =	↓* ↓ ↓ ↓ ↓		
PSC response blocking	= ↓ ↓ ↓				
PSC attention with response blocking	↓ = = ↑				
Non-CA condition			↓		
CA condition			↓		
CA plus contingency modeling condition			↑		
Attention only condition			↑		
Attention enriched condition			↑		
No PSC attention			↓		
Delivery of praise and preferred food items on a variable time		↓* ↓* ↓		↓* ↓*	
Activity or nature of the task					
Altering instructional requests/method of instruction		↓ ↓ ↓ ↓			
Characteristics of the environment					
Music/environment enrich with music and guitar	↑	↑ ↓*		↓*	
PSC access to tangible		↓* ↓	↓*	↓* ↓ ↓*	
PSC no access to tangible			↑*	↑* ↑ ↑	
PSC restricted access to extinction (no interaction)				↑	

PSC contingent reinforcement with or without delivery of auditory cue		↓ ↓		
Access to different tangibles			*	
Access to nonpreferred food items			↑ ↑ ↑	
Structurally matched stimuli with and without music	*[e]* *			
Structurally unmatched stimuli	* * * ↑↑ ↑			
Matched stimuli	↓ ↓ ↓			
Visual and audio stimulation (television)	* = =			
Delivery of praise and preferred food items on a variable time schedule		↓* ↓* ↓	↓* ↓*	
Personal context				
Adding condiments to the consumption of previously rejected food (vegetables)				↑
Vitamin supplement				↓
Sleep deprivation/disruption		↑ ↑		↑
PSC without free access to stereotypy	↑			
PSC with free access to stereotypy	↓ ↓ ↓ ↓ ↓ ↓			

Note: MO = motivating operations; PSC = pre-session condition; CA = contingent attention.
↓ Abolishing effect for participant
↑ Establishing effect for participant.
= No effect for participant.
* Mixed effects for participant.
[*]Behavioral function of this participant serves multiple functions.

10.2.2 Economic evidence

No economic evidence on environmental changes aimed at reducing and managing behaviour that challenges in people with a learning disability was identified by the systematic search of the economic literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

10.2.3 Clinical evidence statements

10.2.3.1 Sensory intervention versus any control

- Very low quality evidence from 3 separate studies (N = 20 to 89) of sensory interventions was either inconclusive or favoured the control across a range of relevant outcomes.

10.2.3.2 Structured activity versus unstructured activity

- Very low quality evidence from a single study (N = 26), showed structured activity was more effective than unstructured activity in reducing targeted behaviour that challenges at the end of treatment and at 6-week follow-up.

10.2.3.3 Motivating operations

- Based on very low quality evidence from a systematic review that included 31 single-n or small-n studies involving 55 participants, the following motivating operations had a clear effect on behaviour that challenges in the predicted direction:
    - the modification of instructional variables produced abolishing effects for escape-maintained behaviour
    - deprivation of attention had an establishing effect on attention maintained behaviour
    - access to attention had an abolishing effect on attention maintained behaviour
    - sleep disruption had an establishing effect on escape-maintained behaviour.
- Changes in the level of attention did not appear to function as a motivating operation for escape-maintained behaviour
- Evidence was inconclusive as to the effect of providing access to different types of tangible reinforcement on escape-maintained behaviour.

10.2.4 Economic evidence statements

No economic evidence on environmental changes aimed at reducing and managing behaviour that challenges in people with a learning disability is available.

10.3 Recommendations and link to evidence

Recommendations	37. Do not offer sensory interventions (for example, Snoezelen rooms) before carrying out a functional assessment to establish the person's sensory profile. Bear in mind that the sensory profile may change. 38. Consider changing the physical and social environment to prevent the development, exacerbation or maintenance of behaviour that challenges. 39. Consider developing a structured plan of daytime activity (as part of the curriculum if the person is at school) that reflects the person's interests and capacity. Monitor the effects on behaviour that challenges and adjust the plan in discussion with the person and their family members or carers.
Relative values of different outcomes	The GDG agreed that a number of outcomes were critical to addressing this review question: targeted behaviour that challenges, rates of reactive interventions, quality of life, and service user and carer satisfaction.
Trade-off between clinical benefits and harms	Reporting of harms was limited but in the case of sensory interventions (such as Snoezelen rooms) there was an indication that the provision of such interventions (which have been in widespread use) may not be beneficial and could be harmful to some people. Increases in structured day time activity are likely to bring benefits with little, if any increase, in harms.
Trade-off between net health benefits and resource use	No economic evidence on environmental changes aimed at reducing and managing behaviour that challenges in people with a learning disability was identified. The provision of specific sensory interventions may result in modest additional costs. The development of structured daytime activities may also increase costs but the magnitude of such activities and the impact this may have on reduced resource use to manage behaviour that challenges are not known.
Quality of evidence	The evidence was of very low quality, based on 4 small RCTs (N = 163) and a single review of single-case and small-n studies.
Other considerations	The GDG reviewed the evidence for 3 different kinds of environmental interventions; sensory interventions, structured daytime activity and motivating operations. The reviews did not find any evidence on the effectiveness of positive behaviour support. The GDG carefully considered the evidence for sensory interventions and the possible harms and judged that they should not be used unless a functional analysis had clearly identified such interventions as likely to be of benefit. Instead, the GDG recognised that some settings could promote behaviour that challenges and saw the benefit of advising staff to consider changing the physical and social environment to prevent this from happening. The very limited evidence for structured daytime activity was acknowledged by the GDG, but drawing on their expert knowledge of the impact of impoverished environments on the likelihood of increases in behaviour that challenges, they decided to recommend that plans for structured daytime activity should be developed.

	The review of motivating operations suggested that the factors emerging from the review should inform the development of a range of interventions to address behaviour that challenges, but rather than develop a separate recommendation on them, the GDG felt that the evidence reviewed should be used to inform the development of recommendations on assessment and interventions covered in Chapters 8 and 11.

11 Psychosocial interventions

11.1 Introduction

Psychosocial interventions are the most commonly reported forms of intervention used for behaviour that challenges in people with a learning disability over the last 50 years. Interventions derived from behavioural models feature most prominently within this overall category of intervention. Behavioural interventions, which involve identifying a range of personal, social and environmental events that precipitate behaviours and the subsequent impact of these behaviours, have evolved significantly since their early use with this population. Although the behavioural model has offered a variety of intervention options, until the mid-late 1980s the use of aversive or punishment-based interventions (when an unpleasant or aversive consequence was delivered contingently upon the occurrence of behaviour that challenges) was often a key element of a number of interventions. Contemporary behavioural interventions have moved away from the use of punishment approaches and have focused instead on changing known antecedents for behaviour that challenges, removing certain triggers where possible (for example, pain from an untreated physical health problem), teaching new skills to replace the function of this behaviour or better enable people to cope with known stressors, and using reinforcement to shape behaviour that is non-challenging. Intervention is based on functional assessment that identifies the precipitants and reinforcers for the behaviour. Behavioural intervention is predicated upon individualised packages of assessment and support. This individual focus is congruent with person-centred approaches, and is central to a model that is based on a recognition that all behaviour that challenges has a meaning or is functional for the person who is presenting with it. Intervention is then based on this identified function as opposed to the topography of behaviour. This individual focus is reflected in the content of empirical literature in this field where single-case studies rather than RCTs and other group designs are predominant.

When causal factors or functions for behaviour are accurately identified, appropriate interventions can be designed. These may include introducing a system of communication for a person who has not been able to understand what is expected of them or to express his or her needs adequately; there may be a need to educate adults (family or professionals) on ways to provide appropriate stimulation and activity to reduce boredom or it may be a change in the broader environment to prevent distress in an individual.

While behavioural approaches historically rejected the focus on internal physiological events or hypothetical constructs such as thoughts and beliefs, recent approaches have combined behavioural and cognitive methods; these have evolved as cognitive behavioural approaches (CBT). This approach is problem focused but also ‘action oriented’ with the aim of helping a person to select specific strategies to address problems. Another development has been the use of anger management approaches (Novaco, 1986), which involve enhanced recognition of individualised triggers for anger in combination with the teaching of coping skills, and which have been widely used over the last 2 decades . More recently, various approaches to parent training (Sanders et al., 2014; Webster-Stratton, 2012) built on social learning models and originally devised for children with conduct disorder have been developed in the field of learning disability.

11.2 Review question: In people with a learning disability and behaviour that challenges, what are the benefits and potential harms associated with psychosocial interventions

aimed at reducing and managing behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 79. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 79: Clinical review protocol summary for the review of psychosocial interventions aimed at reducing and managing behaviour that challenges

Component	Description
Review question(s)	In people with a learning disability and behaviour that challenges, what are the benefits and potential harms associated with psychosocial interventions aimed at reducing and managing behaviour that challenges? (RQ4.2)
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability and behaviour that challenges.
Intervention(s)	All psychosocial interventions, including a broad range of therapies, such as communication interventions, applied behaviour analysis, positive behaviour support and CBT.
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Targeted behaviour that challenges • Adaptive functioning, including communication skills. • Quality of life. • Service user and carer satisfaction.
Study design	RCTs and systematic reviews.
Note. RCTs = Randomised controlled trials.	

11.2.1 Clinical evidence

11.2.1.1 Parent training versus any control

Fifteen RCTs (N = 819) met the eligibility criteria for this review: Aman 2009 (Aman, 2009), Bagner 2007 (Bagner & Eyberg, 2007), Brightman 1982 (Brightman et al., 1982), Hand 2012 (Hand et al., 2012), Leung 2013 (Leung et al., 2013), McIntyre 2008 (McIntyre, 2008), Oliva 2012 (Oliva et al., 2012), Plant 2007 (Plant & Sanders, 2007), Prieto-Bayard 1986 (Prieto-Bayard & Baker, 1986), Reitzel 2013 (Reitzel et al., 2013), Roberts 2006 (Roberts et al., 2006), Roux 2013 (Roux et al., 2013), Sofronoff 2011 (Sofronoff et al., 2011), Tellegen 2013 (Tellegen & Sanders, 2013), Whittingham 2009 (Whittingham et al., 2009). Of the eligible studies, 13 included sufficient data to be included in the evidence syntheses, 1 trial (Prieto-Bayard 1986) included no critical outcome data (N = 20) and 1 trial (Brightman 1982; N = 66) included critical outcomes that could not be included in the meta-analyses because of the way the data had been reported A brief narrative synthesis of Brightman 1982 is given to assess whether the findings support or refute the meta-analyses. An overview of the trials included in the meta-analysis can be found in Table 80.

Summary of findings can be found in Table 81. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of behaviour that challenges (assuming dropouts had not improved)

was conducted. In the sensitivity analysis, all effects remained consistent with the main analysis.

Three studies concerned mixed populations of learning disabled and non-learning disabled participants (Aman 2009, Tellegen 2013, Whittingham 2009). To explore the robustness of the findings, a second sensitivity analysis excluding these 3 studies was conducted. All but 1 effect remained consistent with the main analysis (the removal of Aman 2009 led to insufficient evidence to assess adaptive functioning).

Subgroup analysis was carried out to compare the effectiveness of parent training delivered to individuals with that of parent training delivered to groups. Both sub-groups were shown to be equally effective at reducing targeted behaviour that challenges and increasing carer health and wellbeing.

No data were available for the critical outcomes of quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 80: Study information table for trials included in the meta-analysis of parent training versus any control

	Parent training versus any control
Total no. of studies (N[1])	14 (799)
Study ID	(1) Aman 2009[2] (2) Bagner 2007 (3) Brightman 1982[3,4] (4) Hand 2012 (5) Leung 2013 (6) McIntyre 2008 (7) Oliva 2012 (8) Plant 2007[3] (9) Reitzel 2013 (10) Roberts 2006 (11) Roux 2013 (12) Sofronoff 2011 (13) Tellegen 2013[2] (14) Whittingham 2009[2]
Country	(1 to 3, 6, 9) USA (4) Ireland (5) China (7) Italy (8, 10 to 14) Australia
Diagnosis	(1, 13 to 14) PDD (2) Mild to moderate LD (3) Moderate to severe LD (4, 7) Mild LD (5 to 6, 10 to 12) DD (8) LD (9) Autism
Age (mean)	4-8 (4) Not reported

	Parent training versus any control
Sex (% Female)	15-50 (3, 4, 9) Not reported
Ethnicity (% White)	67-100 (5) 0 (3, 8 to 12, 14) Not reported
IQ (mean)	37-73 (3 to 8, 11 to 14) Not reported
Targeted behaviour that challenges	(1) Irritability (2) Aggression (3 to 14) Not specified
Treatment length (weeks)	8-24 (12) 1
Intervention	(1) Individualised parent training (+ TAU/risperidone) (2) Parent–Child Interaction Therapy (3) Behaviour modification training, 'Steps to Independence' series (4) Parents Plus Children's Programme (5) Triple P Level 4 (6) Incredible Years Parent Training Program-Developmental Disabilities (7) Behavioural parent training (8, 10, 11, 12, 14) Stepping Stones Triple P (9) Functional Behaviour Skills Training program (13) Primary Care Stepping Stones Triple P
Comparison	(1) TAU/risperidone monotherapy (2, 3, 5, 8, 11) Wait list (4, 6, 9, 10, 13, 14) TAU (7, 12) No treatment

Notes: DD = developmental disabilities; LD = learning disability; PDD = pervasive developmental disorder; N = total number of participants; TAU = treatment as usual.
[1] Number randomised.
[2] Study excluded in sensitivity analysis due to mixed sample of learning disabled and non-learning disabled participants.
[3] 3-armed trial; 2 active intervention arms combined in analysis.
[4] Data not reported in a meta-analysable format; findings are described narratively.

1 **Table 81: Summary of findings table for parent training versus any control**

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Any control**	Corresponding risk **Parent training**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.46 standard deviations lower** (0.63 to 0.29 lower)		645 (13 studies)	**moderate**[1]
Targeted behaviour that challenges (severity) - follow-up Follow-up: 26- 52 weeks		The mean targeted behaviour that challenges (severity) - follow-up in the intervention groups was **0.13 standard deviations lower** (0.45 lower to 0.19 higher)		139 (2 studies)	**very low**[1,2,3,4]
Targeted behaviour that challenges (severity, non-improvement) - post-treatment	**883 per 1000**	**592 per 1000** (521 to 680)	**RR 0.67** (0.59 to 0.77)	428 (8 studies)	**moderate**[1]
Targeted behaviour that challenges (frequency) - post-		The mean targeted behaviour that challenges (frequency) - post-treatment in		437 (8 studies)	**low**[1,5]

treatment		the intervention groups was **0.60 standard deviations lower** (0.9 to 0.3 lower)			
Targeted behaviour that challenges (frequency) - follow-up Follow-up: mean 26 weeks		The mean targeted behaviour that challenges (frequency) - follow-up in the intervention groups was **0.36 standard deviations lower** (0.85 lower to 0.14 higher)		64 (1 study)	**very low**[4,6,7]
Targeted behaviour that challenges (frequency, non-improvement) - post-treatment	**948 per 1000**	**597 per 1000** (522 to 692)	**RR 0.63** (0.55 to 0.73)	343 (6 studies)	**low**[1,2]
Adaptive functioning (communication) - post-treatment		The mean adaptive functioning (communication) - post-treatment in the intervention groups was **0.47 standard deviations higher** (0.11 to 0.84 higher)		124 (1 study)	**very low**[2,6,7]
Adaptive functioning (total) - post-treatment		The mean adaptive functioning (total) - post-treatment in the intervention groups was **0.51 standard deviations higher** (0.15 to 0.86 higher)		135 (2 studies)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Most information is from studies at moderate risk of bias
[2] Concerns with applicability - different populations
[3] Optimal information size not met
[4] Publication bias strongly suspected
[5] $I^2 > 40\%$
[6] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[7] Optimal information size not met; small, single study

11.2.1.21 Individual parent training versus group parent training

Two RCTs (N = 144) met the eligibility criteria for this review: Brightman 1982 (Brightman et al., 1982), Chadwick 2001 (Chadwick et al., 2001). Of the 2 eligible studies, 1 trial (N = 78) included sufficient data to be included in the evidence syntheses and 1 trial (N = 53) included critical outcome data that was in a non-meta-analysable format; a brief narrative synthesis is therefore given. An overview of the included trials can be found in Table 82.

Summary of findings can be found in Table 83. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 82: Study information table for trials included in the meta-analysis of head to head parent training interventions

	Individual versus group parent training	Parent + optimism versus parent only training	Enhanced versus standard parent training
Total no. of studies (N[1])	2 (131)	1 (54)	1 (50)
Study ID	(1) Brightman 1982[2,3]	Durand 2013	Plant 2007[2]

	Individual versus group parent training	Parent + optimism versus parent only training	Enhanced versus standard parent training
	(2) Chadwick 2001		
Country	(1) USA (2) UK	USA	Australia
Diagnosis	(1) Moderate to severe LD (2) Severe LD	DD	LD
Age (mean)	(1) 6 (2) 8	4	5
Sex (% Female)	(1, 2) Not reported	15	26
Ethnicity (% White)	(1) Not reported (2) 63	Not reported	Not reported
IQ (mean)	Not reported	Not reported	Not reported
Targeted behaviour that challenges	Not specified	Not specified	Not specified
Treatment length (weeks)	(1) Individual = 12 (1) Group = 12 (2) Individual = 10 (2) Group = 5	Parent + optimism = 8 Parent only = 8	Enhanced = 16 Standard = 10
Intervention(s)	(1) Individual behaviour modification training- 'Steps to Independence' series (1) Group behaviour modification training- 'Steps to Independence' series (2) Individually-based parent training (2) Group based parent training	Optimism training + positive behaviour support Positive behaviour support	Stepping Stones Triple P-Enhanced (SSTP-E) Stepping Stones Triple P-Standard (SSTP-S)

Notes: N = total number of participants; DD = developmental disabilities; LD = learning disability; TAU = treatment as usual.
[1] Number randomised.
[2] 3-armed trial: the 2 active intervention arms were compared in the head to head analysis; waitlist arm excluded.
[3] Data not reported in a meta-analysable format; findings are described narratively.

Table 83: Summary of findings table for individual parent training versus group parent training

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Group parent training**	Corresponding risk **Individual parent training**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.38 standard deviations lower** (1.04 lower to 0.28 higher)		38 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (severity) - follow-up Follow-up: mean 26 weeks		The mean targeted behaviour that challenges (severity) - follow-up in the intervention groups was **0.05 standard deviations lower** (0.7 lower to 0.61 higher)		38 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (frequency) - post-		The mean targeted behaviour that challenges (frequency) - post-treatment in the intervention groups was **0.34 standard deviations lower**		31 (1 study)	**very low**[1,2]

treatment		(1.06 lower to 0.38 higher)		
Targeted behaviour that challenges (frequency) - follow-up Follow-up: mean 26 weeks		The mean targeted behaviour that challenges (frequency) - follow-up in the intervention groups was **0.12 standard deviations higher** (0.59 lower to 0.84 higher)	31 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

11.2.1.31 Parent plus optimism training versus parent training alone

One RCT (N = 54) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Durand 2013 (Durand et al., 2013). An overview of the included study can be found in Table 82.

Summary of findings can be found in Table 84. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

The included study only reported data for completers so a sensitivity analysis for non-improvement of behaviour that challenges (assuming dropouts had not improved) was conducted. In the sensitivity analysis, all effects remained consistent with the main analysis.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 84: Summary of findings table for parent plus optimism training versus parent training alone

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk	Corresponding risk			
	Parent training alone	**Parent plus optimism training**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.8 standard deviations lower** (1.49 to 0.11 lower)		35 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (severity, non-improvement) - post-treatment	**647 per 1000**	**278 per 1000** (123 to 634)	**RR 0.43** (0.19 to 0.98)	35 (1 study)	**very low**[1,2]
Carer satisfaction - post-treatment		The mean carer satisfaction - post-treatment in the intervention groups was **0.22 standard deviations higher** (0.44 lower to 0.89 higher)		35 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 or more criteria sufficient to substantially lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

11.2.1.41 Enhanced parent training versus standard parent training

2 One RCT (N = 75) met the eligibility criteria for this review: Plant 2007 (Plant & Sanders,
3 2007). The included study was composed of 3 arms: 2 active intervention arms and 1 waitlist
4 control arm. Only the active intervention arms were included in the head to head evidence
5 synthesis (N = 50). An overview of the included study can be found in Table 82.

6 Summary of findings can be found in Table 85. The full GRADE evidence profiles and
7 associated forest plots can be found in Appendices O and P.

8 The included study only reported data for completers so a sensitivity analysis for non-
9 improvement of behaviour that challenges (assuming dropouts had not improved) was
10 conducted. In the sensitivity analysis, all but one effect remained consistent with the main
11 analysis: non-improvement in the frequency of behaviour that challenges at 52-week follow-
12 up. When assuming dropouts had not improved, the effect favouring standard training was
13 no longer evident.

14 No data were available for the critical outcomes of adaptive functioning, quality of life or
15 service user satisfaction.

16 The study flow diagram and evidence tables (including methodology checklists) can be found
17 in Appendix N, and exclusion list in Appendix Q.

18 **Table 85: Summary of findings table for enhanced parent training versus standard**
19 **parent training**

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Standard parent training**	Corresponding risk **Enhanced parent training**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.06 standard deviations lower** (0.62 lower to 0.49 higher)		50 (1 study)	**low**[1]
Targeted behaviour that challenges (severity) - follow-up Follow-up: mean 52 weeks		The mean targeted behaviour that challenges (severity) - follow-up in the intervention groups was **0.56 standard deviations lower** (1.18 lower to 0.06 higher)		42 (1 study)	**low**[1]
Targeted behaviour that challenges (severity, non-improvement) - post-treatment	**385 per 1000**	**542 per 1000** (296 to 996)	**RR 1.41** (0.77 to 2.59)	50 (1 study)	**low**[1]
Targeted behaviour that challenges (severity, non-improvement) - follow-up Follow-up: mean 52 weeks	**579 per 1000**	**521 per 1000** (301 to 903)	**RR 0.9** (0.52 to 1.56)	42 (1 study)	**low**[1]
Targeted behaviour that challenges (frequency) - post-treatment		The mean targeted behaviour that challenges (frequency) - post-treatment in the intervention groups was **0.04 standard deviations higher** (0.52 lower to 0.59 higher)		50 (1 study)	**low**[1]
Targeted behaviour that challenges (frequency) - follow-up Follow-up: mean 52 weeks		The mean targeted behaviour that challenges (frequency) - follow-up in the intervention groups was **0.04 standard deviations higher** (0.56 lower to 0.65 higher)		42 (1 study)	**low**[1]
Targeted behaviour that challenges (frequency, non-improvement) - post-treatment	**423 per 1000**	**334 per 1000** (161 to 685)	**RR 0.79** (0.38 to 1.62)	50 (1 study)	**low**[1]

Targeted behaviour that challenges (frequency, non-improvement) - follow-up Follow-up: mean 52 weeks	**211 per 1000**	**347 per 1000** (124 to 979)	**RR 1.65** (0.59 to 4.65)	42 (1 study)	**low**[1]
Carer satisfaction- post-treatment		The mean carer satisfaction- post-treatment in the intervention groups was **0.18 standard deviations higher** (0.38 lower to 0.74 higher)		50 (1 study)	**low**[1]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Optimal information size not met; small, single study

11.2.1.51 Cognitive behavioural intervention versus any control

Seven RCTs (N = 339) met the eligibility criteria for this review: Hagiliassis 2005 (Hagiliassis et al., 2005), McPhail 1989 (McPhail & Chamove, 1989), Nezu 1991 (Nezu, 1991), Singh 2013 (Singh et al., 2013),Taylor 2005 (Taylor et al., 2005), Willner 2002 (Willner et al., 2002), Willner 2013 (Willner et al., 2013). Of the 7 eligible studies, only 4 (N = 281) included sufficient data to be included in the evidence syntheses as 3 trials did not include any critical outcome data (Hagiliassis 2005; McPhail 1989; Willner 2002). An overview of the trials included in the meta-analysis can be found in Table 86.

Summary of findings can be found in Table 87. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of behaviour that challenges (assuming dropouts had not improved) was conducted. In the sensitivity analysis, all effects remained consistent with the main analysis.

No data were available for the critical outcomes of service user or carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 86: Study information table for trials included in the meta-analysis of psychosocial interventions versus any control

	Cognitive behavioural intervention versus any control	Behaviour therapy versus any control
Total no. of studies (N[1])	4 (281)	1 (63)
Study ID	(1) Nezu 1991 (2) Singh 2013 (3) Taylor 2005 (4) Willner 2013	Hassiotis 2009
Country	(1, 2) USA (3 to 4) UK	UK
Diagnosis	Mild LD	LD
Age (mean)	23-38	40
Sex (% Female)	21-36 (3) 0	41
Ethnicity (% White)	(1) 93 (2) 59	95

	Cognitive behavioural intervention versus any control	Behaviour therapy versus any control
	(3, 4) Not reported	
IQ (mean)	57-69 (1, 2) Not reported	Not reported
Targeted behaviour that challenges	(1) Maladaptive social behaviour (2) Aggression (3, 4) Anger	Not specified
Treatment length (weeks)	9-12	26
Intervention	(1) Assertiveness and social problem-solving training (2) Meditation on the Soles of the Feet (3) Cognitive-behavioural anger treatment (4) CBT	Behaviour therapy team (applied behaviour analysis + positive behavioural support)
Comparison	(1, 2) Wait list (3, 4) TAU	TAU
Notes: N = total number of participants; LD = learning disability; TAU = treatment as usual; CBT = cognitive behavioural therapy [1] Number randomised.		

Table 87: Summary of findings table for cognitive behavioural interventions versus any control

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Any control**	Corresponding risk **Cognitive behavioural interventions**			
Targeted behaviour that challenges (severity) - post-treatment Family or carer rated		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.24 standard deviations lower** (0.63 lower to 0.15 higher)		103 (1 study)	**low**[1]
Targeted behaviour that challenges (severity) - follow-up Family or carer rated Follow-up: mean 31 weeks		The mean targeted behaviour that challenges (severity) - follow-up in the intervention groups was **0.03 standard deviations lower** (0.46 lower to 0.4 higher)		83 (1 study)	**low**[1]
Targeted behaviour that challenges (severity, non-improvement) - post-treatment Paid carer rated	**750 per 1000**	**502 per 1000** (292 to 847)	**RR 0.67** (0.39 to 1.13)	38 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (severity) - post-treatment Paid carer rated		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.03 standard deviations lower** (0.48 lower to 0.42 higher)		194 (2 studies)	**low**[3,4]
Targeted behaviour that challenges (severity) - follow-up Paid carer rated Follow-up: 17- 31 weeks		The mean targeted behaviour that challenges (severity) - follow-up in the intervention groups was **0.13 standard deviations lower** (0.58 lower to 0.33 higher)		176 (2 studies)	**low**[3,4]
Adaptive functioning - post-treatment Paid carer rated		The mean adaptive functioning - post-treatment in the intervention groups was **1.32 standard deviations higher** (0.46 to 2.18 higher)		28 (1 study)	**very low**[1,2]
Quality of life - post-treatment		The mean quality of life - post-treatment in the intervention groups was **0.16 standard**		129 (1 study)	**low**[1]

Self rated		**deviations lower** (0.5 lower to 0.19 higher)		
Quality of life - follow-up Self rated Follow-up: mean 31 weeks		The mean quality of life - follow-up in the intervention groups was **0.02 standard deviations lower** (0.35 lower to 0.32 higher)	140 (1 study)	**low**[1]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Optimal information size not met; small, single study
[2] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[3] I2 > 40%
[4] Optimal information size not met

11.2.1.61 Behaviour therapy team versus any control

One RCT (N = 63) of behaviour therapy delivered by a specialist community based team met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Hassiotis 2009 (Hassiotis et al., 2009). An overview of the trials included in the meta-analysis can be found in Table 86.

Summary of findings can be found in Table 88. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or carer and service user satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 88: Summary of findings table for behaviour therapy team versus any control

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk	Corresponding risk			
	Any control	**Behavioural therapy**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.47 standard deviations lower** (0.98 lower to 0.04 higher)		61 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (severity) - follow-up Follow-up: mean 78 weeks		The mean targeted behaviour that challenges (severity) - follow-up in the intervention groups was **0.33 standard deviations lower** (0.85 lower to 0.19 higher)		63 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

11.2.1.73 Psychosocial intervention for sleep problems versus any control

Seven RCTs (N = 389) met the eligibility criteria for this review: Cortesi 2012 (Cortesi et al., 2012), Escalona 2001 (Escalona et al., 2001), Johnson 2013 (Johnson et al., 2013),

Montgomery 2004a (Montgomery et al., 2004), Moss 2014 (Moss et al., 2014), Stores 2004 (Stores & Stores, 2004), Wiggs 1999 (Wiggs & Stores, 1999). Of the 7 eligible studies, 6 (N = 289) included sufficient data to be included in the evidence syntheses and 1 (N = 20) did not include any critical outcome data (Escalona 2001). An overview of the trials included in the meta-analysis can be found in Table 89.

Summary of findings can be found in Table 90. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of behaviour that challenges (assuming dropouts had not improved) and non- satisfied carers (assuming dropouts were not satisfied) was conducted. In the sensitivity analysis, all effects remained consistent with the main analysis.

No data were available for the critical outcomes of adaptive functioning, quality of life and service user satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 89: Study information table for trials included in the meta-analysis of psychosocial interventions for sleep problems versus any control

	Psychosocial intervention versus any control	Face to face versus booklet only
Total no. of studies (N[1])	6 (289)	1 (66)
Study ID	(1) Cortesi 2012[2] (2) Johnson 2013 (3) Montgomery 2004a[3] (4) Moss 2014 (5) Stores 2004 (6) Wiggs 1999	Montgomery 2004a[3]
Country	(1, 2) USA (3, 5 to 6) UK (4) Australia	UK
Diagnosis	(1, 2) Autism (3, 6) Severe LD (4) DD (5) Down Syndrome	Severe LD
Age (mean)	3-12 (3) Not reported	Not reported
Sex (% Female)	(1, 2) 18-21 (3, 5 to 6) 36-52 (4) Not reported	36
Ethnicity (% White)	(1) 99 (2) 73 (3 to 6) Not reported	Not reported
IQ (mean)	Not reported (2) 67	Not reported
Targeted behaviour that challenges	(1 to 6) Sleep problem	Sleep problem
Treatment length (weeks)	(1, 2, 8, 13) 8-13 (3, 5) 1	Face to face = 1 Booklet = 1
Intervention	(1) Cognitive–behavioural therapy (plus Melatonin)[2]	Face-to-face delivered behavioural treatment of sleep

	Psychosocial intervention versus any control	Face to face versus booklet only
	(2) Parent-training (3) Behavioural treatment (4) Sleepwise program (5) Instruction package (6) Tailored behavioural sleep program	problems
Comparison	(1) Melatonin only[2] (2) Attention control (3 to 6) Wait list	Booklet delivered behavioural treatment of sleep problems

Notes: N = total number of participants; LD = learning disability; DD = developmental disabilities; TAU = treatment as usual.
[1] Number randomised.
[2] 4-armed trial: utilised psychosocial intervention + melatonin versus melatonin alone in meta-analysis. The psychosocial only arm and placebo arm were deemed unsuitable comparisons due to the potential 'placebo effect'.
[3] 3-armed trial: the 2 active intervention arms were combined in analyses versus control; waitlist arm not utilised in head to head analyses.

Table 90: Summary of findings table for psychosocial interventions for sleep problems versus any control

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **any control**	Corresponding risk **Sleep interventions**			
Targeted behaviour that challenges (global problem sleep behaviour, non-improvement) - post-treatment	**618 per 1000**	**142 per 1000** (62 to 334)	**RR 0.23** (0.1 to 0.54)	69 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (global problem sleep behaviour) - post-treatment		The mean targeted behaviour that challenges (global problem sleep behaviour) - post-treatment in the intervention groups was **1.05 standard deviations lower** (1.48 to 0.63 lower)		154 (4 studies)	**low**[4,5]
Targeted behaviour that challenges (global problem sleep behaviour) - follow-up Follow-up: 6 to 26 weeks		The mean targeted behaviour that challenges (global problem sleep behaviour) - follow-up in the intervention groups was **0.92 standard deviations lower** (1.6 to 0.24 lower)		130 (3 studies)	**very low**[4,5,6]
Targeted behaviour that challenges (total sleep time) - post-treatment Actigraph		The mean targeted behaviour that challenges (total sleep time) - post-treatment in the intervention groups was **0.62 standard deviations higher** (0.2 to 1.03 higher)		96 (2 studies)	**low**[4,5]
Targeted behaviour that challenges (sleep efficiency) - post-treatment Actigraph		The mean targeted behaviour that challenges (sleep efficiency) - post-treatment in the intervention groups was **0.24 standard deviations higher** (0.26 lower to 0.74 higher)		96 (2 studies)	**low**[4,5]
Targeted behaviour that challenges (total sleep time) - follow-up Actigraph Follow-up: mean 26 weeks		The mean targeted behaviour that challenges (total sleep time) - follow-up in the intervention groups was **0.14 standard deviations higher** (0.44 lower to 0.71 higher)		46 (1 study)	**very low**[1,3]
Targeted behaviour that challenges (sleep efficiency) - follow-up Actigraph Follow-up: mean 26 weeks		The mean targeted behaviour that challenges (sleep efficiency) - follow-up in the intervention groups was **0.11 standard deviations lower** (0.69 lower to 0.46 higher)		46 (1 study)	**very low**[1,3]
Targeted behaviour that		The mean targeted behaviour that challenges		69	

challenges (sleep onset latency) - post-treatment Actigraph		(sleep onset latency) - post-treatment in the intervention groups was **0.59 standard deviations lower** (1.07 to 0.11 lower)		(1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (wake after sleep onset) - post-treatment Actigraph		The mean targeted behaviour that challenges (wake after sleep onset) - post-treatment in the intervention groups was **0.31 standard deviations lower** (1.13 lower to 0.51 higher)		96 (2 studies)	**very low**[4,5,6]
Targeted behaviour that challenges (wake after sleep onset) - follow-up Actigraph Follow-up: mean 26 weeks		The mean targeted behaviour that challenges (wake after sleep onset) - follow-up in the intervention groups was **0.29 standard deviations higher** (0.29 lower to 0.88 higher)		46 (1 study)	**very low**[1,3]
Targeted behaviour that challenges (total sleep time) post-treatment Sleep diary		The mean targeted behaviour that challenges (total sleep time) post-treatment in the intervention groups was **0.3 standard deviations lower** (1.02 lower to 0.42 higher)		30 (1 study)	**very low**[1,3]
Targeted behaviour that challenges (activity score) - post-treatment Sleep diary		The mean targeted behaviour that challenges (activity score) - post-treatment in the intervention groups was **0.28 standard deviations higher** (0.44 lower to 1 higher)		30 (1 study)	**very low**[1,3]
Carer Satisfaction (non-satisfied) - post-treatment	**118 per 1000**	**76 per 1000** (8 to 759)	**RR 0.65** (0.07 to 6.45)	30 (1 study)	**very low**[1,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability- different populations
[3] Optimal information size not met; small, single study
[4] Most information is from studies at moderate risk of bias
[5] Optimal information size not met
[6] I2 > 40%

11.2.1.8 Behavioural intervention for sleep problems delivered face to face versus via written booklet only

Two RCTs (N = 90) met the eligibility criteria for this review: Montgomery 2004a (Montgomery et al., 2004), Montgomery 2004b (Montgomery et al., 2004). Of the 2 eligible studies, 1 (N = 66) included sufficient data to be included in the evidence syntheses and 1 (N = 24) did not include any relevant outcomes (Montgomery 2004b). The included study was composed of 3 arms: 2 active intervention arms and 1 waitlist control arm. Only the active intervention arms were included in the head to head evidence synthesis (N = 42). An overview of the trials included in the meta-analysis can be found in Table 89.

Summary of findings can be found in Table 91. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life and carer or service user satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 91: Summary of findings table for behavioural intervention for sleep problems delivered face to face versus via written booklet only

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk Booklet only	Corresponding risk Face-to-face	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (global problem sleep behaviour) - follow-up Follow-up: mean 26 weeks		The mean targeted behaviour that challenges (global problem sleep behaviour) - follow-up in the intervention groups was **0.07 standard deviations lower** (0.68 lower to 0.53 higher)		42 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

11.2.1.93 Moderators of intervention effectiveness

The evidence search identified 1 systematic review that specifically examined moderators of intervention effectiveness using single-case and small-n (Heyvaert et al., 2012). However, the review did not distinguish between psychological and pharmacological interventions. Therefore, the primary author was invited, and subsequently accepted an offer to conduct a re-analysis for the guideline (labelled here as Heyvaert 2013). The re-analysis included 2 separate analyses: a) psychological interventions (k = 119; N = 238); and b) multi-component interventions (k = 137; N = 269). There was sufficient data to examine, using multi-level meta-analysis, the following predictors of intervention effectiveness: Publication year; study quality; age (in years); gender; diagnosis of autism; target behaviour that challenges – self-injurious behaviour; target behaviour that challenges – stereotyped behaviour; target behaviour that challenges – aggression; target behaviour that challenges – destructive behaviour; target behaviour that challenges – disruptive behaviour; sensory impairment; motor impairment; communicative impairment; and use of functional analysis. The meta-analysis was judged to be of adequate quality because 4 of the 5 methodological quality criteria were met; the search of published primary studies was judged to have been unlikely to identify all relevant studies since many are not published (see Appendix N). With regard to the evidence, because of limitations inherent in single-case and small-n studies (see section 3.5.3), the evidence was graded as very low quality.

Table 92: Study information table for the meta-analysis of moderators of intervention effectiveness

	Heyvaert 2013
Review question/ Aim	Examine moderators of intervention effectiveness for people with a learning disability and behaviour that challenges.
Method used to synthesise evidence	Multi-level meta-analysis (re-analysis of original analysis by categorising studies as psychological or multi-component interventions and conducting meta-analysis for each category separately)
Design of included studies	Single-case and small-n
Dates searched	January 2000 to April 2011
Electronic databases	Eric, Pubmed, and Web of Science (supplemented by hand-searching key journal table of contents and reference lists of

	Heyvaert 2013
	included studies)
No. of included studies (N[1])	Psychological interventions (k = 119; N = 238); multi-component interventions (k = 137; N = 269)
Participant characteristics	People with a learning disability and behaviour that challenges
Intervention	Psychological and multi-component interventions
Comparison	N/A
Outcome	Behaviour that challenges
Review Quality	Adequate
Note. k = number of studies. [1] Number of participants.	

A summary of the included review can be found in Table 92. Further information about the method used can be found in the original paper. The findings from the multi-level meta-analysis can be found in Table 93 and Table 94. In each table, Model 1 is the 3-level random effects regression model without moderators, Model 2 includes all potential moderators, and Model 3 includes only those moderators that were statistically significant in Model 2.

Table 93: Parameter estimates and standard errors for the multilevel meta-analysis of psychological interventions

	Model 1	Model 2	Model 3
Fixed effects			
Mean treatment effect	-2.971 (0.422)***		-3.303 (0.451)***
Moderator effect of:			
Publication year		-0.004 (0.127)	
Study quality		0.0211 (0.367)	
Age		-0.0212 (0.022)	
Gender		-0.540 (0.414)	
Autism		-1.212 (0.405)**	-1.210 (0.347)***
Aggression		1.154 (0.260)***	1.277 (0.182)***
SIB		-0.476 (0.338)	
Stereotyped behaviour		-0.075 (0.812)	
Destructive behaviour		0.112 (0.293)	
Disruptive behaviour		-0.350 (0.349)	
Sensory impairment		1.439 (0.651)*	1.352 (0.640)*
Motor impairment		-0.214 (0.617)	
Communicative impairment		0.671 (0.674)	
Functional analysis		-0.453 (1.415)	
Variance of effect			
Between studies	18.873 (2.906)***	19.916 (3.156) ***	18.414 (2.843)***
Between participants	3.041 (0.441)***	2.9762 (0.476) ***	3.0356 (0.452)***
Residual variance	1.003 (0.0142)***	0.9887 (0.0143) ***	0.9928 (0.0140)***

*Notes: * = p < .05; ** = p < .01; *** = p < .001.*

Table 94: Parameter estimates and standard errors for the multilevel meta-analysis of multi-component interventions

	Model 1	Model 2	Model 3
Fixed effects			
Mean treatment effect	-3.530 (0.404)***		-3.890 (0.412)***
Moderator effect of:			
Publication year		0.028 (0.130)	
Study quality		-0.258 (0.371)	
Age		-0.053 (0.037)	
Gender		-0.026 (0.890)	
Autism		-0.070 (1.049)	
Aggression		1.4883 (0.487)**	0.760 (0.134)***
SIB		0.332 (0.536)	
Stereotyped behaviour		0.414 (0.603)	
Destructive behaviour		0.526 (0.491)	
Disruptive behaviour		0.450 (0.493)	
Sensory impairment		-0.943 (1.959)	
Motor impairment		0.9955 (1.462)	
Communicative impairment		1.474 (1.140)	
Functional analysis		-1.396 (1.045)	
Variance of effect			
Between studies	2.486 (1.288)*	2.295 (1.610)	2.583 (1.317)*
Between participants	35.797 (3.350)***	36.573 (3.680)***	36.117 (3.361)***
Residual variance	1.002 (0.012)***	0.994 (0.0122)***	0.997 (0.0121)***

*Notes: * = p < .05; ** = p < .01; *** = p < .001.*

11.2.2 Economic evidence

11.2.2.1 Systematic literature review

The systematic search of the literature identified 2 studies that assessed the cost effectiveness of psychosocial interventions aimed at reducing and managing behaviour that challenges in people with a learning disability (Felce et al., 2014; Hassiotis et al., 2009). Details on the methods used for the systematic review of the economic literature are described in Chapter 3; full references and evidence tables for all economic evaluations included in the systematic literature review are provided in Appendix S. Completed methodology checklists of the studies are provided in Appendix R. Economic evidence profiles of studies considered during guideline development (i.e. studies that fully or partly met the applicability and quality criteria) are presented in Appendix T.

Hassiotis and colleagues (2011; 2009) evaluated the cost effectiveness of specialist behaviour therapy added to treatment as usual versus treatment as usual alone for the management of behaviour that challenges in adults with a learning disability in the UK. Treatment as usual comprised community learning disabilities teams consisting of psychiatrists, community nurses, occupational therapists, speech and language therapists, physiotherapists and generic psychologists. Teams offered a range of interventions including pharmacotherapy, nursing and enhancement of adaptive skills. The economic analysis was conducted alongside a RCT that was included in the guideline systematic review (Hassiotis 2009). Clinical effectiveness and resource use data were obtained from the study participants (N = 63 for 6 months; 58 for 2-year follow-up). The perspective of the analysis

was the NHS and personal social services. Costs consisted of intervention costs (both specialist behaviour therapy and treatment as usual), costs of non-psychiatric inpatient stays and outpatient appointments, day care and leisure activity costs, costs of adult education and support for voluntary work, costs of contacts with GPs, as well as costs of social workers, community nurses and advocates. National unit costs were used. The primary measure of outcome was the level of behaviour that challenges measured by total and subscale scores on the Aberrant Behavior Checklist (ABC). The duration of the study was 24 months. Outcomes were reported for 6 and 24 months; costs were reported for 2 time periods: 0-6 months & 18-24 months. Discounting was not applied on costs or outcomes.

Over the first 6 months, specialist behaviour therapy was less costly than treatment as usual, although no statistical significance was reached (total mean cost per person was £1,415 for specialist behaviour therapy and £3,615 for treatment as usual in likely 2007 prices; cost difference after adjustment for baseline age, gender, level of learning disability, psychotic disorder, affective disorder, pervasive developmental disorder & total ABC score was -£2,900 with 95% CI -£6,788 to £987). The total mean costs per person over 18-24 months (reported after exclusion of non-psychiatric inpatient services) were moderately higher for specialist behaviour therapy (£5,419 versus £4,271 for treatment as usual, cost difference after adjustment -£815m with 95% CI -£5,629 to £3,986). Specialist behaviour therapy was more effective than treatment as usual, as it resulted in a lower transformed total ABC score at both 6 and 24 months, a difference that reached statistical significance. Therefore specialist behaviour therapy added on treatment as usual appeared to be more cost-effective than treatment as usual alone, as it was more effective in the primary outcome at no additional cost.

The study is directly applicable to the NICE decision-making context. Although the measure of outcome was not expressed in QALYs, the intervention was dominant so it was possible to draw conclusions on cost effectiveness despite the absence of QALY estimates. The study was characterised by potentially serious limitations, including the small study sample and the measurement of costs over 2 time periods of 6 months' duration and not over the whole duration of the study, resulting in costs and outcomes being measured over different periods of time.

Felce and colleagues (2014) evaluated the cost effectiveness of manualised group cognitive behavioural intervention versus wait list for the management of behaviour that challenges in adults with a learning disability in the UK. The cognitive behavioural intervention was delivered by day service staff over 12 weeks. The economic analysis was conducted alongside a cluster RCT conducted in the UK that was included in the guideline systematic review (Willner 2013). The study sample comprised 143 adults with minor to moderate learning disability and problem anger (Willner et al., 2013). Resource use data were collected from researchers, service users and home carers over a 12-week period; unit costs were mainly based on national unit costs, while local costs were used for lay therapists. The time horizon of the analysis was 10 months. The perspective of the analysis was that of the NHS and personal social services. Cost elements included intervention (training and delivery), day services, multidisciplinary meetings of staff held to discuss care plans, other community-based professional services, hospital care, medication for the control of aggression or related behaviour that challenges, accommodation, domiciliary support, or respite care. The primary measure of outcome was the provocation Index as completed by service users; this is a measure of felt response to defined hypothetical situations that may provoke anger. Secondary measures included the provocation index completed by key workers; the Profile of Anger Coping Skills (PACS), a measure of anger coping skills, completed by service users and key workers; the PACS imaginal provocation test (PACS-IPT), a measure of response to actual anger-provoking situations completed by service users; aggressive behaviour; mental health; self-esteem; and quality of life.

Mean total costs were similar for the group CBT and wait list (mean weekly cost per person £970 versus £867 in 2011 prices, respectively; adjusted mean difference: £-22 with 95%CI -

£192 to £147, p=0.795). The intervention had similar effectiveness with wait list, as measured by the primary measure of outcome at 10 months. The intervention was more effective than wait list in a number of secondary outcomes, such as key worker-reported provocation index, PACS and PACS-IPT; other secondary outcomes were not significantly different between group CBT and wait list. Conclusively, cognitive behavioural intervention was better than wait list in a number of secondary outcomes at no additional cost.

The study is directly applicable to the NICE decision-making context. Although outcomes were not expressed in the form of QALYs, the intervention appeared to be equally effective to or more effective than wait list at no additional cost, so it was possible to draw conclusions on cost effectiveness despite the absence of QALY estimates. The study was characterised by potentially serious limitations, including the relatively small study sample, the measurement of costs over a 12-week period, the fact that costs and outcomes did not refer to the same period of time, and the overall short time horizon of the analysis.

In addition to these studies, cost data were available from 3 small pilot studies examining 3 positive behavioural support services in the UK, which were completed during guideline development (Iemmi et al., unpublished data). Although these data do not provide any information on the cost effectiveness of positive behaviour support services, they offer a first indication of the costs associated with such services in the UK and are thus reported in this section. Cost information has been obtained for 3 Positive Behaviour Support Services in Bristol, Halton and Ealing, respectively. An overview of the findings is provided in Table 95.

The positive behavioural support service in Bristol was set up in 2005 and is provided by the North Bristol NHS Trust and funded by a joint commissioning group including the Local Authority social care and special education needs commissioners, and the Clinical Commissioning Group commissioner. Users of the service are children and young people (5-18 years) with a moderate or severe learning disability exhibiting severe levels of behaviour that challenges that are at imminent risk of requiring residential school placements due to school breakdown. The aim of the service is to support the school placements of children and adolescents in the community and to increase the capacity of carers and professionals supporting them. The service, which is led by a clinical psychologist, provides a three-phase intervention comprising assessment, intensive intervention and support, and maintenance/closing case. The intensive intervention and support may include different programmes, for example management of behaviour that challenges, emotional literacy training, functional communication training, continence and self-care, which are individually tailored to children's needs and circumstances and are delivered primarily in special schools. The length and the exact content of the intervention depend on children's individual needs and circumstances. The intervention is provided alongside existing supports, such as short breaks. The mean length of the intervention, estimated based on data from 12 users, was 22 months (range 7 to 42 months). The mean annual cost of the intervention, estimated based on data obtained from 5 users, was £36,405 per child (2012/3 prices). This cost figure includes staff costs (1 clinical psychologist and up to 6 graduate assistant psychologists depending on the child's needs), clinical supervision costs, administrative and travel costs.

The positive behavioural support service in Halton was set up in 2010 and is jointly funded and provided by 3 Local Authorities and Clinical Commissioning Groups (Halton, Knowsley and Saint Helens). Users of the service are children (aged 3 to 17 years) and adults with a moderate or severe learning disability and severe levels of behaviour that challenges. The aim of the service is to maintain people with a learning disability and behaviour that challenges in the community and to increase the coping abilities of carers and professionals supporting them. The service is ran by a management team (comprising an operational director, a clinical supervisor and a principal manager), and an operational team (comprising 5 behaviour analysts, 5 assistant behaviour analysts and 5 support workers). The intervention involves 1 or more of 4 areas of work: early intervention for high risk groups (for example training workshops for carers and professionals working with children and adults with a learning disability and behaviour that challenges); crisis prevention and management

(for example early identification of behaviours that may lead to placement breakdowns); technical support for the most complex cases (for example intensive therapy); placement development (for example returning people in out of area placements to their borough). There are 4 different levels of service response according to the user's level of severity. In people with severe behaviour that challenges, and risk of harm to self or others or risk of placement breakdown (level A), a three-phase service is provided, consisting of assessment, intensive therapy, and maintenance/closing case. In people with severe behaviour that challenges with no risk of harm to self or others or risk of placement breakdown (level B), the service comprises a 1-phase mentoring of professionals from other agencies. In people with moderate behaviour that challenges who are in receipt of care from the appropriate service (level C), the service comprises a one-off consultation for support and advice. In people with moderate behaviour that challenges that are not receiving care from the appropriate service (level D), the service comprises a 1-phase redirection to other services. The length of intervention depends on the individual users' needs. The intervention is provided at home and at school, along with usual care that may include short breaks and residential placements. The estimated mean length of the intervention, based on data from 5 users, was 12 months (range 7 to 18 months). The mean cost of the intervention, as estimated using data from an representative case study, was £14,625 over 15 months (2012/3 prices). This case study comprised an adult requiring level A response. The cost figure includes staff costs (behavioural and assistant behavioural analyst, support worker), clinical supervision costs, administrative and travel costs.

The intensive therapeutic and short break service in Ealing is a collaboration between CAMHS and social care, based within the Ealing Service for Children with Additional Needs and funded by the local authority; the service was first piloted between 2008 and 2009 and provided thereafter. Users of the service are children and adolescents (aged 5 to 17 years) with a learning disability and/or a diagnosis of autism who display severe behavioural challenges, are at imminent risk of requiring a residential placement, and have already been allocated a social worker and receiving short breaks, with family and school both committed to the programme; users must not suffer from acute mental disorders requiring psychiatric hospitalisations. The aim of the programme is to maintain children and young people in the family home and the community and to increase the carer ability to cope. The service is led by a clinical psychologist with social workers allocated to all young people seen within the service. The programme comprises intensive clinical psychology interventions (positive behavioural support, system support, therapeutic interventions) and short breaks. The programme, which is provided in addition to usual care, consists of 4 phases: assessment, intensive therapy, short break and maintenance/closing case. The content of the intervention depends on individual children's needs. The mean length of the programme, estimated based on data from 11 children, was 14 months (range 4 to 27 months). Due to the variability of the interventions provided, the cost of the package of care for the length of the intervention was estimated based on data from 2 case studies: a client with high-level needs and a client with low-level needs. The cost for a person with high-level needs over 5 months of intervention was estimated at £12,301, whereas the cost for a person with low-level needs over 22 months of intervention was estimated at £3,967 (2012/3 prices). These cost figures included staff costs for the intensive clinical psychology interventions (1 clinical psychologist and 1 graduate assistant psychologist), and short break costs.

The above information suggests that there is great variability in costs associated with provision of positive behavioural support services in the UK, depending on the structure and staffing arrangements of the services as well as on the individual users' needs.

Table 95: Overview of 3 positive behavioural support services in the UK (Iemmi et al., unpublished data)

Location	Users	Service	Resource use and cost information (2012/3 prices)

Location	Users	Service	Resource use and cost information (2012/3 prices)
Bristol	Children and young people (5-18 years old) with a moderate or severe learning disability and severe levels of behaviour that challenges, at imminent risk of requiring residential school placements due to school breakdown.	Positive behavioural support 3-phase intervention: assessment; intensive intervention and support; maintenance /closing case. Delivered primarily in special schools. Provided alongside existing supports, such as short breaks.	Intervention delivered by 1 clinical psychologist and up to 5 graduate assistant psychologists Mean length of intervention 22 months (range 7-42, data from 12 users). Mean annual intervention cost £36,405 per child (data from 5 users) Cost figure includes: staff, clinical supervision, administration and travel.
Halton	Children (3 to 17 years old) and adults with a moderate or severe learning disability and severe levels of behaviour that challenges	Positive behavioural support Intervention involves 1 or more of: early intervention for high risk groups; crisis prevention and management; technical support for most complex cases; placement development. 4 levels of service according to user's level of severity: Level A. People with severe behaviour that challenges and risk of harm to self or others or risk of placement breakdown: 3-phase service comprising assessment, intensive therapy, and maintenance/closing case. Level B. People with severe behaviour that challenges with no risk of harm to self or others or risk of placement breakdown: 1-phase mentoring of professionals from other agencies. Level C. People with moderate behaviour that challenges in receipt of care from the appropriate service: one-off consultation for support and advice. Level D. People with moderate behaviour that challenges not receiving care from appropriate service: 1-phase redirection to other services. Intervention provided at home and at school, along with usual care that may include short breaks and residential placements.	Intervention delivered by behavioural and assistant behavioural analyst, and support worker. Mean length of intervention 12 months (range 7-18, data from 5 users). Intervention cost of a representative case study (level A response): £14,625 over 15 months. Cost figure includes: staff, clinical supervision, administration and travel.
Ealing	Children and	Intensive therapeutic and short	Led by a clinical

Location	Users	Service	Resource use and cost information (2012/3 prices)
	adolescents (5 to -17 years old) with a learning disability and/or a diagnosis of autism who display severe behavioural challenges, are at imminent risk of requiring a residential placement, and have already been allocated a social worker and receiving short breaks, with family and school both committed to the programme; users must not suffer from acute mental disorders requiring psychiatric hospitalisations.	break service Programme comprises intensive clinical psychology interventions (positive behavioural support, system support, therapeutic interventions) and short breaks. Provided in addition to usual care 4 phases: assessment, intensive therapy, short break and maintenance /closing case.	psychologist with social workers allocated to all young people. Mean length of programme 14 months (range 4-27, data from 11 children). Cost for a person with high-level needs over 5 months of intervention: £12,301 Cost for a person with low-level needs over 22 months of intervention: £3,967. Cost figures include: staff for the intensive clinical psychology intervention (1 clinical psychologist and 1 graduate assistant psychologist), and short break

11.2.2.2 Economic modelling

Although some limited evidence on the cost effectiveness of cognitive behavioural intervention and behaviour therapy for behaviour that challenges in people with a learning disability is available, the systematic search of the literature identified no economic evidence on parent training as well as on psychosocial interventions for sleep problems. Given the significant resource implications associated with provision of both types of interventions, 2 separate economic models were developed to assess the cost effectiveness of

- Parent training in children and young people with a learning disability and behaviour that challenges
- Psychosocial interventions for sleep problems in children and young people with a learning disability

The study populations in both models were determined by the populations in the RCTs included in the respective systematic literature review undertaken for the guideline.

11.2.2.3 Economic modelling - parent training for children and young people with a learning disability and behaviour that challenges

11.2.2.3.1 Interventions assessed

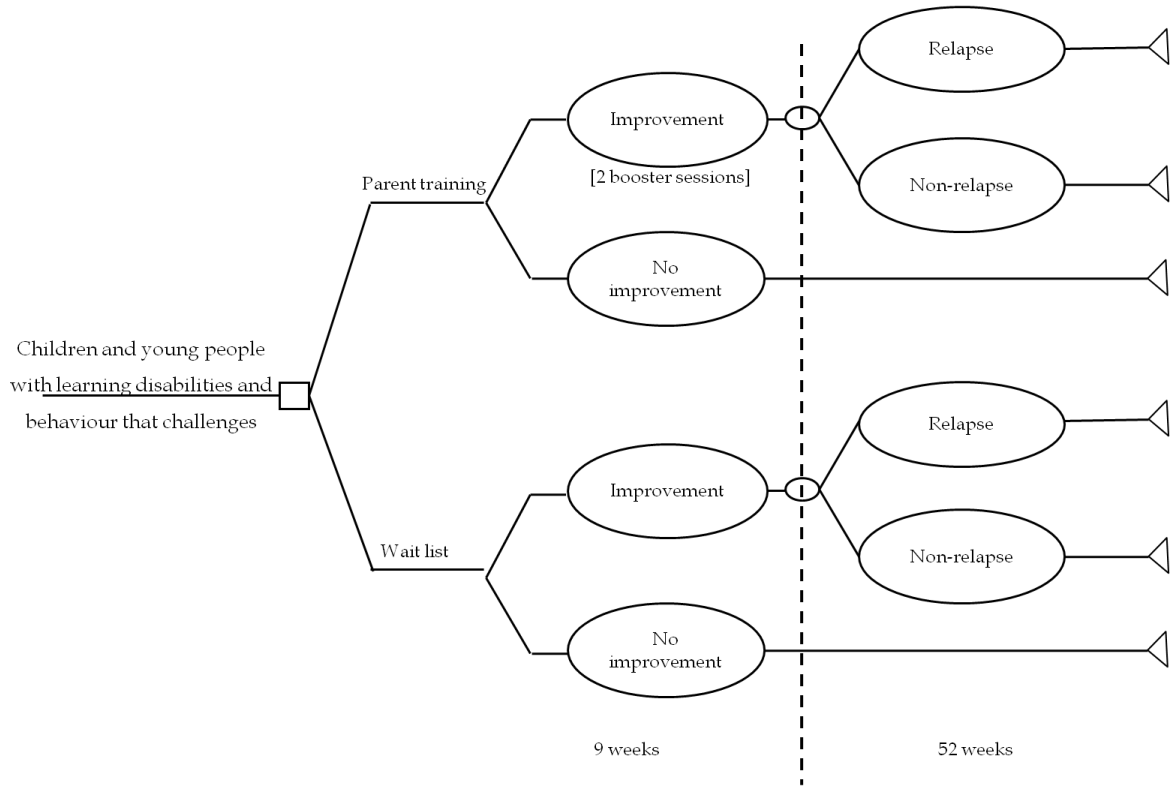
Parent training was compared with wait list. The model considered group parent training because available evidence suggests that there is no difference in the clinical effectiveness between individual and group parent training. Therefore group parent training was selected for modelling as it is more cost-effective than parent training delivered individually (because the intervention cost is lower). Wait list was selected as the comparator as this was the most common control used in the relevant RCTs included in the guideline systematic review. In those RCTs that did not use wait list as a comparator, parent training was predominantly provided in addition to TAU versus TAU alone, so that the control intervention did not incur any extra costs. Therefore, in the vast majority of the RCTs, the comparator was not an

active treatment that would incur extra intervention costs. It should be noted that, ideally, parent training should also be compared with pharmacological interventions that were evaluated in Chapter 12. However, this was not possible as there were no common comparators for parent training and pharmacological interventions that would allow an indirect comparison of their relative effectiveness and, subsequently, the assessment of their relative cost effectiveness: RCTs of parent training for the management of behaviour that challenges in children and young people with a learning disability have mostly used wait list or standard care as a comparator; on the other hand, relevant RCTs of pharmacological interventions has used placebo as control.

11.2.2.3.2 Model structure

A simple decision-tree was constructed using Microsoft Excel 2010 to estimate the cost effectiveness of parent training versus wait list for the management of behaviour that challenges in children and young people with a learning disability. According to the model structure, hypothetical cohorts of families of children and young people with a learning disability and behaviour that challenges received either parent training for 9 weeks or were included in a wait list. At the end of the 9 weeks children and young people either improved in terms of their behaviour that challenges or did not improve. Families of children and young people whose behaviour that challenges improved received 2 booster sessions in the next few months; children and young people whose behaviour that challenges improved could relapse over the following year, or remain improved. Children and young people whose behaviour that challenges did not improve at the end of the first 9 weeks (i.e. at completion of treatment) were conservatively assumed to retain behaviour that challenges over the following year. The time horizon of the model was 61 weeks (9 weeks of treatment and 52 weeks of follow-up). The duration of treatment was consistent with the mean duration of parent training in the RCTs that provided clinical data for the economic analysis. A schematic diagram of the decision-tree is presented in Figure 1.

Figure 1. Schematic diagram of the structure of the economic model evaluating parent training compared with wait list for the management of behaviour that challenges in children and young people with a learning disability



11.2.2.3.3 Costs and outcomes considered in the analysis

The economic analyses adopted the perspective of the NHS and personal social services, as recommended by NICE (NICE, 2012). Costs consisted of intervention costs only, as no data on costs associated with behaviour that challenges in children and young people with a learning disability were identified in the relevant literature. The measure of outcome was the Quality Adjusted Life Year (QALY).

11.2.2.3.4 Clinical input parameters of the economic model

Clinical input parameters included the probability of behaviour that challenges not improving under wait list at 9 weeks, the risk ratio of non-improved behaviour that challenges of parent training versus wait list, and the 1-year probability of relapse to behaviour that challenges.

The guideline systematic review identified 8 RCTs assessing parent training versus wait list for the management of behaviour that challenges in children and young people with a learning disability that reported outcome as improvement in behaviour that challenges regarding its severity (Bagner 2007, Leung 2013, Plant 2007, Roberts 2006, Roux 2013, Sofronoff 2011, Tellegen 2013 and Whittingham 2009). Improvement of behaviour that challenges was defined as a clinically significant change on one of the following scales: the Eyberg Child Behavior Inventory (ECBI) - Problem, the Child Behavior Checklist (CBCL) - Externalising behaviour, or the Developmental Behavior Checklist - Total Behavior Problem (DBC- TBPS). Pooled weighted data from the wait list arms of the 8 RCTs were used to estimate the probability of non-improvement of behaviour that challenges under wait list at 9 weeks, which was utilised in the model. The risk ratio of non-improved behaviour that challenges of parent training versus wait list was derived from meta-analysis these 8 studies.

It must be noted that the economic model utilised the intention-to-treat sensitivity analysis, which assumed that dropouts did not improve.

The 1-year probability of relapse after improvement of behaviour that challenges in children and young people with a learning disability was based on the GDG expert opinion, due to lack of relevant data in the literature. A probability of 0.50 was assumed for parent training and 0.60 for wait list in the base-case analysis. This probability was estimated to be lower in parent training compared with wait list due to the effect of the booster sessions.

11.2.2.3.5 Utility data for estimation of QALYs

In order to express outcomes in the form of QALYs, the health states of the economic model need to be linked to appropriate utility scores. Utility scores represent the Health Related Quality of Life (HRQoL) associated with specific health states on a scale from 0 (death) to 1 (perfect health); they are estimated using preference-based measures that capture people's preferences on the HRQoL experienced in the health states under consideration. Preference-based measures are instruments consisting of a health state classification system, i.e. an instrument that allows determination of the health state of the respondent, and an algorithm that links every health state described by the instrument with a utility score. Utility scores (which express preferences) can be elicited from various population groups (for example, service users, their carers, healthcare professionals or members of the general population). The main methods of valuation are the Visual Analogue Scale (VAS), the Time Trade-Off (TTO) and the Standard Gamble (SG) (Brazier et al., 2007).

The systematic search of the literature identified 3 studies that reported utility scores for children and young people with a learning disability (Carroll & Downs, 2009; Petrou et al., 2010; Petrou & Kupek, 2009). All studies reported utility data relating to a large number of childhood conditions, and provided utility scores associated with the presence of a mild, moderate or severe learning disability without any reference to specific health states within these conditions. These data were not useful in informing the economic model; therefore, these 3 studies were not considered further. In addition to these studies, 1 study was identified (Tilford et al., 2012) that reported utility scores for different health states experienced by children and young people with autism. No information on the IQ of these children was provided in the study; nevertheless, after reviewing the study, the GDG decided to utilise the reported utility data in the economic model as a proxy of the HRQoL of different health states experienced by children and young people with a learning disability.

Tilford and colleagues (2012) reported utility data corresponding to various health states and symptoms associated with autism in children and young people. The study recruited 150 children aged 4-17 years from 2 different sites in the US. All children had a clinical diagnosis of autism meeting DSM-IV-TR criteria (that is, autistic disorder, pervasive developmental disorder not otherwise specified [PDD-NOS] or Asperger's syndrome) and confirmed by scores meeting or exceeding cut-offs for classification with autism on the Autism Diagnostic Observation Schedule (ADOS). Autism-related symptoms (such as sensory issues, social interactions) as well as other behavioural symptoms (such as aggression and hyperactivity) were assessed using the Autism Treatment Network battery. Utility scores were estimated using parents' ratings of their children's HRQoL on the Health Utility Index 3 (HUI3) and the Quality of Well-Being Self-Administered scale (QWB-SA). The HUI is a family of preference-based multi-attribute utility measures (Torrance et al., 1995). The HUI3 health state classification system is the most widely used among the measures of the HUI family, and has been recommended by its developers for the estimation of QALYs in cost-utility analysis. HUI3 covers 8 attributes: cognition, vision, hearing, speech, ambulation, dexterity, emotion and pain; each attribute has 5 or 6 levels of response. Responses to HUI3 can be converted into utility scores using a published algorithm that was developed based on the principles of multi-attribute utility theory, following a valuation survey of members of the general population in Canada; respondents' preferences were elicited using VAS and SG (Feeny et al., 2002). The QWB-SA is an instrument that includes 3 scales of functioning (mobility,

physical activity and social activity) and a measure of 58 symptom and problem complexes; 2 of the symptoms (sexuality and hangovers) were not applicable to younger children with autism and were therefore excluded from the questionnaires. QWB-SA has been valued by 866 community members in the US using VAS (Kaplan & Anderson, 1988).

Table 96 summarises the methods used to derive and value health states associated with autism in children and young people and the resulting utility scores, as reported in Tilford and colleagues (2012). The table includes utility data only for a selection of health states and symptoms of those considered in the study. Health states and symptoms presented in this table are those reflecting or relating closer to states and symptoms considered in economic modelling undertaken for this guideline. The table also includes the level of adjusted statistical significance (p) in the utility scores characterising different severity levels of a symptom. It can be seen that, with the exception of utility scores derived from HUI3 for different severity levels of 'aggression', utility scores based on either HUI3 or QWB-SA can distinguish across different severity levels of all other symptoms included in this table. The authors reported that HUI3 was more sensitive to clinical measures used to characterise children with autism compared with the QWB-SA score and proposed the use of HUI3 for the estimation of QALYs in cost-utility analyses of interventions for children with autism.

Table 96: Summary of methods and utility scores for health states experienced by children and young people with autism

Study	Definition of health states	Valuation method	Population valuing	Health states & corresponding utility scores		
Tilford and colleagues (2012)	HUI3 and QWB-SA profiles of 150 children and young people with autism aged 4-17 years, in the US; profiles constructed for different health states and symptoms associated with autism, based on parents' responses. Diagnosis of autism based on DSM-IV criteria	HUI3 - SG QWB-SA - VAS	504 members of the Canadian general population 866 community members in the US		HUI3 (N = 136)	QWB-SA (N = 140)
				<u>Compulsive behaviours</u>	(p=0.04)	(p=0.02)
				No problem	0.72 (sd 0.19)	0.63 (sd 0.16)
				Minor problem	0.69 (sd 0.23)	0.58 (sd 0.13)
				Moderate problem	0.64 (sd 0.24)	0.58 (sd 0.15)
				Severe problem	0.61 (sd 0.23)	0.53 (sd 0.19)
				<u>Aggression</u>	(p=0.12)	(p=0.03)
				No problem	0.69 (sd 0.21)	0.61 (sd 0.17)
				Minor problem	0.69 (sd 0.22)	0.57 (sd 0.14)
				Moderate problem	0.50 (sd 0.29)	0.49 (sd 0.14)
				Severe problem	0.66 (sd 0.22)	0.55 (sd 0.14)
				<u>Hyperactivity</u>	(p<0.01)	(p=0.03)
				No problem	0.73 (sd 0.26)	0.59 (sd 0.21)
				Mild problem	0.72 (sd 0.20)	0.61 (sd 0.15)
				Moderate problem	0.66 (sd 0.21)	0.61 (sd 0.14)
				Severe problem	0.59 (sd 0.23)	0.52 (sd 0.15)
				<u>Attention span</u>	(p<0.01)	(p<0.01)
				No problem	0.82 (sd 0.14)	0.72 (sd 0.18)
				Mild problem	0.72 (sd 0.19)	0.64 (sd 0.16)
				Moderate problem	0.69 (sd 0.24)	0.57 (sd 0.16)
				Severe problem	0.60 (sd 0.22)	0.55 (sd 0.14)
				<u>Sleep disturbance</u>	(p<0.01)	(p<0.01)
				No problem	0.71 (sd 0.22)	0.64 (sd 0.16)
				Mild problem	0.73 (sd 0.15)	0.55 (sd 0.18)

Study	Definition of health states	Valuation method	Population valuing	Health states & corresponding utility scores		
				Moderate problem	0.55 (sd 0.26)	0.53 (sd 0.12)
				Severe problem	0.61 (sd 0.20)	0.53 (sd 0.11)

1 HUI: Health Utility Index; QWB-SA: Quality of Well-Being Self-Administered Scale; SG: standard gamble; VAS: visual analogue scale

According to NICE guidance on the selection of utility values for use in cost-utility analysis, the measurement of changes in HRQoL should be reported directly from people with the condition examined, and the valuation of health states should be based on public preferences elicited using a choice-based method, such as the TTO or SG, in a representative sample of the UK population. When changes in HRQoL cannot be obtained directly by the people with the condition examined, then data should be obtained from their carers. NICE recommends EQ-5D (Brooks, 1996; Dolan, 1997) for use in cost-utility analyses of interventions for adults; when EQ-5D data are not available, NICE recommends mapping other HRQoL measures to EQ-5D. For economic evaluation of interventions for children, the Institute suggests consideration of alternative standardised and validated preference-based measures of HRQoL that have been designed specifically for use in children (NICE, 2013b).

The study by Tilford and colleagues (2012) provides utility scores based on HUI3 and QWB-SA, but HUI3 appeared to be more sensitive than QWB-SA to clinical measures used to characterise children with autism. Valuation of HUI3 was undertaken using SG, which is a method recommended by NICE, while QWB-SA has been valued using VAS. HUI3 has not been mapped onto EQ-5D in this population. For these reasons the economic models developed for this guideline were populated with HUI3-derived utility scores reported in Tilford and colleagues (2012) for children with autism, which were used as a proxy for children and young people with a learning disability. However, it should be noted that HUI3 has not been designed specifically for use in children. The GDG expressed the opinion that HUI3 is neither directly relevant to the symptoms of children and young people with a learning disability, nor sensitive enough in capturing changes in children's HRQoL. Moreover, HUI3 scores are not directly relevant to the UK context, since valuation was based on the preferences of members of the Canadian population. Nevertheless, given the lack of other appropriate utility data, the utility scores for children with autism derived from HUI3 that were reported in Tilford and colleagues (2012) were used as a proxy for the HRQoL of children and young people with a learning disability in the economic modelling performed to assist development of this guideline.

The guideline economic analysis utilised clinical data on improvement of behaviour that challenges, expressed by a clinically significant change in a number of scales developed to measure this attribute. Tilford and colleagues (2012) reported utility scores corresponding to different levels of aggression, hyperactivity, compulsive behaviour and attention, all of which are related to behaviour that challenges. The changes in utility scores corresponding to different aggression levels were found to be non-significant. Following a review of the available utility data, it was decided to use utility scores for different levels of hyperactivity as a proxy for changes in behaviour that challenges in children and young people with a learning disability. The economic analysis conservatively assumed that at initiation of treatment the HRQoL of the study population corresponded to moderate levels of hyperactivity that improved to mild symptoms following response to treatment. Children that relapsed were assumed to return to the utility score corresponding to moderate symptom levels of hyperactivity. It was assumed that all improvements and decrements in utility occurred linearly between initiation and completion of the 9-week treatment, and between that point and the end of the 52-week follow-up, respectively.

11.2.2.3.6 Cost data

The intervention cost of parent training was calculated by combining relevant resource use (based on data reported in the 8 RCTs included in the guideline systematic review that were considered in the economic analysis) with respective national unit costs, after considering resource use information on group parent training programmes focusing on behaviour management that are available in the UK, as described by Beresford and colleagues (2010). Table 97 presents the details of resource use associated with parent training programmes as reported in each RCT. Table 99 presents an overview of the resource use information

provided by Beresford and colleagues (2010). The economic analysis modelled parent training comprising 8 group sessions lasting 2 hours each; each group was formed by 10 families and was run by a clinical psychologist Band 8a and a mental health nurse Band 5, who acted as co-facilitator. Families whose children showed improvement in their behaviour received another 2 booster group sessions of the same duration. The unit cost for a clinical psychologist band 8a is £134 per hour of client contact (according to Agenda for Change for qualified Allied Health Professionals of the July 2012-June 2013 NHS staff earnings estimates); this cost includes salary, salary oncosts, overheads and capital overheads, but no qualification costs as the latter are not available for clinical psychologists (Curtis, 2013). The unit cost for a mental health nurse band 5 is £74 per hour of face-to-face contact (according to Agenda for Change band 5 of the July 2012-June 2013 NHS staff earnings estimates for qualified nurses); this cost includes salary, salary oncosts, overheads and capital overheads, as well as qualification costs (Curtis, 2013). The intervention cost per child or young person for 8 sessions was estimated at £333 per family (8 sessions x 2 hours x staff unit costs £134+£74 divided by 10 families); when the 2 booster sessions were included, the total intervention cost reached £416.

Table 97: Resource use data reported in RCTs assessing parent training for the management of behaviour that challenges in children and young people with a learning disability that informed the economic model

Study ID	Resource use information
Bagner 2007	12 individual sessions, lasting 60 min each
Leung 2013	6 group sessions lasting 120 min each plus 2 follow-up telephone contacts
Plant 2007	16 individual sessions lasting 60-90 min each
Roberts 2006	10 individual sessions, comprising clinic sessions lasting 120 min each and up to 3-4 home visits lasting 40-60 min each; families with additional needs received a review and feedback session, plus 3 sessions lasting 90 minutes each
Roux 2013	6 group sessions [each group comprising 4-6 families] lasting 120-150 min each and 3 telephone contacts each lasting 15-30 min
Sofronoff 2011	2 seminars lasting 90min each
Tellegen 2013	4 individual sessions lasting 15-105 minutes
Whittingham 2009	5 group sessions [each group comprising 4-5 families]and 4 individual sessions

Table 98. Resource use information on parent training programmes focusing on behaviour management that are available in the UK, as described by Beresford and colleagues (2010)

Programme	Target population	Number / duration of sessions	Group size	Facilitators
ASCEND (ASC – Enhancing Nurture and Development)	Children with autism	11-weekly 2½-hour sessions	Maximum size 20 parents of 8-10 children; best run for parents (≈12-15) of 6-10 children	Qualified therapists (child psychiatrists, clinical psychologists, community psychiatric nurses, etc.) 2 facilitators for groups up to 10; 3-4 for groups >10
Confident Parenting	Children with any disability	6-weekly 2-hour sessions	8 families or 12 participants	3 facilitators drawn from education & clinical psychology (community based learning disability health service)
Cygnet	Children with autism	6-weekly 2½-hour	Maximum 12 parents/carers	2-3 facilitators drawn from range of professional groups including

Programme	Target population	Number / duration of sessions	Group size	Facilitators
		sessions	per group	clinical psychology, education, voluntary sector, and parents
Riding the Rapids	Children with any disability	10-weekly 2-hour sessions	Up to 12 adults per group	1 clinical psychologist, 1 co-facilitator (nurse or teaching staff, input from speech and language therapists)

The intervention cost of wait list was zero. Costs incurred by behaviour that challenges were not included in the analysis due to lack of relevant data, but it is likely that the presence of behaviour that challenges in children and young people with a learning disability incurs considerable additional health and social care costs; such costs may include, for example, costs associated with provision of CAMHS inpatient services, admission to long-term care settings or special education costs.

Table 99 presents the values of all input parameters utilised in the economic model of parent training versus wait list for families of children and young people with a learning disability whose behaviour challenges. As the time horizon of the analysis was 61 weeks, no discounting was necessary.

Table 99. Input parameters utilised in the economic model of parent training versus wait list for the management of behaviour that challenges in children and young people with a learning disability

Input parameter	Deterministic value	Probabilistic distribution	Source of data – comments
Clinical input parameters			
Probability of non-improvement of behaviour that challenges at end of treatment – wait list	0.896	**Beta distribution** α= 199, β= 23	Weighted pooled rate for wait list, guideline meta-analysis (ITT)
Risk ratio of non-improvement of behaviour that challenges, parent training versus wait list	0.72	**Log-normal distribution** 95% CIs: 0.63 to 0.81	Guideline meta-analysis (ITT)
1-year probability of relapse – parent training	0.50	**Beta distribution** α= 50, β= 50	Assumption
1-year probability of relapse – wait list	0.60	α= 60, β= 40	
Utility scores		**Beta distribution**	Tilford et al.,(2012); based on method of moments. Utility score for 'mild hyperactivity' not allowed to fall below that for 'moderate hyperactivity' in the probabilistic model
Mild hyperactivity	0.72	α= 129.92, β= 50.52	
Moderate hyperactivity	0.66	α= 153.82, β= 79.24	
Cost data		**No distributions assigned**	Based on resource use reported in RCTs included in the guideline systematic review (see 11.2.1), relevant information reported in Beresford and colleagues (2010) and the unit costs of clinical psychologist band 8a and mental health nurse band 5 (Curtis, 2013)
Group parent training intervention cost (8 sessions)	£333		
Group parent training – 2 booster sessions	£83		
Wait list intervention cost	£0		

11.2.2.3.7 Handling uncertainty

Model input parameters were synthesised in a probabilistic analysis. This means that model input parameters were assigned probability distributions (rather than being expressed as point estimates), to reflect the uncertainty characterising the available data. Subsequently, 10,000 iterations were performed, each drawing random values out of the distributions fitted onto the model input parameters. Results of the probabilistic analysis (mean costs and QALYs for each intervention) were averaged across the 10,000 iterations. This exercise provides more accurate estimates than those derived from a deterministic analysis (which utilises the mean value of each input parameter ignoring any uncertainty around the mean), by capturing the non-linearity characterising the economic model structure (Briggs et al., 2006).

The probability of non-improvement of behaviour that challenges at completion of treatment (9 weeks) with wait list was assigned a beta distribution. Beta distributions were also assigned to utility values, using the method of moments. The risk ratio of non-improvement of behaviour that challenges for parent training versus wait list was assigned a log-normal distribution. The estimation of distribution ranges was based on the guideline meta-analysis and available data in the published sources of evidence.

The intervention cost of parent training was not assigned a distribution. The cost of group parent training was deemed to be stable and not subject to uncertainty, irrespective of the family's compliance with therapy; this is because participants in a group are not replaced by another person when they occasionally miss one or more sessions or discontinue treatment. Therefore the same resources (in terms of healthcare professional time) are consumed and the full cost of therapy is incurred regardless of whether people attend the full course of treatment or a lower number of group sessions.

Table 99 provides details on the types of distributions assigned to each input parameter and the methods employed to define their range.

In addition, 2 sensitivity analyses were undertaken using the following alternative assumptions:

- parent training was assumed to have a lower risk of relapse (0.40) compared with the base-case scenario (0.50)
- the study population was assumed to have HRQoL corresponding to severe levels of hyperactivity (instead of moderate) at initiation of treatment, as reported in Tilford and colleagues (2012)

11.2.2.3.8 Presentation of the results

Results are presented in the form of the Incremental Cost Effectiveness Ratio (ICER), which is calculated by the following formula:

$$ICER = \Delta C / \Delta E$$

where ΔC and ΔE are the difference in total costs and the difference in effectiveness (QALYs) between 2 interventions, respectively.

In this case the ICER expresses the additional cost per QALY gained associated with provision of parent training in families of children and young people with a learning disability.

In addition, the cost effectiveness acceptability curve (CEAC), which shows the probability of parent training being cost-effective at various cost effectiveness thresholds, including the NICE cost effectiveness thresholds of £20,000 and £30,000/QALY (NICE, 2008), is provided.

Results of the probabilistic analysis are presented in this chapter. Results of the deterministic analysis are provided in Appendix W. Appendix W also provides cost effectiveness planes, showing in graphic form the incremental costs and QALYs of parent training versus wait list.

11.2.2.3.9 Validation of the economic model

The economic model (including the conceptual model and the Excel spreadsheet) was developed by the health economist working on this guideline and checked by a second modeller not working on the guideline. The model was tested for logical consistency by setting input parameters to null and extreme values and examining whether results changed in the expected direction. The results were discussed with the GDG to confirm their plausibility.

11.2.2.3.10 Results

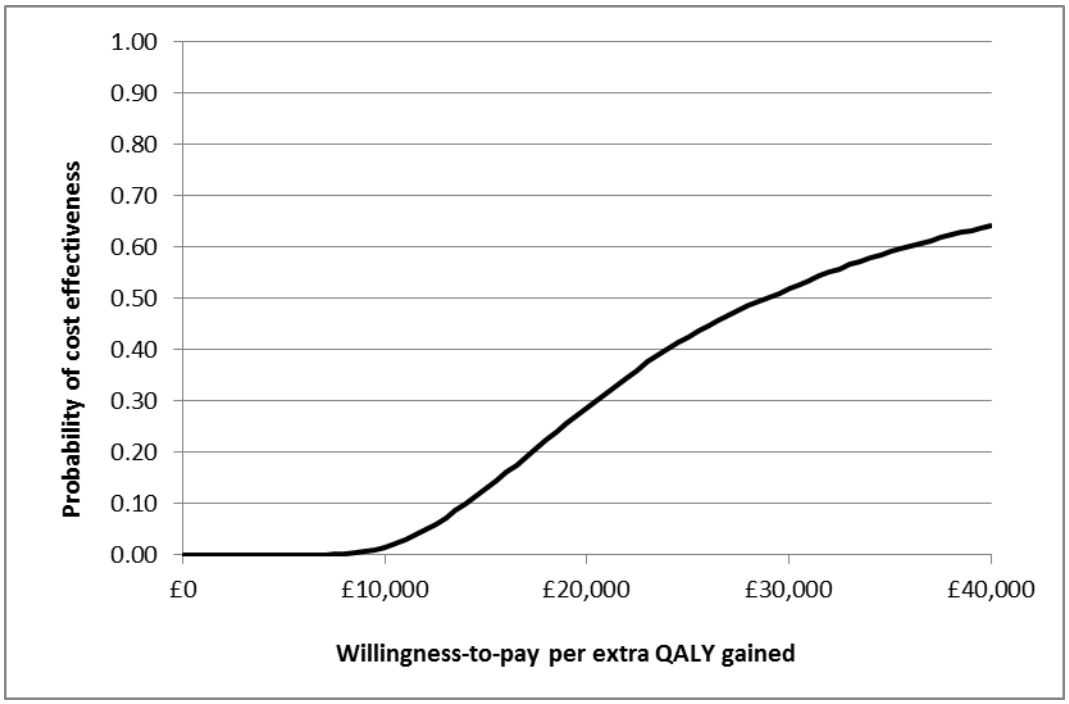
According to the mean probabilistic results, over the 61 weeks of the analysis provision of parent training resulted in 1.33 additional QALYs per 100 children and young people with a learning disability and behaviour that challenges, compared with wait list, at an additional cost of £36,219. The ICER of parent training versus wait list was £27,148/QALY, which is above the lower (£20,000/QALY) but below the upper (£30,000/QALY) NICE cost effectiveness threshold. Full probabilistic results of the base-case economic analysis are presented in Table 100.

Table 100. Mean probabilistic results of economic analysis of parent training for the management of behaviour that challenges in children and young people with a learning disability – mean costs and QALYs for 100 families of children and young people with a learning disability receiving treatment

Intervention	Mean total cost	Mean total QALYs	ICER versus wait list
Group parent training	£36,219	79.28	£27,148/QALY
Wait list	£0	77.94	N/A
Incremental	£36,219	1.33	

The CEAC, shown in Figure 2, suggests that the probability of parent training being cost-effective relative to wait list under the NICE lower and upper cost effectiveness thresholds is 0.29 and 0.52, respectively.

Figure 2. Cost effectiveness acceptability curve of parent training versus wait list for the management of behaviour that challenges in children and young people with a learning disability



Deterministic base case results were overall consistent with probabilistic results. Deterministic results as well the cost effectiveness plane of the analysis are provided in Appendix W.

When a lower risk of relapse over 1 year was assumed for parent training (i.e. 0.40 instead of 0.50), its ICER versus wait list fell at £24,895/QALY and its probability of being cost-effective under the lower and upper NICE cost effectiveness thresholds rose at 0.34 and 0.56, respectively.

When the HRQoL of children and young people was assumed to correspond to severe hyperactivity at initiation of treatment, the ICER versus wait list became £13,037/QALY; the probability of parent training being cost-effective under the lower and upper NICE cost effectiveness thresholds was 0.81 and 0.93, respectively, under this scenario.

11.2.2.3.118 Discussion of findings - limitations of the analysis

The results of the economic model indicate that parent training may be marginally cost-effective for the management of behaviour that challenges in children and young people with a learning disability. However, the cost effectiveness of parent training improves when the long-term benefit is better retained, and, in particular, when the severity of behaviour that challenges is higher at initiation of treatment, as there is more scope for improvement in terms of the children's and young people's HRQoL.

The economic analysis was informed by a meta-analysis of data from 8 RCTs (out of the 14 RCTs included in the respective guideline systematic review) that reported improvement in behaviour that challenges (regarding severity) as a dichotomous outcome. No long-term appropriate follow-up data were available to populate the economic model, and therefore the 1-year probability of relapse following improvement in behaviour that challenges was based on the GDG expert opinion.

Estimation of QALYs was based on utility data derived from HUI3 responses of parents of children with autism in the US; these data were used as a proxy, as no health state-specific utility data for children and young people with a learning disability were identified in the literature. Utility scores for HUI3 have been elicited from members of the Canadian general population and therefore they are not directly applicable to the UK context. More importantly, HUI3 has not been designed for use in children, and may be neither directly relevant to symptoms experienced by children and young people with a learning disability nor adequately sensitive to capture small changes in the HRQoL of this population. Ideally an alternative utility measure should be used for the estimation of QALYs, but at the moment no such measure designed specifically for children and young people with a learning disability and behaviour that challenges is available. Another point for consideration is that the model incorporated exclusively changes in the HRQoL of children and young people with a learning disability and behaviour that challenges. Consideration of the improvement in HRQoL of carers and the family would increase the cost effectiveness of parent training.

The economic model did not include costs associated with the presence of behaviour that challenges in children and young people with a learning disability, due to lack of any relevant data. However, literature suggests that the presence of behaviour that challenges incurs extra costs to health, social and, possibly, educational services (Knapp et al., 2005) and is a common reason for admission to CAMHS inpatient services, long-term care settings or boarding schools; this means that a reduction in the levels of behaviour that challenges as a result of parent training could potentially offset part of (or all) the intervention cost of parent training, so in reality the cost effectiveness of parent training may be considerably higher than that estimated by the guideline economic analysis. It is also likely that the presence of behaviour that challenges in this population incurs extra informal care and other intangible costs to the family, which have not been taken into account in the economic analysis.

Finally, this analysis did not consider other benefits to the family and carers associated with group parent training, arising from meeting with other carers with similar experiences, sharing ideas and receiving peer support.

It should be noted here that the economic analysis modelled only group parent training; individual parent training is less cost-effective, as it is no more effective and incurs higher intervention costs. However, there may be instances where group CBT is not available or not appropriate for some sub-populations, and individual CBT may be the only treatment option to offer.

Taking into account the results and limitations of the analysis, it appears that group parent training may be a cost-effective option for the management of behaviour that challenges in children and young people with a learning disability, especially at more severe levels of behaviour that challenges.

11.2.2.4 Economic modelling – psychosocial and pharmacological interventions for sleep problems in children and young people with a learning disability

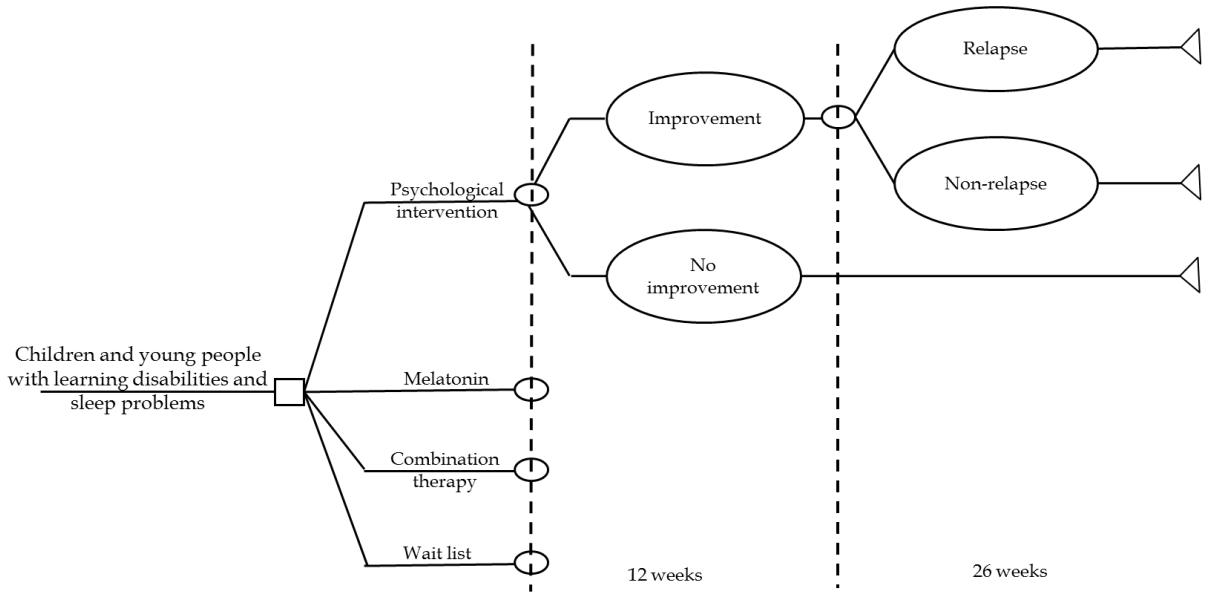
11.2.2.4.1 Interventions assessed

The economic model considered 4 interventions for sleep problems in children and young people with a learning disability: psychosocial intervention, melatonin, combination therapy comprising psychosocial intervention and melatonin, and wait list. Clinical evidence on pharmacological interventions for sleep problems is reported in Chapter 12; however, the detailed methods and results of the economic model for all 4 interventions assessed are provided here for purposes of completeness. The results of the economic analysis that are relevant to pharmacological interventions are summarised in Chapter 12, in the relevant economic section. Wait list was selected as the comparator as this was the most common control used in the relevant RCTs included in the guideline systematic review and still represents standard care in a number of settings.

11.2.2.4.21 Model structure

A simple decision-tree was constructed using Microsoft Excel 2010 to estimate the cost effectiveness of interventions aimed at the management of sleep problems in children and young people with a learning disability. According to the model structure, hypothetical cohorts of children and young people with a learning disability and sleep problems received either psychosocial intervention, melatonin or combination therapy for 12 weeks or were included in a wait list. At the end of the 12 weeks children and young people either experienced an improvement (reduction) in their sleep problems or did not improve. Children and young people whose sleep problems improved could relapse over the following 26 weeks, or remain improved. Children and young people whose sleep problems did not improve at the end of the 12 weeks of therapy were conservatively assumed to retain sleep problems over the following 26 weeks. The time horizon of the model was 38 weeks (12 weeks of treatment and 26 weeks of follow-up). The duration of treatment was consistent with the mean duration of interventions in the RCT that provided most of the clinical data for the economic analysis (Cortesi 2012). A schematic diagram of the decision-tree is presented in Figure 3.

Figure 3. Schematic diagram of the structure of the economic model evaluating psychosocial, pharmacological and combined interventions for the management of sleep problems in children and young people with a learning disability



11.2.2.4.3 Costs and outcomes considered in the analysis

The economic analyses adopted the perspective of the NHS and personal social services, as recommended by NICE (NICE, 2012). Costs consisted of intervention costs only, as no data on costs associated with sleep problems in children and young people with a learning disability was identified in the relevant literature. Moreover, no costs associated with management of side effects of melatonin were incorporated, due to lack of relevant data on the rates of side effects. The measure of outcome was the Quality Adjusted Life Year (QALY).

11.2.2.4.4 *Clinical input parameters of the economic model*

Clinical input parameters included the probability of non-improvement in sleep problems under wait list at 12 weeks, the relative effect of non-improvement in sleep problems for psychosocial intervention versus wait list, the relative risks of non-improvement in sleep problems for melatonin and for combination therapy versus psychosocial intervention, and the 26-week probability of relapse to sleep problems.

No data were available on the probability of non-improvement in sleep problems under wait list, as none of the studies included in the guideline systematic review that used wait list as the control reported dichotomous efficacy data. The only study reporting relevant data was Cortesi 2012, which reported a zero probability of improvement in sleep problems for placebo. The GDG expressed the opinion that this value was rather unrealistic. In the lack of any other relevant data, the economic analysis was run using 4 alternative values for the probability of non-improvement in sleep problems under wait list: 0.900; 0.925; 0.950; and 0.970. The GDG expressed the opinion that the value of non-improvement in sleep problems under wait list is likely to lie within the range of these values.

The guideline systematic review identified 3 RCTs assessing psychosocial intervention versus a non-active control (attention control or wait list) for the management of sleep problems in children and young people with a learning disability, that reported outcomes at the end of the intervention (Johnson 2013, Moss 2014, Wiggs 1999). These studies reported continuous outcomes (global problem sleep outcome), which were summarised in the form of SMD in the guideline meta-analysis. This was subsequently translated into an odds ratio for psychosocial intervention versus wait list using the following formula (Chinn, 2000):

$$\mathrm{LORimprovement} = -\frac{\pi}{\sqrt{3}}\mathrm{SMDimprovement}$$

The probability of non-improvement for psychosocial intervention was subsequently estimated using the following formulae:

$$ODDS_{psych} = (1/OR_{improvement}) * PROB_{WL} / (1 - PROB_{WL})$$

$$PROB_{psych} = ODDS_{psych} / (1 + ODDS_{psych})$$

where $ODDS_{psych}$ the odds for non-improvement of psychosocial intervention; $OR_{improvement}$ the odds ratio of improvement for psychosocial intervention versus wait list, and $PROB_{psych}$ and $PROB_{WL}$ the probability of non-improvement for psychosocial intervention and wait list at end of treatment, respectively.

The risk ratios of non-improvement in sleep problems for melatonin and for combination therapy versus psychosocial intervention were derived from data reported in Cortesi 2012; the economic model utilised the intention-to-treat sensitivity analysis, which assumed that dropouts did not improve.

The 26-week probability of relapse after improvement of sleep problems in children and young people with a learning disability was based on the GDG expert opinion, due to lack of relevant data in the literature. A probability of 0.40 was assumed across all interventions assessed in the economic analysis, following GDG expert opinion.

11.2.2.4.5 *Utility data for estimation of QALYs*

The systematic search of the literature did not identify any studies reporting utility scores for children and young people with a learning disability and sleep problems that are required for the estimation of QALYs in the economic model. However, Tilford and colleagues (2012) reported utility scores for a number of health states relating to symptoms experienced by children and young people with autism, including sleep problems. As described earlier in this

section, given the lack of other appropriate utility data, the GDG decided to utilise the utility data reported by Tilford and colleagues (2012) in the guideline economic modelling as a proxy of the HRQoL of children and young people with a learning disability. Information on the study by Tilford and colleagues (2012) is summarised in Table 101.

The guideline economic analysis utilised data on improvement of global problem sleep behaviour. Tilford and colleagues (2012) reported utility scores corresponding to different levels of sleep problems (no problems, mild problems, moderate problems and severe problems). The utility value for moderate sleep problems was reported to be lower than the utility value for severe sleep problems; the utility value for no sleep problems was reported to be lower than the utility value for mild sleep problems. The economic analysis used the reported utility value for severe sleep problems for children and young people at initiation of treatment, for those not improving and for those relapsing after improvement; and the reported utility value for mild sleep problems for children and young people who improved following intervention. It was assumed that all improvements and decrements in utility occurred linearly between initiation and completion of the 12-week treatment, and between that point and the end of the 26-week follow-up, respectively.

Table 101 presents the values of the clinical and utility input parameters utilised in the economic model of psychosocial, pharmacological and combination therapies for the management of sleep problems in children and young people with a learning disability. As the time horizon of the analysis was 38 weeks, no discounting was necessary.

Table 101. Clinical and utility input parameters utilised in the economic model of psychosocial, pharmacological and combined interventions for the management of sleep problems in children and young people with a learning disability

Input parameter	Deterministic value	Probabilistic distribution	Source of data – comments
Clinical input parameters		**Beta distribution**	
Probability of non-improvement in sleep problems	0.900	α= 39, β= 1	GDG expert opinion due to lack of relevant data; probability distribution based on number of participants in the placebo arm of Cortesi 2012
Wait list (4 scenarios)	0.925	α= 38, β= 2	
	0.950	α= 37, β= 3	
	0.975	α= 36, β= 4	
SMD of improvement – psychosocial intervention versus wait list	-0.85	**Normal distribution** 95% CIs: -1.3 to -0.4	Guideline meta-analysis
Risk ratio of non-improvement		**Log-normal distribution**	Guideline meta-analysis (ITT)
Melatonin versus psychosocial intervention	0.73	95% CIs: 0.58 to 0.92	
Combination therapy versus psychosocial intervention	0.27	95% CIs: 0.16 to 0.47	
26-week probability of relapse – all interventions	0.40	**Beta distribution** α= 40, β= 60	Assumption
Utility scores		**Beta distribution**	Tilford et al., (2012); based on method of moments. Utility score for 'mild sleep problems' not allowed to fall below that for 'severe sleep problems' in the probabilistic model
Mild sleep problems	0.73	α= 178.32, β= 65.96	
Severe sleep problems	0.61	α= 68.32, β= 43.68	

11.2.2.4.61 Cost data

Intervention costs for all therapies were estimated using relevant resource use reported in Cortesi 2012. The other 3 trials that were considered in the economic analysis (Moss 2014, Wiggs 1999 and Johnson 2013) reported information on psychosocial intervention resource use; however, given that the economic analysis was heavily based on the efficacy data reported in Cortesi 2012 and that this study reported detailed resource use data that allowed estimation of the psychosocial intervention cost, it was decided to derive resource use data primarily from this study as well. The psychosocial intervention in Cortesi 2012 was CBT comprising 4 individual sessions lasting 50 minutes each. The study reported 4 additional maintenance sessions that were not considered in the model. Using the unit cost for a clinical psychologist band 8a of £134 per hour of client contact (Curtis, 2013), the mean intervention cost of the psychosocial intervention aiming at managing sleep problems was estimated at £447.

The intervention cost of melatonin was estimated as the sum of the drug acquisition cost and the cost of health professional contacts for monitoring. According to Cortesi 2012, melatonin was administered as controlled release tablets, at a dose of 3mg per day for 12 weeks; monitoring visits lasting 15 minutes each occurred every 2 weeks. In the economic model 3 different formulations of melatonin were tested: modified-release tablets, oral solution and oral suspension. Melatonin oral solution and melatonin oral suspension do not hold a UK product license, and are included in the Drug Tariff under arrangements for payment for Specials and Imported Unlicensed Medicines) (NHS, 2014). Special arrangements for payment of these 2 products were taken into account in the model. Monitoring was estimated to comprise 1 consultant-led paediatrics outpatient visit followed by 5 home visits by community nurses lasting 30 minutes each (150 minutes in total); the unit cost of a consultant-led paediatrics outpatient visit is £172 whereas the unit cost of a community nurse is £70 per hour of home visiting, including travel (Curtis, 2013).

The intervention cost of combination therapy was the sum of melatonin and psychosocial therapy intervention costs. The cost of wait list was zero. Costs associated with sleep problems were not included in the analysis due to lack of relevant data, but it is possible that the presence of sleep problems in children and young people with a learning disability incurs additional health and social care costs, such as GP visits, as well as productivity losses for parents and carers, and intangible costs associated with sleep deprivation, tiredness and lack of energy for the children and young people with a learning disability and sleep problems, their parents and carers.

Table 102 presents the details of resource use, unit costs and total intervention costs of psychosocial, pharmacological and combination therapies for the management of sleep problems in children and young people with a learning disability.

Table 102. Intervention costs of therapies for the management of sleep problems in children and young people with a learning disability

Intervention	Resource use information	Unit cost	Total cost
Psychosocial	4 sessions lasting 50min each	£134/hour	£447
Melatonin 3mg/day	• modified-release tablets • oral solution • oral suspension 1 outpatient paediatrics visit 5 30-min home visits by CN	£65 /12 weeks £211 /12 weeks £410 /12 weeks £172/hour £70/hour	Tablets: £412 Oral solution: £558 Oral suspension: £757
Combination	Sum of resource use for psychosocial intervention (PI) and melatonin (3 formulations, respectively)	As above	PI + tablets: £858 PI + oral solution: £1,005 PI + oral suspension: £1,203

Intervention	Resource use information	Unit cost	Total cost
Wait list	-	N/A	£0
Unit costs taken from (Curtis, 2013) and the (NHS, 2014); CN, community nurse; PI, psychosocial intervention			

11.2.2.4.72 Handling uncertainty

Model input parameters were synthesised in a probabilistic analysis. This means that model input parameters were assigned probability distributions (rather than being expressed as point estimates), to reflect the uncertainty characterising the available data. Subsequently, 10,000 iterations were performed, each drawing random values out of the distributions fitted onto the model input parameters. Results (mean costs and QALYs for each intervention) were averaged across the 10,000 iterations. This exercise provides more accurate estimates than those derived from a deterministic analysis (which utilises the mean value of each input parameter ignoring any uncertainty around the mean), by capturing the non-linearity characterising the economic model structure (Briggs et al., 2006).

The probability of non-improvement of sleep problems at end of treatment (12 weeks) under wait list was assigned a beta distribution. Beta distributions were also assigned to utility values, using the method of moments. The SMD of psychosocial intervention versus wait list was assigned a normal distribution; risk ratios were assigned a log-normal distribution. The estimation of distribution ranges was based on the guideline meta-analysis and available data in the published sources of evidence. Table 103 provides details on the types of distributions assigned to clinical input parameters and utility values and the methods employed to define their range.

Uncertainty in intervention costs was taken into account by assigning different probabilities to the number of monitoring visits (melatonin, combination therapy) or number of sessions (psychosocial intervention, combination therapy) attended by children and young people with a learning disability and sleep problems. These probabilities were determined by completion rates and compliance data reported in Cortesi 2012. The psychosocial intervention had a completion rate of 90%, with completion being defined as having received at least 2 sessions out of the 4. Melatonin had a completion rate also of 90%; non-completers missed administration of more than 20% of the drug. The combination therapy had a completion rate of 95%. The probabilistic distributions that were assigned to the number of visits/sessions of sleep interventions that were determined based on this information are shown in Table 103. In addition to the probabilistic distributions, children and young people receiving melatonin (as monotherapy or in combination with psychosocial therapy) who had only had no or 1 monitoring visit with the community nurse (following 1 outpatient paediatrics visit) were considered to be non-completers and were thus assumed to receive only 50% of the drug.

Table 103. Probabilistic distributions assigned to the number of psychosocial therapy sessions and pharmacological monitoring visits in the economic analysis of interventions for the management of sleep problems in children and young people with a learning disability

Intervention	Probabilistic distributions
Psychosocial	60%: 4 sessions; 30%: 2 or 3 sessions; 10%: 1 session
Melatonin	Distributions apply to community nurse home visits only 50%: 5 visits; 20%: 2 or 3 or 4 visits; 20%: 6 or 7 or 8 visits; 10%: 0 or 1 visits If monitoring visits equal 0 or 1, only 50% of the drug is assumed to be taken
Combination	Psychosocial intervention: 63%: 4 sessions; 32%: 2 or 3 sessions; 5%: 1 session Melatonin:

Intervention	Probabilistic distributions
	Distributions apply to community nurse home visits only 53%: 5 visits; 21%: 2 or 3 or 4 visits; 21%: 6 or 7 or 8 visits; 5%: 0 or 1 visits If monitoring visits equal 0 or 1, only 50% of the drug is assumed to be taken

In addition, a sensitivity analysis was undertaken on the analysis that utilised the 0.900 probability of non-improvement for wait list, using the following alternative assumption:

- the risk of relapse over 26 weeks was concurrently altered for all interventions; a value of zero relapse risk for all interventions and a value of 100% relapse risk for all interventions were tested (instead of the value of 0.40 that was utilised in the base-case scenario)

11.2.2.4.8 Presentation of the results

Results are presented in the form of an incremental analysis, where all options have been ranked from the most to the least effective (in terms of QALYs gained). Options that are dominated by absolute dominance (i.e. they are less effective and more costly than 1 or more other options) or by extended dominance (i.e. they are less effective and more costly than a linear combination of 2 alternative options) are excluded from further analysis. Subsequently, ICERs are calculated for all pairs of consecutive options remaining in analysis.

In addition, results are also presented in the form of net monetary benefits (NMBs) for each intervention. NMB is defined by the following formula:

$$NMB = E * \lambda - C$$

where E and C are the effectiveness (number of QALYs) and costs associated with each intervention, respectively, and λ is the level of the willingness-to-pay per unit of effectiveness, set at the NICE lower cost effectiveness threshold of £20,000/QALY (NICE, 2008). The intervention with the highest NMB is the most cost-effective option (Fenwick et al., 2001).

Finally, the CEAC showing the probability of each intervention being cost-effective at various cost effectiveness thresholds, including the NICE cost effectiveness thresholds of £20,000 and £30,000/QALY, (NICE, 2008) is presented for the analysis utilising a probability of 0.900 for non-improvement under wait list. This is accompanied by the Cost Effectiveness Acceptability Frontier (CEAC), which shows the intervention with the highest mean NMB over different cost effectiveness thresholds, and the probability that this intervention is the most cost-effective among those assessed. The probabilities of cost effectiveness for interventions with the highest NMBs under the lower and upper NICE cost effectiveness thresholds are also provided.

Results of the probabilistic analysis are presented in this chapter. Results of the deterministic analysis are provided in Appendix W. Appendix W also provides cost effectiveness planes, showing in graphic form the incremental costs and QALYs of psychological, pharmacological and combination therapies versus wait list.

11.2.2.4.9 Validation of the economic model

The economic model (including the conceptual model and the Excel spreadsheet) was developed by the health economist working on this guideline and checked by a second modeller not working on the guideline. The model was tested for logical consistency by setting input parameters to null and extreme values and examining whether results changed in the expected direction. The results were discussed with the GDG to confirm their plausibility.

11.2.2.4.10 *Results*

Results of the economic analysis for the 4 scenarios corresponding to the 4 different baseline probabilities of non-improvement under wait list that were utilised in the model are provided in Table 104 and Table 105. Combination therapy is more effective and more costly than any other intervention, followed by melatonin. Psychosocial intervention is the least costly and least effective among active interventions. The results indicate that combination therapy with melatonin being administered in tablets is likely to be the most cost-effective intervention for the management of sleep problems in children and young people with a learning disability, with the exception of the analysis using a 0.900 probability of non-improvement under wait list. Under this scenario the most cost-effective intervention is melatonin in tablets, with the ICER of combination therapy with melatonin in tablets versus melatonin in tablets alone being only slightly above the lower NICE cost effectiveness threshold of £20,000/QALY. At the NICE upper cost effectiveness threshold all active interventions appear to be cost effective compared with standard care, using a 0.900 probability of non-improvement for wait list (according to the cost effectiveness plane presented in Appendix W).

In general, combination therapy with melatonin in tablets and melatonin alone in tablets appear to be cost-effective compared with wait list. Psychosocial intervention and interventions that include melatonin as oral suspension or oral solution (either melatonin monotherapy or combination therapy) do not appear to be cost-effective at the NICE lower cost effectiveness threshold as they rank lower than wait list in terms of cost effectiveness.

The probability of combination therapy (with melatonin in tablets) being cost-effective at the lower NICE cost effectiveness threshold of £20,000/QALY ranged between 39% and 53% (depending on the baseline probability of non-improvement for wait list). At the NICE upper cost effectiveness threshold of £30,000/QALY, combination therapy (with melatonin in tablets) was the most cost-effective intervention with the highest NMB among comparators and a probability of being cost-effective ranging between 63% and 76%. The CEAC and CEAF for the analysis that utilised a 0.900 probability of non-improvement under wait list are shown in Figure 4 and Figure 5, respectively. The CEAC indicates that interventions including melatonin in oral solution or oral suspension had zero probability of being cost effective. The CEAF suggests that at the NICE lower cost effectiveness threshold of £20,000/QALY, melatonin in tablets is the most cost effective intervention, with a probability of being cost effective reaching 28%. At the NICE upper cost effectiveness threshold, combination therapy (melatonin in tablets) appears to be the most cost effective option with a probability of cost effectiveness reaching 63%.

Table 104. Mean probabilistic results of economic analysis of psychosocial, pharmacological and combined interventions for the management of sleep problems in children and young people with a learning disability – mean costs and QALYs per child or young person receiving treatment

Intervention	Probability of non-improvement in sleep problems under wait list									
	0.900					0.925				
	Cost		QALYs		ICER	Cost		QALYs		ICER
	Total	Increm	Total	Increm	(£/QALY)	Total	Increm	Total	Increm	(£/QALY)
Combination – oral suspension	£1,115	£194	0.496	0	Dominated	£1,116	£194	0.495	0	Dominated
Combination – oral solution	£921	£143	0.496	0	Dominated	£922	£143	0.495	0	Dominated
Combination – tablets	£779	£58	0.496	0.019	£20,455	£779	£57	0.495	0.021	£18,683
Melatonin – oral suspension	£721	£189	0.477	0	Dominated	£722	£189	0.474	0	Dominated
Melatonin – oral solution	£532	£139	0.477	0	Dominated	£533	£140	0.474	0	Dominated
Melatonin – tablets	£393	£31	0.477	0.011	£15,496	£393	£31	0.474	0.012	£16,491
Psychosocial intervention	£362	£362	0.466	0.014	Ext dominance	£362	£362	0.462	0.012	Ext dominance
Wait list	£0		0.452		Baseline	£0		0.450		Baseline
Intervention	Probability of non-improvement in sleep problems under wait list									
	0.950					0.975				
	Cost		QALYs		ICER	Cost		QALYs		ICER
	Total	Increm	Total	Increm	(£/QALY)	Total	Increm	Total	Increm	(£/QALY)
Combination – oral suspension	£1,117	£194	0.494	0	Dominated	£1,117	£194	0.497	0	Dominated
Combination – oral solution	£923	£143	0.494	0	Dominated	£923	£143	0.497	0	Dominated
Combination – tablets	£780	£58	0.494	0.023	£17,406	£780	£57	0.497	0.025	£17,393
Melatonin – oral suspension	£722	£189	0.471	0	Dominated	£723	£190	0.469	0	Dominated
Melatonin – oral solution	£533	£139	0.471	0	Dominated	£533	£139	0.469	0	Dominated
Melatonin – tablets	£394	£31	0.471	0.013	Ext dominance	£394	£30	0.469	0.015	Ext dominance
Psychosocial intervention	£364	£363	0.458	0.009	Ext dominance	£364	£364	0.453	0.005	Ext dominance
Wait list	£0		0.449		Baseline	£0		0.447		Baseline

Ext dominance – extended dominance; Increm = incremental

Table 105. Results of probabilistic economic analysis of psychosocial, pharmacological and combined interventions for the management of sleep problems in children and young people with a learning disability – ranking of interventions by Net Monetary Benefit (NMB) per child or young person receiving treatment

Probability of non-improvement in sleep problems under wait list							
0.900		0.925		0.950		0.975	
Intervention	NMB	Intervention	NMB	Intervention	NMB	Intervention	NMB
Melatonin – tablets	£9,153	Combination – tablets	£9,117	Combination – tablets	£9,096	Combination – tablets	£9,061
Combination – tablets	£9,144	Melatonin – tablets	£9,090	Melatonin – tablets	£9,027	Wait list	£8,944
Wait list	£9,039	Wait list	£9,006	Wait list	£8,979	Melatonin – tablets	£8,942
Melatonin – oral solution	£9,014	Combination – oral solution	£8,974	Combination – oral solution	£8,953	Combination – oral solution	£8,918
Combination – oral solution	£9,001	Melatonin – oral solution	£8,950	Melatonin – oral solution	£8,887	Melatonin – oral solution	£8,802
Psychosocial intervention	£8,966	Psychosocial intervention	£8,881	Psychosocial intervention	£8,793	Combination – oral suspension	£8,724
Melatonin – oral suspension	£8,825	Combination – oral suspension	£8,780	Combination – oral suspension	£8,759	Psychosocial intervention	£8,679
Combination – oral suspension	£8,808	Melatonin – oral suspension	£8,761	Melatonin – oral suspension	£8,698	Melatonin – oral suspension	£8,613

Figure 4. Cost effectiveness acceptability curve of sleep interventions for children and young people with a learning disability – using an estimate of 0.900 non-improvement under wait list

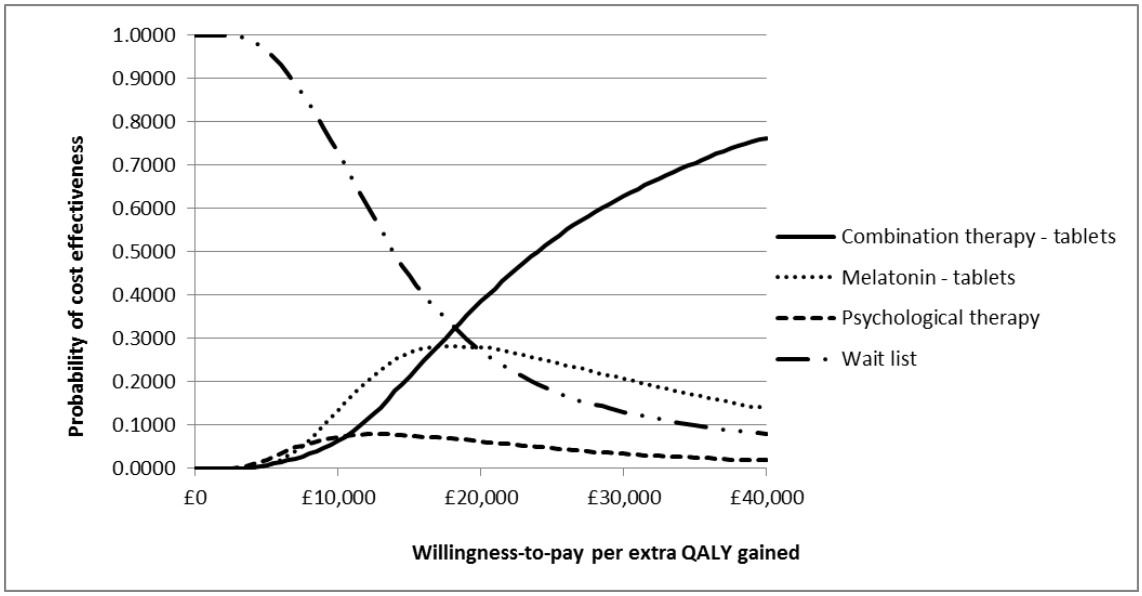
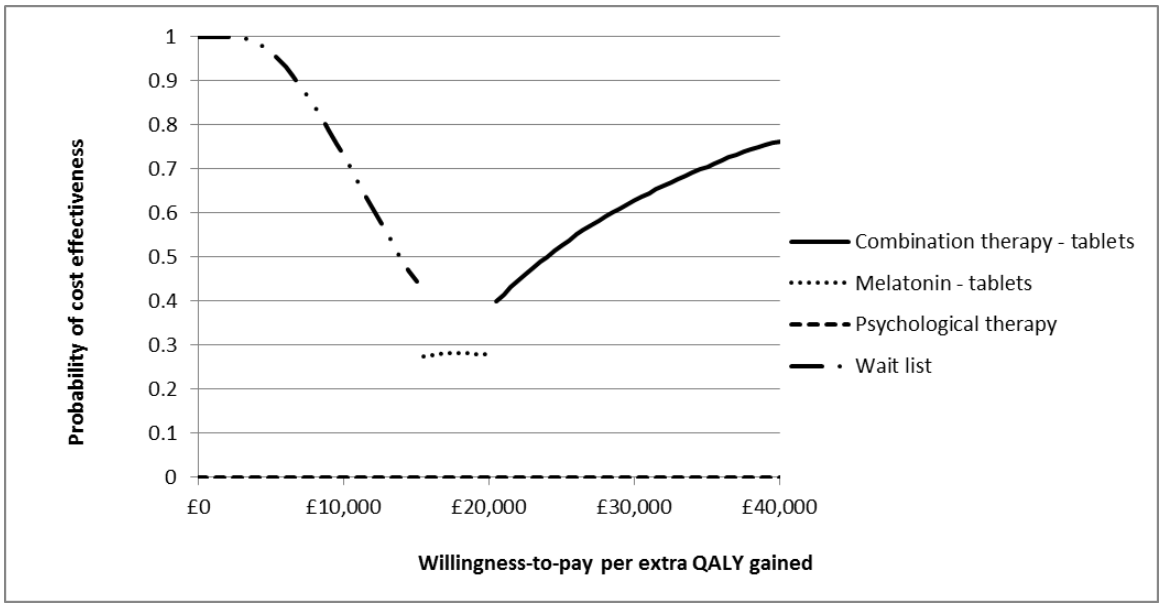


Figure 5. Cost effectiveness acceptability frontier of sleep interventions for children and young people with a learning disability – using an estimate of 0.900 non-improvement under wait list



Deterministic base case results were overall consistent with probabilistic results, although ICERs appeared to be modestly higher. Deterministic results as well the cost effectiveness plane of the analysis for non-improvement under wait list of 0.900 are provided in Appendix W.

When a zero risk of relapse was assumed across all interventions, combination therapy (melatonin in tablets) became the most cost effective intervention at £20,000/QALY, followed by melatonin alone in tablets (ICER of combination therapy versus melatonin £19,971/QALY; ICER of melatonin versus wait list £13,293/QALY; all figures refer to deterministic analysis). At the extreme scenario of all children and young people with sleep problems relapsing following improvement, none of the active interventions was cost effective compared with wait list at the lower NICE cost effectiveness threshold. However, combination therapy and monotherapy with melatonin in tablets were more cost-effective than wait list at the upper NICE cost effectiveness threshold.

11.2.2.4.10 Discussion of findings - limitations of the analysis

The results of the economic model indicate that combination therapy of melatonin in tablets and psychosocial intervention is likely to be cost-effective in the management of sleep problems in children and young people with a learning disability.

The economic analysis was informed by a very limited evidence base: 3 RCTs provided efficacy data on the relative effect of psychosocial intervention versus wait list; relative effects of melatonin and combination therapy were derived from 1 single RCT (Cortesi 2012, 4-armed RCT, N = 160). No long-term follow-up data were available to populate the economic model, and therefore the 26-week probability of relapse following improvement in sleep problems was based on the GDG expert opinion.

Estimation of QALYs was based on utility data derived from HUI3 responses of parents of children with autism in the US; these data were used as a proxy, as no health state-specific utility data for children and young people with a learning disability were identified in the literature. Utility scores for HUI3 have been elicited from members of the Canadian general population and therefore they are not directly applicable to the UK context. More importantly, HUI3 has not been designed for use in children, and may be neither directly relevant to symptoms experienced by children and young people with a learning disability nor adequately sensitive to capture small changes in the HRQoL of this population. Ideally an alternative utility measure should be used for the estimation of QALYs, but at the moment no such measure designed specifically for children and young people with a learning disability and behaviour that challenges is available.

The economic model did not include costs associated with the presence of sleep problems, due to lack of any relevant data. It is possible that the presence of sleep problems in this population incurs extra costs to health and social services; if this is true, then improvement in sleep patterns as a result of sleep interventions could potentially offset part of (or all) the intervention cost, so the cost effectiveness of interventions for the management of sleep problems may be higher than that estimated by the guideline economic analysis. It is also likely that the presence of sleep problems in this population leads to problems in attaining school for the children and young people, productivity losses for the parents, and other intangible costs to the family, which have not been considered in the economic analysis.

The impact of potential side effects from melatonin on costs and HRQoL was not considered in the analysis, due to lack of data on the rates of side effects associated with melatonin and related utility and cost data. Omission of side effects from the model structure may have overestimated the cost effectiveness of melatonin monotherapy and combination therapy.

Taking into account the results and limitations of the analysis, it appears that combination therapy of melatonin in tablets and psychosocial intervention is the most cost-effective option for the management of sleep problems in children and young people with a learning disability. Melatonin alone in tablets is also potentially cost-effective in the management of sleep problems in children and young people with a learning disability.

11.2.3 Clinical evidence statements

11.2.3.1 Parent training versus any control

- Moderate quality evidence from 13 studies (N = 645) suggested that parent training was more effective than control in reducing the severity of targeted behaviour that challenges at the end of intervention.
- Very low quality evidence from 2 studies (N = 139) was inconclusive as to the effectiveness of parent training when compared with control in reducing the severity of targeted behaviour that challenges at up to 52-week follow-up.
- Moderate quality evidence from 8 studies (N = 428) suggested that parent training reduced the risk of not improving the severity of behaviour that challenges at the end of intervention when compared with control.
- Low quality evidence from 8 studies (N = 437) suggested that parent training was more effective than control in reducing the frequency of targeted behaviour that challenges at the end of intervention.
- Very low quality evidence from a single study (N = 64) suggested that parent training was more effective than control in reducing the frequency of targeted behaviour that challenges at 26-week follow-up. However, the precision of this estimate is poor.
- Low quality evidence from 6 studies (N = 343) suggested that parent training reduced the risk of the frequency of behaviour that challenges not being improved at the end of intervention when compared with control.
- Very low quality evidence from up to 2 studies (N = 135) suggested that parent training was more effective than control in increasing communication and adaptive functioning at the end of intervention.
- One trial could not be included in the meta-analysis (N = 66). The authors reported that parent training was more effective than control in reducing targeted behaviour that challenges at end of intervention.

11.2.3.2 Individual parent training versus group parent training

- Very low quality evidence from a single study (N = 31-38) was inconclusive as to the effectiveness of individual parent training, when compared with group parent training, in reducing the severity or frequency of targeted behaviour that challenges at the end of intervention and 26-week follow-up.
- One trial could not be included in the meta-analysis (N = 53). The authors reported no effect of condition on targeted behaviour that challenges at end of intervention or 6-month follow-up.

11.2.3.3 Parent plus optimism training versus parent training alone

- Very low quality evidence from a single study (N = 35) suggested that parent plus optimism training was more effective than parent training alone in reducing the severity of targeted behaviour that challenges at the end of intervention.
- Very low quality evidence from a single study (N = 35) suggested that parent plus optimism training reduced the risk of the severity of behaviour that challenges not being improved at the end of intervention when compared with parent training alone.
- Very low quality evidence from a single study (N = 35) was inconclusive as to the effectiveness of parent plus optimism training, when compared with parent training alone, of increasing carer satisfaction at the end of intervention.

11.2.3.4 Enhanced parent training versus standard parent training

- Very low quality evidence from a single study (N = 50) was inconclusive as to the effectiveness of enhanced parent training, when compared with standard parent training, in reducing the severity of targeted behaviour that challenges at the end of intervention.
- Very low quality evidence from a single study (N = 42) suggested that enhanced parent training was more effective than standard parent training at reducing the severity of targeted behaviour that challenges at 52-week follow-up.
- Low to very low quality evidence from a single study (N = 50) was inconclusive as to the effectiveness of enhanced parent training, when compared with standard parent training, in reducing the risk (of the severity or frequency of behaviour that challenges not being improved) and frequency of targeted behaviour that challenges at the end of intervention and 52-week follow-up.
- Low quality evidence from a single study (N = 50) was inconclusive as to the effectiveness of enhanced parent training, when compared with standard parent training, in increasing carer satisfaction at the end of intervention.

11.2.3.5 Cognitive behavioural intervention versus any control

- When rated by a family member or carer, low quality evidence from a single study (N = 103) suggested that cognitive behavioural intervention was more effective than control at reducing the severity of targeted behaviour that challenges at the end of intervention. However, precision of the estimate is poor and the effect is lost at 31-week follow-up.
- When rated by a paid carer, low quality evidence from 2 studies (N = 194) was inconclusive as to the effectiveness of the cognitive behavioural intervention, when compared with control, in reducing the severity of targeted behaviour that challenges at the end of intervention or up to 31-week follow-up.
- Very low quality evidence from a single study (N = 38) suggested that the cognitive behavioural intervention, when compared with control, reduced the risk of the severity of targeted behaviour that challenges not being improved at end of intervention. However, precision of the estimate is poor.
- Very low quality evidence from a single study (N = 28) suggested that cognitive behavioural intervention was more effective than control in increasing adaptive functioning at the end of intervention.
- Low quality evidence from a single study (N = 129) was inconclusive as to the effectiveness of the cognitive behavioural intervention, when compared with control, in increasing quality of life at both the end of intervention and 31-week follow-up.

11.2.3.6 Behaviour therapy team versus any control

- Very low quality evidence from a single study (N = 61) suggested that the behaviour therapy team was more effective than control in reducing the severity of targeted behaviour that challenges at both end of intervention and 78-week follow-up. However, precision of both estimates was poor.

11.2.3.7 Psychosocial interventions for sleep problems versus any control

- Very low quality evidence from a single study (N = 69) suggested that the psychosocial intervention, when compared with control, reduced the risk of global sleep behaviour not being improved at end of intervention.
- Low quality evidence from up to 4 studies (N = 154) suggested that the psychosocial intervention was more effective than control in reducing global problem sleep behaviour at the end of intervention and up to 26-week follow-up.
- Low quality evidence from up to 2 studies (N = 96) suggested that the psychosocial intervention was more effective than control in increasing actigraph measured total sleep

time at the end of intervention. However, when assessed by carer completed sleep diary and at 26-week follow-up, the evidence was inconclusive.

- Very low quality evidence from 2 studies (N = 96) was inconclusive as to the effectiveness of the psychosocial intervention, when compared with control, in increasing actigraph measured sleep efficiency, and reducing wake after sleep onset, at both the end of intervention and 26-week follow-up.
- Low to very low quality evidence from a single study (N = 69) suggested that the psychosocial intervention was more effective than control in reducing actigraph assessed sleep onset latency at the end of intervention.
- Very low quality evidence from a single study (N = 30) was inconclusive as to the effectiveness of the psychosocial intervention, when compared with control, in reducing night-time activity score at the end of intervention.
- Very low quality evidence from a single study (N = 30) was inconclusive as to the effectiveness of the psychosocial intervention, when compared with control, in reducing the risk of carers being non-satisfied at the end of intervention.

11.2.3.8 Behavioural intervention for sleep problems delivered face to face versus via written booklet only

- Very low quality evidence from a single study (N = 42) was inconclusive as to the effectiveness of the intervention delivered face to face, when compared with booklet only, in reducing problem sleep behaviour at 26-week follow-up.

11.2.3.9 Moderators of intervention effectiveness

- Very low quality evidence from 1 meta-analysis (k = 119; N = 238) suggested that on average the psychological interventions for behaviour that challenges were effective, but the effect varied across participants. Exploring the heterogeneity revealed that psychological interventions were on average less effective for participants with aggression as the type of behaviour that challenges, less effective for participants with a sensory impairment, and more effective for participants with a diagnosis of autism. No other variables, including the use of functional analysis preceding the intervention, were shown to be moderators.
- Very low quality evidence from 1 meta-analysis (k = 137; N = 269) suggested that on average the multi-component interventions for behaviour that challenges were effective, but the effect varied across participants. Exploring the heterogeneity revealed that multi-component interventions were on average less effective for participants with aggression as the type of behaviour that challenges. No other variables, including the use of functional analysis preceding the intervention, were shown to be moderators.

11.2.4 Economic evidence statements

- Low quality evidence from 2 studies (N=206) suggests that psychological interventions (behaviour therapy and CBT) may be cost-effective in the management of behaviour that challenges in adults with a learning disability. Although the evidence is directly applicable to the NICE decision-making context, it is characterised by potentially serious limitations.
- Low quality evidence from 3 pilot studies indicates that there is wide variation in costs associated with provision of positive behavioural support programmes in the UK.
- Low quality evidence from the guideline economic analysis suggests that group parent training for the management of behaviour that challenges in children and young people with a learning disability is potentially cost-effective, especially in children and young people with more severe levels of behaviour that challenges at initiation of treatment.
- Low quality evidence from the guideline economic analysis suggests that combined therapy of melatonin (in tablets) and psychological intervention is potentially the most

cost-effective treatment option for the management of people and young people with a learning disability, according to the guideline economic analysis.

- Melatonin alone in tablets is also potentially cost-effective in the management of sleep problems in children and young people with a learning disability.
- The guideline economic analysis suggests that psychological interventions are not cost-effective for the management of sleep problems in children and young people with a learning disability.
- All guideline economic analyses were characterised by a number of potentially serious limitations relating to limited evidence base (sleep interventions), lack of long-term clinical data, lack of appropriate data on costs associated with behaviour that challenges and sleep problems, omission of the impact of side effects from melatonin on costs and HRQoL, and lack of directly relevant utility data.

11.31 Recommendations and link to evidence

11.3.12 Psychosocial interventions for behaviour that challenges

Recommendations	
	40. Consider parent-training programmes for parents or carers of children with a learning disability who are aged under 12 years and at risk of developing behaviour that challenges. 41. Parent-training programmes should: • be delivered in groups of 10 to 15 parents or carers • be accessible (for example, take place outside normal working hours or in the parent or carer's home or other community-based settings with childcare facilities) • focus on developing communication and social functioning • typically consist of 8 to 12 sessions lasting 90 minutes • follow a developer's manual • employ materials to ensure consistent implementation of the programme. 42. Consider personalised psychosocial interventions that are based on behavioural principles and a functional assessment of behaviour, and consist of: • clear targeted behaviours with agreed outcomes • assessment and modification of environmental factors that could trigger or maintain the behaviour (for example, altering task demands for escape-motivated behaviours and providing a person's preferred member of staff) • addressing staff and family member or carer responses to behaviour that challenges • clearly defined intervention strategies • a clear schedule of reinforcement of desired behaviour and the capacity to offer reinforcement promptly • a specified timescale to meet intervention goals (modifying intervention strategies that do not lead to change within a specified time). 43. Consider individual psychological interventions for adults with an anger management problem. These interventions should be based on cognitive-behavioural principles and delivered individually or in groups over 15–20 hours.

11.3.1.11 Psychosocial interventions for sleep problems

Recommendations	44. Consider behavioural interventions for sleep problems in people with a learning disability and behaviour that challenges that consist of: • a functional analysis of the problem sleep behaviour to inform the intervention (for example, not reinforcing non-sleep behaviours) • structured bedtime routines. 45. Do not offer medication to aid sleep unless the sleep problem persists after a behavioural intervention, and then only: • after consultation with a psychiatrist (or a specialist paediatrician for a child or young person) with expertise in its use in people with a learning disability • together with non-pharmacological interventions and regular reviews (to evaluate continuing need and ensure that the benefits continue to outweigh the risks). If medication is needed to aid sleep, consider melatonin.[f]
Relative values of different outcomes	The GDG specified that all of the following outcomes were critical to decision making: targeted behaviour that challenges, adaptive functioning (including anger control, sleep and communication skills), quality of life, and service user and carer satisfaction.
Trade-off between clinical benefits and harms	The GDG agreed that the evidence generally supports the use of parent training, although long-term follow-up data are needed and there are no data about harms of treatment. The GDG recognised the potential value of early interventions because they equip parents to better manage behaviour so that they may not develop into long-term problems resulting in greater burden for the person, the family and the wider service system. In doing so the GDG drew on their expert knowledge of the good evidence for long-term effects of parent training for children with behavioural problems and the known benefits in other neurodevelopmental disorders (for example, ADHD). In particular, this knowledge was used to provide advice about the group size, number of sessions and other aspects of parent-training programmes. The GDG agreed that based on the evidence and their expert opinion, a personalised psychosocial intervention based on behavioural principles and a functional assessment of behaviour should be offered. In addition, for adults with a learning disability and an anger management problem, consideration should be given to an individual psychological intervention based on CBT. The evidence for psychosocial interventions for sleep and anger management, although of low quality, does support their use for people with a learning disability and behaviour that challenges.
Trade-off between net health benefits and resource use	Limited evidence suggests that psychological interventions may be cost effective in the management of behaviour that challenges in adults with a learning disability.

f This recommendation also appears in section 12.3

	Group parent training is potentially cost effective for the management of behaviour that challenges in children and young people with a learning disability, especially in children and young people with more severe levels of behaviour that challenges at initiation of treatment. Psychological interventions alone are unlikely to be cost effective in the management of sleep problems for a significant number of children and young people with a learning disability; on the other hand, combined therapy of melatonin (in tablets) and psychological intervention appears to be the most cost-effective treatment option for the management of sleep problems in this population. Melatonin alone (in tablets) is also potentially cost effective in the management of sleep problems in children and young people with a learning disability. The GDG considered other benefits resulting from group psychological interventions, such as meeting with other parents and carers experiencing similar situations and exchanging such experiences, sharing ideas and receiving peer support, which was not possible to capture in the guideline economic models. The GDG also considered side effects from melatonin, which were omitted from guideline the economic modelling. The GDG noted that, as costs associated with behaviour that challenges and sleep problems in children and young people with a learning disability (such as costs incurred by health professional contacts, need for special education and residential placements) were not taken into account in the guideline economic models, it was very likely that the cost effectiveness of all interventions versus wait list had been underestimated. Finally, the GDG considered other limitations of the guideline economic analyses, such as the limited evidence base, the lack of long-term clinical data and the lack of directly relevant utility data, which may have affected the results of the economic analyses.
Quality of evidence	Apart from parent training where there is some moderate quality evidence, most evidence was downgraded to low or very low.
Other considerations	In developing the recommendations for sleep problems the GDG carefully considered 2 issues; (1) the problems presented by disturbed sleep for the person with a learning disability and their family and carers throughout the life span, and (2) the need to consider the evidence for the clinical and cost effectiveness of pharmacological interventions for sleep problems (see Chapter 12 and the economic modelling in this chapter). With regard to the first issue, the GDG, drawing on their expert knowledge, decided that it was appropriate to extend the recommendations for the management of sleep problems across the life span and not limit them to children and young people where much of the evidence considered was focused. With regard to the use of medication, and specifically the evidence for superior cost-effectiveness of combined pharmacological and psychological interventions, the GDG was concerned that a recommendation for only combination treatment would mean some people would be reluctant to take up the offer of the interventions and there could be long-term problems in the management of the medication. The GDG therefore decided to first offer a psychological intervention but with combined treatment (with melatonin) as second line if the psychological intervention was not effective.

11.3.2 Research recommendations

4. Are applied behavioural analysis interventions and antipsychotic medication, or a combination of these, effective in reducing the frequency and severity of behaviour that challenges in adults with a learning disability?[g]

g Please note, this research recommendation also appears in section 12.3.1.

12 Pharmacological interventions

12.1 Introduction

Many types of psychotropic medication have been used to manage behaviour that challenges, including antipsychotics, antidepressants, mood stabilisers and sedatives. Despite the diverse underlying aetiologies for the behaviours, medication is mainly utilised in reducing excitation and overt aggression despite the limited evidence for its efficacy in the area of learning disability. The first reports of the use of chlorpromazine in people with a learning disability and behaviour that challenges were published in the 1950s following the successful introduction of antipsychotic medication for the treatment of psychotic disorders. It would appear that a substantial proportion of people with a learning disability in institutional care were in receipt of such medications (Brylewski & Duggan, 2004).

The advent of de-institutionalisation and the implementation of policies encouraging community integration for people with a learning disability may have resulted in some changes in prescribing practice but these are not well understood. However, significant prescribing continues (Robertson et al., 2000), which may be excessive and even unnecessary with long term consequences for the health and wellbeing of the person with a learning disability (Matson et al., 2012; Matson & Neal, 2009).

Antipsychotics are the most frequently prescribed class of psychotropic medication prescribed for as many as two thirds of all people with a learning disability receiving any type of psychotropic medication (Spreat et al., 1997). Local audits and small observational studies of people with a learning disability and developmental disorders who use services suggest that between 21 and 29% may be prescribed antipsychotic medication to manage behaviour that challenges in the absence of a mental disorder such as psychosis or bipolar affective disorder (Doan et al., 2013; Perry et al., 2011). According to a large national audit in the UK, prescription of antipsychotics for behaviour that challenges was significantly higher for those with a more severe learning disability (Paton et al., 2011).

However, some attempts to stop psychotropic medications have shown variable results, with behaviour that challenges re-emerging or discontinuation syndromes being induced (de Leon et al., 2009; Kuijper et al., 2014). There is little evidence for the rates of prescription of other medications such as antidepressants, anxiolytics and mood stabilisers in this population (Deb et al., 2008; Ghosh et al., 2010; Jones et al., 2011).

Although it is accepted that evidence for psychotropic medications in populations with a learning disability and behaviour that challenges is lacking, medication may be used in the long-term if there is intractable and severe aggression or self-injury and where careful monitoring has demonstrated a meaningful benefit that outweighs any harms associated with continuing use.

12.2 Review question: In people with a learning disability and behaviour that challenges, what are the benefits and potential harms associated with pharmacological interventions aimed at reducing and managing behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 106. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 106: Clinical review protocol summary for the review of pharmacological interventions aimed at reducing and managing behaviour that challenges

Component	Description
Review question	In people with a learning disability and behaviour that challenges, what are the benefits and potential harms associated with pharmacological interventions aimed at reducing and managing behaviour that challenges? (RQ4.3)
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability and behaviour that challenges.
Intervention(s)	Pharmacological interventions
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Targeted behaviour that challenges • Adaptive functioning, including communication skills. • Quality of life. • Service user and carer satisfaction. • Adverse events (including sedation/ somnolence/drowsiness, weight outcomes, prolactin level outcomes, seizures, study discontinuation due to adverse events, study discontinuation due to other reasons).
Study design	RCTs and systematic reviews.
Note. RCTs = Randomised controlled trials.	

12.2.1 Clinical evidence

12.2.1.1 Antipsychotics: risperidone versus placebo for behaviour that challenges in children and young people

Five RCTs (N = 355) met the eligibility criteria for this review: Aman 2002 (Aman et al., 2002), Kent 2013 (Kent et al., 2013), RUPP 2002 (Research Units on Pediatric Psychopharmacology (RUPP) Autism Network, 2002), Shea 2004 (Shea et al., 2004), Snyder 2002 (Snyder et al., 2002). All eligible studies included sufficient data to be included in the evidence syntheses. An overview of the trials included in the meta-analysis can be found in Table 107.

Summary of findings can be found in Table 108. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of behaviour that challenges (assuming dropouts had not improved) was conducted. In the sensitivity analysis, all effects remained consistent with the main analysis.

No data were available for the critical outcomes of quality of life or service user and carer satisfaction.

The study flow diagram and evidence tables (including methodology checklists) can be found in Appendix N, and exclusion list in Appendix Q.

Table 107: Study information table for trials included in the meta-analysis of antipsychotics versus placebo in children and young people

	Risperidone versus placebo	Aripiprazole versus placebo
Total no. of studies (N[1])	5 (325)	2 (316)
Study ID	(1) Aman 2002[2]	(1) Marcus 2009[5]

	Risperidone versus placebo	Aripiprazole versus placebo
	(2) Kent 2013[3] (3) RUPP 2002 (4) Shea 2004[2] (5) Snyder 2002[2]	(2) Owen 2009
Country	(1 to 3) USA (4) Canada (5) Worldwide	(1, 2) USA
Diagnosis	(1) Mild to moderate LD (2, 3) Autism (4) PDD & mild to moderate LD (5) Mild to moderate LD[4]	(1, 2) Autism
Age (mean)	7-9	(1) 10 (2) 9
Sex (% Female)	12-34	(1) 11 (2) 12
Ethnicity (% White)	(1, 4, 5) 57-79 (2, 3) Not reported	(1) 71 (2) 74
IQ (mean)	48-70	Not reported
Targeted behaviour that challenges	(1, 4, 5) Conduct problems (2, 3) Irritability	(1, 2) Irritability
Treatment length (weeks)	6-8	(1, 2) 8
Intervention (mean dose; mg/day)	Risperidone (1-1.8)	(1) Aripiprazole (10) (2) Aripiprazole (8.9)
Comparison	Placebo	Placebo

Notes. N = total number of participants; LD = learning disability; mg/day = milligrams per day.
[1] Number randomised.
[2] Meta-analysis based on disaggregated data of participants with IQ ≤ 70, provided upon request from the author.
[3] 3-armed trial: only high dose risperidone and placebo arms utilised.
[4] 2% of participants had borderline intellectual functioning; all others had mild to moderate LD.
[5] Data from high, moderate and low dose conditions combined in meta-analyses.

Table 108: Summary of findings table for risperidone compared with placebo in children and young people

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk Placebo	Corresponding risk Risperidone	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (severity) - post-treatment End-point score		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **1.09 standard deviations lower** (1.39 to 0.79 lower)		257 (4 studies)	**low**[1,2]
Targeted behaviour that challenges (severity) - post-treatment Change score		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.98 standard deviations lower** (1.49 to		66 (1 study)	**very low**[3,4,5]

		0.47 lower)			
Targeted behaviour that challenges (severity, non-improvement) - post-treatment	**850 per 1000**	**357 per 1000** (238 to 544)	**RR 0.42** (0.28 to 0.64)	153 (2 studies)	**low**[1,2]
Adaptive functioning (social) - post-treatment Nisonger Child Behaviour Rating Form - Social Compliance[6]		The mean adaptive functioning (social) - post-treatment in the intervention groups was **0.86 standard deviations higher** (0.42 to 1.3 higher)		155 (3 studies)	**low**[1,2]
Adverse events (elevated prolactin, non-occurrence) - post-treatment	**992 per 1000**	**902 per 1000** (843 to 962)	**RR 0.91** (0.85 to 0.97)	228 (2 studies)	**low**[1,2]
Adverse events (prolactin-related adverse event; oligomenorrhea, non-occurrence) - post-treatment	**1000 per 1000**	**970 per 1000** (890 to 1000)	**RR 0.97** (0.89 to 1.05)	66 (1 study)	**very low**[3,4,5]
Adverse events (prolactin level; ng/ml) - post-treatment		The mean adverse events (prolactin level; ng/ml) - post-treatment in the intervention groups was **3.22 standard deviations higher** (1.68 to 4.75 higher)		241 (3 studies)	**very low**[2,3,4]
Adverse events (weight; kg) - post-treatment Change score		The mean adverse events (weight; kg) - post-treatment in the intervention groups was **0.82 standard deviations higher** (0.57 to 1.06 higher)		282 (3 studies)	**low**[1,2]
Adverse events (weight; kg) - post-treatment Endpoint score		The mean adverse events (weight; kg) - post-treatment in the intervention groups was **0.39 standard deviations higher** (0.16 lower to 0.93 higher)		53 (1 study)	**very low**[3,4,5]
Adverse events (weight gain, non-occurrence) - post-treatment	**993 per 1000**	**904 per 1000** (844 to 954)	**RR 0.91** (0.85 to 0.96)	277 (3 studies)	**very low**[1,2,4]
Adverse events (somnolence/sedation, non-occurrence) - post-treatment	**880 per 1000**	**510 per 1000** (387 to 677)	**RR 0.58** (0.44 to 0.77)	550 (6 studies)	**very low**[1,4,7]
Adverse events (seizure, non-occurrence) - post-treatment	**981 per 1000**	**1000 per 1000** (951 to 1000)	**RR 1.02** (0.97 to 1.08)	101 (1 study)	**very low**[3,5]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**983 per 1000**	**973 per 1000** (944 to 1000)	**RR 0.99** (0.96 to 1.03)	340 (4 studies)	**low**[1,2,4]
Adverse events (discontinuation due other reasons, non-occurrence) - post-treatment	**723 per 1000**	**861 per 1000** (767 to 969)	**RR 1.19** (1.06 to 1.34)	450 (5 studies)	**very low**[1,4,7]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Most information is from studies at moderate risk of bias
[2] Optimal information size not met
[3] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[4] Applicability - different populations
[5] Optimal information size not met; small, single study
[6] Combined adaptive social and compliant/calm subscales
[7] I2 > 40%

12.2.1.2 Antipsychotics: aripiprazole versus placebo for behaviour that challenges in children and young people

Two RCTs (N = 316) met the eligibility criteria for this review: Marcus 2009 (Marcus et al., 2009), Owen 2009 (Owen et al., 2009). All eligible studies included sufficient data to be

included in the evidence syntheses. Marcus 2009 included 3 active intervention arms which were low, high and moderate dose. For the purposes of this review, the 3 groups were combined and compared with the placebo arm.

An overview of the trials included in the meta-analysis can be found in Table 107. Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 109. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of behaviour that challenges (assuming dropouts had not improved) was conducted. In the sensitivity analysis, all effects remained consistent with the main analysis.

No data were available for the critical outcomes of adaptive functioning or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 109: Summary of findings table for aripiprazole compared with placebo in children and young people

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Placebo**	Corresponding risk **Aripiprazole**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.64 standard deviations lower** (0.91 to 0.36 lower)		308 (2 studies)	**very low**[1,2,3]
Targeted behaviour that challenges (severity, non-improvement) - post-treatment	**755 per 1000**	**491 per 1000** (378 to 634)	**RR 0.65** (0.5 to 0.84)	308 (2 studies)	**very low**[1,2,3]
Quality of life - post-treatment		The mean quality of life - post-treatment in the intervention groups was **0.6 standard deviations higher** (0.17 lower to 1.37 higher)		243 (2 studies)	**very low**[1,2,3,4]
Adverse events (elevated prolactin, non-occurrence) - post-treatment	**950 per 1000**	**998 per 1000** (941 to 1000)	**RR 1.05** (0.99 to 1.1)	313 (2 studies)	**very low**[1,2,3]
Adverse events (weight gain; kg) - post- treatment		The mean adverse events (weight gain; kg) - post- treatment in the intervention groups was **0.48 standard deviations higher** (0.17 to 0.8 higher)		216 (1 study)	**very low**[2,5,6]
Adverse events (weight gain; clinically sig., non-occurrence)	**931 per 1000**	**735 per 1000** (661 to 819)	**RR 0.79** (0.71 to 0.88)	313 (2 studies)	**very low**[1,2,3]
Adverse events (sedation, non-occurrence) - post-treatment	**950 per 1000**	**789 per 1000** (722 to 865)	**RR 0.83** (0.76 to 0.91)	313 (2 studies)	**very low**[1,2,3]
Adverse events (seizure, non-occurrence) - post-treatment	**980 per 1000**	**1000 per 1000** (961 to 1000)	**RR 1.03** (0.98 to 1.08)	216 (1 study)	**very low**[2,5,6]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**932 per 1000**	**895 per 1000** (830 to 969)	**RR 0.96** (0.89 to 1.04)	316 (2 studies)	**very low**[1,2,3]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**786 per 1000**	**936 per 1000** (841 to 1000)	**RR 1.19** (1.07 to 1.33)	316 (2 studies)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The

corresponding risk (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Most information is from studies at moderate risk of bias
[2] Applicability - different populations
[3] Optimal information size not met
[4] I2 > 75%
[5] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect.
[6] Optimal information size not met; small, single study

12.2.1.3 Antipsychotics: aripiprazole versus risperidone for behaviour that challenges in children and young people

One RCT (N = 59) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Ghanizadeh 2014 (Ghanizadeh et al., 2014) . An overview of the trial can be found in Table 110. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Summary of findings can be found in Table 111. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 110: Study information table for trials included in the meta-analysis of aripiprazole versus risperidone and olanzapine versus haloperidol in children and young people

	Aripiprazole versus risperidone	Olanzapine versus haloperidol
Total no. of studies (N[1])	1 (59)	1 (12)
Study ID	Ghanizadeh 2013	Malone 2001
Country	Iran	USA
Diagnosis	Autism[2]	PDD + LD[3]
Age (mean)	10	8
Sex (% Female)	19	33
Ethnicity (% White)	Not reported	58
IQ (mean)	Not reported	Not reported
Targeted behaviour that challenges	Irritability	Hyperactivity
Treatment length (weeks)	8	6
Intervention (mean dose; mg/day)	Aripiprazole (5.5)	Olanzapine (10)[4]
Comparison (mean dose; mg/day)	Risperidone (1.1)	Haloperidol (2.5)

Notes. N = total number of participants; LD = learning disability; mg/day = milligrams per day.
[1] Number randomised.
[2] 65% of participants were diagnosed with autism, 13% with Asperger's disorder, 16% PDD-NOS and 2% childhood disruptive behaviour disorder; diagnosis not reported for remainder of sample.
[3] 8% of participants had normal cognitive functioning. All others had mild to severe LD.
[4] Maximum dose.

Table 111: Summary of findings table for aripiprazole compared with risperidone in children and young people

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk Risperidone	Corresponding risk Aripiprazole	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.38 standard deviations higher** (0.14 lower to 0.9 higher)		59 (1 study)	**very low**[1,2,3]
Adverse events (drowsiness, non-occurrence) - post-treatment	**833 per 1000**	**792 per 1000** (617 to 1000)	**RR 0.95** (0.74 to 1.22)	59 (1 study)	**very low**[1,2,3]
Adverse events (seizure, non-occurrence) - post-treatment	**967 per 1000**	**996 per 1000** (909 to 1000)	**RR 1.03** (0.94 to 1.13)	59 (1 study)	**very low**[1,2,3]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**967 per 1000**	**996 per 1000** (909 to 1000)	**RR 1.03** (0.94 to 1.13)	59 (1 study)	**very low**[1,2,3]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**933 per 1000**	**933 per 1000** (812 to 1000)	**RR 1** (0.87 to 1.14)	59 (1 study)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability - different populations
[3] Optimal information size not met; small, single study

12.2.1.43 Antipsychotics: olanzapine versus haloperidol for behaviour that challenges in children and young people

One RCT (N = 12) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Malone 2001(Malone et al., 2001). An overview of the trial can be found in Table 110. Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 112. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 112: Summary of findings table for olanzapine compared with haloperidol in children and young people

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk Haloperidol	Corresponding risk Olanzapine	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (severity) -		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention		12 (1 study)	**very low**[1,2]

post-treatment		groups was**1.4 standard deviations lower** (2.73 to 0.08 lower)			
Adverse events (drowsiness, non-occurrence) - post-treatment	**667 per 1000**	**167 per 1000** (27 to 1000)	**RR 0.25** (0.04 to 1.63)	12 (1 study)	**very low**[1,2]
Adverse events - (weight gain; kg) - post-treatment		The mean adverse events - (weight gain; kg) - post-treatment in the intervention groups was **1.26 standard deviations higher** (0.03 lower to 2.54 higher)		12 (1 study)	**very low**[1,2]
Adverse events (weight gain) - post-treatment	**1000 per 1000**	**850 per 1000** (550 to 1000)	**RR 0.85** (0.55 to 1.31)	12 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 or more criteria sufficient to substantially lower ones confidence in the estimate of effect.
[2] Optimal information size not met; small, single study

12.2.1.5 Antipsychotics: withdrawal of risperidone versus continuation of risperidone for behaviour that challenges in children and young people

One RCT (N = 38) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: RUPP 2005 (Research Units on Pediatric Psychopharmacology (RUPP) Autism Network, 2005). An overview of the trial can be found in Table 113. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Summary of findings can be found in Table 114. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of behaviour that challenges (assuming dropouts had not improved) was conducted. In the sensitivity analysis, all effects remained consistent with the main analysis.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 113: Study information table for trials included in the meta-analysis of withdrawal of antipsychotics versus continuation of antipsychotics in children and young people

	Withdrawal of risperidone versus continuation of risperidone	Withdrawal of aripiprazole versus continuation of aripiprazole
Total no. of studies (N[1])	1 (38)	1 (85)
Study ID	RUPP 2005	Findling 2014
Country	USA	USA
Diagnosis	Autism	Autism
Age (mean)	Not reported	10
Sex (% Female)	Not reported	20
Ethnicity (% White)	Not reported	69

	Withdrawal of risperidone versus continuation of risperidone	Withdrawal of aripiprazole versus continuation of aripiprazole
IQ (mean)	Not reported	Not reported
Targeted behaviour that challenges	Irritability	Irritability
Treatment length (weeks)	8	16
Intervention (mean dose; mg/day)	Withdrawal of risperidone[2]	Withdrawal of aripiprazole[3]
Comparison (mean dose; mg/day)	Continuation of risperidone (2)	Continuation of aripiprazole (9.7)

Notes. N = total number of participants; mg/day = milligrams per day.
[1] Number randomised.
[2]Risperidone maintenance dose reduced by 25% per week over 4 weeks until replaced entirely by placebo on the fourth week.
[3]Participants were switched directly to placebo.

Table 114: Summary of findings table for withdrawal of risperidone compared with continuation of risperidone in children and young people

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Continuation of risperidone**	Corresponding risk **Withdrawal of risperidone**			
Targeted behaviour that challenges (relapse) - post-treatment	**125 per 1000**	**625 per 1000** (162 to 1000)	**RR 5** (1.3 to 19.3)	32 (1 study)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability - different populations
[3] Optimal information size not met; small, single study

12.2.1.63 Antipsychotics: withdrawal of aripiprazole versus continuation of aripiprazole for behaviour that challenges in children and young people

One RCT (N = 85) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Findling 2014 (Findling et al., 2014). An overview of the trial can be found in Table 113. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Summary of findings can be found in Table 115. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 115: Summary of findings table for withdrawal of aripiprazole compared with continuation of aripiprazole in children and young people

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Continuation of aripiprazole**	Corresponding risk **Withdrawal of aripiprazole**			
Targeted behaviour that challenges (relapse) - post-treatment	**341 per 1000**	**522 per 1000** (314 to 871)	**RR 1.53** (0.92 to 2.55)	85 (1 study)	**very low**[1,2,3]
Adverse events (weight gain; clinically sig., non-occurrence)	**951 per 1000**	**980 per 1000** (904 to 1000)	**RR 1.03** (0.95 to 1.12)	85 (1 study)	**very low**[1,2,3]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**1000 per 1000**	**980 per 1000** (920 to 1000)	**RR 0.98** (0.92 to 1.04)	85 (1 study)	**very low**[1,2,3]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**537 per 1000**	**456 per 1000** (295 to 698)	**RR 0.85** (0.55 to 1.3)	85 (1 study)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability - different populations
[3] Optimal information size not met; small, single study

12.2.1.7 Anticonvulsants: topiramate (plus risperidone) versus placebo (plus risperidone) for behaviour that challenges in children and young people

One RCT (N = 40) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Rezaei 2010 (Rezaei et al., 2010). An overview of the trial can be found in Table 116. Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 117. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 116: Study information table for trials included in the meta-analysis of anticonvulsants versus placebo in children and young people

	Topiramate (plus risperidone) versus placebo (plus risperidone)	Valproate versus placebo
Total no. of studies (N[1])	1 (40)	2 (57)
Study ID	Rezaei 2010	(1) Hellings 2005 (2) Hollander 2010
Country	Iran	USA
Diagnosis	Autism	(1) PDD[3] (2) Autism[4]
Age (mean)	8	(1) 11

	Topiramate (plus risperidone) versus placebo (plus risperidone)	Valproate versus placebo
		(2) 9
Sex (% Female)	33	(1) 33 (2) 16
Ethnicity (% White)	Not reported	(1) 90 (2) 30
IQ (mean)	Not reported	(1) 54 (2) 63
Targeted behaviour that challenges	Irritability	(1) Aggression (2) Irritability
Treatment length (weeks)	8	8
Intervention (mean dose; mg/day)	Topiramate (200)[2], Risperidone (2)[2]	(1) Valproate (20)[5] (2) Valproate (375)
Comparison (mean dose; mg/day)	Placebo (N/A), Risperidone (2)[2]	Placebo (N/A)

Notes. N = total number of participants; N/A = not applicable; mg/day = milligrams per day

[1] Number randomised

[2] Maximum dose

[3] 13% of sample had borderline to average intelligence; 87% were diagnosed with a learning disability

[4] 15% of sample had Asperger's syndrome

[5] 20 mg/kg/day

Table 117: Summary of findings table for topiramate (plus risperidone) compared with placebo (plus risperidone) in children and young people

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Placebo plus risperidone**	Corresponding risk **Topiramate plus risperidone**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **1.88 standard deviations lower** (2.63 to 1.12 lower)		40 (1 study)	**very low**[1,2]
Adverse events (sedation, non-occurrence) - post-treatment	**800 per 1000**	**952 per 1000** (744 to 1000)	**RR 1.19** (0.93 to 1.51)	40 (1 study)	**very low**[1,2]
Adverse events (weight at endpoint; kg) - post-treatment		The mean adverse events (weight at endpoint; kg) - post-treatment in the intervention groups was **0.24 standard deviations lower** (0.87 lower to 0.38 higher)		40 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Applicability - different populations

[2] Optimal information size not met; small, single study

12.2.1.81 Anticonvulsants: valproate versus placebo for behaviour that challenges in children and young people

Two RCTs (N = 57) met the eligibility criteria for this review: Hellings 2005 (Hellings et al., 2005), Hollander 2010 (Hollander et al., 2010). All eligible studies included sufficient data to be included in the evidence syntheses. An overview of the trials included in the meta-analysis can be found in Table 116. Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 118. Full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 118: Summary of findings table for valproate compared with placebo in children and young people

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Placebo**	Corresponding risk **Valproate**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.06 standard deviations lower** (0.75 lower to 0.63 higher)		57 (2 studies)	**very low**[1,2,3]
Targeted behaviour that challenges (severity, non-improvement) - post-treatment	**909 per 1000**	**373 per 1000** (191 to 727)	**RR 0.41** (0.21 to 0.8)	27 (1 study)	**very low**[4,5]
Adverse events (weight gain; kg) - post-treatment Change score		The mean adverse events (weight; kg) - post-treatment in the intervention groups was **0.29 standard deviations higher** (0.24 lower to 0.82 higher)		57 (2 studies)	**low**[1,3]
Adverse events (weight gain, non-occurrence) - post-treatment	**714 per 1000**	**564 per 1000** (329 to 971)	**RR 0.79** (0.46 to 1.36)	30 (1 study)	**very low**[4,5]
Adverse events (somnolence/sedation, non-occurrence) - post-treatment	**760 per 1000**	**904 per 1000** (684 to 1000)	**RR 1.19** (0.9 to 1.56)	57 (2 studies)	**low**[1,3]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**1000 per 1000**	**950 per 1000** (830 to 1000)	**RR 0.95** (0.83 to 1.08)	57 (2 studies)	**low**[1,3]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**909 per 1000**	**936 per 1000** (745 to 1000)	**RR 1.03** (0.82 to 1.29)	27 (1 study)	**very low**[4,5]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Most information is from studies at moderate risk of bias
[2] I2 > 40%
[3] Optimal information size not met
[4] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[5] Optimal information size not met; small, single study

12.2.1.91 GABA analogue: piracetam (plus risperidone) versus placebo (plus risperidone) for behaviour that challenges in children and young people

One RCT (N = 40) met the eligibility criteria for this review: Akhondzadeh 2008 (Akhondzadeh et al., 2008). This trial included critical behaviour that challenges outcomes that could not be included in the meta-analyses because of the way the data had been reported; therefore a brief narrative synthesis is given. Data for adverse events are summarised in Table 120.

An overview of the trial can be found in Table 119. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 119: Study information table for trials included in the meta-analysis of piracetam (plus risperidone) versus placebo (plus risperidone) and N-acetylcysteine versus placebo in children and young people

	Piracetam (plus risperidone) versus placebo (plus risperidone)	N-acetylcysteine versus placebo
Total no. of studies (N[1])	1 (40)	1 (33)
Study ID	Akhondzadeh 2008[2]	Hardan 2007
Country	Iran	USA
Diagnosis	Autism	Autism
Age (mean)	7	7
Sex (% Female)	25	7
Ethnicity (% White)	Not reported	Not reported
IQ (mean)	Not reported	Not reported
Targeted behaviour that challenges	Severely disruptive symptoms related to autistic disorder	Irritability
Treatment length (weeks)	10	12
Intervention (maximum dose; mg/day)	Piracetam (800), risperidone (3)	N-acetylcysteine (2700)
Comparison (maximum dose; mg/day)	Placebo (N/A), risperidone (3)	Placebo (N/A)

Notes. N = total number of participants; N/A = not applicable; mg/day = milligrams per day
[1] Number randomised
[2] Data were not reported in a meta-analysable format; findings are described in a narrative summary

Table 120: Summary of findings table piracetam (plus risperidone) versus placebo (plus risperidone) in children and young people

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk Placebo (plus risperidone)	Corresponding risk Piracetam (plus risperidone)	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
Adverse events (drowsiness, non-occurrence) - post-treatment	**550 per 1000**	**649 per 1000** (390 to 1000)	**RR 1.18** (0.71 to 1.97)	40 (1 study)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability - different populations
[3] Optimal information size not met; small, single study

12.2.1.10 Antioxidants: N-acetylcysteine versus placebo for behaviour that challenges in children and young people

One RCT (N = 33) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Hardan 2012 (Hardan et al., 2012) (Hardan et al., 2012). An overview of the trial can be found in Table 119. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Summary of findings can be found in Table 121. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 121: Summary of findings table for N-acetylcysteine compared with placebo in children and young people

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Placebo**	Corresponding risk **N-acetylcysteine (NAC)**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.70 standard deviations lower** (1.46 lower to 0.05 higher)		29 (1 study)	**very low**[1,2,3]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**1000 per 1000**	**930 per 1000** (780 to 1000)	**RR 0.93** (0.78 to 1.11)	33 (1 study)	**very low**[1,2,3]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**667 per 1000**	**933 per 1000** (653 to 1000)	**RR 1.4** (0.98 to 1.99)	33 (1 study)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability - different populations
[3] Optimal information size not met; small, single study

12.2.1.11 Biomedical interventions: omega-3 versus placebo for behaviour that challenges in children and young people

One RCT (N = 13) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Amminger 2007 (Amminger et al., 2007). An overview of the trial can be found in Table 122. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 123. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 122: Study information table for trials included in the meta-analysis of biomedical interventions versus placebo in children and young people

	Omega-3 versus placebo	Ginkgo biloba (plus risperidone) versus placebo (plus risperidone)
Total no. of studies (N[1])	1 (13)	1 (47)
Study ID	Amminger 2007	Hasanzadeh 2012
Country	Austria	Iran
Diagnosis	Autism	Autism
Age (mean)	11	6
Sex (% Female)	0	17
Ethnicity (% White)	Not reported	Not reported
IQ (mean)	Not reported	Not reported
Targeted behaviour that challenges	Irritability	Irritability
Treatment length (weeks)	6	10
Intervention (mean dose; mg/day)	Omega-3 (1500)	Ginkgo biloba (120)[2], risperidone (3)[2]
Comparison (mean dose; mg/day)	Placebo (N/A)	Placebo (N/A), risperidone (3)[2]

Notes. N = total number of participants; N/A = not applicable; mg/day = milligrams per day
[1] Number randomised
[2] Maximum dose

Table 123: Summary of findings table for omega-3 compared with placebo in children and young people

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk Placebo	Corresponding risk Omega-3	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.37 standard deviations higher** (0.79 lower to 1.53 higher)		12 (1 study)	**very low**[1,2,3]

Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**833 per 1000**	**992 per 1000** (650 to 1000)	**RR 1.19** (0.78 to 1.83)	13 (1 study)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability - different populations
[3] Optimal information size not met; small, single study

12.2.1.12 Biomedical interventions: ginkgo biloba (plus risperidone) versus placebo (plus risperidone) for behaviour that challenges in children and young people

One RCT (N = 47) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Hasanzadeh 2012 (Hasanzadeh et al., 2012). An overview of the trial can be found in Table 122. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 124. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 124: Summary of findings table ginkgo biloba (plus risperidone) versus placebo (plus risperidone) in children and young people

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Placebo plus risperidone**	Corresponding risk **Ginkgo biloba plus risperidone**			
Targeted behaviour that challenges (severity) - post-treatment		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.1 standard deviations higher** (0.47 lower to 0.67 higher)		47 (1 study)	**very low**[1,2]
Adverse events (drowsiness, non-occurrence) - post-treatment	**708 per 1000**	**737 per 1000** (517 to 1000)	**RR 1.04** (0.73 to 1.49)	47 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Applicability - different populations
[2] Optimal information size not met; small, single study

12.2.1.131 Antipsychotics: risperidone versus placebo for behaviour that challenges in adults

2 Three RCTs (N = 194) met the eligibility criteria for this review Gagiano 2005 (Gagiano et al.,
3 2005), McDougle 1998 (McDougle et al., 1998), Tyrer 2008 (Tyrer et al., 2008). All eligible
4 studies included sufficient data to be included in the evidence syntheses. Tyrer 2008 was a
5 3-armed trial and compared risperidone, haloperidol and placebo with each other. For the
6 purposes of this review comparison, only risperidone and placebo arms will be utilised (N =
7 58). An overview of the trials included in the meta-analysis can be found in Table 125. See
8 also the study evidence tables in Appendix N and exclusion list in Appendix Q.

9 Further information about both included and excluded studies can be found in Appendices N
10 and Q.

11 Summary of findings can be found in Table 126. The full GRADE evidence profiles and
12 associated forest plots can be found in Appendices O and P.

13 No data were available for the critical outcome of service user and carer satisfaction.

14 See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

15 **Table 125: Study information table for trials included in the meta-analysis of**
16 **antipsychotics versus placebo in adults**

	Risperidone versus placebo	Haloperidol versus placebo
Total no. of studies (N[1])	3 (166)	1 (57)
Study ID	(1) Gagiano 2005 (2) McDougle 1998 (3) Tyrer 2008[2]	Tyrer 2008[5]
Country	(1, 3) Worldwide (2) USA	Worldwide
Diagnosis	(1) Mild to moderate LD[3] (2) Autism or PDD[4] (3) Mild to severe LD	Mild to severe LD
Age (mean)	28-40	40
Sex (% Female)	29-39	38
Ethnicity (% White)	Not reported (2) 77	Not reported
IQ (mean)	55-56 (3) Not reported	Not reported
Targeted behaviour that challenges	(1) Conduct problems (2) Maladaptive behaviours (3) Aggression	Aggression
Treatment length (weeks)	(1) 4 (2, 3) 12	12
Intervention (mean dose; mg/day)	(1, 3) Risperidone (1.6-18) (2) Risperidone (2.9)	Haloperidol (2.9)
Comparison (mean dose; mg/day)	Placebo (N/A)	Placebo (N/A)
Notes. N = total number of participants; LD = learning disability; N/A = not applicable; mg/day = milligrams per day		

	Risperidone versus placebo	Haloperidol versus placebo

[1] Number randomised
[2] 3-armed trial: only risperidone and placebo arms utilised
[3] 16% of participants had borderline intellectual functioning; all others were diagnosed with mild to moderate LD
[4] 26% of participants had IQ ≥ 70
[5] 3-armed trial: only haloperidol and placebo arms utilised

1 **Table 126: Summary of findings table for risperidone compared with placebo in adults**

Outcomes	Illustrative comparative risks* (95% CI) Assumed risk Placebo	Corresponding risk Risperidone	Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
Targeted behaviour that challenges (severity) - post-treatment End-point score; 12 week		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.25 standard deviations lower** (0.94 lower to 0.44 higher)		88 (2 studies)	**low**[1,2]
Targeted behaviour that challenges (severity) - post-treatment Change-score; 12 week		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.44 standard deviations lower** (0.9 lower to 0.02 higher)		74 (1 study)	**very low**[3,4]
Targeted behaviour that challenges (severity) - post-treatment Endpoint-score; 26 weeks[5]		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.16 standard deviations higher** (0.48 lower to 0.81 higher)		37 (1 study)	**low**[4]
Quality of life - post-treatment 12 weeks		The mean quality of life - post-treatment in the intervention groups was **0.27 standard deviations higher** (0.25 lower to 0.79 higher)		58 (1 study)	**low**[4]
Quality of life - post-treatment 26 weeks[5]		The mean quality of life - post-treatment in the intervention groups was **0.2 standard deviations higher** (0.42 lower to 0.82 higher)		40 (1 study)	**low**[4]
Adaptive functioning (social) - post-treatment		The mean adaptive functioning (social) - post-treatment in the intervention groups was **1.36 standard deviations lower** (2.17 to 0.56 lower)		30 (1 study)	**low**[4]
Adverse events (weight gain, non-occurrence) - post-treatment	**1000 per 1000**	**870 per 1000** (690 to 1000)	**RR 0.87** (0.69 to 1.09)	31 (1 study)	**very low**[4,6]
Adverse events (somnolence/sedation, non-occurrence) - post-treatment	**889 per 1000**	**578 per 1000** (249 to 1000)	**RR 0.65** (0.28 to 1.47)	108 (2 studies)	**very low**[2,7]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**1000 per 1000**	**950 per 1000** (870 to 1000)	**RR 0.95** (0.87 to 1.04)	89 (2 studies)	**moderate**[4]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**807 per 1000**	**840 per 1000** (743 to 953)	**RR 1.04** (0.92 to 1.18)	166 (3 studies)	**moderate**[4]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] I2 > 40%
[2] Optimal information size not met
[3] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[4] Optimal information size not met; small, single study

[5] Participants agreed to take the study drug for 12 weeks, with the option of continuing until 26 weeks, unless at 12 weeks other options were preferred. Post-treatment data are therefore provided at both 12 and 26 week end of treatment.
[6] Applicability - different populations
[7] I2 > 75%

12.2.1.142 Antipsychotics: haloperidol versus placebo for behaviour that challenges in adults

One RCT (N = 86) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Tyrer 2008 (Tyrer et al., 2008). Tyrer 2008 was a 3-armed trial and compared risperidone, haloperidol and placebo. For the purposes of this review comparison, only haloperidol and placebo arms will be utilised (N = 57).

An overview of the trial can be found in Table 125. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 127. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 127: Summary of findings table for haloperidol compared with placebo in adults

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk	Corresponding risk			
	Placebo	**Haloperidol**			
Targeted behaviour that challenges (severity) - post-treatment 12 weeks[1]		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.48 standard deviations lower** (1 lower to 0.05 higher)		57 (1 study)	**low**[2]
Targeted behaviour that challenges (severity) - post-treatment 26 weeks[1]		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.25 standard deviations lower** (0.87 lower to 0.37 higher)		40 (1 study)	**low**[2]
Quality of life - post-treatment 12 weeks[1]		The mean quality of life - post-treatment in the intervention groups was **0.17 standard deviations lower** (0.69 lower to 0.35 higher)		57 (1 study)	**low**[2]
Quality of life - post-treatment 26 weeks[1]		The mean quality of life - post-treatment in the intervention groups was **0.18 standard deviations lower** (0.79 lower to 0.43 higher)		41 (1 study)	**low**[2]
Adverse events (seizure, non-occurrence) - post-treatment	**1000 per 1000**	**960 per 1000** (880 to 1000)	**RR 0.96** (0.88 to 1.06)	57 (1 study)	**low**[2]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**1000 per 1000**	**930 per 1000** (820 to 1000)	**RR 0.93** (0.82 to 1.05)	57 (1 study)	**low**[2]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**724 per 1000**	**818 per 1000** (616 to 1000)	**RR 1.13** (0.85 to 1.51)	57 (1 study)	**low**[2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the

intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Patients agreed to take the study drug for 12 weeks, with the option of continuing until 26 weeks, unless at 12 weeks other options were preferred. Post-treatment data are therefore provided at both 12 and 26 week end of treatment.
[2] Optimal information size not met; small, single trial

12.2.1.15 Antipsychotics: risperidone versus haloperidol for behaviour that challenges in adults

One RCT (N = 86) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Tyrer 2008 (Tyrer et al., 2008). Tyrer 2008 was a 3-armed trial and compared risperidone, haloperidol and placebo with each other. For the purposes of this review comparison, only risperidone and haloperidol arms will be utilised (N = 57).

An overview of the trial can be found in Table 128. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 129. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 128: Study information table for trials included in the meta-analysis of risperidone versus haloperidol in adults

	Risperidone versus haloperidol
Total no. of studies (N[1])	1 (57)
Study ID	Tyrer 2008[2]
Country	Worldwide
Diagnosis	Mild to severe LD
Age (mean)	40
Sex (% Female)	38
Ethnicity (% White)	Not reported
IQ (mean)	Not reported
Targeted behaviour that challenges	Aggression
Treatment length (weeks)	12
Intervention (mean dose; mg/day)	Risperidone (1.8)
Comparison (mean dose; mg/day)	Haloperidol (2.9)
Notes. N = total number of participants; LD = learning disability; N/A = not applicable; mg/day = milligrams per day [1] Number randomised [2] 3-armed trial: only risperidone and haloperidol arms utilised	

Table 129: Summary of findings table for risperidone compared with haloperidol in adults

Outcomes	**Illustrative comparative risks* (95% CI)** Assumed Corresponding risk	**Relative effect**	**No of Participants**	**Quality of the evidence**

	risk		(95% CI)	(studies)	(GRADE)
	Haloperidol	**Risperidone**			
Targeted behaviour that challenges (severity) - post-treatment 12 weeks[1]		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.49 standard deviations higher** (0.03 lower to 1.02 higher)		57 (1 study)	**low**[2]
Targeted behaviour that challenges (severity) - post-treatment 26 weeks[1]		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.39 standard deviations higher** (0.28 lower to 1.05 higher)		36 (1 study)	**low**[2]
Quality of life - post-treatment 12 weeks[1]		The mean quality of life - post-treatment in the intervention groups was **0.43 standard deviations higher** (0.09 lower to 0.96 higher)		57 (1 study)	**low**[2]
Quality of life - post-treatment 26 weeks [1]		The mean quality of life - post-treatment in the intervention groups was **0.41 standard deviations higher** (0.23 lower to 1.04 higher)		39 (1 study)	**low**[2]
Adverse events (seizure, non-occurrence) - post-treatment	**964 per 1000**	**1000 per 1000** (906 to 1000)	**RR 1.04** (0.94 to 1.14)	57 (1 study)	**low**[2]
Adverse events (discontinuation due to adverse events) - post-treatment	**929 per 1000**	**966 per 1000** (854 to 1000)	**RR 1.04** (0.92 to 1.18)	57 (1 study)	**low**[2]
Adverse events (discontinuation due to other reasons) - post-treatment	**857 per 1000**	**797 per 1000** (626 to 1000)	**RR 0.93** (0.73 to 1.18)	57 (1 study)	**low**[2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Patients agreed to take the study drug for 12 weeks, with the option of continuing until 26 weeks, unless at 12 weeks other options were preferred. Post-treatment data are therefore provided at both 12 and 26 week end of treatment.
[2] Optimal information size not met; small, single study

12.2.1.161 Antipsychotics: olanzapine versus risperidone for behaviour that challenges in adults

2 One RCT (N = 62) met the eligibility criteria for this review and included sufficient data to be
3 included in the evidence syntheses: Amore 2011 (Amore et al., 2011). An overview of the
4 trial can be found in Table 130. Further information about both included and excluded studies
5 can be found in Appendices N and Q.

6 Summary of findings can be found in Table 131. The full GRADE evidence profiles and
7 associated forest plots can be found in Appendices O and P.

8 No data were available for the critical outcomes of adaptive functioning, quality of life or
9 service user and carer satisfaction.

10 See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

11 **Table 130: Study information table for trials included in the meta-analysis of**
12 **olanzapine versus risperidone in adults**

	Olanzapine versus risperidone
Total no. of studies (N[1])	1 (62)
Study ID	Amore 2011
Country	Italy

	Olanzapine versus risperidone
Diagnosis	Severe LD
Age (mean)	48
Sex (% Female)	27
Ethnicity (% White)	Not reported
IQ (mean)	Not reported
Targeted behaviour that challenges	Aggression
Treatment length (weeks)	24
Intervention (mean dose; mg/day)	Olanzapine (20)
Comparison (mean dose; mg/day)	Risperidone (6)
Notes. N = total number of participants; RCT = randomised controlled trial; LD = learning disability; N/A = not applicable; mg/day = milligrams per day [1] Number randomised	

Table 131: Summary of findings table for olanzapine compared with risperidone in adults

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk **Risperidone**	Corresponding risk **Olanzapine**			
Targeted behaviour that challenges (frequency) - post-treatment		The mean targeted behaviour that challenges (frequency) - post-treatment in the intervention groups was **0.2 standard deviations higher** (0.3 lower to 0.7 higher)		62 (1 study)	**very low**[1,2]
Adverse events (elevated prolactin) - post-treatment	**968 per 1000**	**706 per 1000** (561 to 900)	**RR 0.73** (0.58 to 0.93)	62 (1 study)	**very low**[1,2]
Adverse events (weight gain, non-occurrence) - post-treatment	**903 per 1000**	**777 per 1000** (623 to 966)	**RR 0.86** (0.69 to 1.07)	62 (1 study)	**very low**[1,2]
Adverse events (sedation, non-occurrence) - post-treatment	**839 per 1000**	**772 per 1000** (604 to 990)	**RR 0.92** (0.72 to 1.18)	62 (1 study)	**very low**[2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

12.2.1.17 Antipsychotics: withdrawal of zuclopenthixol versus continuation of zuclopenthixol for behaviour that challenges in adults

Three RCTs (N = 204) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Haessler 2007 (Haessler et al., 2007), Izmeth 1988 (Izmeth et al., 1988), Singh 1992 (Singh & Owino, 1992). An overview of the trials included in the meta-analysis can be found in Table 132. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 133. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 132: Study information table for trials included in the meta-analysis of withdrawal of zuclopenthixol versus continuation of zuclopenthixol in adults

	Withdrawal of zuclopenthixol versus continuation of zuclopenthixol
Total no. of studies (N[1])	3 (204)
Study ID	(1) Haessler 2007 (2) Izmeth 1988 (3) Singh 1992
Country	(1) Germany (2, 3) UK
Diagnosis	Mild to severe LD
Age (mean)	31-36
Sex (% Female)	40-46
Ethnicity (% White)	(1) 100 (2, 3) Not reported
IQ (mean)	(1, 3) Not reported (2) 50
Targeted behaviour that challenges	(1) Aggression (2, 3) Behavioural disorders
Treatment length (weeks)	12
Intervention (mean dose; mg/day)	Withdrawal of zuclopenthixol[2]
Comparison (mean dose; mg/day)	(1) Continuation of zuclopenthixol (11.4) (2) Continuation of zuclopenthixol (119)[3] (3) Continuation of zuclopenthixol (20)[4]
Notes. N = total number of participants; LD = learning disability; N/A = not applicable; mg/day = milligrams per day [1] Number randomised [2] Participants who were in the withdrawal condition received placebo medication [3] Mean dose per week; daily dose not reported [4] Mode dose	

Table 133: Summary of findings table for withdrawal of zuclopenthixol versus continuation of zuclopenthixol in adults

Outcomes	**Illustrative comparative risks* (95% CI)**		**Relative effect (95% CI)**	**No of Participants (studies)**	**Quality of the evidence (GRADE)**
	Assumed risk	Corresponding risk			
	Continuation of zuclopenthixol	**Withdrawal of zuclopenthixol**			

Targeted behaviour that challenges (relapse) - post-treatment	**632 per 1000**	**947 per 1000** (663 to 1000)	**RR 1.5** (1.05 to 2.15)	39 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (severity) - post-treatment End-point score		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.56 standard deviations higher** (0.08 lower to 1.2 higher)		39 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (severity) - post-treatment Change score		The mean targeted behaviour that challenges (severity) - post-treatment in the intervention groups was **0.68 standard deviations higher** (0.24 to 1.11 higher)		85 (1 study)	**very low**[1,2]
Targeted behaviour that challenges (problems in management) - post-treatment	**208 per 1000**	**369 per 1000** (140 to 979)	**RR 1.77** (0.67 to 4.7)	43 (1 study)	**very low**[2,3]
Adaptive functioning (social) - post-treatment		The mean adaptive functioning (social) - post-treatment in the intervention groups was **0.47 standard deviations lower** (0.9 to 0.04 lower)		85 (1 study)	**very low**[1,2]
Adverse events (weight gain; kg) - post- treatment		The mean adverse events (weight gain; kg) - post- treatment in the intervention groups was **0.55 standard deviations lower** (1.19 lower to 0.09 higher)		39 (1 study)	**very low**[1,2]
Adverse events (drowsiness, non-occurrence) - post-treatment	**950 per 1000**	**950 per 1000** (836 to 1000)	**RR 1** (0.88 to 1.15)	42 (1 study)	**very low**[1,2]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**951 per 1000**	**818 per 1000** (676 to 990)	**RR 0.86** (0.71 to 1.04)	204 (3 studies)	**very low**[4,5,6]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**826 per 1000**	**603 per 1000** (273 to 1000)	**RR 0.73** (0.33 to 1.64)	91 (2 studies)	**very low**[4,6,7]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study
[3] Crucial limitation for 1 or more criteria sufficient to substantially lower ones confidence in the estimate of effect
[4] Most information is from studies at moderate risk of bias
[5] I2 > 40%
[6] Optimal information size not met
[7] I2 > 75%

12.2.1.182 Mood stabilisers: lithium versus placebo for behaviour that challenges in adults

3 One RCT (N = 42) met the eligibility criteria for this review and included sufficient data to be
4 included in the evidence syntheses: Craft 1987 (Craft et al., 1987). An overview of the trial
5 can be found in Table 134. See also the study evidence tables in Appendix N and exclusion
6 list in Appendix Q.

7 Further information about both included and excluded studies can be found in Appendices N
8 and Q.

Summary of findings can be found in Table 135. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 134: Study information table for trials included in the meta-analysis of lithium versus placebo in adults

	Lithium versus placebo
Total no. of studies (N[1])	1 (42)
Study ID	Craft 1987
Country	UK
Diagnosis	Mild to moderate LD
Age (mean)	33
Sex (% Female)	31
Ethnicity (% White)	Not reported
IQ (mean)	Not reported
Targeted behaviour that challenges	Aggression
Treatment length (weeks)	12
Intervention (mean dose; mg/day)	Lithium (800)[2]
Comparison	Placebo

Notes. N = total number of participants; LD = learning disability; mg/day = milligrams per day
[1] Number randomised
[2] Starting dose; mean dose not reported

Table 135: Summary of findings table for lithium compared with placebo in adults

Outcomes	**Illustrative comparative risks* (95% CI)** Assumed risk **Placebo**	Corresponding risk **Lithium**	**Relative effect (95% CI)**	**No of Participants (studies)**	**Quality of the evidence (GRADE)**
Targeted behaviour that challenges (frequency, non-improvement)	**700 per 1000**	**273 per 1000** (133 to 574)	**RR 0.39** (0.19 to 0.82)	42 (1 study)	**very low**[1,2]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Optimal information size not met; small, single study

12.2.1.19 Naltrexone versus placebo for self-injurious behaviour in adults

The GDG selected an existing Cochrane review as the basis for this section of the guideline: Rana 2013 (Rana et al., 2013). The systematic review included 5 studies (N = 50): Lewis 1996 (Lewis et al., 1996), Sandman 1990 (Sandman et al., 1990), Symons 2001 (Symons et al., 2001), Thompson 1994 (Thompson et al., 1994), Willemsen-Swinkels 1995 (Willemsen-Swinkels et al., 1995). Of the included studies, 4 reviewed the effectiveness and safety of naltrexone for self-injurious behaviour Sandman 1990, Symons 2001, Thompson 1994, Willemsen-Swinkels (1995). A summary of the included review can be found in Table 135.

Due to differences in study designs (duration, cross-over phases within the studies), heterogeneity of interventions (doses of drugs) and differences in how outcome measures were reported, a meta-analysis was not possible. A brief narrative synthesis is therefore given.

All included studies were prospective, randomised, double-blinded, placebo-controlled trials and had a cross-over design. Included studies were published in peer-reviewed journals between 1990 and 2001. The mean age of included participants was 33 years (range 23-46 years) and 20% were females. All participants were diagnosed with a learning disability. The degree of learning disability was classified as severe to profound in all studies except in Willemsen-Swinkels 1995 where it ranged from mild to profound. The dosage of naltrexone administered was 25-100 mg twice per week in Sandman 1990, 50-100 mg per day in Thompson 1994, 1.5 mg per kilogram per day in Symons 2001 and 50-150 mg per day in Willemsen-Swinkles 1995.

Forms of SIB in the 4 trials included head banging, body hitting, head hitting, hand hitting, self-biting, self-hitting, hair pulling, face-pinching and hitting, self-rubbing, scratching and rocking.

Further information about both included and excluded studies can be found in Rana 2013.

Table 136: Study information table for the systematic review included in the review of antecedent modification

	Rana 2013
Review question/ Aim	To determine the clinical effectiveness of pharmacological interventions in the management of self-injurious behaviour in adults with a learning disability.
Method used to synthesise evidence	Narrative synthesis
Design of included studies	Randomised, double-blinded, placebo-controlled trials with a cross-over design
Dates searched	1948-2012
Electronic databases	(1) Central; (2) MEDLINE; (3) Embase; (4) PsycINFO; (5) CINAHL; (6) Science Citation Index; (7) Social Science Citation Index; (8) Conference Proceedings Citation Index - Science; (9) Conference Proceedings Citation Index - Social science and Humanities; (10) ZETOC; (11) WorldCat; (12) ClinicalTrials.gov; (13) ICTRP
No. of included studies (N[1])	5 (50[2])
Participant characteristics	Adults with LD (mild to profound), aged 18 years or over, presenting with SIB occurring at least during most weeks of the preceding 6 months (as per diagnostic criteria in DC-LD 2001), and without additional psychiatric illness.
Intervention	Pharmacological interventions including any antidepressants, antipsychotics, mood stabilisers, opiate antagonist (naltrexone), beta-blocker (propranolol) and hypnotic (melatonin), regardless of dosage, against placebo.
Comparison	N/A
Outcome	• Frequency, intensity and duration of SIB • Adverse events (effects of medication such as sleepiness, movement disorders, seizures, weight gain, etc.)
Review Quality	High
Notes: SIB = self-injurious behaviour; LD = learning disability [1]Number of participants. [2] The included studies randomised 57 participants; however 7 participants were excluded from the	

	Rana 2013
review as they did not have SIB.	

12.2.1.201 Clomipramine versus placebo for self-injurious behaviour in adults

The GDG selected an existing Cochrane review as the basis for this section of the guideline: Rana 2013 (Rana et al., 2013). The systematic review included 5 studies (N = 50): Lewis 1996 (Lewis et al., 1996), Sandman 1990 (Sandman et al., 1990), Symons 2001 (Symons et al., 2001), Thompson 1994 (Thompson et al., 1994), Willemsen-Swinkels 1995 (Willemsen-Swinkels et al., 1995). Of the included studies, 1 reviewed the effectiveness and safety of clomipramine for self-injurious behaviour: Lewis, 1996. A summary of the included review can be found in Table 135.

The included study was a prospective, randomised, double-blind, placebo-controlled trial and had a cross-over design. The age of included participants ranged from 21 to 39 years and 38% were females. All participants were diagnosed with a severe to profound learning disability. The dosage of clomipramine administered was 3 mg per kilogram per day.

Further information about both included and excluded studies can be found in Rana 2013.

12.2.1.214 Melatonin versus placebo for sleep problems in children

Four RCTs (N = 372) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Braam 2008a (Braam et al., 2008a), Braam 2008b (Braam et al., 2008b), Cortesi 2012 (Cortesi et al., 2012), Gringras 2012 (Gringras et al., 2012). Cortesi 2012 was a 4-armed trial and compared CBT, melatonin, combined treatment and placebo. For the purposes of this review comparison, only melatonin and placebo arms will be utilised (N = 80). An overview of the trials included in the meta-analysis can be found in Table 137. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 138. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of global sleep behaviour (assuming dropouts had not improved) was conducted. In the sensitivity analysis, effects remained consistent with the main analysis.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 137: Study information table for trials included in the meta-analysis of melatonin versus placebo for sleep problems in children

	Melatonin versus placebo	Melatonin versus CBT
Total no. of studies (N[1])	4 (292)	1 (80)
Study ID	(1) Braam 2008a (2) Braam 2008b (3) Cortesi 2012[2] (4) Gringras 2012	Cortesi 2012[4]

	Melatonin versus placebo	Melatonin versus CBT
Country	(1, 2) Netherlands (3) USA (4) UK	USA
Diagnosis	(1) LD (2) Angelman syndrome (3) Autism (4) DD	Autism
Age (mean)	(1) 23 (2 to 4) 7-11	7
Sex (% Female)	(1, 2, 4) 34-63 (3) 18	18
Ethnicity (% White)	Not reported (3) 99	99
IQ (mean)	Not reported	Not reported
Targeted behaviour that challenges	Sleep problem	Sleep problem
Treatment length (weeks)	(1, 2) 4 (3, 4) 12	12
Intervention (mean dose; mg/day)	(1, 2) Melatonin (5)[3] (3) Melatonin (3) (4) Melatonin (6.4)	Melatonin (3)
Comparison	Placebo	CBT

Notes. N = total number of participants; DD = developmental disabilities; LD = learning disability; TAU = treatment as usual.
[1] Number randomised
[2] Four armed trial: only melatonin and placebo arms utilised
[3] Maximum dose
[4] 4-armed trial: only melatonin and CBT arms utilised

1 **Table 138: Summary of findings table for melatonin compared with placebo**

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk	Corresponding risk			
	Placebo	Melatonin			
Targeted behaviour that challenges (global problem sleep behaviour) - post-treatment Children's Sleep Habits Questionnaire		The mean targeted behaviour that challenges (global problem sleep behaviour) - post-treatment in the intervention groups was **1.81 standard deviations lower** (2.39 to 1.23 lower)		66 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (global problem sleep behaviour) - post-treatment Composite Sleep Disturbance Index		The mean targeted behaviour that challenges (global problem sleep behaviour) - post-treatment in the intervention groups was **0.26 standard deviations lower** (0.62 lower to 0.09 higher)		125 (1 study)	**low**[3]
Targeted behaviour that challenges (non-improvement of global problem sleep behaviour) - post-treatment	**1000 per 1000**	**620 per 1000** (480 to 810)	**RR 0.62** (0.48 to 0.81)	66 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges		The mean targeted behaviour that		124	

(sleep efficiency) - post-treatment Actigraph		challenges (sleep efficiency) - post-treatment in the intervention groups was **1.46 standard deviations higher** (0.51 lower to 3.42 higher)		(2 studies)	**very low**[4,5]
Targeted behaviour that challenges (total sleep time) - post-treatment Actigraph		The mean targeted behaviour that challenges (total sleep time) - post-treatment in the intervention groups was **1.01 standard deviations higher** (0.26 lower to 2.28 higher)		125 (2 studies)	**very low**[4,5]
Targeted behaviour that challenges (wake after sleep onset) - post-treatment Actigraph		The mean targeted behaviour that challenges (wake after sleep onset) - post-treatment in the intervention groups was **0.76 standard deviations lower** (1.14 to 0.38 lower)		115 (2 studies)	**moderate**[5]
Targeted behaviour that challenges (sleep onset latency) - post-treatment Actigraph		The mean targeted behaviour that challenges (sleep onset latency) - post-treatment in the intervention groups was **1.23 standard deviations lower** (1.75 to 0.7 lower)		66 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (total sleep time) - post-treatment Sleep diary		The mean targeted behaviour that challenges (total sleep time) - post-treatment in the intervention groups was **0.34 standard deviations higher** (0.37 lower to 1.05 higher)		169 (3 studies)	**low**[5,6]
Targeted behaviour that challenges (number of wakes per night) - post-treatment Sleep diary		The mean targeted behaviour that challenges (number of wakes per night) - post-treatment in the intervention groups was **0.06 standard deviations lower** (0.49 lower to 0.37 higher)		164 (3 studies)	**moderate**[5]
Targeted behaviour that challenges (wake after sleep onset) - post-treatment Sleep diary		The mean targeted behaviour that challenges (wake after sleep onset) - post-treatment in the intervention groups was **0.64 standard deviations lower** (1.03 to 0.25 lower)		172 (3 studies)	**moderate**[5]
Targeted behaviour that challenges (duration of wakes) - post-treatment Sleep diary		The mean targeted behaviour that challenges (duration of wakes) - post-treatment in the intervention groups was **0.23 standard deviations higher** (0.36 lower to 0.82 higher)		163 (3 studies)	**low**[5,6]
Adverse events (somnolence/sedation, non-occurrence) - post-treatment	**868 per 1000**	**868 per 1000** (773 to 990)	**RR 1** (0.89 to 1.14)	146 (1 study)	**low**[3]
Adverse events (discontinuation due to adverse events, non-occurrence) - post-treatment	**974 per 1000**	**983 per 1000** (944 to 1000)	**RR 1.01** (0.97 to 1.06)	146 (1 study)	**low**[3]
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**882 per 1000**	**935 per 1000** (829 to 1000)	**RR 1.06** (0.94 to 1.2)	284 (3 studies)	**low**[5,6]
Adverse events (seizure, non-occurrence) - post-treatment	**987 per 1000**	**997 per 1000** (967 to 1000)	**RR 1.01** (0.98 to 1.05)	146 (1 study)	**low**[3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability- different populations
[3] Optimal information size not met; small, single study
[4] I2 > 75%
[5] Optimal information size not met
[6] I2 > 40%

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12.2.1.221 Melatonin versus CBT for sleep problems in children

One RCT (N = 160) met the eligibility criteria for this review and included sufficient data to be included in the evidence syntheses: Cortesi 2012 (Cortesi et al., 2012). Cortesi 2012 was a 4-armed trial and compared CBT, melatonin, and combined treatment to placebo. For the purposes of this review comparison, only melatonin and CBT arms will be utilised (N = 80). An overview of the included trial can be found in Table 137. See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Further information about both included and excluded studies can be found in Appendices N and Q.

Summary of findings can be found in Table 139. The full GRADE evidence profiles and associated forest plots can be found in Appendices O and P.

As some studies in the meta-analysis only reported data for completers, a sensitivity analysis for non-improvement of sleep onset latency and sleep efficiency (assuming dropouts had not improved) was conducted. In the sensitivity analysis, effects remained consistent with the main analysis.

No evidence was identified in relation to the specific subgroups identified in the review protocol.

No data were available for the critical outcomes of adaptive functioning, quality of life or service user and carer satisfaction.

See also the study evidence tables in Appendix N and exclusion list in Appendix Q.

Table 139: Summary of findings table for melatonin compared with CBT

Outcomes	Illustrative comparative risks* (95% CI)		Relative effect (95% CI)	No of Participants (studies)	Quality of the evidence (GRADE)
	Assumed risk	Corresponding risk			
	CBT	Melatonin			
Targeted behaviour that challenges (global problem sleep behaviour) - post-treatment Children's Sleep Habits Questionnaire		The mean targeted behaviour that challenges (global problem sleep behaviour) - post-treatment in the intervention groups was **0.94 standard deviations lower** (1.45 to 0.44 lower)		67 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (non-improvement of global sleep problem behaviour) - post-treatment	**909 per 1000**	**618 per 1000** (464 to 818)	**RR 0.68** (0.51 to 0.9)	67 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (sleep onset latency) - post-treatment Actigraph		The mean targeted behaviour that challenges (sleep onset latency) - post-treatment in the intervention groups was **0.54 standard deviations lower** (1.03 to 0.05 lower)		67 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (wake after sleep onset) - post-treatment Actigraph		The mean targeted behaviour that challenges (wake after sleep onset) - post-treatment in the intervention groups was **0.73 standard deviations lower** (1.22 to 0.23 lower)		67 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (total sleep time) - post-treatment Actigraph		The mean targeted behaviour that challenges (total sleep time) - post-treatment in the intervention groups was **0.76 standard deviations higher** (0.26 to 1.26 higher)		67 (1 study)	**very low**[1,2,3]
Targeted behaviour that challenges (sleep efficiency) - post-treatment		The mean targeted behaviour that challenges (sleep efficiency) - post-treatment in the intervention groups was		67 (1 study)	**very low**[1,2,3]

Actigraph		**0.89 standard deviations higher** (0.39 to 1.4 higher)			
Adverse events (discontinuation due to other reasons, non-occurrence) - post-treatment	**900 per 1000**	**900 per 1000** (774 to 1000)	**RR 1** (0.86 to 1.16)	80 (1 study)	**very low**[1,2,3]

*The basis for the **assumed risk** (for example, the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI).

CI: Confidence interval; **RR:** Risk ratio;

[1] Crucial limitation for 1 criterion or some limitations for multiple criteria sufficient to lower ones confidence in the estimate of effect
[2] Applicability- different populations
[3] Optimal information size not met; small, single study

12.2.2 Economic evidence

12.2.2.1 Systematic literature review

The systematic search of the literature identified 1 study that assessed the cost effectiveness of psychosocial interventions aimed at reducing and managing behaviour that challenges in people with a learning disability (Romeo et al., 2009). Details on the methods used for the systematic review of the economic literature are described in Chapter 3; full references and evidence tables for all economic evaluations included in the systematic literature review are provided in Appendix S. Completed methodology checklists of the studies are provided in Appendix R. Economic evidence profiles of studies considered during guideline development (i.e. studies that fully or partly met the applicability and quality criteria) are presented in Appendix T.

Romeo and colleagues (2009) evaluated the cost effectiveness of risperidone and haloperidol versus placebo for the management of behaviour that challenges in adults with a learning disability in the UK. The economic analysis was undertaken alongside a multi-country RCT included in the guideline systematic review (Tyrer 2008). The study sample consisted of 86 adults with a learning disability (IQ<75) and behaviour that challenges and aggression. The time horizon of the economic analysis was 26 weeks, and its perspective was societal, including service and informal (unpaid) care costs. Cost elements comprised medication, inpatient care, specialised accommodation, day activities, community-based activities and informal care. Resource use data were collected for a multi-country sub-sample of 58 participants in the trial. National UK unit costs were used. The primary measures of outcome utilised in the economic analysis were the total Modified Overt Aggression Scale (MOAS) score and the total quality of life (QOL-Q) of service users.

The analysis demonstrated that haloperidol was the least costly intervention of those considered in terms of service costs (mean total service costs per person for risperidone, haloperidol and placebo were £15,518, £13,753 and £15,010, respectively, in likely 2006 prices). When costs of informal care were included in the estimation of costs, placebo becomes the least costly intervention (mean total costs per person for risperidone, haloperidol and placebo were £18,954, £17,626 and £16,336, respectively). Haloperidol was shown to be the most effective intervention in terms of reduction in levels of aggression (lowest mean MOAS score per person) and haloperidol was the most effective intervention in terms of quality of life (highest mean QOL-Q score per person). However, differences in costs and outcomes between the interventions were not statistically significant.

In terms of cost effectiveness and under a societal perspective, when using the total MOAS score as an outcome risperidone was dominated by placebo (less effective and more costly). Haloperidol was more effective than placebo at an additional cost of £614 per additional point change on the MOAS. The probability of haloperidol being cost effective compared with

placebo was approximately 50% at zero willingness to pay for an additional point change on MOAS, and roughly 89% for a willingness to pay of £3000 per point improvement in MOAS. When using total QOL-Q score, haloperidol was dominated by placebo. Risperidone was more effective than placebo at an additional cost of £996 per point change on the QOL-Q. The probability of risperidone being cost effective compared with placebo was approximately 52% at any willingness to pay for a 1-point improvement in QOL-Q score. Based on these results, the authors concluded that 'risperidone and haloperidol do not offer good value for money over placebo when service implications, costs and effects on aggression and quality of life associated with treatment are considered' (Romeo et al., 2009).

The study is only partially applicable to the NICE decision-making context, as it has adopted a societal perspective that is wider than the NICE recommended perspective. Moreover, the measure of outcomes was not expressed in QALYs, which made interpretation of findings difficult. The study was judged to have potentially serious limitations, including the small study sample and the relatively short time horizon (26 weeks). Moreover, there were concerns with the quality of the clinical data analysis.

12.2.2.26 Economic modelling

The systematic search of the literature did not identify any evidence on the cost effectiveness of pharmacological interventions for the management of behaviour that challenges in children and young people with a learning disability. Given the efficacy of antipsychotics (risperidone and aripiprazole) for this indication, as shown in the systematic clinical review, and the significant resource implications associated with provision of antipsychotics, an economic model was developed to assess the cost effectiveness of antipsychotics in children and young people with a learning disability and behaviour that challenges. In addition, an economic model that evaluated the cost effectiveness of pharmacological interventions relative to psychological and combination therapies for the management of sleep problems in children and young people with a learning disability was also developed.

12.2.2.27 Economic modelling – antipsychotics for the management of behaviour that challenges in children and young people with a learning disability

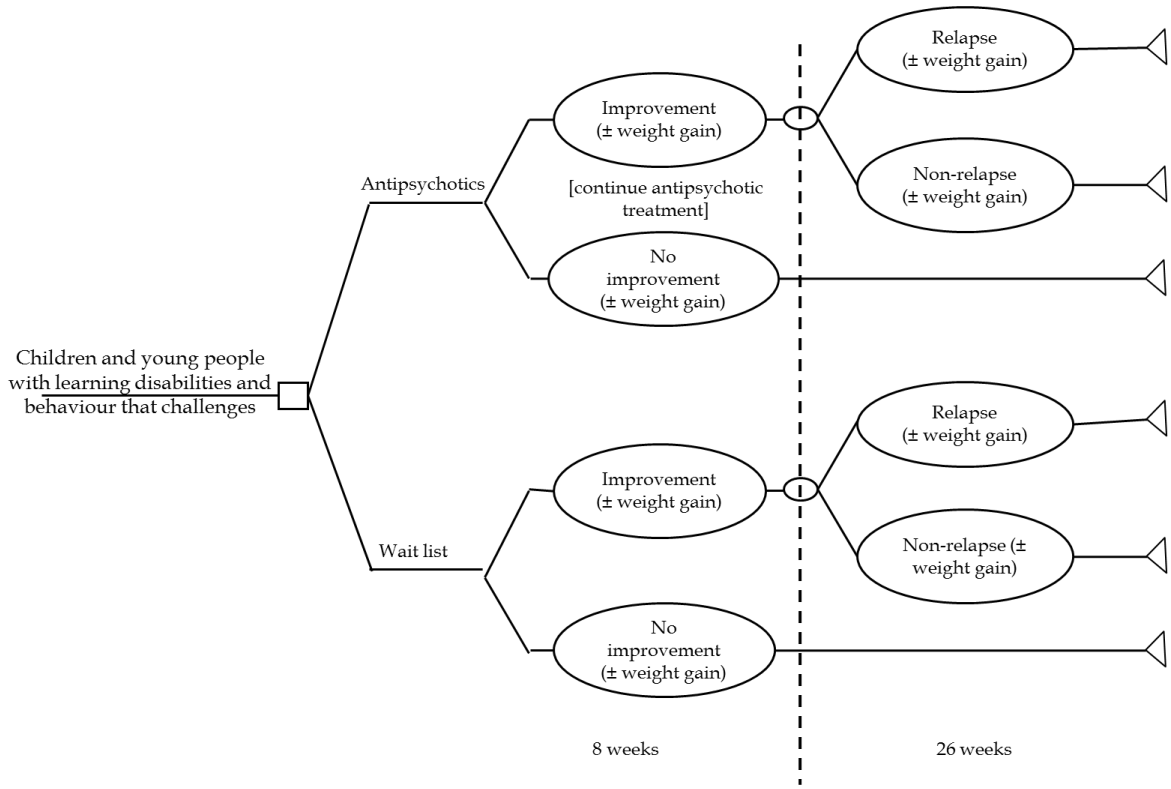
12.2.2.3.1 29 *Interventions assessed*

The evidence on antipsychotics for the management of behaviour that challenges in children and young people with a learning disability that were included in the guideline systematic review came predominantly from RCTs assessing risperidone and/or aripiprazole versus placebo. A small trial (N=12) that compared olanzapine with haloperidol was also identified (Malone 2001), but this evidence was considered too limited to inform an economic model. Consequently, the guideline economic analysis assessed the relative cost effectiveness of risperidone and aripiprazole versus placebo. Risperidone is available in tablets and orodispersible tablets, as well as in oral solution formulation, all of which were considered in the analysis as they entail different acquisition costs. Aripiprazole is available only in tablet formulation which was assessed in the analysis. It should be noted that ideally pharmacological interventions should also be compared with psychological interventions that were evaluated in Chapter 11. However, this was not possible as there were no common comparators for pharmacological and psychological interventions that would allow an indirect comparison of their relative effectiveness and, subsequently, the assessment of their relative cost effectiveness: RCTs of psychological interventions for the management of behaviour that challenges in children and young people with a learning disability have mostly used wait list or standard care as a comparator; on the other hand, relevant RCTs of pharmacological interventions has used placebo as control.

*12.2.2.3.2*1 *Model structure*

2 A simple decision-tree was constructed using Microsoft Excel 2010 to estimate the cost
3 effectiveness of antipsychotics versus placebo for the management of behaviour that
4 challenges in children and young people with a learning disability. According to the model
5 structure, hypothetical cohorts of children and young people with a learning disability and
6 behaviour that challenges received either an antipsychotic or placebo for 8 weeks. At the end
7 of the 8 weeks children and young people either improved in terms of their behaviour that
8 challenges or did not improve. All cohorts were further followed for 26 weeks. Children and
9 young people that had improved during the 8-week antipsychotic treatment continued
10 medication over the follow-up 26-week period. At the end of 26 weeks children and young
11 people that had improved following initial treatment (antipsychotics or placebo) either
12 relapsed or remained improved. Children and young people that had not improved at the end
13 of the first 8 weeks (i.e. at completion of treatment) were conservatively assumed to retain
14 the same levels of behaviour that challenges over the next 26 weeks. Children and young
15 people in both arms of the model could experience weight gain as an adverse event of
16 treatment. Weight gain is one of the most common adverse events of antipsychotic
17 medication, and therefore, given also the availability of clinical and utility data, it was selected
18 out of a range of adverse events associated with antipsychotics, for incorporation into the
19 model structure. The time horizon of the model was 34 weeks (8 weeks of treatment and 26
20 weeks of follow-up). The duration of treatment and follow-up periods was determined by
21 respective time periods in the RCTs that provided clinical data in the economic analysis. The
22 model structure has been adopted from a similar model that was developed to inform the
23 NICE guideline on the management of autism in children and young people (NICE, 2013a). A
24 schematic diagram of the decision-tree is presented in Figure 6.

25 **Figure 6. Schematic diagram of the structure of the economic model evaluating**
26 **antipsychotic drugs compared with placebo for the management of**
27 **behaviour that challenges in children and young people with a learning**
28 **disability**



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12.2.2.3.3 Costs and outcomes considered in the analysis

The economic analyses adopted the perspective of the NHS and personal social services, as recommended by NICE (NICE 2012, The Guidelines Manual). Costs consisted of intervention costs only, as no data on costs associated with behaviour that challenges in children and young people with a learning disability was identified in the relevant literature. Moreover, no extra costs of managing adverse events of medication were considered in the analysis. The measure of outcome was the QALY.

12.2.2.3.4 Clinical input parameters of the economic model

Clinical input parameters included the probability of non-improvement of behaviour that challenges at 8 weeks, the risk ratio of non-improved behaviour that challenges of each antipsychotic (risperidone or aripiprazole) versus placebo, the 24-week probability of relapse after improvement, the risk of (non-)weight gain associated with placebo and the risk ratio of (non-)weight gain of antipsychotics versus placebo.

The guideline systematic review identified 2 RCTs assessing risperidone versus placebo (RUPP 2002 and Shea 2004) and 2 RCTs comparing aripiprazole versus placebo (Marcus 2009 and Owen 2009) for the management of behaviour that challenges in children and young people with a learning disability that reported outcome as improvement in behaviour that challenges regarding its severity. Pooled weighted data from the placebo arms of the 4 RCTs were used to estimate the probability of non-improvement of behaviour that challenges under placebo at 8 weeks, which was utilised in the model. Separate meta-analyses of the risperidone and aripiprazole trials provided the risk ratio of non-improvement in behaviour that challenges of risperidone and aripiprazole, respectively, versus placebo. It must be noted that the economic model utilised the intention-to-treat sensitivity analysis, which assumed that dropouts did not improve.

In addition to the above trials, 1 RCT compared risperidone with aripiprazole (Granizadeh 2013). This trial did not report dichotomous efficacy data that could be used in the economic model, and therefore it was not considered in the economic analysis. The results of the trial indicated that risperidone was more effective than aripiprazole in the management of behaviour that challenges, however, results were not statistically significant.

Two small trials assessed relapse to behaviour that challenges in children and young people that had responded to antipsychotic treatment over an open-label phase and were subsequently either continued on or discontinued from antipsychotic medication (RUPP2005 on risperidone and Findling 2014 on aripiprazole). Data from the antipsychotic continuation arms from these 2 studies were pooled together (due to the small study sample of each study) and used to estimate the 26-week probability of relapse in both pharmacological arms of the economic model, as well as placebo (i.e. antipsychotics and placebo). It should be noted that the relapse data reported for the discontinuation arms of the RCTs (i.e. arms that discontinued the antipsychotic following improvement and received placebo) were not deemed to be relevant to the placebo arm of the economic model, as in discontinuation arms of the trials participants had already received an antipsychotic and discontinued it, whereas in the placebo arm of the economic model children and young people had never been initiated on an antipsychotic.

Data on weight gain were derived from 3 risperidone trials (Aman 2002, Shea 2004 and Snyder 2002) and 2 aripiprazole trials (Owen 2009 and Marcus 2009 that were included in the guideline systematic review. The risk of (non-)weight gain associated with placebo was based on pooled weighted data from the placebo arms of these 5 trials, while the risk ratio of (non-)weight gain for risperidone and aripiprazole versus placebo was derived from separate meta-analyses of the risperidone and aripiprazole trials, respectively.

12.2.2.3.5 Utility data for the estimation of QALYs

A systematic search of the literature was undertaken to identify studies that reported utility scores for children and young people with a learning disability and behaviour that challenges that were required for the estimation of QALYs in the economic modelling undertaken for this guideline. The results of this review are reported in Chapter 11 (section 11.2.2). No studies reporting utility data on distinct health states relating to the condition assessed in this guideline were identified. However, one study was found that reported utility scores for a number of health states relating to symptoms experienced by children and young people with autism, such as hyperactivity, aggression and sleep problems (Tilford et al., 2012); these symptoms are also relevant to children and young people with a learning disability. It should be noted that no information on the IQ of the children in autism that participated in the study was provided. Utility data were derived from parents' responses to HUI3, a preference-based measure that has not been specifically designed for use in children. The GDG expressed the opinion that HUI3 is neither directly relevant to the symptoms of children and young people with a learning disability, nor sensitive enough in capturing changes in children's HRQoL. Moreover, HUI3 scores are not directly relevant to the UK context, since valuation was based on the preferences of members of the Canadian population. Nevertheless, given the lack of other appropriate utility data, the GDG decided to utilise the utility data reported by Tilford and colleagues (2012) in the guideline economic modelling as a proxy of the HRQoL of children and young people with a learning disability. Details on the study by Tilford and colleagues (2012) are provided in Chapter 11 (section 11.2.2).

In consistency with the economic analysis of parent training described in Chapter 11, the economic analysis of antipsychotic treatment for the management of behaviour that challenges used utility scores for different levels of hyperactivity as a proxy for changes in behaviour that challenges in children and young people with a learning disability. The economic analysis conservatively assumed that at initiation of treatment the HRQoL of the study population corresponded to moderate levels of hyperactivity that improved to mild symptoms following response to treatment. Children that relapsed were assumed to return to the utility score corresponding to moderate symptom levels of hyperactivity. It was assumed that all improvements and decrements in utility occurred linearly between initiation and completion of the 8-week treatment, and between that point and the end of the 26-week follow-up, respectively.

Adverse events from medication are expected to result in a reduction in utility scores of children with autism. The economic analysis considered the disutility caused by weight gain, which is one of the most common side effects of antipsychotics. Disutility data associated with the presence of weight gain in children with autism were reported in Tilford and colleagues (2012), but these were generated using QWB-SA and therefore did not meet NICE requirements, as discussed in Chapter 11 (section 11.2.2). Moreover, the study showed discrepancies between utility scores generated using HUI3 and those generated using QWB-SA, and therefore utility scores derived from these 2 measures could not be combined in the economic model. Instead, the economic analysis utilised relevant data from Lenert and colleagues (2004), who reported the disutility caused by weight gain in adults with schizophrenia; HRQoL in this population was measured using the Positive and Negative Syndrome Scale (PANSS), a schizophrenia-specific measure, and utility values were elicited from members of the US public using SG.

Table 140 presents the values of clinical input parameters as well as the utility data that were used to populate the economic model.

Table 140. Clinical input parameters and utility data used to populate the economic model of antipsychotics versus placebo for the management of behaviour that challenges in children and young people with a learning disability

Input parameter	Deterministic value	Probabilistic distribution	Source of data - comments
Clinical input parameters			
Probability of non-improvement of behaviour that challenges at end of treatment – placebo	0.803	**Beta distribution** α= 147, β= 36	Weighted pooled rate for placebo, guideline meta-analysis (ITT)
Risk ratio of non-improvement of behaviour that challenges • risperidone versus placebo • aripiprazole versus placebo	0.46 0.65	**Log-normal distribution** 95% CIs: 0.26 to 0.82 95% CIs: 0.52 to 0.81	Guideline meta-analysis (ITT)
Probability of relapse over 26 weeks – all model arms	0.32	**Beta distribution** α= 19, β= 41	Pooled weighted rate for antipsychotic continuation arms in relapse prevention trials, guideline meta-analysis
Risk of non-weight gain – placebo	0.97	**Beta distribution** α= 241, β= 8	Pooled weighted rate for placebo, guideline meta-analysis
Risk ratio of non-weight gain • risperidone versus placebo • aripiprazole versus placebo	0.91 [0.85, 0.96] 0.79 [0.71, 0.88]	**Log-normal distribution** 95% CIs: 0.85 to 0.96 95% CIs: 0.71 to 0.88	Guideline meta-analysis (ITT)
Utility scores			
Mild hyperactivity Moderate hyperactivity	0.72 0.66	**Beta distribution** α= 129.92, β= 50.52 α= 153.82, β= 79.24	Tilford et al., (2012); distribution estimated using method of moments. Utility score for 'mild hyperactivity' not allowed to fall below that for 'moderate hyperactivity' in the probabilistic model
Weight gain – multiplicative function	0.96	α= 379.99, β= 16.25	Lenert et al., (2004); distribution estimated using method of moments. Value needs to be multiplied by base condition utility score to give the overall utility in the presence of weight gain

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12.2.2.3.61 Cost data

The intervention cost of antipsychotics consists of the drug acquisition cost and the cost of clinical management (healthcare professional time). The intervention cost of placebo comprises the cost of clinical management only. Healthcare professional time was estimated to be the same across all arms of the model, and was therefore excluded from further consideration. Consequently, in the economic analysis the intervention cost of antipsychotics included exclusively drug acquisition costs, while the intervention cost of placebo was zero.

As described earlier, the model considered all 3 available formulations of risperidone (tablets, orodispersible tablets and oral solution) and the only available formulation of aripiprazole (tablets). The daily dosage of drugs was determined by the daily dosage administered in the trials that provided clinical data to the economic model. The acquisition costs of the various formulations of risperidone and of aripiprazole tablets were taken from the Electronic Drug Tariff for England and Wales, April 2014 (NHS, 2014). Daily dosage and drug acquisition costs are presented in Table 141.

Costs incurred by behaviour that challenges were not included in the analysis due to unavailability of relevant data, but it is recognised that behaviour that challenges incurs significant extra costs to health and social care services; such costs may include, for example, costs associated with provision of CAMHS inpatient services, admission to long-term care settings or special education costs. Costs of treating side effects were also not included in the analysis; it is likely that the cost of managing weight gain, which is the only adverse event that was considered in the model structure, is not substantial and in most cases is included in the monitoring costs relating to healthcare professional time, as part of the intervention cost. However, there are other adverse events, such as extrapyramidal symptoms, that require more intensive clinical management and consequently may incur considerable healthcare costs. Omission of costs associated with the presence of behaviour that challenges and with side effects from antipsychotic medication is acknowledged as a limitation of the analysis.

As the time horizon of the analysis was 34 weeks, no discounting of costs and outcomes was necessary.

Table 141. Drug acquisition costs considered in the economic analysis of antipsychotics aimed at behaviour that challenges in children and young people with a learning disability

Drug	Dosage (per day)	Daily cost per person	Notes on estimation of cost (NHS, 2014)
Risperidone – tablets	1.5mg	£0.10	Risperidone (non-proprietary) 0.5mg 20 tablets - £1.05; 1mg 20 tablets – £0.90
Risperidone – oral solution	1.5mg	£0.58	Risperidone (non-proprietary) oral solution 1mg/ml - 100ml - £38.43
Risperidone – orodispersible tablets	1.5mg	£1.57	Risperidone (non-proprietary) 0.5mg 28 orodispersible tablets - £23.32; 1mg 28 orodispersible tablets – £20.61
Aripiprazole – tablets	5mg or 10mg or 15mg	£3.43	Abilify© 5mg or 10mg or 15mg - 28 tablets - £96.04

12.2.2.3.73 Handling uncertainty

Model input parameters were synthesised in a probabilistic analysis. This means that model input parameters were assigned probability distributions (rather than being expressed as point estimates), to reflect the uncertainty characterising the available data. Subsequently, 10,000 iterations were performed, each drawing random values out of the distributions fitted

onto the model input parameters. Results (mean costs and QALYs for each intervention) were averaged across the 10,000 iterations. This exercise provides more accurate estimates than those derived from a deterministic analysis (which utilises the mean value of each input parameter ignoring any uncertainty around the mean), by capturing the non-linearity characterising the economic model structure (Briggs et al., 2006).

The probability of non-improvement of behaviour that challenges following initial treatment with placebo (8 weeks), the 6-month probability of relapse following improvement and the risk of non-weight gain with placebo were assigned a beta distribution. Beta distributions were also assigned to utility values, using the method of moments. The risk ratio of non-improvement of behaviour that challenges for parent training versus wait list was assigned a log-normal distribution. Risk ratios were assigned a log-normal distribution. Drug costs were not assigned a distribution as there is no uncertainty around their cost. The estimation of distribution ranges was based on the guideline meta-analysis and available data in the published sources of evidence.

Table 140 provides details on the types of distributions assigned to each input parameter and the methods employed to define their range.

In addition, 2 sensitivity analyses were undertaken using the following alternative assumptions:

- the risk of relapse over 26 weeks was concurrently altered for all interventions; a values of zero relapse risk for all interventions and a value of 1005 relapse risk for all interventions were tested (instead of the value of 0.32 that was utilised in the base-case scenario)
- the study population was assumed to have HRQoL corresponding to severe levels of hyperactivity (instead of moderate) at initiation of treatment, as reported in Tilford and colleagues (2012)

12.2.2.3.8 Presentation of the results

Results are presented in the form of an incremental analysis, where all options have been ranked from the most to the least effective (in terms of QALYs gained). Options that are dominated by absolute dominance (i.e. they are less effective and more costly than 1 or more other options) or by extended dominance (i.e. they are less effective and more costly than a linear combination of 2 alternative options) are excluded from further analysis. Subsequently, ICERs are calculated for all pairs of consecutive options remaining in analysis.

In addition, as the GDG considered that not all drugs/formulations are suitable to all children and young people with a learning disability and behaviour that challenges, the ICER of each antipsychotic versus placebo was estimated.

Finally, the CEAC which shows the probability of each intervention being cost-effective at various cost effectiveness thresholds, including the NICE cost effectiveness thresholds of £20,000 and £30,000/QALY (NICE, 2008) is presented.

Results of the probabilistic analysis are presented in this chapter. Results of the deterministic analysis are provided in Appendix W. Appendix W also provides cost effectiveness planes, showing in graphic form the incremental costs and QALYs of each intervention versus placebo.

12.2.2.3.9 Validation of the economic model

The economic model (including the conceptual model and the Excel spreadsheet) was developed by the health economist working on this guideline and checked by a second modeller not working on the guideline. The model was tested for logical consistency by setting input parameters to null and extreme values and examining whether results changed

in the expected direction. The results were discussed with the GDG to confirm their plausibility.

12.2.2.3.10 Results

Over the 34 weeks of the analysis, risperidone and aripiprazole resulted in 1.17 and 0.58 additional QALYs, respectively, per 100 children and young people with a learning disability and behaviour that challenges compared with placebo. Risperidone in tablet formulation dominated all other options, as it has the lowest acquisition cost. However, ICERs of all assessed drug/formulation options versus placebo were calculated, as different drugs/formulations of a drug may be indicated for different sub-groups of children and young people with a learning disability and behaviour that challenges, and in such cases their cost effectiveness relative to placebo is relevant.

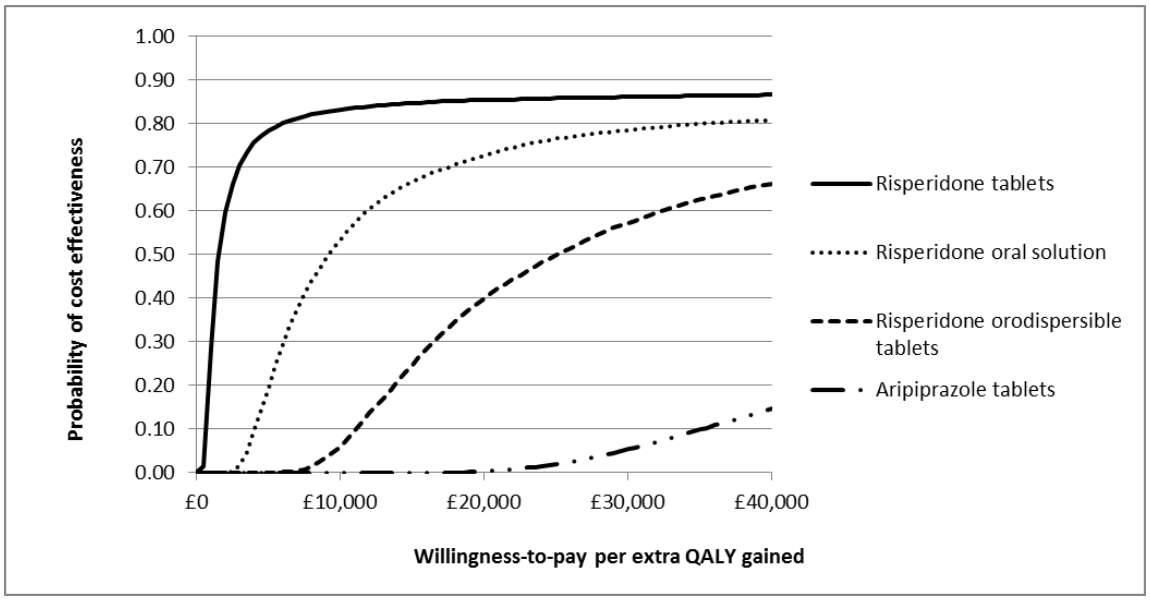
The ICERs of the 3 formulations of risperidone, that is, tablet, oral solution and orodispersible tablet were £1,401/QALY, £8,281/QALY, and £22,537/QALY, respectively. The first 2 ICERs are below the NICE lower cost effectiveness threshold of £20,000/QALY, and the 3rd ICER is above the lower but below the upper NICE cost effectiveness threshold of £30,000/QALY. The ICER of aripiprazole versus placebo is well beyond the NICE upper cost effectiveness threshold of £30,000/QALY, at £49,586/QALY. Full results of the base-case economic analysis are presented in Table 142.

Table 142. Results of economic analysis of antipsychotics versus placebo for the management of behaviour that challenges in children and young people with a learning disability – mean costs and QALYs for 100 children and young people receiving treatment

Antipsychotic drug	Mean cost		Mean QALYs		Incremental analysis (£/QALY)	ICER versus placebo (£/QALY)
	Total	Increm	Total	Increm		
Risperidone – tablets	£1,636	-£8,035	44.91	0	£1,401	£1,401
Risperidone – oral solution	£9,671	-£16,650	44.91	0	Dominated	£8,281
Risperidone – orodispersible tablets	£26,321	-£22,517	44.91	0.59	Dominated	£22,537
Aripiprazole – tablets	£48,838	£48,838	44.32	0.58	Dominated	£84,915
Placebo	£0	0	43.75			

The CEAC shown in Figure 7 illustrates the probability of each antipsychotic drug being cost effective compared with placebo. Full incremental analysis considering all antipsychotics resulted in a CEAC that was very similar to that of risperidone in tablets versus placebo, given that this treatment option dominated all other antipsychotic drug formulations in incremental analysis. The CEAC suggests that the probability of risperidone –tablets, risperidone – oral solution, risperidone – orodispersible tablets and aripiprazole being cost-effective each compared with placebo was 0.85, 0.73, 0.40 and 0.00, respectively, under the NICE lower cost effectiveness threshold; under the NICE upper cost effectiveness threshold this probability for each drug/formulation rose at 0.86, 0.79, 0.57 and 0.05, respectively.

Figure 7. Cost effectiveness acceptability curve of each antipsychotic versus placebo for the management of behaviour that challenges in children and young people with a learning disability



When the risk of relapse over 26 weeks was assumed to be zero, risperidone in tablets remained the most cost effective drug, dominating all other drug treatments and having an ICER versus placebo of £1,191/QALY. The ICERs of the other drug formulations versus placebo were £7,041/QALY for risperidone oral solution, £19,164 for risperidone orodispersible tablet, and £68,493/QALY for aripiprazole tablets.

When the risk of relapse over 26 weeks was assumed to be 1, conclusions did not changed compared with base case analysis: risperidone in tablets remained the most cost effective drug, dominating all other drug treatments and having an ICER versus placebo of £2,258/QALY. The ICERs of the other drug formulations versus placebo were £13,350/QALY for risperidone oral solution, £36,334 for risperidone orodispersible tablet, and £177,339/QALY for aripiprazole tablets.

When the HRQoL of children and young people was assumed to correspond to severe hyperactivity at initiation of treatment, all ICERs were reduced. Risperidone in tablets still dominated all other drug treatment options considered in the analysis. The ICER of each drug formulation versus placebo became £633/QALY for risperidone tablets, £3,740/QALY for risperidone oral solution, £10,179 for risperidone orodispersible tablet, and £32,005/QALY for aripiprazole tablets.

12.2.2.3.11 Discussion of findings - limitations of the analysis

The results of the economic model indicate that, overall, antipsychotics are likely to be a cost-effective intervention for the management of behaviour that challenges in children and young people with a learning disability. In particular, risperidone either in tablets or oral solution was shown to be cost-effective, whereas the analysis indicated that aripiprazole is unlikely to be cost-effective at its current cost; nevertheless, the cost effectiveness of aripiprazole is expected to improve with higher severity of behaviour that challenges at initiation of treatment. The drug acquisition cost is an important driver of cost effectiveness, as more expensive drugs or formulations of the same drug are less cost-effective than options with lower acquisition cost (and possibly not cost-effective under NICE criteria). Of the drugs and drug formulations assessed, risperidone in tablet formulation was the least

costly and most cost-effective option. However, there may be instances where other formulations of risperidone or other antipsychotics may be more appropriate for some children and young people with a learning disability and behaviour that challenges, depending on the drug's side effect profile, contra-indications and other individual circumstances. The cost effectiveness of antipsychotics (in particular aripiprazole) improves when the severity of the behaviour that challenges is higher at initiation of treatment, as there is more scope for improvement in terms of the children's and young people's HRQoL.

The model considered a very limited number of antipsychotics that were assessed in the trials included in the guideline systematic review. The economic analysis was informed by 2 meta-analyses of efficacy data derived from 4 RCTs that reported improvement in behaviour that challenges (regarding severity) as a dichotomous outcome. Limited follow-up data derived from 2 trials were available. Regarding adverse events, the economic model considered the risk for weight gain and the resulting decrements in utility. Weight gain was selected for incorporation in the model structure as it is one of the most common adverse events associated with antipsychotic medication, and relevant clinical and utility data were available to populate the model. However, antipsychotic medication is linked to a number of other adverse events, such as extrapyramidal symptoms or elevation in prolactin levels, all of which have a negative impact on the HRQoL of children and young people with a learning disability and most likely incur extra healthcare costs for their management. These parameters (disutility due to adverse events other than weight gain and costs of management of adverse events) were not taken into account in the model. It should be noted that different antipsychotics have different side effect profiles, and this may potentially affect their relative cost effectiveness.

Estimation of QALYs was based on utility data derived from HUI3 responses of parents of children with autism in the US; these data were used as a proxy, as no health state-specific utility data for children and young people with a learning disability were identified in the literature. Utility scores for HUI3 have been elicited from members of the Canadian general population and therefore they are not directly applicable to the UK context. More importantly, HUI3 has not been designed for use in children, and may be neither directly relevant to symptoms experienced by children and young people with a learning disability nor adequately sensitive to capture small changes in the HRQoL of this population. Ideally an alternative utility measure should have been used for the estimation of QALYs, but at the moment no such measure designed specifically for children and young people with a learning disability and behaviour that challenges is available. The model also utilised disutility data associated with weight gain. These data were based on analysis of PANSS scores of adults with schizophrenia and subsequent elicitation of preferences for schizophrenia-related health states from members of the US public. Consequently, these data are not directly relevant to children and young people with a learning disability, but they were nevertheless utilised in the economic model due to lack of any other relevant data. Another point for consideration is that the model incorporated exclusively changes in the HRQoL of children and young people with a learning disability and behaviour that challenges. Consideration of the improvement in HRQoL of carers and the family would most probably increase the cost effectiveness of antipsychotics.

Costs incurred by behaviour that challenges were not included in the analysis due to unavailability of relevant data. However, behaviour that challenges requires extra healthcare resources for its management (Knapp et al., 2005) and is a common reason for admission to CAMHS inpatient services, long-term care settings or boarding schools. It is also likely that the presence of behaviour that challenges in this population incurs extra intangible as well as informal care costs to the family, which have not been taken into account in the economic analysis. This means that the cost effectiveness of antipsychotics for the management of behaviour that challenges in children and young people with a learning disability is probably higher than that estimated by the guideline economic analysis.

Taking into account the results and limitations of the analysis, it appears that antipsychotics, in particular those available as generics, are likely to be a cost-effective option for the management of behaviour that challenges in children and young people with a learning disability. Antipsychotics that currently have high acquisition costs, such as aripiprazole, are less likely to be cost-effective.

12.2.2.4 Economic modelling – melatonin for the management of sleep problems in children and young people with a learning disability

An economic model was constructed for this guideline, aiming to assess the relative cost effectiveness of 4 interventions (psychosocial intervention, melatonin, combination therapy of psychosocial intervention and melatonin, and wait list) for the management of sleep problems in children and young people with a learning disability. Detailed methods and results are provided in Chapter 11 (section 11.2.2.2). The results of the analysis indicated that combination therapy of melatonin in tablets and psychosocial intervention is the most cost-effective option for the management of sleep problems in children and young people with a learning disability. Melatonin alone in tablets is also potentially cost-effective in the management of sleep problems in children and young people with a learning disability. The analysis was characterised by a number of limitations, including the limited evidence base, lack of long-term clinical data, lack of appropriate data on costs associated with sleep problems, omission of the impact of side effects from melatonin on costs and HRQoL, and lack of directly relevant utility data.

12.2.3 Clinical evidence statements

12.2.3.1 Antipsychotics: risperidone versus placebo for behaviour that challenges in children and young people

- Low quality evidence from 4 studies (N = 257), suggested that risperidone was more effective than placebo in reducing the severity of targeted behaviour that challenges at the end of intervention as measured by end point scores when compared with placebo. This effect was also found with change from baseline scores (k = 1; N = 66).
- Low quality evidence from 2 studies (N = 153) suggested that risperidone reduced the risk of the severity of targeted behaviour that challenges not being improved at the end of intervention when compared with placebo
- Low quality evidence from 3 studies (N = 155), suggested that risperidone was more effective than placebo at improving adaptive social functioning at the end of intervention when compared with placebo.
- Low to very low quality evidence from up to 3 studies (N = 241) suggested that risperidone increased the risk of participants having elevated prolactin levels, and that those treated with risperidone had higher levels of prolactin when compared with placebo at the end of intervention.
- Low to very low quality evidence from up to 3 studies (N = 282) suggested that risperidone was associated with greater weight gain when based on change from baseline and endpoint scores than placebo at the end of treatment. However, the precision of the estimate based on endpoint scores was poor.
- Very low quality evidence from 6 studies (N = 550) suggested that risperidone was associated with increased levels of sedation and somnolence when compared with placebo.
- Very low quality evidence from 5 studies (N = 450) suggested that risperidone was associated with a reduced risk of study discontinuation due to reasons other than adverse events when compared with placebo.

12.2.3.21 Antipsychotics: aripiprazole versus placebo for behaviour that challenges in children and young people

- Very low quality evidence from 2 studies (N = 308), suggested that aripiprazole was more effective than placebo in reducing the severity of targeted behaviour that challenges at the end of intervention when compared with placebo.
- Very low quality evidence from 2 studies (N = 308), suggested that aripiprazole reduced the risk of the severity of targeted behaviour that challenges not being improved at the end of intervention when compared with placebo.
- Very low quality evidence from 2 studies (N = 243) suggested that aripiprazole was more effective than placebo in increasing quality of life at the end of intervention. However, the precision of this estimate is poor.
- Very low quality evidence from 2 studies (N = 313) was inconclusive as to whether aripiprazole was associated with elevated prolactin levels when compared with placebo at the end of intervention.
- Very low quality evidence from up to 2 studies (N = 313) suggested that aripiprazole was associated with greater levels of weight gain and increased the risk of clinically significant weight gain when compared with placebo at the end of intervention.
- Very low quality evidence from 2 studies (N = 313) suggested that aripiprazole increased the risk of sedation when compared with placebo at the end of intervention.
- Very low quality evidence from 2 studies (N = 316) suggested that aripiprazole was associated with a reduced risk of study discontinuation due to reasons other than adverse events when compared with placebo.

12.2.3.23 Antipsychotics: aripiprazole versus risperidone for behaviour that challenges in children and young people

- Very low quality evidence from a single study (N = 59) suggested that aripiprazole was less effective than risperidone in reducing the severity of targeted behaviour that challenges at the end of intervention. However, the precision of this estimate is poor.

12.2.3.24 Antipsychotics: olanzapine versus haloperidol for behaviour that challenges in children and young people

- Very low quality evidence from a single study (N = 12), suggested that olanzapine was more effective than haloperidol in reducing the severity of behaviour that challenges at the end of intervention.
- Very low quality evidence from a single study (N = 12), suggested that olanzapine increased drowsiness to a greater extent than haloperidol. However, the precision of this estimate was poor.
- Very low quality evidence from a single study (N = 12) suggested that olanzapine increased weight gain to a greater extent than haloperidol.

12.2.3.25 Antipsychotics: withdrawal of risperidone versus continuation of risperidone for behaviour that challenges in children and young people

- Very low quality evidence from a single study (N = 32), suggested that participants who initially responded to treatment with risperidone and were subsequently withdrawn from this intervention were at an increased risk of demonstrating the targeted behaviour that challenges when compared with participants who continued treatment.

12.2.3.26 Antipsychotics: withdrawal of aripiprazole versus continuation of aripiprazole for behaviour that challenges in children and young people

- Very low quality evidence from a single study (N = 85), suggested that participants who initially responded to treatment with aripiprazole and were subsequently withdrawn from

this intervention were at an increased risk of demonstrating the targeted behaviour that challenges when compared with participants who continued treatment. However, the precision of this estimate is poor.

12.2.3.7 Anticonvulsants: topiramate (plus risperidone) versus placebo (plus risperidone) for behaviour that challenges in children and young people

- Very low quality evidence from a single study (N = 40), suggested that combined treatment with topiramate and risperidone was more effective in reducing the severity of targeted behaviour that challenges at the end of intervention when compared with combined treatment with placebo and risperidone.

12.2.3.8 Anticonvulsants: valproate versus placebo for behaviour that challenges in children and young people

- Very low quality evidence from 2 studies (N = 57) was inconclusive as to the effectiveness of valproate, when compared with placebo, in reducing the severity of targeted behaviour that challenges at the end of intervention.
- Very low quality evidence from a single study (N = 27), suggested that valproate reduced the risk of the severity of targeted behaviour that challenges not being improved at the end of intervention when compared with placebo.

12.2.3.9 GABA analogue: piracetam (plus risperidone) versus placebo (plus risperidone) for behaviour that challenges in children and young people

- One trial could not be included in the meta-analysis of behaviour that challenges outcomes due to the format in which data were presented (N = 40). The authors reported that combined treatment with piracetam and risperidone reduced the severity of targeted behaviour that challenges at end of intervention to a greater extent than combined treatment with placebo and risperidone.

12.2.3.10 Antioxidants: N-acetylcysteine versus placebo for behaviour that challenges in children and young people

- Very low quality evidence from a single study (N = 29), suggested that N-acetylcysteine was more effective than placebo in reducing the severity of behaviour that challenges at the end of intervention. However, the precision of this estimate is poor.

12.2.3.11 Biomedical interventions: omega-3 versus placebo for behaviour that challenges in children and young people

- Very low evidence from a single study (N = 12) was inconclusive as to the effectiveness of omega-3, when compared with placebo, in reducing the severity of behaviour that challenges at the end of intervention.

12.2.3.12 Biomedical interventions: ginkgo biloba (plus risperidone) versus placebo (plus risperidone) for behaviour that challenges in children and young people

- Very low evidence from a single study (N = 47) was inconclusive as to the effectiveness of combined treatment with ginkgo biloba and risperidone, when compared with combined treatment with placebo and risperidone, in reducing the severity of targeted behaviour that challenges at the end of intervention.

12.2.3.13 Antipsychotics: risperidone versus placebo for behaviour that challenges in adults

- Low quality evidence from 2 studies (N = 88) was inconclusive as to the effectiveness of risperidone, when compared with placebo, in reducing the severity of targeted behaviour that challenges at the end of a 12 and 26 week intervention as measured by end point scores when compared with placebo.
- Very low quality evidence from a single study (N = 74), suggested that risperidone was more effective than placebo in reducing the severity of targeted behaviour that challenges at the end of a 12 week intervention as measured by change from baseline scores when compared with placebo. However, the precision of this estimate is poor.
- Low quality evidence from a single study was inconclusive as to the effectiveness of risperidone, when compared with placebo, in improving quality of life at the end of a 12 (N = 58) and 26 (N = 40) week intervention.
- Low quality evidence from a single study (N = 30), suggested that risperidone was more effective than placebo in improving adaptive social functioning at the end of a 12 week intervention.
- Very low quality evidence from 2 studies (N = 108) suggested that risperidone increased the risk of somnolence and sedation when compared with placebo. However, the precision of this estimate was poor.

12.2.3.14 Antipsychotics: haloperidol versus placebo for behaviour that challenges in adults

- Low quality evidence from a single study (N = 57), suggested that haloperidol was more effective than placebo in reducing the severity of targeted behaviour that challenges at the end of a 12 week intervention. However, the precision of this estimate is poor.
- Low quality evidence from a single study (N = 40) was inconclusive as to the effectiveness of haloperidol, when compared with placebo in reducing the severity of targeted behaviour that challenges at the end of a 26 week intervention.
- Low quality evidence from a single study was inconclusive as to the effectiveness of haloperidol, when compared with placebo in improving quality of life at the end of a 12 (N = 57) and 26 (N = 41) week intervention.

12.2.3.15 Antipsychotics: risperidone versus haloperidol for behaviour that challenges in adults

- Low quality evidence from a single study (N = 57), suggested that risperidone was less effective than haloperidol in reducing the severity of behaviour that challenges at the end of a 12 week intervention although the precision of this estimate is poor. Moreover, at the end of a 26 week intervention, low quality evidence was inconclusive (N = 36) as to the effectiveness of risperidone over haloperidol in reducing the severity of behaviour that challenges.
- Low quality evidence from a single study suggested that risperidone was more effective than haloperidol in improving quality of life at the end of a 12 (N = 57) and 26 (N = 39) week intervention. However, the precision of both estimates are poor.

12.2.3.16 Antipsychotics: olanzapine versus risperidone for behaviour that challenges in adults

- Very low quality evidence from a single study (N = 62) was inconclusive as to the effectiveness of olanzapine, when compared with risperidone, in reducing the frequency of behaviour that challenges at the end of intervention.
- Very low quality evidence from a single study (N = 62) suggested that risperidone was associated with elevated prolactin levels when compared with olanzapine.

12.2.3.17 Antipsychotics: withdrawal of zuclopenthixol versus continuation of zuclopenthixol for behaviour that challenges in adults

- Very low quality evidence from a single study (N = 39), suggested that participants who initially responded to treatment with zuclopenthixol and were subsequently withdrawn from this intervention were at an increased risk of demonstrating the behaviour that challenges when compared with participants who continued treatment.
- Very low quality evidence from 2 studies (N =124), suggested that withdrawal of zuclopenthixol was less effective than continuation of zuclopenthixol in reducing the severity of behaviour that challenges as measured by end point scores and change from baseline scores at the end of intervention. However, the precision of this estimate is poor.
- Very low quality evidence from a single study (N = 43) was inconclusive as to the effectiveness of withdrawal of zuclopenthixol when compared with continuation of zuclopenthixol in reducing the risk of participants presenting behaviour that challenges in the form of staff reported problems in management at the end of intervention.
- Very low quality evidence from a single study (N = 85), suggested that withdrawal of zuclopenthixol was less effective than continuation of zuclopenthixol in improving adaptive social functioning at the end of intervention.
- Very low quality evidence from a single study (N = 39), suggested that withdrawal of zuclopenthixol was associated with lower weight gain when compared with continuation of zuclopenthixol at the end of intervention.
- Very low quality evidence from a single study (N = 42) was inconclusive as to whether continuation of zuclopenthixol increased drowsiness to a greater extent than withdrawal of zuclopenthixol.
- Very low quality evidence from up to 3 studies (N = 204) suggested that withdrawal of zuclopenthixol was associated with increased risk of study discontinuation due to adverse events and discontinuation due to other reasons when compared with continuation of zuclopenthixol. However, the precision of this estimate was poor.

12.2.3.18 Mood stabilisers: lithium versus placebo for behaviour that challenges in adults

- Very low quality evidence from a single study (N = 42), suggested that lithium reduced the risk of the severity of targeted behaviour that challenges not being improved at the end of intervention when compared with placebo

12.2.3.19 Naltrexone versus placebo for self-injurious behaviour in adults

- Trials could not be included in the meta-analysis due to differences in study designs, dose and outcome format. The authors of Symons 2001 (N = 4) reported that naltrexone reduced the frequency of targeted behaviour that challenges in 3 of the 4 participants at the end of intervention when compared with placebo. Similarly, the authors of Sandman 1990 (N = 4) reported that naltrexone reduced targeted behaviour that challenges in all participants. Evidence from both studies was very low quality.
- The authors of Thompson 1994 (N = 8) reported that when compared with placebo, naltrexone reduced the number of days of high frequency self-injurious behaviour and increased the number of days of low frequency self-injurious behaviour. However, the effects of naltrexone differed depending on the form and location of self-injury. Evidence was very low quality.
- The authors of Willemsen-Swinkels 1995 (N = 26) reported that neither the single dose nor long-term treatment with naltrexone had any beneficial effects on targeted behaviour that challenges. Evidence was very low quality.

12.2.3.20 Clomipramine versus placebo for self-injurious behaviour in adults

- One trial could not be included in the meta-analysis due to the format in which data were presented (N = 8). The authors of Lewis 1996 reported no benefit of clomipramine, when compared with placebo, on the severity or frequency of the targeted behaviour that challenges at the end of intervention. The evidence was of very low quality.

12.2.3.21 Melatonin versus placebo for sleep problems in children

- Very low quality evidence suggested that melatonin was more effective than placebo at reducing global problem sleep behaviour when measured by both the Children's Sleep Habit Questionnaire (k = 1; N = 66) and the Composite Sleep Disturbance Index (k = 1; N = 125) at end of intervention. However, the precision of the estimate for the Composite Sleep Disturbance Index was poor.
- Very low quality evidence from a single study (N = 66) suggested that melatonin reduced the risk of problem sleep behaviour not being improved at the end of intervention when compared with placebo.
- Very low quality evidence from 2 studies (N = 125) suggested that melatonin was more effective than placebo at increasing actigraph assessed sleep efficiency and total sleep time at end of intervention. However, the precision of both estimates was poor.
- Moderate quality evidence from up to 3 studies (N = 172) suggested that melatonin was more effective than placebo at reducing both actigraph and sleep diary assessed wake after sleep onset at end of intervention.
- Very low quality evidence from a single study (N = 66) suggested that melatonin was more effective than placebo at reducing actigraph assessed sleep onset latency at the end of intervention.
- Low quality evidence from 3 studies (N = 169) suggested that melatonin was more effective than placebo at increasing sleep diary assessed total sleep time at the end of intervention.
- Moderate quality evidence from 3 studies (N = 164) was inconclusive as to the effectiveness of melatonin when compared with placebo at reducing sleep diary assessed number of wakes per night and duration of wakes at the end of intervention.
- Moderate quality evidence from 3 studies (N = 173) suggested that melatonin was more effective than placebo at reducing wake after sleep onset at the end of intervention.

12.2.3.22 Melatonin versus CBT for sleep problems in children

- Very low quality evidence from a single study (N = 67) suggested that melatonin was more effective than CBT at reducing global problem sleep behaviour at end of intervention.
- Very low quality evidence from a single study (N = 67) suggested that melatonin reduced the risk of sleep onset latency not being improved at the end of intervention when compared with CBT.
- Very low quality evidence from a single study (N = 67) suggested that melatonin was more effective than CBT at reducing actigraph assessed sleep onset latency and wake after sleep onset at end of intervention.
- Very low quality evidence from a single study (N = 67) suggested that melatonin was more effective than CBT at increasing actigraph assessed total sleep time and sleep efficiency at end of intervention.
- Very low quality evidence from a single study (N = 80) suggested that melatonin was not associated with an increased risk of study discontinuation when compared with placebo.

12.2.14 Economic evidence statements

- Low quality evidence from 1 single study (N=86) suggests that risperidone and haloperidol are unlikely to be cost-effective in adults with a learning disability and behaviour that challenges. Evidence is based on an analysis that has not used the QALY as the measure of outcome and conclusions depended on the measure of outcome used and the willingness to pay for an additional unit of benefit.
- Low quality evidence from the guideline economic analysis suggested that risperidone either in tablets or oral solution was cost-effective in the management of behaviour that challenges in children and young people with a learning disability.
- According to the guideline economic analysis, aripiprazole was not cost-effective in the management of behaviour that challenges in children and young people with a learning disability; nevertheless, its cost effectiveness is expected to improve once the drug becomes available in generic form.
- Low quality from the guideline economic analysis suggests that melatonin in tablets is likely to be more cost-effective than psychological intervention and wait list in the management of sleep problems in children and young people with a learning disability.
- Combined therapy of melatonin (in tablets) and psychological intervention appears to be the most cost-effective treatment option for the management of people and young people with a learning disability.
- All guideline economic analyses were characterised by a number of potentially serious limitations relating to limited evidence base, lack of long-term clinical data, lack of appropriate data on costs associated with behaviour that challenges and sleep problems, lack of (or limited) consideration of the impact of side effects of drugs on HRQoL and costs, and lack of directly relevant utility data.

12.3 Recommendations and link to evidence

Recommendations	
	46. Consider medication for people with a learning disability and behaviour that challenges if: • the person has a coexisting mental or physical health problem (see recommendation 33) **or** • psychosocial, psychological or other interventions alone do not produce change within the specified time **or** • the risk to the person or others is very severe. Only offer medication in combination with psychosocial, psychological or other interventions. 47. When prescribing medication for behaviour that challenges, take into account side effects and develop a care plan that includes: • a rationale for medication, explained to family members and carers • how long the medication should be taken for • a strategy for reviewing the prescription and stopping the medication. 48. Consider antipsychotic medication for behaviour that challenges if psychological or other interventions are insufficient or cannot be delivered alone because of the severity of risk to self or others. Antipsychotic medication

should initially be prescribed and monitored by a specialist (an adult or child psychiatrist, or a neurodevelopmental paediatrician with expertise in learning disabilities) who should:

- identify the target behaviour
- decide on a measure to monitor effectiveness (for example, direct observations, the Aberrant Behaviour Checklist or the Adaptive Behaviour Scale), including frequency and severity of the behaviour and impact on functioning
- start with a low dose and use the minimum effective dose needed
- only prescribe a single drug
- review the effectiveness and any side effects of the medication after 3–4 weeks
- stop the medication if there is no indication of a response at 6 weeks
- not prescribe p.r.n. (as-needed) medication for more than 4 weeks
- review the medication if the person's environmental or personal circumstances change.

49. When choosing which antipsychotic medication to offer, take into account side effects, acquisition costs, the person's preference (or that of their family member or carer, if appropriate) and response to previous antipsychotic medication.

50. If there is a positive response to antipsychotic medication:
    - conduct a full multidisciplinary review after 3 months and then at least every 6 months covering all prescribed medication (including effectiveness, side effects and plans for stopping)
    - only continue to offer medication that has proven benefit.

51. When prescribing is transferred to primary or community care, or between services, the specialist should give clear guidance to the practitioner responsible for continued prescribing about:
    - which behaviours to target
    - monitoring of beneficial and side effects
    - taking the lowest effective dose
    - how long the medication should be taken for
    - plans for stopping the medication.

52. For the use of rapid tranquillisation, follow the NICE guideline on violence and aggression (update in progress; publication expected May 2015).

	53. Do not offer medication to aid sleep unless the sleep problem persists after a behavioural intervention, and then only: • after consultation with a psychiatrist (or a specialist paediatrician for a child or young person) with expertise in its use in people with a learning disability • together with non-pharmacological interventions and regular reviews (to evaluate continuing need and ensure that the benefits continue to outweigh the risks). If medication is needed to aid sleep, consider melatonin.[h]
Relative values of different outcomes	The GDG agreed that a number of outcomes were critical to addressing this review question: behaviour that challenges, sleep problems, harms (for example weight gain, raised hormone levels and seizures), sedation, discontinuation, quality of life, and service user and carer satisfaction.
Trade-off between clinical benefits and harms	The benefits of medication, principally antipsychotic medication on behaviour that challenges were demonstrated in this review but outcomes were mainly short-term and data on long-term benefits were sparse. There was evidence of harms including weight gain and raised prolactin levels as well as evidence of sedation; data on other potential long-term harms were absent. The evidence for the use of antipsychotic medication for children was of better quality than that for adults but the concerns about potential harms (for example raised prolactin levels) were also higher. Data for other medication other than antipsychotics were very limited with the exception of melatonin for sleep problems.
Trade-off between net health benefits and resource use	Limited evidence failed to demonstrate that antipsychotics are cost effective in the management of behaviour that challenges in adults with a learning disability. Risperidone appears to be cost effective in the management of behaviour that challenges in children and young people with a learning disability, regardless of the formulation used. In contrast, aripiprazole does not appear to be a cost-effective treatment option; nevertheless, its cost effectiveness is expected to improve once aripiprazole becomes available in generic form. Melatonin (in tablets) is likely to be more cost-effective than psychological intervention and wait list in the management of sleep problems in children and young people with a learning disability. Combined therapy of melatonin (in tablets) and psychological intervention appears to be the most cost-effective treatment option for the management of people and young people with a learning disability. The GDG noted that, as costs associated with behaviour that challenges and sleep problems in children and young people with a learning disability (such as costs incurred by health professional contacts, need for special education and residential placements) were not taken into account in the guideline economic models, it was very likely that the cost effectiveness of all drug treatment options had been underestimated. On the other

h This recommendation also appears in section 11.3

	hand, the GDG took into account the fact that the economic models did not capture reductions in HRQoL and costs associated with management of adverse events from medication, apart from the impact of weight gain on HRQoL. This is likely to have biased guideline economic analyses in favour of drugs. Finally, the GDG considered other limitations of the guideline economic analyses, such as the limited evidence base, the lack of long-term clinical data and the lack of directly relevant utility data, which may have affected the results of the economic analyses.
Quality of evidence	The evidence for almost all comparisons for all medication was very low or low. Considerable caution is required in the interpretation of the data. Further problems may arise as a result of publication bias.
Other considerations	The GDG faced a number of problems in developing recommendations on the use of medication for behaviour that challenges: (1) the low quality of most of the evidence and (2) the evidence of potential harms, which was in line with known harms from much larger datasets (for example the use of antipsychotic medication in adults with severe mental illness). Importantly the GDG was aware of the significant concerns of service users and carers about the potential over use of medication to manage behaviour that challenges and the limited review and monitoring of medication once prescribed, In addition the GDG was also aware that the evidence was limited but better for use in children and young people than in adults, which was set against the greater concerns about potential harms to children. Having carefully reviewed the evidence, the GDG decided that there was a place for the use of antipsychotic medication but that its use should be limited in the following ways. It should only be used where no or limited benefit has been derived from a psychosocial intervention or where there is an immediate need to prevent harm to the self or others from severe behaviour that challenges. Use of antipsychotics should be also be very closely reviewed and monitored and stopped if no benefit is demonstrated. The GDG was also clear that if as part of the assessment of behaviour that challenges a mental disorder was identified then the pharmacological treatment of that should follow existing NICE guidance. The GDG also considered whether to recommend a particular antipsychotic drug (the best available evidence was for risperidone) but decided not to do so because they were concerned that limiting choice in the absence of evidence of effect for a range of other drugs might limit access to a beneficial intervention if there was no response to a particular drug. With the exception of melatonin for sleep problems there was insufficient evidence to recommend the use of drugs other than antipsychotics. The GDG decided to recommend melatonin for use in the management of sleep problems, in combination with psychosocial interventions (see Chapter 11 for further details)

12.3.12 Research recommendations

5. Are applied behavioural analysis interventions and antipsychotic medication, or a combination of these, effective in reducing the frequency and severity of behaviour that challenges in adults with a learning disability?[i]

i Please note, this research recommendation also appears in section 11.3.2.

1

13 Reactive strategies

13.1 Introduction

Reactive strategies are actions, responses and planned interventions in response to the presentation of identifiable behaviour that challenges. Reactive strategies have the aim of bringing about immediate behavioural change in an individual or establishing control over a situation so that risk associated with the presentation of the behaviour is minimised or eradicated. Reactive strategies may take a number of forms and can include environmental, psychosocial and restrictive interventions such as physical holds, mechanical and manual restraint, seclusion and 'time out' or the use of emergency medication. It is suggested that up to half of people with a learning disability who display behaviour that challenges may be subject to reactive strategies (Paley, 2013).

Reactive strategies do not aim to achieve long-term behaviour change, however those strategies that are aversive or punitive have the potential to change an individual's behaviour through negative association with displaying particular behaviours. Much research in the 1970s and 1980s focused on alternatives to punishment and aversive strategies. More recently interventions that focus on upholding an individual's human rights have come to the fore. Such approaches treat people with dignity and respect, have an ethical basis and are delivered alongside proactive strategies in order to reduce the likelihood of behaviour that challenges. Reactive strategies are more likely to be effective in the context of good person-centred planning that recognises the situations, environment, social settings or interpersonal environments that are associated with a higher likelihood of behaviour that challenges and seeks to affect change in those settings. Traditional behaviour support planning typically draws on a menu of reactive strategies including: environmental change; stimulus control, cessation or introduction; preferred activities; preferred interactions/people; distraction, diffusion and de-escalation.

Guidance issued on the subject of behavioural support, reactive strategies and restrictive practices has taken on a generic health and social care focus where previously specific guidance for people with a learning disability and behaviour that challenges was published (Paley, 2013). However, the focus has continued to be on the principles of least restrictive alternatives, proportionality to the risks posed by the behaviour and gradient approaches to any reactive or restrictive interventions, considering restrictive interventions a last resort.

13.2 Review question: In people with a learning disability and behaviour that challenges, what are the benefits and potential harms of 'reactive strategies' aimed at managing behaviour that challenges?

The review protocol summary, including the review question and the eligibility criteria used for this section of the guideline, can be found in Table 143. A complete list of review questions and review protocols can be found in Appendix F; further information about the search strategy can be found in Appendix H.

Table 143: Clinical review protocol summary for the review of reactive strategies aimed at reducing and managing behaviour that challenges

Component	Description
Review question	In people with a learning disability and behaviour that challenges, what are the benefits and potential harms of 'reactive strategies' (including physical restraint, mechanical restraint, confinement, and containment and seclusion) aimed at managing behaviour that

Component	Description
	challenges? (RQ4.4)
Population	Children, young people and adults with a mild, moderate, severe or profound learning disability and behaviour that challenges.
Intervention(s)	All reactive strategies, including physical restraint, mechanical restraint, confinement, and containment and seclusion.
Comparison	• Treatment as usual • No treatment, placebo, waitlist control, attention control • Any alternative management strategy
Critical outcomes	• Targeted behaviour that challenges • Rates of manual restraint • Rates of seclusion • Quality of life • Service user and carer satisfaction.
Study design	RCTs and systematic reviews.
Note. RCTs = Randomised controlled trials.	

13.2.11 Clinical evidence

No RCTs or systematic reviews of RCTs met the eligibility criteria for this review. A search for other systematic reviews identified only 1: Heyvaert 2014 (Heyvaert et al., 2014). An overview of the included systematic review can be found in Table 144. The review included 59 single-case or small-n studies (N = 94): Atcheson 2006 (Atcheson, 2006), Borrero 2002 (Borrero et al., 2002), Cameron 1996 (Cameron et al., 1996), Cannella-Malone 2008 (Cannella-Malone et al., 2008), Carr 2002 (Carr et al., 2002b), Chung & Cannella-Malone 2010 (Chung & Cannella-Malone, 2010a), Dura 1991 (Dura, 1991), Fisher 1996 (Fisher et al., 1996), Fisher 1997 (Fisher et al., 1997), Fisher 1998 (Fisher et al., 1998), Fox 2008 (Fox et al., 2008), Graff 1999 (Graff et al., 1999), Hanley 1998 (Hanley et al., 1998), Hanley 2000 (Hanley et al., 2000), Irvin 1998 (Irvin et al., 1998), Jena 1995(Jena, 1995), Jena 1999(Jena, 1999), Kahng 2001(Kahng et al., 2001), Kelley 2002 (Kelley et al., 2002), Kerth 2009 (Kerth et al., 2009), Lalli 1996 (Lalli et al., 1996), Le & Smith 2002 (Le & Smith, 2002), LeBlanc 1997 (LeBlanc et al., 1997), Lerman & Iwata 1996 (Lerman & Iwata, 1996), Lerman 1997 (Lerman et al., 1997), Lerman 2003 (Lerman et al., 2003), Lindberg 1999 (Lindberg et al., 1999), Luiselli 1991 (Luiselli, 1991), Luiselli 1998 (Luiselli, 1998), Matson & Keyes 1990 (Matson & Keyes, 1990), Mazaleski 1994 (Mazaleski et al., 1994), McCord 2001 (McCord et al., 2001), McCord 2005 (McCord et al., 2005), McKerchar 2001 (McKerchar et al., 2001), Moore 2004 (Moore et al., 2004), Mueller & Kafta 2006 (Mueller & Kafka, 2006), Northup 1997 (Northup et al., 1997), O'Connor 2003 (O'Connor et al., 2003), Piazza 1998 (Piazza et al., 1998), Rapp & Miltenberger 2000 (Rapp & Miltenberger, 2000), Rapp 2000 (Rapp et al., 2000), Rapp 2001 (Rapp et al., 2001), Reid 1993 (Reid et al., 1993), Richman 1998 (Richman et al., 1998), Roane 2001 (Roane S, 2001), Rolider 1991 (Rolider et al., 1991), Roscoe 1998 (Roscoe et al., 1998), Sisson 1993 (Sisson et al., 1993), Smith 1992 (Smith et al., 1992), Smith 1996 (Smith et al., 1996), Smith 1999 (Smith et al., 1999), Tarbox 2002 (Tarbox et al., 2002), Thompson 1998 (Thompson et al., 1998), Thompson 1999 (Thompson et al., 1999), Toole 2003 (Toole et al., 2003), Turner 1996 (Turner et al., 1996), Van Houten 1993 (Van Houten, 1993), Vollmer 1994 (Vollmer et al., 1994), Zhou 2000 (Zhou et al., 2000). Of the 59 included studies, 20 were identified through the search of electronic databases and 39 were identified through the manual hand search of relevant journals. Fifty-eight studies were published in peer reviewed journals between 1990 and 2010 and one study (Atcheson 2006) was a dissertation from the University of North Texas.

The 59 included studies included 94 participants. Of the included participants, 2% had mild learning disability, 4% moderate, 22% severe, 59% profound and 13% unspecified. The mean age of participants was 24 years (range = 3 to 58) and 51% were female. In 87% of

cases, the targeted CB type was internal maladaptive behaviour. A summary of the review can be found in Table 144 and Appendix N.

Further information about included and excluded studies can be found in Heyvaert 2014.

Using the Single-Case Experimental Design (SCED) Scale (Tate et al., 2008), the methodological quality of the 59 included studies was 7.31 (SD = 1.15; range = 4–9) out of a possible 11 (high scores represent better quality).

A sensitivity analysis was conducted to investigate influence of an outlying case on overall effect size: the conclusions regarding the main statistical analysis and the moderator analysis are the same for the full data set as for the data set without the one outlier.

The meta-analysis was judged to be of adequate quality because 4 of the 5 methodological quality criteria were met; the search of published primary studies was judged to have been unlikely to identify all relevant studies since many are not published (see Appendix N). With regard to the evidence, because of limitations inherent in single-case and small-n studies (see section 3.5.3), the evidence was graded as low quality.

Table 144: Study information table for the systematic review included in the review of reactive interventions

	Heyvaert 2014
Review question/ Aim	To evaluate the effectiveness of reactive interventions (including physical, mechanical and environmental restraint) for reducing behaviour that challenges
Method used to synthesise evidence	Multilevel meta-analysis • In addition, a moderator analysis was conducted to assess the moderating effect of 5 participant variables and 2 study variables.
Design of included studies	Single-case and small-n
Dates searched	January 1990 to September 2011
Electronic databases	Academic Search Premier, Cumulative Index to Nursing and Allied Health Literature, Embase, Education Resources Information Center, Medline, PsycINFO, PubMed and Web of Science.
Additional search methods	Manual hand search of the 32 relevant journals
No. of included studies (N[1])	59 (94)
Participant characteristics	People with a learning disability and behaviour that challenges
Intervention	Interventions responding to behaviour that challenges involving the limitation or restriction of movement or mobility: • Personal/ physical/ manual restraint • Mechanical restraint • Environmental restraint including seclusion, isolation, confinement and time-out. Excluded chemical restraint interventions and natural therapeutic holding interventions.
Comparison	N/A
Outcome	Targeted behaviour that challenges
Review Quality	Adequate
[1] Number of participants.	

The findings from the multi-level meta-analysis can be found in Table 145. In the table, Model 1 is the 3-level random effects regression model without moderators, Model 2 includes

all potential moderators, and Model 3 includes only those moderators that were statistically significant in Model 2.

Table 145: Parameter estimates and standard errors for the multilevel meta-analysis of reactive strategies

	Model 1	Model 2	Model 3
Fixed effects			
Mean treatment effect	-3.16 (0.45)***		-2.20 (0.60)***
Moderator effect of:			
Age		-0.01 (0.03)	
Gender		-1.96 (0.83)*	-1.88 (0.82)*
Type of behaviour that challenges		0.22 (0.78)	
Intellectual disabilities level		-0.99 (0.67)	
Restraint type		0.18 (0.58)	
Publication year		-0.01 (0.11)	
Study quality		-0.11 (0.46)	
Variance of effect			
Between studies	3.49 (2.27)	2.32 (1.66)	3.05 (2.19)
Between participants	12.21 (2.50)***	9.82 (2.07)***	11.88 (2.45)***
Residual variance	1.00 (0.02)***	1.00 (0.02)***	1.00 (0.02)***

*Notes: * = p < .05; ** = p < .01; *** = p < .001.*

13.2.2 Economic evidence

No economic evidence on reactive strategies aimed at reducing and managing behaviour that challenges in people with a learning disability was identified by the systematic search of the economic literature undertaken for this guideline. Details on the methods used for the systematic search of the economic literature are described in Chapter 3.

13.2.3 Clinical evidence statements

- In one systematic review with 59 included studies (94 participants), there was very low quality evidence that reactive strategies (restrictive interventions) may be effective in reducing behaviour that challenges when compared with not using reactive strategies. The effect varied across participants, but not studies.
- Based on the same review, there was very low quality evidence from a moderator analysis that reactive strategies, on average, appeared to be more effective for female than for male participants. The evidence suggested that age, type of behaviour that challenges, learning disabilities level, type of reactive strategy, publication year, and study quality were unlikely to be strongly associated with intervention effectiveness.

13.2.4 Economic evidence statements

No economic evidence on reactive strategies aimed at reducing and managing behaviour that challenges in people with a learning disability is available.

13.3 Recommendations and link to evidence

Recommendations	54. Only use reactive strategies for people with a learning disability and behaviour that challenges as a last resort and together with the proactive interventions described in 9.5, 10.3 and 11.3. When risks to self or others are significant or breakdown in the

	person's living arrangements is very likely, consider using reactive strategies as an initial intervention and introduce proactive interventions once the situation stabilises. 55. Plan reactive strategies from an ethically sound basis and use a graded approach that considers the least aversive and restrictive alternatives first. Encourage the person and their family members or carers to be involved in planning and reviewing reactive strategies whenever possible. **56.** If a restrictive intervention is used as part of a reactive strategy, carry out a thorough risk assessment. Take into account: • any physical health problems and physiological contraindications to the use of restrictive interventions, in particular manual and mechanical restraint • any psychological risks associated with the intervention • any known biomechanical risks, such as cardiovascular and musculoskeletal risks • any sensory sensitivities, such as a high or low threshold for pain or touch. 57. Ensure that any restrictive intervention is accompanied by a restrictive intervention reduction programme, as part of the long-term behaviour support plan, to reduce the use of and need for restrictive interventions. 58. Ensure that planned restrictive interventions: • take place within the appropriate legal framework of the Human Rights Act 1998, the relevant rights in the European Convention on Human Rights, the Mental Health Act 1983 and the Mental Capacity Act 2005, including the supplementary code of practice on deprivation of liberty safeguards • are in the best interest of the person to protect them or others from immediate and significant harm • are a reasonable, necessary and proportionate response to the risk presented. 59. Regularly review and reassess the safety, efficacy, frequency of use and continued need for reactive strategies. Document their use as part of an incident record and use this in personal and organisational debrief procedures to inform future behaviour support planning and organisational learning.
Relative values of different outcomes	The GDG agreed that a number of outcomes were critical to addressing this review question: targeted behaviour that challenges, rates of manual restraint, rates of seclusion, quality of life, and service user and carer satisfaction.

Trade-off between clinical benefits and harms	Reactive strategies in this review produced benefits which likely outweigh harms. However, the GDG was aware of the possible harms that could arise from the use of restrictive interventions, which include the loss of liberty and possible physical harms that might arise from manual or mechanical restraint. Reporting of harms was limited in the studies included in the systematic review and this is addressed in the other considerations below.
Trade-off between net health benefits and resource use	No economic evidence in this area is available. The interventions considered in this review may incur varying costs for their implementation, associated with staff time and training, and appropriate room space and/or equipment (for example, mechanical or environmental restraint). The GDG judged that provision of such interventions may result in benefits that outweigh costs; the main benefit of such interventions is a reduction in severe behaviour that challenges that is difficult to manage otherwise and which may pose an immediate risk to the service user and people involved with the person's care. However, decisions need to be made on the basis of safety of people with a learning disability and behaviour that challenges, their carers, family and health and social care staff and also consideration of human rights and compliance with existing legislation.
Quality of evidence	No RCTs met the eligibility criteria for this review, and therefore, a systematic review of single-case and small-n studies that focused on the effectiveness of restraint interventions for behaviour that challenges among people with a learning disability was used. The included studies were judged individually to be of adequate quality. Nevertheless, although the evidence was not formally graded it would be fair to consider it as no more than very low quality, primarily due to the potential for publication bias and inconsistency.
Other considerations	The evidence for a variety of reactive strategies suggested benefit but evidence on possible harms associated with the interventions was limited. In addition the range of interventions in the reviewed studies varied considerably and they were carefully designed to address specific behaviour that challenges. The GDG agreed that these interventions could be of real value. In addition the GDG was also aware of the potential benefits of medication in the short-term management of severe behaviour that challenges that might present an immediate risk to a person or others involved in their care. The GDG also had concerns that reactive strategies could be misused or delivered badly with potentially harmful effects. Taking these factors into account the GDG therefore decided to set out a series of key principles to guide the use of reactive strategies for the management of behaviour that challenges, including using the least restrictive and safest methods, having a basis in sound ethical and legislative practice and the need for regular review and reduction in the reactive intervention as soon as is feasible.

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14 References

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