Appendix S: Evidence tables of economic evaluations

A.1 Interventions aimed at the prevention of behaviour that challenges in people with learning disabilities

A.1.1 Psychosocial interventions for adaptive behaviour

A.1.1.1 References to included studies

1. Chasson GS, Harris G, Harris GE. Cost comparison of early intensive behavioral intervention and special education for children with autism. Journal of Child and Family Studies 2007; 16(3): 401-413
2. Jacobson JW, Mulick JA, Green J. Cost-benefit estimates for early intensive behavioral intervention for young children with autism - General model and single state case. Behavioral Interventions 1998; 13(4): 201-226. EXCLUDED
3. Motiwala SS, Gupta S, Lilly MB, Ungar WJ, Coyte PC. The Cost-Effectiveness of Expanding Intensive Behavioural Intervention to All Autistic Children in Ontario. Healthcare Policy 2006; 1(2):135-151.
4. Peters-Scheffer N, Didden R, Korzilius H, Matson J. Cost comparison of early intensive behavioral intervention and treatment as usual for children with autism spectrum disorder in the Netherlands. Research in Developmental Disabilities 2012; 33(6): 1763-1772.

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Chasson et al., 2007 US Cost analysis	Interventions: Early Intensive Behavioural Intervention for 3 years (EIBI) Standard educational service for children with autism, comprising special education for 18 years	Children with autism, aged 4 years at the start of analysis Economic modelling Source of resource use and unit costs: state estimates (Texas) based on assumptions and personal communication Source of clinical effectiveness data (proportion of children receiving EIBI who improve and do not require special education): estimates based on published literature	Costs: EIBI, special education (state-budgeted, local, federal, and private); regular education costs omitted since common in both arms (baseline, standard costs) Mean cost per child: EIBI: $151,500 Standard educational service: $360,000 Cost difference per child: -$208,500	N/A	Perspective: public (state, local, federal) & private – confined to intervention costs Currency: US$ Cost year: probably 2004 Time horizon: 18 years Discounting: not applied Applicability: partially applicable Quality: potentially serious limitations

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Jacobson et al., 1998 US Cost analysis	Interventions: Early Intensive Behavioural Intervention (EIBI) for children with autism No intervention	Children with autism or pervasive developmental disorder (PDD), aged 3 years at the start of analysis Economic modelling Source of resource use and unit costs: state estimates (Pennsylvania) based on published literature Source of clinical effectiveness data (effectiveness of EIBI): estimates based on assumptions – different values tested to estimate financial benefits	Costs: EIBI, regular, special and intensive special education, , family support services, supplemental security income/aid to dependent children (SSI/ADC), adult developmental disability services, adult home- and community based services, intensive adult community services, adult institutional services, supported work services, supported wages Total net cost of EIBI per person (from 3 to 55 years): For effectiveness of EIBI 20% (normal functioning) -$ 656,385 For effectiveness of EIBI 30% (normal functioning) -$798,251 For effectiveness of EIBI 40% (normal functioning) -$940,118 For effectiveness of EIBI 50% (normal functioning) -$1,081,984	NA	Perspective: societal (public & wages) Currency: US$ Cost year: 1996 Time horizon: 52 years Discounting: possibly 3%, except SSI/ADC which was discounted at 1,5% Applicability: partially applicable Quality: very serious limitations (no intervention implicitly assumed to lead to zero levels of normal functioning)

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Motiwala et al., 2006 Canada Cost effectiveness analysis	Interventions: Expansion of 3 years of Early Intensive Behavioural Intervention to all eligible children (EIBI) Standard service, including 3 years of EIBI (37% of eligible children) and no intervention (63% of eligible children) No intervention	Children with autism, aged 2-5 years Economic modelling Source of resource use and unit costs: provincial government data (Ontario, Canada) Source of clinical effectiveness data (proportion of children with normal functioning, semi-dependent and very dependent): published literature and further assumptions	Costs: EIBI cost (training costs of therapists; contractual payments to service providers; salaries, benefits & overheads incurred by provincial civil servants), educational and respite services, adult day programmes, accommodation, supported employment Mean total cost per person: EIBI: $960,595 Standard service: $995,074 No intervention: $1,014,315 Primary measure of outcome: number of dependency-free years per person Number of dependency-free years per person: EIBI: 14.0 Standard service: 11.2 No intervention: 9.6	EIBI dominant over standard service and no intervention Standard service dominant over no intervention Results sensitive to EIBI efficacy and discount rate	Perspective: public (provincial government in Canada) Currency: Canadian$ Cost year: 2003 Time horizon: up to 65 years of age Discounting:3% Applicability: partially applicable Quality: potentially serious limitations

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Peters-Scheffer et al., 2012 Netherlands Cost analysis	Interventions: Early Intensive Behavioural Intervention (EIBI) plus treatment as usual (TAU) TAU alone	Children with autism of preschool age Economic modelling Source of resource use and unit costs: national data and assumptions Source of clinical effectiveness data (proportion of children with normal functioning, semi-dependent and very dependent): review of published meta-analyses – selection of data based on their applicability to the Dutch setting / naïve addition of data across treatment arms and further assumptions	Costs: EIBI (personnel, capital assets, transportation, materials and supplies), educational services, speech therapy & physiotherapy, daytime activities and care, social benefits for parents, payments for future adult living expenses, day programs or supported work, sheltered environment services Mean total cost per child: EIBI: €2,578,746 TAU: €3,681,813 Difference: -€1,103,067	EIBI less costly than TAU Using more optimistic data for TAU: cost difference: -€250,761	Perspective: public services Currency: Euros (€) Cost year: likely 2011 Time horizon: up to 65 years of age Discounting: not undertaken Applicability: partially applicable Quality: potentially serious limitations

A.1.2 Health awareness interventions

A.1.2.1 Reference to included study

Romeo R, Knapp M, Morrison J, Melville C, Allan L, Finlayson J, Cooper SA (2009) Cost estimation of a health-check intervention for adults with intellectual disabilities in the UK. Journal of Intellectual Disability Research, 53(5), 426-39.

AND

Cooper SA, Morrison J, Melville C, Finlayson J, Allan L, Martin G, Robinson N (2006) Improving the health of people with intellectual disabilities: outcomes of a health screening programme after 1 year. Journal of Intellectual Disability Research, 50(Pt 9), 667-77.

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Romeo et al., 2009B UK Cost consequenceanalysis	Interventions: Health-check intervention (HCI) comprising 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; referral algorithms to intellectual disabilities services Standard care (SC)	Adults with learning disability registered with primary care services Cohort study with matched controls Source of effectiveness & resource use data: cohort study with matched controls (Cooper et al., 2006; N=100) Source of unit costs: national sources & further estimates	Costs: intervention (equipment & staff time), primary, inpatient, outpatient & specialist intellectual disability services, other healthcare services, daytime activities (unsupported & supported paid employment, voluntary work, adult education classes, day centres and additional support), respite care, aids and adaptations, paid and unpaid care. Total cost of intervention per person: £82 Total mean service cost (SD) per person: HCI: £9,412 (£6,899); SC: £10,091 (£7,775) Bootstrapped cost difference: -£679 (95%CI -£3,429 to £2,292) Total mean carer support cost (SD) per person: HCI: £40,673 (£27,978); SC: £62,766 (£44,320) Bootstrapped cost difference: -£22,093 (95%CI -£35,394 to -£7,571) Total cost (SD) per person: HCI: £50,085 (£30,824); SC: £72,857 (£48,679) Bootstrapped cost difference: -£22,772 (95%CI -£37,569 to -£6,400) Measures of outcome: levels of health need detection, met new health needs, met health promotion and monitoring needs Mean number of new health needs per person:	HCI was dominant (better outcomes at lower cost)	Perspective: societal (services and care support) Currency: GB£ Cost year: 2003 Time horizon:12 months Discounting: not needed Participants matched with controls for age, gender and level of learning disability Costs collected prospectively for intervention group and retrospectively for control group Applicability: directly applicable Quality: potentially serious limitations

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
			HCI 4.80, SC 2.26, p<0.001 Mean number of met new health needs per person: HCI 3.56, SC 2.26, p<0.001 Level of met health promotion needs & health monitoring needs greater for HCI (p< 0.001 and p=0.039, respectively)		

A.2 Interventions aimed at reducing and managing behaviour that challenges in people with learning disabilities

A.2.1 Psychosocial interventions aimed at reducing and managing behaviour that challenges in people with learning disabilities

A.2.1.1 References to included studies

1. Hassiotis A, Robotham D, Canagasabey A, Romeo R, Langridge D, Blizard R, Murad S, King M (2009) Randomized, single-blind, controlled trial of a specialist behavior therapy team for challenging behavior in adults with intellectual disabilities. American Journal of Psychiatry 166:1278-1285.

AND

Hassiotis A, Canagasabey A, Robotham D, Marston L, Romeo R, King M (2011) Applied behaviour analysis and standard treatment in intellectual disability: 2-Year outcomes. British Journal of Psychiatry 198:490-491.

2. Felce D, Cohen D, Willner P, Rose J, Kroese B, Rose N, Shead J, Jahoda A, Macmahon P, Lammie C, Stimpson A, Woodgate C, Gillespie D, Townson J, Nuttall J, Hood K (2014) Cognitive behavioural anger management intervention for people with intellectual disabilities: costs of intervention and impact on health and social care resource use. Journal of Intellectual Disability Research doi: 10.1111/jir.12112

AND

Willner P, Rose J, Jahoda A, Stenfert Kroese B, Felce D, MacMahon P, Stimpson A, Rose N, Gillespie D, Shead J, Lammie C, Woodgate C, Townson JK, Nuttall J, Cohen D, Hood K (2013) A cluster randomised controlled trial of a manualised cognitive-behavioural anger management intervention delivered by supervised lay therapists to people with intellectual disabilities. Health Technology Assessment 17(21).

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Hassiotis et al., 2009 & 2011 UK Cost effectiveness analysis	Interventions: Specialist behaviour therapy plus treatment as usual (SBT+TAU) TAU alone, comprising community intellectual disabilities teams; each team includes psychiatrists, community nurses, occupational therapists, speech & language therapists, physiotherapists and generic psychologists. Teams offer a range of interventions including pharmacotherapy, nursing and enhancement of adaptive skills	Adults with any severity of intellectual disability and challenging behaviour RCT [Hassiotis 2009] Source of clinical effectiveness data: RCT (N=63 for 6 months; 58 for 2-year follow-up) Source of resource use data: RCT (N=63 for 6 months; 58 for 2-year follow-up) Source of unit costs: national unit costs	Costs: Intervention (SBT and TAU), non-psychiatric inpatient stays, outpatient appointments and day care, leisure activities, adult education, support for voluntary work, contact with GPs, social workers, community nurses & advocates. Costs over 0-6 months: Total cost (SD) per person: SBT + TAU: £1,415 (£1,349) TAU: £3,615 (£8,239) Cost difference after adjusting for baseline age, gender, level of learning disability, psychotic disorder, affective disorder, pervasive developmental disorder & total ABC score: –£2,900 (95% CI –£6,788 to £987) Costs over 18-24 months (non-psychiatric inpatient services excluded): Total cost (SD) per person: SBT + TAU: £5,419 (£7,660) TAU: £4,271 (£7,612) Cost difference after adjustment: -£815 (95% CI -£5,629 to £3,986) Primary measure of outcome: challenging behaviour measured by total and subscale scores on the Aberrant Behavior Checklist (ABC)	SBT+TAU more effective in the primary outcome at no additional cost	Perspective: NHS & PSS Currency: GB£ Cost year: likely 2007 Time horizon:6 & 24 months; costs reported for 2 time periods: 0-6 months & 18-24 months Discounting: not applied Costs and outcomes measured over different periods of time Applicability: directly applicable Quality: potentially serious limitations

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
			Difference in the transformed total ABC score: 6 months: -0.89 (95% CI= -1.74 to -0.04) 24 months: -0.88 (95% CI = -1.66 to -0.11)		

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Felce et al., 2014 UK Cost consequence analysis	Interventions: Manualised group CBT anger management intervention, delivered by day service staff over 12 weeks (CBT) Treatment as usual waiting list (WL)	Adults with minor to moderate intellectual disability and problem anger RCT [Willner 2013] Source of clinical effectiveness data: Cluster RCT (N=143) Source of resource use data: RCT (n=133) – data collected from researchers, service users and home carers Source of unit costs: national unit costs,	Costs: 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 challenging behaviour, accommodation, domiciliary support or respite care Mean total cost per person per week (SD): CBT: £970 (£700); WL: £867 (£592) Adjusted mean difference: £-22 (95%CI -£192 to £147, p=0.795) Primary measure of outcome: Provocation Index (PI) as completed by service users, a measure of felt response to defined hypothetical situations that may provoke anger Secondary measures: PI completed by key workers; Profile of Anger Coping Skills (PACS), a measure of anger coping skills, completed by service users and key workers; PACS imaginal provocation test (PACS-IPT), a measure of response to actual anger-provoking situations completed by service users; aggressive	CBT better than WL in a number of secondary outcomes at no additional cost	Perspectives: NHS & PSS Currency: UK£ Cost year: 2010-11 Time horizon: 10 months; costs were measured over a 12-week period Discounting: not needed Applicability: partially applicable Quality: potentially serious limitations

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
		local costs for lay therapists	behaviour; mental health; self-esteem; quality of life Mean self-reported PI (SD) at 10 months: CBT: 41.4 (23.78); WL: 45.1 (17.46) Adjusted mean difference: -2.8 (95% CI -7.4 to 1.7) p=0.210 Key worker-reported PI, PACS, PACS-IPT: significantly improved for CBT; other secondary outcomes: not significantly different between CBT and WL		

A.2.2 Pharmacological interventions aimed at reducing and managing behaviour that challenges in people with learning disabilities

A.2.2.1 References to included study

Romeo R, Knapp M, Tyrer P, Crawford M, Oliver-Africano P (2009) The treatment of challenging behaviour in intellectual disabilities: cost-effectiveness analysis. Journal of Intellectual Disability Research 53:633-643.

AND

Tyrer P, Oliver-Africano PC, Ahmed Z, Bouras N, Cooray S, Deb S, Murphy D, Hare M, Meade M, Reece B, Kramo K, Bhaumik S, Harley D, Regan A, Thomas D, Rao B, North B, Eliahoo J, Karatela S, Soni A, Crawford M (2008) Risperidone, haloperidol, and placebo in the treatment of aggressive challenging behaviour in patients with intellectual disability: a randomised controlled trial. The Lancet 371, 57-63.

AND

Tyrer P, Oliver-Africano P, Romeo R, Knapp M, Dickens S, Bouras N, et al (2009) Neuroleptics in the treatment of aggressive challenging behaviour for people with intellectual disabilities: a randomised controlled trial (NACHBID). Health Technology Assessment 13(21)

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
Romeo et al., 2009A & Tyrer et al.., 2008 & 2009 UK Cost effectiveness analysis	Interventions: Risperidone 1mg/d (max 2mg/d) Haloperidol 2.5mg/d (max 5mg/d) Placebo	Adults with learning disability (IQ<75) and challenging behaviour and aggression RCT [Tyrer 2008] Source of clinical effectiveness data: RCT (N=86 randomised, n=56 for MOAS, n=60 for QOL-Q) Source of resource use data: RCT (N=58 for 6 months; no data collected from participants in Australia) Source of unit costs: national unit costs	Costs: medication, inpatient care, specialised accommodation, day activities, community-based activities, informal care. Total mean service cost (SD) per person: Risperidone: £15,518 (£13,084) Haloperidol: £13,753 (£13,316) Placebo: £15,010 (£9,115) Total mean cost (SD) per person, including informal care: Risperidone: £18,954 (£13,502) Haloperidol: £17,626 (£12,883) Placebo: £16,336 (£8,918) Measures of outcome: total Modified Overt Aggression Scale (MOAS) score; total quality of life (QOL-Q) Total mean MOAS score (SD) /person: Risperidone: 7.5 (9.9) Haloperidol: 3.9 (8.4) Placebo: 6.0 (8.1) Total mean QOL-Q score (SD) /person: Risperidone: 74.4 (11.7) Haloperidol: 69.7 (11.0)	Using total MOAS score: Placebo dominates risperidone Haloperidol vs. placebo: £614 /additional point change on the MOAS Probability of haloperidol being more cost-effective than placebo: ≈50% for WTP=0, ≈89% for WTP=£3000 /point improvement in MOAS Using total QOL-Q: Placebo dominates haloperidol Risperidone vs. placebo: £996 /point change on the QOL-Q Probability of risperidone being more cost-effective than placebo: ≈52% for any WTP for one point improvement in QOL-Q	Perspective: societal (services and informal care) Currency: GB£ Cost year: likely 2005/6 Time horizon:26 weeks Discounting: not needed Applicability: partially applicable Quality: potentially serious limitations

Study Country Study type	Intervention details	Study population Study design Data sources	Costs: description and values Outcomes: description and values	Results: Cost-effectiveness	Comments
			Placebo: 71.9 (12.9) Differences in costs & outcomes between interventions not statistically significant		