

Myalgic encephalomyelitis (or encephalopathy) / chronic fatigue syndrome: diagnosis and management

[A] Evidence reviews for information, education and support for people with ME/CFS and their families and carers

NICE guideline <number>

Evidence reviews underpinning recommendations and research recommendations in the NICE guideline

November 2020

Draft for Consultation

*These evidence reviews were developed
by the National Guideline Centre*

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Contents

Contents	4
1 Information, education and support for people with ME/CFS and their families and carers	5
1.1 Review question	5
1.1.1 Introduction	5
1.1.2 Summary of the protocol	5
1.1.3 Methods and process	5
1.1.4 Qualitative evidence	6
1.1.5 Summary of studies included in the qualitative evidence	7
1.1.6 Qualitative evidence synthesis	17
1.1.7 Economic evidence	38
2 The committee’s discussion and interpretation of the evidence	39
2.1 The quality of the evidence	39
2.2 Findings identified in the evidence synthesis	39
2.3 Cost effectiveness and resource use	48
Appendix A – Review protocols	49
Appendix B – Literature search strategies	57
B.1 Clinical search literature search strategy	57
Appendix C Qualitative evidence study selection	62
Appendix D – Qualitative evidence	63
Appendix E – GRADE-CERQual tables	93
Appendix F – Excluded studies	121
References	125

1 Information, education and support for 2 people with ME/CFS and their families 3 and carers

4 1.1 Review question

5 What information, education and support do people with ME/CFS and their families and
6 carers need?

7 1.1.1 Introduction

8 Current guidance acknowledges that a supportive and collaborative relationship between
9 health care practitioners and the person with a confirmed diagnosis of ME/CFS, together with
10 accurate information, facilitates effective management of the condition. The care and support
11 needs of people with ME/CFS varies from very little to continual care depending on severity
12 of illness, stage of diagnosis, and ability to self-manage.

13 There is currently no specific guidance on what should be included in or the content of any
14 information, the efficacy of different support approaches, or the educational needs of people
15 with ME/CFS, their families and carers. This review aims to explore and identify the
16 information, education and support needs of people with ME/CFS and their families and
17 carers.

18 1.1.2 Summary of the protocol

19 For full details see the review protocol in Appendix A.

20 Table 1: Characteristics of review question

Objective	To identify the information, education and support required by people with or who are suspected of having ME/CFS and their families and carers.
Population and setting	Adults, children and young people who are diagnosed with ME/CFS, or who are suspected of having ME/CFS by their primary clinician and their families and carers.
Context	Perceptions of people with or who are suspected of having ME/CFS of the information, education and support they require.
Review strategy	Synthesis of qualitative research, following a thematic analysis approach. Results presented in narrative and in table format with summary statements of main review findings. Quality of the evidence will be assessed by a GRADE CerQual approach for each review finding.

21

22 1.1.3 Methods and process

23 This evidence review was developed using the methods and process described in
24 [Developing NICE guidelines: the manual](#). Methods specific to this review question are
25 described in the review protocol in appendix A and the methods document.

26 Declarations of interest were recorded according to [NICE's conflicts of interest policy](#).

1 1.1.4 Qualitative evidence

2 1.1.4.1 Included studies

3 Fifteen qualitative studies were included in the review;^{15, 18, 22, 25, 32, 38, 43, 58, 61, 69, 87, 89, 116, 130, 131}
4 these are summarised in Table 2 below. Key findings from these studies are summarised in
5 Section 1.1.6.1 below. See also the study selection flow chart in Appendix C, study evidence
6 tables in Appendix D, and excluded studies lists in Appendix F.

7 Adults

8 Nine studies were identified on the information, education and support needs of adults with
9 ME/CFS and their families and carers. Of these, eight included adults with ME/CFS and one
10 study both adults with ME/CFS and their carers.

11 Children and young people

12 Six studies were identified on the information, education and support needs of children and
13 young people with ME/CFS and their families and carers. Of these, four studies included
14 children and young people with ME/CFS, one study children and young people with ME/CFS
15 and their families and carers, and one study the mothers of children and young people with
16 ME/CFS. Two of the studies excluded severely affected individuals. One study was
17 conducted exclusively with parent carers caring for children from 5 years old to adults (the
18 majority caring for adult daughters). The information reported in this study was considered to
19 contribute to emerging themes relevant to both strata of adults, and children and young
20 people.

21 In line with the review protocol the evidence relevant to adults is reported separately to
22 children and young people. The severity of ME/CFS was mixed or unclear. No evidence was
23 identified for social care professionals caring for people with ME/CFS.

24 As a large number of papers were identified for this review, inclusion was halted once data
25 saturation was reached. Data saturation is the point at which no new themes or data
26 contributing to themes emerged from the studies found to match the review protocol. These
27 studies are listed in Table 8.

28 Where 'CFS/ME' has been used in the evidence review, it is in order to reflect the
29 terminology used in the included studies.

30 1.1.4.2 Excluded studies

31 See the excluded studies in Appendix F.

32

1 1.1.5 Summary of studies included in the qualitative evidence

2 **Table 2: Summary of studies included in the review**

Study	Design	Population	Research aim	Comments
Bayliss 2016 ¹⁵	Semi-structured interviews and thematic analysis followed by theory-driven analysis.	<p>Patients (n=11), mean age (range): 46 (27 to 71) years; GPs (n=8)</p> <p>Patients were recruited from participating GP practices where GPs had been given access to an online 'CFS/ME' training module; that involved patient resource packs for use in consultation with new and existing 'CFS/ME' patients.</p> <p>UK</p> <p>Stratum: adults, severity mixed or unclear</p>	To explore the extent to which 'CFS/ME' training and resources can be implemented in routine primary care, leading to a better understanding of the barriers and facilitators to the adoption and integration of new practices associated with medically unexplained conditions.	<p>In line with the review protocol, themes emerging from the information provided by the patient population have only been extracted.</p> <p>Not all interviewed GPs had fully engaged in the training or research: 6/8 GPs interviewed had participated in the training, although not all had completed the online test and downloaded their completion certificate.</p> <p>ME/CFS diagnosis: Searches of GP practice databases were conducted by the research team to identify individuals with an existing diagnosis of 'CFS/ME'. GPs were asked to review these lists and to exclude patients with other conditions, or other factors that may account for their fatigue.</p>
Beasant 2014 ¹⁸	In-depth semi-structured face-to-face interviews and thematic analysis.	N=12 adolescents; male/female 3/9; age mean (SD) 13.9 (1.6)	To understand the experiences of adolescents and families	Specialist Medical Intervention and Lightning Evaluation (SMILE) study designed to test

Study	Design	Population	Research aim	Comments
		<p>years; illness duration median (IQR) 13 (9 to 18) months; mildly or moderately affected by ME/CFS, 5 were interviewed post randomisation but before receiving the SMILE study intervention, and 7 after the intervention.</p> <p>N=13 mothers; 5 mothers were interviewed at all three time points, 8 took part in one-off interviews: 4 post randomisation and 4 after their child received an intervention.</p> <p>UK</p> <p>Stratum: children and young people, mild/moderate severity</p>	<p>in accessing and using a specialist service and to explore whether or not they value referral to a specialist service for young people with 'CFS/ME'.</p>	<p>the feasibility and acceptability of recruiting adolescents to a randomised controlled trial (RCT) comparing specialist medical care with specialist medical care and the Lightning process.</p>
Brigden 2018 ²²	<p>In-depth semi-structured interviews (face-to-face or via Skype) and thematic analysis.</p>	<p>Adolescents recruited from a specialist paediatric 'CFS/ME' service.</p> <p>N=9; male/female: 3/6; mean age (SD): 14.89 (1.9) years, at different</p>	<p>To gather the views of adolescents with 'CFS/ME' to explore what they access online for information and support, and how this influences the way they cope with the condition.</p>	<p>Inclusion criteria: a diagnosis of 'CFS/ME' (NICE CG53 criteria), age 12-17 years and self-identified as having used the internet for 'CFS/ME'. Exclusion criteria: insufficient proficiency in English to</p>

Study	Design	Population	Research aim	Comments
		<p>stages of the condition; mean number of months from initial assessment to interview (SD): 12.98 (7.98), range 4 to 25) months.</p> <p>UK</p> <p>Stratum: children and young people, severity mixed or unclear</p>		<p>participate in an interview or severely affected.</p>
<p>Broughton 2017²⁵</p>	<p>Semi-structured interviews (six face-to-face, 10 via telephone) and thematic analysis.</p> <p>Cross-sectional design using opportunity sampling.</p>	<p>Adults who were completing treatment for ME/CFS at one of three outpatient NHS specialist 'CFS/ME' services.</p> <p>N=16; 87.5% female, 12.5% male. Median age of participants: 43 (range 24-62). Median self-reported duration of illness: 7.5 years (range 1-17). The sample was representative of patients treated by the 3 services during 2014 (median age 40, 81% female), except for longer duration of illness.</p>	<p>To explore the experiences of 'CFS/ME' patients who were completing programmes of treatment at three NHS specialist 'CFS/ME' services in England.</p>	<p>NHS specialist 'CFS/ME' services followed NICE guidelines for diagnosis and management of 'CFS/ME', offering patient centred programmes aiming to increase patients' physical, emotional and cognitive capabilities whilst managing the impact of symptoms. CBT and GET are the two main evidence-based therapies which (or components of which) are used in conjunction with techniques aimed at managing activity, sleep hygiene and relaxation. Patients also receive practical support around employment and the benefits system. Services shared a philosophy</p>

Study	Design	Population	Research aim	Comments
		UK Stratum: adults, severity mixed or unclear		of rehabilitation aimed at 'recovery' or 'significant improvement', whilst acknowledging that this would not be attained by all patients.
Chew-Graham 2008 ³²	Semi-structured interviews and thematic analysis	Family physicians (n=14; mean age: 48, SD: 12 years) and patients (n=24; mean age: 48, SD: 12 years) participating in a RCT of 2 nurse-led interventions in primary care (FINE trial) UK Stratum: adults, severity mixed or unclear	To explore how patients with 'CFS/ME' and family physicians conceptualise and understand this condition and how their understanding might affect the primary care consultation.	FINE trial was a primary-care-based RCT examining self-help treatment and pragmatic rehabilitation for patients with ME/CFS. To be included in the trial, registered patients with 'CFS/ME' referred by physicians in 44 primary care trusts in North West England, had to fulfil the Oxford inclusion criteria for 'CFS/ME', score 70% or less on the SF-36 physical functioning scale and 4 or more on the 11-item Chalder fatigue scale. In line with the review protocol, themes emerging from the information provided by the patient population have only been extracted.
De Carvalho Leite ³⁸	Focus groups (n=6) and semi-structured interviews (n=35) and (data-led) thematic analysis.	Adults with 'CFS/ME' (n=35), purposively selected to include a diverse range of illness	To investigate the impact of 'CFS/ME' on people from varied social background, including	Six of the 35 participants were purposively selected to include a diverse range of illness

Study	Design	Population	Research aim	Comments
		<p>severity, duration, social variation (age, gender, ethnic background and socio-economic conditions) and year of diagnosis.</p> <p>UK</p> <p>Stratum: adults, severity mixed or unclear</p>	<p>those from ethnic minorities, and what challenges may be posed to health care practitioners in providing appropriate and equitable care for this condition.</p>	<p>severity, for both an initial focus group discussion and a later one-to-one interview.</p> <p>The study was part of the National Observatory of people with 'CFS/ME' in England, which aims to produce and to facilitate epidemiological and social research, in response to the needs of these people so as to fill a major gap in the evidence of the occurrence and the impact of this disease.</p>
Devendorf 2018 ⁴³	<p>Mixed-methods design; qualitative analysis of participants' open-ended survey responses from a previous project that examined illness severity, stigma, physician interactions and depression.</p>	<p>Patients who self-identify as having ME/CFS and endorsed suicidal ideation (SI) but did not meet depression criteria</p> <p>N=29; 79.3% female, 20.7% male. Mean age: 51.48 years old. Mean score for the BDI-PC: 2.38; one participant endorsed active SI (i.e. score of 3), 28 participants endorsed passive SI (i.e. score of 1).</p> <p>USA</p>	<p>An exploratory study to explore the relationship between ME/CFS and suicidal ideations, including quality of life, loss of function, isolation and hopelessness.</p>	<p>The study was hosted online using Research Electronic Data Capture.</p>

Study	Design	Population	Research aim	Comments
		Stratum: adults, severity mixed or unclear		
Hannon 2012 ⁵⁸	Semi-structured interviews and grounded theory approach.	Health practitioners (GPs n=9, practice nurses n=5, 'CFS/ME' specialists n=4), Carers (n=10), patients (n=16), aged 28-71 UK Stratum: adults, severity mixed or unclear	To develop an education and training intervention to support practitioners in making an early diagnosis of 'CFS/ME' and supporting patients in the management of their symptoms.	For the purpose of this review only the information disclosed by patients and carers was extracted.
Harris 2017 ⁶¹	Semi-structured interviews and thematic analysis.	Adolescents (n=11); mean age (range): 15 (13-17 years); 8 female UK Stratum: children and young people, severity mixed or unclear (severe/very severe likely excluded)	To explore what adolescents felt had caused their problems with eating, whether they were triggers and maintaining factors and what interventions they felt would be helpful.	Adolescents were given the option to be interviewed alone or with a parent and seven chose to have their mothers present, all of whom contributed to the interview. Information disclosed by the parents relevant to the aim of this review has also been extracted. The sample was drawn from a 'CFS/ME' specialist hospital service providing regional support for assessment and treatment of over 300 children aged 5-19 years. Potential

Study	Design	Population	Research aim	Comments
				<p>participants were identified as being eligible by their clinician.</p> <p>Adolescents who were housebound i.e. potentially severe/ very severe cases were excluded.</p>
Jelbert 2010 ⁶⁹	Semi-structured interviews and thematic analysis (interpretative phenomenological analysis).	<p>Five adolescents who were considered to have recovered from ME/CFS.</p> <p>N=5; 4 female, 1 male. Mean age: 15.2 years (range 13-18 years). Only adolescents who had been discharged within the last year were included. All participants reported having experienced ME/CFS symptoms for a duration of between 1.5 and 2 years.</p> <p>UK</p> <p>Stratum: children and young people, severity mixed or unclear</p>	To gain an understanding of adolescents' illness experiences of ME/CFS, from its beginning to its end, to identify themes that have implications for clinical practice and raise further questions for formal investigation in quantitative studies.	Adolescents were chosen on the basis of having met diagnostic criteria for CFS as assessed by a consultant paediatrician in the paediatric outpatient clinic the study took place.

Study	Design	Population	Research aim	Comments
McDermott 2011 ⁸⁷	Semi-structured interviews and analysed using the constant comparative method	Patients (n=20) newly referred to a specialist 'CFS/ME' service by their GP; mean age (range): 39 (22-60) years. UK Stratum: adults, severity mixed or unclear	To explore the hopes and expectations of patients newly referred to a 'CFS/ME' Service in the South of England.	
Mihelicova 2016 ⁸⁹	Secondary analysis of parent-carer interview narratives using Interpretative phenomenological analysis (IPA) (i.e. thematic analysis)	Parent-carers (n=19) to children from 5-year old to adults; 12 mothers and 7 fathers, primarily caring for adult daughters. UK Stratum: age and severity mixed or unclear	To give voices to those who care for individuals with ME and are often stigmatised and inform future research to ensure that parent-carers of individuals with ME have adequate resources and support.	Interviews had been published in the book 'Lost Voices from a Hidden Illness' (Boulton, 2008); a collection of passages from individuals with ME, their significant others and carers that aims to raise awareness of the impact of ME and to allow individual voices to be heard.
Ryckeghem 2017 ¹¹⁶	Semi-structured interviews using open explorative thematic coding (thematic analysis).	A purposive sample of patients was selected through the department of General Internal Medicine at the University Hospital Ghent to achieve maximum variation. A convenience sample of GPs was recruited from different provinces in Belgium.	To explore the experiences and expectations of 'CFS' patients and GPs to develop the potential role of an advanced nurse practitioner (ANP) at the diagnostic care path of abnormal fatigue developed for regional transmurial implementation	A definitive diagnosis was established following a multi-disciplinary discussion in the diagnostic process. The views of patients are only extracted for this review.

Study	Design	Population	Research aim	Comments
		<p>Patients (n=15); median age (range): 45 (33-59 years); GPs (n=15); median age (range): 49 (31-62 years).</p> <p>Belgium</p> <p>Stratum: adults, severity mixed or unclear</p>	<p>in the Belgian provinces of East and West Flanders.</p>	
Taylor 2005 ¹³¹	<p>Focus group interviews, open-ended questionnaires, progress notes, and from a program evaluation questionnaire.</p>	<p>Adults with ME/CFS meeting the Fukuda criteria for CFS, who were participating in a research project aimed to evaluate a participant-designed rehabilitation program.</p> <p>N=47; 45 female, 2 male. Mean age: 46.9 years (SD 10.4). Seven participants were in full-time work, seven in part-time work and 33 were not working. Eight participants were minority ethnicity, 39 were non-minority. All participants met the CDC Fukuda <i>et al</i> (1994) criteria for ME/CFS.</p>	<p>To determine what aspects of the disability experience of persons with CFS are explained by the social model of disability, and what aspects of disability fall outside or contradict central tenets of the social model.</p>	<p>Data for this study emerged from a federally funded research project that developed and evaluated a participants-driven program for individuals with 'CFS', implemented at a centre of independent living.</p>

Study	Design	Population	Research aim	Comments
		USA Stratum: adults, severity mixed or unclear		
Taylor 2017 ¹³⁰	Semi-structured interviews (n=8 conducted at home; n=1 over the phone) and thematic analysis.	Young people (n=9) with a primary diagnosis of 'CFS/ME' and co-morbid low mood; 88.9% female; median age (range): 14 (14-15) years; median illness duration (range): 12 (8.5-37.5) months; median HADS depression score (range): 11.5 (10.5-12.5); median HADS anxiety score (range): 12 (9.5-14). Median Chalder fatigue scale score (range): 29 (25-31); mean disability/SF-36 physical function score (range): 45 (30-50) UK Stratum: children and young people, severity mixed or unclear	To explore the experiences of young people with 'CFS/ME' and depression in order to understand their views on why low mood developed, the impact of having low mood and what they had found to be helpful and unhelpful in treatment.	Diagnosis established by specialist paediatric service, after a thorough assessment which included screening for other disorders associated with fatigue (NICE 2007; RCPH, 2004). Co-morbid low mood was defined as a depression sub-scale score of >9 on HADS either at assessment or at subsequent treatment appointments with the specialist 'CFS/ME' service. 78% (7/9) had <40% school attendance i.e. 2 days or fewer per week.

1 See Appendix D for full evidence tables.

2 1.1.6 Qualitative evidence synthesis

3 1.1.6.1 Summary of the qualitative evidence

4 Table 3: Review findings for Adults with ME/CFS, their families or carers

Main findings	Statement of finding
Information needs	
Information about ME/CFS ^{25, 32, 43, 58, 87, 116, 132}	Patients and carers needed general information about ME/CFS for both themselves and others (families, friends, employers and practitioners), to enable them to develop accurate expectations about the future, relieve feelings of distress caused by the general lack of sufficient information both patients and carers experienced and educate others.
Type of information: online & evidence-based ^{15, 32, 58}	Patients and carers could benefit from information leaflets, information and resources that are evidence-based and available online and in DVD format, as well as from alternative sources such as media representations of people with ME/CFS.
Advice on symptom management & treatment options ^{32, 38, 43, 58, 87, 89, 131}	Patients and carers need guidance on symptom management (including self-help strategies and lifestyle advice) and effective and diverse treatment options to choose from, including biomedical and alternative treatments other than those currently offered by the health care system, lack of which can lead to financial costs and even suicidal thoughts.
Support needs	
Need for understanding & advocacy ^{15, 25, 38, 43, 58, 89, 116, 131}	Patients and carers needed understanding and recognition of ME/CFS from the medical community, employers, colleagues, family and friends, lack of which was unhelpful in the workplace, in accessing healthcare and social support and implicated their well-being and social relationships.
Support with acceptance ²⁵	Patients need support with acceptance of their new life circumstances, their diagnosis and the likely prognosis of their illness in order to maximise the benefit received from treatment.
Support during difficult phases of ME/CFS ^{25, 89, 116}	The time following diagnosis of ME/CFS and the early stages of treatment were difficult times for patients who experienced feelings of distress, difficulty with acceptance of diagnosis, of advice given and occasionally a lack of guidance while phases of relapse were particularly difficult for carers.
Support from specialists ^{15, 25, 87, 131}	Patients hoped to be referred to specialist services to overcome the barriers to diagnosis and treatment they encountered in primary care and those who ultimately were, had benefited in ways including diagnosis, validation and information provision.
Help accessing support ^{38, 43, 58, 131}	Patients and carers needed sign posting to relevant contacts that would help address the consequences of ME/CFS, including knowledgeable physicians, information on existing benefits and support they were entitled to and help accessing them.

Main findings	Statement of finding
Financial support ^{38, 43}	Financial support was crucial for the illness and life-management of ME/CFS and social relationships while having to work due to a lack of financial resources or financial constraints arising due to a lack of other forms of support had physical consequences and negatively influenced coping.
Ongoing health professional support ^{15, 25, 32, 116}	Patients identified the need to establish a close, ongoing relationship with their health professional who would be involved in the long-term management of ME/CFS and accompany and advise them at all stages of the care process.
Social support ^{25, 43, 58, 87}	Patients and carers felt isolated, struggling to maintain social relationships and could benefit from social interactions and mutual patient support.
Practical support with daily living & social care ^{38, 43}	Patients needed practical support for both themselves and their carers, including help with personal and domestic tasks and access to social care, lack of which implicated their ability to manage their lives and maintain their family roles.
Further needs	
Need for tailored and accessible hospital care ^{25, 38, 58}	Patients emphasised the importance of hospital care that would be tailored to their needs, the severity of their symptoms and other commitments, in terms of accessibility and flexibility in the frequency, the duration and the mode of attendance to medical appointments.
Need for a diagnosis ^{38, 87}	A diagnosis of ME/CFS- that was often gained through private or alternative health services- enabled patients to gain advice from health professionals and take action towards symptom improvement while a lack of a diagnosis exacerbated their psychological and financial pressures and impeded their access to social services.
Need for a positive diagnosis & future direction ^{15, 58, 87}	Patients reflected on the importance of a positive direction for the future and on the need for the ME/CFS diagnosis to be framed in a positive way to enable them to maintain hope for improvement.
Patient support during medical consultations ⁵⁸	GP consultations were challenging for patients who could benefit from the presence of their caregiver.
Advice (for carers) on how to support patients ⁵⁸	Carers need advice on how to support patients with ME/CFS as they lacked an understanding of the illness and discussed the danger of giving inappropriate advice or support to patients, exacerbating their symptoms.

1 See Appendix E for full GRADE-CERQual tables.

2 1.1.6.2 Narrative summary of review findings

3 1.1.6.2.1 Information education and support for adults with ME/CFS, their families or 4 carers

5 Review finding 1: Information about ME/CFS

6 Patients and carers felt they had a limited understanding of 'CFS/ME' and lacked general
7 information about the illness, having received little or conflicting information. They report not
8 having a sufficient overview of what to expect. This often led to false hopes and expectations
9 about the illness, treatment and prognosis and led to distress and worry. Moving away from
10 the idea of cure towards goals related to management of 'CFS/ME' was linked to obtaining

1 information and acquiring knowledge about the condition. Patients who expressed confidence
2 in their level of knowledge about 'CFS/ME' also tended to express less anxiety about the future
3 and less concern or distress about waiting for an appointment with specialist services.
4 Understanding the complexity of the illness, especially the interaction between different
5 factors, was perceived by many as a route out of this cycle of fatigue. Some patients also
6 expressed hopes that specialist services would be able to advise them on which factors were
7 relevant to the illness.

8 Patients frequently reported their families, friends, co-workers and employers also lacked an
9 understanding of ME/CFS which led to a lack of empathy, frustration and often implicated their
10 relationship with patients who often felt obligated to educate them. To bypass limited clinical
11 knowledge and actively engage in consultations, patients reported having to seek additional
12 sources of evidence about 'CFS/ME' to bring to physicians.

13 Explanation of quality assessment: Moderate concerns over methodological limitations with
14 moderate concerns in two studies (due to concerns over the appropriateness of the data
15 collection method of one study that was a follow-up to a quantitative study with open-ended
16 online responses and due to the potential influence of the researcher on the findings not being
17 discussed and lack of transparency on the analysis process in the other study), minor concerns
18 over two studies (due to the potential influence of the researcher on the findings not being
19 discussed in one study and concerns over data analysis due to data richness with findings
20 mostly supported by single quotes in both studies), very minor concern in one study (due to
21 the potential influence of the researcher on the findings not being discussed) and no concerns
22 over two studies; no concerns about coherence with consistent information emerging across
23 studies; minor concerns about relevance due participants in one study being self-identified as
24 having ME/CFS rather than having been diagnosed according to accepted criteria but no
25 similar concerns in the other six contributing studies and participants of one study consisting
26 of people recruited in a RCT; no concerns about adequacy, the finding being supported by
27 sufficient information across the contributing studies. Overall assessment of confidence was
28 moderate due the methodological limitations and concerns about relevance identified in the
29 contributing studies.

30 **Review finding 2: Type of information: online & evidence-based**

31 Patients and carers in one study stated that a brief leaflet outlining the symptoms and the
32 evidence for management options would be useful at diagnosis. However, most patients
33 suggested that information resources should be made available online and therefore
34 accessible to all and suggested websites (such as NHS Choices) that they believed would be
35 useful for the resources to be linked to in order to be easily accessible. Patients welcomed
36 evidence-based resources of information as there are currently issues with identifying reliable
37 information on the internet and evidence-based information resources were reported to have
38 an impact on the friends, family and colleagues of the patients, improving relationships and
39 strengthening support networks.

40 DVD case studies were seen as particularly important in helping patients and carers to
41 understand that others shared their experiences, and their format allowed those who found it
42 difficult to read to access the information. As a result of this information some patients felt that
43 they needed to visit their practice less frequently.

44 Nevertheless, some patients were concerned that by placing the resources online, GPs would
45 be let off managing the condition in primary care and reported that they wished to bring
46 information from the internet to the consultation in order to gain a diagnosis from a health
47 professional or family physician. Self-help groups and web sites were seen to provide
48 important sources of evidence that patients could take to their family physician.

49 Alternative sources of evidence that were seen as helpful included representations of
50 'CFS/ME' expressed within the media, as media personalities lent credibility to the condition
51 and their positive attributes relieved patients from being culpable.

1

2 Explanation of quality assessment: Minor concerns over methodological limitations with minor
3 concerns in two studies (due to the potential impact of the researcher on the findings not being
4 discussed in one study and due to concerns over data analysis due to data richness with
5 themes mostly supported by single quotes in both studies) and very minor concerns in the
6 other study (due the potential influence of the researcher not being discussed); moderate
7 concerns about coherence due to differences both between studies and within studies in
8 regards to the types of information patients preferred, with some patients and carers reporting
9 on the usefulness of leaflets, others reporting on the usefulness of information available online,
10 with some worrying online information would negatively impact the management of ME/CFS
11 in primary care and others talking about alternative evidence sources from the media; very
12 minor concerns about relevance (due to participants in one study consisting of people recruited
13 in a RCT) that were too minor to lower the confidence rating; no concerns about adequacy.
14 Overall assessment of confidence was moderate due to the methodological concerns and
15 concerns around coherence.

16 **Review finding 3: Advice on symptom management & treatment options**

17 ME/CFS patients and carers reported experiences characterised by a lack of treatment
18 planning, treatment recommendations and advice on symptom management or even having
19 received inappropriate treatment recommendations. They sought advice on effective self-
20 help strategies, reporting dilemma about whether to push themselves to continue activities or
21 whether to rest or take time off, and advice on diet, the effects of stress on symptoms, how to
22 best cope with continued employment or return to work.

23 Due to a lack of knowledge in the medical community and the lack of support they experienced
24 from the health care system, patients and carers described how, having been left to find their
25 own information, they encountered difficulty finding appropriate treatment, with some seeking
26 help from private professionals or paying for alternative therapies. Those desperate for relief
27 of feelings of pain or illness reported finding treatments such as massage, osteopathy, dietary
28 advice and acupuncture helpful, and it caused ongoing frustration that such interventions were
29 not funded by either the NHS or by a private health insurance for 'CFS/ME'.

30 Patients felt that the health-care system should explore useful interventions and suffered
31 from a lack of control over choices of treatment for managing their illness, which they saw as
32 due to both a lack of resources in the National Health and social systems and the relative
33 lack of recognition or value given to their own experience with illness. They criticized the
34 continued focus on cognitive-behavioural and graded exercise therapies which attribute the
35 illness to psychological causes and called for more physiological research to develop
36 effective biomedical treatments and destigmatize the field. Some patients retained for a new
37 treatment as a rational to continue to live while others experienced moments where the
38 unbearable physical pain of the illness led them to view suicide as a logical escape.

39 Explanation of quality assessment: Moderate concerns over methodological limitations with
40 serious concerns over one study (related to the use of public data where a lack of detail on
41 the methods of participant recruitment and primary data collection implicating our ability to
42 assess bias), moderate concerns over one study (due to concerns over the appropriateness
43 of the data collection method, the study being a follow-up to a quantitative study with open-
44 ended online responses) and minor and very minor concerns in the majority of studies (minor
45 concerns in two studies due to concerns over data analysis due to data richness with findings
46 mostly supported by single quotes in two studies and the potential influence of the
47 researcher not being discussed in one study, and very minor concerns in two studies due to
48 the influence of the researcher not being discussed) and no concerns over one study; no
49 concerns about coherence; very minor concerns over relevance with concerns over
50 applicability of one study where participants were self-identified as having ME/CFS and had
51 not been diagnosed according to accepted criteria and one study where parent carers of
52 ME/CFS patients were not limited to people caring for adults, being counterbalanced by the

1 fact that the same information emerged from studies with no such concerns, one of which
2 included patients from diverse social and ethnic backgrounds, degrees of illness severity and
3 duration; no concerns over adequacy. Overall assessment of confidence was moderate due
4 to the methodological limitations and minor concerns over relevance.

5 **Review finding 4: Need for understanding & advocacy**

6

7 When reflecting on the role of their GP, ME/CFS patients wanted to be believed. Both
8 patients and carers described their frustration when GPs or practice nurses did not recognise
9 the seriousness of their symptoms or questioned the legitimacy of the condition and those
10 who felt believed GPs described how important this was to their wellbeing. They revealed
11 how facing barriers in gaining recognition of their illness from health professionals both
12 delayed or reduced access to support and greatly exacerbated emotional pressures. Even
13 when bed-bound, patients encountered unsupportive attitudes from health professionals
14 which greatly undermined their chances of wider belief and support. Patients highlighted how
15 lack of access to social care and practical support was exacerbated when health
16 practitioners would not recognise their illness, making a profound impact on their ability to
17 carry out their family caregiving roles. Many encountered doctors that were trained to view
18 ME/CFS as psychiatric, which was dismaying and caused disappointment when doctors
19 made exercises recommendations and psychological attributions or inferences

20 Patients that were referred to specialist services valued being taken seriously and reported
21 positive experiences where GPs had been supportive, while patients reporting less positive
22 experiences described barriers in accessing specialist services including having to take a
23 proactive role in asking for diagnostic tests due to a lack of their GPs' belief in ME/CFS.
24

25 Patients also faced incredulity and resentment in the workplace, which made them reluctant to
26 report their disability status and request accommodations and tended to push themselves to
27 avoid negative reactions, having to leave their jobs or accepting less-demanding or less-
28 sophisticated work. Because of the constant disbelief they were extremely ambivalent about
29 the process of getting others to make allowances for them, setting limits on asking for help.
30 Some patients had felt the need to 'hide' symptoms of 'CFS/ME', particularly from their
31 employer, for example using annual leave to manage symptom flare up for a number of years.
32

33 Patients reported that family members, friends and co-workers viewed them as individuals
34 who had no obvious manifestation of symptoms and impairment. This combined with a lack
35 of medical validation meant that family members, friends and co-workers, tended to feel
36 confused, and were prone to question whether the symptoms and impairments were real.
37 Negative responses and a lack of understanding from those not affected by ME/CFS were
38 distressing, created tension and often threatened relationships of both patients and parent
39 carers who felt isolated and hoped for change in the way ME/CFS is received.

40 Explanation of quality assessment: Moderate concerns over methodological limitations with
41 serious concerns over one study (related to the use of public data, where lack of detail over
42 participant recruitment and the primary data collection method implicated our ability to
43 assess bias) and moderate concerns over two studies (due to the potential influence of the
44 researcher on the findings not being discussed and lack of transparency on the analysis
45 process in one study and due to concerns over the appropriateness of the data collection
46 method of one study that was a follow-up of a quantitative study with data emerging from
47 open-ended online survey answers) but minor concerns in one study (due to the influence of
48 the researcher not being discussed and concerns about data analysis due to data richness
49 with findings mostly supported by single quotes), very minor concerns in two studies (due to
50 the potential influence of the researcher on the findings not being discussed) and no
51 concerns over one study; no concerns about coherence; very minor concerns over relevance
52 associated with two studies, due to participants in one study being self-identified as having
53 ME/CFS and parent carers of ME/CFS patients in the other study not being limited to people

1 caring for adults, being counterbalanced by the fact that the same information emerged from
2 studies with no such concerns that included patients from diverse social and ethnic
3 backgrounds, degrees of illness severity and duration; no concerns about adequacy. Overall
4 assessment of confidence was moderate due to the methodological concerns identified.

5 **Review finding 5: Support with Acceptance**

6 Time appeared to influence acceptance of the diagnosis, with some participants recalling a
7 gradual acceptance that treatment might not be curative. The importance of acceptance in
8 obtaining the most benefit from treatment was highlighted and participants discussed a need
9 to accept changes to their lives as a result of developing ME/CFS, and reflected upon what
10 they had lost or relinquished, including social networks, employment, career and study
11 aspirations and independence. When discussing personal responses patients viewed as key
12 to overcoming challenging periods during treatment, they highlighted being open, positive,
13 proactive, willing to try anything, being able to take a leap of faith and having perseverance.
14 They particularly highlighted the importance of being 'willing to change' and being prepared
15 to say goodbye to their old life completely in order to engage fully with treatment.

16 Explanation of quality assessment: No concerns over methodological limitations identified in
17 the contributing study; minor concerns about coherence with the theme not clearly emerging
18 from the data as the importance of acceptance and willingness to change were highlighted
19 but not explicitly identified as areas patients need support in; no concerns over relevance;
20 minor concerns over adequacy with sufficient information on the topic available from one
21 study. Overall assessment of confidence was moderate due to concerns about coherence
22 and adequacy.

23 **Review finding 6: Support during difficult phases of ME/CFS**

24 Diagnosis was a difficult time for patients who recalled feeling angry, distressed, frustrated
25 and fearful and that the diagnosis represented a life sentence. Accepting the diagnosis was
26 difficult because of patients' own negative preconceptions about ME/CFS and the reactions
27 of others. A need for intensive support and follow-up after completion of the diagnostic care
28 path emerged, with many participants having experience the feeling of being left in the dark
29 afterwards for example having to search themselves for caregivers in their neighbourhood.
30 Some patients found initial stages of treatment difficult and explained how during early
31 stages of treatment, advice given by clinicians felt counter-intuitive, and was a departure
32 from the way that symptoms and 'boom and bust cycles' had been self-managed prior to
33 accessing services.

34 Parent carers reported feelings of uncertainty regarding diagnosis and prognosis following
35 phases of remission and relapse and experienced difficulty in seeing their son/daughter
36 relapse, particularly when there has been improvement prior to relapse, which offered hope
37 that was then lost and exacerbated the anxieties of living in uncertainty.

38 Explanation of quality assessment: moderate concerns over methodological limitations with
39 serious concerns in one study (associated with the use of public data where lack of detail on
40 the method of primary data collection and participant recruitment limited our ability to assess
41 the risk of bias), moderate concerns over one study (due to the potential influence of the
42 researcher on the findings not being discussed, concerns over data analysis due to a lack of
43 sufficient information on the data analysis process and concerns over data richness with
44 findings mostly supported by single quotes) and no concerns over the third contributing
45 study; minor concerns about coherence due to the finding emerging from three studies and
46 participants in different studies reporting difficulty or a need for support with different phases
47 of the illness: post diagnosis (two studies), relapse (one study); very minor concerns over
48 relevance due to participants in one study being carers of patients of various ages not limited
49 to adults that were considered too minor to lower the confidence rating as the theme also
50 emerged from two studies with no similar concerns; minor concerns over adequacy due to
51 issues with data richness in one study but sufficient information to support the theme overall.

- 1 Overall assessment of confidence was low due the concerns over methodological limitations,
- 2 coherence and adequacy.

3 **Review finding 7: Support from specialists**

4 Patients' experiences were characterised by absence of treatment planning and treatment
5 recommendations and there was continued and ongoing dissatisfaction with treatment when
6 it was administered by a physician that did not specialise in 'CFS', as patients encountered
7 misinformation, misdiagnosis and inappropriate treatment recommendations. They wanted
8 more access to specialist services, with some recognising that GPs didn't have the time to
9 manage their condition.

10 Patients that had been referred to specialist services reported that their unsuccessful
11 attempts to resolve symptoms using strategies available in primary care led to their referral to
12 specialist services and they recalled having had hopes and expectations of referral and
13 treatment including confirming diagnosis and managing symptoms better. Patients
14 completing treatment at NHS specialist services felt they had benefited and reported that
15 they had their diagnosis confirmed when they were assessed by the specialist services,
16 which provided information and explanation of 'CFS/ME', simultaneously validating and
17 normalising their experiences and symptoms. All participants felt they had benefited from
18 accessing specialist service.

19 Explanation of quality assessment: very minor concerns over methodological limitations with
20 very minor concerns over two studies (due to the potential influence of the researcher on the
21 findings not being discussed) and no further concerns in the other two contributing studies;
22 no concerns about coherence with the information reported being consistent across studies;
23 no concerns over relevance; no concerns over adequacy. Overall assessment of confidence
24 was high as the methodological concerns identified were too minor to lower the confidence
25 rating.

26 **Review Finding 8: Help accessing support**

27 Patients had difficulty accessing helpful health-care providers and many encountered disdain,
28 disbelief and lack of knowledge. They described long and frustrating histories of their attempts
29 to access necessary information and services to help them address the consequences of their
30 impairment while finding helpful physicians that could provide a diagnosis and appropriate
31 services often took years of shuffling through various doctors. Both patients and carers
32 highlighted the need for sign posting from the GP, information on local support groups, advice
33 on benefits and referrals to the third sector, reporting that most GPs and practice nurses did
34 not have details of relevant contacts. Applying for welfare benefits while affected by 'CFS/ME'
35 was also difficult for patients as the nature of their symptoms meant that complicated forms
36 having to be completed to start the process were especially daunting. Patients frequently found
37 the benefits system complicated and confusing, something to 'fight' rather than a source of
38 support. Not being able to predict what they were entitled to, people found out in a hit-and miss
39 way, making arduous applications for benefits that were often refused. Many participants,
40 especially from non-White groups, expressed their needs for much more information on
41 entitlement to help focus their applications on attainable benefits. Unable to claim any benefits
42 whether through lack of support from benefits staff or their current financial situation, patients
43 were often bereft of resources, and enforced total dependence on family or partner. Problems
44 acquiring disability income, concerns about requesting workplace accommodation and
45 difficulties accessing community-based resources (such as meal-delivery programs and
46 specialised transportation options) were also reported. This was because patients had difficulty
47 convincing their physicians of the need for such resources, because they were unaware of
48 these resources or because their health care professionals lacked knowledge of how and why
49 they might benefit from such resources. Where benefits were successfully secured, these were
50 always perceived as helpful. For people beginning to recover and wanting to increase their
51 activities gradually to include limited part time work, income support was seen as too inflexible
52 to allow this. The benefit stopped if people started work, even when they could not work

1 sufficient hours to earn enough money to support themselves. To obtain essential benefits they
2 needed to represent themselves as very impaired, yet in attempting to move back into
3 employment or education, they had to represent themselves as minimally affected.

4 Explanation of quality assessment: Minor concerns over methodological limitations with
5 moderate concerns in one study (due to concerns over the appropriateness of the data
6 collection method, the study being a follow-up to a quantitative study with open-ended online
7 responses), but minor concerns in one study (due to the potential influence of the researcher
8 on the findings not being discussed and concerns over data analysis due to data richness with
9 findings mostly supported by single quotes), very minor concerns in another study (due to the
10 potential influence of the researcher on the findings not being discussed) and no concerns in
11 the fourth contributing study; no concerns about coherence, the theme clearly emerging from
12 four studies; very minor concerns over relevance due to concerns identified in one study (due
13 to participants being self-identified as having ME/CFS) but no concerns over the
14 representativeness of the sample in any of the other contributing studies; no concerns over
15 adequacy. Overall assessment of confidence was moderate due to the methodological
16 limitations.

17 **Review Finding 9: Financial support**

18 Financial support was identified as crucial for illness-and life-management, and to maintain
19 education and social relationships. Lacking financial resources made it difficult for patients to
20 cope with ME/CFS, since they had to exert energy to work causing physical consequences.
21 Patients described many financial constrains arising in the absence of other forms of support
22 to live with ME/CFS and there were primary consequences of impoverishment and secondary
23 consequences for social standing, relationships and future entitlements, while limited incomes
24 imposed hard choices about what money would be spent on which depts.

25 Explanation of quality assessment: Moderate concerns over methodological limitations with
26 moderate concerns in one study (due to concerns over the appropriateness of the data
27 collection method, the study being a follow-up to a quantitative study with open-ended online
28 responses) and very minor concerns in the other study (due to the potential impact of the
29 researcher on the findings not being discussed); no concerns about coherence with nothing to
30 lower the confidence rating; minor concerns over relevance associated with one of the studies
31 (due to participants being self-identified as having ME/CFS) but the theme also emerging from
32 a study with no similar concerns (reported by patients from diverse social and ethnic
33 backgrounds, degrees of illness severity and duration); minor concerns over adequacy with
34 limited information supporting the finding in one study. Overall assessment of confidence was
35 low due to concerns over methodological limitations, relevance and adequacy.

36 **Review finding 10: Ongoing health-professional support**

37 Patients wanted their GP to be accessible and actively involved in the longer-term
38 management of their condition and reported disengaging from primary care where support was
39 not received. Many patients noted they were not seen by the same medical doctor or
40 caretakers at intake and feedback consultations and emphasised the need for someone who
41 accompanies, informs, advises and assists them at all stages of the care process with whom
42 they can build a relationship of trust, who can because of his specialised knowledge and skills,
43 identify their needs and expectations and answer them. Having an established relationship
44 with a physician was important and highly valued in both the diagnosis and management of
45 the condition while not having such an ongoing relationship was reported to make it difficult to
46 achieve agreement about the symptoms and the diagnosis as patients were often unable to
47 demonstrate the extent of their condition within a consultation at times they may not be
48 experiencing symptoms

49 Explanation of quality assessment: minor concerns over methodological limitations with
50 moderate concerns over one study (due to the potential impact of the researcher on the
51 findings not being discussed and concerns over data analysis with lack of sufficient information

1 on the data analysis process and concerns over data richness with some findings supported
2 by single quotes) but minor concerns over one study (due to concerns over data richness with
3 findings mostly supported by single quotes), very minor concerns over another study (due to
4 the potential impact of the researcher on the findings not being discussed) and no concerns
5 over the fourth contributing study; no concerns about coherence with a clear theme emerging
6 in the contributing studies; no concerns over relevance; no concerns over adequacy, the theme
7 being supported by sufficient information across the studies. Overall assessment of confidence
8 was moderate due to the methodological concerns identified.

9 **Review finding 11: Social support**

10 Patients were physically and mentally isolated struggling to maintain relationships because of
11 their symptoms, with many losing close friends and using social media and online forums,
12 which were helpful, but not commensurate with face-to-face interactions. Feelings of isolation
13 were also reported by carers who had to devote a lot of time to attend patients' needs. Those
14 completing treatment at NHS specialist services recalled group sessions positively, with
15 benefits including relating to other patients, the opportunity to share experiences and stories,
16 receiving support from group members, supporting others, hearing about their experiences
17 and having their own personal experiences and symptoms validated and normalised. Some
18 thought it important that a group 'clicked' in order for the benefits of mutual support to be
19 realised. Hearing from or about individuals who had recovered or substantially improved
20 following ME/CFS was perceived by patients as a potential source of encouragement which
21 could be offered by specialist services.

22 Explanation of quality assessment: minor concerns over methodological limitations with
23 moderate concerns over one study (due to concerns over the appropriateness of the data
24 collection method, the study being a follow-up to a quantitative study with open-ended online
25 responses) but minor limitations in one study (due to the potential influence of the researcher
26 not being discussed and concerns over data analysis due to data richness with themes mostly
27 supported by single quotes), very minor limitations in one study (due to the potential influence
28 of the researcher on the findings not being discussed) and no concerns over the fourth
29 contributing study; no concerns about coherence with nothing to lower the confidence rating;
30 minor concerns over relevance with moderate concerns in one study (due to participants being
31 self-identified as having ME/CFS) but with no similar concerns identified in any other
32 contributing study; no concerns over adequacy with sufficient information supporting the theme
33 across contributing studies. Overall assessment of confidence was moderate due to minor
34 concerns over methodological limitations and relevance.

35 **Review finding 12: Practical support with daily living & social care**

36 Lacking caregiver resources made it difficult for patients to cope, since they had to exert energy
37 to work themselves causing physical consequences. Patients felt hope, validation and
38 compassion when there were support systems present to help with daily living activities, with
39 a supportive loved one for instance reported to save a patient valuable resting time. Practical
40 support for personal care, family roles, independent living and support for carers was extremely
41 important for people with moderate to severe illness. Many reported needing help with all
42 personal and domestic tasks: with moving around the house, getting out of bed and chairs,
43 washing and dressing, feeding and self-care, running a home, including meals preparation,
44 shopping or cleaning and how these intensified with child care. Where people could not find
45 alternative ways of getting support for practical tasks, the home environment, where they had
46 to spend most of their time, became not their refuge but a further source of stressful
47 experiences of deteriorating well-being. Social care was of paramount importance for patients'
48 life priorities which often included sustaining their own roles as family caregivers. They
49 highlighted how lack of access to social care and practical support, which is the case for most
50 people with ME/CFS, made a profound impact on their ability to carry out their family caregiving
51 roles, particularly as parents, raising questions about how people will be able to manage their

1 lives with the debilitating symptoms they reported. Without social care support, often partners,
2 parents and sometimes children had to become carers.

3 Explanation of quality assessment: Minor concerns over methodological limitations with
4 moderate concerns in one study (due to concerns over the appropriateness of the data
5 collection method, the study being a follow-up to a quantitative study with open-ended online
6 responses) but very minor concerns in the other contributing study where the majority of the
7 information supporting the theme emerged from (due to the role of the researcher not being
8 discussed); no concerns about coherence with nothing to lower the confidence rating; very
9 minor concerns over relevance as although patients of one study were self-identified as having
10 ME/CFS, the sample of the study contributing the most information to this theme included
11 patients from diverse social and ethnic backgrounds, degrees of illness severity and duration,
12 recruited through ME/CFS support groups and selected by clinicians and there were no
13 concerns over the accuracy of their diagnosis; no concerns over adequacy with sufficient
14 information supporting the theme. Overall assessment of confidence was moderate due to the
15 concerns over methodological limitations and concerns over relevance were too minor to
16 further lower the confidence rating.

17 **Review finding 13: Need for tailored and accessible hospital care**

18 Patients highlighted the importance of flexibility when making medical appointments and
19 accommodating treatment programmes around their commitments, lack of which could be a
20 barrier to attendance. They noted travel during the early stages of the illness or when
21 symptoms were severe could be incredibly hard with participants finding the journey stressful
22 and needing to recover after appointments. Some discussed the importance of good public
23 transport links to the specialist service, whilst others felt that they would not have been able to
24 attend appointments without use of a car. Concerns were raised about the ability of those
25 severely affected by 'CFS/ME' to access specialist services. Flexibility in the frequency and
26 mode of appointments was valued by participants because of travel burden and symptom
27 fluctuation. The option of having some appointments by telephone was highly valued,
28 particularly when symptom severity or travel problems made attendance difficult. Skype was
29 also mentioned as a possibility

30 Patients who had hospital care also described their need for designated wards for ME/CFS,
31 with environments adapted to their needs, as in keeping light and noise levels low. Some
32 highlighter the limited time for consultation as a barrier to appropriate care provision and
33 another reason for seeking support outside the NHS.

34 Explanation of quality assessment: Very minor concerns over methodological limitations with
35 minor concerns in one study (due to the potential impact of the researcher on the findings not
36 being discussed and concerns over data analysis due to data richness, with findings supported
37 by single quotes) but very minor limitations in one study (due to the potential impact of the
38 researcher on the findings not being discussed) and no concerns over the third contributing
39 study; no concerns about coherence, the finding clearly emerging from the three studies; no
40 concerns about relevance; minor concerns about adequacy with information supporting the
41 finding in one study being limited. Overall assessment of confidence was moderate due to
42 minor concerns over adequacy and methodological concerns were too minor to lower the
43 confidence rating.

44 **Review finding 14: Need for a diagnosis:**

45 Patients described gaining diagnostic clarity as a step towards regaining self-respect, being
46 able to explain the problem to others and as a positive move towards regaining control of their
47 lives or taking constructive action to improve their symptoms. Achieving a diagnosis was seen
48 as a crucial milestone for most participants as it often led to advice from doctors and other
49 health care professionals with particular knowledge of CFS/ME. However, patients often
50 encountered oppositional health service responses and some therefore decided to use private
51 or alternative health services as a way of getting diagnosis or help, often exacerbating stress,

1 uncertainty and financial pressures. It was also reported that until a diagnosis was gained,
2 social services could not assess their needs

3 Explanation of quality assessment: very minor concerns over methodological limitations with
4 very minor concerns in both contributing studies (due to the role of the researcher not being
5 discussed); no concerns about coherence with nothing to lower the confidence rating; no
6 concerns about relevance; no concerns about adequacy with sufficient information to support
7 the theme emerging from the studies. Overall assessment of confidence was high as concerns
8 over methodological limitations were too minor to lower the confidence rating.

9 **Review finding 15: Need for a positive diagnosis and future direction**

10 When reflecting on the role of their GP, patients wanted to receive a positive diagnosis. They
11 highlighted the need for the diagnosis to be given in a positive way to maintain hope that
12 symptoms can improve. Hearing from or about individuals who had recovered or substantially
13 improved following ME/CFS was perceived as a potential source of encouragement which
14 could be offered by specialist services, while media accounts of individuals who were very
15 severely disabled appeared to be a key source of anxiety. When describing their expectations,
16 a clear tension was evident between wanting and believing in hope for the future and fear that
17 their illness would not improve or might deteriorate, and patients hoped that referral to a
18 specialist service would give them positive direction for the future.

19 Explanation of quality assessment: minor concerns over methodological limitations with very
20 minor concerns in two contributing studies (due to the potential impact of the role of the
21 researcher not being discussed) and minor concerns in one study (due to the potential impact
22 of the researcher on the findings not being discussed and concerns about data analysis due
23 to data richness with themes supported by single quotes); no concerns about coherence with
24 nothing to lower the confidence rating; no concerns about relevance; minor concerns about
25 adequacy with limited information supporting the theme in one study but sufficient information
26 emerging from the other studies. Overall assessment of confidence was moderate due to
27 methodological limitations and minor concerns over adequacy.

28 **Review finding 16: Patient support during medical consultations**

29 Patients and carers described how visiting a GP can be a challenging experience, with patients
30 describing difficulty in remembering or articulating their symptoms and how they would take a
31 carer or family member with them to make sense of the consultation. Patients and carers
32 described the important role that carers play in the management of the illness, which included
33 support during a GP consultation.

34 Explanation of quality assessment: minor concerns over methodological limitations in the
35 contributing study (due to the potential impact of the researcher on the findings not being
36 discussed and data analysis because of concerns over data richness with themes mostly
37 supported by single quotes); no concerns about coherence with the theme clearly emerging
38 from the data with nothing to lower the confidence rating; no concerns over relevance;
39 moderate concerns over adequacy with relatively limited information to support the theme.
40 Overall assessment of confidence was low due the concerns over data richness impacting the
41 quality of the study's methodology and adequacy.

42 **Review finding 17: Advice (for carers) on how to support patients**

43 Carers appeared to have a lack of understanding of 'CFS/ME' and were frustrated by the
44 patient's symptoms. They discussed the danger of giving inappropriate advice or support to
45 patients, exacerbating symptoms or pushing the patient to do too much too soon.

46 Explanation of quality assessment: minor concerns over methodological limitations in the
47 contributing study (due to the potential impact of the researcher on the findings not being
48 discussed and concerns about data analysis over data richness with themes supported by
49 single quotes); no concerns about coherence, with the theme clearly emerging from the data

1 and nothing to lower the confidence rating; no concerns about relevance; serious concerns
 2 about adequacy with very limited information from the study supporting the theme. Overall
 3 assessment of confidence was very low due to concerns over methodological limitations and
 4 adequacy.

5 **1.1.6.2.2 Information education and support for children and young people with**
 6 **ME/CFS, their families or carers**

7 **Table 4: Review findings for Children and young people**

Main findings	Statement of finding
Information needs	
Information about ME/CFS ^{18, 23, 61, 69, 89}	A lack of information about ME/CFS and particularly about the symptoms and prognosis of the illness reported by both adolescent patients and their parents contributed to negative and undesirable feelings of uncertainty, fear and worry for both groups of people, while learning about the condition was reported to be helpful, contributing to feelings of validation in adolescents.
Sources of information: need for digital resources ²³	Adolescents used website resources they considered reliable but preferred patient-led sites and social media that contained experiential accounts which provided a great source of support that could be accessed at any time and facilitated coping.
Types of information ²³	Having in-depth, reliable and relatable, information that is patient friendly and regularly updated can be therapeutic for adolescents.
Support needs	
Need for validation & advocacy ^{18, 23, 69, 89}	A lack of understanding and validation of their experience from both medical professionals and their social environment was reported by young people with ME/CFS who felt the need to talk about their illness and often utilised online resources to help others understand; recognition and acknowledgment of the condition from specialist services resulted in a sense of relief in adolescents and parents.
Support with acceptance & adaptation ^{18, 23, 89, 130}	The acceptance of and adaptation to the lifestyle changes brought by ME/CFS, including changes in their identity, activities and social relationships, appeared challenging for young patients and carers and often impacted their mood and well-being.
Management strategies & support with implementation ^{18, 89}	A lack of resources for management was reported by parent-carers and where available, specialist guidance on management of ME/CFS and medical care strategies- although potentially difficult to integrate- were reported to positively impact the life of both young people and their families.
Mutual patient support ^{23, 69, 130}	Young people with ME/CFS reported on the importance of developing a connection with other patients either online or offline which provided support, alleviated feelings of isolation and benefited recovery.
Educational support ^{18, 130}	Lack of support within young peoples' educational environment was a source of anxiety and low mood, while better communication between healthcare and education

Main findings	Statement of finding
	providers enabled patients and carers to gain support and accommodate their needs.
Further needs	
Need for a diagnosis ^{18, 69}	The period before receiving a formal diagnosis was particularly difficult for adolescents with ME/CFS and their carers, while gaining a diagnostic label facilitated steps towards recovery and access to educational support.
Tailored approach to care ^{18, 130}	The importance of a tailored treatment approach and its impact on recovery was highlighted by adolescent patients and adolescent patient mothers
Referral to specialist service ¹⁸	Young people with ME/CFS and their mothers reported how accessing specialist ME/CFS services had benefited them in terms of diagnosis, information and guidance, psychological support, symptom management, treatment and recovery as well as in terms of access to educational support.

1

2 Review finding 1: Information about ME/CFS

3 Young people appeared to experience difficulty with a lack of information around the condition
4 from medical professionals involved. Adolescents talked about the intention to fact-find, with
5 reference to carrying out research and to know more about the condition. Reading facts about
6 symptoms was reported to be helpful by adolescents who identified with the information and
7 felt validated by this. Many negative and undesirable emotions were described relating to
8 various aspects of the young people's experience of 'CFS', including the uncertainty around
9 the illness diagnosis and prognosis, with participants reporting that not knowing why they were
10 undergoing many tests made them feel worried and not knowing what to expect made them
11 feel scared. Mothers of adolescents with ME/CFS reported there was a lack of initial guidance
12 or information around the illness and day-to-day management of symptoms that lead to initial
13 confusion and uncertainty about their child's health and possible diagnosis. Parents reported
14 feelings of uncertainty in regards to prognosis, talking about fears of death when their
15 son/daughter was at his/her worst and parents of adolescents with ME/CFS experiencing
16 eating difficulties felt it would help to know eating problems are 'normal' in this population and
17 may help them understand the illness further.

18

19 Explanation of quality assessment: Moderate concerns over methodological limitations with
20 moderate concerns in one study (due to the role of the researcher not being discussed and
21 serious concerns over data richness), serious concerns in another study (associated with the
22 use of public data, where the lack of detail on the primary data collection methods and
23 participant recruitment impacted our ability to assess risk of bias) but minor concerns in three
24 studies (due to the role of the researcher not being discussed in two studies along with lack of
25 sufficient detail over the data analysis process in one study and minor concerns over data
26 richness in the other study and due to the very small sample size and homogenous population
27 of one study); no concerns about coherence; moderate concerns over relevance with minor
28 concerns identified in all contributing studies (due to one study including parent carers of
29 ME/CFS patients that were not limited to the children and young people age stratum, due to
30 the very small sample size and homogenous population of people who attended the same
31 clinic that was limited to people that had recovered, the sample of one study being limited to
32 people with ME/CFS who experienced eating difficulties, one study excluding severely affected
33 individuals and the sample in one study consisting of participants from a feasibility RCT); no
34 concerns about adequacy with sufficient information supporting the theme. Overall
35 assessment of confidence was low due to the methodological limitations of the contributing
36 studies and concerns over relevance.

1 Review finding 2: Sources of information: need for digital resources

2 Adolescents with ME/CFS sought 'official' sites to establish facts about the condition and
3 talked about the status of those sites as 'reliable'. For the majority, this included National
4 Health Service (NHS) websites, and in some cases also Action for ME, Association of Young
5 people with ME/Chronic Fatigue and the ME Association. However, they reported only using
6 the NHS sites a few times with most moving on to explore patient-led and peer-led sites
7 containing subjective, experiential accounts. This included health forums, but also sites that
8 were not necessarily health-related, including Facebook, Instagram, Blogs and YouTube. In
9 contrast to NHS sites, these were accessed regularly and over the long-term, such as a few
10 times per week or every day. For most participants it was the patient-led/peer-led sites that
11 were associated with coping. Anecdotes and endorsements appeared to promote beliefs in
12 the efficacy of strategies, providing inspiration to try out the new strategies. The fact that
13 these sites could be rapidly accessed at any time seemed to provide a great sense of
14 support. Being able to interact with these sites in a quick and undemanding way through a
15 shared language of 'likes' and 'comments' was less demanding and more flexible than offline
16 relationships especially in the context of a disabling and fluctuating illness. Technological
17 affordances including videos were described as facilitating a sense of relationship and the
18 fact that these sites could be rapidly accessed at any time seemed to provide a great sense
19 of support.

20 Explanation of quality assessment: Minor concerns over methodological limitations in the
21 contributing study (due to the role of the researcher not being discussed and lack of details
22 on the analysis process followed); no concerns about coherence with the theme clearly
23 emerging from the study and nothing to lower the confidence rating; minor concerns over
24 relevance as the study excluded severely affected individuals; no concerns about adequacy
25 with sufficient information to support the theme. Overall assessment of confidence was
26 moderate due to concerns over methodology and relevance.

27 Review finding 3: Types of information

28 Adolescents with ME/CFS talked about the status of official websites they were accessing as
29 'reliable', but felt that NHS sites were not user-friendly, as they used medical terminology,
30 lacked depth and their content remained unchanged. Regularly accessed sites (i.e. patient-
31 led/peer-led) used ingroup terms and phrases which were accessible and appealing, were
32 considered to offer greater level of depth and were constantly updated. They preferred the
33 numerous accounts and story-telling approach of patient-led/peer-led and non-health-related
34 sites and which provided support with the psychological difficulties they often experienced,
35 encouraged them to open up and seek help offline and the technological affordances of
36 videos.

37 Explanation of quality assessment: Minor concerns over methodological limitations in the
38 contributing study (due to the role of the researcher not being discussed and lack of details
39 on the analysis process followed); no concerns about coherence with nothing in the study to
40 lower the confidence rating in the finding; minor concerns about relevance as the study
41 excluded severely affected individuals; no concerns about adequacy with sufficient
42 information to support the theme. Overall assessment of confidence was moderate due to
43 concerns over methodological limitations and relevance.

44 Review finding 4: Need for validation and advocacy

45 Young people appeared to experience difficulty with a lack of understanding and awareness
46 around the condition from medical professionals involved. They referred to a fear or actual
47 experiences of disbelief and judgment from others (including teachers and peers), which was
48 particularly difficult for some. Lack of understanding and invalidation from friends was
49 discussed but individuals talked about showing their friends online information to legitimise
50 their illness and help their peers understand and adapt to the illness. Adolescents referred to
51 specialist ME/CFS services reported that specialist medical care was positive, as it enabled

1 them to talk about their illness while their mothers reported that the specialist service
2 recognised and acknowledged the young person's condition, resulting in a sense of relief and
3 reassurance.

4 Parents reported they were often met with disbelief and hostility from medical professionals,
5 that close friends and other family members were sceptical and perplexed by the illness and
6 as a result may find themselves disconnected from others who may not respond well.
7 Parents felt alienated and reported being estranged from friends and family as the role of
8 carers became their priority. Because of a lack of understanding by those who are not
9 immediately impacted by the illness, carers were left feeling alone in their world. Parents
10 report hoping for change in terms of the way ME is received and wish for understanding from
11 others.

12 The theme was also supported by participants' behaviour outside interviews described in one
13 study. As reported by authors, the impression given in participants' comments prior the
14 interviews was that they felt it was important to recount their story so that the full extent of
15 their difficulties might be recognised, listened to and learnt from by others.

16 Explanation of quality assessment: Moderate concerns over methodological limitations with
17 minor concerns in three studies (due to the role of the researcher not being discussed in two
18 studies along with a lack of sufficient detail on the data analysis process in one study,
19 concerns over data richness in the other study and due to the very small sample size and
20 homogenous population of people who attended the same clinic in one study) but serious
21 limitations in the fourth contributing study (related to the use of public data where a lack of
22 detail on participant recruitment and the primary data collection methods implicated our
23 ability to assess risk of bias); no concerns about coherence with nothing to lower the
24 confidence rating; moderate concerns over relevance with minor concerns across
25 contributing studies (due to the exclusion of severely affected individuals from two studies,
26 one of which also consisted of people who had been previously recruited in an RCT, one
27 study including parent carers of ME/CFS patients that were not limited to the children and
28 young people age stratum, due to the aforementioned very small sample size and
29 homogenous population one study that was also limited to people that had recovered, whose
30 views may differ from those with active ME/CFS) ; no concerns about adequacy. Overall
31 assessment of confidence was low due to methodological limitations and concerns about
32 relevance.

33 **Review finding 5: Support with acceptance & adaptation**

34 Young people with ME/CFS reported that 'CFS/ME' put a strain on normal adolescent life,
35 such as their identity and friendships and that accepting that for a time they must reduce their
36 activity levels and adopt a routine was challenging. They commonly reported that ME/CFS
37 symptoms prevented, restricted or interfered with their activities. Not being able to do
38 activities they fully enjoyed or engage in them fully provoked negative emotions such as low
39 mood, frustration, boredom and hopelessness.

40 ME/CFS was also reported to impact various aspects of the parents' identity, such as their
41 roles within their families, relationships and aspects of their lives outside of their families
42 such as their careers. Parents reported taking on roles as carers, advocates for the illness
43 and their children and support their children as educators. Parents described the ways in
44 which their roles within their families have changed to include caretaking responsibilities and
45 the way this has affected their own well-being. The role of parental carers is a balance of
46 providing day-to-day care for the person who is ill while providing affection. Parents may
47 struggle to find ways to provide affection to their children who are often in great pain; in
48 essence 'ME' may change the way in which parents relate to their children. Taking a new
49 role results in much of the parent's time being allotted to caretaking responsibilities. As a
50 result of parents taking a new caretaking role, they reported being distanced from
51 relationships with other family members, from their hobbies and careers. They described how
52 their relationship with their spouse and children changed and distancing is manifested in the

1 relationships within the family unit, with parents reporting that their family plans and habits
2 have changed.

3 Explanation of quality assessment: Moderate concerns over methodological limitations with
4 minor concerns in three studies (due to the role of the researcher not being discussed in two
5 studies along with a lack of sufficient detail on the data analysis process in one study and
6 concerns over data richness in the other study, and due to concerns over potential selection
7 bias in the third study as recruitment across a multidisciplinary team meant reasons for
8 declining participation were not clear) but serious concerns over the fourth contributing study
9 (related to the use of public data where a lack of detail on participant recruitment and the
10 primary data collection methods implicated our ability to assess risk of bias); no concerns
11 about coherence, the theme clearly emerging in the studies; moderate concerns over
12 relevance due to minor concerns over all four contributing studies (with severe cases of
13 ME/CFS excluded from two studies and the sample of one of which also consisting of
14 participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the
15 RCT, with the sample of one study being limited to ME/CFS patients with co-morbid low
16 mood and that of one study including carers of ME/CFS patients that were not limited to the
17 children and young people age stratum); no concerns over adequacy with sufficient
18 information to support the theme. Overall assessment of confidence was low due to concerns
19 over methodological limitations and relevance.

20 **Review finding 6: Management strategies & support with implementation**

21 Parents reported a lack of treatments and resources for managing 'ME'. Young people
22 reported that guidance on how to manage their condition given from specialist medical care
23 brought structure and a sense of normality back to their lives. A few of the mothers of these
24 young people noted that specialist medical care strategies had an impact on the whole family
25 and could be difficult to integrate with their routine lifestyle. Some mothers felt that the
26 'CFS/ME' service reinforced symptom management strategies that they had been trying to get
27 their child to follow and felt that their child would be more likely to listen if techniques were
28 legitimised by a health-care professional.

29 Explanation of quality assessment: Moderate concerns over methodological limitations with
30 minor concerns in one study (due to the role of the researcher not being discussed and data
31 richness) but serious concerns in the other contributing study (associated with the use of public
32 data and lack of detail on how the sample was derived, the methods of primary data collection
33 and inability to assess bias); no concerns about coherence with the theme being well grounded
34 in the data with coherent information emerging; minor concerns over relevance with due to
35 minor concerns in both studies (due to the sample in one study consisting of parent carers that
36 were not limited to the stratum of children and young people and the sample of one study
37 consisting of participants from a feasibility RCT which may differ from ME/CFS patients not
38 recruited in the RCT); minor concerns over adequacy associated with aforementioned
39 concerns over data richness. Overall assessment of confidence was low due to methodological
40 limitations and concerns over relevance and adequacy.

41 **Review finding 7: Mutual patient support**

42 Adolescents with ME/CFS felt that building supportive networks, including developing
43 relationships with other young people with ME/CFS could be beneficial and talked about the
44 potential of feeling understood and less alone. Some experienced difficulty talking about the
45 psychological difficulties they often experienced with family, friends and clinicians and reading
46 stories online to seek support. They described the loneliness of the condition and how
47 spending time on patient-led websites helped them develop a connection with others like them
48 and a sense of community which made them feel understood and alleviated this isolation.
49 Young people also reported that meeting and talking to others with 'CFS' was helpful in the
50 management of and recovery from the illness.

1 Explanation of quality assessment: Moderate concerns over methodological limitations with
2 minor limitations in all contributing studies (due to the role of the researcher not being
3 discussed and lack of details on the analysis process followed in one study, due to the very
4 small sample size and homogenous population of people who attended the same clinic in one
5 study and due to potential selection bias in the other study as recruitment across a
6 multidisciplinary team meant reasons for declining participation were not clear); no concerns
7 about coherence the theme clearly emerging from three studies; moderate concerns over
8 relevance with minor concerns across contributing studies (due to adolescents with severe
9 ME/CFS excluded from one study, all participants having recovered from ME/CFS and
10 potentially holding different opinions to those in the active stage of ME/CFS in one study and
11 participants in one study having co-morbid low mood potentially holding different opinions to
12 those who do not); no concerns over adequacy. Overall assessment of confidence was low
13 due to methodological limitations and concerns over relevance.

14 **Review finding 8: Educational support**

15 Young people felt that better support from education systems could have helped. They
16 described schools and colleges as inflexible, unhelpful, un-empathetic and invalidating and
17 identified this as a cause of increased anxiety and low mood. They particularly reported that
18 teachers had not been very supportive. Mothers of those who had accessed a ME/CFS
19 specialist service discussed the beneficial way in which the service opened channels of
20 dialogue between health-care professionals and education providers in a variety of ways. A
21 letter provided by the 'CFS/ME' service confirming a diagnosis enabled mothers to legitimately
22 take their child out of school, request funding for home schooling and more generally inform
23 and gain support from teachers when managing reduced attendance.

24 Explanation of quality assessment: Minor concerns over methodological limitations in both
25 contributing studies (due to the role of the researcher not being discussed and concerns over
26 data richness in one study, potential selection bias in the other study as recruitment across a
27 multidisciplinary team meant reasons for declining participation were not clear); no concerns
28 about coherence, the theme clearly emerging from the data; minor concerns relevance
29 associated with both contributing studies (due to the sample in one study consisting of
30 participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the
31 RCT and due to patients in the other study having co-morbid low mood potentially limiting the
32 representativeness of their views for ME/CFS patients without low mood); moderate concerns
33 about adequacy with relatively limited information supporting the theme across studies. Overall
34 assessment of confidence was low due to concerns over methodological limitations, relevance
35 and adequacy.

36 **Review finding 9: Need for a diagnosis:**

37 Adolescents described a long period of diagnostic uncertainty as being particularly difficult
38 both in terms of feeling helpless and in terms of feeling disbelieved. Arriving at a diagnostic
39 label for the illness appeared to be a helpful experience for constructing their own meaning
40 and dispelling disbelief from themselves and from others. Mother of adolescents described
41 how a formal diagnosis enabled positive change and steps towards a managed recovery and
42 how confirmation of diagnosis enabled mothers to legitimately take their child out of school,
43 request funding for home schooling and more generally inform and gain support from
44 teachers when managing reduced attendance

45 Explanation of quality assessment: Minor concerns over methodological limitations in both
46 contributing studies (due to the role of the researcher not being discussed and concerns over
47 data richness in one study, due to the small sample size and homogenous population of
48 participants who attended the same clinic of the other study); no concerns about coherence
49 with the theme very clearly emerging from the data in both contributing studies; minor
50 concerns over relevance associated with both contributing studies (due to the sample of one
51 study consisting of participants from a feasibility RCT which may differ from ME/CFS patients
52 not recruited in the RCT; participants in the other study having recovered from ME/CFS and

1 therefore possibly not holding similar opinions to those in the stage of active ME/CFS); minor
2 concerns over adequacy due to the relatively limited information supporting the finding in the
3 studies. Overall assessment of confidence was moderate due to minor concerns over
4 methodological limitations, relevance and adequacy.

5 **Review finding 10: Tailored approach to care**

6 Young people found different treatment approaches helpful and emphasised the importance
7 of an individualised approach. Some talked about finding CBT helpful while others recognised
8 that AM or the combination of CBT with medication could be helpful approaches. The
9 importance of a tailored approach was also highlighted by mothers of adolescent patients who
10 reported that the tailored, patient-centred specialist medical intervention they gained access
11 to via referral to a specialist service enabled positive change and steps towards managed
12 recovery.

13 Explanation of quality assessment: Minor concerns over methodological limitations in both the
14 contributing studies (due to the role of the researcher not being discussed and concerns over
15 data richness in one study and potential selection bias in the other study as recruitment across
16 a multidisciplinary team meant reasons for declining participation were not clear); no concerns
17 about coherence with nothing to lower the confidence rating; minor concerns over relevance
18 with minor concerns in both contributing studies (due to the sample in one study consisting of
19 participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the
20 RCT and because patients in the other study had co-morbid low mood which potentially limits
21 the representativeness of their views for ME/CFS patients without low mood and it is unclear
22 whether the information on the importance of an individualised approach to care reported is in
23 relation to their ME/CFS or low mood); moderate concerns over adequacy with relatively limited
24 information supporting the theme. Overall assessment of confidence was low due to concerns
25 over methodological limitations, relevance and adequacy.

26 **Review finding 11: Referral to specialist services**

27 Prior to accessing specialist services, mothers of adolescent patients with ME/CFS
28 experienced a lack of initial guidance or information around the illness and day-to-day
29 management of symptoms that led to confusion and uncertainty about their child's health and
30 possible diagnosis. Specialists provided guidance on how to manage ME/CFS which was
31 reported to bring structure and a sense of normality back to lives of young people. Mothers
32 reported the initial assessment appointment with the 'CFS/ME' service as a positive
33 experience, which was useful and helpful and how referral to a specialist service gave families
34 access to an informative team of experts, for some a formal diagnosis, and for all a tailored,
35 patient centred specialist medical intervention that had not been available earlier which
36 enabled positive change and steps towards managed recovery.

37 Mothers reported that the specialist service recognised and acknowledged the young person's
38 condition, resulting in a sense of relief and reassurance. Mothers felt that symptoms were now
39 understood and that they would receive help. Adolescents reported that specialist medical care
40 was positive, as it enabled them to talk about their illness.

41 Some mothers felt that the 'CFS/ME' service reinforced symptom management strategies that
42 they had been trying to get their child to follow and felt that their child would be more likely to
43 listen if techniques were legitimised by a health-care professional. A few mothers also noted
44 that specialist medical care strategies had an impact on the whole family and could be difficult
45 to integrate with their routine lifestyle.

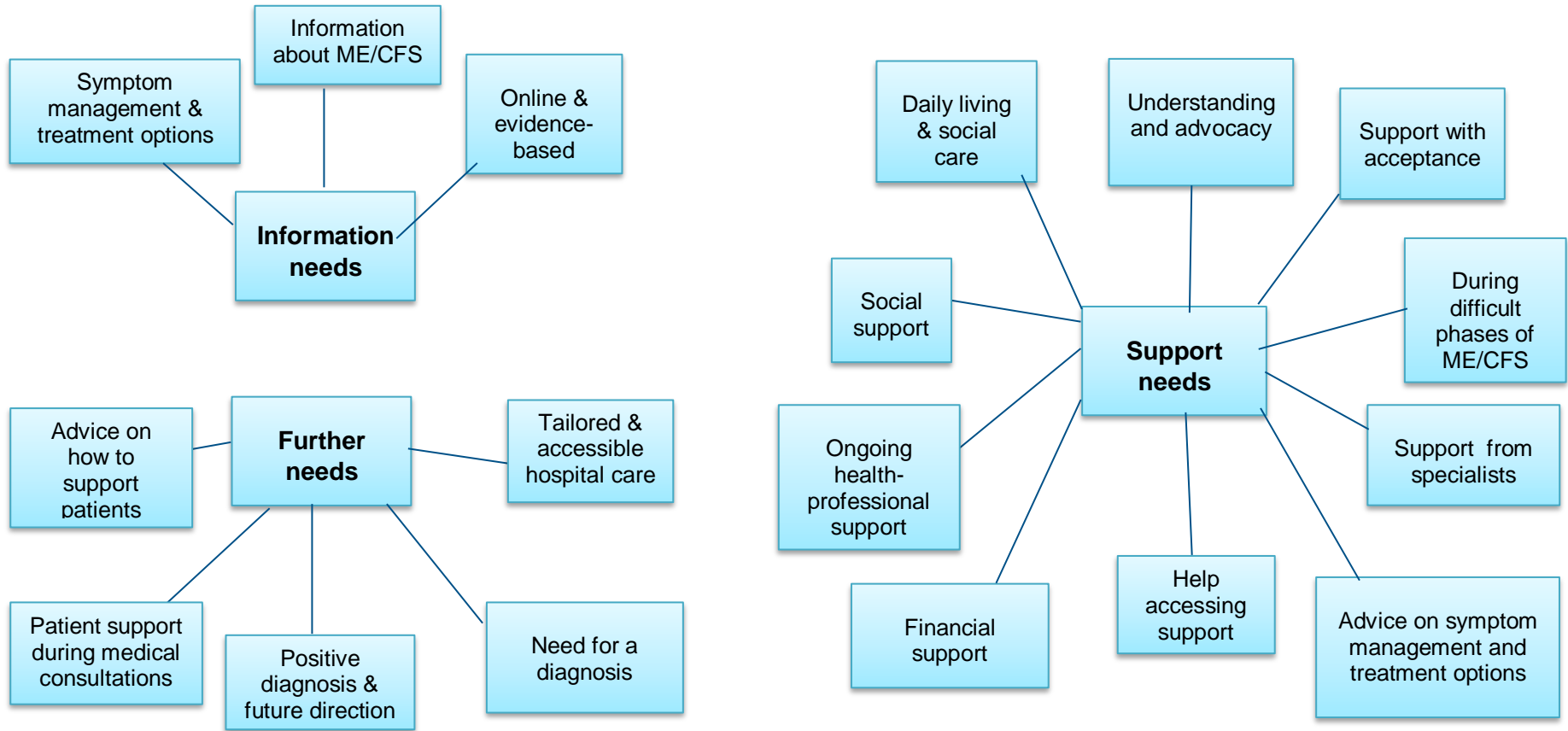
46 Mothers also discussed the beneficial way in which the 'CFS/ME' service opened channels of
47 dialogue between health-care professionals and education providers in a variety of ways. A
48 letter provided by the 'CFS/ME' service confirming a diagnosis enabled mothers to legitimately
49 take their child out of school, request funding for home schooling and more generally inform
50 and gain support from teachers when managing reduced attendance.

1 Explanation of quality assessment: minor concerns over methodological limitations in the
2 contributing study (due to the role of the researcher not being discussed and concerns over
3 data richness); no concerns about coherence with nothing to lower the confidence rating;
4 moderate concerns over relevance with referral to specialist services emerging as a positive
5 and useful experience exclusively from a study aiming to examine that and by people who had
6 accessed a specialist service and not reported as a need of young people with ME/CFS in
7 other studies; no concerns over adequacy with sufficient information to support the theme
8 emerging from the study. Overall assessment of confidence was moderate due to the concerns
9 over methodological limitations and relevance.

10

1

Figure 1: Theme map of review findings (adults)

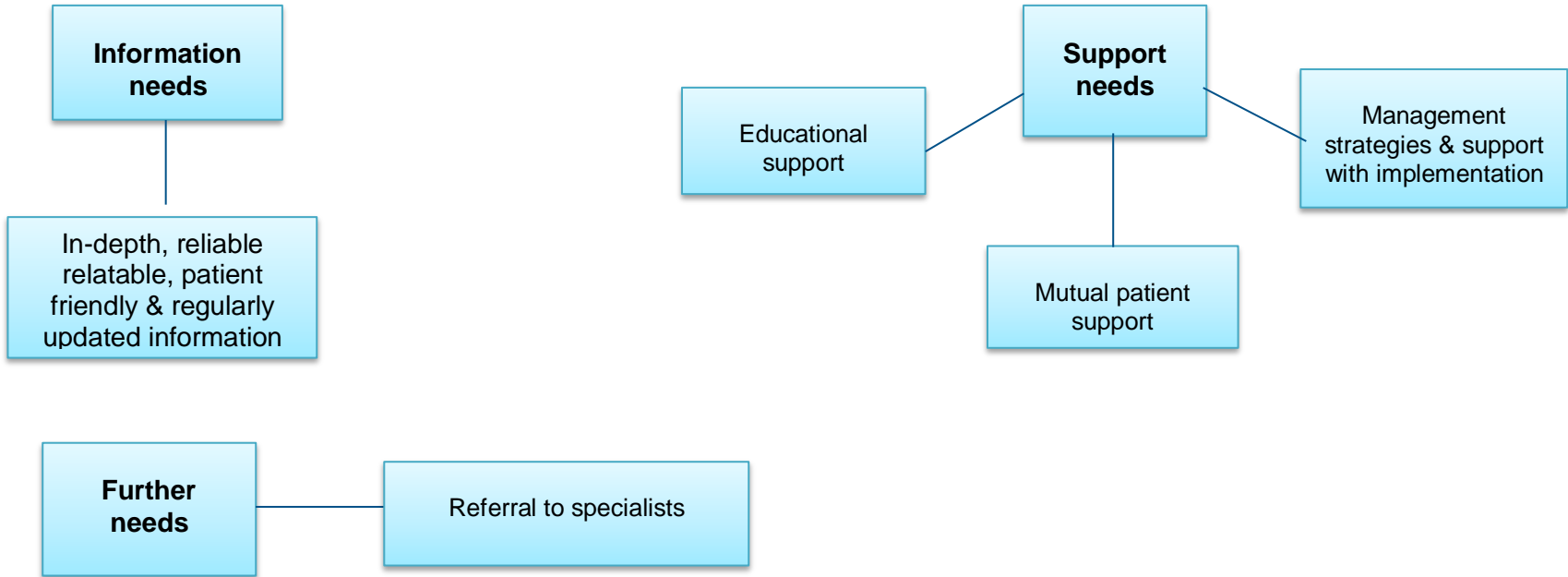


2

3

1

Figure 2: Theme map of review findings (additional themes in children/young people)



2

1 1.1.7 Economic evidence

- 2 The committee agreed that health economic studies would not be relevant to this review
- 3 question, and so were not sought.

1 **2 The committee's discussion and** 2 **interpretation of the evidence**

3 The committee discussed this evidence with the findings from the evidence reviews on
4 evidence report C: access to care, evidence report B: information for health and social care
5 professionals, evidence report D: diagnosis and evidence report I: multidisciplinary teams
6 and the report on Children and Young people (Appendix 1) and people with severe ME/CFS
7 (Appendix 2). Where relevant these sources are noted.

8 **2.1 The quality of the evidence**

9 Fifteen qualitative studies were included in the review. The majority of the studies were
10 conducted with adults with ME/CFS, one study included the views of family and carers. Six
11 studies were conducted on adolescents, two of which included the families and carers of
12 participants and two of which excluded severely affected individuals. There was one study
13 conducted exclusively on parent carers caring for children from 5 years old to adults (the
14 majority caring for adult daughters). The information reported in this study was considered to
15 contribute to emerging themes relevant to both strata of adults, and children and young
16 people.

17 Confidence in the review findings ranged from high to very low. Main reasons for
18 downgrading were methodological limitations and relevance. The most common
19 methodological limitations identified were insufficient reporting of the role of the researcher in
20 the data collection, insufficient reporting or limitations of the methods of data analysis and
21 insufficient data presented to support the findings. There were concerns regarding the
22 relevance of findings from studies which included subgroups of the review population. For
23 example, the experiences of people with self-identified ME/CFS, those with eating difficulties,
24 suicidal ideation, low mood, and those participating in clinical trials. Although this evidence
25 cannot be generalised to the wider ME/CFS population the committee agreed that it was
26 useful to help to build a more complete picture of the information, education and support
27 needs of all people with ME/CFS.

28 Two studies explicitly stated that people with severe ME/CFS were excluded, and the
29 committee considered it unlikely that other studies would have included people with severe
30 ME/CFS due to the difficulties in conducting research in this population.

31 Some findings were based on evidence from a small number of studies, this indicates
32 coherence was less clear with concerns about the adequacy of data.

33 The committee placed greater weight on high and moderate confidence findings than low
34 and very low confidence findings during discussion of the evidence. However, they
35 acknowledged that some lower confidence findings reflected their own experience and
36 should not be overlooked. The committee agreed the findings highlighting the importance of
37 signposting to relevant contacts, financial support, support from caregivers during medical
38 consultations and advice for carers on how to support people with ME/CFS were all
39 commonly identified as areas information was hard to find by people with ME/CFS.

40 The committee agreed evidence from the adult population reflected their knowledge and
41 experience about children and young people and could be used to support their decision
42 making for children and young people.

43 **2.2 Findings identified in the evidence synthesis**

44
45 Themes identified for children and young people broadly mirrored those identified for adults.
46 Findings unique to children and young people are highlighted.

1 Information needs

2

3 *Understanding ME/CFS*

4

5 Evidence suggested that people with ME/CFS and their carers need general information
6 about ME/CFS for both themselves and others (families, friends, employers and
7 practitioners), to enable them to develop accurate expectations about the future, relieve
8 feelings of distress caused by the general lack of sufficient information and to educate others
9 in order to gain understanding and empathy. The time following diagnosis of ME/CFS and
10 the early stages of treatment were difficult and people experienced feelings of distress,
11 difficulty with acceptance of diagnosis, of advice given and occasionally a lack of guidance,
12 while phases of relapse were particularly difficult for carers. The committee agreed that being
13 informed about ME/CFS, benefits people with ME/CFS and their carers in many ways.
14 Therefore, the committee recommended that people are provided with up to date information
15 throughout the diagnostic and care pathway and this information should be tailored to the
16 person's individual circumstances and expanded or repeated as required. The committee
17 agreed that as part of the information provided at the start when ME/CFS is suspected, it is
18 important to explain to people presenting with possible symptoms that there is no diagnostic
19 test for ME/CFS and that it is recognised on clinical grounds alone.

20

21 Evidence suggested that a positive direction for the future and the ME/CFS diagnosis being
22 framed in a positive way was important to people with ME/CFS and enabled them to maintain
23 hope for improvement. The committee discussed the ethical considerations regarding health
24 care professionals taking 'positive' or 'optimistic' approaches. It was considered that striking
25 a balance between fostering realistic hope without exaggerating the chances of recovery and
26 creating false hope was essential. It was the experience of the committee that people with
27 ME/CFS appreciate honesty from health care professionals. The committee also considered
28 it vital that when filling out assessment forms for financial, health and social support, people
29 can be honest about how their condition affects them and not pressured to be optimistic.
30 Therefore, the committee decided to recommend that people are given information about
31 ME/CFS that is balanced and realistic about living with the condition and possible outcome.
32 They recommended people with ME/CFS are advised about the unpredictability of the
33 condition, its impact on their life and need for adjustments, aggravating factors, relapse and
34 remission and possible ways to manage symptoms. They hoped that recommendations on
35 managing relapse would alleviate the difficulties experienced by people with ME/CFS and
36 carers during these periods.

37

38

39 Evidence suggested people with ME/CFS need support with acceptance of their new life
40 circumstances, their diagnosis and the likely prognosis of their illness in order to maximise the
41 benefit received from treatment. The committee noted the evidence highlighting the
42 importance of acceptance and willingness to change did not explicitly identify them as areas
43 in which people need support. The committee also noted that acceptance was also identified
44 in another review (Evidence review C: access to care) as a barrier to care and that
45 acceptance of the condition is a personal process. It was hoped that and recommendations
46 on providing information to people with ME/CFS would increase understanding of the
47 condition.

48

49

50 *Management of ME/CFS*

51

52 Evidence suggested that people need guidance on symptom management. They wanted
53 information on effective and diverse treatment options to choose from, including biomedical
54 and alternative treatments other than those currently offered by the health care system. The
55 committee considered this finding alongside the lack of evidence of clinical effectiveness for
56 many of the interventions reviewed in this guideline. The committee acknowledged the

1 frustration that people felt towards not being given information about all available treatment
2 options, and recognised the risk of people receiving information from resources that give
3 false hope and can result in people investing energy and money on interventions that have
4 no evidence of benefit. The committee agreed it was important that people with ME/CFS are
5 provided with all the information about risks and benefits regarding interventions for symptom
6 management to make an informed decision on whether they may be helpful.
7 Recommendations related to information for people with ME/CFS on interventions for
8 symptom management are discussed further in Evidence review G-Non-pharmacological
9 management. The committee were aware that lack of autonomy over treatment choices has
10 been reported by people with ME/CFS. They noted this was supported in the report Appendix
11 1 - Children and young people and in Evidence review G-Non-pharmacological management
12 People reported their experiences of being offered unhelpful treatments that they later
13 declined, and of having repeatedly been given advice that was unhelpful and so not followed.
14 This was sometimes perceived by healthcare professionals as getting in the way of their
15 recovery. While noting that every NICE guideline states people have the right to be involved
16 in discussions and make informed decisions about their care the committee agreed it was
17 important to make a recommendation to reinforce the importance of patient choice and the
18 right to decline or withdraw from any part of their management without it affecting any other
19 part of their care.

20

21 *Delivery of information*

22

23 Evidence suggested that people could benefit from information and resources that are
24 evidence-based and delivered in various formats, such as information leaflets, DVDs, and
25 online information as well as from alternative sources such as media representations of
26 people with ME/CFS. The committee considered the variety of different formats by which
27 information could be delivered and the potential advantages and disadvantages of each. For
28 example, written materials could be taken away and read at a time when the person has
29 more capacity to absorb the information and could be shared with friends/family/colleagues.
30 The committee noted the review finding was downgraded due to concerns regarding
31 coherence, as there were differences both between and within studies in the formats that
32 people preferred. This was in line with the committee's experience of individual preferences
33 and different formats being effective for different people. The committee noted that the NICE
34 guideline on patient experience (CG138) includes recommendations on giving people
35 information in accessible formats, including both written and oral. However, they considered
36 that people with ME/CFS could benefit from a wider variety of options. Therefore, the
37 committee recommended that information be made available in a variety of formats (for
38 example written materials, electronic and audio).

39

40 Information needs of children and young people

41

42

43 Findings related to the information needs of children and young people with ME/CFS were
44 broadly similar to those of adults. A specific finding related to children and young people was
45 not knowing why they were undergoing many tests or what to expect made them feel scared.
46 The committee considered that for this reason, maintenance of good communication
47 between health care professionals and children and young people with ME/CFS is an
48 important principle of care. They recommended that the voice of the child or young person is
49 heard by taking a child-centered approach with the communication focusing on them and
50 discussing and regularly reviewing with the child or young person how they want to be
51 involved in making decisions about their care (with the acknowledgment that parents and
52 carers may act as advocates).

53

54 *Delivery of information*

55

1 Evidence suggested that adolescents used website resources they considered reliable but
2 preferred patient-led sites and social media that contained experiential accounts. These
3 provided a great source of support, could be accessed at any time and facilitated coping.
4 Evidence also suggested having in-depth, reliable and relatable information that is patient
5 friendly and regularly updated can be therapeutic for adolescents. Despite the low quality of
6 the evidence, these findings reflected the experience of the committee. It was considered
7 that social media in particular could help young people feel to connected to others during
8 times when social contact is limited. This was supported by the finding that young people
9 with ME/CFS also reported on the importance of developing a connection with other patients
10 either online or offline and how this alleviated feelings of isolation and benefited recovery.
11 The committee noted the evidence was based on adolescents and no evidence was
12 identified in children. It was also discussed that as well as age, the potential benefits may
13 depend on symptom severity, cognitive ability, individual circumstances and preferences.
14 Therefore, the committee recommended that when providing information for children and
15 young people with ME/CFS, account should be taken of their developmental stage and level
16 of understanding, any disabilities or communication needs. Different formats such as one-to-
17 one discussion, group discussions, written materials and pictures, play, art and music
18 activities and digital media, for example social media should be used where appropriate.

19

20 Support needs

21

22 *Being believed*

23

24 Evidence suggested that people with ME/CFS and their carers needed understanding and
25 recognition of ME/CFS from the medical community, employers, colleagues, family and
26 friends. Not being believed was unhelpful in the workplace, in accessing healthcare and
27 social support, and impacted on their well-being and social relationships. Disbelief of the
28 legitimacy of the condition and the need for validation are themes that run throughout the
29 qualitative evidence reviews across the guideline, as well as the reports on children and
30 young people (Appendix 1), people with severe ME/CFS (Appendix 2) and Dr Muirhead's
31 expert testimony (Appendix 3). The committee prioritized the awareness of ME/CFS and
32 placed it at the front of the guideline. The first section of the guideline sets out principles of
33 care for people with ME/CFS, clearly stating that healthcare professionals should be aware
34 that ME/CFS is a real chronic medical condition that is complex and can have a significant
35 impact on people's quality of life and this reality should be acknowledged to the person with
36 ME/CFS. The committee hoped that these recommendations would lead to wider belief and
37 support from other important people such as employers, colleagues, family members and
38 friends.

39

40

41 *ME/CFS support groups*

42

43 Evidence suggested the time following diagnosis of ME/CFS and the early stages of
44 treatment were difficult and people experienced feelings of distress and difficulty with
45 acceptance of diagnosis. There was uncertainty about advice given to them and sometimes
46 a lack of any guidance. Phases of relapse were particularly difficult for carers to understand
47 and cope with. Despite the low quality of the evidence, the committee considered this finding
48 to be consistent with their own experience and recognised that some people may require
49 extra resources for information and support. The committee also considered this in relation to
50 the finding that people with ME/CFS and their carers felt isolated, struggling to maintain
51 social relationships and could benefit from social interactions and mutual patient support.

52

53 Evidence suggested that people with ME/CFS and their carers needed sign posting to
54 relevant contacts that would help address the impact of ME/CFS, including knowledgeable
55 physicians, information on existing benefits and support and help accessing them. Evidence
56 also suggested that advice on financial support was crucial for illness and life-management

1 of ME/CFS and social relationships. Continuing to work due to a lack of financial resources
2 or financial constraints and without support had physical consequences and negatively
3 influenced coping.

4
5 The committee considered that although the evidence supporting these findings was of low
6 quality, information and support from patient support groups can be invaluable to people with
7 ME/CFS. The committee was aware of unregulated sites that offered questionable advice
8 and claims of treatments for ME/CFS. The committee agreed the importance of making
9 people aware of the support available, so made recommendations to give people with
10 ME/CFS information about self-help groups, support groups and other local and national
11 resources and advice about accessing and applying for benefits.

12
13 The committee could not signpost to any one source about ME/CFS, but they noted that in
14 Evidence review B – Information for health and social care professionals, health care
15 professionals (HCPs) from specialist services report using information resources produced
16 by patient groups such as Action for ME or the ME Association when giving advice to people
17 diagnosed with ME/CFS. In addition, the committee acknowledged that information and
18 support for people with severe or very severe ME/CFS was limited and they were aware of
19 the 25% M.E group, a patient group providing advice for people with severe or very severe
20 ME/CFS.

21
22 *Social care support*

23 Evidence suggested that people with ME/CFS needed practical support for both themselves
24 and their carers, including help with personal and domestic tasks and access to social care,
25 lack of which impacted on their ability to manage their lives and maintain their family roles.

26
27 Support needs of families and carers

28
29 The committee discussed the impact of ME/CFS on other family members acknowledging
30 that it can affect all members of a family (including siblings) and all aspects of family life.
31 They noted the impact varied according to the symptoms and their severity, and could
32 fluctuate in alignment with the severity of ME/CFS. The committee noted there is a
33 psychological, physical and emotional impact on families and carers and described below
34 some of the ways they can be affected.

35 Family members might have to take on more (or all) responsibilities in order to allow the
36 person with ME/CFS to effectively manage their energy envelope. This may include:

- 37
- 38 • help with personal care - from providing a bowl of water to aid washing to helping with
 - 39 mobility and transfers to/from the bath; assisting to stand from the toilet and from
 - 40 lying to sitting to standing, etc
 - 41 • cooking and serving meals
 - 42 • household chores, such as cleaning
 - 43 • shopping
 - 44 • supporting medication use, such as prompting the person when to take medication or
 - 45 helping them prepare medications
 - 46 • providing information to HCPs and benefits assessors, where the person with
 - 47 ME/CFS has given consent, including any changes in functional ability or symptoms
 - 48 • supporting non-pharmacological interventions, such as passive movement and
 - 49 stretching
 - 50 • maintaining a safe and comfortable environment in and out of the home, such as low
- lighting and low noise

- 1 • planning ahead and prioritising activities and tasks, to reduce the burden on the
2 person with ME/CFS. This may include cancelling planned activities as circumstances
3 or symptoms change
- 4 • Wage earning
- 5 • Childcare including the school run - and the effect on relationship with the child if
6 they are unable to participate in activities.
- 7 • Driving - and especially night driving
- 8 • Attending medical appointments to help with getting to the appointment and
9 comprehending the information given.
- 10 In addition, feelings of guilt and stress about doing/not doing what is best; anxiety caused by
11 confusion and lack of understanding of the condition; tiredness from taking on extra
12 responsibilities, and lack of sleep, may all contribute to:

- 13 • stress, exhaustion, and an inability to function properly at work or in education,
14 • worry that they should be doing more, better, differently,
15 • inability or lack of desire to socialise and engage in 'normal' relationships,
16 • relationship breakdowns,
17 • isolation from wider family and friends,
18 • loss of income.

19 Siblings of a young person with ME/CFS will be particularly vulnerable to becoming
20 distracted from school, being tired and inattentive, missing out on opportunities because no-
21 one can support, or accompany them. They may stop bringing friends home because of
22 embarrassment of what friends might see or perceive. They are likely to worry about the
23 future burden on themselves of looking after their sibling when parents are no longer able to
24 do so. Their life choices are likely to be influenced by the impact ME/CFS is having on their
25 family situation.

26
27 Young children of someone with ME/CFS will experience the same impacts as siblings. In
28 addition, even where there is another parent in the house, will play their part as a young
29 carer. They may miss some of their education or not reach their potential.

30
31 The committee agreed that it was important that people and their families and carers were
32 aware of the support available to them. The committee considered that some people may be
33 reluctant to or have reservations about engaging with social care support services due to
34 previous disbelief about the severity of the illness and the level of impact on day-to-day
35 functioning. They noted there are sensitivities in this area, particularly with children and
36 young people and their families (see the discussion on safeguarding in evidence review B:
37 Information for health and social care professionals). The committee wanted to be clear that
38 these recommendations are about accessing support and health care professionals should
39 discuss sensitively with the person and their family and carers how social care may benefit
40 them. If the person decides they would like to access social care services, they and their
41 family and carers should be made aware how to self-refer for a social care assessment or if
42 they prefer a referral should be made by the health or social care professional. The
43 committee made recommendations sign posting to different assessments and support.
44 These included social care needs assessment, highlighting to parents and carers the
45 Children and Families Act 2014 and for young carers ,the Young Carers (Needs
46 Assessment) Regulations 2015.

47 In the committee's experience health and social care professionals were not always aware
48 about the support available to carers and families of people with ME/CFS and they
49 referenced the NICE guideline on supporting adult carers.

50

51

52 *Specialist ME/CFS services*

53

1 Evidence suggested a diagnosis of ME/CFS, often gained through private or alternative
2 health services, enabled patients to gain advice from health professionals and take action
3 towards symptom improvement while a lack of a diagnosis exacerbated psychological and
4 financial pressures and impeded access to social services. People with ME/CFS hoped to be
5 referred to specialist services to overcome the barriers to diagnosis and treatment they
6 encountered in primary care and those who ultimately were, had benefited in ways including
7 diagnosis, validation and information provision. (see Evidence review I: Multidisciplinary
8 care) The importance of receiving an accurate and timely diagnosis is a theme running
9 throughout several of the qualitative reviews in the guideline and specific recommendations
10 related to this are discussed in the Evidence review D:Diagnosis. The committee noted that
11 this finding highlighted the importance of support for people with suspected ME/CFS during
12 the pre-diagnostic stage. Recommendations for pre-diagnostic management are discussed in
13 the Evidence review E:Strategies pre diagnosis.

14

15 *Continuity of care*

16

17 Evidence suggested that people with ME/CFS need to establish a close, ongoing relationship
18 with their health professional who would be involved in the long-term management of
19 ME/CFS and accompany and advise them at all stages of the care process. The committee
20 noted that the theme of lack of continuity of care was identified as a barrier to care as well as
21 a support need and this fed in to discussion of the potential implications for people with
22 ME/CFS and the rationale for the recommendation to allocate a single point of contact to the
23 person with ME/CFS. see Evidence review C: Access to care and Evidence review I:
24 Multidisciplinary care.

25

26 *Advocacy and support with communication*

27

28 Evidence suggested that GP consultations were challenging for people with ME/CFS and
29 they could benefit from the presence of their caregiver. The committee discussed the
30 different ways in which GP consultations could be challenging, including articulating
31 symptoms and absorbing and remembering information. The committee considered that the
32 presence of a family member or care giver could offer support to the person with ME/CFS in
33 several ways. They can help to articulate the experiences of the person with ME/CFS, take in
34 or note down information which can later be relayed to the person if they are struggling with
35 concentration during the consultation, and ask questions as well as making the person with
36 ME/CFS feel less anxious and more confident. Therefore, a recommendation was made to
37 involve family members and carers (as appropriate) in discussions and care planning if the
38 person with ME/CFS chooses for them to be included. It was noted that people with ME/CFS
39 can appear quiet but are still able to take in information and they require respect from health
40 care professionals by being spoken to as well as their family members/carers. The
41 committee noted that as good practice health care professionals should check the person's
42 understanding of each consultation and offer a summary as appropriate to the person's
43 needs. The importance of accommodations in medical consultations also emerged in the
44 report Appendix 2 – People with severe ME/CFS, where people highlighted the importance of
45 more flexibility with appointments, home visits and the use of technology such as tele-
46 consults and emails which are rarely offered and appear to be an essential communication
47 tool to support people with severe ME/CFS.

48

49

50 Support needs of children and young people

51

52 *Being believed*

53

54 The theme of the need for validation was echoed in the evidence identified for children and
55 young people with ME/CFS. The negative impacts of having experienced disbelief and
56 judgement from medical professionals, as well as family members, peers and teachers were

1 particularly evident in this group. The committee discussed the possible reasons for this. It
2 was suggested that children/young people are continually entering different developmental
3 stages with changing behaviours that may be difficult to distinguish between 'normal' and
4 'illness' behaviors. Adults, on the other hand, are more likely to be able to explain their
5 symptoms in terms of their premorbid experiences and abilities. An example was given of a
6 teenager laying in bed for long periods of time, which could be interpreted as 'normal
7 teenage behaviour', but for an adult who has always gone to bed and risen at the same time
8 for several years, laying in bed for long periods of time would be more likely to be viewed as
9 uncharacteristic and a potential indicator of illness. The committee considered it to be
10 important that children and young people, are believed and listened to. This supported the
11 recommendation to ensure that the voice of the child or young person is heard by taking a
12 child-centered approach and discussing how they want to be involved in making decisions
13 about their care. The committee also considered that health professionals should be aware
14 that children and young people with ME/CFS may have had negative experiences and to
15 approach communication with the child or young person with sensitivity. A recommendation
16 was made to raise awareness that children and young people may have experienced
17 significant emotional impact from social stigma, disbelief and delay in receiving a diagnosis
18 and this may result in a hesitation to engage in health and social care services.

19

20 *Education and training*

21

22 Evidence suggested that lack of support within young people's educational environment was
23 a source of anxiety and low mood, while better communication between healthcare and
24 education providers enabled patients and carers to gain support and accommodate their
25 needs. Despite the low quality of the evidence supporting the finding, the committee
26 considered it very important as in their experience overall there is currently very little support
27 available for young people and children with ME/CFS in supporting their educational needs.
28 They noted that support from individual education providers is highly variable.

29

30 The committee discussed the potential consequences of a lack of support and understanding
31 from education providers. They were aware that without alternative education options and
32 adaptations some children and young people may leave education with potentially negative
33 consequences on their future. It was highlighted by the committee that there is a legal
34 responsibility to ensure children receive an education, but this does not mean that they must
35 attend a school.

36

37 The committee acknowledged that schools and colleges are high stimulus environments with
38 high physical, cognitive and emotional demands and time in school results in a high level of
39 energy expenditure. The committee agreed that communication between health and social
40 care professionals and training and education services is key to enable understanding of the
41 needs and impairments of people with ME/CFS and provision of appropriate educational
42 support. Therefore, the committee recommended that health and social care professionals
43 should work and communicate closely with social care, training and education services to
44 enable them to understand the needs and impairments of children and young people with
45 ME/CFS, ensure a common understanding of the aims of the person with ME/CFS and
46 discuss a flexible approach to training and education.

47

48 The committee discussed the adjustments and adaptations that could support someone in
49 education or training. These should be planned with the person with ME/CFS and their
50 families and health, social care and education services. The committee noted that ME/CFS
51 can be an invisible condition, people with ME/CFS may appear well and it can be difficult for
52 people to understand that adaptations are needed. They noted that children and young people
53 as a rule want to fit in and not be singled out from their peers. This can result in children and
54 young people overexerting themselves.

55

1 All plans for adjustments and adaptations need to be flexible and may need adjusting
2 frequently, potentially daily and especially at the beginning and during post exacerbations.
3 Plans range from reduced hours and workload with a reduced curriculum, excluding physical
4 education and sports sessions. Core subjects may be prioritised and then subjects the child
5 or young person enjoys. All study time should be time led and not task led, homework
6 reduced and prioritized with extensions. Assistance and time extensions for exams are
7 important to consider. As noted schools and colleges are high stimulus environments and
8 regular rest breaks through day with a quiet designated area, use of time out cards, physical
9 support by keeping classrooms close together, use of a lift pass, use of wheelchairs and
10 some work at home online are useful adaptations. In some children and young people with
11 ME/CFS there may be times (for example, when symptoms are worse) when a move to
12 online and home schooling may be required.

13
14 There are also circumstances where education should be completely suspended for a period
15 of time, as the person may not have the capacity for any form of education, for example
16 during periods of severe illness. The committee noted that any return to education should
17 tailored to the individual's circumstances and be carefully monitored to avoid any flares or
18 relapse.

19
20 The committee noted that education, health and care plans for people up to the age of 25
21 years can be requested by parents, school or local authorities, but that many people are
22 unaware of the availability of these plans. The committee noted these are legal documents
23 outlining the support a child or young person will receive to meet their special needs across
24 education, health and social care. The committee agreed that parents and carers need to be
25 provided with this information and made a recommendation to give parents and carers
26 information on about education, health and care plans and how to request one from their
27 local authority.

28
29 The committee considered that although education is an important aspect of daily living,
30 there are other important aspects such as home and social activities. The committee agreed
31 that it was important that children and young people have a balance between time spent on
32 education, or training, home and family life, and social activities decided to highlight this in
33 the recommendations.

34 The committee discussed the lack of understanding about ME/CFS in children and young
35 people and reduced or non-attendance from school can result in safeguarding concerns
36 being raised and this is discussed in Evidence review B – Information for health and social
37 care professionals.

38 39 Access to care

40
41 Evidence suggested that hospital care tailored to individual needs, symptom severity and
42 other commitments, in terms of accessibility and flexibility in the frequency, duration and
43 mode of attendance to medical appointments was valued by people with ME/CFS. Themes
44 of individualized care and flexibility around the frequency, duration and mode of medical
45 appointments ran throughout several of the qualitative reviews and are discussed along with
46 the recommendations elsewhere (Evidence review C: Access to care)

47 48 Information and education needs of families and carers

49
50 The committee noted there was limited evidence directly referring to the information,
51 education and support needs of families and carers. The committee discussed this was an
52 important area and they were aware that some family members and friends, particularly
53 when someone first has symptoms, can find it difficult to understand ME/CFS and the impact
54 of the symptoms. This was reflected in the evidence with carers reporting they could be
55 frustrated by the symptoms people had and lacked an understanding of the illness. They

1 recognised the danger of giving inappropriate advice or support, exacerbating symptoms or
2 pushing the person with ME/CFS to do too much too soon. This is also supported by the
3 Children and young people's report, where symptoms were reported to worsen where people
4 had been given unhelpful advice (by health-care professionals) such as to go out every day
5 despite not feeling good enough to do so. The report highlights how being pushed and not
6 being given time to rest or recover from symptoms as a result of other people's unrealistic
7 expectations, including people from the family and school of the person with ME/CFS, could
8 act against effective management.

9
10 The committee considered that families and carers benefit from the opportunity to obtain
11 accurate information about the condition and about ways that they can help the person with
12 ME/CFS and therefore made a recommendation to that they should be given this information.
13 The committee agreed that the sections of the guideline on Principles of care for people with
14 ME/CFS and Information and support provided valuable information for families and carers of
15 people with ME/CFS. In the information and support section of the guideline the
16 recommendations include providing information, where appropriate, to families and carers,
17 not only people with ME/CFS.

18 The committee noted that information leaflets for carers do exist, but it is unclear whether
19 these have been formally evaluated and therefore there may be a risk of misinformation.
20 The committee were aware of unregulated sites that offered questionable advice and claims
21 of treatments for ME/CFS. As with the information for people with ME/CFS the committee
22 could not signpost to any one source about ME/CFS, but they noted that in Evidence review
23 B – Information for health and social care professionals. HCPs from specialist services,
24 report using information resources produced by patient groups such as Action for ME or the
25 ME Association when giving advice to people diagnosed with ME/CFS. In addition, the
26 committee acknowledged that information and support for people with severe ME/CFS was
27 limited and they were aware of the 25% M.E group, a patient group providing advice for
28 people with severe ME/CFS.
29

30 In the context of families and carers the committee acknowledged that the recommendations
31 highlighting the importance of advocacy and support with communication for people with
32 ME/CFS also support family members and carers. Some committee members were aware of
33 parents that had felt there were occasions when had been perceived as 'pushy' or 'over
34 anxious'. They were wary of their actions being misinterpreted and as a result cautious in
35 how they pursued support for their child. Some parents felt there was an underlying fear that
36 they may be accused of maltreatment or abuse if they are persistent in advocating for their
37 child. Safeguarding is discussed in detail in Evidence review B – Information for health and
38 social care professionals
39

40 **2.3 Cost effectiveness and resource use**

41 Cost effectiveness evidence was not sought as this was a qualitative review. The
42 recommendations generally provide guidance regarding the content of information and
43 support specific to people with ME/CFS and their families in line with the general principles of
44 provision of information already established in the existing NICE Patient Experience
45 Guideline and so were not considered likely to have a substantial resource impact over and
46 above this.

47

1 Appendix A – Review protocols

2 Review protocol for information, education and support for people with suspected and/or diagnosed ME/CFS, their families and 3 carers

ID	Field	Content
0.	PROSPERO registration number	CRD42019152083
1.	Review title	What information, education and support do people with/suspected of having ME/CFS and their families and carers need?
2.	Review question	What information, education and support do people with/suspected of having ME/CFS and their families and carers need?
3.	Objective	To identify the information, education and support required, as identified by people with/suspected of having ME/CFS and their families and carers.
4.	Searches	<p>The following databases will be searched:</p> <ul style="list-style-type: none"> • Embase • MEDLINE • CINAHL • PsychINFO <p>Searches will be restricted by:</p> <ul style="list-style-type: none"> • English language

		<p>The searches may be re-run 6 weeks before the final committee meeting and further studies retrieved for inclusion if relevant.</p> <p>The full search strategies will be published in the final review</p>
5.	Condition or domain being studied	ME / CFS
6.	Population	Adults, children and young people who are diagnosed with ME/CFS, or who are suspected of having ME/CFS by their primary clinician and their families and carers.
7.	Intervention/Exposure/Test	Information, education and support that patients, their families and carers require.
8.	Comparator/Reference standard/Confounding factors	NA
9.	Types of study to be included	Qualitative studies (e.g. transcript data collected from focus groups / semi structured interviews)
10.	Other exclusion criteria	Exclusion: Quantitative studies (i.e. closed questionnaire surveys)
11.	Context	N/A
12.	Primary outcomes (critical outcomes)	Themes emerging from qualitative data

13.	Secondary outcomes (important outcomes)	Not applicable
14.	Data extraction (selection and coding)	<p>EndNote will be used for reference management, sifting, citations and bibliographies. All references identified by the searches and from other sources will be screened for inclusion. 10% of the abstracts will be reviewed by two reviewers, with any disagreements resolved by discussion or, if necessary, a third independent reviewer.</p> <p>The full text of potentially eligible studies will be retrieved and will be assessed in line with the criteria outlined above.</p> <p>A standardised form will be used to extract information from studies (see Developing NICE guidelines: the manual section 6.4).</p> <p>Additional qualitative studies will be added to the review until themes within the analysis become saturated; i.e. studies will only be included if they contribute towards the development of existing themes or to the development of new themes. The point at which data saturation is reached will be noted within the review.</p>
15.	Risk of bias (quality) assessment	<p>Risk of bias will be assessed using the appropriate checklist as described in Developing NICE guidelines: the manual:</p> <p>For this review the CASP qualitative checklist will be used to assess risk of bias of individual studies.</p> <p>A sample of 10% of the critical appraisals will be quality assured by a second reviewer. Disagreements between the review authors over the risk of bias in particular studies will be resolved by discussion, with involvement of a third review author where necessary.</p>
16.	Strategy for data synthesis	<p>The synthesis of qualitative data will follow a thematic analysis approach. Information will be synthesised into main review findings. Results will be presented in a detailed narrative and in table format with summary statements of main review findings.</p>

		GRADE CERQual will be used to synthesise the qualitative data and assess the certainty of evidence for each review finding.
17.	Analysis of sub-groups	<p>Stratification:</p> <ul style="list-style-type: none"> • Children/young people vs. adults • People with severe ME/ less severe ME (as defined by the studies)
18.	Type and method of review	<input type="checkbox"/> Intervention <input type="checkbox"/> Diagnostic <input type="checkbox"/> Prognostic <input checked="" type="checkbox"/> Qualitative <input type="checkbox"/> Epidemiologic <input type="checkbox"/> Service Delivery <input type="checkbox"/> Other (please specify)
19.	Language	English
20.	Country	England
21.	Anticipated or actual start date	01/05/19
22.	Anticipated completion date	01/03/20

23.	Stage of review at time of this submission	Review stage	Started	Completed
		Preliminary searches	<input type="checkbox"/>	<input checked="" type="checkbox"/>
		Piloting of the study selection process	<input type="checkbox"/>	<input checked="" type="checkbox"/>
		Formal screening of search results against eligibility criteria	<input type="checkbox"/>	<input type="checkbox"/>
		Data extraction	<input type="checkbox"/>	<input type="checkbox"/>
		Risk of bias (quality) assessment	<input type="checkbox"/>	<input type="checkbox"/>

		Data analysis	<input type="checkbox"/>	<input type="checkbox"/>
24.	Named contact	<p>5a. Named contact National Guideline Centre</p> <p>5b Named contact e-mail CFSME@nice.org.uk</p> <p>5e Organisational affiliation of the review National Institute for Health and Care Excellence (NICE) and the National Guideline Centre</p>		
25.	Review team members	<p>From the National Guideline Centre:</p> <ul style="list-style-type: none"> • Dr Kate Kelley [Guideline lead] • Ms Maria Smyth [Senior systematic reviewer] • Ms Melina Vasileiou [Systematic reviewer] • Dr Richard Clubbe [Systematic reviewer] • Dr Karin van Bart [Systematic reviewer] • Mr David Wonderling [Health economist] • Ms Agnes Cuyas [Information specialist] • Ms Kate Ashmore [Project manager] 		
26.	Funding sources/sponsor	This systematic review is being completed by the National Guideline Centre which receives funding from NICE.		
27.	Conflicts of interest	All guideline committee members and anyone who has direct input into NICE guidelines (including the evidence review team and expert witnesses) must declare any potential conflicts of interest in line with NICE's code of practice for declaring and dealing with conflicts of interest. Any relevant interests, or changes to interests, will also be declared publicly at the start of each guideline committee meeting. Before each meeting, any potential conflicts of interest will be considered by the guideline committee		

		Chair and a senior member of the development team. Any decisions to exclude a person from all or part of a meeting will be documented. Any changes to a member's declaration of interests will be recorded in the minutes of the meeting. Declarations of interests will be published with the final guideline.
28.	Collaborators	Development of this systematic review will be overseen by an advisory committee who will use the review to inform the development of evidence-based recommendations in line with section 3 of Developing NICE guidelines: the manual . Members of the guideline committee are available on the NICE website: [NICE guideline webpage].
29.	Other registration details	N/A
30.	Reference/URL for published protocol	[Give the citation and link for the published protocol, if there is one.]
31.	Dissemination plans	<p>NICE may use a range of different methods to raise awareness of the guideline. These include standard approaches such as:</p> <p>Notifying registered stakeholders of publication</p> <p>Publicising the guideline through NICE's newsletter and alerts</p> <p>Issuing a press release or briefing as appropriate, posting news articles on the NICE website, using social media channels, and publicising the guideline within NICE.</p>
32.	Keywords	Patients experience, information
33.	Details of existing review of same topic by same authors	N/A
34.	Current review status	<input checked="" type="checkbox"/> Ongoing

		<input type="checkbox"/> Completed but not published <input type="checkbox"/> Completed and published <input type="checkbox"/> Completed, published and being updated <input type="checkbox"/> Discontinued
35..	Additional information	N/A
36.	Details of final publication	www.nice.org.uk

1

2

1 Appendix B – Literature search strategies

2 This literature search strategy was used for the following review question:

- 3 • What information, education and support do people with ME/CFS and their families and
4 carers need?

5 The literature searches for this review are detailed below and complied with the methodology
6 outlined in Developing NICE guidelines: the manual.⁹²

7 For more information, please see the Methodology review published as part of the
8 accompanying documents for this guideline.

B.1.9 Clinical search literature search strategy

10 Searches were constructed using a PICO framework where population (P) terms were
11 combined with Intervention (I) and in some cases Comparison (C) terms. Outcomes (O) are
12 rarely used in search strategies for interventions as these concepts may not be well
13 described in title, abstract or indexes and therefore difficult to retrieve.

14 Searches for patient views were run in Medline (OVID), Embase (OVID), CINAHL, and
15 PsycINFO (ProQuest).

16 **Table 4: Database date parameters and filters used**

Database	Dates searched	Search filter used
Medline (OVID)	1946 – 23 June 2020	Exclusions
Embase (OVID)	1974 – 23 June 2020	Exclusions
The Cochrane Library (Wiley)	Cochrane Reviews to 2020 Issue 6 of 12 CENTRAL to 2020 Issue 6 of 12	None
CINAHL, Current Nursing and Allied Health Literature (EBSCO)	Inception – 23 June 2020	None
PsycINFO (ProQuest)	Inception – 23 June 2020	Exclusions
Epistemonikos (The Epistemonikos Foundation)	Inception - 23 June 2020	None

17 Medline (Ovid) search terms

1.	Fatigue Syndrome, Chronic/
2.	chronic* fatigue*.ti,ab.
3.	((fatigue* adj2 (disorder* or syndrome* or post viral or postviral or immune dysfunction* or post infection* or postinfection*)).ti,ab.
4.	((myalgic or post infection* or postinfection*) adj (encephalomyelitis or encephalopathy)).ti,ab.
5.	((ME adj CFS) or (CFS adj ME) or CFIDS or PVFS).ti,ab.
6.	(Systemic Exertion Intolerance Disease or SEID).ti,ab.
7.	((CFS adj SEID) or (SEID adj CFS) or (ME adj CFS adj SEID) or (ME adj SEID) or (SEID adj ME)).ti,ab.
8.	((Orthostatic intolerance or postural orthostatic tachycardia syndrome or postural tachycardia syndrome or POTS) adj6 (CFS or chronic* fatigue* or ME or myalgic or SEID or systemic exertion)).ti,ab.

9.	((Post-exertional or postexertional) adj2 malaise).ti,ab.
10.	(neurasthenic neuroses or epidemic neuromyasthenia or neurataxia or neuroasthenia or neurasthenia).ti,ab.
11.	((atypical or simulating or resembling) adj poliomyelitis).ti,ab.
12.	((chronic adj2 epstein Barr virus) or CEBV or CAEBV or chronic mononucleosis).ti,ab.
13.	xenotropic murine leukemia virus-related virus.ti,ab.
14.	effort syndrome*.ti,ab.
15.	((akureyri or iceland or tapanui or royal free or royal free hospital) adj disease*) or ((yuppie or yuppy or tapanui) adj flu)).ti,ab.
16.	or/1-15
17.	letter/
18.	editorial/
19.	news/
20.	exp historical article/
21.	Anecdotes as Topic/
22.	comment/
23.	case report/
24.	(letter or comment*).ti.
25.	or/17-24
26.	randomized controlled trial/ or random*.ti,ab.
27.	25 not 26
28.	animals/ not humans/
29.	exp Animals, Laboratory/
30.	exp Animal Experimentation/
31.	exp Models, Animal/
32.	exp Rodentia/
33.	(rat or rats or mouse or mice).ti.
34.	or/27-33
35.	16 not 34
36.	limit 35 to English language

1 Embase (Ovid) search terms

1.	chronic fatigue syndrome/
2.	chronic* fatigue*.ti,ab.
3.	(fatigue* adj2 (disorder* or syndrome* or post viral or postviral or immune dysfunction* or post infection* or postinfection*)).ti,ab.
4.	((myalgic or post infection* or postinfection*) adj (encephalomyelitis or encephalopathy)).ti,ab.
5.	((ME adj CFS) or (CFS adj ME) or CFIDS or PVFS).ti,ab.
6.	(Systemic Exertion Intolerance Disease or SEID).ti,ab.
7.	((CFS adj SEID) or (SEID adj CFS) or (ME adj CFS adj SEID) or (ME adj SEID) or (SEID adj ME)).ti,ab.
8.	((Orthostatic intolerance or postural orthostatic tachycardia syndrome or postural tachycardia syndrome or POTS) adj6 (CFS or chronic* fatigue* or ME or myalgic or SEID or systemic exertion)).ti,ab.
9.	((Post-exertional or postexertional) adj2 malaise).ti,ab.
10.	(neurasthenic neuroses or epidemic neuromyasthenia or neurataxia or neuroasthenia or neurasthenia).ti,ab.

11.	((atypical or simulating or resembling) adj poliomyelitis).ti,ab.
12.	((chronic adj2 epstein Barr virus) or CEBV or CAEBV or chronic mononucleosis).ti,ab.
13.	xenotropic murine leukemia virus-related virus.ti,ab.
14.	effort syndrome*.ti,ab.
15.	((akureyri or iceland or tapanui or royal free or royal free hospital) adj disease*) or ((yuppie or yuppy or tapanui) adj flu)).ti,ab.
16.	or/1-15
17.	letter.pt. or letter/
18.	note.pt.
19.	editorial.pt.
20.	case report/ or case study/
21.	(letter or comment*).ti.
22.	or/17-21
23.	randomized controlled trial/ or random*.ti,ab.
24.	22 not 23
25.	animal/ not human/
26.	nonhuman/
27.	exp Animal Experiment/
28.	exp Experimental Animal/
29.	animal model/
30.	exp Rodent/
31.	(rat or rats or mouse or mice).ti.
32.	or/24-31
33.	16 not 32
34.	limit 33 to English language

1 Cochrane Library (Wiley) search terms

#1.	MeSH descriptor: [Fatigue Syndrome, Chronic] this term only
#2.	chronic* fatigue*.ti,ab
#3.	(fatigue* near/2 (disorder* or syndrome* or post viral or postviral or immune dysfunction* or post infection* or postinfection*)):ti,ab
#4.	((myalgic or post infection* or postinfection*) near/1 (encephalomyelitis or encephalopathy)):ti,ab
#5.	((ME near/1 CFS) or (CFS near/1 ME) or CFIDS or PVFS):ti,ab
#6.	(Systemic Exertion Intolerance Disease or SEID):ti,ab
#7.	((CFS near/1 SEID) or (SEID near/1 CFS) or (ME near/1 CFS near/1 SEID) or (ME near/1 SEID) or (SEID near/1 ME)):ti,ab
#8.	(Orthostatic intolerance or postural orthostatic tachycardia syndrome or postural tachycardia syndrome or POTS)
#9.	((Post-exertional or postexertional) near/2 malaise):ti,ab
#10.	(neurasthenic neuroses or epidemic neuromyasthenia or neurataxia or neuroasthenia or neurasthenia):ti,ab
#11.	((atypical or simulating or resembling) near/1 poliomyelitis):ti,ab
#12.	((chronic epstein Barr virus) or CEBV or CAEBV or chronic mononucleosis):ti,ab
#13.	xenotropic murine leukemia virus-related virus:ti,ab
#14.	effort syndrome*:ti,ab
#15.	((akureyri or iceland or tapanui or "royal free" or "royal free hospital") near/1 disease*):ti,ab

#16.	((yuppie or yuppy or tapanui) near flu):ti,ab
#17.	(or #1-#16)

1 CINAHL (EBSCO) search terms

S1.	(MH "Fatigue Syndrome, Chronic")
S2.	chronic* fatigue*
S3.	(fatigue* n2 (disorder* or syndrome* or post viral or postviral or immune dysfunction* or post infection* or postinfection*))
S4.	((myalgic or post infection* or postinfection*) and (encephalomyelitis or encephalopathy))
S5.	((ME and CFS) or (CFS and ME) or CFIDS or PVFS)
S6.	(Systemic Exertion Intolerance Disease or SEID)
S7.	((CFS and SEID) or (SEID and CFS) or (ME and CFS and SEID) or (CFS and ME and SEID) or (ME and SEID) or (SEID and ME))
S8.	((Orthostatic intolerance or postural orthostatic tachycardia syndrome or postural tachycardia syndrome) and (CFS or chronic* fatigue* or ME or myalgic or SEID or systemic exertion))
S9.	((Post-exertional or postexertional) n2 malaise)
S10.	(neurasthenic neuroses or epidemic neuromyasthenia or neurataxia or neuroasthenia)
S11.	((atypical or simulating or resembling) and poliomyelitis)
S12.	(chronic epstein Barr virus or chronic mononucleosis)
S13.	xenotropic murine leukemia virus-related virus
S14.	effort syndrome*
S15.	((akureyri or iceland or tapanui or royal free or royal free hospital) and disease*) or ((yuppie or yuppy or tapanui) and flu))
S16.	S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15

2 PsycINFO (ProQuest) search terms

1.	(((chronic* fatigue*) OR (fatigue* NEAR2 (disorder* OR syndrome* OR post viral OR postviral OR immune dysfunction* OR post infection* OR postinfection*)) OR ((myalgic OR post infection* OR postinfection*) NEAR1 (encephalomyelitis OR encephalopathy)) OR ((ME NEAR1 CFS) OR (CFS NEAR1 ME) OR CFIDS OR PVFS) OR (Systemic Exertion Intolerance Disease OR SEID) OR ((CFS NEAR1 SEID) OR (SEID NEAR1 CFS)) OR ((ME NEAR1 CFS NEAR1 SEID) OR (ME NEAR1 SEID) OR (SEID NEAR1 ME)) OR ((Orthostatic intolerance OR postural orthostatic tachycardia syndrome OR postural tachycardia syndrome OR POTS) NEAR6 (CFS OR chronic* fatigue* OR ME OR myalgic OR SEID OR systemic exertion)) OR (neurasthenic neuroses OR epidemic neuromyasthenia OR neurataxia OR neuroasthenia OR neurasthenia) OR ((atypical OR simulating OR resembling) NEAR1 poliomyelitis)) OR (((chronic NEAR2 epstein Barr virus) OR CEBV OR CAEBV OR chronic mononucleosis) OR (xenotropic murine leukemia virus-related virus) OR (effort syndrome*)) OR ((akureyri OR iceland OR tapanui OR royal free OR royal free hospital) NEAR1 disease*) OR ((yuppie OR yuppy OR tapanui) NEAR1 flu) OR MAINSUBJECT.EXACT.EXPLODE("Chronic Fatigue Syndrome")) AND (stpe.exact("Scholarly Journals") AND la.exact("ENG") AND po.exact("Human") NOT (me.exact("Empirical Study" OR "Quantitative Study" OR "Longitudinal Study" OR "Clinical Trial" OR "Qualitative Study" OR "Prospective Study" OR "Followup Study" OR "Literature Review" OR "Retrospective Study" OR "Systematic Review" OR "Meta Analysis") AND po.exact("Human"))
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3 Epistemonikos search terms

1.	(advanced_title_en:((advanced_title_en:((chronic* fatigue* syndrome*) OR (fatigue* syndrome* OR fatigue* disorder* OR postviral fatigue* OR post viral fatigue* OR fatigue* immune dysfunction OR post infection fatigue* OR postinfection fatigue*)) OR (encephalomyelitis OR encephalopathy) OR ("ME/CFS" OR "CFS/ME" OR "CFIDS"
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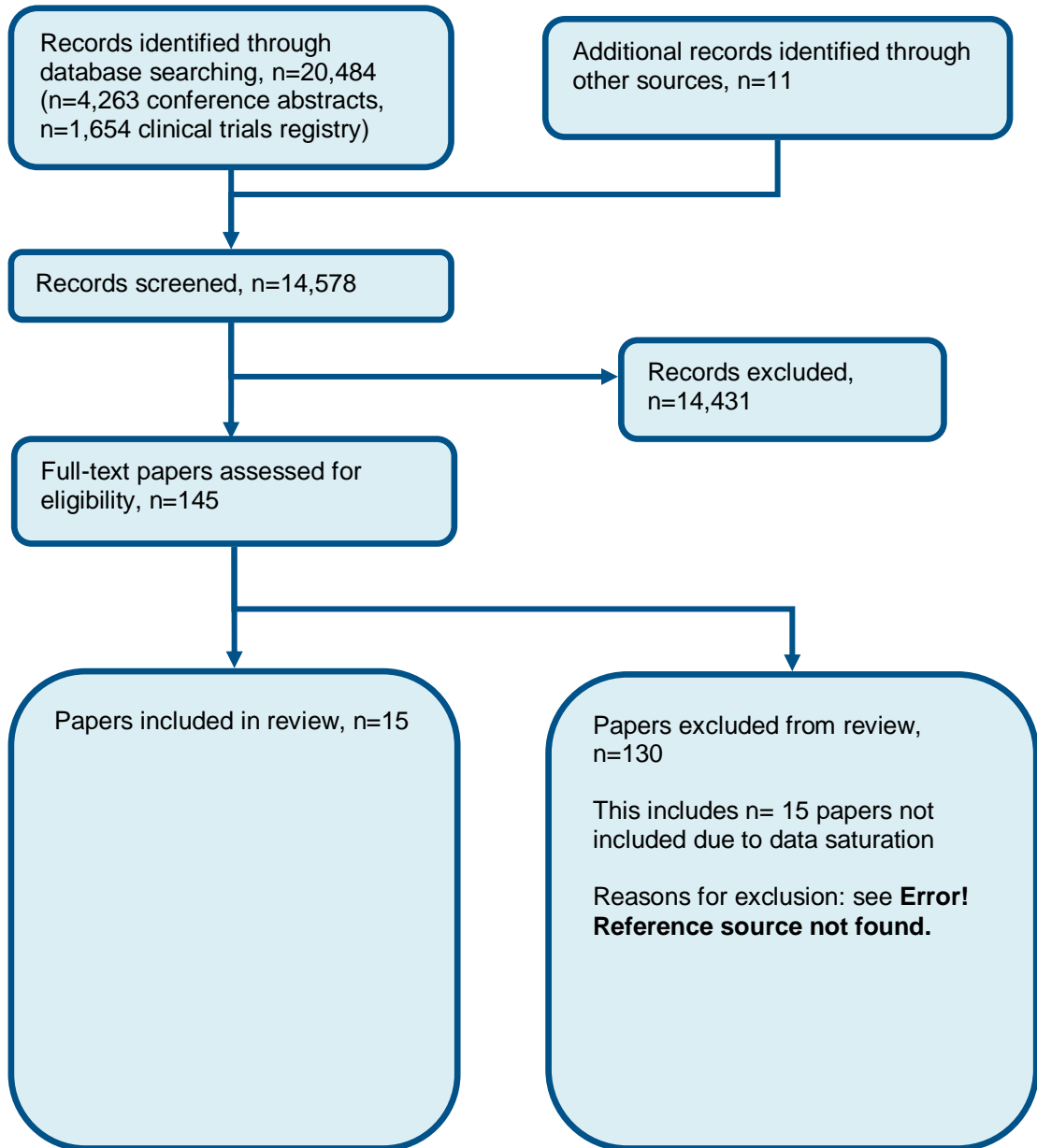
	<p>OR "PVFS") OR (Systemic Exertion Intolerance Disease OR SEID) OR ((CFS AND SEID) OR (SEID AND CFS) OR (ME AND CFS AND SEID) OR (ME AND SEID) OR (SEID AND ME)) OR (Orthostatic intolerance OR postural orthostatic tachycardia syndrome OR postural tachycardia syndrome OR POTS) OR ((Post-exertional OR postexertional) AND malaise) OR (neurasthenic neuroses OR epidemic neuromyasthenia OR neurataxia OR neuroasthenia OR neurasthenia) OR (atypical poliomyelitis OR simulating poliomyelitis OR resembling poliomyelitis) OR (chronic epstein Barr virus OR CEBV OR CAEBV OR chronic mononucleosis) OR (xenotropic murine leukemia virus-related virus) OR (effort syndrome*) OR (akureyri OR iceland disease OR tapanui OR royal free disease) OR (yuppie flu OR yuppy flu OR tapanui flu)) OR advanced_abstract_en:((chronic* fatigue* syndrome*) OR (fatigue* syndrome* OR fatigue* disorder* OR postviral fatigue* OR post viral fatigue* OR fatigue* immune dysfunction OR post infection fatigue* OR postinfection fatigue*)) OR (encephalomyelitis OR encephalopathy) OR ("ME/CFS" OR "CFS/ME" OR "CFIDS" OR "PVFS") OR (Systemic Exertion Intolerance Disease OR SEID) OR ((CFS AND SEID) OR (SEID AND CFS) OR (ME AND CFS AND SEID) OR (ME AND SEID) OR (SEID AND ME)) OR (Orthostatic intolerance OR postural orthostatic tachycardia syndrome OR postural tachycardia syndrome OR POTS) OR ((Post-exertional OR postexertional) AND malaise) OR (neurasthenic neuroses OR epidemic neuromyasthenia OR neurataxia OR neuroasthenia OR neurasthenia) OR (atypical poliomyelitis OR simulating poliomyelitis OR resembling poliomyelitis) OR (chronic epstein Barr virus OR CEBV OR CAEBV OR chronic mononucleosis) OR (xenotropic murine leukemia virus-related virus) OR (effort syndrome*) OR (akureyri OR iceland disease OR tapanui OR royal free disease) OR (yuppie flu OR yuppy flu OR tapanui flu)))) OR advanced_abstract_en:((advanced_title_en:((chronic* fatigue* syndrome*) OR (fatigue* syndrome* OR fatigue* disorder* OR postviral fatigue* OR post viral fatigue* OR fatigue* immune dysfunction OR post infection fatigue* OR postinfection fatigue*)) OR (encephalomyelitis OR encephalopathy) OR ("ME/CFS" OR "CFS/ME" OR "CFIDS" OR "PVFS") OR (Systemic Exertion Intolerance Disease OR SEID) OR ((CFS AND SEID) OR (SEID AND CFS) OR (ME AND CFS AND SEID) OR (ME AND SEID) OR (SEID AND ME)) OR (Orthostatic intolerance OR postural orthostatic tachycardia syndrome OR postural tachycardia syndrome OR POTS) OR ((Post-exertional OR postexertional) AND malaise) OR (neurasthenic neuroses OR epidemic neuromyasthenia OR neurataxia OR neuroasthenia OR neurasthenia) OR (atypical poliomyelitis OR simulating poliomyelitis OR resembling poliomyelitis) OR (chronic epstein Barr virus OR CEBV OR CAEBV OR chronic mononucleosis) OR (xenotropic murine leukemia virus-related virus) OR (effort syndrome*) OR (akureyri OR iceland disease OR tapanui OR royal free disease) OR (yuppie flu OR yuppy flu OR tapanui flu)) OR advanced_abstract_en:((chronic* fatigue* syndrome*) OR (fatigue* syndrome* OR fatigue* disorder* OR postviral fatigue* OR post viral fatigue* OR fatigue* immune dysfunction OR post infection fatigue* OR postinfection fatigue*)) OR (encephalomyelitis OR encephalopathy) OR ("ME/CFS" OR "CFS/ME" OR "CFIDS" OR "PVFS") OR (Systemic Exertion Intolerance Disease OR SEID) OR ((CFS AND SEID) OR (SEID AND CFS) OR (ME AND CFS AND SEID) OR (ME AND SEID) OR (SEID AND ME)) OR (Orthostatic intolerance OR postural orthostatic tachycardia syndrome OR postural tachycardia syndrome OR POTS) OR ((Post-exertional OR postexertional) AND malaise) OR (neurasthenic neuroses OR epidemic neuromyasthenia OR neurataxia OR neuroasthenia OR neurasthenia) OR (atypical poliomyelitis OR simulating poliomyelitis OR resembling poliomyelitis) OR (chronic epstein Barr virus OR CEBV OR CAEBV OR chronic mononucleosis) OR (xenotropic murine leukemia virus-related virus) OR (effort syndrome*) OR (akureyri OR iceland disease OR tapanui OR royal free disease) OR (yuppie flu OR yuppy flu OR tapanui flu))))))</p>
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1 Appendix C Qualitative evidence study selection

Figure 3: Flow chart of qualitative study selection for the review of information, education and support for people with ME/CFS, their families and carers



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1 Appendix D – Qualitative evidence

2

Study	Bayliss 2016 ¹⁵
Aim	To explore the extent to which ‘CFS/ME’ training and resources can be implemented in routine primary care, leading to a better understanding of the barriers and facilitators to the adoption and integration of new practices associated with medically unexplained conditions.
Population	<p>GPs from practices from seven PCTs in North West England who were given access to an online ‘CFS/ME’ training module (hosted by the Royal College of General Practitioners RCGP website) that involved patient resource packs for use in consultation with new and existing ‘CFS/ME’ patients, who had completed training.</p> <p>Patients recruited from participating GP practices. Searches of GP practice databases were conducted by the research team to identify individuals with an existing diagnosis of ‘CFS/ME’. GPs were asked to review these lists and to exclude patients with other conditions, or other factors that may account for their fatigue.</p> <p>Patients (n=11), mean age (range): 46 (27 to 71) years; GPs (n=8)</p>
Setting	Primary care
Study design	Qualitative interview study
Methods and analysis	<p>Individual face-to-face semi-structured interviews were conducted in the participants’ home, using topic guides that had been developed from a review of the literature and research team (including patient and carer research partners) discussions. Patient interviews focused on their views of ‘CFS/ME’ patient resource and their experience with their GP before and after practice had access to the online training.</p> <p>Interviews were digitally recorded and transcribed verbatim. Analysis was conducted in parallel with the interviews and was inductive, using components of thematic analysis. Transcripts were read, annotated and categorised independently by researchers of different professional backgrounds and patient and carer research partners. A further theory-driven analysis of the data guided by four main constructs of NPT was conducted separately by all researchers and the final analysis was agreed through discussion.</p>
Findings	<p>Support from GPs</p> <p>a) Need to be believed: When reflecting on the role of their GP, patients wanted to be believed. Patients with varying severity and time since diagnosis described how the provision of reliable evidence based information meant that their GP was validating their ‘CFS/ME’; this enabled them to self-manage their condition.</p>

Study	Bayliss 2016¹⁵
	<p>b) Need for a positive diagnosis: When reflecting on the role of their GP, patients wanted to receive a positive diagnosis</p> <p>c) Availability & involvement: They also wanted their GP to be accessible and actively involved in the longer term management of their condition. Where support was not received, patients reported disengaging from primary care.</p>
	Information resources for patients, families & carers
	<p>a) Online resources: DVD case studies were seen as particularly important in helping patients and carers to understand that others shared their experiences, and the format allowed those who found it difficult to read to access the information. As a result of this information some patients felt that they needed to visit their practice less frequently. Patients suggested that resources should be made available online and therefore accessible to all. Patients suggested websites that they believed would be useful for the resources to be linked to in order to be easily accessible. These included NHS Choices website, YouTube or HealthTalk on-line and RCGP.</p> <p>b) Evidence-based information pack: The resources were also reported to have had an impact on the friends, family and colleagues of the patients interviewed. In some cases, the provision of evidence based information improved relationships and strengthened support networks. Patients stated the resource pack would be of greatest benefit to newly diagnosed patients. However, a number of patients who had the condition for a number of years reported that a comprehensive pack of information allowed them to consolidate their knowledge and sometimes learn something new. An evidence-based resource of information was welcomed as there are currently issues with identifying reliable information on the internet.</p> <p>c) Information from reliable sources: Some patients were concerned that by placing the resources online, GPs would be let off managing the condition in primary care and sceptical attitudes would continue. Patients therefore reported that they wished to bring information from the internet to the consultation in order to gain a diagnosis from a health professional. Participants had done this in the past, and GPs welcomed this where information was from a reliable source.</p>
	Referral to specialist services: Patients wanted more access to specialist services, with some recognising that GPs didn't have the time to manage their condition.
Limitations and applicability of evidence	<p>Very minor limitations due to the role of the researcher not being discussed.</p> <p>No concerns over applicability</p>

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Study	Beasant 2014¹⁸
Aim	To understand the experiences of adolescents and families in accessing and using a specialist service, and explore whether or not they value referral to a specialist service for young people with 'CFS/ME'
Population	Mothers and adolescents diagnosed with 'CFS/ME' by paediatric 'CFS/ME' specialist service (aged 12-18 years, mildly or moderately affected i.e. not house bound), referred to large regional specialist 'CFS/ME' service in South West England, participating in the SMILE

Study	<p>Beasant 2014¹⁸</p> <p>study (designed to test the feasibility and acceptability of recruiting adolescents to a RCT comparing specialist medical care with specialist medical care and the Lighting Process).</p> <p>Mothers n=13; adolescents n=12, male: 3 (25%), female: 9 (75%), mean age (SD): 13.9 (1.6); median illness duration (IQR): 13 (9 to 18) months</p>
Setting	Large regional specialist ‘CFS/ME’ service in South West England.
Study design	Qualitative interview study
Methods and analysis	<p>In-depth qualitative interviews were conducted using a topic guide to ensure similar areas were covered but with sufficient flexibility to enable participants to raise topics of interest to them, covering questions concerning experiences of the initial clinical assessment appointment, study participation and the interventions that young people received. Families were interviewed at three possible time points: after initial assessment, at the specialist clinic and before randomisation, after randomisation but before the intervention, and after the intervention. Adolescents were interviewed once at one of these time points for not more than 20 minutes. Parent interviews lasted for 20-60 min and were conducted at a convenient location, usually at the participant's home.</p> <p>Interviews were audio-recorded and transcribed verbatim.</p> <p>Data were analysed using thematic analysis using techniques of constant comparison. Data analysis was an ongoing and iterative process, commencing soon after data collection started and informing further sampling and data collection. Two members of the research team analysed ~10% of the data independently to compare coding and enhance its reliability. Descriptive accounts were produced, and theoretical explanations for behaviours, opinions and decisions were developed.</p>
Findings	<p>Information about ‘CFS/ME’ & its management</p> <p>Mothers felt that there was a lack of initial guidance or information around ‘CFS/ME’ and day-to-day management of symptoms before accessing the specialist service, leading to initial confusion and uncertainty about their child’s health and possible diagnosis of ‘CFS/ME’</p> <p>Guidance on management:</p> <p>Young people reported that guidance on how to manage their condition given from specialist medical care brought structure and a sense of normality back to their lives.</p> <p>Tailored approach to treatment:</p> <p>Mothers reported that the tailored, patient-centred specialist medical intervention they gained access to via referral to a specialist service, had not been available earlier and enabled positive change and steps towards managed recovery.</p> <p>Referral to specialist services</p> <p>The majority of mothers reported the initial assessment appointment with the ‘CFS/ME’ service as a positive experience, which was useful and helpful. Referral to a specialist service gave families access to an informative team of experts, for some a formal diagnosis,</p>

Study	Beasant 2014¹⁸
	and for all a tailored, patient centred specialist medical intervention that had not been available earlier. This enabled positive change and steps towards a managed recovery.
	Need for validation for patients
	Mothers reported that the specialist service recognised and acknowledged the young person’s condition, resulting in a sense of relief and reassurance. Mothers felt that symptoms were now understood and that they would receive help.
	Need to talk about ‘CFS/ME’
	Half the adolescents reported that specialist medical care was positive, as it enabled them to talk about their illness.
	Health professional support in the implementation of management strategies for carers
	Some mothers felt that the ‘CFS/ME’ service reinforced symptom management strategies that they had been trying to get their child to follow, and felt that their child would be more likely to listen if techniques were legitimised by a health-care professional. A few mothers also noted that specialist medical care strategies had an impact on the whole family and could be difficult to integrate with their routine lifestyle.
	Support with acceptance
	It was reported that accepting that for a time adolescents must reduce their activity levels and adopt a routine was challenging.
	Educational support
	It was reported by young people that teachers had not been very supportive and mothers discussed the beneficial way in which the ‘CFS/ME’ service opened channels of dialogue between health-care professionals and education providers in a variety of ways. A letter provided by the ‘CFS/ME’ service confirming a diagnosis enabled mothers to legitimately take their child out of school, request funding for home schooling and more generally inform and gain support from teachers when managing reduced attendance.
Limitations and applicability of evidence	Minor limitations due to the role of the researcher not being discussed, data richness (with some findings supported by single quotes). Minor concerns over applicability due to the research aim of the study and representativeness of the sample considering it consisted of feasibility RCT participants which may differ from eligible patients not recruited to a trial.

1

Study	Brigden 2018²²
Aim	To gather the views of adolescents with ‘CFS/ME’ to explore what they access online for information and support, and how this influences the way they cope with the condition.
Population	Adolescents recruited from a specialist paediatric ‘CFS/ME’ service. Inclusion criteria: a diagnosis of CFS/ME (NICE CG53 criteria), age 12-17 years and self-identified as having used the internet for ‘CFS/ME’. Exclusion criteria: insufficient proficiency in English to participate in an interview or severely affected.

Study	Brigden 2018²²
	Characteristics: n=9; male/female: 3/6; mean age (SD): 14.89 (1.9) years, at different stages of the condition; mean number of months from initial assessment to interview (SD): 12.98 (7.98), range 4 to 25) months.
Setting	Specialist paediatric 'CFS/ME' service
Study design	Qualitative interview study
Methods and analysis	<p>In-depth qualitative interviews were conducted using a semi-structured topic-guide that was developed to answer the research question in line with the literature on coping; contained open-ended questions and were conducted by MSc student in Health Psychology covering qualitative methods who received practical training and guidance through supervision around the development of the topic guide and interview style. Participants were encouraged to talk for as long as they needed and Interviews were audio-recorded and transcribed verbatim and anonymised.</p> <p>Thematic analysis was carried out using the stages proposed by Braun and Clarke. Four transcripts were double coded and two senior researchers collaborated on the development of themes and interpretations, informed by the literature on coping.</p>
Findings	<p>Facts/info about the condition</p> <p>a) Symptoms: Participants explicitly talked about the intention to fact-find, with reference to carrying out research and to know more about the condition; some indicate that reading the facts about symptoms was helpful; they identified with the information and felt validated by this.</p> <p>b) Treatments: Some participants felt there was not enough information about treatments.</p> <p>c) Behavioural coping: Although a few participants reported reading about self-management approaches on official websites, for most participants it was the patient-led/peer-led sites that were associated with coping. Participants reported that forums were spaces where participants could learn about coping strategies from others. Such as establishing sleep routines, activity management, pacing, meditation, supplementing schooling with online education, taking exams at home and methods to monitor activity.</p> <p>Sources of information: Digital resources</p> <p>a) Official websites: Around the time of diagnosis participants sought out 'official' sites to establish facts about the condition and talked about the status of those sites as 'reliable'. For the majority, this included National Health Service (NHS) websites, and in some cases also Action for ME, Association of Young people with ME/Chronic Fatigue and the ME Association 'CFS/ME' charities.</p> <p>b) Patient-led/peer-led sites & social media: Participants only used the NHS sites a few times and most moved on to explore patient-led and peer-led sites containing subjective, experiential accounts. This included health forums, but also sites that were not necessarily health-related, including Facebook, Instagram, Blogs and YouTube. In contrast to NHS sites, these were accessed regularly and over the long-term, such as a few times per week or every day. For most participants it was the patient-led/peer-led sites that were associated with coping. Anecdotes and endorsements appeared to promote beliefs in the efficacy of strategies, providing inspiration to try out the new strategies.</p>

Study	Brigden 2018 ²²
	<p>Types/Characteristics of information sources</p> <p>a) User-friendly & reliable: Participants talked about the status of those sites as ‘reliable’. Participants felt that the NHS sites were not user-friendly; they used medical terminology, lacked depth and were static-the content remained unchanged.</p> <p>b) In-depth & updated: Sites reported to be accessed regularly (i.e. patient-led/peer-led) used ingroup terms and phrases which were accessible and appealing, were considered to offer greater level of depth and were constantly updated.</p> <p>c) Relatable: Participants preferred the story-telling approach of patient-led/peer-led and non-health-related sites, the numerous accounts and the technological affordances of videos.</p>
	<p>Social support (via digital resources)</p> <p>Participants described the loneliness of the condition. Through spending time on these sites, they developed ‘connection’ with others and a sense of community, which alleviated this isolation; they experienced a sense of being able to relate to others like them, feeling understood and validated. Certain technological affordances were described as facilitating a sense of relationship. The fact that these sites could be rapidly accessed at any time seemed to provide a great sense of support. Participants stated they could interact with these sites in a quick and undemanding way through a shared language of ‘likes’ and ‘comments’. The online world was less demanding and more flexible than offline relationships especially in the context of a disabling and fluctuating illness.</p>
	<p>Coping with stigma</p> <p>Several participants raised the issues of comorbid psychological difficulties, such as anxiety and low mood, and the support that online sites provided. Some indicated these were difficult subjects to talk about with family, friends and clinicians, but that it was therapeutic to engage with this material online. Participants explained that reading stories online encouraged them to open up and seek help offline.</p>
	<p>Adaptation/ Establishing normality (in the face of an isolating and disabling condition)</p> <p>Patients reported that ‘CFS/ME’ put a strain on normal adolescent life, such as their identity and friendships. The digital spaces allowed them to adapt and maintain normality. Online resources were supportive spaces where patients could explore and maintain aspects of identity unrelated to their condition. They talked about and connected with others about typical adolescent hobbies and things that defined them unrelated to ‘CFS/ME’.</p>
	<p>Relationship support/ Need for validation/legitimacy</p> <p>Participants also talked about the role of the internet in relation to supporting offline friendships. Lack of understanding and invalidation from friends was discussed but individuals talked about showing their friends online information to legitimise their illness and help their peers understand and adapt to the illness.</p>
Limitations and applicability of evidence	<p>Minor limitations due to the role of the researcher not being discussed, data analysis due to the limited details provided.</p> <p>Minor concerns over applicability, the study being of potentially limited applicability to severely affected adolescents since they were not included in the sample.</p>

Study	Broughton 2017²⁵
Aim	To explore the experiences of ‘CFS/ME’ patients who were completing programmes of treatment at three NHS specialist ‘CFS/ME’ services in England.
Population	<p>Adults completing/concluding treatment at one of three outpatient NHS specialist ‘CFS/ME’ services (median age 43, range 24-62 years; median self-reported illness duration 7.5 years, range 1-17).</p> <p>N=16; male: 12.5%, female: 87.5% median age (range): 43 (24-62) years; median self-reported illness duration (range): 7.5 (1-17) years</p> <p>Participants recruited between July-September 2014, who completed a course of treatment within this period, returning a Consent to Contact Form. Exclusion criteria: age <18 years; too severely affected to be able to participate in interviews; unable to provide informed consent; unable to read and understand the patient information sheet and consent forms; or not diagnosed with ‘CFS/ME’ as a primary diagnosis.</p>
Setting	Three outpatient NHS specialist ‘CFS/ME’ services in England.
Study design	Cross-sectional design using semi-structured interviews to explore patients’ experiences.
Methods and analysis	Six face-to-face (conducted at the participant's home) and 10 telephone semi-structured interviews lasting from 23 to 57 min (mean length 32 min) with questions about the patient journey before, during and at the end of receiving specialist medical care. All interviews began with the open question: "Tell me about your CFS/ME" and participants were encouraged to guide discussion and introduce their own topics of interest. Interviews were audio-recorded, transcribed and analysed thematically (by two researchers). Techniques of constant comparison informed the analysis and identification of themes.
Findings	<p>Patient information about ‘CFS/ME’</p> <p>Many participants recalled having a limited understanding of ‘CFS/ME’ prior to accessing specialist services, having received little or conflicting information about the illness. Some participants attributing their early hopes and expectations about treatment to lack of knowledge about ‘CFS/ME’ and to being unsure about what to expect from a specialist service. Moving away from the idea of cure towards goals related to management of ‘CFS/ME’ was linked to obtaining information and acquiring knowledge about the condition.</p> <p>Need for validation of experiences & symptoms from:</p> <p>a) GPs: All participants were referred to CFS/ME specialist services by their GPs and reported varied experiences before referral to specialist services. Participants with positive experiences reported that their GPs had been very supportive; they valued being taken seriously and recognised the key role that their HP had played. Participants with less positive experiences described barriers in accessing specialist services including having to take a proactive role in asking for diagnostic tests and GPs’ lack of belief in CFS/ME</p> <p>b) Employers: Some participants had felt the need to 'hide' symptoms of ‘CFS/ME’, particularly from their employer, for example using annual leave to manage symptom flare up for a number of years</p> <p>Support with acceptance of diagnosis & adaptation:</p>

Study	Broughton 2017 ²⁵
	<p>Although some patients described feeling relieved that diagnosis provided an answer and ruled out other conditions, it was a difficult time for the majority. Participants recalled feeling angry, distressed, frustrated and fearful and that the diagnosis represented a life sentence. Accepting diagnosis of a contested condition was difficult for some; because of participants own negative preconceptions about 'CFS/ME' and the reactions of others. These patients discussed feeling under pressure to convince or prove the validity of their experiences. Time appeared to influence acceptance, with some participants recalling a gradual acceptance that treatment might not be curative. The importance of acceptance in obtaining the most benefit from treatment was highlighted and participants discussed a need to accept changes to their lives as a result of developing ME/CFS, and reflected upon what they had lost or relinquished, including social networks, employment, career and study aspirations and independence.</p>
	<p>Support during early stages of treatment:</p>
	<p>Half the participants recalled finding initial stages of treatment difficult. Many discussed personal responses they believed were key to overcoming challenging periods during treatment. Characteristics described included being open, positive, proactive, willing to try anything, being able to take a leap of faith and having perseverance. They explained how during early stages of treatment advice given by clinicians felt counter-intuitive, and was a departure from the way that symptoms and 'boom and bust cycles' had been self-managed prior to accessing services. Participants highlighted the importance of being 'willing to change' and being prepared to say goodbye to their old life completely in order to engage fully with treatment.</p>
	<p>Realistic goal setting (towards management instead of cure)</p>
	<p>Participants recalled that clinicians assisted with and encouraged the development of new goals which had not been held prior to accessing specialist services. Some viewed these as vital to treatment success, representing a shift in focus towards management rather than cure. New goals were described as smaller, a lot more realistic and more sensible, involving breaking down existing goals, lowering expectations and focusing on the day to day rather than the future.</p>
	<p>Flexibility in medical care (appointments):</p>
	<p>Participants discussed accessibility in terms of being able to attend appointments and accommodate treatment programmes around their commitments. The majority of participants were pleased with the practical accessibility of clinics, describing journeys as being manageable or easy. However all participants mentioned accessibility could be a barrier to attendance. Whilst all reported the ease of access to the clinic improved overtime as symptoms improved, travel during the early stages could be incredibly hard with participants finding the journey stressful and needing to recover after appointments. Some discussed the importance of good public transport links to the specialist service, whilst others felt that they would not have been able to attend appointments without use of a car. Some participants discussed the need for assistance to attend appointments, including help from partners or friends, particularly when symptoms were severe. Others said that work commitments could be a barrier to attending appointments; they noted accessing the clinic would have been difficult if experiencing severe symptoms and concerns were raised about the ability of those severely affected by 'CFS/ME' to access specialist services. Flexibility in the frequency and mode of appointments was valued by participants; with two saying they appreciated being offered later appointments because of travel burden and symptom fluctuation. The option of having some appointments by telephone was highly valued, particularly when symptom severity or travel problems made attendance difficult. Skype was also mentioned as a possibility</p>

Study	Broughton 2017²⁵
	Psychological support:
	a) Mutual support: The majority of participants recalled group sessions positively, with benefits including relating to other patients, the opportunity to share experiences and stories, receiving support from group members, supporting others, hearing about their experiences and having their own personal experiences and symptoms validated and normalised. Some thought it important that a group clicked or gelled in order for the benefits of mutual support to be realised; use of humour, and having things in common were recalled as facilitators of group bonding.
	b) Professional support: Relationships with clinical staff working at the 'CFS/ME' service were highly valued by participants, particularly those accessing one-to-one treatment sessions. Many participants had positive experiences and comments about the clinicians they had worked with. Participants discussed the value and importance of being believed.
	c) Post-discharge support: The majority of participants were worried about their ability to cope following discharge and were concerned about the level of support that would be available. Participants had concerns about life after discharge and some said they would prefer to retain links with specialist services. Some felt they would miss the routine and structure of attending a specialist service or would struggle to maintain progress independently. Strong negative reactions to discharge were expressed by several participants in the context of their experiences before attending the specialist service, when they felt unsupported by GPs, family, friends, colleagues and employers and had experienced stigma and lack of understanding.
	Referral to specialist services:
	Many participants had their 'CFS/ME' diagnosis confirmed when they were assessed by the specialist services. For many participants specialist services provided information and explanation of 'CFS/ME', simultaneously validating and normalising participants' experiences and symptoms. All participants felt they had benefited from accessing specialist service. The majority recalled having had hopes and expectations of referral and treatment including to confirm diagnosis and manage symptoms better.
Limitations and applicability of evidence	No concerns over methodological limitations. No concerns over applicability.

1

Study	Chew-Graham 2008³²
Aim	To explore how patients with 'CFS/ME' and family physicians conceptualise this condition and understand it and how their understanding might affect the primary care consultation.
Population	A purposive sample of family physicians and patients participating in a randomized-controlled trial of 2 nurse-led interventions for 'CFS/ME' in primary care were recruited. For the trial, family physicians in 44 primary care trusts in North West England were invited to participate by referring to the trial those registered patients who had 'CFS/ME'. Patients were considered eligible if they were aged 18 years or older, fulfilled the Oxford inclusion criteria for 'CFS/ME', scored 70% or less on the SF-36 physical functioning scale and 4 or more on the 11-item Chalder fatigue scale

Study	Chew-Graham 2008 ³²
	<p>Family physicians: n=14; 7 male, 7 female; mean age: 48, SD: 12 years; one of the family physicians' practice was not participating in the FINE trial.</p> <p>Patients: n=24; 11 male, 13 female; mean age: 48, SD: 12 years; months since CFS diagnosis range: 1-240, median: 40.5</p>
Setting	Family physicians and registered patients were from 44 primary care trusts in North West England
Study design	Qualitative interview study
Methods and analysis	<p>Semi-structured interviews were conducted by one author at the patients' home and physicians place of work (1 physician was interviewed at home). Interviews lasted between 16 and 72 minutes (median duration= 38 minutes). An interview guide providing a flexible framework for questioning and exploring a number of areas: models of illness, appearance of symptoms, reaching a diagnosis, the consultation and doctor-patient encounters, was used. The interviewer combined open-ended questions to elicit free responses with focused questions for probing and prompting. Digitally recorded interviews were transcribed verbatim by a professional transcribing service, with transcripts checked against the tape by the interviewing author.</p> <p>Analysis proceeded in parallel with the interviews and was inductive taking an interpretative stance. Coding was iterative and informed by the accumulating data and continuing thematic analysis. Coding and interpretation was undertaken individually by four authors.</p>
Findings	<p>Advice on management</p> <p>Some patients described the experience of the family physicians who had been unable to offer any advice to help them understand or manage their condition.</p> <p>Information on 'CFS/ME' (from alternative sources outside the clinical domain)</p> <p>To bypass the sticking point of limited clinical knowledge and actively engage in consultations, patients had to move beyond clinical knowledge and seek additional sources of evidence about 'CFS/ME'. Helpful alternative sources of evidence included representations of 'CFS/ME' expressed within the media. Media personalities lent credibility to the condition, and their positive attributes relieved patients from being culpable. Patients described how they would take information from the Internet to their family physician to bring some knowledge to the consultation and initiate action from the physician, with self-help groups and web sites proving important sources of evidence that patients could take to their family physician. Patients used this method to engage family physicians in a dialog and as means of accessing other treatments and services and the importance of the family doctor listening to the evidence patients sought from other sources was emphasised.</p> <p>Established relationship with physician</p> <p>Some patients believed it was important in both the diagnosis and management of their condition to have an established relationship with their family physician. Not having such an ongoing relationship with their family physician was reported by the patients to make it difficult to achieve agreement about the symptoms and the diagnosis, because the primary physician had no prior knowledge of them;</p>

Study	Chew-Graham 2008³²
	They reported on their inability to demonstrate the extent of their condition beyond the snapshot view revealed in the consultation being unable to establish that symptoms come and go and that the condition is invisible on good days.
Limitations and applicability of evidence	Minor limitations due to concerns over data richness with some findings supported by limited quotes. Minor concerns over applicability due to the sample which consisted of people recruited in a RCT.

1

Study	De Carvalho Leite 2011³⁸
Aim	To investigate the impact of 'CFS/ME' on people from varied social background, including those from ethnic minorities, and what challenges may be posed to health care practitioners in providing appropriate and equitable care for this condition.
Population	Adults with 'CFS/ME', recruited through relevant support groups, community organisations and centres, purposively selected to include a diverse range of illness severity, duration, social variation (age, gender, ethnic background and socio-economic conditions) and year of diagnosis. n=35; aged 18-55; male/female: 8/27; illness duration for the majority was 7≤ than years
Setting	
Study design	Qualitative inquiry using in-depth semi-structured interviews
Methods and analysis	In depth semi-structured one-to-one interviews (n=35) and focus group discussions: six of the 35 participants were purposively selected (to include a diverse range of illness severity), for both an initial focus group discussion and the later one-to-one interview. These were tape-recorded and transcribed verbatim. The Focus group with six people with 'CFS/ME' was used to identify the main themes and issues to be explored more deeply in the subsequent interviews. It took place in a quiet room and lasted for two hours, with a break for refreshment and rest. The group was conducted by a researcher, while another researcher supported the group dynamics, observed and took notes to facilitate later analysis. The discussion was managed as a conversation, encouraging participants to tell their own stories to help articulate their ideas about the experience of living with 'CFS/ME'. Three broad areas of inquiry reflected in guide questions were used as starting points to encourage story-telling and discussion to facilitate the emergence of story line narratives within these areas: a) becoming ill and being diagnosed; b) the impact of living with 'CFS/ME'; and c) self-management and being managed within health and social care services. Story telling allowed themes to emerge, without being fixed to a set research agenda. The sequence and wording of questions were decided in the course of the discussion to respond to participants' preferences and conversational styles.

Study	<p>De Carvalho Leite 2011³⁸</p> <p>One-to-one semi-structured interviews of about 45 minutes (up to a maximum of 3 interviews per participant (45 interviews in total) were conducted with the 35 participants by a researcher at the participant’s home or another place convenient for them.</p> <p>Thematic analysis was used on both the focus group and interview datasets. The focus group data transcripts were analysed by four researchers, who together identified the main storylines and emerging thematic areas of support needs, and then adapted question guides for one-to-one interviews.</p> <p>The interviews transcripts were analysed by five researchers who first independently read and re-read the transcripts to identify and extract words and text sections which appeared to describe experiences of living with CFS/ME and encountering health and social services. They independently selected, focused and condensed the data in tabulated written notes with codes. Three researchers met to compare the reliability of codes and agree the developed coding scheme. New codes were developed before comparative subject analysis. Finally a wider group of researchers drew conclusions for the whole dataset</p>
Findings	<p>Validation& Recognition of symptoms</p> <p>a) By practitioners: Participants revealed how they were facing distinctive illness-related barriers in gaining recognition of their illness. Their encounters with health professionals were reported as often problematic in ways which both delayed or reduced access to support (to manage the illness required people to gain access to appropriate health expertise which in turn, could affect the likelihood of gaining family and wider social support) and greatly exacerbated emotional pressures. There were reiterated experiences of not being listened to by health-care practitioners. This often posed particular problems in the earliest stages of the condition. Nearly all participants, from both white and non-white groups spoke of their illness not being taken seriously by GPs, with individual symptoms being dismissed, perhaps as a ‘virus’ or as a common cold. Many experienced this as a profound lack of acknowledgement. This was reflected in participants across all ethnic groups wanting health care practitioners to have the time to help the patient feel ‘empowered’ and ‘believed’, to increase their sense of inclusion and acknowledgment. Even when bed-bound, participants encountered unsupportive attitudes from health professionals which greatly undermined their chances of wider belief and support. Participants highlighted how lack of access to social care and practical support was exacerbated when health practitioners would not recognise their illness, making a profound impact on their ability to carry out their family caregiving roles.</p> <p>b) By patients themselves: Lack of recognitions could be especially difficult for people from ethnic minority groups in which such illnesses were less commonly identified as self-recognition and belief of the symptoms and experiences was problematic.</p> <p>Need for a diagnosis</p> <p>Achieving a diagnosis was seen as a crucial milestone for most participants. Where this led to advice from doctors and other health care professionals with particular knowledge of ‘CFS/ME’, this was almost invariably a positive experience (e.g. one participant commented on his luck in gaining a prompt GP diagnosis, leading to coordinated care and support from his manager, which allowed him to work part-time within his capabilities and to gain sick leave and retirement as the illness progressed’). Most participants however, found doctors saying they could not help, resulting in their feeling abandoned to fight the problem by themselves. Participants most often encountered oppositional health services responses and some therefore decided to use a private or alternative health</p>

Study	De Carvalho Leite 2011³⁸
	services as a way of getting diagnosis or help, often exacerbating stress, uncertainty and financial pressures. It was also reported that until a diagnosis was gained, social services could not even assess their needs
	Need for variability and choice in illness management
	Participants felt that the health-care system should explore useful interventions and suffered from a lack of control over choices of treatment for managing their illness, which they saw as due to both lack of resources in the National Health and social systems and relative lack of recognition or value given to their own experience with illness. Participants desperate for relief of feelings of pain or illness reported finding treatments such as massage, osteopathy, dietary advice and acupuncture helpful, and it caused ongoing frustration that such interventions were not funded by either the NHS or by a private health insurance for 'CFS/ME.'
	Need for tailored hospital care
	People who had hospital care described their need for designated wards for 'CFS/ME, with environments adapted to their needs, as in keeping light and noise levels low. Some highlighter the limited time for consultation as a barriers to appropriate care provision and another reason for seeking support outside the NHS.
	Practical support
	Practical support for personal care, family roles, independent living and support for carers was invariably seen as extremely important for people with moderate to severe illness. Many reported needing help with all personal and domestic tasks: with moving around the house, getting out of bed and from chairs, washing and dressing, feeding and self-care, running a home, including meals preparation, shopping or cleaning and how these intensified with child care. Where people could not find alternative ways of getting support for practical tasks, the home environment, where they had to spend most of their time, became not their refuge but a further source of stressful experiences of deteriorating well-being.
	Access to social care support
	Social care was of paramount importance for most participants' life priorities which often included sustaining their own roles as family caregivers. They highlighted how lack of access to social care and practical support was exacerbated when health practitioners would not recognise their illness, making a profound impact on their ability to carry out their family caregiving roles, particularly as parents. Participants described how 'most people with 'CFS/ME' don't get social care' raising serious questions about how people might go and manage their lives with the debilitating symptoms they reported. Without social care support, often partners, parents and sometimes children had to become carers.
	Financial Support
	Financial support was consistently identified as crucial for illness-and life-management, and to maintain education, social relationships and maternal well-being. All participants described many financial constrains arising in the absence of other forms of support to live with 'CFS/ME'. There were primary consequences of impoverishment, but then secondary consequences of for social standing, relationships and future entitlements. Limited incomes imposed hard choices about what money would be spent on, which depts. To run up

Study	De Carvalho Leite 2011³⁸
	Information and support accessing welfare benefits
	Participants described the frustrations of attempting to apply for welfare benefits while affected by ‘CFS/ME’. The nature of ‘CFS/ME’ symptoms (with extreme fatigue, pain and cognitive impairment), meant that complicated forms having to be completed even to start the process were especially daunting. Participants frequently found the benefits system complicated and confusing, something to ‘fight’ rather than a source of support. Not being able to predict what they were entitled to, people found out in a hit-and miss way, making arduous applications for benefits that were often refused. Many participants, especially from non-White groups, expressed their needs for much more information on entitlement to help focus their applications on attainable benefits. Some participants, unable to claim any benefits whether through lack of support from benefits staff or their current financial situation, experienced profound effects on their lives, being bereft of resources, and enforced total dependence on family or partner. Where not getting benefit and feeling too unwell to sign on for unemployment benefit, people could then experience a lack of acknowledged civil status and, without being credited for National Insurance payments, worried about the implications for later claiming pensions. Where benefits were successfully secured, these were always perceived as helpful.
	Support accessing employment
	For people beginning to recover and wanting to increase their activities gradually to include limited part time work, income support was seen as too inflexible to allow this. The benefit stopped if people started work, even when they could not work sufficient hours to earn enough money to support themselves. To obtain essential benefits they needed to represent themselves as very impaired, yet in attempting to move back into employment or education, they had to represent themselves as minimally affected.
Limitations and applicability of evidence	Very minor limitations due to the potential influence of the researcher on the findings not being explored. No concerns about relevance with patients from diverse social and ethnic backgrounds and various degrees of illness severity and duration being represented in the sample.

1

Study	Devendorf 2018⁴³
Aim	To investigate factors, other than depression that explain suicidal ideation, including quality of life, loss of functioning, isolation, and hopelessness about prognosis.
Population	Patients who self-identify as having ME/CFS and endorsed suicidal ideation (SI) but did not meet depression criteria; recruited through patient advocacy websites, newsletters, social media and Internet forums. N=29; 79.3% female, 20.7% male. Mean age: 51.48 years old. Mean score for the BDI-PC: 2.38; one participant endorsed active SI (i.e. score of 3), 28 participants endorsed passive SI (i.e. score of 1).
Setting	The study was hosted online, with participants recruited from patient advocacy websites, newsletters, social media and internet forums.

Study	Devendorf 2018 ⁴³
Study design	Mixed-methods design; qualitative analysis of participants' open-ended survey responses from a previous project that examined illness severity, stigma, physician interactions and depression (McManimen <i>et al</i> , 2018).
Methods and analysis	<p>After analysing participants' quantitative responses to the Beck Depression Inventory for Primary Care (BDI-PC), the authors qualitatively analysed participants' open-ended responses that followed the previously completed survey. Participants could clarify or expand upon their survey responses through an open-ended format.</p> <p>Analysis was conducted in the following steps: (1) multiple readings of the data; (2) open coding; (3) developing a final code-book; (4) applying the final code-book, while considering the whole context of each response; (5) establishing inter-rater reliability; and (6) finalizing and categorising codes into themes and sub-themes.</p>
Findings	Need for financial resources:
	Lacking financial resources made it difficult for patients to cope, since they had to exert energy to work themselves causing physical consequences.
	Need for caregiver resources:
	Lacking caregiver resources made it difficult for patients to cope, since they had to exert energy to work themselves causing physical consequences. Participants felt hope, validation and compassion when there were support systems present to help with daily living activities, with a supportive loved one for instance reported to save a patient valuable resting time.
	Need for/Access to knowledgeable/helpful physicians:
	Many patients lacked access to helpful physicians and for those who had positive healthcare experiences, having a knowledgeable physician provided hope, encouragement and relief. Participants lacked access to helpful health-care providers and many encountered disdain, disbelief and lack of knowledge. Finding helpful physicians took years of shuffling through 'ignorant' doctors.
	Need for validation (from HCPs) & acceptance of ME/CFS as non-psychiatric:
	19 patients commented on their dissatisfaction with health-care providers which were likely driven by disregard for ME and CFS. Most encountered doctors that were trained to view ME and CFS as psychiatric, which was dismaying. Disappointment ensued when doctors vocalized psychological attributions or inferences, with exercises recommendations and psychological attributions or inferences.
	Need for new helpful biomedical treatments
Participants called for more physiological research to develop medicinal treatment and destigmatize the field. They did not believe their illness would resolve naturally and most were hopeless that new, effective treatments would be developed in their lifetime. They criticized the continued focus on cognitive-behavioural and graded exercise therapies because they attribute the illness to psychological causes and felt that ongoing debates were a hindrance toward biomedical progress for biomedical treatments. Some patients retained for a new treatment as a rationale to continue to live. Six participants said there were moments when their illness causes overwhelming physical pain that is unbearable and with no helpful treatment, participants viewed suicide as a logical escape.	
	Information on ME/CFS for family and friends:

Study	Devendorf 2018⁴³
	Participants frequently encountered misunderstandings about ME and CFS from friends, family, healthcare providers and the public. They were frustrated when friends and family misunderstood and downplayed the severity of ME and CFS. It was reported that friends and family gave participants unsolicited advice about treatments and receiving this unwarranted advice frustrated participants because it often conveyed that others misunderstood or delegitimized ME and CFS. Participants felt obligated to educate others to clarify misunderstandings, provide context and serve as advocates.
	Social support
	Participants were physically and mentally isolated and struggled to maintain relationships because of their fatigue, post-exertional malaise, pain and cognitive impairments. Several lost close friends. Many used social media and online forums, which were helpful, but not commensurate with face-to-face interactions.
Limitations and applicability of evidence	Moderate methodological limitations due to the appropriateness of the data collection method, the study being a follow-up to a quantitative study with open-ended online responses. Moderate concerns over applicability due to participants being a subset of a previous quantitative study who were self-identified as ME/CFS (not diagnosed according to accepted criteria) with suicidal ideations but not depression.

1

Study	Hannon 2012⁵⁸
Aim	To develop an education and training intervention to support practitioners in making an early diagnosis of 'CFS/ME' and supporting patients in the management of their symptoms.
Population	Health practitioners (GPs n=9, practice nurses n=5, 'CFS/ME' specialists n=4), Carers (n=10), patients (n=16), aged 28-71 were recruited via purposive sampling of GP practices, advertisements through existing 'CFS/ME' support groups, community groups and via the patient co-investigator or through specialist 'CFS/ME' services in the NHS in response to a project flyer. Patients and carers included n=12 BME (black minority ethnic) group participants.
Setting	Patients and carers were recruited through 'CFS/ME' support groups, community groups, specialist 'CFS/ME' services in the NHS. A purposive sample of BME group patients were also recruited from South Asian third sector groups in Greater Manchester and personal visits to community groups. Practitioners were recruited via a purposive sample of GP Practices and Primary Care Trusts.
Study design	Qualitative interview study
Methods and analysis	Individual semi-structured interviews were conducted face-to-face using topic guides: patient/carer interview focus included experiences of being diagnosed, support received in primary care. Interviews were audio-recorded and transcribed verbatim. Initially inductive analysis was conducted using thematic analysis in line with modified grounded theory approach, using open coding; a deductive approach was then taken when data fully analysed.

Study	Hannon 2012 ⁵⁸
Findings	Need for validation (from medical community)
	Patients and carers described their frustration when GPs or practice nurses did not recognise the seriousness of their symptoms or questioned the legitimacy of the condition. Patients who felt believed by their GP described how important this was to their wellbeing.
	Need for positively framed diagnosis
	Patients reported a diagnosis allowed them to tell family, friends and their employer that they had a label for their symptoms. All patients highlighted the need for the diagnosis to be given by the GP in a positive way to maintain hope that symptoms can improve.
	Advice on symptom management
	Patients described having been given a diagnosis of ‘CFS/ME’ without any advice on symptom management or support. They described how they had been left to find their own information and persuade the GP to meet their needs.
	Need for alternative therapies
	Some patients also sought help from private professionals or paid for alternative therapies as they did not feel that they received any support from the NHS and this often led to financial costs.
	Direction or Sign-posting towards sources of support/ relevant contacts or support/ Help accessing support
	Patients and carers highlighted the need for sign posting from the GP, information on local support groups, advice on benefits and referrals to the third sector, however most GPs and practice nurses did not have details of relevant contacts.
	Sources of information (printed & audio-visual)
	Patients and carers stated that a brief leaflet outlining the symptoms and the evidence for management options would be useful at diagnosis. For patients finding it too difficult to read leaflets, a DVD would be preferred. Patients said they would like an overview of the evidence base on the DVD and case studies of patients and carers to gain an understanding of how others live with the condition.
	Patient support from carers during consultations
	Patients and carers described how visiting a GP can be a challenging experience, with patients describing difficulty in remembering or articulating their symptoms and how they would take a carer or family member with them to make sense of the consultation. Patients and carers described the important role that carers play in the management of the illness, which included support in the home and during a GP consultation.
	Flexibility in medical appointments
Some patients also highlighted a need for flexibility when making appointments.	
Support for carers	
a) Psychological support: Carers explained how they could feel isolated as they had to devote so much time to the patient in order to attend to their needs.	
b) Information about ‘CFS/ME’: Some carers lacked understanding of ‘CFS/ME’ and were frustrated by the patient's symptoms.	

Study	Hannon 2012⁵⁸
	c) Advice on how to support patients: Carers discussed the danger of giving inappropriate advice or support to patients, exacerbating symptoms or pushing the patient to do too much too soon.
Limitations and applicability of evidence	Minor methodological limitations due to the role of the researcher not being discussed, data analysis because of data richness with themes mostly supported by single quotes. No concerns over applicability.
Study	Harris 2017 ⁶¹
Aim	To explore what adolescents felt had caused their problems with eating, whether there were triggers and maintaining factors and what interventions they felt would be helpful.
Population	Adolescents with a primary diagnosis of ME/CFS, aged between 12-18 years who experienced at least one of the following: difficulty with eating, frequent nausea, lack of appetite, weight loss, abdominal pain, bloating, diarrhoea or constipation. The sample was drawn from a 'CFS/ME' specialist hospital service providing regional support for assessment and treatment of over 300 children a year in the Gloucester, Bristol, Wiltshire and Somerset areas, covering a population of 400,000 children aged 5-19 years (Office of national statistics, 2011).
Setting	'CFS/ME' specialist hospital service
Study design	Qualitative interview study
Methods and analysis	Semi-structured interviews were conducted. Interview topics included the adolescents' experience of eating; the factors they felt caused and exacerbate eating difficulties and what they believed were helpful strategies. Interviews lasted approximately 30 minutes and were audio-recorded, transcribed verbatim and analysed thematically using techniques of constant comparison which commenced soon after data collection and informed further interview protocols. All adolescents were given the option to be interviewed alone or with a parent and seven chose to have their mothers present, all of whom contributed to the interview. Verbatim quotes from participants and mothers are used to illustrate themes.
Findings	External support for eating difficulties: Eating difficulties impacted the adolescents' health affecting their weight, fatigue and their broader social and family circumstances. Mothers reported feeling 'scared', 'awful', 'angry', 'stressed' and 'terrified' by the dramatic weight loss and 'real change' in their child's health as a consequence of poor eating circumstances. Some of the families described seeking external, professional support for the adolescents eating difficulties. Five adolescents took prescribed sickness or stomach acid relief medication which they found helpful; however, it was not common to have been offered medication to relieve their symptoms which frustrated some adolescents.

Study	Hannon 2012⁵⁸
	Education about 'CFS/ME' & its associated eating difficulties:
	Parents felt it would help to know eating problems are 'normal' in this population and may help them understand the illness further.
	Practical advice for dealing with eating difficulties:
	Practical advice such as 'recipe booklets', 'cooking classes' and 'guidelines on what I should and shouldn't eat' were suggested as possible helpful interventions both by adolescents and their parents.
Limitations and applicability of evidence	Moderate limitations due to the role of the researcher and issues with data richness. Minor concerns over applicability due to the population being limited to adolescents with ME/CFS and their parents, who experienced eating difficulties; findings may not be equally relevant to the wider population of ME/CFS who did not experience such difficulties.

1

Study	Jelbert 2010⁶⁹
Aim	To provide a qualitative perspective of adolescents' experiences of ME/CFS
Population	Five adolescents who were considered to have recovered from ME/CFS. Participants were chosen on the basis of having met diagnostic criteria for CFS as assessed by the consultant paediatrician running the paediatric outpatient clinic through which they were approached. Recovery was represented by a clinical decision to discharge. N=5; 4 female, 1 male. Mean age: 15.2 years (range 13-18 years). Only adolescents who had been discharged within the last year were included. All participants reported having experienced ME/CFS symptoms for a duration of between 1.5 and 2 years.
Setting	Paediatric outpatient clinic, UK
Study design	Semi-structured interviews and thematic analysis (interpretative phenomenological analysis).
Methods and analysis	Data was collected through individual semi-structured interviews. All participants chose to be interviewed at home. Points of reference were agreed around areas that were felt likely to be of particular pertinence to young people with ME/CFS, including impact of CFS on everyday living, coping with ME/CFS, and impact of ME/CFS on self-identity. Additionally, the researcher sought to explore adolescents' understanding of ME/CFS and the impact of ME/CFS on their future hopes and plans. The interview therefore opened with broader questions around participants' general experiences of CFS: "Can you start by describing what CFS is to me?" and "Can you now tell me what CFS was like for you?", followed by further questions and prompts around the additional areas described. All interviews were audiotaped and transcribed in full. Transcripts were read several times by the interviewer, noting initial thoughts and then tentatively identifying initial themes, or summary phrases, from these notes. Emergent themes were listed and then progressively organized into clusters as possible connections between them were identified. The researcher revisited the transcript a number of

Study	Jelbert 2010⁶⁹
	times to check that connections identified within the data made sense and did not detract from the essence of the primary source material. Cross-validation, through the presentation of the material to a second qualitative researcher not involved in the study, was carried out to prompt discussion and achieve interrater agreement, aiming to avoid individual researcher bias. Member validation was also carried out, comprising of a summary of the results being given to all five participants to check that the themes identified were felt to match their actual experience
Findings	<p>Need for understanding (from medical & social community)</p> <p>Young people appeared to experience difficulty with a lack of understanding and awareness around the condition from medical professionals involved. All but one of the young people referred to a fear or actual experiences of disbelief and judgment from others (including teachers and peers), which proved to be particularly difficult for some. The impression given in participants' comments prior the interviews was that they felt it was important to recount their story so that the full extent of their difficulties might be recognised, listened to and learnt from by others.</p> <p>Need for a diagnostic label</p> <p>Many described a long period of diagnostic uncertainty as being particularly difficult both in terms of feeling helpless and in terms of feeling disbelieved. Arriving at a diagnostic label for the illness appeared to be a helpful experience for constructing their own meaning and dispelling disbelief from themselves and from others.</p> <p>Information on CFS (prognosis)</p> <p>Many of the young people appeared to experience difficulty with a lack of information around the condition from medical professionals involved. Many negative and undesirable emotions were described relating to various aspects of the young people's experience of CFS, including the uncertainty around the illness diagnosis and prognosis, with participants reporting not knowing why they were undergoing many tests made them feel worried and not knowing what to expect making them feel scared.</p> <p>Social support (family, friends & other patients)</p> <p>All young people identified support from parents and friends as having been particularly important to their management of and recovery for CFS and most felt that meeting and talking to others with CFS was helpful.</p> <p>Support during transition from ill to well/ with reintegration</p> <p>Some people appeared to find reintegration and their transition from ill to well more difficult than others, reporting getting used to the illness and feeling scared about what it would be not to have it.</p>
Limitations and applicability of evidence	<p>Minor methodological limitations due to small sample size and homogenous population of participants who attended the same clinic.</p> <p>Minor concerns about applicability due to all participants having recovered from ME/CFS and therefore possibly not holding similar opinions to those in the stage of active ME/CFS.</p>

Study	McDermott 2011⁸⁷
Aim	To explore the hopes and expectations of patients newly referred to a 'CFS/ME' Service in the South of England.
Population	<p>Patients consecutively referred to a specialist 'CFS/ME' service in the South of England by their GP between June and October 2009. The service framework comprised: a diagnosis by a specialist doctor (GP with special interest in 'CFS/ME') for patients with no previous diagnosis or uncertainty regarding diagnosis; self-help advice on strategies to improve symptoms, provided in the form of a Lifestyle Management Group Programme led by an Occupational Therapist or Nurse Specialist (5 x 2 hours sessions held over 10 weeks).</p> <p>N=20; mean age (range): 39 (22-60) years; male/female: 3/17; 2/10 were members of a patient support group.</p>
Setting	ME/CFS specialist NHS services, South of England
Study design	Qualitative interview study
Methods and analysis	<p>Semi-structured telephone interviews were conducted by a researcher, using a semi-structured interview guide that was developed and piloted with the help of researchers, specialist 'CFS/ME' health professionals, GP colleagues and four individuals with 'CFS/ME'. Participants were asked about their hoped and expectations of referral and their beliefs about the nature, prognosis and appropriate management of 'CFS/ME'.</p> <p>Interviews were audio-taped, transcribed and analysed using drawing on the principles of constant comparative analysis, with initial findings from each interview being used to inform and guide subsequent interviews. Five transcripts were analysed by the primary researcher to draw the preliminary list of codes while the two fellow researchers independently coded the same data to enhance dependability of results. Based on initial codes, the emerging themes were tested on the full data corpus of 20 interviews and all authors checked the validity and consistency of coding in several data sessions (involving the discussion of data fragments and refining the coding scheme)</p>
Findings	<p>Need for specialist advice</p> <p>Patient participants reported that they had been referred because they were trying to find a pathway through the illness; the need for specialist advice had generally been suggested either by the GP or by the patient themselves, after attempting unsuccessfully to resolve symptoms using strategies available in primary care.</p> <p>Need for diagnosis</p> <p>All participants described gaining diagnostic clarity as a step towards regaining self-respect, being able to explain the problem, to others and as a positive move towards regaining some control of their lives or taking constructive action to improve their symptoms</p> <p>Information about 'CFS/ME' & prognosis</p> <p>Some participants perceived the period of waiting for specialist diagnosis as a time of great uncertainty and lack of information. Many participants felt they had little knowledge about 'CFS/ME', In some cases, participants feared that 'CFS/ME' was a degenerative disease comparable with Parkinson's disease or multiple sclerosis. Lack of clarity about 'CFS/ME' and likely prognosis appeared to be a key source of distress and worry. The few participants who expressed confidence in their level of knowledge about 'CFS/ME' also</p>

Study	McDermott 2011⁸⁷
	tended to express less anxiety about the future and less concern or distress about waiting for the appointment. Participants tended to perceive their illness as complex, they volunteered a range of possible contributory factors and expressed hopes that the service would be able to advise them on which were relevant to the illness. Gaining specialist guidance to understand the complexity of the illness, especially interaction between different factors, was perceived by many as a route out of this cycle of fatigue.
	Advice on management
	a) Self-help & alternative strategies: Participants expectations of the specialist service tended to focus on hoping for advice on effective self-help strategies; they reported that a key expectation of their referral was to find out whether they were doing the right thing in terms of how they were dealing with their illness (e.g. they reported a sense of dilemma about whether to push themselves to continue activities or whether to rest or take time off). Advice was also sought on the use of alternative therapies and participants expressed a willingness to try out new strategies suggested by the specialist service.
	b) Lifestyle advice: Advice was also sought on diet, the effects of stress on symptoms, how to best cope with continued employment or return to work.
	Need for positive direction for the future
	From other patients: Hearing from or about individuals who had recovered or substantially improved following 'CFS/ME' was perceived as a potential source of encouragement which could be offered by specialist services. Media accounts of individuals who were very severely disabled by 'CFS/ME' appeared to be a key source of anxiety.
	From specialist service/ clinicians: When describing their expectations, a clear tension was evident between wanting and believing in hope for the future and fear that their illness would not improve or might deteriorate. Participants hoped that referral to a specialist service would give them positive direction for the future.
Limitations and applicability of evidence	Very minor limitations due to the role of the researcher not being discussed. No concerns over applicability.

1

Study	Mihelicova 2016⁸⁹
Aim	To give voices to those who care for individuals with ME and are often stigmatised and inform future research to ensure that parent-carers of individuals with ME have adequate resources and support.
Population	Parent individuals caring for ME/CFS patients of various ages, ranging from a 5-year old child to adults; primarily parents caring for adult daughters. N=19; 12 mothers and 7 fathers.
Setting	Public domain

Study	Mihelicova 2016⁸⁹
Study design	Secondary analysis of qualitative interviews
Methods and analysis	<p>Data were collected from interviews published in <i>Lost Voices from a Hidden Illness</i> (Boulton, 2008), a book offering a collection of passages from individuals with ME, their significant others, and carers that aims to raise awareness of the impact of ME and to allow individual voices to be heard. Parent accounts were scanned and transferred into NVivo 9.2, which was used for the present analysis. Two authors new to the area of ME/CFS separately coded the data and three authors with extensive research experience in the area provided guidance and feedback throughout the project.</p> <p>Data were analysed using interpretative phenomenological analysis (IPA) methodology, with thematic trends being noted. Data was consolidated through a merging process where coders brought their analyses together. Themes that were not found across both coders were retained.</p>
Findings	<p>Adaptation:</p> <p>a) Dealing with identity change: ME was reported to impact various aspects of the parents' identity, such as their roles within their families, relationships and aspects of their lives outside of their families such as their careers. Parents reported taking on roles as carers, advocates for the illness and their children and support their children as educators. Parents described the ways in which their roles within their families have changed and the way this has affected their own well-being. The parent role is redefined to include caretaking responsibilities, which may impact well-being. The role of parental carers is a balance of providing day-to-day care for the person who is ill while providing affection. Parents may struggle to find ways to provide affection to their children who are often in great pain; in essence ME may change the way in which parents relate to their children. Taking a new role results in much of the parent's time being allotted to caretaking responsibilities.</p> <p>b) Dealing with changes in the perception of time: Most parents mentioned an evolving relation to time. They describe some days as being repetitive with long to-do lists and multiple carer responsibilities resulting in the experience of changes in how time is felt.</p> <p>c) Changes in family relationships/ dynamics: As a result of parents taking a new caretaking role, they reported being distanced from relationships with other family members, from their hobbies and careers. They described how their relationship with their spouse and children changed and distancing manifested in the relationships within the family unit. Distancing extends to the way a family, as a unit, spends their time with parents reporting that their family plans and habits have changed.</p> <p>Need for advocacy from medical & social community and connection:</p> <p>Parents reported they were often met with disbelief and hostility from medical professionals, that close friends and other family members were sceptical and perplexed by the illness and as a result may find themselves disconnected from others who may not respond well. Parents felt alienated and reported being estranged from friends and family as the role of carers became their priority. Because of a lack of understanding by those who are not immediately impacted by the illness, carers were left feeling alone in their world. Parents report hoping for change in terms of the way ME is received and wish for understanding from others.</p>

Study	Mihelicova 2016⁸⁹
	Dealing with uncertainty/ Support during difficult times: remission/relapse
	Parents reported feelings of uncertainty in regards to diagnosis and prognosis, particularly following phases of remission and relapse. Parents discuss difficulty in seeing their son/daughter relapse, particularly when there has been improvement prior to relapse, which offered hope that was then lost and exacerbated the anxieties of living in uncertainty.
	Information on prognosis: fear of death & feelings of uncertainty
	Parents reported feelings of uncertainty in regards to prognosis; they talk about fears of death when their son/daughter was at his/her worst.
	Information on treatment and ways to support patients (e.g. energy management strategy, anti-viral medication)
	Parents report a lack of treatments and resources for managing ME. Due to a lack of knowledge in the medical community, patients may encounter difficulty with finding appropriate treatment and support for their children. One parent did describe effective symptom management strategies and improvement after certain treatments. Considering their child's energy expenditure and helping him avoid over-exerting himself was an effective way to manage the illness. The same parent also talked about seeing significant improvement in their young son when he was treated with anti-virals by a medical herbalist
Limitations and applicability of evidence	Serious limitations related to the use of public data due to lack of details on how the sample was derived, the methods of primary data collection and inability to assess bias. No concerns over applicability

1

Study	Ryckeghem 2017¹¹⁶
Aim	To explore the experiences and expectations of patients with chronic fatigue syndrome and general practitioners to develop the potential role of an advanced nurse practitioner at the diagnostic care path of abnormal fatigue developed for regional transmural implementation in the Belgian provinces of East and West Flanders.
Population	A purposive sample of patients was selected through the department of General Internal Medicine at the University Hospital Ghent to achieve maximum variation. A convenience sample of GPs was recruited from different provinces in Belgium. Patients (n=15); median age (range): 45 (33-59 years), n=14 female ; GPs (n=15); median age (range): 49 (31-62 years), n=7 female.
Setting	University Hospital Ghent
Study design	Qualitative interview study

Study	Ryckeghem 2017 ¹⁶
Methods and analysis	<p>Individual semi-structured interviews were conducted over 9 months in 2014-2015, using interview guide questions developed through an extensive literature review. Interviews took place at the patients' home (n=12) or at the University Hospital Ghent (n=3), lasted on average 1 hour 4 minutes and were audio-recorded and transcribed verbatim. All interviews started with the same question: 'Could you tell me more about how it all started?'</p> <p>Interviews conducted with GPs took place in their practice and lasted on average 27 minutes. GP interviews started with the question: 'What is your professional experience with patients diagnosed with CFS?'</p> <p>Thematic analysis (open explorative thematic coding) was used.</p>
Findings	<p>Patient information about CFS Patients feel they lack general information about the disease and they are not offered a clear model of explanation.</p> <p>Patient information on/ Understanding of the way forward (following diagnosis) When participating in the care path, most patients experience they do not have sufficient overview of what to expect. They know they have to undergo several tests, but often find the information very superficial. They often suffer from information overload, hampering clear understanding. Following the diagnosis, patients reported they often don't know how to proceed.</p> <p>Need for consistent guidance in the care process/ potentially in the form of an intermediator Many patients noted they were not seen by the same medical doctor or caretakers at intake and feedback consultations in the referral centre. Therefore, they emphasize the need for someone who accompanies them, informs them advises them and instructs and assists them at all stages of the care process. Most patients also experience they need someone where they can turn to and with whom they can build a relationship of trust, someone who shares their views and who they can tell their story to, who can because of his specialised knowledge and skills, identify their needs and expectations and answer them.</p> <p>Need for support/ follow-up post-diagnosis Patients need intensive support and follow-up after completion of the diagnostic care path at the referral centre. Nevertheless most experience the feeling of being left in the dark afterwards and usually having to search themselves for caregivers in their neighbourhood.</p> <p>Need for Empathy/Validation/recognition Patients find it important to inform the environment (family, friends, colleagues and employers and caregivers so CFS patients receive more recognition and credibility</p> <p>Punctuality of reporting/ care & timely communication Patients experience they sometimes have to wait a long time before receiving reports of examinations performed, reporting having to visit doctors and services and asking for reports several times and still not receiving what they asked for. They felt they should not have to ask for reports.</p>

Study	Ryckeghem 2017¹¹⁶
	Information for the patient's environment
	<p>a) Information for families: People within the patients' direct environment have little understanding of the disease and the disease progression, making patients feel that their family, friends and colleagues have difficulties empathizing with them.</p> <p>b) Information for employers/ occupational support: Patients experience similar difficulties for employers if they are still at work. Although they do receive advice from the referral centre to make adjustments at work, patients often describe these adjustments as 'impossible to implement'</p>
Limitations and applicability of evidence	<p>Moderate limitations due to the role of the researcher, concerns over data analysis due to the lack of detail and concerns over data richness with some findings supported by single quotes.</p> <p>No concerns over applicability.</p>

1

Study	Taylor 2005¹³¹
Aim	To determine what aspects of the disability experience of persons with CFS are explained by the social model of disability, and what aspects of disability fall outside or contradict central tenets of the social model.
Population	<p>Adults with ME/CFS, who were participating in a research project aimed to evaluate a participant-designed rehabilitation program. All participants met the CDC Fukuda <i>et al</i> (1994) criteria for ME/CFS.</p> <p>N=47; 45 female, 2 male. Mean age: 46.9 years (SD 10.4). Seven participants were in full-time work, seven in part-time work and 33 were not working. Eight participants were minority ethnicity, 39 were non-minority.</p>
Setting	A centre of independent living in the United States
Study design	Qualitative study on data from focus group interviews, open-ended questionnaires, progress notes, and from a program evaluation questionnaire.
Methods and analysis	<p>Data for this study emerged from a federally funded research project that developed and evaluated a participant-driven program for individuals with ME/CFS. The study was a participatory research project in which clients actively identified their service needs, shaped the services they received, and decided the criteria by which the services would be evaluated. For each client, qualitative data were collected over a period of 12 months. Data were drawn from the following sources: (1) Focus Groups; (2) End-of-Group Reflections Form; and (3) Progress Notes.</p> <p>During Focus groups participants were educated about the social model and were asked about their experiences with CFS within social contexts of home, work and community, their interactions with health care providers, family, friends and peers with and without disabilities. End-of-Group Reflections Form questionnaire was distributed at the end of each group meeting and included questions</p>

Study	Taylor 2005¹³¹
	<p>such as ‘Was there anything in particular about the independent living philosophy, advocacy, empowerment, or sense of community that you learned in today’s group?’</p> <p>Analysis was based on the grounded theory approach and followed a qualitative comparative method. This type of analysis involves going back and forth between the emerging data findings and ongoing data collection. This process allows for the themes that emerge from the findings to be checked for counter instances, more fully explored, and further developed. Triangulation was used to achieve confidence in the findings by comparing information within and across data collection methods, across participants, and across time.</p>
Findings	Information on ‘CFS/ME’ for patients
	Most participants found information on CFS outside medical care (through the internet or self-help groups) and then took the information to their physician)
	Information on ‘CFS/ME’ for social environment (families, friends and co-workers)
	Participants reported that family members, friends and co-workers viewed them as individuals that had no obvious manifestation of the disability and who often had extreme variability in symptoms and impairment. They noted that friends had difficulty understanding the fluctuation in the severity of the impairment. Participants were typically unsure as to how to respond to their family member’s or friend’s requests for understanding or help. Participants reported that friends’ lack of understanding made both parties frustrated or angry, strained or resulted in termination of friendships, with a number of people reporting they no longer had many, if any, close friends.
	Need for validation
	<p>a) Medical community: Participants consistently reported that when they sought help for their condition by health care providers, most health professionals were either relatively ignorant or incredulous of CFS, leading to disbelief in the legitimacy of CFS as a medical entity. Many talked about how they had to screen and select their health care providers based on their willingness to recognise their condition. Others often engaged in activity aimed at informing both health professionals and lay persons about CFS.</p> <p>b) From family & friends: Participants reported that family members, friends and co-workers viewed them as individuals who had no obvious manifestation of symptoms and impairment. This combined with a lack of medical validation meant that family members, friends and co-workers, tended to feel confused, and were prone to question whether the symptoms and impairments were real. Negative responses from family members were distressing, created tension and often threatened relationships. Every participant reported strained relationships with friends as well as having lost relationships as a result of the friends’ lack of understanding of their disability.</p> <p>c) In the workplace: Participants faced incredulity and resentment in the workplace, making them reluctant to report their disability status and request accommodations and tended to push themselves to avoid negative reactions, having to leave their jobs or accepting less-demanding or less-sophisticated work. Because of the constant disbelief participants were extremely ambivalent about the process of getting others to make allowances for them, setting limits or asking for help.</p>
	Need for treatment advice from specialists

Study	Taylor 2005¹³¹
	Participants reported experiences characterised by absence of treatment planning and treatment recommendations. Most reported continued and ongoing dissatisfaction with their treatment when it was administered by a physician that did not specialise in CFS. Along the way they encountered misinformation, misdiagnosis and inappropriate treatment recommendations.
	Support locating & accessing appropriate services
	Most participants described long and frustrating histories of their attempts to access necessary information and services to help them address the consequences of their impairment, reporting they sought treatment for their symptoms and impairments from an average of six physicians before they were ultimately diagnosed. Finding a physician who could provide appropriate services was often tricky.
	Information on benefits & community-based resources and support accessing them
	Participants reported problems acquiring disability income, concerns about requesting workplace accommodation and difficulties accessing community-based resources (such as meal-delivery programs and specialised transportation options). This was because participants had difficulty convincing their physicians of the need for such resources, because they were unaware of these resources or because their health care professionals lacked knowledge of how and why they might benefit from such resources.
Limitations and applicability of evidence	No concerns over methodological limitations. No concerns over applicability.

1

Study	Taylor 2017¹³⁰
Aim	To explore the experiences of young people with 'CFS/ME' and depression in order to understand their views on why low mood developed, the impact of having low mood and what they had found to be helpful and unhelpful in treatment.
Population	Young people aged between 12 and 18 years first assessed between 2013 and 2015 by the Royal National Hospital for Rheumatic Diseases, Bath UK paediatric 'CFS/ME' service (a large regional and national, NHS specialist service); diagnosed with 'CFS/ME' after a thorough assessment which included screening for other disorders associated with fatigue (NICE, 2007; RCPCH, 2004). n=9, 88.9% female; median age (range): 14 (14-15) years; median illness duration (range): 12 (8.5-37.5) months; median HADS depression score (range): 11.5 (10.5-12.5); median HADS anxiety score (range): 12 (9.5-14). Median Chalder fatigue scale score (range): 29 (25-31); mean disability/SF-36 physical function score (range): 45 (30-50)
Setting	Royal National Hospital for Rheumatic Diseases, Bath UK (paediatric 'CFS/ME' specialist service)
Study design	Qualitative interview study
Methods and analysis	Semi-structured interviews were conducted with eight patients at home and one participant on the phone (six were interviewed alone and three with a parent present), using a topic guide focusing on young persons' response to the depression items on the HADS questionnaire, why young people felt they had become low in mood, whether their 'CFS/ME' preceded or followed their low mood,

Study	<p>Taylor 2017¹³⁰</p> <p>factors contributing to low mood, what treatment strategies were helpful and unhelpful and whether anything else would have helped. The topic guide was used flexibly and the interviewer used open questions while following its structure. Interviews were digitally recorded and transcribed verbatim.</p> <p>Data were independently coded by three researchers who met together to combine codes into themes. Where there was little difference in coding between researchers it was discussed further and one researcher led the final decision. Transcripts were thematically analysed on NVivo using techniques of constant comparison.</p>
Findings	<p>Adaptation: Impact of difficulties on normative processes of being a teenager</p> <p>a) Restricted activity: Patients commonly talked about the relationship between ‘CFS/ME’ symptoms, the management of symptoms and changes in activity and mood. Some presented a linear relationship between symptoms (e.g. fatigue, pain, cognitive impairment) which prevented, restricted or interfered with their activities. This impacted their mood; not being able to do activities they fully enjoyed or engage in them fully provoked negative emotions such as low mood, frustration, boredom and hopelessness. Often their negative appraisals meant they were unable to take pleasure from activities they did partake.</p> <p>b) Social changes: All participants talked about the challenges of negotiating their social environments and wider systems (friendship groups, families and the education system) due to ‘CFS/ME’; they described unhelpful behaviours, negative cognitions and difficult emotions related to these challenges. The majority talked about negative experiences with peers as a result of having CFS; highlighted a general inability to sustain social relationships, experienced a lack of understanding negative attitudes and described a failure of their friends to accommodate and adapt to their condition. They described the emotional impact of those social changes and talked about fears of social judgment, perceptions of not fitting in and feelings of loneliness, frustration, anxiety, low mood and loss. Participants described their responses to these challenges, describing strategies such as lack of communication, passive communication, withdrawal and avoidance. Young people talked about the impact of ‘CFS/ME’ on their family and family dynamics and how this made them feel low; about their family suffering as a result of their condition and about increased dependence on their family.</p>
	<p>Peer support from other patients/ Developing a patient network & interpersonal skills:</p> <p>A number of participants indicated that building supportive peer groups, in particular developing connections with others who have ‘CFS/ME’, could be a helpful intervention. Developing interpersonal skills to help their peer groups understand and adapt to the condition was also highlighted. Participants felt building supportive networks, including developing relationships with other young people with ‘CFS/ME’ could be beneficial and talked about the potential of feeling understood and less alone.</p>
	<p>Supportive education systems</p> <p>Young people felt that better support from education systems could have helped. Almost all participants described schools and colleges as inflexible, unhelpful, un-empathetic and invalidating and identified this as a cause of increased anxiety and low mood.</p>
	<p>Information on (Individualised) helpful approaches to managing low mood</p>

Study	Taylor 2017 ¹³⁰
	Young people found different approaches helpful; the importance of an individualised approach was emphasised. Some talked about finding CBT helpful; the combination of CBT with medication was also discussed. Some recognised that AM could be a helpful approach. Young people generally did not mind taking medication providing they found it helpful.
Limitations and applicability of evidence	Minor concerns over methodological limitations due to potential selection bias as recruitment across a multidisciplinary team meant reasons for declining participation were not clear. Minor concerns over applicability due to the study sample having co-morbid low mood potentially limiting their representativeness of the wider ME/CFS population of young people.

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2

1 Appendix E – GRADE-CERQual tables

2 Table 5: Summary of evidence for information education and support for adults with ME/CFS, their families or carers.

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Information about ME/CFS					
7	Semi-structured interviews (5 studies) with thematic analysis (3 studies), constant comparative method (1 study) and grounded theory analysis (1 study); Focus group interviews with grounded theory analysis (1 study); qualitative analysis of open-ended	Patients and carers reported they needed general information about ME/CFS for both themselves and others (families, friends, employers and practitioners), to enable them to develop accurate expectations about the future, relieve feelings of distress caused by the general lack of sufficient information both patients and carers experienced and educate others.	Limitations	Moderate concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	survey responses (1 study)				

- 1 Five studies with very minor to moderate issues; limitations due the potential impact of the researcher on the findings not being discussed in (Hannon 2012; McDermott 2011;
- 2 Ryckeghem 2017), due to a lack of transparency on the analysis process in one study (Ryckeghem 2017), due to concerns over the appropriateness of the data collection
- 3 method of one study that was a follow-up to a quantitative study with open-ended online responses (Devendorf 2018); due to concerns over data analysis due to data
- 4 richness with findings mostly supported by single quotes in two studies (Chew-Graham 2008; Hannon 2012); minor concerns over relevance due to participants in one study
- 5 being self-identified as having ME/CFS rather than having been diagnosed according to accepted criteria (Devendorf 2018) and participants of one study consisting of people
- 6 previously recruited in a RCT (Chew-graham 2008) but no further concerns in any other contributing study.

7

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Type of information: Online & evidence-based					
3	Semi-structured interviews (3 studies) with thematic analysis (1 study) followed by theory-driven analysis (1	While leaflets were reported to be useful for patients and carers at diagnosis, information available online and (an information overview and case studies) in DVD format were reported to facilitate accessibility to all patients, including those who have difficulty to read although some worried the availability of information online would negatively impact the management of ME/CFS in primary care; evidence-based online resources could also benefit patients who wish to bring knowledge to their consultations as well as their social environment; representations	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	Moderate concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	study) and grounded theory analysis (1 study)	of the illness expressed through the media as media personalities also provide helpful sources of evidence for patients.			

1 Three studies with very minor and minor issues; methodological limitations due to the potential impact of the researcher on the findings not being discussed in two studies
2 (Bayliss 2016, Hannon 2012) and due to concerns over data analysis in two studies due to data richness with themes mostly supported by single quotes (Chew-Graham
3 2008, Hannon 2012); moderate concerns about coherence due to differences both between studies and within studies in regards to the types of information patients
4 preferred, with some patients and carers reporting on the usefulness of leaflets (Hannon 2012), others reporting on the usefulness of information available online (Bayliss
5 2016; Chew graham 2008; Hannon 2012) and in DVD format (Bayliss 2016; Hannon 2012), with some worrying online information would negatively impact the management
6 of ME/CFS in primary care (Bayliss 2016) and others talking about alternative evidence sources from the media (Chew graham 2008); very minor concerns over relevance
7 due to participants in one study consisting of people recruited in a RCT (Chew-graham 2008).

8

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Advice on symptom management & treatment options					
7	Semi-structured interviews (4 studies) that included focus groups (1 study)	Patients and carers need guidance on symptom management, including self-help strategies and lifestyle advice and effective and diverse treatment options to choose from including biomedical and alternative treatments other than those currently offered by the health care system, lack of which led to financial costs and even suicidal thoughts.	Limitations	Moderate concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	analysed using thematic analysis (2 studies) , grounded theory approach (1 study), constant comparative method (1 study); qualitative analysis of open-ended survey responses (1 study); secondary analysis of narratives and thematic analysis (1 study); focus group interviews and grounded theory		Adequacy	No concerns about adequacy	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	approach (1 study)				

1 Six studies with very minor to serious issues; limitations due to the potential influence of the researcher on the findings not being discussed in three studies (De Carvalho
2 Leite; Hannon 2012; McDermott 2011) due to concerns over data analysis due to data richness with findings mostly supported by single quotes in two studies (Chew-Graham
3 2008; Hannon 2012), due to concerns over the appropriateness of the data collection method of one study that was a follow-up to a quantitative study with open-ended online
4 responses (Devendorf 2018); limitations related to the use of public data in one study where a lack of detail on the methods of participant recruitment and primary data
5 collection implicated our ability to assess bias (Mihelicova 2016); very minor concerns over relevance due to concerns over one study where participants were self-identified
6 as having ME/ (Devendorf 2018) and one study where parent carers of ME/CFS patients were not limited to people caring for adults (Mihelicova 2016), being counterbalanced
7 by the fact that the same information emerged from studies with no such concerns, one of which included patients from diverse social and ethnic backgrounds, degrees of
8 illness severity and duration (De Carvalho Leite 2011).

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Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Need for understanding & advocacy					
8	Semi structured interviews (5 studies) and focus groups (1 study) analysed	Patients and carers needed understanding and recognition of ME/CFS from the medical community, employers, colleagues, family and friends, lack of which was unhelpful in the workplace, in accessing healthcare and social support and implicated their well-being and social relationships.	Limitations	Moderate concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	with thematic analysis (4 studies), followed by theory-driven analysis (1 study) or analysed with grounded theory (1 study); qualitative analysis of open-ended survey responses (1 study); secondary analysis of interview narratives and thematic analysis (1 study); focus groups and grounded theory approach (1 study).		Adequacy	No concerns about adequacy	

1 Six studies with very minor to serious issues; methodological limitations due to the role of the researcher not being discussed in four studies (Bayliss 2016, De Carvalho Leite,
 2 Hannon 2012, Ryckeghem 2017), concerns over the appropriateness of the data collection method due to one study being a follow-up of a quantitative study with data
 3 emerging from open-ended online survey answers (Devendorf 2018), concerns about data analysis due to data richness with themes mostly supported by single quotes in
 4 one study (Hannon 2012) and lack of transparency on the analysis process (Ryckeghem 2017), limitations related to the use of public data in one study where a lack of detail
 5 on the methods of participant recruitment and primary data collection implicated our ability to assess bias (Mihelicova 2016); very minor concerns about relevance with
 6 concerns over one study where participants were self-identified as having ME/CFS (Devendorf 2018) and parent carers of ME/CFS patients in one study (Mihelicova 2016)
 7 not being limited to people caring for adults being counterbalanced by the fact that the same information emerged from studies with no such concerns, which included
 8 ME/CFS patients from diverse social and ethnic backgrounds, degrees of illness severity and duration (De Carvalho Leite 2011)

9

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Support with acceptance					
1	Semi-structured interviews and thematic analysis	Patients need support with acceptance of their new life circumstances, their diagnosis and the likely prognosis of their illness in order to maximise the benefit received from treatment.	Limitations	No concerns about methodological limitations	MODERATE
			Coherence	Minor concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Minor concerns about adequacy	

10 One study with no particular methodological issues; minor concerns about coherence due to the theme not clearly emerging from the data as the importance of acceptance
 11 and willingness to change were highlighted but not explicitly identified as areas patients need support with; minor concerns about adequacy with sufficient information on the
 12 topic available from the study.

13

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Support during difficult phases of ME/CFS					
3	Semi-structured interviews and thematic analysis (2 studies) and secondary analysis of narrative interviews using thematic analysis (1 study)	The time following diagnosis of ME/CFS and the early stages of treatment were difficult times for patients who experienced feelings of distress, difficulty with acceptance of diagnosis, of advice given and occasionally a lack of guidance while phases of relapse were particularly difficult for carers.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	Minor concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	Minor concerns about adequacy	

- 1 Two studies with moderate and serious issues; methodological associated with the use of public data in one study, where lack of detail on the method of primary data
- 2 collection and participant recruitment limited our ability to assess the risk of bias (Mihelicova 2016), due to the potential influence of the researcher on the findings not being
- 3 discussed, concerns over data analysis due to a lack of sufficient information on the data analysis process and concerns over data richness with findings mostly supported by
- 4 single quotes in one study (Ryckeghem 2017); minor concerns about coherence due to the finding emerging from three studies and participants in different studies reporting
- 5 difficulty or a need for support with different phases of the illness: post diagnosis (two studies), relapse (one study); very minor concerns over relevance due to participants in
- 6 one study being carers of patients of various ages that were not limited to the adult age stratum (Mihelicova 2016); minor concerns about adequacy due to issues with data
- 7 richness in one study.

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Support from specialists					
4	Semi-structured interviews and thematic analysis (2 studies) that was followed by theory-driven analysis (1 study); semi-structured interviews and constant comparative method (1 study); focus group interviews and grounded theory analysis (1 study)	Patients hoped to be referred to specialist services to overcome the barriers to diagnosis and treatment they encountered in primary care and those who ultimately were, had benefited in ways including diagnosis, validation and information provision.	Limitations	Very minor concerns about methodological limitations	HIGH
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	No concerns about adequacy	

1 Two studies with very minor issues; limitations due to the potential influence of the researcher not being discussed (Bayliss 2016; McDermott 2011).

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Help accessing support					
4	Semi-structured interviews and grounded theory approach (1 study) that included focus groups and thematic analysis (1 study); qualitative analysis of open-ended survey responses (1 study); Focus group interviews and grounded theory (1 study)	Patients and carers needed sign posting to relevant contacts that would help address the consequences of ME/CFS, including knowledgeable physicians, information on existing benefits and support they were entitled to and help accessing them.	Limitations	Minor concerns about limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	No concerns about adequacy	

- 1 Three studies with very minor to moderate issues; methodological limitations due to concerns over the appropriateness of the data collection method in one study being a
- 2 follow-up to a quantitative study with open-ended online responses (Devendorf 2018); concerns over data analysis due to data richness with themes mostly supported by
- 3 single quotes in one study (Hannon 2012); due to the potential influence of the researcher on the findings not being discussed in two studies (De Carvalho Leite 2011;

- 1 Hannon 2012); very minor concerns over relevance due to participants in one study being self-identified as having ME/CFS (Devendorf 2018) but no concerns over the
- 2 representativeness of the sample in other contributing studies.
- 3

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Financial support					
2	Semi-structured interviews and focus groups with thematic analysis (1 study) and qualitative analysis of open-ended survey responses (1 study)	Financial support was crucial for the illness and life- management of ME/CFS and social relationships while having to work due to a lack of financial resources or financial constrains arising due to a lack of other forms of support, had physical consequences and negatively influenced coping.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	Minor concerns about adequacy	

- 4 Two studies with very minor and moderate issues; methodological limitations due to concerns over the appropriateness of the data collection method in one study being a
- 5 follow-up to a quantitative study with open-ended online responses (Devendorf 2018), due to the potential impact of the researcher on the findings not being discussed in one
- 6 study (De Carvalho Leite 2011); minor concerns over relevance due to participants in one study being self-identified as having ME/CFS (Devendorf 2018) but the theme also
- 7 reported by patients. from diverse social and ethnic backgrounds, degrees of illness severity and duration (De Carvalho Leite 2011); minor concerns over adequacy with
- 8 limited information supporting the finding in one study (Devendorf 2018).

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Ongoing health professional support					
4	Semi-structured interviews and thematic analysis (4 studies) that was followed by theory-driven analysis (1 study)	Patients identified the need to establish a close, ongoing relationship with their health professional who would be involved in the long-term management of ME/CFS and accompany and advise them at all stages of the care process.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	No concerns about adequacy	

- 1 Three studies with very minor to moderate issues; methodological limitations due to the potential impact of the researcher on the findings not being discussed in two studies
- 2 (Bayliss 2016; Ryckeghem 2017) and lack of sufficient information on the data analysis process in one study (Ryckeghem 2017), concerns over data richness with some
- 3 findings supported by single quotes in two studies (Chew-Graham 2008; Ryckeghem 2017).

4

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Social Support					
4	Semi-structured interviews (3)	Patients and carers felt isolated, struggling to maintain social relationships and could benefit from social interactions and mutual patient support.	Limitations	Minor concerns about methodological limitations	MODERATE

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	studies) with grounded theory approach (1 study), constant comparative method (1 study) and thematic analysis (1 study) and qualitative analysis of open-ended survey responses (1 study)		Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	No concerns about adequacy	

1 Three studies with very minor to moderate issues; limitations due to the potential influence of the researcher not being discussed in two studies (Hannon 2012, McDermott
2 2011), concerns over data analysis due to data richness with themes mostly supported by single quotes in one study (Hannon 2012), concerns over the appropriateness of
3 the data collection method in one study being a follow-up to a quantitative study with open-ended online responses (Devendorf 2018); minor concerns over relevance due to
4 participants in one study being self-identified as having ME/CFS (Devendorf 2018) but no similar concerns identified in any other contributing study

5

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Practical support with daily living & social care					
2	Semi-structured interviews and focus groups and thematic analysis (1 study); qualitative analysis of open-ended survey responses (1 study)	Patients needed practical support for both themselves and their carers, including help with personal and domestic tasks and access to social care, lack of which implicated their ability to manage their lives and maintain their family roles.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	No concerns about adequacy	

1 Two studies with very minor and moderate issues; methodological limitations due to concerns over the appropriateness of the data collection method in one study being a
2 follow-up to a quantitative study with open-ended online responses (Devendorf 2018), the potential influence of the researcher not being discussed (De Carvalho Leite 2011);
3 very minor concerns about relevance as although patients of one study were self-identified as having ME/CFS (Devendorf 2018), the sample of the other contributing study,
4 where the majority of the information supporting the theme emerged from, included patients from diverse social and ethnic backgrounds, degrees of illness severity and
5 duration recruited through ME/CFS support groups and selected by clinicians and there were no concerns over the accuracy of their diagnosis (De Carvalho Leite 2011).

6

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Need for tailored and accessible hospital care					
3	Semi-structured interviews (3 studies) that also involved focus groups (1 study), analysed using thematic analysis (2 studies) and grounded theory approach (1 study)	Patients emphasised the importance of hospital care that would be tailored to their needs, the severity of their symptoms and other commitments, in terms of accessibility and flexibility in the frequency, the duration and the mode of attendance to medical appointments and treatment.	Limitations	Very minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Minor concerns about adequacy	

- 1 Two studies with very minor to minor issues; methodological limitations due to the role of the researcher not being discussed in two studies (De Carvalho Leite 2011, Hannon 2012) and concerns over data analysis due to data richness, with findings supported by single quotes in one study (Hannon 2012); minor concerns over adequacy with limited
- 2 information supporting the finding in one study (Hannon 2012).
- 3

4

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Need for a diagnosis					
2	Semi structured interviews and constant comparative method (1 study); Semi-structured interviews and focus group discussions and thematic analysis (1 study)	A diagnosis of ME/CFS- that was often gained through private or alternative health services- enabled patients to gain advice from health professionals and take action towards symptom improvement while a lack of a diagnosis exacerbated their psychological and financial pressures and impeded their access to social services.	Limitations	Very minor concerns about methodological limitations	HIGH
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	No concerns about adequacy	

1 Two studies with very minor issues; methodological limitations due the role of the researcher not being discussed in both studies (De Carvalho Leite 2011, McDermott 2011)

2

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Need for a positive diagnosis and future direction					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
3	Semi-structured interviews with thematic analysis followed by theory driven analysis (1 study), constant comparative method (1 study) and grounded theory approach (1 study).	Patients reflected on the importance of a positive direction for the future and on the need for the ME/CFS diagnosis to be framed in a positive way to enable them to maintain hope for improvement.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Minor concerns about adequacy	

- 1 Three studies with very minor to minor issues; methodological limitations due to the role of the researcher not being discussed in three studies (Bayliss 2016, Hannon 2012
- 2 McDermott 2011), due to concerns over data analysis in one study due to issues with data richness and themes mostly supported by single quotes (Hannon 2012); minor
- 3 concerns about adequacy with limited information supporting the theme in one study (Hannon 2012).

4

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Patient support during medical consultations					
1	Semi-structured interviews and grounded theory approach	GP consultations were challenging for patients who could benefit from the presence of their caregiver.	Limitations	Minor concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

1 One study with minor issues; limitations due to the potential influence of the researcher not being discussed and data analysis with concerns over data richness as themes were mostly supported by single quotes in the contributing study (Hannon 2012); moderate concerns over adequacy with relatively limited information supporting the theme.

3

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Advice (for carers) on how to support patients					
1	Semi-structured interviews and grounded	Carers need advice on how to support patients with ME/CFS as they lacked an understanding of the illness and discussed the danger of giving inappropriate advice or support to patients, exacerbating their symptoms.	Limitations	Minor concerns about methodological limitations	VERY LOW
			Coherence	No concerns about coherence	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	theory approach		Relevance	No concerns about relevance	
			Adequacy	Serious concerns about adequacy	

- 1 One study with minor issues; limitations due to the potential influence of the researcher on the findings not being discussed and data analysis with concerns over data
- 2 richness as themes were mostly supported by single quotes in the contributing study (Hannon 2012); serious concerns over adequacy with very limited information supporting
- 3 the theme.

4 Table 6: Summary of evidence for information education and support for children and young people with ME/CFS, their families or
5 carers

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Information about ME/CFS					
5	Semi-structured interviews and thematic analysis (4 studies) and secondary analysis of interview narratives	A lack of information about ME/CFS and particularly about the symptoms and prognosis of the illness reported by both adolescent patients and their parents contributed to negative and undesirable feelings of uncertainty, fear and worry for both groups of people, while learning about the condition was reported to be helpful, contributing to feelings of validation in adolescents.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	No concerns about adequacy	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	and thematic analysis (1 study).				

1 Five studies with minor to serious issues; overall moderate concerns over methodological limitations due to the role of the researcher not being discussed in three studies
 2 (Beasant 2014, Brigden 2018, Harris 2017), due to the use of public data in one study, where the lack of detail on the primary data collection methods and participant
 3 recruitment impacted our ability to assess risk of bias (Mihelicova 2016), lack of sufficient detail over the data analysis process (Brigden 2018), concerns over data richness in
 4 one study (Beasant 2014) and due to the very small sample size and homogenous population of one study (Jelbert 2010) ; moderate concerns over relevance with minor
 5 concern identified in all contributing studies, due to one study including parent carers of ME/CFS patients that were not limited to the children and young people age stratum
 6 (Mihelicova 2016), due to the very small sample size and homogenous population of people who attended the same clinic that was limited to people that had recovered
 7 (Jelbert 2010), the sample of one study being limited to people with ME/CFS who experienced eating difficulties (Harris 2017), two studies excluding severely affected
 8 individuals (Beasant 2014, Brigden 2018) and the sample in one study consisting of participants from a feasibility RCT (Beasant 2014).

9

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Sources of information: need for digital resources					
1	Semi-structured interviews and thematic analysis	Adolescents used website resources they considered reliable but preferred patient-led sites and social media that contained experiential account which provided a great source of support that could be accessed at any time and facilitated coping.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
			Adequacy	No concerns about adequacy	

1 One study with minor issues; methodological limitations due to the role of the researcher not being discussed and lack of details on the analysis process followed (Brigden
2 2018); minor concerns over relevance as the study excluded severely affected individual.
3

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Types of information					
1	Semi-structured interviews and thematic analysis	Having in-depth, reliable and relatable, information that is patient friendly and regularly updated can be therapeutic for adolescents.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	No concerns about adequacy	

4 One study with minor issues; methodological limitations due to the role of the researcher not being discussed and lack of details on the analysis process followed (Brigden
5 2018); minor concerns over relevance as the study excluded severely affected individuals.
6

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Need for validation & advocacy					
4	Semi-structured interviews and thematic analysis (3 studies) and secondary analysis of interview narratives and thematic analysis (1 study).	A lack of understanding and validation of their experience from both medical professionals and their social environment was reported by young people with ME/CFS who felt the need to talk about their illness and often utilised online resources to help others understand, while recognition and acknowledgment of the condition from specialist services resulted in a sense of relief in adolescents and parents.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	No concerns about adequacy	

1 Three studies with minor issues and one study with serious issues; methodological limitations due to the role of the researcher not being discussed in two studies (Beasant
2 2014, Brigden 2018), lack of sufficient detail on the data analysis process in one study (Brigden 2018, concerns over data richness in one study (Beasant 2014), due to the
3 very small sample size and homogenous population of people who attended the same clinic in one study (Jelbert 2010) and concerns related to the use of public data where
4 a lack of detail on participant recruitment and the primary data collection methods implicated our ability to assess risk of bias (Mihelicova 2016); moderate concerns over
5 relevance due to the exclusion of severely affected individuals from two studies (Beasant 2014, Brigden 2018), one study consisting of people who had been previously
6 recruited in an RCT (Beasant 2014), one study including parent carers of ME/CFS patients that were not limited to the children and young people age stratum (Mihelicova
7 2016), due to the aforementioned very small sample size and homogenous population of one study that was also limited to people that had recovered, whose views may differ
8 from those with active ME/CFS (Jelbert 2010).

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Support with acceptance & adaptation					
4	Semi-structured interviews and thematic analysis (2 studies); secondary analysis of interview narratives and thematic analysis (1 study); focus groups and grounded theory approach (1 study)	The acceptance of and adaptation to the lifestyle changes brought by ME/CFS, including changes in their identity, activities and social relationships, appeared challenging for young patients and carers and often impacted their mood and well-being.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	No concerns about adequacy	

1 Three studies with minor issues and one study with serious issues; limitations due to the role of the researcher not being discussed in two studies (Beasant 2014; Brigden
2 2018), lack of sufficient detail on the data analysis process in one study (Brigden 2018), concerns over data richness in one study (Beasant 2014), concerns over potential
3 selection bias in one study as recruitment across a multidisciplinary team meant reasons for declining participation were not clear (Taylor 2017), and concerns related to the
4 use of public data in one study where a lack of detail on participant recruitment and the primary data collection methods implicated our ability to assess risk of bias (Mihelicova
5 2016); serious concerns over relevance as severe cases of ME/CFS were not included in two studies (Beasant 2014; Brigden 2018) and the sample of one study consisting of
6 participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the RCT (Beasant 2014), one study specifically included ME/CFS patients with co-
7 morbid low mood (Taylor 2017) and one study included carers of ME/CFS patients that were not limited to the children and young people age stratum (Mihelicova 2016).

8

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Management strategies & support with implementation					
2	Semi-structured interviews and thematic analysis (1 study) and secondary analysis of interview narratives and thematic analysis (1 study).	A lack of resources for management was reported by parent-carers and where available, specialist guidance on management of ME/CFS and medical care strategies- although potentially difficult to integrate- were reported to positively impact the life of both young people and their families.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	Minor concerns about adequacy	

- 1 Two studies with minor and serious issues; methodological limitations due to the role of the researcher and concerns over data richness in one study (Beasant 2014), and
 - 2 associated with the use of public data and lack of detail on how the sample was derived, the methods of primary data collection and inability to assess bias (Mihelicova
 - 3 2016); minor concerns over relevance with the sample of one study consisting of parent carers that were not limited to the stratum of children and young people (Mihelicova
 - 4 2016) and the sample of the other study consisting of participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the RCT (Beasant 2014);
 - 5 minor concerns over adequacy due to concerns over data richness.
- 6

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Mutual patient support					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
3	Semi-structured interviews and thematic analysis (3 studies)	Young people with ME/CFS reported on the importance of developing a connection with other patients either online or offline which provided support, alleviated feelings of isolation and benefited recovery.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	No concerns about adequacy	

1 Three studies with minor issues; limitations due to the role of the researcher not being discussed and lack of details on the analysis process followed (Brigden 2018), due to
 2 the very small sample size and homogenous population of people who attended the same clinic in one study (Jelbert 2010) and due to potential selection bias in the other
 3 study as recruitment across a multidisciplinary team meant reasons for declining participation were not clear (Taylor 2017); moderate concerns over relevance due to the
 4 population of one study excluding severely affected individuals (Brigden 2018), all participants having recovered from ME/CFS and potentially holding different opinions to
 5 those in the active stage of ME/CFS in one study (Jelbert 2010) and participants in one study having co-morbid low mood potentially holding different opinions to those who
 6 do not (Taylor 2017).

7

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Educational support					
2	Semi-structured interviews	Lack of support within young peoples' educational environment was a source of anxiety and low mood, while better communication between healthcare and education providers	Limitations	Minor concerns about methodological limitations	LOW

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
	and thematic analysis (2 studies)	enabled patients and carers to gain support and accommodate their needs.	Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

- 1 Two studies with minor issues; methodological limitations due to the role of the researcher not being discussed and concerns over data richness in one study (Beasant 2014),
 2 potential selection bias in the other study as recruitment across a multidisciplinary team meant reasons for declining participation were not clear (Taylor 2017); minor concerns
 3 over relevance due to the sample of one study consisting of participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the RCT (Beasant
 4 2014) and due to patients in the other study having co-morbid low mood, potentially limiting the representativeness of their views for ME/CFS patients without low mood
 5 (Taylor 2017); moderate concerns over adequacy with relatively limited information supporting the theme in the contributing studies.

6

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Need for a diagnosis					
2	Semi-structured interviews and thematic analysis (2 studies)	The period before receiving a formal diagnosis was particularly difficult for adolescents with ME/CFS and their carers, while gaining a diagnostic label facilitated steps towards recovery and access to educational support.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
			Adequacy	Minor concerns about adequacy	

- 1 Two studies with minor issues; methodological limitations due to the role of the researcher not being discussed and concerns over data richness in one study (Beasant 2014),
- 2 due to the small sample size and homogenous population of participants who attended the same clinic of the other study (Jelbert 2010); minor concerns over relevance due to
- 3 the sample of one study consisting of participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the RCT (Beasant 2014), due to participants
- 4 in the other study having recovered from ME/CFS and therefore possibly not holding similar opinions to those in the stage of active ME/CFS (Jelbert 2010); minor concerns
- 5 about adequacy due to the relatively limited information supporting the finding in the studies.

6

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Tailored approach to care					
2	Semi-structured interviews and thematic analysis (2 studies)	The importance of a tailored treatment approach and its impact on recovery was highlighted by adolescent patients and adolescent patient mothers.	Limitations	Minor concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

1 Two studies with minor issues; methodological limitations due to the role of the researcher not being discussed and concerns over data richness in one study (Beasant 2014),
 2 potential selection bias in the other study as recruitment across a multidisciplinary team meant reasons for declining participation were not clear (Taylor 2017); minor
 3 concerns over relevance with minor concerns in both contributing studies due to the sample in one study consisting of participants from a feasibility RCT which may differ from
 4 ME/CFS patients not recruited in the RCT (Beasant 2014) and because patients in the other study had co-morbid low mood which potentially limits the representativeness of
 5 their views for ME/CFS patients without low mood and it is unclear whether the information on the importance of an individualised approach to care reported is in relation to
 6 their ME/CFS or low mood (Taylor 2017); moderate concerns over adequacy with relatively limited information supporting the theme.

7

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Referral to specialist services					
1	Semi-structured interviews and thematic analysis	Young people with ME/CFS and their mothers reported how accessing specialist ME/CFS services had benefited them in terms of diagnosis, information and guidance, psychological support, symptom management, treatment and recovery as well as in terms of access to educational support.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	No concerns about adequacy	

8 One study with minor issues; methodological limitations due to the role of the researcher not being discussed and concerns over data richness (Beasant 2014); moderate
 9 concerns over applicability with referral to specialist services emerging as a positive and useful experience exclusively from a study aiming to examine that and by people who
 10 had accessed a specialist service and not reported as a need of young people with ME/CFS in other studies

11

12

1 Appendix F – Excluded studies

2 Table 7: Studies excluded from the qualitative review

Reference	Reason for exclusion
Aikman 1995 ¹	Unable to obtain paper
Anderson 1997 ²	No relevant themes
Anderson 2012 ⁴	Incorrect study design (non-PICO systematic review)
Antcliff 2018 ⁵	No relevant themes
Arrol 2008 ⁶	No relevant themes
Asbring 2001 ⁷	Incorrect population: included CFS patients but majority had fibromyalgia diagnosis and results were not analysed separately
Asbring 2002 ⁹	Incorrect population: mixed CFS and fibromyalgia patients
Asbring 2004 ⁸	Incorrect population: mixed CFS and fibromyalgia patients
Ashby 2006 ¹⁰	No relevant themes
Ax 1998 ¹²	No relevant themes
Beaulieu, 2000 ¹⁹	No relevant themes
Bayliss 2014 ¹⁴	Incorrect study design (non-PICO systematic review)
Bayliss 2014 ¹⁶	Not a primary study
Bazelmans 2005 ¹⁷	Incorrect study design: questionnaire; no extractable themes
Bennett 2007 ²⁰	No extractable themes; analysis focused on the identification of symptoms experienced by patients.
Brady 2016 ²¹	Incorrect population: mixed population of people with ME/CFS and type 1 and 2 diabetes
Brooks 2012 ²⁴	Incorrect study design: included interviews but findings are based on questionnaire i.e. cross-sectional data with no qualitative analysis; no relevant themes
Bulow 2003 ²⁶	Incorrect population: interviews of patients with CFS or a related diagnosis in which fatigue was a significant part of their suffering
Caplan 2001 ²⁷	Incorrect study design: patient story
Chernow 2008 ²⁸	Unable to obtain paper
Cheshire 2020 ²⁹	No relevant themes
Chew-Graham 2010 ³¹	Population did not meet protocol: GPs
Clarke 1999 ³³	No relevant themes
Clements 1997 ³⁵	No relevant themes
Costello 1998 ³⁶	Unable to obtain paper
Gan 2010 ⁵²	Incorrect population: caregivers of people with acquired brain injury
Davison 1997 ³⁷	Incorrect study design: article
De Silva 2013 ³⁹	Not primary study
Dennison 2010 ⁴⁰	No relevant themes
Devendorf 2017 ⁴²	Population does not match protocol: ME/CFS specialists
Devendorf 2019 ⁴¹	Population does not match protocol: ME/CFS specialists
Drachler 2009 ⁴⁵	Incorrect study design (non-PICO systematic review)
Everett 2002 ⁴⁷	Population does not match protocol: school teachers of adolescents with ME/CFS
Fowler 2005 ⁴⁹	No relevant themes; incorrect population: inadequate definition (children experiencing 'disabling fatigue' classified as CFS).
Friedberg 1998 ⁵¹	Unable to obtain paper

Reference	Reason for exclusion
Friedberg 2016 ⁵⁰	Incorrect population: majority diagnosed with unexplained chronic fatigue, not CFS; no relevant themes
Gotts 2016 ⁵⁴	No relevant themes
Gray 2003 ⁵⁵	No relevant themes
Guise 2007 ⁵⁷	No relevant themes
Guise 2010 ⁵⁶	No relevant themes
Hareide 2011 ⁵⁹	No relevant themes
Harris 2016 ⁶⁰	Incorrect study design (non-PICO systematic review)
Higginson 2008 ⁶³	Incorrect population: not ME/CFS
Horrocks 2015 ⁶⁴	Unable to obtain paper
Hart 2000 ⁶²	No relevant themes
Horton 2010 ⁶⁷	Population does not match protocol: Health professionals
Horton-Salway 2002 ⁶⁵	Incorrect study design: article
Horton-Salway 2004 ⁶⁶	Incorrect study design: article
Jason 2015 ⁶⁸	Incorrect study design: article
Jensen 2001 ⁷⁰	Unable to obtain paper
Keech 2015 ⁷¹	No relevant themes; study employed qualitative methods to devise a self-reported psychometric measure for fatigue
Kendrick 2016 ⁷²	Incorrect study design: questionnaire measures
Kisely 2002 ⁷³	Incorrect study design: evaluation of web-based information
Larun 2007 ⁷⁵	Incorrect study design (non-PICO systematic review)
Larun 2011 ⁷⁴	No relevant themes
Lee 2000 ⁷⁶	Unable to obtain paper
Lee 2001 ⁷⁷	Incorrect population: insufficient definition of CFS, patients described to have chronic fatigue and weakness
Levine 1997 ⁷⁸	Analysis does not meet protocol: quantitative analysis and no extractable themes.
Lian 2016 ⁷⁹	No relevant themes
Lin 2009 ⁸⁰	No relevant themes
Littrel 2012 ⁸²	Unable to obtain paper
Lingard 2014 ⁸¹	No relevant themes
Lombaard 2005 ⁸³	No relevant themes
Marks 2016 ⁸⁵	Population does not match protocol: Health professionals
McInnis 2015 ⁸⁸	Incorrect population: included CFS patients but majority had fibromyalgia diagnosis rather than CFS and results were not analysed separately
Moore 2000 ⁹¹	Incorrect study design: combination of quantitative and qualitative methodology with results from statistical and thematic analysis not reported separately and no extractable themes.
Njolstad 2019 ⁹³	No relevant themes
Olson 2015 ⁹⁴	No relevant themes
Ong 2005 ⁹⁵	No relevant themes
Parslow 2015 ⁹⁶	No relevant themes
Parslow 2017 ⁹⁸	Incorrect study design (non-PICO systematic review)
Parslow 2017 ⁹⁹	No relevant themes
Parslow 2018 ⁹⁷	No relevant themes
Pemberton 2014 ¹⁰¹	No relevant themes

Reference	Reason for exclusion
Pemberton 2014 ¹⁰⁰	No relevant themes
Picariello 2017 ¹⁰²	No relevant themes
Pinikahana 2002 ¹⁰³	No relevant themes
Pinxsterhuis 2015 ¹⁰⁵	Incorrect study design (non-PICO systematic review)
Pinxsterhuis 2015 ¹⁰⁴	No relevant themes
Prins 2000 ¹⁰⁶	Analysis does not meet protocol: qualitative responses used to support quantitative questionnaire analysis; no extractable themes
Raine 2004 ¹⁰⁷	Incorrect population: GPs perceptions of CFS and irritable bowel syndrome; no relevant themes
Ray 1995 ¹⁰⁹	Incorrect study design: questionnaires and quantitative analysis
Ray 1998 ¹⁰⁸	No relevant themes
Reme 2013 ¹¹⁰	No relevant themes
Reynolds 2006 ¹¹²	Incorrect study design: qualitative analysis of three narratives: o relevant themes
Reynolds 2008 ¹¹³	No relevant themes
Reynolds 2010 ¹¹¹	Incorrect population: self-reported ME/CFS that was not confirmed; no relevant themes
Richards 1998 ¹¹⁵	Incorrect study design: questionnaires and no qualitative analysis to allow the extraction of themes
Richards 2002 ¹¹⁴	No relevant themes
Sachs 2001 ¹¹⁷	Incorrect analysis: no thematic analysis, mostly observations from attending consultations and their transcripts
Saltzstein 1998 ¹¹⁸	Incorrect analysis: semi structured interview results reported quantitatively
Schoofs ¹¹⁹	Incorrect population: mixed ME/CFS and fibromyalgia population
Sidi-Ali-Mebarek 2009 ¹²⁰	No relevant themes
Snell 2001 ¹²¹	Incorrect study design: Qualitative case study of two patients with no extractable themes
Soderlund 2000 ¹²³	No relevant themes
Soderlund 2005 ¹²²	No relevant themes
Son 2015 ¹²⁴	No relevant themes; specific to Traditional Korean Medicine (TKM) or focused on symptoms/experience
Stenhoff 2015 ¹²⁵	Incorrect population: medical students
Stormorken 2015 ¹²⁶	No relevant themes
Sturge-Jacobs 2002 ¹²⁷	Incorrect population: people with Fibromyalgia
Sunnquist 2017 ¹²⁸	Incorrect study design: quantitative (survey)
Swoboda 2006 ¹²⁹	Incorrect population: mixed population of self-identified people with CFS, multiple chemical sensitivities and Gulf War Syndrome
Tevens 2004 ¹³³	Incorrect population: women with fibromyalgia and CFS
Theorell 1999 ¹³⁴	Incorrect study design: reports questionnaire results quantitatively only
Travers 2008 ¹³⁵	No relevant themes
Tuck 1998 ¹³⁶	No relevant themes
Tuck 2000 ¹³⁷	Incorrect study design: questionnaires and no qualitative analysis to allow the extraction of themes
Ward 2008 ¹³⁹	No relevant themes
Ware 1993 ¹⁴⁰	No relevant themes
Ware 1998 ¹⁴¹	No relevant themes

Reference	Reason for exclusion
Ware 1999 ¹⁴²	No relevant themes
Whitehead 2006 ¹⁴³	No relevant themes
Whitehead 2006 ¹⁴⁴	No relevant themes
Williams 2016 ¹⁴⁵	No relevant themes
Wilson 2011 ¹⁴⁶	Incorrect population: experiencing chronic fatigue due to long-term conditions other than chronic fatigue syndrome

1

2 **Table 8: Studies identified but not included in the qualitative review due to saturation**
3 **being reached**

Reference
Anderson 2014 ³
Ax 1997 ¹³
Ax 2002 ¹¹
Clarke 2000 ³⁴
Chew-Graham 2011 ³⁰
Donalek 2009 ⁴⁴
Edwards 2007 ⁴⁶
Fisher 2013 ⁴⁸
Gilje 2008 ⁵³
Lovell 1999 ⁸⁴
McCue 2004 ⁸⁶
Missen 2012 ⁹⁰
Velleman 2016 ¹³⁸
Winger 2014 ¹⁴⁷
Woodward 1995 ¹⁴⁸

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