National Institute for Health and Care Excellence

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

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

NICE guideline NG201

Evidence reviews underpinning recommendations and research recommendations in the NICE guideline

August 2021

Final

These evidence reviews were developed by the National Guideline Centre

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

1.1 Review question

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

1.1.1 Introduction

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

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

1.1.2 Summary of the protocol

For full details see the review protocol in Appendix A.

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.

1.1.3 Methods and process

This evidence review was developed using the methods and process described in Developing NICE guidelines: the manual. Methods specific to this review question are described in the review protocol in appendix A and the methods document.

Declarations of interest were recorded according to NICE's conflicts of interest policy.

1.1.4 Qualitative evidence

1.1.4.1 Included studies

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

Adults

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

Children and young people

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

In line with the review protocol the evidence relevant to adults is reported separately to children and young people. The severity of ME/CFS was mixed or unclear.

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

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

1.1.4.2 Excluded studies

See the excluded studies in Appendix F.

1.1.5 Summary of studies included in the qualitative evidence

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.	Patients (n=11), mean age (range): 46 (27 to 71) years; GPs (n=8) 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. UK Stratum: adults, severity mixed or unclear	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.	In line with the review protocol, themes emerging from the information provided by the patient population have only been extracted. 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. 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.
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
		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. 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. UK Stratum: children and young people, mild/moderate severity	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'.	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.
Brigden 2018[22]	In-depth semi-structured interviews (face-to-face or via Skype) and thematic analysis.	Adolescents recruited from a specialist paediatric 'CFS/ME' service. N=9; male/female: 3/6; mean age (SD): 14.89 (1.9) years, at different	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.	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

Study	Design	Population	Research aim	Comments
		stages of the condition; mean number of months from initial assessment to interview (SD): 12.98 (7.98), range 4 to 25) months. UK Stratum: children and young people, severity mixed or unclear		participate in an interview or severely affected.
Broughton 2017[25]	Semi-structured interviews (six face-to-face, 10 via telephone) and thematic analysis. Cross-sectional design using opportunity sampling.	Adults who were completing treatment for ME/CFS at one of three outpatient NHS specialist 'CFS/ME' services. 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.	To explore the experiences of 'CFS/ME' patients who were completing programmes of treatment at three NHS specialist 'CFS/ME' services in England.	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

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
		severity, duration, social variation (age, gender, ethnic background and socio-economic conditions) and year of diagnosis. UK Stratum: adults, severity mixed or unclear	those from ethnic minorities, and what challenges may be posed to health care practitioners in providing appropriate and equitable care for this condition.	severity, for both an initial focus group discussion and a later one-to-one interview. 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.
Devendorf 2018[43]	Mixed-methods design; qualitative analysis of participants' open-ended survey responses from a previous project that examined illness severity, stigma, physician interactions and depression.	Patients who self-identify as having ME/CFS and endorsed suicidal ideation (SI) but did not meet depression criteria 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). USA	An exploratory study to explore the relationship between ME/CFS and suicidal ideations, including quality of life, loss of function, isolation and hopelessness.	The study was hosted online using Research Electronic Data Capture.

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
				participants were identified as being eligible by their clinician. Adolescents who were housebound i.e. potentially severe/ very severe cases were excluded.
Jelbert 2010[69]	Semi-structured interviews and thematic analysis (interpretative phenomenological analysis).	Five adolescents who were considered to have recovered from ME/CFS. 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. UK Stratum: children and young people, severity mixed or unclear	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 transmural 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
		Patients (n=15); median age (range): 45 (33-59 years); GPs (n=15); median age (range): 49 (31-62 years). Belgium Stratum: adults, severity mixed or unclear	in the Belgian provinces of East and West Flanders.	
Taylor 2005[131]	Focus group interviews, open-ended questionnaires, progress notes, and from a program evaluation questionnaire.	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. 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 *et al* (1994) criteria for ME/CFS.	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.	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.

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.

See Appendix D for full evidence tables.

1.1.6 Qualitative evidence synthesis

1.1.6.1 Summary of the qualitative evidence

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, 131]	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 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 constrains 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.

See Appendix E for full GRADE-CERQual tables.

1.1.6.2 Narrative summary of review findings

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

Review finding 1: Information about ME/CFS

Patients and carers felt they had a limited understanding of 'CFS/ME' and lacked general information about the illness, having received little or conflicting information. They report not having a sufficient overview of what to expect. This often led to false hopes and expectations about the illness, treatment and prognosis and led to distress and worry. 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. Patients who expressed confidence in their level of knowledge about 'CFS/ME' also tended to express less anxiety about the future and less concern or distress about waiting for an appointment with specialist services. Understanding the complexity of the illness, especially the interaction between different factors, was perceived by many as a route out of this cycle of fatigue. Some patients also expressed hopes that specialist services would be able to advise them on which factors were relevant to the illness.

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

Explanation of quality assessment: Moderate concerns over methodological limitations with moderate concerns in two studies (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 responses and due to the potential influence of the researcher on the findings not being discussed and lack of transparency on the analysis process in the other study), minor concerns over two studies (due to the potential influence of the researcher on the findings not being discussed in one study and concerns over data analysis due to data richness with findings mostly supported by single quotes in both studies), very minor concern in one study (due to the potential influence of the researcher on the findings not being discussed) and no concerns over two studies; no concerns about coherence with consistent information emerging across studies; minor concerns about relevance due participants in one study being self-identified as having ME/CFS rather than having been diagnosed according to accepted criteria but no similar concerns in the other six contributing studies and participants of one study consisting of people recruited in a RCT; no concerns about adequacy, the finding being supported by sufficient information across the contributing studies. Overall assessment of confidence was moderate due the methodological limitations and concerns about relevance identified in the contributing studies.

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

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

DVD case studies were seen as particularly important in helping patients and carers to understand that others shared their experiences, and their 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.

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

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

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

Review finding 3: Advice on symptom management & treatment options

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

Due to a lack of knowledge in the medical community and the lack of support they experienced from the health care system, patients and carers described how, having been left to find their own information, they encountered difficulty finding appropriate treatment, with some seeking help from private professionals or paying for alternative therapies. Those 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'.

Patients 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 a lack of resources in the National Health and social systems and the relative lack of recognition or value given to their own experience with illness. They criticized the continued focus on cognitive-behavioural and graded exercise therapies which attribute the illness to psychological causes and called for more physiological research to develop effective biomedical treatments and destigmatize the field. Some patients retained for a new treatment as a rational to continue to live while others experienced moments where the unbearable physical pain of the illness led them to view suicide as a logical escape.

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

Review finding 4: Need for understanding & advocacy

When reflecting on the role of their GP, ME/CFS patients wanted to be believed. Both 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 and those who felt believed GPs described how important this was to their wellbeing. They revealed how facing barriers in gaining recognition of their illness from health professionals both delayed or reduced access to support and greatly exacerbated emotional pressures. Even when bed-bound, patients encountered unsupportive attitudes from health professionals which greatly undermined their chances of wider belief and support. Patients 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. Many encountered doctors that were trained to view ME/CFS as mental health condition , which was dismaying and caused disappointment when doctors made exercises recommendations and psychological attributions or inferences.

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

Patients also faced incredulity and resentment in the workplace, which made 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 they were extremely ambivalent about the process of getting others to make allowances for them, setting limits on asking for help. Some patients 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.

Patients 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 and a lack of understanding from those not affected by ME/CFS were distressing, created tension and often threatened relationships of both patients and parent carers who felt isolated and hoped for change in the way ME/CFS is received.

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

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

Review finding 5: Support with Acceptance

Time appeared to influence acceptance of the diagnosis, 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. When discussing personal responses patients viewed as key to overcoming challenging periods during treatment, they highlighted being open, positive, proactive, willing to try anything, being able to take a leap of faith and having perseverance. They particularly 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.

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

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

Diagnosis was a difficult time for patients who recalled feeling angry, distressed, frustrated and fearful and that the diagnosis represented a life sentence. Accepting the diagnosis was difficult because of patients' own negative preconceptions about ME/CFS and the reactions of others. A need for intensive support and follow-up after completion of the diagnostic care path emerged, with many participants having experience the feeling of being left in the dark afterwards for example having to search themselves for caregivers in their neighbourhood. Some patients found initial stages of treatment difficult and 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.

Parent carers reported feelings of uncertainty regarding diagnosis and prognosis following phases of remission and relapse and experienced 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.

Explanation of quality assessment: moderate concerns over methodological limitations with serious concerns in one study (associated with the use of public data where lack of detail on the method of primary data collection and participant recruitment limited our ability to assess the risk of bias), moderate concerns over one study (due to the potential influence of the researcher on the findings not being 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 single quotes) and no concerns over the third contributing study; minor concerns about coherence due to the finding emerging from three studies and participants in different studies reporting 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 one study being carers of patients of various ages not limited to adults that were considered too minor to lower the confidence rating as the theme also emerged from two studies with no similar concerns; minor concerns over adequacy due to issues with data richness in one study but sufficient information to support the theme overall.

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

Review finding 7: Support from specialists

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

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

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

Review Finding 8: Help accessing support

Patients had difficulty accessing helpful health-care providers and many encountered disdain, disbelief and lack of knowledge. They described long and frustrating histories of their attempts to access necessary information and services to help them address the consequences of their impairment while finding helpful physicians that could provide a diagnosis and appropriate services often took years of shuffling through various doctors. Both 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, reporting that most GPs and practice nurses did not have details of relevant contacts. Applying for welfare benefits while affected by 'CFS/ME' was also difficult for patients as the nature of their symptoms meant that complicated forms having to be completed to start the process were especially daunting. Patients 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. Unable to claim any benefits whether through lack of support from benefits staff or their current financial situation, patients were often bereft of resources, and enforced total dependence on family or partner. Problems acquiring disability income, concerns about requesting workplace accommodation and difficulties accessing community-based resources (such as meal-delivery programs and specialised transportation options) were also reported. This was because patients 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. Where benefits were successfully secured, these were always perceived as helpful. 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.

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

Review Finding 9: Financial support

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

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

Review finding 10: Ongoing health-professional support

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

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

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

Review finding 11: Social support

Patients were physically and mentally isolated struggling to maintain relationships because of their symptoms, with many losing close friends and using social media and online forums, which were helpful, but not commensurate with face-to-face interactions. Feelings of isolation were also reported by carers who had to devote a lot of time to attend patients' needs. Those completing treatment at NHS specialist services 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' in order for the benefits of mutual support to be realised. Hearing from or about individuals who had recovered or substantially improved following ME/CFS was perceived by patients as a potential source of encouragement which could be offered by specialist services.

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

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

Lacking caregiver resources made it difficult for patients to cope, since they had to exert energy to work themselves causing physical consequences. Patients 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. Practical support for personal care, family roles, independent living and support for carers was 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 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. Social care was of paramount importance for patients' life priorities which often included sustaining their own roles as family caregivers. They highlighted how lack of access to social care and practical support, which is the case for most people with ME/CFS, made a profound impact on their ability to carry out their family caregiving roles, particularly as parents, raising questions about how people will be able to manage their

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

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

Review finding 13: Need for tailored and accessible hospital care

Patients highlighted the importance of flexibility when making medical appointments and accommodating treatment programmes around their commitments, lack of which could be a barrier to attendance. They noted travel during the early stages of the illness or when symptoms were severe 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. 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 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.

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

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

Review finding 14: Need for a diagnosis:

Patients 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 control of their lives or taking constructive action to improve their symptoms. Achieving a diagnosis was seen as a crucial milestone for most participants as it often led to advice from doctors and other health care professionals with particular knowledge of CFS/ME. However, patients often encountered oppositional health service responses and some therefore decided to use private or alternative health 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 assess their needs.

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

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

When reflecting on the role of their GP, patents wanted to receive a positive diagnosis. They highlighted the need for the diagnosis to be given in a positive way to maintain hope that symptoms can improve. Hearing from or about individuals who had recovered or substantially improved following ME/CFS was perceived as a potential source of encouragement which could be offered by specialist services, while media accounts of individuals who were very severely disabled appeared to be a key source of anxiety. 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, and patients hoped that referral to a specialist service would give them positive direction for the future.

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

Review finding 16: Patient support during medical 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 during a GP consultation.

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

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

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

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

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

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

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.

Review finding 1: Information about ME/CFS

Young people appeared to experience difficulty with a lack of information around the condition from medical professionals involved. Adolescents talked about the intention to fact-find, with reference to carrying out research and to know more about the condition. Reading facts about symptoms was reported to be helpful by adolescents who identified with the information and felt validated by this. 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 that not knowing why they were undergoing many tests made them feel worried and not knowing what to expect made them feel scared. Mothers of adolescents with ME/CFS reported there was a lack of initial guidance or information around the illness and day-to-day management of symptoms that lead to initial confusion and uncertainty about their child's health and possible diagnosis. Parents reported feelings of uncertainty in regard to prognosis, talking about fears of death when their son/daughter was at his/her worst and parents of adolescents with ME/CFS experiencing eating difficulties felt it would help to know eating problems are 'normal' in this population and may help them understand the illness further.

Explanation of quality assessment: Moderate concerns over methodological limitations with moderate concerns in one study (due to the role of the researcher not being discussed and serious concerns over data richness), serious concerns in another study (associated with the use of public data, where the lack of detail on the primary data collection methods and participant recruitment impacted our ability to assess risk of bias) but minor concerns in three studies (due to the role of the researcher not being discussed in two studies along with lack of sufficient detail over the data analysis process in one study and minor concerns over data richness in the other study and due to the very small sample size and homogenous population of one study); no concerns about coherence; moderate concerns over relevance with minor concerns 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, 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, the sample of one study being limited to people with ME/CFS who experienced eating difficulties, one study excluding severely affected individuals and the sample in one study consisting of participants from a feasibility RCT); no concerns about adequacy with sufficient information supporting the theme. Overall assessment of confidence was low due to the methodological limitations of the contributing studies and concerns over relevance.

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

Adolescents with ME/CFS sought '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. However, they reported only using the NHS sites a few times with most moving 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. The fact that these sites could be rapidly accessed at any time seemed to provide a great sense of support. Being able to interact with these sites in a quick and undemanding way through a shared language of 'likes' and 'comments was less demanding and more flexible than offline relationships especially in the context of a disabling and fluctuating illness. Technological affordances including videos were described as facilitating a sense of relationship and the fact that these sites could be rapidly accessed at any time seemed to provide a great sense of support.

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

Review finding 3: Types of information

Adolescents with ME/CFS talked about the status of official websites they were accessing as 'reliable', but felt that NHS sites were not user-friendly, as they used medical terminology, lacked depth and their content remained unchanged. Regularly accessed sites (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. They preferred the numerous accounts and story-telling approach of patient-led/peer-led and non-health-related sites and which provided support with the psychological difficulties they often experienced, encouraged them to open up and seek help offline and the technological affordances of videos.

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

Review finding 4: Need for validation and advocacy

Young people appeared to experience difficulty with a lack of understanding and awareness around the condition from medical professionals involved. They referred to a fear or actual experiences of disbelief and judgment from others (including teachers and peers), which was particularly difficult for some. 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. Adolescents referred to specialist ME/CFS services reported that specialist medical care was positive, as it enabled

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

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.

The theme was also supported by participants' behaviour outside interviews described in one study. As reported by authors, 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.

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

Review finding 5: Support with acceptance & adaptation

Young people with ME/CFS reported that 'CFS/ME' put a strain on normal adolescent life, such as their identity and friendships and that accepting that for a time they must reduce their activity levels and adopt a routine was challenging. They commonly reported that ME/CFS symptoms prevented, restricted or interfered with their activities. 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.

ME/CFS was also 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 to include caretaking responsibilities and the way this has affected their own 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. 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 is manifested in the

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

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

Review finding 6: Management strategies & support with implementation

Parents reported a lack of treatments and resources for managing 'ME'. 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. A few of the mothers of these young people noted that specialist medical care strategies had an impact on the whole family and could be difficult to integrate with their routine lifestyle. 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.

Explanation of quality assessment: Moderate concerns over methodological limitations with minor concerns in one study (due to the role of the researcher not being discussed and data richness) but serious concerns in the other contributing study (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); no concerns about coherence with the theme being well grounded in the data with coherent information emerging; minor concerns over relevance with due to minor concerns in both studies (due to the sample in one study consisting of parent carers that were not limited to the stratum of children and young people and the sample of one study consisting of participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the RCT); minor concerns over adequacy associated with aforementioned concerns over data richness. Overall assessment of confidence was low due to methodological limitations and concerns over relevance and adequacy.

Review finding 7: Mutual patient support

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

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

Review finding 8: Educational support

Young people felt that better support from education systems could have helped. They described schools and colleges as inflexible, unhelpful, un-empathetic and invalidating and identified this as a cause of increased anxiety and low mood. They particularly reported that teachers had not been very supportive. Mothers of those who had accessed a ME/CFS specialist service discussed the beneficial way in which the 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.

Explanation of quality assessment: Minor concerns over methodological limitations in both contributing studies (due to the role of the researcher not being discussed and concerns over data richness in one study, potential selection bias in the other study as recruitment across a multidisciplinary team meant reasons for declining participation were not clear); no concerns about coherence, the theme clearly emerging from the data; minor concerns relevance associated with both contributing studies (due to the sample in one study consisting of participants from a feasibility RCT which may differ from ME/CFS patients not recruited in the RCT 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); moderate concerns about adequacy with relatively limited information supporting the theme across studies. Overall assessment of confidence was low due to concerns over methodological limitations, relevance and adequacy.

Review finding 9: Need for a diagnosis:

Adolescents 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. Mother of adolescents described how a formal diagnosis enabled positive change and steps towards a managed recovery and how confirmation of 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.

Explanation of quality assessment: Minor concerns over methodological limitations in both contributing studies (due to the role of the researcher not being discussed and concerns over data richness in one study, due to the small sample size and homogenous population of participants who attended the same clinic of the other study); no concerns about coherence with the theme very clearly emerging from the data in both contributing studies; minor concerns over relevance associated with both contributing studies (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; participants 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); minor concerns over adequacy due to the relatively limited information supporting the finding in the studies. Overall assessment of confidence was moderate due to minor concerns over methodological limitations, relevance and adequacy.

Review finding 10: Tailored approach to care

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

Explanation of quality assessment: Minor concerns over methodological limitations in both the contributing studies (due to the role of the researcher not being discussed and concerns over data richness in one study and potential selection bias in the other study as recruitment across a multidisciplinary team meant reasons for declining participation were not clear); no concerns about coherence with nothing to lower the confidence rating; minor 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 ME/CFS patients not recruited in the RCT and because patients in the other study had co-morbid low mood which potentially limits the representativeness of 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 their ME/CFS or low mood); moderate concerns over adequacy with relatively limited information supporting the theme. Overall assessment of confidence was low due to concerns over methodological limitations, relevance and adequacy.

Review finding 11: Referral to specialist services

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

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. Adolescents reported that specialist medical care was positive, as it enabled them to talk about their illness.

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.

Mothers also 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.

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

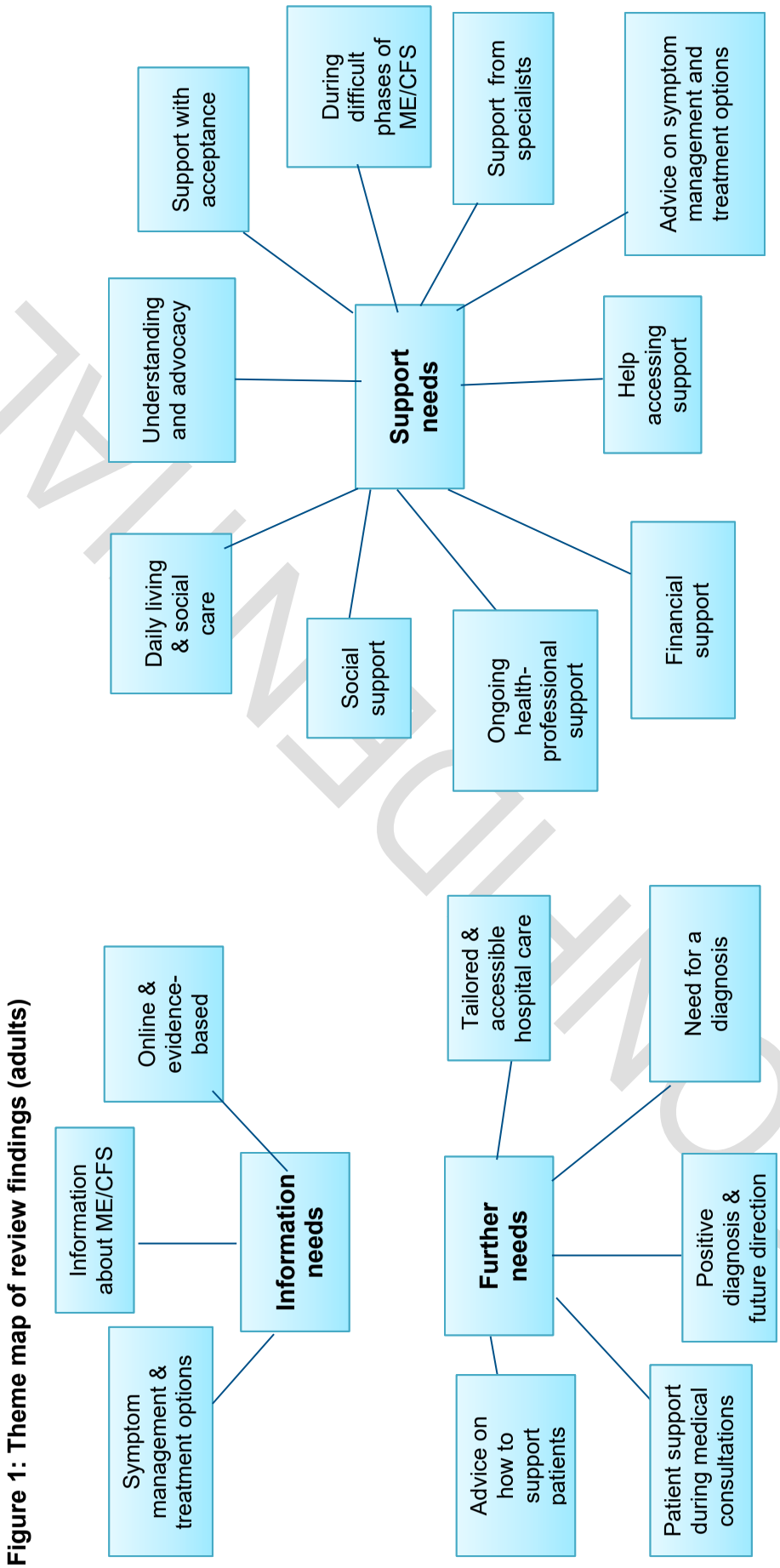


Figure 1: Theme map of review findings (adults)

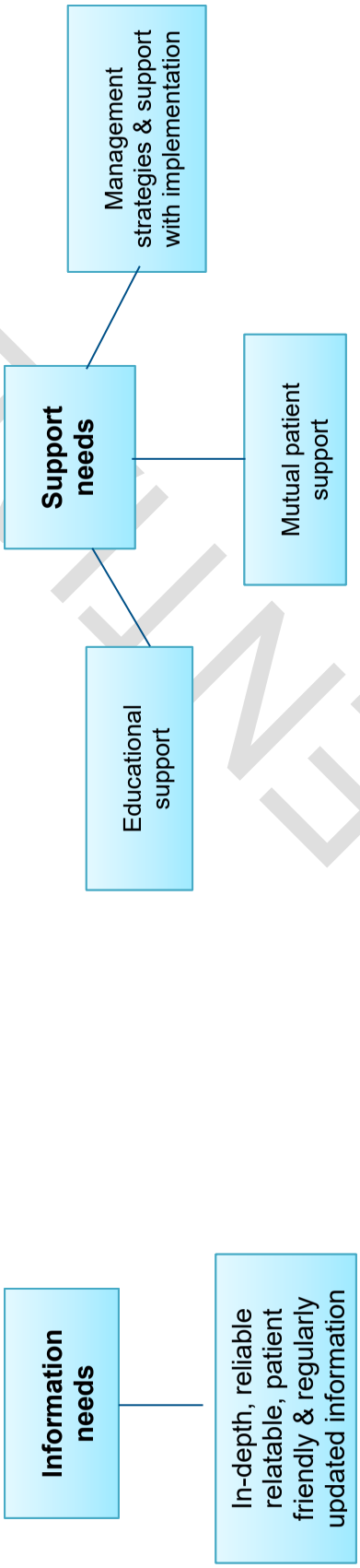


Figure 2: Theme map of review findings (additional themes in children/young people)

1.1.7 Economic evidence

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

2 The committee's discussion and interpretation of the evidence

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

2.1 The quality of the evidence

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

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

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

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

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

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

2.2 Findings identified in the evidence synthesis

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

Information needs

Understanding ME/CFS

Evidence suggested that people with ME/CFS and their carers need 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 and to educate others in order to gain understanding and empathy. The time following diagnosis of ME/CFS and the early stages of treatment were difficult and people 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. The committee agreed that being informed about ME/CFS, benefits people with ME/CFS and their carers in many ways. Therefore, the committee recommended that people are provided with up-to-date information throughout the diagnostic and care pathway and this information should be tailored to the person's individual circumstances and expanded or repeated as required. The committee agreed that as part of the information provided at the start when ME/CFS is suspected, it is important to explain to people presenting with possible symptoms that there is no diagnostic test for ME/CFS and that it is recognised on clinical grounds alone.

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

Evidence suggested people with ME/CFS 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. The committee noted the evidence highlighting the importance of acceptance and willingness to change did not explicitly identify them as areas in which people need support. The committee also noted that acceptance was also identified in another review (Evidence review C: access to care) as a barrier to care and that acceptance of the condition is a personal process. It was hoped that and recommendations on providing information to people with ME/CFS would increase understanding of the condition.

Management of ME/CFS

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

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

Delivery of information

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

<u>Information needs of children and young people</u>

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

Delivery of information

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

Support needs

Being believed

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

ME/CFS support groups

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

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

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

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

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

Social care support
Evidence suggested that people with ME/CFS needed practical support for both themselves and their carers, including help with personal and domestic tasks and access to social care, lack of which impacted on their ability to manage their lives and maintain their family roles. It was noted that not everyone with ME/CFS has a family or other informal carer. Some are entirely dependent on paid care.

Support needs of families and carers

The committee discussed the impact of ME/CFS on other family members acknowledging that it can affect all members of a family (including siblings) and all aspects of family life. They noted the impact varied according to the symptoms and their severity and could fluctuate in alignment with the severity of ME/CFS. The committee noted there is a psychological, physical and emotional impact on families and carers and described below some of the ways they can be affected. Family members might have to take on more (or all) responsibilities in order to allow the person with ME/CFS to effectively manage their energy limits. This may include:

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

- planning ahead and prioritising activities and tasks, to reduce the burden on the person with ME/CFS. This may include cancelling planned activities as circumstances or symptoms change
- Wage earning
- Childcare including the school run - and the effect on relationship with the child if they are unable to participate in activities.
- Driving - and especially night driving
- Attending medical appointments to help with getting to the appointment and comprehending the information given.

In addition, feelings of guilt and stress about doing/not doing what is best; anxiety caused by confusion and lack of understanding of the condition; tiredness from taking on extra responsibilities, and lack of sleep, may all contribute to:

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

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

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

The committee agreed that it was important that people and their families and carers were aware of the support available to them. The committee considered that some people may be reluctant to or have reservations about engaging with social care support services due to previous disbelief about the severity of the illness and the level of impact on day-to-day functioning. They noted there are sensitivities in this area, particularly with children and young people and their families (see the discussion on safeguarding in evidence review B: Information for health and social care professionals). The committee wanted to be clear that these recommendations are about accessing support and health care professionals should discuss sensitively with the person and their family and carers how social care and social services may benefit them. If the person decides they would like to access social care services, they and their family and carers should be made aware how to self-refer for a social care assessment or if they prefer a referral should be made by the health or social care professional. The committee made recommendations sign posting to different assessments and support. These included social care needs assessment, highlighting to parents and carers the Children and Families Act 2014 and for young carers, the Young Carers (Needs Assessment) Regulations 2015. In the committee's experience health and social care professionals were not always aware about the support available to carers and families of people with ME/CFS and they referenced the NICE guideline on supporting adult carers.

Specialist ME/CFS services

Evidence suggested a diagnosis of ME/CFS, 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 psychological and financial pressures and impeded access to social services. People with ME/CFS 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. (see Evidence review I: Multidisciplinary care) The importance of receiving an accurate and timely diagnosis is a theme running throughout several of the qualitative reviews in the guideline and specific recommendations related to this are discussed in the Evidence review D: Diagnosis. The committee noted that this finding highlighted the importance of support for people with suspected ME/CFS during the pre-diagnostic stage. Recommendations for pre-diagnostic management are discussed in the Evidence review E: Strategies pre diagnosis.

Continuity of care

Evidence suggested that people with ME/CFS 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. The committee noted that the theme of lack of continuity of care was identified as a barrier to care as well as a support need and this fed into discussion of the potential implications for people with ME/CFS and the rationale for the recommendation to allocate a named contact to the person with ME/CFS. see Evidence review C: Access to care and Evidence review I: Multidisciplinary care.

Advocacy and support with communication

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

<u>Support needs of children and young people</u>

Being believed

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

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

Education and training

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

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

The committee acknowledged that schools and colleges are high stimulus environments with high physical, cognitive and emotional demands and time in school results in a high level of energy expenditure. The committee acknowledged that that school is a place of activities beyond academic education and that the social element of attending school in person should not be dismissed. The committee agreed that communication between health and social care professionals and training and education services is key to enable understanding of the needs and impairments of people with ME/CFS and provision of appropriate educational support. Therefore, the committee recommended that health and social care professionals should work and communicate closely with social care, training and education services to enable them to understand the needs and impairments of children and young people with ME/CFS, ensure a common understanding of the aims of the person with ME/CFS and discuss a flexible approach to training and education.

The committee discussed the adjustments and adaptions that could support someone in education or training. These should be planned with the person with ME/CFS and their families and health, social care and education services. The committee noted that ME/CFS can be an invisible condition, people with ME/CFS may appear well and it can be difficult for people to understand that adaptions are needed. They noted that children and young people

as a rule want to fit in and not be singled out from their peers. This can result in children and young people overexerting themselves or not wanting to use aids like wheelchairs.

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

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

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

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

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

Work/employment

Just as children and young people need supporting with education and training, adults need supporting with work and employment. It is important that people are given the opportunity to discuss work and how, if appropriate, what might be making return to work challenging. Adjustments and adaptions may be required to support them (for example, flexible hours, working from home, special equipment) through their occupational health department at work. The Access to work scheme may be able to assist employers with paying for any adjustments.

Access to care

Evidence suggested that hospital care tailored to individual needs, symptom severity and other commitments, in terms of accessibility and flexibility in the frequency, duration and

mode of attendance to medical appointments was valued by people with ME/CFS. Themes of individualized care and flexibility around the frequency, duration and mode of medical appointments ran throughout several of the qualitative reviews and are discussed along with the recommendations elsewhere (Evidence review C: Access to care).

Information and education needs of families and carers

The committee noted there was limited evidence directly referring to the information, education and support needs of families and carers. The committee discussed this was an important area and they were aware that some family members and friends, particularly when someone first has symptoms, can find it difficult to understand ME/CFS and the impact of the symptoms. This was reflected in the evidence with carers reporting they could be frustrated by the symptoms people had and lacked an understanding of the illness. They recognised the danger of giving inappropriate advice or support, exacerbating symptoms or pushing the person with ME/CFS to do too much too soon. This is also supported by the Children and young people's report, where symptoms were reported to worsen where people had been given unhelpful advice (by health-care professionals) such as to go out every day despite not feeling good enough to do so. The report highlights how being pushed and not being given time to rest or recover from symptoms as a result of other people's unrealistic expectations, including people from the family and school of the person with ME/CFS, could act against effective management.

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

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

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

2.3 Cost effectiveness and resource use

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

2.4 Other factors the committee took into account

The committee discussed the lack of research including pregnant women, childbirth and post-natal care in all areas of the guideline. This committee noted there is a general lack of information available about how to support women with ME/CFS and their partners during pregnancy through to the post-natal period. The committee agreed that women with ME/CFS can have very different experiences of pregnancy and childbirth on their symptoms. The committee agreed they did not have the expertise to make any specific recommendations but considered that the focus in the guideline on personalised care and regular review of care should prompt the necessary planning required for pregnant women through to and including the post-natal period.

To raise awareness of this gap in the evidence pregnant women and women in the post-natal period have been specified in the population for the self-management strategies, sleep management strategies, and dietary strategies research recommendations.

Appendix A Review protocols

Review protocol for information, education and support for people with suspected and/or diagnosed ME/CFS, their families and 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	The following databases will be searched: • Embase • MEDLINE • CINAHL • PsychINFO Searches will be restricted by: • English language

		The searches may be re-run 6 weeks before the final committee meeting and further studies retrieved for inclusion if relevant. The full search strategies will be published in the final review
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)	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. The full text of potentially eligible studies will be retrieved and will be assessed in line with the criteria outlined above. A standardised form will be used to extract information from studies (see Developing NICE guidelines: the manual section 6.4). 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.
15.	Risk of bias (quality) assessment	Risk of bias will be assessed using the appropriate checklist as described in Developing NICE guidelines: the manual: For this review the CASP qualitative checklist will be used to assess risk of bias of individual studies. 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.
16.	Strategy for data synthesis	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.

		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	Stratification: • Children/young people vs. adults • People with severe ME/ less severe ME (as defined by the studies)
18.	Type and method of review	☐ Intervention ☐ Diagnostic ☐ Prognostic ☒ Qualitative ☐ Epidemiologic ☐ Service Delivery ☐ 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	☐	☑
		Piloting of the study selection process	☐	☑
		Formal screening of search results against eligibility criteria	☐	☐
		Data extraction	☐	☐
		Risk of bias (quality) assessment	☐	☐

		Data analysis	☐	☐
24.	Named contact	**5a. Named contact** National Guideline Centre **5b Named contact e-mail** CFSME@nice.org.uk **5e Organisational affiliation of the review** National Institute for Health and Care Excellence (NICE) and the National Guideline Centre		
25.	Review team members	From the National Guideline Centre: • 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	NICE may use a range of different methods to raise awareness of the guideline. These include standard approaches such as: Notifying registered stakeholders of publication Publicising the guideline through NICE's newsletter and alerts 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.
32.	Keywords	Patients experience, information
33.	Details of existing review of same topic by same authors	N/A
34.	Current review status	☒ Ongoing

		☐ Completed but not published ☐ Completed and published ☐ Completed, published and being updated ☐ Discontinued
35..	Additional information	N/A
36.	Details of final publication	www.nice.org.uk

Appendix B Literature search strategies

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

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

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

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

B.1 Clinical search literature search strategy

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

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

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

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

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

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)

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

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 (stype.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"))

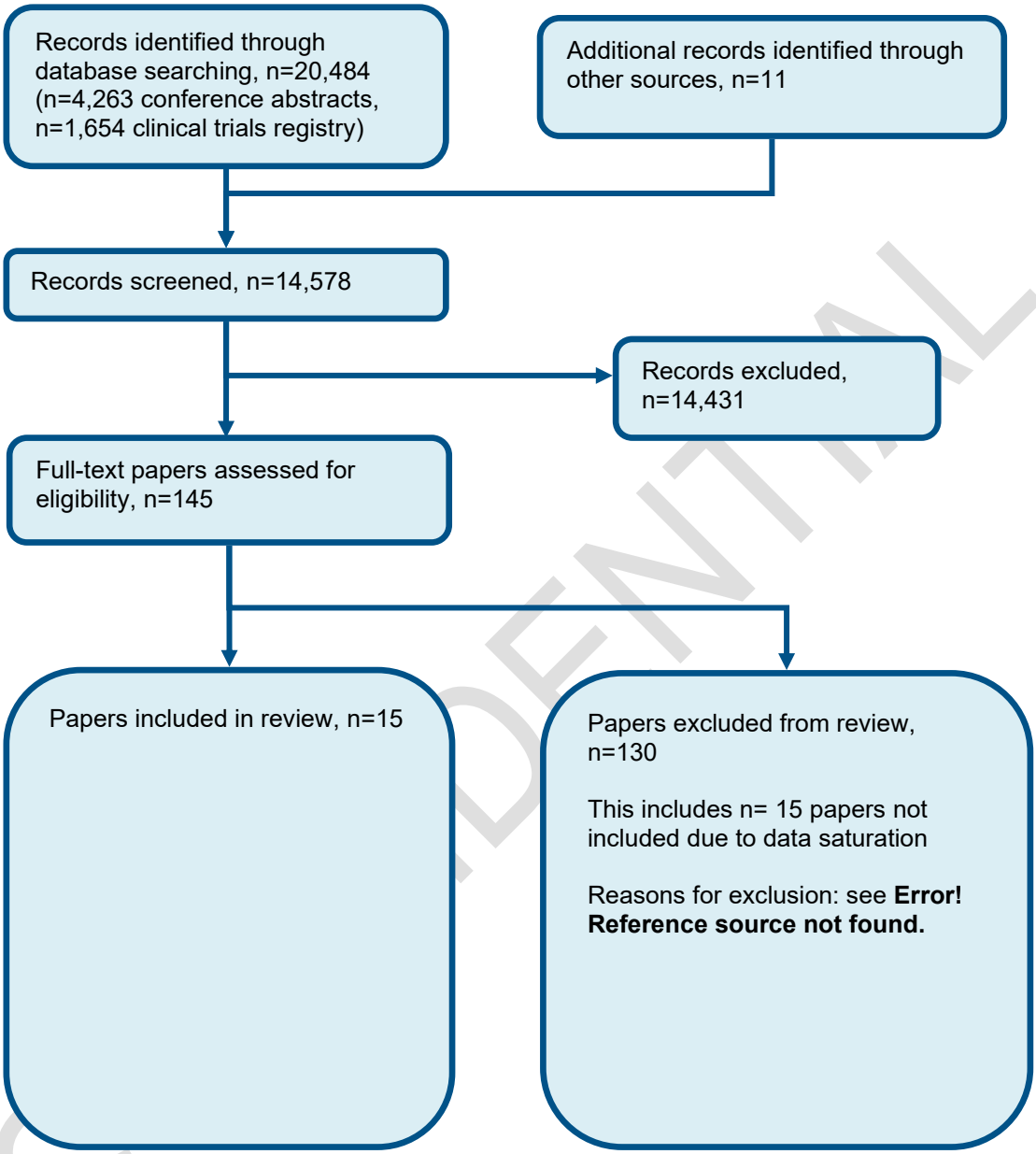
Epistemonikos search terms

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"

	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)))))

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



Appendix D Qualitative evidence

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	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. 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. Patients (n=11), mean age (range): 46 (27 to 71) years; GPs (n=8)
Setting	Primary care
Study design	Qualitative interview study
Methods and analysis	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. 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.
Findings	**Support from GPs** a) **Need to be believed:** When reflecting on the role of their GP, patents 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.

Study	Bayliss 2016[15]
	b) **Need for a positive diagnosis:** When reflecting on the role of their GP, patents wanted to receive a positive diagnosis
	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.
	Information resources for patients, families & carers
	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.
	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.
	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.
	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	Very minor limitations due to the role of the researcher not being discussed. No concerns over applicability

Study	Beasant 2014[18]
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	Beasant 2014[18]
	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). 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
Setting	Large regional specialist 'CFS/ME' service in South West England.
Study design	Qualitative interview study
Methods and analysis	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. Interviews were audio-recorded and transcribed verbatim. 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.
Findings	**Information about 'CFS/ME' & its management**
	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'
	Guidance on management:
	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.
	Tailored approach to treatment:
	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.
	Referral to specialist services
	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,

Study	Beasant 2014[18]
	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.

Study	Brigden 2018[22]
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 2018[22]
	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	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. 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.
Findings	**Facts/info about the condition**
	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.
	b) **Treatments:** Some participants felt there was not enough information about treatments.
	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.
	Sources of information: Digital resources
	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.
	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.

Study	Brigden 2018[22]
	Types/Characteristics of information sources
	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.
	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.
	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.
	Social support (via digital resources)
	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.
	Coping with stigma
	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.
	Adaptation/ Establishing normality (in the face of an isolating and disabling condition)
	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'.
	Relationship support/ Need for validation/legitimacy
	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.
Limitations and applicability of evidence	Minor limitations due to the role of the researcher not being discussed, data analysis due to the limited details provided. Minor concerns over applicability, the study being of potentially limited applicability to severely affected adolescents since they were not included in the sample.

Study	Broughton 2017[25]
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	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). 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 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.
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	**Patient information about 'CFS/ME'** 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. **Need for validation of experiences & symptoms from:** 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 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 **Support with acceptance of diagnosis & adaptation:**

Study	Broughton 2017[25]
	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.
	Support during early stages of treatment:
	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.
	Realistic goal setting (towards management instead of cure)
	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.
	Flexibility in medical care (appointments):
	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

Study	Broughton 2017[25]
	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.

Study	Chew-Graham 2008[32]
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]
	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. Patients: n=24; 11 male, 13 female; mean age: 48, SD: 12 years; months since CFS diagnosis range: 1-240, median: 40.5
Setting	Family physicians and registered patients were from 44 primary care trusts in North West England
Study design	Qualitative interview study
Methods and analysis	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. 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.
Findings	**Advice on management**
	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.
	Information on 'CFS/ME' (from alternative sources outside the clinical domain)
	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.
	Established relationship with physician
	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;

Study	Chew-Graham 2008[32]
	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.

Study	De Carvalho Leite 2011[38]
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	De Carvalho Leite 2011[38]
	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. 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. 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
Findings	**Validation& Recognition of symptoms**
	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.
	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.
	Need for a diagnosis
	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

Study	De Carvalho Leite 2011[38]
	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 2011[38]
	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.

Study	Devendorf 2018[43]
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 *et al*, 2018).
Methods and analysis	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 and open-ended format. 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.
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 rational 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 2018[43]
	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.

Study	Hannon 2012[58]
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 General 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 2012[58]
	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.
	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.

Study	**Jelbert 2010[69]**
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.

Study	Jelbert 2010[69]
	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 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	**Need for understanding (from medical & social community)**
	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.
	Need for a diagnostic label
	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.
	Information on CFS (prognosis)
	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.
	Social support (family, friends & other patients)
	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.
	Support during transition from ill to well/ with reintegration
	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.
Limitations and applicability of evidence	Minor methodological limitations due to small sample size and homogenous population of participants who attended the same clinic.

Study	Jelbert 2010[69]
	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.

Study	McDermott 2011[87]
Aim	To explore the hopes and expectations of patients newly referred to a 'CFS/ME' Service in the South of England.
Population	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). N=20; mean age (range): 39 (22-60) years; male/female: 3/17; 2/10 were members of a patient support group.
Setting	ME/CFS specialist NHS services, South of England
Study design	Qualitative interview study
Methods and analysis	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'. 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)
Findings	**Need for specialist advice**
	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.
	Need for diagnosis
	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

Study	McDermott 2011[87]
	Information about 'CFS/ME' & prognosis
	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 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.

Study	Mihelicova 2016[89]
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.

Study	Mihelicova 2016[89]
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 design	Secondary analysis of qualitative interviews
Methods and analysis	Data were collected from interviews published in Lost Voices from a Hidden Illness (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. Data were analysed used 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.
Findings	**Adaptation:**
	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.
	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.
	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 in 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.
	Need for advocacy from medical & social community and connection:

Study	Mihelicova 2016[89]
	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.
	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

Study	Ryckeghem 2017[116]
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.

Study	Ryckeghem 2017[116]
	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
Methods and analysis	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?' 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?' Thematic analysis (open explorative thematic coding) was used.
Findings	**Patient information about CFS**
	Patients feel they lack general information about the disease and they are not offered a clear model of explanation.
	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.
	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.
	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.
	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
	Punctuality of reporting/ care & timely communication

Study	Ryckeghem 2017[116]
	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.
	Information for the patient's environment
	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.
	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'
Limitations and applicability of evidence	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. No concerns over applicability.

Study	Taylor 2005[131]
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	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 *et al* (1994) criteria for ME/CFS. 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.
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	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.

Study	Taylor 2005[131]
	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 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?' 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.
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
	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.
	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.
	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.**

Study	Taylor 2005[131]
	Need for treatment advice from specialists
	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.

Study	Taylor 2017[130]
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

Study	Taylor 2017[130]
	questionnaire, why young people felt they had become low in mood, whether their 'CFS/ME' preceded or followed their low mood, 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. 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.
Findings	**Adaptation: Impact of difficulties on normative processes of being a teenager**
	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.
	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.
	Peer support from other patients/ Developing a patient network & interpersonal skills:
	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.
	Supportive education systems
	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.
	Information on (Individualised) helpful approaches to managing low mood

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.

Appendix E GRADE-CERQual tables

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)				

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; 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 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 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 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 previously recruited in a RCT (Chew-graham 2008) but no further concerns in any other contributing study.

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.			

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 (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 2008, Hannon 2012); moderate concerns about coherence due to differences both between studies and within studies in regards to the types of information patients preferred, with some patients and carers reporting on the usefulness of leaflets (Hannon 2012), others reporting on the usefulness of information available online (Bayliss 2016; Chew graham 2008; Hannon 2012) and in DVD format (Bayliss 2016; Hannon 2012), with some worrying online information would negatively impact the management 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 due to participants in one study consisting of people recruited in a RCT (Chew-graham 2008).

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**	**Finding**	**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)				

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 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 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 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 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 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 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 illness severity and duration (De Carvalho Leite 2011).

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**	**Finding**	**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	

Six studies with very minor to srious issues; methodological limitations due to the role of the researcher not being discussed in four studies (Bayliss 2016, De Carvalho Leite, 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 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 one study (Hannon 2012) and lack of transparency on the analysis process (Rycheghem 2017), 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 collection implicated our ability to assess bias (Mihelicova 2016); very minor concerns about relevance with 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) 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 ME/CFS patients from diverse social and ethnic backgrounds, degrees of illness severity and duration (De Carvalho Leite 2011)

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 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	

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 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 topic available from the study.

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	

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 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 discussed, concerns over data analysis due to a lack of sufficient information on the data analysis process and cocnerns over data richness with findings mostly supported by 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 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 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 richness in one study.

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**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	

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**	**Finding**	**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	

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 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 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;

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 representativeness of the sample in other contributing studies.

Study design and sample size			Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**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	

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 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 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 reported by patients. from diverse social and ethnic backgrounds, degrees of illness severity and duration (De Carvalho Leite 2011); minor concerns over adequacy with 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	

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 (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 findings supported by single quotes in two studies (Chew-Graham 2008; Ryckeghem 2017).

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**	**Finding**	**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	

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 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 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 participants in one study being self-identified as having ME/CFS (Devendorf 2018) but no similar concerns identified in any other contributing study

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	

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 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); 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, where the majority of the information supporting the theme emerged from, included patients from diverse social and ethnic backgrounds, degrees of illness severity and 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).

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	

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 information supporting the finding in one study (Hannon 2012).

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	

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)

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	

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 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 concerns about adequacy with limited information supporting the theme in one study (Hannon 2012).

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	

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.

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	

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 richness as themes were mostly supported by single quotes in the contributing study (Hannon 2012); serious concerns over adequacy with very limited information supporting the theme.

Table 6: Summary of evidence for information education and support for children and young people 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					
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).				

Five studies with minor to serious issues; overall moderate concerns over methodological limitations due to the role of the researcher not being discussed in thtree studies (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 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 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 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 (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 (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 individuals (Beasant 2014, Brigden 2018) and the sample in one study consisting of participants from a feasibility RCT (Beasant 2014).

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	

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 2018); minor concerns over relevance as the study excluded severely affected individual.

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	

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 2018); minor concerns over relevance as the study excluded severely affected individuals.

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	

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 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 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 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 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 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 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 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	

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 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 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 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 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 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-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).

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	

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 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 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 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); minor concerns over adequacy due to concerns over data richness.

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**	**Finding**	**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	

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 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 study as recruitment across a multidisciplinary team meant reasons for declining participation were not clear (Taylor 2017); moderate concerns over relevance due to the population of one study excluding severely affected individuals (Brigden 2018), all participants having recovered from ME/CFS and potentially holding different opinions to 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 do not (Taylor 2017).

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**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	

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), 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 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 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 (Taylor 2017); moderate concerns over adequacy with relatively limited information supporting the theme in the contributing studies.

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	

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), 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 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 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 about adequacy due to the relatively limited information supporting the finding in the studies.

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	

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), 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 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 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 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 their ME/CFS or low mood (Taylor 2017); moderate concerns over adequacy with relatively limited information supporting the theme.

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	

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 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 had accessed a specialist service and not reported as a need of young people with ME/CFS in other studies

Appendix F Excluded studies

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 2013[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[132]	Incorrect population: women with fibromyalgia and CFS
Theorell 1999[133]	Incorrect study design: reports questionnaire results quantitatively only
Travers 2008[134]	No relevant themes
Tuck 1998[135]	No relevant themes
Tuck 2000[136]	Incorrect study design: questionnaires and no qualitative analysis to allow the extraction of themes
Ward 2008[138]	No relevant themes
Ware 1993[139]	No relevant themes
Ware 1998[140]	No relevant themes

Reference	Reason for exclusion
Ware 1999[141]	No relevant themes
Whitehead 2006[142]	No relevant themes
Whitehead 2006[143]	No relevant themes
Williams 2016[144]	No relevant themes
Wilson 2011[145]	Incorrect population: experiencing chronic fatigue due to long-term conditions other than chronic fatigue syndrome

Table 8: Studies identified but not included in the qualitative review due to saturation 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[137]
Winger 2014[146]
Woodward 1995[147]

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