National Institute for Health and Care Excellence

Final

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

[B] Information, education and support for health and social care professionals

NICE guideline NG206

Evidence reviews underpinning recommendations and research recommendations in the NICE guideline

October 2021

Final

These evidence reviews were developed by the National Guideline Centre

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Introduction

ME/CFS is a condition which may have a profound and long-lasting effect on the lives of those affected by it. It affects approximately between 150,000 to 250,000 people in the UK. It is not generally included in the training curriculums of health and social care professionals. Different sets of diagnostic criteria, different names for the condition (or possibly conditions), and continuingly emerging research regarding aetiology, pathogenesis and treatment all contribute to confusion on the part of practitioners. As a consequence, many health and social care professionals are ill-equipped to manage people with this multifaceted condition. It is important to identify the information, education and support needs of health and social care professionals and how best to address these needs.

The committee used both reviews to inform their recommendations in these areas. The committee discussion of the evidence and interpretation is in section 3.

1. Information, education and support

1.1. Review question

What information, education and support do health and social care professionals who provide care for people with ME/CFS need?

1.1.1. 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 health and social care professionals caring for people with or who are suspected of having ME/CFS.
Population and setting	• Health and social care professionals caring for someone with or who are suspected of having ME/CFS. • Perspectives of people with ME/CFS and the families and carers of people with ME/CFS about the information, education and support needs of health and social care professionals who provide care.
Context	Perceptions from health care professionals and people with or who are suspected of having ME/CFS about the information, education and support needed by health care and social care professionals.
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.

1.1.2. 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.

1.1.3. Qualitative evidence

1.1.3.1. Included studies

Sixteen qualitative studies were included in the review;[19, 24, 30, 31, 40-42, 45, 57, 66, 68, 84, 106, 115, 124, 130] these are summarised in Table 2 below. Key findings from these studies are summarised in Section Table 7 and Table 8below. See also the study selection flow chart in Appendix B, study evidence tables in Appendix D, and excluded studies lists in Appendix F.

Adults

The evidence presented is from studies which included health care professionals caring for adults with ME/CFS (n=9); adults with ME/CFS (n=4); and significant others* of people with ME/CFS (n=1). Findings emerging from those studies are presented together as common themes emerged from the different study populations. One study[19] included all three relevant populations; health professionals, people with ME/CFS, and 'significant others' .

*The term ‘significant others’ is used in the study to describe people identified by person with ME/CFS such as friends, partners, spouses, adult children.

Children and young people

The evidence in relation to children and young people is from studies which included adolescents who had recovered from ME/CFS (n=1), and health care professionals caring for children and young people with ME/CFS (n=1).

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

A large number of papers were identified for this review. Studies were included until saturation of themes was reached. Data saturation is the point at which no new themes, or data contributing to themes emerged from the studies. Studies not included due to saturation being reached are listed in Table 11.

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

1.1.3.2. Excluded studies

See excluded studies in Appendix F.

1.1.4. Summary of studies included in the qualitative evidence

Table 2: Summary of studies included in the review

Study	Design	Population	Research aim	Comments
Beaulieu 2000[19]	Mixture of structured and semi structured questions, analysed using thematic analysis.	Health professionals including general practitioners, mental health professionals (one of whom was not a physician), infectious disease specialists, immunologists and rheumatologists, recruited following identification by people with 'CFS' participating in the study. N=15; male/female 10/5; had been in practice from six to seventeen years and individually had seen from six to almost one hundred cases. People who were English-speaking and who had a diagnosis of 'CFS' from a medical doctor, recruited from physicians' practices, support groups and identified by leaders of associations. N=43; male/female 16/27; 26% were in school or working full or part time; mean age at onset was 34.2 years (range 15 to 58 years); people had been ill for an average of seven years. Significant others including friends, parents, spouses, adult children, and a sibling, recruited following identification by people with 'CFS' participating in the study.	To examine multiple perspectives on stigmatization and legitimation of 'CFS'.	

Study	Design	Population	Research aim	Comments
		N=23; male/female not reported; 69% were working. Canada Stratum: adults/mixed population		
Broughton 2017[24]	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. UK Stratum: adults/mixed population	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. Cognitive Behavioural Therapy (CBT) and Graded Exercise Therapy (GET) are the two main evidence-based therapies which (or components of which) are used in conjunction with techniques aimed at managing activity,

Study	Design	Population	Research aim	Comments
				sleep hygiene and relaxation. Patients also receive practical support around employment and the benefits system. Services shared a philosophy of rehabilitation aimed at 'recovery' or 'significant improvement', whilst acknowledging that this would not be attained by all patients.
Chew-Graham 2008[31]	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/mixed population	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.
Chew-Graham 2010[30]	Semi-structured interviews and thematic analysis (using an iterative approach).	GPs working in practices participating in the FINE trial (n=22). UK Stratum: adults/mixed population	To explore GPs' beliefs about the value of the label of 'CFS/ME', implications of the diagnosis and attitudes towards patients with this condition.	FINE trial was a primary-care RCT examining self-help treatment and pragmatic rehabilitation for

Study	Design	Population	Research aim	Comments
				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.
Devendorf 2017[41]	Semi-structured interviews and (deductive) thematic analysis.	Mixed sample of people with different specialties, working with adults and children and adolescents. Physicians who were experts in the ME and CFS field (n=10); mean age (SD): 65 (12) years. USA Stratum: adults/mixed population	To explore views of physicians with expertise in 'ME and CFS' to define and measure recovery from 'ME and CFS'	Experts were determined by their 'ME and CFS' patient experience, research contributions, and overall involvement in the field (e.g. running 'ME and CFS 'specialty clinics, participating on committees.

Study	Design	Population	Research aim	Comments
Devendorf 2019[40]	sSemi-structured phone-based interviews with physicians and analysing the data using deductive thematic analysis.	Physicians specialising in ME/CFS of diverse medical specialties (n=10) and other physicians (n=3), not identified as ME/CFS specialists. n=13, males: 9, females: 4; mean age 60 years. For years in practice, three physicians had 30 or more years, seven had 20-29 years, one had 10-19 years and two had 1-9 years of medical experience. USA Stratum: adults/mixed population	To explore physicians views on the challenges to studying and approaching recovery, to examine these challenges in-depth and provide recommendations that will improve how researchers and practitioners approach the study and quantification of ME and CFS recovery.	Participants were recruited via non-probabilistic, purposive sampling. Specialists were defined by their extensive patient experience, research contributions and significant involvement in the field. The sample was diverse in their medical specialties: epidemiology (n=1), geriatrics (n=1), infectious diseases (n=1), neurology (n=1), internal medicine (n=2), psychiatry (n=2), general medicine (n=3), and paediatrics (n=5); three physicians identified with two medical specialties.
Devendorf 2018[42]	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).	An exploratory study to explore the relationship between ME/CFS and suicidal ideations, including quality of life, loss of function, isolation and hopelessness.	

Study	Design	Population	Research aim	Comments
		USA Stratum: adults/mixed population		
Edwards 2007[45]	Interpretative phenomenological analysis of semi-structured interviews.	People diagnosed with ME/CFS by a medical professional. N=8; all women. Age range: 37-55 years. Illness duration range: 18 months to 12 years. Inclusion criteria: over 18 years of age, speak English as a first language, diagnosed with ME/CFS by a medical professional, have suffered ME/CFS symptoms for at least one year, consider ME/CFS as their main health problem, and currently experiencing symptoms of at least moderate severity. All but one had stopped working due to ME/CFS. UK Stratum: adults/mixed population	To explore the experiences and difficulties of people living with ME/CFS.	
Hannon 2012[57]	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/mixed population	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.	
Horton 2010[66]	Semi-structured interviews and thematic analysis.	Health care professionals (HCP) who had been nominated by people with ME/CFS who	To explore the nature of professional 'best practice'	

Study	Design	Population	Research aim	Comments
		had taken part in an associated England-wide study of their support needs. N=6; gender not reported. Three participants were from specialist services (medicine, occupational therapy, physiotherapy) and three were from non-specialist services (medicine, occupational health, holistic practice). 36 people with ME/CFS nominated eight HCPs as having provided them with particularly helpful or effective care and six agreed to participate. One HCP was named by six different people with ME/CFS. UK Stratum: adults/mixed population	in working with people with ME/CFS.	
Jelbert 2010[68]	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	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
Marks 2016[84]	Semi-structured interviews and thematic analysis (grounded theory methodology).	Paediatricians, physiotherapists and clinical psychologists working in two NHS organisations in the UK: a hospital outpatient paediatric service, and a specialist centre providing inpatient and outpatient care for young people ME/CFS. N=10; 7 female, 3 male. Mean age not stated. Medical specialties were as follows: paediatricians (n=4), physiotherapists (n=3), and clinical psychologists (n=3). All had a minimum of 3 years' experience of working with ≥3 young people with ME/CFS. UK Stratum: children and young people	To explore HCPs experiences of working with children and adolescents with ME/CFS so as to develop an understanding of the processes relating to how they understand the condition.	
Raine 2004[106]	Qualitative analysis of transcripts of facilitated group discussions.	General practitioners randomly selected from the Department of Health's general practitioner database. N=46; male/female 29/17; mean age 46.9 years; had worked for an average of 14.8 years in general practice, and 9 were affiliated to a medical school. UK Stratum: adults/mixed population	To compare general practitioners' perceptions of chronic fatigue syndrome and irritable bowel syndrome and to consider the implications of their perceptions for the use of psychological treatments.	
Ryckeghem 2017[115]	Semi-structured interviews using open	A purposive sample of patients was selected through the department of General Internal	To explore the experiences and	A definitive diagnosis was established

Study	Design	Population	Research aim	Comments
	explorative thematic coding (thematic analysis).	Medicine at the University Hospital Ghent to achieve maximum variation. A convenience sample of GPs was recruited from different provinces in Belgium. Patients (n=15); median age (range): 45 (33-59 years); GPs (n=15); median age (range): 49 (31-62 years). Belgium Stratum: adults/mixed population	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 in the Belgian provinces of East and West Flanders.	following a multi-disciplinary discussion in the diagnostic process.
Stenhoff 2015[124]	Face-to-face semi-structured interviews and inductive thematic analysis.	Undergraduate medical students in years 3, 4 and 5 at the University of Manchester, UK. N=21; 7 female, 14 male. Mean age: 22 years old. Four were third-year students, 11 were fourth-year students and six were fifth (final)-year students. Participants were recruited through the university's student-net, poster adverts around campus and via personal contact. Sampling ended at saturation in a staged approach, with two students turned away at the end of the study. UK Stratum: adults/mixed population	To investigate medical students' beliefs, attitudes and knowledge of ME/CFS.	
Taylor 2005[130]	Focus group interviews, open-	Adults with ME/CFS meeting the Fukuda criteria for CFS, who were participating in a	To determine what aspects of the disability	Data for this study emerged from a

Study	Design	Population	Research aim	Comments
	ended questionnaires, progress notes, and from a program evaluation questionnaire.	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. USA Stratum: adults/mixed population	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.	federally funded research project that developed and evaluated a participants-driven program for individuals with 'CFS', implemented at a centre of independent living.

See Appendix D for full evidence tables.

1.1.5. Summary of the qualitative evidence

Table 3: Review findings for health care professionals caring for adults with ME/CFS.

Main findings	Statement of finding
Health care professionals' awareness and knowledge of ME/CFS[19, 24, 30, 31, 40-42, 45, 57, 106, 115, 124, 130]	There is need for increased training for healthcare professionals (HCPs) to increase knowledge of ME/CFS and its management. HCPs often lack the knowledge or awareness to be able to diagnose and manage patients with ME/CFS. This often delays diagnosis and referral and means that patients can be mismanaged. This was expressed by both HCPs and people with ME/CFS. There is a need for improved education of HCPs about ME/CFS and an increased presence of the disease in the medical curriculum.
Consensus on diagnostic criteria[19, 40, 41, 57]	The lack of a confirmed consensus on the diagnostic criteria for ME/CFS meant that there was confusion among HCPs when consulted with symptoms. HCPs expressed the need for agreed case definitions for both diagnosis and recovery.
Symptom measures[31, 41]	The lack of agreed tests and measurements for ME/CFS symptoms mean that HCPs are reluctant to make a diagnosis based on limited clinical signs and struggle to assess recovery.
Clinical pathway[30, 66, 106]	There is need for a clearer clinical management pathway for ME/CFS. HCPs are often sure of where to refer patients once a diagnosis has been reached. ME/CFS specialists express concern at the lack of referrals to their services made by GPs.
Training[57, 66, 124]	HCPs highlighted the need for training in how to diagnose and manage ME/CFS, with a preference for an internet-based course. GPs suggested that ME/CFS specialist services should support GPs by providing them with information and training. There is currently little or no formal training on ME/CFS in the medical curriculum, with students claiming their knowledge often comes from media.
Information resources[57, 66]	Some HCPs expressed the need for a resource that can be used during consultation to educate and reassure patients when diagnosed with ME/CFS, for example an online video resource. 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.
Support from specialist services[66, 115]	There is seen to be a lack of communication between GPs and referral centres, with a need for increased feedback and sharing of information from specialist services. Specialist services need to be more visible and provide education and information for GPs.
Information about support groups[19, 57, 66]	HCPs are often unable to recommend support groups because they had little knowledge or information about them.
Exposure to people with ME/CFS[31, 41, 66]	HCPs find that contact with ME/CFS sufferers outside of the clinical setting improved their understanding of the condition. For example, phone conversations or observation of patients living with ME/CFS in their daily lives allowed HCPs to make better understand symptoms and make decisions about management.

See Appendix E for full GRADE-CERQual tables.

1.1.5.1. Narrative summary of review findings and quality

Information, education and support needs of health care professionals caring for adults with ME/CFS.

Review finding 1: Health care professionals' awareness and knowledge of ME/CFS

Both healthcare professionals (HCPs) and patients with ME/CFS reported that there was a general lack of knowledge and belief in ME/CFS from HCPs and in the healthcare system. HCPs admitted to having little clinical information available to them and were unprepared by their medical training and continuing education to diagnose and manage ME/CFS. They often had to seek information from non-clinical sources. There was variation among HCPs about their beliefs in the aetiology of ME/CFS, with physicians variously attributing the condition to psychosocial factors or physiological theories.

Several studies indicated that people with ME/CFS do not think HCPs are adequately prepared from their professional training to diagnose and manage ME/CFS. This meant that patients experienced varied, often negative, experiences when reporting to GPs with symptoms. Patients reported having to consult multiple GPs before having their symptoms taking seriously and eventually receiving a diagnosis of ME/CFS. Some people reported having to take information to their GPs to educate them about ME/CFS, having learned about the condition through media and online research.

Medical students reported that there was little to no education around ME/CFS in the medical curriculum. People with ME/CFS also felt that more teaching about ME/CFS on the UK undergraduate curriculum was required.

Explanation of quality assessment: minor concerns over methodological limitations with minor concerns over four of the contributing studies (due to concerns over data analysis with data often supported by single quotes in two studies, due to the potential influence of the researcher on the findings in one study where half of the participants were given a systematic review on the effectiveness of mental health interventions prior to the data collection and due to concerns over the recruitment strategy in one study where recruitment of participants was done through responses to an advertisement, therefore risking over-representation of students who are more informed or have stronger views on ME/CFS); moderate concerns over three studies (due to concerns over participant recruitment with selection of HCP participants by ME/CFS patients in one study and concerns over data analysis with coding and analysis undertaken by a single researcher in that study, due to the role of the researcher not being discussed and concerns over data analysis due to a lack of sufficient detail and some themes supported by single quotes in one study and 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); no methodological concerns over four studies. No concerns about the coherence of the finding. Minor concerns over relevance with moderate concerns over two studies (due to participants in one study being a subset of a previous quantitative study who were self-identified as having ME/CFS rather than diagnosed according to accepted criteria and due to concerns over the applicability of one study conducted on the Belgian health care system to the NHS setting); but minor concerns in six of the contributing studies (due to the research aim driving the theme being different to that of the current review in three studies, due to participants having been previously recruited in a RCT in two studies, due to concerns over relevance of one study that was published prior to new guidelines and diagnostic criteria, due to concerns over the small and homogenous sample size and lack of representation of Health professionals in the sample of one study, and due to the population of medical students all attending the same medical school rather than practicing HCPs in one study); no concerns over relevance in five studies. No concerns over adequacy. Overall assessment of confidence was moderate due to methodological limitations and relevance.

Review finding 2: Consensus on diagnostic criteria

HCPs expressed the need for an agreed diagnostic criteria and case definitions of ME/CFS. Depending on the criteria and case definitions used by different HCPs, patients may be diagnosed differently between services, often delaying appropriate treatment for some patients. Frequently HCPs talked about how a diagnosis of ME/CFS was made only by exclusion in absence of positive diagnostic criteria.

It was also reported that there is a need for HCPs to better understand the relationship between ME/CFS and depression, where misdiagnoses are possible and patients can be referred to the wrong services as a result.

While some HCPs found the 2007 NICE ME/CFS guidelines helpful, they thought that the lack of any diagnostic test giving conclusive proof of the condition impacted both patients and practitioners, and that until such a test was developed, the existence of the condition would remain in doubt amongst some HCPs.

For some HCPs there was confusion around labels for ME/CFS, with some unclear of the difference between ME, CFS, chronic fatigue or post viral fatigue. HCPs found the CDC diagnostic guidelines unclear and unhelpful as a diagnosis of exclusion.

Explanation of quality assessment: minor concerns over methodological limitations with moderate limitations in one study (due to concerns over participant recruitment with selection of HCP participants by ME/CFS patients and over data analysis with coding and analysis undertaken by a single researcher) but minor limitations in two studies (due to concerns over data analysis with data often supported by single quotes) and no concerns in the other contributing study; no concerns about coherence; minor concerns over relevance due to the findings in two studies being driven by the studies' original aim that differed from that of the current review and due to the contribution of an older study potentially losing relevance (e.g. published prior to new guidelines and diagnostic criteria); no concerns about adequacy. Overall assessment of confidence was moderate due to the methodological limitations and relevance.

Review finding 3: Symptom measures

HCPs reported that they found it frustrating that they could not measure how a patient was affected by their condition. Without definitive tests available to them, HCPs are often reluctant to make a diagnosis considering the 'invisible' nature of ME/CFS symptoms and the disparity between clinical signs and subjective symptoms. HCPs hoped for more integration of useful physiological measures in assessing ME/CFS but knew that this was difficult considering the lack of an identified biomarker.

Some HCPs considered recovery to be when patients no longer meet diagnostic criteria, given the lack of measures for assessing recovery available to them. Some HCPs used exercise tests as objective measures, but others were sceptical of the sensitivity of these kinds of tests. HCPs therefore identified the need for consensus case definitions on how to measure recovery in ME/CFS patients, to ensure there was consistency across services.

Explanation of quality assessment: very minor concerns over methodological limitations with minor concerns in one study (due to data analysis with themes mostly supported by single quotes) but no further concerns identified; no concerns about coherence; minor concerns about relevance due to the aim of the contributing studies driving the theme being different from that of the current review and due to participants in one study having been previously recruited in a RCT; no concerns about adequacy with sufficient information to support the theme. Overall assessment of confidence was moderate due to very minor and minor concerns across two domains of quality assessment.

Review finding 4: Clinical pathway

HCPs thought the label of ME/CFS could be problematic because it does not offer a clear management pathway for the HCP or the patient. This was described as another reason why GPs are reluctant to make a diagnosis of ME/CFS, and will often use the label as a last resort. Some GPs did not know where to refer patients once a diagnosis of ME/CFS was reached, considering specialist services to be fragmented and often not having the required contact information. This was also true of mental health services, with HCPs' lack of knowledge about mental health services cited as a reason why ME/CFS patients were often not referred to receive these types of intervention.

Explanation of quality assessment: minor concerns over methodological limitations with minor concerns in two studies (due to the potential influence of the researcher on the findings in one study and concerns over data analysis with findings mostly supported by single quotes in one study) and no concerns over the third contributing study; no concerns about coherence; no concerns about relevance as concerns over the representativeness of the sample of one study due to participants having been previously recruited in a RCT were too minor to lower our overall assessment of relevance; no concerns about adequacy with sufficient information supporting the theme. Overall assessment of confidence was high due to the concerns over methodological limitations being too minor and no further concerns identified.

Review finding 5: Training

HCPs identified the need for training to address the lack of understanding and belief of the ME/CFS among professionals. Some GPs and nurses suggested that an easily accessible online training resource would be useful and might be favoured over face-to-face training due to the perception that for many ME/CFS was a low priority and HCPs might not want to dedicate a lot of time to training. HCPs and specialists agreed that specialist services were ideally placed to provide the training that GPs and nurses need to recognise and manage ME/CFS.

Explanation of quality assessment: minor concerns over methodological limitations with minor concerns in all three contributing studies (due to concerns over data analysis with findings mostly supported by single quotes in two studies and due to concerns over participant recruitment in one study); no concerns over coherence; very minor concerns over relevance due to the population of one study being medical students rather than practicing HCPs and the homogeneity of that population as all students were attending the same medical school at the University of Manchester, but no similar concerns in any of the other studies; no concerns about adequacy. Overall assessment of confidence was moderate due to minor concerns over methodological limitations and very minor concerns over relevance.

Review finding 6: Information resources

GPs and nurses said it would be useful to be able to print information on symptom management from an online resource, or show an online video, during consultation. It was suggested that a DVD or similar video resource might be useful for those patients who struggle to read written resources because of fatigue, concentration and memory problems.

Several studies identified the media as a common source of information about ME/CFS to both HCPs and patients with ME/CFS. However, it was also observed that debate in the media and influence from individuals' opinions could also lead to scepticism about the legitimacy of the condition.

HCPs from specialist services reported finding standard information packs and DVDs useful, often recommending leaflets produced by patient support organisations such as *Action for ME* and *ME Association*. Specialists also reported referring people to the *Citizen's Advice*

Bureau or *Disability Information and Advice Line* for advice on disability-related support matters, as well as expressing the importance of providing information for employers of ME/CFS.

Explanation of quality assessment: very minor concerns over methodological limitations with very minor concerns over both contributing studies (due to data analysis with themes mostly supported by single quotes) that were considered too minor to lower our overall confidence in the finding; no concerns about coherence; no concerns over relevance; no concerns about adequacy with sufficient information to support the theme. Overall assessment of confidence was high as methodological limitations were very minor and no further concerns were identified.

Review finding 7: Support from specialist services

GPs suggested that it was helpful to have the support of ME/CFS specialists. GPs often lacked confidence in making an ME/CFS diagnosis alone and referring a patient to secondary care assisted in achieving a diagnosis. However, GPs reported limited availability of helpful services that could support them.

A number of GPs and nurses said they were unaware of specialist ME/CFS services. ME/CFS specialists expressed frustration that GPs in their region often did not refer patients to their services. HCPs had often difficulties referring to specialists due to the fragmentation of services and lack of collaboration between the two services. Patients also showed concern about long waiting times for specialist services and suggested that increased communication between primary and secondary care might allow GPs to better manage them.

Specialist HCPs emphasised that there was a need for specialist services to be more 'visible' and for them to provide training and education for other HCPs. Noting in particular GPs, due to their lack of knowledge and awareness and direct contact with people with ME/CFS. Specialists were found to have the experience and expertise required to support GPs and other HCPs in reaching an ME/CFS diagnosis, giving advice on appropriate medication, providing services such as specialist Occupational Therapy and supporting patients to apply for benefits.

Explanation of quality assessment: moderate concerns over methodological limitations with minor and moderate concerns in the two contributing studies (due to concerns over data analysis with findings supported by single quotes in both studies and the potential influence of the researcher on the findings not being discussed in one study); no concerns about the coherence of the findings; minor concerns over relevance due to concerns over the applicability of one study that had been conducted in the Belgian health system to the NHS setting; no concerns about adequacy with sufficient information to support the theme. Overall assessment of confidence was moderate due to methodological limitations and concerns over relevance.

Review finding 8: Information about support groups

When HCPs wanted to highlight healthcare services and information to their patients, such as local support groups and advice on benefits, they were unable to do because they did not have details of relevant contacts. Other HCPs said that they were hesitant to recommend ME/CFS support groups because they had little knowledge of them, or opinion was divided over whether these support groups were harmful or helpful.

Explanation of quality assessment: minor concerns over methodological limitations with moderate concerns over one study (due to concerns over participant selection and data analysis with coding and analysis by a single researcher) and minor concerns over two studies (due to concerns over data analysis with some findings supported by single quotes); no concerns about the coherence of the finding; very minor concerns over relevance due to information in one study being driven by the study's original aim that differed from that of the

current review; no concerns over adequacy. Overall assessment of confidence was moderate due to concerns over methodological limitations as concerns over relevance were too minor to lower the overall confidence rating .

Review finding 9: Exposure to people with ME/CFS

HCPs observed that the most valuable source of evidence available to them had been observation of patients outside of the clinical setting. Seeing the activities and personal life of someone with ME/CFS helped HCPs to recognise and understand what it's like to live with the condition. For example, regular phone conversations with patients helped HCPs to observe the coming and going of symptoms and progress of management. Conversations with patients' 'significant others', that is, partners, family members and friends, could sometimes give a more accurate account of the patient's life and impact of ME/CFS symptoms than could be expressed subjectively by the person with ME/CFS.

Patient feedback was identified by HCPs as important to understand why someone may have stopped making appointments. It is sometimes unclear to HCPs whether this is because they have improved or whether there are other circumstances why they stopped engaging. This lack of information gives an inaccurate picture about the course ME/CFS takes and how patients recover.

Exposure to new presentations of ME/CFS is considered important for improving primary care practice, enabling HCPs to recognise the condition and develop confidence in their diagnostic skills. This includes careful history-taking and listening carefully and patiently to presentation of symptoms.

Explanation of quality assessment: very minor concerns over methodological limitations with no concerns over the majority of the contributing studies and minor concerns over one study (due to concerns over data analysis with some data supported by single quotes); no concerns about the coherence of the finding; very minor concerns about relevance due to the population of one study having been previously recruited for a RCT with a different research to that of this review; no concerns about adequacy. Overall assessment of confidence was high with concerns over methodological limitations and relevance being too minor to lower the confidence rating.

Table 4: Review findings for health care professionals caring for children and young people with ME/CFS

Main findings	Statement of finding
Health care professionals' awareness and knowledge of ME/CFS[68, 84]	HCPs often lack the knowledge or awareness to be able to diagnose and manage children and young people with ME/CFS.
Consensus on diagnostic criteria[84]	HCPs caring for children and young people find difficulty in reaching a diagnosis of ME/CFS, with uncertainty around diagnostic criteria and appropriate labels for young people presenting with symptoms.
Clinical pathway[84]	For HCPs caring for children and young people there is uncertainty regarding appropriate and effective treatment pathways for patients after diagnosis.
Training[84]	HCPs caring for children and young people need standardised specialist training around ME/CFS to ensure that there is consistency across services.

Information, education and support needs of health care professionals caring for children and young people with ME/CFS.

Review finding 1: Health care professionals' awareness and knowledge of ME/CFS

Paediatric specialists HCPs caring for children and young people with ME/CFS reported that they experienced uncertainty about ME/CFS due to a lack of understanding of its symptoms and the underlying aetiology. Young people with ME/CFS described the difficulties they had experienced with HCPs who lacked understanding and awareness of their condition and provided little helpful information.

Explanation of quality assessment: minor concerns over methodological limitations with minor limitations in both contributing studies (due to concerns over the small sample size and recruitment strategy in both studies); no concerns about the coherence of the findings; minor concerns over relevance due to population of one study consisting of recovered patients whose views may differ from patients with active ME/CFS; no concerns about adequacy. Overall assessment of confidence was moderate due to minor concerns over methodological limitations and relevance.

Review finding 2: Consensus on diagnostic criteria

HCPs caring for children and young people with ME/CFS found it difficult to identify and appropriate label ME/CFS. There was thought to be diagnostic variability different HCPs when diagnosing children with ME/CFS due to differing understandings of the condition and the lack of a definitive test, resulting in a difficult diagnostic process for the patient. There is also inconsistency in the labels applied to children with ME/CFS, with confusion around the terms 'chronic fatigue' and 'CFS'.

Explanation of quality assessment: minor concerns over methodological limitations in the contributing study (due to concerns over the recruitment strategy); no concerns about the coherence of the finding; no concerns about relevance; minor concerns about adequacy due to support from a single study with a small sample size. Overall assessment of confidence was moderate due to minor concerns over methodological limitations and adequacy.

Review finding 3: Clinical pathway

HCPs caring for children and young people with ME/CFS described their uncertainty around appropriateness and effectiveness of treatment pathways. HCPs believed that the choice of label given to a young person influenced the subsequent treatment and recovery pathways.

Explanation of quality assessment: minor concerns over methodological limitations in the contributing study (due to concerns over the recruitment strategy); no concerns about the coherence of the finding; no concerns about relevance; minor concerns about adequacy due to support from a single study with a small sample size. Overall assessment of confidence was moderate due to minor concerns over methodological limitations and adequacy.

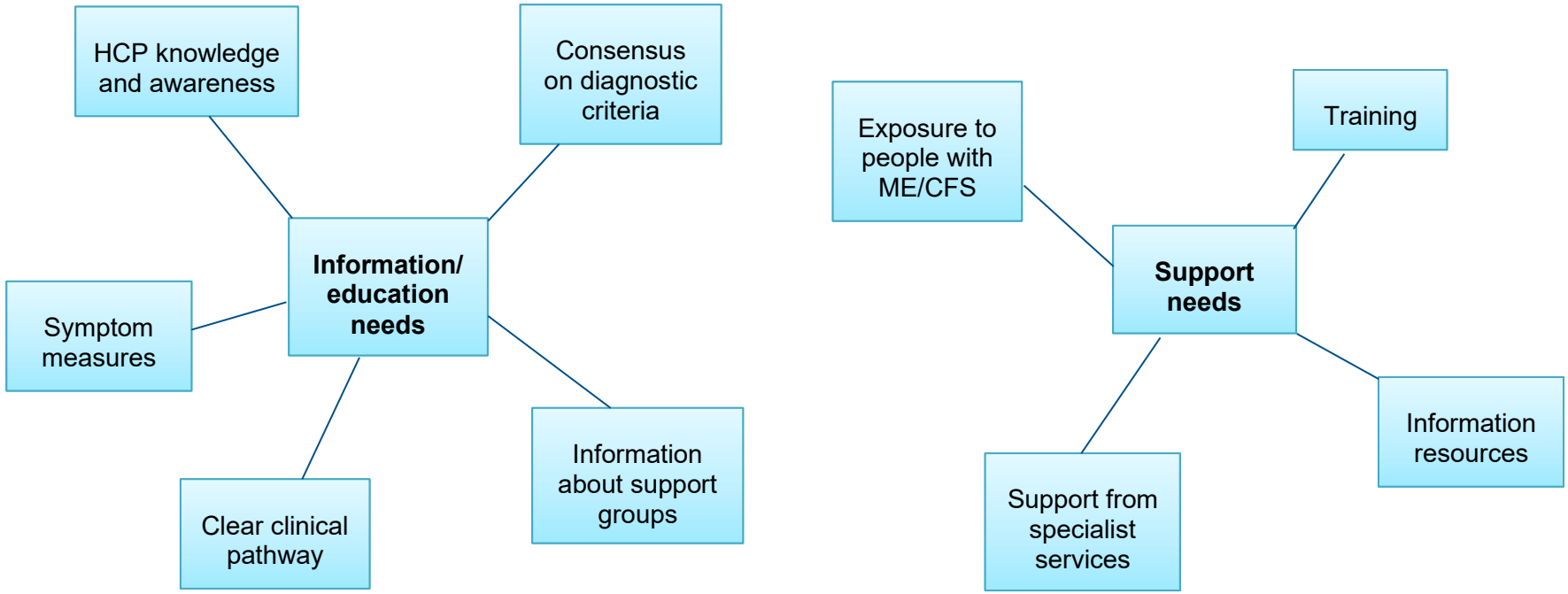
Review finding 4: Training

HCPs caring for children and young people with ME/CFS described the need for standardised specialist training to address lack of understanding and uncertainty around the condition and to ensure that there is consistency of treatment across health care services.

Explanation of quality assessment: minor concerns over methodological limitations in the contributing study (due to concerns over the recruitment strategy); no concerns about the coherence of the findings; no concerns about relevance; minor concerns about adequacy due to support from a single study with a small sample size. Overall assessment of

confidence was moderate due to minor concerns over methodological limitations and adequacy.

Figure 1 Theme map of review findings



Source/Note: No additional themes identified in children/young people that differed from those identified in adults.

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1.1.6. Economic evidence

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

2. Barriers and facilitators to providing information, education and support

2.1. Review question

What are the barriers and facilitators to providing information, education and support for health and social care professionals?

2.1.1. Summary of the protocol

Table 5: PICO characteristics of review question

Objective	To identify the barriers and facilitators to the providing of information to people with ME/CFS
Population and setting	• Health and social care professionals caring for people with ME/CFS • People with ME/CFS and their families and carers.
Context	Perceptions from health care professionals and people with or who are suspected of having ME/CFS about the barriers and facilitators to providing information, education and support.
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.

For full details see the review protocol in Appendix A.

2.1.2. 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.

2.1.3. Qualitative evidence

2.1.3.1. Included studies

Fifteen qualitative studies were included in the review;[13, 15, 18, 19, 22, 24, 31, 37, 40, 57, 66, 115, 124, 130, 147] these are summarised in Table 6 below. Key findings from these studies are summarised in the clinical evidence summary below (Table 7). See also the study selection flow chart in Appendix C, study evidence tables in Appendix D, and excluded studies lists in Appendix F.

The evidence came from studies conducted with health care professionals caring for adults with ME/CFS (n=3), adults with ME/CFS and their carers (n=6), or a mix of HCPs and adults with ME/CFS/carers (n=6). Two studies were identified that focused on children and young people with ME/CFS; these have been stratified for thematic analysis. Ten of the twelve studies featuring people with ME/CFS had a mixed or unclear severity of ME/CFS, while two studies explicitly excluded people with severe ME/CFS[22].

The evidence from the health care professionals and the adults with ME/CFS is reported together as common themes were identified in both populations.

2.1.3.2. Excluded studies

For full list of excluded studies see Appendix F.

2.1.1. Summary of studies included in the qualitative evidence

Table 6: Summary of studies included in the evidence review

Study	Design	Population	Research aim	Comments
Ax 1997[13]	Semi-structured interviews at the participant's home, analysed by content analysis.	Study 1: n=9 people with ME/CFS, mean age (SD, range): 44.2 (5.21, 16-68) years; male/female: 3/6; mean illness durations (range): 7.89 (1-14) years Study 2: n=9 people with ME/CFS, mean age (SD, range): 44.5 (7.67,34-55) years; male/female: 1/8; mean illness duration (range): 7.7 (1-19) years UK	To describe ways in which physicians and people with ME/CFS communicated their cognitions and illness beliefs which form the bases of their treatment expectations and the consequences of such interactions in terms of future treatment choices.	This report was based on two separate studies that were part of a larger project on illness adjustment.
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. This included patient resource packs for use in consultation with new and existing 'CFS/ME' patients. UK	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.	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.

Study	Design	Population	Research aim	Comments
Beasant 2014[18]	In–depth semi structured face to face interviews with thematic analysis.	N=12 adolescents; male/female 3/9; age mean (SD) 13.9 (1.6) 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	To understand the experiences of adolescents and families 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'.	Specialist Medical Intervention and Lightning Evaluation (SMILE) study designed to test 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.
Beaulieu 2000[19]	Mixture of structured and semi-structured interviews, analysed by thematic analysis.	Health professionals including general practitioners, mental health professionals (one of whom was not a physician), infectious disease specialists, immunologists and rheumatologists, recruited following identification by people with 'CFS' participating in the study. N=15; male/female 10/5; had been in practice from six to seventeen years and individually had seen from six to almost one hundred cases. People who were English-speaking and who had a diagnosis of 'CFS' from a medical doctor, recruited from physicians practices, support	To examine multiple perspectives on stigmatization and legitimation of 'CFS'.	

Study	Design	Population	Research aim	Comments
		groups and identified by leaders of associations. N=43; male/female 16/27; 26% were in school or working full or part time; mean age at onset was 34.2 years (range 15 to 58 years); people had been ill for an average of seven years. Significant others including friends, parents, spouses, adult children and a sibling, recruited following identification by people with 'CFS' participating in the study. N=23; male/female not reported; 69% were working. Stratum: adults/mixed population Canada		
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 stages of the condition; mean number of months from initial assessment to interview (SD): 12.98 (7.98), range 4 to 25) months. UK	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 participate in an interview or severely affected.
Broughton 2017[24]	Semi-structured interviews (six face-to-face, 10 via telephone) and thematic analysis.	Adults who were completing treatment for ME/CFS at one of three outpatient NHS specialist 'CFS/ME' services.	To explore the experiences of 'CFS/ME' patients who were completing programmes of treatment at three NHS specialist	NHS specialist 'CFS/ME' services followed NICE guidelines for diagnosis and management of 'CFS/ME', offering patient centred programmes aiming to increase

Study	Design	Population	Research aim	Comments
	Cross-sectional design using opportunity sampling.	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. UK Stratum: adults/mixed population	'CFS/ME' services in England.	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 of rehabilitation aimed at 'recovery' or 'significant improvement', whilst acknowledging that this would not be attained by all patients.
Chew-Graham 2008[31]	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/mixed population	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.
De Carvalho Leite 2011[37]	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 severity, duration, social variation (age, gender, ethnic background and	To investigate the impact of 'CFS/ME' on people from varied social background, including those from ethnic minorities, and what	Six of the 35 participants were purposively selected to include a diverse range of illness severity, for both an initial focus group

Study	Design	Population	Research aim	Comments
		socio-economic conditions) and year of diagnosis. UK	challenges may be posed to health care practitioners in providing appropriate and equitable care for this condition.	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 2019[40]	Semi-structured phone-based interviews with physicians and analysing the data using deductive thematic analysis.	Physicians specialising in ME/CFS of diverse medical specialties (n=10) and other physicians (n=3), not identified as ME/CFS specialists. n=13, males: 9, females: 4; mean age 60 years. For years in practice, three physicians had 30 or more years, seven had 20-29 years, one had 10-19 years and two had 1-9 years of medical experience. USA	To explore physicians views on the challenges to studying and approaching recovery, to examine these challenges in-depth and provide recommendations that will improve how researchers and practitioners approach the study and quantification of ME and CFS recovery.	Participants were recruited via non-probabilistic, purposive sampling. Specialists were defined by their extensive patient experience, research contributions and significant involvement in the field. The sample was diverse in their medical specialties: epidemiology (n=1), geriatrics (n=1), infectious diseases (n=1), neurology (n=1), internal medicine (n=2), psychiatry (n=2), general medicine (n=3), and paediatrics (n=5); three physicians identified with two medical specialties.
Hannon 2012[57]	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/mixed population	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.	

Study	Design	Population	Research aim	Comments
Horton 2010[66]	Semi-structured interviews and thematic analysis.	Specialist (n=3) and non-specialist (n=3) health care professionals who had been nominated by people with ME/CFS who had taken part in an associated England-wide study of their support needs. N=6; gender not reported. Three participants were from specialist services (medicine, occupational therapy, physiotherapy) and three were from non-specialist services (medicine, occupational health, holistic practice). 36 people with ME/CFS nominated eight HCPs as having provided them with particularly helpful or effective care and six agreed to participate. One HCP was named by six different people with ME/CFS. UK	To explore the nature of professional 'best practice' in working with people with ME/CFS.	
Ryckeghem 2017[115]	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. Patients (n=15); median age (range): 45 (33-59 years),n=14 female ; GPs (n=15); median age	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.	

Study	Design	Population	Research aim	Comments
		(range): 49 (31-62 years), n=7 female. Belgium		
Stenhoff 2015[124]	Face-to-face semi-structured interviews and inductive thematic analysis.	Undergraduate medical students in years 3, 4 and 5 at the University of Manchester, UK. N=21; 7 female, 14 male. Mean age: 22 years old. Four were third-year students, 11 were fourth-year students and six were fifth (final)-year students. Participants were recruited through the university's student-net, poster adverts around campus and via personal contact. Sampling ended at saturation in a staged approach, with two students turned away at the end of the study. UK Stratum: adults/mixed population	To investigate medical students' beliefs, attitudes and knowledge of ME/CFS.	.
Taylor 2005[130]	Focus group interviews, open-ended questionnaires, progress notes, and a program evaluation questionnaire, with thematic analysis using grounded theory approach	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	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
		minority ethnicity, 39 were non-minority. USA Stratum: adults/mixed population		
Woodward 1995[147]	Two related investigations: qualitative interviews with GPs and a longitudinal study comprising three qualitative interviews with patients.	General practitioners, N=20; male/female: 9/11. People diagnosed by doctors as having CFS. N=50; male/female: 10/40; females mean age (range): 36.4 (13 to 64) years; males mean age (range): 39.2 (25 to 53) years. Australia	To examine doctors' and patients' views on the risks and benefits of the symptomatic diagnosis of CFS.	

See Appendix D for full evidence tables.

2.1.2. Summary of the qualitative evidence

Table 7: Review findings, adults with ME/CFS, severity mixed or unclear

Main findings	Statement of finding
Facilitator: Communication/ relationship between HCP and patient[40,66,115,31,19,147]	Building a relationship between HCP and patient allows better provision of information and support, and frequent contact improves understanding of ME/CFS for both HCP and patient.
Facilitator: Referral to specialist services[24,37,15]	Specialist services were seen as the best provider of information and support for people with ME/CFS, with referral to specialists providing a positive experience after a long road to diagnosis.
Facilitator: Online training resource[15]	HCPs valued online training resources that showed how to work with people with ME/CFS in a consultation setting, with several GPs finding video resources particularly useful for this.
Barrier: Limited specialist referral options[15]	While referral to specialists and other support services was seen a route to provide information and support, GPs often do not know when or where they should refer people with ME/CFS.
Barrier: Limited knowledge of support groups[57,130,19]	HCPs often do not have sufficient information to be able to refer people with ME/CFS to support groups and can be hesitant to do so because of mixed beliefs about their effect on the patient.
Barrier: Lack of training[124,57]	There is a lack of training and education available for HCPs, GPs in particular, on how to manage people with ME/CFS, beginning with an absence of ME/CFS on the university medical curriculum.
Barrier: Reluctance of GPs to training[15]	Some GPs are reluctant to take on the management of people with ME/CFS, preferring to refer to secondary care specialists, and do not always engage with ME/CFS training when offered.
Barrier: Consultation time constraints[15]	Due to the complexity of ME/CFS and its symptoms, HCPs often find that the nature of clinic time and short consultation lengths do not allow for effective communication and support.
Barrier: cognitive and physical functioning of ME/CFS patients and the impact to engage with services[66]	People with ME/CFS, particularly those with severe ME/CFS, have limited energy and combined with the impact of cognitive difficulties find it difficult to engage and receive information, and support from HCPs.
Barrier: Information overload[115]	People with ME/CFS can sometimes experience an overload of information during the care process which can negatively affect their understanding of the condition.
Barrier: Fear of negative reactions[19,13]	HCPs can be hesitant to provide information and discuss psychological factors around ME/CFS with patients due to concerns about patients' possible negative reactions.
Barrier: Uncertainty and lack of confidence in information[19]	Uncertainties associated with ME/CFS mean that HCPs are often unsure about the reliability of information they have, making them cautious and hesitant when explaining the condition.

See Appendix E for full GRADE-CERQual tables.

2.1.2.1. Narrative summary of review findings: people with ME/CFS, severity mixed or unclear

Facilitators

Review finding 1: Communication/relationship between HCP and patient

The importance of building a relationship between the HCP and person with ME/CFS for providing support was highlighted. Not having ongoing contact with an individual HCP made the communication of information and provision of support difficult as it took time to reach agreement and understanding about symptoms and fluctuation of the condition. Some patients expressed the need for a dedicated individual who could provide continuity by accompanying them to consultations and to inform, advise, instruct and assist them at all stages of the care process.

HCPs who followed up with their patients regularly by email, over the phone or through home visits found that this communication benefited both the HCP and the patient. It allowed the HCP to gain feedback about their practice and the support it provides, as well as improving the HCP's knowledge of ME/CFS by better understanding the course and fluctuation of the illness. Phone contact in particular was necessary and appropriate for specialists to provide support to people with severe ME/CFS who are house bound.

Explanation of quality assessment: moderate concerns over methodological limitations with serious concerns in one study (due to the lack of discussion of the potential impact of the researcher on the findings not being discussed and participant recruitment was unclear and risk of bias in the data analysis since type of analysis and details are not provided), moderate concerns in two studies (due to the lack of discussion of potential impact of role of the researcher on the findings in one study and concerns over data analysis in both studies due to a lack of sufficient detail and findings mostly supported by single quotes in one study and due to the analysis being done by a single researcher in the other study and concerns over participant recruitment in one study) and minor concerns in three studies (due to the role of the researcher and minor concerns over data analysis with findings mostly supported by single quotes) ; no concerns about coherence; moderate concerns of relevance due to indirectness of the study's research aims (three studies), indirect population samples due to participants having been previously recruited in a different study (two studies), concerns over the relevance of one study (conducted in the Belgian healthcare setting) to the NHS setting, and due to time since publication of two studies conducted in 1995 and 2000 prior the development of new guidelines and diagnostic criteria; no concerns about adequacy. Overall assessment of confidence was moderate due to the methodological limitations and concerns over relevance identified.

Review finding 2: Referral to specialist services

People with ME/CFS found that they received the most information and support when they referred to specialist services. These specialist services provided information and an explanation of ME/CFS which had been previously difficult to find from GP services. Diagnosis is a key step in this process, with most people with ME/CFS finding that diagnosis was a key milestone which led to a positive experience in which they received useful advice and support from health care professionals with particular knowledge of ME/CFS.

This theme is supported by three studies with an overall assessment of high confidence. Explanation of quality assessment: very minor concerns over methodological limitations in two studies (due to the lack of discussion of role of the researcher) and no limitations in the other contributing study; no concerns about coherence; very minor concerns about relevance (only one study had very minor concerns due to a population who may have already received support and/or information prior to the study); no concerns about adequacy of information supporting the theme. Overall assessment of confidence was high as concerns over

methodological limitations and relevance were very minor and did not lower the confidence rating.

Review finding 3: Online training resource

HCPs valued online training resources that improved their knowledge of ME/CFS and prepared them to provide information and build positive relationships with people with ME/CFS. One study found that an online training module that used video clips to show how a GP can work with a person with ME/CFS within a consultation setting was particularly valued by GPs.

Explanation of quality assessment: very minor concerns over methodological limitations in the contributing study (due to the lack of discussion of the role of the researcher); no concerns about coherence; no concerns about relevance; moderate concerns of adequacy due to the broadly-applicable theme being found only in a single study. Overall assessment of confidence was moderate due to concerns over adequacy and concerns over methodological limitations being too minor to lower the confidence rating.

Barriers

Review finding 4: Limited specialist referral options

While referral to specialist services was considered one of the best ways to support and provide information to people with ME/CFS, this is often limited by few referral options. In these cases, GPs were unsure when they should refer, where they should refer or what the specialist services could offer. Contact between GPs and specialist services could also be complicated by changes in specialist services such as redesigns or loss of contact with ME/CFS specialists who retire or relocate.

Explanation of quality assessment: very minor concerns over methodological limitations in the contributing study (due to the lack of discussion of the potential influence the researcher on the findings not being discussed); no concerns about coherence; no concerns about relevance; and moderate concerns of adequacy due to the broadly-applicable theme being found only in a single study. Overall assessment of confidence was moderate due to concerns over adequacy, as methodological limitations were too minor to further lower the confidence rating.

Review finding 5: Limited knowledge of support groups

Patients and carers highlighted the need for more signposting from their GP to information on local support groups, advice on benefits and referrals to the third sector, however most GPs and practice nurses do not have details of these contacts or are reluctant to make recommendations due to lack of knowledge of support groups. People with ME/CFS reported problems with gaining access to disability income, workplace accommodations or community-based resources because their GPs were unaware of such resources or because the physicians were unconvinced of the need for this type of support.

HCPs attitudes towards support groups varied, with most clinicians hesitant to recommend support groups because they had little knowledge of them and some believing that support groups could be harmful as well as helpful, depending on their approaches and the individual patient. Many saw this problem as not knowing how an individual would be affected by the methods of these external groups. For example, some patients could become devastated after being exposed to the worst scenarios of ME/CFS.

Explanation of quality assessment: minor concerns over methodological limitations with moderate limitations in one study (due to concerns over participant selection and data analysis (coding and analysis by a single researcher), but minor concerns over one study (due to the role of the researcher and lack of data richness with findings mostly supported by single quotes) and no concerns over the third contributing study; no concerns about

coherence; minor concerns about relevance due to participants in one study being recruited from a previous study with a different research aim (evaluating a participant-designed rehabilitation program) and concerns due to its year of publication (2000) that preceded current guidelines and diagnostic criteria, but no concerns in the other two contributing studies; no concerns about adequacy. Overall assessment of confidence was moderate due to minor concerns over methodological limitations and relevance.

Review finding 6: Lack of training

There is a lack of training and education around ME/CFS available for HCPs, GPs in particular, and this contributes to a lack of knowledge among HCPs and therefore a lack of opportunity to educate people with ME/CFS who attend their services. ME/CFS specialists highlighted a training need in primary care, where GPs and practice nurses have varying degrees of understanding of ME/CFS and some question its legitimacy. This lack of training begins at university, where ME/CFS is largely absent from the medical curriculum.

Explanation of quality assessment: minor concerns over methodological limitations with minor concerns in both contributing studies (due to potential selection bias in one study due to recruitment of participants in response to an advertisement therefore risking over-representation of people who are more informed or have stronger views on ME/CFS and due to the lack of discussion of role of the researcher and lack of data richness with findings mostly supported by single quotes in the other study); no concerns about coherence; minor concerns about relevance due to indirect population of one study that included medical students rather than practicing healthcare professionals; minor concerns about adequacy due to the broadly-applicable theme being based on only two studies. Overall assessment of confidence was moderate due to minor concerns over methodological limitations, relevance and adequacy.

Review finding 7: Reluctance of GPs to training

Some GPs show reluctance to manage people with ME/CFS and in one study it was found that some practices were unwilling to engage in training on ME/CFS management. Reasons for lack of engagement include scepticism about ME/CFS and the complexity of managing the condition and working with patients and their families. Some GPs were seen to prefer referral to special services than to manage and support people with ME/CFS themselves, implying that the long-term commitment to manage this type of patient was too much for primary care professionals and that this was best left to secondary care specialists. Other reasons given for lack of engagement with ME/CFS included the small number of patients with the condition, pressures on time within a consultation and suggestion that ME/CFS was not a priority.

Explanation of quality assessment: very minor concerns over methodological limitations in the contributing study (due to the role of the researcher not being discussed; no concerns about coherence; no concerns about relevance; moderate concerns of adequacy due to the broadly-applicable theme being found only in a single study. Overall assessment of confidence was moderate due to concerns over adequacy and concerns over methodological limitations being too minor to further lower the confidence rating.

Review finding 8: Consultation time constraints

For GPs, the nature of clinic time and consultation length was a limiting factor for delivery of information and support. Both GPs and patients reported that a ten-minute consultation was not sufficient for the patient to communicate the complexity of their experience or for GPs to deliver information and manage the person with ME/CFS.

Explanation of quality assessment: very minor concerns over methodological limitations in the contributing study (due to the lack of discussion of the role of the researcher not being discussed); no concerns about coherence; no concerns about relevance; moderate concerns of adequacy due to the broadly-applicable theme being found only in a single study. Overall

assessment of confidence was moderate due to concerns over adequacy and concerns over methodological limitations being too minor to further lower the confidence rating.

Review finding 9: Cognitive and physical functioning of ME/CFS patients (particularly those with severe ME/CFS) and the impact on receiving information

For people with severe ME/CFS, their ability to receive information and support from specialists was particularly affected as a result of reduced cognitive and physical functioning, with some people with severe ME/CFS unable to communicate effectively. This was seen to be extremely challenging for HCPs, particularly specialists, who did not know how to address this problem.

Explanation of quality assessment: minor concerns over methodological limitations in the contributing study (due to the role of the researcher and concerns over data analysis with some findings supported by single quotes); no concerns about coherence; no concerns about relevance; moderate concerns about adequacy due to the broadly-applicable theme being found only in a single study. Overall assessment of confidence was low due the methodological limitations and concerns over adequacy identified.

Review finding 10: Information overload

People with ME/CFS explained that they could sometimes suffer from an information overload from health care services, which could hamper their understanding of the condition. It was expressed that providing the right information at the right time was important to support people with ME/CFS.

Explanation of quality assessment: moderate concerns over methodological limitations in the contributing study (due to the role of the researcher, and concerns over data analysis with a lack of sufficient detail and findings mostly supported by single quotes; no concerns about coherence; moderate concerns about relevance due to setting (Belgian healthcare service); moderate concerns about adequacy to the broadly-applicable theme being briefly described in a single study. Overall assessment of confidence was very low due to moderate concerns over methodological limitations, relevance and adequacy.

Review finding 11: Fear of negative reactions

When patients' views differed from the advice that HCPs gave them, the advice could be met with anger, particularly when HCPs advised to find psychological or psychiatric support. HCPs sometimes hesitated to discuss psychological factors around ME/CFS with patients due to concerns about patients' possible reactions. This could even include reluctance to discuss concurrent psychological disorders that they detected. Because of this perception of people with ME/CFS as resistant to any suggestion of psychological disorder, HCPs often try to avoid stigmatising explanations; however, they also felt that avoiding labels and detailed discussions may leave the patient with a sense of ambiguity.

Explanation of quality assessment: moderate concerns over methodological limitations with moderate concerns in both contributing studies (due to role of the researcher, lack of detail on data analysis method one study, concerns over participant recruitment and data analysis as coding and analysis was done by a single researcher in the other study); no concerns about coherence; minor concerns about relevance due to concerns over one study (with main emerging findings being driven by the study's original aims (to explore multiple perspectives on stigmatisation and legitimation of ME/CFS and its year of publication (2000) preceded present guidelines and diagnostic criteria) but no concerns over the other contributing study; minor concerns about adequacy due to broadly applicable theme being based on two studies. Overall assessment of confidence was low due to concerns over methodological limitations, relevance and adequacy.

Review finding 12: Uncertainty and lack of confidence in information

Uncertainties associated with ME/CFS mean that HCPs are often uncertain about the accuracy and reliability of the information they have, making them cautious and uneasy when explaining the condition. This was seen to be due to conflicting medical findings and opinions about ME/CFS, complicated by the lack of diagnostic tests or laboratory findings. Some HCPs felt that their credibility was at stake if their explanations were proven wrong in the future, resulting in physicians using a cautious and tentative tone when delivering an explanation of the illness to patients.

Explanation of quality assessment: moderate concerns over methodological limitations in the contributing study (due to concerns over participant recruitment and data analysis with coding and analysis by a single researcher); no concerns about coherence; moderate concerns about relevance due to main emerging findings being driven by the study's original aims (to explore multiple perspectives on stigmatisation and legitimation of ME/CFS) and concerns over relevance arising from the fact that the finding emerged from a single study that due to its year of publication (2000) present guidelines and diagnostic criteria; moderate concerns about adequacy due to the broadly-applicable theme emerging from only one study. Overall assessment of confidence was very low due to moderate concerns over methodological limitations, relevance and adequacy.

Table 8: Review findings, children and young people with ME/CFS, severity mixed or unclear

Main findings	Statement of finding
Facilitator: Referral to specialist services[18]	Referral to specialist services provided children and young people with ME/CFS and their parents with information and support, as well as a letter allowing educational adjustments.
Facilitator: Digital social support[22]	Digital social support websites such as health forums and other social media sites provide quick, simple and undemanding access to social support, reducing isolation.
Barrier: Unhelpful or unrelatable NHS information resources[22]	NHS resources lack the accessibility and relatability provided by patient- and peer-led websites in terms of language and narrative approach used.

See Appendix E for full GRADE-CERQual tables.

2.1.2.2. Narrative summary of review findings: children and young people with ME/CFS, severity mixed or unclear

Facilitators

Review finding 1: Referral to specialist services

When children and young people with ME/CFS and their parents received a referral to specialist services, they were given access to experts who provided them with information about the condition, guidance on management, sometimes an official diagnosis, and tailored patient-centred specialist medical support that they had not received previously. ME/CFS specialist services also provided confirmatory communication to schools that allowed mothers to legitimately take their child out of school, request funds for home schooling, or make other appropriate supportive adjustments in cooperation with teachers.

Explanation of quality assessment: minor concerns over methodological limitations in the contributing study (due to the unclear relationship between the researcher and participants and concerns over data richness with findings mostly supported by single quotes); no concerns about coherence; minor concerns about relevance due to indirect study aims (to understand the experiences of accessing and using specialist services) and lack of clarity

around which intervention the findings relate to and the representativeness of the sample considering it consisted of feasibility RCT participants which may differ from eligible patients not recruited to a trial; moderate concerns about adequacy due to the broadly-applicable theme being found only in a single study. Overall assessment of confidence was low due to concerns over methodological limitations, relevance and adequacy.

Review finding 2: Digital social support

Children and young people with ME/CFS expressed a preference for online information and social support. Accessible and appealing social support sites included health forums as well as non-health-related sites such as Facebook, Instagram, blogs and YouTube. The speed at which they could access these resources at any time provided a great sense of support and reduced feelings of loneliness and isolation. Specific features of such support sites that children and young people found appealing were the shared language of likes and comments to give a connection to others. This online social system was seen as less demanding and more flexible than offline relationships in the context of a disabling and fluctuating illness.

Explanation of quality assessment: minor concern over methodological limitations in the contributing study (due to role of the researcher not being discussed and lack of details on the data analysis); no concerns about coherence; minor concerns about relevance due to the study not including severely affected adolescents; and moderate concerns about adequacy due to the broadly-applicable theme being found only in a single study. Overall assessment of confidence was low due to concerns over methodological limitations, relevance and adequacy.

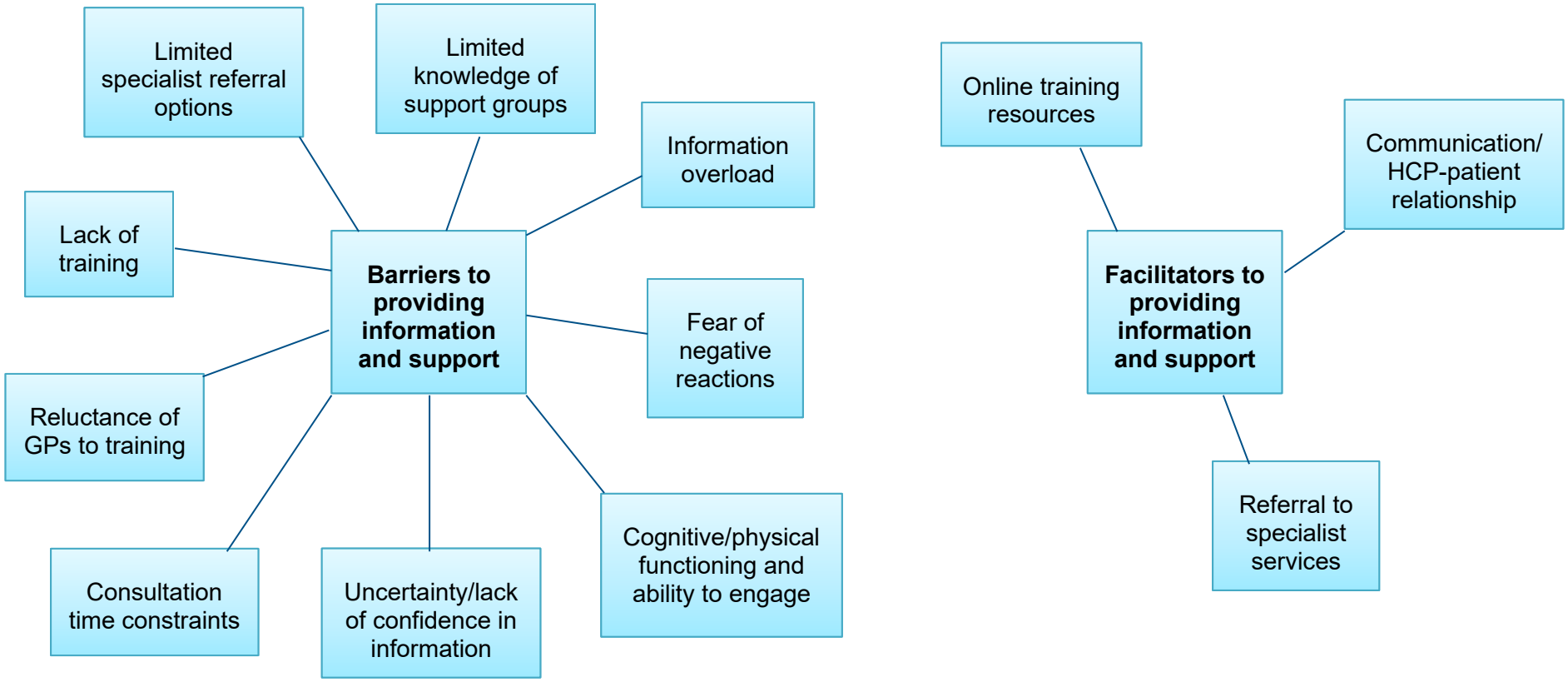
Barrier

Review finding 3: Unhelpful or unrelatable NHS information resources

Children and young people with ME/CFS found that NHS websites providing information on ME/CFS were not user-friendly because they used medical terminology, lacked depth and were not up to date. By comparison, this population felt that patient- and peer-led websites were more helpful and reliable, using terms and phrases that were more accessible and appealing and offering greater depth. There was a preference for the story-telling approach used by patient- and peer-led resources and the videos used by these sites.

Explanation of quality assessment: minor concerns over methodological limitations in the contributing study (due to lack of discussion of the role of the researcher and lack of details on the data analysis); no concerns about coherence; minor concerns about relevance due to the study not including severely affected adolescents; moderate concerns about adequacy due to the broadly-applicable theme being found only in a single study. Overall assessment of confidence was low due to concerns over methodological limitations, relevance and adequacy.

Figure 2 Theme map of review findings (adults)



Source/Note: Some themes could be considered barriers or facilitators to providing information, education and support depending on their presence/absence, e.g. ability of people with ME/CFS to engage with health care services.

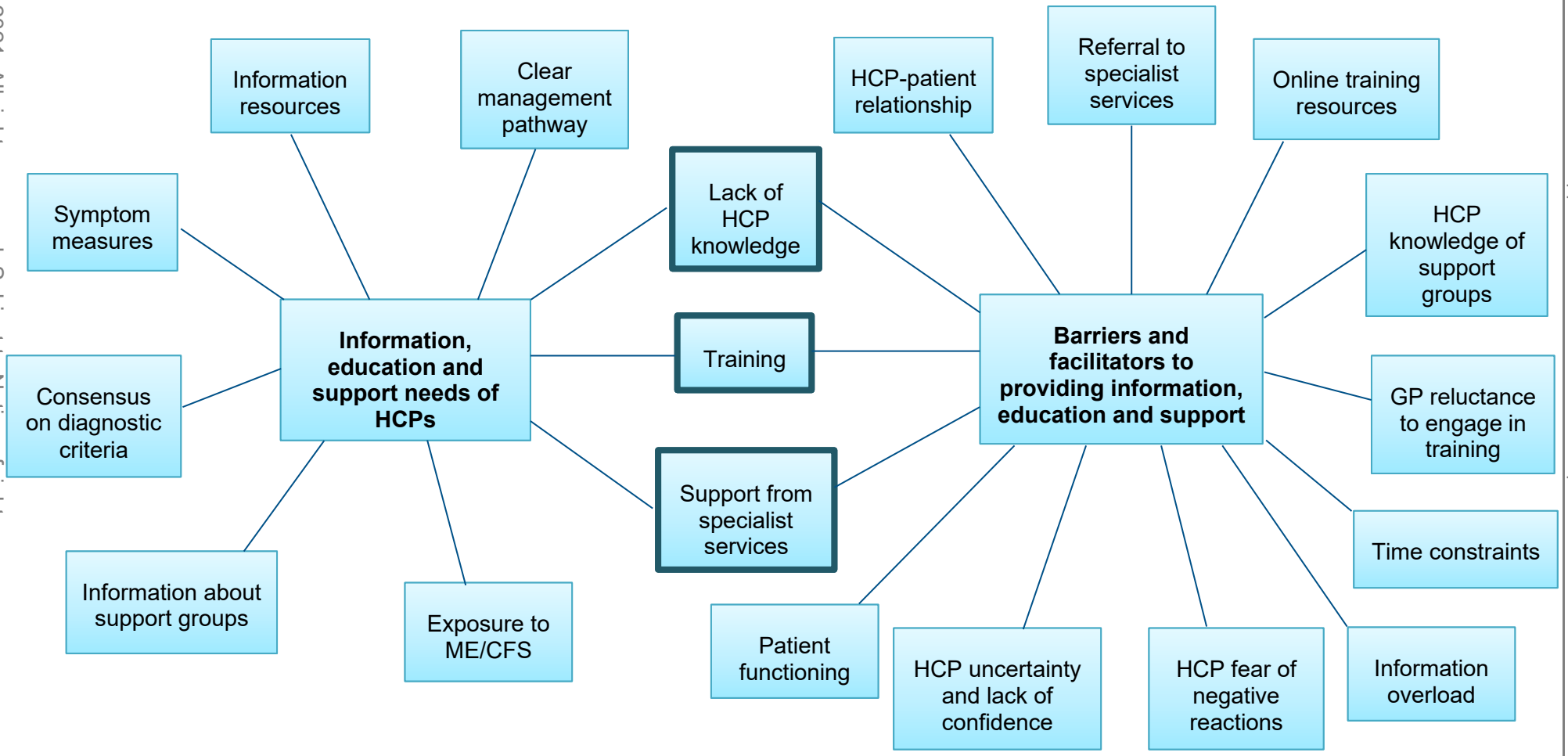
Figure 3: Theme map of review findings (children/young people)

Source/Note: Only additional themes identified in children/young people that differed from those identified in adults are displayed here.

2.1.3. Economic evidence

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

Figure 4 Map of overlapping themes in the review findings



3. The committee's discussion and interpretation of the evidence

The committee discussion of the review on information, education and support for health and social care professionals caring for people with ME/CFS and the review on barriers and facilitators to providing information to people with ME/CFS and the expert testimony on information, education and support for health and social care professionals caring for people with ME/CFS (see Appendix 3: Expert testimonies) are included here.

The committee discussion was also informed by the findings from the evidence reviews on evidence report C: access to care, evidence report A: information for people with ME/CFS, evidence report D: diagnosis and evidence report I: multidisciplinary teams. Where relevant these sources are noted.

3.1. The quality of the evidence

Information, education and support for health and social care professionals caring for people with ME/CFS

Sixteen qualitative studies were included in the review. The majority of the evidence was based on health care professionals (HCPs) caring for adults with ME/CFS and adults with ME/CFS. One study included adolescents who had recovered from ME/CFS and one study included health care professionals caring for children and young people with ME/CFS. One study included 'significant others' of people with ME/CFS. No evidence was identified for social care professionals caring for people with ME/CFS.

Confidence in the review findings ranged from high to moderate. Main reasons for downgrading were methodological limitations and relevance. The most common methodological limitations identified were insufficiently rigorous data analyses, with findings being supported by single quotes and limited explanations and the relationships between the researchers and participants not being adequately reported.

Most findings were from studies that had different research aims to the aim of this review. Several findings were directly applicable and despite minor concerns regarding their relevance reducing the committee's confidence in those findings, the committee agreed they contributed useful information that could support decision making. There were concerns regarding the relevance of findings from some studies on subgroups of the review population, such as women with ME/CFS and those with suicidal ideation. One study was based on medical students rather than practicing health care professionals caring for people with ME/CFS. The committee considered that although these views may not be directly applicable to practicing health care professionals, they were valuable to build a more complete picture of the information, education and support needs of health care professionals working with people with ME/CFS.

Some findings, particularly those identified for children and young people were based on evidence from a small number of studies, which meant that coherence was less clear and there were some concerns about the adequacy of data.

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.

Barriers and facilitators to providing information to people with ME/CFS

Fifteen qualitative studies were included in the review. The majority of the evidence was based on health care professionals caring for adults with ME/CFS and adults with ME/CFS.

Two studies were based on children and young people with ME/CFS. Ten of the twelve studies on patients/carers involved people with a mixed or unclear severity of ME/CFS, while two studies excluded people with severe ME/CFS.

Confidence in the review findings ranged from high to very low. Main reasons for downgrading were methodological limitations, relevance and adequacy. The most common methodological limitations identified were insufficient reporting of methods of data analysis, insufficient data presented to support findings, for example single quotes only and the relationships between the researchers and participants not being adequately considered or reported.

There were concerns regarding the relevance of findings from studies that had different research aims to the aim of this review, however findings were deemed to be directly applicable, and the committee agreed they contributed useful information that could support decision making. There were concerns regarding the relevance of findings from the studies that excluded people with severe ME/CFS, although the committee reflected that none of the studies were likely to have included people with severe or very severe ME/CFS due to difficulties conducting research in this group. One study was based on medical students rather than practicing health care professionals and one study was based on the Belgian healthcare setting. This evidence may not be directly applicable to practicing health care professionals in the NHS, but they were valuable to build a more complete picture of the information, education and support needs of health care professionals working with people with ME/CFS.

Six of the studies were published over a decade ago and the committee discussed their relevance to current practice. The committee agreed that many of the issues identified by health and social care professionals and people with ME/CFS then are the same today.

The majority of the findings were based on single studies; online training resources, limited specialist referral, reluctance of GPs to training, consultation time constraints, capacity of ME/CFS patients, information overload, and uncertainty and lack of confidence in the information. Coherence was less clear in these findings with concerns about the adequacy of data supporting them.

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 some were also identified in other qualitative reviews across the guideline. For example, the length of consultations does not allow for effective communication and support was rated ‘low’, but in the review of barriers and facilitators to care for people with ME/CFS (see evidence report C: access to care) time limited consultations were identified as a barrier to the provision of appropriate care, and there was moderate confidence in this finding.

3.2. Findings identified in the evidence syntheses

The review questions outlined at the start of this report are an exploration of the needs of health and social care professionals for information, education and support and a separate question exploring in more detail how this can be implemented and achieved. It is clear from the reviews that many studies addressed both perspectives and 10 studies are included in both reviews. Figure 4 illustrates the overlapping themes in the 2 reviews.

To avoid duplication the committee discussion of the findings from both reviews are reported under the following headings: Information and education needs (knowledge about ME/CFS, understanding living with ME/CFS), support from specialist services, and provision and delivery of training. The needs are described and then the barriers and facilitators highlighted. Themes identified for children and young people broadly mirrored those identified for adults. Findings unique to children and young people are highlighted.

The committee considered that the findings identified in these reviews were consistent with their experience and the expert testimony.

Information and education needs

Knowledge about ME/CFS

Lack of knowledge and beliefs about ME/CFS and the need for education and training to address these emerged as an important theme in both reviews. In the information and support review evidence suggested that HCPs have a lack of knowledge and belief in ME/CFS. This results in a lack of confidence and ability to diagnose and manage people with ME/CFS, leading to delays in diagnosis and referral and mismanagement of patients. This was expressed by both HCPs and people with ME/CFS. Lack of knowledge, understanding and belief in ME/CFS is a theme echoed in the evidence identified on children and young people and in evidence report C: access to care and evidence report A: information for people with ME/CFS.

In the barriers and facilitators review lack of training and education across healthcare practitioners in how to manage ME/CFS was identified as a barrier to HCPs providing information and support to people with ME/CFS. The improved education of HCPs about ME/CFS and an increased presence of training about the condition in the medical curriculum was identified as key to address the lack of understanding and belief about ME/CFS. This theme was echoed in the evidence identified on children and young people and in evidence report C: access to care and evidence report A: information for people with ME/CFS.

These findings were reflected by Dr Muirhead in her expert testimony. She described experiencing a significant change in her understanding and beliefs about ME/CFS after becoming ill with ME/CFS. Her experience has been that the information, education and support provided by medical bodies is mostly outdated, misleading and not in line with patient experience. In particular, she expressed concerns that ME/CFS training and education is not mandatory, is merged with medically unexplained symptoms and is based on theories of deconditioning and fear avoidance of exercise. This lack of and misinformation leads to incongruity between an HCP's understanding of the illness and the experience of patients. This disparity can compromise the relationship between people with ME/CFS and the healthcare professionals they meet. In the worst case poor training and poor information may cause harm.

The committee considered the qualitative evidence of the lack of knowledge and awareness of ME/CFS among HCPs and the need for more and better training, alongside the expert testimony. The experiences recounted by Dr Muirhead resonated with several members of the committee, particularly the lay members, many of whom had shared similar experiences. The committee discussed the potential harms of the lack of awareness, knowledge and the misunderstanding among HCPs to the care of people with ME/CFS and agreed that people with ME/CFS should have their care and treatment delivered by or as a minimum overseen by health and social care practitioners who have training and experience in ME/CFS relevant to their role. They agreed anyone delivering care to people with ME/CFS should have access to training and maintain continuous professional development in ME/CFS. The committee discussed that evidence for social care professionals is lacking but this was equally as important to consider. The committee agreed the training recommendations should apply to social care professionals but recognised this might be different in some of the content from the training for HCPs. To reinforce the importance of training and education the committee made a recommendation directed at service providers that access to training for people with ME/CFS should be provided for all staff that have contact with or deliver care to people with ME/CFS and should include information on what ME/CFS, including diagnosis, management and monitoring, and the experiences of people with ME/CFS.

Suspecting and diagnosing ME/CFS

In the information, education and support review, evidence suggested there the lack of a confirmed consensus on the diagnostic criteria for ME/CFS meant that there was confusion among HCPs in when to suspect or diagnose ME/CFS. HCPs expressed the need for agreed case definitions for both diagnosis and recovery. The lack of agreed tests and measurements for ME/CFS symptoms mean that HCPs are reluctant to make a diagnosis based on limited clinical signs and struggle to assess recovery. Similarly, HCPs caring for children and young people find difficulty in reaching a diagnosis of ME/CFS, with uncertainty around diagnostic criteria and appropriate labels.

The committee considered these findings alongside the review on diagnostic criteria (see evidence report D: diagnosis). The committee noted that diagnostic criteria vary between centres and sometimes a hybrid of different criteria and clinical judgement used. The committee discussed different symptom measurement scales for diagnosis and recovery, including the Checklist Individual Strength and the DePaul Symptom Questionnaire, but there was little consensus on the value of these measures. Furthermore, no evidence was identified on the accuracy of these scales for predicting diagnosis in the review of predictive tests/signs/symptoms (see evidence report E: strategies before diagnosis). The committee acknowledged there is uncertainty around how to identify and diagnose someone with ME/CFS and noted that in their experience this can result in people being undiagnosed and seeing many doctors and specialities until they meet a clinician with enough knowledge to recognise and diagnose ME/CFS. The criteria for suspecting and diagnosing ME/CFS is discussed in the report on diagnosis. The committee agreed that with recommendations in this guideline on suspecting ME/CFS and with training GPs should have the confidence to consider a ME/CFS diagnosis and refer to a ME/CFS specialist for confirmation of the diagnosis (see evidence report I: multidisciplinary care for discussion on referral to specialist ME/CFS services).

Management of ME/CFS

In the information and support review evidence suggested there is need for a clearer clinical management pathway for ME/CFS. The committee considered this finding alongside the lack of evidence of clinical effectiveness for many of the interventions reviewed in this guideline. The committee discussed the frustration and sometimes anxiety that could be experienced by health and social care professionals when they are unable to cure or even improve the symptoms of people with ME/CFS.

The committee acknowledged the frustration of non-specialists in ME/CFS not knowing what to do or where to get support but also the helplessness of patients not receiving access to the expertise and management they require. The committee agreed that training should include education on the lack of evidence of any curative interventions, but also focus on ways that health and social care professionals can provide care, including advice on symptom control, managing relapse, reviewing and monitoring, access to services and tailoring access according to the person's needs. The committee considered the guideline recommendations on management of ME/CFS and managing relapse, including specific recommendations for people with severe or very severe ME/CFS and children and young people, should increase knowledge and confidence regarding management.

The committee discussed providing guidance on specific areas to include in training programmes. They recognised that the content of training programmes should be specific to the role of the practitioner, for example it is appropriate a GP should have training on how to provisionally diagnose ME/CFS but this would not be relevant to a social worker. They acknowledged that if interpreted rigidly providing a list of areas for training can be counterproductive with only the topics listed included. Taking this into account the committee decided a list would not be useful in this context but recommended that training should be relevant to the professional's role so they are able to care for people in accordance with this guideline. The committee were aware of the availability of out of date training materials and

emphasised that training should reflect current knowledge in ME/CFS, and this should include understanding what ME/CFS is.

Providing information for people with ME/CFS

Throughout the reviews in the guideline, it is clear that HCPs lack knowledge about ME/CFS (in evidence report C: access to care and evidence report A: information for people with ME/CFS.) and this is followed by a lack of confidence in providing resources for people with ME/CFS.

In the barriers and facilitators review evidence suggested that HCPs are often unable to recommend support groups or give advice on benefits because they have little knowledge or information about them or relevant contacts. The committee agreed that this was their experience and considered this in relation to the benefits of such information and advice for people with ME/CFS and their families and carers identified in other reviews. The committee noted there are many groups that offer information for people with ME/CFS and some that are unmoderated and suggest advice that is controversial and can be unhelpful.

In both reviews evidence suggested that HCPs need trustworthy good quality resources that can be used during consultations to educate and reassure patients when diagnosed with ME/CFS. HCPs from specialist services reported 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. The committee agreed that training for HCPs should include information on accessing support services (for example, patient support groups) so they can pass this information on to patients.

This committee noted this was particularly pertinent to children and young people with ME/CFS and the difficulties they have in accessing relevant and accessible information. In the barriers and facilitators review evidence suggested that NHS resources lack the accessibility and relatability provided by patient- and peer-led websites in terms of language and narrative approach used. Evidence also suggested that digital social support websites such as health forums and other social media sites provide quick, simple and undemanding access to social support, reducing isolation. These findings were also identified in the review of information, education and support needs of people with ME/CFS and discussed further in that report and support the recommendation to consider using different formats such as digital media, including social media where appropriate when providing information for children and young people with ME/CFS.

In the barriers and facilitators review evidence suggested that specialist services were seen as the best provider of information and support. This was supported by the finding that referral to specialist services provided children and young people with ME/CFS and their parents with information and support, as well as a letter allowing educational adjustments.

Understanding about living with ME/CFS

HCP –patient relationship

In the barriers and facilitators review evidence suggested that building a relationship between the HCP and person with ME/CFS allows better provision of information and support and frequent contact improves understanding of ME/CFS for both parties. The committee noted that good and ongoing HCP-patient relationships were highlighted as a support need in evidence report A: information for people with ME/CFS, as a facilitator in evidence report C: access to care and as important to a positive experience of several interventions in the review of experiences of interventions for ME/CFS (see evidence report G: non pharmacological management).

The committee considered this theme was in keeping with existing models of good clinical communication and therapeutic relationships and were mindful of these when making recommendations on principles of care emphasising the importance of taking the time to

build supportive, trusting and empathetic relationships. Specifically, the committee cross referred to the principles on communication, information giving and shared decision making in the NICE guidelines on patient experience in adult NHS services and people's experience in adult social care services. Existing models of good clinical communication and therapeutic relationships also guided the development of recommendations regarding the content, approach and delivery of management interventions.

In the barriers and facilitators review evidence suggested that HCPs can be hesitant to provide information and in particular discuss psychological factors around ME/CFS due to concerns about patients' possible negative reactions. The committee discussed the sensitivities around the topic of psychological symptoms and considered evidence from other reviews that people may have experienced disbelief in the past or the implication that their condition is, 'all in their head'. It was also acknowledged that although there are several theories, the definitive causation of ME/CFS is still unknown. The committee agreed that a holistic approach which takes into account the person's needs is most appropriate to assessment, provision of information and management, and the recommendations throughout the guideline reflect this.

The committee discussed the difference between patients' expectations and their experience with HCPs and the problems this causes. The committee agreed that training should take a holistic view and HCPs should be encouraged to listen to patients and the experience of their symptoms, acknowledging to the person with ME/CFS the reality and potential impact of ME/CFS and their symptoms.

In the information, education and support review evidence suggested that experience of working with people with ME/CFS enabled HCPs to recognise the condition and develop confidence in their diagnostic skills. Contact with people with ME/CFS outside of the clinical setting improved HCPs understanding of the lived experience of the condition and raised awareness about the challenges that some people with ME/CFS face. Some of the committee members knew of people with ME/CFS who are reluctant to engage with health and social services and have stopped contact after they have been unable to get the help or support they need. The committee noted that GPs are also unlikely to have had significant experience with people with the most severe symptoms or to have seen people when they are experiencing severe symptoms as these people are often unable to attend appointments (also see Evidence report C: access to care). The committee considered it important that training includes education on the impact of ME/CFS on people living with the condition as well as their families and carers and tailoring access according to the person's needs (see evidence report C: access to care).

Giving information to people with ME/CFS

In the barriers and facilitators review evidence suggested that as a result of reduced physical and cognitive functioning people with ME/CFS can have difficulties in absorbing information from HCPs and can sometimes experience an overload of information which can negatively affect their understanding of the condition.

The committee were familiar with the concept of information overload from their experience and the need for adaptions when delivering information to people with ME/CFS. Time constraints in primary care were also identified as a barrier to diagnosis and care, with evidence suggesting that due to the complexity of ME/CFS and its symptoms, HCPs often find that the short length of consultations do not allow for effective communication and support.

The committee considered that these findings supported the recommendations about educating HCPs about understanding ME/CFS and also the principles of care outlined in the recommendations. In particular checking the person's understanding of each consultation, offering a summary as appropriate to the person's needs, adapting the timing, length and

frequency, of appointments and treatments and ensuring information is available in a variety of formats (see evidence report C: access to care).

Support from specialist services

Support from specialist services was identified from two perspectives in both reviews: access to services for people with ME/CFS and access to advice. The committee noted the need for access to specialist support was identified by both HCPs and people with ME/CFS and this theme was echoed throughout several of the guideline reviews.

Access to services

In the information and support review evidence suggested that there is need for a clearer clinical management pathway for ME/CFS. HCPs are often unsure of where to refer patients once a diagnosis has been reached. ME/CFS specialists also expressed concern at the lack of referrals to their services made by GPs.

Uncertainty around appropriateness and effectiveness of treatment pathways was also shown by the evidence on HCPs caring for children and young people. The committee noted that the concern expressed by HCPs regarding the lack of a clear management pathway was similar to the concern expressed by people with ME/CFS. In the information, education and support review evidence suggested that GPs found it helpful to have support from ME/CFS specialists, but that there was limited availability of specialist services, or they were unaware of them. Specialist HCPs on the other hand expressed frustration that GPs in their region often did not refer patients to their services and emphasised that there was a need for these services to be more 'visible' and provide training and education for other HCPs. People with ME/CFS also showed concern about long waiting times for specialist services and suggested that increased communication between primary and secondary care might allow better management by GPs.

Based on the qualitative evidence on the availability of specialist services and lack of HCP knowledge of them, the committee noted that information and training for GPs should include advice on when to refer a patient to specialist ME/CFS services and how to access them. The committee noted there is variation and inequity in access to specialist ME/CFS services in England and Wales with paediatric services being particularly limited.

Access to advice

In the information and support review GPs suggested that ME/CFS specialist services should support primary care by providing them with information and training. Medical students reported that there is little or no formal training on ME/CFS in the medical curriculum and that their knowledge often comes from media.

The committee discussed who should provide training for health and social care professionals and the appropriate level of specialist involvement. It was suggested that primary and secondary care services should work together to develop a training program and that training for generalists/GPs should be provided or at least supported by specialist services. The committee also reflected that there was a need for standardised specialist training to ensure that there is consistency across services. Based on the expert testimony by Dr Muirhead and the experience of the committee about outdated training materials, it was considered the content and training methods should be evidence based and that trainers should have proven skills, knowledge and experience in the particular area of training.

Delivery of training

In the barriers and facilitator review evidence suggested that some GPs are reluctant to take on the management of people with ME/CFS, preferring to refer to secondary care specialists,

and do not always engage with ME/CFS training when offered. The committee discussed why this might be the case and considered that if GPs where underconfident about their knowledge about ME/CFS and their skills to manage people's symptoms then it would be understandable they would refer to secondary care specialists ensuring the best care for their patients. The committee reflected that the training recommendations should help address the lack of knowledge and apprehension that GPs have about managing ME/CFS. The reluctance to engage training was seen as multifactorial but as mentioned above GPs have limited time and the committee agreed that making training resources accessible to GPs was critical in ensuring their uptake. The committee agreed that this finding could be applied to all health and social care professionals and that knowledge and training about ME/CFS was limited.

As is the case with any training programmes the committee agreed it was important to ensure they are of high quality and agreed that training programmes should have evidence based content, developed and supported by specialist services with input from people with ME/CFS and run by trainers who are experienced and knowledgeable about ME/CFS. It was suggested that ideally assessment should be part of the training. The committee recognised it is the responsibility of professional bodies to develop training requirements for their members and the inclusion of ME/CFS in undergraduate and postgraduate curricula.

In support of this, in the barriers and facilitators review HCPs valued online training resources that showed how to work with people with ME/CFS in a consultation setting, with several GPs finding video resources particularly useful for this. The committee noted the preference identified in the evidence for online training and noted that training can be delivered through online courses such as E-learning and online videos.

3.3. Cost effectiveness and resource use

Cost effectiveness evidence was not sought as both reviews questions related to qualitative evidence.

The committee considered that the review's findings demonstrated the need for better training and education for health care professionals about ME/CFS. This was also supported by qualitative evidence from other guideline chapters including those covering barriers to diagnosis and barriers to care. The committee recommended that new training programmes be developed that are based on the latest evidence and patient experience. The cost effectiveness of this training is uncertain but insufficient training of health care professionals has clearly led to delayed diagnosis and poor quality of life for many patients. Any initial costs for training should save downstream costs that result from delayed diagnosis and inappropriate management of symptoms.

Another theme that was the need for specialist services as a source of information and support for health care professionals but also the need for a clear clinical management pathway. This was echoed in the reviews of barriers to diagnosis and care and in the review of patient information needs. The committee recommended that a specialist multidisciplinary team to confirm diagnosis, establish a treatment plan and provide support for primary care services. The cost effectiveness of a specialist multidisciplinary team is uncertain, but the uneven provision of specialist services has been identified by patients and staff as a key contributor to delayed diagnosis and poor patient outcomes.

3.4. Other factors the committee took into account

Safeguarding

The committee discussed how a lack of knowledge and understanding about ME/CFS has led to people not being believed and this has had negative consequences particularly for

children and young people, and their families. The committee agreed it was very important to make recommendations raising awareness on this topic.

The committee recognised that safeguarding is an issue that has been part of the ME/CFS landscape in a way that is different to other chronic illnesses and disabilities because people with ME/CFS commonly report that they are not believed. Some of the committee members were aware of families of children with ME/CFS not being believed for years and the persistent feeling of a low level of threat of accusation of making their child's illness up. This has led to parents having to explain repeatedly to health and social care professionals why their children cannot engage in activities or have reduced attendance at school. This has resulted in mistrust and acrimony between people with ME/CFS, their families and health and social care services. This mistrust and breakdown in relationships between people with ME/CFS, their families and health and social care professionals causes long-term damage with a reluctance for people with ME/CFS to engage with health and social care services. The committee made recommendations to raise awareness of the impact prejudice and not being believed can have on an individual and their families and their subsequent fear of engagement in health and social care involvement.

The committee members noted that while the issue of safeguarding is not solely about children and young people most of the concerns the committee were aware of related to children and young people with ME/CFS.

There has been considerable controversy over the use of child protection procedures and care proceedings in children and young people with suspected or diagnosed ME/CFS. The committee acknowledged the devastating emotional and physical impact this stressful experience can have on children, young people and their families. The committee noted that once child protection concerns are raised the process is very difficult to reverse or stop even when it has been concluded that there is no harm.

The committee agreed it was important that recommendations address some of the common misconceptions that may have led to child protection concerns, and to provide guidance on this topic.

One of the key themes identified throughout the evidence reviews (see evidence review A: information and support for people with ME/CFS) and the additional evidence (see Appendix 1, 2 and 3) provided in this guideline is the lack of understanding, education and knowledge that health and social care practitioners have about ME/CFS. This is turn leads to a reluctance to diagnose ME/CFS and delays in diagnosis (see evidence review D: diagnosis).

The committee noted these factors have also been highlighted by the ME/CFS community as contributing to safeguarding concerns being raised. Committee members knew of examples where a lack of understanding about ME/CFS, its fluctuating nature, its range of possible symptoms and severity had led to inappropriate safeguarding concerns being raised in families.

In addition to the recommendations in the guideline on diagnosis and education for health and social care practitioners, the committee made a consensus recommendation to dispel some of the common misconceptions that have been held about when to suspect a potential safeguarding issue. The important ones the committee identified are discussed below.

Physical symptoms that may not fit into a commonly recognised illness pattern

The committee agreed that the wide range of symptoms of ME/CFS and their interaction is complex and cannot be explained by current knowledge of organic pathology. The presentation of ME/CFS is diverse, often with multiple symptoms which could – in the absence of other typical features of ME/CFS - give grounds for concern. Where core features of ME/CFS are present other symptoms are a common occurrence as part of the illness. The committee noted that many of the symptoms of ME/CFS are general and are evident in a wide range of conditions. For example, the profound fatigue after exertion is a feature of

ME/CFS. However, it is easily confused with fatigue in mental health conditions such as depression or in other diseases such as MS, cardiac failure and muscle disorders. This complexity and lack of understanding about the core features of ME/CFS, and the variety of symptoms and severity that can accompany them can result in health care professionals raising concerns.

More than one child or family member with ME/CFS

The committee noted that the aetiology of ME/CFS is not clear and the role of genetics, environmental factors and infections remain unknown. An assumption that the possibility of more than one person in a household is unlikely is not substantiated.

Exercising choice over treatment by the child, young person or parent/carer

The children and young people report found that young people and their parents often knew more about ME/CFS than the health and social care professional they met, and this could result in them disagreeing with the professionals about the advice and treatments they were offered. The committee were aware of safeguarding concerns being raised when young people with ME/CFS or their parents/carers had refused part of a management plan (because parents/carers considered the management plan not to be in the child's best interest). The right for the person to make choices about their treatment and not to be penalised is emphasised throughout the guideline (see principles of care) and the committee agreed that it should be clear that disagreeing with any part of a management plan is not in itself an indicator of abuse.

Parents/carers communicating for the child or young person and reduced or non-attendance at school/college

The committee were aware that parents/carers communicating for the child or young person and reduced or non-attendance at school have been red flags for child protection concerns.

Communication

The committee discussed how a lack of understanding and knowledge about how ME/CFS symptoms (for example, fatigue, post exertional symptom exacerbation (PESE), cognitive difficulties) can affect the ability to communicate and can result in these misconceptions. The committee gave examples of their own experience as parents: they want to protect their children from worsening their symptoms, which they know is more likely to happen in a situation where the child is already anxious or stressed and they can see their child is struggling to communicate. This is particularly challenging in cases of people with severe or very severe ME/CFS, where communication is difficult, and parents or carers inevitably communicate with health care professionals on behalf of the person with ME/CFS.

In addition, the committee noted that in children or young people that have had previous negative experiences with health and social care workers it is not surprising that they want their parents or carers to advocate and communicate for them. This can be interpreted as reinforcing misconceptions that the parent does not let their child talk. The committee discussed that this could result in parents finding it difficult to advocate for their child without being seen as overprotective or pushy. This results in the withdrawal of seeking support and can have a long term impact on both care and education.

School attendance

The committee noted that the key to understanding school attendance in a child with ME/CFS is proportionality. Any interpretation of the appropriateness of help with communication and to school attendance must be within the context of the rest of the child or young person's life. The committee have experience where there is the perception that if a child is in school then "everything is okay". However, being physically there and appearing well does not mean the child or young person does not struggle to maintain any other activity

outside of school. Children may struggle to maintain attendance at school but then at home are so exhausted that they are unable to maintain any activity of simple play or interaction. Their post exertional fatigue is exhibited at home.

The committee discussed that the parent or carer experience could be very different from that of the school. The nature of PESE means people are rarely seen at their worst which can be confusing for people that have little understanding about ME/CFS and may result in miscommunication and a perception that the parent or child is exaggerating. This may result in parents making the decision to reduce or adjust school attendance in the best interests of their child which can raise concerns. Reduced or non-attendance is often perceived to be school avoidance (but is usually a result of being unable to attend, either because they are too unwell or the school have not been supportive or proactive in finding the best ways to manage education).

Safeguarding assessments and assessments under the Mental Health Act or Mental Capacity Act

The committee agreed to underpin these recommendations on safeguarding by recommending that health and social care practitioners that have training and experience in ME/CFS should be directly involved in safeguarding assessments on people diagnosed with or suspected of having ME/CFS. It is recognised that there are emergency situations where an urgent assessment is needed to prevent harm and in these circumstances health and social care practitioners that have training and experience in ME/CFS should be involved as soon as possible.

The committee hoped that recommending that health and social care practitioners that have training and experience in ME/CFS should be directly involved in safeguarding assessments would help reduce some of inappropriate safeguarding concerns that are raised. The involvement of a person who has received training in ME/CFS in the assessment of safeguarding concerns is important because safeguarding issues are complex and the features of ME/CFS can easily raise false safeguarding concerns. The committee noted that fabricated illness is very rare and a diagnosis that should only be arrived at with great caution.

The committee recognised it was important that children and young people at risk of maltreatment and abuse should be identified and receive the help and support they need. The committee made a consensus recommendation to raise awareness that recognising and responding to possible child maltreatment, abuse and neglect is complex and should be considered in the same way for children and young people with ME/CFS as with any child with a chronic illness or disability. The NICE guidelines on Child maltreatment: when to suspect maltreatment in under 18s and Child abuse and neglect are referenced.

The committee were aware of people with ME/CFS that had been subject to Mental Health Act or Mental Capacity Act assessments and noted that the appropriateness of the assessments and outcomes has been challenged. The committee discussed that this can be a result of a lack of understanding about ME/CFS and agreed to make a consensus recommendation similar to the one on safeguarding assessments. Assessments should involve health and social care professionals who have training and experience in ME/CFS and in an emergency situation health and social care professionals who have training and experience in ME/CFS should be contacted in the next 24 hours.

People with severe or very severe ME/CFS

The committee agreed it was important to recognise that health and social care professionals with little understanding of ME/CFS symptoms and their severity can misinterpret the needs of people with severe or very severe ME/CFS as a matter for concern, for example, the need for a low stimulus room as enforced social isolation, lack of self-care as neglect, inability to digest food as an eating disorder. On this basis the committee made a consensus

recommendation that people with severe or very severe ME/CFS are at risk of their symptoms being confused with signs of abuse or neglect.

Pregnancy, childbirth and post-natal care

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.

Appendices

Appendix A Review protocols

Review protocol for Information, education and support for health and social care professionals

ID	Field	Content
	Scope	6 Information, education and support for health and social care professionals
	Draft review question	6.1 What information, education and support do health and social care professionals who provide care for people with ME/CFS need?
0.	PROSPERO registration number	CRD42019152080
1.	Review title	What information, education and support do health and social care professionals who provide care for people with/suspected of having ME/CFS need?
2.	Review question	What information, education and support do health and social care professionals who provide care for people with/suspected of having ME/CFS need?
3.	Objective	To identify the information, education and support required, as identified by health and social care professionals caring for people with/suspected of having ME/CFS, people with ME/CFS and the families and carers of people with ME/CFS.
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	Health and social care professionals caring for someone with/suspected of having ME/CFS. Perspectives of people with ME/CFS and the families and carers of people with ME/CFS about the information, education and support needs of health and social care professionals who provide care.

7.	Intervention/Exposure/Test	Information, education and support that health and social care professionals who provide care for people with/suspected of having ME/CFS 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

		• Severe vs. less severe (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: https://www.nice.org.uk/guidance/indevelopment/gid-ng10091

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

Review protocol for Barriers and facilitators to providing information, education and support

ID	Field	Content
0.	PROSPERO registration number	CRD42019152089
1.	Review title	What are the barriers and facilitators to providing information, education and support to people with ME/CFS for health and social care professionals?
2.	Review question	What are the barriers and facilitators to providing information, education and support to people with ME/CFS for health and social care professionals?
3.	Objective	To identify the barriers and facilitators to the providing of information to people with ME/CFS
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 final submission of the review and further studies retrieved for inclusion if relevant. The full search strategies for MEDLINE database will be published in the final review.
5.	Condition or domain being studied	ME / CFS
6.	Population	Health and social care professionals caring for people with ME/CFS, people with ME/CFS and their families and carers.
7.	Intervention/Exposure/Test	Perceptions, experiences and views of health and social care professionals, people with ME/CFS and their families and carers of the assisting factors and hurdles during the process of providing information, education and support

8.	Comparator/Reference standard/Confounding factors	N/A
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 (ie 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 • Severe ME/CFS vs. less severe (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: https://www.nice.org.uk/guidance/indevelopment/gid-ng10091
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 questions:

- What information, education and support do health and social care professionals who provide care for people with ME/CFS need?
- What are the barriers and facilitators to providing information, education and support for health and social care professionals?

The literature searches for this review are detailed below and complied with the methodology outlined in Developing NICE guidelines: the manual.[91]

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 9: 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 5: Flow chart of qualitative study selection for the review of information, education and support for health and social health professionals

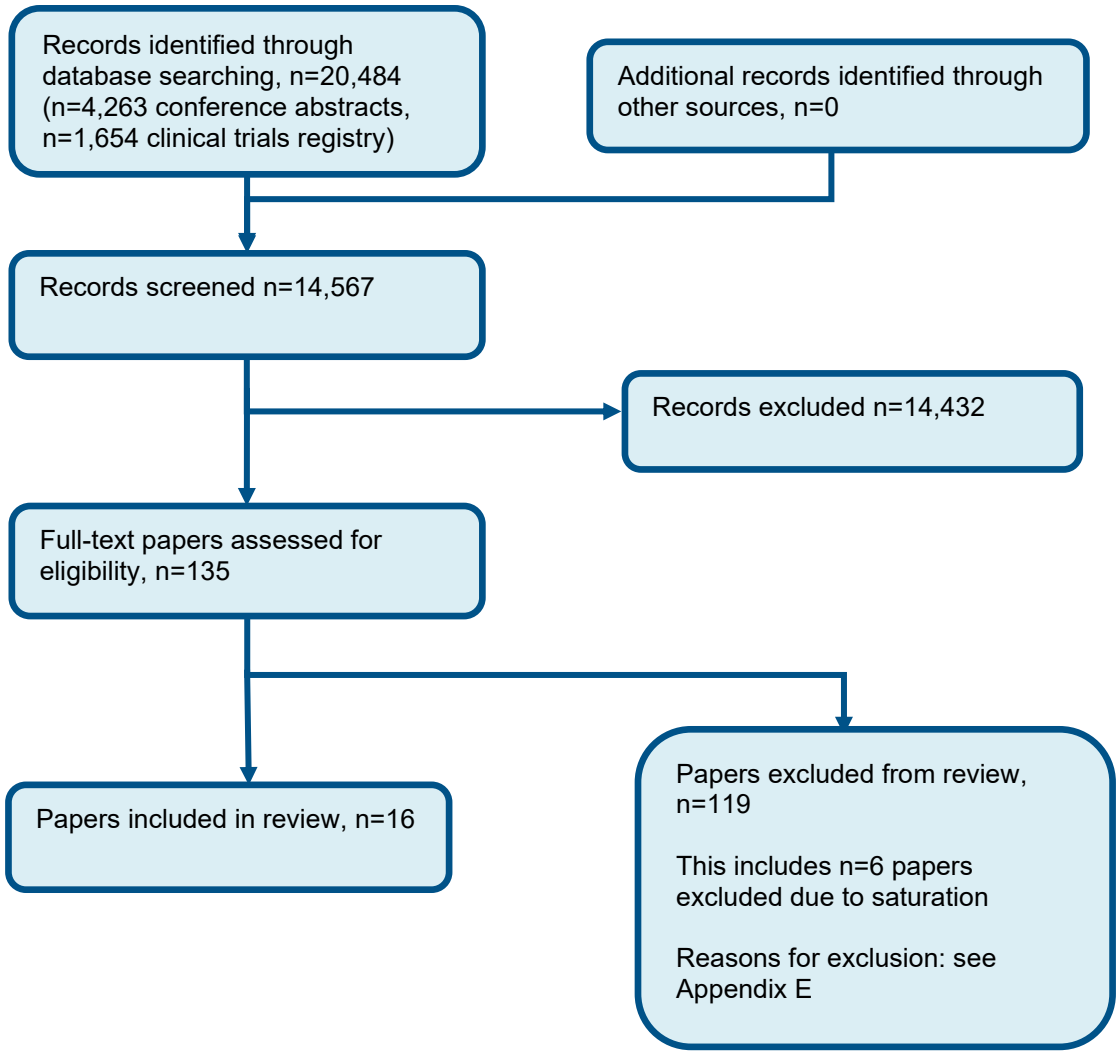
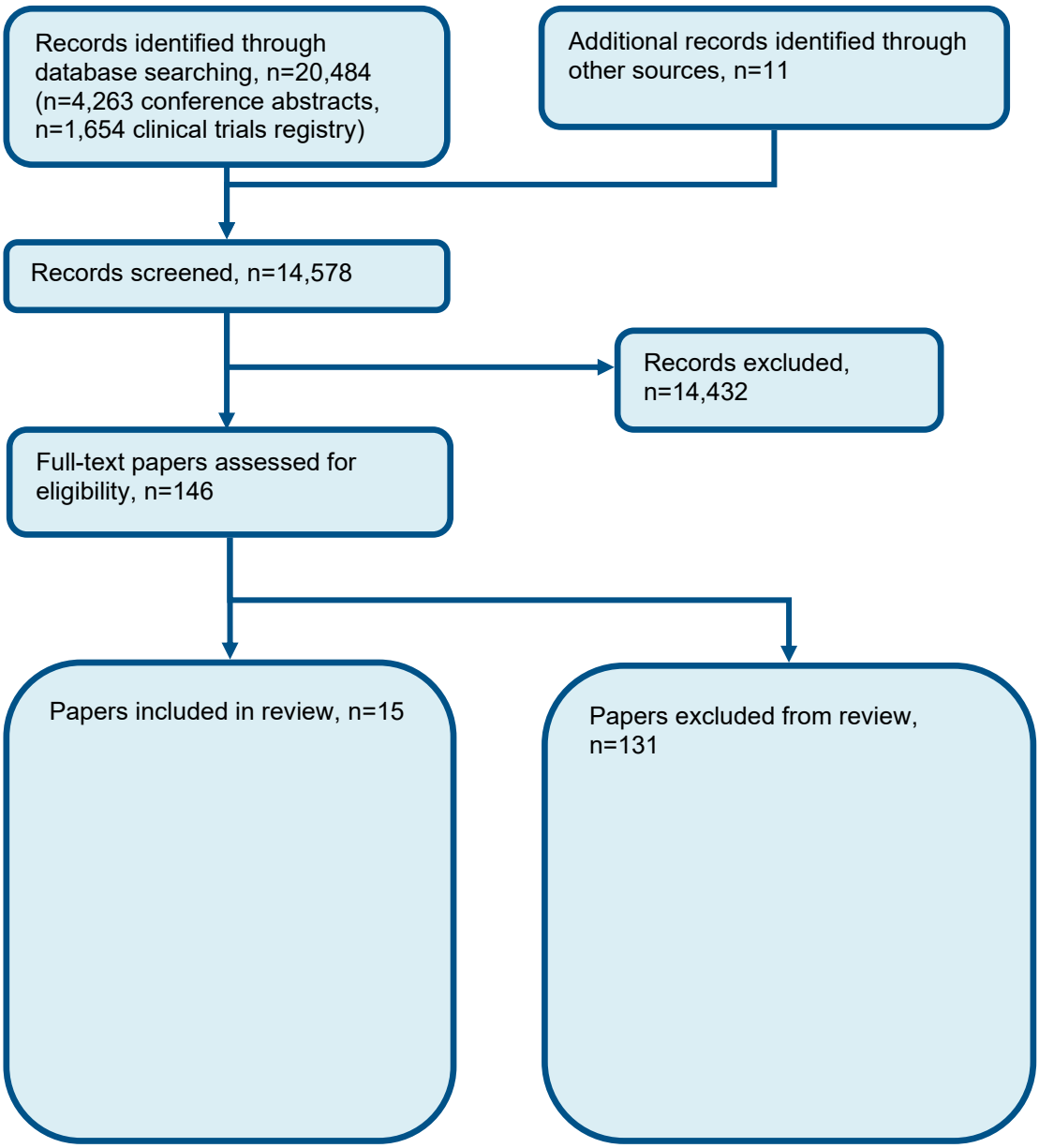


Figure 6: Flow chart of qualitative study selection for the review of barriers and facilitators to providing information, education and support



Appendix D Qualitative evidence

Information, education and support for health and social health professionals

Study	Beaulieu 2000[19]
Aim	To examine multiple perspectives on stigmatization and legitimation of 'CFS'.
Population	Health professionals including general practitioners, mental health professionals (one of whom was not a physician), infectious disease specialists, immunologists and rheumatologists, recruited following identification by people with 'CFS' participating in the study. N=15; male/female 10/5; had been in practice from six to seventeen years and individually had seen from six to almost one hundred cases. People who were English-speaking and who had a diagnosis of 'CFS' from a medical doctor, recruited from physicians' practices, support groups and identified by leaders of associations. N=43; male/female 16/27; 26% were in school or working full or part time; mean age at onset was 34.2 years (range 15 to 58 years); people had been ill for an average of seven years. Significant others including friends, parents, spouses, adult children and a sibling, recruited following identification by people with 'CFS' participating in the study. N=23; male/female not reported; 69% were working.
Setting	McGill University, Montreal
Study design	Qualitative interview study
Methods and analysis	Mixture of structured and semi structured questions related to approach to diagnosing, explaining and treating 'CFS', views on support groups and alternative therapies, whether thinking had changed over time, impressions of typical and atypical patients and challenges in dealing with people with ME/CFS(doctors); symptom experiences, the impact on roles and functioning, beliefs about cause, attempts to manage the illness through help seeking and treatment and reactions from health professionals (people with 'CFS'); knowledge about people with ME/CFS' experiences, ideas about cause and treatments, how having someone close with 'CFS' affected their lives (significant others).

Study	Beaulieu 2000[19]
	78% of those who agreed to face to face interviews also consented to taping and tapes were transcribed. For telephone interviews and interviews in which people refused to be taped, notes of key words and phrases were taken. These notes were elaborated as soon as possible after the interviews. Interviews took place in people's homes, their offices, the researcher's office, or in neutral public places such as coffee shops or parks. A few doctors were interviewed by telephone. Interviews were analysed using thematic analysis. Transcripts of each interview were summarized according to the broadest content areas of questions. Summaries were then pooled according to categories and read and reread for recurring themes and variations in the first gross categories.
Findings	**Awareness of the reality of ME/CFS**
	Some people with ME/CFS went through a relatively quiescent period with regards to help seeking until they, or their doctors, heard of 'CFS' and reconsidered the case in light of this information. In several instances, people were diagnosed only after they had suggested 'CFS' to doctors. Other people who continued to request medical certificates were met with grudging agreement or bluntly told that they were shirkers. Many people were also shocked to hear other physicians disparaging the diagnosing doctor. Some retrospectively explained doctors' sceptical behaviours as a function of the Medical ignorance of 'CFS' that was widespread until the late 1980s. No clinician suggested that CFS patients were malingering, but some specifically underlined why they thought patients were not, e.g. sudden onset of illness and protracted recovery of otherwise well-functioning young people, evidence of actively seeking to resolve the problem, reluctance to enter the sick role, no indication of secondary gains, colleagues who had considerable clinical experience with the illness and expert opinions in the literature.
	Consensus on diagnostic label
	Doctors who gave a diagnosis used the labels CFS, myalgic encephalomyelitis (ME), post vital fatigue, or descriptive diagnoses such as "fatigue with possible depressive etiology", or if pain was a prominent complaint "regional pain syndrome". Regardless of the chosen label, their language was cautious. Some avoided a label and discussions of etiology, but in so doing they may have left an ambiguous impression. Some clinicians were doubtful whether CFS was a new and separate illness, but believed it to be a new name for post-viral-fatigue, neurasthenia or asthenia. Some friends and relatives in health or related fields wondered if 'CFS' was merely a 'catch all 'label that doctors used to bide their ignorance of what was wrong and to placate anxious patients. These friends and relatives suggested that the illness is so vaguely defined that "you can hook two or three different medical diagnoses and put Chronic Fatigue Syndrome on it". From their insider position as health professionals, they were a little cynical that doctors were giving patients a label just to shut [them] up".
	Clear guidelines for diagnosis
	Diagnostic difficulties stemmed from the lack of clear guidelines, the fact that 'CFS' is a diagnosis of exclusion, and people with ME/CFS' resistance to psychiatric evaluations. Some doctors thought the CDC case definition might be the answer to their diagnostic

Study	Beaulieu 2000[19]
	difficulties. A few tried to apply the CDC criteria, but became disillusioned about its usefulness when they saw patients who they were convinced had the illness but who did not meet all the criteria.
	Cause of ME/CFS
	All clinicians acknowledged that the cause of 'CFS' is unknown. Clinicians variously considered CFS a physical, psychosomatic, or nonspecific disorder, or an illness combining both physical and psychological factors, based on experiences with people with ME/CFS and a selective weighting of the medical literature. They underlined the fact that neither their own views nor those in the literature have firm empirical support. The hypotheses were unrelated to clinicians' specialities and usually reached after struggling with different causal perspectives. In some cases, they remain highly unstable; even clinicians who believed their etiological hypotheses were stable could waver when the illness' refractoriness to treatment challenged their identities as healers.
	Explanation of ME/CFS
	The uncertainties associated with the illness made clinicians insecure about the accuracy of their information, hence the cautious and uneasy tone of their explanations. If they did not contextualize and qualify their explanations and were proven wrong in the future, their credibility was at stake.
	Test results
	It was the experience of people with ME/CFS that if abnormalities were found, doctors could not specify the links between these findings and their symptoms. While patients could accept specialists' reports of normal test results, they were not reassured by well-meaning doctors who suggested there was no cause for concern.
	Information about support groups
	Most clinicians would hesitate to recommend support groups because they had little knowledge of these groups. Others hedged because they believed support groups could be harmful or helpful, depending on their approaches and depending on individual patients. For these clinicians, the problem was that they had no way of determining how a given patient would be affected. One recounted patients being devastated after being exposed to the worst scenarios in support groups and believing such fates were inevitable. Others surmised that support groups could contribute to chronic disability because they "medicalize patients' distress", "reinforce illness behaviours", "institutionalize illness" and "possibly encourage dependency". One wondered whether patients didn't "pick up symptoms" from such groups. Another gave a cautious nod to groups who were "more or less involved with the mainstream" of medical thinking.
	Understanding of the lived experience of 'CFS'
	Some people with ME/CFS found that doctors who were thought to be well informed about 'CFS' abandoned patients after making the diagnosis, because they said nothing could be done. They may have been knowledgeable, but they did not understand people with ME/CFS' experiences. They did not understand how frightening it was to go through the experience of having a poorly understood illness without medical support.

Study	Beaulieu 2000[19]
Limitations and applicability of evidence	Moderate methodological limitations due to concerns over participant selection (with HCP participants directly selected by ME/CFS patients) and data analysis (coding and analysis by a single researcher). Very minor concerns over applicability as main findings emerging are driven by the study's original aims to explore multiple perspectives on stigmatization and legitimation of 'CFS'.

Study	Broughton 2017[24]
Aim	To explore patients' experiences for ME/CFS by capturing the perspective of patients who have been treated by NHS specialist ME/CFS services in England.
Population	Adults who were completing treatment for ME/CFS. 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.
Setting	Three outpatient NHS specialist ME/CFS services.
Study design	Semi-structured interviews and thematic analysis.
Methods and analysis	Six participants were interviewed face-to-face in their own homes whilst ten participants were interviewed via telephone. Interview length ranged from 23 to 57 minutes, with a mean length of 32 minutes. The study adhered to a 'participatory' qualitative research paradigm, using an inductive approach which was dirven by the data, and which did not hypothesis about potential findings. The semi-structured interview protocol was developed through consultation with a Patient Reference Group affiliated with Action for ME. All interviews began with the open question: "Tell me about your ME/CFS" and participants were encouraged to guide discussion and introduce their own topics of interest. Techniques of constant comparison informed the analysis and the identification of themes. Transcripts were coded thematically before an iterative process was used to agree a final structure of themes and subthemes.
Findings	**Patient-reported theme: Lack of knowledge of GPs**
	All participants were referred to ME/CFS specialist services by their GPs. Participants reported varied experiences before referral. Participants with negative experiences of this process described a number of barriers to accessing specialist services, including lack of information, having to take a proactive role in asking for diagnostic tests, and GPs' lack of "awareness", "knowledge" or "belief" in ME/CFS.

Study	Broughton 2017[24]
Limitations and applicability of evidence	No concerns over methodological limitations. Very minor concerns over applicability as the population is only ME/CFS patients and does not take into account the views of HCPs. However, the patients were recruited from a representative range of NHS specialist services making their opinions directly applicable to NHS-based HCPs.

Study	Chew-Graham 2008[31]
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	Family physicians who participated in a randomised controlled trial (FINE): 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 who participated in a randomised controlled trial (FINE): 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	**Information & consensus on measures of the illness**
	Family physicians expressed frustrations that they could not measure how the patient was affected by their condition. It was so-called 'invisible' and the symptoms seemed out of proportion to the signs leading some to doubt the condition and the genuineness of its presentation. The inability to demonstrate the extent of their condition beyond the snapshot view revealed in the consultation meant that patients were unable to establish that symptoms come and go and that the condition is invisible on good days. Family physicians described how they ran a battery of tests, which invariably returned negative results. With no manifest sign of patients' symptoms and

Study	Chew-Graham 2008[31]
	no confirmation of a diagnosis, the physicians would often reach clinical impasse. Patients were aware their condition was invisible from a biomedical perspective.
	Understanding of the condition & medical training (diagnosis & management)
	Family physicians admitted having limited clinical understanding about 'CFS/ME' available to them, causing them to question the existence of the condition. Patients were aware that the medical community disagreed over the existence of the condition and also that family physicians had limited clinical knowledge about 'CFS/ME'. They believed they were unprepared by their medical training and continuing education to diagnose and manage 'CFS/ME' and they acquired evidence from sources outside the clinical domain. Their training enabled them to exclude a physical cause for the patients' symptoms but doubt and limited knowledge about 'CFS/ME' made the diagnosis uncertain.
	Sources of information:
	a) **Social knowledge/Need for exposure to people with ME/CFS:** A source of evidence open to family physicians was their observation of patients outside the clinical setting of the consultation. Patients' activities and behaviours, if witnessed could potentially support the notion of the existence of the condition. For some family physicians seeing patients with the condition led them to conclude it existed and they recognised that by working with patients with 'CFS/ME', they came to learn about the condition. Patients were aware that achieving understanding through a significant other constituted powerful and convincing evidence of the existence of 'CFS/ME'. Some family physicians reported that they developed an understanding of the condition only after they had known someone socially (other than a patient) who had it, and as with media personalities, the status or credibility of the significant other determined how persuasive the evidence was. For family physicians an extremely convincing source of evidence was personal experience of the condition, with one physician reporting her knowledge was initially influenced by her sister who had 'CFS/ME', but far more powerful evidence came from being diagnosed herself with 'CFS/ME'.
	b) **Media:** Representations of 'CFS/ME' expressed within the media provided a useful source of evidence for both family physicians and patients to build a model for the condition. Media personalities lent credibility to the condition and their positive attributes relieved patients from being culpable.
Limitations and applicability of evidence	No concerns over methodological limitations Minor concerns over applicability due to the research aim and sample which consisted of people recruited in a RCT (FINE trial).

Study	Chew-Graham 2010[30]
Aim	To explore GPs' beliefs about the value of the label of 'CFS/ME', implications of the diagnosis and attitudes towards patients with ME/CFS.
Population	GPs (n=22) recruited via purposive sampling through practices participating in the FINE trial.

Study	Chew-Graham 2010[30]
	46 GPs were invited by letter but 22 agreed to be interviewed.
Setting	GP practices in North-West England
Study design	Qualitative interview study
Methods and analysis	Semi-structured interviews lasting between 10 to 72 minutes (median duration 34 minutes) were conducted using an interview guide. This provided a flexible framework for questioning and explored a number of areas: ideas about the cause of 'CFS/ME', previous experience of patients with 'CFS/ME', how the diagnosis of 'CFS/ME' was achieved, and their role in management of those patients. The interviewer combined open questions to elicit free responses with focused questions for probing and prompting. Interviews were digitally recorded and transcribed verbatim. Analysis proceeded in parallel with the interviews and was inductive, taking an interpretative stance. Coding was iterative and was informed by the accumulating data and continuing thematic analysis. Thematic categories were identified at initial interviews which were then tested or explored at subsequent interviews were disconfirmatory evidence was sought. Interpretation and coding of data was undertaken by three researchers individually and themes were agreed through discussion with the whole team.
Findings	**Support defining & understanding 'CFS/ME'**
	GPs described a struggle, trying to make sense of a difficult set of symptoms and attributed different causes to the illness. There was also some debate over whether 'CFS/ME' actually existed as a medical condition. Such beliefs about 'CFS/ME' necessarily will lead to difficulties in labelling the symptoms or making a diagnosis.
	Need for clear management pathway
	The majority of GPs felt that the label of 'CFS/ME' could be harmful because it did not offer a clear management pathway for either the GP or the patient. This can cause GPs to be reluctant to make the diagnosis of 'CFS/ME'. A number of GPs reported frustration with supporting patients once a diagnosis was made implying that 'CFS/ME' was difficult to manage as no 'cure' was possible.
	Support from secondary care: diagnosis & management
	Those GPs who felt that making the diagnosis or labelling the patient's condition was helpful suggested that referring the patient to secondary care could potentially assist in achieving a diagnosis and providing support to GPs who lack confidence in making the diagnosis alone. GPs however reported experiences of limited availability of potentially helpful places to support them in either making the diagnosis or managing the patient.
	Support post-diagnosis (& recognition)
	GPs alluded to the difficulties they had experienced working with patients with CFS/ME once the diagnosis was agreed. The role of supporting the patient was stressed by respondents with a number of GPs reporting frustrations with this work implying that 'CFS/ME' was difficult to manage as no 'cure' was possible and that the work invested in working with such patients is largely unrecognised.

Study	Chew-Graham 2010[30]
Limitations and applicability of evidence	No concerns over methodological limitations. Minor concerns over applicability due to the sample which consisted of people recruited in a RCT.

Study	Devendorf 2017[41]
Aim	To explore views of physicians with expertise in ME and CFS to define and measure recovery from ME and CFS
Population	Physicians who were experts in the ME and CFS field were recruited using a non-probabilistic, purposive sample approach. Experts were determined by their ME and CFS patient experience, research contributions, and overall involvement in the field (e.g. running ME and CFS specialty clinics, participating on committees. N=10; male/female 8/2; mean age (SD): 65 (12) years; with more than 16 years of experience in ME and CFS; the primary patient population of half of the participants were adults; the primary patient population of half of the participants were children and adolescents. Physicians f varied in specialty: paediatrics (n=3); neurology and infectious diseases , family medicine , general internal medicine, internal medicine and geriatric medicine, immunologist, paediatrics and infectious diseases, internal medicine and epidemiology
Setting	DePaul University, Chicago
Study design	Qualitative interview study
Methods and analysis	Semi-structured interviews were conducted over the phone (except for one that was conducted via email), using questions adapted from previous qualitative investigation of recovery with patients that tackled physicians thoughts on: the likelihood of recovery, defining recovery, measuring recovery, treatment approaches, and predictors of prognosis. Interviews were audio-recorded and transcribed verbatim. Interviews were analysed using deductive thematic analysis. Interviews were coded by two researchers and this was followed by initial theme and subtheme development
Findings	**Consensus on case definitions**
	Many physicians considered recovery as no-longer meeting diagnostic criteria-though case definitions were not always specified.
	Consensus on definition of recovery
	Physicians acknowledged the subjectivity of recovery, which they felt breaks down to semantics and is often mislabelled. They expressed that both physicians and patients mistake significant improvement as recovery or people use the phrases ‘partial’ and ‘full recovery’.

Study	Devendorf 2017[41]
	Measures of recovery
	Opinions differed on a number of measures of recovery. For instance while some advocated using exercise tests as objective measures, others critiqued the sensitivity of such tests; however all emphasised that measuring recovery from ME and CFS should be multi-faceted. Nearly all physicians emphasised general functioning as a recovery measure with many assessing functioning in terms of hours of daily functioning; some compared their ME and CFS patients to their healthy patients; one mentioned using the functional disability inventory; most measured functional improvement using patient premorbid levels of functioning. It was every physician's goal to get their patients back in work or school, in terms of measuring recovery however, some felt work and school status were effective indicators of functioning, while others noted the limitations of such measures; on the one hand these were viewed as positive therapeutic outcomes and on the other hand they were viewed as 'all or nothing' measures by several.
	Definition and importance of improvement
	Depending on the illness severity, some physicians considered working part-time as significant improvement, while others considered working full-time-but without the ability to perform other activities-as significant improvement. Further physicians felt that aiming for significant improvement was a worthy and obtainable goal, while several conveyed that their patients felt hopeless, powerless and frustrated with their illness and that reaching a significant level of functioning can empower patients, alleviate uncertainty, and restore a level of normalcy. A few physicians who noted the unlikelihood of full recovery, would be happy if their patients reached a near-recovery level of functioning, contingent on some medications or coping
	Most problematic symptoms
	Physicians were aware of the multiplicity and variation of symptoms that patients can experience. Most physicians conceptualised recovery as complete symptom remission and inferred symptoms like PEM, fatigue, sleep issues, pain and neurological issues (e.g. brain fog) to be the most problematic. Some physicians noted the importance of monitoring symptoms over time and to consider contexts like work, school and physical activity.
	Consensus on assessment of symptomatology: physical functioning measures
	Many physicians commented on the general use of laboratory exercise measures to assess fatigue, PEM, and other symptomatology. Some supported using exercise tests in research and practice, while others mentioned their limitations. One participant specified and advocated for the 2 day exercise tests (also known as CPET), noting that test's merits of being objective and able to measure ME and CFS severity; one suggested using actigraphy as an objective research measure; another mentioned the ability to climb flights of stairs as an exercise test. Other physicians, while not rendering exercise measures useless, felt they lacked sensitivity, were not the best representations of everyday functioning, and were confounded by motivation. One research-minded physician felt patients tend to improve cognitively, and exercise tests might exacerbate patients' symptoms.
	Need for physiological measures in assessing ME/CFS
	Several physicians hoped for more integration of physiological measures in assessing ME and CFS- this can be difficult however, since there is no universal, identified biomarker and physicians provided a full medical history to rule out other manifestations. Some

Study	Devendorf 2017[41]
	examined orthostatic intolerance using the NASA 10-minute Lean Test, one research-minded physician suggested using CPET to measure blood pressure, heart rate and aerobic capacity and taking blood samples to measure natural killer cells. Some felt future research should identify gene-expression markers in patients.
Limitations and applicability of evidence	Minor methodological limitations due to data analysis with themes mostly supported by single quotes. Minor concerns over applicability as main findings emerging are driven by the study's original aims to explore physicians' views on recovery.

Study	Devendorf 2019[40]
Aim	To explore physicians' views on the challenges to studying and approaching recovery of ME/CFS patients.
Population	Physicians who treat myalgic encephalomyelitis and chronic fatigue syndrome. N=13; 4 female, 9 male. Mean age: 60 years old. Experience in practice: 4 had 30 or more years, 7 had 20-29 years and 2 had 1-9 years medical experience. Medical specialties were as follows: epidemiology (n=1), geriatrics (n=1), infectious disease (n=1), neurology (n=1), internal medicine (n=2), psychiatry (n=2), general medicine (n=3), and paediatrics (n=5) (3 physicians identified with two specialties).
Setting	Phone- or email-based interviews.
Study design	Qualitative approach, conducting semi-structured interviews with physicians and analysing the data using deductive thematic analysis.
Methods and analysis	Semi-structured interviews were conducted over the phone (or via email for one participant) following verbal consent (or written consent via email). Interviews asked physicians about their general thoughts on recovery from ME and CFS, particularly regarding definition, measurement and study of recovery. Questions were inspired by online patient discussion boards discussing the PACE trial, and were generated based on discussions with an expert in the field. Follow-up questions were asked to expound upon participants' responses. Interviews were audio-recorded (at a mean length of 31 minutes), transcribed verbatim and verified for accuracy. Deductive thematic analysis was used in a six-step approach to explore challenges to studying recovery from ME and CFS. Coders searched for meanings and patterns by reading and rereading participants' transcripts while revisiting audio recordings. Inter-rater reliability was found to be good based on 20% of the interviews (n=3).
Findings	**Theme: Consensus on diagnostic criteria, case definitions and ME/CFS aetiology.**
	There is a need for consensus on inclusion criteria for ME/CFS diagnosis. Depending on the case definition used by physicians, patients may be diagnosed differently between providers, therefore delaying appropriate treatment and affecting clinicians' views of patient recovery. There is also variation in the etiological views of clinicians with regard to ME/CFS, with some participants variously believing in physiological causes, temporary causes and psychiatric causes. This affects how physicians treat ME/CFS patients. There

Study	Devendorf 2019[40]
	is also a lack of understanding of how ME/CFS relates to depression, which can skew recovery rates and cause misdiagnosis. Diagnosing patients with depression when they really have ME/CFS may have detrimental effects as this process is inherently stigmatizing, delegitimizing and damaging to patients because they may inadvertently seek inappropriate care. In summary, responses in this area highlighted two needs for practitioners: a need for a consensus on ME/CFS diagnostic criteria and agreed clinical case definitions, and a need for clinicians to better understand the relationship between ME/CFS and depression to improve diagnoses.
	Theme: Patient feedback/communication for better understanding of ME/CFS progression, treatment and recovery.
	Clinicians participating in this study acknowledged that there is often uncertainty as to why patients stop making appointments. Reasons might include patients being unable to afford treatment, lack of time to make or attend an appointment or transferring to another provider. It is often unclear to clinicians whether a patient has stopped making appointments because they have improved. Particularly due to fluctuation of symptoms and their severity, there is a need to track patients via for example phone calls or email. The increased communications suggested here are suggested to benefit both the patient and the clinician, as the clinician gains feedback on their approach, learns more about the course of ME/CFS and finds out which prescribed treatments have been most effective for the individual patient.
Limitations and applicability of evidence	No concerns over applicability or methodological limitations.

Study	Devendorf 2018[42]
Aim	An exploratory study to explore the relationship between ME/CFS and suicidal ideations, including quality of life, loss of function, isolation and hopelessness.
Population	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).
Setting	The study was hosted online, with participants recruited from patient advocacy websites, newsletters, social media and internet forums.

Study	Devendorf 2018[42]
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	**Patient-reported theme: Lack of knowledge of GPs and health system**
	The majority of participants commented on their dissatisfaction with healthcare providers. Many said they encountered disdain, disbelief and a lack of knowledge from HCPs. Most encountered doctors who were trained to view ME/CFS as psychiatric. Several participants emphasised the need to educate doctors on ME/CFS, perhaps incorporating the topic into medical curriculums in the United Kingdom, the Netherlands, Canada and the United States. Participants called for more physiological research to develop medical treatments and destigmatise the field. Some patients emailed their doctors studies and publications to encourage them to learn about ME/CFS.
Limitations and applicability of evidence	Moderate methodological limitations due to 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	Edwards 2007[45]
Aim	To explore the experiences and difficulties of people living with ME/CFS.
Population	People diagnosed with ME/CFS by a medical professional. N=8; all women. Age range: 37-55 years. Illness duration range: 18 months to 12 years. Inclusion criteria: over 18 years of age, speak English as a first language, diagnosed with ME/CFS by a medical professional, have had ME/CFS symptoms for at least one year, consider ME/CFS as their main health problem, and currently experiencing symptoms of at least moderate severity. All but one had stopped working due to ME/CFS.
Setting	United Kingdom, diagnosed in the NHS

Study	Edwards 2007[45]
Study design	Interpretative phenomenological analysis of semi-structured interviews.
Methods and analysis	The first eight participants who responded during recruitment were interviewed. This number yielded sufficiently 'rich' data for analysis. Analysis was done as a series of steps through which themes and the relationships between them were identified. This was done first within, then across individual accounts. Constant checking led to each level of the analysis being verified or modified and ultimately enhanced by other levels.
Findings	**Patient-reported theme: Lack of knowledge from GPs**
	Participants described feeling angry and let down by the health profession. For some participants health professionals were interpreted as trying their best but lacking in knowledge and understanding. For others, experiences with health professionals were extremely negative. Such experiences increased participants' feelings of being disempowered, helpless and hopeless and undermined their belief in their ability to cope.
Limitations and applicability of evidence	Minor methodological limitations due to small sample size and homogeneity of population (all participants were women with ME/CFS). Minor concerns over applicability due to the concerns over the small and homogenous sample size and lack of representation of Health professionals in the sample.

Study	Hannon 2012[57]
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; practitioner interviews focused on current practice in the diagnosis and management of patients with 'ME/CFS', attitudes towards ME/CFS and training and education needs; Specialist 'CFS/ME'

Study	Hannon 2012[57]
	practitioner interviews focused on the needs of patients and asked for comments on existing 'CFS/ME' resources. 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.
Findings	**Awareness of ME/CFS**
	The GP and practice nurse respondents expressed varying degrees of understanding of 'CFS/ME' and some questioned whether 'CFS/ME' was a legitimate illness; they were unaware of the evidence base for this condition or believed the symptoms could be explained by a psychological problem or secondary gain. Those who did recognise it as a legitimate illness were aware that some of their colleagues fail to identify this condition which can lead to inappropriate diagnosis. Patients and carers explained how they took information to their GP in an attempt to raise their awareness of the condition. A gap in knowledge was also recognised by 'CFS/ME' specialists who highlighted a training need in primary care.
	Training needs
	Across patient and health professional interviews there was acknowledgment of the value of GPs recognising 'CFS/ME' as a legitimate condition and to have a sound understanding of the condition. Although some GPs and practice nurses were hesitant to prioritise time for face-to-face training due to the low priority given to 'CFS/ME', they suggested that they might engage in training if they thought that it would help them to reduce the number of consultations with patients who repeatedly present with symptoms of 'CFS/ME'. Practitioners described the need for easily accessible IT based training.
	Information on treatment
	Some GPs and practice nurses used the label as a last resort and with reluctance because making the diagnosis did not lead to obvious treatment approaches and they believed that there was no cure for 'CFS/ME'.
	Consensus on diagnostic criteria
	Practitioners described how the diagnosis of 'CFS/ME' was made by exclusion due to the lack of positive diagnostic criteria.
	Need for information on healthcare contacts
	Patients and carers highlighted the need for sign posting from their 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.
	Information for and support from specialist services:
	Health professionals described difficulties with referral to secondary care due to fragmented services and a lack of collaboration and a number of GPs and practice nurses were unaware of specialist ME/CFS services. Others had referred their patients to the specialist

Study	Hannon 2012[57]
	service, but lacked an understanding of what these services can offer patients. Patients were concerned around the long waiting time to attend specialist services and it was also suggested that improved communication between primary care and the specialist service may enable the GP to manage the patient during this period.
	Need for (online/digital) resources:
	The use of a template built into the GP computer system was suggested as being potentially useful to aid practitioners to conduct a symptom check-list and carry out the necessary investigations and view management options. GPs and practice nurses wanted to be able to print information on symptom management from an on-line resource during the consultation. GPs also suggested that a DVD would be useful for those patients who would struggle to read written resources because of fatigue and concentration and memory problems.
	Need for longer duration of consultations
	HPs recognised that a 10 minute consultation with a patient with 'CFS/ME' can be challenging due to the variety and complexity of symptoms. A ten minute consultation was also seen as a potential barrier to diagnosis by 'CFS/ME' specialists as GPs may not be able to gain a complete understanding of the variety of symptoms patients can experience and the impact of those on their life.
Limitations and applicability of evidence	Minor methodological limitations due to data analysis with themes mostly supported by single quotes. No concerns over applicability.

Study	Horton 2010[66]
Aim	To explore the nature of professional 'best practice' in working with people with ME/CFS.
Population	Health care professionals who had been nominated by people with ME/CFS who had taken part in an associated England-wide study of their support needs. N=6; genders not reported. Three participants were from specialist services (medicine, occupational therapy, physiotherapy) and three were from non-specialist services (medicine, occupational health, holistic practice). 36 people with ME/CFS nominated eight HCPs as having provided them with particularly helpful or effective care and six agreed to participate. One HCP was named by six different people with ME/CFS.
Setting	UK (East of England and London)
Study design	Qualitative study using semi-structured interviews.
Methods and analysis	Five interviews were conducted face-to-face ad one by telephone. Semi-structured interviews were based on a topic guide developed to reflect research literature identifying key aspects of service user and HCP experiences of ME/CFS and to deploy a framework of

Study	Horton 2010[66]
	question types (e.g. experience, opinion, feeling). The following topics were covered in interviews: i) general experiences of working with people with ME/CFS; ii) enabling people to access information and resources; iii) recognising and responding to the needs of people with ME/CFS; iv) enabling people to take an active role; and v) experiences of working with people from ethnic minorities, or from manual or routine occupations, or who have a severe condition. Interviews lasted between a half to a full hour and all were audio-recorded. Audio recordings were transcribed in full using English orthography according to an agreed protocol. To maintain anonymity of the participants, transcripts were labelled simply as Health Care Practitioner number 1 to 6. Codes were created from the first two transcripts as a basis for iterative thematic analysis. Themes and sub-themes were identified and developed by the individual researchers and a two-stage process of cross-checking and discussion was used to validate the analysis. Two validation meetings were held in which the main themes were presented to 23 people living with ME/CFS, family carers and ten HCPs. Comments from these groups showed strong accord with the findings of the study.
Findings	**Theme: Training for GPs and the role of specialist services**
	Specialist HCPs emphasised that there was a need for specialist services to be more 'visible' and to provide education for other HCPs, GPs especially, because there is a lack of knowledge about the condition in the GP population. This was thought to be because GPs lacked frequent exposure to these patients. Specialists had both experience and expertise to be able to support GPs and other HCPs in reaching or confirming a diagnosis, giving advice on appropriate medication, or providing services such as specialist Occupational Therapy. Specialists were involved in supporting people applying for benefits, often trying to help other agencies understand the variability inherent in the condition. HCPs described frustration at the lack of recognition or common acknowledgement of the condition by society and its institutions, such as health or benefits agencies, and poor access to resources such as CBT or other psychological services when they were thought to be necessary.
	Theme: Sources of information and resources
	Specialist practitioners were very clear in saying that not all sources of information were to be trusted. For some people with ME/CFS the internet may provide valuable information on specialist services, for others it may be hard to access and a source of confusing and misleading information. HCPs therefore reported exercising care about where they direct people for information which will help them understand or explain their condition to others. HCPs from specialist services reported using standard information packs and DVDs, and directing people to local support groups or expert patient programmes. They also reported recommending leaflets produced by *Action for ME* or the *ME Association*, and referring people to the *Citizen's Advice Bureau* (CAB) or *Disability Information and Advice Line* services (DIAL UK) for advice on disability-related support matters such as benefit or mobility issues. They highlighted the importance of providing appropriate and accurate information for employers of people with ME/CFS as well as employees with the condition.
	Theme: Diagnosis

Study	Horton 2010[66]
	It was acknowledged that reaching a firm diagnosis of ME/CFS can be challenging for GPs working in primary care. Although HCPs thought that the NICE guidelines were proving helpful, they saw the lack of any diagnostic test giving conclusive proof of the condition as impacting on practitioners and patients alike. One view was that until such a test is developed the existence of the condition will remain in doubt amongst some HCPs. Exposure to new presentations of ME/CFS was considered important for improving primary care practice. It enabled HCPs to recognise the condition and develop confidence in their diagnostic skills. Very careful history-taking, listening carefully and patiently to presentation of symptoms, with appropriate investigation were all considered vital elements of practice.
	Theme: Professional values
	Specialist HCPs identified a core minority group of GPs in their region who made referral to their service, but contrasted these GPs with the many who did not understand ME/CFS, and who see it as a psychological rather than a physical condition. They reported whole practices as having decided that ME/CFS did not exist and that many GPs would never make a referral to a specialist service. Participant HCPs reported how some patients told them that their GP openly stated their lack of belief in the existence of ME/CFS. All participants emphasised the importance and powerful therapeutic value of listening. Time limits in the primary care system often constrain patients from recounting their full story. Participants reiterated the need for practitioners to be knowledgeable, empathetic, inventive and capable of learning, acknowledging the patient's condition and taking it seriously. They should be able to respond flexibly to people's needs, accommodate the difficulties inherent in the condition that affect concentration and/or physical access, remain positive and engender a trusting relationship.
Limitations and applicability of evidence	Minor methodological limitations due to concerns over data analysis with some themes supported by single quotes. No concerns over applicability

Study	Jelbert 2010[68]
Aim	To provide a qualitative perspective of adolescents' experiences of ME/CFS
Population	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.
Setting	Paediatric outpatient clinic, UK

Study	Jelbert 2010[68]
Study design	Semi-structured interviews and thematic analysis (interpretative phenomenological analysis).
Methods and analysis	Data was collected through individual semi-structured interviews. All participants chose to be interviewed at home. Points of reference were agreed around areas that were felt likely to be of particular pertinence to young people with ME/CFS, including impact of CFS on everyday living, coping with ME/CFS, and impact of ME/CFS on self-identity. Additionally, the researcher sought to explore adolescents' understanding of ME/CFS and the impact of ME/CFS on their future hopes and plans. The interview therefore opened with broader questions around participants' general experiences of CFS: "Can you start by describing what CFS is to me?" and "Can you now tell me what CFS was like for you?", followed by further questions and prompts around the additional areas described. All interviews were audiotaped and transcribed in full. Transcripts were read several times by the interviewer, noting initial thoughts and then tentatively identifying initial themes, or summary phrases, from these notes. Emergent themes were listed and then progressively organized into clusters as possible connections between them were identified. The researcher revisited the transcript a number of 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	**Patient-reported theme: Lack of information, understanding and awareness from HCPs**
	Many of the young people expressed that they experienced difficulty with a lack of information, understanding and awareness around the condition from medical professionals involved.
Limitations and applicability of evidence	Minor methodological concerns due to small sample size and homogenous population of participants who attended the same clinic. 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	Marks 2016[84]
Aim	To explore health care professionals' experiences of working with children and adolescents with ME/CFS so as to develop an understanding of the processes relating to how they understand the condition.
Population	Paediatricians, physiotherapists and clinical psychologists working in two NHS organisations in the UK: a hospital outpatient paediatric service, and a specialist centre providing inpatient and outpatient care for young people ME/CFS.

Study	Marks 2016[84]
	N=10; 7 female, 3 male. Mean age not stated. Medical specialties were as follows: paediatricians (n=4), physiotherapists (n=3), and clinical psychologists (n=3). All had a minimum of 3 years' experience of working with ≥3 young people with ME/CFS.
Setting	Two NHS organisations in the UK: a hospital outpatient paediatric service, and a specialist centre providing inpatient and outpatient care for young people ME/CFS.
Study design	Semi-structured interviews and thematic analysis (grounded theory methodology).
Methods and analysis	The study followed grounded theory methodology, whereby theory is derived from data, facilitating in-depth analysis of HCPs' understanding of 'CFS/ME' by moving beyond 'what' they understand to ascertain the 'when', 'where', 'how' and 'why' of their understanding. A semi-structured interview schedule was developed by the research team. The initial schedule focussed on how participants referred to and understood ME/CFS, exploring thoughts about aetiology, maintaining factors and effective recovery. Following data analysis, the schedule was modified and focussed on the emerging theory. The audio-recorded interviews were conducted by the primary researcher and varied between 28 and 83 minutes in length. Interviews were transcribed and analysed consecutively, reflecting the continuous interaction between data collection and analysis. Line-by-line coding highlighted emergent concepts, and transcripts were simultaneously analysed to facilitate analytical and interpretive triangulation. Concepts were constantly compared both within and between transcripts, and these were grouped into categories describing data at a more abstract level. Axial coding was used to explore the relationships between categories. Theory was refined through 'selective coding' where a core category emerged, and a provisional model was proposed outlining how contexts produce particular beliefs, which generate certain actions and consequences. Emergent findings were validated by asking subsequent participants to judge the reasonableness of these and inviting them to expand the theory further. Data collection ceased when theoretical saturation was reached.
Findings	**Core theme: working with uncertainty**
	Health care professionals (HCPs) acknowledged a lack of understanding of ME/CFS compared to other health conditions. Unknown aetiology, limited evidence and research contradictions contributed to uncertainty.
	a) Making sense of ME/CFS
	Due to uncertainty and lack of empirically grounded understanding, HCPs endeavoured to 'make sense of ME/CFS' by developing their own understanding. Regarding aetiology, all recognised the contribution of physiological and psychological factors, however differences appeared in the emphasis given to these. Some HCPs believed in an undiscovered physiological cause, while others

Study	Marks 2016[84]
	believed symptoms served particular functions (e.g. avoiding anxiety). Credence given to physiological or psychosocial factors was often consistent with the participant's professional background. All HCPs reflected on moving from an original position of scepticism of ME/CFS to strongly believing in the condition, attributing this change to clinical experience. Personal beliefs were rooted in a professional's clinical experience, training and life. Few referred to the role of research in developing these perspectives, and variation in personal beliefs was attributed to uncertainties and individual experience.
	b) Diagnosis and choice of label given
	HCPs described uncertainty in appropriately identifying and labelling ME/CFS. Intrinsically linked with a professional's understanding of ME/CFS is the explanation given to families, and the subsequent choice of labels used to account for difficulties. However, a young person is likely to be referred to other HCPs who could conceptualise the difficulties differently and participants described significant **diagnostic variability** that resulted in a 'difficult' and 'challenging' diagnostic process given the need to safety diagnose in the absence of a definitive test. There was inconsistency in the use of the terms 'Chronic Fatigue' and 'CFS'. For some, these were synonymous, but others felt the latter conveyed increased symptom severity, or that the terms differentiated between fatigue rooted in physiological factors and fatigue stemming from psychosocial issues. Young people presenting to services with medically unexplained fatigue could receive one of a range of labels including 'ME/CFS', 'CFS', Chronic Fatigue', 'Chronic Pain' and 'MUPS'; difficulties could also be conceptualised and labelled as 'depression' and 'anxiety'. Some HCPs described feeling more comfortable giving a diagnosis of medically unexplained physical symptoms (MUPS) rather than ME/CFS, because of not being able to provide a clear aetiology.
	c) Pathways to intervention
	HCPs also described uncertainty regarding the appropriateness and effectiveness of treatment pathways. The choice of label given to a young person influenced subsequent intervention. The experience of receiving a diagnosis, and the explanation around it, was pivotal in families' acceptance of the diagnosis and label. The pathway to recovery varied as a consequence of the label given. HCPs described the importance of 'reconceptualising cases', illustrating the reflective nature of their practice and the continuous feedback between clinical experiences and making sense of ME/CFS.
	Summary/conclusion
	These themes highlighted the need for greater consistency across services, which may be achieved through **standardised specialist training** to ensure that HCPs are provided with, and work from, the same information.

Study	Marks 2016[84]
Limitations and applicability of evidence	Minor methodological limitations due to small sample size and recruitment skewing towards HCPs with positive attitudes towards ME/CFS (as participants were recruited on the basis of how they informed and validate emerging theory). No concerns about applicability.

Study	Raine 2004[106]
Aim	To compare general practitioners' perceptions of chronic fatigue syndrome and irritable bowel syndrome and to consider the implications of their perceptions for the use of psychological treatments.
Population	General practitioners randomly selected from the Department of Health's general practitioner database. N=46; male/female 29/17; mean age 46.9 years; had worked for an average of 14.8 years in general practice, and 9 were affiliated to a medical school.
Setting	NHS England
Study design	Qualitative analysis of transcripts of facilitated group discussions.
Methods and analysis	Participants were each sent a series of clinical scenarios involving patients with chronic fatigue syndrome or irritable bowel syndrome—for example, one scenario concerned the appropriateness of behavioural therapy in a patient who believes that chronic fatigue syndrome has an organic cause. The doctors were asked to rate their level of agreement with using mental health interventions. Two of the four groups were also given a systematic review of the effectiveness of mental health interventions for chronic fatigue syndrome and irritable bowel syndrome. The participants of each group met for a facilitated discussion in 4 groups of between 9 and 12, which lasted approximately 4 hours, where they explored any differences in opinion. Discussions were conducted according to a protocol comprising a description of the nominal group process to be followed, instructions to be given to each group, and explanations of the terms used in the questionnaire. Meetings were audiotaped and later transcribed and field notes were also taken. The analysis of the transcribed data involved independent scrutiny by two authors of the initial transcripts and journal notes to draw up a preliminary list of themes. The two authors then met to compare and discuss identified themes. Interpretations were also appraised by the other authors. Grounded theory was used to identify provisional themes by using the respondents' own concepts, then using these themes iteratively, applying them to later transcripts to allow the emergence of an analytical theory suited to the context.
Findings	**Knowledge about effective treatments**

Study	Raine 2004[106]
	For most of the participants, choosing appropriate treatments for chronic fatigue syndrome was like groping in the dark—either not knowing who to refer to or just "feeling hopeless and more hopeless". They might therefore consider mental health interventions only as part of a process of trying a range of treatments: "You would do anything for these patients".
	Knowledge about mental health interventions
	Main reasons for not referring patients for mental health interventions included lack of familiarity with mental health treatments "Medics don't really understand what psychologists do" and a perceived lack of local mental health resources.
Limitations and applicability of evidence	Minor methodological limitations due to the role of the researcher (half of the participants were given a systematic review on the effectiveness of mental health interventions prior to the data collection). Minor concerns over applicability as main findings emerging are driven by the study's original aims to compare general practitioners' perceptions of chronic fatigue syndrome and irritable bowel syndrome and to consider the implications of their perceptions for the use of psychological treatments.

Study	Ryckeghem 2017[115]
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 (ANP) 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. Inclusion criteria: diagnosis of CFS after multidisciplinary discussion in the diagnostic process; age above 18 years; given informed consent; Dutch as mother tongue. A convenience sample of GPs was recruited from different provinces in Belgium. Inclusion criteria: given informed consent, Dutch as mother tongue. 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 between May 2014 and January 2015 by the main investigator, using interview guide questions developed through an extensive literature review.

Study	Ryckeghem 2017[115]
	Patient 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 started with the same question: 'Could you tell me more about how it all started?' GP interviews took place in their practice, lasted on average 27 minutes and started with the question: 'What is your professional experience with patients diagnosed with CFS?' All interviews were audio-recorded and transcribed verbatim. Thematic analysis (open explorative thematic coding) was used. Analyses were conducted using Nvivo Version 10. Open coding was used; then a code tree was drafted, rendering data more manageable and well-organized. Thematic analysis was used to find similarities or contradictions in the experiences and expectations.
Findings	**Information about 'CFS/ME'**
	GPs experience mixed emotions following the diagnosis. They tend to believe the patient and try to show a sense of understanding for their problem, but are frustrated about their inability to solve the problem. They describe CFS as a complex diagnosis, emphasizing its multifactorial nature with problems in the psychological, physical and social domain. They acknowledge that very little is known about the disease.
	Support fulfilling their intermediary role between patients and multidisciplinary teams
	a) Resources: (after completion of the diagnostic care pathway) The referral centre passes a coordinating role on to the GP, who has an intermediary role between the patient and members of the multidisciplinary team. Most GPs felt that because of lack of time- they cannot fulfil this task; and most experienced they need someone who can take over some coordinative task, so an optimal guidance of the patient can be guaranteed beyond the referral centre. Patients also experience that this role is often difficult to carry out by the GP.
	b) Improved communication & feedback from referral centre: Overall, GPs reported there is lack of communication between the referral centre and the GP and they get little or delayed feedback. As a result they are often dependent on the patient for information, which complicates the implementation of this coordinating role. They also experience they have to wait long time before receiving reports of examinations performed.
Limitations and applicability of evidence	Moderate methodological limitations due to the role of the researcher not being discussed, concerns over data analysis (due to a lack of sufficient detail and some themes supported by single quotes) Moderate concerns over applicability to NHS due to setting (specific application to Belgian healthcare system).

Study	Stenhoff 2015[124]
Aim	To investigate medical students' beliefs, attitudes and knowledge of ME/CFS.
Population	Undergraduate medical students in years 3, 4 and 5 at the University of Manchester, UK. N=21; 7 female, 14 male. Mean age: 22 years old. Four were third-year students, 11 were fourth-year students and six were fifth (final)-year students. Participants were recruited through the university's student-net, poster adverts around campus and via personal contact. Sampling ended at saturation in a staged approach, with two students turned away at the end of the study.
Setting	University of Manchester medical school
Study design	Qualitative; face-to-face semi-structured interviews and inductive thematic analysis.
Methods and analysis	Face-to-face semi-structured interviews lasting between 15 and 50 minutes (mean duration = 22 minutes) were conducted. Participants were interviewed individually between December 2010 and May 2011. A brief topic guide was used to explore beliefs about ME/CFS, encounters with people living with ME/CFS, views on the management of ME/CFS, societal attitudes towards ME/CFS and learning needs. Interviews were digitally recorded and transcribed verbatim. Participants' names were removed from transcripts and participants were assigned an identification number. Transcripts were analysed using thematic analysis incorporating aspects of grounded theory, for example, constant comparative methods and memo writing. The data were analysed within a realist framework which aims to report experiences, meanings and the reality of participants. Transcripts were initially read and reread. After familiarisation and immersion into the data, descriptive line-by-line coding was undertaken, allowing the researchers to organise the data into meaningful groups. As different themes emerged, earlier transcripts were read and discussed by four researchers. Discussion among several researchers with different perspectives served to increase the trustworthiness of the analysis. Upon completing initial coding, transcripts were examined for broader themes. Themes were coded taking an interpretive stance and were data-driven. The themes were 'defined and refined' to create a hierarchy of superordinate themes and sub-themes. Memo writing was undertaken throughout the data analysis, allowing for the refinement ideas and enabling the constant comparative approach. Concurrent data collection and analysis allowed the emerging analysis to influence and shape further data collection.
Findings	**Theme: Training needs**
	Summary: All students in the study reported they had no formal training in ME/CFS. Students made useful suggestions about how training could be best delivered. All participants reported having received no formal ME/CFS training to date. Participants commented that it was 'brushed under the rug' or 'skimmed over' within the medical curriculum. Although students were unsure of the prevalence of ME/CFS, students felt that course leaders prioritised conditions based on prevalence and surmised this may explain why ME/CFS had not been included. Others felt that

Study	Stenhoff 2015[124]
	ME/CFS was not included because the condition was too controversial, complex or unclear. Despite this, students reported that they and their course mates would find training to be beneficial in their future roles. Where training was desired, students wanted introductions on recognising, diagnosing and managing patients with ME/CFS. Some left their knowledge was so limited that any information would be valuable. Students reported that self-directed study made up a substantial part of their undergraduate course. Students felt that questions related to the condition either be 'skimmed over' or not covered at all. As a result, some students felt that ME/CFS training was more suited to a lecture or seminar, suggesting that this would need to be made compulsory or examined. In order to engage students and change negative attitudes towards ME/CFS, students suggested that delivery of teaching around ME/CFS should stress the impact of the condition of patients' lives. They suggested that meeting patients would promote more positive attitudes towards the condition. Some felt that ME/CFS training could be made engaging by making teaching interactive and being taught by specialists who understood the condition.
	Theme: Limited knowledge but many opinions
	Summary: Although students were aware of ME/CFS and able to offer many opinions regarding its aetiology and prognosis, they reported that their knowledge was limited. Participants reported having limited knowledge of ME/CFS and found it difficult to describe symptoms of ME/CFS beyond tiredness or fatigue. Some admitted that they were merely speculating by using a literal interpretation of the term 'chronic fatigue syndrome'. Students viewed ME/CFS as having no known pathology and perceived ME/CFS to be a diagnosis of exclusion. Some students viewed ME/CFS as being caused by biological factors, such as genetics or by a physical disease process such as a viral infection. Some students viewed the problem as being caused by personality type or hypersensitivity or saw ME/CFS as being 'medically unexplained'. Other students held more complex biopsychosocial models of ME/CFS and recognised that the condition may be multifactorial. Four participants who were psychology intercalating students described more detailed and developed cognitive and behavioural maintenance models of ME/CFS which reflected more certainty around an understanding of ME/CFS. When considering prognosis, some students perceived ME/CFS to be a lifelong condition. However, some felt that outcomes varied between patients depending on the severity of the condition, the patient's personal situation, illness beliefs and coping strategies, demonstrating a more biopsychosocial understanding of the illness.
	Theme: Influences on medical students' attitudes (origins of HCPs' negative beliefs towards ME/CFS)
	Summary: Students expressed difficulty understanding ME/CFS within the biomedical framework that still prevailed within medicine. In the absence of training, students acquired their knowledge of ME/CFS largely from informal sources. Clinical tutors were particularly influential in shaping students' beliefs and attitudes towards ME/CFS.

Study	Stenhoff 2015[124]
	Students articulated that the legitimacy of ME/CFS patients' symptoms was at times questioned by themselves and by their fellow students. Negative attitudes were held towards patients with ME/CFS; these attitudes were expressed both implicitly and explicitly. Implicit negative attitudes were revealed where students compared everyday tiredness and the symptoms of ME/CFS. ME/CFS patients were viewed as being time-wasters or malingerers or as having a second agenda. Others perceive ME/CFS to be associated with poor coping skills or as being related to an individual's personality or 'laziness'. Students claimed that they encountered these negative attitudes from clinical tutors and that doctors who trained them in clinical settings were highly influential figures. Often students took the opinions of these trainers as unquestionable truths as they viewed their seniors as being more experienced and knowledgeable. This authority of medical knowledge meant that negative and dismissing attitudes were often 'passed down' to students. In many cases, doctors made negative throwaway comments and gestures regarding ME/CFS patients that students subsequently internalised. Students gained some of their understanding of ME/CFS from the media. Media coverage was often highly stigmatising and derogatory towards individuals with the condition and cast further doubt on the genuineness of ME/CFS patients. Conversely, students who had personal knowledge of someone with ME/CFS reported that this experience had positively changed their opinions and attitudes towards ME/CFS, expressing more empathetic attitudes towards individuals living with ME/CFS. Specifically, students felt that previous knowledge of a person *before* the onset of ME/CFS helped to legitimise the illness.
Limitations and applicability of evidence	Minor methodological limitations due to recruitment of participants through responses to an advertisement, therefore risking over-representation of students who are more informed or have stronger views on ME/CFS. Minor concerns about applicability due to the population of medical students rather than practicing HCPs and the fact that all students were attending the same medical school at the University of Manchester.

Study	Taylor 2005[130]
Aim	To examine the adequacy of the social model for explaining the disability experience of persons with ME/CFS.
Population	Adults with ME/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.
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.

Study	Taylor 2005[130]
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. Analysis of the data 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. Several strategies were used to achieve confidence in the findings. Data were triangulated by comparing information within and across data collection methods, across participants, and across time. Member checking with the participants was done to assure that the evolving understanding of their disability experience accurately reflected their views.
Findings	**Patient-reported theme: Lack of knowledge from HCPs**
	The participants in this study consistently reported that when they sought help for their condition from health care providers, most health care professionals were either relatively ignorant or incredulous of ME/CFS. Consequently, most participants reported experiences characterized by: knowledge about ME/CFS; outright disbelief in the legitimacy of ME/CFS as a medical entity; lack of validation of participants' described impairment and symptoms; absence of treatment planning and treatment recommendations; tendency to overemphasise psychological and social variables as possible causes of the symptoms; tendency to overprescribe psychotropic medications; tendency to view exercise and psychotherapy as the only non-pharmacological treatments for ME/CFS. Participants reported that they sought treatment for their CFS symptoms and impairments from an average of six physicians before they were ultimately diagnosed with CFS. After being diagnosed with CFS, most participants reported continued and ongoing dissatisfaction with their treatment, particularly when it was administered by a physician that did not specialize in CFS. Along the way they encountered misinformation, misdiagnosis, and inappropriate treatment recommendations. Finding a physician who could provide appropriate services was often tricky for participants. Most participants found information on CFS outside medical care (through the Internet or self-help groups) and then took the information to their physicians. One participant reported that she took articles to her doctor to convince and inform him about CFS. Many participants talked about how they had to screen and select their health care providers based on their willingness to recognize their condition. Participants also talked about the lack of knowledge in occupational therapy about ME/CFS. An important aspect of service access for the participants was the relative absence of any rehabilitation services. For instance, none of the subjects in this study had received occupational therapy within the past 12 months.
Limitations and applicability of evidence	No concerns about applicability or methodological limitations.

Barriers and facilitators to information, education and support for health and social health professionals

Study	Ax 1997[13]
Aim	To describe ways in which physicians and people with ME/CFS communicated their cognitions and illness beliefs which form the bases of their treatment expectations and the consequences of such interactions in terms of future treatment choices.
Population	People diagnosed with ME, CFS, or PVFS by a medical practitioner. This paper was a qualitative report based on two separate studies which were part of a larger project on illness adjustment. Participants for these studies were recruited through several London-based ME support groups. Study 1: n=9, mean age (SD, range): 44.2 (5.21, 16-68) years; male/female: 3/6; mean illness durations (range): 7.89 (1-14) years Study 2: n=9, mean age (SD, range): 44.5 (7.67,34-55) years; male/female: 1/8; mean illness duration (range): 7.7 (1-19) years
Setting	Patients recruited through ME support groups; interviews conducted in participants' home
Study design	Qualitative interview study
Methods and analysis	Semi-structured interviews were conducted at the participants' home, covering issues concerning adaptation to CFS and support received. That included questions about the doctor-patient relationship before and after diagnosis, satisfaction with the treatment received and about people with ME/CFS' views on their doctors and the health service. Interviews lasted about 90 minutes, were tape-recorded and transcribed. Data was analysed by content analysis
	Barrier: Anger at GPs' advice
	Advice offered by GPs was not always appreciated by patients, especially if it involved giving up important activities. Advice to find psychological or psychiatric support was also not greeted with enthusiasm by patients' whose illness model including maladaptive behaviour was unacceptable and clashed with their own view of CFS as a physical and uncontrollable condition. Divergent opinions were associated with the development of extreme anger among patients' towards doctors and other health professionals who did not believe their illness was 'real' , were arrogant or gave them bad advice and led them to reject medical and health professionals and increased their sense of self-reliance (expressed in terms of increased self-management of symptoms).
Limitations and applicability of evidence	Moderate limitations due to the role of the researcher, lack of detail on the method of data analysis. No concerns over applicability.

Study	Bayliss 2016[15]
Aim	Following the development of an online training module for GPs, and an information pack and DVD for patients, this study explored the extent to which these 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	Individuals with an existing diagnosis of 'CFS/ME', recruited from participating GP practices. Patients with other conditions, or other factors that may account for their fatigue were excluded. 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 (n=11); male/female 2/9; mean age (range): 46 (27-74) years; GPs (n=8)
Setting	Participants' homes, UK
Study design	Semi structured interviews with thematic analysis.
Methods and analysis	Patient interviews focused on their views on the 'CFS/ME' patient resource and their experience with their GP before and after the practice had access to the online training. Interviews were digitally recorded and transcribed. Analysis was conducted in parallel with the interviews and was inductive, using components of thematic analysis. Thematic categories were identified in initial interviews and then explored in subsequent interviews. Main categories were compared across interviews and reintegrated into common themes. Interview transcripts were read, annotated, and categorised independently by researchers of different professional backgrounds and patient and carer research partners to increase reliability of the analysis. Open coding was used initially. It was agreed that theoretical saturation across the data sets was achieved when no new themes emerged during the final interviews.
Findings	**Barrier: Reluctance of GPs**
	The research team experienced difficulty recruiting GP practices for training. Reasons given for the lack of engagement included a level of scepticism about ME/CFS and the complexity of managing the condition and working with patients and their families. One GP highlighted the divide within their profession, with those who will manage patients with complex conditions such as 'CFS/ME', and those who prefer to refer on. There was an implication that, for some, the level of commitment required to manage patients over the longer term is too much for a primary care professional, and that 'CFS/ME' should be managed in secondary care by specialists. Reasons that GPs gave for a lack of commitment to the ME/CFS training included the small number of patients with the condition, pressures on time within a consultation and suggestion that ME/CFS was not a priority.

Study	Bayliss 2016[15]
	Facilitator: Referral to specialist services
	A recommendation that came from the study was a call for greater investment in secondary care services. For example, most GPs interviewed in the study reported that training highlighted the complexity of the condition. They therefore believed that it would be more appropriate for ME/CFS to be managed by a specialist service. Patients also wanted more access to specialist services, with some recognising that GPs didn't have the time to manage their condition.
	Barrier: Limited specialist referral options
	Limited referral options were seen as a barrier to successfully working with patients to manage ME/CFS. The resources in this study were designed with the aim of increasing the referral of more severe cases of ME/CFS to secondary care services. During the time of the study, a specialist with an interest in ME/CFS retired and other ME/CFS services were redesigned. GPs therefore remained unsure when they should refer, where to refer and what the specialist services could offer.
	Facilitator: Online training resource
	GPs valued the online training resource used as part of the study as it provided the information required for GPs and patients to work in partnership to prioritise symptoms and develop a management plan over a number of consultations. GPs who completed training said that their knowledge of ME/CFS improved and it helped them establish a positive relationship with their patients as they felt they now had something to offer. The video clips on the online training module that showed how a GP can work with a patient within a consultation were particularly valued by a number of GPs.
	Barrier: Lack of opportunity for reinforcement after training
	GPs believed that a barrier that prevented them working with ME/CFS patients was that after completion of the training they had difficulty remembering key messages due to limited opportunities to diagnose the condition because it was seen as relatively rare.
	Barrier: Limitations of consultation
	Where opportunities arose to use what they had learnt from training, GPs and patients reported a lack of time within a ten minute consultation. Patients felt unable to explain the complexity of their condition to their GP. Without the opportunity to relay this information, patients struggled to work with their GP to manage their symptoms. GPs reported rarely using the information packs with ME/CFS patients who had been diagnosed for some time.
Limitations and applicability of evidence	Very minor methodological limitations due to the potential influence of the role of the researcher No concerns regarding applicability.

Study	Beasant 2014[18]
Aim	To understand the experiences of adolescents and families in accessing and using a specialist service and to explore whether or not adolescents and their mothers value referral to a specialist service for young people with 'CFS/ME'.
Population	Adolescents taking part in the Specialist Medical Intervention and Lightning Evaluation (SMILE) study and their mothers. Participants were eligible for the SMILE study if they had been diagnosed with 'CFS/ME', were aged between 12 and 18 years and were mildly or moderately affected by the condition; that is, they were not house bound (NICE, 2007). Purposive sampling to ensure that interviews included a range of participants in terms of age, sex, socioeconomic circumstance and ethnicity as well as families from both intervention arms. N=12 adolescents; male/female 3/9; age mean (SD) 13.9 (1.6) years; illness duration median (IQR) 13 (9 to 18) months; 5 were interviewed post randomisation but before receiving the 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.
Setting	Participants' homes, UK
Study design	Semi structured interviews with thematic analysis
Methods and analysis	Families were interviewed at three possible time points: after initial assessment before randomisation, after randomisation before the intervention, and after the intervention. Adolescents with 'CFS/ME' were interviewed once at one of these time points for not more than 20 min; parent interviews lasted for 20–60 min. A checklist of topics was used to ensure that similar areas were covered in each interview (experiences of the initial clinical assessment appointment, study participation and the interventions) but with sufficient flexibility to enable participants to raise topics of interest to them. Interviews were audio-recorded and transcribed. Data items were systematically assigned codes using the qualitative data organisation package NVivo and analysed thematically 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.
Findings	**Facilitator: Referral to specialist services**
	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. This enabled positive change and steps towards a managed recovery. Some mothers felt that the 'CFS/ME' service reinforced symptom management strategies that they had been trying to get their child to follow, and that they felt their child would be more likely to listen if techniques were legitimised by a health-care professional. Half the adolescents reported that specialist medical care was positive, as it enabled them to talk about their illness and gave guidance on how to manage their condition. A letter provided by the 'CFS/ME' service confirming a diagnosis enabled

Study	Beasant 2014[18]
	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 methodological limitations due to unclear relationship between the researcher and participants, potential issues with data richness with some findings supported by single quotes. Minor concerns regarding applicability due to study's aim to understand the experiences of accessing as well as using a specialist service (some participants had not yet used the service) and unclear which intervention the findings relate to and the representativeness of the sample considering it consisted of feasibility RCT participants which may differ from eligible patients not recruited to a trial.

Study	Beaulieu 2000[19]
Aim	To examine multiple perspectives on stigmatization and legitimation of CFS.
Population	Health professionals including general practitioners, mental health professionals (one of whom was not a physician), infectious disease specialists, immunologists and rheumatologists, recruited following identification by people with CFS participating in the study. N=15; male/female 10/5; had been in practice from six to seventeen years and individually had seen from six to almost one hundred cases. People who were English-speaking and who had a diagnosis of CFS from a medical doctor, recruited from physicians' practices, support groups and identified by leaders of associations. N=43; male/female 16/27; 26% were in school or working full or part time; mean age at onset was 34.2 years (range 15 to 58 years); people had been ill for an average of seven years. Significant others including friends, parents, spouses, adult children and a sibling, recruited following identification by people with CFS participating in the study. N=23; male/female not reported; 69% were working.
Setting	McGill University, Montreal
Study design	Qualitative interview study
Methods and analysis	Mixture of structured and semi structured questions related to approach to diagnosing, explaining and treating CFS, views on support groups and alternative therapies, whether thinking had changed over time, impressions of typical and atypical patients and challenges in dealing with people with ME/CFS (doctors); symptom experiences, the impact on roles and functioning, beliefs about cause,

Study	Beaulieu 2000[19]
	attempts to manage the illness through help seeking and treatment and reactions from health professionals (people with CFS); knowledge about people with ME/CFS' experiences, ideas about cause and treatments, how having someone close with CFS affected their lives (significant others). 78% of those who agreed to face to face interviews also consented to taping and tapes were transcribed. For telephone interviews and interviews in which people refused to be taped, notes of key words and phrases were taken. These notes were elaborated as soon as possible after the interviews. Interviews took place in people's homes, their offices, the researcher's office, or in neutral public places such as coffee shops or parks. A few doctors were interviewed by telephone. Interviews were analysed using thematic analysis. Transcripts of each interview were summarized according to the broadest content areas of questions. Summaries were then pooled according to categories and read and reread for recurring themes and variations in the first gross categories.
Findings	**Barrier: HCP cautiousness due to uncertainty around ME/CFS**
	Conflicting medical findings and opinions about CFS left practicing clinicians with a fundamental problem on how to think about and manage an illness in which patients' self-reports are largely uncorroborated by physical examination and laboratory findings. Without specific positive findings or medical consensus to lend authority to diagnosing, explaining, and treating CFS, practicing clinicians felt the impact of this illness most clearly in their dealings with people with ME/CFS. The uncertainties associated with the illness made clinicians insecure about the accuracy of their information, hence the cautious and uneasy tone of their explanations. If they did not contextualize and qualify their explanations and were proven wrong in the future, their credibility was at stake. The tone of explanations of CFS clinicians gave to patients was cautious and tentative. The content reflected clinicians' attempts to preserve patients' morale and avoid stigmatizing explanations on the one hand, while maintaining their own credibility on the other.
	Barrier: Fear of negative reactions to psychological discussions
	Concerns about patients' reactions to any suggestions of psychological factors could reach the point where clinicians hesitated even to discuss concurrent affective disorders that they detected. Clinicians had perceptions of CFS as an illness that could ruin lives and perceptions of people with ME/CFS as resistant to any suggestion of a psychological disorder. As a result, they tried to avoid clearly stigmatizing explanations; some avoided a label and discussions of etiology. But in so doing, they may have left an ambiguous impression.
	Facilitator: Communication between HCP and patient
	Through regular office or home visits and social connections to patients, clinicians gained varying degrees of information about the psychosocial aspects of patients' lives. Part of the mix on which the etiological hypotheses of clinicians were founded, were observations.
	Barrier: HCP reluctance to refer to support groups

Study	Beaulieu 2000[19]
	Clinicians' reactions to support groups varied over a narrower range from cautious to leery. Most would hesitate to recommend support groups because they had little knowledge of these groups. Others hedged because they believed support groups could be harmful or helpful, depending on their approaches and depending on individual patients. For these clinicians, the problem was that they had no way of determining how a given patient would be affected. One recounted patients being devastated after being exposed to the worst scenarios in support groups and believing such fates were inevitable. Others surmised that support groups could contribute to chronic disability because they "medicalize patients' distress", "reinforce illness behaviours", "institutionalize illness" and "possibly encourage dependency". One wondered whether patients didn't "pick up symptoms" from such groups. Another gave a cautious nod to groups who were "more or less involved with the mainstream" of medical thinking.
Limitations and applicability of evidence	Moderate methodological limitations due to concerns over participant selection (with HCP participants directly selected by ME/CFS patients) and data analysis (coding and analysis by a single researcher). Minor concerns over applicability as main findings emerging are driven by the study's original aims to explore multiple perspectives on stigmatization and legitimation of CFS and due to the study being published prior to new guidelines and diagnostic criteria.

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. 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.

Study	Brigden 2018[22]
	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	**Barrier: Unhelpful NHS resources (compared to alternative peer-led sources)**
	Participants felt that the NHS sites were not user-friendly; they used medical terminology, lacked depth and were static-the content remained unchanged. Sites reported to be accessed regularly (i.e. patient-led/peer-led) used in group terms and phrases which were accessible and appealing, were considered to offer greater level of depth and were constantly updated. 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.
	Facilitator: Social support via digital resources
	Participants described the loneliness of the condition. Through spending time on social websites, 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.
Limitations and applicability of evidence	Minor methodological limitations due to the role of the researcher, lack of details on the data analysis. Minor concerns over applicability due to the study not including severely affected adolescents.

Study	Broughton 2017[24]
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

Study	Broughton 2017[24]
	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	**Barrier/Facilitator: 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 methodological limitations. Minor concerns over applicability due to population sample being recruited from people completing ME/CFs treatment on the NHS who may have already received support and/or information. Excluded severely affected (those who were too severe to participate in interviews).

Study	Chew-Graham 2008[31]
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	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

Study	Chew-Graham 2008[31]
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	**Barrier/Facilitator: 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. 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 methodological limitations due to lack of data richness with some findings supported by single quotes. Minor concerns over applicability due to the research aim and sample which consisted of people recruited in a RCT (FINE trial).

Study	De Carvalho Leite 2011[37]
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/M', 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	Participants recruited via ME/CFS support groups, community organisations and centres and interviews conducted at the participants' home.
Study design	Qualitative inquiry using in-depth semi-structured interviews

Study	De Carvalho Leite 2011[37]
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. 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	**Barrier/Facilitator: Diagnosis and referral**
	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'). Until a diagnosis was gained, social services could not even assess patients' needs in order for them to gain access to social care support.

Study	De Carvalho Leite 2011[37]
Limitations and applicability of evidence	Very minor limitations due to the role of the researcher on the findings not being discussed. 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 2019[40]
Aim	To explore physicians' views on the challenges to studying and approaching recovery, to examine these challenges in-depth and provide recommendations that will improve how researchers and practitioners approach the study and quantification of ME and CFS recovery.
Population	Physicians specialising in ME/CFS of diverse medical specialties (n=10), recruited via non-probabilistic, purposive sampling. Specialists were defined by their extensive patient experience, research contributions and significant involvement in the field. Other physicians (n=3), not identified as ME/CFS specialists (one paediatrician, two psychiatrists) were also recruited. n=13, males: 9, females: 4; mean age 60 years. For years in practice, three physicians had 30 or more years, seven had 20-29 years, one had 10-19 years and two had 1-9 years of medical experience. The sample was diverse in their medical specialties: epidemiology (n=1), geriatrics (n=1), infectious diseases (n=1), neurology (n=1), internal medicine (n=2), psychiatry (n=2), general medicine (n=3), and paediatrics (n=5); three physicians identified with two medical specialties.
Setting	The place of work of the recruited physicians is not specified. The study was conducted at DePaul university in the USA.
Study design	Qualitative interview study
Methods and analysis	Semi-structured phone-based interviews (one via email) (mean duration 31 minutes). Interviews asked physicians about their general thoughts on recovery from ME/CFS-defining, measuring and studying recovery. Questions were inspired by online, patient discussion boards discussing the PACE trial. Interviews were audio-recorded, transcribed verbatim and verified for accuracy. Transcripts were analysed using deductive thematic analysis by two researchers.
Findings	**Facilitator: Good communication and follow-up** Some physicians followed up with their patients over phone or email. This communication benefits both the physician and patient. Physicians gain feedback about their practice while learning about the course of ME/CFS. Meanwhile, patients feel supported by their doctor, save money and avoid the risk of a symptom flare.
Limitations and applicability of evidence	Minor methodological limitations due to the role of the researcher and lack of data richness with themes mostly supported by single quotes.

Study	Devendorf 2019[40]
	Minor concerns over applicability as main findings emerging are driven by the study's original aims to explore physicians' views on recovery.

Study	Hannon 2012[57]
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 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	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; practitioner interviews focused on current practice in the diagnosis and management of patients with ME/CFS, attitudes towards ME/CFS and training and education needs; Specialist 'CFS/ME' practitioner interviews focused on the needs of patients and asked for comments on existing 'CFS/ME' resources. 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.
Findings	**Barrier: Need for training in primary care**
	A gap in knowledge was recognised by 'CFS/ME' specialists who highlighted a training need in primary care. The GP and practice nurse respondents expressed varying degrees of understanding of 'CFS/ME' and some questioned whether 'CFS/ME' was a legitimate illness; they were unaware of the evidence base for this condition or believed the symptoms could be explained by a psychological problem or secondary gains. Patients and carers explained how they took information to their GP in an attempt to raise their awareness of the condition.
	Barrier: Limited information for support referral

Study	Hannon 2012[57]
	Patients and carers highlighted the need for sign posting from their 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.
Limitations and applicability of evidence	Minor limitations due to the role of the researcher and lack of data richness with themes mostly supported by single quotes. No concerns over applicability.

Study	Horton 2010[66]
Aim	To explore the nature of professional 'best practice' in working with people with ME/CFS.
Population	Health care professionals who had been nominated by people with ME/CFS who had taken part in an associated England-wide study of their support needs. N=6; genders not reported. Three participants were from specialist services (medicine, occupational therapy, physiotherapy) and three were from non-specialist services (medicine, occupational health, holistic practice). 36 people with ME/CFS nominated eight HCPs as having provided them with particularly helpful or effective care and six agreed to participate. One HCP was named by six different people with ME/CFS.
Setting	UK (East of England and London)
Study design	Qualitative study using semi-structured interviews.
Methods and analysis	Five interviews were conducted face-to-face ad one by telephone. Semi-structured interviews were based on a topic guide developed to reflect research literature identifying key aspects of service user and HCP experiences of ME/CFS and to deploy a framework of question types (e.g. experience, opinion, feeling). The following topics were covered in interviews: i) general experiences of working with people with ME/CFS; ii) enabling people to access information and resources; iii) recognising and responding to the needs of people with ME/CFS; iv) enabling people to take an active role; and v) experiences of working with people from ethnic minorities, or from manual or routine occupations, or who have a severe condition. Interviews lasted between a half to a full hour and all were audio-recorded. Audio recordings were transcribed in full using English orthography according to an agreed protocol. To maintain anonymity of the participants, transcripts were labelled simply as Health Care Practitioner number 1 to 6. Codes were created from the first two transcripts as a basis for iterative thematic analysis. Themes and sub-themes were identified and developed by the individual researchers and a two-stage process of cross-checking and discussion was used to validate the analysis. Two validation meetings

Study	Horton 2010[66]
	were held in which the main themes were presented to 23 people living with ME/CFS, family carers and ten HCPs. Comments from these groups showed strong accord with the findings of the study.
Findings	**Barrier: Disease severity**
	A very small proportion of people seen by specialists HCPs were living with a severe condition and were significantly unwell, confined to home, or bedbound in a darkened room, unable to communicate. This was reported to be extremely challenging even by the specialist HCPs who may have very few helpful suggestions.
	Facilitator: Communication between HCP and patient
	Specialist HCPs would visit people with serious condition at home, or if appropriate maintain contact by phone, especially to offer support for the family
Limitations and applicability of evidence	Minor methodological limitations due to the role of the researcher and data richness with some findings supported by single quotes. No concerns over applicability.

Study	Ryckeghem 2017[115]
Aim	To explore the experiences and expectations of patients with chronic fatigue syndrome and general practitioners to develop the potential role of an advanced nurse practitioner at the diagnostic care path of abnormal fatigue developed for regional transmural implementation in the Belgian provinces of East and West Flanders.
Population	A purposive sample of patients was selected through the department of General Internal Medicine at the University Hospital Ghent to achieve maximum variation. A convenience sample of GPs was recruited from different provinces in Belgium. Patients (n=15); median age (range): 45 (33-59 years), n=14 female; GPs (n=15); median age (range): 49 (31-62 years), n=7 female.
Setting	University Hospital Ghent
Study design	Qualitative interview study
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?'

Study	Ryckeghem 2017[115]
	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	**Barrier: Information overload**
	Providing information to the patient at the right time about what matters is important. Some patients suffer from information overload, hampering a clear understanding.
	Barrier: Lack of consistency or a dedicated HCP during consultations
	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, advices them and instructs and assists them at all stages of their care process.
	Barrier: Lack of communication
	Overall, GPs reported there is lack of communication between the referral centre and the GP and they get little or delayed feedback. As a result they are often dependent on the patient for information, which complicates the implementation of this coordinating role. They also experience they have to wait long time before receiving reports of examinations performed.
Limitations and applicability of evidence	Moderate limitations due to the role of the researcher and concerns over data analysis due to lack of sufficient detail and some themes supported by single quotes). Moderate concerns over applicability to NHS due to setting (specific application to Belgian healthcare system).

Study	Stenhoff 2015[124]
Aim	To investigate medical students' beliefs, attitudes and knowledge of ME/CFS.
Population	Undergraduate medical students in years 3, 4 and 5 at the University of Manchester, UK. N=21; 7 female, 14 male. Mean age: 22 years old. Four were third-year students, 11 were fourth-year students and six were fifth (final)-year students. Participants were recruited through the university's student-net, poster adverts around campus and via personal contact. Sampling ended at saturation in a staged approach, with two students turned away at the end of the study.
Setting	University of Manchester medical school

Study	Stenhoff 2015[124]
Study design	Qualitative; face-to-face semi-structured interviews and inductive thematic analysis.
Methods and analysis	Face-to-face semi-structured interviews lasting between 15 and 50 minutes (mean duration = 22 minutes) were conducted. Participants were interviewed individually between December 2010 and May 2011. A brief topic guide was used to explore beliefs about ME/CFS, encounters with people living with ME/CFS, views on the management of ME/CFS, societal attitudes towards ME/CFS and learning needs. Interviews were digitally recorded and transcribed verbatim. Participants' names were removed from transcripts and participants were assigned an identification number. Transcripts were analysed using thematic analysis incorporating aspects of grounded theory, for example, constant comparative methods and memo writing. The data were analysed within a realist framework which aims to report experiences, meanings and the reality of participants. Transcripts were initially read and reread. After familiarisation and immersion into the data, descriptive line-by-line coding was undertaken, allowing the researchers to organise the data into meaningful groups. As different themes emerged, earlier transcripts were read and discussed by four researchers. Discussion among several researchers with different perspectives served to increase the trustworthiness of the analysis. Upon completing initial coding, transcripts were examined for broader themes. Themes were coded taking an interpretive stance and were data-driven. The themes were 'defined and refined' to create a hierarchy of superordinate themes and sub-themes. Memo writing was undertaken throughout the data analysis, allowing for the refinement ideas and enabling the constant comparative approach. Concurrent data collection and analysis allowed the emerging analysis to influence and shape further data collection.
Findings	**Barrier: Lack of training**
	All participants reported having received no formal ME/CFS training to date. Participants commented that it was 'brushed under the rug' or 'skimmed over' within the medical curriculum. Although students were unsure of the prevalence of ME/CFS, students felt that course leaders prioritised conditions based on prevalence and surmised this may explain why ME/CFS had not been included. Others felt that ME/CFS was not included because the condition was too controversial, complex or unclear. Despite this, students reported that they and their course mates would find training to be beneficial in their future roles. Where training was desired, students wanted introductions on recognising, diagnosing and managing patients with ME/CFS. Some felt their knowledge was so limited that any information would be valuable. Students reported that self-directed study made up a substantial part of their undergraduate course. Students felt that questions related to the condition would either be 'skimmed over' or not covered at all. As a result, some students felt that ME/CFS training was more suited to a lecture or seminar, suggesting that this would need to be made compulsory or examined. In order to engage students and change negative attitudes towards ME/CFS, students suggested that delivery of teaching around ME/CFS should stress the impact of the condition of patients' lives. They suggested that meeting patients would promote more positive attitudes towards the condition. Some felt that ME/CFS training could be made engaging by making teaching interactive and being taught by specialists who understood the condition.

Study	Stenhoff 2015[124]
Limitations and applicability of evidence	Minor methodological limitations due to recruitment of participants through responses to an advertisement, therefore risking over-representation of students who are more informed or have stronger views on ME/CFS. Minor concerns about applicability due to the population of medical students rather than practicing HCPs and the fact that all students were attending the same medical school at the University of Manchester.

Study	Taylor 2005[130]
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, thematic analysis using grounded theory approach.
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. 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?’

Study	Taylor 2005[130]
	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	**Barrier: Lack of support referral**
	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 applicability or methodological limitations.

Study	Woodward 1995[147]
Aim	To examine doctors' and patients' views on the risks and benefits of the symptomatic diagnosis of CFS.
Population	General practitioners recruited with the assistance of the Canberra branch of the Royal Australian College of General Practitioners (RACGP). N=20; male/female: 9/11 People diagnosed by doctors as having CFS N=50; male/female: 10/40; females mean age (range): 36.4 (13 to 64) years; males mean age (range): 39.2 (25 to 53) years.
Setting	Royal Australian College of General Practitioners (RACGP)
Study design	Two related investigations: qualitative interviews with GPs; a longitudinal study comprising three qualitative interviews with patients.
Methods and analysis	General practitioners were asked about their views on CFS and the difficulties it created for them in their practices.

Study	Woodward 1995[147]
	Patients were interviewed in-depth three times over a period of 2 years (1990-1992); they were asked to describe the characteristics of their illness over time, their history of medical investigations, the social consequences of their illness over time, their history of medical investigations, the social consequences of their illness and their own approaches to managing their illness. Interview schedules for both investigations were provided prior to the interview to allow participants time to reflect on their answers. At the interview, they could speak to their notes if they had made any (60% of the participants with CFS had done so) or discuss the questions in a less structured way. At the end of each interview, the schedule was reviewed to ensure that all questions had been addressed. Interviews were conducted and taped by one interviewer. They were later transcribed, coded and analysed. To ensure reliability and consistency in the coding process, several interviews were recorded some weeks after they were initially coded. Analysis of the qualitative data was facilitated by the use of a computer program designed for that purpose (NUDIST)
Findings	**Facilitator: HCP and patient communication/relationship building**
	Some doctors had found ways to manage the scientific uncertainties that were reported to surround CFS, by adopting a collaborative approach to providing care and said they were committed to working with patients' views about their health, indicating a desire to understand the world of their clients. They tried to develop a relationship with them, where all aspects of the persons' health might be discussed, including the alternative treatments they might seek. They dealt with their concerns about care and accommodated scientific uncertainty either by becoming 'case managers' or 'sounding boards' for their patients. They monitored health changes, gave emotional support and encouragement and passed on relevant research data or advice based on other patients' experiences. They reported they had not learned these responses in their medical training, but had learned to offer this sort of care through exposure to many patients with chronic illness, experience of illness either in themselves or in a family member, or by witnessing the changes due to an unexplained illness in a previously healthy patient.
Limitations and applicability of evidence	Serious methodological limitations due to the role of the researcher not being discussed, selection bias as participant recruitment is unclear, risk of bias in the data analysis since type of analysis and details are not provided. Minor concerns over applicability due to the age of the study in context of progression of views and treatment of ME/CFS.

Appendix E GRADE-CERQual tables

Summary of evidence: Information, education and support for health care professionals (stratum: adults/mixed or unclear age; mixed or unclear severity)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Health care professionals' awareness and knowledge of ME/CFS					
13	Semi-structured interviews and thematic analysis (9 studies), group discussions and thematic analysis (2 studies), mixed structured and semi-structured interviews and thematic analysis (1 study), mixed methods qualitative analysis of open-ended survey	There is need for increased training for HCPs to increase knowledge of ME/CFS and its management. Health care professionals (HCPs) often lack the knowledge or awareness to be able to diagnose and manage patients with ME/CFS. This often delays diagnosis and referral and means that patients can be mismanaged. This was expressed by both HCPs and people with ME/CFS. There is a need for improved education of HCPs about ME/CFS and an increased presence of the disease in the medical curriculum.	Limitations	Minor 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
	responses (1 study)				

Seven studies with minor to moderate issues; methodological limitations due to concerns over data analysis with data supported by single quotes in three studies (Devendorf 2017; Hannon 2012; Ryckeghem 2017) and lack of sufficient detail provided (Ryckeghem 2017), due to the potential influence of the researcher on the findings in one study where half of the participants were given a systematic review on the effectiveness of mental health interventions prior to the data collection (Raine 2004),. due to concerns over the recruitment strategy in one study where participants were selected through responses to an advertisement, therefore risking over-representation of students who are more informed or have stronger views on ME/CFS (Stenhoff 2015), due to concerns over participant recruitment with selection of HCP participants by ME/CFS patients in one study and concerns over data analysis with coding and analysis undertaken by a single researcher (Beaulieu 2000), 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); minor concerns over relevance due to participants of one study being a subset of a previous quantitative study who were self-identified as having ME/CFS rather than diagnosed according to accepted criteria (Devendorf 2018), due to the research aim driving the theme being different to that of the current review in three studies (Beaulieu 2000; Chew-Graham 2008; Devendorf 2017), due to participants having been previously recruited in a RCT in two studies (Chew-Graham 2008; Chew-Graham 2010), due to concerns over relevance of one study that was published prior to new guidelines and diagnostic criteria (Beaulieu 2000), due to concerns over the small and homogenous sample size and lack of representation of Health professionals in the sample of one study (Edwards 2007), due to the population of medical students all attending the same medical school rather than practicing HCPs in one study (Stenhoff 2015) and due to concerns over the applicability of one study conducted on the Belgian healthcare system to the NHS setting (Ryckeghem 2017)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Consensus on diagnostic criteria					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
4	Semi-structured interviews and thematic analysis (3 studies); mixed structured and semi-structured interviews and thematic analysis (1 study)	The lack of a confirmed consensus on the diagnostic criteria for ME/CFS meant that there was confusion among HCPs when consulted with symptoms. HCPs expressed the need for agreed case definitions for both diagnosis and recovery.	Limitations	Minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Three studies with minor to moderate issues; methodological limitations due to concerns over participant recruitment with selection of HCP participants by ME/CFS patients in one study and concerns over data analysis with coding and analysis undertaken by a single researcher (Beaulieu 2000) and due to concerns over data analysis with data often supported by single quotes in two studies (Devendorf 2017; Hannon 2012); minor concerns over relevance due to the findings in two studies being driven by the studies' original aim that differed from that of the current review (Beaulieu 2000; Devendorf 2017) and due to the contribution of an older study (Beaulieu 2000) potentially losing relevance (e.g. published prior to new guidelines and diagnostic criteria).

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Symptom measures					
2	Semi-structured interviews and thematic analysis (2 studies)	The lack of agreed tests and measurements for ME/CFS symptoms mean that HCPs are reluctant to make a diagnosis based on limited clinical signs and struggle to assess recovery.	Limitations	Very minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Two studies with minor issues; methodological limitations due to concerns over data analysis with data mostly supported by single quotes in one study (Devendorf 2017); minor concerns over relevance due to the aim of both contributing studies driving the theme being different to that of the current review (Chew-Graham 2008; Devendorf 2017) and due to participants of one study having been previously 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**
Clinical pathway					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
3	Semi-structured interviews and thematic analysis (2 studies), qualitative analysis of transcripts of facilitated group discussions (1 study)	There is need for a clearer clinical management pathway for ME/CFS. HCPs are often sure of where to refer patients once a diagnosis has been reached. ME/CFS specialists express concern at the lack of referrals to their services made by GPs.	Limitations	Minor methodological limitations	HIGH
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	No concerns about adequacy	

Two studies with minor issues; methodological limitations due to the potential influence of the researcher on the findings in one study (Raine 2004) and concerns over data analysis with findings mostly supported by single quotes in one study (Horton 2010) that were too minor to lower the confidence rating; concerns over the sample of one study consisting of people previously recruited in an RCT were too minor to lower the confidence rating (Chew-Graham 2010)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Training					
3	Semi-structured interviews and thematic analysis (2 studies)	HCPs highlighted the need for training in how to diagnose and manage ME/CFS, with a preference for an internet-based course. GPs suggested that ME/CFS specialist services should support GPs by providing them with information and training. There is currently little or no formal training on ME/CFS in the medical curriculum, with students claiming their knowledge often comes from media.	Limitations	Minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Three studies with minor issues; methodological limitations due to concerns over data analysis with findings mostly supported by single quotes in two studies (Hannon 2012; Horton 2010) and due to concerns over participant recruitment in one study (Stenhof 2015); very minor concerns over relevance due to the population of one study (Stenhof 2015) being medical students rather than practicing HCPs and the homogeneity of that population as all students were attending the same medical school at the University of Manchester, but no similar concerns in any of the other studies;

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Information resources					
2	Semi-structured interviews and thematic analysis (2 studies)	Some HCPs expressed the need for a resource that can be used during consultation to educate and reassure patients when diagnosed with ME/CFS, for example an online video resource. 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.	Limitations	Very minor methodological limitations	HIGH
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	No concerns about adequacy	

Two studies with minor issues; methodological limitations due to data analysis with findings mostly supported by single quotes in both contributing studies (Hannon 2012; Horton 2010), that were considered too minor to lower our overall confidence in the finding.

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Support from specialist services					
2	Semi-structured interviews and thematic analysis (2 studies)	There is seen to be a lack of communication between GPs and referral centres, with a need for increased feedback and sharing of information from specialist services. Specialist services need to be more visible and provide education and information for GPs.	Limitations	Moderate methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Two studies with minor to moderate issues; limitations due to concerns over data analysis with findings supported by single quotes in two studies (Horton 2010; Ryckenghem 2017) and due to the potential influence of the researcher on the findings not being discussed in one study (Ryckeghem 2017); minor concerns over relevance due to concerns over the applicability of one study that had been conducted in the Belgian health system to the NHS setting (Ryckeghem 2017).

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Information about support groups					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
3	Semi-structured interviews and thematic analysis (2 studies) mixed structured and semi-structured interviews and thematic analysis (1 study)	HCPs are often unable to recommend support groups because they had little knowledge or information about them.	Limitations	Minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Three studies with minor to moderate issues; methodological limitations due to concerns over participant selection and data analysis with coding and analysis by a single researcher in one study (Beaulieu 2000) and due to concerns over data analysis with some findings supported by single quotes in two studies (Hannon 2012; Horton 2010); very minor concerns about relevance due to information in one study being driven by the study's original research aim that differed from that of the current review (Beaulieu 2000)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Exposure to people with ME/CFS					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
3	Semi-structured interviews and thematic analysis (3 studies)	HCPs find that contact with people with ME/CFS outside of the clinical setting improved their understanding of the condition. For example, phone conversations or observation of patients living with ME/CFS in their daily lives allowed HCPs to make better understand symptoms and make decisions about management.	Limitations	Very minor methodological limitations	HIGH
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Two studies with minor issues; methodological limitations due to concerns over data analysis with some data supported by single quotes in one study (Horton 2010); very minor concerns over relevance due to the population of one study having been previously recruited in a RCT with a different aim to that of the present review (Chew-Graham 2008).

Summary of evidence: Information, education and support for health care professionals (stratum: children/young people)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Health care professionals' awareness and knowledge of ME/CFS					
2	Semi-structured interviews and thematic analysis (2 studies)	Health care professionals (HCPs) often lack the knowledge or awareness to be able to diagnose and manage children and young people with ME/CFS. This is supported by the opinions of both young people with ME/CFS and HCPs who care for them.	Limitations	Minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Two studies with minor issues; methodological limitations due to concerns over the small sample size and recruitment in both contributing studies (Jelbert 2010; Marks 2016); minor concerns over relevance due to the population of one study consisting of recovered patients whose views may differ from patients 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**
Consensus on diagnostic criteria					
1	Semi-structured interviews and thematic analysis (1 study)	HCPs caring for children and young people find difficulty in reaching a diagnosis of ME/CFS, with uncertainty around diagnostic criteria and appropriate labels for young people presenting with symptoms.	Limitations	Minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Minor concerns about adequacy	

One study with minor issues; methodological limitations due to concerns over the recruitment strategy (Marks 2016); minor concerns over adequacy due to support from a single study with a small sample size.

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Clinical pathway					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
1	Semi-structured interviews and thematic analysis (1 study)	For HCPs caring for children and young people there is uncertainty regarding appropriate and effective treatment pathways for patients after diagnosis.	Limitations	Minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Minor concerns about adequacy	

One study with minor issues; methodological limitations due to concerns over the recruitment strategy (Marks 2016); minor concerns over adequacy due to support from a single study with a small sample size.

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

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
1	Semi-structured interviews and thematic analysis (1 study)	HCPs caring for children and young people need standardised specialist training around ME/CFS to ensure that there is consistency across services.	Limitations	Minor methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Minor concerns about adequacy	

One study with minor issues; methodological limitations due to concerns over the recruitment strategy (Marks 2016); minor concerns over adequacy due to support from a single study with a small sample size

Barriers and facilitators

Summary of evidence, adults with ME/CFS, severity mixed or unclear

Study design and sample size			Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
Facilitator: Communication/ relationship between HCP and patient					
6	Semi-structured interviews and thematic analysis (4 studies); mixture of structured and semi-structured interviews and thematic analysis (1 study); combination of qualitative interviews with GPs and a longitudinal study comprising three qualitative interviews with patients (1 study)	Building a relationship between HCP and patient allows better provision of information and support, and frequent contact improves understanding of ME/CFS for both HCP and patient.	Limitations	Moderate concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	No concerns about adequacy	

Six studies with minor to serious issues; methodological limitations were due to role of the researcher not being discussed in four studies (Devendorf 2019, Horton 2010, Ryckeghem 2017, Woodward 1995), lack of data richness with findings mostly supported by single quotes in three studies (Devendorf 2019, Chew-graham 2008;), concerns over participant recruitment/selection in three studies (Ryckeghem 2017, Beaulieu 2000, Woodward 1995) and risk of bias from data analysis method in three studies (Beaulieu 2000, Ryckeghem 2017, Woodward 1995);
Moderate concerns over relevance that were due to indirect research aims of the individual studies with findings emerging driven by the study's original aims (Devendorf 2019, Chew-graham 2008, Beaulieu 2000), indirect population sample that had originally been recruited for a different study (Horton 2010, Chew-graham 2008), concerns over the applicability of one study (conducted in Belgium) to the NHS setting (Ryckeghem 2017) and because views reported in two studies could be considered dated due to time since publication (Woodward 1995; Beaulieu 2000)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Facilitator: Referral to specialist services					
3	Semi-structured interviews and thematic analysis (1 study); Semi-structured interviews and thematic analysis followed by theory-driven analysis (1 study); Focus groups and semi-structured interviews with thematic analysis (1 study)	Specialist services were seen as the best provider of information and support for people with ME/CFS, with referral to specialists providing a positive experience after a long road to diagnosis.	Limitations	Very minor concerns about methodological limitations	HIGH
			Coherence	No concerns about coherence	
			Relevance	Very minor concerns about relevance	
			Adequacy	No concerns about adequacy	

Two studies with very minor issues; methodological limitations due to the role of the researcher (De Carvalho Leite 2011, Bayliss 2016); very minor concerns about relevance because only one of three studies had minor concerns of applicability due to recruitment of sample from people completing ME/CFS treatment on the NHS who may have already received support and/or information (Broughton 2017)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Facilitator: Online training resource					
1	Semi-structured interviews and thematic analysis followed by theory-driven analysis (1 study)	HCPs valued online training resources that showed how to work with people with ME/CFS in a consultation setting, with several GPs finding video resources particularly useful for this.	Limitations	Very minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with very minor issues; methodological limitations due to the role of the researcher not being discussed,(Bayliss 2016) moderate concerns about coherence due the theme being found only in a single study (Bayliss 2016)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Limited specialist referral options					
1	Semi-structured interviews and thematic analysis followed by theory-driven analysis	While referral to specialists and other support services was seen a route to provide information and support, GPs often do not know when or where they should refer people with ME/CFS.	Limitations	Very minor concerns about methodological limitations	MODERATE
			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**
Barrier: Limited specialist referral options					
			Relevance	No concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with very minor issues; methodological limitations due to the role of the researcher, (Bayliss 2016); moderate concerns about adequacy with the theme being found in only one study

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Limited knowledge of support groups					
3	Semi-structured interviews and grounded theory approach (1 study); mixture of structured and semi-structured with thematic analysis (1 study); focus group interviews, open-ended questionnaires, progress notes, and a program evaluation questionnaire, with thematic analysis	HCPs often do not have sufficient information to be able to refer people with ME/CFS to support groups and can be hesitant to do so because of mixed beliefs about their effect on the patient.	Limitations	Minor 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**
	using grounded theory approach (1 study)				

Two studies with minor to moderate issues; methodological limitations due to concerns over participant selection and data analysis (coding and analysis by a single researcher) in one study (Beaulieu 2000) and due to role of the researcher and lack of data richness with findings mostly supported by single quotes in one study (Hannon 2012 and no concerns over the third study; minor concerns over relevance due to the indirect research aim of one study and its year of publication (2000) which preceded present guidelines and diagnostic criteria (Beaulieu 2000)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Lack of training					
2	Semi-structured interviews and inductive thematic analysis (1 study); semi-structured interviews and grounded theory approach (1 study)	There is a lack of training and education available for HCPs, GPs in particular, on how to manage people with ME/CFS, beginning with an absence of ME/CFS on the university medical curriculum.	Limitations	Minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	Minor concerns about adequacy	

Two studies with minor issues; methodological limitations due to potential selection bias in one study due to recruitment of participants through responses to an advertisement therefore risking over-representation of people who are more informed or have stronger views on ME/CFS (Stenhoff 2015), the role of the researcher and lack of data richness with findings mostly supported by single quotes (Hannon 2012); minor concerns over relevance due to the indirectness of the population of one study that included medical students rather than practicing HCPs (Stenhoff 2015); minor concerns over adequacy due to broadly-applicable theme based on two studies (Stenhoff 2015; Hannon 2012)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Reluctance of GPs to training					
1	Semi-structured interviews and thematic analysis followed by theory-driven analysis	Some GPs are reluctant to take on the management of people with ME/CFS, preferring to refer to secondary care specialists, and do not always engage with ME/CFS training when offered.	Limitations	Very minor concerns about methodological limitations	MODERATE
			Coherence	No concerns about coherence	
			Relevance	No concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with very minor issues; methodological limitations due to the role of the researcher not being discussed, (Bayliss 2016); moderate concerns about adequacy due to broadly-applicable theme being found only in a single study(Bayliss 2016)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Consultation time constraints					
1	Semi-structured interviews and thematic analysis followed by theory-	Due to the complexity of ME/CFS and its symptoms, HCPs often find that the nature of clinic time and short consultation lengths do not allow for effective communication and support.	Limitations	Very minor concerns about methodological limitations	MODERATE
			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
	driven analysis (1 study)		Relevance	No concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with very minor issues; methodological limitations due to the role of the researcher not being discussed (Bayliss 2016); moderate concerns about adequacy due to broadly-applicable theme being found only in a single study (Bayliss 2016)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	Design		Criteria	Rating	Overall assessment of confidence
Barrier: cognitive and physical functioning of ME/CFS patients and the impact on engaging with services					
1	Semi-structured interviews and thematic analysis	People with ME/CFS, particularly those with severe ME/CFS, have limited capacity to receive information, education and support from HCPs due to inability to communicate effectively.	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; methodological limitations due to the role of the researcher and concerns over data analysis with findings mostly supported by single quotes (Horton 2010); moderate concerns over adequacy due to broadly-applicable theme being found only in a single study (Horton 2010)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Information overload					
1	Semi-structured interviews and thematic analysis	People with ME/CFS can sometimes experience an overload of information during the care process which can negatively affect their understanding of the condition.	Limitations	Moderate concerns about methodological limitations	VERY LOW
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with moderate issues; methodological limitations due to the role of the researcher and concerns over data analysis with a lack of sufficient detail and findings mostly supported by single quotes (Ryckeghem 2017); moderate concerns over relevance due to setting (Belgian healthcare service); moderate concerns about adequacy to broadly-applicable theme being only briefly described in a single study (Ryckeghem 2017)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**		**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Fear of negative reactions					
2	Semi-structured interviews analysed by content analysis (1 study); mixture of structured and semi-structured interviews,	HCPs can be hesitant to provide information and discuss psychological factors around ME/CFS with patients due to concerns about patients' possible negative reactions.	Limitations	Moderate concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	

Study design and sample size			Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
	analysed by thematic analysis (1 study)		Adequacy	Minor concerns about adequacy	

Two studies with moderate issues; methodological limitations due to the: role of the researcher, lack of detail on data analysis method (Ax 1997), concerns over participant recruitment and data analysis with coding and analysis by a single researcher (Beaulieu 2000); minor concerns over relevance due to main emerging findings of one study (Beaulieu 2000) being driven by the study's original aims to explore multiple perspectives on stigmatization and legitimation of CFS and its year of publication (2000) which preceded present guidelines and diagnostic criteria but no concerns over the other contributing study; minor concerns over adequacy due to broadly-applicable theme based on two studies (Ax 1997; Beaulieu 2000)

Study design and sample size			Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Uncertainty and lack of confidence in information					
1	Mixture of structured and semi-structured interviews, analysed by thematic analysis	Uncertainties associated with ME/CFS mean that HCPs are often unsure about the reliability of information they have, making them cautious and hesitant when explaining the condition.	Limitations	Moderate concerns about methodological limitations	VERY LOW
			Coherence	No concerns about coherence	
			Relevance	Moderate concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with moderate issues; methodological limitations due to concerns over participant recruitment and data analysis (coding and analysis by a single researcher) (Beaulieu 2000); moderate concerns over relevance due to main emerging findings being driven by the study's original aims to explore multiple perspectives on stigmatization and legitimation of CFS (Beaulieu 2000) and the fact that the finding emerged from a single study that due to its year of publication (2000) preceded present guidelines and diagnostic criteria; moderate concerns over adequacy due to broadly-applicable theme being found only in a single study (Beaulieu 2000)

Summary of evidence, children and young people with ME/CFS, severity mixed or unclear

Study design and sample size			Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
Facilitator: Referral to specialist services					
1	Semi structured interviews with thematic analysis	Referral to specialist services provided children and young people with ME/CFS and their parents with information and support, as well as a letter allowing educational adjustments.	Limitations	Minor concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with minor issues; methodological limitations due to unclear relationship between the researcher and participants, and concerns over data richness with findings mostly supported by single quotes (Beasant 2014); minor concerns over relevance due to study's aim to understand the experiences of accessing as well as using a specialist service (some participants had not yet used the service) and unclear which intervention the findings relate to (Beasant 2014) and concerns over and the representativeness of the sample considering it consisted of feasibility RCT participants which may differ from eligible patients not recruited to a trial ; moderate concerns over adequacy due to broadly-applicable theme being found only in a single study (Beasant 2014)

Study design and sample size			Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
Facilitator: Digital social support					

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
1	Semi-structured interviews and thematic analysis	Digital social support websites such as health forums and other social media sites provide quick, simple and undemanding access to social support, reducing isolation.	Limitations	Minor concerns about methodological limitations	LOW
			Coherence	No concerns about coherence	
			Relevance	Minor concerns about relevance	
			Adequacy	Moderate concerns about adequacy	

One study with minor issues; methodological limitations due to the role of the researcher and lack of detail on the data analysis (Brigden 2018); minor concerns over relevance due to the study not including severely affected adolescents; moderate concerns over adequacy due to broadly-applicable theme being found only in a single study (Brigden 2018)

Study design and sample size		Finding	Quality assessment		
Number of studies contributing to the finding	**Design**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
Barrier: Unhelpful or unrelatable NHS information resources					
1	Semi-structured interviews and thematic analysis	NHS resources lack the accessibility and relatability provided by patient- and peer-led websites in terms of language and narrative approach used.	Limitations	Minor concerns about methodological limitations	LOW
			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**	**Finding**	**Criteria**	**Rating**	**Overall assessment of confidence**
			Adequacy	Moderate concerns about adequacy	

One study with minor issues; methodological limitations due to the role of the researcher and lack of detail on the data analysis (Brigden 2018); minor concerns over relevance due to the study not including severely affected adolescents; moderate concerns about adequacy due to broadly-applicable theme being found only in a single study (Brigden 2018)

Appendix F Excluded studies

Clinical studies

Table 10: Studies excluded from the qualitative review: information and support

Reference	Reason for exclusion
Aikman 1995[1]	Thesis, unable to obtain paper
Anderson 1997[2]	No relevant themes
Anderson 2012[4]	Incorrect study design (non-PICO systematic review)
Anderson 2014[3]	No relevant themes
Antcliff 2018[3]	No relevant themes
Asbring 2001[7]	Incorrect population (majority not ME/CFS)
Asbring 2002[7]	Incorrect population (included people with fibromyalgia – 13 FM, 12 ME/CFS)
Asbring 2004[7]	Incorrect population (included people with fibromyalgia – 13 FM, 12 ME/CFS)
Ashby 2006[10]	No relevant themes
Ax 1997[13]	No relevant themes
Ax 1998[12]	No relevant themes
Ax 2002[11]	No relevant themes
Bayliss 2014[14]	No relevant themes
Bayliss 2014[16]	Secondary analysis of already included study (Hannon 2012)
Bazelmans 2005[17]	No relevant themes; Incorrect study design
Beasant 2014[18]	No relevant themes
Bennett 2007[20]	No relevant themes
Brady 2016[21]	Incorrect population
Brigden 2018[22]	No relevant themes
Brooks 2012[23]	Incorrect study design (findings based on cross-sectional/questionnaire data); No relevant themes
Bulow 2003[25]	Insufficient ME/CFS diagnosis; Incorrect study design/analysis
Caplan 2001[26]	Narrative article
Chernow 2008[27]	Thesis, unable to obtain paper
Cheshire 2020[28]	No relevant themes
Chew-Graham 2011[29]	No relevant themes
Clarke 1999[32]	No relevant themes
Clements 1997[34]	No relevant themes
Costello 1998[35]	Thesis, unable to obtain paper
Davison 1997[36]	No relevant themes
De Carvalho 2011[37]	No relevant themes
De Silva[38]	Secondary analysis of an already included study (Hannon 2012)
Dennison 2010[39]	No relevant themes
Donalek 2009[43]	No relevant themes

Reference	Reason for exclusion
Drachler 2009[44]	Incorrect study design (non-PICO systematic review)
Everett 2002[46]	Incorrect population (secondary school teachers)
Fisher 2013[47]	No relevant themes
Fowler 2005[48]	Incorrect study design (quantitative analysis, no themes)
Friedberg 1998[50]	Book chapter, not available
Friedberg 2016[49]	Incorrect population (majority had 'unexplained chronic fatigue'; Emphasis on quantitative analysis; No relevant themes
Gan 2010[51]	Incorrect population
Gilje 2008[52]	No relevant themes
Gotts 2016[53]	No relevant themes
Gray 2003[54]	No relevant themes
Guise 2007[56]	No relevant themes
Guise 2010[55]	Incorrect study design/analysis; No relevant themes
Hareide 2011[58]	No relevant themes
Harris 2016[59]	Incorrect study design (non-PICO systematic review)
Harris 2017[60]	No relevant themes
Hart 2000[61]	No relevant themes
Higginson 2008[62]	Incorrect population
Horrocks 2015[63]	Book chapter, not available
Horton-Salway 2002[64]	Article; Incorrect study design/analysis
Horton-Salway 2004[65]	Article; Incorrect study design/analysis
Jason 2015[67]	Article
Jensen 2001[69]	Thesis, unable to obtain paper
Keech 2015[70]	Incorrect study design/analysis; No relevant themes
Kendrick 2016[71]	Incorrect study design; No relevant themes
Kisely 2002[72]	Incorrect study design (evaluation of web-based information)
Larun 2007[74]	Incorrect study design (non-PICO systematic review)
Larun 2011[73]	No relevant themes
Lee 2000[75]	Thesis, unable to obtain paper
Lee 2001[76]	Incorrect population (sample described as chronic fatigue and weakness)
Levine 1997[77]	Incorrect study design/analysis
Lian 2016[78]	No relevant themes
Lingard 2014[80]	No relevant themes
Littrell 2012[81]	Thesis, unable to obtain paper
Lombaard 2005[82]	No relevant themes
Lovell 1999[83]	No relevant themes
McDermott 2011[86]	No relevant themes
McInnis 2015[87]	Incorrect population
Mihelicova 2016[88]	No relevant themes
Missen 2012[89]	No relevant themes

Reference	Reason for exclusion
Moore 2000[90]	Incorrect study design (combined statistical and thematic analysis, not reported as qualitative)
Njolstad 2019[92]	No relevant themes
Olson 2015[93]	No relevant themes
Ong 2005[94]	No relevant themes
Parslow 2015[95]	No relevant themes
Parslow 2017[97]	Incorrect study design (non-PICO systematic review)
Parslow 2017[98]	No relevant themes
Pemberton 2014[99]	No relevant themes
Pemberton 2014[100]	No relevant themes
Picariello 2017[101]	No relevant themes
Pinxsterhuis 2015[104]	Incorrect study design (non-PICO systematic review)
Pinxsterhuis 2015[103]	No relevant themes
Prins 2000[105]	Incorrect study design/analysis (no thematic analysis)
Ray 1995[108]	Incorrect study design (quantitative, questionnaires)
Ray 1998[107]	No relevant themes
Reme 2013[109]	No relevant themes
Reynolds 2006[111]	No relevant themes
Reynolds 2008[112]	No relevant themes
Reynolds 2010[110]	Incorrect population; No relevant themes
Richards 1998[114]	Incorrect study design (quantitative, questionnaires)
Richards 2006[113]	No relevant themes
Sidi-Ali-Mebarek 2009[119]	Thesis, unable to obtain paper
Snell 2001[120]	Incorrect study design (case study)
Soderlund 2000[122]	No relevant themes
Soderlund 2005[121]	Incorrect study design (results combined with quantitative data)
Stormorken 2015[125]	No relevant themes
Sturge-Jacobs 2002[126]	Incorrect population
Swoboda 2006[128]	Incorrect population
Taylor 2017[129]	No relevant themes
Tevens 2004[131]	Thesis, unable to obtain paper
Theorell 1999[132]	Incorrect study design (quantitative, questionnaires)
Travers 2008[133]	No relevant themes
Tuck 1998[134]	No relevant themes
Tuck 2000[135]	Incorrect study design (quantitative, questionnaires)
Velleman 2016[136]	No relevant themes
Ward 2008[137]	No relevant themes
Ware 1998[139]	No relevant themes
Whitehead 2006[142]	No relevant themes
Williams 2016[144]	No relevant themes

Reference	Reason for exclusion
Winger 2014[146]	No relevant themes

Table 11: Studies identified but not extracted due to saturation

Reference
Arrol 2008[6]
Clarke 2000[33]
McCue 2004[85]
Pinikahana 2002[102]
Schoofs 2004[118]
Whitehead 2006[143]

Table 12: Studies excluded from the qualitative review: barriers and facilitators to providing information and support

Reference	Reason for exclusion
Aikman 1995[1]	Thesis, unable to obtain paper
Anderson 2014[3]	No relevant themes
Anderson 1997[2]	No relevant themes
Anderson 2012[4]	Incorrect study design (non-PICO systematic review)
Antcliff 2016[5]	No relevant themes
Arroll 2008[6]	No relevant themes
Asbring 2001[7]	Incorrect population (majority not ME/CFS)
Asbring 2004[8]	Incorrect population (included people with fibromyalgia – 13 FM, 12 ME/CFS)
Asbring 2002[9]	Incorrect population (included people with fibromyalgia – 13 FM, 12 ME/CFS)
Ashby 2006[10]	No relevant themes
Ax 2002[11]	No relevant themes
Ax 1998[12]	No relevant themes
Bayliss 2014[16]	Secondary analysis of already included study (Hannon 2012)
Bayliss 2014[14]	No relevant themes
Bazelmans 2005 [17]	No relevant themes; Incorrect study design
Bennett 2007[20]	No relevant themes
Brady 2016[21]	Incorrect population

Reference	Reason for exclusion
Brooks 2013[23]	Incorrect study design (findings based on cross-sectional/questionnaire data); No relevant themes
Bulow 2003[25]	Insufficient ME/CFS diagnosis; Incorrect study design/analysis
Caplan 2001[26]	Narrative article
Chernow 2008[27]	Thesis, unable to obtain paper
Cheshire 2020[28]	No relevant themes
Chew-Graham 2011[29]	No relevant themes
Chew-Graham 2010[30]	No relevant themes
Clarke 1999[32]	No relevant themes
Clarke 2000[33]	No relevant themes
Clements 1997[34]	No relevant themes
Costello 1998[35]	Thesis, unable to obtain paper
Davison 1997[36]	No relevant themes
Dennison 2010[39]	No relevant themes
De Silva 2013[38]	No relevant themes
Devendorf 2017[41]	No relevant themes
Devendorf 2018[42]	No relevant themes
Donalek 2009[43]	No relevant themes
Drachler 2009[44]	Incorrect study design (non-PICO systematic review)
Edwards 2007[45]	No relevant themes
Everett 2002[46]	Incorrect population (secondary school teachers)
Fisher 2013[47]	No relevant themes
Fowler 2005[48]	Incorrect study design (quantitative analysis, no themes)
Friedberg 1998[50]	Book chapter, not available
Friedberg 2016[49]	Incorrect population (majority had 'unexplained chronic fatigue'; Emphasis on quantitative analysis; No relevant themes
Gan 2010[51]	Incorrect population
Gilje 2008[52]	No relevant themes
Gotts 2016[53]	No relevant themes
Gray 2003[54]	No relevant themes

Reference	Reason for exclusion
Guise 2010[55]	Incorrect study design/analysis; No relevant themes
Guise 2007[56]	No relevant themes
Hareide 2011[58]	No relevant themes
Harris 2016[59]	Incorrect study design (non-PICO systematic review)
Harris 2017[60]	No relevant themes
Hart 2000[61]	No relevant themes
Higginson 2008[62]	Incorrect population
Horrocks 2015[63]	Book chapter, not available
Horton-Salway 2002[64]	Article; Incorrect study design/analysis
Horton-Salway 2004[65]	Article; Incorrect study design/analysis
Jason 2015[67]	Article
Jelbert 2010[68]	No relevant themes
Jensen 2001[69]	Thesis, unable to obtain paper
Keech 2015[70]	Incorrect study design/analysis; No relevant themes
Kendrick 2016[71]	Incorrect study design; No relevant themes
Kisely 2002[72]	Incorrect study design (evaluation of web-based information)
Larun 2011[73]	No relevant themes
Larun 2007[74]	Incorrect study design (non-PICO systematic review)
Lee 2000[75]	Thesis, unable to obtain paper
Lee 2001[76]	Incorrect population (sample described as chronic fatigue and weakness)
Levine 1997[77]	Incorrect study design/analysis
Lian 2016[78]	No relevant themes
Lin 2009[79]	No relevant themes
Lingard 2014[80]	No relevant themes
Littrell 2012[81]	Thesis, unable to obtain paper
Lombaard 2005[82]	No relevant themes
Lovell 1999[83]	No relevant themes
Marks 2016[84]	No relevant themes
McCue 2004[85]	No relevant themes

Reference	Reason for exclusion
McDermott 2011[86]	No relevant themes
McInnis 2015[87]	Incorrect population
Mihelicova 2016[88]	No relevant themes
Missen 2012[89]	No relevant themes
Moore 2000[90]	Incorrect study design (combined statistical and thematic analysis, not reported as qualitative)
Njolstad 2019[92]	No relevant themes
Olson 2015[93]	No relevant themes
Ong 2005[94]	No relevant themes
Parslow 2017[97]	Incorrect study design (non-PICO systematic review)
Parslow 2017[98]	No relevant themes
Parslow 2015[95]	No relevant themes
Parslow 2018[96]	No relevant themes
Pemberton 2014[100]	No relevant themes
Pemberton 2014[99]	No relevant themes
Picariello 2017[101]	No relevant themes
Pinikahana 2002[102]	No relevant themes
Pinxsterhuis 2015[104]	Incorrect study design (non-PICO systematic review)
Pinxsterhuis 2015[103]	No relevant themes
Prins 2000[105]	Incorrect study design/analysis (no thematic analysis)
Raine 2004[106]	No relevant themes
Ray 1995[108]	Incorrect study design (quantitative, questionnaires)
Ray 1998[107]	No relevant themes
Reme 2013[109]	No relevant themes
Reynolds 2010[110]	Incorrect population; No relevant themes
Reynolds 2006[111]	No relevant themes
Reynolds 2008[112]	No relevant themes
Richards 1998[114]	Incorrect study design (quantitative, questionnaires)
Richards 2006[113]	No relevant themes
Sachs 2001[116]	Incorrect study design; No thematic analysis

Reference	Reason for exclusion
Saltzstein 1998[117]	Incorrect study design (reported quantiatively)
Schoofs 2004[118]	Incorrect population (ME/CFS and fibromyalgia)
Sidi-Ali-Mebarek 2009[119]	Thesis, unable to obtain paper
Snell 2001[120]	Incorrect study design (case study)
Soderlund 2000[122]	No relevant themes
Soderlund 2005[121]	Incorrect study design (results combined with quantitative data)
Son 2015[123]	No relevant themes
Stormorken 2015[125]	No relevant themes
Sturge-Jacobs 2002[126]	Incorrect population
Sunnquist 2017[127]	Incorrect study design (survey reported quantitatively)
Swoboda 2006[128]	Incorrect population
Tevens 2004[131]	Thesis, unable to obtain paper
Taylor 2017[129]	No relevant themes
Theorell 1999[132]	Incorrect study design (quantitative, questionnaires)
Travers 2008[133]	No relevant themes
Tuck 2000[135]	Incorrect study design (quantitative, questionnaires)
Tuck 1998[134]	No relevant themes
Velleman 2016[136]	No relevant themes
Ward 2008[137]	No relevant themes
Ware 1998[139]	No relevant themes
Ware 1999[140]	No relevant themes
Ware 1993[138]	No relevant themes
Webb 2011[141]	No relevant themes
Whitehead 2006[143]	No relevant themes
Whitehead 2006[142]	No relevant themes
Williams 2016[144]	No relevant themes
Wilson 2011[145]	Incorrect population (experiencing chronic fatigue due to other long-term conditions)
Winger 2014[146]	No relevant themes

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