

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

Appendix 1: Involving children and young people in developing a NICE guideline on myalgic encephalomyelitis / chronic fatigue syndrome: diagnosis and management

NICE guideline NG206

Oxford Clinical Allied Technology and Trial Services Unit

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Final

*This guideline was developed by the
National Guideline Centre*

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1 Involving children and young people

An integral part of developing NICE guidelines is the involvement of people with direct experience of the condition and all guideline committees have lay members. The ME/CFS guideline committee has 5 lay members, including 2 parents with experience of children and young people with ME/CFS and 1 young person. However, during the scoping process it was identified there was limited published evidence directly from the children's and young people's perspective. For this topic it was considered crucial that the experiences, perspectives and opinions of children and young people with ME/CFS should inform the guideline and interviews were conducted with children and young people.

The consultation was commissioned by NICE and carried out by the Oxford Clinical allied technology and trial services unit.

The Oxford Clinical allied technology and trial services unit was awarded the commission after an open tender process. An invitation to tender was sent to the guideline registered stakeholders and posted on the Royal College of Physicians website. Applicants had to submit a written proposal outlining how they met the research brief, submitted bids were shortlisted and shortlisted bidders were then interviewed by telephone. The process was led by the National Guideline Centre (NGC) and overseen by a subgroup from the ME/CFS guideline committee. The subgroup comprised of the ME/CFS guideline chair, vice chair, the NGC guideline lead, two lay members and representation from the NICE Patient and Public Involvement unit. The subgroup shortlisted the bids, interviewed the shortlisted bidders and awarded the tender.

See Section 2 for the report and sections 3 and 4 for how the committee used the report to support their decision making.

2 Report

2.1 Abstract

2.1.1 Background

A new guideline for Myalgic encephalomyelitis (or encephalopathy)/chronic fatigue syndrome (ME/CFS): diagnosis and management is currently being developed by the National Institute for Health and Care Excellence (NICE) to inform health and social care practice in England. It is essential that NICE guidelines are informed by the views of those receiving care. This report presents findings from a consultation with young people affected by ME/CFS. The consultation aimed to explore the views and experiences of young people on the following topics identified from the guideline scope (add reference): Identification and assessment before diagnosis, diagnosis of ME/CFS, management of ME/CFS, monitoring and review, information, education, and support for people with suspected or diagnosed ME/CFS and their families and carers, and information, education and support for health and social care professionals.

The consultation was commissioned by NICE and carried out by the Oxford Clinical allied technology and trial services unit: Kim Chapman, Sophie Lawrie, Dr Shelly Coe, Sheera Sutherland, Professor Helen Dawes (Oxford Brookes University), Dr Andrew Soundy (University of Birmingham).

2.1.2 Methods

A qualitative approach using hermeneutic phenomenology and situated within a subtle realistic world view was undertaken. The consultation comprised qualitative thematic analysis of data collected from 12 interviews and 3 online surveys completed between September and October 2019. A total of 16 young people took part (10 female, 6 male), ranging in age from 11 to 18 years. A topic guide comprising the pre-defined themes was used to structure the sessions, which consisted of discussion points. Data collection was informed by a pilot consultation group of young people. Key findings were shared with all participants.

2.1.3 Findings

The views expressed often reflected the individual nature of the condition severity and age of participants, but core themes emerged from interviews and included the patient journey to diagnosis and subsequent development of management and coping strategies, as well as concerns of a limited understanding of the illness and its management from the HCPs involved in their care pathway. A number of the themes highlighted potentially modifiable factors, both positive and negative, that could be addressed within the care pathway to improve the care of young people with ME/CFS. Factors were physical, psychological, contextual and social, and included support and information for young people with ME/CFS and HCPs for diagnosis and management, support for HCPs in how to better interact with patients and family members and bespoke approaches to management.

2.2 Background

Myalgic encephalomyelitis (ME) and Chronic Fatigue Syndrome (CFS) are serious and chronic, debilitating conditions characterised by immune, neurological and cognitive impairment, sleep abnormalities, and autonomic dysfunction, resulting in significant functional impairment accompanied by a pathological level of fatigue. The impacts of ME/CFS are far-reaching and have considerable personal, social and economic consequences affecting both individuals living with ME/CFS as well as their families.

More widely researched in adults, there is little research in children and young people due to difficulties in diagnosis, discussed in the commentary provided by Geraghty et al., 2018. An earlier consultation designed and undertaken by children and young people, their parents and health and educational professionals was facilitated by the UK charity Action for M.E. The findings indicated high levels of distress and hopelessness across the sample.

2.3 Methods

2.3.1 Consultation aims

This consultation was commissioned by NICE as part of their work to develop a new guideline for Myalgic encephalomyelitis (or encephalopathy)/chronic fatigue syndrome (ME/CFS).

The aim of this consultation was to recruit and facilitate focus groups of young people who have been affected by ME/CFS so that their perspectives informed the development of the new guideline. The key aspect of the focus groups was to allow children and young people with ME/CFS to provide insight about their perspectives on specific questions and issues identified by both the committee based on the guideline scope and themselves. The following topics were identified from the guideline scope (add reference): Identification and assessment before diagnosis, diagnosis of ME/CFS, management of ME/CFS, monitoring and review, information, education, and support for people with suspected or diagnosed ME/CFS and their families and carers, and information, education and support for health and social care professionals.

Specific requirements set out by committee included a geographical spread across England and to include young people with severe ME/CFS to ensure their voices were heard.

2.3.2 Consultation design

The consolidated criteria for reporting qualitative research (COREQ; Tong et al, 2007) was used to frame the methods section of this report. A qualitative approach using hermeneutic phenomenology and situated within a subtle realistic world view was undertaken. This paradigmatic stance focuses on the common realities expressed by individuals. The view point recognizes that an individual has unique experiences however it also identifies that across a number of people common realities are present. The focus of this study was to identify the common realities.

Reflexivity

Author SL undertook all interviews. She was a female in her 20's who was not known to the participants except for information gained regarding the study and an understanding that she was a researcher from Oxford Brookes University.

2.3.3 Sampling

Eligibility criteria

The following inclusion criteria was adopted; (a) children/young people between the ages of 9-18 years, (b) having the capacity to provide assent/consent assessed by the researcher, and (c) People with ME/CFS often have co-morbid entities and for this study were acceptable for inclusion. Common comorbidities include conditions such as fibromyalgia, myofascial pain, temporomandibular joint syndrome, irritable bowel syndrome (IBS), Raynaud's phenomenon, depression, migraine, allergies, and sicca syndrome.

The following exclusion criteria was adopted; (a) unable to communicate or participate safely (as determined by their parent/carer), (b) unwilling to provide assent, were too severely affected by their condition such that taking part could cause significant long-term detriment to their health, (c) had serious thoughts of self-harm or suicide (as reported by parent/carer), and (d) presented with primary psychiatric disorders or substance abuse (as reported by parent/carer).

Sample size

Several considerations were made in regard to determining the sample size. We identified that the study aim was focused on specific experiences and the group of individuals were from a relatively heterogeneous group of young people. Because of the unique nature of the experiences and the similarity of expressions and the quality of dialogue following the first five interviews we identified that a lower number of participants was required to achieve data saturation of theme (Malterud et al., 2016). This approach was consistent with Guest et al., (2006) who stated that twelve interviews in a group of relatively heterogeneous individuals should be enough to achieve data saturation. With this in mind, we continued to interview 14 children/young people and were able to achieve data saturation within a specific number of themes.

2.3.4 Recruitment and procedures

We aimed to conduct three to four focus groups comprising of no more than four young people. The locations were undefined at the time of recruitment to allow for flexibility and accommodate the needs of the participants. Age appropriate recruitment materials were prepared. This included participant information sheet (PIS), assent/consent form and data privacy notice for 9-15year olds and 16-18year olds and PIS for the parent/carer (Appendix 6.1). An animated recruitment notice video was also created.

Feedback from the Advisory Group suggested that many young people, in particular those with severe ME/CFS would be unable and/or unwilling to travel to central locations. We therefore planned to hold individual interviews with those who expressed an interest in taking part but declined to attend a focus group.

As a partner organisation, Action for ME distributed the PIS and data privacy notice via email to their member directory, these were also posted to the Action for ME website and via their social media page (Facebook). In accordance with the General Data Protection Regulation (GDPR), a gatekeeper letter was provided by Action for ME to confirm that members had provided permission to be sent such material. As gatekeeper letters could not be obtained from additional charities, Action for ME was the only organisation involved in the recruitment process. Young people and their parent/carer were required to contact research team member SL via telephone or email to express their interest in taking part. During a pre-arranged call, SL answered any questions about the consultation, confirmed eligibility and that the PIS had been reviewed at least 48 hours prior to the call. A further call was arranged if less than 48 hours had elapsed to allow ample time for PIS review. Verbal informed assent/consent was received by SL during the telephone call and documented (Appendix

6.2). The assent/consent forms were signed and dated (including the time) by SL then posted to participants for them to sign, date and return in a stamped addressed envelope provided. Any forms with an incomplete signature or date were sent back with a request to complete fully. Young people under the age of 16 completed the assent forms and their parent/carer provided consent on their behalf.

At the time of obtaining consent, SL discussed the consultation options with participants and their parent/carer. During the same call demographic information was collected (age, gender, ME/CFS severity (reported by parent/carer) as well as contact information (address, email and telephone number).

SL received assent/consent from 16 young people to take part in the consultation. No focus groups were conducted. Individual interviews were planned to take no longer than 60 minutes. All participants were offered the chance to pause or stop the interview at any time and resume at a time convenient to them and a parent/carer was present in all interviews where requested. All interviews were audio recorded and transcribed for analysis. In total 12 interviews and 3 online surveys were completed between September and October 2019. One young person withdrew from the consultation without taking part in any form of interview due to the severity of their symptoms (severe).

2.3.5 Data collection tool and development

The committee provided pre-defined themes for the consultation based on the guideline scope.. The themes were shared among the Advisory Group for review and discussion. A guide was created by the Advisory Group, which consisted of questions addressing each theme as well as possible prompts for SL to use to fully explore the participant's thoughts and experiences. For several of the themes the questions and prompts were made age appropriate (9-15 years old; 16-18 years old).

During the consenting process participants were offered the opportunity to take part in a focus group or individual interviews. The preference for individual interviews was overwhelming therefore no focus groups were undertaken. The online survey was offered as a third option for those participants who were unable to take part in either focus group or individual interviews.

Prior to recruitment, the interview guide was piloted in several young people. This was achieved by undertaking a single cognitive interview (speak aloud) as a pilot of the interview schedule. This ensured that the questions asked were understood as intended by the recipient. Modifications were made to the questions and prompts where necessary. The final interview schedule has 5 themes and 16 questions. See appendix 6.3.

2.3.6 Data analysis

The data was analysed for thematic content, which fit the selected methodology. This involved six steps: (1) transcribing the data, (2) generating initial codes, (3) searching for themes, (4) reviewing themes, (5) defining and naming themes and (6) producing the report.

In order to minimise bias and as a quality assurance method Phases 1-3 involved blinded analysis to ensure a quality check of analysis. This ensured the most common themes were extracted and agreement was made at this stage regarding the definition and scope of each theme. The first five transcripts were read by three members of the team (SC, AS, SS) who then discussed the transcripts and agreed on a coding framework to organise the data and to facilitate analysis of themes within and between transcripts for the following interviews. There were no instances where an agreement was not met between the researchers.

2.3.7 Trustworthiness and Rigor

Quality enhancement strategies were employed that reflected a world view including reflexivity, a clear exposition of methods including an audit trail (available upon request) and negative case analysis (Pope and Mays, 2000). Further analysis followed as indicated by Morse (2015). These include; (a) negative case analysis presented first to AS then to the steering group to establish criticality within themes, (b) peer debriefing occurring as the researchers defended cases, (c) a presentable audit trail for the steering group, (d) triangulation across different levels including theory and investigator and finally (e) clarifying researcher bias across the three main sources. To improve robustness, codes, themes and subthemes were discussed within the research team (HD, KC).

2.3.8 Ethical considerations

The purpose of the consultation was to explore the thoughts and experiences of children and young people living with ME/CFS to inform the new guideline. The consultation involved recruitment through third party partner Action for ME and involved interviews and an online survey. Through use of the Health Research Authority (HRA) decision tool <http://www.hra-decisiontools.org.uk/research/> and further communication with the HRA the consultation was not deemed to be research. However, due to its sensitive nature, this project and documentation was reviewed by the Oxford Brookes University Research Ethics Committee (UREC) through expedited process, reference E19022. The approved document can be provided by the lead author.

2.4 Findings

2.4.1 Participant characteristics

Participants of the consultation included ten females and six males. This is consistent with data that suggests higher prevalence of ME/CFS in females. The age of the participants ranged from 11-18 years of age. All participants had received a diagnosis of ME/CFS and reported none of the excluded medical conditions. The reported severity of young people included: mild (1), mild/moderate (3), moderate (3), moderate/severe (3), and severe (6). Overall, the consultation met the geographical spread requirement with one young person taking part from each of the following postcode district (denoted by the postcode prefix): Brighton, Bristol, Chelmsford, Chester, Gloucester, Hemel, Hull, Ipswich, Kent, Northampton, Oldham, Reading, Southampton, Stockport, Swindon, and Telford.

2.4.2 Thematic framework and consultation findings

A total of three themes were identified.

2.4.3 Theme 1: The patient journey to diagnosis

This theme represents the experiences of the illness detailed by individuals before diagnosis. It reflects the challenges faced and illustrates the common symptoms experienced and the subsequent process of identifying a diagnosis with a focus on interactions and engagement with health care professionals. Two sub-themes were included in this; (a) early experiences and symptoms and (b) development of symptoms.

Sub-theme 1.1: Early experiences and symptoms

This sub theme describes the experiences of symptoms very early on to illustrate the participant's memories of how the illness began to present itself. Early experiences were identified as resolvable short-term illnesses. Examples of this would include the identification of a cold, pain, headache or fatigue. Initially participants would gradually distinguish the

experience from a typical or known experience or events which were understood, to identifying them as something that was unknown. Despite this, the symptoms were assumed and treated as something common which would end. Investigation and treatment related action was limited because individuals perceived that symptoms were only a short term event and would finish soon.

Symptoms were differentiated as different to the normal presentation because they would continue for longer. Individuals identified that the symptoms would linger. For instance, ME4 described the experience as: “Just kind of being drained, not feeling revived by sleep”. The most noticeable and distinguishing symptom was feeling tired or drained and ME7 stated “as it went on I started to get more and more sick, it felt less like a flu or a cold kind of thing, I just felt completely drained.”. ME10 stated “Well I got a headache, and then things just got worse, like I started feeling dizzy and sick and ... I was sick quite a few times the first few, I don’t know months or year”.

One experience mentioned but not by all, was feeling angrier or increased loss of temper (ME5).

ME11 stated: “So I think I’ve been ill since I was a baby, but I only started like really like recognising it when I was 12, and I got ill with glandular fever – I never really got any better. So like I would come home from school and just sleep, and I’d be falling behind on my schoolwork, and I’d just sit in lessons and I couldn’t like do the work cos I couldn’t think because my brain was so foggy ... and then I started getting really bad pains in my arms and legs”.

Sub-theme 1.2: Development of symptoms

This sub-theme considered the worsening of symptoms, as participants began to associate what was happening with a more serious illness. The time taken to appreciate the illness is reflected by ME11 who recognised the symptoms and illness from much earlier in life but only after experiencing glandular fever did a diagnosis come “*I’ve been ill since I was a baby, but I only started like really like recognising it when I was 12, and I got ill with glandular fever – I never really got any better.*” The descriptions from participants focused on an increase in symptoms by length of time and frequency of experiences, as well as intensity of experiences. This experience is articulated by ME10 who stated “*I got a headache, and then things just got worse, like I started feeling dizzy and sick and ... I was sick quite a few times the first few, I don’t know months or year.*”

The understanding was limited by individuals not having a diagnosis quickly. Over a longer time period individuals would experience progressive or more devastating symptoms, linked to more serious illness like flu or glandular fever e.g., “Like three weeks [with the symptoms], and then I felt like really bad, and then I discovered I had pneumonia after that time” (ME2). The understanding of experiences and the process and how to manage it was difficult initially for all participants. For instance, ME5 stated “it felt like my body was suddenly made of bricks or lead – it took a lot more energy for me to move. My head just felt really inflamed and kind of achy all the time. I couldn’t concentrate for long enough, I lost track of my words and what I was saying”. The impact is clearly identified by ME13 who stated: “*My whole life is affected all the time. I don’t go to school. I can’t go out with my friends. I can only ever see 1 friend at a time and for only half an hour. Then I can’t see anyone for a long time because afterwards it makes me worse.*”

Symptoms frequently and progressively impacted participants’ meaningful activities and relationships. Most commonly this was reflected by increased periods of time off school and not being able to see friends. The primary reason for this was due to feeling too tired. The impact of the ME was clearly articulated by ME15: “*Every single thing, like school, friendships, relationships, like every single part of my life...there was a time where I couldn’t walk to school, I’d take a taxi, or you know I would go on a scooter so I didn’t have to walk.*”

And when I was really really severely affected with ME I couldn't go to the toilet and I couldn't get out of bed, and I would have my meals in bed - so I couldn't go downstairs and... eat."

Often the school concerned would initially require the participant to attend lessons or restrict the ability to rest, this is highlighted by participant ME2 stated: *"There was people that were trying to help me because I was ill, but then when I said I wanted to have a break and I wanted to go home... they weren't taking me back to the office. They would say oh wait 5 minutes, and then they kept on saying that, and then I just couldn't handle it"*.

Participants identified a need for earlier diagnosis, the main reason for this was to reduce the extreme experiences of symptoms. Some expressed anger and lack of management due to lack of knowledge from health care professionals about what happened. Health care professionals would obtain a diagnosis most often through a process of elimination. For instance, that the illness was not abdominal migraines or the flu. HCPs identified a need often to refer to other specialties when a diagnosis could not be obtained. Limited advice from HCPs meant that some individuals began to consider the condition themselves for instance ME3 identified *"I didn't really get much kind of information or support [from HCPs] ...most of my information and knowledge about my condition came from the research that my mum was doing"*

2.4.4 Theme 2: Factors which negatively influence coping

During the patient journey to diagnosis, participants identified several factors which would influence coping. Four sub-themes were identified; (a) HCPs limited understanding of the illness, (b) interactions which prevented management and coping, (c) the nature and intensity of symptoms and subsequent needs, and (d) ineffective experiences of treatment and management.

Sub-theme 2.1: Health Care Professionals (HCPs) limited understanding of the illness

Before diagnosis individuals identified a lack of advice or reassurance from HCPs that was able to support the individual in coping and managing their condition. It was identified across participants that HCPs required time to work out what advice to provide. For instance, ME4 stated: *"they obviously were kind of interested in what I was going through, but they didn't ever give me any kind of advice or any kind of reassurance."* Multiple visits to HCPs were often required just to convince them there was a problem and ensure further investigation was undertaken. This required a process of repeated explanation of symptoms and was often in itself a tiring experience. Some individuals identified that their diagnosis occurred after a year or 18 month wait. In one case and at the time of the interview ME2 had privately undertaken blood tests in Germany which provided a diagnosis but they did not have that diagnosis in the UK and therefore could not be treated for it. ME15 has a similar experience in the delay of diagnosis and stated *"3 ½ months after I started feeling really ill I got the diagnosis privately. But then 7 months after I'd gotten ill I got the diagnosis by NHS – so it did take quite a long time for the NHS to diagnose me with ME or CFS."* The implication of not having a diagnosis would impact on the individual's well-being. For instance, ME7 stated: *"I wasn't diagnosed I couldn't really access any of the help or support or gain a better understanding of what was happening to me, because I wasn't diagnosed."*

At worst the interactions between the participant and HCP could deny the experiences reported by participants. This could be undertaken in different ways including;

(a) An inability to recognise or acknowledge experiences or provide empathy for participants. For instance, ME6 stated: *"while they may not have been able to give me any support and make me feel better, I wish they could have said 'Right, you're in pain, and I understand, and we don't really know what to do about it' - but all they ... seem to be in denial about what I was going through."* And the impact of this created anguish for participants for instance ME14 stated: *"The specialist person didn't listen to me, she even wrote things that weren't*

right, she was always trying to make things sound better than they were. She made me feel bad, I hated going.”

(b) coming back to a particular diagnosis which was wrong or being taken from one HCP to another without progress. For instance, ME7 stated: *“so he (health care professional) was very kind of stuck on the thing that it was kind of just a flu-like thing, trying to put it down to something else”*. The impact of being moved around different HCPs had a significant impact on participants and meant some felt abandoned by the health service. This was demonstrated by ME9 who stated: *“We were getting unsatisfactory response from our GP – they basically fobbed us off. Even after it was shown that it wasn’t an anxiety related issue, they still didn’t provide us with an answer for it... we were cut adrift effectively and ended up fending for yourselves - and as a result we were totally in control and had no support...we were the ones that had to come up with the idea ourselves and broach it with them – which put us in an incredibly scary and vulnerable position”*.

(c) the HCP focusing on certain aspects which the participant didn’t find helpful. For instance, ME1 stated: *“there’s weirdness when standing, which ... kind of insomnia and shifted sleep, so having just said that they focus on sleep a lot was a bad thing...but I think they should ask about other symptoms”*.

(d) when knowledge and information was provided that was incorrect. For instance, ME9 noted: *“So when we were first diagnosed we had a meeting with XXX as I mentioned, and she told us that three quarters of teenagers with the illness would recover from it in 2 to 3 years I think she said ... no 6 months she said actually ... which she has since denied saying in fact, but both me and my mother remember it quite distinctly and wrote it down”*

Sub-theme 2.2: interactions which prevented management and coping

There were several identified problems which acted against effective management. These included;

(a) an expectation from other stakeholders (school, family) and HCPs that the symptoms would end and could be explained as a result of a slightly more serious illness like flu. For instance, ME3 illustrates this point *“I don’t remember people really being concerned about it...when the flu eventually did go and I was just left exhausted ... I assume they just thought it was the after effects of just the run of the mill flu that would go away eventually.”*

(b) Accommodation of needs generally and required time away from school, but not being given time to rest or recover from symptoms. For instance, ME1 stated *“the secondary school was quite like no missing school, you can’t miss school. You shouldn’t be missing more than like six days”*

(c) Accommodation of needs at school and during activities and not being allowed to rest for recovery for instance ME4 stated: *“I mean I was trying to go to school, but actually it was making me so much worse trying. Because if I had a good day then I’d go in, but then I’d literally have a bad week afterwards because of payback.”*

(d) experiencing unrealistic expectations from others for instance ME7 stated: *“School was a bit unsupportive at first because you know like I was saying they kind of push you to be able to do that. So therefore, I pushed myself, and that was damaging to my health because I couldn’t do what I wanted to do and everyone else wanted me to do”*.

(e) a repeated cycle of doing too much and having to be supported in an alternative way. Most often this related to experiences of attending school for instance ME2 stated: *“I kept on going in every day and then going back out again, like in the middle of school, like every time– someone had to pick me up, it was very hard.”*

These problems were likely influenced by the belief that others had about the problems being experienced. For instance ME10 had a mixed response from people around them with some thinking the individual was faking the illness: *“Well a lot of the people in my school did. I think some adults were a bit dubious at least, and my brother joked about it a lot. I don’t know, I think my dad was a bit just annoyed, mum was fine I think.”*

In a similar way ME12 stated: *“Well because it took so long for me to get diagnosed I sort of had to tell people well you know it’s meant to be this, but I can’t give you any concrete proof that it is – and I had quite a few people, and some of my friends actually who thought that I was sort of making the whole thing up”*

Sub-theme 2.3: the nature and intensity of symptoms and subsequent needs

Individuals identified the importance of understanding the frequency and intensity of symptoms. For instance, symptoms could change frequently as ME3 states: *“I’d also just like to say – my symptoms change so often, they change a lot and it’s never one thing that is the most important things that stays the most important thing for very long.”* Further to this, individuals identified the nature of intense and dominating symptoms as something which illustrated the impact. For instance, ME3 stated: *“I’ve got headaches and exhaustion and stuff. So yeah at the moment ... yeah my top two which affect me the most are my headaches and the post exertional malaise...I was off school for 3 weeks at the beginning, and then I tried to go back into school full time and on the second day I fainted and was sent home”.*

Associated with this was the importance of considering the contexts and factors which create the symptoms and understanding what not to do. This helped identify what coping and management of the illness should consider. For instance, ME6 stated: *“Because I’ll feel terrible and dizzy and tired, I’ll feel worn out and I haven’t even done much, mostly just getting to the car. It’s a problem because it’s ... like when we’re on the road I can hear and feel the car moving, so that kind of messes with my head and I have sort of a dulling pain in my head”.*

Sub-theme 2.4: ineffective experiences of treatment and management

Various errors or problems with advice were identified. These included:

- (a) Advice being too generic. For instance, ME5 identified that healthcare professionals would provide factual advice and education through printed sheets but not understand the illness. This could be because a diagnosis was not given.
- (b) Health care professionals did not understand the outcomes and impact of the treatment or advice they were providing. For instance, ME6 stated *“They didn’t understand that it was cause and effect, that when I done something it would have an effect on how I felt.”*
- (c) The use of HCPs that were not specialists. This could make the treatment ineffective or attempting tests which could not be carried out. For instance, ME3 recalled a referral for a graded exercise test despite being in a wheelchair and being referred but refused initially for CBT.
- (d) A perception from participants that HCPs were not able to cope with the inability to create change. For instance, ME1 stated: *“the person [HCP] I was seeing couldn’t cope with the fact that they couldn’t fix me or what it was that really went wrong as such, but it kind of went ...it sort of ended up with me getting quite frustrated, because it was sort of like it felt to me like the person I was seeing was kind of like blaming me”.*

Another participant (ME2) identified experiences of interactions where they were repeatedly told the same advice despite no change across time; *“They just kept on saying... they kept on saying you have to go out every day – which I didn’t feel good enough to do– they kept on*

just saying everything wrong, they just weren't right at all. They were almost like literally they just, they weren't helping, they were just getting in the way or something".

ME2 and ME3 identified a range of treatments they had experienced, in the end both had to refuse these treatment because they were making their symptoms worse.

(e) interactions which did not consider the impact on the participant. This included a need for family support within interactions as ME3 states; *"my mum wasn't allowed to come in with me ... it was quite scary, it was the first time I'd ever been in to a medical appointment without my mum"*. ME12 highlights the lack of appropriate support and as a consequence the need to self-manage, stating; *"I felt...that once we'd got the diagnosis that was it, we were left on our own."* This is re-iterated by ME11 *"there wasn't much support from doctors, it's all just been like self-management"*.

2.4.5 Theme 3: Coping strategies

During the patient journey to diagnosis, participants identified several strategies experienced which aided coping and were able to positively benefit their bio-psychosocial well-being. Five codes were identified; (a) support provision from school, (b) pacing and energy saving strategies (c) efficiency of referral (d) supportive advice and (e) practical changes.

Sub-theme 3.1: support provision from school

Several participants identified interactions and support from their school or teacher that aided their ability to cope. This occurred in different ways including:

(a) Decisions that were made to allow the participant to undertake school work from home or within a specialist service at the hospital, and the school being understanding of this for instance, ME3 recalls the following: *"So they believed me when I said that something was wrong, and they saw me like in tears when I was trying to come in to school, but I was just too exhausted. And my head of year immediately agreed to a part time timetable – so I was very lucky there"*.

(b) An adapted timetable for the participant was identified as extremely valuable. For instance, ME7 stated: *"They did allow me to do the 70% timetable because my attendance wasn't good doing the ... that made my attendance a bit better because you know I had time to rest in school."*

(c) the ability to be able to select the pace of activities and being able to undertake activities gradually. For instance, ME5 stated: *"They told me that I needed to go in like for an hour each time, and then I started to increase that, they helped me increase it like ... for example like every month I increased my time by a bit more, which included (increasing school)"*.

(d) one participant highlighted the value of a home tutor stating that without this support that progression at school would be impossible.

(e) finally one student (ME2) identified and described the value of a selected and comfortable environment like on their sofa at home and being taught one to one in that setting as ideal.

Sub-theme 3.2: Pacing and energy saving strategies

Participants consistently identified the importance of understanding the significance and impact of ME/CFS on energy levels. For instance, ME11 reflected back on the lessoned learned about energy conservation and management: *"it's hard to explain, but doing anything causes that much more energy. So for example if I was to do a 10 minute walk, like back when I was really ill, that would have just been in it, like I couldn't walk my dog, literally most of my days were going from my bedroom downstairs to the sofa and then sleeping."* ME14 provides an illustration of the more severe cases of those with ME/CFS and explained the

large impact on their daily life; *"I sleep 13 hours a day, I try to get out of bed each day. I can't dress I'm too tired, I can't bathe unaided. My life is not good. I try to study each day but that uses all of my energy for the day. My tiredness, PEM, pain and mobility are my big problem"*.

How to get better and feel better was important to and experiences of this advice was highly valued across individuals. Most often this meant knowing when to rest and not take part in activities, but it also included planning, pacing and activity management. For instance, ME3 stated *"Yeah, so pacing helped save energy for things that I wanted to do - so socialising can count as that - and lifestyle changes, planning ahead to save energy for things I need to do."* Further to this ME4 stated *"I started doing activity management ... that's what he said ... which was very good and probably helped quite a lot too at the start, but I feel like it could have been given to me earlier as well - cos it's only a piece of paper that we printed off at home"*. A lot of these tasks were to manage fatigue and tiredness as ME11 explains; *"So I will do something and then rest, and depending on the severity of someone's illness they might want to rest more or pace more. So now I can do a full day at school with like 20 minute ... not a full day but part time ... and I'll do like 20 minutes rest like throughout, or like at break it'll be a rest - so like just time for my body just to catch up with itself."*

Some advice was identified as being received from HCPs. For instance, ME5 identified the value of pacing sheets... *"They helped me so I could like make sure that I was on my timetable, like make sure ... yeah, they helped me ... I don't know, it just helped me go in more."* In a similar way ME7 identified the value of advice and strategies received stating; *"the therapist was more about managing things, you know like graded exercise therapy, and CBT was mentioned as well. So like she was more about supporting, management of the condition. And yeah that was really good, she put me on different ... you know did like diaries of things that ... like activity diaries to see what affected it and how I could manage, and then she put me on graded exercise to kind of increase stamina and like how long it would take me to feel fatigued and that kind of thing."*

It is important to note that ME4 did not allow people to talk to provide advice about managing symptoms as they believed they would be better in the future and would not be ill indefinitely.

Sub-theme 3.3: Efficiency of referral and treatment

Participants valued interactions that could positively identify the diagnosis or provide access to individuals who could support this. The efficiency of the referral process was highlighted and valued as part of this, as was the knowledge that effective treatment can begin once this has been identified. Several participants identified the value of seeing HCPs in a private setting stating *"Appointments with private specialists tend to come through a lot quicker, which is a lot easier because it means I get to see people quicker"* (ME3). Another participant (ME15) highlighted the importance of contacting a specialist ME service stating: *"once my mum got the diagnosis then the private doctor, he contacted that information and advice thing, and they told us to go to the ME children's service, which has been absolutely amazing ...it really gave us an understanding of ME because we didn't know how to pace or how to manage my energy, and we didn't really understand how that worked."*

Individuals highlighted the value of certain forms of treatment. This included treatment from physiotherapists. For instance: *"Interviewer: Okay so when you first started to go to the physio, did it help with the muscle pains? ME5 Response: Yeah well she gave me certain exercises that I need to do and things, so it kind of helped."*

Sub-theme 3.4: Supportive advice

Participants highlighted the value of social support in different ways. This included: (a) being known to an individual, being understood over time and knowing the medical records were identified as important. The contrast of this was identified by ME11 who stated: *"The first thing we were told was there's no cure. We were told it's a lifelong condition that might get*

better on its own, but I should learn how to live with it. Yeah, and then from then there wasn't much support from doctors, it's all just been like self-management.”, (b) receiving support that was personalised and informative was highly valued, (c) provision of emotional support and listening from HCPs for instance ME1 stated this as a need within the interview: “is there a service or something you can point me in the direction of that can sort of just give me someone who I can sort of talk to about the fact that I'm sad about the fact I'm missing out on life and that I'm having to give up stuff....sort of CBT kind of thing, which isn't what I needed and all the people who have suggested it have later come back or a different doctor who is a psychologist, you know knows about these things, has gone sorry you don't really need that kind of thing”.

It should be noted that support from friends could be a problem as individuals could feel so tired that even keeping in contact via social media was hard. Finally individuals highlighted the importance of advice from doctors around rest and energy conservation and the need to believe what is happening for instance, ME13 stated: *“Doctors must tell you to rest. They must listen and tell you to rest until you know what is wrong. They need to take it seriously. It feels like you might die and you are scared and when they tell you to go home and take a tablet it is really upsetting. It makes people not believe you because the Doctors should know what is best to do”*

Sub-theme 3.5: Practical changes

Considerations for longer term change was identified as a requirement by participants. It was suggested that years ahead could be considered. Changes undertaken by participants included;

(a) Installing a stair lift or wet room to make it more suitable. A stair lift for energy conservation was especially important for ME3 who stated *“A stair lift [at home] is something that would have helped me massively 3 or 4 years ago”.*

(b) Consideration by service providers to accommodate the needs of participants in not being able to attend services because of limited energy. For instance, ME11 stated: *“I think doctors should do home visits if the patient isn't well enough to go. Because a lot of the times like the doctors ... like we've had to travel to my consultant before because they've moved him, and at times it's been tough getting to him cos of I've just been unwell.”*

(c) A need for service providers to be more aware of the various resources available to support individuals with ME. This could include knowing about forums, support groups and specialist centres, supported by ME14: *“I think doctors should make more awareness about the forums and the support groups that are available, because we found them on our own, and I think they might have been more useful if we found them earlier on so we can talk to other parents about it”.*

2.5 Discussion

Summary

Interviews identified a number of modifiable factors within the current care pathway that could be targeted to improve the health, wellbeing and quality of life of young people diagnosed with ME/CFS. Participants were both male and female, young people aged 11-18 with ME/CFS from across the UK with mild to severe disease. The views expressed often reflected the individual nature of the condition severity and age of participants, but core themes emerged from interviews and included the patient journey to diagnosis and subsequent development of management and coping strategies, as well as concerns of a limited understanding of the illness and its management from the HCPs involved in their care

pathway. A number of the themes highlighted potentially modifiable factors, both positive and negative, that could be addressed within the care pathway to improve the care of young people with ME/CFS. Factors were physical, psychological, contextual and social, and included support and information for young people with ME/CFS and HCPs for diagnosis and management, support for HCPs in how to better interact with patients and family members and bespoke approaches to management. Key points for recommendations include training of HCPs (disease, management, interactions); training of schools, information of management for young people with ME/CFS.

Journey to diagnosis

A core theme from interviews was the experience of symptoms prior to diagnosis, to illustrate the participant's memories of how the illness began to present itself. Early experiences were often identified as resolvable short-term illnesses such as colds and flu. Symptoms were over time differentiated from being normal colds or flu, because they would continue for longer and progressed to other symptoms including a feeling of being completely drained with pain and heaviness in limbs, dizziness, foggy thinking and in one individual labile mood. This may be an important reflection for consideration by HCP in the early consideration of a diagnosis of ME/CFS. The participants' journey to diagnosis involved reflections of the gradual worsening of both physical and cognitive symptoms, as participants began to associate what was happening with the presence of a more serious illness. Over a longer time period individuals reported experiencing progressive and more devastating symptoms, linked to more serious illnesses such as flu or glandular fever. Symptoms were reported to frequently and progressively impact on participants' ability to engage in meaningful activities and relationships.

Presentation of ME/CFS can be seen in children both gradually and abruptly. Acute onset of fever and viral-like symptoms is common along with a history or minor relapsing illness over time (Rowe et al., 2017). Known illnesses such as infectious mononucleosis may also be a risk factor for ME/CFS in young people (Katz et al., 2009).

Management and coping strategies

Factors that were highlighted to negatively affect coping were the impact of the attitudes of HCPs and other stakeholders (teachers, peers) who were perceived to have limited understanding of the illness and its impact. An overriding feeling of frustration at the lack of understanding of the disease, the severity of symptoms and disease management was also a recurrent theme with young people reflecting on participating or being encouraged to participate in activities that were thought to be unachievable or detrimental. Further frustration was expressed in the variability in the nature and intensity of symptoms and subsequent needs. Frustration was expressed at both a personal level and also in individual's reactions with other individuals and their difficulty in understanding an altering presentation in symptoms from day to day. The frustration of others in not being able to help was also felt by the young people, having a detrimental effect on their ability to cope with the condition. Finally a huge challenge for the young people was having to deal with a lack of positive treatments and exposure to ineffective treatment and management approaches for the disease.

Positive coping strategies included support provision from school such as altering timetables, the importance of pacing and energy saving strategies from HCPs, timely and well managed referrals to appropriate HCPs and the benefit of social support and a more supportive attitude when giving advice. The positive impact of social media forums and private healthcare were often reported, as were benefits from practical changes to the home or school setting.

Coping and management strategies in children and young people with ME/CFS reported by Parslow et al., (2015) included self-learning a balance of activity and saving up energy for

activities, a description used by specialist therapeutic services. Rowe et al., (2017) suggests training in coping skills for young patients and highlights the importance of the relationship between the school, student, family and the clinical team.

Interactions with HCPs and other stakeholders

Participants highlighted a need for better management from HCPs. Factors highlighted to have a negative impact on coping included a limited understanding of the illness, interactions which prevented management and coping, the nature and intensity of symptoms and subsequent needs, and ineffective experiences of treatment and management. Within this theme there was consideration of the need for an earlier diagnosis, in order to reduce the extreme experiences of symptoms and emotional distress. Some participants expressed anger with the poor management and lack of knowledge of health care professionals. Participants noted that health care professionals appeared to obtain a diagnosis most often through a process of elimination. HCPs identified a need often to refer to other specialities when a diagnosis could not be obtained. Limited advice from HCPs meant that some individuals began to search for a diagnosis. Young people reported that HCPs did not appear to understand the condition, with a lack of advice or reassurance from HCPs in how to coping and managing symptoms. There were a number of issues highlighted with schools who had a limited understanding of the condition and its management such as need to rest or timetabling flexibility. Indeed the participants identified the importance of a better understanding of the frequency and intensity of symptoms and the changing and variability of symptoms and needs.

Meta synthesis from Bayliss et al., (2014) highlighted patient frustration around failure of GPs to diagnose or validate ME/CFS and a belief that medical teams lacked the time or therapeutic expertise to provide adequate emotional support.

Consultation strengths and limitations

The consultation was carried out and managed by a research team who are experienced in a range of research methodologies and in working across a range of different conditions including ME/CFS. The team was supported by a diverse Advisory Group which consisted of counselling psychologists with experience in working with people with ME/CFS, physiotherapists within the NHS who work with young people in the fatigue service, the parent of a child with CFS, a Head Teacher providing education for pupils with medical needs, as well as representation from Action for ME. By including the online element we were able to include the most severely affected children/young people.

Limitations of the consultation will be reflective of qualitative research in a relatively small heterogeneous sample. Due to predefined numbers required by in the commission and the short time scale there were limitations on the sample size. We were however able to recruit above the target 14 (16 consented, 15 completed). The time scale and lack of engagement from third party organisations meant there were limitations on the recruitment. Despite this, the included participants were reflective of a range of geographies across England, genders and condition severities. The UREC Chair received communication from a concerned parent that the design of the study and the exclusion criteria that children/young people should not take part if doing so would cause pain or discomfort would mean that those severely affected would not take part and thus skew the data. Discussion with the research team was encouraged for anyone interested in being involved in the consultation and children/young people who reported severe ME/CFS did take part. However, we accept that there were limitations on those children/young people that were able to participate. Qualitative methodology and inherent bias of the researchers but a number of strategies were implemented to reduce bias. These included the use of the topic guide with prompts for interviews and a single interviewer, third party transcription of all audio recordings and blind analysis transcripts by three researchers (separate to the interviewer). In accommodating

those children/young people most severely affected the use of online survey was utilised rather than individual interviews which may have limited the depth of response.

The consultation should be reviewed and interpreted with this in mind.

Acknowledgments

Firstly, the research team would like to thank all the children and young people who gave their time to take part in the interviews, and their families who provided their support during the interviews. Their willingness to take part in the consultation and the open way in which spoke about their experiences has enabled us to

We would like to acknowledge Action for ME for their support in helping us recruit to the consultation. We would also like to acknowledge the Advisory Group for their support and expertise in the development of the consultation process.

2.6 References

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3 Drawing on the report to inform the recommendations

Two members of the research team from the Oxford Clinical allied technology and trial services unit presented the findings of the report to the committee. Prior to the meeting the committee had received the study report. The themes that emerged from the report were taken into consideration alongside identified evidence when drafting recommendations. This was the most applicable evidence for a number of topics and influenced the recommendations directly. Where relevant this is referenced in the committee discussions of the evidence reviews.

4 The committee's overview of the research

The committee assessed the report and agreed that it was a well conducted and reported study led by a team experienced in qualitative methodology and supported by a diverse Advisory Group. The committee noted the sample was diverse and recruited from across England, including both females and males and young people with different severities of ME/CFS.

The lay committee members noted that the report findings reflected their own experiences.

Although the limitations of the research are described well in the report the committee noted the additional points:

- the diagnosis of ME/CFS was self-report
- it was unclear if all the sample were recruited from Action for ME potentially representing only one group of young people with similar views
- it was unclear if the participants were currently under NHS care and if the experiences reflected current care.
- all the young people had their parents present, The committee recognised this was important to support the young people but their parent's presence may have influenced their responses
- children with ME/CFS and children and young people with very severe ME/CFS were not represented the committee recognised the difficulties with recruiting and researching these groups.
- data collection methods had to be adapted to ensure the project was accessible. This resulted in a mix of data collection with face to face interviews and online interviews being conducted.
- the research team were restricted by the areas of the scope and the time to conduct the research. This did not allow for deeper probing and questioning potentially missing some important issues.

This was taken into account when considering the findings of the research.

Appendix A: Report Consultation information leaflets

The following pages contain the individual information leaflets that were provided to children and young people (aged 9-15 and aged 16-18) and parents as part of the recruitment process.

NICE

National Institute for
Health and Care Excellence

INFORMATION SHEET

NICE Consultation: Guidelines for Children and Young People

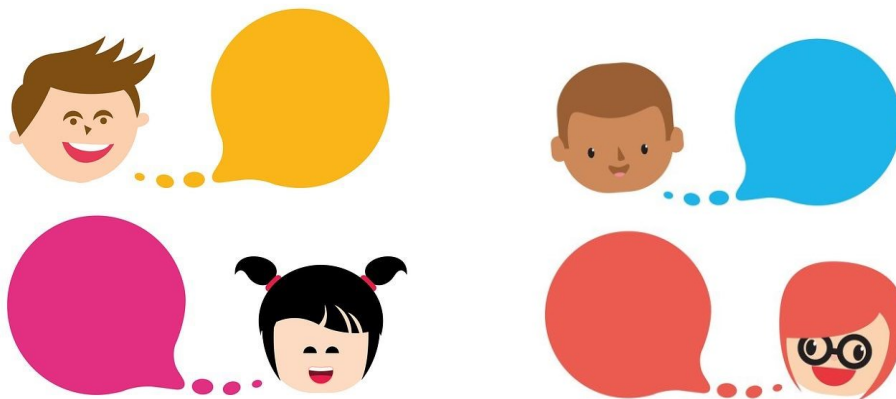
9-15 years

Full title of Project: Involving children and young people in developing a NICE guideline on Myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) diagnosis and management.

You are being invited to take part in a research study. Before you decide whether or not to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully.

What is the purpose of the study?

We are asking people like yourself about how to improve care for children and young people with ME/CFS. To take part, children or young people will attend a group or telephone call to talk about how children and young people like to be supported. We will ask you questions about your experiences of healthcare and will give you the opportunity to discuss and share these experiences in a safe space. The information will be used to inform the development of new national guidelines for the care of children and young people.



Why have I been invited to participate?

You have been invited to take part because you are between 9 and 15 years of age and have been told by a doctor that you have ME/ CFS. Your parent/carer will also be able to tell us this.

Do I have to take part?

It is up to you to decide whether or not to take part.

If you do decide to take part, you will be given this information sheet and a privacy notice (this tells you what we do with any information you give us) to keep. You will also be asked to sign a form saying that you want to take part.

If you take part in the focus group, you can leave the study at any point and information that you have already given will be used in the study. If you are taking part in an interview, you can leave the study at any point and you can ask the researchers to remove any information you have given up to the point that the information is analysed/de-identified (so that we don't know what information has come from you).

What will happen to me if I take part?

If you take part, you will attend ONE group chat/video call, depending on which you prefer. This will be either be at Oxford Brookes University or there will be the option to take place over a video call. Your voice will be recorded during the group chat/video call using an audio-recorder. Your parent or carer will be asked the same questions as you but they will complete an online survey instead of taking part in the focus group or interview. .

What are the possible disadvantages and risks of taking part?

The focus group or interview you take part in will not take more than one hour. You may start to feel tired but if you do you will be able to take a rest in a quiet space and start again when you are ready. We will check that you are ok during the focus group or interview and if you tell us you are upset or we think you might be upset we have a process we will follow. You will be given the chance to have a break, you can then continue or if you do not wish to continue and are still upset we will advise you to speak to your parent, GP or healthcare provider. With your permission we can also contact these people for you. With your permission we will contact you after the focus group or interview to check how you are. You can also contact us if you feel upset after the focus group or interview.

What are the possible benefits of taking part?

Taking part in the study may not help you. However, you will have an opportunity to influence national guidelines. Doctors, nurses and other healthcare professionals will use these guidelines to improve care, possibly for you in the future and for similar children and young people.

Will what I say in this study be kept confidential?

Only the researchers and other young people attending the group will know your views. Your name and any information that may identify you will NOT be given to others or published in our reports.

If we become worried about your safety or the safety of others we will make sure you get the support you need.

What should I do if I want to take part?

If you would like to take part in this research study, you can do so by contacting the research team by using the contact details below.

What will happen to the results of the research study?

A report will be produced for NICE. This will include what we find out from the information provided by young people attending our focus groups/telephone calls. In the future the findings may also be used to produce published research articles. If we are allowed and if you or your parent/carer has given us an email address, we can email you with a summary of the study. The charities involved in the study will also show this on their websites.

Who is organising and funding the research?

The Royal College of Physicians is funding the research.

Who has reviewed the study?

This research has been approved by the University Research Ethics Committee at Oxford Brookes University.

Contact for Further Information

To ask about taking part in the study please contact:

For any other questions contact the Principal Investigator on the project:

If you worried about the way this study has been conducted, you should contact the Chair of the University Research Ethics Committee on ethics@brookes.ac.uk.

Thank you!



INFORMATION SHEET

NICE Consultation: Guidelines for Children and Young People

16-18 years

Full title of Project: Involving children and young people in developing a NICE guideline on Myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) diagnosis and management.

You are being invited to take part in a research study. Before you decide whether or not to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully.

What is the purpose of the study?

We are carrying out a consultation about how to improve care for children and young people with ME/ CFS. To take part, young people will attend a focus group or interview to discuss how children and young people like to be supported. This group will ask you questions about your experiences of healthcare and will give you the opportunity to discuss and share these experiences in a safe space. The information will be used to inform the development of new national guidelines for the care of children and young people with ME/ CFS.



Why have I been invited to participate?

You have been invited to take part because you are 16 to 18

years of age and have been told you have a clinical diagnosis of ME/ CFS. We will ask you or your parent/carer to confirm that you meet all of the inclusion criteria and none of the exclusion criteria.

You must:

- be aged between 9-18 years
- live anywhere in England and Wales; and
- have experience of living with mild, moderate and /or severe ME/CFS (<https://www.nice.org.uk/guidance/cg53/chapter/Appendix-D-Definitions-used-in-this-guideline>)
- have been diagnosed with ME/CFS by a healthcare professional - self reported by participant/parent/carer
- have capacity to consent/assent and parental consent/ assent

You must not:

- be unable to communicate or participate safely (as determined by your parents/carers)
- be unwilling to provide assent
- be too severely affected by your condition such that taking part could cause significant and long-term detriment to your health. This will be discussed openly and any reservations on either side will be raised before being included in the study.
- have serious thoughts about self-harm or suicide, as reported by you or your parents/carer.
- have primary psychiatric disorders and substance abuse, as reported by you or your parents/carer.

Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part, you will be given this information sheet and a privacy notice (this tells you what we do with any information you give us) to keep. You will also be asked to sign a form saying that you want to take part. If you take part in the focus group, you can leave the study at any point and information that you have already given will be used in the study. If you are taking part in an interview, you can leave the study at any point and you can ask the researchers to remove any information you have given up to the point that the information is analysed/de-identified (so that we don't know what information has come from you).

What will happen to me if I take part?

If you take part, you will be invited to attend ONE focus

group at Oxford Brookes University or interview, either at a chosen location, or by video call depending on which is most convenient for you. You will be asked questions about your ME/CFS related to the following themes; diagnosis, management, how your ME/CFS is monitored and reviewed, and available information, education and support. Your voice will be recorded during the group consultation using an audio-recorder. Your parent or carer will be asked the same questions as you but they will complete an online survey instead of taking part in the focus group or interview. Your travel expenses to attend the focus group or interview will be fully reimbursed.

What are the possible disadvantages and risks of taking part?

The focus group or interview you take part in will not take more than one hour. You may start to feel tired but if you do you will be able to take a rest in a quiet space and start again when you are ready. We will check that you are ok during the focus group or interview and if you tell us you are upset or we think you might be upset we have a process we will follow. You will be given the chance to have a break, you can then continue or if you do not wish to continue and are still upset we will advise you to speak to your parent, GP or healthcare provider. With your permission we can also contact these people for you. With your permission we will contact you after the focus group or interview to check how you are. You can also contact us if you feel upset after the focus group or interview.

What are the possible benefits of taking part?

There are no direct benefits of taking part. However, you will have an opportunity to influence national guidelines. Doctors, nurses and other healthcare professionals will use these guidelines to improve care for similar children and young people.

Will what I say in this study be kept confidential?

Only the focus group/interview team and other young people attending the group will know your views. Your name and any information that may identify you will NOT be given to others or published in our reports.

If we become worried about your safety or the safety of others we will make sure you get the support you need.

What should I do if I want to take part?

If you would like to take part in this research study, you can do so by contacting a member of the research team using the contact details below.

What will happen to the results of the research study?

A report will be produced for NICE. This will include what we find out from the information provided by young people attending our focus groups/interviews. In the future, the findings may also be used to produce published research articles. If we are allowed and if you or your parent/carer has given us an email address, we can email you with a summary of the study. The charities involved in the study will also show this on their websites.

Who is organising and funding the research?

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Research Ethics Committee at Oxford Brookes University.

Contact for Further Information

To ask about taking part in the study please contact: v

For any other questions please contact the Principal Investigator on the project:

If you worried about the way this study has been conducted, you should contact the Chair of the University Research Ethics Committee on ethics@brookes.ac.uk.

Thank you!



INFORMATION SHEET

NICE Consultation: Guidelines for Children and Young People

Parents and carers

Full title of Project: Involving children and young people in developing a NICE guideline on Myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) diagnosis and management.

Your child/adolescent or a child/adolescent you care for is being invited to take part in a research study. Before you decide whether or not it is appropriate for them to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. If they choose to take part, you will also be given the opportunity to be involved yourself.

What is the purpose of the study?

In order to best meet the needs of young people with ME/CFS we plan to develop a guideline that is representative of the voices and experiences of young people and those directly involved in their care which reflects their lived experience. We intend to build on the findings of a collaborative consultation. The consultation was designed and undertaken by children and young people, their parents and health and educational professionals and it was facilitated by UK charity Action for M.E. The aim of the consultation was to assess the perceived needs of children and young people in order to cope better with the impact of living with ME/CFS.

We are carrying out a consultation about how to improve care for children and young people with ME/CFS. To take part, we are looking for young people aged between 9 and 18 willing to attend a focus group/interview to discuss how they like to be supported. This group will ask the young people questions about their experiences of healthcare and will give them the opportunity to discuss and share their experiences in a safe space. The information will be used to inform the development of new national guidelines for the care of children and young people.

Why have I been invited to participate?

You have been invited because you have expressed an interest in hearing about ME/CFS research via Action for M.E or you have seen the study advertised on other ME/CFS relevant websites.

Your child/ adolescent must:

- be aged between 9-18 years
- live anywhere in England and Wales; and
- have experience of living with mild, moderate and /or severe ME/CFS (<https://www.nice.org.uk/guidance/cg53/chapter/Appendix-D-Definitions-used-in-this-guideline>)
- have been diagnosed with ME/CFS by a healthcare professional - self reported by participant/parent/carer
- have capacity to consent/assent and parental consent/ assent

Your child/adolescent must not:

- be unable to communicate or participate safely (as determined by their parents/carers)
- be unwilling to consent/assent
- not be too severely affected by their condition such that taking part could cause significant and long-term detriment to their health. This will be discussed openly and any reservations on either side will be raised before including a participant in the study.
- have serious thoughts about self-harm or suicide, as reported by parents/carer.
- presence of primary psychiatric disorders and substance abuse , as reported by parents/carer.

Please discuss openly any reservations with the research team.

To take part in the online questionnaire your child/adolescent must be taking part in a focus group/interview.

Does your child/ adolescent have to take part?

It is up to you and your child/young person to decide whether or not to take part. If you do decide to take part, you will be given this information sheet and a privacy notice (this tells you what we do with any information you give us) to keep. You will also be asked to sign a consent form allowing your child/ young person you care for to take part. If your child/young person you are responsible for takes part in the focus group, you can withdraw from the study at any point and information that they have already given will be used in the study. If they are taking part in an interview, you can withdraw from the study at any point and you can ask the researchers to remove any information that has been given up to the point that the information is analysed/de-identified. If your child/adolescent chooses to take part, you do not have to take part in the online questionnaire. If you and your child do not feel confident to communicate, verbally in English please discuss with a member of the research team.

What will happen to me if I take part?

If your child/ young person takes part, you will attend ONE focus group at Oxford Brookes University or alternatively an interview, which will take place at a location chosen by you or by phone depending on which is most convenient for you. We will record the voice of your child/ young person using an audio recorder. As a parent or carer you will be asked the same questions as your child but these will be completed separately and via an online questionnaire. This will keep the time of the interview to a minimum and ensure we can fully capture both your experiences as a parent or carer and those of your child/ young person.

The topics of the focus groups/interviews/online questionnaire will include:

- Identification of the child/adolescents condition and the assessment process before diagnosis;
- The process of the diagnosis of ME/CFS;
- The management and support they (and their families) have received regarding their ME/CFS;
- How their ME/CFS is monitored and reviewed;
- What information, education, and support for people with suspected or diagnosed ME/CFS and their families and carers was/is available and how helpful it was
- What information, education and support for health and social care professionals is needed to help them manage their condition well.

What are the possible disadvantages and risks of taking part?

The focus group or interview your child/young person you are responsible for takes part in will not take more than one hour. They may start to feel tired but if this happens, they will be able to take a rest in a quiet space and start again when they are ready. The focus group or interview they will take part in will not take more than one hour. The online survey you can choose to take part in should not take longer than 20 minutes. We will check that the child/young person you are responsible for is ok during the focus group or interview and also that you are ok while you complete the survey. If we think you/they might be upset or if we are told that you/they are upset, we have a process we will follow. You/they will be given the chance to have a break, you can then continue or if you/they do not wish to continue and are still upset we will advise you/they to speak to you, a GP or mental health provider. With your/their permission we can also contact these people for you.

With your/their permission we will contact you/them after the focus group or interview to check how you are. You/they can also contact us if you feel upset after the focus group or interview. We will reimburse your travel costs for attending the focus group or interview.

What are the possible benefits of taking part?

There are no direct benefits of taking part. However, you will have an opportunity to influence national guidelines. Doctors, nurses and other healthcare professionals will use these guidelines to improve care for similar children and young people.

Will what I say in this study be kept confidential?

Only the focus group/interview team and other young people attending the group will know your child's views. Your child/adolescent's name and any information that may identify your child will NOT be given to others or published in our reports. Data we collect from you via the online questionnaire will be anonymous.

All information collected about your child/adolescent will be pseudonymised. Access to the data codes will be only by the lead researcher working on

this study. Access to computer files will be by password only with restricted access and data and codes will be kept in locked filing cabinets. Data generated by the study will be retained in accordance with the University's policy on Academic Integrity. The data generated in the course of the research will be kept securely in paper or electronic form for a period of ten years after the completion of a research project.

What should I do if I want to take part?

If your child/adolescent would like to take part in this research study, he or she can do so by contacting the research using the contact details below.

What will happen to the results of the research study?

A report will be produced for NICE. This will include what we find out from the information provided by young people attending our consultation groups. In the future, the findings may also be used to produce published research articles. If we are allowed and if you or your parent/carer has given us an email address, we can email you with a summary of the study. The charities involved in the study will also show this on their websites.

Who is organising and funding the research?

The Royal College of Physicians are funding this study.

Who has reviewed the study?

This research has been approved by the University Research Ethics Committee at Oxford Brookes University.

Contact for Further Information

To ask about taking part in the study please contact:

For any other questions please contact the Principal Investigator on the project:
If you worried about the way this study has been conducted, you should contact the Chair of the University Research Ethics Committee on ethics@brookes.ac.uk.

Thank you for taking time to read this information sheet.

Appendix B: Consultation consent forms

The following pages contain the individual assent/consent forms that were used by the research team to receive informed consent. Where appropriate, assent was received from children and young people aged 9-15, consent was received from young people aged 16-18 and parents/carers.



Assent Form (9-15 years old)

(To be completed once a parent/guardian has consented for a child younger than 16 years old)

Title of Project: Involving children and young people in developing a NICE guideline on Myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) diagnosis and management.

Principal Investigators:

Participant Identification Number for this study:

| Child (or if unable, parent on their behalf) /young person to circle all they agree with: | Please circle |
|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------|
| <ul style="list-style-type: none">I have read and understood the information sheet [version1 12 Jul 2019] for 9-15 Years Old | Yes/No |
| <ul style="list-style-type: none">I have had time to ask questions and have had these answered | Yes/No |
| <ul style="list-style-type: none">I know that I can stop doing the study at any time without giving reason. Information I give may still be used by the researchers | Yes/No |
| <ul style="list-style-type: none">I understand that I will not have my name on any of the final papers or presentations and my data will be kept safe and secure | Yes/No |
| <ul style="list-style-type: none">I agree to be in this study | Yes/No |

If any answers are 'no' or you don't want to take part, don't sign your name.

If you do want to take part, you can write your name below:

| | | |
|-----------------------------------|-------|-------------------------|
| Name of participant | Date | Participant's signature |
| _____ | _____ | _____ |
| Name of researcher taking consent | Date | Researcher's signature |
| _____ | _____ | _____ |

CONSENT FORM (16-18 year old)

NICE Consultation: Guidelines for Children and Young People

Full title of Project: Involving children and young people in developing a NICE guideline on Myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) diagnosis and management.

Principal Investigators:

Please initial box

1. I confirm that I have read and understand the information sheet [version 1 12 Jul 19] for the above study and have had the opportunity to ask questions and have them answered.
2. I understand that I can withdraw at any time, without giving reason. Information I provide before I withdraw may be used by the researchers.
3. I understand that I will not have my name on any of the final papers or presentations and my data will be kept safe and secure.
4. I agree to my data being used in research publications to be included in NICE guideline development.
5. I agree to the interview/focus group being audio recorded
6. I know that the information I provide can only be protected within the limitations of the law.
7. I agree to take part in the above research study

5 Please initial box

- | | 6 | Yes | No |
|---------------------------------------------------------------------------------------------------------------------------------------------|----------|--------------------------|--------------------------|
| 8. I agree to the use of pseudonymised quotes in publications | | <input type="checkbox"/> | <input type="checkbox"/> |
| 9. Would you like to be contacted for future research relevant to ME/CFS/Physical activity? If yes, please provide your email address below | | <input type="checkbox"/> | <input type="checkbox"/> |

.....

| | | |
|-----------------------------------|-------|-----------|
| _____ | _____ | _____ |
| Name of Participant | Date | Signature |
| _____ | _____ | _____ |
| Name of Researcher taking consent | Date | Signature |

CONSENT FORM (parent on behalf of child/adolescent)

NICE Consultation: Guidelines for Children and Young People

Full title of Project: Involving children and young people in developing a NICE guideline on Myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) diagnosis and management.

Principal Investigators:

Please initial box

1. I confirm that I have read and understand the information sheet [version 1 12 Jul 2019] for the above study and have had the opportunity to ask questions and have them answered.
2. I understand that I can withdraw my child/ adolescent at any time, without giving reason. Information I provide before I withdraw consent may still be used.
3. I understand that in all instances where results are disseminated, my child's/ adolescent's identity will remain pseudonymous and that all data will be treated as confidential (within the limitations of the law)
4. I agree to my child's/ adolescent's data being used in research publications to be included in NICE guideline development.
5. I agree to contact my child's/ adolescent's GP if I have any health or medical concerns to check that there is no reason why they should not take part in the study and to let the researchers know if it is advised that my child does not take part
6. I agree my child/ adolescent to take part in the above research study.
7. I agree to the interview/focus group being audio recorded

7Please initial box

8 Yes No

8. I agree to the use of pseudonymised quotes in publications

9. Would you like to be contacted for future research relevant to ME/CFS/Physical activity? If yes, please provide your email address below

.....

Name of Participant

Date

Signature

Name of Researcher taking consent

Date

Signature

Appendix C: Consultation group topic guide

The topic guide with focus group/interview questions and prompts for children and young people aged 9-15 and young people aged 16-18 can be found on the following page.

| Topic area | Age:16- 18 | Prompts | Age: 9- 15 | Prompts |
|-------------------------------------------------------|---------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------|
| Identification and assessment before diagnosis | Can you tell us about when you first started to feel unwell/poorly before you were told you had ME/CFS? | <i>When did you first notice a change in your health? What symptoms appeared? What was people's understanding of what you were experiencing?</i> | Can you tell us about when you first started to feel unwell/poorly | <i>What changed in how you were feeling? Did people understand what you meant?</i> |
| | When you first started feeling unwell with ME/CFS what effect did this have on your life? | <i>For example how did your family or school friends feel/respond when you were diagnosed?</i> (health, symptoms, school/college, friends, family) | When you first felt unwell how did this make you feel? | <i>Did anything change with your school, family or friends?</i> |
| | Can you tell us about your experience with healthcare professionals? | <i>Say what we mean by healthcare professionals</i> <i>Was there anything that went well or not so well?</i> | Can you tell us what it was like when you first talked to doctors or nurses about how you were feeling? | <i>Say what we mean by healthcare professionals</i> <i>Was there anything that went well or not so well?</i> <i>How did you feel?</i> |

| | | | | |
|-----------------------------|--------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------|
| Diagnosis of ME/CFS | How long did it take for you to be diagnosed with ME/CFS? | <i>If it was a long time, what was the impact?</i> | Did it take a long time to find out what your illness is? | <i>If it was a long time, how did this make you feel?</i> |
| | Could you tell us how you were diagnosed? | <i>Which medical staff did you see? Was there anything that went well or not so well?</i> | Could you tell us how you were told you were unwell? | <i>Which medical staff did you see? Was there anything that went well or not so well?</i> |
| | What support was made available to you at the time of diagnosis? | <i>Were you offered any specific services or information? Was there anything that went well or not so well?</i> | Was there any help offered to you when you were diagnosed? | Were you offered any specific services or information? Was there anything that went well or not so well? |
| Management of ME/CFS | Can you tell us about the types of treatment have you used since your diagnosis of ME/CFS? | | Can you tell us about what things you have tried to make you feel better? | |
| | Were there any treatments that you believe had the most effect on your symptoms/ health or | | What things helped you? Is there anything that didn't help? | <i>Did it help symptoms/anything else?</i> |

| | | | | |
|------------------------------|------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| | recovery? Is there anything that didn't help? | | | |
| | Can you tell us about any treatments that helped with participation in school or socialising with friends? | | Can you tell us about anything that helped you with school or staying in touch with friends? | |
| | Can you describe the impact your symptoms have on everyday life? | <i>Are there any symptoms, that have more of an impact than others?</i> | Can you describe what things are affected when you feel unwell? | <i>Which unwell feelings? Who does it affect?</i> |
| | Can you tell us about your experience with healthcare professionals now? | <i>Do you/did you see people regularly? Is it well organised? Do you see different people? Are your needs met? Do you feel in control of what happens to you? Does it take a lot of time? What is the impact on the things you want to do?</i> | Can you tell us about your experience with healthcare professionals now? | <i>Is it well organised? Do you see different people? Are your needs met? Do you feel in control of what happens to you? Does it take a lot of time? What is the impact on the things you want to do?</i> |
| Monitoring and review | Has anyone talked to you about your managing illness, and what might happen in the future? | | Has anyone talked to you about managing your illness in the future? | |

| | | | | |
|-------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------|---------------------------------------------------------------------------------------------------------------------|--------------------------------------|
| Information, education and support | Can you tell us about what information and support you were given about ME/CFS? | <i>How did you receive it? Was it useful?</i> | Can you tell us what you have been told about your condition? | <i>Who told you? Was it helpful?</i> |
| | Describe what information, education or support would you like or do you think would have been helpful before gaining a formal diagnosis? | How about during treatment? | If you could go back in time what would you want to tell yourself about your illness? | |
| | In an ideal world, what would you have wanted healthcare professionals to have told you or done for you? | | If you had a friend with the same illness as you what would you like doctors or nurses to tell them or do for them? | |
| | What kinds of support would you like to have going forward? | | What kinds of support would you like to have? | |

