

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

Appendix 2: Involving adults with severe ME/CFS symptoms in developing a NICE guideline on Myalgic encephalomyelitis/Chronic fatigue syndrome: diagnosis and management

NICE guideline <number>

University of Manchester Centre for Primary Care

November 2020

Draft for Consultation

*This guideline was developed by the
National Guideline Centre*

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1 **Involving people with severe ME/CFS**

2

3 An integral part of developing NICE guidelines is the involvement of people with direct
4 experience of the condition and all guideline committees have lay members. The ME/CFS
5 guideline committee has 5 lay members with varying experience of severe ME/CFS either
6 directly or as parents of children and young people who have had periods of severe ME/CFS.
7 However, during the scoping process it was identified there was limited published evidence
8 directly from the perspective of people with severe ME/CFS. For this topic it was considered
9 crucial that the experiences, perspectives and opinions of people with severe ME/CFS
10 inform the guideline and an online questionnaire was conducted with people with severe
11 ME/CFS.

12 The consultation was commissioned by NICE and carried out by the University of
13 Manchester Centre for Primary Care.

14 The University of Manchester Centre for Primary Care was awarded the commission after an
15 open tender process. An invitation to tender was sent to the guideline registered
16 stakeholders and posted on the Royal College of Physicians website. Applicants had to
17 submit a written proposal outlining how they met the research brief, submitted bids were
18 shortlisted and shortlisted bidders were then interviewed by telephone. The process was led
19 by the National Guideline Centre (NGC) and overseen by a subgroup from the ME/CFS
20 guideline committee. The subgroup comprised of the ME/CFS guideline chair, vice chair, the
21 NGC guideline lead, two lay members and representation from the NICE Patient and Public
22 Involvement unit. The subgroup shortlisted the bids, interviewed the shortlisted bidders and
23 awarded the tender.

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

26

2 ¹ Report

2 ² 2.1 Summary

3 ³ 2.1.1 Background

4 Myalgic encephalomyelitis (ME) and/or chronic fatigue syndrome (CFS) is a poorly understood
5 illness that affects approximately 250,000 people in the UK. It is estimated that around 25% of
6 adults with ME/CFS experience severe symptoms or illness presentation. These people are
7 often housebound or bedbound for long periods, sometimes lasting many years. At the most
8 severe end of the ME/CFS symptom spectrum, patients report fatigue after minimal effort, post-
9 exertional malaise, prominent cognitive deficits, intolerance of light, noise and other stimuli,
10 and a host of other symptoms. This cohort represent the most challenging sub-group within
11 the ME/CFS population. Given the severity of symptoms reported, many of these people find
12 it difficult to access medical and social care.

13 A new guideline for Myalgic encephalomyelitis (or encephalopathy)/chronic fatigue syndrome
14 (ME/CFS): diagnosis and management is currently being developed by the National Institute
15 for Health and Care Excellence (NICE) to inform health and social care practice in England.
16 It is essential that NICE guidelines are informed by the views of those receiving care. This
17 report presents findings from a consultation with people with severe ME/CFS symptoms. The
18 consultation aimed to explore the views and experiences of people with severe ME/CFS
19 symptoms on the following topics identified from the guideline scope: Identification and
20 assessment before diagnosis, diagnosis of ME/CFS, management of ME/CFS, monitoring
21 and review, information, education, and support for people with suspected or diagnosed
22 ME/CFS and their families and carers, and information, education and support for health and
23 social care professionals. The consultation was commissioned by NICE and carried out by
24 the University of Manchester Centre for Primary Care: Professor Aneez Esmail, Dr Keith
25 Geraghty, Dr Charles Adeniji and Dr Stoyan Kurtev.

26 ²⁶ 2.1.2 Methods

27 The project employs a survey instrument to gauge the views of people with severe ME/CFS,
28 within the context of an exploratory study design encompassing inductive and qualitative
29 methods. Ethical approval for this project was sought and granted from the University of
30 Manchester Research Ethics Committee (2019-7763-12089). A series of research questions
31 were derived from an embedded set of research objectives formulated from consultation with
32 the guideline committee and based on the guideline scope. These questions were imputed into
33 an electronic survey that was pilot tested on 10 patients from a convenience sample and then
34 opened nationally to people with severe ME/CFS during November 2019, using social media
35 promotion and advertising via patient organisations. Approximately 1600 persons clicked on
36 the survey link, 343 started the survey, 124 completed all questions and from these we
37 retrieved 60 complete responses, including meeting our inclusion criteria of self-reported
38 severe status and ME/CFS confirmed by a medical professional. We used thematic and
39 narrative methods to analyze and synthesize collected data.

40 ⁴⁰ 2.1.3 Findings

41 We identified a clear cohort of people with severe ME/CFS. Almost 2/3rds of the cohort stated
42 their illness started suddenly, the rest reported gradual onset. 90% of respondents are female
43 and the majority of respondents recall an infection as the trigger for their ME/CFS. 97% of
44 participants are unable to work full time or continue in study, most cannot work part-time either.
45 100% report difficulty attending social events. Approximately half of the participants are unable
46 to walk outside or undertake tasks such as shopping, whilst the other half report difficulties
47 with such activities. Most receive care support from family members, while 1/3rd report having

1 a funded care assistant. Many participants do not receive funded care support. Most
2 participants receive some form of disability benefit, but many report difficulties and delays
3 accessing such payments. Nearly 2/3^{rds} of the participants report a lack of social care support,
4 other than disability benefits.

5 Many people with severe ME/CFS report anger and frustration engaging with the medical
6 profession, a significant proportion find getting a diagnosis an arduous task and are reporting
7 that doctors have little knowledge of the illness. Some participants report positive experiences,
8 often building supportive relationships with their general practitioner. Many of the sample
9 reported moving from moderate illness status to severe over the course of the illness. A
10 number of severe patients have tried therapies such as cognitive behavioural therapy (CBT),
11 graded exercise therapies (GET) and variants of pacing therapy (PT). Many have also tried
12 psychotherapy or counselling, physiotherapy and alternative therapies. GET ranked highest
13 for negative responses, followed by CBT and physiotherapy. Responses to CBT and
14 physiotherapy are mixed, whilst pacing receives the largest positive response rank.

15 Most severe patients take regular medications to control symptoms, often pain killers, sleep
16 aids and anti-depressants, as well as a wide range of other drugs. Severe patients often find
17 it difficult to attend hospital visits and are extremely fearful of hospital in-patient care, even for
18 non-ME/CFS related health complaints, due to a lack of understanding and accommodation of
19 their needs. Despite this, many patients want regular follow-up and monitoring, particularly
20 specialist care, including dealing with symptoms such as pain and postural orthostatic
21 intolerance (POTS). Many of the participants have had ME/CFS for a long time, they follow
22 research and developments in the field, some attempt to communicate these to GPs. The
23 majority appear pragmatic, they know that understanding for the illness is progressing. Most
24 call for more research, particularly biomedical research, and a move away from a focus on
25 psycho-social factors. Most of the cohort report finding it difficult to secure social care and most
26 say they want enhanced support and GP involvement with disability benefit claims and illness
27 management, particularly the use of technologies such as tele-consultations from home.
28 People with severe ME/CFS are generally keen to inform doctors, social workers and carers
29 about their specific needs.

30 Many people with severe ME/CFS remain unwell for years or decades. Only 8% of our cohort
31 report improving over time. Of the rest, approximately 1/3rd remain stable and 1/3rd continue to
32 deteriorate, whilst others report fluctuations. Many participants experience extremely
33 debilitating symptoms, some remain bedbound and unable to walk, the majority are
34 homebound. Many of the participants report difficulties standing (orthostatic intolerance),
35 aversion to stimuli such as light, noise or enhanced cognitive load. Many of the participants
36 report vulnerability to crashing or deterioration after emotional or physical effort and stress.
37 Participants report that pushing beyond limits, often via participating in graded exercise therapy
38 or physiotherapy, results in some type of negative symptom response that can last from days
39 to months, and many report associated psychological distress with such relapses.

40 **2.1.4 Discussion**

41 High levels of distress, frustration and anger, aimed mostly at the medical profession are
42 reported. ME/CFS is a complex illness that is poorly understood among medical and social
43 care professionals. Participants reported challenges in accessing medical care, such as the
44 need for home-visits. Participants want medical practitioners to receive more training on the
45 illness and its impact on quality of life. Participants want earlier diagnosis and more access to
46 specialist secondary medical care and social care.

47 Participants at the severe end of the ME/CFS spectrum report little benefit from treatments
48 such as CBT or GET. Many report pacing therapies as their treatment of choice. These findings
49 conflict with evidence from randomized controlled trials but are in line with evidence from
50 patient surveys. However, severe ME/CFS patients are often absent from trials of CBT or GET,
51 and there are very few trials of pacing therapies. The most negative comments and stories

1 from participants are related to psycho-behavioural treatments, particularly Graded Exercise
2 Therapy. A number of respondents expressed strong views that these treatments are not
3 appropriate and are harmful. Some patients state that CBT helps deal with the psychological
4 stresses that are part of chronic illness, particularly anxiety and depression. Pacing appears
5 to ameliorate symptoms or prevent deterioration, but most severe patients report little
6 improvement in their illness status over the long-term.

7 Many participants perceive that the medical profession view ME/CFS as a predominantly
8 psychological illness. Many suggest that there is too little focus on existing or new biomedical
9 research on causes and pathogenesis of the illness. Despite their poor health, many
10 participants remain optimistic and are actively engaged in following developments in the field,
11 such as the forthcoming NICE updating of treatment recommendations in the guideline.
12 Involving patients with severe ME/CFS in health care planning is difficult given the limitations
13 the illness imposes on them, thus innovative inclusion methods should be considered or
14 designed to involve this sub-set of ME/CFS patients. Doctors and allied health professionals
15 should adopt a flexible and concordant approach when dealing with these patients. Most
16 people with severe ME/CFS want to form better working relationships with their primary care
17 physician and secondary care specialists.

18

2.2¹⁹ Introduction

20 The prevalence of ME/CFS in the general UK adult population is around 0.5% (NICE, 2007;
21 Nacul et al., 2011), however higher rates of 1-2% are sometimes cited. It is estimated that
22 approximately 25% of people with ME/CFS experience severe symptoms or illness
23 presentation (Strassheim et al., 2018). Severely afflicted patients are often housebound and/or
24 bedbound for large amounts of time and have more intense, diverse and persistent
25 symptomology, including pain, fatigue, malaise after minimal exertion, intolerance to light or
26 noise, and cognitive complaints (Pendergrast et al., 2016). Housebound patients are often
27 socially isolated, are unable to work or continue in education, and may experience anxiety and
28 depression. This group within the wider ME/CFS population represent the most challenging
29 cases of the illness. They are difficult to access from a research perspective. Expert knowledge
30 and careful consideration are needed to engage these patients. In a study of several chronic
31 diseases including cancer, stroke, schizophrenia, and renal failure, patients with ME/CFS have
32 the lowest median quality of life (Falk Hvidberg et al., 2015). Patients with severe ME/CFS
33 report lower functional abilities than patients with Type II diabetes mellitus, congestive heart
34 failure, multiple sclerosis, and end-stage renal disease (Buchwald et al., 1996; Pendergrast et
35 al., 2016).

36 The guideline will be aimed at supporting health and social care professionals, including those
37 working or providing input into educational and occupational health services, commissioners
38 and people with suspected or diagnosed ME/CFS, their families and carers and the public.
39 Specific consideration will be given to people with severe ME/CFS symptoms.

40 NICE acknowledge that any involvement from adults with severe ME/CFS symptoms needs to
41 occur within an ethical framework in which the most severely afflicted participants are offered
42 an opportunity to participate in the development of appropriate treatment guidelines for this
43 illness. NICE recognize that this group of participants needs to be afforded an opportunity to
44 provide feedback about the medical and social care they receive in accordance with NICE
45 Patient and Public Involvement Policy.

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

1 **2.3 Aim of the project**

2 The aim of this project was to recruit and explore the opinions of people who have severe
3 ME/CFS so that their perspectives informed the development of the new guideline

4 This project utilises an exploratory study design to engage patients with severe ME/CFS in
5 order to offer the Guideline Committee insights into the needs and perspectives of adults with
6 severe ME/CFS symptoms, including providing high quality data on issues of importance to
7 those severely affected by ME/CFS. The project is specifically tailored to take account of the
8 unique needs of patients with severe ME/CFS. Adults with severe ME/CFS symptoms are an
9 underserved group with symptoms that may result in being confined to their homes, and for
10 some, being bedbound for long periods of time.

11 **2.3.1 Research objectives**

12 The key aspect of the project was to give people with severe ME/CFS the opportunity to
13 provide insight about their perspectives on specific questions and issues identified by the
14 committee based on the guideline scope. The following topics were identified from the
15 guideline scope: Identification and assessment before diagnosis, diagnosis of ME/CFS,
16 management of ME/CFS, monitoring and review, information, education, and support for
17 people with suspected or diagnosed ME/CFS and their families and carers, and information,
18 education and support for health and social care professionals.

19 **2.4 Background**

20 **2.4.1 Difficulties getting a diagnosis**

21 Many sufferers anecdotally report problems getting an early diagnosis and appropriate medical
22 care. We conducted a literature search and found scant research on 'diagnosis of ME/CFS',
23 specifically how long it takes patients to get a diagnosis and the process patients go through
24 to get a diagnosis. One study using general practice data shows that ME/CFS patients present
25 to GPs with characteristic symptoms years or decades before receiving a confirmatory
26 diagnosis (Collin et al., 2017). Our review of research studies and patient surveys revealed
27 consistent themes. ME/CFS patients often take years to get a confirmatory diagnosis,
28 physicians are reluctant to diagnose and/or lack confidence in diagnosis and treatment as a
29 result of inadequate training and limited clinical exposure. A GP survey by Bowen et al. finds
30 that 48% of GPs do not feel confident with making a diagnosis of CFS/ME (Bowen et al., 2005).
31 Raine et al. found that UK GPs negatively stereotype ME/CFS patients with having
32 'undesirable traits' that cause frustration for doctors (Raine, 2004). One Swedish study found
33 that physicians view ME/CFS as a condition below 'disease status' with views that patients are
34 illness-focused, demanding and medicalizing (Asbring and Narvanen, 2003). There are
35 conflicting models of the illness that generate confusion and acrimony for doctors and patients.
36 As a result, despite patients reporting that getting a diagnosis is the single most helpful event
37 for them in managing their condition (Drachler Mde et al., 2009), many sufferers turn to online
38 patient groups for support, disengage with traditional medical care and attempt to manage their
39 condition without medical support.

40 **2.4.2 Difficulties defining and diagnosing severe ME/CFS**

41 There are no clear biomarkers to easily aid a physician in the diagnosis of ME/CFS; often a
42 diagnosis of the condition is made after excluding other possible causes for the patients' fatigue
43 and other symptoms (Komaroff, 2015; Fischer et al., 2014). In addition, different research
44 groups use different diagnostic criteria to assess ME/CFS status (Nacul et al., 2017), thus we
45 observe fluctuating prevalence rates in the literature – and a knock-on impact on diagnosis of
46 ME/CFS at the clinic level (Geraghty and Adeniji, 2019). In an illness that is hard to diagnose

1 and mimics generalised fatigue, which is a characteristic complaint in many other chronic
 2 illnesses or mental health states, such as depression, severe ME/CFS patients might represent
 3 the 'clearest' cohort of ME/CFS. However, researchers and clinicians do not have a universally
 4 accepted instrument to assess severity. Researchers in this field use a wide range of scales
 5 to assess symptoms – the *DePaul Symptom Survey* (Jason et al., 2015) and the *Chalder*
 6 *Fatigue Scale* (Chalder et al., 1993) measure symptom profile and fatigue. Researchers also
 7 use quality of life scales, mostly SF-36 to assess functional status and disability levels (Ware
 8 et al., 2007), but none of these instruments offer clinicians an accessible or simplistic ME/CFS
 9 severity-rating or tool to use in clinical settings.

10 2.4.3 Lack of focus on severe ME/CFS

11 Despite almost 25% of all sufferers being classed as severe (Group, 2002), it is estimated that
 12 as little as 0.5% of the entire ME/CFS literature base covers 'severe ME/CFS' (Abbot, 2014).
 13 Patient charity groups have raised concerns about the neglect of this under-served patient
 14 group. A 2002 Chief Medical Officer's Report highlighted this finding and the lack of focus on
 15 those patients that are bedbound or housebound "*Severely ill are severely overlooked; just*
 16 *ignored and invisible*" Section 2.3.1 (Group, 2002). Action for M.E.'s 2014 '*M.E. Time to deliver*'
 17 patient group survey report found that:

- 18
- 19 • 96% of respondents with severe M.E. said they had stopped or reduced household
- 20 tasks
- 21 • 95% had stopped or reduced social contact
- 22 • 74% require full or part-time care
- 23 • 70% were no longer able to leave their home independently.

24 An international study looked at adults (18 years or over) with ME/CFS who are confined to
 25 their homes due to severe symptomatology compared with non-housebound sufferers
 26 (Pendergrast et al., 2016). The researchers used the DePaul Symptom Questionnaire to
 27 assess ME/CFS symptoms and the SF-36 to measure health impact on physical/mental
 28 functioning. Findings indicated that the housebound group (severe sufferers) represented one
 29 quarter of the sample and were significantly more impaired with regards to physical functioning,
 30 bodily pain, vitality, social functioning, fatigue, post-exertional malaise, sleep, pain, neuro-
 31 cognitive, autonomic, neuro-endocrine and immune functioning compared to individuals who
 32 were not housebound (Pendergrast et al., 2016). Understanding the differences between
 33 housebound and not housebound groups holds implications for doctors and health planners.
 34 Important patient characteristics are extracted from the above study and are summarised in
 35 Table 1.

36 **Table 1: Characteristics of international cohorts of ME/CFS patients**

Key Characteristics	UK Newcastle	US DePaul	EU Norway 1*
No. of Participants	100	216	175
Female v Male	85% f.	84% f.	87% f.
Caucasian vs. other ethnicity	99% c.	98% c.	99% c.
mean age	45	52	43
Receive disability	30%	57%	84%
Currently working *full time or part-time	37.5%	13.5%	10%
University or college Qualifications	50%	75%	50%

1 1. **Two samples quoted in study (we used 1 sample, both are similar), includes homebound and not.*

2

3 Table 1 demonstrates a number of important characteristics of severe ME/CFS sufferers. The
4 majority of sufferers appear to be female, which is in line with epidemiological studies that
5 continually show a higher female ratio. The average age of participants is in the mid-40s.
6 Caucasians appear over-represented in research studies, as are college graduates and
7 professionals. Whilst around one in ten severe sufferers continue in employment in the US or
8 Norway, almost one third of the UK cohort continued to work full-time or part-time. The UK
9 cohort held the lowest level of disability benefits claimed across the three international cohorts,
10 perhaps exemplifying challenges UK sufferers face accessing social care support.

11

12 **2.5 Study methodology**

13 **2.5.1 Survey rationale**

14 Prof. Newton and colleagues at Newcastle University undertook a study to define the
15 prevalence of severe CFS/ME and its clinical characteristics in the North East of England
16 (Strassheim et al., 2018) – from 483 questionnaire packs requested only 63 were returned by
17 patients, showing how difficult it is engage patients with severe ME/CFS in research studies.
18 These patients have very specific needs and difficulties, such as difficulties with cognitive
19 function and extreme fatigue. With this in mind, standard face-to-face interviews are extremely
20 difficult to conduct, requiring home visits that can be difficult for people with severe ME/CFS.
21 In order to reach people with severe ME/CFS in the timeframe of the project, between June
22 2019 and December 2019, we opted for a survey. It was not feasible to undertake face-to-face
23 interviews with enough ME/CFS patients given their geographical spread, the impact this might
24 have on bedbound patients and the limited resources available to us. We considered tele-
25 interviews with ME/CFS patients but later rejected this option after preliminary feedback from
26 people with ME/CFS that interviews of any length of time, between 15 minutes to 1 hour, would
27 cause considerable fatigue and increase other symptoms. Given our research objectives we
28 opted for an online survey. The benefits of an online survey were two-fold: first, it allows access
29 to a wide number of responders located across England and Wales; second, it could be
30 completed by respondents in their own time, thus minimising the burden on patients with
31 severe ME/CFS.

32 **2.5.2 Questionnaire development**

33 Following discussions with the guideline committee and a review of relevant literature, we
34 formulated a survey questionnaire with a series of questions that covered our main research
35 objectives (page 8). These objectives aligned with the guideline scope and the committee's
36 identified areas of special interest for patients with severe ME/CFS (Appendix 1). We opted for
37 a series of predominantly semi-structured questions to allow respondents to elaborate on their
38 experiences of care, diagnosis and management. Each question was specifically structured to
39 allow respondents to provide their responses with the minimum of effort in terms of cognitive
40 load or time-effort. The questionnaire could be completed by the patient, with the help of a care
41 assistant or family member if needed – thus it allowed the patient to complete the questionnaire
42 in their own time. This, we believed, would cause the minimum of stress and would allow for
43 the best possible results. *SurveySelect* software (2019 TM) was used to administer the survey.
44 Table 2 provides an overview of the link between scoping areas identified by the guideline
45 committee and our research objective areas of interest and key areas of focus within our survey
46 instrument.

1 **Table 2: Overview of research areas, objectives and linked details attended to in**
 2 **survey**

Scoping Area	Research Objective: Exploring	Explored in Questionnaire with patients with severe ME/CFS
Identification and assessment before diagnosis	1. <i>Participants' views on illness identification and assessment before diagnosis</i>	Their experience of: <ul style="list-style-type: none"> • Initial illness <ul style="list-style-type: none"> ○ Being believed • Initial illness and impact on life (including family, friends, school, college, university, work) • Initial contact with a health and social care professional about symptoms • What worked well • What didn't work well
Diagnosis of ME/CFS	2. <i>Participants' experiences of diagnosis of ME/CFS</i>	Their experience of: <ul style="list-style-type: none"> • Continuing illness and severe ME/CFS • Continuing illness and impact on life (including family, friends, work, college, university) • Contact with health and social care professionals to get a diagnosis, approach taken • Time to get a diagnosis • What worked well • What didn't work well
Management of ME/CFS	3. <i>Participants' experiences of management of ME/CFS</i>	Their experience of: <ul style="list-style-type: none"> • Interventions (benefits and harms) <ul style="list-style-type: none"> ○ For ME/CFS and symptomatic relief ○ Outcomes: benefits and harms ○ If offered interventions have not been taken up, why • Contact with health and social care professionals and services <ul style="list-style-type: none"> ○ Are your basic needs met? ○ Co-ordination of care ○ Referral to specialists ○ Hospitalisation ○ Involvement in decision making <ul style="list-style-type: none"> ▪ Feelings of control and choice ○ Access to services <ul style="list-style-type: none"> ▪ Access to appointments and getting to appointments (distance to clinics) ▪ Home visits ▪ Support services (mobility aids) • What worked well

		<ul style="list-style-type: none"> ○ Experience of recovery if appropriate ○ Experience of reintegration if appropriate (for example, work, friendship groups) ● What didn't work well <ul style="list-style-type: none"> ○ Experience of relapse
Monitoring and review	<i>4. Participants' experiences of monitoring and review of ME/CFS</i>	<p>Their experience of:</p> <ul style="list-style-type: none"> ● Continuing care ● Follow up <p>Who does this? Information about prognosis or future planning</p>
Information, education, and support for people with suspected or diagnosed ME/CFS and their families and carers	<i>5. Participants' experiences of information, education, and support for ME/CFS as well their families and carers</i>	<p>Their experience of:</p> <ul style="list-style-type: none"> ● Accessing information, education and support <ul style="list-style-type: none"> ○ What was useful and what wasn't ○ Information and support networks
Information, education and support for health and social care professionals.	<i>6. Participants' views on Information, education and support for health and social care professionals.</i>	<p>Their experience of:</p> <ul style="list-style-type: none"> ● Knowledge of the health and social care professionals <ul style="list-style-type: none"> ○ Where do you think they get information from ● Health and social care professionals' attitude to ME/CFS ● Do they have the ability to provide support and what has been useful

1

2 2.6 Sample selection

3 The respondents involved in this survey were recruited in two phases, a pilot test phase and
4 an open survey phase. To ensure confidentiality, respondents were given questionnaire
5 questions with no names or signature input requirements.

6 2.6.1 Pilot test phase participant recruitment

7 In this first phase we invited a select number (n=10) of people with severe ME/CFS to
8 participate. This group were drawn from a mix of direct contacts between Dr. Geraghty and a
9 number of patients and contacts with a number of patient advocacy organizations, asking them
10 to select a small number of their members to trial our survey (*a convenience sample*). This
11 group were given access to the online survey and were encouraged to give feedback on the
12 survey and the process of completing it. Our aim was to test the survey, to ascertain if it was
13 viable, to see if patients with severe ME/CFS had any specific problems or concerns

- 1 completing the questions. Feedback received proved valuable insights into how respondents
- 2 might experience survey completion.

3 **2.6.2 Main purposeful recruitment**

4 Given the short time-frame available to our team to conduct this project (between June and
5 December 2019) we opted for a *purposeful sampling* strategy. Our survey was advertised via
6 social media (using Twitter and University of Manchester website). Given the scope of NICE
7 guidelines, we restricted our survey to patients living in England and Wales. Patient advocacy
8 and charity organizations were also contacted and asked to advertise our survey to their online
9 platforms. This dual approach of open-survey and targeting of charity groups, helped widen
10 coverage of our survey to members of ME/CFS patient representative groups and non-
11 members. To take part in the survey participants had to meet the following inclusion criteria:

- 12 1. Have a confirmed diagnosis of ME/CFS from either a GP or NHS specialist.
- 13 2. Be adults age 18 or over residing in England or Wales.
- 14 3. Self-identify as suffering from 'severe ME/CFS'.
- 15 4. Consent via signing of a consent form & reading of a participant information sheet.

16 **2.6.3 Target sample**

17 Purposive samples are commonly used for of non-probabilistic sampling and their size typically
18 relies on the concept of "*saturation*," or the point at which no new information or themes are
19 observed in the data. Guest et al. suggest saturation occurs within the first twelve interviews
20 (Guest et al., 2006), however other qualitative methodologists suggest 20+ respondents to
21 establish credible findings (Hagaman and Wutich, 2017). We opted for a survey methodology
22 over interviews and thus aimed for higher numbers of respondents, circa 50 complete
23 responses to garner a credible sample, before beginning data analysis.

24 **2.7 Research ethics and confidentiality**

25 Formal ethical approval for this project was sought and granted from the University of
26 Manchester Research Ethics Committee (2019-7763-12089). Given the nature of the project
27 – a commissioned project not recruiting patients from the NHS, NHS ethical approval was not
28 required. Our project involved a range of ethical considerations. Our team highlighted the need
29 for ensuring respondent confidentiality and awareness of the specific needs of patients with
30 severe ME/CFS as important ethical considerations. In undertaking this project, all members
31 of the research team adhered to the core principles of research ethics outlines in the Belmont
32 Report (1979):

33

- 34 1. Respect for research participants, their health status and confidentiality.
- 35 2. Beneficence, awareness of participants circumstances and limitations and efforts to
36 minimize distress in the interview process.
- 37 3. Justice, fairly and accurately representing the feedback given by participants.

38 **2.7.1 Disclosure and informed consent**

39 All potential participants were asked to sign a Consent Form in order to begin the survey
40 (Appendix 2). In addition, each participant received a detailed Participant Information Sheet
41 (PIS) (Appendix 3) that we attached to the beginning of the survey and respondents had to
42 positively click that they had read this before proceeding to answer survey questions. This PIS
43 was downloadable as a PDF for participants to retain. We were particularly concerned that

1 completing the survey might be an onerous and exhausting task for people with severe
2 ME/CFS, thus we gave detailed instructions that the survey could be completing over different
3 days and we gave instructions for responders to get help if needed and not to endure symptom
4 flare. Participants were encouraged to contact a member of our research team if they have
5 any prior questions or concerns, or problems, completing the survey. We received many
6 emails, discussed in our findings section.

7 **2.8 Data analysis**

8 **2.8.1 Data collection and synthesis**

9 Data processing entailed data collection via survey responses, data cleaning to exclude
10 ineligible participants, thematic analysis of responses, data coding, analysis and interpretation.
11 A database of responses was extracted from our *SurveySelect* programme and exported to
12 Microsoft Excel. Both Excel and Microsoft Word were used to tabulate data and undertake
13 analysis. We utilised a mix of *Thematic Content & Narrative Analysis* to arrive at meaningful
14 interpretations of the collected data (Glaser and Strauss, 2017). Our aim was to draw out
15 recurrent themes (frequent response or patterns), whilst allowing for individual narratives to
16 also emerge, that accurately and fairly represented the views of patients with severe ME/CFS.
17 The process of data synthesis and interpretation involved:

- 18 ▪ Two researchers undertook data collection and collation.
- 19 ▪ Responses from questionnaires were collated into a project file by exporting a dataset
20 from *SurveySelect* tool to MS Excel.
- 21 ▪ Each researcher read over responses allocated to them.
- 22 ▪ Text responses to key questions or series of questions were categorised as relevant to
23 our specific research objectives. Themes were identified and coded applying Ryan and
24 Bernard's (2003) repetition approach to identify themes (Ryan and Bernard, 2000).
- 25 ▪ A senior member of the research team with expertise in qualitative methods reviewed
26 completed data outputs to assess consistency and the quality of analysis.
- 27 ▪ Both data sheets were merged into a single report using headlines to cover the main
28 themes to emerge from the overall dataset (creating a coherent narrative that includes
29 quotes from respondents to support reported themes).
- 30 ▪ All members of the research team approved the data entries in our report.

31 **2.9 Findings**

32 **Participant characteristics and classification**

33 Approximately 1600 persons clicked on our survey link and opened the survey. From these
34 343 started the survey, 124 completed, 219 did not complete, 60 self-reported clearly as
35 severe, 27 self-reported moderate-to-severe, 33 self-reported as moderate-to-mild and 4 had
36 no confirmed diagnosis. For the purposes of our analysis we decided to take only those 60
37 that had severe illness status with a confirmed diagnosis of ME/CFS. Respondents' ages
38 spanned 19 years old to 80 years old, with a mean age of 50 and SD = 13.4 years (Figure 1).
39 The majority of the respondents were female (50/60), 9 were male and 1 classified themselves
40 as non-binary.

41 **Participant responses**

42 In this section, we present participant responses with reference to identified themes and sub-
43 themes. We include direct quotations from participant responses to expand on our summarised
44 themes and to contextualise the meaning behind these themes. We use participant code
45 identifier numbering to differentiate respondents.

1 2.9.1 Participants' views on illness identification and assessment before 2 diagnosis (including current status)

3 Illness onset

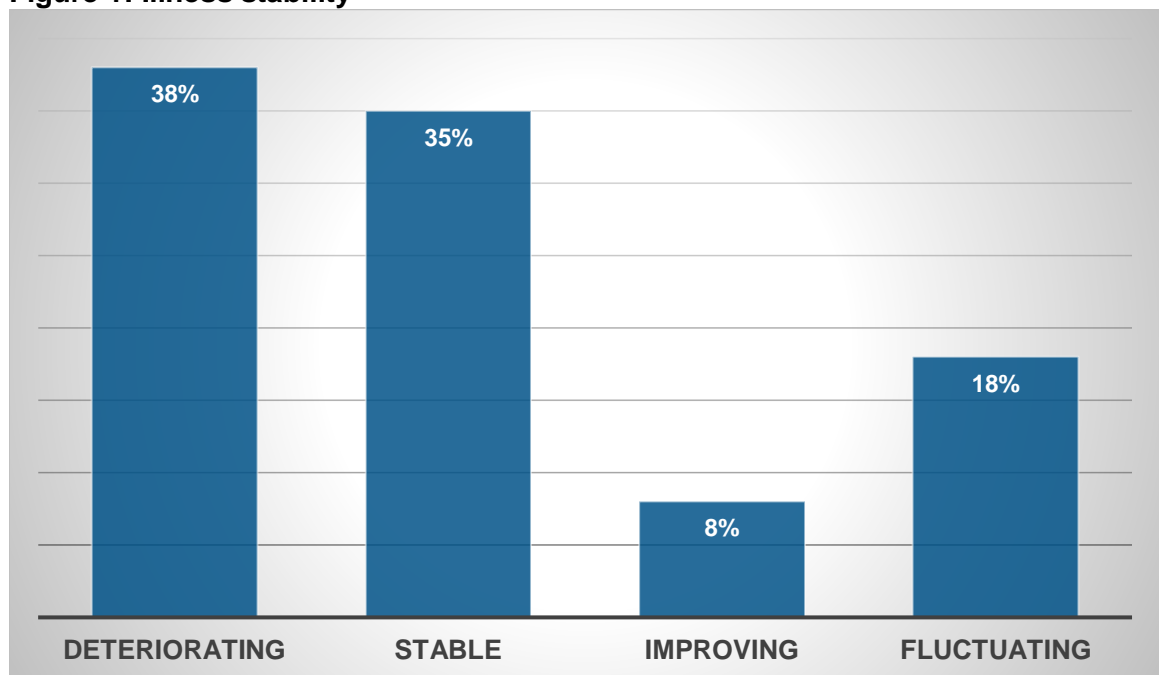
4 Almost two thirds of respondents indicated that the illness started suddenly (38/60), while the
5 rest report the illness had a gradual onset of worsening symptoms (22). Many respondents
6 report a singular infection event, such as glandular fever, as the trigger, and gradual worsening
7 of their condition in the subsequent months and years.

8 Illness stability/progression

9 We asked early on in our survey questionnaire whether patients felt their illness was stable
10 and whether they had periods of remission or improvement (we ask a similar question in Q39
11 but include symptom fluctuations after the treatment section – it is reported below in Section 4
12 with similar results as reported here).

13 38% of respondents stated their illness had generally gotten worse over time (Figure 1). 35%
14 remain stable (moderate-to-severe) over time, 18% tend to fluctuate. Only 8% report generally
15 improving over time.

Figure 1: Illness stability



16

17 The proportion of self-reported fluctuating condition also increases with decreasing severity of
18 the illness, just like the self-reported improving condition. This can be due to the fact that
19 participants experience ill health without any periods of relief, meaning that their condition
20 cannot fluctuate between bad and better, while with decreasing severity it is easier for the
21 condition to fluctuate between bad and better.

22 Education or work status with illness

23

- 1 • 58 out of 60 respondents said their ME/CFS is preventing them from continuing in work
2 or study.
3 • 3 said they could continue to work but on a reduced level (partially).
4 • 1 respondent gave a 'no' answer and 1 gave a 'nondescript' answer.

5 **Social event participation**

- 6
7 • 59 out of 60 respondents said they were unable to attend social events.
8 • 1 stated that they are partially impeded.
9 • Social isolation is a key indicator of severe status.

10 **Outside activities (walking or shopping)**

- 11
12 • Half of respondents (30) report that they are not able at all to go outside for any
13 activities.
14 • The other half (30) report that they are only able to do so partially for limited periods,
15 often minutes only.

16 **Support from family members**

17 Three quarters of the respondents (44) state that they receive support from family members,
18 while a quarter (15) state that they do not receive such support, either because the family are
19 unable or unwilling to provide it.

20 **Support from a care assistant**

21 One third (19) of the respondents state they have a care assistant, typically provided to them,
22 but in some cases paid for by them. 1/3rd (17) state they don't have a care assistant, but
23 typically would like to have one if it could be provided. Around 1/3rd (21) state that their carer
24 is one or multiple family members. 1 reports being on a waiting list for a carer, 1 has had a
25 carer in the past and 1 can't find a suitable person to provide care because of too many special
26 requirements.

27 **2.9.2 Participants' experience of diagnosis of ME/CFS**

28 **Time to first diagnosis**

29 We found large variability in the time it took to get diagnosed, ranging from 2 months to 21
30 years. The mean value excluding extreme cases of over 10 years, is 24 months (SD=22.6
31 months). There are 3 extreme cases (11 years, 11 years and 21 years).

32 **Getting a diagnosis: GP, specialist or other**

33 The majority of respondents indicated that their diagnosis was given by a medical professional:
34

- 35 • GP (23),
36 • Specialist (13) and
37 • ME/CSF clinic (8)
38 • Consultant (7)
39 • Other: neurologist 3, paediatric specialist 3, immunologist 2, rheumatologist 1.
40

1 Experiences of getting diagnosed

2 About a third of respondents indicated that they were reasonably satisfied with the process of
3 getting ME/CFS diagnosed (23).

4 1577773 *“most doctors haven't heard of myalgic encephalomyelitis. they certainly haven't*
5 *been trained to know the tests to diagnose it. I was lucky I found one.”*

6 The most frequent complaint among others related to the process taking too long (25) and not
7 feeling believed by doctors (14). 8 indicated that they had to suggest the diagnosis to the GP
8 or specialist, while 5 indicated that they had to get the diagnosis privately. 7 indicated that they
9 were diagnosed initially with depression, 1 with anaemia, 1 with anxiety and 1 with
10 hypothyroidism.

11 *Our numbers do not add up to 60 because most respondents had multiple complaints.

12 1560014 *“I was made to feel it was somehow my fault I was still ill; At that time we were*
13 *told that you couldn't have ME if you had underactive thyroid.”*

14 1574885 *“He seemed dismissive about anything that fell outside of the strict boundaries*
15 *he had regarding cfs/me.”*

16 1577409 *“My then GP was very nice, she said she didn't believe in ME but that in my*
17 *case she was prepared to diagnose ME.”*

18 1577978 *“NHS was worse than useless. Left me without a correct diagnosis for 2 years.*
19 *As a result I had to keep working full time and got gradually worse and worse.*
20 *They accused me of being depressed which was ironic as that was the only*
21 *symptom I didn't have. In the end I had to see a private physician, who was the*
22 *first doctor who actually listened to me.”*

23 Many respondents said that getting a diagnosis gave them a sense of relief and legitimacy.

24 1578853 *“Good to be validated. The dx of ME as it was called back then seemed to*
25 *legitimise me in the eyes of doctors! Myalgic encephalomyelitis sounded*
26 *serious to them...”*

27 Others talked about the practicalities of a diagnosis, for example making claims for benefits or
28 private insurance claims to support them whilst unwell.

29 1579148 *“I paid for a specialist diagnosis as I needed this to make an insurance claim.”*

30 Factors that patients felt helped in getting a diagnosis

31 The main factor is the positive attitude from the GP or specialist setting the diagnosis. 14
32 respondents mentioned the GP being informed and educated as an important factor, 11
33 indicated the importance of the GP being supportive and listening to their concerns.

34 1582572 *“Being able to see a GP who had significant knowledge about the illness. Many*
35 *doctors had failed to spot it in the months/years before then despite me being*
36 *seriously ill.”*

37 The second major factor was the patient's own efforts – doing research and asking questions
38 of medical professionals (16), including perseverance in trying to persuade the doctor that this
39 is the right diagnosis. Secondary factors were paying to get a diagnosis privately (7) and having
40 clear symptoms (7). 2 respondents mention having a family history of the illness as a facilitating
41 factor, and 1 mention being a child as a facilitating factor, since this draws more sympathy from
42 the doctors and makes them put more effort in the investigations.

1 1574735 *“Going to the doctor many times to complain that I couldn't function, although
2 this wasn't a positive thing because it went on for three years and was very
3 upsetting because they were very rude to me. They got fed up with me and the
4 nurse would sulk and be angry when I went to see her for the blood tests that
5 the ME clinic asked for, for the referral....Having a local ME service with a GP
6 who knew the diagnostic criteria for ME was helpful because it meant I could
7 get a diagnosis.”*

8 **Factors that delayed diagnosis**

9 A quarter of the respondents said there were no factors that delayed their diagnosis. The main
10 factor indicated in delays (about half of the sample) is uninformed GP or specialist (32).
11 Secondary factors are ambiguous symptoms (5) and NHS waiting times (5). Some
12 respondents complained about not being believed as a factor (3) and the lack of clarity in the
13 diagnostic criteria (3). The lack of an ME/CFS specialist locally is also mentioned (2) as well
14 as the lack of remote support for patients who are unable to travel for investigations (2). Other
15 patients mention the lack of availability of private specialists and their own denial of the illness
16 as factors. A typical story,

17 1582331 *“If I had been taken seriously the first few times I saw a GP I could have been
18 diagnosed years earlier, and may not be so ill now. The waits between
19 appointments delayed my diagnosis massively...It took me a very long time to
20 meet a GP who suggested it, and he was not confident in his knowledge of it at
21 all. Even now when I see a different GP they know very little and sometimes
22 misunderstand basic parts of it very often.”*

23 **Physician acceptance of ME/CFS diagnosis and support of patient**

24 52 respondents said their doctor agreed with their diagnosis, while only 8 said that their doctor
25 did not agree. Less than half (25/60) said their GP was supportive and a little over half (34)
26 said their GP was unsupportive of their diagnosis and illness. 4 respondents said that they had
27 to change their GP. Patients report changing GPs either purposefully or because of life
28 circumstances and having different experiences with them.

29 **2.9.3 Participants' experience management and treatment of ME/CFS**

30 **Prescribed medications/drugs**

31 In question 19, we asked if patients had been prescribed any drugs by their doctor or a
32 specialist, specifically for their ME/CFS or related symptoms. To our surprise, given there are
33 no recommended drug treatments for ME/CFS, only 7 out of 60 respondents said 'no' to drug
34 prescriptions for their ME/CFS symptoms. Some gave a yes answer without details, but the
35 majority gave long lists of drugs taken, often spanning many years, even decades (see quotes
36 below). We were initially going to list the drugs mentioned but this became too complex given
37 lists were often long and patients talked about drugs currently taken and drugs taken in the
38 past, thus we decided to record the drug classes most often mentioned by respondents. These
39 are:

40 **Table 3: Drugs taken to treat illness/symptoms**

Most Mentioned Medications	Lesser Mentioned Medications
<ul style="list-style-type: none"> • Pain Medications – often amitriptyline, Pregabalin, Gabapentin, codeine, lyrica, NSAIDs and others 	<ul style="list-style-type: none"> • Migraine – Sumatriptan • Anti-virals – acyclovir • Proton Pump Inhibitor – Omeprazole

- **Sleep Medications** – Zopiclone and others
- **Anti-depressants** – often not named
- **Anxiety (also muscle spasm) – Diazepam**
- **Steroids – hydrocortisone**
- **Vitamin supplements – most Vit B12, Mg, Vit C**
- Anti-histamine – cetirizine
- Beta-blocker – propranolol
- Postural Orthostatic Intolerance meds (POTS) - Pyridostigmine
- Mast Cell Activation meds (MCAS) - Ketotifen

1 Some typical patient comments about drugs taken,

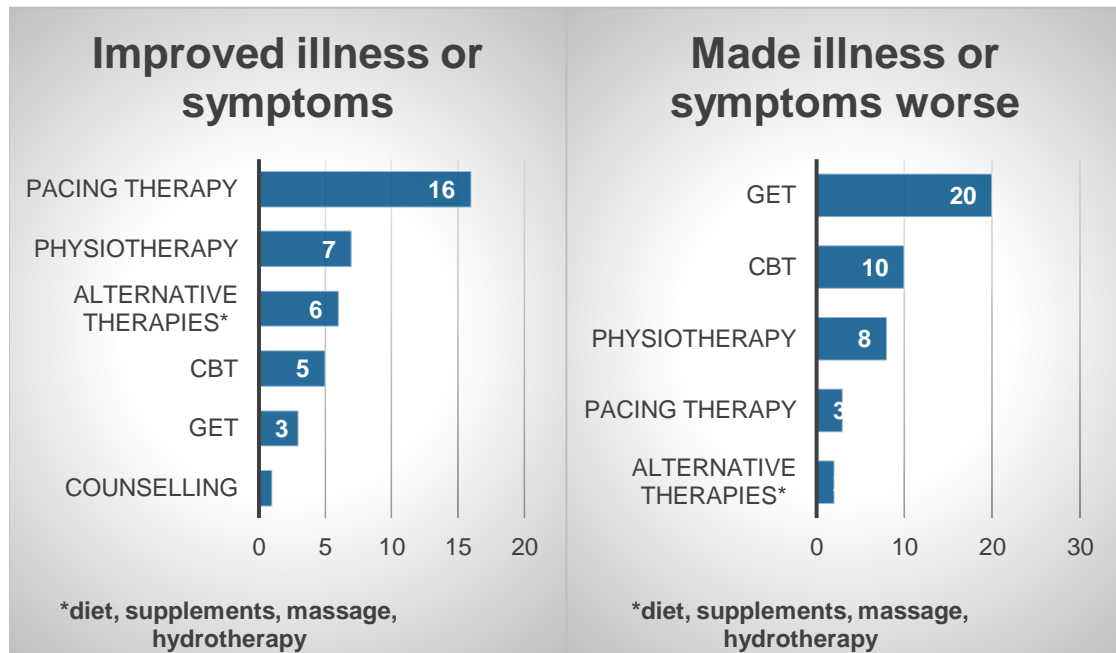
- 2 1560312 *“So many over decades. My medical drugs are an archaeological document of*
3 *fads in treatment*1. *Antidepressants of different types*2. *Antivirals -valtrex,*
4 *amantidine*3. *Pain - codeine, lycica, 4. Experimental - ivig, kutapressin,*
5 *magnesium injections, b125. Spasms -baclofen*6. *Antibiotics*7. *Steroids,*
6 *Prednisone, hydrocortisone.”*
- 7 1585039 *“Amitriptyline alleviates headaches; Baclofen reduces muscle spasm;*
8 *Clonazepam reduces muscle spasm; Gabapentin reduces shooting, stabbing*
9 *and gnawing breakthrough pain; Hydrocortisone made a big difference in my*
10 *energy levels since the day I started taking it. Lanzaprazole - helps the nausea;*
11 *Nimidopine- marked improvement in cognitive function:- ability to word-find,*
12 *improvement in vocabulary and structuring sentences. Been on it for 10 years;*
13 *Pizotifen helps to control the migraines; Sumatriptan tablets/nasal spray/IM*
14 *injection works to a degree.”*

15 **Interventions: treatments and responses**

16 We asked in questions 21 and 22, whether participants had undertaken any specific treatment
17 or been prescribed a treatment for their ME/CFS and whether such treatments made things
18 better, worse or had no change. We specifically listed CBT, GET and Pacing, as these are the
19 three most common treatments reported in the literature, but we asked respondents to list any
20 other treatments if they had tried something other than the above. Respondents gave a lot of
21 information across both questions, so in our analysis we had to combine answers across both
22 questions.

23 Results were mixed. Overall, respondents mostly gave negative or neutral responses. We
24 detail these below (Figure 3). The therapies respondents most often tried were CBT, GET,
25 Pacing Therapy (either self-directed or guided by a health professional), physiotherapy,
26 psychotherapy, counselling, and alternative medicine or therapies. It is important to note that
27 a significant number of respondents stated that they had not tried CBT, GET or other therapies,
28 or that these made no difference if tried. 23 respondents said they either did not try the therapy
29 recommended, or it made no difference. In table 3 below we list what therapies improved
30 symptoms most often or made symptoms worse most often, as reported by the number of
31 participants. We can observe an inverse relationship, that correlates, between both sides
32 (improved or worsened).

Figure 2:



Number of people reporting

1 **Cognitive behavioural therapy (CBT)**

2 Overall, responses about CBT and symptoms changes were slightly negative to neutral, 30
3 respondents out of a cohort of 60 said they had tried CBT (50%), and of these, 10 respondents
4 stated it made things worse, whilst 5 stated it helped and 15 stated it had no impact of neutral
5 change.

6 1579580 *"CBT helped with my depression. The loss of my previous life has been difficult
7 to come to terms with".*

8 1586506 *"CBT made me worse since I was taught to view my illness as a false belief,
9 and therefore repeatedly ignored post-exertional malaise until it developed into
10 permanent worsening of my symptoms."*

11 1560014 *"I dragged myself to CBT sessions because I was assure(d) it would make me
12 better...They made me permanently more ill."*

13 **Graded Exercise therapy**

14 GET in all forms generates the most negative responses. 26 respondents said they had tried
15 GET in Q21 and in Q22 20 participants reported it made them feel worse, 3 stated it helped
16 or improved symptoms and the rest had neutral change. The remainder of the 60 cohort had
17 not tried GET.

18 1560312 *"The early type was to increase activity regardless if how one functioned. I never
19 recovered back to the activity level I had when I started The 2nd type of GET
20 was to increase on when stabilised but this never worked as the disease got
21 worse the more I did."*

22 1579580 *"GET was given by a physio who was sent from Stockport. They were really
23 supportive and very knowledgeable regarding ME so exercise was very gentle.
24 I understood that I needed to move my legs and arms to prevent muscle
25 wastage."*

1 1582572 “The GET was unhelpful and reduced my overall level of activity.”

2 1586506 “GET made me worse every time I tried it - and I kept trying it throughout my
3 twenties.”

4 **Physiotherapy**

5 Responses about physiotherapy were mixed, 8 respondents stated that it made them worse,
6 whilst 7 reported it helped. Respondent 1575763 who had reported some benefit using CBT
7 for anxiety tried physio with profoundly negative comments:

8 “I saw a physio at my local NHS centre. I had gone from my bed, to my wheelchair to
9 the appointment. At the appointment she looked me in the face, repeatedly telling me
10 ‘ME is all in the mind, you’re making it up’ whilst instructing me to attempt to do arm
11 push-ups on the arm rests of the wheelchair. This was both physically and emotionally
12 damaging. I have lost all faith in medical professionals.”

13 Respondent 157563’s loss of faith in health professionals following a bad experience is not an
14 uncommon reported event. Participants that experience a distressing experience may mistrust
15 health professionals and often disengage from all formal treatments.

16 Respondent 1585039 reports moving between two types of physiotherapy for their very severe
17 ME/CFS. Their experience exemplifies how subtle changes in approach bring about differing
18 results and how understanding of ME/CFS is essential for a good relationship between
19 therapist and patient,

20 “I was bed bound and the emphasis was solely on sitting me out in a chair. This
21 approach exacerbated the ME. My physiotherapy was then taken over by a
22 neurophysiotherapist who worked with MS and Parkinson's patients. She did passive
23 and then active assisted exercises to put all the joints through their range of movement.
24 She also did chest physio which improved the oxygen levels in my body. If I was very
25 fatigued she would do massage. She came to the house three times per week for eight
26 years and over time I progressed to walking with a frame. Setbacks were frequent,
27 often due to my response to external noise as well as infection etc. After each set back
28 she would have to start again with the passive exercises and then build up again. A lot
29 of the work was on muscle memory. The physio was sympathetically done and without
30 it I would not have been able to make the progress I have.”

31 1574735 recounts a similar experience,

32 “I was given physiotherapy exercises to do by the ME/CFS clinic but these caused me
33 to crash for a month, so once I recovered I saw a non-specialist physiotherapist at my
34 university who pared down the exercises to make them more efficient, gentle and
35 manageable for me. These help with core strength, joint mobility and muscle stiffness
36 which I find beneficial in reducing musculoskeletal pains that result from my limited
37 mobility, but it does not have any impact on my ME symptoms. I have to be extremely
38 careful with it because I can only do a tiny but of very gentle physiotherapy each day
39 before it makes me feel really unwell.”

40 **Pacing Therapy**

41 Respondents rated Pacing Therapy as most useful in terms of stabilizing or improving
42 symptoms – 16 reporting improvement, while only 3 reported becoming worse using pacing.
43 We note here that respondents tried different forms of pacing therapies – Adaptive Pacing
44 Therapy (APT) similar to that used in clinical trials such as the PACE Trial, self-directed pacing
45 and other variants.

46 1576479 “Pacing is a good strategy- stay within energy envelope and very occasionally
47 bounce the boundaries to see if underlying health has improved enough to do

1 *more. I got back to normal activity levels doing this, though how my underlying*
2 *health improved is a mystery. Any treatment with an emphasis on increasing*
3 *activity to a schedule reveals that the therapist fundamentally misunderstands*
4 *the nature of ME.”*

5 1574735 *“I have been visiting NHS ME/CFS clinics 2-3 times a year since my*
6 *diagnosis, who give me advice on pacing. I find pacing helpful to minimize*
7 *flare-ups, longer-term worsening and reduce the frequency and intensity of*
8 *crashes, but it has not led to any improvement in symptoms overall.”*

9 Some patients noted the difference between pacing and GET.

10 1579730 *“The pacing course encouraged increasing activity, which for me wasn't*
11 *appropriate. I deteriorated to bedbound within 18 months of diagnosis.” Pacing*
12 *requires staying within limits and gentle testing of boundaries, whereas GET*
13 *requires pushing past limits and increasing activity steadily.”*

14 **Counselling and alternative therapies**

15 Many patients reported that they had tried counselling. Responses were generally mixed for
16 counselling, some found it of benefit, while others reported no change. For example:

17 1578170 *“Counselling helped me deal with some of the frustration and loss involved with*
18 *having a chronic illness but it did not improve my symptoms. In fact, sometimes*
19 *the cognitive effort of counselling sometimes left me feeling very unwell*
20 *physically for several days after an appointment.”*

21 The types of alternative therapies most often mentioned were massage therapy, hydrotherapy,
22 dietary changes, including supplements, and meditation/yoga. Results were generally mixed
23 to neutral, having overall positive to neutral impact on symptoms.

24 **Treatment refusal**

25 In question 23, we asked patients if they had ever refused a treatment and why. 35
26 respondents out of 60 said they had refused treatment or would again if treatment were offered
27 (22 never refused and the rest stated non-applicable). Common reasons given for not trying
28 were 'being too ill' or deeming the treatment 'inappropriate'. Unsurprisingly, severe patients
29 are often too unwell to attend out-patient treatments such as CBT or GET. Respondent
30 1577539 writes, *“GET...assessed as too ill for it. I couldn't even have got to the appointments*
31 *often enough.”* The same patient did try physiotherapy privately and found it of some help.

32 **Specialist treatments: in-hospital care and outpatient**

33 In question 24, we asked if patients ever had any other specialist treatment, with a list of
34 examples such as tube-feeding or IV fluids, treatments one might expect patients to potentially
35 experience at the more severe end of the ME/CFS spectrum. 46/60 said no. From those who
36 gave answers the following treatments were ranked:

- 37 ▪ Tube Feeding (or variants of feeding support)
- 38 ▪ IV Fluids
- 39 ▪ Speech Therapy

40 *we suspect if we listed more examples, respondents may have added more items to their
41 responses.

42 **In-hospital stays**

43 28 respondents said they had no hospital in-stays, 32 recorded at least one hospital in-stay,
44 mostly for non-ME/CFS related health complaints, cancer treatment, surgery, infections and
45 investigations.

1 A number of respondents reported being anxious or fearful of needing to go into hospital,
 2 expressing the view that hospital staff would not understand their ME/CFS and needs.
 3 Respondent 1580186 writes,

4 *"No - its my biggest fear. If I had a non-terminal illness (I would chose to die*
 5 *rather than cope with undergoing something like chemo for instance, I could*
 6 *never cope with it) that was very painful, I have no idea how I would cope with*
 7 *a hospital admission - people around me drains me within minutes."*

8 1582572 *"I have avoided all in-patient stays including checking myself out."*

9 1575763 *"I do not feel safe in hospital. I avoid A&E and hospital at all cost. My illness is*
 10 *treated poorly."*

11 1560312 *"Only 1 hospital stat as hospitals are dangerous places for PWME (people with*
 12 *ME)."*

13 *In Section 5-6 below we review doctors' knowledge of ME/CFS.

14

15 **2.9.4 Contact with health and social care professionals and services**

16 **GP home visits**

17 We asked respondents whether their GP visits them at home if they are unable to attend a GP
 18 surgery. 30 patients said yes, their GP visits them at home, 28 said no, they can't get a home
 19 visit and 2 said they hadn't asked for a visit. It was surprising that almost 50% of participants
 20 could not get a home visit given their severe status, often being homebound. We also detected
 21 a worrying trend – in the group that answered 'yes', a fair number said that it was becoming
 22 increasingly difficult to get a home visit and GPs were very reluctant to offer this service:

23 1575763 *"They did in the past but currently do not."*

24 1578170 *"No. The one time I insisted on a home visit, and the doctor very reluctantly*
 25 *agreed, I heard her muttering expletives down the phone - in German!!"*

26 1579197 *"Yes now but I was once told by one of the GPs at the surgery we don't make*
 27 *home visits to people with M.E."*

28 1578559 *"Reluctantly. They fight it every time despite me being so housebound. Because*
 29 *I *could* force myself there, despite the toll it would take, they insist that I do*
 30 *unless I cannot get up from the bed at all."*

31 **Special accommodations for patients attending GP or hospital appointments**

32 Again, results are mixed. However, GPs appear to be more accommodating than hospital
 33 care/specialists. GPs allow phone consultations, use of email and home-visits, whereas these
 34 appear to be absent in specialist care. 26 respondents state that they had not been offered
 35 any special arrangement or accommodation whilst attending their GP or hospital care. 30
 36 patients said they had been offered some accommodation.

37 1560411 *"My GP will visit at home if requested. I no longer attend specialist hospital*
 38 *appointments as they, in their own words, tell me there is nothing they can*
 39 *offer."*

40 **Patient control and choice**

41 We asked in question 32, if patients make choices about the types of care they receive or take.
 42 Respondents answered this question from different perspectives – some talking about medical

1 care, others about access to social care, and others about their personal funds that allow them
2 to pay for certain treatments. The majority said they were involved or had choices, but then
3 added that there are no real choices to make, given there are few available treatments. A
4 selection of responses:

5 1560014 *"I make decisions about my care...To be quite honest, one size does not fit all.
6 What is suitable for some illnesses is not suitable for me."*

7 1583136 *"There is no care - there is no choice. I feel that I am seen as a heart-sink
8 patient."*

9 1578559 *"I make all the decisions about my care with family support, but there are few
10 options to choose from."*

11 1586506 *"Since my family is comfortably off, I am able to make my own choices.
12 However, there is almost no suitable care available for patients with M.E. even
13 if you are able to pay."*

14 **Taking patients' views on board in treatment**

15 In question 33, we asked patients whether doctors consult them and take their views on board
16 during treatment. Results were mixed. We coded 'yes' for patient's views mostly taken on
17 board, 'no', for mostly not, and 'mixed' for mixed responses: we had 20 no, 19 yes, 9 mixed
18 and the rest non-applicable. Many answers did include mixed experiences. One example of a
19 positive response reveals that good relationships with open communication and patient
20 participation are key:

21 1580063 *"To be fair, some do. I had an excellent gynaecologist who also seemed quite
22 familiar with ME. We considered a hysterectomy at one point as I suffered
23 excessive bleeding. He was able to discuss how ME affects recovery and it
24 definitely factored in his advice to me. My ME consultants have been kind,
25 supportive and very helpful. They treat the relationship as a partnership. I am
26 lucky that I have been able to afford choice in going private."*

27 More negative responses centre around dismissive doctors, not feeling listened to and a lack
28 of physician knowledge.

29 1583769 *"They have often said over the years that are constrained by NICE Guidelines.
30 One particular GP in the surgery will consult me and be willing to listen and the
31 Community Neuro Rehab Team also do, to some extent."*

32 Many respondents talk about trying to educate their GP or specialist about the condition.

33 1578864 *"They mainly like to impose their views on me. I try to educate them, most know
34 I know more of ME than they do however do not like to admit it."*

35 1584124 *"Yes but those I have encountered have less knowledge about ME than I do.
36 We are 'lucky' to get one who actually thinks we are ill."*

37 **2.9.5 Participants' experiences of monitoring and review of ME/CFS**

38 **GP or specialist monitoring and review of patients**

39 We asked respondents if their ME/CFS is regularly monitored by either a GP or hospital specialist.
40 41 respondents said 'no' they did not have regular GP monitoring, 13 said 'yes' they did, and
41 the rest said either not applicable or did not give an answer. From those who did give an
42 answer of 'yes', the main method for GP follow-up was a home visit or telephone consultation.
43 11 said they had specialist monitoring (these 11 include those who gave a yes to GP follow-

1 up), again mostly via hospital visits or tele-consultation. The rest of the cohort had no specialist
2 follow-up or gave a non-applicable answer. Typical patient comments include:

3 1586506 *“No. Any doctor visit makes me ill (even a home visit), so it is only worthwhile*
4 *for a specific purpose.”*

5 1584124 *“GP comes once every 3 months. This is very good by ME standards BUT in*
6 *context of how ill we actually are, this is pathetic. I have not had any specialist*
7 *person in many years.”*

8 1583136 *“No, I see the gp only for problems caused by the ME, such as Reynaud's or*
9 *meds reviews once a year.”*

10 In a separate question (Q45) we asked patients whether or not they felt their illness receives
11 ‘adequate ongoing medical support’. Results were even worse than medical monitoring above.
12 55 respondents said no or disagreed, while only 5 said yes or agreed.

13 Some patients commented that given GPs have so little to offer they don’t often ask for help.

14 1574885 *“No - although some of that is down to me, I have anxiety around doctor*
15 *appointments and I am able to manage it alone, and as I have not been offered*
16 *anything that would improve my health situation previously I see no point in*
17 *regularly attending doctors appointments when I am not really well enough for*
18 *it.”*

19 1569304 *“No although I understand GPs are limited in what they know about the illness*
20 *(as they obviously need to have knowledge of a wide range of illnesses) and by*
21 *what they can offer.”*

22 **Patient improvement or deterioration**

23 We asked respondents about their illness stability after treatment in question 39. Therefore,
24 we feel Section 4 ‘ongoing experiences after diagnosis and treatment’ is the best place to
25 report our findings.

26 Many respondents report long periods of gradual small improvement followed by relapse and
27 “crashing down” after some health-related event and failing to recover after that. A little less
28 than half (26) patients report that their condition is relatively stable and about the same number
29 (25) that it is slowly deteriorating. The majority of those in the stable category classify
30 themselves as very severe and state that the reason why their condition does not change is
31 because it can hardly get any worse, i.e., they are stuck at the upper end of the scale. About
32 a quarter of the respondents (13) report fluctuations in their condition, which can be small-
33 scale (on the scale of individual days or even within a day) or larger-scale (on the scale of
34 months), with 1 reporting seasonal fluctuations – feeling better in the summer. Slow
35 improvement is reported by only 5 respondents (8%) and another 2 report slow improvement
36 from a very severe to a severe but manageable condition. Another common theme is the
37 reporting of singular or multiple “sharp crash” events, typically due to illness, accident or similar
38 events with significant impact on health. 9 respondents report sharp crashes punctuating the
39 overall trend, which can be stable or slowly deteriorating, and some report fluctuations besides
40 the overall trend. The numbers do not add up to 60 as respondents typically report more than
41 one type of event.

1 **2.9.6 Participants' experiences of information, education and support for**
 2 **people with suspected or diagnosed ME/CFS and their families and**
 3 **carers**

4 **Information and educational material available about the illness**

5 We asked in question 47, whether respondents felt there is enough information or educational
 6 material available relating to the illness. 48 of the 60 said 'no', 11 said 'yes' and the remainder
 7 were undecided or gave no answer. The majority of respondents stated clearly that there was
 8 either no information on the illness provided to them from the NHS or health professionals, or
 9 that any information available was deeply flawed. Respondents blamed a focus on the
 10 psychology of ME/CFS and psychological treatments such as CBT and GET. Even for the few
 11 patients that said there was adequate information available, they often said this came from
 12 charity groups or international sources, not the NHS. Many respondents said they got
 13 information from charity groups. Here is a sample of typical responses:

14 1582572 *"It feels there is quite a lot of information out there, but much of it is inaccurate*
 15 *or unhelpful and the crucial information is not being got across to those who*
 16 *need it. Information on things like PEM and aerobic capacity does not seem to*
 17 *be understood properly even by some 'experts'."*

18 1580063 *"I think the information is unclear, the advice is either incorrect or misleading.*
 19 *The assumption that most patients improve in time is unfounded. This makes it*
 20 *impossible for the patient to adjust and handle practical matters like finances,*
 21 *relationships with employers and family and friends. This has a hugely*
 22 *detrimental effect in the long term."*

23 1578343 *"No. The lack of funding for research into the condition and the psycho-somatic*
 24 *model of treatment is more detrimental to informed education than it is helpful*
 25 *(in my opinion)."*

26 1561885 *"absolutely not. There is far too much information focusing on the physiological*
 27 *side of things and non explaining the true reality."*

28 1578170 *"There is lots of information if you look online often provided by ME charities or*
 29 *voluntary organisations. However, the information available through the NHS is*
 30 *insufficient and frequently incorrect."*

31 **How accessible is information and educational material about ME/CFS and sources?**

32 The majority of participants stated that where information of educational materials existed,
 33 these were accessible. The most frequent sources used were:

- 34 • Online materials and sources
 35 • Via charities, either on or offline
 36 • Via social media and patient forums

37 *NHS sources were almost never mentioned.

38 **Peer support: membership of an ME/CFS patient group**

39 Most respondents said they belonged to such a group (only 12 respondents out of 60 were not
 40 a member of a support group). The 25% ME Group and the ME Association were the two
 41 charities mentioned most, however many respondents mentioned being part of online support
 42 groups, such as Facebook or local ME/CFS groups.

1 Support from a social worker/social services

2 Nearly two thirds of the respondents (38) stated that they do not have any social care support,
3 with few saying that they are paying for care privately and a couple saying that it is currently
4 being arranged. About one third (21) said that they have social carer, but 6 of them complained
5 that the service provided is inadequate and only 2 report being very satisfied with the service.

6 *As we noted in Section 1 'Support from Care Assistant,' many patients are cared for by family
7 members.

8 Many respondents report a battle to access social care and a lack of awareness of the illness
9 among social workers.

10 1584124 *"I had an assessment some years ago. Refused funded care. ME team*
11 *supported me but had no success. No social worker. Probably I am too ill for a*
12 *long talk with one even if offered and I doubt their training on ME amount to*
13 *anything at all."*

14 1585039 *"Had support from social services. It was exhausting, stressful. They failed to*
15 *do what they agreed to and were disorganised & inaccurate care plans etc."*

16 1580186 *"No - I choose to pay out of my PIP for my carers - I don't want to have to go*
17 *through regular social services assessments and cope with the fear of the*
18 *service being withdrawn for lack of Government funding."*

19 1577409 *"Local social services don't involve clients in writing or developing care plans*
20 *so they bear little relevance to the support needed."*

21 Types of social support accessed

22 6 report receiving household aids for bathroom, kitchen and bed. 6 report receiving part-time
23 personal and domestic care by a care assistant and 1 reports receiving full-time care. Individual
24 respondents report receiving hydrotherapy, psychological therapy and nutritional support.

25 Use of mobility scooter

26 Nearly two thirds (38) report having or using an electric or manual wheelchair. 12 report having
27 a scooter. 10 report having no aids and 4 report having other types of aids – walking sticks,
28 bed lever, stairlift.

29 Social care payments/benefits received

30 In question 36, we asked respondents whether they had received any government disability or
31 sickness benefits. The majority, 56 respondents, stated they had. 2 stated they had not, *1 of
32 those who gave a yes answer said they were currently "too ill" to apply, whilst 2 gave non-
33 descript answers. It was not possible to accurately collate what benefits were received given
34 some respondents answered with a 'yes', whilst others gave details. Of those who provided
35 details we note that Disability Living Allowance (DLA), Personal Independence Payment (PIP),
36 Employment Support Allowance (ESA) or some combination of these, such as ESA and DLA.
37 A fair number of respondents reported stress dealing with applications for benefits.

38 1580063 *"ESA. I applied for DLA and was refused, but was granted Incapacity Benefit. I*
39 *was later transferred to ESA. I should be entitled to at least some element of*
40 *PIP, but I simply haven't got the capacity to cope with yet another form and*
41 *more hassle."*

42 1583814 *"No, they stopped my disability it's in appeal."*

1 Barriers and difficulties in accessing social care

2 The most common complaint (18) is that the process is too difficult. Respondents use
 3 expressions like “gruelling and punishing”, “awful”, “interrogation”, “stressful”, “accused of
 4 lying”, “nightmare”, “major fight”, “bullied”, “harassed”, “treated like criminal”, “cruel campaign”,
 5 “physically, cognitively and emotionally exhausting”, “a lot of stressful form filling”, “total
 6 nightmare”. 12 respondents complain that the process has had negative impact on their health.
 7 11 have complaints about the assessment process, stating that the assessors did not
 8 document objectively their condition. 13 stated that they had some difficulties with accessing
 9 care and benefits, but that it worked out for them in the end. 20 respondents state that they
 10 received the benefits only after going through an appeal process. Minority groups of
 11 respondents stated that they were denied benefits (5), they had to give up trying to get them
 12 (3), the GP did not support their claim (4) and that they did not even want to try (1).

13 *Only 5 out of 60 stated that they did not encounter problems in the process.

14 1582331 “Yes, many.

15 *Universal Credit: I have to turn in fit notes every few weeks until a Work Capability*
 16 *Assessment is done. It has been over a year and I still have not had one. I have to get*
 17 *a phone appointment with my doctor (can only manage about 1x a month), ask for a fit*
 18 *note, get someone else to pick up the fit note for me, register it on the online account*
 19 *(very hard for me), then get someone else to bring it to the job centre for me. Every 3*
 20 *weeks. It is the biggest cause of PEM for me at the moment, and I can't even manage*
 21 *it as often as they want me to. There's been times I put the date in wrong by one day*
 22 *on the online account, managed to get someone to take it in for me, then had it refused*
 23 *because of it and sent back to me again because the job centre people won't edit the*
 24 *date on my claim. I've had threats of being sanctioned during time periods I was so ill I*
 25 *was not able to use a computer to turn one in. I've had times I couldn't afford my internet*
 26 *bill, so couldn't register the fit note - there is no way I can get to a library to do it there.*
 27 *Countless issues like that.*

28 *Social Care: I had a family worker from the council, who didn't believe in ME/CFS. 2*
 29 *charity workers made a referral to social care for me as she wasn't helping at all, my*
 30 *first two were denied because I had involvement from another council service. Because*
 31 *of the wait times involved with every aspect of my care right now, my social workers*
 32 *haven't been able to do much before they change again. Because of this I have the*
 33 *manager of the team helping me all the time, but she is very busy, and I've gone through*
 34 *3 others in the past few months. I am constantly having to go through the situation over*
 35 *and over every time they change, which is very hard for me.”*

36 Patient suggestions about what doctors and care or social support workers can do to 37 support them

38 We asked respondents in question 46, what more they feel doctors or care support workers
 39 could do to assist those living with ME/CFS. We had a range of informative answers, some
 40 very detailed. Below we list a summary of the common themes within these answers:

- 41 • More empathy, understanding and respect to patient and the illness.
- 42 • More and better training of doctors and allied health profession – information booklets,
 43 training courses, severe patient case studies.
- 44 • More flexibility – especially with appointments.
- 45 • Home visits & use of technology such as tele-consults.
- 46 • More support – especially home social support and social care benefits.

- 1 • More follow up – particularly specialist review and management of symptoms,
2 specifically POTs.
3 • More detail and coverage within the NICE guidelines.

4 **2.9.7 Participants' views on of information, education and support for health**
5 **and social care professionals**

6 **Doctors' knowledge of ME/CFS**

7 We asked respondents whether or not their primary care doctor or hospital specialist had
8 knowledge of ME/CFS. Results were predominantly poor. We coded answers Y for positive
9 knowledge, N for lacking knowledge and M for mixed results, such as different doctors both
10 having knowledge and lacking knowledge. Results were N=34, Y=10, M=16.

11

12 1556935 *"I have generally had to educate any doctors I've been involved with."*

13

14 1560014 *"Absolutely not. The advice I always got was try harder, put more effort in.*
15 *Exercise!"*

16

17 1568000 *"No, all GPs I have ever seen know of it but don't know anything about it. The*
18 *specialist I saw followed the NICE guidelines, believed it was caused by*
19 *deconditioning and GET was not harmful. Which I now know is not true. I was*
20 *also told to take a multivitamin and that out of all the alt therapies the lightning*
21 *process was the most promising. Which seems ridiculous knowing what I now*
22 *know."*

23

24 More positive accounts include:

25

26 1580063 *"My specialist is very knowledgeable and experienced in managing ME. I see*
27 *him privately outside the NHS."*

28 1580186 *"GP - pretty good. M.E. clinic O.T - very good."*

29

30 Typical mixed (M) account:

31

32 1576475 *"Hospital specialist very knowledgeable but Primary Care useless & not well*
33 *informed."*

34

35 **Awareness and understanding among health professionals**

36 We asked respondents for their suggestions about what could be done to improve raising
37 awareness of the illness among health and social care professionals. Most gave expansive

- 1 answers to this question, writing full paragraphs in the answer. The key themes to emerge from
2 our analysis are:
- 3 • More and better training of doctors and allied health profession – information
4 booklets, training courses, severe patient case studies.
 - 5 • A focus on ME/CFS as a physical disease rather than on CFS which is
6 associated with a psychological syndrome
 - 7 • Widening the focus of debate away from focus on CBT and GET treatments
8 – viewed as stigmatizing and harmful
 - 9 • Update NICE guidelines to reflect their experiences
 - 10 • Improving doctors' attitudes and empathy with severe patients.

11 Most respondents asked for more training and education of doctors, starting at medical school
12 and extending into GP level training, to view the illness as a serious debilitating illness, similar
13 to diseases such as multiple sclerosis.

14 1578560 *“As I said before CCG's need to commit to GP awareness and education. This
15 has to be a priority, especially with the lack of specialist services. As part of my
16 role as a patient rep working with the NHS patients cite poor GP care as their
17 main concern. They describe falling into a black hole of lack of care. I know
18 exactly what they mean. I don't go to my GP about new or worsening ME
19 symptoms, there is no point.”*

20 1578864 *“I feel in medical school students should go out and visit those who are amongst
21 the worst cases of ME and see how real it is prior to teaching them how to deal
22 with it.”*

23 1582331 *“I really want doctors to be taught more about ME/CFS. There are still doctors
24 who think it isn't real, or believe patients are mentally ill or hypochondriacs. I am
25 aware not an awful lot about this disease is known yet, but the knowledge of
26 even the most basic parts of it is very lacking in health and social care
27 professionals considering how severe and how common it can be. More of my
28 social workers have known about it than my GPs. Which is not the way it should
29 be at all. My social workers have done more for my health than a GP has in 10
30 years. I am massively grateful of course, but it should not be this way at all.”*

31 Some respondents recounted they had tried to inform their GPs, offering leaflets or booklets,
32 however GPs were often dismissive. Changing GP attitudes via awareness training was also
33 highlighted as a priority, such as visiting severe patients at home, interacting with charity
34 groups and reviewing the latest research, particularly on the biology of ME/CFS and its impact
35 on participants.

36

37 **Professional empathy and respect for patients**

38 We asked whether respondents felt they had been treated with respect and
39 empathy/understanding by health and social care professions in question 57. Their answers
40 were informative. Results were mixed. We coded answers 'yes' for mostly, 'no' for mostly not,
41 and 'm' for mixed, meaning that different professionals are seen acted differently (y and n). 13
42 respondents gave an affirmative answer that they had been treated with respect and empathy,
43 20 respondents said no, whilst 20 said their experience was mixed (y and n) and 7 said it was
44 not applicable.

45 Poor

- 1 1577409 *"No. Lack of belief in ME has meant GPs are rude and behave badly if I raise*
2 *ME treatment or symptoms. I now won't go to the GP on my own, after I was*
3 *shouted at a few times."*
- 4 1579810 *"No. I have been treated with contempt by the NHS. My previous GP hardly*
5 *deigned to talk to me."*
- 6 1579825 *"I have had many many bad experiences where I have been dismissed,*
7 *belittled, made to feel I am at fault for not being able to*
8 *understand/communicate with the doctor, irrational for being upset, responsible*
9 *for becoming and staying ill, making up symptoms or trying to get attention, had*
10 *my needs and experiences with drugs, especially withdrawal, dismissed etc."*
- 11 Mixed
- 12 1580863 *"Doctors and hospital consultants are the worse can be very dismissive and*
13 *sometimes plain arrogant and rude. Nurses vary, I have had really nice kind*
14 *caring ones and others not so."*
- 15 1577978 *"Last time I went my GP was one of the good ones and he was excellent. A*
16 *nurse practitioner however said oh we don't get much ME these days, I don't*
17 *think people get that anymore! Implying it was a 1980's fad."*
- 18 1583769 *"Some just dismiss ME as insignificant others are respectful and understanding.*
19 *Things have improved over the years. From my experience it's now 50/50."*
- 20 Good
- 21 1578864 *"Yes...I have also had in my home during an emergency The Acute Care team*
22 *who treated me wonderfully. Three times they came and each time I was treated*
23 *with dignity and respect."*

24 **Patients' free comments: additional information**

25 In our last question, 59, we asked respondents if there was any other relevant information they
26 wished to share not covered in other questions. We allowed respondents space to detail
27 anything they felt was relevant. In total, 46 respondents gave feedback in this question, while
28 14 opted not to. 10 of the 46 gave feedback on the process of data collection itself, the majority
29 talked about how taxing and exhausting it is to complete a survey like ours with severe
30 ME/CFS. Others thanked us and NICE for undertaking this study,

- 31 1578559 *"Thank you for taking an interest in our condition and the often poor quality of*
32 *life for patients because of the lack of appropriate care."*

33 The remaining 36 gave substantive feedback. 24 respondents complained about the poor
34 quality of care they received from the NHS and GP services and gave a range of
35 recommendations to improve things. These were on the common themes we identified
36 throughout our survey – training of doctors, sympathy and understanding, accommodation of
37 special needs, such as quiet rooms in GP practices, help with or reduction of paperwork, not
38 pushing for more exercise (GET), equality of care with e.g., MS and Parkinson's. For instance,

- 39 1575763 *"In my 19yrs of living with ME, most of these years with severe ME, I have never*
40 *had a care plan. I have face much medical abuse, neglect and gaslighting. I*
41 *have very little support and no action for my ME. I have lost nearly 20 years of*
42 *my life and had no help, that is a sobering thought. We live in a first world*
43 *country yet I am constantly left to fight for the same level of medical care others*
44 *receive because of ignorance, misunderstanding and poor guidelines. The*
45 *government has not funded biomedical research since 2012, yet other countries*
46 *are thriving with it. We have one of the best health systems in the world yet I do*

1 *not feel safe to go to hospital because of how I am treated because of my*
 2 *illness. This must change. We need support. Drs are bound by guidelines. They*
 3 *must change. Education and Reid needed and mostly, patients need help.”*
 4 (complete answer)

5 10 respondents gave advice for improvements in the research on ME, mainly the suggestion
 6 to refocus attention away from psychological and psychiatric research to biological causes and
 7 mechanisms.

8 1574735 *“I really think it is extremely important that the current biopsychosocial model of*
 9 *ME (on which CBT and GET but also the form of pacing currently taught in ME*
 10 *clinics) is completely done away with. It isn't helpful and it causes so much harm*
 11 *to patients, and misunderstanding among medical professionals which*
 12 *negatively impacts our care in a serious way which can lead to neglect,*
 13 *mistreatment, inappropriate advice, worsening of our condition, isolation, loss*
 14 *of benefits, preventable mental health problems (trauma), and suicides. We*
 15 *really need to do away with all of the unnecessary suffering caused by this*
 16 *outdated and unscientific model. Medical education is a priority, as is updating*
 17 *the ME clinics so they are staffed by medical professionals who actually*
 18 *understand the biological nature of the illness, the latest biomedical research,*
 19 *and the best approaches to clinical management of symptoms e.g. through*
 20 *pharmaceuticals and appropriate pacing/physiotherapy, not the kind that is*
 21 *currently used. These for me are top priorities, because we can't go on as we*
 22 *have been for decades. There has been too much unnecessary suffering*
 23 *already.”* (complete answer)

24 A few respondents mentioned the need to improve NICE guidelines, for example respondent
 25 1579730 – *“NICE guidelines need to acknowledge [that] CBT/GET are not adequate and are*
 26 *potentially dangerous to patients. The focus should be on supportive and pragmatic care.”* 3
 27 respondents expressed complaint with the benefits system of support for the patients,
 28 reinstating the points made in question 38. 2 respondents make an explicit request that patients
 29 should be believed by the doctors.

30 *Our numbers do not add up to 36 because in a few cases participant responses cover two
 31 coded categories.

32 2.9.8 Summary of salient findings

33 Table 4: Points from each scoping areas

34

Salient Summary Points	
Section 1	<ul style="list-style-type: none"> The majority of respondents are female and our full cohort had ages from 19-80, with more than half between 30-60 years old.
<i>Patient Characteristics</i>	<ul style="list-style-type: none"> Over half of severe patients report that their illness started suddenly after some infection. Around 1/3rd of severe patients report deterioration of the condition over time, another 1/3rd report fluctuations, whilst just 8% report improvement. The majority of severe ME/CFS patients are unable to work or study, are homebound and do not participate in social events, or walk outside for anything more than very short distances.

	<ul style="list-style-type: none"> • Most severe ME/CFS patients are cared for by family members or a care assistant.
Section 2 <i>Diagnosis</i>	<ul style="list-style-type: none"> • Most participants experience delays in getting a confirmatory diagnosis, with time to first diagnosis averaging 2 years but spanning anything from 2 months to 21 years in extreme cases. • Most participants are diagnosed by a GP or hospital-based specialist or at an ME/CFS clinic. • Approximately 1/3rd of respondents indicate that they are reasonably satisfied with the process of getting ME/CFS diagnosed, but approximately 2/3rds report being unsatisfied. The most common complaint is delay in diagnosis or not being believed. • Many respondents state that getting a diagnosis is positive and offers them legitimacy and access to social support. • GP or specialist awareness of ME/CFS is rated as the most important factor in speedy diagnosis of delay in diagnosis. • Around half of GPs remain uninformed and skeptical of the illness and many patients report feeling unsupported, a small number change GPs because of this, others limit their interactions with doctors.
Section 3 <i>Treatment & Management</i>	<ul style="list-style-type: none"> • A significant number of patients with severe status are too ill to try recommended therapies. • Many respondents have tried multiple therapies – CBT, GET, Pacing, Physio, and alternative approaches. • Results are generally negative for CBT, GET and Physio for the majority of respondents, however a small percentage do find some elements of CBT, GET and Physio useful, particularly when they are tailored to their individual needs. • Medical professionals sometimes give assurances to patients that they will improve if they follow CBT or GET, yet this does not often happen and may cause distress to the patient if they fail to improve or worsen in any way, they may also lose faith in health professionals. • Many participants reported losing faith in medical professionals after a bad or distressing experience and going off to try alternative approaches. Health professionals need to treat ME/CFS with understanding and maintain open and respectful communication. Where health practitioners express anger or frustration with patients, trust is often lost and the therapeutic relationship also. • Pacing, whether self-directed or supported by a professional, appears to help a higher percentage of patients, but not all respondents. • Almost half of all severe participants struggle to get a GP home-visit and the trend seems to be increasing. • Many severe patients find going to hospital a distressing experience, or they avoid hospital visits. The specific needs of patients with severe ME/CFS need to be considered more by hospital staff when these patients are being treated in-hospital, even for illnesses other than ME/CFS.

	<ul style="list-style-type: none"> Whist GPs offer home visits, tele-consults and use of email technology, hospital consultants rarely offer this – yet use of such technology appears to be an essential tool for severe ME/CFS participants to access care and services. There are few treatments available for patients, thus they have limited choices about the care they receive.
<p>Section 4</p> <p><i>Long-term Care</i></p>	<ul style="list-style-type: none"> Most patients are not monitored or reviewed regularly by either a GP or hospital specialist. Most participants report an initial illness followed by long periods of ill health, with some fluctuations, with a general pattern for most of a gradual decline in function over time, often years, whilst few patients recover, although a fair portion improve or stabilize. Other illnesses, secondary to ME/CFS and/or life stresses, can cause downward spiral and decline in function.
<p>Section 5</p> <p><i>Awareness & Support</i></p>	<ul style="list-style-type: none"> Many ME/CFS patients with severe status join patient support groups, often online local groups. Patients want better training of doctors and allied health professionals – information booklets, training courses, severe patient case studies. Patients find it difficult to access social care support, particularly there is a lack of tailored support for their specific needs. Patients often offer suggestions for improving care, commonly better professional training, more access to home visits, more regular follow-up and specialist monitoring and help with social care and home support. Patients often keep up-to-date with research developments.
<p>Section 6</p> <p><i>Important Factors in Care and Management</i></p>	<ul style="list-style-type: none"> The majority of severe ME/CFS participants state that their doctors lack knowledge of the illness, therefore greater emphasis needs to be placed on training, beginning in medical school and extending to specialist post-graduate training. Training should include face-to-face contact with severe patients and asking doctors to become cognizant of emerging research evidence on bio-physiological abnormalities associated with the illness. Patients often ask for NICE guidelines to include severe patients' accounts of the illness and their experiences of treatments such as CBT, GET, Pacing Therapy and Physiotherapy. Patients want less focus to be placed on psycho-social facets of illness and call on health professionals to be provided with information on new developments in ME/CFS. Many patients report that they are not treated with respect or empathy by health professionals, doctors and allied health workers.

1 2.10 Discussion

2 2.10.1 A unique patient cohort with unique challenges

3 The ME/CFS literature provides credible evidence that approximately 25% of all sufferers fall
4 into the category of 'severe ME/CFS'. However, no clear and widely accepted definition of
5 severe ME/CFS is given in the literature, thus severe generally refers to sufferers who are
6 mostly home-bound, often bedbound for the majority of the day, and are severely functionally
7 impaired using standardised quality of life instruments. Epidemiological prevalence estimates
8 (that commonly vary between 0.1% to 1%) suggest that there are around 250,000 people living
9 with ME/CFS in the UK. This would suggest that approximately 62,500 sufferers may
10 experience severe illness presentation. A general practice with a population of 10,000 patients
11 is likely to have 30–40 patients with ME/CFS and around half of these are likely to fall into the
12 moderate-to-severe category and may need input from specialist services (Group, 2002).

13

14 Findings from this survey show that many participants may move between severe and
15 moderate levels throughout the course of the illness, most gradually getting more unwell and
16 restricted over the years, whilst others improve and a small percentage recover. We noted that
17 many survey respondents self-classify as moderate, that we excluded in our analysis, who
18 may fall into the severe category – we suspect that these patients do not wish to self-identify
19 as 'severe ME/CFS' and retain a level of optimism about their illness status. There are no
20 simple methods available to clinicians or patients to assess illness severity. Research tools
21 such as the *DePaul Symptom Survey* of the *Chalder Fatigue Scale* do not specifically measure
22 illness severity. There is a need for development of a severity scale for ME/CFS.

23

24 ME/CFS may be hard to diagnose and is largely defined by generalised fatigue, a characteristic
25 of many other chronic illnesses, idiopathic complaints and is also associated with affective
26 disorders such as depression. Severe ME/CFS patients arguably represent the clearest cohort
27 of 'ME/CFS' cases. These patients experience most of the cardinal symptoms associated with
28 the illness, such as fatigue, pain, sleep intolerance, malaise after minimal exertion, intolerance
29 to light or noise, and cognitive complaints. From our survey, we found that severe sufferers
30 are often socially isolated, are unable to work, or reduce work to part-time or less, often
31 discontinue in education, although with a mean age of 34 years in ME/CFS onset in our survey
32 and a mean age of 45 years in other studies of severe ME/CFS (Pendergrast et al., 2016),
33 many participants have completed third level education before the illness begins. Many severe
34 participants report mental health complaints, such as depression and anxiety, co-morbid to
35 ME/CFS. However, the Pendergrast et al. international study of severe ME/CFS cohorts
36 revealed no significant differences in prevalence rates of comorbid psychiatric conditions
37 (major depression, bipolar disorder, anxiety, schizophrenia, eating disorders, and substance
38 abuse) between individuals who were housebound and those who were not housebound – a
39 surprising finding given differences in social isolation and symptomology associated with
40 severe illness presentation.

41

42 Severe sufferers within the wider ME/CFS population, represent the most challenging cases
43 of the illness, particularly for community physicians to manage in primary care. Expert
44 knowledge and careful consideration are needed to engage these patients, yet many studies
45 reveal that many GPs lack training on the illness and lack of confidence in dealing with these
46 patients (Raine et al., 2004). GPs also hold certain value-judgements about these patients,
47 that they are difficult to treat or are combative (Raine et al., 2004).

1 **2.10.2 Their call for recognition and support**

2 Findings from this survey reveal concern among participants that doctors and allied health and
3 social care professionals do not understand their illness or acknowledge their suffering. Many
4 participants report not feeling believed, feeling vulnerable, and having to battle doctors for
5 support, including home visits, referrals to specialist care or with social care applications. This
6 is particularly noteworthy, given ME/CFS patients experience extreme fatigue, emotional
7 fragility and cognitive complaints. Such problems with care and support have been identified
8 in previous reports on ME/CFS (Group, 2002; NICE, 2007). Given that severe patients have
9 lower quality of life scores compared with many other serious chronic health complaints and
10 illnesses, such as multiple sclerosis or diabetes, there is a need to bridge the support gap that
11 currently exists.

12 The present study provides a snapshot insight into a combative and acrimonious relationship
13 that exists between a portion of severe ME/CFS patients and their doctors. Patients report
14 difficulties getting a diagnosis and suffering severe symptoms for long periods without medical
15 intervention. Previous research has demonstrated that a good relationship with general
16 practitioners from the onset of the illness is essential for avoiding progression to severe
17 presentation of the illness (Pheby and Saffron, 2009) and that getting a diagnosis is the single
18 most helpful event in managing the condition (Drachler Mde et al., 2009), yet current levels of
19 knowledge of ME/CFS among doctors and allied health workers appears inadequate and a
20 cause of ongoing concern and distress for sufferers.

21 **2.10.3 Accessing care and the needs gaps**

22 Patients with severe ME/CFS experience more symptoms, to a higher intensity and for
23 prolonged periods, compared with mild and moderate patients. Unsurprisingly, those patients
24 within the severe category have worse prognosis for recovery. Severe patients may have the
25 greatest need for medical support and intervention but often have the greatest trouble
26 accessing this support, given their housebound status, pain and fatigue symptomology – an
27 classic example of the inverse care law (Tudor Hart, 1971). Our findings mirror those of a
28 survey by patient charity Action for ME that revealed that less than 50% of bedridden patients
29 are monitored by a medical practitioner and 60% are often too unwell to travel to a clinic, yet
30 many GPs refuse home visits. This survey confirms that severe ME/CFS patients have
31 problems accessing both primary care support and specialist care. Despite early management
32 of the illness being an important factor in preventing onset of severe presentation (Pheby and
33 Saffron, 2009). Severe patients also find it physically and emotionally difficult to access all
34 available care, whether medical, psychological, or social. Many participants reported relying
35 on family members, friends and carers for support.

36 **2.10.4 Severe patients absent from research studies**

37 Severe ME/CFS patients are often absent across the majority of research studies on this
38 illness. It is noteworthy, that being bedbound or housebound precludes most severe sufferers
39 from taking part in research studies, whether that be exercise physiology studies or clinical
40 trials of psycho-behavioural therapies. The largest conducted randomised controlled trial of
41 CBT and GET (PACE) required patients to attend multiple therapy sessions in a clinic setting
42 (White et al., 2011). Such trials require ambulatory patients well enough to attend. Wearden et
43 al. attempted to overcome this problem by delivering a combination of CBT and GET to patients
44 at home via practice nurses, but this trial reported much lower levels of benefit (Wearden et
45 al., 2010) using such therapies. We had a strong response to our survey call and received
46 many emails from patients. We found that severe sufferers are keen to get involved in research
47 but are often ignored, meaning patient surveys are often their only avenue to communicate
48 their needs.

49

1 **2.10.5 Problems with diagnosis**

2 Despite published guidelines for medical professionals to follow to diagnose ME/CFS (Baker
3 and Shaw, 2007), the literature shows that diagnosis remains a challenge for medical
4 professionals and patients often have to wait long periods for a confirmatory diagnosis. Bansal
5 et al. suggest that the ubiquity of general fatigue as a presenting complaint in general practice
6 (around 30% of patients experience some fatigue) makes it difficult for UK GPs to differentiate
7 idiopathic fatigue or fatigue related to other common health complaints from the illness
8 ME/CFS (Bansal, 2016). UK doctors may employ the Oxford Criteria (Sharpe et al., 1991)
9 and/or NICE guidelines (NICE, 2007) to aid in making a diagnosis. The absence of biomarkers
10 to identify ME/CFS means it remains an illness of exclusion diagnosed clinically. This may
11 partly explain why diagnosis is often delayed with an average of 2 years in our study, but this
12 average is much longer than current diagnostic guidelines of 4-6 months after the onset of core
13 symptoms (NICE, 2007). Diagnostic delays may also be caused by doctors, particularly GPs,
14 lacking knowledge of the illness, challenging patients on the origins and severity of their
15 symptoms, denying accommodations such as home-visits and combative doctor-patient
16 relationships.

17 **2.10.6 What patients say helps, what doesn't and what they want**

18 Many of our respondents were too unwell to participate in treatments such as cognitive
19 behavioural therapy (CBT) or graded exercise therapy (GET). However, for those who
20 undertake such therapies, few participants reported significant benefit from such therapies.
21 Many participants state that psycho-behavioural treatments are inappropriate, particularly
22 GET. A large proportion report that GET causes a worsening of symptoms. This finding runs
23 contrary to evidence from clinical trials that report few adverse outcomes with GET (Dougall et
24 al., 2014). However, as we noted above, patients with severe ME/CFS status are often absent
25 from clinical trials. A small percentage of patients state that CBT helps with the psychological
26 stresses that comes with chronic illness.

27 The greatest proportion of our patient cohort state that pacing therapies help most often, but
28 mainly to ameliorate symptoms or prevent deterioration – most severe patients report little
29 change in their illness status over the long-term. This finding may fit with Jason's Envelope
30 Theory (Jason et al., 2013), of staying within energy limits until stronger. This may be
31 particularly useful for severe sufferers. However, a question does arise as to how doctors and
32 allied health professionals, particularly physiotherapists and occupational therapists, can
33 support these patients in moving limbs and avoiding deconditioning. Long periods of
34 confinement to the home will result in profound loss of physical conditioning and other
35 problems, physical and psychological. There may be a need to develop tailored exercise or
36 movement programmes other than GET, given many patients report problems with GET,
37 perhaps some form of supportive physiotherapy as described by patients in '*Participants'*
38 *experiences of management and treatment of ME/CFS*' p23-24.

39 Many patients report having tried alternative therapies, but again the majority report that these
40 treatments only help manage symptoms. Many of our survey cohort recognise that anxiety and
41 depression are common mental health complaints that can arise during the course of their
42 illness, but many are unwilling to seek help with these complaints because they believe the
43 medical profession views ME/CFS as a predominantly psychological illness and disclosure of
44 mental health complaints might bias their doctors views of the illness. GPs should consider
45 alternatives to CBT only for dealing with mental health complaints in ME/CFS.

46 Many severe participants mention the need for home-based visits with GPs, tele-calls with GPs
47 and hospital specialists, as very important to them. GPs and hospital-based staff should
48 consider using such technologies with these patients. Patients also talk about the need for
49 specialist follow-up and monitoring, and support with symptoms, particularly orthostatic
50 intolerance (often called POTs).

1 The importance of social support, in all its forms, home care assistance, support from family
2 and friends, occupational therapy, support from local and national agencies with disability
3 benefits and mobility aids – emerged as an important theme in our study. We found that many
4 severe ME/CFS patients join patient support groups, but not all join national groups; many
5 participate in online forums and local patient groups. Almost all of our sample claimed disability
6 or social care benefits, but a large number recount difficulties in accessing such benefits. Many
7 patients want doctors to do more to support them with their claims, including providing medical
8 evidence and letters of support.

9 Blease et al. wrote about how ME/CFS patients often feel a sense of injustice, both epistemic
10 and hermeneutic, that their illness is not understood and they feel unsupported. We detected
11 patient frustration at not being believed and having their symptoms dismissed. We believe
12 doctors could do more to support patients and small changes in patient management and
13 doctor-patient communication might overcome many of the issues we identified.

14 **2.10.7 Building better doctor-patient relationships with severe ME/CFS patients**

15 Severe ME/CFS patients wish to be treated with dignity, respect and empathy. Many report
16 difficult and distressing experiences with doctors and other health and social care
17 professionals. There is a lack of understanding of the illness and often a lack of empathy for
18 the patient and their plight. Empathy is important in building effective doctor-patient
19 relationships and has been shown to improve outcomes, particularly in general practice
20 (Derksen et al., 2013). Empathy lowers patients' anxiety and distress and delivers significantly
21 better clinical outcomes (Derksen et al., 2013), and given anxiety and distress are common
22 complaints for severe ME/CFS patients, doctors must do more to avoid causing or adding to
23 patients' distress. All ME/CFS sufferers are at enhanced risk of suicide (Roberts et al., 2016),
24 but severe ME/CFS sufferers are particularly vulnerable to severe depression and suicide
25 given they experience the highest levels of social isolation and debilitating symptoms, therefore
26 health professionals need to take extreme care in communicating with and managing these
27 patients.

28 Many severe patients become 'expert patients' and attempt to inform their GP or specialist,
29 however whilst some doctors form good relationships with their patients, others fail to engage
30 these patients. Raine talks about how GPs often lack confidence in dealing with ME/CFS
31 patients and how they view these patients as challenging and combative (Raine, 2004). GPs
32 and other health professionals clearly need more training on the illness and face-to-face patient
33 exposure, particularly within patients' homes to see the impact the illness has on people with
34 severe ME/CFS. Participants in our survey consistently stated that doctors need to stay
35 abreast of the latest developments in ME/CFS research.

36

37 **2.11 Study strengths and limitations**

38 One of the main strengths of this study is that it has been carried out by an experienced team
39 of professional researchers with expert knowledge of myalgic encephalomyelitis/chronic
40 fatigue syndrome and clinical experience. Our research team included two medical doctors
41 and a nurse. The objectives and methods applied throughout are clearly specified, and thus
42 enhance the reliability and credibility of findings. Given that other studies reported difficulties
43 in recruiting and engaging patients with severe ME/CFS, such as poor response rate reported
44 by Newton and colleagues at Newcastle (Strassheim et al., 2018) or high time and cost
45 demands reported by Lacerda and colleagues at LSHTM (Lacerda et al., 2018), our study
46 opted for a survey methodology that allowed us to engage severe ME/CFS patients within a
47 short timeframe and the accessibility of a survey enhanced our response rate, we had far more
48 responses than required, in excess of 340 in under 1 week. Our survey method also minimised
49 input requirements on the part of respondents compared to face-to-face interview with a home
50 visit.

1 Our study has some inherent limitations. First, the use of a survey questionnaire as a data
2 collection instrument may exclude very severe participants who are too unwell to complete
3 such surveys. We attempted to overcome this limitation by instructing respondents to seek the
4 assistance of family members, friends or care assistants, to complete the survey. Surveys of
5 this kind are open to response bias. Patients who have recovered from ME/CFS or who have
6 moved from severe to moderate or mild symptoms might be unwilling to engage in such
7 studies. There is also a risk that patients with negative experiences of medical care are more
8 likely to participate than those with positive experiences, however this factor applies to all such
9 patient experience research. We attempted to overcome such bias by advertising our survey
10 via social media to widen participation beyond patient advocacy group members. We
11 emphasized that responses would remain confidential and we were careful not to encourage
12 negative responses by using neutrally framed semi-structured and open-ended questions that
13 allowed respondents to give a full account of their views and experiences, both positive and
14 negative.

15 We did not obtain confirmation of ME/CFS status by independent medical professional
16 assessment, instead we relied on respondents to attest to their ME/CFS. This is common
17 practice in this field, given it is often too costly and time consuming to medically screen every
18 patient for a confirmatory diagnosis. However, respondents in our survey had to confirm if they
19 had an ME/CFS diagnosis from a medical professional. Respondents who did not, were
20 excluded from our analysis.

21 Another potential bias within our survey concerns the self-rating of 'severe ME/CFS'. It was
22 not feasible within the remit of our study to measure the severity levels of each respondent's
23 illness. Indeed, measuring the severity of ME/CFS is inherently difficult given the lack of
24 biomarkers or guidelines to assess severity. To avoid complexity, we opted to allow
25 participants to self-rate as 'severe', however we did structure questions in order to explore
26 severity factors, such as asking participants if they were housebound or not, if they could or
27 not and so on. We found that a large percentage of patients self-classified as 'moderate' who
28 may well meet a 'severe' categorisation. For example, some moderate respondents stated
29 they were housebound and spent most of the day in bed. However, we excluded these patients
30 due to their 'moderate' self-classification, but they could perhaps have been included. As we
31 had enough responses with clear severe status to analyse, exclusion of moderate participants
32 has no impact on our findings, but it does reveal that patients' severity status is highly
33 subjective and patients may be poor judges of their actual functional status. Finally, our
34 thematic analysis of responses is open to selection or interpretation bias. We sought to
35 minimise such bias by using two independent researchers to extract and analyse data,
36 overseen by a senior academic. We also included direct quotations from patient respondents
37 alongside data and themes identified and presented.

38 **2.12 Acknowledgments**

39 This project would not have been possible without the support of ME/CFS patient participants.
40 We would like to thank those participants who helped pilot test our survey and all the
41 participants who completed the survey. We understand that completing the many questions
42 was often very taxing for many sufferers, thus we are sincerely grateful to everyone who took
43 part, including carers and family members who assisted. We would also like to thank Prof. Leni
44 Jason at DePaul University in the US for his general advice on running a survey of this kind
45 and the best methods for patient engagement and data analysis. He is an international expert
46 in ME/CFS who has conducted multiple patient surveys. We are grateful to the University of
47 Manchester Ethics Committee for their helpful comments on the project. We wish to thank Kate
48 Kelley, Operations Director at the Royal College of Physicians, for her support and insights
49 throughout the consultation. We would also like to thank NICE and the ME/CFS Guideline
50 Committee for commissioning the project.

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3 Drawing on the report to inform the 2 recommendations

3 A member of the NGC technical team presented the findings of the report to the committee
4 and 1 member of the research team was available on the telephone to answer questions
5 from the committee. The committee had received the study report two weeks before the
6 meeting. The themes that emerged from the report were taken into consideration alongside
7 other identified evidence when drafting recommendations. This was the most applicable
8 evidence for a number of topics and influenced the recommendations directly. Where
9 relevant this is referenced in the committee discussions of the evidence reviews.

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4 ¹ The committee's overview of the research

² The committee assessed the report and agreed it is a valuable report providing information
³ on a population that are very difficult to identify and access. The lay committee members
⁴ noted that the report findings that reflected their own experiences and is reflective of other
⁵ surveys seeking the views of people with severe ME/CFS.

⁶ Although the limitations of the research are described well in the report the committee noted
⁷ the additional points:

- ⁸ • the sample was a self-selected group
- ⁹ • the diagnosis of ME/CFS and severity of the condition was self-reported. The
¹⁰ definition of severe could be different for different people making it difficult to attribute
¹¹ a commonality to the results
- ¹² • people with very severe ME/CFS are unlikely to have participated. The committee
¹³ recognised the difficulties with recruiting and researching people with severe
¹⁴ ME/CFS.
- ¹⁵ • the empirical basis for the project was not clear. The methodology was described as
¹⁶ qualitative and a survey and a mixed methods approach was described in the results
¹⁷ with limited qualitative analysis
- ¹⁸ • it isn't clear who had therapies, what therapies they had, or when or where these
¹⁹ where implemented and the relationship to their symptoms at the time. This made it
²⁰ difficult for the committee to attribute any positive or negative effect to the therapies
²¹ mentioned in the report.
- ²² • the research team were restricted by the areas of the scope and the time to conduct
²³ the research, This did not allow for deeper probing and questioning potentially
²⁴ missing some important issues.
- ²⁵ • the survey could be completed with assistance from a family member/carer. The
²⁶ committee recognised and supported the rationale for this, but also acknowledged
²⁷ that it may have influenced the responses.
- ²⁸ • issues of sample size and data saturation were discussed in relation to qualitative
²⁹ interview studies, but there was no clear rationale for the sample size selected for the
³⁰ design that was used.

³¹ This was taken into account when considering the findings of the research.

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1 Appendix A: Research guide and 2 questionnaire 3

Scope area	Review questions	Area to be explored in interviews/questionnaires with people with severe ME
Identification and assessment before diagnosis	<ul style="list-style-type: none"> • What are the most clinically effective and cost effective precautionary management strategies that should be adopted while being assessed for a diagnosis of ME/CFS? • In people with suspected ME/CFS, what are the criteria used to establish a diagnosis? • What are the barriers and facilitators to the diagnosis of ME/CFS? 	<p>Experience of :</p> <ul style="list-style-type: none"> • Initial illness <ul style="list-style-type: none"> ◦ Being believed • Initial illness and impact on life (including family, friends, school, college, university, work) • Initial contact with a health and social care professional about symptoms • What worked well • What didn't work well
	Questions	<ol style="list-style-type: none"> 1. At what age were you first diagnosed? 2. How long have you had ME/CFS? 3. Have you received a firm diagnosis of ME/CFS? 4. - Was this from a GP, specialist or other - specify? 5. How long did it take you to get a diagnosis? 6. Did your illness start suddenly, or gradually worsen over time? 7. What was your experience of getting a diagnosis (please detail)? 8. What factors do you feel helped you get a diagnosis? 9. Did any factors delay your diagnosis? 10. Did your primary care doctor/GP agree with a diagnosis of ME/CFS and offer you appropriate support? 11. Is your illness relatively stable, have you experienced any periods of improvement or remission? 12. Would you classify yourself as mild, moderate or severe ME/CFS currently? 13. Did or does your illness prevent you from: 14. ...Attending school or college/university/training or work? 15. ...Participate in social events? 16. Are you able to get outside your home to shop or undertake outside activities? 17. Do you receive support from family members? 18. Do you have a carer or care assistant? (how often per week)

Diagnosis of ME/CFS	<ul style="list-style-type: none"> • What are the predictive accuracies of specific tests, or clinical symptoms/signs, to identify people who will subsequently be given a definitive diagnosis of ME/CFS? • In people with suspected ME/CFS, what are the criteria used to establish a diagnosis? • What are the barriers and facilitators to the diagnosis of ME/CFS? 	<p>Experience of :</p> <ul style="list-style-type: none"> • Continuing illness and severe ME/CFS • Continuing illness and impact on life (including family, friends, work, college, university) • Contact with health and social care professionals to get a diagnosis, approach taken • Time to get a diagnosis • What worked well • What didn't work well
	Questions	Above
Management of ME/CFS	<ul style="list-style-type: none"> • What is the clinical and cost-effectiveness of pharmacological interventions for people with ME/CFS? • What is the clinical and cost-effectiveness of non-pharmacological interventions for people with ME/CFS? (includes self-management strategies) • In people with ME/CFS, what is the clinical and cost-effectiveness of different models of multidisciplinary care? • What are the barriers and facilitators to the care of people with ME/CFS? (will include access to care) 	<p>Experience of:</p> <ul style="list-style-type: none"> • Interventions (benefits and harms) <ul style="list-style-type: none"> ○ For ME/CFS and symptomatic relief ○ Outcomes: benefits and harms ○ If offered interventions have not been taken up, why • Contact with health and social care professionals and services <ul style="list-style-type: none"> ○ Are your basic needs met? ○ Co-ordination of care ○ Referral to specialists ○ Hospitalisation ○ Involvement in decision making <ul style="list-style-type: none"> ▪ Feelings of control and choice ○ Access to services <ul style="list-style-type: none"> ▪ Access to appointments and getting to appointments (distance to clinics) ▪ Home visits ▪ Support services (mobility aids) • What worked well <ul style="list-style-type: none"> ○ Experience of recovery if appropriate ○ Experience of reintegration if appropriate (for example, work, friendship groups) • What didn't work well <ul style="list-style-type: none"> ○ Experience of relapse
	Questions	<p>19. Have you been prescribed any drugs by your doctor or a specialist specifically for your ME/CFS or related symptoms – list?</p> <p>20. Have these helped improved symptoms? – please specify</p> <p>21. Have you been offered any other treatments for your illness?</p> <ul style="list-style-type: none"> - Cognitive Behavioural Therapy (CBT) - Graded Exercise Therapy (GET)

		<ul style="list-style-type: none"> - Pacing Therapy (Adapted Pacing Therapy – APT) - Physiotherapy - Other therapies (please specify) <ol style="list-style-type: none"> 22. For each treatment or therapy undertaken, please detail if this therapy helped, made no difference, or made symptoms worse? 23. Have you ever refused to undertake a specific therapy or treatment (please specify which ones and your reasons for not undertaking)? 24. Have you ever required specialist support such as tube feeding, IV fluids, speech therapy, - please list? 25. Have you ever tried any alternative treatments or therapies (not offered by your doctor or the NHS) y/n – please specify which ones? E.g. massage, supplements, psychotherapy, and so on. 26. Did any of these alternative treatments or therapies help improve your symptoms, did any make things worse? 27. Does your GP visit you at home, if you are unable to attend a GP practice/surgery? 28. Are you able to attend hospital appointments or appointments with specialists? 29. Do GPs/specialists offer any alternative arrangements if you are unable to attend? 30. Are hospital staff aware of your condition and do they accommodate your needs on hospital visits? 31. Have you had any hospital in-stays – how many per year or since you developed the illness? 32. Do you make decisions about your care, do you feel you are able to make choices about the types or care you receive? 33. Do doctors consult you and take your views on board during treatment? 34. Have you had any support form a social worker/social services? 35. What type of support or care do you receive from them? 36. Did you receive any Government disability of sickness benefits? 37. Do you use mobility aids or a mobility scooter? 38. Have you encountered any difficulties in accessing social care and or sickness benefits – please specify any issues? 39. Has your illness and symptoms remained relative stable or changed since it began – specify if it has remained relatively the same, has worsened over time, has improved over time, or fluctuates frequently? 40. Have you been able to return to work, study if prevented to previously? 41. Have you received any special medical or social care assistance that has helped you undertake work or education/training?
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		<p>42. Have you been able to take part in social activities recently if prevented previously?</p> <p>43. What types of medical or social support have been most useful to you in managing your illness - please specify?</p>
Monitoring and review	<ul style="list-style-type: none"> • What is the most clinically and cost-effective method of monitoring/reviewing people with ME/CFS? • What are the barriers and facilitators to the care of people with ME/CFS? (will include access to care) 	<p>Experience of :</p> <ul style="list-style-type: none"> • Continuing care • Follow up <p>Who does this? Information about prognosis or future planning</p>
	Questions	<p>44. Is your ME/CFS regularly monitored by either a GP or hospital specialist, if so who and how often?</p> <p>45. Do you feel your illness receives adequate ongoing medical support?</p> <p>46. What more do you feel your doctors or care support workers could do to assist you living with ME/CFS?</p>
Information, education, and support for people with suspected or diagnosed ME/CFS and their families and carers	<ul style="list-style-type: none"> • What information, education and support do people with ME/CFS and their families and carers need? • What information, education and support do people with suspected ME/CFS and their families need before formal diagnosis? 	<p>Experience of :</p> <ul style="list-style-type: none"> • Accessing information, education and support <ul style="list-style-type: none"> ○ What was useful and what wasn't ○ Information and support networks
	Questions	<p>47. Do you feel there is enough information or educational material available relating to your illness?</p> <p>48. How accessible is this information or educational material?</p> <p>49. Where did you go to get or access information or educational material – please detail?</p> <p>50. Are you a member of an ME/CFS patient organisation or support group – please detail which ones?</p> <p>51. What information or educational material have you found most useful to you in dealing with your illness?</p> <p>52. Is the material you used tailored for your needs?</p> <p>53. Is there material available tailored to family members and carers?</p>
Information, education and support	<ul style="list-style-type: none"> • What information, education and support do health and social 	<p>Experience of :</p> <ul style="list-style-type: none"> • Knowledge of the health and social care professionals

<p>for health and social care professionals.</p>	<p>care professionals who provide care for people with ME/CFS need?</p> <ul style="list-style-type: none"> • What are the barriers and facilitators to providing information, education and support for health and social care professionals? 	<ul style="list-style-type: none"> ○ Where do you think they get information from • Health and social care professionals attitude to ME/CFS • Do they have the ability to provide support and what has been useful
	<p>Questions</p>	<p>54. Is or was your primary care doctor or hospital specialist knowledgeable about ME/CFS – please detail?</p> <p>55. If you feel their knowledge or awareness was lacking in any way, what could be done to improve raising awareness of the illness among health and social care professionals?</p> <p>56. When visiting your GP or hospital were you able to convey any special needs or requirements to staff, were these needs accommodated e.g. quiet area, short waiting time, and so on?</p> <p>57. Were you treated with respect and empathy/understanding by health and social care professions – please detail any response?</p> <p>58. What do health and social care professionals need to specifically take into account when dealing with patients with severe ME/CFS – please detail any response?</p> <p>59. Is there any other relevant information you wish to share that is not covered in the questions above – please feel free to detail in this section?</p>

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Appendix B: Consent form

UoM Participant Consent Form

This research project is compliant with the GDPR (General Data Protection Regulation) and the Data Protection Act 2018.

Title of Research

Involving adults with severe ME/CFS symptoms in developing a NICE guideline on Myalgic encephalomyelitis (or encephalopathy)/chronic fatigue syndrome: diagnosis and management

If you are happy to participate please complete and sign the consent form below

	Activities	Initials
1.	I confirm that I have read the attached information sheet (study information sheet version 1, 1 st Oct 2019) for the above study and have had the opportunity to consider the information and ask questions and had these answered satisfactorily.	
2.	I understand that my participation in the study is voluntary and that I am free to withdraw at any time without giving a reason and without detriment to myself. I understand that it may not be possible to remove all of the data I provide, from the project once it has been anonymised and forms part of the data set or a report. I agree to take part on this basis.	

3.	I agree that any data collected may be published in anonymous form in academic books, reports or journals.	
4.	I understand that data collected during the study may be looked at by individuals from The University of Manchester or regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data	
5.	I agree that any anonymised (that does not identify me) data collected as part of this study may be shared with researchers at other institutions.	
6.	I agree that researchers from Manchester or given permission by the research team running this study may contact me in future about other research projects. (this is not expected but we are asking just in case there is ever a need to contact you in future)	
7.	I agree that the researchers may retain my contact details in order to be able to remove me from the study if I change my mind regarding my participation.	
8.	Where I have assistance filling in my answers to questions in the survey, this will be indicated.	
9.	I understand that there may be instances where during the course of the study information or data is revealed which means that the researchers will be obliged to break confidentiality and this has been explained in more detail in the information sheet.	
10.	I agree to take part in this study.	

Data Protection

The personal information we collect and use to conduct this research will be processed in accordance with data protection law as explained in the Participant Information Sheet and the Privacy Notice for Research Participants.

Name of Participant

Signature

Date

Name of the person taking consent

Signature

Date

[Consent forms will be securely kept at the University of Manchester. Online version consent forms will be electronically stored, whereas hardcopy consent forms will be stored in our secure offices at the University.]

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1 Appendix C: Participant information sheet

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University of Manchester Research Participant Information Sheet

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9 Title of Research

10 Involving adults with severe ME/CFS symptoms in developing a NICE guideline on Myalgic encephalomyelitis (or
11 encephalopathy)/chronic fatigue syndrome: diagnosis and management

12 You are being invited to take part in a research study to explore the views and needs of patients with severe
13 ME/CFS. This project has been commissioned by the National Institute of Health and Care Excellence (NICE).
14 Before you decide whether to take part, it is important for you to understand why the research is being
15 conducted and what it will involve. Please take time to read the following information carefully before deciding
16 whether to take part and discuss it with others if you wish. Please ask if there is anything that is not clear or if
17 you would like more information. Thank you for taking the time to read this.

18 About the research

19 ➤ Who will conduct the research?

20 The project is being led by Dr. Keith Geraghty, working with colleagues at the University of Manchester Centre
21 for Primary Care.

22 ➤ What is the purpose of the research?

23 The purpose of the study is to explore the needs of patients with severe ME/CFS. Our wish is to better understand
24 the needs and views of patients with severe ME/CFS presentations and to provide NICE with up-to-date
25 information that might help inform the NICE Guideline Committee as they undertake a review of treatment
26 guidelines for this illness. We plan to recruit a selection of patients, in the time period available to us between
27 October 2019 and November 2019 and to write up a report based on our findings that we will pass to NICE at
28 the end of the project.

29 ➤ Will the outcomes of the research be published?

30 We also hope to publish a research paper from this project.

31 ➤ Who has reviewed the research project?

32 This project has been reviewed by The University of Manchester Research Ethics Committee (September 2019).

33 ➤ Who is funding the research project?

34 The National Institute of Health and Care Excellence

35 What would my involvement be?

36 ➤ What would I be asked to do if I took part?

37 We are asking people with severe ME/CFS to take part in this study. This involves completing a short online
38 survey. We anticipate that the survey will take you between 30 minutes to 1 hour to complete, however you may
39 not want to do this in one sitting if it makes you feel unwell or aggravates your symptoms. We advise that you
40 pace yourself and complete the survey in your own time. A family member or carer can assist you if needed. The
41 survey will ask you a range of short questions about your illness, your care needs and your experiences of
42 accessing health and social care.

43 ➤ Will I be compensated for taking part?

44 We are not offering any compensation for taking part as we do not have funding for this. We greatly appreciate
45 your participation in this project.

46 ➤ What happens if I do not want to take part or if I change my mind?

47 It is up to you whether or not you decide to take part. If you decide to take part, you will be given this information
48 sheet and a consent form to sign (confirm you agree). You can contact us at any stage if you do not wish to take
49 part of if you wish to withdraw from the study. However, it will not be possible to remove your data from the

1 project once it has been anonymised as we will not be able to identify your specific data. This does not affect
2 your data protection rights. If you decide not to take part, you do not need to do anything further.

3 Data Protection and Confidentiality

4 ➤ What information will you collect about me?

5 In order to participate in this research project we will need to collect information that could identify you, called
6 “personal identifiable information”. Specifically we will need to collect:

- 7 • Your name
- 8 • Age
- 9 • sex
- 10 • How long you have suffered from ME/CFS
- 11 • Other less identifiable information

12 ➤ Under what legal basis are you collecting this information?

13 We are collecting and storing this personal identifiable information in accordance with data protection law which
14 protect your rights. These state that we must have a legal basis (specific reason) for collecting your data. For this
15 study, the specific reason is that it is “a public interest task” and “a process necessary for research purposes”.

16 ➤ What are my rights in relation to the information you will collect about me?

17 You have a number of rights under data protection law regarding your personal information. For example, you
18 can request a copy of the information we hold about you. If you would like to know more about your different
19 rights or the way we use your personal information to ensure we follow the law, please consult our Privacy Notice
20 for Research.

- 21 • Will my participation in the study be confidential and my personal identifiable information be
22 protected?

23 In accordance with data protection law, The University of Manchester is the Data Controller for this project. This
24 means that we are responsible for making sure your personal information is kept secure, confidential and used
25 only in the way you have been told it will be used. All researchers are trained with this in mind, and your data
26 will be looked after in the following way:

27 Important note: UoM requires identifiable data to be anonymised as soon as the objectives of the project allow.
28 The standard retention period for data once anonymised is 5 years unless funders or regulators have specified
29 longer retention requirements.

30 Only the study team at The University of Manchester will have access to your personal information, but they will
31 anonymise it as soon as possible. Your name and any other identifying information will be removed and replaced
32 with a random ID number. Only the research team will have access to the key that links this ID number to your
33 personal information. Your consent form and contact details will be retained for 5 years (electronic copies will
34 be securely kept at our University data storage facility and hardcopies will be kept in a locked office within our
35 faculty building. Data may be transferred electronically between researchers on and off-site, however only data
36 that has removed personal identifiers will be shared in this way. All data sharing will involve password protected
37 files.

38 We have a duty of care to participants which includes breaking confidentiality if you disclose information that
39 indicates that your health and well-being are at serious risk. In such cases we might share the relevant
40 information with qualified medical and or social care professionals.

41 Data Sharing Requests from Other Parties (other than our research team):

42 When you agree to take part in a research study, the information you provide may be liable to data sharing
43 requests from other researchers and interested parties. We will only share data that does not include any
44 personal identifiers (such as your name or contact details). We will only share data if requesters can guarantee
45 data security, with a plan for data storage. We will only share data if you have given consent to do so.

46 Opt-Out Reminder: You are able to opt out of this study within 2 weeks after you complete the online survey.
47 After this time your data may form part of a report or dataset that cannot be changed. You can have your
48 personal details deleted at anytime. We will retain your contact details on a secure University server/storage
49 facility in order to be able to remove you in future, if you so wish. This data will be kept for 5 years, before being
50 destroyed/deleted permanently.

51 Please also note that individuals from The University of Manchester or regulatory authorities may need to look
52 at the data collected for this study to make sure the project is being carried out as planned. This may involve
53 looking at identifiable data. All individuals involved in auditing and monitoring the study will have a strict duty
54 of confidentiality to you as a research participant.

55 What if I have a complaint?

1 You may contact any member of our research team at any time if you have a complaint or concern about your
2 participation in this study or any other matter relating to this study. Email contact details are provided below.
3 CONTACT DETAILS FOR COMPLAINTS:
4 Dr. KEITH GERAGHTY or Prof. ANEEZ ESMAIL (Project principal investigator and Project lead)
5 Email: keith.geraghty@manchester.ac.uk
6 Email: Aneez.esmail@manchester.ac.uk
7 Tel: +44(0) 161 306 3990
8 If you wish to make a formal complaint to someone independent of the research team or if you are not satisfied
9 with the response you have gained from the researchers in the first instance then please contact
10 The Research Governance and Integrity Officer, Research Office, Christie Building, The University of Manchester,
11 Oxford Road, Manchester, M13 9PL, by emailing: research.complaints@manchester.ac.uk or by telephoning
12 0161 275 2674.
13 If you wish to contact us about your data protection rights, please email dataprotection@manchester.ac.uk or
14 write to The Information Governance Office, Christie Building, The University of Manchester, Oxford Road, M13
15 9PL at the University and we will guide you through the process of exercising your rights.
16 You also have a right to complain to the Information Commissioner's Office about complaints relating to your
17 personal identifiable information Tel 0303 123 1113
18
19 CONTACT DETAILS:
20 If you have any queries about the study or if you are interested in taking part then please contact the
21 researcher(s)
22 Dr. KEITH GERAGHTY or Prof. ANEEZ ESMAIL (Project principal investigator and Project lead)
23 Email: keith.geraghty@manchester.ac.uk
24 Email: Aneez.esmail@manchester.ac.uk
25 Tel: +44(0) 161 306 3990
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1 Appendix D: Abbreviations

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CBT	cognitive behavioural therapy
CCBT	computerised cognitive behavioural therapy
CDC	Centers for Disease Control
CFS	chronic fatigue syndrome
CI	confidence interval
CMO	Chief Medical Officer
DoH	Department of Health
EBV	Epstein–Barr virus
ECG	electrocardiogram
ESA	employment support allowance
GDG	Guideline Development Group
GET	graded exercise therapy
GRP	Guideline Review Panel
HCP	healthcare professional
LSHTM	London School of Hygiene and Tropical Medicine
ME	myalgic encephalomyelitis or myalgic encephalopathy
ME/CFS	Myalgic encephalomyelitis (or encephalopathy)/chronic fatigue syndrome
MRI	magnetic resonance imaging
NCC-PC	National Collaborating Centre for Primary Care
NHS	National Health Service
NICE	National Institute for Health and Clinical Excellence
NSAID	non-steroidal anti-inflammatory drug
PCT	Primary Care Trust
PIP	Personal Independence Payment
PIS	Participant Information Sheet
PVFS	post-viral fatigue syndrome
QALY	quality-adjusted life year

QoL	quality of life
RCGP	Royal College of General Practitioners
RCT	randomised controlled trial
SG	support group
SMC	standard medical care
SSRI	selective serotonin reuptake inhibitor
UC	Universal Credit

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