

Counting The Cost

Chronic Fatigue
Syndrome/Myalgic
Encephalomyelitis

28th September 2017

Full Report



The purpose of this report is to:



Support **improved NHS and societal understanding** of chronic fatigue syndrome / myalgic encephalomyelitis (CFS/ME);



Highlight **current inequalities** of care and support



Identify the **economic implications** of the condition – not just to the NHS, but also to UK society as a whole.

About this report

It is hoped that policy makers and commissioners will use this information to make decisions on the planning and funding of CFS/ME services and research.

For the sake of clarity and relevance to UK public health, we adopt the nomenclature of 'CFS/ME' throughout this report as a catch-all term, unless specifically quoting from sources that have used singular or other terminology.

The views expressed in this report are those of the authors alone. All facts have been checked for accuracy as far as possible.

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Julia Manning, Founding Director
2020health

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Foreword

David Butcher, Chairman

“As someone who has suffered from CFS/ME I understand how appalling this illness is, how it can devastate the lives of sufferers, their carers and their families.”



For the past 20 years there has been extensive argument in the UK about the causes and diagnostic criteria for this illness. For some considerable time, the conventional wisdom was that this illness was purely psychological in origin.

The World Health Organisation has clearly classified CFS/ME as a neurological disease in its International Classification of Diseases (ICD), section G93.3.

Despite this clarity, there is still a lack of universal agreement about CFS/ME in the UK. This has led to a paralysis of research into both the bio-medical causes of and treatments for CFS/ME, and the research that has been done has focused primarily on the psychological side.

The time has come for a proper research strategy for CFS/ME, looking at both bio-medical causes and treatments. In order to commence a dialogue with government and other interested parties, it is essential for everyone to be on the same page. To achieve that degree of agreement will be a challenge, but I believe the first step in that process is to start a new public conversation about this horrible illness.

The purpose of this report by the health think tank 2020health, sponsored by the Optimum Health Clinic, is to do just that. Nothing concentrates the mind like money. This is the first cost of illness study of CFS/ME to the UK economy combining direct costs (including primary and secondary care contacts, prescription and over the counter medications, and complimentary treatments) and indirect costs (including work productivity losses, informal care and welfare payments). The results are staggering.

In commissioning this report, our hope is that we can:

- Demonstrate clearly all the costs of CFS/ME to the UK economy;
- Use this report to start a new public conversation about the illness;
- Start a dialogue with all interested parties to create a new strategy to research the bio-medical causes of and treatments for CFS/ME.

The Optimum Health Clinic Foundation

Registered charity number: 1131664

Executive summary



“ Greater disability than those with type 2 diabetes, congestive heart failure, back pain/sciatica, lung disease, osteoarthritis, multiple sclerosis, and even most cancers.

Chronic fatigue syndrome (CFS), also known as myalgic encephalomyelitis (ME), is a complex, fluctuating condition characterised by emotional, mental and physical fatigue.

Accompanying symptoms typically include postexertional malaise or incapacitation, memory and concentration problems, musculoskeletal pain, headaches, sore throat, painful swollen lymph nodes and sleep disturbance (Fukuda, 1994; Carruthers, 2003). The National Institute of Health and Care Excellence (NICE) estimates the prevalence of CFS / ME to be ‘at least’ 0.2% to 0.4% of the UK population,

implying up to 1 in 250 people affected, or 260,000 in total.

Quality of life research suggests that the well-characterised CFS/ME sufferer may experience on average greater disability than those with type 2 diabetes, congestive heart failure, back pain/sciatica, lung disease, osteoarthritis, multiple sclerosis and even most cancers (Nacul et al., 2011a). Severe sufferers are largely housebound, the very severe confined to a bed most of the time and reliant on carers for all their needs, day and night (ME Association, 2007). In prolonged severe illness, associated psychological and physical health risks increase, including postural

hypotension, deep venous thrombosis, osteoporosis, deconditioning and pressure sores (NICE, 2007).

The average length of the illness is around six years, though some people live with CFS/ME for decades (Nisenbaum et al., 2000). In addition to the significant and protracted suffering caused by CFS/ME, patients may experience further psychological distress resulting from clinical and public scepticism, even stigma, still common in the UK (Action for ME: Time to deliver survey, 2014). A 2008 patient-group survey suggested that one third of GPs were not supportive in CFS/ME cases (Gibson et al., 2011); another survey found GPs on the whole expressing 'little confidence in positively attributing the label of CFS/ME to a patient and their symptoms' (Chew-Graham et al., 2010). The causes of CFS/ME remain unknown – a frustration to patients and clinicians alike.

Calls for action

CFS/ME sufferers are probably among the most marginalised patients in the UK. NICE's Guideline Development Group noted 'anecdotal reports of people with severe CFS/ME not seeing medical practitioners for many years' (NICE, 2007). Even now, more than one third of specialist adult CFS/ME services in the NHS provide no support to severely affected patients (McDermott et al., 2014).

Our own investigations suggest that some 14,000 people are referred to publicly-funded specialist CFS/ME services each year in the UK, with NHS running costs at around £14 million.¹ Approximately three quarters of people referred are diagnosed with CFS/ME.

In England, we estimate the number diagnosed in specialist services to be in the region of 10,000. Though the number of services does not appear to have risen by much in recent years², services themselves appear to have expanded slightly. This is potentially good news for people with CFS/ME whose CCG, health board or trust is providing the service; our FOI responses however reveal often minimal

referrals 'out of area' for CFS/ME patients, meaning that inequalities of access remain significant.

If just a small minority of CFS/ME sufferers have access to full specialist services in any particular year, it should also be noted that the average time to assessment in specialised services is three years four months (NOD, 2011). This can hardly be described as timely access.

It is likely that a lack of clinical specialism in CFS/ME is attributable in part to the trend of under-investment in chronic conditions generally (Monitor, 2013), and also a lack of appreciation as to the costs and societal implications of CFS/ME to the UK.

Our study

We undertook a comprehensive UK cost-of-illness study on CFS/ME, based on recorded patient data from both specialised services and primary care. Data were found in (i) economic evaluations within clinical trials for CFS/ME, and (ii) cross-sectional economic studies of CFS/ME. Only (peer-review) papers from the UK were included due to significant differences between the UK and other countries in regards to health care system structure, employment, earnings and benefits. Papers from other countries were used to corroborate findings.

With little data on welfare payments received by recruited patients, we also contacted the Department of Work and Pensions for estimates on Employment





Specialised CFS/ME services

Based on financial data obtained from 35 specialised CFS/ME services in the UK, service running costs average at just under £1,000 per referral, with 75% of those referred receiving a CFS/ME diagnosis.

A number of services reported an average of 8–10 clinical contacts (quoted range of 1 – 24 contacts) during the course of a year. Eight services reported running costs at less than £100,000 per annum.

2020health, 2016

“ *The total cost to the UK economy of CFS/ME in 2014/15 was at least £3.3 billion*

and Support Allowance (ESA) and Disability Living Allowance (DLA) payments to people with CFS/ME as a primary disabling condition.

According to our weighted analysis, the total cost to the UK economy of CFS/ME in 2014/15 was at least £3.3 billion, assuming a cautious estimate of 0.4% prevalence within the UK population.³ In our unweighted analysis, we found an average cost per person with CFS/ME of £16,966. These figures account for health care costs, the majority of disability-related welfare payments, productivity losses and unpaid informal care. We were unable to capture all CFS/ME costs. Missing costs included productivity losses among carers themselves, through reduced hours in employment, and costs associated with ‘presenteesim’ (productivity losses due to working while unwell). The true costs of CFS/ME to the UK are therefore likely to be much higher.

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Implications for the NHS and wider society

Health boards, CCGs and trusts that have not invested in CFS/ME expertise may be running false economies.

Our economic analysis revealed NHS spending on people with CFS/ME to be in the region of £542 million. Drawing on matched sample findings by Lin et al. (2011), this amounts to well over £300 million more than a ‘non-fatigued’ population.

Just 3% of the £542 million applies to the running of joined up, specialised services. Clinicians with CFS/ME specialism are not of course exclusive to such services, but it is highly probable that the NHS is spending substantial amounts of money on the non-specialised treatment of CFS/ME.

CFS/ME services

The specific advantages of a joined up, specialised CFS/ME service have not yet been systematically evaluated. However, there are strong reasons why commissioners need to consider investment in specialist CFS/ME care.

First is the economic reason. If a CCG, trust or health board has decided not to commission a specialist CFS/ME service they are still faced with potentially substantial expenditure on CFS/ME support, symptom management and treatment. Expenses associated with specialist care may not be much more than non-specialist care, and yet hold greater promise for return on investment, even in the short to medium term.

Second, we would not expect sufferers of (for example) MS, diabetes or heart failure to be advised, supported

£14 million

UK NHS spend on dedicated, specialised CFS/ME services

£542 million

Total UK health service spend on people with CFS/ME

£3.3 billion

Annual cost of CFS/ME to the UK

and treated by non-specialists. NICE claims that approximately half of all people with CFS/ME 'need input from specialist services' (NICE, 2007) – such is the complexity of the condition, especially among the moderately, severely and long-term affected.

Third, equality of access is a core value of the NHS. That many severely affected, housebound people with CFS/ME receive negligible or even no support from specialised services is no doubt distressing to both patients and their families. For sufferers across the range of CFS/ME severity, there is evidence that out of area referrals do not bring equality of access, running counter to NHS principles.

CFS/ME Research

The funding of CFS/ME research needs to be re-evaluated in light of the immense economic implications of the condition, which has greatest prevalence among the working-age population. The DWP alone pays out well in excess of £100 million annually in ESA and DLA payments to people with a primary disabling condition of CFS/ME; productivity losses (of patients and carers) mean lower revenues for businesses and government of a far greater order. Stronger research emphasis has the potential to diminish the economic impact of CFS/ME to wider society in the longer term.

Well-designed research is the best means by which new frontiers can be explored in CFS/ME care. It holds promise not just for patient outcomes, but could also resolve some of the disagreement between patient organisations and medical authorities on the nature of CFS/ME, which is confusing to patients and potentially steering some away from specific treatment options (Hossenbaccus & White 2013). In this respect, research needs to be designed in collaboration with CFS/ME patient organisations, drawing on patient insight and lived experience.

Conclusion

There is some outstanding work being done in support of CFS/ME sufferers across the UK, by local NHS and by patient-support charities, and also in the sphere of research. **But the picture in the UK as a whole is one of grossly unequal care, marginalised and sometimes forgotten patients, and in the light of our findings, probable false economies.**

Impact of greater access and quality of care may be discernible well within a funding cycle. Commissioners and central government need to reconsider funding decisions and organise CFS/ME services and research as appropriate to a treatable condition that has far-reaching societal and economic implications for the UK.

20-40

The peak age of onset of CFS/ME. (Capelli et al., 2010)

81%

Proportion of carers who are husbands, wives or partners. (Nacul et al., 2011)

76%

Proportion of CFS / ME sufferers who are female (Collin, 2011)

85%

Proportion of CFS/ME sufferers who have experienced some form of lost employment due to the illness. (PACE trial, 2012)

Introduction

It is perhaps surprising that only comparatively recently has CFS/ME become accepted as a genuine illness with unique features and symptoms, and therefore distinguishable from any other disease.

In the UK, clinicians received limited official guidance on CFS/ME until the late 1990s. At the request of the Chief Medical Officer, The Royal Colleges of Physicians, Psychiatrists and General Practitioners undertook a joint report on chronic fatigue syndrome in 1996 (revised 1997), and the Department of Health (DH) followed with the commissioning of an independent CFS/ME Working Group in 1998. The Working Group submitted its report to the Chief Medical Officer in 2002, detailing how the NHS might best provide care for people of all ages with CFS. Two years later the DH made £8.5 million available to create 12 new NHS CFS/ME service centres and 28 support teams for people with CFS/ME in England. That same year (2004), the National Institute of Health and Care Excellence (NICE) was tasked with creating clinical guidelines for the diagnosis and management of chronic fatigue syndrome, which it published in 2007.

Though the medical community is more informed on CFS/ME than previously, debate continues on cause, diagnosis, pathophysiology and treatment (Mallet et al., 2016). Certainly, recent research suggests many GPs have little confidence in diagnosing CFS/ME (Chew-Graham et al., 2010; Hannon et al., 2012), and by no means all of those who do identify the condition have referral opportunity to specialists, let alone multidisciplinary CFS/ME services (McDermott et al, 2014).

Studies have shown that individuals are much more likely to experience symptom improvement, even full recovery, when treated (Cairns & Hotopf, 2005;

White et al., 2013). There is also compelling evidence that the prolonged and debilitating nature of CFS/ME results in significant economic costs to society, owing in particular to indirect costs stemming from lost productivity and informal care (Jason et al., 2003; Collin et al., 2011). It can therefore be argued that aside from the clinical and moral arguments, there is an economic incentive to see CFS/ME sufferers – most of whom are of working and parenting age – given timely, appropriate treatment, so to diminish the negative repercussions on the UK economy.

In this report we draw on ‘costs of illness’ literature, cost-effectiveness studies and Freedom of Information (FoI) requests to produce an estimate of total direct and indirect costs of CFS/ME to the UK. It is our hope that policy makers and commissioners will be able to use this information to make decisions on the planning and funding of CFS/ME services and research.

Report structure

In Part 1, we discuss the nature of CFS/ME, its potential triggers and prevalence, and also the personal and societal impact of the condition. We then explore treatment pathways and conclude with an overview of services and access in the UK.

In Part 2, we present our macroeconomic study on CFS/ME, with data on direct and indirect costs drawn from peer-reviewed academic literature, with further evidence supplied through Freedom of Information (FOI) contact with specialist NHS CFS/ME services, trusts, health boards and the Department of Work and Pensions.

Methodology

In our research for Part 1 of this report we looked to peer-review and grey literature, and NHS and NICE guidance, to gauge clinical understanding and patient experience of CFS/ME. A rapid evidence review

conducted through the PubMed search engine enabled us to access meta-analyses and literature reviews on key aspects of CFS/ME epidemiology, including clinical criteria, prevalence studies and gender balance, as well as evidence of NHS services and treatment. Description of available treatment options was largely informed by NICE guidance. We also issued FOI requests to CFS/ME services throughout the UK to compare current referral rates to those estimated by earlier studies.

For Part 2 of this report, the cost-of-illness study, we conducted a rapid review of the literature in peer-review journals to identify studies quantifying health care resource use and/or productivity losses associated with CFS/ME. Data was found in (i) economic evaluations within clinical trials for CFS/ME, and (ii) cross-sectional economic studies of CFS/ME. Only

papers from the UK were included due to significant differences between the UK and other countries in regards to health care system structure, employment, earnings and benefits. Papers from other countries were however used to corroborate findings.

Complementing this work, we issued FOI requests to the Department of Work and Pensions to understand current expenditure on welfare payments to individuals with a primary disabling condition of CFS/ME. We also drew information from the above mentioned FOI requests to NHS specialised CFS/ME services to estimate specific expenditure in this area.

A Steering Group (see Appendix C) met three times during the project period to ensure we maintained focus and rigour of research, and drew relevant, objective conclusions.



PART 1:

Understanding a complex illness

1. 'Chronic fatigue syndrome' and 'myalgic encephalomyelitis'

The terms chronic fatigue syndrome (CFS) and myalgic encephalomyelitis (ME) have been used to describe a complex multi-systemic disorder characterised by chronic and intense fatigue and not explained by ongoing exertion or any existing medical condition. Typical symptoms include postexertional malaise, memory or concentration problems, muscle pain, joint pain, headaches, sore throat, painful swollen lymph nodes and sleep disturbance (Fukuda et al., 1994; Carruthers et al., 2003).

The median length of illness in adults is around six years, though for some, CFS/ME lasts decades (Nisenbaum et al., 2000). It has been also been suggested that at any one time up to 25% of CFS/ME cases may be classed as 'severe' (DH, 2002), where the individual becomes largely confined to house, wheelchair or bed, and dependent on carers for many or all of their needs. In prolonged severe illness, associated psychological and physical health risks increase, including postural hypotension, deep venous thrombosis, osteoporosis, deconditioning and pressure sores (NICE, 2007).

The causes of CFS/ME are unknown, although several factors have been suggested, including immunological, genetic, viral, neuroendocrine and psychological (Bagnall et al., 2002). Among the most common apparent triggers are infection and stressful life events (Kato et al., 2006; Faro et al., 2016). Most experts consider the condition heterogeneous and probably activated by a variety of factors (NICE, 2007; Holgate et al., 2011).

CFS/ME in children and adolescents

Children and adolescents with CFS/ME experience the common core symptoms, but otherwise they may present differently from adults.

In a study of UK and Dutch clinical cohorts, children under 12 years of age were found less likely to have cognitive symptoms and more likely to present with a sore throat. Adolescents (12–18) were more likely to have headaches and less likely to have tender lymph nodes, palpitations, dizziness, general malaise and pain, compared to adults. Adolescents were more likely to have comorbid depression but less likely to have anxiety, compared to adults (Collin et al., 2015).

Only recently have these differences in presentation been recognised and studied, meaning that clinical guidelines, teaching and definitions for paediatric patients have been largely based on descriptions of CFS/ME in adults (Collin et al., 2015).

1.1 Clinical criteria of CFS/ME

There are many sets of clinical criteria available to define CFS and/or ME; Brurberg et al. identified no less than 20 in 2014. The most widely used case definition is probably that of the Centre for Disease Control and Prevention (CDC), 1994 (shown in Appendix B of this report).

The multiplicity of clinical criteria, and indeed various preferences for specific definitions such as 'ME' or 'SEID' (systemic exertion intolerance disease),⁴ owes to the fact that the cause or causes of CFS/ME are unknown and there are no objective tests to identify the condition. In the UK, clinical guidance is provided by the National Institute of Health and Care Excellence (NICE), whose guidelines and criteria for the diagnosis of CFS/ME are shown below.

NICE guidelines for diagnosing CFS/ME

Guidelines issued in 2007 from the National Institute for Health and Care Excellence (NICE) state doctors should consider diagnosing CFS/ME if a person has fatigue and all of the following apply:

- it is new or had a clear starting point (it has not been a lifelong problem)
- it is persistent or recurrent, or both
- it is unexplained by other conditions
- it substantially reduces the amount of activity someone can do
- it feels worse after physical activity

The person should also have one or more of these symptoms:

- difficulty sleeping or insomnia
- muscle or joint pain without inflammation
- headaches
- painful lymph nodes that are not enlarged
- sore throat
- poor mental function, such as difficulty thinking
- symptoms getting worse after physical or mental exertion
- feeling unwell or having flu-like symptoms
- dizziness or nausea
- heart palpitations without heart disease

A diagnosis of CFS/ME can only be confirmed by a clinician after other conditions have been ruled out. The symptoms listed above must have persisted for at least four months in an adult and three months in a child or young person.

NICE's criteria are not as stringent as those of the Centre for Disease Control, 1994 (see Appendix B). Under CDC guidelines, 'CFS' should be considered only after six months of symptom duration, rather than four, and requires the presence of 'four or more' of eight named symptoms, as opposed to just 'one or more' of ten named symptoms.

Since CFS/ME symptoms are similar to those present in a number of other medical conditions, diagnostic criteria recommend a process of 'diagnosis by exclusion' over a set period. This unfortunately leaves individuals suffering, and attempting to manage, symptoms for several months (at a minimum) before any treatment for recovery may even be considered. And yet, symptoms at early onset can be severe: manifestation of CFS/ME is not necessarily linear. Moreover, following referral to services, there may be a further wait of many months before treatment actually begins.

1.2 Professional and public awareness

In 1980, most GPs did not accept the existence of chronic fatigue syndrome (Ho-Yen & McNamara, 1991). A lack of clinical belief in the condition was only exacerbated by the media promoting the term ‘Yuppie Flu’ during the 1980s (and beyond), an entirely unhelpful and stigmatising term that in any case falsely suggested predominant occurrence among burned-out high flyers.

In 1990, a study involving practices located within the Highland health board and Western Isles health board found 29% of GPs either undecided (22%) or doubtful (7%) as to the existence of chronic fatigue syndrome (Ho-Yen and McNamara, 1991). This indicated some improvement of GP acceptance of the condition over recent years, although the survey was confined to a narrow geographical area and thus not necessarily representative of the UK.

A larger survey in England in 2005, with 811 GP respondents across three regions, found that:

48% of GPs did not feel confident with making a diagnosis of CFS/ME and 41% did not feel confident in treatment. 72% of GPs accepted CFS/ME as a recognisable clinical entity and those GPs had significantly more positive attitudes.

Some 28% of GPs in this survey either did not accept, or were sceptical of, CFS/ME as a ‘recognisable clinical entity’ – very similar findings to those of Ho-Yen 15 years earlier. And yet, by this time several case definitions of CFS/ME had been supported by strategic medical bodies (e.g. in the USA, Canada and UK), and official statements had been made to the NHS on CFS/ME as a genuine medical condition (CFS Working Group / Chief Medical Officer 2002).

Since this time there has been limited published evidence of increasing GP capability in diagnosing and treating CFS/ME. A 2008 patient-group survey, reported in a RCGP paper on CFS (Gibson et al., 2011), suggested that one third of GPs were not supportive in CFS/ME cases; another survey found GPs on the whole

expressing ‘little confidence in positively attributing the label of CFS/ME to a patient and their symptoms’ (Chew-Graham et al., 2010).

When in 2014 Action for ME asked 50 GPs in Dumfries and Galloway, Fife and Highland about their educational needs around CFS/ME:

A large majority (82%) said they had not undertaken any training on ME-CFS, while nearly two thirds (66%) told us they were not aware of the SGPS [Scottish good practice statement on ME-CFS].

“ The disbelief and controversy over CFS /ME that exists within the professions has done nothing to dispel public disbelief in the existence of such a seemingly varied and inconstant illness.”

CFS/ME Working Group, 2002

With regard to initial medical education, a recent small UK study of medical students revealed little



understanding of CFS/ME deriving from the curriculum, with knowledge they did possess deriving largely from informal sources. Students expressed difficulty understanding chronic fatigue syndrome within a traditional biomedical framework (Stenhoff et al., 2015).

Recent efforts to improve awareness within the primary setting include an online training module for GPs, available on the Royal College for General Practice website (rcgp.org). However, because GPs see few people with CFS/ME, proportionally, their capacity for developing specific expertise remains limited (Bayliss et al., 2016).

Past clinical scepticism around CFS/ME has certainly not helped foster public recognition and compassion. The media has played an important role in this, and unfortunately ignorance persists in some quarters. In January 2016, The Sun published the headline, 'New research reveals that yuppie flu hits one in 50 teenagers'. Only after

Views on CFS/ME

Three national UK newspapers, UK ME websites, and UK medical websites and textbooks were analysed during 2010 for views on the nature of chronic fatigue syndrome.

Those that considered CFS/ME illness to be physical;

- 89% (32 of 36) of ME patient organizations,
- 58% (42/72) of newspaper articles,
- 24% (9/38) of medical authorities

63% (24/38) of medical authorities regarded the illness as both physical and psychological.
(Hossenbaccus, Z. & White, P. D. 2013)

the charity Action for ME protested against use of the stigmatising term 'yuppie flu' did The Sun change its online headline. Action for ME said the term was particularly ill-informed because, 'the research [from the University of Bristol] found that children from poorer families – not those "young upwardly-mobile professionals" – were more likely to have ME/CFS' (Chowdhury, 2016).

It is also important to note significant disagreement between CFS/ME patient organisations and medical authorities on the nature of CFS/ME (see insert), which will be further confusing patients and potentially steering some away from specific treatment options (Hossenbaccus and White 2013).

2. Personal, family and societal impact of CFS/ME

Eighteen months ago I was "normal". I worked 40+ hours a week, volunteered [for charity] in my spare time...and was always on the go. Almost overnight that all changed. I can no longer work. In fact, I am rarely able to leave the house, and spend up to 23 hours a day in bed. I have seen the world I know and love crumble around me; my family have become my carers; my relationship has suffered; and I have lost my connection to friends and the outside world, becoming a recluse.

(Stratton, 2015)

The historic lack of understanding, awareness and recognition of CFS/ME has no doubt created stigma, stress and added distress among many sufferers (Green et al., 1999). The ‘well characterised’ CFS/ME sufferer – experiencing persistent emotional, mental, and physical fatigue accompanied by a range of neurological and psychological problems – may experience on average greater disability than those with type 2 diabetes, congestive heart failure, back pain/sciatica, lung disease, osteoarthritis, multiple sclerosis and even most cancers (Nacul et al., 2011a). Little wonder that the CFS/ME patient can feel burdened by a sense of hopelessness, all the more so without opportunity of access to suitably qualified CFS/ME clinicians.

CFS/ME has profound impact on carers also, the majority being husbands, wives or partners, though parents and children may be the principal carer. With higher CFS/ME prevalence among women, it is common to find informal care provided by men. The emotional pressures on those caring for people with CFS/ME can be intense (Nacul et al., 2011a).

There is some evidence that with CFS/ME comes a significantly greater likelihood of suicide. One study in the USA found the three highest causes of death among those with CFS/ME to be cancer (37.8%), heart disease (19%) and suicide (19%) (Hornig, 2014). This was a small study only (examining 59 deaths), but with suicide representing just 1.6% of deaths from all causes in the general US population, the survey indicated a pressing need for further research in this area. DePaul University, Chicago, launched an ambitious ME and CFS Mortality Study in 2014, which is ongoing.

As already indicated, the impact on personal finances, both of sufferer and carer(s), is profound in the majority of CFS/ME cases. According to the National Outcomes Database, some 58% of individuals with CFS/ME who access specialist services are temporarily or indefinitely unemployed (Collin, 2011). In the PACE trial of 2011, 85% of participants were found to have experienced some form of lost employment due to the illness (McCrone et al., 2012). Moreover:

‘...families of patients must bear the costs of informal care, often reducing their own working hours. In young adults, disruption of education reduces productivity in later years.’ (Collin et al., 2011).

We explore in some detail the nature of cost in Part 2. But important to note are the wider economic implications of CFS/ME: reduced productivity among both sufferers and their carers (revenue and taxation), together with government spend on NHS services and welfare payments, underscores CFS/ME as a concern of individuals, of families and of society as a whole.

“ Dealing with multiple disappointments as the recovery process ebbs and flows over a long period of time is exhausting, as well as the sadness at the loss of opportunity and pain that your loved one is having to endure.”

Ian (carer), whose daughter has been living with CFS/ME for 11 years.

3. The demographics of CFS /ME

3.1 CFS/ME prevalence in the UK

Case definitions have an important bearing on prevalence estimates. In recognition of this fact some studies have contrasted several case definitions among single populations to attempt a more accurate estimate of prevalence (Johnston et al., 2013a). The issue is further complicated by the fact that patient self-reporting of the condition commonly results in significantly higher prevalence figures than those obtained in medical settings (Johnston et al., 2013b).

This is true of prevalence studies in the UK, as shown in Table 1.1, which contrasts findings from five studies, with prevalence estimates ranging from 0.2% to 2.6%, contingent on case definition and recruitment methodology.

When the National Institute of Health and Care Excellence (NICE) published its recommendations on CFS/ME in 2007, it stated that a lack of epidemiological data for the UK (at the time) meant that prevalence data needed to be extrapolated from other countries to arrive at a more informed deduction. Indeed, due to the limitations of individual prevalence studies, which tend to be confined to specific geographical localities, meta-analyses are a necessary reference point for estimating likely prevalence. NICE concluded that evidence suggested a population prevalence of 'at least' 0.2–0.4%.

Self-reporting studies, which in the UK have consistently estimated prevalence at just above 2%, or 1 in 50, give some context to NICE's 'at least' qualification. After all, both clinical assessment and population study questionnaires are reliant on the individual's subjective feedback – there are no diagnostic tests that prove CFS/ME. A more recent meta-analysis of CFS/ME in adults from countries worldwide (all studies using the 1994 CDC case definition) found pooled prevalence of self-reported CFS/ME at 3.28%, while results from seven studies using clinical assessment alone found adult prevalence at 0.76% (Johnston et al., 2013b).

Table 1.1. Self-reported and clinically-reported CFS/ME prevalence, UK.

| Reporting | Study | CFS/ME case definitions | Age range | Prevalence |
|--------------------------------|--------------------------|---|-----------|------------|
| Self-reported prevalence | Wessely et al., 1997 | CDC (1988/94), Oxford (1991) and Australian (1990) | 18–45 | 2.6% |
| | Cho et al., 2009 | CDC (1994) | 18–45 | 2.1% |
| | Bhui et al. 2011 | CDC (1994) | 16–74 | 2.3% |
| Clinically-reported prevalence | Ho-Yen and McNamara 1991 | Ho-Yen, 1990 | All ages | 0.13%. |
| | Nacul et al. 2011b, | CDC (1994), Canadian (2003) and Epidemiological (ECD, 2008) | 18–64 | 0.2% |

UK studies in paediatrics are also important. The Avon Longitudinal Study of Parents and Children (ALSPAC), 2016, published findings that CFS/ME affects 1.9% of 16-year-olds and is positively associated with higher family adversity. ALSPAC, as a large, parent/child-reporting study, found authorised half-day absences among CFS/ME sufferers to be three times greater than non-CFS/ME: 53.9 half-day absences per academic year, compared with 18.3 in the group without CFS/ME (Collin et al., 2016).

For clinically-assessed young people with CFS/ME, school absences appear to be much higher. In 2011 the National Outcomes Database reported that 58% of children and young people with CFS/ME attend school ≤ 2 days per week (Collin, 2011).

In conclusion, if incidence of CFS/ME in the UK remains uncertain, it is not unreasonable to assume prevalence somewhere between 0.2%–0.6% of the UK population as a whole (and up to 0.76 of the working-age population), taking into consideration recent epidemiological data and analyses drawn from clinical assessments.

3.2 Severity levels

In the UK, NHS Choices define three distinct levels of CFS/ME severity, as follows:

- mild – you're able to care for yourself, but may need days off work to rest
- moderate – you may have reduced mobility, and your symptoms can vary; you may also have disturbed sleep patterns and need to sleep in the afternoon
- severe – you're able to carry out minimal daily tasks, such as brushing your teeth, but have significantly reduced mobility, and may also have difficulty concentrating

(NHS Choices: Chronic fatigue syndrome)

NHS Choices' categorisation omits a fourth severity level, 'very severe'. Whereas 'severe' may imply largely housebound, in a 'very severe' case the individual would be bed-bound most of the time, perhaps even tube fed, suffering significant impairment of both physical and cognitive functioning and reliant on carers for all their needs, day and night (ME Association, 2007).

Few studies have captured CFS/ME prevalence broken down by severity, perhaps due to the questionable clinical utility of this knowledge, given that symptom severity fluctuates over time (NICE, 2007). Evidence

found through a process of rapid review is summarised in Table 1.2.

Strong variance among surveys may be explained by a dissimilarity of severity scoring and recruitment methodologies. Nevertheless, these results bring into question the estimate by NICE that up to 25% of CFS/ME sufferers are 'severe' at any one time. Findings suggest that this estimate may have been informed by national support groups, whose membership is perhaps less likely to include a balanced (indicative) proportion of individuals with mild CFS/ME.

It must not be forgotten however that even mild CFS/ME can be significantly disruptive to normal life. Even if 60% of people with CFS/ME suffer 'mild' symptoms at any one time (indicated by Pheby & Saffron, 2009), this group still has reduced capacity in every-day tasks, the workplace, education and training, and parenting.

Only through a large population study will epidemiologists be able to understand the distribution of severity levels among CFS/ME sufferers. It could be a valuable undertaking in order to understand to what extent the NHS is delivering equitable care to the population with CFS/ME.

3.3 Age and gender balance

The peak age of onset of CFS/ME is between 20 and 40 years (Capelli et al., 2010), and it is a curious fact of



Table 1.2. CFS/ME severity prevalence, UK studies

| Study | Mild | Moderate | Severe / very severe |
|--------------------------------|------|----------|----------------------|
| 1. Ho-Yen & McNamara, 1991 | 19% | 53% | 29% |
| 2. Dorset CFS/ME Project, 2006 | - | - | 10% |
| 3. Pheby & Saffron, 2009 | 60% | 28% | 12% |
| 4. Wearden et al., 2010 | - | - | 11% |
| 5. Action for ME survey, 2014 | 36% | 42% | 22% |

Notes:

1. Ho-Yen & McNamara, 1991: 289 participants in Scotland, Highland and Western Isles health boards.
2. McDermott M. 2006. Dorset CFS/ME Project. Conference proceedings of the British Association of CFS /ME (2006).
3. Pheby & Saffron, 2009. Enrolled 1104 participants with CFS/ME, of which 1037 were identified according to severity. Defined severity groups by Barthel scoring⁵ and housebound/bedbound status.
4. Wearden et al., 2010. Nurse led, home based self-help treatment for patients in primary care with chronic fatigue syndrome: randomised controlled trial.
5. Action for ME survey (2014). Recruited 2018 people with CFS/ME from within and beyond organisation membership.⁶

this condition that women are more commonly affected than men. The UK's National Outcomes Database (NOD) suggests that 76% of clinically-assessed adults with CFS/ME are female. Presentation in adolescents (12–18) also sees a strong gender bias (82% female), although in children under 12, gender balance is more equal (57% female) (Collin et al., 2015).

NOD data derives from specialised services. As we do not know whether men and women are equally inclined to attend services for assessment and treatment, it is difficult to know whether these results are indicative of the wider CFS/ME population.

Population studies involving random digit-dialling in the USA have also shown greater female susceptibility. A population study in Georgia in fact suggested stark differences in prevalence by gender across geographic strata. In the metropolitan area, CFS/ME prevalence in women was 11.2 times that in men, whereas in the urban and rural populations the female-to-male ratios of CFS/ME prevalence were 1.7 and 0.8, respectively (Reeves et al., 2007).

Gender as a risk factor for CFS/ME has been explored, without conclusive results. While studies have found measurable differences between women and men with CFS/ME, and that women are more likely than men to experience severe symptoms, illness patterns suggestive of different pathophysiological processes between the sexes have not been found (Tseng and Natelson, 2004; Faro et al., 2016).

4. Treatment options

The NHS states that treatments offered should depend on how CFS/ME affects the individual, with the understanding that what works for one person may not work for another (NHS Choices/Treatment). However, whilst NICE has issued guidance on treatment options for CFS/ME, not all recommended treatments are available to all CFS/ME sufferers due to the variability of service access, as previously noted.

NICE divides CFS/ME treatment into four categories:

1. Therapeutic interventions
2. Pharmacological interventions
3. Dietary interventions and supplements
4. Complementary therapies

NICE do not recommend all cited interventions, with some lacking an evidence base, and it has to be acknowledged that tensions exist among CFS/ME support groups, charities and patients on the one hand, and clinicians, researchers and policy advisors on the other, as to what treatment, or combination of treatments, should be promoted and valued in terms of outcomes and cost-effectiveness.

It is not the purpose of this report to compare and contrast interventions and outcomes. The topic is vast and beyond project scope. We therefore limit ourselves to a brief description of interventions – not exhaustive – which people with CFS/ME are accessing within and beyond the NHS and, to varying extents, reporting as helpful. We reference the NICE literature (Management 6.1–6.7) throughout 5.1 to 5.4, and cite references external to NICE as appropriate.

4.1 Therapeutic interventions

The primary interventions for CFS/ME approved by NICE are cognitive behavioural therapy (CBT) and graded exercise therapy (GET). Both involve a collaborative patient–clinician approach and have been shown, through randomised control trials, to be moderately effective treatments (Malouff et al., 2008;

PACE, 2011).⁷ NICE recommends these interventions in the treatment of mild and moderate CFS/ME, while principles of CBT and GET may be incorporated into a care programme for people with severe symptoms.

CBT aims to reduce the levels of symptoms, disability and distress associated with CFS/ME. It is intended to enable validation of the person's symptoms and condition and develops awareness of thoughts, expectations or beliefs about CFS/ME. It also addresses lifestyle choices and stress management, explores problem solving and goal setting, and treats any associated or comorbid anxiety, depression or mood disorder.

GET involves physical assessment, baseline measurement and meaningful goal-setting and education. Physical activity is individually tailored with planned increases in the duration of exercise, followed, in turn, by an increase in intensity when the individual is able, with the objective of improving symptoms and functioning. Personnel involved in this care include occupational therapists, physiotherapists and rehabilitation care assistants.

Pacing, or 'adaptive pacing therapy' (APT), is a self-management approach, drawing on some concepts used in 'activity management', where periods of activity are balanced with periods of rest. Many people with CFS/ME have reported pacing as helpful, despite a lack of evidence base in trials (NICE, 2007; PACE, 2011). Pacing is a prime example of stakeholder disagreement: in Action for M.E.'s 2014 survey of more than 2,000 CFS/ME sufferers, 85% of respondents who had used pacing considered it 'a little or very helpful'.

Therapeutic interventions also extend to sleep management, which includes sleep hygiene, using behavioural approaches, and changes in environmental conditions, introduced to improve the quality of sleep. NICE also recommends that relaxation techniques, such as guided visualisation or breathing techniques, should be suggested (as appropriate) for the management of pain, sleep problems and comorbid stress or anxiety. Such interventions may be used in

combination with components of CBT or GET (NICE Guidance 1.6.2.5).

4.2 Pharmacological treatments

NICE states there is 'equivocal and limited evidence on the overall benefits of pharmacological treatments for CFS/ME'. A recent review of management issues with CFS/ME confirmed that effective medications for the illness remain elusive, especially in the treatment of the primary symptom of fatigue (Bourke, 2015).

Pharmacological treatments are however used extensively in the NHS to aid relief of other symptoms of CFS/ME. For example, over-the-counter painkillers can help ease muscle pain, joint pain and headaches. Antidepressants, specifically amitriptyline, may be considered for people with pain or sleep problems (NHS Choices).

4.3 Dietary interventions and supplements

A balanced diet with all the essential nutrients is of course vital for optimum health, and diet should be examined as part of a person's overall CFS/ME assessment.

The issue of 'dietary intervention', specifically an exclusion diet or dietary manipulation, is quite different. While NICE does not specifically recommend their use, it is acknowledged that 'many people find them helpful in managing symptoms, including bowel symptoms'.

Supplements used by people with CFS/ME have included vitamin B12, vitamin C, magnesium, NADH (nicotinamide adenine dinucleotide) and multivitamins and minerals. While NICE does not approve their use for treating the symptoms of the condition, it admits that some people have reported finding these helpful as a part of a self-management strategy for their symptoms. NICE also recognises that some people with CFS/ME need supplements because of a restricted dietary intake or nutritional deficiencies (NICE, 1.4.7).

4.4 Complementary therapies

NICE does not recommend complementary therapies as effective treatments for CFS/ME, although, as with supplements, it acknowledges that some people with CFS/ME 'choose to use such therapies for symptom control, and find them helpful' (NICE, 1.4.7.1). In fact, complementary and alternative medicines (CAMs) are popular with patient groups, with one recent study finding 74% patient organisation support for CAMs in the treatment of CFS/ME (Mallet et al., 2016).

Complementary therapies are only occasionally found in NHS services, although acupuncture is now used in many NHS general practices, as well as in the majority of pain clinics and hospices in the UK (NHS Choices). Mindfulness, using breathing and meditation techniques, is also emerging as an offered intervention.⁸ CAMs, in practice, are probably used by a large minority of CFS/ME sufferers (25% to 33%, according to the PACE trial, 2011), with most paying privately for the treatment.

4.5 Delivery of treatments

NICE guidelines for diagnosis and treatment of CFS/ME recommend that referral to specialist care should be offered within six months of presentation for mild CFS/ME, within three to four months for moderate CFS/ME and immediately for (suspected) severe CFS/ME (NICE, 2007).

In most places in Scotland, Wales and Northern Ireland, as well as in many CCG areas in England, CFS/ME care is coordinated by GPs who refer to individual services as they deem appropriate. GPs have access to resources to help diagnosis and decision making. The 'Scottish Good Practice Statement on ME-CFS',

for example, supplies a diagnostic, management and referral algorithm (care pathway) for GPs.

It is in theory ideal for treatment to be delivered through a specialised multi-disciplinary CFS/ME service, so to ensure an integrated, non-duplicating approach with the involvement of CFS/ME experts in relevant fields. Access to experienced clinicians is otherwise not guaranteed.

Whether care is delivered within the primary care setting, under a trust's specialised service or even privately, there needs to be good communication and regular contact between the (coordinating) healthcare professional and the individual. The healthcare professional should use their clinical judgement to tailor the investigations and interventions required to individual need (NICE CG53; 6.1)

“ Healthcare professionals should explain that no single strategy will be successful for all patients, or during all stages of the condition.”

NICE, CFS/ME Clinical Guidelines, 2007

Many specialised services offer treatments based on graded activity management strategies within a cognitive behavioural framework (shsc.nhs). A structured programme may cover topics such as:

- Monitoring activity levels
- Establishing baselines
- Identifying appropriate graded increases
- Moderating patterns of activity and rest
- Sleep management
- Relaxation training
- Movement and stretching
- Understanding and managing symptoms
- Understanding medication

- Dietary advice
- Coping with flare ups
- Identifying and overcoming blocks to progress

(Sources: leedspft.nhs; kmpt.nhs)

The MDT delivering CFS/ME advice and treatment may variously include a GP with a Special Interest (GPwSI), consultant physician/psychiatrist, specialist nurse, occupational therapist, physiotherapist, clinical psychologist, dietician and perhaps a counsellor. In rare cases, as in the service provided by The Royal London Hospital for Integrated Medicine, a mindfulness facilitator is also available.

It appears that in the majority of cases, just one course of treatment is offered to patients through an NHS specialised service, with patient contacts with services perhaps averaging between 8 and 12 during the course of a year.⁹ Interventions such as CBT and GET may be delivered one-to-one or in group settings.

Private practice

CFS/ME sufferers may pay privately for treatment. Possible reasons for private expenditure include lengthy NHS waiting times, lack of access to CFS/ME specialists or preferred treatment options, and lack of support following completion of an NHS treatment pathway. There is some evidence that those who receive a course of CFS/ME treatment through the NHS are often offered no follow-up support (Action for M.E. 2014 survey).

There are a variety of institutions across the UK that provide private outpatient or inpatient ('residential') care for CFS/ME sufferers. These range from single treatment specialities to MDT services, including integrative medicine and other mind-body-spirit approaches.

5. CFS/ME support and services in the NHS

A general practice with 10,000 patients is likely to include up to 40 people with CFS/ME; half of these

people will need input from specialist services.

NICE, 2007

“Over a third of specialist adult ME/CFS services in the NHS provide no service to severely affected patients, and a further 12 per cent of trusts provide only minimal or occasional help to housebound patients, primarily due to lack of funding.”

(McDermott et al., 2014)

As a population, CFS/ME sufferers are probably among the most marginalised patients in the UK. NICE's Guideline Development Group noted 'anecdotal reports of people with severe CFS/ME not seeing medical practitioners for many years' (NICE, 2007). GP awareness of CFS/ME is often lacking, while some remain sceptical of the condition (Chew-Graham et al., 2010). And across all levels of severity, only a small minority of CFS/ME sufferers have access to full specialist services in any particular year. Among these patients, the average time to assessment is three years

four months (Collin, 2011).

The situation for severely affected CFS/ME sufferers may not have improved in recent years, in spite of increased NHS awareness. A recent study found 33% of specialist CFS/ME services in England offering no service for housebound patients (McDermott et al., 2014). Support may be even less in Wales, Scotland and Northern Ireland, where dedicated services appear even more scarce.

Action for M.E.'s 2014 survey of more than 2,000 CFS/ME sufferers (all severities) found that more than half (54%) had not attended an NHS CFS/ME clinic in the past five years (Action for M.E., 2014). Even though many specialised services (most run by trusts) accept referrals across CCG borders, in reality, numbers referred out of area can be less than 20, even single figures, across an entire CCG region in any given year.¹⁰

We found 56 specialised CFS/ME services in the UK operating during the accounting years 2013–15 (England 51, Wales 1, Scotland 3, Northern Ireland 1). Available treatments, staff and patient contacts varied considerably. Based on 54 FOI request responses (or feedback) received from trusts, health boards and community interest companies (CICs)¹¹ between

In Scotland, NHS Greater Glasgow and Clyde (NHSGG&C) has set up **the NHS Centre for Integrative Care**, which provides a comprehensive 90-minute integrative CFS/ME assessment in the form of an outpatient clinic. A pathway is agreed with both the clinician and the patient, with programmes including **Mindfulness Based Cognitive Therapy (MBCT)**; **Moving into Balance** (aiming to balance energy, rebuild stamina and reduce stress); and both **Art therapy and Music therapy**.

Source: 2020health correspondence with NHSGG&C, 2016.

March and July 2016, we estimate that some 14,000 people are referred to NHS CFS/ME services each year in the UK, with around three quarters diagnosed as CFS/ME cases, and with NHS running costs at around £14 million.¹² Less than one quarter of responding specialised CFS/ME services reported activity in delivering treatments to children under the age of 16.

In England, we estimate the number diagnosed in specialist services to be in the region of 10,000. Though the number of services does not appear to have risen in recent years (with similar numbers of services decommissioned and newly commissioned), services themselves appear to have expanded slightly in size. A previous estimate based on data from 2008–10 indicated that each year in England some 11,000 adults are referred, 9000 are assessed and 7000 adults receive a diagnosis of CFS/ME (Collin et al., 2012).

Since treatment outcomes have not been systematically compared with those of primary care, it should not be suggested that patients seen in such services always receive better care. CFS/ME experts are not exclusive to dedicated CFS/ME services. In Wales, just one health board, Betsi Cadwaladr University Health Board (BCUHB), commissions a dedicated CFS/ME service. However in Powys, people with CFS/ME can access services through the Pain and Fatigue Management Service, where they receive a full biopsychosocial assessment and create a care plan in collaboration with a clinician. For some complex cases, attendance at a three week residential programme may be an option.

While dedicated CFS/ME services are also infrequently found in Scotland (11 out of 14 health boards have no specific CFS/ME service),¹³ NHS boards are advised to observe the 'Scottish Good Practice Statement on ME–CFS', which states the necessity of adopting a 'holistic approach to care and symptom control', defined as 'comprehensive patient care that considers the physical, psychological, social, economic and spiritual needs of the patient and his or her response to the illness' (SGPS, 2010).

It is very difficult to judge the overall effectiveness of CFS/ME care in the UK, although some evidence on specialised services has been gathered by the CFS/ME National Outcomes Database (NOD). The NOD holds the largest CFS/ME disease register in the world, with assessment data on more than 7,000 adults and 1,500 children with CFS/ME. A recent NOD-based study found that patients who attend NHS specialist CFS/ME services were experiencing moderate improvements in fatigue, anxiety and depression, but were seeing limited improvements in physical function as compared with CFS/ME patients in the PACE trial (2011). The authors could not determine whether these results were due to differences in the delivery or the content of treatments offered by NHS services (Crawley et al., 2013).

It has been suggested that both GPs and patients would like to see the development of an online resource that provides instant access to advice from CFS/ME specialists, so to help reduce the emotional and physical burden currently experienced by those with CFS/ME who are either left with no support or on long waiting lists for secondary care services (Bayliss et al, 2016).

Whilst 'virtual' access to CFS/ME specialists may be highly valued, there remains a pressing need for greater physical access to specialist care, with wider availability of CFS/ME expertise in each and every commissioning region of the UK. Furthermore, there is still much work needed to understand the value of treatment regimens that take a fully holistic approach to CFS/ME management and recovery, bearing in mind that trials have found the NHS-recommended interventions of CBT and GET to be just 'moderately effective' for the treatment of CFS/ME (Crawley et al., 2013).

6. Summary (Part 1)

There is very little equity in terms of CFS/ME support and treatment in the UK. GPs see few people with CFS/ME, proportionally, and their ability to develop specific expertise is therefore limited; moreover,

access to healthcare professionals with appropriate skills and expertise in CFS/ME management remains extremely patchy throughout the UK, especially for the most severely affected. In England, access to specialist care may have become more inequitable with the dissolution of PCTs and formation of CCGs, since individual commissioning regions have increased by 40% and 'out of area' referrals to specialist CFS/ME services are often in single or very low double figures. Whilst the numbers of referrals may have risen, the majority of access still derives from the close geographical vicinity of the specialised service.

Chronic conditions are a soft target in an age of NHS austerity, but for treatable conditions the decision not to invest can be a false economy. Commissioning decision making would benefit from a wider understanding of NHS costs associated with CFS/ME. How much does the condition cost the NHS, with and

without the £14m investment in specialised services? It is also important to understand whether government funding of CFS/ME research, the exact investment of which is unknown,¹⁴ is planned appropriately in relation to the full economic impact of CFS/ME to the UK. Part 2 of this report studies the wider financial and societal context in order to inform this debate.

PART 2:

The economic impact of CFS/ME in the United Kingdom: direct and indirect costs

Introduction

CFS/ME has significant economic implications for individuals and society as a whole. Studies of the economic impact of CFS/ME in the UK have provided evidence of direct costs (including primary and secondary care contacts, prescription and over-the-counter medications, and complimentary treatments) and indirect costs (including work productivity losses, informal care and welfare payments) (McCrone et al., 2003; Sabes-Figuera et al., 2010). No previous study, to our knowledge, has attempted to combine data from multiple studies in order to pool findings, refine estimates (by mitigating the potentially distorting effects of specific methodologies) and apply costing data across the UK as a whole. The true costs of CFS/ME to the UK are therefore unknown.

Methods

Data for costs of CFS/ME

Cost of illness studies commonly use population based databases to calculate (i) the prevalence of the disease, and (ii) health care resource use and productivity losses. CFS/ME is poorly coded in population based databases, in part due to a lack of understanding as to the causes of CFS/ME (Prins et al., 2006, Chew-Graham et al., 2008); a cost of illness study using this data was therefore not possible. In the UK, the CFS/ME National Outcomes Database (NOD) is a particularly valuable resource but does not capture data for CFS/ME patients outside of specialist services.

To understand costs corresponding to the wider CFS/ME population, we conducted a rapid review of the literature in peer review journals to identify studies quantifying health care resource use and/or productivity losses associated with CFS/ME. Data were found in (i) economic evaluations within clinical trials for CFS/ME, and (ii) cross-sectional economic studies of

CFS/ME. Only papers from the UK were included due to significant differences between the UK and other countries in regards to health care system structure, employment, earnings and benefits. Papers from other countries have however been used to corroborate findings.

Data on health care resource use, medication, informal care and productivity were extracted from the five papers. For health care resource use other than medication the average number of contacts with services was inputted. Where papers reported only costs of health care, costs were divided by unit costs reported in the paper to calculate the original number of service contacts. Health care resource use, rather than costs, was used so that 2014/15 health care unit costs could be applied consistently across different studies (see Table 2.1 for unit costs used). For trials, where possible, only data collected at baseline before patients received the intervention, or data corresponding to treatment as usual, were used to attempt to describe the cost of standard care. If this was not possible, data collected at later follow-up periods in the trial were used. The average economic cost of CFS/ME in the UK for 2014/15 was calculated from the average for each category of costs across papers.

We also calculated a weighted number of contacts per week for each health care resource type to weight for the number of patients with CFS/ME in each paper, so that papers with larger sample sizes were more heavily represented in the final calculation. The average weekly cost of health care resource was calculated by dividing the average use of each resource type (e.g. outpatient appointments, GP contacts) by the number of weeks data was collected and multiplying resource use by number of participants in the relevant study. The weekly total contacts for each type of

Table 2.1: Health care resources use unit costs in 2013/14 GBP

| Resource | Unit cost | Source |
|-------------------------|-----------|--------------------|
| Inpatient | £608 | PSSRU |
| Outpatient | £112 | PSSRU |
| Accident and Emergency | £108 | Reference costs |
| Daycase Surgery | £704 | PSSRU |
| Day Facility | £32 | PSSRU |
| GP – Surgery | £40 | PSSRU |
| GP Home | £82 | PSSRU |
| Practice Nurse | £14 | PSSRU |
| District Nurse | £45 | PSSRU |
| Occupational Therapist | £31 | PSSRU |
| Physiotherapist | £38 | PSSRU |
| Counsellor/Psychologist | £177 | Reference costs |
| Nutritionist | £80 | Reference costs |
| Phlebotomy | £149 | Reference costs |
| Complementary therapies | £48 | McCrone et al 2012 |
| Pharmacist | £88 | PSSRU |
| Psychiatrist | £51 | PSSRU |
| Neurologist | £174 | PSSRU |

health care resource use were then summed across studies and divided by the total number of participants that contributed data to that type of health care resource use across all studies, enabling calculation of the weekly average weighted resource use per patient. This was multiplied by the costs in Table 2.1 to calculate the average weighted weekly cost of health care resource per patient with CFS/ME. This was multiplied by 52 to extrapolate to 1 year.

Medication costs, informal care costs and lost earnings were inputted into Excel as total costs reported in cross-sectional studies or weighted averages across trial arms for trials. Medication costs were updated to 2014/15 prices using the retail prices index (ONS 2016). The cost of informal care and lost earnings was updated to 2014/15 prices using percentage change in median earnings between the year of the cost analysis and 2014/15 (ONS 2016).

Welfare payments

Limited information is available on incapacity benefits for CFS/ME. Information on payments for Disability Living Allowance and Employment and Support Allowance was obtained from the UK Department of Work and Pensions (DWP). Data from papers were compared with data from DWP to corroborate findings.

Sources for CFS/ME prevalence

Estimated prevalence of a condition has very specific implications for a costing study. As described in Part 1, NICE estimates a CFS/ME population prevalence of 0.2–0.4%, based on international evidence (NICE, 2007). More recently, a meta-analysis of seven prevalence studies of CFS/ME in adults from countries worldwide (all using the 1994 CDC case definition) suggested clinically-reported prevalence among adults to be higher, at 0.76% (Johnston et al., 2013b).

We used prevalence estimates of 0.2% and 0.4% to calculate the total economic cost of health care, medication and informal care. The prevalence figure of 0.76% of CFS/ME in working age adults was used to calculate the cost of productivity losses.

Extrapolated costs are based on a UK population projection for mid-2014 of 64.6 million, of which 42 million (65%) were aged 15 to 64 (ONS 2015).

Results

Details of the five UK papers identified that met the inclusion criteria for the analysis are reported in Table 2.2. The majority of papers (three) are cross-sectional studies; three papers recruited patients from primary care and two from specialist CFS/ME services. Of the two trial papers, Richardson et al., (2013) did not collect data at baseline, only at 20 weeks and 70 weeks after randomisation. There was a significant amount of missing data at 70 weeks so only data collected at 20 weeks were included. One study reported lost earnings only (Collin et al., 2011), the other four papers reporting health care resource use, informal care and lost earnings. McCrone et al. (2012) also report the percentage of patients that receive welfare payments and the cost of a predefined specialist services package for CFS/ME.

Table 2.2. Characteristics of studies included in analysis

| First Author (year) | Year of cost data | Trial or cross-sectional | Number of CFS/ME patients | Services recruited from | Follow-up duration (weeks) | Data reported |
|----------------------|-------------------|--------------------------|---------------------------|---------------------------------------|----------------------------|---|
| Collin (2011) | 2010 | Cross-sectional | 1991 | Specialist Services, England | 52 | Lost earnings |
| McCrone (2003) | 2000 | Cross-sectional | 44 | Primary Care, London | 12 | Health care costs, informal care, lost earnings |
| Sabes-Figuera (2010) | 2006 | Cross-sectional | 222 | Primary Care, London and South Thames | 26 | Health care resources, informal care, lost earnings |
| McCrone (2012) | 2009 | Trial | 640 | Specialist Services, UK | 26 | Health care resources, informal care, lost earnings, percentage receiving welfare |
| Richardson (2013) | 2008 | Trial | 296 | Primary Care, North West England | 20 | Health care resources, informal care, lost earnings |

Data derived from patients attending specialised services needed to be treated with caution. Not all patients with CFS/ME are referred to a specialist service, and both mild and very severe (housebound) are most likely underrepresented; Collin et al. (2011) estimate that 11.54 per 100,000 of the total UK population are referred to specialist services.

To calculate productivity losses the five studies all used the human capital approach, multiplying time off work by earnings, to calculate the cost of lost productivity. The human capital approach is widely criticised for over estimating the true cost of loss of productivity to the economy as it does not take into account that some replacement occurs when people leave (Pritchard and Sculpher, 2000). This was adjusted for in Sabes-Figuera by multiplying the total by 80%. We have applied this same adjustment to the other four studies.

Table 2.3 reports the average cost of health care, medication, informal care and productivity losses across the five studies adjusted to 2014/15 GBP prices. The four studies that identify health care use report very similar costs, with little difference between costs reported in McCrone et al (2012), who recruited patients with CFS/ME from specialist services, and the other three studies that recruited from primary care. Medication, informal care and productivity costs differ widely across the five studies. In particular the two studies that recruited from specialist services (McCrone et al., 2012 and Collin et al., 2011) appear to report significantly higher productivity losses, suggesting higher productivity losses for more severe and longer-suffering patients. This hypothesis is supported by the findings of Collin et al. (2011), that greater levels of fatigue are associated with cessation of employment.

It is not clear why McCrone et al. (2003) report significantly higher informal care costs, although a shorter follow-up period of 12 weeks may have enabled them to collect more reliable data on the involvement of carers of people with CFS/ME. Lower productivity losses in this study may be attributed to the fact that lost earnings from long term unemployment (employment cessation begun prior to the study) were not captured. These issues serve to highlight the fact that costing differences between the studies in the areas of medication, informal care and productivity losses are due in part to the different methodologies employed.

Using the data in Table 2.3, calculating productivity losses as higher for patients in specialist services and assuming a prevalence of CFS/ME of 0.4%, the total economic cost of health care in the UK is £357 million, this figure including £38 million for medication. The total cost of informal care is £2,140 million and £873 million

Table 2.3. Average costs of health care, medication, informal care and productivity losses across five studies for 2014/15 (GBP)

| Study | Health care costs (weekly average) | Health care costs (yearly average) | Medication costs (yearly average) | Informal care costs (yearly average) | Productivity losses (yearly average) | Total |
|----------------------|------------------------------------|------------------------------------|-----------------------------------|--------------------------------------|--------------------------------------|----------------|
| Collin (2011) | | | | | £18,522 | £18,522 |
| McCrone (2003) | £31 | £1,604 | £72 | £17,549 | £1,983 | £21,207 |
| McCrone (2012) | £24 | £1,262 | £108 | £6,349 | £12,985 | £20,704 |
| Richardson (2013) | £21 | £1,072 | £366 | £6,926 | £7,372 | £15,735 |
| Sabes-Figuera (2010) | £19 | £1,002 | £47 | £2,308 | £5,306 | £8,663 |
| Average | £24 | £1,235 | £148 | £8,283 | £9,234 | £16,966 |

for productivity losses, the lower value for productivity losses based on the assumption that only 65% of the population are of working age, whereas informal care costs are applied to the population across all age groups. The total economic cost of CFS/ME in 2014/15 was £3,372 million, excluding incapacity benefits discussed below.

If we assume a prevalence of CFS/ME at the lower end of the estimations of 0.2% the total cost of health care (including medication) is £180 million, the cost of informal care £1,070 million and the cost of productivity losses £463 million, for a total cost to the economy of CFS/ME in 2014/15 of £1,713 million.

If we assume that the prevalence of CFS/ME is at the higher end of 0.76%, but for working age adults only, the cost to the economy of CFS/ME in 2014/15 could be as high as £4,744 million, the majority as a result of the cost of informal care (£2,643 million), followed by productivity costs (£1,612 million) and health care costs (all inclusive) of £441 million.

Comparison with weighted average costs

Table 2.4 reports similar results to Table 2.3, but weighting for different sample sizes in each study, so that studies with larger sample sizes contribute more to the average than smaller studies. The results are similar to just using averages across studies, suggesting consistency in the results across different studies regardless of sample size. The cost of health care is slightly higher however, potentially due to double counting of specialist services as a result of different categories of health care use being used across different studies. The results remain broadly the same though. This is because informal care and productivity account for the majority of the cost to the economy of CFS/ME.

Table 2.4. Total weighted average cost across five studies of CFS/ME in the UK, 2014/15

| RESOURCE | Number studies reporting values | Weighted average yearly cost | CFS/ME prevalence of 0.4% | CFS/ME prevalence of 0.2% | CFS/ME prevalence of 0.76% (working age adults only) |
|---------------------------|---------------------------------|------------------------------|---------------------------|---------------------------|--|
| Inpatient | 3 | £157 | £40,594,459 | £20,297,230 | £50,134,157 |
| Outpatient | 3 | £252 | £65,070,452 | £32,535,226 | £80,362,008 |
| Accident and Emergency | 3 | £84 | £21,669,184 | £10,834,592 | £26,761,442 |
| Daycase Surgery | 1 | £111 | £28,760,569 | £14,380,285 | £35,519,303 |
| Day Facility | 1 | £11 | £2,905,108 | £1,452,554 | £3,587,808 |
| GP - Surgery | 4 | £366 | £94,535,118 | £47,267,559 | £116,750,870 |
| GP Home | 1 | £16 | £4,089,393 | £2,044,697 | £5,050,401 |
| Practice Nurse | 3 | £11 | £2,971,424 | £1,485,712 | £3,669,708 |
| District Nurse | 2 | £1 | £289,646 | £144,823 | £357,713 |
| Occupational Therapist | 1 | £15 | £3,910,543 | £1,955,272 | £4,829,521 |
| Physiotherapist | 4 | £102 | £26,443,302 | £13,221,651 | £32,657,478 |
| Counsellor / Psychologist | 2 | £250 | £64,620,337 | £32,310,169 | £79,806,116 |
| Nutritionist | 1 | £152 | £39,188,696 | £19,594,348 | £48,398,040 |
| Phlebotomy | 1 | £149 | £38,551,691 | £19,275,845 | £47,611,338 |
| Complementary therapies | 3 | £194 | £50,061,367 | £25,030,683 | £61,825,788 |
| Pharmacist | 1 | £42 | £10,805,125 | £5,402,562 | £13,344,329 |
| Psychiatrist | 1 | £9 | £2,370,442 | £1,185,221 | £2,927,496 |

| | | | | | |
|--|---|---------|-----------------------|-----------------------|-----------------------|
| Neurologist | 1 | £14 | £3,560,780 | £1,780,390 | £4,397,563 |
| Medication | 4 | £159 | £41,016,562 | £20,508,281 | £50,655,454 |
| Total health care cost (including medication) | | £2,095 | £541,414,198 | £270,707,099 | £668,646,535 |
| Standardised medical care | 1 | £148 | £1,103,261 | £1,103,261 | £1,103,261 |
| Total all health care (inc specialist & meds) | | | £542,517,459 | £271,810,360 | £669,749,796 |
| Informal Care | 4 | £6,155 | £1,590,307,280 | £795,153,640 | £1,964,029,490 |
| Productivity losses primary care | 3 | £6,134 | £1,000,478,885 | £485,378,805 | £1,927,659,028 |
| Productivity losses specialist services | 2 | £17,175 | £83,221,940 | £83,221,940 | £83,221,940 |
| Total productivity | | £23,309 | £1,083,700,825 | £568,600,745 | £2,010,880,969 |
| Total cost | | | £3,216,525,564 | £1,635,564,745 | £4,644,660,255 |

Incapacity benefits

McCrone et al. (2012) was the only study to report a percentage of people who claimed incapacity and illness disability benefit. Thirteen percent of patients (83 of the 641 patients) reported receiving income benefits 6 months before randomisation and 28% (177 of 641 patients) reported receiving illness disability benefit or disability living allowance (DLA).

This information is for specialist services only. If the percentage receiving income benefits (now called employment and support allowance (ESA)) is extrapolated to all working age adults with CFS/ME, assuming a prevalence of 0.4%, then 27,747 adults would receive income benefits. This is a higher number than that reported by the DWP of 21,120 people receiving income benefits under the category of "other disorders of the brain",¹⁵ in which CFS/ME is included. If instead the assumption is that 13% of working age adults accessing specialist services receive ESA, and for those that access primary care only half that, 6.5%, receive ESA, the total number of working age adults with CFS/ME that receive ESA is 11,187.

Collin et al (2011) report that the median duration of unemployment for someone with CFS/ME accessing specialist services is 11 months. Based on this finding and a weekly ESA payment of £109, the total cost in 2014/15 of ESA payments for CFS/ME in the UK may have been in the region of £50 million.

The 2014/15 figure for DLA payments to individuals with a primary disabling of CFS/ME has been released by the DWP in response to a Freedom of Information request.¹⁶ The DWP estimates that there were some 14,000 CFS/ME claimants with a case in payment on Disability Living Allowance in 2014/15, at a cost of £64 million.

The similarity of costs between ESA and DLA in relation to CFS/ME is not unexpected. Across all conditions, overall net payments of ESA/IB/SDA and DLA in 2014/15 were:

ESA/IB/SDA 13.8bn (13,807m)

DLA (not including PIP) £13.8bn (13,798m)

Source: DWP Gross Benefit Expenditure, 2014/15

The ESA/IB/SDA to DLA ratio across all conditions was thus close to 1:1 in 2014/15. Information considered, we estimate that welfare benefits for CFS/ME sufferers specifically were in excess of £110m in 2014/15. This figure almost certainly falls short of the total paid out by government in benefits related to CFS/ME, bearing in mind additional costs associated with claimants of (i) Personal Independence Payments (PIP), (ii) Carer's Allowance and Attendance Allowance, and (iii) housing benefit, where these are due to CFS/ME as a primary disabling condition.

In the McCrone et al. 2012 study, six percent of patients also reported payments from income protection schemes or private pensions. It is unclear what the cost to the economy is of these payments.

Discussion

The total cost to the UK economy of CFS/ME in 2014/15 was approximately £3.3 billion in our weighted analysis, assuming a prevalence of CFS/ME of 0.4% of the population, with an average cost per person with CFS/ME of £16,966 in the unweighted analysis. The true cost though could be as high as £4.8 billion and as low as £1.7 billion. These results are dependent on the actual prevalence of CFS/ME in the UK and the methodology used to calculate the total figure.

The findings of this study are slightly lower (proportionally) than those of recent studies of costs of CFS/ME to the US economy. Jason et al (2008), using CFS/ME prevalence data of 0.42%, estimated total direct and indirect costs between \$18.7 billion and \$24 billion for the USA as a whole. Reynolds et al (2004), using a point prevalence of 235 per 100,000 (0.24% prevalence), calculated the annual total value of lost productivity alone at \$9.1 billion, representing about \$20,000 per person with CFS/ME. Lin et al., (2011) estimated productivity and health care costs in the state of Georgia alone at US\$1.6 billion, though based on a much higher prevalence estimate of 2.5%.

Our study reports the total cost of CFS/ME to the UK population as a whole, including both children and adults above the working age. The weighted analysis

associates higher costs with informal care in comparison with productivity losses, apparently in disagreement with four out of five study findings. However, whereas informal care can be associated with CFS/ME sufferers of all ages, work-place productivity losses cannot.

Informal care and health care services can be considered substitute goods, in that a decrease in the supply of one may result in an increase in the other (McCrone, 2009). Additionally, professional care may have been sought more often by people with no informal care support. Whether care was provided to meet their needs is unknown.¹⁷

In the five papers, there was no information on the cost of health care services for children with CFS/ME. It is possible that we have overestimated the cost of primary and secondary care services for children given the lower prevalence among 0–18s as a whole.

Conversely, this analysis has probably underestimated some key costs. Productivity losses among carers themselves, through reduced hours in employment (Collin et al., 2011), were not captured. Further, applying the CFS/ME prevalence estimate of 0.4% equally across all age groups has likely resulted in an underestimate of productivity losses among the working-age population, since above-average prevalence would be expected among this group, given that the peak age of onset of CFS/ME is between 20 and 40 years (Capelli et al., 2010).

Another important consideration is that all studies included in this analysis captured productivity costs associated with absenteeism only; i.e. productivity losses from sick leave and discontinuation of employment. What is not measured is presenteeism – reduced productivity at work and increased likelihood of errors made through affected judgement, due to an illness. The absence of presenteeism data means productivity losses are likely to be significantly underestimated.

Strengths and limitations

As far as we are aware, this is one of the first studies to estimate the costs of CFS/ME to the UK economy.

One of the strengths of this study is that a number of sources of information have been used to corroborate findings. The only assumption that has a significant impact on the data is the prevalence of CFS/ME in the UK, although we based our analysis on conservative estimates derived through clinical assessments, not on self-reported prevalence.

This analysis has a number of limitations. The papers chosen were based on a rapid review rather than a full systematic review. As a result papers published in smaller journals may not have been included.

Across all studies the reporting of productivity costs was limited by the methodology used. All studies used the human capital approach and recognised its inherent limitations, although only one study adjusted for the potential over reporting of costs associated with productivity losses. We however made a similar adjustment for all studies in our analysis.

The data also comes from a number of different types of studies and populations, with slightly different methods used for analysis. In particular, reporting of health care resources differed between studies in a way that made synthesis challenging. This may have resulted in some double counting and errors in calculating costs. That the total health care cost was broadly similar across the four studies would suggest this did not have a significant impact on the results. There were also significant differences in the values reported for informal care, in particular McCrone et al. (2003), and productivity losses (Collin et al. 2011). We therefore needed to exercise caution in applying these data across methodologies so as not to inflate results disproportionately. For this reason, the findings of the weighted analysis are potentially the more robust.

The results from studies with follow-up of less than 12 months were extrapolated well beyond duration of their follow-up. It is not clear that people have

consistent costs over the full 12 months, even though the median length of illness in adults is around six years.

Conclusion

The direct and indirect costs associated with CFS/ME amount to at least £3.3 billion per year in the UK, a significant amount particularly given the relatively low prevalence of the condition. The economic impact of the condition has important implications in terms of investment planning in CFS/ME services and also UK research funding.

The UK health services spend approximately £542 million on people with CFS/ME. Drawing on matched sample findings by Lin et al. (2011), this amounts to well over £300 million more than a 'non-fatigued' population. Based on freedom of information requests responses obtained in 2016,¹⁸ we estimate that UK public spending on specialised CFS/ME services is currently in the region of £14 million per annum, just 3% of the £542m total.

CFS/ME expertise should not be thought of as exclusive to specialised services. Probable, however, is that many – and perhaps the majority – of local health services are spending considerable sums of money on non-specialised CFS/ME care.

More research is required to understand the true value of specialised CFS/ME services as compared with 'treatment as usual' by non-specialists, as well as the medium to long-term cost benefits of such services. However, NICE has made clear that around half of all people with CFS/ME 'need input from specialist services' (NICE, 2007), such are the complexities and highly disabling effects of the condition. Moreover, we would not expect sufferers of (for example) MS, diabetes or heart failure to be advised, supported and treated by non-specialists. We should expect no less for people with CFS/ME.

Further research, and therefore greater research funding, is also needed to move forward. This is a responsibility of government, which through the DWP

pays out well in excess of £100 million in ESA and DLA payments to people with a primary disabling condition of CFS/ME;¹⁹ productivity losses impact business and government revenues many times more.

Both NHS commissioners and central government need to reconsider funding decisions and organise CFS/ME services and research as appropriate to a treatable condition that has far-reaching societal and economic implications for the UK.

CFS/ME in numbers

- 260,000 The number of CFS/ME sufferers in UK
- 81% Proportion of carers who are husbands, wives or partners (Nacul et al., 2011)
- 16% Proportion of carers who are parents or children (Nacul et al., 2011)
- 85% Proportion of CFS/ME sufferers who have experienced some form of lost employment due to the illness (PACE trial, 2012)
- 20 – 40 The peak age of onset of CFS/ME (Capelli et al., 2010)
- 76% Proportion of CFS/ME sufferers who are female (Collin, 2011)
- 1:50 Possible prevalence of CFS/ME among 16 year-olds in the UK (Collin et al., 2016)
- £14 million UK spend on dedicated, specialised CFS/ME services
- £542 million Total UK health service spend on CFS/ME
- £3.3 billion Annual cost of CFS/ME to the UK

APPENDIX A:

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APPENDIX B:

Chronic fatigue syndrome case definition; CDC, 1994

Centre for Disease Control and Prevention: CFS case definition, 1994

Chronic fatigue syndrome (CFS) is a debilitating and complex disorder characterised by intense fatigue that is not improved by bed rest and that may be worsened by physical activity or mental exertion. People with CFS often function at a substantially lower level of activity than they were capable of before they became ill. The cause or causes of CFS have not been identified, and no specific diagnostic tests are available. Therefore, a CFS diagnosis requires three criteria:

1. The individual has had severe chronic fatigue for six or more consecutive months that is not due to ongoing exertion or other medical conditions associated with fatigue (these other conditions need to be ruled out by a doctor after diagnostic tests have been conducted)
2. The fatigue significantly interferes with daily activities and work
3. The individual concurrently has four or more of the following eight symptoms:
 - post-exertion malaise lasting more than 24 hours
 - unrefreshing sleep
 - significant impairment of short-term memory or concentration
 - muscle pain
 - pain in the joints without swelling or redness
 - headaches of a new type, pattern, or severity
 - tender lymph nodes in the neck or armpit
 - a sore throat that is frequent or recurring

These symptoms should have persisted or recurred during 6 or more consecutive months of illness and they cannot have first appeared before the fatigue.

APPENDIX C:

Steering group members

| Name | Role/Organisation |
|------------------------|--|
| Dr Dennis Cox | GP and Medico-Legal Specialist |
| Lisa Rodrigues CBE | Writer, Coach and Mental Health Campaigner |
| Dr Alastair Santhouse | Consultant in Psychological Medicine, South London and Maudsley NHS Foundation Trust, London |
| Dr Anna-Maria Volkmann | Health Psychologist and Medical Anthropologist, UCL |
| David Butcher | Chairman, The Optimum Health Clinic Foundation |
| Alex Howard | Founder and CEO, The Optimum Health Clinic |
| Matt James | Assistant Director, 2020health |
| Julia Manning | Founding Director, 2020health |
| Jon Paxman | Senior Researcher, 2020health |

The participation, expertise and advice of the steering group were greatly valued.

The findings and conclusions of this report are many and should not be understood as necessarily reflecting the professional opinion of all steering group members, outside of 2020health.

APPENDIX D:

Endnotes

1 Information, mainly via Freedom of Information Requests, obtained from 54 (of 56 known) specialist CFS/ME services, run by trusts, health boards and community interest companies, throughout the UK. 53 services returned referral activity data, 35 of those including information on running costs. Average costs extrapolated according to estimated numbers referred across all 56 services. (2020health, 2016.)

2 We found 51 services operating in England during the period 2013–15. According to Collin, S. et al., 2012, there were 49 in operation between 2008–10.

3 Further studies on prevalence have been undertaken since NICE produced its estimates in 2007. A meta-analysis by Johnston et al. (2013), examining seven studies using clinical assessment, found adult prevalence at 0.76%.

4 'SEID' was proposed by the US Institute of Medicine (IOM) in 2015, partly due to a concern that the 'CFS' label was liable to trivialise the seriousness of the condition and promote misunderstanding of the illness. The IOM also deemed the term myalgic encephalomyelitis as inappropriate due to 'a lack of evidence for encephalomyelitis (brain inflammation) in patients with this disease', and because myalgia (muscle pain) is 'not a core symptom of the disease' (IOM, 2015). See: [http://www.thelancet.com/journals/lancet/article/PIIS0140-6736\(15\)60270-7/fulltext](http://www.thelancet.com/journals/lancet/article/PIIS0140-6736(15)60270-7/fulltext)

5 Barthel scoring questionnaire is a point-scoring system that assesses ability in the following areas: bowels, bladder, grooming, toilet use, feeding, transfer, mobility, dressing, stairs, bathing.

6 Survey participants were recruited through the Action for ME's InterAction magazine (for members), and more broadly through the charity's website and social media.

7 The authors acknowledge some differing opinion on CBT and GET in the treatment of CFS/ME. For example: Twisk & Arnoldus (2012); Shepherd / meassociation: 'Patient reaction' online article, 18/01/2016.

8 For example: <http://www.humber.nhs.uk/services/chronic-fatigue.htm>

9 2020health direct contact with trusts, 2016.

10 Referrals to specialised services collated via freedom of information requests, 2020health, 2016.

11 CICs are not obliged to respond to FOIs. Certain information was withheld by some.

12 This figure is based on average known costs across 35 specialised services in England. Source: 2020health freedom of information requests, 2016.

13 2020health FOI requests to Scottish Health Boards, 2016.

14 Total government research spend on CFS/ME is unknown because specific expenditure by one of the major funders of UK CFS/ME research – the National Institute for Health Research (NIHR) Clinical Research Network (CRN) – cannot be disaggregated from total CRN expenditure. See: <http://www.publications.parliament.uk/pa/cm201314/cmhansrd/cm140109/text/140109w0001.htm>

15 (UK) Department for Work and Pensions (Statistical services). FOI response. 2016-1978, 22 June 2016

16 (UK) Department for Work and Pensions Central Freedom of Information Team, FOI response 1404, 29 April, 2016.

17 By 'professional care' we mean health and/or social care. Since there was little evidence of professional social care contacts in the five studies, we contacted seventeen councils (at random) through freedom of information requests for data on social care provision to people with CFS/ME. Because mandatory recording of health conditions does not include CFS/ME, councils were unable to identify associated costs, even though some confirmed they were delivering support services to people with CFS/ME.

18 Estimate based on FOI request responses and feedback (March–July 2016) received from 53 of 56 (known) UK specialist CFS/ME services – run by trusts, health boards and Community Interest Companies (CICs) – of which 35 disclosed both costs and numbers referred.

19 Estimate based on DWP FOI responses (2020health, 2016) and findings by McCrone et al. (2012).