# The Perceived Impact of Healthcare Stigma and Marginalisation on Illness Burden: The Lived Experiences of People with Myalgic Encephalomyelitis / Chronic Fatigue Syndrome (ME/CFS).

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**Abstract**

Myalgic encephalomyelitis / chronic fatigue syndrome (ME/CFS) is a chronic, disabling yet medically contested condition widely associated with social and healthcare-related stigma and marginalisation. Although stigma and associated phenomena are broadly associated with increased illness burden, no known studies have explicitly explored healthcare-related stigma and marginalisation and impact on illness burden, as experienced by people with ME/CFS. This study aims to contribute to this research gap. Taking an idiographic approach, semi-structured interviews with five people with ME/CFS were conducted, transcribed verbatim and analysed using Interpretative Phenomenological Analysis (IPA). Three superordinate themes were identified: 'navigating the paradoxes of ME/CFS healthcare', 'lack of patient-centred ME/CFS healthcare' and 'ripple effect of stigma across multiple life domains'. For the purpose of this paper, the latter two themes are presented and discussed. Participants reported repeated instances of their illness narrative being dismissed by healthcare professionals in favour of a psychologised model of ME/CFS. Healthcare-related stigma was found to impact detrimentally (directly or indirectly) on physical, psychological, occupational, financial and social life domains. Findings are grounded in extant literature, implications for practice and policy are discussed and future directions are suggested. Future research should consider more socio-demographically diverse or targeted samples combined with an intersectional approach to stigma and marginalisation within the ME/CFS population. Additionally, research is needed focusing upon the experiences of people with severe/very severe and enduring ME/CFS.

**The Perceived Impact of Healthcare Stigma and Marginalisation on Illness Burden: The Lived Experiences of People with Myalgic Encephalomyelitis / Chronic Fatigue Syndrome (ME/CFS).**

## 1 Introduction

Myalgic encephalomyelitis / chronic fatigue syndrome (ME/CFS) is a chronic, disabling condition characterised by profound fatigue exacerbated by exertion, often accompanied by cognitive impairment, sleep disturbances, gastro-intestinal dysfunction, widespread pain and autonomic dysfunction (Carruthers & van de Sande, 2012; Goudsmit et al., 2009). An estimated 150,000 to 250,000 people in the UK have ME/CFS (Shepherd & Chaudhuri, 2019), and the illness is associated with substantial functional impairment across physical, occupational and social domains (Taylor, 2005). Up to 25% of people with ME/CFS are so severely affected that they are predominantly or entirely bedbound and unable to perform basic personal tasks (CFS/ME Working Group, 2002). Research suggests that people living with ME/CFS demonstrate lower health-related quality of life and/or greater functional impairment than people living with cancer, stroke, congestive heart failure, multiple sclerosis and end-stage renal disease (Komaroff et al., 1996; Falk Hvidberg et al., 2015; Schweitzer et al. 1995; Kingdon et al., 2018). A particular complexity of ME/CFS involves its contested status as a legitimate medical condition (Clarke & James, 2003), associated with aetiological uncertainties alongside lack of biomarkers (Fischer et al., 2014). Medical uncertainty is associated with stigmatisation and marginalisation of people with ME/CFS, both broadly within society, and more particularly within healthcare (Anderson et al., 2012).

Stigma has been conceptualised as a multi-faceted construct where “labelling, stereotyping, separation, status loss, and discrimination occur together in a power situation that allows them” (Link & Phelan, 2001, p.377). Separation of ingroup from outgroup engenders a process of social marginalisation and exclusion whereby stigmatised persons are typically relegated to the fringes of society in depriving them of rights and resources accorded to dominant social groups (Major & O’Brien, 2005). The role of power in such processes is pertinent, since it has been argued that stigma can only arise when a power differential exists (Link & Phelan, 2001). The practitioner-patient relationship within healthcare represents such a power differential (Kidd & Carel, 2017; Strickler, 2009).

Healthcare systems draw upon explanatory models of health and illness defining who is ill and who is not, whilst healthcare professionals (HCPs) are accorded authority to control access to legitimate illness identities and subsequent healthcare benefits, by virtue of their training, social position and assumed epistemic authority (Carel & Kidd, 2014; Illich, 1975). Stigma and marginalisation of patients in healthcare has been explicated through the theoretical framework of epistemic injustice (Kidd & Carel, 2017). Rooted within the work of Fricker (2007), epistemic injustice occurs when the capacity of an individual or group as a ‘knower’ is discredited, and can be further conceptualised as testimonial and hermeneutical injustice (Fricker, 2007). Testimonial injustice involves the discrediting of an individual’s experience and knowledge as a result of negative

stereotyping, usually predicated upon stereotyped minority group membership. Whilst testimonial injustice might be argued to operate on an individual (person-to-person) level, hermeneutical injustice can be understood as a structural phenomenon, arising when socially dominant explanatory (hermeneutical) models fail to capture the meaning-making of a minority group (Fricker, 2007). Epistemic injustice, like stigma, thus arises from a power differential. Although all patients may be subject to epistemic injustice (Carel & Kidd, 2014), it might apply even more to ME/CFS as a contested diagnosis (Stone, 2018). It has been argued that people with ME/CFS are subjected to both testimonial and hermeneutical injustice in healthcare, largely due to the inability of the biomedical model to fully conceptualise ME/CFS and the subsequent application a biopsychosocial model which attempts to fill the explanatory gap (Blease et al., 2017).

The biomedical model addresses biological factors in health and illness, privileging objectively diagnosable, anatomically distinct disease over indistinct multi-systemic conditions (Wade & Halligan, 2004). An increasing body of biomedical research demonstrates neuroimmune and energy metabolism abnormalities in ME/CFS (Nakatomi, et al., 2014; Maes & Twisk, 2010; Cortes Rivera et al., 2019). However, debates and confusion over nomenclature, a highly heterogeneous patient population and numerous case definitions (potentially delineating different patient sub-groups or distinct clinical entities) confound the research picture and hamper biomedical conceptualisation (Nacul et al., 2019; Jason et al., 2016). Tendencies towards dualistic thinking in healthcare may result in illnesses which do not fully fit the biomedical model being re-constructed as predominantly or wholly psychological (Horton-Salway, 2002), arguably an example of hermeneutical injustice. In the case of ME/CFS, lack of solid biomedical explanatory framework has given rise to the application of the biopsychosocial (BPS) model (Engel, 1977) in understanding and treating the illness.

The BPS model acknowledges psychological and social factors in health and illness alongside biological influences, and was originally proposed to address healthcare inequities deemed to arise from a reductionist, dualistic biomedical model (Engel, 1977/1992). In conceptualising and treating ME/CFS, the BPS model has been combined with cognitive-behavioural principles to suggest that biopsychosocial influences can be further categorised as predisposing, precipitating and perpetuating factors (Deary et al., 2007). However, biological factors within this model receive little attention (an infectious agent may or may not precipitate ME/CFS), whilst psychological factors are emphasised across predisposing, precipitating and perpetuating factors (Sharpe et al., 1997; Deary & Chalder, 2006). BPS proponents believe that ME/CFS is predominantly perpetuated by aberrant illness beliefs and fear of exacerbating symptoms, leading to activity avoidance (Wessely et al., 1989; Knoop et al., 2010); however, empirical evidence of fear-avoidance is lacking (Gallagher et al., 2005; Tack, 2019). Whilst people with ME/CFS tend to attribute their illness to biological causes, BPS-inspired literature frames such attributions as aberrant illness beliefs (Wessely, 1990/1997) and suggests that physical attributions may be associated with poorer treatment outcomes (Picariello et al., 2017). The BPS model of ME/CFS has thus been criticised for ignoring indications of biological pathology, psychologising the illness and discounting patient narratives, contributing to epistemic injustice and fuelling healthcare-related stigma and marginalisation (Geraghty & Blease, 2019; Geraghty & Esmail, 2016). Such criticisms are most strongly voiced with respect to cognitive behavioural therapy (CBT)

and graded exercise therapy (GET), BPS-inspired treatment interventions for ME/CFS that aim to challenge aberrant illness beliefs and encourage people with ME/CFS to increase activity despite potential symptom exacerbation (Geraghty & Blease, 2019; Bavinton et al., 2004). Whilst data from multiple ME/CFS patient surveys reveal that most respondents prefer pacing (MEA, 2010; MEA, 2015), a means of managing symptoms by balancing rest and activity within personal limitations (Jason et al., 2008), NICE guidelines currently recommend CBT and GET as primary interventions for ME/CFS (NICE, 2018), despite a questionable and controversial evidence base.

A number of randomised clinical trials (RCTs) and systematic reviews (Price et al., 2008; Larun et al., 2016) have suggested that CBT and GET are moderately effective in treating ME/CFS, thus informing and solidifying current NICE recommendations. These studies have been challenged on bias and methodological grounds (Laws, 2017; Vink & Vink-Niese, 2018). One such RCT, the PACE trial (White et al., 2011), attracted criticism when independent data re-analyses suggested that the level of effectiveness of CBT and GET was approximately two-thirds lower than originally reported (Wilshire et al., 2017). RCT data are also inconsistent with patient survey data suggesting that CBT brings about no benefit in the majority of respondents, whilst GET results in deterioration in the majority of respondents (MEA, 2010; MEA, 2015; Geraghty, Hann & Kurtev, 2019). Additionally, some (not all) survey respondents report experiencing these treatments as inappropriately psychologising, dismissive, coercive and distressing (MEA, 2015). Although patient surveys carry potential biases (Shepherd, 2017), this data demonstrates that generalisations of intervention effectiveness across the whole ME/CFS population can be inaccurate and harmful. Exclusion of patient narratives from the evidence-based practice (EBP) model (Sackett et al., 1996), contrary to the model's ethos (Spring, 2007), is arguably an example of healthcare-related testimonial injustice. Indeed, there have been calls for more patient-centred, phenomenologically-informed, approaches to ME/CFS and wider healthcare (Blease et al., 2017; Carel & Kidd, 2014). Above-mentioned survey findings suggest that stigma and marginalisation in healthcare practice may increase the overall illness burden of some people with ME/CFS, where burden is defined as the personal cost of the illness on a physical, psychological, social, economic and occupational level. Whilst patient experiences of stigma and marginalisation within healthcare are widely reported within ME/CFS literature, the potential impact on illness burden remains largely unexplored.

Experiences of stigma and marginalisation are widely documented in ME/CFS literature (Åsbring & Närvänen, 2002; Anderson et al., 2012). Dickson et al. (2007) explored delegitimising experiences of 14 people with ME/CFS both with respect to healthcare encounters and significant others, using interpretative phenomenological analysis (IPA). One theme, 'negotiating a diagnosis', details participants' perception that their moral character was questioned through being perceived by HCPs as malingering and seeking to avoid everyday responsibilities. Perceived HCP minimisation of concerns and coercion into taking psychotropic medication was also reported, despite participants challenging psychological illness attributions and the effectiveness of such medication. Finally, anger, frustration, distress and sense of delegitimisation were reported; one participant believed she would be treated very differently if she had a (biomedically) legitimised health condition. The authors conclude that experiences of delegitimisation are as great a burden for people with ME/CFS as the

illness itself (Dickson et al., 2007). Although the authors note that recruiting participants through an alternative therapy clinic might have biased the sample against conventional medicine, participants' motivations in seeking alternative treatments might further underline the marginalisation of people with ME/CFS from conventional healthcare (de Carvalho Leite et al., 2011). Additionally, research capturing HCPs' attitudes towards and perceptions of people with ME/CFS broadly supports such patient experiences (Chew-Graham et al., 2009).

Negative stereotyping of ME/CFS by HCPs is well documented within qualitative research: ME/CFS is variously framed as an inability to face existential challenges of life, a lack of work ethic and stoicism, and a burdensome, 'heartsinky' patient group (Chew-Graham et al., 2010; Raine et al., 2004). Additionally, Thomas and Smith (2005) conducted a survey of GPs' beliefs surrounding ME/CFS (*n*=45), finding that just under half of GP respondents did not believe that ME/CFS exists. Discourse analytical research has shown how some HCPs use the BPS model to construct ME/CFS as psychogenic or psychosomatic (Horton-Salway, 2002), thus justifying psychological treatment despite mounting biological indications. The high level of dissatisfaction reported by people with ME/CFS in healthcare, alongside a perceived lack of HCP clinical understanding and interpersonal skills (Gilje et al., 2008), is supported by research highlighting need for greater HCP education and training in ME/CFS (Chew-Graham et al., 2010; Stenhoff et al., 2015).

Limited ME/CFS literature demonstrates more positive patient experiences, associated with patients feeling respected and listened to, alongside HCP willingness to learn (Ong et al., 2005; Drachler et al., 2009). Patient survey data (MEA, 2015) suggest that improved HCP understanding of ME/CFS (notably as a physical illness), alongside a patient-centred approach, is associated with improved patient experiences. Mixed findings might be explained by individual differences across patients, HCPs and researchers. Patient symptom severity and absence/presence of psychological comorbidity may impact on effectiveness and acceptability of interventions: RCTs of CBT and/or GET tend to exclude severely affected patients, and may use case definitions which over-select patients with psychological comorbidities (White et al., 2011; Nacul et al., 2019). Further, research suggests that HCPs have more positive attitudes towards people with ME/CFS when they are known personally (Horton et al., 2010), arguably highlighting the role of prejudice in stigma and marginalisation (Link & Phelan, 2001). Researcher bias might also impact (Wilshire, 2017); studies detailing notably positive patient healthcare experiences are co-authored by proponents of the BPS model of ME/CFS (Picariello et al. 2017; Broughton et al., 2017). Finally, differing methodologies and research questions may produce different findings. Most studies, with the exception of Dickson et al. (2007), do not focus explicitly on negative healthcare experiences and do not use methodological approaches with strong idiographic emphasis (such as IPA). Given the heterogeneity of the ME/CFS population, this may result in over-generalisation of cross-case findings lacking nuances of individual experience (Smith, 2004). The overall research picture suggests largely negative healthcare encounters in this patient group, supported by those who experience more positive encounters describing themselves as 'fortunate' or 'lucky' (Gilje et al. 2008; Dickson et al., 2007) .

Whilst stigma and marginalisation within healthcare are well-documented within ME/CFS research, there are to the author's knowledge no primary qualitative studies focusing explicitly on the

potential impact of such experiences on illness burden. However, alongside previously noted patient survey data (MEA, 2010; MEA, 2015), there are suggestions within ME/CFS research that epistemic injustice, stigma and marginalisation may increase illness burden. People with ME/CFS report identity issues arising from lack of legitimised diagnosis, alongside loss of self-esteem, self-efficacy and feelings of hopelessness/helplessness associated with disconfirming healthcare encounters (Åsbring 2001; Larun & Malterud, 2007; Edwards et al., 2007). Participant report demonstrates how restrictions placed on access to medical investigations for new or increased symptoms, apparently associated with negative stereotyping of ME/CFS, can lead to physical harms (Gilje et al., 2008). Although limited, such research is consistent with BPS-inspired literature, which discourages HCPs from over-investigating physical health complaints with the suggestion that this is colluding with patients' aberrant illness beliefs (Stanley et al., 2002). Reported loss of patient trust in HCPs, arising from experience of stigma and marginalisation (Dickson et al., 2007) may be associated with patient withdrawal from healthcare (Åsbring & Närvänen, 2002), carrying health-related risks. Finally, the suggestion that stigma can increase illness burden might be explicated through a biological framework. In wider (non-ME/CFS) literature, experiences related to stigma and marginalisation (such as discrimination) have been associated with physical and mental ill-health via a hypothesised mechanism of allostatic load (Pascoe & Smart Richman, 2009; Quinn & Chaudoir, 2015). Allostasis refers to the adaptive (neuro-immuno-endocrine) mechanisms within the body that facilitate stability in the face of stressors, whilst allostatic load describes the under- or over-stimulation of such mechanisms, often relating to chronic stress and associated with disease (McEwen, 1998). Some research has found that people with ME/CFS have higher allostatic load relative to healthy controls (Maloney et al., 2006).

Clearly, the potential impact of stigma and marginalisation on the illness burden of people with ME/CFS is worthy of explicit and focused exploration. Although there is limited mention of social factors in the BPS conceptualisation of ME/CFS, primarily the influence of benefits and social support (Bentall et al., 2002; Band et al., 2014), the potential role of stigma as a social factor is rarely mentioned (Van Houdenhove & Luyten, 2008). Given the above-mentioned lack of patient input into EBP and calls for a more phenomenological approach to healthcare, an approach which focuses on the lived experiences of people with ME/CFS is indicated. This study therefore aimed to explore lived experiences of stigma and marginalisation in healthcare, and perceived impact on illness burden, in a sample of people with ME/CFS. Additionally, the study aimed to explore what factors might contribute to improved healthcare, and potentially lessen illness burden. The research questions were as follows:

1. What are the experiences of people with ME/CFS regarding stigma and marginalisation in primary and secondary NHS healthcare?

2. What impact have experiences of stigma and marginalisation in healthcare had on the illness burden of people with ME/CFS?

3. From the perspective of people with ME/CFS, how might healthcare be improved?

# 2 Method

## *2.1 Methodology*

A qualitative approach was deemed necessary and appropriate to address research questions exploring participant experiences and meaning-making (Smith, 2015). Interpretative phenomenological analysis (IPA) was used both as methodology and analytical method. Phenomenology, IPA's primary epistemological position (Smith, 2004), concerns how individuals experience phenomena; working primarily from the participant's frame of reference (inductively) was considered appropriate for a patient group subject to epistemic injustice. IPA's dual focus on idiographic and collective narrative, alongside ability to elucidate complex, multi-faceted constructs (Larkin et al., 2006), was considered appropriate to explore experiences of stigma. The interpretative aspect of IPA acknowledges that individual capacity to 'know' is dependent upon the sociocultural context of the knower: a double hermeneutic is created as the researcher seeks to make sense of the participant's meaning-making (Smith & Osborn, 2008). The impact of the researcher's self upon the research is thus underlined, highlighting a need for reflexivity throughout the research process (Shaw, 2010).

## *2.2 Reflexivity*

As a person with ME/CFS and a therapist who has worked with people with ME/CFS, I acknowledge that my background has indubitably impacted upon the research process. Personal and professional experience of healthcare stigma and marginalisation within this clinical population has played a prominent role in informing research questions. Notably, I do not subscribe to a psychological model of ME/CFS. As an advocate of social justice with an interest in healthcare ethics, I believe that current ME/CFS healthcare raises concerns in these respects. A critical psychology perspective with an explicit focus upon stigma and marginalisation, inevitably encompassing more negative healthcare experiences, was deemed necessary to illuminate and examine these concerns (Teo, 2015).

## *2.3 Participants*

A purposive sample of five participants (see table 1) was recruited via the ME Association, a UK-based ME/CFS patient organisation (see section 2.4). IPA can only generate rich and nuanced data at both an idiographic and collective level with small samples (Smith, 2004) and a sample size of five is justified from a number of perspectives (Smith et al., 2009; Pietkiewicz & Smith, 2010).

**Table 1**

*Participant Demographic Data*

| **Participant** (pseudonym) | **Age** | **Ethnicity** | **Gender** | **Education level** | **Employment status** | **Years since diagnosis*** (reported in January 2020) | **Severity of ME/CFS** (self-rated)* |
|---|---|---|---|---|---|---|---|
| Ivor | 67 | white | man | Bachelor's degree | Retired on ill-health grounds | 17 years | moderate |
| Marie | 33 | white | woman | A-levels | Unable to work due to ill-health | 2 years 3 months | moderate |
| Elizabeth | 48 | white | woman | Bachelor's degree (Working towards a PhD when developed ME/CFS) | Unable to work due to ill-health | 20 years (Note: 24 years since symptom onset) | very severe |
| Jane | 59 | white | woman | HND | Unable to work due to ill-health | 6 years 5 months (Also diagnosed with post-viral fatigue syndrome in 2001) | moderate |
| Art | 51 | white | man | Master's degree | Unable to work / retired on ill-health grounds | 3 years 9 months | moderate-severe |

***** Period between symptom onset and diagnosis ranged from months to years; exact data was not collected

* Severity assessed using the ME Association Disability Rating Scale (MEA, 2016)

Mean age = 52 years  Mean years since diagnosis = 9 years 11 months

Inclusion criteria comprised adults (18 years +), fluent in English, with a self-reported medical diagnosis of ME/CFS, for whom ME/CFS was the primary health issue. Exclusion criteria included diagnosed psychological or psychiatric comorbidities and vulnerable adults, with a view to protecting participants and reducing biases arising from differential diagnosis or misdiagnosis (Brurberg et al., 2014). A comorbid diagnosis of fibromyalgia also excluded participants from the study; although high rates of comorbidity are reported between fibromyalgia and ME/CFS (Jackson & MacLeod, 2017),

levels of healthcare-related stigma and marginalisation across the two patient groups may differ (Åsbring & Närvänen, 2003).

### *2.4 Procedure*

Participants were recruited through the ME Association, after gaining permission to advertise the study on the organisation's website and social media. Interested parties responding to the advertisement were sent further study information alongside a consent form (detailing confidentiality and data protection provisions) and a demographics questionnaire to complete prior to interview (appendices 1, 2, 3 & 4). Interviews were conducted as described in section 2.5. Given that cognitive dysfunction is a common component of ME/CFS, participants were also emailed a broad idea of interview questions in advance of interview (appendix 7). Post-interview debrief (appendix 6) provided details of information and support services, with a view to participant protection (BPS, 2009/2014). Interviews were digitally recorded and, given IPA's foregrounding of the meaning of linguistic encounters rather than linguistic intricacies, interviews were transcribed verbatim, a denaturalised approach (Oliver et al., 2005). The study gained ethical approval from the University of Derby.

### *2.5 Data Collection*

Interviews were conducted via Skype (*n*=3), telephone (*n*=1) and face-to-face (*n*=1) according to participant location and preference. Participants were given opportunity to complete the interview over more than one meeting, take breaks during interview and encouraged to stop when necessary, according to their health needs. Four participants each had one interview, lasting an average of 70 minutes. One participant (Elizabeth) had two interviews with combined length of 109 minutes. Consistent with IPA's underpinning of phenomenology and co-construction of knowledge, semi-structured interviews were chosen for their combination of structure and flexibility (Howitt, 2016; Willig, 2013); participants were encouraged to lead whilst the researcher had opportunity to pursue salient points. Each interview was thus uniquely shaped by researcher questions and participant responses. The topic guide (appendix 5), broadly informed through literature, served as a broad framework (Brocki & Wearden, 2006); although continuously re-evaluated in light of previous interviews (Smith & Osborn, 2008) no amendments were considered necessary. Data saturation (Saunders et al., 2018) was deemed inconsistent with IPA's iterative approach; the scope of data collection was guided by balancing analytical depth and breadth within time constraints (Smith et al., 2009).

### *2.6 Data Analysis*

Analysis proceeded via steps outlined in Smith and Osborn (2008), broadly delineated as (1) looking for themes, (2) connecting themes and (3) continuing the analysis with other cases.

(1) The first transcript was read several times before proceeding to annotation (associations, questions, summaries, preliminary interpretations) in the left-hand margin. *Example: no explanation of illness, no answers, having to 'fit a script'.* The right-hand margin was used to develop emergent themes, moving from annotation to a higher level of abstraction, making theoretical connections, whilst ensuring themes were grounded in raw data. *Example: need for individually-tailored healthcare.*

(2) Connections were sought between emergent themes, resulting in clustering and collapsing of themes alongside development of superordinate concepts. *Example: lack of patient-centred care.* A table of themes was produced, listing superordinate themes and subordinate themes, data extracts supporting these themes and location of extracts in the transcript.

(3) In continuing the analysis, each transcript was taken afresh with an attempt to bracket off previous analytical insights (Smith & Osborn, 2015) and separate tables of themes were produced for each case. A final table of superordinate and subordinate themes was then constructed, accounting for convergences and divergences across and within cases. For example, divergences in experiences of stigma expressed by different participant themes were synthesised under a higher-level theme sufficiently broad to account for divergences ('Ripple effect of stigma across multiple life domains'). Triangulation, with a view to enriching understanding rather than seeking convergence, was provided through the research supervisors who reviewed transcripts and tables of themes, ensuring that themes were grounded in the raw data.

### *2.7 Study Quality*

Whilst the concepts of validity, reliability and statistical generalisability are used to assess the quality of quantitative research, the concept of trustworthiness has been deemed more appropriate for qualitative studies (Shenton, 2004). Trustworthiness has been defined as consisting of credibility, confirmability, transferability and dependability (Lincoln & Guba, 1985). Credibility, the level of congruence between research findings and reality, was increased through supervisory analyst triangulation, with a view to enriching understanding as opposed to seeking convergence (Willig, 2013). Ensuring analysis was grounded in raw data (via continual referral to participant quotes and context) increased credibility and also confirmability, the degree to which findings are shaped by participant data as opposed to researcher bias. Potential researcher biases were also countered through reflective journaling, supervision and audit trail (Shenton, 2004; see section 4.3). Case-to-case transferability, the extent to which findings are applicable to other contexts, was addressed through thick description of the phenomena under investigation and broader research process. Dependability, the extent to which a different researcher could repeat the study, was addressed through detailed methodological description.

## 3 Findings

Three superordinate themes were identified: 'navigating the paradoxes of ME/CFS healthcare', 'lack of patient-centred ME/CFS healthcare' and 'ripple effect of stigma across multiple life domains' (see table 2). For the purpose of this paper, the latter two themes and accompanying sub-themes are presented and discussed.

**Table 2**

*Master Table of Themes*

| Superordinate Theme<br>*Description* | Subordinate Theme<br>*Description* |
|---|---|
| 1 Navigating the paradoxes of ME/CFS healthcare<br><br>*There were repeated instances of participants experiencing healthcare as running counter to the expected ethos. Participants drew on self and others in filling the gap in HCP knowledge of ME/CFS and in healthcare provision, whilst navigating healthcare interventions that were generally experienced as not fit for purpose.* | 1.1 Onus of managing ME/CFS falls to the patient and others<br><br>*There were recurring examples of participants drawing on self and others outside of primary and secondary care to manage their illness. Where this was not possible, greater marginalisation and dissatisfaction with healthcare resulted.* |
| | 1.2 Limited HCP knowledge and patient as educator<br><br>*There were recurring accounts of lack of HCP insight into the lived experience of ME/CFS and lack of HCP knowledge of ME/CFS as a biomedical entity. Such accounts were associated with some participants seeking to educate HCPs about their illness.* |
| | 1.3 Lack of fit-for-purpose, needs-based healthcare<br><br>*There were recurring descriptions of healthcare interventions being unfit for the needs of people with ME/CFS, particularly those at the severe end of the spectrum.* |
| 2 Lack of patient-centred ME/CFS healthcare<br><br>*There were repeated examples of HCPs dismissing patient testimony. Healthcare models and HCP stereotyping were experienced as being prioritized over patients, whilst lack of HCP-patient care partnership and suggestion of political agendas were associated with loss of patient trust. In rarer instances where the patient was foregrounded, experience of healthcare was more positive and HCP-patient relationships appeared improved.* | 2.1 Struggling to be heard: Healthcare models and HCP stereotyping before patients<br><br>*There were repeated accounts of HCPs putting psychologically informed healthcare models and negative stereotypes before the patient, associated with a dismissal or misrepresentation of patient narratives.* |

| | | |
|---|---|---|
| | | 2.2 Lack of patient-HCP care partnership<br><br>*There were repeated participant accounts of lack of care partnership, in particular lack of involvement in discussions over treatment rationale and potential risks, with some suggestion of participants feeling pressured into treatments and blamed for not improving.* |
| | | 2.3 Healthcare hierarchies, politics and loss of patient trust<br><br>*There was suggestion across participants of the relative authority of HCPs compared to patients, which for some participants was associated with a loss of trust when combined with negative healthcare experiences. For one participant, loss of trust was also associated with a strong theme of perceived underlying political agenda taking precedence over patients.* |
| 3 Ripple effect of stigma across multiple life domains<br><br>*There were recurrent examples of stigma and increasing marginalisation impacting negatively upon participants across physical, psychological, occupational, social and financial domains. Much of this impact was associated with inadequate or stigmatising healthcare practice, policy, provision and discourse.* | | 3.1 Impact of healthcare-related stigma on other social determinants of health<br><br>*Stigma associated with healthcare practice, policy, provision and discourse was shown to impact negatively on factors recognized as social determinants of health (outside of healthcare provision).These included social support levels, economic stability, meaningful employment, housing and education.* |
| | | 3.2 Healthcare-related psychological distress and physical harms<br><br>*HCP disbelief and dismissal of patient narratives, alongside participants' physical deterioration from healthcare interventions, were shown to have a negative psychological impact across most participants. Stigma-related psychological distress appeared to increase physical illness burden for some participants.* |

### *3.1 Lack of Patient-Centred ME/CFS Healthcare (Theme 2)*

There were repeated examples of HCPs dismissing patient testimony. Healthcare models and HCP stereotyping were experienced as being prioritized over patients, whilst lack of HCP-patient care partnership and suggestion of political agendas were associated with loss of patient trust. In rarer instances where the patient was foregrounded, experience of healthcare was more positive and HCP-patient relationships appeared improved.

#### 3.1.1 Struggling to be Heard: Healthcare Models and HCP Stereotyping Before Patients (Sub-theme 2.1)

All participants experienced their understanding of ME/CFS being dismissed by HCPs, though the pervasiveness of such experiences varied. The most frequently described experience was one of ME/CFS being conceptualised by HCPs through a psychological or psychiatric lens. Participants communicated a sense of knowing their bodies and rejected such conceptualisations:

> "I tried to get a diagnosis through my GP and they just didn't believe it – it was the yuppie flu thing. I spent several years going to different GPs in the practice trying to get different opinions… but they were all much the same…you're depressed and you're suffering from anxiety…It wasn't anxiety, I wanted to know what was wrong with me." (Ivor, 55-60)

The 'yuppie flu thing' represents an inaccurate, psychologised perspective of ME/CFS prevalent within society at the time of Ivor's diagnosis. The 'several years' spent in search of legitimisation, a period during which Ivor reported being 'fobbed off' (112) by anti-depressants, suggests that repeated dismissal of Ivor's narrative delayed his diagnosis. Although the term 'yuppie flu' has lost social prevalence, all participants perceived the label of CFS as misrepresenting and minimising their illness experience:

> "I believe I would be taken more seriously by GPs etc if I did not have a CFS diagnosis. I can be so ill sometimes, extreme difficulty breathing, chest pains, tremors, mini-blackouts, falling over etc and they do not entertain the thought of referring me to a specialist. Someone else presenting with these symptoms would be given some examination at least." (Elizabeth, 1030-1035)

In contrast to Elizabeth's lived experience of ME/CFS, predominated by extreme, debilitating physical symptoms, Elizabeth recounted numerous psychologising encounters in both primary and secondary (ME/CFS specialist) care. Beyond repeated recommendations of anti-depressants, Elizabeth noted how HCPs questioned her moral character: "you're attention-seeking, you're dramatic, you're just making it up, you're a liar, you're a malingerer – I've been called all of these things" (Elizabeth, 109-111). Elizabeth's experience is one where ME/CFS is conflated with a mental health condition, which in turn is misrepresented as signalling personal moral failure ('you're a liar, you're a malingerer'). The use of third person ('you're a liar…') may suggest linguistic and psychological distancing from a construction of self that Elizabeth does not share. Whilst it is arguably easier to dismiss patient testimony when the patient can be framed as lacking a moral compass, some participants raised the potential of cognitive dysfunction, characteristic of ME/CFS, to undermine patient credibility in healthcare encounters. Art discussed how he planned for appointments days in advance to manage this and described how this aspect of ME/CFS made him feel "such an idiot" (388). The generally reported struggle to be heard in healthcare encounters seemed to be compacted for some

participants by the complexity of ME/CFS: “it’s a very difficult condition to explain simply and quickly” (Jane, 90-91). However, in contrast to repeated accounts of disconfirming encounters, there were rarer examples of patient narrative being valued by HCPs. Art, who noted how he had almost always felt believed (though not necessarily understood) by HCPs, described himself as “fortunate” (170) in experiencing healthcare as primarily positive. One instance of Ivor’s healthcare experiences (of a specialist ME/CFS group management course) stood out amongst all participant narratives:

> “She [ME/CFS clinic HCP] came along to one of the sessions and she said ‘I’m going to tell you how I think you can cope, but this isn’t gospel, it’s all sorts of emerging ideas …please give me your feedback and if you have sort of negative feedback I’d love to hear that, positive feedback I’d love to hear it’. You know, she was willing to learn and that was brilliant.” (Ivor, 240-245)

Ivor’s perception of being involved in his healthcare as ‘brilliant’ is supported by his account of feeling “positive” (252) and “confident” (249) as a result. There is an underlying suggestion here of the importance of care partnership to Ivor; care partnership is explored in the following theme.

#### 3.1.2 Lack of Patient-HCP Care Partnership (Sub-theme 2.2)

All participants, with the exception of Art, described a general lack of being involved in their care, in particular vis-à-vis discussions over treatment rationale and associated potential harms. There was uncertainty across most participants as to whether treatments had involved CBT and/or GET elements or principles alongside confusion over terminology (for example, GET versus activity management). Marie recounted how her repeated questions to specialist clinic HCPs were left unanswered: “if you don’t fit their script, you’re stuck” (Marie, 780-781). There was suggestion that treatment benefits were oversold whilst side-effects were downplayed, alongside an acknowledged need for reporting of harm protocols for BPS interventions. Both Ivor and Elizabeth found that GET was framed by HCPs as a ‘cure’, a claim that stood in stark contrast to their experience:

> “Graded exercise therapy was something the physiotherapist was insistent on doing and I tried it and it set me back at least two years in my coping. I was just absolutely washed out, I couldn’t work - I was off work for several months”. (Ivor, 276-279)

The ‘insistence’ of the physiotherapist suggests that Ivor felt somewhat coerced into trying GET, with the result that his health greatly deteriorated. Whilst Jane felt able to tell her HCP that a graded activity approach was not working for her, she noted how other patients might have felt “pressured” (173) to follow HCP advice. Although Art did not echo this, he commented on being ‘determined to stick out’ clinic advice (395), even though he noted a cost in terms of increased symptoms. Ivor’s experience of GET is mirrored and amplified by Elizabeth’s account:

> “I got no explanation [of treatment rationale] it was a case of ‘I want objective measures that you’re improving – you have to do this or the course ends’. And I told him this is going to make me really ill […] I was actually having seizures it was that bad – the relapse from that.” (Elizabeth 158-163)

It is noteworthy that both Elizabeth and Ivor recounted being positioned by HCPs as uncompliant with treatment (unmotivated or obstructive) and felt blamed for their failure to improve. The narrative of blame

was particularly strong in Elizabeth's case, something she associated with an underlying treatment philosophy: "with physiotherapy you go into their department and on the wall it says 'you get out what you put in'. So that implies it's your fault it didn't work" (Elizabeth, 122-124). In this respect it is interesting that Art appeared to blame himself for struggling to find a baseline for graded activity: "I think I've proved so many times that I'm not very good at this yet – I keep crashing" (Art, 305-306). Art also raised the question of whether socio-demographic factors might impact on quality of healthcare encounter:

> "For the consultant, I can only imagine what it would be like for somebody much younger than me, who didn't perhaps have that background [Art's profession], who didn't have the language skills, or the education, or whose first language wasn't English … I wonder what they would have got from those conversations." (Art, 216-221)

The importance of care partnership was explicitly or implicitly present in all participants' accounts, underlined by Marie's response to being asked how healthcare could be improved:

> "Just with the ME service being able to ask questions, to get answers, even if it's let me look at that, think about that and come back with a plan rather than wait and see if it comes up." (Marie, 774-776)

There is perhaps some suggestion here of a wish for greater HCP transparency or congruence vis-à-vis limitations of HCP knowledge ('let me look at that, think about that'). Participant perspectives on transparency and congruence within healthcare are touched upon in the following theme.

#### 3.1.3 Healthcare Hierarchies, Politics and Loss of Patient Trust (Sub-theme 2.3)

All participants alluded to the relative social and epistemic authority of HCPs compared to patients, although participants varied as to how they felt this impacted on their healthcare experiences. Epistemic hierarchy was for some participants marked by patient testimony being treated as a less authoritative form of knowledge compared to medical 'evidence' or HCP opinion. Such hierarchy appeared to be associated for some participants with the inability to 'prove' the seriousness of ME/CFS due to lack of diagnostic biomarkers. Whilst Jane mentioned structures potentially "too big for us to be able to influence" (577), Elizabeth's narrative was pervaded with perception of HCP power and overarching political agenda prioritised over patients:

> "The objective is to increase the length of a window of time in which we are functional so that we can go back to work... and providing CBT/GET therapies which are claimed to have a high improvement rate so it's the patient's fault if it doesn't work. It appears the PACE trial statistical analysis was geared to produce these results." (Elizabeth, 1003-1008) … "It's all utterly corrupt, agenda-driven rubbish." (Elizabeth, 856)

Elizabeth's experience of healthcare as 'utterly corrupt, agenda-driven rubbish' stands in stark contrast to expected healthcare ethics and ethos. Such contrast appeared to be associated with Elizabeth's expressed loss of trust in HCPs and the healthcare system, a sentiment shared by most participants. Some participants expressed reluctance to engage with healthcare services unless in an absolute emergency: "unless I'm dying I just won't go now because I know I'll be told it's just the ME" (Marie, 704-705). Marie's loss of trust was compacted by her complaint of new symptoms being ascribed by her GP to ME/CFS, a dynamic reported by

most participants. Although Marie challenged this, her symptoms went without further investigation and she was later hospitalised with a non-ME/CFS medical issue. Elizabeth linked her loss of trust in part to incongruence between HCPs' words and actions: "They go through the first thing of 'oh we don't think it's a mental problem' and then pretty much behave towards you as though it is" (Elizabeth, 905-907). It is noteworthy that Elizabeth raised a formal complaint about her healthcare which was treated with less credibility than the perspective of the HCP concerned.

### *3.2 Ripple Effect of Stigma Across Multiple Life Domains (Theme 3)*

There were recurrent examples of stigma and increasing marginalisation impacting negatively upon participants across physical, psychological, occupational, social and financial domains. Much of this impact was associated with inadequate or stigmatising healthcare practice, policy, provision and discourse, particularly with regard to the influence of healthcare on welfare provision.

#### 3.2.1 Impact of Healthcare-Related Stigma on Other Social Determinants of Health (Sub-theme 3.1)

The potential of stigma and its sequalae to impact across multiple life domains was noted in the accounts of all participants, albeit to varying degrees. All participants reported loss or marginalisation: disruption or loss of employment and education, threatened loss of home, loss of expected family roles, loss of friendships and relationships. It is noteworthy that these areas can be conceptualised as social determinants of health (meaningful employment, education, housing, social support). It is equally notable that some of these losses were explicitly associated by participants with disbelief and dismissal in healthcare encounters, in turn associated with inadequate and inappropriate healthcare provision. Marie described how trying to manage the expectations of a specialist ME/CFS clinic programme increased her physical health burden, requiring shifting of family roles and increased need of a family carer: "her life gets affected as much as mine does" (Marie, 332). Marie also described how disbelief and lack of legitimacy in healthcare caused difficulties when navigating the benefits system, with a detrimental ripple effect on finances and housing. Such obstacles to securing welfare provision were reported by other participants and associated with healthcare-related disbelief:

> "I didn't feel believed and certainly the [DWP assessment] report that they wrote assumed so much – you can do this and therefore you can do it all the time. There was no thought of can you do it continuously or can you do it again tomorrow. And I ended up going to tribunal for that to come back. The whole DWP experience was quite traumatic with not being believed because of the medical condition." (Jane, 318-323)

Here, Jane highlights how lack of medical understanding of a fluctuating health condition perpetuated disbelief in benefits assessing, creating a traumatic struggle to secure financial support. Art appeared to be the least impacted; although he experienced economic stress, notably associated with an unsatisfactory DWP assessment and rejection of benefit claim, he eventually obtained ill-health retirement, reporting that this allowed him to focus on managing his health. However, Art indirectly underlined the potential detrimental influence of healthcare discourse on broader social attitudes towards ME/CFS:

> "Certainly my colleagues and even my closest friends see it [ME/CFS] as tiredness […] even when they experience like now I'm struggling to find words and I'm losing my voice a bit – they attribute that to tiredness. So an understanding that it isn't tiredness would be useful for healthcare." (Art, 487-493)

Art notes the lack of understanding of ME/CFS in society more broadly and then links this to a need for healthcare professionals to better understand the condition. Art thus appears to consider legitimisation of ME/CFS within a healthcare context as a way of countering negative and damaging attitudes in a broader social context. A similar dynamic of influence was highlighted by Ivor, who explained how his wife could not accept his illness, believing it "was all yuppie flu and it was all in the mind" (407-408). The impact of such disbelief on Ivor's life was great: "She [Ivor's wife] said she couldn't cope with me anymore – and we are currently going through a divorce" (427-428). Ivor's wife's view of ME/CFS as yuppie flu mirrors Ivor's experience of healthcare attitudes at the time of his diagnosis, as previously discussed, again suggesting that healthcare stigma can perpetuate broader social stigma. Accordingly, greater understanding of ME/CFS amongst HCPs was underlined by all participants as a factor that would improve healthcare (forming a core part of sub-theme 1.2). The potential ripple effect of healthcare-related stigma is summed up by Marie: "I understand how they [people with severe ME] can end up housebound and bedbound, because they've got nobody trying to help them" (514-516). The most extreme example comes from Elizabeth:

> "When you're disabled for a long time, and living on a very small amount of money, you get further and further … your life is eroded … further and further away from normal. I didn't have kids, I haven't been in a relationship for at least 15 years – it just adds to it. My life is so far removed from normal. It's very isolating." (Elizabeth, 687-691)

The potential of stigma and subsequent marginalisation to create a vicious cycle of ever-reducing life chances is powerfully summarised in 'your life is eroded'. Elizabeth had no social support network, no children or intimate relationship, was struggling financially, unable to work, and had been forced to give up her PhD (through ill-health) many years previously. Elizabeth attributed all these things to the impact of living with ME/CFS, combined with inadequate and discriminatory health (and social) care policy, practice and provision. Elizabeth commented that she approached her GP for advice on how to access social care (notably, in the absence of disability benefits) but was not given any assistance. It should be asked what kind of impact such losses might have upon psychological well-being; this is explored as part of the next theme.

#### 3.2.2 Healthcare-Related Psychological Distress and Physical Harms (Sub-theme 3.2)

Anger, fear, anxiety, low mood, frustration and feelings of vulnerability were all reported as a result of healthcare and associated experiences of benefits assessments, across all participants except Art. Experiences of being dismissed and misrepresented (as previously discussed) were for Ivor associated with feelings of unworthiness: "I just felt as if I wasn't being treated as a human being" (Ivor, 128-129). This was echoed by Jane's account of the impact of a DWP assessment of her sense of self:

> "You just feel as if you are not as worthy as somebody else … that you're being made out to be a scrounger. I have had several conversations with two or three very close friends afterwards, almost

> asking their opinion of what they think about it […] I was trying to see what they thought of me." (Jane, 339-343)

The use of 'just' in both Ivor's and Jane's accounts, along with Jane's use of third person 'you' ('you are not as worthy…') suggest a need to minimise or create psychological distance from the pain of feeling like a less worthy human being. Feelings of unworthiness are often associated with the social emotion of shame; although shame was not reported, both guilt and loss of confidence at not being able to contribute on an occupational level were reported by some participants. Jane's need to seek reassurance from friends suggests the power and influence of medical discourse and the extent to which Jane's perception of disbelief and dismissal caused her to doubt herself. Accordingly, "acceptance and belief" (725) was noted by Jane as something that could improve healthcare experiences. It is noteworthy that much of the anxiety and stress reported by participants was associated with (unprompted) accounts of negotiating the benefits system, with Jane noting how having to "fight my corner" (368) added to her overall illness burden. Jane also noted how benefits assessments seemed to be "geared towards the mental health side of things" (496), which she contrasted with her contextualised understanding of anxiety as a reasonable response to financial uncertainty. Marie used the word 'fight' eight times to describe her struggle to for adequate healthcare. It could be questioned how such fighting might impact upon a person with a physical illness exacerbated by exertion; it is thus interesting that Marie reported that stress and frustration (apparently associated with having to fight for a satisfactory level of care) seemed to exacerbate her physical symptoms. Elizabeth's account represents a powerful example of this dynamic: "I claimed something called PIP in 2016 and … dealing with that depth of corruption made me extremely ill and it took about two years to recover from" (Elizabeth, 661-663).

Further accounts of increased physical illness burden were associated with the effects of certain healthcare interventions. Marie and Ivor commented on how being prescribed what they deemed to be unsuitable medications carried negative physical side-effects, whilst Ivor and Elizabeth described anxiety and stress as a result of their physical deterioration associated with GET. Art, who reported the lowest levels of distress overall (and the highest levels of HCP belief), was in the early stages of getting to grips with specialist clinic guidance. The majority of participants commented on how accessibility issues around ME/CFS clinics, alongside suitability of environment, impacted negatively on their physical symptoms, with Art highlighting the irony of a healthcare event causing a deterioration in health. The most powerful account of harm came from Elizabeth who used the word 'abuse' three times to describe her experience of health and social care, commenting: "They are just trying to kill you off – that's what it feels like – trying to kill you off" (Elizabeth, 809-810).

## 4 Discussion

This study sought to explore the experiences of stigma and marginalisation in healthcare, and impact on illness burden, as experienced by people with ME/CFS, alongside factors which might contribute to improved healthcare. Participants described repeated instances of their testimony being dismissed by HCPs, notably in favour of a psychologised understanding of ME/CFS. To varying degrees, participants described losses, limitations and harms across physical, psychological, social, financial and occupational domains, some of which were explicitly associated with dismissive healthcare encounters and unsatisfactory

healthcare provision. Rarer participant reports of HCPs valuing patient testimony appeared to coincide with lower overall health burden, most clearly in the psychological domain. Such findings are supported by factors which participants felt could improve healthcare: HCP belief, acceptance, improved understanding of ME/CFS and a more collaborative care partnership. These findings are discussed through the lens of biopsychosocial theory and literature, stigma research, and the framework of epistemic injustice.

### *4.1 Grounding in Wider Literature*

Participant accounts of HCP psychologisation, dismissal and misrepresentation in this study are consistent with wider qualitative research (Chew-Graham et al., 2009; Raine et al., 2004) and may be associated with the empirically unvalidated privileging of the psychological pillar within the BPS model of ME/CFS (Geraghty et al., 2019). The lack of safeguards against privileging one BPS pillar over another has been criticised as a form of unregulated eclecticism which may facilitate biases of those who draw upon the model (Ghaemi, 2009); it is notable that the BPS model of ME/CFS has been developed and promoted primarily by those favouring a psychological or psychiatric understanding of illness (Deary et al., 2007). Misrepresentation (psychologisation) and negative stereotyping (framing patients as malingering, dramatic and uncooperative) on grounds of contested illness identity leads to HCP dismissal of patient testimony, a form of testimonial injustice (Fricker, 2007). Such dismissal is compacted by hermeneutical impoverishment whereby neither patients nor HCPs have adequate explanatory models to capture the ME/CFS illness experience. The concept of people with ME/CFS having to 'fit the script' suggests that experiences which cannot be accommodated by dominant explanatory frameworks are distorted or denied by HCPs. Given the social and epistemic power of HCPs (Fricker, 2007; Kidd & Carel, 2017), epistemic injustice in healthcare can be enacted in the form of discrimination, exemplified by the suggestion in this study and more broadly that people with ME/CFS feel they are treated differently to other patients because of their diagnosis (Dickson et al., 2007). Epistemic injustice is frequently bound up with socio-demographic disadvantage (Fricker, 2007) and it has been argued that there is conceptual overlap between epistemic injustice and stigma (Buchman et al., 2017). A particular example of epistemic injustice and stigma as illustrated in this study is that of blaming the patient.

ME/CFS patient survey data captures patient accounts of HCPs blaming patients for non-improvement (MEA, 2015); this is supported by research capturing HCP stereotyping of people with ME/CFS as lazy and lacking motivation (Stenhoff et al., 2015; Chew-Graham et al., 2009). Research has shown that the BPS model in ME/CFS can be used to shift health-related accountability from HCPs onto patients (Horton-Salway, 2002). Whilst this may be associated with HCP feelings of powerlessness, uncertainty and a resistance to traditional patient-HCP roles being challenged (Raine et al. 2004; Horton-Salway, 2002), this study indicates that blaming the patient may be associated with healthcare discourse, notably an overarching ideology within ME/CFS healthcare of 'you get out what you put in'. Such ideology is consonant with BPS literature which emphasises the importance of personal effort and motivation in overcoming catastrophising cognitions and fear-avoidance (Picariello et al., 2017). There are indications that such ideology and associated blame occur within healthcare more broadly (at least, in the case of patients with contested illness) and may arise from mainstream, neoliberal associations between productivity and success, alongside political and economic agendas, notably within the context of welfare reform (Shakespeare et al., 2017; Briant et al., 2013). Essentially, when patients fail to fit dominant assumptions and explanatory models, lack

of alternatives may lead to criticism and blame of patients (as unmotivated, obstructive or malingering) rather than a critique of existing hermeneutical frameworks. The psychological health burden of epistemic injustice and stigma, highlighted in this study, is supported more widely (Larun & Malterud, 2007; OxCATTS, 2019). Of particular note, the questioning or doubting of the self in response to persistent epistemic injustice has been considered through a lens of gaslighting (Berenstain, 2016). This study also supports wider research in suggesting that more positive experiences are associated with HCP belief, willingness to learn and better understanding of ME/CFS (Drachler et al., 2009). Consistent with stigma research, this study demonstrates that healthcare stigma can impact on multiple life domains beyond the psychological.

The study's findings demonstrate that stigmatising healthcare encounters can impact on domains that might be conceptualised as social determinants of health (SDH; CSDH, 2008). The detrimental effect of enacted stigma within the arena of one SDH (healthcare provision) upon other SDHs that might otherwise represent protective factors (meaningful employment, housing, education, social support, economic stability) indicates a potential ripple effect of stigma and resultant marginalisation (Link & Phelan, 2006; Friedman, 2020). This is supported by literature suggesting that stigma and systematic epistemic injustice can have pervasive influences across multiple life domains, reducing a person's life chances (Link & Phelan, 2001; Fricker, 2007). Some instances of ripple effect in this study are overt and occur at least partly at an individual level; the impact of healthcare-related stigma and discrimination on benefits assessing was particularly clear, supported by participant account in wider research suggesting that navigating the benefits system increased physical and psychological health burden (Drachler et al., 2009). However, this study also suggests that a less overt ripple effect may involve the impact of healthcare discourse, associated with structural factors such as policy and political frameworks, on broader negative social attitudes towards ME/CFS (Drachler et al., 2009; Briant et al., 2013). This serves to demonstrate that healthcare-related stigmatisation and marginalisation can occur in the absence of stigmatising practice on a HCP level (Link & Phelan, 2001). The potential physical harms of enacted epistemic injustice and stigma warrant particular attention.

Physical harms resulting from stigma and epistemic injustice are demonstrated within this study, and to some extent more widely, through HCP insistence upon inappropriate treatment despite patient challenges and through lack of treatment associated with dismissal of patient testimony (Gilje et al., 2008; MEA, 2015). This suggests both that EBP within ME/CFS healthcare insufficiently draws upon patient narrative, contrary to its purported ethos (Sackett et al., 1996), and that the principle of patient-centred healthcare (NICE, 2018; NHS, n.d.) does not routinely apply in the case of ME/CFS. Clinical coercion and lack of transparency over treatment rationale and potential harms (Dickson et al., 2007; Kindlon, 2011) also raise questions about informed consent (Geraghty & Blease, 2018). It has been argued that the imperatives of informed consent may be neglected in cases where patients are considered irrational and thus epistemically unreliable (Sherwin, 1998); this might hold relevance for people with ME/CFS given that the illness is believed by BPS proponents to be perpetuated by "catastrophic illness beliefs" (Deary et al., 2007, p.787). Physical harms and risk of further marginalisation may arise from patient withdrawal from healthcare, illustrated in this study and more broadly and associated with loss of patient trust (Dickson et al., 2007; MEA, 2015; OxCATTs, 2019). Reluctance to engage with healthcare services may also be understood as a way of coping with identity threats posed by stigma and discrimination (Major & O'Brien, 2005). Finally, increased physical ill-health resulting from the chronic, uncontrollable stressor of stigma cannot be ruled out. Some

research suggests that increased allostatic load (from chronic, cumulative stress) is associated with greater severity and frequency of symptoms in people with ME/CFS (Goertzel et al., 2006); it is noteworthy that the most severely affected participant in this study (Elizabeth) also reported the most enduring, dismissive and psychologising healthcare encounters. The diversity of experiences across participants in this study (with a bias towards unsatisfactory experiences) is supported more widely (MEA, 2015; Gilje et al., 2008). Such variation may be to some extent elucidated through an intersectionality lens.

Experience of stigma and marginalisation may be influenced by intersection of multiple layers of socio-demographic disadvantage (Banks, 2018; Frost, 2011). Art, being a man with a high level of education and relative economic stability (intersection of gender, economic and educative relative advantage) may be less vulnerable to stigma and marginalisation than Elizabeth (intersection of gender and economic disadvantage, educatively marginalised through disability). It should however be noted that an additive approach to intersectionality is over-simplistic (Crenshaw, 1989). Gendered experiences of healthcare suggest that women are more likely to be dismissed and/or psychologised than men (Hoffmann & Tarzian, 2001; Richman & Jason, 2001), whilst ME/CFS was historically (re-)framed by psychiatrists as hysteria on the grounds that it disproportionately affected women (McEvedy & Beard, 1970). It is thus noteworthy that Art reported almost always being believed by HCPs, whilst Elizabeth's experience was one of systematic disbelief. Stigma and epistemic injustice arise out of a power differential (Link & Phelan, 2001; Fricker, 2007) and intersectionality can be seen as intersecting gradients of social and identity power. Given the association between social power and epistemic trustworthiness (Fricker, 2007), it may be that greater epistemic trustworthiness is accorded by HCPs to patients who can be positioned at the intersection of multiple social advantage. Additionally, variables such as age at illness onset may impact; non-ME/CFS research has shown that higher levels of self-reported stigma are associated with earlier illness onset (Świtaj et al., 2009). From this perspective, it is noteworthy that Elizabeth (who recounted the most marked experiences of stigma) reported the earliest age at illness onset of all participants.

### *4.2 Methodological Critique*

The small sample size allowed an in-depth exploration and idiographic focus which would not otherwise have been possible. Qualitative research does not aim to be generalisable in a probabilistic-statistical sense (Smith, 2018) and the study aims to "demonstrate existence, not incidence" (Smith et al., 2009, p.30). The study however may be considered analytically generalisable in supporting themes, findings and concepts found in extant literature (Smith, 2018). Thick description may also facilitate a case-to-case transferability to contexts outside of ME/CFS healthcare (Polit & Beck, 2010), for example through elucidating processes by which certain patient groups are stigmatised and marginalised. Finally, the study may offer naturalistic generalisability (Smith, 2018) in resonating with readers' personal and professional experiences. There are however some limitations to the study.

Members of patient organisations may have more severe and enduring experiences of ME/CFS relative to the wider ME/CFS population (Shepherd, 2017), may take a more critical stance towards the medical establishment and/or may be more willing to participate in critical research. This, accompanied by the study's explicit focus on stigma and marginalisation, may be argued to bias the sample towards greater experience of stigma and marginalisation. However, there was notable diversity in the sample in terms of

severity of symptoms, illness duration and degree of experience of stigma and marginalisation. Furthermore, exclusion criteria included a diagnosis of fibromyalgia and/or any psychological or psychiatric condition, for reasons outlined in section 2.3. Feedback from people with ME/CFS expressing an interest in the study suggested that these exclusion criteria may have excluded people with more complex, severe and enduring experiences of ME/CFS, alongside more negative healthcare experiences. Finally, more severely affected patients are often excluded from research and marginalised from health and social care (McDermott et al., 2014). Taken together, these considerations suggest that the study has not captured the full extent of healthcare-related stigma and marginalisation within this patient group. The study lacked socio-demographic diversity from some perspectives, notably as regards race and ethnicity; all participants were white. Research suggests that racial and ethnic disadvantage can impact on stigma within ME/CFS and wider healthcare, notably increasing likelihood of being psychologised (Annamma et al., 2013; de Carvalho Leite et al., 2011); this again suggests that the study did not fully capture the phenomenon under investigation. Finally, the study did not consider individual participant factors (for example self-stigma, self-efficacy, locus of control) in influencing the phenomenon under investigation. Psychologisation of ME/CFS emphasises purported individual factors at the detriment of social and societal factors, and this study sought to address this bias.

### *4.3 Reflexivity*

As a person with ME/CFS, I continuously reflected upon potential advantages and drawbacks to being part of the researched group (Coyle, 2016; Wilkinson & Kitzinger, 1996). I drew upon reflective journaling, audit trail, supervision (including analyst triangulation) and skills from therapy training (attending to transference/countertransference) to guard against entanglement of personal experiences, beliefs and values with those of participants (Shenton, 2004; Berger, 2015). Equally, I acknowledge that personal and professional experience in ME/CFS (see section 2.2) has coloured the research process. My background, combined with a reflexive approach, might be considered a form of prolonged engagement (Lincoln & Guba, 1985), facilitating greater sensitivity to context (Yardley, 2015). Equally, I acknowledge that participants draw upon multiple social identities beyond illness, many of which I might not share (Lyons, 2016). My therapy background and inclination to work interpretatively indubitably influenced the research process. For example, in interview I sometimes found myself tentatively interpreting a participant remark and inviting feedback to facilitate depth (e.g. ‘sounds like a lonely space to be in?’). It is noteworthy that participants were able to correct such tentative interpretations as they deemed necessary; I consider this indicative of trust and rapport arguably facilitated through my identification as a person with ME/CFS (Berger, 2015).

### *4.4 Implications for Practice and Policy and Future Directions*

The study highlights a need for improved HCP education in ME/CFS, as supported in extant literature (Unger et al, 2016). Beyond increasing understanding of ME/CFS as a biomedical entity and providing insight into the lived experience of ME/CFS, this study also indicates a need for greater emphasis on critical reflexivity (Fricker, 2007; Wilson, 2020) in HCP education and continued professional development. Critical reflexivity training should encourage HCPs to critically reflect upon personal bias and stereotyping (individual level factors) and the limitations as well as strengths of their training models and underlying philosophy (structural factors). Such critical self-awareness in HCPs might foster a degree of

epistemic humility which may counter epistemic injustice; epistemic humility has also been proposed as an ingredient which may facilitate more collaborative care partnership (Buchman et al., 2017). Epistemic humility may also foster a willingness to listen to and learn from patients; listening to patients has been suggested as a means by which HCPs can regain the trust of people with ME/CFS (Shepherd, 2017). Consideration should be given to involving patients as educators in HCP training (Chew-Graham et al., 2010). Additionally, imperatives of informed consent, understood as processual rather than a one-off event, should be underlined in HCP education, whilst ways in which informed consent can be ensured in therapeutic contexts require more attention.

On a policy-making level, greater awareness is indicated of how structural factors can subject patients to epistemic injustice and inadequate healthcare (Kidd & Carel, 2017). In particular, the values and assumptions implicit in EBP, and how these translate into practice, deserve critical scrutiny (Goldenberg, 2006). Accepting the patient narrative as a valuable form of evidence as per the EBP model (Sackett et al., 1996) and involving patients in the development of policies that affect their care (Carel & Kidd, 2014) is indicated; the outcome of the current review of NICE (2018) guidelines on ME/CFS will provide an indicator on progress in this respect. Given that NICE guidelines and NHS ethos are predicated upon patient-centred care, exploration of factors which block the translation of policy and ethos into practice is indicated. As regards healthcare models, this study broadly supports research suggesting that the BPS model of ME/CFS and associated interventions can cause harm to patients (Geraghty & Blease, 2019; Blease et al., 2017); research funding should prioritize high-quality biomedical studies alongside research exploring alternatives to or revisions of the BPS model of ME/CFS. The suggestion of a phenomenologically-informed approach (Blease et al., 2017) should be seriously considered. The BPS model of ME/CFS should account for a previously unacknowledged social factor, that of healthcare-related stigma and marginalisation, to some extent perpetuated by the BPS model itself.

Further research into stigma and marginalisation of people with ME/CFS should draw upon more socio-demographically diverse samples, taking a targeted approach to include participants with multiple social disadvantage. Research should be conducted through a lens of (non-additive) intersectionality combined with an institutional factors approach (Gkiouleka et al., 2018); this might help to elucidate individual differences whilst acknowledging the broader socio-political context of stigma. Further ME/CFS research is also indicated exploring healthcare experiences of people who have multiple diagnoses, especially where those diagnoses (such as fibromyalgia and mental health conditions) are associated with stigma, alongside more targeted research exploring healthcare experiences of those at the most severe end of the ME/CFS spectrum. Research on more severely affected patients should consider stigma and marginalisation in social care and potential impact on illness burden (25% ME Group, 2010).

Overall, the study's findings suggest that healthcare stigma, bound up with epistemic injustice, can lead to marginalisation within healthcare services and across multiple domains, reducing life chances and increasing illness burden. Intersection of social disadvantage, along with age of illness onset, may impact on experience of stigma and marginalisation and this warrants further research. People with ME/CFS are subjected to both individual and structural level stigma, to both testimonial and hermeneutical injustice, and can be considered "doubly wronged" (Fricker, 2007, p.159). Implications for practice and policy alongside future directions have been discussed. Greater emphasis within HCP education on critical reflexivity and

(processual) informed consent is recommended, alongside broader examination of individual and structural factors perpetuating epistemic injustice in healthcare and blocking the translation of patient-centred care principles into practice. Valuing patient testimony as a source of evidence is central to these recommendations.

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## Appendices

**Appendix 1**

***Participant Invitation***

**Invitation to Take Part in ME/CFS Research**

If you are an adult living with ME/CFS, and you live in Gloucestershire, you are invited to take part in a research study that I am conducting as part of my studies in psychology at the University of Derby.

**Study Title**

Stigma and marginalisation in the healthcare encounter and perceived impact on illness burden: The lived experiences of people with myalgic encephalomyelitis / chronic fatigue syndrome (ME/CFS).

**What is the study about?**

The study aims to explore the experiences of people with ME/CFS regarding stigma and marginalisation in healthcare encounters, in both primary and secondary care*. The study will also explore how such healthcare encounters impact on overall illness burden (physical and emotional health, overall well-being) from the perspective of people with ME/CFS. Finally, the study will explore what factors might contribute to more positive healthcare encounters, again from the perspective of people with ME/CFS.

Participation in this study is entirely voluntary and, if you wish to take part, you will be asked to meet with the researcher to talk about the above topics. This is referred to as an interview. There are no right or wrong answers to questions; rather, the emphasis is on allowing you to respond to the researcher's broad questions in a way that is meaningful to you. The duration of the interview can vary, but is generally expected to take around 60 minutes.

**Who can take part?**

You can take part in this study if:

- You have a medical diagnosis of ME/CFS.
- You are 18 years of age or over.
- You live in Gloucestershire.
- ME/CFS is your primary (main) diagnosis and health concern.
- You are fluent in English.
- You have experience of both primary and secondary healthcare for ME/CFS.*

*Primary care would include your GP, whilst secondary care would include specialist ME/CFS care services, for example cognitive behavioural therapy (CBT) and graded exercise therapy (GET).

**Who cannot take part?**

There are few cases where, unfortunately, you cannot take part in this study. This is in order to protect participants from harm, and to ensure that the study explores experiences of people with ME/CFS as opposed to other health conditions. You cannot take part in this study if:

- You have a diagnosed mental health condition (for example, depression, anxiety, or any other mental health diagnosis).

- You have a medical diagnosis of fibromyalgia.
- You consider yourself to be a vulnerable adult or adult at risk.

**What should I do if I would like to know more?**

If you are interested in taking part in the study, please contact the researcher, **Jo Hunt**, via email: **j.hunt21@unimail.derby.ac.uk.**

Best wishes,

Jo Hunt.

**Appendix 2**

***Participant Information Sheet***

**Participant Information Sheet**

Thank you for expressing an interest in a research study that I am conducting as part of my studies in psychology at the University of Derby.

**Study Title:**

Stigma and marginalisation in the healthcare encounter and perceived impact on illness burden: The lived experiences of people with myalgic encephalomyelitis / chronic fatigue syndrome (ME/CFS).

**What is the study about?**

The study aims to explore the experiences of people with ME/CFS regarding stigma and marginalisation in healthcare encounters, in both primary and secondary care. The study will also explore how such healthcare encounters impact on overall illness burden (physical and emotional health, overall well-being) from the perspective of people with ME/CFS. Finally, the study will explore what factors might contribute to more positive healthcare encounters, again from the perspective of people with ME/CFS.

**Do I have to take part?**

Participation in this study is entirely voluntary. If you decide to participate, you have the right to withdraw from the study, without giving reason and without consequence, up to two weeks after you have taken part in the interview. After this time, data be analysed, and it will no longer be possible to withdraw. You will be asked to choose a pseudonym (false name) which will be used in data analysis and write-up in order to protect your anonymity. If you wish to withdraw within two weeks from interview, you should contact the researcher (details below), with your pseudonym, requesting to withdraw from the study. Your data will then be deleted and will not be used in the research.

**What happens to me if I take part?**

If you wish to participate in the study, you will be asked to meet with the researcher to talk about the above topics. This is referred to as an interview. There are no right or wrong answers or expectations about what you should say; rather, the emphasis is on allowing you to respond to the researcher's broad questions in a way that is meaningful to you. The duration of the interview can vary but is generally expected to take around 60 minutes; interviews will be recorded to assist accurate analysis of data. Please be aware that this kind of interview is not the same as counselling or therapy, and the researcher cannot act in the role of counsellor or therapist. However, it is important that you feel comfortable during interview, and the researcher will be able to signpost you to sources of information, support and guidance in case you feel that you should need it.

**Will my participation in this study be kept confidential?**

Your data (personal details and data generated through interview) will be processed and stored in accordance with ethical and legal guidelines outlined within the General Data Protection Regulation (part of the Data Protection Act 2018), within the British Psychological Society's code of research ethics, and within Derby University's code of scientific practice. This is with a view to safeguarding your anonymity and keeping your data confidential. Electronic data (including interview recordings) will be stored and processed on a password protected computer to which only the researcher has access, whilst paper-based documents will be kept in locked storage when not in use and will not leave the work premises of the researcher.

You will be asked to choose a pseudonym (false name) which will be used throughout data analysis and write-up to protect your anonymity. Recordings, transcripts (written versions of the recording) and analysis notes will be labelled only with your pseudonym; any identifying details will be removed. Your personal details (name, contact details) will be kept separate from data generated through interview and will not be used in analysis and write-up. Data collection will be limited to that which is necessary to fulfil the aims of the research; only the researcher and research supervisor will have access to this information. Under GDPR provisions, your data will be kept securely for a period of seven years and then safely destroyed. If you withdraw from the research, which you can do up to two weeks post interview, your data will be destroyed and not used in the research.

**What will happen to the results of the research study?**

The study is part of my studies in psychology at Derby University. The study report will be read and assessed by my research supervisor (details below) and a second assessor. The study report may also be published or further disseminated, and the study's findings will be shared with you. The study report, including any quotes from interview, will be written up in such a way that you are not identifiable, in order to maintain your anonymity.

**Contact for further information**

Further information on the study (study aims, what to expect, data protection etc.) can be obtained from:

Researcher: **Jo Hunt j.hunt21@unimail.derby.ac.uk**

If you have any concerns relating to this study please contact:

Research Supervisor: **Dr Dan Herron D.Herron@derby.ac.uk 01332 597749**

Thank you for taking the time to read this sheet. I hope that you feel able to take part in the study. If you have any further questions about the study, please let me know. If you are happy to take part in the study, please read and complete the consent form.

**Appendix 3**

***Participant Consent Form***

**Consent Form**

**Study Title:**

Stigma and marginalisation in the healthcare encounter and impact on illness burden: The lived experiences of people with myalgic encephalomyelitis / chronic fatigue syndrome (ME/CFS).

| | **Please INITIAL each statement you agree to** |
|---|---|
| 1. I confirm that I have read and understood the participant information sheet for the above study. | ☐ |
| 2. I have had the opportunity to ask any questions. | ☐ |
| 3. I understand that my participation is voluntary and that I am free to withdraw up to two weeks after participating in interview without giving any reason. | ☐ |
| 4. I confirm that I am aged 18 or over. | ☐ |
| 5. I confirm that I have a medical diagnosis of ME/CFS. | ☐ |
| 6. I confirm that ME/CFS is my primary diagnosis / health concern. | ☐ |
| 7. I confirm that I do not have any diagnosed mental health conditions. | ☐ |
| 8. I confirm that I do not have a medical diagnosis of fibromyalgia. | ☐ |
| 9. I confirm that I have experience of both primary and secondary healthcare for ME/CFS. | ☐ |
| 10. I understand that the interview will be recorded and transcribed, and that the transcripts will not include any personal identifying information. | ☐ |

11. I understand that the recordings and anonymised transcripts will be stored securely for seven years after the end of study declaration. ☐

12. I understand that quotes from the audio-recording may be included in future reports or publications, but that these will be anonymous and I will not be identified. ☐

13. I understand that the researcher will be collecting data from my participation in this study as described in the information sheet. ☐

14. As a researcher I am bound by ethical and legal guidelines on data protection and while this allows me to use your data, it also means I have obligations towards you to:

- not seek more information from you than what is essential and necessary for the study;
- make sure that you are not identified by the data by anonymising it using a pseudonym chosen by you;
- use your anonymised data only for the purposes of this study and for any relevant publications that arise from it;
- store data safely on a password-protected system to which only the named researcher has access;
- not keep your information for longer than is necessary (usually for seven years);
- safely destroy your data by shredding or permanently deleting them.

The University of Derby will act as the Data Controller for this study. This means that the University is responsible for looking after your information and using it properly. Researchers on the project with access to the data are highly qualified and experienced and have been very careful to ensure the security of your data. The study was approved for its ethical standards by The University of Derby Human Sciences Research Ethics Committee. However, in the unlikely event that you feel you need to make a complaint regarding the use of your information, you can contact the Data Protection Officer at the University of Derby: James Eaglesfield (01332) 591762 or the Information Commissioners Office 0303 123 1113.

Further information on the study (study aims, what to expect, data protection etc.) can be obtained from:

Researcher: **Jo Hunt j.hunt21@unimail.derby.ac.uk**

If you have any concerns relating to this study please contact:

Research Supervisor: **Dr Dan Herron D.Herron@derby.ac.uk 01332 597749**

at the University of Derby, Kedleston Road, Derby DE22 1GB

I understood and consent to the above. ☐

15. I agree to take part in the study. ☐

Please print your name, sign and date below if you have read and agreed to all the above, and wish to participate in this study.

Participant name: ..... Signature: ..... Date: .....

Researcher name: ..... Signature: ..... Date: .....

**Appendix 4**

***Demographic Questionnaire***

**Demographic Questionnaire**

I would like to collect some data as part of the study which will enable me to better understand the context of your healthcare experiences, and to gain a more holistic understanding of you as a person. This is particularly helpful when considering stigma and marginalisation, since social categories (demographics) may impact on a person's experiences of stigma and marginalisation.

The data that you provide me with will not be linked to your name or any identifying details, to protect your anonymity. In the study report, I will include an overview of participant demographics, but will not include any of your personal details (name, contact details, any identifying data) which will treated as confidential at all times as described in the participant information sheet. Please tick or circle your responses below.

1. What is your age? (Please write your age below, or write 'prefer not to say')

2. How would you describe your ethnicity and race? (Please feel free to specify further within the broad categories listed here).

- Black
- Asian
- White
- Mixed
- Other (please specify)
- Prefer not to say

3. What gender do you identify as?

- Woman
- Man
- Non-binary
- Other (please specify)
- Prefer not to say

4. What is the highest level of education you have completed?

- No schooling completed
- Secondary school education (GSCEs / O levels)
- Secondary school education (A levels)
- Diploma
- Foundation degree or HND
- Bachelor's degree
- Master's degree
- PhD / Doctorate
- Prefer not to say

5. What is your employment status?

- Employed for wages
- Self-employed
- Unable to work
- Out of work and looking for work
- Out of work but not currently looking for work

- A homemaker
- A student
- Retired
- Other (please specify)
- Prefer not to say

6. How long have you been diagnosed with ME/CFS? (Years and / or months)

7. How would you rate the severity of your symptoms, using the below scale? If your symptoms fluctuate from day to day (or hour to hour) please try to give an indication of how you feel your symptoms are most of the time. If you feel the severity of your symptoms crosses two categories (e.g. moderate-severe) please circle or tick all categories which apply. Please read the below scale before giving your response based on the following options.

- Very severe
- Severe
- Moderate
- Moderate to mild

The MEA Disability Rating Scale, 2016

(Taken from the website of the ME Association, https://www.meassociation.org.uk/2016/05/the-mea-disability-rating-scale-2016/)

**VERY SEVERE**

---

**100% DISABLED:** Severe symptoms – often on a continual basis. Cognitive function (i.e. short-term memory, concentration, attention span) is likely to be very poor. Bedridden and incapable of living independently. Requires a great deal of supervision and practical support – including disability aids such as a hoist or a stair lift – with all aspects of personal care (i.e. feeding, dressing, washing) on a 24-hour basis.
**90% DISABLED:** Severe symptoms, often including marked cognitive dysfunction, for much or all of the time. Bedridden and housebound for much or all of the time. Has considerable difficulties with all aspects of personal care. Unable to plan or prepare meals. Requires practical support and supervision on a 24-hour basis.

---

**SEVERE**

---

**80% DISABLED:** Moderate to severe symptoms for most or all of the time. Only able to carry out a very limited range of physical activities relating to personal care without help. Requires help with meal planning and preparation. Frequently unable to leave the house and may be confined to a wheelchair when up, or spends much of the day in bed. Unable to concentrate for more than short periods of time. Usually requires daytime and night-time supervision.
**70% DISABLED:** Moderate to severe symptoms for most or all of the time. Confined to the house for much or all of the time. Normally requires help with various aspects of personal care and meal planning and preparation, possibly on a 24-hour basis. Very limited mobility. May require wheelchair assistance.

---

**MODERATE**

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**60% DISABLED:** Moderate symptoms for much or all of the time. Significant symptom exacerbation follows mental or physical exertion. Not usually confined to the house but has significant restrictions on mobility

when outside and may require wheelchair assistance. Likely to require help with aspects of personal care and meal preparation – but not necessarily on a full-time basis. Requires regular rest periods during the day. Unable to resume any meaningful regular employment or education.
**50% DISABLED:** Moderate symptoms for much or all of the time. Symptom exacerbation follows mental or physical exertion. Not usually confined to the house but mobility restricted to walking up to a few hundred yards at best. May require help with some aspects of personal care. May require help with meal planning and preparation. Requires regular rest periods during the day. Able to carry out light activities (i.e. housework, desk work) linked to normal daily living for short periods but not able to resume regular employment or education.
**40% DISABLED:** Moderate symptoms for some or much of the time. Normally able to carry out most activities linked to personal care and normal daily living, but may require assistance with meal preparation. May be able to cope with some work-related tasks for short periods – provided they are not mentally or physically strenuous – but not able to resume regular work or education.

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**MODERATE TO MILD**

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**30% DISABLED:** Fluctuating level of mild to moderate symptoms. Normally able to carry out all aspects of personal care and to plan and prepare meals. Able to walk short distances on a regular basis. May be able to return to work on a flexible or part-time basis – provided adjustments are made to cope with physical activity or cognitive problems. May have to stop leisure or social pursuits to resume work or education.
**20% DISABLED:** Normally only mild symptoms at rest but exacerbation will follow activity. Able to carry out all aspects of personal care and to plan and prepare meals. Able to walk short to medium distances (i.e. up to half a mile) on a regular basis. Normally able to return to flexible or part-time work or education.
**10% DISABLED:** Generally well with only occasional mild symptoms. No problems with personal care or daily living. Mobility and cognitive functions may still be restricted but almost back to previous levels. May be able to return to full-time work or education.
**0% DISABLED:** Fit and well for at least the past three months. No symptoms at rest or after exertion. Capable of full-time work or education.

Thank you for taking the time to complete this questionnaire.

**Appendix 5**

***Topic Guide for Participant Interviews***

**Topic Guide**

Interview questions are informed through prior research (notably Geraghty & Blease, 2019; Geraghty & Blease, 2018). These papers highlight ME/CFS patients' difficulties accessing medical care, the 'sick role', and social support along with high levels of dissatisfaction with quality of care, negative responses to CBT and GET, lack of informed consent in treatments, challenges to patient narrative and potential psychological harm.

Questions in regular type, prompts in italics.

**Introduction**

Introduce self. The aim of this interview is to explore your experiences of stigma and marginalisation in healthcare encounters, as a person with ME/CFS. I will also ask you about how such healthcare encounters may have impacted on your overall illness burden (physical and emotional health, overall well-being). Finally, I will ask you about what factors you think might contribute to more positive healthcare encounters. There are no right or wrong answers or expectations about what you should say; rather, the emphasis is on allowing you to respond to my questions in a way that is meaningful to you.

**Opening questions**

1. How do you prefer to refer to ME/CFS? (ME, CFS, ME/CFS etc.)

(I will then try to use the same nomenclature as the participant – ME/CFS is used throughout the topic guide as default)

2. Tell me about how you came to be diagnosed with ME/CFS? (adapted from Dickson et al., 2007 – starting with a broad and less sensitive question before digging deeper, seeking to build trust and rapport)

3. What does ME/CFS mean to you?

*- How did your diagnosis make you feel?*
*- How do you make sense of having ME/CFS? (could link this back in part to preferred name for ME/CFS)*

**Experiences of healthcare encounters**

4. Tell me about the kind of healthcare encounters / experiences you have had, as somebody diagnosed with ME/CFS?

*- Primary care*
*- Secondary care / specialist care services*

5. How did these experiences make you feel? / What did they mean to you?

6. How involved did you feel in these encounters?

   *- To what extent did you feel that your ideas about and understanding of your illness were taken on board?*
   *- To what extent did you feel able to ask questions and / or raise concerns?*
   *- How did this make you feel? / What did this mean to you?*

7. To what extent were the reasons underpinning any treatment options explained to you (to what extent were you clear on why you were being offered these treatments)? By treatment I mean medication, psychological therapy or physiotherapy, or any other advice.

*- How did this make you feel? / What did this mean to you?*

8. To what extent were potential advantages and potential drawbacks of treatments explained to you?

*- How did this make you feel? / What did this mean to you?*

**Potential impact of healthcare encounters on illness burden**

9. Do you feel that these encounters have had an impact on your illness burden? If so, in what way? (explain illness burden – experience of ME/CFS, mental and physical health, symptoms, overall well-being)

10. How did these encounters make you feel? / What did these encounters mean to you?

*- About yourself (self-relating)*
*- About yourself in relation to others*
*- About your illness*
*- About healthcare professionals and the healthcare system in general*

*- Any short-term impact?*
*- Any longer term impact?*
*- Psychological, social, physical aspects*

**Factors that might contribute to a more positive healthcare encounter**

11. What would an ideal healthcare encounter look like from your perspective?

*- Include aspects beyond medical / physical treatment or cure, in view of the current lack of either of these.*
*- What things would you have liked to change about the healthcare encounters you've told me about (and why)*

12. What would such a healthcare encounter (ideal or more positive encounter) mean to you? / How would such a healthcare encounter make you feel?

*- About yourself (self-relating)*
*- About yourself in relation to others*
*- About your illness*
*- About healthcare professionals and the healthcare system in general*

**Closing question**

13. Is there anything else you would like to add to what we have talked about today?

Thank you very much for taking the time to share your experiences with me today, and for participating in this study.

Topic Guide References

Dickson, A., Knussen, C., & Flowers, P. (2007). Stigma and the delegitimation experience: An interpretative phenomenological analysis of people living with chronic fatigue syndrome. *Psychology & Health*, *22*(7), 851–867.

Geraghty, K. J., & Blease, C. (2019). Myalgic encephalomyelitis/chronic fatigue syndrome and the biopsychosocial model: a review of patient harm and distress in the medical encounter. *Disability And Rehabilitation*, 1–10.

Geraghty, K.J. & Blease, C. (2018). Cognitive behavioural therapy in the treatment of chronic fatigue syndrome: A narrative review on efficacy and informed consent. *Journal of Health Psychology*, *23*(1), 127-138.

**Appendix 6**

***Participant Debrief***

**Debrief Sheet**

**Study Title:**

Stigma and marginalisation in the healthcare encounter and perceived impact on illness burden: The lived experiences of people with myalgic encephalomyelitis / chronic fatigue syndrome (ME/CFS).

Thank you for giving your time to contribute to this study.

**What is the study about?**

The study aims to explore the experiences of people with ME/CFS regarding stigma and marginalisation in healthcare encounters, in both primary and secondary care. The study will also explore how such healthcare encounters impact on overall illness burden (physical and emotional health, overall well-being) from the perspective of people with ME/CFS. Finally, the study will explore what factors might contribute to more positive healthcare encounters, again from the perspective of people with ME/CFS.

**Will my participation in this study be kept confidential?**

Please be reminded that your data (personal details and data generated through interview) will be processed and stored in accordance with ethical and legal guidelines outlined within the General Data Protection Regulation (part of the Data Protection Act 2018), within the British Psychological Society's code of research ethics, and within University of Derby's code of scientific practice. This is with a view to safeguarding your anonymity and keeping your data confidential. Electronic data will be stored and processed on a password protected computer to which only the researcher has access, whilst paper-based documents will be kept in locked storage when not in use and will not leave the work premises of the researcher. A pseudonym (false name) chosen by you will be used throughout data analysis and write-up to protect your anonymity; your personal details (name, contact details) will be kept separate from data generated through interview and will not be used in analysis and write-up. Data collection will be limited to that which is necessary to fulfil the aims of the research; only the researcher and research supervisor will have access to this information. Under GDPR provisions, your data will be kept securely for up to seven years; it will then be deleted / destroyed.

**Do I still have to take part?**

Your participation in the study is very much appreciated but if you decide that you would like to withdraw your data from the study, you can do so without giving any reason and without consequence. Please note that this withdrawal can only take place up to two weeks following the day that you participated in the research interview; after this time, it will be too late as the data will have been analysed. You can withdraw your data by contacting me with your chosen pseudonym via email at: j.hunt21@unimail.derby.ac.uk. In case of withdrawal, your data will be deleted /destroyed and not used in the research. This pseudonym is indicated below for your convenience.

Pseudonym: .....

**What if I have further questions or concerns?**

Further information on the study (study aims, what to expect, data protection etc.) can be obtained from:

Researcher: **Jo Hunt j.hunt21@unimail.derby.ac.uk**

If you have any concerns relating to this study please contact:

Research Supervisor: **Dr Dan Herron D.Herron@derby.ac.uk 01332 597749**

If your participation in this study has raised any issues relating to physical or emotional health, disability matters and / or social support that you would like to discuss further, you can contact the following organisations:

- ME Connect: Support line of the ME Association, offers support, information and signposting to anyone affected by ME/CFS.

Telephone: 0344 576 5326 (available every day, during the hours of 10am-12noon, 2pm-4pm and 7pm-9pm)

Email available via online form at: https://www.meassociation.org.uk/contact-meconnect/

- Samaritans: Emotional support 24 hours day, 365 days a year.

Telephone: 116123. Email: jo@samaritans.org (emails are usually responded to within 24 hours).

- BACP (British Association for Counselling and Psychotherapy)

BACP provides a therapist directory where you can search for counsellors and psychotherapists in your area, with areas of interest and specialisms to suit your needs: https://www.bacp.co.uk/search/Therapists

You can also check that any counsellor or psychotherapist you choose is BACP registered: https://www.bacp.co.uk/search/Register

- Your GP is a recommended point of contact for any physical and / or mental health concerns
- In Gloucestershire, NHS Let's Talk (IAPT) have a self-referral route for primary care talking therapies (via email form or telephone) – without having to be GP-referred

Website: https://www.talk2gether.nhs.uk/contact-us/ Telephone: 0800 073 2200

Finally, I would like to thank you again for your participation in this study.

**Appendix 7**

***Information Relating to Interview Questions, Given to Participants Before Interview***

The interviews will be very loosely structured around three broad questions. There are no right or wrong answers to these questions; rather, you should answer them in a way that is meaningful for you. The questions may be phrased differently in interview and may not always follow in the same order; each interview will be unique because I try to be led by you within the parameters of the broad research questions. I will also be asking follow-up questions within each broad interview question; this will depend to some extent on what you tell me and so cannot be planned in advance. The three research questions are:

1) What are your experiences, as a person living with ME/CFS, regarding stigma and marginalisation in healthcare encounters, in both primary and secondary (specialist) care?

The kinds of areas I will be exploring include: how involved you feel or felt in your care, how informed you feel or felt about the reasons for any treatments offered, to what extent you felt able to ask questions and / or raise concerns.

2) What impact (if any) do you feel these healthcare encounters have had on your overall illness burden?

Illness burden may be defined as your overall sense of wellbeing, including physical and emotional health, as well as social aspects such as quality of relationships with others.

3) What factors do you think would contribute to more positive healthcare encounters, as a person living with ME/CFS?

Additionally, before we move into the above topics, I will also ask you to tell me about how you came to be diagnosed with ME/CFS, to give some context to our interview.