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Citation: Strassheim, Victoria, Newton, Julia L. and Collins, Tracy (2021) Experiences of Living with Severe Chronic Fatigue Syndrome/Myalgic Encephalomyelitis. *Healthcare*, 9 (2). p. 168. ISSN 2227-9032

Published by: MDPI

URL: <https://doi.org/10.3390/healthcare9020168> <<https://doi.org/10.3390/healthcare9020168>>

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1 Article

2 Experiences of living with Chronic Fatigue 3 Syndrome/Myalgic Encephalomyelitis

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15 Received: date; Accepted: date; Published: date

16 **Abstract:** Chronic Fatigue Syndrome/Myalgic Encephalomyelitis (CFS/ME) is a rare disease with no
17 known etiology. It affects 0.4% of the population, 25% of which experience the severe and very
18 severe categories, defined as being wheelchair-, house- and bed-bound. Currently, the absence of
19 biomarkers necessitates a diagnosis by exclusion, which can create stigma around the illness. Very
20 little research has been conducted with the partly defined severe and very severe category of
21 CFS/ME. This is in part because the significant health burdens experienced by these people create
22 difficulties engaging in research and healthcare provision as it is currently delivered. This
23 qualitative study explores the experiences of five individuals living with CFS/ME in its most severe
24 form through semi-structured interviews. A six-phase themed analysis was performed using
25 interview transcripts, which included identifying, analysing and reporting patterns amongst the
26 interviews. Inductive analysis was performed, coding the data without trying to fit it into a pre-
27 existing framework or pre-conception, allowing the personal experiences of the five individuals to
28 be expressed freely. Overarching themes of ‘Lived Experience’, ‘Challenges to daily life’ and
29 ‘Management of the condition’ were identified. These themes highlight factors that place people at
30 greater risk of experiencing the more severe presentation of CFS/ME. It is hoped that these insights
31 will allow research and clinical communities to engage more effectively with the severely affected
32 CFS/ME population.

33 **Keywords:** ME/CFS 1; severe 2; very severe 3; housebound 4; qualitative 5; interview 6; experience
34 7.

35

36 1. Introduction

37 Myalgia Encephalomyelitis (CFS/ME) is a rare disease with no known etiology [1, 2]. Its cause
38 is unknown[3] and studies suggest it affects 0.4% of the population [4]. Criteria have been produced
39 to identify clinical characteristics[5]; however, diagnosis remains by exclusion due to the absence of
40 biomarkers[6] which has led to significant stigma[7].

41 Diagnostic criteria have evolved, with better understanding of the condition. The International
42 Consensus in 2011[8] developed from a growing understanding of the condition and has led to the
43 identification of heterogeneous groups within the CFS/ME population.

44 Subgroups had previously been defined by Cox et al[9, 10]. The categories are mild, moderate,
45 severe and very severe and were implemented in the National Institute for Health and Care
46 Excellence guidelines. Severe and very severe CFS/ME individuals are wheelchair, house or
47 bedbound, due to the severity of their symptom burden.

48 The severe and very severe CFS/ME population find it difficult to access their wider
49 environments. This creates difficulty for them to engage in research and healthcare provision as it is
50 currently delivered. Therefore, this group are classed as hard to reach[11]. It is suspected that 10-25%
51 of the 0.4% CFS/ME population are in the severe to very severe category. This figure has been
52 supported by the CFS/ME charities and evidenced in the research literature [12, 13]. It has been
53 estimated that there are up to 40 people with CFS/ME in each GP practice across England, 25% of
54 which are severe and very severe[14]. Therefore, across England alone there are approximately 82,000
55 very vulnerable, severe and very severely affected CFS/ME individuals housebound and bedbound
56 that may not be currently not receiving appropriate treatment.

57 Quantitative and qualitative data is limited due to the hard to reach nature of the severely
58 affected CFS/ME population. However, this is slowly changing, and texture is being added to the
59 objective evidence published[15-18] . In recent years more qualitative research has been performed
60 with the CFS/ME population. However, the gap in knowledge regarding severely and very severely
61 CFS/ME remains. Reviewing the research and evidence as it is published and reflecting on previous
62 trials leads to a better understanding of how variables impact each other. The PACE trial was one
63 such trial[19, 20]. It used operational diagnostic criteria to identify 600 patients to be randomised to
64 one of four treatment pathways: specialist care, plus or minus adaptive pacing therapy, Cognitive
65 Behavioural Therapy (CBT) and Graded Exercise Therapy (GET). The aim was to gather scientific
66 evidence to demonstrate the outcomes of each treatment. The research was planned and executed
67 prior to the 2011 International Consensus Criteria[8], which identified the heterogeneity of the
68 condition, categorising mild, moderate, severe and very severe[9, 10]. Within the Pace trial, the
69 eligibility assessment and consent for treatment was the outcome 6-minute walk test, which would
70 have precluded severe and very severe patients from taking part in the study[21].

71 In addition to exclusion from major research, it is emerging that people with CFS/ME experience
72 discrimination from healthcare professionals and wider society[14, 22]. This can be partially
73 attributed to a lack of understanding around the condition . CFS/ME is an illness not a disease and as
74 such currently has no identifiable pathology [27]. This does not sit well within the current biomedical
75 illness model that has been historically taught within medical schools[23]. Large institutions such as
76 Universities, Hospitals, Health and social care agencies function within clear objective, evaluative
77 models in which theories and ideas are critiqued and scrutinised. Evaluative models shift the
78 physiological state to defence which is incompatible with creativity and expansive theories[24].

79 People with CFS/ME that take a biomedical approach[23] may feel vulnerable without
80 biomarkers to authenticate their illness or referral to specialist services to endorse complex
81 overlapping symptoms[25]. The lack of identifiable pathology can result in psychological labelling or
82 somatisation[23]. This lack of understanding can contribute to a sense of blame shifting, where
83 patients may feel that they are held accountable for their poor health[23].

84 For therapy to work an individual must first accept their situation[26] and feel believed[18]. This
85 can be impeded when healthcare professionals, the structure they work within and wider society do
86 not acknowledge the limits of medical science and so continue to require hard evidence to support a
87 diagnosis, rather than treat and manage symptoms[23, 25, 27, 28]. The therapeutic relationship may
88 be further jeopardised if an individual with CFS/ME does not have the energy required to express
89 themselves effectively, particularly in a stressful situation where their illness may not be believed[18].

90 This article expands on a 2016 two phase pilot study. The project aimed to understand the
91 feasibility of severe and very severe CFS/ME individuals engaging with research, whilst scoping and
92 defining the prevalence of CFS/ME in the region. In that study, 2.5% of the 2,500 severe CFS/ME
93 population in the northeast of England were identified and characterised. The study also explored
94 the quality of life, symptom burden and impact of severe CFS/ME, collating a database from a postal
95 survey in phase 1[29]. Using the database, five individuals with severe and very severe CFS/ME

96 were identified for phase 2 of the study. Attending the participant in their own home to limit the
97 research burden on the CFS/ME individual and aid pacing strategies is novel to this research area.
98 The patient and public involvement and methodology for the entire study is presented elsewhere[29].
99 This paper presents findings from the five qualitative interviews.

100 1.2. Aims

101 To explore the personal experience and understanding of individuals with CFS/ME.
102 To identify overarching themes that may highlight factors putting people at greater risk of
103 experiencing the more severe presentation of CFS/ME.
104 To provide a better understanding of this population to allow healthcare and research
105 communities to engage with individuals more effectively.

106 2. Materials and Methods

107 2.2. Methodology

108 A qualitative methodology involving in-depth interviews and drawing on phenomenology was
109 selected. The interviews followed a semi-structured format[30], with interview schedules designed
110 and worded to establish rapport and explore the area of concern in an open and flexible manner[31].

111 The aim of the interviews was to understand the perspective of the individual experience[32], to
112 uncover personal meaning[31, 33].

113 This group have limited presence in the research literature, and the methodology was
114 explorative. By understanding the personal experience of individuals within the severe and very
115 severe CFS/ME community, future qualitative research may be better focused with an improved
116 understanding of practice, service and research delivery to this population.

117 2.3.1. Data Sampling

118 The sample was purposive[34], being defined as having severe CFS/ME as identified from the
119 database of self-reported CFS/ME individuals. How the participants were recruited is described in
120 some detail elsewhere [29]. However, for completeness, 483 questionnaire packs with an expression
121 of interest (EOI) to be involved in future studies was posted out. 425 packs via ME North East. Of
122 those 483 packs, 63 were returned in various stages of completion. Within the returns were over 40
123 completed expression of interest forms to be involved in future studies.

124 Resource was a consideration when recruiting the five participants to understand the feasibility
125 of engaging severely affected CFS/ME individuals with research. The returned EOI came from a large
126 geographical area, to be covered by one researcher. The fluctuating and unpredictable health of the
127 cohort was also considered. Therefore, participants were approached who lived near the research
128 base to limited disruption, if appointments had to be rescheduled at short notice. Participants within
129 the designated area were contacted until five had been recruited. These participants each agreed to
130 four home visits over a three-month period. In each case, the first visit was to obtain consent and
131 perform autonomic tests; the second visit was cognitive testing; the third was an interview and finally
132 a physical assessment.

133 The five participants completed a second consent form and patient information sheet, specific
134 to phase 2, which detailed the agreement to have results published in a peer reviewed journal.

135 The ethical principle of non-maleficence to do no harm[35] was a primary aim in planning the
136 research.

137

138 2.3.2. Data Collection

139 A pre organised date and time was agreed, with the understanding that it could be rescheduled
140 if necessary. Some participants gave specific instructions as to how to arrive at the home, in order
141 reduce noise and stimulation.

142 Each interview lasted approximately one to two hours. Two patients had planned in advance
143 and had compiled additional information to support the interview process and to reduce cognitive
144 strain. A third patient found following a conversation too difficult and supplied a report that they
145 had compiled over the course of their illness, which VS (author/researcher) read aloud as part of the
146 interview and the individual corrected or expanded on as they felt necessary. The youngest
147 participant, who had been ill since her teens, requested that her mother be present to help answer
148 any questions.

149 The interviews were conducted and recorded in conjunction with the collection of field notes
150 [36-38]. This was to increase the trustworthiness and rigour of the data as well as the transferability
151 of the findings[38].

152 2.3.3. Data Analysis

153 Thematic analysis drew on a grounded theory approach, with a method of inductive
154 analysis[39]. Data was coded without trying to fit it into a pre-existing framework. The researchers
155 were implicit in the process, taking a constructivist style to develop theories based on the data, not a
156 pre-defined question to answer. The aim was to explore the individuals' experiences with severe
157 CFS/ME.

158 Braun and Clark describe a six phased approach which was followed [39]: Initial familiarisation,
159 generating codes, searching for themes, reviewing the themes and codes, defining and naming the
160 codes and finally writing the report. The software package NVivo version 12 was used to organise
161 and analyse the data.

162 The transcripts were reviewed and codes were created and grouped into similar themes. Themes
163 were then grouped for similarities and reduced to a manageable number.

164 Reflexivity was employed through conducting the analysis and interpretation of the findings
165 with a second (qualitative) researcher (TC). The process was inductive, the data produced was
166 broad and rich.

167 2.4. Rigour

168 A research team with diverse background, knowledge and skills was created to collect and
169 analyse the data. The team had the collective ability to (1). access this hard to reach, community, (2).
170 collect the data whilst monitoring and limiting the impact the research might have on the participants
171 and (3). analyse the output. It was through this pooling of skills that this research was made possible.
172 The potential for research bias is recognised and was limited through co-author collaboration of the
173 research team to increase the trustworthiness of the analysis[38].

174 A clinically reasoned decision was taken not to have participants validate the researcher's
175 transcripts. It is understood that 'member checking' increases the internal validity and credibility of
176 research[38]. However, this was outweighed by the need to adhere to the ethical principle of non-
177 maleficence, to do no harm[35]. Participants were very fragile, and so it was felt that additional home
178 visits would impact health and function too greatly. However, the Chief Executive Officer of the
179 charity ME North East, who was instrumental in accessing the interviewed individuals, was sent a
180 summary of the findings on behalf of the participants.

181 2.5. Ethical Considerations

182 Full ethics approval was granted by North East-Newcastle and North Tyneside 2 Research Ethics
183 Committee. The participants had provided separate informed consent for both phase one and two of
184 the study.

185 Pseudonyms have been given to each of the participants to protect their identities and maintain
186 confidentiality

187 3. Results

188

Table 1. Participant Characteristics.

Pseudonym	Age range	Length of illness	Gender	Living arrangements	Support	Dependents	Background
JANE:	36-44	7 years	Female	Terraced house, predominantly bedbound, upstairs toilet.	Husband and mother	2 primary aged children	Very physically active prior to illness, professional, mother of 2 married.
DAVID:	36-45	18 years	Male	Semi-detached house. Predominantly housebound, living mostly in his bedroom.	Ageing parents	0	Single. Completed A-levels and managed to do office work between episodes of ill health, until early 20's. Had been very active prior to ill health.
ABI:	36-45	20 years	Female	Lived in a bungalow. Predominantly house bound.	Husband and ageing parents nearby	0	Married. Had been very active prior to ill health.
LORRAINE :	56-65	37 years	Female	Bungalow. Completely bedbound in a bedroom.	24-hour social care	0	Single, professional, unable to work. Had been very active academically prior to ill health.
HELEN:	16-25	6 years	Female	Lived in a house with parents and older sister. Housebound, except for occasional outing assisted by family.	Working age parents and an older sister	0	Single, education abandoned due to ill health. Notable to work or continue education due to symptoms burden.

189 3.1. Findings

190 The characteristics of the five participants are presented in Table 1. Individual circumstances
191 created very diverse presentations, despite each participant being within the severe or very severe
192 CFS/ME category.

193 Three overarching themes were identified from the initial codes. Within two of these themes,
194 sub themes were identified (See Table 2).

195 Each of these themes is now discussed in turn.

196

197

Table 2. Themes and subthemes.

Theme	Sub theme	Concepts within Sub theme
Lived experience	History and Initial Presentation	
	Impact of Illness	
Challenges to Everyday Life	Intrinsic	Physical Processing/Psychological Cognitive
	Extrinsic	
Management		

198

199 3.1.1. Theme 1: Lived Experience

200 The illness experiences of each of the five participants were very different, with one shared
 201 feature, the impact the illness had had on their lives. This overarching ‘Lived Experience’ theme
 202 explores the experience of living with severe CFS/ME and incorporates two sub-themes: history and
 203 initial presentation and the impact the illness has had on each participant.

204 3.1.2. History and Initial Presentation

205 This sub theme considers family history, comorbidities, life events, age at onset of illness, initial
 206 presentation and advice. The participants became ill at different points in their life, with different
 207 resources and burdens to manage their illness with. This led to different expressions of the illness
 208 reflecting diverse lives and values and unique personal biopsychosocial frameworks.

209 For all the individuals there was a recognised pre-existing vulnerability to becoming ill. A trigger
 210 then led to the development of multiple symptoms. This is illustrated in Table 3.

211

Table 3. Characteristics of early illness.

Pseudonym	Pre-existing vulnerability	Trigger	Initial Symptoms	Time to diagnose	Transitions of Coping strategy
Jane	Young mum, full time worker, transitioning point in life.	Complex. Viral infections, birth of son.	Myalgia, pain, viral infections.	3 years	Persisting to Acceptance
David	Never returned to pre- glandular fever energy levels. Began working full time. Contracted influenza B. Transitioning point in his life.	Influenza B.	Post viral fatigue, aches and pains.	3 months to diagnose post viral fatigue.	Avoidance ‘Head in the sand like an ostrich’.

Abi	Not listening to body.	Tic bite. Working as a gym and aerobics instructor. Just kept going.	Overly hot and sweating during routine exercise. Exhausted, muscles hurting, fatigue.	Unknown	Persisting to acceptance of situation.
Lorraine	Limited support network. Establishing herself in her new career. Transitioning point in her life	Viral.	Orthostatic intolerance, fatigue, weakness.	Unknown	Persisting and boom and busting towards acceptance.
Helen	Exam time, transitioning point in her life.	Migraines and hypersomnia	Headaches, migraines, and hypersomnia	Unknown	Degree of persisting, boom and busting due to limited experience.

212 There was also evidence that for some participants precipitating behaviours and circumstances
 213 made managing this complex cluster of symptoms, that define the illness, difficult. For several, it was
 214 an active life. For example, Lorraine became ill after completing her honours degree. She continued
 215 to struggle, living alone whilst trying to establish a new career and complete a post graduate degree.
 216 Similarly, Jane was a busy full-time working Mum of a sick baby who required regular hospital
 217 admissions.

218 For the individuals who became ill as young adults, multiple burdens were not a factor, however
 219 they had not created robust coping strategies to manage a debilitating long-term condition before the
 220 illness was triggered. As can be seen from Table 1, David and Helen had not had the opportunities
 221 to establish themselves in a workplace or higher education to gain life experience before
 222 circumstances called on them to manage their illness.

223 Two of the five participants expressed how the initial presentation of this illness was difficult to
 224 describe to healthcare professionals, as there were often multiple competing and overlapping
 225 symptoms. For example, Abi described feeling *'tired exhausted, muscles were hurting. Felt poisoned, more
 226 than ill, horrible feeling all over, like that, but I was just feeling weird and wrong'*.

227 When the illness was triggered in a transitional phase of life e.g. new jobs/careers, new parents,
 228 it was difficult to understand the cause and effect. Jane stated: *'In hindsight it wasn't a normal type of
 229 tiredness. I just didn't have the words or assertiveness to convince anyone'*.

230 The initial presentation was often vague, and the initial advice received from healthcare
 231 professionals was often to keep going, stay active and get fitter.

232 Most participants described a 'persisting' or 'boom or bust' behaviour pattern during a sustained
 233 period in which they attempted, unsuccessfully to regain their previous 'normal'. For example, Jane
 234 changed jobs, trying to get a healthier balance: *'I took a month off, when I was to get myself well, get myself
 235 fit. Doctors' advice was to exercise in the hope that, you know, start of the new job physically better, physically
 236 fitter'*.

237 Helen was given similar advice and being younger it was her Mum who directed the activity; -
 238 *'I did try taking her to the park, running around, desperately thinking she needs to exercise, she
 239 needs exercise. And then she was wiped out for days and days and days.'* Exercise intolerance
 240 appears to be present for multiple individuals, however without an objective physical cause it is
 241 difficult to identify. Each of the participants deteriorated whilst trying to get fitter or be 'normal'.

242 There appears to be a consistent pattern of boom/bust and persistence leading to deterioration,
 243 with consequent periods of bedrest to alleviate symptoms, resulting in deconditioning and limited
 244 function which can be profound.

245 At best, energy fluctuations allow individuals to experience only fleeting moments of 'normal'.
246 For example, Jane stated she was active for 2% of the day when she washed and toileted, whereas
247 Lorraine was completely bedbound and dependent on assistance just to sit up in bed.

248 For all or many of the participants, tasks such as personal hygiene and food preparation are
249 limited to basic needs. Overlaying complexities of orthostatic symptoms, allergies and sensitivities
250 all impact activities of daily living. This was demonstrated by David being limited to a bowl of
251 cornflakes on the days his ageing mother could not assist him to make a meal, which was becoming
252 more frequent. Jane reluctantly admitted she could possibly make a meal for herself twice a month
253 and at times had gone days without food when her carers had been away, she was simply too ill to
254 do any more than the essentials. Nutrition had been prioritised out of that energy calculation.

255 Sleep was also affected. Two individuals described parasomnias, sleeping 22 hours per day for
256 long periods of time or being awake but unable to move. Lorraine related her poor sleep to autonomic
257 disturbance: *'hyperadrenergic - over heating and waking hourly. Bad dreams, hallucinations'*

258 Jane expressed that sleep was beyond her control due to family circumstances, with *'Two small
259 children who can be ill and climb into bed.'*

260 3.2. Impact on Life

261 This sub-theme illustrates the impact on life including function, nourishment, sleep and social
262 isolation. Individuals acknowledge the confining effects of their illness. Some found it difficult to live
263 with meaning as this required energy and effort which they did not have. Therefore, life was a
264 passage of time, without having the resources to take action. For David, *'the biggest problem for people
265 with ME, you are in limbo, nobody knows what to do with you. What life, because it's more about filling time
266 than actually living, because living requires doing things with much effort'*.

267 However, Jane had adapted and altered her thoughts to accept her situation, prioritising her
268 energy for the activities that gave her life meaning and value, demonstrating resilience to her
269 situation:

270 *'I have all the people and things that are important in life and you know like relationships and the love
271 you have got in your family, I still have that so my life has been boiled down to the most important
272 bits that are still here. If I was going to lose things from my life, it would be my work, and yeah it
273 would be reluctantly to be able to go outside and have a life, but it's these special relationships that I
274 cherish that make me feel happy and content'*.

275 3.2.1. Theme 2: Challenges to Everyday life

276 This overarching theme comprises of two sub themes: Intrinsic – those elements that were
277 fundamental to the individual's physiology or psychology and extrinsic - those elements that
278 operated from outside the individual.

279 3.2.2. Intrinsic Concepts

280 Within this sub-theme were three distinct concepts. The physical concept included the effect of
281 activity, adrenaline response/orthostatic intolerance, allergies and sensitivities and baseline level of
282 activity/unpredictable fluctuations. Processing/psychological concept encompassed belief about
283 cure, personal views, views about management, communication and energy balance. Finally,
284 concepts comprising cognitive impairment, dissociation/detachment, and mental health.

285 Each individual's ability to function was limited to their personal capacity, which fluctuated
286 extremely and created difficulties to plan and manage basic day to day activities such as washing,
287 toileting and in some instances feeding, particularly with the added symptom of allergies and
288 sensitivities. For example, Lorraine said, *'digesting food is an energy challenge'*, and Abi found, *'In the food
289 department (allergies), the big one is nickel- which limits food diversity'*.

290 It appears that when baseline energy levels are so low and capacity is so limited, there is a
291 frequent tendency to experience the challenge of everyday activities as a physiological threat.

292 Participants often access the sympathetic nervous system just to carry out basic functions such as
293 personal hygiene and eating. It seems this often leads to dysautonomia, a disruption of the autonomic
294 nervous system that regulates the heart, blood vessels, digestion and breathing. This disruption could
295 be due to activity and/or orthostatic challenge. For example, Jane said, *'I am in bed 100% of the time.*
296 *When active, pain feels worse, feel weak, feeling hot, cold, fevery, kind of heart palpitation, breathlessness,*
297 *shaky'.*

298 The management of physiological and physical limitations was often bound by thoughts and
299 perceptions of the participant's experience, which was in turn often limited by their condition. The
300 ability to process and communicate beliefs about their situation were limited by their energy levels.
301 Lorraine said:

302 *'You get to a point in a relationship where you actually need to say 'this is what is going on with my*
303 *illness....' and then you have to eat and we never get a chance, there's no time for conversation. All*
304 *my emotions are around, are set to one side, all the loss, all the bereavement, loss of self, loss of life,*
305 *loss of opportunity, loss of the living, so family relationships are just set to one side, there isn't time*
306 *to process those emotions to ever have them'.*

307 The processing ability appeared to be further impacted by cognitive impairments. Individuals
308 recognised this, however the strategies they put in place created an increased energy expenditure.

309 Compounding these challenges was the concerning presence of dissociation and detachment,
310 expressed by four of the five participants, along with, in some cases, loss of identity. For example:
311 *'I feel detached and confused. I suffer from disconnectedness, so I don't feel physically present - don't*
312 *concentrate on that- having a blister and gritting your teeth. I don't know, I can't remember' (Lorraine).* *'There*
313 *is numbness most of the time, a contentedness. Then the rest of the time- frustration, disappointment, fear. My*
314 *body being unresponsive. Just staring into space. I don't feel myself anymore, sense of self or identity... I have*
315 *accepted, peaceful, but I struggle with I don't feel like myself anymore' (Jane).*

316 3.3.3. Extrinsic Concepts

317 This sub-theme relates to the influences outside of the individual's control that harm their ability
318 to function in the world given their illness. Such influences include the benefits system, lack of
319 professional understanding and prejudicial views.

320 Most social challenges came from a lack of understanding within society as to the nature of
321 CFS/ME or the extent to which it can impact. This lack of understanding can lead to prejudicial views
322 and preconceptions that impede an individual's access to healthcare and social services. Abi for
323 example conveyed an experience she had had in an accident and emergency department, having been
324 taken from her Health Centre to Hospital in an ambulance. Once she arrived, the attitude of the staff
325 changed when they realised, she was a frequent visitor *'I passed out. I woke up and was being dragged*
326 *along the floor by two orderlies and this nurse screaming at me'.*

327 Abi received the care she required once her tachycardia was identified.

328 Lorraine, too, recognised this engrained culture. *'It takes years to erode institutionalised discrimination*
329 *and prejudice which are unhealthy and negative for both victim and perpetrator'.*

330 Health and social care require a diagnosis to be current in order for an individual to be eligible
331 for social support. This support is binary: there is no grading within the system. This creates a
332 difficulty when there is not a biomedical marker to identify the illness. The repeated cycle of having
333 to demonstrate ill-health impacts an individual's ability to manage and improve from that health
334 issue. It also undermines continuity of recognition of that ongoing issue. For example, Lorraine said *'I*
335 *lack a current diagnosis so I can't get my benefits. They keep saying no current diagnosis'.* Similarly, for
336 David, *'After probably a year to a year and a half of having ME, I was improving, doing much better, but then*
337 *the benefits agency reviewed and decided I was fit for work. I lasted 6 months working as admin in a restaurant*
338 *before I crumbled.....[following repeated appeals the benefits withdrawal decision was repealed]. I should never*
339 *have been taken off (benefits) in the first place'.*

340 3.3.4. Theme 3: Management of the Condition

341 This theme incorporates managing and coping strategies, relief from symptoms, understanding
342 acceptance, acceptance of professional lack of understanding, social media, GP attitudes and healthy
343 carer beliefs.

344 The five individuals interviewed managed their condition within the confines of the means at
345 their disposal, both intrinsic and extrinsic. Their strategies were founded on a subjective
346 understanding of their capabilities, their illness and their resources. Participants relied on support
347 from family members, social media and the internet to gain information they required. Social media
348 was used to maintain contact with friends. For example, Abi used 'Twitter with friends' and Helen
349 used the internet as she transitions from sleep to wakefulness, 'In bed 30 mins am on phone waking up,
350 googling'.

351 Whilst the internet alleviates the social isolation, it can distil and reinforce beliefs. For example,
352 Abi reported, 'I have never been to a CFS clinic and I am glad, because I won't want to do GET
353 (Graded Exercise Therapy). I know from experience, physically pushing past what you feel, it made
354 me worse, so I wouldn't want to entertain that'.

355 In terms of social support, Helen described how she was reliant on the availability of her
356 working parents to take her out to socialise. However, their availability also had to coincide with
357 when she was well enough. Helen's parents appeared to have had sacrificed their social life to prevent
358 her from being left alone and isolated. Her family appeared to plan their lives around her illness, as
359 far as they could, whilst also maintaining their income. The focussed pressure on caregivers was
360 substantiated by Jane, who explained it was her husband, the income provider, who worked full time,
361 was the main caregiver to two primary aged children, and was also her care giver. Therefore, it was
362 not only the CFS/ME participants who had reduced their lives to necessary priorities, but also their
363 caregivers and families.

364 In addition to social support, healthcare professionals were acknowledged as people who could
365 help.

366 Abi reported the occasional doctor who understands, whilst Helen and her mother were very
367 keen to praise their current GP as 'fabulous' because they understood Helen's situation. This relieved
368 a lot of stress. Lorraine found that individuals who were not fixed in their beliefs of the condition
369 were most beneficial: 'People who are genuine, non-judgemental and open minded' (Lorraine).

370 This often left a very narrow path to navigate. This in turn may limit the potential for an
371 individual with CFS/ME to improve. For example, Abi stated 'I have learnt it is pointless, some people
372 won't listen and there is no point. I have tried to fight back and been called neurotic. I was really
373 frustrated, not neurotic'. Similarly, for David reported:

374 'Resignation - it's not a great surprise after all these years. You hear about how much progress
375 they make with this and that and the other and you think yeah, but there is obviously an
376 awful lot of conditions where nothing changes for decades'.

377 4. Discussion

378 Our first aim was to explore the personal experience and understanding of individuals with
379 CFS/ME. This was achieved through open questioning and exploration of the participant's views of
380 their reality.

381 The second aim was to identify overarching themes that may help identify risk factors that place
382 people at greater threat of experiencing the more severe presentation of CFS/ME. Many of the
383 participants demonstrated previously identified risk factors for expressing the severe form of
384 CFS/ME: a delayed diagnosis[22, 28]; problems accessing social security[22, 40] and poor
385 relationships with doctors or health professionals[14, 22, 23].

386 This small qualitative study has identified other common factors, which need further research
387 to clarify and confirm. For example, demonstration of deterioration as the individual initially
388 attempted to get fitter or remain 'normal'. This may be an indication of an unidentified exercise
389 intolerance. It appears that the point in a person's life when the illness presents is of importance.
390 Several participants were moving from one phase of life to another. For example, school exam time,

391 moving from school or higher education to work life, or following the birth of a child. It appears the
392 severe presentation may manifest when the illness coincides with a transitioning time in a person's
393 life. Another common factor is the relationship between burden and resource. Those with dependents
394 or many responsibilities and a limited support network appear to be more vulnerable to the severe
395 expression of the illness. Or those individuals who had a support network, but remained to some
396 degree dependent on carers, not able to establish independence due to the illness.

397 The final aim of this study was to provide a better understanding of this population to allow a
398 research community to engage with them more effectively. This has been addressed to an extent by
399 Kingdon [14]. We have taken a phenomenological approach to report the lived experience of five
400 individuals with CFS/ME. These findings cannot be generalised, however it is possible that they are
401 transferable to other individuals in a similar position. Here will expand on how these findings may
402 be applied to the evolving understanding.

403 All five of the participants had vague initial presentations that they found difficult to explain,
404 illustrating the experience of living with poorly understood illness. Despite a fatigue presentation
405 they were actively encouraged to keep going and push through or had themselves tried to regain
406 their former life. Maladaptive sickness patterns have been recognised in chronic illness [41] and the
407 recommendation of exercise in the presence of fatigue is increasingly acknowledged as detrimental.
408 Inappropriate advice may promote unhealthy pacing behaviours of 'boom and bust' and persistence
409 [42]. It is suggested that this ultimately leads to deconditioning through the over training exercise
410 curve, which is recognised in athletes but remains an under researched area[43]. Fatigue self-efficacy
411 improves outcomes [41], however the confidence to self-manage fatigue must be fostered gradually
412 and immediately if unhealthy adaptive behaviours are to be avoided.

413 The timing of the illness appears to have importance. When illness occurs at a young age, school
414 attendance is reduced, seriously affecting intellectual and social development[16, 44]. This is
415 illustrated by Helen and to some extent David, who continued to be dependent on their parents, their
416 carers into adulthood. This combination can further impact managing this complex illness [15].
417 Another critical factor is if the illness occurs during a transition, e.g. from professional to working
418 mother, when tiredness is expected. This may make diagnosis more difficult: transitioning life stages
419 produce confounding factors that confuse a biomedical assessment.

420 All the participants followed a deteriorating pattern. It appears there comes a point when
421 burdens exceed resources and the opportunity to improve is extinguished. People then experience
422 physiological threats, resulting fight or flight reactions or dissociative responses. Dissociation is
423 described within poly vagal theory as losing a sense of presence resulting in experiencing a
424 disconnection and a lack of continuity between thoughts, memories, surroundings and actions[45].

425 It is concerning that at least two of the participants reported symptoms of dissociation. It is
426 suspected in the three others. Benign aspects of existence were experienced as threats so extreme that
427 the ability to be present was lost. Acceptance has been identified as a precursor for any therapeutic
428 intervention to succeed. However, it is proposed that for acceptance to occur, a person must feel safe
429 and present within their physical environment. This has significant implications for management
430 and rehabilitation.

431 Intrinsic challenges to everyday life are further compounded by the extrinsic burdens. All five
432 of the participants reported poor or limited interactions with healthcare professionals during their
433 illness. Negative attitudes towards CFS/ME by medical professionals are repeatedly reported[23, 46].
434 After many attempts at trying and failing to navigate the health and social care systems, with an
435 imbalance of energy, resources and burdens, some individuals experiencing CFS/ME eventually
436 appear to accept their limitations and that of their health professionals.

437 All of the participants were forced to give up education or employment. In work and educational
438 institutions, the lack of understanding and provision for people with CFS/ME creates obstacles for
439 people with the illness to remain in those environments. CBT and GET do not restore the ability of a
440 person with CFS/ME to work [47]. People with CFS/ME who cannot remain in employment need to
441 access the benefits system. As CFS/ME does not sit within the current biomedical model of health and
442 social care, this creates issues navigating the benefits system. This was reported by two of the five

443 participants. It has been recorded that the benefits system in the UK does not meet the needs of people
444 with CFS/ME leaving them socially isolated and/or increasingly dependent on friends and family.
445 The distress of navigating the system often exacerbates health conditions[40]

446 The five participants managed their condition as well as resources allowed, both intrinsic and
447 extrinsic. At times this unfortunately meant accepting the limits of the system in which they found
448 themselves.

449 Four of the five study participants had received specialist support during their illness. However,
450 this support was not always valued. One participant was receiving support at the time of the study.
451 All the participants presented with complex multi-faceted issues that impacted every component of
452 the biopsychosocial model. Their ability had declined to the extent where it impacted every aspect of
453 their life: physical function, diet, sleep and social interaction.

454 The participants were heavily reliant on the internet to source management strategies. This
455 often-distilled illness beliefs. Health literacy has been shown to be a challenge in vulnerable
456 groups[48]. However, we do not understand how severely affected CFS/ME individuals use health
457 literature because they are so under researched.

458 It appears that severely affected CFS/ME individuals must lead a very disciplined and limited
459 existence in order to manage symptom burden within their intrinsic and extrinsic limitations. It is an
460 open question as to whether such limits impact their ability to be psychologically flexible and resilient
461 in their outlook.

462 5. Conclusions

463 This study is novel as it has accessed this hard to reach population group and recorded their
464 experience. Most of the participants had received some form of specialist CFS/ME support or had
465 access to the healthcare services. However, their experiences ranged from accepting the limitations
466 of the service to having a very negative view.

467 CFS/ME is a medically unexplained illness lying at the boundaries of understanding within the
468 legacy biomedical model. An illness where there is no single, simple cause or theoretical model, no
469 clear mind/body division and no definitive classification[1] does not sit easily in the current
470 healthcare system. The CFS/ME presentation conflicts with the current health and social care model
471 [1, 2]. The severe CFS/ME presentation sits outside the model and therefore is not acknowledged.

472 This illness ranks low within primary care as it is not life threatening [23]. It is however,
473 potentially life shortening[14]. There are certainly physical and mental health symptoms which are
474 often disregarded or missed within the complex presentation[14] and reports suggest that 88 suicides
475 have been partly attributed to CFS/ME between 2001 and 2016. However, it has been noted that it is
476 not necessarily intrinsic factors that lead to suicide, but a combination of extrinsic factors, which
477 include a lack of medical care and social support, failure to control key symptoms and inadequate
478 financial help. Depression is not always a feature in CFS/ME related suicide[49].

479 Pathway focused institutional cultures are not predisposed to embrace the ambiguities inherent
480 in adopting the more holistic biopsychosocial model, where outcomes are more difficult to define
481 and evaluate. The resulting continued narrow biomedical focus of the current social care system
482 results in neither the healthcare professional nor the CFS/ME patient feeling safe with each coming
483 from a position of defence when they communicate[23, 27, 45]. People with the severe expression of
484 CFS/ME appear to avoid the harm of the current health and social care system by purposely
485 withdrawing from it. This reduces opportunities for rehabilitation and is an area for further study.

486 Individuals with severe CFS/ME live on the peripheries of society, at the edges of the research
487 bell curve[50]. They do not belong within 'normal' expectations and they do not have the energy to
488 try to fit[51], therefore they remain socially, medically and financially isolated. The role of
489 environment has been discussed within the international classification of function. Disability has
490 been acknowledged as a socially created problem which can limit freedom by failure to provide the
491 resources and opportunities needed to make participation feasible[52]. This paradigm must be
492 explored further if we are to better understand and provide adequate health and social care for the

493 severe CFS/ME population or other people experiencing ‘illness’ that does not fall into the biomedical
494 model.

495 The findings of this study aim to assist understanding of the needs of the Severe CFS/ME
496 population. Currently, the healthcare system and research community is failing to provide resources
497 and opportunities for this group to engage, and so enable the positive outcome of increased
498 independence. Longer periods of intervention, home visits and telephone consultations and in
499 extreme cases inpatient rehabilitation in specialist services are effective evidenced interventions in
500 the research literature [10, 16, 44, 53] . Such services would meet the needs of CFS/ME individuals,
501 much better than the status quo which often forces patients to meet the needs of the system in order
502 to secure the care that they need.

503 A re-evaluation of the approach taken to CFS/ME and other unexplained illness is ever more
504 urgent given the upcoming surge in numbers of long haul COVID-19 individuals. A major symptom
505 of such long haul COVID is fatigue[54, 55]. Research and healthcare communities have much
506 experience to share and further research to perform, particularly in the area of health, social care and
507 societal attitudes allowing vulnerable ill people to remain valued members of society.

508
509

510 **Limitations**

511 The thematic analysis aspect of this research studies a small number of participants in depth, giving
512 a rich presentation. The participants were from a small geographical area and may not be
513 representative of the wider CFS/ME community.

514 It is recommended that further research is conducted with a larger sample of participants across
515 a wider geographical area of the United Kingdom. Adequate financial and time provision must be
516 allocated to allow severe and very severe CFS/ME individuals to engage in future projects. Part of
517 future research regarding CFS/ME must explore the wider biopsychosocial factors that lead to the
518 severe expressions of fatigue. The goal being to identify risk factors that affect the deterioration of the
519 condition within different life phases and aid earlier detection of those at risk of the severe and very
520 severe expression of CFS/ME and adequate provision of healthcare.

521

522 **Author Contributions:** Conceptualization, J.L.N.; V.S. and T.C.; methodology, T.C. and V.S. validation, V.S. and
523 T.C.; formal analysis, V.S. and T.C.; investigation, J.L.N.; V.S. and T.C.; resources, X.X.; data curation, V.S. and
524 T.C.; writing—original draft preparation, VS.; writing—review and editing, VS, TC and JLN.; visualization, X.X.;
525 supervision, TC and JLN.; project administration, VS.; funding acquisition, VS.

526 **Funding** This study was supported by ME Research UK (SCIO charity number SC036942).

527 **Acknowledgments:** We thank ME Research UK for funding the study that allowed this pilot project. Researcher
528 and Health Psychologist Practitioner, Professor Vincent Deary for his guidance and insights into the
529 presentations of some of the participants. We would also like to thank ME North East, all the local support
530 groups and in particular, the participants who contributed to this study.

531 **Conflicts of Interest:** The authors declare no conflict of interest.

532 **Publisher’s Note:** MDPI stays neutral with regard to jurisdictional claims in published maps and institutional
533 affiliations.



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