Relational and psychological aspects of the experience of chronic fatigue syndrome for patients and their significant others: A grounded theory study

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1.1. Abstract

Social processes have been investigated as a maintaining factor in chronic fatigue syndrome, also known as myalgic encephamyelitis (CFS/ME), and the responses of the significant others (SOs) of people with CFS/ME (PwCFS/ME) have been shown to be associated with illness outcomes. Experiences of PwCFS/ME and their significant others have been explored, but with limited depth and/or breadth. This study aimed to develop a nuanced understanding of relationships between PwCFS/ME and their SOs, describing emotional and relational characteristics, processes and consequences. 15 participants consisting of PwCFS/ME (*n*=9) and SOs of PwCFS/ME (*n*=6) completed a total of 26 audio-recorded, semi-structured interviews which were analysed according to grounded theory methodology. Four categories are presented: *Onset, The Problem Pattern, Cohesion and Acceptance,* with *The Problem Pattern* being the core category. Within these, patterns of family functioning are identified and discussed in relation to family systems theory and models of family cohesion. Three key dimensions of cohesion are identified in the data: *disengagement, enmeshment,* and *healthy cohesion*, as are the emotional experiences that accompany these relational patterns. Future research and interventions that incorporate the significant other should aim to improve togetherness, autonomy, and acceptance in relationships, with the goal of improving quality of life within the limitations of the illness.

Keywords: chronic fatigue syndrome, significant other, relational processes, grounded theory, qualitative, family cohesion, acceptance.

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1.3. Contents

2. Introduction

Research suggests that people with chronic fatigue syndrome/myalgic encephalomyelitis (referred to throughout as CFS/ME and as PwCFS/ME meaning a person or people with CFS/ME) are affected by the responses of their significant others (SO). There is evidence that the similar constructs of ‘solicitous responses’ and ‘emotional overinvolvement’ have a detrimental impact on the long-term health outcomes of PwCFS/ME, as do ‘negative responses’ and ‘critical comments’ (Band, Barrowclough, & Wearden, 2014; Kerns & Rosenberg, 1995; Romano, Jensen, Schmaling, Hops, & Buchwald, 2009; Verspaandonk, Coenders, Bleijenberg, Lobbestael, & Knoop, 2015). These constructs will be defined and discussed in the literature and referred back to frequently throughout the thesis. There is a lack, however, of evidence around the relational patterns and emotions that give rise to and are a consequence of these responses. There is also limited discussion in the literature around what more constructive responses or relational patterns might look like, and what PwCFS/ME and their SOs may be able to do to improve their responses.

This grounded theory study offers an exploration of the relational patterns experienced by people who self-reported living with CFS/ME and their SOs. I argue here that relationships in the context of CFS/ME exist on a continuum of family cohesion, with disengagement and enmeshment at the two extremes. Both extremes lead to the same set of consequences: social isolation, feelings of anger and frustration and/or guilt. Participant accounts illustrate some of the ways PwCFS/ME and their SOs were able to find ways forwards and move towards improvements, and we see that more balance in family systems is necessary. Ultimately, the most desired and helpful approach to life and illness for the dyad emerges as that of acceptance.

The aim of grounded theory is to account for behaviour patterns by generating a theory, ensuring that the theory is relevant and useful for the subjects of the theory

(Glaser, 1978). The method was developed by Glaser & Strauss (1967) with the aim of providing a method for constructing new theory instead of testing ideas that have been preconceived. This meant that theory should be grounded in empirical data and not in existing theory and literature, and thus the early recommendation was that a researcher should not engage with literature before collecting data. It was asserted that concepts should be allowed to ‘emerge’ in the data and that ideas from existing literature should not be allowed to contaminate the research product, with the researcher remaining objective and thus separate from the research data.

Later development of grounded theory methods such as by Strauss & Corbin (1990) recognised that researchers bring professional and personal experience to their research. They recommended that researchers should be permitted to engage with literature at all stages of the research process within grounded theory method, provided that they remained mindful of not imposing upon their emergent theory. The idea that a researcher should be separate from the data was maintained. However, in constructivist grounded theory, put forward by Charmaz (2014), there is recognition that it is not possible to avoid influence from the researcher, and that the data generated is the result of an interactional process and shared experience, whilst theory is a product of the researcher’s perspective. Therefore, a more pragmatic approach to literature is recommended in constructivist grounded theory. Charmaz recognised that it is impractical in most cases for a researcher not to engage with literature at all prior to undertaking research. Indeed, ethics committees, PhD procedures, and funders require evidence that there is a need for a particular piece of research, and this evidence is found in the literature. A researcher would usually engage with literature in order to refine the research question and aims, but according to Charmaz they should not allow themselves to be directed by previous studies or theories in their interpretations of the research data. Instead, they should follow a process of constant comparison between sets of data, constructed concepts, and allow the data to direct the exploration of existing literature.

Therefore, in accordance with constructivist grounded theory, I have opted to present this thesis in a chronological sense with regards to the way in which I engaged with the literature. The literature reviews in Chapter 3 and Chapter 4 offer a historical overview of CFS/ME and a scoping literature review respectively. These chapters take the reader up to the point of data collection. The subsequent literature and theories with which I engaged are presented as integrated discussion alongside the results, as this enabled me to illustrate the data that led me to explore certain concepts and the related literature.

The use of labels in CFS/ME is a subject of much controversy (Jason, Taylor, Plioplys, Stepanek, & Shlaes, 2002; Jason, Taylor, Stepanek, & Plioplys, 2001; Picariello, Ali, Moss-Morris, & Chalder, 2015). It has become common practice in research and clinical practice for CFS/ME to be used (Baker & Shaw, 2007; Chew-Graham, Dowrick, Wearden, Richardson, & Peters, 2010; NICE, 2007), and I therefore refer to CFS/ME throughout. Pseudonyms are also used throughout the document and personal details are omitted in order to protect the confidentiality of the research participants.

3. History of CFS/ME

3.1. Overview of CFS/ME

CFS/ME has a complex history and many other names by which the same set of symptoms have previously been known. In order to make sense of the history and many complexities of this illness, it is first necessary to understand aspects of its nature. From that point, we then explore the history of CFS/ME and how it came to be understood today. Whilst there are as many as 20 case definitions of CFS/ME in use (Brurberg, Fønhus, Larun, Flottorp, & Malterud, 2014), the most frequently used and cited is that developed for the Centers for Disease Control and Prevention (CDC) by Fukuda et al. (1994) (Brurberg et al. 2014). Therefore, I refer the Fukuda et al. (1994) case definition as the authoritative source throughout this thesis regarding diagnostic criteria.

3.1.1. Symptoms. The most prominent feature of CFS/ME is severe and persistent fatigue that is debilitating and not alleviated by sleep or rest. This is usually accompanied by a combination of other symptoms including musculoskeletal pain, and/or difficulty sleeping, post-exertional malaise, painful or swollen lymph nodes, and cognitive dysfunction such as reduced concentration and memory (Fukuda et al., 1994).

Many PwCFS/ME describe their experience of CFS/ME as being like a very bad case of influenza that will not go away (Moss-Morris & Petrie, 2000). The combination and severity of symptoms varies from person to person, and some or all of the aforementioned symptoms may be present (Fukuda et al., 1994). In addition, many PwCFS/ME experience psychiatric illness such as anxiety or depression whilst also suffering with CFS/ME (Cella, White, Sharpe, & Chalder, 2013). However, it is common for PwCFS/ME to point out that their physical symptoms emerged first; many PwCFS/ME argue that their subsequent depressive and/or anxiety symptoms are reactive and experienced in response to the life-limiting disabilities that CFS/ME has caused them (Surawy, Hackmann, Hawton, & Sharpe, 1995).

3.1.2. Prevalence. Studies investigating the prevalence rate of CFS/ME in the general population in the early 21st century estimated it to be between 0.2% and 0.41%, with some variance explained by the differing criteria used for diagnosis (Afari & Buchwald, 2003; Reid et al., 2000; Reyes et al., 2003). There has been no difference in prevalence found between people of different socioeconomic status or ethnic groups in systematic population surveys; however, females are at greater risk of developing the illness than males (Reid et al., 2000). Global prevalence is estimated at 0.6%, according to the Oxford criteria (NHS Centre for Reviews and Dissemination, 2002). The most common age of onset ranges from 20 to 40 years of age, but people of all ages may be diagnosed with CFS/ME (Engleberg, 2002).

3.1.3. Definitions and diagnosis. Writing an exhaustive historical overview of CFS/ME is a challenging task, not least because it has been linked to so many other medical illnesses and labels, and to a certain extent this continues to be the case today. A major problem in defining CFS/ME is the lack of certainty around the cause, how it is acquired, and its pathophysiology. As Hyde states (1992), 'if you know the exact cause, the method of acquiring and the pathophysiology of a disease, one does not need a definition...if we have a definitive test for a specific disease, there is little need for a definition' (p.5-6). In considering its definition, it is useful to be aware of as full a range of names as possible by which the illness or illnesses closely resembling it have been known. This helps us make sense of the many avenues that have been explored in research into CFS/ME. It also separates the variety of names into meaningful categories, offering a framework for considering the various aspects and nature of this illness. Hyde et al. (1992) present a compilation of the full range of names. An adaptation of this is available in

Table 1. Names for CFS/ME, with some further additional names added that have been encountered.

Table 1. *Names for CFS/ME*

Neuromyasthenia names	Neuromyasthenia, Neurasthenia, Epidemic neuromyasthenia, Epidemic pseudo myasthenia, Sporadic postinfectious neuromyasthenia, Neurocirculatory asthenia.
Names based on location of epidemic outbreak	Iceland disease, Akureyri disease, Coventry disease, Tapanui flu, Otago mystery disease, Royal Free disease, Lake Tahoe mystery disease, Lyndonville chronic mononucleosis, the English disease;
Myalgic Encephalomyelitis names	Myalgic encephalomyelitis (M.E.), benign encephalomyelitis, benign myalgic encephalomyelitis, benign subacute encephalomyelitis, epidemic myalgic encephalomyelitis or encephalomyelopathy, acute infective encephalomyelitis, epidemic diencephalomyelitis, lymphoreticular encephalomyelopathy;
Myalgia names	Epidemic malaise, persistent myalgia following sore throat, Damadian's ache, Myofascial syndrome, Muscular rheaumatism, Fibromyalgia syndrome, Fibromyositis, Fibrositis, Epidemic myositis, Lymphocytic meningo encephalitis with myalgia and rash, Syndrome polyalgique idiopathique diffuse (S.P.I.D.);
Personal names	Da Costa's Syndrome, Beard's disease;
Symptom-based names	Chronic fatigue syndrome (CFS), La Spasmophilie (France), Raggedy Ann Syndrome, the English sweats, Effort syndrome, Tetanie chronic idiopathique;
Bacterial names	Chronic brucellosis, Chronic lyme disease;
Combined virus/symptom names	Post-viral fatigue syndrome (PVFS), Persistent viral fatigue syndrome.
Immune-based names	Chronic immune activation syndrome (CAIS), Chronic immune dysfunction syndrome (CIDS), Low natural killer cell syndrome (Japan), Multiple chemical sensitivity syndrome, Ecological disease, Allergic fatigue syndrome, Antibody negative lupus, Antibody negative lyme disease, Chronic activated immune dysfunction syndrome (CAIDS), Chronic fatigue and immune dysfunction syndrome (CFIDS), Naxalone-reversible monocyte dysfunction syndrome (NRMDS);
Epstein-Barr Virus based names	Chronic Epstein-Barr virus syndrome (CEBV), Chronic active Epstein-Barr virus infection (CAEBV), Virus epidemic in recurrent waves, Chronic mononucleosis, Familial chronic mononucleosis, Chronic infectious mononucleosis, chronic active Epstein-Barr virus infection, Chronic mononucleosis-like syndrome;
Hypothalamic names	Epidemic vegetative neuritis, Neurocirculatory asthenia, vasoregulatory asthenia, vasomotor instability, vasomotor neurosis, Habitual chronic hyperventilation syndrome;
Poliomyelitis names	A disease resembling or simulating poliomyelitis, atypical poliomyelitis, abortive poliomyelitis, encephalitis simulating poliomyelitis, encephalitis resembling poliomyelitis, postpolio syndrome, posterior poliomyelitis, sensory poliomyelitis.
War-based names	Gulf war syndrome
The Atypical names	Atypical multiple sclerosis, Atypical migraine;
Media names	Yuppie flu, Yuppie plague;
Miscellaneous names	Soldier's heart, Epidemic vasculitis syndrome.

Note. Adapted from Hyde, Goldstein & Levine, 1992, p.3

I have referred throughout this thesis to CFS/ME as an illness because that is the term participants used to communicate their conceptualisation of what it is, but it should be noted that Hyde (2003) argues that CFS/ME is a state of chronic fatigue rather than a disease or illness. Although as many as 20 case definitions have been found to exist (Brurberg et al., 2014), the authors of the four key published definitions have repeatedly made clear that their definition is of a syndrome (collection of symptoms) and not a specific disease or illness (Hyde, 2003). In spite of this, there is much confusion among PwCFS/ME, doctors, and in the USA among insurers, who misunderstand it to be a disease with a specific set of treatments and outcomes (Hyde, 2003). The four most prominent published definitions are as follows:

- 1988 - CDC Criteria (Holmes et al., 1988)
- 1990 - Lloyd, Hickie, Broughton, Spencer and Wakefield created a definition widely used in Australia. (Lloyd et al, 1990; Reid et al., 2000)
- 1991 – Oxford Criteria (Sharpe et al., 1991)
- 1994 - NIH/CDC definitions - a copy of the first definition with a few modifications (Fukuda et al., 1994)

In England, the National Institute for Health and Clinical Excellence (NICE) Guidelines (NICE, 2007) are used by medical practitioners to determine a diagnosis and make treatment decisions. However, as previously noted, Brurberg, Fohnus, Larun, Flottorp & Malterud (2014) concluded that the most frequently applied case definition in CFS/ME research studies is the CDC-1994/Fukuda (Fukuda et al. 1994). Interestingly, recommendations in the NICE guidelines are that it should be diagnosed within 4 months of onset in adults and 3 months in children (NICE, 2007), rather than the six months specified in earlier criteria (Fukuda et al, 1994).

3.2. Pre-1900 history

3.2.1. Febricula. Straus (1991) reported that one of the earliest known reports of a CFS/ME-like illness was published in 1750 by Sir Richard Manningham. According to Manningham's definition of febricula, the symptoms included "little low, continued fever...little transient chilliness...listlessness with great lassitude and weariness all over the body...little flying pains...sometimes the patient is a little delirious and forgetful" (Manningham, 1750, p. 52-3, as cited in Straus, 1991). Manningham also highlighted similarities between febricula and the symptoms of illnesses described by Hippocrates and others, as well as discussing its apparent prevalence among sedentary academic-types and women from wealthy families (as cited in Straus, 1991). Straus outlined the precipitating factors as Manningham saw them to include grief, viral infection such as a cold, and over-thinking, whilst diagnoses applied to a similar set of symptoms included "hypo, spleen, fever on the spirits, nervous fever, hysteric fever, and the vapours" (p. 52).

3.2.2. Florence Nightingale. Florence Nightingale (1820-1910), an Englishwoman credited as being the founder of modern nursing and reformer of hospitals, has been posthumously involved in the naming controversy for CFS/ME due to evidence of her having suffered from a debilitating illness that went undiagnosed following her return home from the Crimean War (Young, 1995). Some claim her symptoms were similar to those of CFS/ME; consequently, groups of PwCFS/ME have adopted her birthday on 12th May as International CFS/ME Awareness Day (Jason et al., 2002). However, closer inspection of Nightingale's reported symptoms calls into question the possibility of her suffering from CFS/ME. Nightingale believed her illness was typhus (Nightingale & Baly, 1991), whilst Cope (1958, as cited in Young, 1995) and Pickering (1974, as cited in Young, 1995) maintained that her illness was not caused by organic disease and favoured a diagnosis of psychoneurosis. Her official biographer, Edward Cook, wrote that even though she ‘worked like Hercules’ she had heart dilation and neurasthenia (Cook, p.446, as cited in Baly, 1996). More recently however, Young (1995) and Dossey (2010) argued the case for a diagnosis of chronic brucellosis. In a letter in the BMJ (Baly, 1996), Hyde is quoted as stating his belief that it ‘was the basis of Miss Nightingale's invalidism’. However, Baly goes on to comment that the sheer volume of work Nightingale produced whilst ill made it unlikely that she in fact had CFS/ME. It is also noted that there is strong evidence to support the claim that Nightingale exaggerated her illness and the letter concludes with a statement that her illness was ‘multifaceted and compounded by the advice and treatment of the day’ (Baly, 1996).

3.2.3. Neurasthenia. The late 1800s saw debate amongst physicians over nervous asthenia, which was later called neurasthenia. In 1868, Prof. Austin Flint (Flint, 1868) outlined the condition nervous asthenia, a name coined by his colleague Prof. Fordyce Barker. Flint noted that this debilitating condition of the nervous system was not known to be related to anaemia or a disorder of vital functions. He concluded that it was the result of over-activity of the nervous system following intellectual or emotional challenges. Investigation of the main body organs found no evidence of disease and people frequently presented with no well-defined ailment, yet, Flint acknowledged, the morbid condition clearly existed. His description of the symptoms, ‘languor, lassitude, want of buoyancy, aching of the limbs, and mental depression...wakeful during the night...’ echo those of the current diagnostic criteria for CFS, as does Flint's description of the post-exertional malaise experienced by people presenting with this condition (Flint, 1868, p.739).

Flint's recommendations for the treatment of nervous asthenia were not dissimilar to the approach taken today by western medicine, discussed later; initial, temporary and complete rest from all responsibilities was suggested, but only for long enough to give the nervous system time to recuperate. Finally, Flint emphasised the importance of a return to activity, including mental exercise, but in moderation, and of taking care of the body through careful management of the diet, wake-sleep cycle, outdoor pursuits, bathing, tonics, and limited use of tobacco, which is similar to modern treatments such as graded exercise therapy and adaptive pacing therapy.

The following year, the American neurologist Brian Beard (1869) wrote of neurasthenia, a ‘condition of the system’ that he viewed as ‘exhaustion of the nervous system’ and the most frequent ‘cause and effect of disease’ of his time (p.217). The term, which he derived from Greek, can be literally interpreted as meaning ‘want of strength in the nerve’. Beard acknowledged that although the term was new and had been developed for the purpose of raising the profile of nervous exhaustion among the medical community, the symptomology had begun to be recognised under other terms previously, including what Flint described in 1868 as nervous asthenia. Beard drew comparisons with anaemia, suggesting parallels in some symptoms such as insomnia, headaches and neuralgia and also noted that both anaemia and neurasthenia were of greater prevalence in well-educated, civilised communities, ‘part of the compensation for our progress and refinement’ (p. 217). Much like today, some doctors recommended rest whilst others advocated exercise for people diagnosed with neurasthenia (Beard, 1869). Beard's works on neurasthenia were welcomed by the middle classes of America and Europe and respected in the developing field of psychiatry. His concepts endured until the 1930s, when neurasthenia came to be thought of as indistinguishable from affective illness and was no longer diagnosed as a condition in its own right (Straus, 1991).

3.3. 20th century history

3.3.1. Post-polio syndrome. Postpolio syndrome occurs among 25-80% of people who suffer an acute poliomyelitis attack early in life (Hyde et al., 1992). The symptoms of post-polio syndrome are identical to those listed in the diagnostic criteria for CFS/ME, but it is considered by some to be a separate illness from CFS/ME because it is seen as having a more progressive nature. However, some researchers argue that postpolio syndrome is essentially CFS/ME occurring in an individual who has previously survived poliomyelitis (Hyde et al., 1992).

3.3.2. Epidemic outbreaks. In the years following the eradication of neurasthenia as a diagnosis, chronic fatigue had all but disappeared from the radar of the medical profession; not having a name or known aetiology, it was omitted from medical text books and ignored in research (Ware, 1992). However, Wessely (1997) notes that fatigue syndromes and fatigued people must still have existed during this time, and that a few papers describing fatigue-like symptoms did emerge. Certainly, there were epidemic outbreaks of fatigue-like illnesses, the earliest recorded case in 1934 being among doctors and nurses in a number of hospitals in Los Angeles, USA (Jenkins, 1991). This was reported as an incidence of poliomyelitis, the symptoms of which included cramps, muscle weakness, involuntary muscle contractions, clonic movements and muscle inco-ordinations, as well as back pain, excessive sweating, dry skin, hyperaesthesia, sensitivity to cold, vasomotor and trophic disturbances. A third of cases among adults involved inflammation of joints, and many experienced emotional instability. However, it was milder but more extensive than polio. Similar symptoms were reported in the late 1930s in an outbreak affecting patients and staff in community and military hospitals in Switzerland, and among nurses at Harefield Sanatorium, England, in 1939 (Jenkins, 1991). At least twenty more similar epidemic outbreaks were reported between 1934 and 1958 in the United States, Denmark, Iceland, Australia, Greece, England, South Africa and Germany under a number of names including Icelandic disease, acute infective encephalomyelitis, epidemic vegetative neuritis, and Royal Free disease (Shafran, 1991).

Royal Free disease was the name given to an outbreak of a fatigue-like illness at The Royal Free Hospital in England in 1955. The outbreak affected mostly just the staff, particularly female nurses, with greater prevalence among residential staff, and on 25th July 1955 the hospital had to be closed. It didn't reopen again until early October, by which point more that 300 people had been affected. Initially, glandular fever was considered as a diagnosis, but tests failed to support this. As paralysis developed among some sufferers, polio was also investigated as a possible diagnosis, but tests on cerebrospinal fluid found no evidence of polio. In the absence of any other diagnosis, a viral infection of the central nervous system was hypothesised (McEvedy & Beard, 1970b).

A year later, a Lancet article entitled "A New Clinical Entity?" discussing the puzzling Royal Free disease proposed the term benign myalgic encephalomyelitis, with characteristics differentiating it from "poliomyelitis, epidemic myalgia, glandular fever, the forms of epidemic encephalitis already described, and, need it be said, hysteria" (Lancet, 1956, as cited by McEvedy & Beard, 1970b, p.7). However, McEvedy and Beard (1970b) concluded that the disease was unlikely to have been caused by encephalitis, owing to the fact that there had not been any deaths during the epidemic, which would be expected in encephalitis. Similarly, they noted that few high temperatures were recorded among those who fell ill, and there were no comas. No evidence of encephalitis was found in blood or cerebrospinal fluid, neither were there biological markers suggesting or associated with organic dysfunction of the central nervous system. This led to the proposal of an hysterical epidemic by way of explanation for the Royal Free disease. However, McEvedy and Beard (1970a) did not believe that all similar epidemics were necessarily hysteria; they suggested altered medical perceptions due to the fear of a polio epidemic led to oversensitivity to symptoms, meaning that mild and otherwise usually unreported symptoms such as nausea and vomiting were documented. Briggs and Levine (1994) offered an alternative conceptualisation; although the similarities in the clinical presentation of each outbreak made it plausible to group them as the same or similar illnesses, variations in reported neurological features lead Briggs and Levine to conclude that subtypes should be investigated, with more detailed analysis of the neurological features specific to each case.

Jenkins (1991) reports that a further notable outbreak of a fatigue-like illness occurred between August 1970 and January 1971 at London's Hospital for Sick Children, Great Ormond Street, during which time 145 members of staff became ill. The illness was called epidemic neuromyasthenia (ENM) and in 1976-1978, a number of children with similar symptoms were reported at the same hospital. It was concluded that the illness may in fact be quite common, but may be misdiagnosed as psychiatric illness (Dillon, 1978, as cited in Jenkins 1991).

3.3.3. Myalgic encephalomyelitis. In the 1980s, unexplained chronic fatigue was hypothesised as a reaction to modern lifestyles and was dubbed 'yuppie flu' because of its apparent prevalence amongst young professionals. It seemed a popular and widely accepted theory, but the suggestion that its cause was psychological and not biological was rejected by sufferers (Wessely, 1997). A range of environmental factors were considered as having a causal role such as pollutants, diets, allergens, viruses and toxins, and a range of new names were suggested for the unfathomable illness, including post-viral fatigue syndrome and chronic immune deficiency syndrome. In 1986, the first diagnostic criteria for myalgic encephalomyelitis, or M.E., were published by Dr. Melvin Ramsay who took the decision to drop the previously used 'benign' from the name because of the level of disability experienced by its sufferers.

Table 2. The meaning of Myalgic Encephalomyelitis*,* presents the meaning behind this name and its relevance to the symptoms (Hyde et al., 1992).

Table 2. *The meaning of Myalgic Encephalomyelitis*

Segment of Name	**Meaning**
My	Muscle
Algic	Pain
Encephalo	Brain
Myel	spinal cord
Itis	Inflammation (there is limited evidence of inflammation in research to date, and so it has been proposed 'opathy' may be a better suffix which would denote that there is pathology, so the name would be Myalgic Encephalomyelopathy)

3.3.4. Chronic fatigue syndrome. The 1980s also saw two further outbreaks of an illness that closely resembled the previous outbreaks. These outbreaks were initially linked to Epstein-Barr virus (EBV) and were frequently diagnosed as chronic Epstein-Barr virus syndrome, because 'many, but not all, patients had Epstein-Barr virus antibody profiles' (Holmes et al., 1988). However, a causal link between the chronic symptoms and the Epstein-Barr virus has been questioned since. Although some studies found statistically significant associations between the chronic symptoms and Epstein-Barr virus, the associations were found to be as strong or stronger between the symptoms and the measles virus, herpes simplex 1 and 2 and cytomegalovirus (Holmes et al., 1988). Between 1985 and 1988, chronic Epstein-Barr virus syndrome was being diagnosed by doctors on a frequent basis, but Holmes et al (1988) believed that this was being done on the basis of diagnostic criteria that were not well defined. In 1988, Holmes et al. deemed it inappropriate to diagnose the chronic fatigue illness with a name that infers a specific causal agent. They provided a working case definition for the CDC and proposed the new name chronic fatigue syndrome. Their aim was to make diagnosis and research with patient populations more structured, systematic processes that acknowledged the lack of correlation between the chronic fatigue symptoms and any one specific causal agent. This name was proposed because it offered a more accurate description of the most prominent of all symptoms, the chronic fatigue (Holmes et al., 1988) and it does not invoke assumptions about the aetiology of the illness (Lewis et al., 1994).

In 1994, Fukuda et al. proposed a 'Comprehensive Approach to its Definition and Study' which offered a conceptual framework for treatment and research into the chronic fatigue syndrome. It recommended a revised approach to the clinical evaluation of people presenting with fatigue, a new case definition, and suggestions for subgrouping patient populations. All of these definitions recognise this as an illness located in an individual – none consider the impact on or of relationships.

3.4. Pathophysiology

The search for an organic cause and an understanding of the organic nature of CFS/ME could be considered thus far to have provided more answers about what CFS/ME is not than about what it is. There has been no single cause identified, and the mechanisms by which this condition is explained are complex (White, 2004). The body of research seeking to understand the biology of the illness, its symptoms and possible biomarkers is ever-growing and has included exploration of the possibility of dysfunction in the nervous system, the neuroendocrine system and/or the immune system (Klimas, Broderick, & Fletcher, 2012). Where initially, viral theories seemed promising, hypotheses focusing on the immune system, central nervous system (CNS) and brain have since taken precedence (Moss-Morris & Petrie, 2000). Here, we consider the key pathophysiological explorations that have taken place in research in recent years, with findings supporting the theory that CFS/ME is an illness in which multiple systems of the body are implicated.

An important point of note when considering the findings of any research into the pathology of CFS/ME is the complexity of CFS/ME population samples. Frequently contradictory findings could be a consequence of the fact that the CFS/ME population is not homogeneous. There are a range of difficulties in diagnosing the illness, there is a lack of consistent abnormalities with symptoms varying widely in presence and severity, and illness onset varies from gradual to acute (Masuda & Munemoto, 2002; Masuda & Nakayama, 2002).

A viral infection is commonly reported by PwCFS/ME as a precipitating factor in onset of their illness. In the late 1980s, this became a particular focus of research along with the use of the name ‘post-viral fatigue syndrome’ (PVFS) (Moss-Morris & Petrie, 2000). Certainly, persistent fatigue often lasting longer than 6 months is a symptom experienced by many infected with a viral or bacterial infection (Bansal et al. 2012) and the aetiological role of viruses in the development of CFS/ME has received support in research; in the 1980s, many people with PVFS were found to have been infected by an enterovirus (Yousef et al., 1988). Evidence of infection with enterovirus RNA was found in skeletal muscles of some people with fatigue, and in some cases these people tested positively up to 20 years post-onset (Archard, Bowles, Behan, Bell, & Doyle, 1988). A number of other viruses have been associated with CFS/ME, including Epstein Barr virus, Cytomegalovirus, Retrovirus, Human Herpes virus, and hepatitis C virus, but there is a lack of consistent evidence of a pathogenic relationship between any of these viruses and CFS/ME, as much research demonstrating a positive relationship is met with contradictory findings in other studies (Engleberg, 2002).

Although there is a growing body of research investigating the possibility that immune dysfunction could be a key factor in CFS/ME (Patarca-Montero, Antoni, Fletcher, & Klimas, 2001; Stewart et al., 2003) and abnormalities have been discovered, there is currently no consistent evidence of immune dysfunction as a primary cause of CFS/ME (Bansal et al. 2012). Although the theory of immune dysfunction as a causal factor has been investigated widely, results have been contradictory (Bassi, Amital, Amital, Doria, & Schoenfeld, 2008; Lorusso et al., 2009; Lyall, Peakman, & Wessely, 2003; Vernon & Reeves, 2005). What is also unclear is whether the defects that have been found to be present in the immune systems of CFS/ME are causal or a result of the illness, but studies have found therapies that target modulation of the immune system to be somewhat effective (Bansal et al. 2012).

Many studies analysing gene expression in PwCFS/ME have found that genes known to be associated with the immune system and defence are prominent in this population (Fang et al., 2006; Grans, Nilsson, & Evengard, 2005; Kaushik et al., 2005; Vernon, Unger, Dimulescu, Ravjeevan, & Reeves, 2002; Whistler et al., 2005). Eleven common pathways have been identified in studies investigating, and these have been found to be interrelated, which serves as evidence that complex biological mechanisms are implicated in CFS/ME (Fang et al., 2006). However, due to methodological issues, a number of gene expression studies cannot be considered reliable, and there is consequently still a long way to go before we have a clear understanding of the role of genetic factors and the metabolic pathways involved in the development and perpetuation of CFS/ME (Kerr et al., 2008).

Cerebral changes have been identified amongst PwCFS/ME. For example, one study found significant reductions in the volume of global grey matter in PwCFS/ME in comparison to healthy controls, with the decline found to be associated with reduced physical activity. Whilst this serves as evidence of the key role of the central nervous system (CNS) in CFS/ME, it remains unclear whether this is a cause or consequence of the illness (de Lange et al., 2005). Further research then sought to test whether the changes (known as cerebral atrophy which means cell loss) could be reversible by comparing the response of PwCFS/ME with low grey matter volume and healthy controls following CBT intervention. Findings demonstrated an increase in grey matter volume amongst PwCFS/ME following CBT intervention, specifically within the prefrontal cortex. These findings are concurrent with the theory of brain plasticity, and demonstrate an interactional relationship between macroscopic cortical plasticity and behaviour. (de Lange et al., 2008)

There is also evidence supporting a hypothesis of autonomic nervous system (ANS) dysfunction; adolescents with CFS/ME have been found to report significantly more dysregulation of the ANS than healthy controls, including changes in skin colour, feeling too warm, sweating and shivering. These participants were also found to have higher heart rates, tympanic temperature and higher levels of norepinephrine and epinephrine than healthy controls, and abnormal responses in tests designed to measure thermoregulatory responses. (Wyller et al., 2007)

Following positive findings in studies investigating functioning of the neuroendocrine system in fibromyalgia, it is hypothesized that disorders of the stress hormonal system – the hypothalamic-pituitary-adrenal axis (HPA) and related hormone production – could be present in some cases of CFS/ME as well as in mechanisms in the ANS that serve to regulate the neuroendocrine system (Wysenbeck, Shapira, & Leibovici, 1991).

3.5. Aetiology

Despite the attempt to locate a pathophysiological basis for CFS/ME, aetiological models actually suggest a combination of biopsychosocial factors as a more complete explanation for its onset, an approach that is known as the biopsychosocial model of illness (Alonso, 2004). In the case of CFS/ME, the precipitating factors were most often found to include an acute illness, usually viral, coinciding with stressors such as work-related stress or difficulties in personal relationships (Surawy et al, 1995). Where the individual was already struggling with such psychosocial stressors, it was proposed that a minor physical illness could be the 'last straw' (Surawy et al., 1995, p. 538) preceding a downward spiral into an almost complete loss of ability to cope.

In relating this explanation to cognitive theory (Beck, 1979), Surawy et al. (1995) proposed that the 'critical incident' which serves to activate the individual's assumptions around their worth, strength and achievement could be seen to be their altered self-perception as they become unable to perform to their usual high standards, and their inability to accept this in themselves. Surawy et al. (1995) explained that this 'critical incident' leads the individual to try even harder to perform, in the face of increasing levels of exhaustion. Ultimately, this strategy eventually fails and the individual enters ‘a state of chronic exhaustion, frustration and demoralisation' (p. 538).

Hyde (2003) also proposed a list of 'host factors' that he found frequently cause people to have become temporarily or chronically 'immune-compromised'. Hyde found that prior to the onset of CFS/ME, people have usually experienced one or more of the following:

- Exhaustion often caused by doing too much, overworking or frequent nightshifts
- Repeated episodes of infectious disease
- Recent immunisation
- Illness or experience of trauma

- Exposure to toxic chemicals

Within the biopsychosocial model of CFS/ME, it is conceptualised that following the onset of a fatigued state, this is also then perpetuated by a combination of social, physiological, behavioural, cognitive and emotional factors (Moss-Morris, 2005; Surawy et al., 1995; Wessely, Butler, Chalder, & David, 1991). It is proposed that as individuals focus on their symptoms, they often become fixated on organic explanations and on a belief that they have CFS/ME or a postural syndrome (Surawy et al., 1995). Surawy et al. (1995) hypothesise that some of the symptoms reported by PwCFS/ME once stuck in the fatigue cycle, including muscle pain, difficulties with concentration and the fatigue itself, are a consequence of physical deconditioning following periods of inactivity and emotional difficulties.

PwCFS/ME then seek to avoid anything that exacerbates symptoms, for example in some cases this may mean getting out of bed or getting dressed, in other cases this may mean forms of physical exercise such as walking, and although the avoidance serves to reduce symptoms in the short term, the long-term consequence is that physical and mental activity becomes even less tolerable for the PwCFS/ME as their threshold reduces through the periods of inactivity. As PwCFS/ME are also likely to fluctuate between periods of inactivity and periods of attempting to function at pre-morbid levels, they then experience repeated failure to perform, causing further distress and strengthening their belief in their illness as unbeatable. Eventually, the PwCFS/ME gives up trying to return to their pre-morbid levels of functioning and instead becomes preoccupied with symptoms and a sense of increasing disability. As this develops further, the PwCFS/ME becomes demoralised and begins to accept that they may never recover (Surawy et al. 1995). This must be an incredibly frustrating and seemingly hopeless process for the PwCFS/ME and those around them to go through, with feelings of resignation and stagnation being a possible consequence.

3.6. Assessment & differential diagnosis

Due to the lack of a pathognomonic signs and diagnostic tests, CFS/ME is diagnosed via a process of exclusion. Individuals are subjected to a battery of tests and assessments which screen for as many alternative explanations, medical and psychiatric, as is possible through routine testing, and once all alternative explanations have been ruled out, a diagnosis of CFS/ME may be made, provided the individual meets the diagnostic criteria (Fukuda et al., 1994).

There is limited discussion of the differences between acute and gradual onset of CFS/ME in the definitions. For this reason, individuals with apparent gradual onset CFS/ME can be diagnosed as such according to the 1988, 1994 or UK definitions; however, CFS/ME is often a misdiagnosis, whereby the PwCFS/ME has another underlying illness that remains undetected (Hyde, 2003). The implications of this could be that other underlying illnesses go untreated, and medical professionals may be reluctant to undertake further tests for other illnesses once a diagnosis of CFS/ME has been made. Indeed, Hyde (2003) believes that further testing at later stages of the illness would in fact reveal that a proportion of PwCFS/ME actually have a major illness that has gone undetected following the CFS/ME diagnosis. Interestingly, Hyde points out, if any major illness other than CFS/ME is diagnosed, then the CFS/ME diagnosis is usually removed. Hyde reports that he spends approximately one and a half days examining each person who presents with symptoms of CFS/ME, mapping the PwCFS/ME's organs, structures and systems, which he claims allows for more informed decisions about the illness process than most physicians are able to make. Hyde consequently puts forward a convincing argument for more in-depth testing of PwCFS/ME, stating that whilst the necessary testing may, at first sight, seem costly at around £10,000, in fact the average cost to the economy through long term loss of earnings for the individuals who fall ill is much greater.

Following their findings that PwCFS/ME have a high prevalence of psychiatric disorders that often go unrecognised, Lane, Manu, & Matthews (1991) recommend that the assessment of CFS/ME should also incorporate a full and structured psychiatric evaluation. They also found that PwCFS/ME often experienced lifelong functional symptoms beyond those recognised in the case definition of CFS/ME. It may be the case that individuals feel that they cannot ask for further support with symptoms once a diagnosis of CFS/ME has been made – they may believe that what they have been offered is all that is available and consequently attempt to cope alone with symptoms that could have been alleviated through intervention. For example, where a healthy individual might experience symptoms such as difficulties with sleeping, depression or anxiety and access health services for episodic support, a PwCFS/ME may see symptoms such as these as part of their illness and feel that additional services are not available to them other than those already offered.

In a study assessing the efficacy of the cognitive behavioural approach to CFS/ME, (Surawy et al., 1995) participants with CFS/ME did not report feelings of anxiety or depression, despite the fact that somatic symptoms associated with such emotional disorders were often present. They found any reports of anxious or depressed mood amongst the participants with CFS/ME were reported to be a consequence of their fatigue and associated symptoms and not the root of the problem.

3.7. Prognosis & recovery

Just as the illness experience of CFS/ME is subjective, so is the concept of recovery. Longitudinal studies have shown that although some people diagnosed with CFS/ME improve over time, a majority experience symptoms for many years, with some never fully returning to levels of functioning that they consider to represent full recovery (Fukuda et al., 1994; Peterson, Schenck, & Sherman, 1991; Wilson et al., 1994), although there is a lack of clarification in the literature over what ‘full recovery’ might mean in CFS/ME (for example see Cairns & Hotopf, 2005). Although most PwCFS/ME eventually improve, often gradually, it is rare for adults with CFS/ME who experience moderate to major illness to return to their pre-morbid level of functioning (Cairns & Hotopf, 2005; Joyce, Hotopf, & Wessely, 1997). PwCFS/ME who remain debilitated by two years after illness onset are the least likely to make a full recovery, although the majority of children and adolescents with a diagnosis of CFS/ME do recover substantially if given appropriate care and support in coping with their symptoms (Marshall, 1999).

4. Psychosocial factors: A Scoping Literature Review

4.1. Scoping Literature Review: Method

The purpose of the scoping review was to identify key findings and map the current evidence available prior to defining the research questions and aims. Whilst the area of interest was predefined (psychosocial aspects of CFS/ME), the scoping review enabled the identification of key concepts and gaps in the literature. Several procedures were undertaken in the literature review process.

Prior to the first draft of this scoping literature review, database searches of peer-reviewed journal articles were conducted on Web of Science and Google Scholar using a wide range of search terms. The first search undertaken was using the search phrase *chronic fatigue syndrome.* Further more refined searches included the phrase *chronic fatigue* alongside the following terms: *attribution, cognitive, illness perceptions, family, significant other, spouse, partner, experience, qualitative,* and further searches were conducted using the terms *medically unexplained illness, chronic pain* and *chronic illness* alongside search terms related to the area of interest (eg *Illness perceptions*). Papers were sorted into the themes of *biological*, *psychological, treatment*, *social*, *significant other*, and *other* based on the main focus of the paper. The papers judged to be of most relevance and importance (for example, based on relevance of titles, number of citations and/or dates of publication) in each theme were then printed, read and annotated, with more detailed themes identified within each paper and used to develop an understanding of the narrative within the literature so far. Summary cover-sheets were systematically added to each paper and used to draft a coherent synthesis of the key relevant literature. During this process, the reference sections for each key paper were also searched in order to identify any additional articles or books not picked up in the database searches, and any new articles which came to light were also analysed and categorised as described.

Key books and guidance documents that were cited in journal articles and/or were available in the library at the University of Hull were either purchased or accessed as e-books or through the library, and the relevant chapters were analysed. The reference sections of these books were also searched. The most relevant published text used during this review most comprehensively was Moss-Morris and Petrie (2000), *Chronic Fatigue Syndrome*, which provided an excellent overview of the knowledge to date of psychological and social aspects of chronic fatigue syndrome up to the year of publication.

The initial scoping review concluded in early 2015 prior to the refinement of the research design. However, during the data collection and analysis process, further exploration of the literature was undertaken, guided by concepts arising from the data. This process was more organic and therefore less systematic than the previous literature search, but it was extensive and involved the exploration of a broad range of concepts and terms that arose in the analytic process. The findings from these literature reviews are presented and discussed in relation to the research data in each of the findings chapters.

Following the completion of data collection and during the writing-up of findings in 2018, an up-to-date literature review was conducted, guided by the research questions, aims, and the concepts and categories presented in the findings. This process followed the same database search method outlined above (Google Scholar and Web of Science), but search terms were expanded to include specific topics of interest based on the research findings. A detailed spreadsheet was kept, summarising each paper and grouping all papers into the following themes: *Key Researchers, Key Texts*, *Significant Others, Self & Identity, Social Support, Other issues* (including *precipitating* and *perpetuating factors* and *avoidance behavior), Expressed Emotion, Attribution & Control, Guilt, Causes of CFS/ME, Acceptance & Mindfulness, Rationale,* and *Onset &*

Diagnosis. Many papers were duplicated across themes if they reported issues that were of relevance to more than one theme. This process enabled a thorough audit of all papers relevant to the research undertaken, and although it was not practical to include every piece of literature ever written on each subject, it ensured that the most recent and/or relevant advances were incorporated either within the scoping review (if necessary) or within the relevant discussion sections of the findings.

The following scoping literature review presents the findings of the first stage of the literature review outlined above, but with some minimal refinement and relevant additions following the final review of the literature in 2018. Finally, the aims that were developed for the subsequent research out of the emergent gaps in the literature are presented.

4.2. The cognitive behavioural models

In the absence of a clear biological cause or test for CFS/ME, the preferred paradigm within which most researchers and clinicians now work is that of the cognitive behavioural and biopsychosocial model (Dickson, Knussen, & Flowers, 2007). Responding to illness by reducing levels of activity has been found to be a mechanism of adaptation and protection when people are unwell (Hart, 1988; Kelley et al., 2003). Because in CFS/ME, worsening of symptoms is usually experienced as a result of exertion, most adapt to their illness by reducing activity levels. This is considered in psychiatry to be a maladaptive response in CFS/ME that serves to perpetuate the illness. Further, the attribution of illness to physical causes has been found to lead PwCFS/ME to further avoid physical activity in an attempt to manage symptoms, thereby causing a vicious cycle of deconditioning and depressed mood. (Wessely et al., 1991; Wessely, David, Butler, & Chalder, 1989)

As momentum gathered for this as a hypothesis, which takes into account fear avoidance and deconditioning as interacting factors in CFS/ME (Deale, Chalder, Marks,

& Wessely, 1997; Prins, Bleijenberg, & Bazelmans, 2001), a cognitive behavioural model of CFS/ME emerged. Within the early model proposed by Wessely et al. (1991), the possibility of an organic cause was not completely disregarded; rather, initial onset was considered to be generally precipitated by an organic factor such as a virus. This acute organic illness is thought to then interact with psychological and behavioural responses as part of the fear-avoidance-deconditioning cycle so that illness and symptoms are perpetuated (Deale, Chalder, & Wessely, 1998; Wessely et al., 1991). The cycle leads to a state of chronic physiological dysregulation in which sleep/wake cycles, muscle functioning and deconditioning all play a key role (Deary, Chalder, & Sharpe, 2007).

Surawy et al. (1995) extended this cognitive behavioural model through their work which uncovered psychological processes as predisposing and precipitating, as well as perpetuating, factors. During their clinical work with 100 individuals who met the criteria for CFS/ME, cognitive therapy methods were used to elicit information about participant cognitions in relation to their illness. They found three major themes emerged, which can be summarised as follows:

- Specific activities make them feel worse and they should instead be resting.
- Frequent comparison of current abilities with pre-morbid abilities, particularly in relation to activity levels or performance standards.
- The opinions of others are important, and they don't want to be seen as 'lazy' or unable to cope.

They also found consistent underlying assumptions amongst participants, which followed two themes:

- A focus on high standards, and a failure to meet high standards indicates 'failure as a person.' (p.537)

- Psychological strength was perceived by participants as important, so they preferred not to admit to what they saw as weaknesses.

These assumptions led participants to believe that they must achieve high standards without complaint or signs of weakness, and that the respect of others depended on their ability to do this. It is suggested that this leads the PwCFS/ME to focus on the somatic aspects of the illness and discount the emotional aspects, further perpetuating their symptoms. Findings suggest that individuals who hold assumptions that they must meet the high standards of others without showing signs of weakness or inability to cope are particularly vulnerable to the development of CFS/ME. Explorations of the childhood experiences of PwCFS/ME revealed that these beliefs and assumptions arose through interactions with caregivers who placed high value on the meeting of expectations, and that these expectations often involved high standards. Participants reported that parental recognition, acceptance and affection were dependent upon the child's ability to meet these high expectations, and admissions of difficulty or anxiety were not tolerated (Surawy et al. 1995).

Surawy et al. (1995) also reported that their clinical observations and enquiry with both PwCFS/ME and families revealed that PwCFS/ME were perfectionist, achievement-orientated personality types who set high standards for themselves and were concerned about the opinions of others. Their lifestyles prior to the onset of their illness were characterised by long-term efforts to meet the expectations of others and their own high standards. It became clear that typical PwCFS/ME who are achievement-orientated personality types become conflicted by their belief in the need to reduce activity, and consequently a cycle of ‘all-or-nothing’ behaviour begins (Spence, Moss-Morris, & Chalder, 2005).

4.3. Attribution

Within Leventhal's Self-Regulatory Model of illness (SRM) (see Leventhal, Meyer, & Nerenze, 1980), an individual who is living with an illness assigns meaning to their illness experience; these meanings are underpinned by their perceptions about the illness, and their perceptions are influenced their emotional state. This then becomes a cycle as their perceptions then start to further influence their emotional state.

As they attempt to return to 'normal' functioning, coping strategies are employed, but the coping strategies selected are dependent upon their perceptions about the illness and their emotional state. They then evaluate the effectiveness of these coping strategies based on whether they are successful in restoring 'normal' functioning. This model therefore illustrates that an individual's illness perceptions and the subsequent coping strategies have a direct impact on their psychological well-being. A significant proportion of variance in levels of distress has been shown to be explained by illness perceptions in a range of illnesses including chronic illness (e.g. Cartwright, Endean, & Porter, 2009; Evans & Norman, 2009; Llewellyn, McGurk, & Weinman, 2007; Millar, Purushotham, McLatchie, George, & Murray, 2005; Traeger et al., 2009).

Surawy et al. (1995) argue that within the cognitive model of CFS/ME the individual's inability to accept their failure to cope means that they also struggle to accept any role for social or psychological stressors in their illness, and instead seek biological explanations. Focusing on symptoms and holding negative beliefs about cause, prognosis and recovery have been found to be associated with poorer clinical outcomes for PwCFS/ME (Moss-Morris & Chalder, 2003; Moss-Morris, Sharon, Tobin, & Baldi, 2005). PwCFS/ME have been found to believe that symptoms indicate that their illness is worsening, and that activities that increase the severity of symptoms are harmful or a potential cause of relapse (Warwick & Salkovskis, 1990).

This is consistent with findings demonstrating that compared with healthy controls, PwCFS/ME are more likely to make somatic attributions for symptoms such as headaches and fluctuations in weight that are actually normal, common, everyday symptoms, and are less likely to attribute symptoms to psychological causes (Butler, Chalder, & Wessely, 2001; Dendy, Cooper, & Sharpe, 2001). Cho, Bhugra & Wessely (2008) assert that when people become ill, they want to know why, and the answers and reasons they find influence their cognitions and behaviours in relation to their illness experience.

Following research demonstrating a tendency among PwCFS/ME to favour a physical disease explanation for their symptoms and reject a psychological explanation (Matthews, Manu, & Lane, 1989; Powell, Dolan, & Wessely, 1990), further investigations reveal that PwCFS/ME who strongly believe in an organic cause for the illness fare worse in terms of clinical outcomes and disability, whereas those who are open to psychosocial interventions see greater improvements in their disability (Moss-Morris, Petrie, & Weinman, 1996). This seems to suggest that the way PwCFS/ME perceive their illness strongly correlates with their ability to overcome it. On the contrary, Deale, Chalder & Wessely (1998) found that attributions to a physical illness were less influential than was previously thought. Their findings suggest that positive outcomes from CBT treatment in CFS/ME are associated with alterations in behaviour so that avoidance behaviour is reduced, and not with changes to the causal attributions made by PwCFS/ME.

It is likely that the cultural setting within which one experiences life with CFS/ME influences the attributions one makes, although research is limited in this area. Cho, Bhugra & Wessely (2008) found that British PwCFS/ME perceived their illness as more chronic and attributed their illness to a physical cause more frequently than Brazilian PwCFS/ME. This may be related to a number of psychosocial factors such as the design, model and delivery of healthcare services in different cultures, general attitudes towards illness including stigma around mental health issues, and influences from media, patient groups and published resources. A range of influences may interact to alter the perception of illness and the preferences of PwCFS/ME in how they construct their understanding and explanations of their experience.

4.4. Treatment

Currently the only evidence-based treatments for CFS/ME are psychosocial interventions. The UK National Institute for Health and Clinical Excellence (NICE) (NICE, 2007) recommends treatment with cognitive behaviour therapy (CBT) and/or graded exercise therapy (GET). However, surveys have revealed a view among PwCFS/ME that these treatments can cause harm and specialist medical care (SMC) and adaptive pacing therapy (APT) are therefore often favoured by PwCFS/ME (White et al. 2011).

APT is an approach to treatment that favours the envelope theory of CFS/ME (Pesek, Jason, & Taylor, 2000; White et al., 2011). Envelope theory views the illness as an organic illness which renders individuals lacking in pre-morbid energy levels. It does not consider the illness to be reversible according to behavioural changes, and instead focuses on management of and adaptation to the illness by careful planning of activity levels so as not to ‘use’ more than 70% of their conceptual energy reserves, thereby avoiding fatigue. In this way, it aims to aid natural recovery. Patients use diaries to track and plan activities, develop an understanding of what exacerbates symptoms, build in plenty of rest time, and gradually work towards increasing activity levels if the PwCFS/ME feels able (Pesek et al., 2000; White et al., 2011).

GET is based upon the theories of deconditioning and exercise intolerance which hypothesise that physiological changes are caused or worsened by deconditioning through the avoidance of activity in an effort to reduce symptoms; however, this in turn causes an increased perception of effort, and activity levels are further reduced as a result. Within this model, it is assumed that the changes and effects of deconditioning are reversible through efforts to gradually increase activity levels thereby decreasing symptoms and disability levels. (Fulcher & White, 1997; Moss-Morris et al., 2005)

Treatment with CBT for CFS/ME is based on the theory of fear avoidance, which proposes that a PwCFS/ME becomes trapped in a cycle of avoiding activity and fear of then engaging in activity. Within this model, it is assumed that the illness is reversible through altering a PwCFS/ME's cognitive responses and therefore the associated behaviours in which they engage, as it is believed these are linked to the physiological processes that allow the fatigue cycle to continue (Deale et al., 1997; Prins et al., 2001). Although it was initially intended that CBT might alter the causal attributions made by PwCFS/ME, it is in fact thought to improve outcomes through altering avoidance behaviours (Deale et al., 1998). It is notable that despite the recognition that the views and opinions of others are key in the CBT formulations of CFS/ME, the interventions recommended are all based on individual work with the patient.

The 2011 PACE trial (White et al., 2011), a CFS/ME multicenter, randomised, four-group study, compared treatment with CBT, GET and APT each delivered alongside SMC with the delivery of SMC alone, examining the potential for harm as well as for improvement. It also compared APT with CBT and GET across subgroups of participants who met different diagnostic criteria. 641 PwCFS/ME were involved in the study across six specialist CFS/ME clinics, and outcomes were assessed up to one-year post-randomisation.

Self-reported improvements of at least 2 points in fatigue and 8 points in physical functioning were achieved by 42% of participants in the APT group, 59% for CBT, 61% for GET and 45% SMC. There was improvement among more participants in the CBT group compared with APT or SMC, and among more participants in the GET group compared with APT and SMC. There was no difference in improvement between the APT and SMC groups. More participants in the CBT and GET groups rated themselves as much better or very much better at 52 weeks than in the APT and SMC groups. There was no difference between the groups in the number of participants who considered themselves to be worse following treatment, with as many as 9% across each group. Compared to the APT and SMC groups, participants in the CBT and GET groups also had better scores for work, social adjustment, depression and sleep disturbance, post-exertional malaise was lower, anxiety levels were lower than the SMC group.

When added to SMC, CBT and GET appear to have greater success in reducing fatigue and improving physical functioning. Therefore, the findings support CBT and GET as effective treatments for CFS/ME when added to SMC, and fail to provide evidence to support pacing (APT) as a first-line therapy (White et al., 2011).

4.5. Social support

No illness is exclusively physiological; every illness exists and the experience of it is influenced by, social context, and certainly in the case of CFS/ME, the social environment is known to influence the severity of symptoms (Ware & Kleinman, 1992). There is a great deal of controversy surrounding CFS/ME, and PwCFS/ME are highly aware of the stigma around their illness (Looper & Kirmayer, 2004). Indeed, Ware (1992) noted that PwCFS/ME are more often perceived by others to be malingerers whose illness is really all in the mind. PwCFS/ME report becoming isolated and lonely, experiencing a loss of friendships as friends become impatient and cynical about their illness (Dickson et al., 2007). In the face of this isolation and as their lives become more limited by disability as a result of their illness, they experience fewer opportunities for social interactions and become ever more isolated (Dickson et al. 2007). Further, as a consequence of their illness they experience loss of material possessions, jobs, social support networks and recreational activities (Assefi, Coy, Uslan, Smith, & Buchwald, 2003). It is possible that this puts more pressure on their closest relationships, and there is a growing body of research exploring how PwCFS/ME experience their relationship with a partner (Asbring, 2001; Dickson et al., 2007; Dickson, Knussen, & Flowers, 2008; Drachler et al., 2009), and of spousal or couple experiences in chronic illness (Eriksson & Svedlund, 2006; Söderberg, Strand, Haapala, & Lundman, 2003) and chronic pain (Cano, Johansen, & Geisser, 2004; Leonard, Cano, & Johansen, 2006; Newton-John & Williams, 2006). It seems clear that the wider family support system and environment is also a highly influential factor in CFS/ME worthy of consideration (Dickson et al., 2007; Donalek, 2009). PwCFS/ME have been shown to sometimes experience delegitimation from their SO (Dickson et al., 2007). They are particularly vulnerable to distress if they feel their SO fails to understand their illness and does not provide validation for their suffering; feeling delegitimised by their SO appears to be more distressing for PwCFS/ME than the delegitimation they experience from the medical profession (Dickson et al. 2007).

The impact of SO responses on CFS/ME outcomes has also been explored. Solicitous responses are defined as demonstrating concern for the PwCFS/ME's physical condition, comforting them, and/or discouraging them from doing things or doing things for them (Kerns & Rosenberg, 1995), and have been found to be associated with increased fatigue and disability (Brooks, Daglish, & Wearden, 2013; Romano et al., 2009; Schmaling, Smith, & Buchwald, 2000; Verspaandonk et al., 2015). Negative responses, defined as the expression of anger and frustration at the PwCFS/ME (Kerns & Rosenberg, 1995) have also been associated with poorer psychological outcomes for PwCFS/ME as well as increased depression (Romano et al., 2009) but high levels of frustration could be a result of difficulties in communication between PwCFS/ME and their SO, and in expressing and meeting one another's needs.

Band, Barrowclough and Wearden (2014) expanded upon previous research to explore SO responses in broader dyadic relationships; although the majority of SOs in the study were partners, the sample did include several parent/child relationships and one sister. Their work explored SO responses using a framework called ‘expressed emotion’, developed to assess family communication patterns, in particular, how relatives of people with psychiatric conditions talk about them (Vaughn & Leff, 1976). Extensive studies into the EE construct have revealed that high EE is a predictor of poorer clinical outcomes in a range of conditions (Butzlaff & Hooley, 1998), including social anxiety disorder (e.g. Garcia-Lopez, Muela, Espinosa-Fernandez, & Diaz-Castela, 2009), schizophrenia (e.g. Leff, Kuipers, Berkowitz, Eberlein-Vries, & Sturgeon, 1982), depression (e.g. Hooley, Orley, & Teasdale, 1986; Vaughn & Leff, 1976), eating disorders (e.g. Szmukler, Eisler, Russell, & Dare, 1985) and dementia (Safavi, Berry, & Wearden, 2017). A post-data collection review of literature suggests that high EE has also been related to higher levels of SO burden and distress in dementia (Safavi, Berry, & Wearden, 2018).

Within the EE framework, SOs are categorised as being high or low in EE based on evidence for their behavior across three dimensions - Critical Comments, Emotional Overinvolvement (EOI) and Hostility (Vaughn & Leff, 1976). High Expressed Emotion is defined as being hostile, high in critical comments and a level of intolerance to the PwCFS/ME, although family members are thought to believe they are being helpful when behaving in this way. Families with high EOI tend to exhibit over-protective behavior, self-sacrifice, and can be emotionally intrusive. Hostility in the SO is characterised by a negative attitude towards the PwCFS/ME in which the family may believe the PwCFS/ME has control over their disorder and it is their choice not to get better. Where there is a combination of both Hostility and EOI, family attitudes can be characterised within the EE framework by critical attitudes – they may believe that the

PwCFS/ME is not to blame for their illness but may still criticise the PwCFS/ME's behavior in relation to their illness (Vaughn & Leff, 1976). Hostility and Criticism have been found to be highly correlated (Chambless, Bryan, Aiken, Steketee, & Hooley, 1999) and factor analysis conducted by Chambless et al. (1999) revealed that EE consists instead of Criticism, EOI and the third factor of Positivity, which consists of Warmth and Positive Comments.

The factors of the EE construct were considered by Band et al. (2014) to map onto the concepts that have been gaining pace in CFS/ME research; solicitous responses were considered similar to EOI in the EE framework, and negative responses were viewed as similar to the criticism construct in EE. Band et al. explained that within the EE framework a critical comment might be coded in data when there is evidence of strong tonal criticism or disapproval of the PwCFS/ME. Where an SO is seen to over-identify with the PwCFS/ME, displaying self-sacrificing behaviours which could also be interpreted as being overprotective and emotionally exaggerated (Vaughn & Leff, 1976), this would be coded as an example of EOI. High levels of SO critical comments were found by Band et al. (2014) to be predictive of increased fatigue severity and increased depression in the PwCFS/ME; parents were more likely to score highly for EE in the domain of EOI than partners were and high EE in SOs was found to be associated with poorer outcomes in fatigue severity and depression longitudinally.

However, what was lacking in this work was an exploration and explanation of the relationship dynamics underlying these behaviours. Why did SOs demonstrate EOI, and how did both PwCFS/ME and SOs feel in this situation? High EE tells us that there are many emotions to be explored, but it does not tell us why these emotions evolved or about the interactional effect of these emotions. Similarly, the findings may provide evidence of critical comments, but they do little to illustrate for us what this experience is like for a PwCFS/ME, or even the factors that lead to an SO behaving in this way. For a deeper understanding, qualitative exploration of the perspectives of both SOs and PwCFS/ME are necessary.

Although research considering the perspective of SOs in the context of CFS/ME is more limited, Kelly, Soderlund, Albert, & McGarrahan (1999) incorporated SOs into a study of social support among 41 PwCFS/ME and found that half of the 25 SOs reported that the impact on their life had been negative. Family roles were disrupted, and participants experienced anger and difficulties with finance. Ax, Gregg and Jones (2002) undertook a comprehensive qualitative study in which they explored the difficulties experienced by 17 people caring for someone with CFS/ME at three stages of the illness: after onset but prior to diagnosis (Stage 1), the period after diagnosis (Stage 2), and the period around the time of the study (Stage 3). Amongst these were husbands, wives, a mother, a child and friends. Methods of analysis in the study are unclear, but it seems that the data was analysed using thematic analysis. Findings indicated that SOs felt their lives were only slightly limited by the illness, but that all the carers had to make changes in their own lives to adapt to and cope with the illness, resulting in eventual acceptance, although this was conceptualised more as a form of resignation. Themes identified across the three stages include: Caring tasks and difficulties, Acceptance, Meaning of a diagnosis, Beliefs about the origin of CFS, and Acceptance and adjustment. Four major themes and 16 subthemes are presented. However, the paper attempts to present such a large number of themes and subthemes that it lacks depth and lacks comparison of types of relationships. All participants are referred to as ‘carers’ and therefore any potential differences in terms of relational patterns are not identified. Instead, the study focuses on commonalities in the data, presenting most of the subthemes with quantifying statement such as:

> *"While 11 carers suggested that they accepted the reality of and severity of the symptoms before the diagnosis, they felt powerless to help the sufferer."* (P. 37)

More recently, Brooks, King and Wearden (2014) conducted an in-depth qualitative study exploring the experience of CFS/ME from both the PwCFS/ME and their SOs' perspective. In-depth semi-structured interviews were conducted with two PwCFS/ME and their spouses and Interpretative Phenomenological Analysis (Smith, Flowers, & Larkin, 2009) of the data was conducted. Findings suggested that interactions with others outside of the relationship was a central issue for participants. Two themes relating to couples' interactions with others were presented: Interactions with healthcare professionals and Interactions with the social world. The couples who participated had different experiences with healthcare professionals – one couple felt that they had a collaborative relationship with health professionals whilst the other couple felt let down by services. In interactions with the social world, it was asserted that all participants felt there was a lack of understanding of CFS/ME. One couple was viewed as having an 'effective marital partnership' (p.14) which the participants believed helped in coping with the condition, whilst the other couple had more difficulties, with one withdrawing and the other feeling socially isolated. However, because it was conducted with just four participants, this study was limited in its breadth. As a qualitative study it of course did not seek generalisability, but it is possible in qualitative research to theorise about patterns in the data. Although comparisons were made between the two couples, because of the small sample size it was not possible for the authors to theorise about whether these could be patterns which could apply more broadly in different types of relationships in CFS/ME.

4.6. Aims

The aim of the present study was to extend the work of Band et al. (2014) and Brooks et al.’s (2014) model of investigating CFS/ME from the perspective of the PwCFS/ME and their SO. It sought to provide more nuanced explanations of the dynamics at play that were described by Band et al. (2014), and to employ similar strategies to Brooks et al. (2014) but with a broader sample in order to theorise about relational patterns. This research also employs ideas from the work of Ax et al. (2002) by exploring a range of relationship types, but seeks to compare and contrast the relational experiences in these dyads, identifying differences as well as similarities in relational patterns with a view to developing an explanatory model to inform future research and clinical practice.

5. Methodology

5.1. Introduction

This study is a qualitative grounded theory study which consisted of semi-structured interviews in participant homes or over the telephone. Data collection took place between May 2015 and November 2016. 15 participants consisting of PwCFS/ME (*n*=9) and SOs of PwCFS/ME (*n*=6) completed a total of 26 audio-recorded semi-structured interviews. Participants completed a diagrammatic task in between interviews based on research topics provided by the researcher. This was then used as a prompt during second interviews. Data was transcribed by a team of three students but was analysed by the researcher independently guided by grounded theory methodology.

This chapter will describe the research question, aims, design, philosophical paradigm and procedures. Much of this section is written in the first person, as is customary in reporting much qualitative research. This is because, as is explained in more detail later, the decision to undertake qualitative research cannot be separated from the researcher's position, assumptions and aims, and indeed these must be recognised as implicated and influential in the research process (Charmaz, 2014). This section provides a personal account as to how I reached crucial decisions in designing and undertaking the research.

5.2. Research question & aims

The previous chapter outlined the rationale for the exploration of the relationship between PwCFS/ME and their SOs in the context of CFS/ME. The lack of understanding in this area led to the development of the following research question, to be addressed in this study:

1. How do people with CFS/ME and their SOs experience their relationship in the context of illness?

The initial aims of the research were:

1. To develop a more nuanced understanding and explanation of the difficulties in relationships between PwCFS/ME and their SO in the context of CFS/ME than currently offered in the literature.
2. To describe in detail the emotional processes that may be underlying previous findings in research regarding SO responses.
3. To develop a model that explains the characteristics, processes and consequences of relationships between PwCFS/ME and their SO in the context of CFS/ME.

5.3. Research design

Previous research suggests that in CFS/ME strained relationships between PwCFS/ME and their SOs correlated with poorer clinical outcomes (Band et al., 2014; Band, Wearden, & Barrowclough, 2015; Wearden, Tarrier, Barrowclough, Zastowny, & Rahill, 2000). I became interested in the changes in family dynamics in response to CFS/ME and identified a gap in research illustrated in the previous chapter around understanding in more detail what the difficulties are in dynamics between PwCFS/ME and their SO. I identified a need to understand what does and what does not work in family dynamics in CFS/ME – what factors are conducive to effective relationship functioning in CFS/ME, and what factors are barriers?

I wanted to explore people's experiences of relationships in the context of CFS/ME. My intention was to understand their thoughts, feelings and behaviours, the reasons behind these, the meaning they applied to them and to their illness, and the way that these factors changed their relationship dynamics over the course of their illness. As I began to develop an understanding of qualitative research, I realised that because of the subjective nature of what I wanted to investigate, and the emphasis on participants' own interpretations and meaning, this topic would be best explored through qualitative methodology (Braun & Clarke, 2013).

In identifying the most suitable qualitative approach aligned with the research question and aims, Grounded theory method (GTM) (Glaser & Strauss 1967) was chosen over narrative, phenomenological, case study and ethnographic approaches. This was because the aims of the research were to understand relational processes and to develop a theoretical model to explain these. Table 3. provides a summary of the major applications of each of these approaches and demonstrates why grounded theory was the approach best suited to my research aims.

Table 3. *Five main approaches to qualitative research*

Inductive Qualitative Approach	**Purpose**
Narrative	Collect, capture and retell participants' stories
Phenomenological	Capture participants experiences, examining how they make sense of them
Grounded Theory	Develop a model or theory to aid understanding of phenomenon, situation or process
Case Study	In-depth study of well-defined case or phenomenon using multiple data sources
Ethnography	Explore a group or phenomenon through observation in their natural environment

Note. Creswell (2012) *Qualitative Inquiry and Research Design. Choosing Among Five Approaches* (3rd).

At present, limited theory exists around PwCFS/ME-SO relationships in CFS/ME. The nature of CFS/ME mean that theories developed in the context of other illnesses should not be assumed applicable. Whilst there is evidence to suggest that PwCFS/ME and SO relationships are associated with clinical outcomes (e.g. Band et al., 2014, 2015), a deeper understanding is required around how this interaction manifests in the lives of PwCFS/ME and their SOs. GTM allowed for exploration from a predominantly participant-led perspective. GTM allows space for the researcher to construct theory from the data rather than selecting pre-existing theory and working within it (Urquhart, 2013). This study presents a theory that is grounded in the data but draws on existing theory as a helpful and evidence-based way of understanding and explaining processes. The systematic process of constant comparison across the data set and with existing theory allowed deeper exploration of the relevance of current theory in relation to concepts emerging from the data (Charmaz, 2014).

5.4. Philosophical paradigm

Because of the recognition of the researcher as central to the research process and findings in qualitative research, it is important for a researcher to recognise, analyse and reflect on and explain the values and experiences that have shaped their approach to research (Braun & Clarke, 2013; Urquhart, 2013). It is also necessary for the researcher to analyse and understand their own worldview so that it can then be measured up against theoretical paradigms. After all, "paradigms are overarching philosophical systems" and they "represent belief systems that attach the user to a particular worldview" (Denzin & Lincoln 2011, p.5). This worldview is greatly influenced by the racial, class, gender, cultural and community perspectives with which the researcher has deeply engaged throughout their lives (Denzin & Lincoln, 2011). It is also important to recognise that in qualitative research, the instrument of enquiry is the researcher (Patton, 2002). The choice of topic, data collection procedures and the analytic process are affected by the researcher's experiences, values, opinions and intentions (Braun & Clarke, 2013). It is therefore of vital importance for the researcher to recognise, define and reflect on how their background, beliefs and biases have affected every aspect of their research. This is should be done throughout the research process through employing reflexivity (Schwandt, 1997), which will be discussed later, but it is also necessary to discuss how my presuppositions affected my early methodological decisions in the research design process (Braun & Clarke 2013).

5.4.1. Researcher background. I am a 35-year-old white British psychology PhD student with an MSc Psychology Conversion Award and a BA (Hons) Ethnomusicology. I also worked as an Assistant Psychologist for Humber NHS Foundation Trust in an acute adult mental health inpatient unit during the data collection and analysis period. I have previously worked as a primary school teacher and in various positions in business, and have been a volunteer with Samaritans for five years. Prior to undertaking my PhD, I had no professional experience of qualitative or quantitative research or of working with PwCFS/ME. However, I have a family member who has been living with CFS/ME for over 16 years, and know several other people who have previously received a diagnosis of the illness. I approached the study of CFS/ME with curiosity and an open mind, and used reflection and supervision throughout the study to become more mindful of my own biases, preconceptions, assumptions and beliefs as concepts and questions presented themselves.

5.4.2. Philosophical positioning. My explorations of qualitative research have enabled me to realise that whilst I recognise the usefulness of positivist approaches in answering certain questions, I also see the value of interpretivist approaches in seeking to understand human experience and meaning (Denzin & Lincoln, 2011). Being introduced to the concept of social constructivism was particularly pivotal for me in altering the way I think so that my philosophical positioning shifted. Whilst I identify strongly with many of the assumptions of relativist and interpretivist paradigms, I also have an appreciation for the contribution of positivist approaches to the development of psychology as a scientific discipline. However, I have found that I value subjectivity, and don't believe it is possible for a researcher to be completely removed from their research so that there is no bias in the design, data or findings. I also have much more fluid concepts of what constitutes and constructs 'truth' and 'reality' than positivist approaches allow for. Therefore, the combination of my background in psychology alongside my own skills as a researcher and interest in qualitative methodology lead me to conclude that I am philosophically positioned somewhere between positivist and interpretivist approaches, with respect for the merits of both approaches. For these reasons, I consider myself to mostly think and work within the ontological paradigm of critical realism, and my epistemological approach is that of a contextualist (King & Horrocks, 2010), although when conducting quantitative research I am able to adopt a positivist approach to answer appropriate research questions.

Meeting my research aims required an understanding of how and why things happened in relationships in CFS/ME. I therefore came to understand that this research in particular required me to work within an Interpretivist paradigm, specifically that of social constructivism. I have outlined my ontological, epistemological and axiological stance below to demonstrate why:

- *Ontological stance:* I believe that there are multiple realities that can be developed through interaction between participants and researchers.
- *Epistemological stance:* I view the researcher as integral to the creation of 'reality' in research, so that reality is in fact co-created between the researcher and the participant(s).
- *Axiological stance:* I view beliefs and values as socially constructed prior to, within and beyond the research interview.

5.4.3. Constructivist grounded theory. The original Grounded Theory approach was set out by Glaser & Strauss (Glaser & Strauss, 1967). However, they had disagreements about a number of methodological issues, later going their separate ways to propose altered approaches (Glaser, 1978, 1992; Strauss & Corbin, 1998; Anselm Strauss & Corbin, 1990). More recently the constructivist approach proposed by Charmaz (2014) has gained popularity as a more pragmatic way to apply the method. Charmaz proposes that there is an 'objectivist' and therefore positivist approach to grounded theory, distinguishable from interpretivist, or ‘constructivist’ grounded theory. This study presents a blend of interpretivist and constructivist approaches to grounded theory, although epistemologically the constructivist approach most resonates with me. Although it is not wholly consistent with the constructivist approach to grounded theory, it is perhaps most closely aligned with a constructivist approach. Table 4. replicates a table presented by Charmaz (2014, p.236) clearly outlining the differences between objectivist and constructivist grounded theory, from which I worked.

Table 4. *Objectivist v constructivist grounded theory: comparisons and contrasts*

Objectivist grounded theory	**Constructivist grounded theory**
Foundational Assumptions • Assumes an external reality • Assumes discovery of data • Assumes conceptualizations emerge from data analysis • Views representations of data as unproblematic • Assumes neutrality, passivity, and authority of the observer.	**Foundational Assumptions** • Assumes multiple realities • Assumes mutual construction of data through *interaction* • Assumes the researcher constructs categories • Views representation of the data as problematic, relativistic, situational, and partial • Assumes the observer’s values, priorities, positions and actions affect views.
Objectives • Aims to achieve context-free generalisations • Aims for parsimonious, abstract, conceptualizations that transcend historical and situational locations • Aims to create theory that fits, works, has relevance, and is modifiable. (Glaser)	**Objectives** • Views generalisations as partial, conditional, and situated in time, space, positions, action and interactions • Aims for interpretive understanding of historically situated data • Specifies range of variation • Aims to create theory that has credibility, originality, resonance, and usefulness.
Implications for Data Analysis • Views data analysis as an objective process • Sees emergent categories as forming the analysis • Sees reflexivity as one possible data source • Gives priority to researcher’s analytic categories and voice.	**Implications for Data Analysis** • Acknowledges subjectivities throughout the data analysis • Views co-constructed data as beginning the analytic direction • Engages in reflexivity throughout the research process • Seeks and re-represents participants’ views and voices as integral to the analysis.

Note. Adapted from Charmaz (2014). *Constructing Grounded Theory* (2nd), P. 236.

The most salient point for me in learning more about constructivist grounded theory was reaching an understanding of what constitutes objectivism in grounded theory. If I was an objectivist, I would interrogate the data with demonstrable rigour in an attempt to make the methodology defensible when compared with quantitative methods. I would assume that in undertaking research I am discovering an external reality that exists independent of my influence and social action as a researcher; and I would assume that if I apply the published methods and steps for conducting grounded theory, then I will achieve robust theoretical explanations. Charmaz points out, ‘given these assumptions, objectivist proponents would argue for a stricter adherence to grounded theory steps than constructivists do.’ (2014, p.237).

5.4.4. Memo-writing & reflexivity. Reflexivity is a central issue in all stages of qualitative research (Braun & Clarke, 2013; Anselm Strauss & Corbin, 1990) and especially during the process of theoretical coding and theory-building (Urquhart, 2013). Firstly, it allows the researcher to reflect on how their own values and behaviours may impact upon the research process, in data collection, analysis, interpretation and reporting. Secondly, it aids the process of constant comparison, and memo-writing enables the development of theory through continuous reflection and recording of insights (Denzin & Lincoln, 2011). Reflection may be especially important for novice researchers, but conducting research is a journey of continuous improvement, rather than mastery, and reflection on one’s own thoughts and behaviours as a researcher facilitates that process (Glesne & Peshkin, 1992). Reflexivity is an important learning tool that enables insight and growth as a researcher and aids thoughtful interpretation; it also enables one to monitor their own development of thought, which is a vital process in qualitative research (Watt, 2007). Charmaz (2014) states ‘without engaging in reflexivity, researchers may elevate their own tacit assumptions and interpretations to ‘objective’ status’ (p.238).

5.5. Research methods

5.5.1. Sampling technique. A purposive sampling strategy called *maximum variation* was employed in this study, the aim of which is to capture a wide range of perspectives in order to represent the diversity of the field (Flick, 2009). However, Flick points out that sampling decisions must always involve a process of balancing width and depth of the sample or data with the availability of resources such as time and money. This sampling method was selected because it was hoped that it would enable the identification of patterns occurring across a wide range of participants and types of relationships, and the delimitation of the possibilities of the developing theories. It allowed flexibility in terms of who could participate regarding age, gender, relationship status, characteristics of the illness, length of time since illness onset, diagnosis and recovery, if any. Data collection methods were designed to enable those more disabled by their illness to participate. It also meant that in investigating SO relationships, I aimed to recruit individuals who included parents, spouses and siblings. I was also interested in hearing from those who did not have parents or spouses, or whose relationships had broken down as a consequence of their illness as I believed interesting and comparative insights could be constructed. Inclusion and exclusion criteria are outlined in Table 5. *Inclusion criteria* and Table 6. *Exclusion criteria*

This sampling strategy is particularly useful in the context of CFS/ME because of the fact that it is diagnosed in many different ways and according to several different criteria. Most should have been diagnosed following a period of illness of six months or more, accompanied by the symptoms described in the CDC definition and negative test results for any other illness that could produce similar symptoms. However, in reality many PwCFS/ME go undiagnosed for a significant period of time. Many self-diagnose or attend private clinics where they eventually receive the diagnosis. Further, it seems that many people who have received a diagnosis may have been misdiagnosed, and in many ways the diagnosis is used as a 'catch-all' for a broad range of differing illnesses that fall under the same umbrella because of the symptom of fatigue (Hyde, 2003). Therefore, attempting to recruit a homogenous sample in CFS/ME disregards the inherent subjectivity and heterogeneity of the label and illness.

Table 5. *Inclusion criteria*

Inclusion criteria	**Rationale**
Eligible participants had to be 16 or over. No upper age limit was specified.	This was a study of adults. Therefore, participants under the age of 16 were not eligible.
Fluent English speaking and listening skills.	No resources for translation available.
A self-reported diagnosis of CFS/ME provided by a health professional, or the SO of somebody with a diagnosis of CFS/ME.	This is a subjective study of the experience of PwCFS/ME and their SO. Many people who identify as having CFS/ME may not have a formal diagnosis; the intention of this study is to be relevant to those who identify as having CFS/ME. However, in requesting that participants have received a formal diagnosis, this increases the likelihood of participants meeting a formal set of criteria for CFS/ME.

Table 6. *Exclusion criteria*

Exclusion criteria	**Rationale**
People under the age of 16.	This was a study of adults. Therefore, participants under the age of 16 were not eligible.
People without fluent English speaking and listening skills.	No resources for translation available.
People who do not report that a health professional diagnosed their CFS/ME; SOs who are unable to report that their SO has a formal diagnosis of CFS/ME.	This is a subjective study of the experience of PwCFS/ME and their SO. Many people who identify as having CFS/ME may not have a formal diagnosis; the intention of this study is to be relevant to those who identify as having CFS/ME. However, in requesting that participants have received a formal diagnosis, this increases the likelihood of participants meeting a formal set of criteria for CFS/ME.
Inpatients in any kind of hospital.	For the safety and wellbeing of participants.
People with capacity to provide informed consent.	Individuals who do not have capacity to understand the information about the study and make a reasoned decision about whether or not they wish to participate cannot provide informed consent and therefore it is not ethical to include them.

Traditionally, in grounded theory, the decision about sample size and when to stop sampling should be made on the basis of *theoretical saturation* (Glaser & Strauss, 1967), and indeed theoretical saturation is what grounded theorists should aim for according to its founders (Glaser & Strauss, 1967; Charmaz, 2014). This was conceptualised by Glaser & Strauss (1967) as the point at which no new categories are being generated during analysis of data and the researcher is seeing the same comments made again and again. At this point a researcher should make the judgement about whether their categories are saturated (Glaser, 1992). However, there are varied descriptions and conceptualisations about what constitutes theoretical saturation as well as inconsistencies in its operationalisation in the grounded theory literature and in the broader qualitative literature (Saunders et al., 2018). Charmaz (2014) warns researchers that the credibility of a study is affected by claims of saturation, and that frequently grounded theorists use terms such as saturation without critical consideration of its meaning and value. These thoughts echo the views of Morse (1995) who asserts that some researchers claim saturation without providing sufficient evidence of what led them to that judgement, and indeed Saunders et al. (2018) provide more recent examples of research where this could be seen to be the case.

A challenge highlighted by Dey (1999) is the imprecision of saturation as a term and concept, which Dey suggests is because it is reliant on the researcher to reach a decision that categories are saturated by the data. Dey prefers instead to conceptualise categories as *suggested* by the data and therefore suggests a more suitable term than saturation might be ‘theoretical sufficiency’ (p.257).

A further point made by Strauss & Corbin (1998) is that saturation is not a ‘point’ to be reached as such; rather, it is about the degree of saturation achieved. They also argued that sometimes, when an elegant theory is in development, it is important to know when more data could become counter-productive because it may be limited in its ability to add anything further to the overall story the researcher seeks to portray in the theory they are developing. This perspective has since been supported by Nelson (2016*)* who agrees that the problem with the term saturation is that it infers a point of completeness to be achieved. Nelson proposes focusing instead on and whether the data collected enables the depth of understanding necessary for the development of theoretical categories. Meanwhile, it is the view of Saunders et al., (2018; p.1901) that the focus on saturation as a point in time leads to ‘uncertainty and equivocation’, particularly where researchers claim saturation and yet report having continued data collection and coding to confirm or validate their claim of saturation. Saunders et al. also point to the flawed logic of saturation as a concept because it requires a researcher to make a judgement about that which they have not observed based on what they have already observed.

In this study, a criteria for identifying similarities and differences were developed during the data collection and analysis process, so that the decision about where to sample from next could be reached, in accordance with the application of maximum variation sampling. A number of participants came forward very quickly, each of whom offered a different type of dyadic situation. Each new case sampled held the possibility of contributing new insights for the further elaboration or refinement of theoretical ideas (Flick, 2009; Glaser & Strauss, 1967). With the Constructivist approach to grounded theory (Charmaz, 2014) at the forefront of my mind, I considered the following issues when making decisions about sampling and about when to stop collecting data:

- Do I have sufficient breadth in the sample to have confidence that the data represents a range of cases and viewpoints?
- Does the data offer ample comparative cases?
- Do these comparisons offer further insight in the process of theoretical development?
- How are the theoretical categories linked, and are there further questions arising according to those links that warrant further exploration?

- If there are other directions that the categories take me in, do these categories add depth and/or breadth? If they add breadth but not depth, then do they extend the theory beyond what is useful or practical within the scope of this work?
- As the theoretical categories matured, I also asked myself would collecting further data enhance the current theory usefully, or just add more evidence of the categories I already had?

I made judgements during the selective and theoretical coding processes about which categories to focus on (discussed in Section 5.8.1.), as well as asking further questions later in the research process (highlighted at the beginning of Chapter 9), and thus whether to seek further data for the development or exploration of certain concepts and categories. There were other directions that the concepts and categories could have taken me in; for example, I could have explored a range of categories relating to day-to-day living and patient-GP relationships. However, it was my view that these categories diluted the usefulness of the theoretical development by posing the risk of spreading the story too thinly, and extending beyond the research questions. The point at which I had sufficient breadth in the sample, with ample comparative cases, maturing theoretical development, and sufficient ‘evidence’ for each theoretical category was the point at which I stopped collecting data.

5.5.2. Recruitment methods. Community-dwelling participants from Hull, East Yorkshire and North Lincolnshire were recruited via word-of-mouth, through the researcher contacting local CFS/ME support groups in the North of England, through contacts at the University, social media and online marketing. A website with information about the study was also set up. No incentives were offered for participation.

During this process, I also made multiple attempts to engage with the local NHS CFS/ME service with a view to collaborating for recruitment in order to gain a broader sample, and to gain a deeper understanding of local service provision. Contact was made via the research supervisor, by a clinical psychologist in my clinical post, and directly by the researcher in attempts to arrange a meeting to discuss collaboration. The CFS/ME service did say that they were interested in the study but subsequent attempts to arrange a meeting went unanswered and ultimately, I ceased trying.

A majority of the support in publicising the research was received from one CFS/ME Support Group in the North of England who agreed to notify its members about the study. A senior member of the group contacted other support group members by telephone if they believed they would be able and willing to participate, informing them of the study. The senior group member then contacted me by telephone with participant contact details if they had consented to them doing so. 10 of the 15 participants recruited for this study were recruited as a result of communication with this group. The rest were recruited by word-of-mouth, via university contacts, social media and online marketing and had no links to the support group.

Once participants had received information about the study, they either contacted the researcher by telephone, or if somebody else had first spoken with them and been given permission to pass their details on to the researcher, they were then contacted by the researcher by telephone. The study was discussed verbally over the telephone, following which the Participant Information Sheet (see Appendix 2), and a document detailing the procedure and a list of interview topics (see Appendices 3 & 4) were then sent to the potential participant via email or by post. Having read this information, if they agreed to participate then the first face-to-face or telephone interview was arranged by the researcher by telephone. Informed consent was obtained in writing either before or at the beginning of the first interview.

5.5.3. Participants. Participants were 15 community-dwelling adults consisting of people who had a self-reported diagnosis of CFS/ME from a health professional

(PwCFS/ME) (*n*=9) and their SOs (SOs) (*n*=6). The sample consisted of nine females and six males. This convenience sample was 100% white British who lived in or originated from the North of England. Two triads, three dyads and three individuals participated. The mean age of all participants at first interview was 51.93 years (range 21-82). Further participant demographic information is outlined in Table 7. *Participant demographic information.*

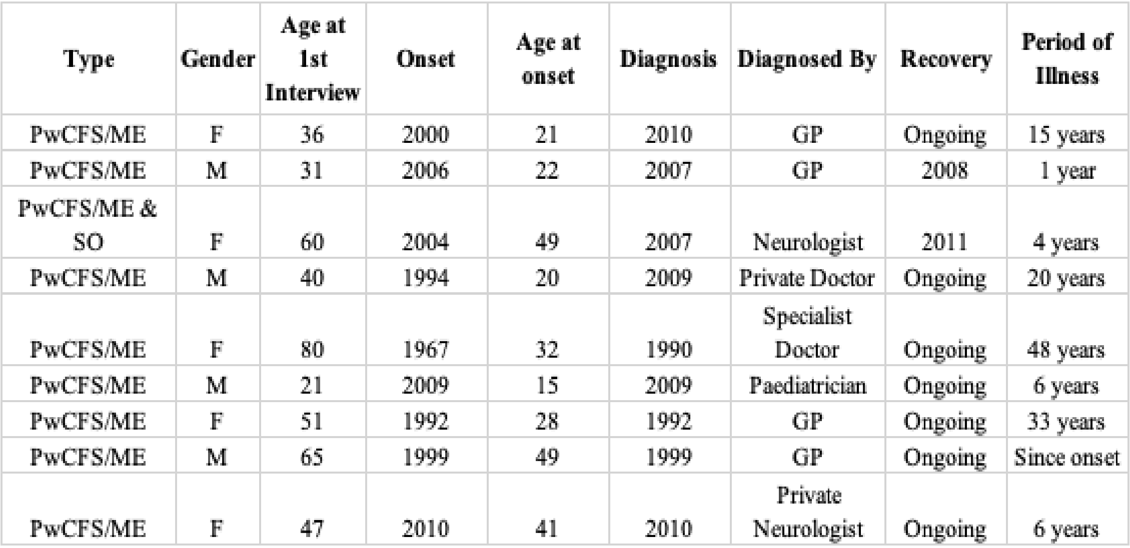
Table 7. *Participant demographic information*

Type	**Gender**	**Age at 1st Interview**	**Education**	**Ethinic Origin**	**Marital Status**	**Living Situation at time of interview**
PwCFS/ME	F	36	Postgraduate Certificate	White British	Single	Alone
PwCFS/ME	M	31	Degree	White British	Living with Partner	With partner
Participating as a PwCFS/ME & as an SO	F	60	Degree	White British	Married	With spouse
SO	M	65	Professional Qualifications	White British	Married	With spouse
PwCFS/ME	M	40	NVQ L2	White British	Single	With parents
SO	F	61	Degree	White British	Married	With son and spouse
PwCFS/ME	F	80	Professional Qualifications	White British	Married	With spouse
SO	M	82	O'levels	White British	Married	With spouse
PwCFS/ME	M	21	GCSEs	White British	Single	With Mother
SO	F	54	Degree	White British	Divorced	With son
SO	F	24	Degree	White British	Single	With friends
PwCFS/ME	F	51	Degree	White British	Single	Alone
SO	F	62	NNEB	White British	Married	With spouse
PwCFS/ME	M	65	Postgraduate Certificate	White British	Married	With spouse
PwCFS/ME	F	47	PhD	White British	Married	With spouse and two children

PwCFS/ME. Four were male and five were female. The mean age at first interview was 47.88 (range 21-80). The mean age at illness onset was 30.7 (range 15-

49) and the mean length of illness was 16.5 years (range 1-48). Further information about illness term and diagnosis is outlined in Table 8. Illness information.

Table 8. *Illness information*



Type	Gender	Age at 1st Interview	Onset	Age at onset	Diagnosis	Diagnosed By	Recovery	Period of Illness
PwCFS/ME	F	36	2000	21	2010	GP	Ongoing	15 years
PwCFS/ME	M	31	2006	22	2007	GP	2008	1 year
PwCFS/ME & SO	F	60	2004	49	2007	Neurologist	2011	4 years
PwCFS/ME	M	40	1994	20	2009	Private Doctor	Ongoing	20 years
PwCFS/ME	F	80	1967	32	1990	Specialist Doctor	Ongoing	48 years
PwCFS/ME	M	21	2009	15	2009	Paediatrician	Ongoing	6 years
PwCFS/ME	F	51	1992	28	1992	GP	Ongoing	33 years
PwCFS/ME	M	65	1999	49	1999	GP	Ongoing	Since onset
PwCFS/ME	F	47	2010	41	2010	Private Neurologist	Ongoing	6 years

SOs. All SO participants were an SO of one of the participants who had CFS/ME; five of the SOs were female and two were male. Two were mothers of PwCFS/ME, two were husbands of PwCFS/ME, one was a wife of a PwCFS/ME, and one was a sister. The mean age at first interview was 58 (range 24-82).

5.5.4. Resources and costs. This research has been funded by the University of Hull. The researcher is in receipt of a stipend as part of a PhD Studentship. There were no costs other than travel arrangements and administrative resources, all of which were covered by the stipend, studentship, and departmental office supplies.

5.5.5. Plan for reporting findings. Following completion of the PhD, it is intended that this research will lead to publication in an academic journal to be identified by the researcher and research supervisors. Participants will be provided with a lay summary of the study and its findings as well as links to academic publications.

5.5.6. Ethical considerations.

Participant rights and wellbeing. Written informed consent was obtained and participants were assured that no identifying personal information would be shared (see Appendix 1). To ensure confidentiality, participants were provided with a number prior to participation, with which their data could be identified without recording names. Participants were made aware of the risks and benefits of participation and informed of their right to withdraw for up to 24 hours following the final interview. They were also informed of their right not to answer specific questions of their choosing, and of the lack of provision for on-going support following participation. Participants were also provided with a participant information form and a debrief sheet (See Appendix 2). Within the Participant Information Sheet it was specified that only the second interview would be audio-recorded and analysed. However, this was discussed with each participant at the beginning of each interview and participants articulated that they believed it was valuable to also capture the content of the first interview as an important aspect of the narrative. Every participant gave verbal consent for each interview to be audio-recorded, transcribed and analysed. The use of the anonymised data in future academic publications and dissemination was also discussed with participants and verbal consent was provided.

There was a small risk of physical and emotional distress to the participants through participants becoming tired due to spending time talking, and due to talking about potentially emotive topics. The researcher discussed with participants prior to and during interviews how they could prepare themselves physically and emotionally for each interview and what they might do following the interview to minimise any physical discomfort. A number of participants did become distressed during the interview; they were offered the opportunity to stop the interview and given time to recover in line with their needs. They were also sufficiently debriefed following both interviews by the researcher and were provided with contact details for Let's Talk Hull, Relate, Samaritans, NHS Direct and Carers UK where they were able to access further support for issues raised during the study. An agreement was reached between the researcher and research supervisor that if participants disclosed any risk, the researcher would inform the research supervisor at the earliest opportunity so that they could decide together about the best course of action, in line with University and NHS Safeguarding policies. This was not necessary. The researcher received support from the research supervisor throughout the study and was able to discuss any difficulties, ethical concerns and emotional consequences of the work in the confidential, professional and supportive sessions conducted between the researcher and research supervisor.

Ethical approval. Ethical approval was granted by the Research Ethics Committee within the Department of Psychology at the University of Hull on 9th March 2015 (Reference number 490152-1423848796).

5.5.7. Data management during the project.

This data management plan is also documented in the University of Hull Generic Data Management Plan proforma, completed by the Researcher. This is a working document and is therefore attached at the end of this thesis as Appendix 17. The key features of the data management plan are outlined below.

Hard Data. Personal data and participant consent forms were stored in paper form only, in a locked drawer, in a locked filing cabinet, in a locked office at the University of Hull. Copies of transcripts were printed out and annotated as part of the research process. These were stored in an office in the Researcher's home during the data analysis phase. Upon completion of the data analysis, these were taken to the

Researcher's locked office at the University of Hull where they are stored in a locked filing cabinet.

Digital Audio Data. Audio recordings were made on a MacBook Computer with Firewall and FileVault enabled. These were immediately transferred onto an encrypted hard drive belonging to the Researcher, and all other copies were deleted. Audio recordings of interviews were then stored on this encrypted hard drive. Audio recordings will remain on the hard drive until the thesis has been submitted and accepted by the examiners as complete, at which point the audio recordings will be permanently deleted.

Digital Written Data. Anonymised research data (transcripts) and NVivo files were stored in password-protected files, on a password hard drive, accessible only to the researcher, the research supervisor, and two university-approved research associates. The Research Associates received training from the Researcher in safe data management and confidentiality in line with the procedures outlined in this data management plan. When documents (Word documents and NVivo files) were in use, this took place on a MacBook Pro belonging to the Researcher with FileVault, Firewall, and antivirus software enabled, or on University Desktop PCs on the University Campus on the Researcher's personal University drive. When documents were in use by the research associates, they were transferred via the University Box file storage system and were briefly stored on password-protected personal computers belonging to the research associates. Once completed and returned to the researcher via Box, they were deleted from the research associates' computers, and downloaded onto the researcher's encrypted hard drive and deleted from Box. Memos and analytical writing was stored as password-protected files, on an encrypted hard drive belonging to the researcher.

Back-up. All of the above documents were backed up on a secure, access-restricted personal drive on the University of Hull network.

5.5.8. Long-term data management plan.

These procedures are also documented in the University of Hull Generic Data Management Plan proforma (Appendix 17).

Hard Data. All hard data (consent forms, personal data, field notes, copies of memos) will be stored in a locked filing cabinet, in a locked office belonging to the Researcher in the Department of Psychology at the University of Hull. Should the Researcher leave the University, this data will be handed over to the Department of Psychology for safe storage for six years after the end of the PhD.

Digital Data. All audio data will be destroyed by being deleted from the encrypted hard drive and university server upon completion of the PhD. Following acceptance by the examiners of this thesis as complete, the anonymised data (transcripts) and NVivo files will be uploaded onto a private folder as encrypted, confidential documents on the cloud storage system, Figshare. The Researcher will have responsibility for ensuring this data is destroyed six years after the end of the PhD. Once stored on Figshare, all data stored on the hard drive and backed up on the Researcher's personal university drive will be fully deleted.

5.6. Procedure

5.6.1. Data collection plan. Interviews were chosen as the method of data collection over alternatives such as focus groups due to the confidential and intimate nature of the topics I wished to explore. I concluded that participants may feel more comfortable talking about their relationship experiences either alone or with their SO than they might in a group setting. Each participant was interviewed twice. The main purpose of the research was to focus on experiences of PwCFS/ME/SO relationships in the context of CFS/ME, and this was the focus of the second interview. The first interview was added to the research design following consultation with PwCFS/ME during the research design process. PwCFS/ME were found to be keen to share the details of their illness, path to diagnosis and engagement with health professionals, and to explain what life was like before their illness. Whilst these issues first appeared not to be directly related to the research question, it became clear that it was necessary to allow space for exploration of these topics in order to gain a picture of the context within which the PwCFS/ME and SO relationship operates. The first interview was added to the procedure in order to develop rapport and provide the platform to explore issues important to participants through the trajectory of the illness. The second interview focused on the relationship between the participant and their SO in response and relation to CFS/ME. A minimum of two weeks between interviews was required based on participant availability, but no maximum time frame between interviews was stipulated. Second interviews needed to be arranged flexibly based on participant needs.

5.6.2. Interviews conducted. A total of 26 audio-recorded semi-structured interviews were conducted. 25 of these were undertaken within a 10-month period between May 2015 and February 2016. One final interview was undertaken 9 months later in November 2016 as the final participant had become unwell shortly after her first interview and wanted to wait until she felt emotionally strong enough to complete her second interview. 14 participants completed two interviews and one participant completed one interview only (Sarah, sister of Sean). This was because she was not present at the first interview, but upon arriving at the second interview she was present and wanted to participate. As she would be the first sibling to participate, I felt this was a good opportunity to broaden the sample in line with the sampling approach so I agreed to her participation.

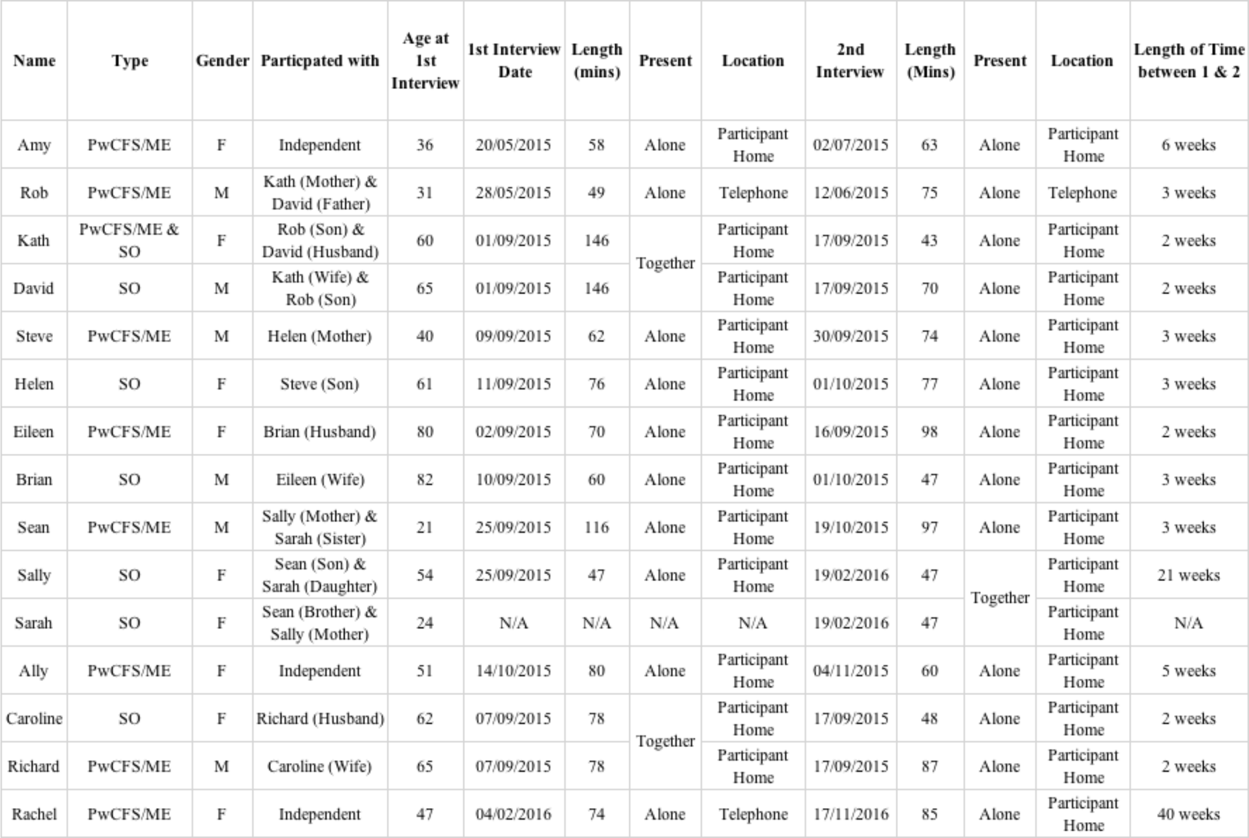
5.6.3. Interview location and attendees. Interviews took place over the telephone (*n*=3) or face-to-face in the participant's home (*n*=23). Two participants preferred the interviews to be undertaken over the telephone, one because of geographical location (he was from the East Riding but lived in Birmingham at the time of the interview) and the other because of time commitments. Some interviews were conducted in pairs (n=3) and the rest were conducted with individuals (*n*=23). Two dyads chose to be interviewed together for the first interview but, in order to ensure both members of the dyad contributed equal amounts of interview content in the second interview, it was requested by the researcher that they undertake the second interviews separately. As previously explained, one further participant (Sarah, sister of Sean) joined her mother (Sally) at the second interview only.

5.6.4. Interview length. Prior to interview, participants were briefed that each appointment was expected to last approximately one hour, but that this was flexible based on their needs. The duration was dependent upon the flow of the conversation and was predominantly participant-led, although the researcher made efforts to move the interview along at an appropriate pace using the topic guide. Interviews lasted a mean average of 83.86 minutes (range 43-146 minutes). The mean length of first interviews was 95 minutes, (range 47-146 minutes). The mean length of second interviews was 72.71 minutes (range 43-98). This time did not include the non-recorded discussions at the start and finish of each appointment. During the earliest interviews, in an attempt to allow the interview to be as participant-led as possible I did not always manage the structure of the interview well enough in order to ensure the content was relevant to the research question. I allowed several of the interviews to go off on tangents which meant that large portions of the data were not as useful in addressing the research question, although all data was transcribed and analysed. At later interviews, I managed the interviews gently to keep them on course.

5.6.5. Time between interviews. The intended length of time between interviews 1 and 2 was approximately 2-3 weeks, but was dependent on participants' needs and availability. The mean length of time between interviews with all 15 participants included was 6 weeks (range 1-40). However, there were two participants for whom the gap was especially long (Rachel, 40 weeks and Caroline, 21 weeks). In both cases, this was because the participants cancelled their second interview due to illness and then said they would get in touch when they felt well enough and had availability. Excluding these participants, the mean length of time between interviews was three weeks (range 1-6).

Further details of interview dates, times, lengths between interviews, and who they participated with are summarised in Table 9. *Interview information.*

Table 9. *Interview information.*



Name	**Type**	**Gender**	**Particpated with**	**Age at 1st Interview**	**1st Interview Date**	**Length (mins)**	**Present**	**Location**	**2nd Interview**	**Length (Mins)**	**Present**	**Location**	**Length of Time between 1 & 2**
Amy	PwCFS/ME	F	Independent	36	20/05/2015	58	Alone	Participant Home	02/07/2015	63	Alone	Participant Home	6 weeks
Rob	PwCFS/ME	M	Kath (Mother) & David (Father)	31	28/05/2015	49	Alone	Telephone	12/06/2015	75	Alone	Telephone	3 weeks
Kath	PwCFS/ME & SO	F	Rob (Son) & David (Husband)	60	01/09/2015	146	Together	Participant Home	17/09/2015	43	Alone	Participant Home	2 weeks
David	SO	M	Kath (Wife) & Rob (Son)	65	01/09/2015	146		Participant Home	17/09/2015	70	Alone	Participant Home	2 weeks
Steve	PwCFS/ME	M	Helen (Mother)	40	09/09/2015	62	Alone	Participant Home	30/09/2015	74	Alone	Participant Home	3 weeks
Helen	SO	F	Steve (Son)	61	11/09/2015	76	Alone	Participant Home	01/10/2015	77	Alone	Participant Home	3 weeks
Eileen	PwCFS/ME	F	Brian (Husband)	80	02/09/2015	70	Alone	Participant Home	16/09/2015	98	Alone	Participant Home	2 weeks
Brian	SO	M	Eileen (Wife)	82	10/09/2015	60	Alone	Participant Home	01/10/2015	47	Alone	Participant Home	3 weeks
Sean	PwCFS/ME	M	Sally (Mother) & Sarah (Sister)	21	25/09/2015	116	Alone	Participant Home	19/10/2015	97	Alone	Participant Home	3 weeks
Sally	SO	F	Sean (Son) & Sarah (Daughter)	54	25/09/2015	47	Alone	Participant Home	19/02/2016	47	Together	Participant Home	21 weeks
Sarah	SO	F	Sean (Brother) & Sally (Mother)	24	N/A	N/A	N/A	N/A	19/02/2016	47		Participant Home	N/A
Ally	PwCFS/ME	F	Independent	51	14/10/2015	80	Alone	Participant Home	04/11/2015	60	Alone	Participant Home	5 weeks
Caroline	SO	F	Richard (Husband)	62	07/09/2015	78	Together	Participant Home	17/09/2015	48	Alone	Participant Home	2 weeks
Richard	PwCFS/ME	M	Caroline (Wife)	65	07/09/2015	78		Participant Home	17/09/2015	87	Alone	Participant Home	2 weeks
Rachel	PwCFS/ME	F	Independent	47	04/02/2016	74	Alone	Telephone	17/11/2016	85	Alone	Participant Home	40 weeks

5.6.6. Interview structure. A topic guide (see Appendix 3.) relating to the research question was drawn up and given to participants prior to the first interview in order to prepare them for what they would be asked to talk about, and give them time to think about and begin to construct their own understanding of their experiences prior to the interview, especially useful because of the cognitive difficulties that some

PwCFS/ME can experience. At the end of the first interview, participants were given a topic guide for the second interview, provided in advance of the second interview for the same reasons as for the first interview (see Appendix 4.). They were also provided with instructions for completing a spider diagram (Appendix 5.) around the proposed interview topic guide in the two to three weeks between the first and second interviews.

5.6.7. Spider diagram. Most frequently, innovative interviewing methods are used in research with children, young people and families. Graphic elicitation has been acknowledged as having the potential to encourage interviewees to elicit contributions that by other means may be more difficult to obtain (Crilly, Blackwell, & Clarkson, 2006; James, Jenks, & Prout, 1998). For this reason, I asked participants to develop a spider diagram based around the suggested topics for the second interview, in between the two interviews. A spider diagram is also known as a ‘mind map’ and involved using topics written on paper as the starting point for exploration of thoughts and ideas relating to them, with the ability to explore further each idea as it arises. Participants were given instructions (See Appendix 5) that asked them to use the topic guide as a prompt for them to think about their own relationship experiences in the context of CFS/ME. 13 of the 15 participants completed this task with the exception of one who only attended a second interview, and another who drafted a written report under the topic headings instead of a diagram. Some of the participants gave their spider diagrams to me at the end of their interview although this was not a requirement and was at the participant’s discretion. Examples are included in Appendix 6. The preparation of the spider diagrams allowed the participant opportunities to consider the topics of the second interview in between the first and second interview. The intention was that this may lead to deeper, more considered insights from the participant's perspective, and that the ideas they articulated through the use of the spider diagram would be more stable and consistent, having been mulled over for an extended period of time. It also served to create structure in a complex and emotive topic; many participants disclosed that they found it cathartic to create a record of their thoughts and feelings. This is consistent with Holstein & Gubrium's (2004) assertion that an interview can be 'a site of, and occasion for, producing reportable knowledge' (p.141).

5.6.8. Field notes & memos. I took handwritten field notes during the interview including recording participant demographic information and highlighting key points that stood out as important. This also enabled the recording of questions about the data as it being collected in comparison with other participant data. I maintained a reflexive journal throughout the research process. Memos were also used immediately following interviews as a means of recording immediate ideas and reflections on the interview and to begin to develop theoretical exploration. Examples of field notes, journal notes and memos are included in Appendices 7, 8 and 9 respectively.

5.6.9. Audio recordings. All 26 interviews were recorded using software called Logic Pro on a MacBook Pro belonging to myself, the researcher, with the verbal consent of participants at the beginning of each interview. Following the interviews, the Logic projects were bounced into MP3 audio files ready for transcription. One interview could not be bounced into an MP3 as the audio file was corrupted due to a recording fault (the first interview with the first research participant, Amy, aged 36). The data from that interview was therefore not usable and the participant was informed at the second interview.

5.6.10. Transcription. 25 of the 26 interviews were transcribed following the corruption of one recording. Transcription was undertaken in MS Word and in some cases Dragon Dictation software was used. Two voluntary Undergraduate Research Associates were appointed within the Department of Psychology to assist in the transcription of interview data. I provided them with training and guidance to ensure they adhered to confidentiality and ethical requirements, knew how to use Dragon, and to help them understand the research and transcription process. Initially, it was intended that only the second interview would be recorded, transcribed and analysed for the purpose of the PhD because it was thought that within this interview would exist the data most relevant to the research question. However, in discussion with the PhD supervisor prior to the first interview, it was agreed that the topics explored in the two interviews could not really be meaningfully separated. Therefore, this was discussed with participants at the beginning of each interview and consent provided to record, transcribe and analyse both interviews by all participants. As ethical approval for this study was required at a departmental level and specific details of the research design were not required as part of the ethical approval process, no ethical amendments were required in order for this decision to be taken.

5.7. Data management strategies

A qualitative data analysis software (QDAS) package called NVivo was used to assist the data analysis process. There are a number of QDAS packages available; however, NVivo was available free of charge through the University of Hull, as was an introductory course in its use in qualitative research, which I undertook. NVivo was initially developed by qualitative researchers and it lends itself well to GTM in terms of its functionality (Bazeley & Jackson, 2013). It is important to note that it is not possible for software to perform qualitative data analysis; a researcher must rely on their skill, knowledge of and engagement with the data, and their ability to work with the data and software within the context of their chosen methodology (Coffey & Atkinson, 1996). Whilst the processes of coding depend upon the recommended procedural practices within a given methodological choice, the processes employed in the use of NVivo to organise and interrogate these codes and data files are common to all qualitative methods. A researcher is therefore able to learn about the general principles of using NVivo and then apply the principles to their own project (Bazeley & Jackson, 2013).

I used NVivo to undertake the initial line-by-line coding process (outlined in section 5.8) However, in later stages of the analytic process, I preferred to move on to paper and consequently printed out transcripts and coded them by hand. The reasons for this decision are described in more detail in section 5.8.1 *The coding process.*

5.8. Data analysis process

In keeping with constructivist grounded theory methodology as described by Charmaz (2014), it was originally intended that each interview would be transcribed and analysed before embarking on another interview, so that the findings from an interview can be allowed to shape the questions and highlight issues worthy of further exploration in time for the next interview. In this way, a researcher allows emerging concepts within the data to become a focus as further interviews take place.

However, as discussed earlier, an influx of willing participants wished to participate in a very short space of time, each of whom was a suitable participant within the maximum variation sampling strategy. Despite efforts, it became impractical to keep up with transcribing and analysing the data at the rate at which participants were willing to participate. Transcription and analyses were undertaken simultaneously, but not completed in time for each new interview. In spite of this, the use of memos, field notes and my proximity to the data as the researcher meant that although the formal coding process had not taken place with all data, informal concepts were emerging during reading and transcription process, and I was able to utilise this and allow it to shape the new interviews I conducted. I made links and drew comparisons during interviews and between them using memos and journalling, and continued to do this as coding was undertaken.

Other key characteristics of GTM to which I adhered in this study include the following:

- *Theory-building.* My intention was to work inductively, building theory from the data upwards by constructing and relating concepts.
- *Theory grounded in the data.* Other than my background as a psychologist and personal experience with CFS/ME, I tried not to apply preconceived ideas to my research. I monitored this via reflexivity (discussed below). It was necessary to undertake a review of literature in order to develop a rationale for undertaking research, but upon reaching a decision about this, I stopped engaging with theoretical literature until I reached the point of theoretical coding.
- *Emergent concepts that lead the process.* Further data collection was based on emergent concepts. Data collection ended when no new conceptualisations were emerging in the analytic process.
- *Systematic.* Although the processes of analysis and conceptualisation did not happen in the way it was originally planned, analysis was systematic, and began as soon as data was available. Analysis and conceptualisation did take place simultaneously via a process of constant comparison which is central to GTM. The discussion of the coding process below explains in more detail how this was undertaken.
- *Theoretical memos.* A series of data-set specific memos were kept during the research process. In addition, further memos were written around particular concepts of interest emerging in the data as well on issues requiring further reading.
- *Reflexivity.* A research journal was also kept detailing the analytical process itself. Initially, all memo-writing and journalling was undertaken within NVivo enabling it to be easily linked to codes, concepts and data sets and to be easily retrieved when reconsidering any of these. Later in the process, when the coding process moved to paper, these were instead completed using MS Word.

5.8.1. The coding process. The main strands of GTM outlined earlier differ in the recommendations of procedures and the guidelines within which one ought to operate if one is to undertake a 'true' grounded theory study. In reality, qualitative research and grounded theory lend themselves well to flexibility, adaptation and pragmatism. It can be applied according to the aims and requirements of the researcher and the research. As such, I discussed whether my approaches were acceptable with the research supervisor at each stage. However, as a novice researcher and grounded theorist, it is important to have an awareness of the main strands so that educated decisions about how to undertake the research and the analysis can be made in line with traditional expectations of the application of GTM.

The focus of grounded theory is to develop a substantive theory through the identification of causal relationships between categories (Charmaz, 2014; Glaser & Strauss, 1967). In their 1990 book, Strauss & Corbin presented one coding paradigm and warned that if researchers do not use this one paradigm, their analysis will be lacking in density and precision (Strauss & Corbin, 1990), although they did backtrack somewhat on this insistence some years later (Strauss & Corbin, 1998). Following the split of Glaser and Strauss in 1990 (Urquhart, 2013), Glaser went on to place emphasis on allowing concepts to emerge and not forcing data into preconceived ideas, thus offering a range of options for relating emergent categories (Glaser, 1992). This enables a broader way of thinking about research data and the phenomena of interest than may be found within the Straussian tradition of grounded theory (Urquhart, 2013). My work was initially guided following the Glaserian tradition but with a degree of pragmatism, but the major influence was from the constructivist approach recommended by Charmaz (2014). I undertook three stages of coding: open, selective and theoretical.

Open coding. This is the process by which codes are applied to the data as ideas are constructed based on the data. Ideally, the researcher should try to undertake this in an openminded way, and take steps to limit the influence of preconceived ideas or assumptions, such as memo-writing and reflection. In this way, the data is allowed to breathe so that the categories that are developed are based on what the data actually tells us, rather than what the researcher thinks it should tell us.

In keeping with Glaser's recommendations around open coding (Glaser, 1978), I began by coding the first 5-6 sets of data in every way possible. Whilst I kept the research aims and question in mind throughout, I attempted to allow other stories to be told by the data so that the potential for identifying future research directions was maximised. The process involves labelling segments of data, be it words, lines, phrases or paragraphs, with 'codes' deemed appropriate by the researcher. As these codes begin to build, the researcher then begins grouping these codes into larger codes. The purpose of this process is to identify what is important in the research based on the data. I undertook this process through 'line-by-line' coding; that is, to work through each data set line-by-line and code any idea that emerges, regardless of its relevance to the research question. Examples of my line-by-line coding are provided in Appendix 10. This is a time-consuming process, but is an essential initial step in the early stages of a grounded theory study in order to avoid premature framing of problems and areas of interest. It also ensures that as a researcher you get to know your data intimately and your findings can be easily defended due to the fact that you know your data in great depth. As I moved on through the data sets, it became less necessary to conduct coding on a line-by-line basis. At this point, I moved into selective coding. Appendix 11. *Early coding structure* outlines the major codes and concepts that had been developed at the point at which I moved away from line-by-line coding and into selective coding.

Selective coding. This process begins once core categories have become established and the focus of subsequent coding is to flesh-out the related categories. The open coding process opens so many potential avenues of exploration, that the process of selective coding does require the researcher to become comfortable with making decisions about which aspects of the analysis to fully exploit and which to bypass at this stage, as it becomes impossible to explore and report every option that your data presents. In my work, I found that there were a number of important categories emerging, each of which could have become a core category. However, in order to refine my thinking and analyses, I returned to my research question and research aims so that decisions could be made about which thread to follow in the data. In doing so, I was able to formulate a theory grounded in both the research question and the data.

It was during the selective coding phase that I moved away from using NVivo and started coding by hand with highlighter pens and annotations, post-its and cut-outs of data. Examples of these are provided in Appendix 12. The reason for this move was that as I began coding selectively, this became an increasingly creative process and less systematic and process-driven. I started to feel frustrated and limited by the linear way in which the data was visible on a computer screen. I wanted to work more directly with the data, to be able to compare and contrast excerpts more easily, to move through the data and codes in a more tangible way, and to have a clearer sense of the structure of the data. I was finding that the process of viewing the data through a computer monitor was limiting my ability to think and work creatively with the data. By printing transcripts and code lists, I was able to draw and make links between codes, concepts and data excerpts; I used highlighters and handwritten notes to annotate reflections and ideas alongside the data as well as specific codes. I used scissors and large sheets of paper to cut and stick sections of data and memos. In this way, I was able to spend considerable time comparing codes to one another and grouping them into meaningful categories.

During this phase of coding I drew on my personal understandings and interpretations, and I also found the software limiting in my ability to develop conceptualisation whilst ensuring I stayed grounded in data. At this point, I started to draw conceptual maps and integrative diagrams – during the analytic process these were drawn and redrawn multiple times as my thinking evolved. These maps were pivotal in helping me to condense my ideas, understand how concepts were related to one another and to the research question and then communicate these with the research supervisor. I have included a selection from different stages in the research process in Appendix 16.

It was also during the process of selective coding that the decision not to undertake any further interviews was reached between the Research Supervisor and the Researcher. This was because new codes that were emerging from new data served only to further evidence the categories that had already been constructed, and offered no new insights. Categories were already well-evidenced through previous coding.

Theoretical coding. Theoretical coding according to Glaser (1978) is the stage in the analytical process within which links are established between categories composed of many interrelated codes. The relationships between codes and categories are examined in order to build theory. During this process, theoretical memos are extremely useful for recording ideas, cross-referencing between codes, and developing insight (Charmaz, 2014; Glaser, 1978). Examples of theoretical memos are provided in Appendix 13.

5.8.2. Supervision. Throughout the data collection and analysis process, I met with the research supervisor on a regular basis (once per month on average). During our meetings, I brought data excerpts which we discussed, coded and compared. I discussed my process and coding with the supervisor and he advised on literature with which I might engage. We engaged in reflexive discussions about the data collection and analysis process. An example of a supervision record from the 6-month review meeting is included in Appendix 14. We used creative strategies to generate insights about the data including the use of prompt cards to interrogate the data, and an online program called RepGrid which allows for exploration of concepts and ideas, and exploration of relationships between concepts. Notes from this are included in Appendix 15.

5.8.3. Theorising and writing. I began writing the thesis as soon as I started the analytic process, but this took many different forms before it evolved into the form it now takes. When I reached the point at which I had developed a collection of connected concepts, I met with my research supervisor with the specific purpose of reviewing these. I had drawn out another set of conceptual maps that encapsulated everything I had come to understand and wished to communicate about the participants' stories. We spent time reflecting on these, moving through them and considering how the concepts related to one another. Following this, I returned to writing with renewed vigour and a more refined model structurally. The concepts that I had defined at earlier stages remained but many were combined and/or re-named according to the new structure and new links we had drawn between them. During this phase of the writing process, I expanded my understanding of psychological formulation and began to explore clinical applications of psychology such as systemic theory. This process requires theoretical integration of existing theory with my own ideas drawn from the data. The theory presented in the finished thesis took its final shape during the writing process as I continued to engage with a range of literature in a way that was led by the data and my ideas about it. The final theory presented and evidenced in the following chapter is a refined version of the many ideas constructed throughout the process of research. The concepts from which it has been constructed converged and were reshaped many, many times through the systematic, rigorous, yet creative and complex process that I have discovered grounded theory to be.

6. Results: Overview

6.1. Introduction

This chapter sets out my findings alongside relevant discussion with reference to literature. The purpose in setting it out with integrated discussion is to maintain the flow of the narrative, and allow for exploration of specific concepts alongside consideration of broader, overarching theories that are of relevance throughout. The decision to provide a summary of key theories and integrated discussion was driven by grounded theory methodology. The literature chapters presented the literature with which I engaged prior to undertaking the research. However, much of the theory and literature discussed alongside the findings were data-driven explorations. I conducted analysis, constructed concepts and then returned to literature and theory to understand where these data-derived concepts fitted into the wider discussion in the literature.

In presenting my findings, I first present an overarching ‘formulation of the problem’ which introduces the challenges and experiences of participants within a psychologically informed framework. This is followed by a diagrammatic representation of my grounded theory that arises from this problem formulation, and a brief overview of each concept within it. Before moving on to present evidence and discuss each concept, I provide a brief summary of several psychological theories to which I will refer frequently throughout my presentation of the findings, particularly in relation to the Problem Pattern as defined in my theory. Following the next section which provides the formulation of the Problem & introduction to the model, the results chapters are structured as follows:

Category A: Onset

Category B: Core Category - The Problem Pattern

Category C: Balance

Category D: Acceptance

6.2. Formulation of the problem

6.2.1. Approach to formulation. I opted to use a psychological formulation approach as a guide for systematic thinking about the data as a whole when formulating the problem. Doing so offered a framework for constructing and presenting the problem and aided the process of linking concepts, offering a logical structure and aligning the work with clinical practice and applied psychology.

Considering the volume of research in CFS/ME that draws on the CBT approach, and the subsequent research into CBT as a treatment for CFS/ME, a CBT approach to formulation may have made sense. However, I felt it would be unlikely to reveal original insights. Consequently, I investigated a range of approaches to formulation in order to identify the best fit for my research aims and the type of data I had collected.

I settled on a systemic formulation framework (Dallos & Stedmon, 2006) which places emphasis on meaning and recognises language as central in the construction of understanding between individuals. Within this framework, the role of the therapist is viewed as an integral part of this construction, which is aligned with social constructivism and indeed a constructivist grounded theory approach (Charmaz, 2014). My leaning towards social constructivism and the systemic nature of interviewing dyads lead me to conclude that a systemic approach was a good fit (Johnston & Dallos, 2006). This is not a true systemic formulation, which would usually be developed in the context of family therapy. In clinical practice, a trained team of therapists would ideally work with several members of a family in a clinical setting to formulate collaboratively. I am not a therapist, I have no formal systemic training and this formulation has not been constructed in therapy or developed collaboratively with participants. However, borrowing from a systemic approach to formulation enabled a systematic, thorough yet concise description of the overarching problem and findings. During analysis and writing of the findings, I did not apply a systemic framework to coding and theorising as I felt this would be constricting and would not allow creativity in the analytic process, but systemic theories were held in mind and compared with the concepts I constructed.

6.2.2. Deconstructing the problem. The research question set out to explore the relationships between PwCFS/ME and their SO in the context of CFS/ME. CFS/ME can be understood to be both an individual and interpersonal difficulty. This is because of its impact on the physical and mental wellbeing of the individual and the subsequent impact on the family. I came to view the PwCFS/ME as having been forced to relinquish previous roles and responsibilities, whilst others had to step in to support the PwCFS/ME and sometimes to take up the relinquished roles of the PwCFS/ME. As the illness progressed, roles changed gradually and previously constructed individual, dyadic and family identities seemed unwillingly broken down and grudgingly reconstructed in the process of adaptation to illness and disability, to the dismay of the PwCFS/ME and those close to them who often experienced a deep sense of loss.

Contextually, the narratives were constructed within the home environment and rooted within the family lives of the participants initially. During the analytic process, I was able to locate these narratives, and the meanings communicated within them as I interpreted them, in the contexts and assumptions of broader social networks and conventions, ideologies, functional institutions, and to a lesser extent, national and global factors. In brief, the dyads under study experienced significant and often distressing changes in their lives and relationships as a consequence of an illness for which there is no known cure. There are limited treatment options with inconsistent availability and inconclusive evidence for successful outcomes. Participant perceptions were that the medical establishment did not really know what to do with them and research suggests that it ultimately deligitimises their suffering, eventually rejecting them (Dickson et al., 2007). Meanwhile, participants articulated a sense that society is confused and ill-informed about CFS/ME, imposing ignorant views and opinions upon PwCFS/ME.

6.2.3. Problem-maintaining patterns and feedback loops. Many participants explained that they had experienced significant stress in their lives prior to illness onset and it is therefore fair to assume that the dyadic relationship may have already been affected by this. However, I concluded that it was only following illness onset that the roles of carer and cared for were fully adopted, and this was the point at which the presenting problem under study evolved. PwCFS/ME faced a gradual and devastating loss of the life they previously knew; SOs grappled with the difficulty of trying to support the PwCFS/ME without angering them or worsening their condition in any way. I theorised that it became difficult for the SO to strike a balance between doing too much and not doing enough, so as to support constructively and not disable and disempower, and that in these conditions, tensions arose. Some dyads appeared never to have found this balance, others handled it by putting emotional distance between them. Destructive but often unspoken patterns of anger, blame, guilt and resentment were apparent on both sides of the dyad that I considered to be stuck in this pattern.

The PwCFS/ME's coping behaviours mirrored this in trying to balance doing too much and not doing enough – this has been constructed and reported in much previous research and is well-known as the 'boom & bust' cycle (Moss-Morris, 2005). It was described and labeled as such by participants, illustrating both the commonalities in the illness across the sample, but also demonstrating the importance of language in shaping how participants constructed their narratives retrospectively about their patterns of behaviour and experience.

6.2.4. Constraining belief systems & narratives. With demands on both members of the dyad – physical, emotional, financial, practical – many relationships in the context of CFS/ME could be interpreted as becoming problem-saturated, as could their narrative of their lives as individuals, as a dyad, and as a family. This in turn served to exacerbate the negative and often depressive emotions experienced by the PwCFS/ME. The PwCFS/ME frequently reported suffering perpetual, disabling physical symptoms. There were clear emotional implications and practical limitations that accompanied the sudden onset of disability and loss of independence. With uncertainty as to whether this loss was temporary or permanent, it appeared difficult for individuals to integrate or adapt to their new circumstances. Meanwhile, if their relationships were able to be maintained, the SO's life was also deeply influenced by the PwCFS/ME's illness. They tried to support the PwCFS/ME and the family whilst also maintaining their responsibilities such as work, all the while watching the PwCFS/ME's illness developing, and often feeling powerless to help. SOs described experiencing distress as a consequence and these difficulties extended into the broader family dynamics as both the PwCFS/ME and their SO engaged in coping behaviours and relational patterns that affected the functioning of the family system.

As they sought to develop their own narrative around what they were or had been experiencing, the PwCFS/ME attempted to make sense of their illness despite generally inconclusive explanations from healthcare professionals. Many appeared to become focused on their perception that their experience of physical symptoms indicated an organic cause for their illness that just had not yet been found, and they searched for evidence confirming that their illness was 'physical'. Previous research suggests that this belief is counterproductive and predicts less favourable clinical outcomes (Moss-Morris, 2005).

6.2.5. Transitions, emotions and attachments. At the core of the stories told by participants lay their depictions of the emotional impact of becoming disabled and cared for (PwCFS/ME) or becoming a carer (SO). Meeting each other's needs within the dyadic relationship became uncharted territory and a challenging process of trial and error. Normal patterns of relating and attachment were put under enormous strain, and fractures in a range of relationships in the lives of participants seemed common, especially where they were met with less-than-supportive family members, friends, colleagues, GPs, or authorities. I suggest that for the PwCFS/ME this led to increased levels of social isolation and withdrawal, whilst for the SO it caused ripples across their life as they tried to maintain their levels of functioning in the face of a ruptured family system and a sense of bewilderment about their loved one's situation. Some SOs maintained social lives and roles in the community, but many described a significant reduction in their commitments beyond the relationship in order to meet the demands of their situation at home, thus impacting upon their sense of self.

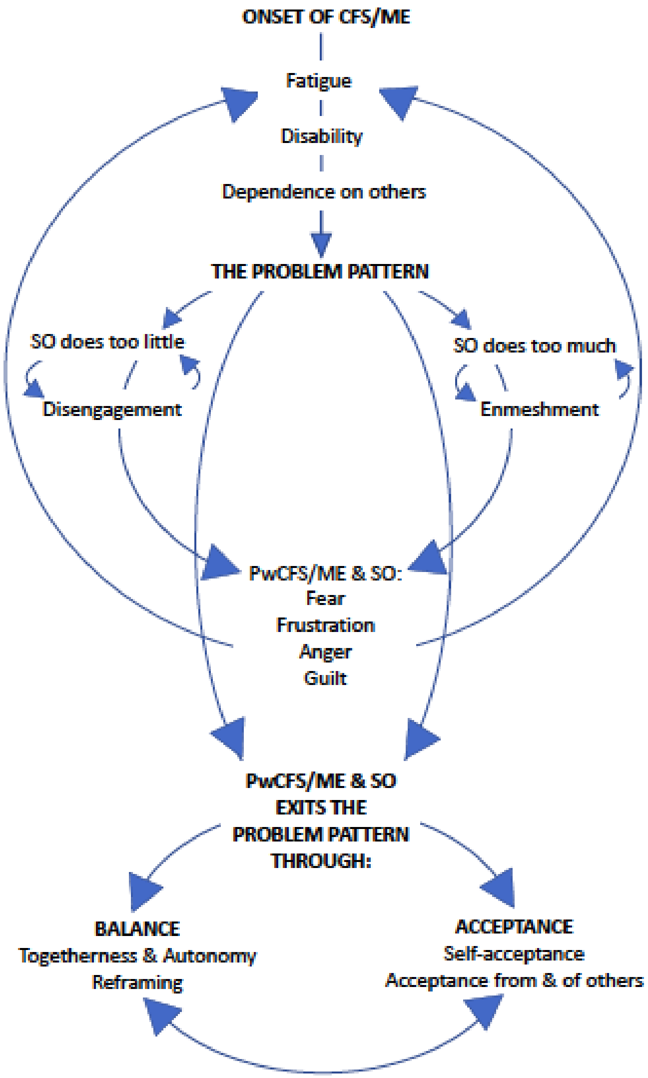
In participants' narratives I found what I perceived to be expressed, but often suppressed, feelings of guilt and resentment towards each other and towards the world beyond the dyad, as well as anger and frustration. These emotional experiences and their causes are broken down and described alongside an analysis of the relationship between the members of the dyad.

6.2.6. Exceptions. Some participants spoke of healthy, happy, loving relationships that eased their burdens and were conducive to living a good life following or in spite of illness. Even in relationships where this was not the case, many participants spoke fondly of holidays and days out as an exception to or escape from their struggles, and these emerged as a time of altered perspectives amongst participants due to the break from their perceived reality. It also became clear that in dyads where communication was portrayed as being positive and constructive, both members had been able to rebuild, rediscover or maintain a sense of independence and autonomy. Many also spoke of this being possible due to a process of reflection reframing and acceptance. Where acceptance appeared to have been reached or was a recognised work-in-progress, participants communicated having achieved a level of balance in supporting/being supported. Positive emotional and relational experiences were communicated, and participants appeared to be finding ways to live within the limitations of illness. These experiences are analysed and represented in Concepts C. Balance and D. Acceptance, in which stories of joy, love and acceptance within the limitations of illness were expressed by participants.

6.2.7. Theoretical model. Through the process of coding the data and moving from codes to conceptualisation, I developed multiple diagrammatic representations for the concepts, examples of which are available in Appendix 16. In the final stages of the analytic process, these diagrams were combined to present one integrative model which supports and illustrates my argument that CFS/ME is largely a relational experience, and that interactions with others may be factors in the maintenance of and effective coping with the illness. This model is depicted in Figure 1: *The Problem Pattern & Exits: A model for understanding the psychological and relational experiences of PwCFS/ME and their SO.* The central feature of the model is the problematic relational patterns that emerge in CFS/ME, named 'The Problem Pattern'.

The Problem Pattern & Exits model (Figure 1) provides a theory for understanding the cyclical nature of the illness in terms of psychological and relational factors. All PwCFS/ME and their SOs in this study got caught in this cycle at some point in their illness, and eventually travelled down one of the two paths out of the cycle – they got stuck in the problem pattern, or they discovered exits from it in that enabled a process of acceptance to begin. In this section I present a high-level description of the pattern and related categories (emboldened in the model) and concepts.

Figure 1. *The Problem Pattern & Exits: A model for understanding the psychological and relational experiences of PwCFS/ME and their SO*



6.2.8. Introduction to categories.

Category A. Onset. This concept refers to the initial months of illness onset and the interpersonal processes surrounding diagnosis. Individuals face a range of social and psychological challenges and conflicts relating to this period. This category and the dimensions discussed here could generate an entire thesis in and of itself. It is presented here predominantly as a means of placing the model in context, and is therefore less analytical and more descriptive than other concepts in order to promote economy and enable a stronger analytic focus on later concepts that are of greater direct relevance to the research question.

Category B. Core Category: The Problem Pattern. This is the core category in this research and is the category around which the other three categories are based. Although in CFS/ME there is usually a range and combination of debilitating symptoms, the most prominent is invariably fatigue. It became clear that this experience of debilitating fatigue cycles was significantly disabling for those individuals who experienced it. They felt less able to do what they had previously done, and their levels of activity significantly decreased. With this disability came a reliance on others to support the PwCFS/ME in managing the aspects of life that they were no longer able to manage. For example, many were often no longer able to work, cook for the family, clean, support the family or even take care of themselves. Dependence on others impacted upon the PwCFS/ME and the SO, and different dyads coped in different ways with this dependence. In comparing the data, I started to notice an apparent difficulty for the PwCFS/ME and their SO as they settled into their emerging roles as dependent and carer - striking a balance between doing (or expecting the SO to do) enough to be helpful and supportive but not disabling further. We will see the constructs of enmeshment and disengagement as particularly problematic relational patterns in CFS/ME, and how these are related to doing too much and doing too little. All PwCFS/ME described a period early on in their illness which could be interpreted as feelings of depression, although most were keen to point out that it was in response to, and not precipitating, their illness. For some, this depression was also coloured by fear, frustration and anger. Guilt was also recognised as a powerful and prevalent emotion communicated in the data.

Category C. Balance. In this section, I provide evidence of how families exited the problem pattern in the context of CFS/ME. This section highlights the positive relational experiences that were evident in some narratives and shows how participants saw these as being linked to their ability to cope with their illness. I also highlight how relationships that achieve a balance between togetherness and autonomy are consequently viewed as having healthy levels of cohesion, and how they are distinct from those that are disengaged or enmeshed.

Category D. Acceptance. The final category is the phase that some PwCFS/ME and their SOs moved into once they found exits from the problem pattern. The individual and their family may never fully recover in the sense that they may never return to pre-morbid levels of functioning, but some dyads had found ways to live the life they now have with some levels of peace and acceptance, and some described experiencing joy and thankfulness for the positive things they have in their lives, despite the presence of the illness.

7. Category A: Onset

This category, Onset, explains the ways PwCFS/ME and their SOs make sense of their illness. The focus of this research is the experience and functioning of the PwCFS/ME/SO dyadic relationship in the context of CFS/ME. However, as previously explained, it became apparent during the research planning phase that participants would want to feel that I as a researcher understood what the illness meant for them. The PwCFS/ME wanted to tell me about how they first became ill, their struggle to understand what was happening to them, and their suffering as they became more and more isolated as individuals. They also wanted to express the frustrations they had felt during the process of seeking a diagnosis and treatment. This seemed particularly important information for them to share with me before being able to talk about the deeply personal experiences within their relationships. Even though this material did not at first appear to directly address the research question, I quickly realised that this aspect of their story was essentially the context within which the dyadic relationship existed. Without this contextual information, my research would lack depth and insights into the dyadic relationship itself would be limited. Further, this concept is not just about how PwCFS/ME became ill and were diagnosed; it is about understanding how they make sense of CFS/ME and what it means to them. We have to understand this in order to understand the impact it has on their lives.

This first concept emerges predominantly from the first interviews, and from the participants' need to tell their story of onset and diagnosis. It is beyond the scope of this piece of work to explore the full detail of predisposing, precipitating and perpetuating factors in participants' illness; this was not the focus of the research. The most prominent factors in the data are presented here to offer a flavour of participants' narratives. Because this concept aims to describe the experience of illness onset and receiving a diagnosis from the perspective of participants, it is less analytical than later concepts. Offering in-depth analysis of this data would not directly answer the research question. However, to ignore it would leave the subsequent theory lacking its foundations.

Although CFS/ME is an illness that varies greatly from person to person, the experience of onset has striking similarities for most people who eventually receive a diagnosis of CFS/ME. Because the focus here is on the early months of the illness experience – namely, symptoms, diagnosis and treatment – this section is heavily weighted in the stories of PwCFS/ME, to the exclusion of SO participants. This was also a dynamic that I observed in the initial interviews. Some of the earlier dyads I interviewed undertook the first interview in pairs. However, on both occasions I found that the SO barely spoke up at all – the narrative was dominated by the PwCFS/ME, so the content from those first interviews in which I focused on the period of onset is heavily weighted in PwCFS/ME's perspective. This may be reflective of the dynamic within the home environment during the period of illness onset. It suggests that the needs of the PwCFS/ME may become a priority, whilst the SO is required to meet those needs and possibly feels as though they do not have a right to choose or complain. This may contribute to a feeling of imbalance within relationships, and give rise to feelings of resentment and frustration. These concepts are explored later in Category B. The Problem Pattern.

7.1. Early onset: A Flu-like illness.

Participant accounts of the onset of illness portrayed narratives of having previously been an active person in the pre-illness period, and having experienced viral illness often alongside a habit of overworking, for example:

> *"In January 1991 I got a fluey-kind of virus and it took a while to recover; I had to have four weeks off work. On reflection I think I recovered completely from that one then in January 1995 I got a fluey-kind of viral illness, which took me longer to recover from and I had something like half a term off work and I asked the question, 'Did I ever fully recover from that one… In 1999 which is the one that wiped me out, I had six months off work and was still not well and the GP I was seeing at the time used the six-month definition for viral-based ME…"* (PwCFS/ME, Richard)

Most participants with CFS/ME believed that initially they had the flu or something similar and held on to the hope that it would pass as quickly as it started. However, after several weeks or months of illness and no sign of recovery, they became engulfed in a slow, painful period of realisation that their illness was perhaps something more complex than they first thought, and that normal life was unlikely to resume any time soon. They spoke frequently of their expectations of recovery that failed to be met, and of vain attempts to return to normal life and levels of functioning:

> *"I don't think my CFS/ME at that point was very bad at all but…I had labyrinthitis I think at that point, and that seemed to trigger it. Erm… [long pause] I can't exactly remember the order of things…I think it was a virus…then I just never seemed to recover from it..."* (PwCFS/ME, Sean)

These descriptions reflect the body of literature around this aspect of the illness. For example, Moss-Morris & Petrie (2000) reported that the early, pre-diagnosis stage of CFS/ME is usually characterised by symptoms that resemble a viral illness. This has been shown to often follow a period of stress, overworking or overtraining, or illness due to a virus (Surawy et al., 1995). Ultimately, the individual who becomes ill is often seen to have been overdoing it for a considerable length of time before they become ill, as they are likely to have a personality that places value on achievement and high standards (White & Schweitzer, 2000).

7.2. Fatigue and sleep disturbance: which came first?

Disruption to the body clock was perceived as being especially problematic in the early months of illness for most participants. They described it as though sleep was something that either *happens to* them or *does not happen to* them; none of the participants appeared to have a sense of agency in their sleep/wake cycle during illness onset. Participants described struggling with their sleeping and waking patterns due to constant feelings of fatigue alongside unrefreshing sleep. Whilst some described a pattern in which they felt they wanted to sleep all the time, others found it difficult to sleep at night which in turn had further negative consequences on the body.

> *"What happened was I had a virus and I over exercised and then I really started to feel physically unwell and I couldn't sleep…which has always been one of the biggest issues for me."* (PwCFS/ME, Steve)

> *"I was running at quite an elite level so at least twice a day, and I was just finding I was not sleeping well and feeling really tired and my performance in running was going down and down. I thought it was due to overtraining and not getting enough food into me… I felt quite rubbish and thought 'I'm training well' but my heart rate was getting higher all the time which suggested some kind of virus, and I felt really tired and drained and I was running at the same pace as usual but it was killing me. Ended up not sleeping well and having blood

> *sugar problems. I had to sleep after lunch and literally couldn't stay awake."* (PwCFS/ME, Rob)

Whether sleep disturbance is a precipitating or perpetuating factor in CFS/ME is a matter of debate. Although there is evidence to suggest that sleep disturbance is linked with fatigue in CFS/ME and that this is indeed a common occurrence in the illness (Krupp, Jandorf, Coyle, & Mendelson, 1993), sleepiness and fatigue are both subjective, interrelated but separate phenomena despite being used interchangeably as terms in clinical practice (Shen, Barbera, & Shapiro, 2006). In fact, excessive daytime sleepiness (EDS) is a condition in itself, and the features of fatigue and EDS overlap so that distinguishing between the two presents challenges (Shen et al., 2006). A further complication is that sleep disturbance has also been found to be a symptom of depression (Nutt, Wilson, & Paterson, 2008). Therefore, determining the role of sleep disturbance in the onset of CFS/ME is much like a 'chicken-and-egg' problem in that it is hard to determine which came first, sleep disturbance or other symptoms, although it seems clear that sleep disturbance and other symptoms are interrelated and compound one another in CFS/ME.

7.3. Social and psychological factors.

Participants clearly identified the role of emotional stress in the development of their illness, appearing to believe that social stressors contributed to illness onset. Overworking, exhaustion and relationship difficulties were cited as key 'causes' of illness by several participants in this study. However, they appeared to demonstrate somewhat limited recognition of the fact that they had some agency in this, as though things were beyond their choice and control, and it was *happening to* them.

> *"There was a lot going on when I was diagnosed with this. Erm…. [Sigh] in the April of that year I ended a relationship, we'd been together for seven years and had lived together. I ended it in April, it was quite traumatic…So I think I, not got over it, but did every hour I could overtime and just worked and worked and worked…, I worked like a lunatic. And I used to get a lot of respiratory tract infections. I was a heavy smoker at the time and had a course of antibiotics and I just rang up when they finished and went again. In November I ended up having three lots of antibiotics, and I had no strength at all and I didn't know what was happening to me…"* (PwCFS/ME, Ally)

Ally describes here coping with grief at the loss of her relationship by working a lot of hours. It seems in her explanation that she does recognise that this was her way of coping, it is possible that her behavior meant that she failed to process her grief and became stuck because she did not allow herself to experience the pain of grief (Worden, 2003). Her use of the word 'lunatic' suggests that she knew what she was doing was not healthy. Yet she later seems to absolve herself of any responsibility in the phrase *'I didn't know what was happening to me'*. Rachel's story echoes Ally's description of overworking and earlier descriptions of disrupted sleep cycles. Rachel describes how social factors left her feeling under such pressure that the choice to live in a healthy way was taken away from her. Her ability to cope and function was impaired by what she believed was being forced upon her by others;

> *"I think that it was definitely external pressures making me stressed and not being able to sleep and I just kept on working even though I was really, really stressed and tired, I just powered on through it, I tried to put things in place so I could reduce workload and stuff but it wasn't…I wasn't able to do that*

> *and in the end I had a massive car crash because I was just running on errr… I was just mental (laughs). I don't mean that I meant to crash the car, it was an accident, but I was clearly not functioning in a way that somebody driving a car should be because I was so exhausted and so I had a massive car crash and have never really been the same since."* (PwCFS/ME, Rachel)

What Ally and Rachel are describing here is *their response* to external pressures - maladaptive coping styles and a sense of duty regarding overworking. Both participants describe social pressures that lead to certain behaviours in their approach to work. They use language of mental illness – 'lunatic' and 'mental' - to describe their behaviour prior to the onset of their illness. This seems to suggest that on reflection they both understood that they were behaving in ways that were having a detrimental effect on their health and wellbeing.

This pattern of behavior in the face of stress relates to research around personality in CFS/ME, which has revealed a common theme in CFS/ME of people overworking and burning out. Surawy et al. (1995) suggested that people who are predisposed to CFS/ME tend to be perfectionists and high achievement orientated people for whom other peoples' opinions are of great importance in maintaining their self-esteem. Therefore, they impose high standards upon themselves. Kath's excerpt below succinctly evidences the viral causal explanation discussed earlier alongside her own clear assertion that she wanted to be seen to be coping despite significant stressors and physical illness, as per Surawy et al.'s (1995) suggestion:

> *"Mine started with a shingles virus that was not diagnosed… I went back to work after two weeks not knowing what it was, not being given the antivirals that you're supposed to have after that, erm…I remember one of my students*

> *saying to me 'Are you sure you should be at work? You don't look very well', I said 'Oh well I'm here and coping' and that's my attitude - I just thought it was some virus and I was over it but I never really looked up after that, did I? I had a busy year because I was working, and there was a lot of pressure at work with paperwork, new systems…and then we moved here, so we had all the stress of moving…"* (PwCFS/ME, Kath)

Surawy et al. (1994) found this pattern of pressing on in the face of stressors that hinder people's ability to function and cause them stress to be common among PwCFS/ME, leading to exhaustion. Surawy et al. proposed that by following this pattern of overworking and then breaking down physically, they protect their self-esteem. They are able to attribute their functional disability to physical illness, instead of having to admit to being unable to cope psychologically or behaviourally with the external pressures, for example by putting a stop to excessive demands on their time, which would not be aligned with their beliefs about expectations of themselves.

7.4. Confusion and disbelief: what's happening to me?

PwCFS/ME described a period during the initial onset of illness of not knowing what was happening to them; indeed, this phrase featured frequently in the data. They faced disabling physical symptoms that affected their ability to function in the way they were used to, and invariably endured a battery of medical tests and consultations from which no conclusions could be drawn. It became evident that the overwhelming feelings in the absence of a diagnosis, were utter confusion about what was happening to them, and a sense of disbelief.

For example, Amy started to experience symptoms of illness very early on in a new relationship whilst at university. Her story of the onset of her illness communicated a sense of confusion and frustration in both herself and her SO. She described confusion at her sudden disconnect with what her body was doing and its apparent decline as they both struggled to make sense of her symptoms. The emotional consequences of her illness within the context of their young relationship, and the absence of any means of communicating their distress with one another, lead them to drifting apart emotionally:

> *"Quite early on in the relationship I just suddenly fainted which I'd never done previously, so my body was obviously not in a great place…so physically I think things started to happen that I couldn't understand so obviously that made me a bit more paranoid, a bit kind of not sure what was happening…and I think that probably made me turn a bit more for emotional support when he probably didn't have a clue what was going on because we didn't really talk about what was happening, I didn't really know myself."* *(*PwCFS/ME, Amy)

This phrase 'I didn't know what was happening to me' came up again and again throughout the interviews, but particularly in reference to the early onset of the illness. Many participants expressed feelings of bewilderment and confusion, and complete desperation at being given no answers by medical professionals.

Kath provided a description of the numerous, and fruitless, tests and consultations with a range of specialists that she endured, and of her determination to seek answers and solutions for herself in the absence of answers from professionals. Despite being a solution-focused, pro-active person, her desperation to find answers drove her to distraction and intense frustration, so that she eventually began to question her own sanity:

> *"…I went back and I think I lost my rag with them by this time because I was going for quite some time…it was very frustrating because you were reiterating the same story over and over again to different people, how you feel and all the symptoms you are going through but nobody comes up with any answers so you become very mentally frustrated, and then because you've got a brain in your head… you start trying to piece it together yourself and trying to rationalise it and then you get to the point when nobody is coming up with anything…so then you start to worry about things that are not there because nobody is coming up with an answer to why you feel like this, you feel at death's door and I was still struggling to carry on…"* (PwCFS/ME, Kath)

These excerpts can be linked to theories of grief and loss; Although the full extent of their loss may not have been fully processed at this point in their illness, PwCFS/ME have by now faced a relatively abrupt loss of the life they previously knew, and a loss of their health and of previously perceived 'certainties' in life. They are now living in uncertainty and are lacking explanations and solutions. They may also at this point be grieving their loss of work, meaningful occupation, and previous ways of relating to others to which their illness has become a barrier. Within Parkes' Phases of Grief model (1972), the first of the four phases proposed is Shock and Numbness, which is followed by Yearning and Searching. These phases appear to map onto participant descriptions of the early days of illness in which I argue they experienced disbelief followed by a desperate searching for answers.

7.5. The meaning in a diagnosis.

Because part of the criteria for diagnosing CFS/ME is that it is an illness that has persisted for six months or more (Fukuda et al. 1994), this initial period of confusion usually lasts at least six months, although for many it remains undiagnosed for many more months or even years. Receiving a diagnosis is problematic because of the individual's understanding of what a diagnosis is and what it is not. There appears to be discord between the very nature of CFS/ME as both a label and an illness, how the medical community see this, and how PwCFS/ME see it. For some participants it seemed clear that their interpretation in hindsight was that their illness was CFS/ME from the start, and that doctors failed to diagnose quickly:

> *"You know, I was getting the sick note saying post viral fatigue, I had a colleague saying well this thing you've got sounds like that ME thing and I queried that with GP and he said 'Well no, you've got post-viral fatigue, ME is a more long-term and rather more serious condition' but then after six months he used that definition…A big problem is that they don't give the ME label until then and therefore you don't really get the best advice in terms of how to manage the condition…"* (PwCFS/ME, Richard)

Similarly, Steve explained his belief that the advice he received from his GP actually served to worsen his physical symptoms:

> *"Well it's difficult… I didn't know what was going on when I became ill. I was very confused. And I went to my GP and then my health deteriorated when I went to my GP."* (PwCFS/ME, Steve)

Steve went on to explain that the reason his health deteriorated after seeing his GP was because the GP did not offer a diagnosis and instead advised him to exercise more. Steve and his Mum both described how he had diligently followed the GP's advice, but that this had served to debilitate him yet further so that he ended up bedbound:

> "*We went to the doctors and they prescribed antidepressants [to him]. Now I did look on the internet, I did research you know, I put his symptoms in and I was looking, and I actually went back to the doctor with a leaflet about chronic fatigue…the doctor dismissed it and said 'No it's not that'…he was prescribed antidepressants, he was told to do exercise, and he got pushed beyond his limits really and at the end of the day discovered not 'til quite a few years later it was chronic fatigue he had so we were all getting him to do things really what [sic] was making him more ill…I knew there was something seriously wrong so I went to the doctor asking about it and the doctor just said it wasn't that and I had to accept it…if he'd have said to me and Steve 'Look we think it's chronic fatigue', I think we'd have gone down a different path. Steve suffered for years with depression and anxiety because that's what he was labelled and it wasn't until years down the line that we got the diagnosis of chronic fatigue. So, all those years it could have been dealt with in a different way…"* (SO, Helen)

Kath described how both she and her son Rob became ill but did not receive a diagnosis of CFS/ME for quite some time:

> *"I think the both of us, I'm sure I'm speaking partly for what I've experienced but also what I saw with [Rob], it is very slow to be diagnosed, I think that's the first issue that comes up to my mind…"* (PwCFS/ME & SO, Kath)

CFS/ME is a diagnosis of exclusion which means that people who eventually receive a diagnosis must have tested negatively on a range of other tests before their GP can reasonably diagnose CFS/ME. It is not a disease in and of itself, but rather it is a collection of medically unexplained symptoms (Fukuda et al., 1994). The criteria for diagnosis mean that essentially the illness is not CFS/ME until it has persisted for six months or more according to Fukuda et al. (1994), or four months according the NICE guidelines (2007). Therefore, in the early days of the illness what people experience is not technically CFS/ME because it would not meet the criteria for the label (Fukuda et al., 1994; Holmes et al., 1988; NICE, 2007; Sharpe, 1991; Wilson et al., 1994). Doctors cannot provide an earlier diagnosis for this very reason. The excerpts presented suggest that upon receiving a diagnosis or having self-diagnosed, many PwCFS/ME believe they have a knowable disease called CFS/ME, in the way that one contracts flu or a common cold, and they believe that this is what it was from the moment they first became ill. The disparity here between the criteria for diagnosis and what people come to believe the diagnosis means could be seen to be the root cause of much frustration on the part of both GPs and PwCFS/ME.

Also worthy of consideration here is the fact that illness perceptions have been shown to be crucial in shaping how an individual copes with symptoms (Leventhal, Nerenz, & Steele, 1984). Leventhal et al. (1980) proposed the self-regulatory model of illness behaviour which explains the role of illness perceptions in determining coping strategies. It is summarised by Moss-Morris & Petrie (2000) as consisting of four components:

> *"The cognitive representation of the illness, the emotional response to the illness and treatment, the coping directed by the illness representation, and the individual's appraisal of the coping outcome"* (p.92).

When a diagnosis is provided by a doctor, individuals construct a representation of what that diagnosis and illness means which then determines their behavioural and emotional responses (Petrie & Weinman, 1997). The coherent view of illness is thought to be constructed of five components: Identity - the label and symptoms viewed by the PwCFS/ME as ‘the disease’; Cause - the ideas an individual holds about the causal factors of their illness; Time-line – their beliefs about how long they are likely to be ill for (acute, chronic or episodic); Consequences – how the illness affects their life and functioning; and Cure-control – how they manage their illness and whether they might be able to control or recover from it. Beliefs that an illness will last a long time and symptoms are numerous have been shown to be associated with poorer outcomes and limited beliefs in the ability of the PwCFS/ME to control or cure their illness (Moss-Morris & Petrie, 2000).

This provides at least some explanation as to why the period of onset is so confusing for PwCFS/ME. Not only are they suffering disabling symptoms and the distress that accompanies this, but they also face uncertainty about what their illness is, and their illness representation is in a constant state of flux. They cannot therefore make informed decisions about how best to cope. It is not until a label is applied by a person perceived to be in a position of authority that they can form a coherent view of their illness. Indeed, when interviewing 50 PwCFS/ME over a two-year period, Woodward, Broom & Legge (1995) found that PwCFS/ME felt having a coherent diagnosis was enabling for them whilst a lack of an explanation for their problems had negative effects for them.

Unfortunately, it seems as though many PwCFS/ME seek the diagnosis for reasons of attribution. Despite the existence of many a diagnosis for other non-biological conditions and the lack of evidence for an organic cause for CFS/ME, a diagnosis of CFS/ME seems to provide PwCFS/ME with some validation that they have a physical illness. This provides them with a solution to the problem of attribution; being able to attribute their symptoms to a physical cause alleviates blame – much like Suraway et al.'s (1994) suggestion that this protects their self-esteem, because it means they do not have to admit they cannot cope, a diagnosis of CFS/ME from a GP alludes to a physical illness in the minds of many PwCFS/ME, and therefore perhaps they can absolve themselves of responsibility for the development of their illness. PwCFS/ME have also been shown to be less likely than people diagnosed with depression and other medical illnesses to attribute their illness to internal psychological factors such as their behaviour or emotional state (Moss-Morris, 1997; Weinman, Petrie, Moss-Morris, & Horne, 1996) although I have presented evidence that some participants did acknowledge their own behaviours as being factors in the onset of their illness. Moss-Morris (1997) found that 70 per cent of illness was attributed to physical causes among PwCFS/ME. Physical attributions were initially linked to poorer outcomes in CFS/ME and this became an area of research in CFS/ME that received a lot of attention (Moss-Morris, 1997). However, Deale, Chalder & Wessely (1998) presented evidence which suggested physical attributions may not play such a crucial role in clinical outcomes as first thought.

7.6. I've been diagnosed, what now?

Despite evidence that suggests a diagnosis helps to shape illness perceptions and attributions, participant narratives suggested that even in the presence of a diagnosis, confusion persists. Perhaps this is in the absence of a clear explanation of what the diagnosis means. Although the diagnosis of CFS/ME can offer PwCFS/ME at least some answers to the question 'What's happening to me?', arrival at the diagnosis is only one stop on the journey, and it brings with it further complications. Ally explained it as though her doctor's sick note was somewhat detached from her experience –it was just a label that bore no meaning for her, and so her experience remained completely baffling, despite having been provided with a diagnosis:

> *"I didn't know what was happening to me, and the doctor kept writing chronic fatigue syndrome on the sick note."* (PwCFS/ME, Ally)

This may be indicative of a wider problem in CFS/ME – a diagnosis may only give rise to more questions for PwCFS/ME, and offers few answers. Sadly, for the PwCFS/ME, once the diagnosis has been offered, there seems to be variation in the support offered depending on whom the PwCFS/ME sees, and where they live. NHS resources are scarce, suggesting that funding decisions do not always go in the PwCFS/ME's favour. For example, one PwCFS/ME was hardly able to access any funded treatments:

> *"I had a number of tests, a scan and MRI, and electrode test for my nerves, and various blood tests and stuff to come to the diagnosis and then he shared that information with my GP and suggested a number of treatments most of which could not be funded..."* (PwCFS/ME, Rachel)

Meanwhile, participants living in another county had been offered opportunities to see a psychiatrist, and were sometimes but not always referred to a specialist CFS/ME service with the local mental health NHS trust. Most participants described being open to meeting a psychiatrist, but often described having been told by them that there is nothing 'wrong' with them with regard to their mental health or that they cannot do anything for them (see excerpts below:

> *"[The GP] said oh I think you'd better go to a psychiatrist which they often do you see and so I saw the psychiatrist, she was lovely and she said no, no there's nothing wrong with you, it's your lifestyle"* (PwCFS/ME, Eileen)

> *"The GP did refer me to the psychiatrist … and I went to see a lady there but it took a year and a half, but strangely enough what happened was 'Well we'll send you to the ME clinic'…"* (PwCFS/ME, Kath)

> *"The problem I had with it all was that it was all just so confusing, I mean it was always anxiety and depression, mood disorder [the psychiatrist] said, whatever that means…but they completely ignored the physiology… and they even admitted it to me, [the psychiatrist] admitted 'We've never been able to help you.'"* (PwCFS/ME, Steve)

This last excerpt could also be considered supportive of the argument that physical attribution leads to poorer outcomes, and hints at the cross-over between CFS/ME symptoms and mental health difficulties.

Not all participants were referred for psychiatric or psychotherapeutic input however. Ally, who had elected her GP as her SO and who felt that he had been highly supportive of her, seemed not to have been offered a referral to a psychiatrist or to the CFS/ME clinic, and appeared upset and confused when I posed the question during her interview:

Interviewer	*Did you receive help at the ME clinic? The NHS one.*
Ally:	*I don't think there is one.*
Interviewer:	*There is, in [city], I think it is at [address]*

Ally: *Oh, I’ve never heard of it!*

Interviewer: *Has your GP offered support at all like CBT?*

Ally: *Mm. No. He offered me the pain clinic at [hospital] but getting there was just too much really. I think they would have offered [pause] acupuncture. Myself I’ve had acupuncture, and reiki which worked, I probably shouldn’t have it since I’m [religious denomination] but it really worked. The acupuncture relaxed me, I’ve had hypnotherapy, I had a chap come for weeks and weeks but I didn’t know there was an ME clinic.*

Interviewer: *Yeah, they provide services through the NHS so psychotherapy is available, advice on pacing…*

Ally: *Pacing and breathing. They are the best things really. I didn’t know there was an ME clinic.*

Interviewer: *Is your GP someone you have had a good relationship with and felt supported and understood by?*

Ally: *Yeah absolutely. But he said to me a long time ago that many people don’t believe in it [CFS/ME].*

With inconsistent service availability and a limited number of evidence-based treatments, GPs are in a challenging position. They must mediate between a frustrated and distressed individual who desperately wants help, and a lack of empirical understanding of how medical professionals can help PwCFS/ME. Their job is to help alleviate suffering, yet they have little to offer. In this situation, many offer medication that aims to assist in symptom management such as painkillers, sleeping aids, and antidepressants, but some participants in this study stated that these rarely help:

> *"I went to the doctor, he gave me an antidepressant, I can't remember, it was a strong one, bandied about at the time…and I just collapsed with that after taking it, I couldn't keep upright and then I must've given up on that."* (PwCFS/ME, Eileen)

> *"I had to try another month on another antidepressant…because 'that's what we do with people with ME and if we look at your physical notes…and we saw you've not been taking antidepressants, well you've not being doing anything right to cure the ME' - was the attitude at the time… I probably was bit angry at that stage, and bit angry at the medical establishment at large, not the GP, I was specifically seeing he was very understanding…I knew it wouldn't [help], and my GP knew that, but you know, 'We've got to jump through this particular hoop' is what it is all about."* (PwCFS/ME, Richard)

Healthcare professionals' attitudes, beliefs and knowledge of CFS/ME have become a focus of research in recent years. Deale & Wessely (2001) found that two-thirds of PwCFS/ME reported dissatisfaction with their care. In a study of the beliefs of GPs and PwCFS/ME around the illness, Thomas and Smith (2005) sent surveys to 120 GP surgeries in Wales in which only half of the 48 GPs who responded believed in the existence of CFS/ME. They also reported that there is a lack of specialist knowledge around CFS/ME in primary care. Marks, Huws & Whitehead (2015) suggested that healthcare professionals are 'working with uncertainty', and make sense of CFS/ME and decisions about their management of these individuals on the basis of their previous experiences. However, Marks et al. also suggested that this influences the labels applied to individuals and the interventions offered. This is reflected in the evidence presented in this study; participant perceptions were that healthcare professionals communicate feeling at a loss, or are disbelieving, leading to inconsistent and sometimes inappropriate care provision, and a growing sense of frustration for both parties. There is hope though; Bowen, Pheby, Charlett, & McNulty (2005) found that a GP who recognised CFS/ME as a clinical entity also had a more positive attitude towards the condition, and this was a predictor of improved prognosis.

In many illnesses, the process of getting a diagnosis can be difficult, frustrating and lengthy as symptoms remain unexplained and anxiety about the possible causes builds. In the absence of a diagnosis, prognosis remains unknown, knowing what to do to feel better remains a mystery and many PwCFS/ME can feel anxious. Despite the fact that NICE guidelines highlight the need for a decisive and confident early diagnosis of CFS/ME in primary care, GPs consistently demonstrate a reluctance to diagnose. In a study which consisted of interviews with 20 GPs regarding their views on diagnosing CFS/ME, Woodward, Broom & Legge (1995) found that 70% of the GPs interviewed expressed reluctance to diagnose an individual as having CFS/ME due to the lack of certainty around the etiology of the illness, and due to a general belief that such a diagnosis may 'become a disabling self-fulfilling prophecy' (Woodward, Broom & Legge, 1995, p.325). Due to the lack of financial incentives for GPs to diagnose and manage CFS/ME effectively, many PwCFS/ME do not experience the standard of treatment set out in the NICE Guidelines (Lester & Campbell, 2010; Lester, Hannon, & Campbell, 2011). My study would suggest that this may still be the case.

Woodward, Broom & Legge (1995) found that PwCFS/ME surveyed about their views around diagnosis in primary care felt it had been enabling to be provided with a single coherent diagnosis and that it was helpful to have an explanation for their difficulties. Further, following semi-structured interviews with PwCFS/ME and carers, Hannon et al., (2012) found that the recognition of CFS/ME as a legitimate illness was of paramount importance to PwCFS/ME and carers, as was feeling that they were believed by healthcare professionals. Diagnosis can be seen to be one form of validation for people struggling to come to terms with a complex, disabling and controversial illness.

The following excerpt from a book written by an Australian TV Presenter who had CFS/ME for a number of years, Leigh Hatcher, perfectly sums up this debate:

> *"I finished my first year of illness none the wiser about what 'it' was. How fiercely I yearned for some kind of clarity or label! One specialist told me a story that had particular resonance for me. A woman who had suffered significant ill health for three years without a diagnosis came to see him. In the end he discovered cancer of the pancreas-undoubtedly a death sentence. But her overwhelming response was one of relief and gratitude that at last she had a label and wasn't going crazy. I knew how she felt."* (Hatcher, 2005, p. 37)

In order to understand why diagnosis is seen to be so important, it is useful to think about diagnosis in CFS/ME in comparison to another chronic illness, Multiple Sclerosis (MS). MS is a similarly complex condition that is difficult to diagnose because it cannot be confirmed by a single clinical feature or diagnostic test (McDonald et al., 2001). Research has shown that people who have neurological symptoms that remain undiagnosed are likely to conduct their own information search through reading literature and talking with acquaintances, and this can lead them to develop anxiety around their condition, believing that it could be a more life-threatening diagnosis, or else they self-diagnose as having MS. Uncertainty also causes problems for people with MS in coping with their illness and symptoms (Wollin, Dale, Spenser, & Walsh, 2000).

Where people with MS are given more information about the management of their illness, it has been shown that they can somewhat overcome the feelings of uncertainty, and thus the provision of information is recognised as key to illness management (Somerset, Campbell, Sharp, & Peters, 2001; Vickrey et al., 2000). Early diagnosis in MS has been demonstrated in some studies to reduce anxiety and distress (Burnfield & Burnfield, 1978; Mushlin, Mooney, Grow, & Phelps, 1994; O'Connor, Detsky, Tansey, & Kucharczyk, 1994). Considering this evidence, it is fair to conclude that in CFS/ME, people may benefit from earlier diagnosis. However, the period from onset to diagnosis in CFS/ME is likely to last significantly longer than the four months of symptomology required in order to meet the NICE (2007) diagnostic criteria or the six months required to meet the CDC criteria (Fukuda et al., 1994). In a primary care trial, Chew-Graham, Dowrick, Wearden, Richardson, & Peters (2010) found that the average time from symptom onset to diagnosis in CFS/ME was 3.6 years. Within my sample, specific dates of onset and diagnosis were not collected, but from the approximate years of onset and diagnosis provided by participants, five were diagnosed within approximately one year of onset, one was diagnosed approximately three years following onset, and three further participants were diagnosed 10, 15 and 32 years following onset. This data provides an average of 7.4 years from onset to diagnosis, although this must be viewed with caution as the sample size is small (*n*=9) and the range is large (<1 year – 32 years).

What happens once a diagnosis has been reached, either formally or via self-diagnosis? Certainly, from the descriptions provided by participants, it seemed for most of them that a significant amount of time and energy went into receiving a diagnosis. The PwCFS/ME is perhaps at least armed with information about what their illness is not once they receive their diagnosis. But regarding what their illness is, they are often left to make sense of it for themselves. Huibers & Wessely (2006) noted that diagnosis may only be useful in CFS/ME if it marks the beginning of therapeutic intervention and not the end, but studies have also shown that primary health care professionals are ill-equipped to deal with CFS/ME once a diagnosis has been made (e.g. Bowen et al., 2005). As there is no cure and limited evidence about how best to manage the condition, if we consider my findings alongside the findings of existing research then it may be fair to conclude that a formal diagnosis does little to help PwCFS/ME other than offering a focus for their personal research into how to manage their symptoms. As we see in this study, making sense of their illness can become a full time project for PwCFS/ME and their SOs, and learning how to manage it is a process of trial and error.

7.7. Participant construction of their narrative of illness experience

The excerpts presented in this section illustrate key factors perceived by participants to have contributed to the onset of CFS/ME. They also provide a glimpse of the rationalisation and reasoning process that people go through as time passes and they try to make sense of their illness. Many of the participants had been ill with CFS/ME for many years when they undertook these interviews, so reflecting on the early days of their illness with the benefit of hindsight enabled them to tell these stories in ways that reflect what they have come to believe about CFS/ME and their own experiences. They had all spent considerable time trying to make sense of their illness. This meant that they were able to see or construct patterns in their illness trajectory and behavior that may not have been apparent to them when they were in the throes of illness onset. Had I been able to interview them during the initial period of illness, I believe it is less likely that they would be able to so clearly connect the stresses and strains of their social world, lifestyle and coping responses to their experience of feeling unwell. When one is in the thick of a debilitating illness, surviving daily struggles becomes the main focus and broader, overarching patterns are not obvious until much later, when looking back and reflecting. It is at that point that one becomes able to construct a story around what happened to them, and connect dots in ways that it is unlikely would have been viewed as being connected if interviewed at that time.

7.8. Final thoughts

This section also provides evidence that the symptoms and illness experience is consistent with CFS/ME research to date, and that the sample is therefore typical. My expectations from the consultation process that my sample would want to provide context were accurate. They wanted me to hear their story because they wanted to be understood. They wanted to convey what their experience was like because they were so used to not being believed and understood. Many of them also felt that they had rarely had an opportunity like this to tell their story in a context where they felt listened to and understood.

Narratives suggest that the slow, painful process of making sense of the illness and experience takes place in the context of stigma and delegitimation. We see then that whilst the early months of illness are coloured by confusion, frustration, medical tests and a fight to understand what is happening, the arrival at a diagnosis of CFS/ME signals a new phase of illness experience for both the PwCFS/ME and the SO. The diagnosis may at first offer some relief as it provides answers to some questions for people who receive it, and possibly offers at least some form of a (much-disputed) explanation, but it certainly does not offer solutions. Instead, it marks the start of a difficult journey through CFS/ME for the PwCFS/ME and their SOs which for some may lead them into eventual acceptance and possibly even a return to pre-morbid levels of functioning, but for others will be a downward spiral into life-long illness from which they may never recover.

As we move into the next concept, we shift our focus on diagnosis to the narrated experience of living with CFS/ME for both the PwCFS/ME and their SO (often their carer). We begin to consider the relational dynamics at play and see how the trajectory of illness can be affected by these dynamics.

8. Category B. Core Category: The Problem Pattern

Having provided a high-level description of each aspect of the core category of the Problem Pattern in the earlier introduction, I now present evidence for the Problem Pattern. I present each aspect in detail alongside text excerpts and in-depth analysis, illustrating how I constructed the categories and concepts that led to the development of the grounded theory. I link this to the literature and to existing theories selected for their relevance. The first two concepts in this category, Fatigue and Disability focus on the experience of the PwCFS/ME but I then move into concepts that illustrate the experience of the SO too.

8.1. Fatigue: Communicating experience of the cycle of fatigue

In CFS/ME, fatigue is the central and most debilitating symptom of the illness and was therefore one of the most prominent topics of discussion across the interviews. It is described by PwCFS/ME as something far beyond day-to-day tiredness. It traps them in a cycle of exhaustion in which they feel the need to rest, and yet the less they do, the less they feel able to do, so their general physical capabilities are lessened. In the co-construction of the participant narratives (between participants and I as the researcher within the interview context), participants used linguistic strategies to convey their experiences to me in ways that they felt I could understand. They used metaphors, analogies and comparisons as a means of making sense of their experience, and of communicating that experience by relating it to previous similar feelings or states that they had experienced in their lives. This was also a technique employed to induce empathy in me. My empathising was based on my ability to relate their experience to my own experiences; I reached my own level of understanding about their suffering by bringing to my mind something that they had viewed likely to be a common experience between us. It enabled me to draw upon my own memories of illness and use my imagination during the interview so that we could both feel we had reached shared understanding of their experience. As I draw on their narratives to tell their stories of struggle and fatigue, I will also highlight their use of language to communicate experience and attempt to be understood. In doing so, they often use analogies to assist others in empathising.

In an attempt to explain her illness experience to me, Ally drew upon a commonly experienced disabling illness to which most people can relate, influenza:

> *"...just fatigue upon fatigue, like having flu all the time."* (PwCFS/ME, Ally)

This is a comparison that PwCFS/ME often make when they seek to help others reach an understanding of the illness experience (Moss-Morris & Petrie, 2000). It is important to PwCFS/ME to feel that those around them understand that their symptom experience is extreme and is far removed from normal, everyday experiences of fatigue. They communicate this to others using commonly understood analogies.

The disruption to the body clock discussed in Category A. Onset was echoed by a number of PwCFS/ME. For example, Sean reflected on how unsettling it can be to have an irregular sleep/wake cycle that is not aligned with those around him, hinting at the social isolation and negative feelings that this causes:

> *"There have been times where I have woken at seven at night and gone to bed at six or seven in the morning. I get about 10-12 hours' sleep a night. It doesn't feel good waking up when everyone else is going to bed."* (PwCFS/ME, Sean)

In highlighting his sleeping patterns, Sean infers that he recognises that these are 'abnormal' habits that cause him to feel separated from the lives of others in his family. Rob also referred to difficulties with his wake/sleep cycle:

> *"I had to sleep after lunch and literally couldn't stay awake. I missed out on running because I felt so tired but a different type of tiredness, a completely different type which is hard to describe really. Then I was sleeping in the day and going to bed at night and waking up literally every 20 minutes and I would wake up in the morning feeling like a bus had hit me. Getting out of bed was hard and I had no energy to do anything."* (PwCFS/ME, Rob)

Again, Rob describes wake/sleep cycles that were at odds with the normal daily patterns of those around him, putting him out of synchronisation with others in the family home. For example, his Mum and Dad were up early in the morning and working during the day on week days. The expression '*feeling like I'd been hit by a bus*' is a metaphor that is used commonly in the English language, which I interpreted as feeling completely drained physically and unable to move upon waking. Rob tried to maintain patterns of waking and sleeping but his physical experience and daytime fatigue made this difficult. Rob's description is also an example of unrefreshing sleep which is a criterion for CFS/ME (Fukuda et al., 1994). His use of the phrase '*it's hard to describe'* illustrates the difficulties PwCFS/ME have in communicating the extremities of their experience of fatigue to people close to them who have not been through the same thing.

Participant descriptions suggested that in many cases the fatigue and body clock disruption was of an extreme nature, so that the PwCFS/ME felt they must confine themselves to bed rest, thus isolating themselves within the family home, and increasing their dependency upon others. In the most severe cases they described feeling unable do anything; even going to the toilet or washing can be too much:

> *"I was just in bed the whole time, I was really struggling to get up. I would get up in the morning…I'd sit at the breakfast table with [my children] for as long as I could, which was maybe ten minutes because sitting up was just really hard, and then I would go back up to bed; sometimes I didn't come downstairs and we would have breakfast upstairs…and they would go to nursery and to work and I would just stay in bed and he would leave stuff next to the bed so I had something to eat and water… And that would be really hard because it would take me ages, if I need to go to the loo, it would take me ages to get to the bathroom, it's just in the next room but it could just take me a whole morning just to get to the bathroom, if I got up I was dizzy and I fainted, and if I tried to walk I'd be dizzy and I'd faint… I remember showering being really difficult…looking back it was really, really difficult and it was really acute I think for about six months then it started to get a little bit easier."* (PwCFS/ME, Rachel)

The language of this narrative centres on themes of struggle and difficulty as Rachel has attempted to construct a condensed linguistic representation of six months of her most debilitating fatigue experience. Her construction painted for me a dark picture of a life that had ground to a halt and shrunk into the confines a bedroom, bathroom and kitchen. Rachel is also attempting to convey her efforts to maintain some form of structure in her day by getting up in the morning to see her children, despite the fact that she experienced this as extremely difficult and physically uncomfortable. Her story of fatigue illustrates her need to virtually drop out of society completely during this acute phase. But even worse, with such a high degree of disability, the impact on her family members also comes into view, as does the mechanism by which Rachel started to become isolated from family life. As a mother, being unable to look after her two children and having to rely on her husband to support her with even her most basic self-care needs due to such extreme fatigue was something that she articulated as being a devastating experience. We shall see as we move through each aspect of the model how this degree of disability has consequences in relationships.

Rob was an athlete and described himself as having exceptional levels of physical fitness prior to the onset of his illness. He described why, despite his usually high levels of motivation for physical activity, it was so hard to fight the fatigue and attempt to increase activity:

> *"I wasn't bed ridden. Like I said, getting out of bed was horrible, it was like I was hungover. I could do things but I would fall asleep and I was that tired that I couldn't fight falling asleep. Like, there would be a football match on that I'd really want to watch but I'd be that tired that I couldn't keep my eyes open. That very, very rarely happens now. But yeah, I was struggling with day-to-day things because I could do something for a bit but then I just had to rest and sleep."* (PwCFS/ME, Rob)

It is interesting to note that even in the case of someone who has historically pushed his body to its limits and had no problems with motivation levels or physical fitness, breaking out of this cycle of fatigue seemed to be such a struggle. We see the term 'struggling' used to describe the experience, and a comparison drawn with a common experience, that of a hangover, in an attempt to communicate with me the experience of fatigue as it was for him. Other phrases that Rob used to communicate his experience, such as *'I couldn't keep my eyes open'* and *'I couldn't fight falling asleep'* are descriptive phrases or metaphors commonly used in the English language to communicate common physical experiences between people.

The cycle of fatigue: A discussion. The cycle that this level of fatigue and the consequential inactivity leads to has been a subject of research in recent decades– at this level of functioning, any slight increase in activity would likely tire the body, so that the experience of fatigue could seem to worsen. Wessely et al. (1989) first hypothesised that physical deconditioning may be a perpetuating factor in CFS/ME, compounded by the experience of depression encountered during this acute phase of the illness. This has been linked to avoidance behaviours in chronic pain; attempts to prevent further discomfort can lead to fear-avoidance. PwCFS/ME have been found to avoid activity that they deem likely to worsen symptoms, and it has been suggested that this further decreases physical fitness (White et al., 2011). However, studies of physical fitness in CFS/ME have shown conflicting results. For example, De Lorenzo et al., (1998) found evidence of physical and cardiovascular deconditioning among PwCFS/ME compared with healthy controls, whilst Bazelmans, Bleijenberg, Van Der Meer, & Folgering (2001) found no evidence of a significant difference in physical fitness levels between PwCFS/ME and healthy controls. These coping patterns can also cause difficulties in relationships because others can feel as though they do not know what to expect from the PwCFS/ME. The fluctuations can give the impression that the PwCFS/ME might be malingering when it suits them, or some may view PwCFS/ME as simply needing to try harder – if they were able to do it yesterday, then why can they not do it today? Family members may find it difficult to know how to help, what to say, and it may make any kind of planning difficult.

Final thoughts on the concept of fatigue. The excerpts presented illustrate the centrality of the experience of fatigue to the illness, and the apparent need for participants to feel that their level of fatigue was understood by me as the researcher as being significantly worse than an everyday experience of fatigue. This was of importance to PwCFS/ME because they often felt that others did not or could not understand what it was like. Certainly, I can say anecdotally that frequently when describing my research to people, many would joke 'Oh, I've got chronic fatigue', which highlighted the prevalence of a general level of fatigue amongst busy people in modern society, and an apparent misunderstanding of the extremities of fatigue in CFS/ME for many people who have limited knowledge of the illness. For those with CFS/ME I gained a sense that it was necessary for them to communicate the extent of their fatigue in order to distinguish their experience from the normal fatigue experienced by many people regularly, because they wanted to feel that I understood the extent of their suffering. For this reason, fatigue has been placed at the centre of the Problem Pattern as it reflects the fact that fatigue is where it begins, but it is also what it leads to; '*fatigue upon fatigue'* as articulated by Ally. It sparks the beginning of a vicious cycle, with the experience of intense fatigue and reduced activity leading to disability and dependence on others, the next aspect within the problem pattern to be discussed. We will see what this experience of fatigue means for relationships as the following concepts are described and discussed.

8.2. Disability: losing ability & being disabled

'Disability' is a label that refers to an individual's handicap or to a condition that limits someone's physical or mental functioning in the long term, and may hinder their day-to-day activities ("Definition of disability under the Equality Act 2010," n.d.). It is sometimes used interchangeably with the word 'impairment'. Because the concept of disability was controversial among my participants, I refer to their self-reported lack of or loss of ability as 'impairment' here. The concept of disability is also discussed.

8.2.1. Boom & bust. Evidence of all-or-nothing behaviour, known among participants as ‘boom and bust’, was present among participants and in many cases also appeared to contribute in the end to increased activity limitation on a long-term basis and to greater long-term disability:

> *“In ‘99, I was told ‘you should do what you know you can do’, alright, now my definition of knowing what I could do was doing too much…And once I went back [to work], I still had these symptoms…erm…I was pulling myself up the stairs by the bannister…”* (PwCFS/ME, Richard)

> *“I was… Pushing myself, erm, booming and busting all the time but you see I didn’t know…”* (PwCFS/ME, Steve)

> *“I think I just bumbled along for quite some time, and then I’d have periods where I did feel stronger and I could do more and I got into this bad behaviour of boom and bust. The moment I felt slightly better I’d go like a mad woman and do all these things I couldn’t have done before. I went on like that for many years…because when you haven’t had a lot of strength for a long time and suddenly get a little bit you just want to do everything! It’s just the nature of it really. But that’s the worst thing you can do...I was overjoyed I could do things…I was just like flogging a dead horse really trying to work and boom and bust really…”* (PwCFS/ME, Ally)

Unfortunately, when trapped in this cycle, recovery is desperately desired, but becomes less and less likely. As people move in and out of the ‘boom and bust’ phases of this cycle, it seems they move backwards and forwards from a place of hope and motivation to a place of dejection, depression and frustration, and this significantly impacts upon their beliefs about their illness and symptoms, and seemingly hinders their chance of recovery:

> *"I was doing this boom-bust cycle where I was thinking 'I don't feel quite as horrible today' so I tried cramming as much into the day as possible, so…I was just trying to do too much when I felt better, but then I was bedbound again. At that point I got into that cycle of bedbound, housebound, going out a bit…"* (PwCFS/ME, Sean)

So, it seems from the excerpts above that whilst all-or-nothing behaviour may suggest alternating between activity limitation and overdoing it, in bigger-picture terms all-or-nothing behaviour may be one stage of the progression into long term disability in CFS/ME, rather than one specific way of coping and managing.

This pattern of behaviour has consequences in relationships because it is a factor that contributes to dependence on others, and to the emotional challenges and stressors that this can bring into relationships within the family. But also, it can lead others to feel frustration and confusion about what the PwCFS/ME can and cannot do and why. Again, SOs can sometimes feel that they want the PwCFS/ME to try harder. For example, Sean's sister Sarah articulated feelings of frustration she felt at her brother's degree of inactivity, which she did not necessarily equate to disability as is illustrated by the apparent doubt about how her brother was handling his illness that can be seen in her words. I interpreted this as also attributing a degree of responsibility to Sean for both the perpetuation of his illness and his failure to improve. Also apparent in the following excerpt are Sarah and Sean's mother Sally's assumptions about how the situation could be handled differently by Sean, and by them, and also about the potential for conflict in attempting to navigate these issues:

Sarah:	*I think you've been good cop and I've been bad cop because I just go in and get at him really and initially he doesn't react well but after a while he thinks and thinks maybe she's right. That may not be the right way of doing it but it makes him think of his behaviour once, like one time I came back from uni and he hadn't showered and I said look this isn't what we do, get yourself sorted. Things like that, just… I don't know…*
Sally:	*I try be positive and persuade him and coax him because I don't want direct conflict. As much as I go upstairs and think 'urgh I'm so annoyed about something', I don't want to argue with him and send him back to square one.*
Interviewer:	*But you think the conflict sometimes is a good thing?*
Sarah:	*Yeah.*
Sally:	*She's on a shorter fuse than me.*
Interviewer:	*But it does sometimes have a positive impact?*
Sally:	*Yeah, it highlights what I'm trying to work towards it what he knows he should be doing but [Sarah] bulldozes straight through to it.*
Sarah:	*I'm very direct with my comments. I won't accept when he says I am doing this but I tell him straight how I see it.*
Interviewer:	*Do you think that works?*

Sarah: *Initially he gets annoyed and gets into ultra-stubborn mode but after a few days he says you were right or something.*

Sally: *I wish I could have somebody with me who could motivate me when I get really exhausted to go into him exactly the same time every morning and say 'right you've got 30 minutes before you get up for breakfast then you have an hour after that, but not pushing too much, to have a shower and get dressed, then maybes a walk around the block with the dog, then it will be lunch, then a break'…*

Sarah describes here how she made clear to Sean her expectations about how his days should be structured if he wished to see improvements in functioning. She and her mother together explained how they each fostered different dynamics with Sean in the ways in which they communicated with him and the expectations they placed upon him, and a belief that the combination of the two approaches might be helping him. However, they both experienced frustrations and found themselves lacking in confidence with what to do for the best.

Within the CBT model of CFS/ME it has been proposed that symptom experience interacts with cognitions and behavioural choices, which in turn has been shown to be associated with the perpetuation of symptoms. The beliefs individuals hold about their symptoms and about the best ways to cope with them is a key driver behind their behaviour (Knoop, Prins, Moss-Morris, & Bleijenberg, 2010; Nijs, Almond, De Becker, Truijen, & Paul, 2008; Vercoulen et al., 1998). When individuals focus on and/or catastrophise their symptoms, or believe that the symptoms are indicative of physical harm, they engage in one of two types of behaviours: 'All-or-nothing' behaviour, or limiting activity (Moss-Morris, 2005). Behaviours are adopted in response to symptom severity, with PwCFS/ME being more likely to limit activity when their fatigue and/or pain symptoms are perceived to be subjectively high, whereas all-or-nothing behaviour has been shown to be adopted when PwCFS/ME are feeling better compared to their worse phases of impairment (Band, Barrowclough, Caldwell, Emsley, & Wearden, 2017).

8.2.2. Activity limitation. In further support of the findings of Band et al. (2017), participants described becoming less able to take care of their own needs when pain and fatigue symptoms were high, and in the more severe cases, or when individuals were at their worst, even simple things like washing their hair, preparing a meal or having a conversation were depicted as being too much to manage.

The most severely affected had been housebound or even bedbound for a significant period of time, in some cases longer than 20 years, and had gone through a long and painful process of realising the extent of their impairment. In many cases, this had a significant impact on family members. PwCFS/ME experienced this as a complex process of loss – a loss of previous abilities, of their former self and of the day-to-day life they had imagined for themselves but not been able to live. Whether they were bedbound, housebound, or able to maintain some contact with the outside world, their life became drastically limited in comparison to the life they previously knew:

> *"I was just running on adrenaline for nearly a year and it just totally wiped me out…I couldn't even brush my own teeth…Couldn't even stand up to go to the loo…you can hear the anger and the emotion in my voice now, I'm still really in touch with that, how badly cheated I was, because it wasn't just my health, it was my little children who were two (crying)… And I couldn't brush*

> *their hair… Or help them… Or look after them… Because I was so ill (breaking down)…"* (PwCFS/ME, Rachel)

We see in Rachel's excerpt evidence of the activity limitation process in action when she was in a particularly acute phase of his illness, supporting the findings of Band et al. (2017). This is also evident in the following excerpt from Sean's interview:

> *"My sister was still around in the house and she was helpful and my mum obviously. But during the day I was alone just with the dog which was a bit rough when you're alone and feeling that ill. I could have used support at that point. They'd bring me food in the morning but I'd just stay in bed and the bathroom all day because I couldn't get myself downstairs and I didn't know how to try and it was … kind of rough."* (PwCFS/ME, Sean)

Sean's description of not being able to get himself down the stairs and not knowing 'how to try' is of interest. Clearly, he knew how to navigate stairs prior to his illness; being unable to get up and down the stairs was more likely a consequence of his subjective experience of his illness and his perceived level of impairment than of a physical inability to use stairs. His belief that he could not get himself down the stairs could be seen to be limiting his activity and capabilities.

A further example of activity limitation can be seen in the following excerpt from Ally. As she described to me what a typical day meant for her, it became clear that she had removed herself from the social world almost completely, not even choosing to watch television. This reduction of her world and activities had become normal for her; she had developed a routine to accommodate her illness:

> "*Usually around seven or just whenever I wake up, I'll go in the kitchen and get this liquid iron and have something to eat, wait 20 minutes and get a hot drink then. Maybe erm… I might watch the headlines then I'm back in bed for two or three hours, sometimes I read the newspapers online to avoid the depressing stuff, have three hours sleep and get up again and warm up a drink then back in bed. That's it really."* (PwCFS/ME, Ally)

Not only has Ally created a lifestyle for herself that limits her activity and isolates her from the world 'outside', she also actively avoids any flow of incoming information that could serve to further influence her low mood. Ally did not initially elaborate much on how she gets by, but she disclosed that she has a carer who does her food shopping, cleans and prepares her meals. She is confined to her bedroom and living room, and that is the extent of her world in a seemingly downward spiral of activity limitation, disability and isolation:

> *"[Long pause] Erm… horrendous fatigue. Mm. Very sensitive to light and noise, and smell and maybe a tone of voice - I can't bear it if someone's loud. I can't manage much telly or radio. And it's getting more and more things. My world is getting smaller and smaller and smaller… erm…"* (PwCFS/ME, Ally)

In all three instances their experience of fatigue and other symptoms led to their perception that they could not engage in these activities. They felt unable to meet their own basic needs such as going to the toilet or getting food and believed that doing so would be highly uncomfortable and possibly dangerous, with severe consequences. Because of these beliefs, they behaved in ways that aimed to limit the physical discomfort they thought these activities would cause, thus avoiding the potential consequences of exertion which they believe could worsen their condition. These examples support previous evidence of the CBT model of CFS/ME (Butler et al., 2001; Knoop et al., 2010; Moss-Morris, 2005; Nijs et al., 2008; Surawy et al., 1995; Vercoulen et al., 1998).

Activity limitation: A discussion. Although many PwCFS/ME believe that coping by resting and avoiding activity will improve their condition and limit their suffering, possibly leading to a cure eventually (Moss-Morris & Petrie, 2000), research suggests that this may not be the case. Coping by accommodating the illness has been found to be positively related to impairment, and disengaging with activities has been found to be positively related to higher levels of emotional disturbance (Ray, Jefferies, & Weir, 1995). Coping by avoiding activity been found to differentiate people with depression from PwCFS/ME, and it has been suggested that it may be one of the defining features of CFS/ME (Moss-Morris, 1997). Activity avoidance has also been found to be associated with higher levels of dysfunction (Moss-Morris, 1997; Ray et al., 1995; Ray, Jefferies, & Weir, 1997), increased fatigue severity (Vercoulen et al., 1998) and increased pain intensity (Nijs et al., 2008). Further, studies have shown that a reduction in activity limitation is a key mechanism through which symptoms improve following intervention with CBT (Heins, Knoop, Burk, & Bleijenberg, 2013; Wearden & Emsley, 2013). Particularly in cases of lengthy periods of illness, Ray et al., (1997) (1997) found that the less PwCFS/ME felt able to do, the less they were then able to do. This is related to the aforementioned controversial theory that physical deconditioning occurs in CFS/ME and causes a perpetuating cycle of disability and decreasing physical fitness (Lorenzo, 1998; Wessely et al. 1989). Therefore, some evidence suggests that PwCFS/ME reduce their capabilities further through believing themselves to be disabled, and this may be the process that Ally is going through that is illustrated by her description: "*My world is getting smaller and smaller and smaller*". This suggests that coping by limiting activity also has consequences in relationships. As individuals reduce their activities, they reduce their social interaction and social world. The less social interaction they have, they less they feel able to do. This will be explored further in later concepts.

However, there is also evidence that coping by limiting activity could have some potentially positive outcomes. Moss-Morris (1997) found that limiting activity was positively associated with a sense of control over illness. It has been suggested that in limiting activity levels, PwCFS/ME increase the predictability of their condition and may also successfully control their symptoms to some degree (Ray et al. 1997). In the case of Ally, her life and day-to-day experience is portrayed as low-stress, low-stimulation, and highly predictable. Moss-Morris & Petrie (2000) also suggested that the belief in the need to manage the illness by reducing activity may offer a feeling of legitimacy in removing themselves from stressful situations. If we refer back to earlier excerpts from Ally and Rachel (p. 99- 101) in which they described their stressful lives and social relationships prior to illness onset, these examples could be seen to support Moss-Morris & Petrie's suggestion.

8.2.3. Disability as a label. Whilst all participants described high levels of functional impairment, most were reluctant to apply the term 'disabled' to themselves or to identify as having a disability. However, they were usually accepting of the fact that they needed the kind of state financial aid afforded to those with a disability. These include the Disability Living Allowance, access to facilities for people with a disability, and parking badges. Accessing these services required that they were first willing to identify as having a disability, at least in a formal sense. However, the acceptance and use of the term 'disabled' was controversial for participants. The following example illustrates these issues:

Interviewer: *Do you use the word 'disabled' or 'disability'?*

Rachel: *No, I never use it, no, I don't like it.*

Interviewer: *Do you view yourself as disabled?*

Rachel: *That's really hard, I know that I am, I know that I am, and I use the disability route to get access to what I need, like blue badge parking, like seating at concerts, like seating on a train.*

Interviewer: *And why don't you like the disabled label?*

Rachel: *Erm…I don't know, it's the 'dis', it's the 'dis' – this guy on the radio Peter White, I think we talked about him last time, when he says 'It's no triumph, no tragedy, it's just how it is', it's just how it is for me, it's just how it is, and it is hard, but I don't like the 'dis' in the 'abled', because actually, I do work, I do contribute to society and contribute to the family in my own relaxed way.*

The period in her illness to which Rachel is referring in this excerpt is her present experience at the time of the interview, and was far removed from the period of acute impairment that was described in the excerpt on pages 122-123. At the time of interview, Rachel had been gradually improving over a period of several years, and had reached a point where she had shaped a life in which she was able to be a mother, wife and friend and was able to work and contribute to the family financially within the limitations of her illness. This will be discussed in more detail in later concepts. Of particular interest here is the meaning of the word disability for Rachel, and her reluctance to take on those meanings herself. Her words also suggest that she recognises that societal assumptions exist about someone who is disabled being of less value to society, less able to participate or contribute, and less able to have as fulfilling a family life as people who are not disabled. Even though, having met her, I feel certain that Rachel would not regard herself as someone who believes people who are disabled do not contribute to society, her argument regarding her reluctance to identify with the label seems to suggest otherwise.

As we have seen, the definition of disability relates to impairment that is long term and affects daily physical functioning. Whilst most participants were currently experiencing long term impairment that affected their daily physical functioning, a reason that some PwCFS/ME did not often identify as 'disabled' may be linked to whether they view their impairment as permanent, as well as to their self-concept:

Sean: *It's taken a lot for me to accept the labels or whatever that's associated. I don't see myself as disabled.*
Interviewer: *Do you get disability benefit?*
Sean: *Yeah.*
Interviewer: *Do you feel OK about that?*
Sean: *It's necessary in the short term. I don't want to be like that any longer than I have to. But I have taken full benefit of everything available.*

Identification with the label 'disabled' may seem like an admission of resignation to the illness, recognising their level of impairment as stable. Whilst some participants did appear to have done this, others did not. Rachel seemed to identify with a notion of acceptance of her situation, whatever that might be at a given time, without applying preconceived societal frameworks or limiting labels to her abilities and functioning, and Sean appears in this excerpt to be accepting of his current state and the social and financial needs arising from it, but neither identified as being disabled.

Disability as a Label: A discussion. There are many societal misconceptions about disability, and these have been addressed to some extent by the social model of disability (Oliver, 1983, as cited in Oliver, Sapey, & Thomas (2012). The social model of disability proposes that disability is a consequence of the way society is organised, as opposed to the medical model which views people as disabled by their impairments or differences, thus locating the disability within the individual (Marks, 1997). The perceived reluctance of some PwCFS/ME to identify as disabled may be a response to their belief that identifying as such places the disability within them, although research suggests that a majority of PwCFS/ME attribute their illness to physical causes (Ware & Kleinman, 1992; Wessely & Powell, 1989; White, Lehman, Hemphill, Mandel, & Lehman, 2006) and feel relief upon diagnosis (Brooks et al., 2014). It therefore seems counterintuitive that some may then be reluctant to adopt the 'disability' label. Future research investigating possible links between physical illness attributions and a willingness to adopt the label 'disabled' could offer interesting insights.

8.2.4. Final thoughts on the concept of Disability. Disability has been shown to be a complex concept in relation to CFS/ME. The evidence presented here illustrates that many PwCFS/ME experience at least a period in which they engage in all-or-nothing behaviour, and a period of severe impairment as a consequence of their illness. When considered alongside previous findings from the CBT model of CFS/ME, the data I have presented appears to offer descriptive evidence of the process of perpetuation of disability via activity limitation and avoidance behaviours. It also points towards a belief among PwCFS/ME that engaging in activities could cause harm and worsen their level of impairment in the long term, and that their perception of their level of impairment is influenced by their beliefs about the consequences of engaging in activity – hence, it is a vicious cycle in which perceived disability perpetuates disability. However, the data presented also offers some evidence that individuals may engage in these behaviours in an effort to try and control their illness. They believe that rest is helpful and necessary, with potentially better outcomes than pushing themselves through pain and discomfort, and there is some limited evidence that this could have positive outcomes (Moss-Morris, 1997; Ray et al 1997; Moss-Morris & Petrie, 2000).

The perceived reluctance to identify as disabled may be related to the perception of the meaning of the word 'disabled' among PwCFS/ME. Despite this, the excerpts suggest that PwCFS/ME do wish to be viewed by social support systems as 'disabled' in order to be able to access certain services and financial benefits in the short term, but this may be viewed as a temporary solution to a temporary problem. Thus, the reluctance to accept this impairment as stable which may be associated with their belief that rest may lead to an eventual cure, despite the fact that this is contradicted by research (Moss-Morris, 1997; Ray, Jefferies & Weir, 1997; Ray et al. 1995; Vercoulen et al., 1998; Nijs et al., 2008; Heins, Knoop, Nurk & Bleijenberg, 2013; Wearden & Emsley, 2013).

Ultimately, being disabled has consequences in relationships because it can lead to dependence on others and can place the SO in the position of being a carer and may fundamentally affect the nature of the relationship. Therefore, the reluctance to identify as being disabled may also be indicative of PwCFS/ME being reluctant to relinquish a sense of independence, or perhaps of struggling to accept their need for support from others. A lack of acceptance could suggest that they are in the disbelief phase of grieving for their pre-morbid levels of functioning, and possibly also grieving for their loss of independence. Disability and dependence on others have systemic implications because it leads PwCFS/ME to be unable to fulfil the roles they previously filled, and requires others to step in and fill the gaps. As we shall see, this can lead to complex family dynamics and a range of difficult emotions.

8.3. Dependence: Adjusting to dependence.

With impairment and/or disability there often also comes some level of dependence on others and so it is at this point that I begin to introduce the perspectives of the SOs, presenting dependence as an interpersonal process that must be navigated by the PwCFS/ME and SO, as well as the wider family. Indeed, in the following excerpts and concepts to follow, I will present an argument that although the suffering of the PwCFS/ME is clear, the entire family has to come to terms with a new reality. As we explore the concept of 'dependence' in more detail, we will see how the new roles of dependent and caregiver are also fraught with challenges for both the PwCFS/ME and the SO.

8.3.1. Dependence on formal carers. Participants with CFS/ME described how they had to rely upon the support and help of others to get by during their worst periods of illness. For example, we have already considered the descriptions provided by Rachel, whose husband fed her and helped her shower; Sean, who relied upon his mother and sister to bring him food in the morning; and Ally who required the help of a carer to take care of her basic needs such as preparing food. Sometimes, this dependence required input from formal carers, which emerges in the narratives as difficult to accept for the PwCFS/ME:

> "*When I was at my worst there was talk of carers coming in to look after me, we didn't end up doing it because of me being stubborn.*" (PwCFS/ME, Sean)

Despite Sean living with his sister and mother, the family investigated carers as a further support provision. It is interesting that we have previously seen Sean stating that this period was ‘rough’ and that he felt isolated and could have used extra support, and yet he states here that he actively discouraged input from outside others.

Ally did not have an SO to support her, and so for her, carers were the only option for her day-to-day support needs:

> *“I do have practical support. It’s a difficult one really. Shopping, cleaning. At the moment she comes twice but she might come more. Twice a week, sometimes three or four times. We’re [Ally and the carer] looking at her coming more regularly but she’s just moved house. Erm… yes, it’s a difficult one because you do need the help but the intrusion is just really… you wonder whether it’s worth it really. I take responsibility for that because I did broach the subject with her, I haven’t told her much about ME, she knows I’m not good on my legs, she’s a good person but she loves to talk so as soon as she comes in she’ll start. She’s not loud, she’s not animated but she talks and talks and my head is like an iceberg and it all pings off of it, and I can’t find the strength to say ‘just be quiet please’. It’s been going on two and a half years, and its only now I’ve felt the need to say something. But I don’t want to lose her, she’s a good person and drives me to hospital and all that… it started with an agency, I won’t say the name, but the agency have never really been that clever… the left hand never knew what the right hand was doing. It was all wishy washy and I felt so degraded. It was bad… for a start your independence is gone… They sent me a schedule, but in the end I just wanted them out of my life because it’s just more hassle.”* (PwCFS/ME, Ally)

Ally experiences dependence on others as distressing and intrusive, and the dependence leads to her feeling unable to advocate for her own interpersonal preferences in her own home for fear of losing something that she knows she desperately needs. This led her to feeling ‘degraded’, and to conclude that she had lost her sense of independence entirely. It was clearly a distressing experience for her relying on the provision of agency carers to meet her day-to-day needs.

8.3.2. Dependence on informal family caregivers. In instances where there was a SO to provide support, the whole family was affected by the level of disability and subsequent dependence on others. PwCFS/ME often described themselves to me as not wanting to be ‘takers’. The experience of being dependent left them feeling a sense of loss as well as feeling indebted to their SO for all that they had to do for them just to help them get through the day, and for the emotional strain that their dependence placed upon the dyad:

> *“I want to be a woman and his wife, not a dependent, like an old person. We have elderly relatives who are quite old; his Dad had a stroke and came to live with us over the summer, and it’s really tough constantly having to care for another grown up.”* (PwCFS/ME, Rachel)

> *“Most conversations now are to do with my illness. We rarely have conversations about anything else…I resent it a bit. It’s not what I want… If I was normal I would be... The relationship would be more equal at this point. It’s still very mother and dependent child at this point, and I wish it wasn’t. I wish I could do more to help around the house...”* (PwCFS/ME, Sean)

The parent/child dynamic was not just restricted to dyads that were parent/child. Rachel, who was for the purposes of this research a PwCFS/ME who also inhabited roles as a wife and a mother in her family system hinted at her level of dependence as affecting the family to the extent that it is like ‘having another child in the house’:

> *“Oh God, I’d just be like a third child in the house, sometimes I am that dependent. I can be so unwell that sometimes I need help with getting in the bath, getting out the bath, washing my hair, it’s just like having a toddler.”* (PwCFS/ME, Rachel)

This comparison of dependence as an adult with the dependence of a child was also noted by Helen, who is the Mother of PwCFS/ME Steve, when she described how she viewed the dynamic of her relationship with her 40-year old son who lived with her due to his illness:

> *“It was like having a child again…the relationship with me and [Steve] is that I’m his carer…I cook, clean, do his washing, you know, sometimes I will put something in the washer, but I cook his meals - in the past he has cooked his meals but then his arms ache or whatever, he loses concentration, the tea towel was too far near the hob and, do you know, things like that?”* (SO, Helen)

Helen’s use of the phrase ‘*his arms ache or whatever’* hints at a sense of frustration and is somewhat dismissive, which suggests that she may have grown weary of his inability to undertake simple tasks of self-care. This description does not suggest disability, it suggests a lack of self-sufficiency. However, it also suggests that perhaps there may be an element of dis-enabling in the relationship – he attempts to do things for himself but makes mistakes. Her comments on his mistake or her habit of taking over limit his willingness to push himself. He then knows that if he attempts to do things for himself but does not do them well or according to his Mother's standards, she will do them for him instead. Helen may therefore be encouraging her son's dependence upon her. However, it is also possible that Steve knows that his mother will step in to help him so that he can rest more. For someone who feels isolated and ill, the feeling of being taken care of may be a strong source of comfort.

The altered relationship and change in roles so that one member of the dyad became the dependent and the other as a carer was not necessarily restricted to just one family member, as was illustrated by a participant named Sarah who was sister to Sean. Sean had identified his mother, Sally, as his SO, but during my second interview with Sally, Sarah was also at home and asked to participate in Sally's interview. During this interview she stated:

> *"I think our relationship changed as [Sean] became ill because instead of a sibling relationship I was his carer as well…which is fine because I will do things for him but it definitely changed things because this frustration is there because I can see that he should be doing things that he just wasn't doing and I wanted him to eat and be up at a decent time and he wasn't. He was just in bed all the time, and I could see the effects of him being in bed all the time."* (SO, Sarah)

Sarah's phrasing 'our relationship changed as Sean became ill' suggests that the journey into becoming a carer was gradual and possibly unconscious. Here, Sarah is giving herself the label of 'carer'.

Dependence: A discussion. Dependence requires the input of others and therefore it generates a dynamic within which the illness exists. I have already presented research that has explored the impact of SO responses, which highlighted two core types of responses: solicitous and negative. Both types of responses have been identified as leading to poorer outcomes for PwCFS/ME (Brooks et al., 2013; Kerns & Rosenberg, 1995; Romano et al., 2009; Schmaling et al., 2000; Verspaandonk et al., 2015). On the one hand, Helen's responses evidenced by the excerpt above could be considered to be solicitous and therefore may be contribute to the perpetuation of Steve's illness. On the other hand, as his only source of support, it could be viewed as the only thing that is keeping him going and preventing a mental health crisis. However, Verspaandonk et al (2015) concluded that a focus on fatigue and impairment in conversations with SOs may decrease the ability of PwCFS/ME to engage with CBT interventions fully because it decreases their ability to be autonomous and active. Although Steve had not attempted CBT, it is possible that his mother's responses may unintentionally hinder his success in treatment if he did engage.

Similarly, if we refer to Sean's excerpt in which he stated that conversations revolve around his illness, we see further evidence that Sean's SOs may be unintentionally hindering his chances of achieving improved outcomes when in treatment. Despite Sarah's belief that her approach to helping her brother may be helpful, previous research suggests that both her approach (negative response) and his mother's approach (solicitous) may have negative consequences for Sean (Brooks et al., 2013; Romano et al., 2009; Schmaling et al., 2000; Verspaandonk et al., 2015), although these studies explored responses in couple relationships, whereas Sarah is Sean's sister and Sally is his mother.

Band et al. (2014) applied the EE framework, exploring SO responses in broader dyadic relationships including partners, parent/child relationships and one sibling relationship. High EE (emotional overinvolvement - EOI, critical comments and hostility) was associated with poorer outcomes. Families with high EOI tend to be over-protective, self-sacrificing and emotionally intrusive. Hostility is characterised by a negative attitude towards the PwCFS/ME relating to a belief that the PwCFS/ME can control their disorder. A combination of hostility and EOI within relationships leads to critical attitudes – the family members may not blame the PwCFS/ME for their illness, yet may criticise their behaviour relating to it (Vaughn & Leff, 1976).

It seems from Sarah's description that she sometimes used one style of response and then at other times she used another: on the one hand, accommodating her brother's illness by comforting him and doing things for him, as her mother did (solicitous responses/EOI), but on the other hand sometimes expressing frustration at him (negative responses/critical comments). But predominantly, she focused on pushing him by criticising his behaviour and expressing frustration, whereas Sally is clear that she finds it difficult to be anything other than solicitous/EOI. Because both approaches may, if measured, be found to rate highly for EE, it seems likely that both Sarah and Sally's responses could be counterproductive, and based on previous findings that high EE correlates with poorer outcomes in a range of conditions (e.g. Garcia-Lopez et al., 2009; Hooley et al., 1986; Leff et al., 1982; McFarlane et al., 1995; Vaughn & Leff, 1976), it is therefore possible that in their attempts to help Sean, Sarah and Sally may both simultaneously be inadvertently contributing to poorer outcomes in the long term. This begs the question, what might be a better approach? This question is addressed in Category C. Balance and Category D. Acceptance.

Also worthy of note here is Sperry's assertion that CFS/ME can be especially disruptive if it occurs at a transitional point of the family life cycle (2012). The Family Life Cycle is a model of change and development which proposes that specific types of families typically experience common patterns which are influenced by shifts in the demands of internal and external pressures, requiring adaptation. It is not necessarily relevant to all emergent family models such as single-parent families, blended families, or couples without children (Sperry, 2012). It proposes six key stages through which families transition: Leaving home, forming a stable couple relationship, Birth of children, Living with adolescents/Mid years relationship, Launching children, and Retirement and old age (Dallos & Draper, 2010; Sperry, 2012). Through each stage, families are thought to strive to maintain the family structure and sense of identity whilst also adapting and evolving in response to demands. (Dallos & Draper, 2010). There are three levels of the system proposed: at the centre of the system is the 'Nuclear family' – the immediate family members. This exists at the centre of the 'Extended family' which is the broader family unit. The family exists within the 'Social System' and the networks, institutions and perceived cultural expectations that exist within a given family's social system. Intergenerational traditions have also been highlighted as important aspects in understanding family patterns of relating and functioning (Carter & McGoldrick, 1988).

Sperry (2012) suggests that in the case of CFS/ME, a parent with a physical impairment during the 'Birth of children' phase of the family life cycle is essentially lost as a parent. Sperry posits that the family becomes, in a practical sense, a single-parent family due to the level of activity limitation and perceived physical impairment. This view may be controversial, as it suggests that an individual is no longer capable of parenting if they are physically impaired. A better way to view this might be to see the PwCFS/ME as limited in their ability to contribute to family functioning in the ways that they had previously. It should also be noted, however, that not all PwCFS/ME experience such acute levels of disability. CFS/ME is unlikely to have this level of impact on the families is all cases, but certainly it seems there is some evidence in my sample that this can be the case. For example, for Rachel's family her illness occurred shortly after childbirth and so we see how her level of impairment hindered family adjustment at this crucial stage of development in the family system. However, it is interesting to note that there can be some positive outcomes; Soderberg, Strand, Haapala

& Lundman (2003) found that husbands of women with fibromyalgia (an illness often compared to CFS/ME) reported feeling more involved in the upbringing of their children because their wife’s illness necessitated greater childrearing responsibilities for the husbands.

Final thoughts on the concept of dependence. In presenting the concept of dependence, I have highlighted the relational and systemic nature of CFS/ME by demonstrating that the illness experience, that is, fatigue, disability and subsequent dependence on family members, does not exist separately in the PwCFS, independent from the family. It impacts the SO, and in turn, their coping strategies and behaviours have an impact on the PwCFS, as has been highlighted in recent studies (Brooks et al., 2013; Romano et al., 2009; Schmaling et al., 2000; Verspaandonk et al., 2015). But, as was highlighted by Sperry (2012) and more recently by Band et al (2014), this is not restricted to couple relationships, neither is it limited to just one family member; the whole family system is affected by the dependence that CFS/ME necessitates. In presenting the perspective of a PwCFS/ME’s sister who was not identified as the primary SO by the PwCFS/ME, my work extends evidence for the impact of CFS/ME on the family system, and also points towards interactions with others in the family system as being highly influential in the management of CFS/ME. The excerpts presented from Sarah, Sally and Sean support the findings of Band et al (2014) and therefore also provide evidence of high EE in some dyads in the context of CFS/ME. As we move into the following concepts, I will unpack EOI a little more by exploring an interactional aspect of relationships that is particularly present in EOI, enmeshment (Khafi, Yates, & Sher-Censor, 2015), and was especially evident in two dyads in particular (Steve and Helen; Sally and Sean).

8.4. Family coping processes: Too much support and too little support.

Supporting a PwCFS/ME is a challenge, even in relationships where interactions are more positive and the relationship seems to function in ‘healthy’ ways. Levels of functioning may fluctuate daily or even hourly, people may have different levels of functioning depending on how much rest they have had and what they have been doing. In these conditions, finding a routine and rhythm is difficult and nobody can really know what to expect. The family finds itself constantly having to evaluate each day, often several times within a day, how much support to offer or how much to do, so as not to cause friction in the relationship. In these strained conditions, relationships are put under considerable pressure; the daily struggle can leave SOs feeling that they do not know quite what to do for the best and are ‘walking on eggshells’ (SO, Sally):

> *“…I think his illness has taken the fun out of the relationship most of the time and it’s made things difficult now particularly as he’s got older that you’re walking on eggshells. You’re trying to keep the peace, encourage, support and push a little bit, but we do go from one crisis to the next quite honestly.”* (SO, Sally)

> *“He’s never been violent or anything but he ignores me, he shouts and bangs doors, do know what I mean? (Pauses) but I leave it to his choice what he does…I make him a chicken broth and he does that, just boils it in a pan, and he does wash the pan and dish but when I cook an evening meal his plate just goes in the sink and I do the washing up with my husband. He’s not always like that, sometime he will make himself something, we seem to go down a path and stay there for a while but then something will change and we’ll go down another path…It could be just [Steve]’s mood, his mood for the better. Or something you*

> *say. I can never shout at him saying 'Will you just do that' because I know there will be repercussions like the anxiety."* (SO, Helen)

> *"I think he [boyfriend] almost felt sorry for me, 'cause he couldn't understand what was going on and he could see that I wasn't very well but he didn't know what to do and anything he tried wouldn't really help. Erm, so yeah then after coming back, shortly after, we split up…"* (PwCFS/ME, Amy)

It is apparent in these narratives that SOs wanted to help, and wanted to make the situation in some way better for the PwCFS/ME, but they perhaps also wanted to keep the peace and maintain the homeostasis within the family system. It seems that sometimes they may feel that they are being taken advantage of or that the relationship is unfair and not equal. However, in other situations, it seemed to be the case that the SO just did not know what to do, as is illustrated by Amy's narrative, and in the following example where Rob describes how he thinks his dad might feel as a SO trying to offer support:

> *"My Dad probably felt a bit, to be fair he probably felt a bit helpless really, it's a bit like well what, you know, you know what I mean? I mean what can you do really? I mean…he used to do things, he used to you know obviously do things round the house and try and help you, you know, with things like that…'cause he's always very good…he's a helpful person, he will help people, you know…he would do anything to help you out…but I think he just felt he was probably, he didn't really know what to do to help, you know what I mean? That was, that was obviously, and like I say, that's not a criticism towards him at all because you know, it's, you know, he, he just didn't really know what to help, and you know he did what he could…"* (PwCFS, Rob)

It is in the way Rob spoke that he really struggled to evaluate his Dad's input, and to articulate that evaluation. He found it difficult to say something about his father's strategies for helping him through his illness that he felt would be a negative evaluation, but it is also clear that his Dad did what he could but with limited knowledge about what might be best, he felt lost. I got the sense that the SOs in these examples would try anything they could think of to ease the PwCFS/ME suffering, but to little avail. Recognising or communicating needs was revealed as a constant process of trial and error which gives rise to tension and frustrations:

> Interviewer: *What could your mum and sister do now to keep those relationships healthy?*
>
> Sean: *Encourage me rather than try to fix.*
>
> Interviewer: *With CFS/ME it's very difficult to fix, so anyone trying to fix is going to get frustrated.*
>
> Sean: *It's that question mum asks, what can I do to help? It's encourage [SIC] and support that I need, it's hard to find the balance.*

I viewed the process of trying to get it right as being like a dance whereby the PwCFS/ME and their SO manoeuvre around one another cautiously. I theorised that there are three sets of social circumstances, conceptualised simply as 'Too much support', 'Too little support' and 'Striking and maintaining a balance'. The latter is the most positively constructive set of actions that a PwCFS/ME and their SO can co-construct, and it can lead to improved outcomes for both, so that they can dance together. 'Too much support' and 'Too little support' on the other hand, contribute to the problem pattern because they perpetuate problems. As we have already seen in some of the examples presented so far, if people around the PwCFS/ME give too much support, the PwCFS/ME stops doing, or trying to do, things they might otherwise have been able to do. On the other hand, if the SO gives too little support, or if there is not an SO around, then the PwCFS/ME may be insufficiently supported and they try to do things they perhaps ought not to be doing, making them feel more unwell.

8.4.1. Too much support: High cohesion or enmeshment? Within the sample I noted that in dyads where women were in the role of carer, the narratives suggested that they took on far more emotionally than the male participants whose role was as a carer. In order to illustrate the concept of offering too much support, I will predominantly draw upon the content of interviews with two mother/son dyads where the son was the PwCFS/ME, and in which I saw aspects of enmeshment and emotional over-involvement (EOI) as a thread running throughout the eight interviews conducted with these individuals. The individuals concerned are Sean and his mother Sally, and Steve and his mother, Helen.

Relational patterns: Sean & Sally. SOs can have a considerable influence on the illness beliefs, journeys through healthcare services, and access to treatment that a PwCFS/ME experiences. In this first excerpt, Sally refers frequently to ‘we’ meaning herself and her son, illustrating her close involvement in every aspect of her son’s journey. She constructs her experience of supporting her son as a ‘fight’ against another – health services - which she appears to view as one group entity:

> “*We couldn’t get any help from doctors so we got him referred and we got him the diagnosis that he was gluten intolerant, and changed his diet and then he got some nutrition and that did improve his condition. But again, that was me fighting for it and being called an overprotective, fussy mother… I don’t feel that [health services] blame me, I think they thought I was fussy and that it*

> *was all in his head, and if I backed off he'd be better, rather than 'I know my child and I know what he wants'. He was a sportsman and what a joker he was, and look at him now. You know?"* (SO, Sally)

Sally had previously told me that she is coeliac so it was of interest to me that she reported being instrumental in his receiving a diagnosis of gluten intolerance – upon reflection, I wondered why having that diagnosis was of importance, as improved nutrition and a gluten-free diet could have been trialed without having first received a diagnosis if she had suspicions. In a sense, his diagnosis of being gluten intolerant made them more alike. Her use of the word 'we' I interpreted as indicative of her perception that she and Sean were one unit fighting against the others (health services). Her description suggests to me that she felt she was being judged by others for her involvement in trying 'find a solution', and that she was aware that there may be other ways of responding to the situation. But she rationalised her behaviour and involvement on the basis of the quality of her relationship with her son, so her belief that she knows her son better than anyone else knows him concludes with her assumption that she should know better than anyone else what is best for him. This perception influenced her thought processes, beliefs and subsequent actions, and therefore has consequences for both her and Sean, as well for those who come into contact with the family.

The fixation on diagnoses and on receiving validation and support from external sources was also a consistent thread running through Sally and Sean's interviews, as is illustrated in the following extract:

> *"It has been easy to slip through cracks…. If I hadn't persevered with GPs and found one that's understanding I'd just be left hanging. I know people who haven't had a good GP for years and have just been left alone. The only*

> *reason I didn't get to that point was because of my mum's bullheadedness really. She ran around, made phone calls, and bullied people into talking about it, trying to find a solution. I didn't have the energy at that point, or confidence and will, to do that for myself. So yeah… if Mum hadn't done that I would have been isolated to the full extent with no support at all."* (PwCFS, Sean)

The core feature of this excerpt that particularly jumped out for me was Sean's description of Sally's approach to helping him. I interpreted this as Sally trying to advocate for Sean, but this advocating had developed into a sense of desperation in trying to help her son, translating into desperate action. The strong terminology Sean used, 'bullheadedness', 'ran around', 'bullied people' is suggestive of frustration and subsequent aggression, firm and fixed beliefs and expectations, but also of Sally having taken the problem on as her own. Whilst Sean reports that this approach yielded positive outcomes for him, in a separate part of his interview he disclosed that there had been consequences for his Mum, also hinting at his illness serving a function for his Mum in her construction of her own identity:

> *"Erm… she… after the divorce she kind of [long pause] I don't know… she just decided that her life was just going to be caring for me and my sister and she didn't have much of a social life then and that was what she chose at the time. When we got older and realised she'd been doing that, isolating herself in ways, we encouraged her to stop that. Ever since I've become ill she has become as isolated as me really."* (PwCFS/ME, Sean)

Sean's illness had started in his early teenage years. His portrayal of his mother's life and purpose as being built around her role as a mother and carer, and he and his sister's concerns about the vacuum this may have been creating, led me to be curious about the function of Sean's illness in the family dynamic. Writing in my research journal after the second interview with Sally and Sean, I began commenting on this curiosity:

> *"It feels like every time [Sean] pipes up and advocates for himself, states what he needs, or what he feels, he thinks he has gone off topic. He loses confidence in what he is saying and looks to me for redirection, reassurance. Is this part of his role and the dynamic he has with his mother? Does he need to look to her for reassurance to know how to feel? Does he feel he has the right, or the ability, to express his feelings? It seemed to me that he was acutely self-aware. This reminded me of my interviews with [Steve] whose behaviours indicated similar processes."*

I had also noted that towards the end of the first interview with Sean I had gained a sense that Sean was indirectly expressing a feeling of suffocation by his mother. I interpreted his words and communication as an indication that her behaviour towards him felt over-protective to him, an indication of emotional overinvolvement, but also that he felt that she really depended upon him and his sister and that this was limiting his ability to develop a sense of freedom and independence. For example:

> Sean: *It's been so long now for my mum caring for me that she feels a duty or responsibility of care no matter what I say to convince her that I would be okay for her and my sister go for a few days to see my aunt. It took me a lot of convincing, a lot of promising… I don't know how different my mum is to other carers. Even though she does*

a lot to support me, she still feels guilty for going out to work for the day and leaving me without anyone else. That's difficult for all of us even though I know she has to do it. She wants to spend as much time here as she can even at cost to her. If there was something available I would encourage her to do it for sure.

Interviewer: *And how would that affect you? Do you think it would change you?*

Sean: *It would bring some pressure off me, it sounds selfish but yeah.*

Interviewer: *If your mum was happier do you think it would make you happier too?*

Sean: *Definitely yep. I'm conscious that one of the reasons she isn't social and doesn't do things she enjoys is because she thinks it would make me unhappy.*

Interviewer: *Have you spoken to her about this?*

Sean: *Yeah*

Interviewer: *And what was the outcome?*

Sean: *Not much. I don't phrase it well when I talk to her about it. We end up arguing because it comes out wrong. She feels like I'm rejecting her help which isn't what I mean, or want her to leave me to it, or… it's back to the trying to fix things again… I want her to encourage me to get better in my own time. (Pauses) Redirect me…*

Interviewer: *Well, I was asking how it would help you if your mum was happier.*

Sean: *Okay. It would be a positive impact. I want her to be out socialising. I want her to be able to trust me to be more independent. There's times where she doesn't trust me to be in the house alone. It's ridiculous to me. I have told her, and my GP that even though I have suicidal thoughts, I wouldn't act on them because of everyone else but she needs constant reassurance that that's the case. It's not just that, it's like if I collapsed whilst she was out or something. She won't go on holiday to a different country without me or without my sister being here.*

This description suggests that Sean felt he lacked autonomy; where an individual is autonomous, they are able to make decisions independently following a period of personal reflection, without involving others. Being able to make decisions autonomously gives individuals a sense of control over events (Sensen, 2013), which will be discussed later. Perhaps because Sean's illness onset was during his teenage years, the years in which one would normally be going through the process of individuation, he had missed out on the social opportunities to develop in a such a way. This may be one reason for Sean's struggle to be and to feel autonomous. I viewed this situation as a 'double-edged sword' because without his mother's emotional closeness and practical advocacy, Sean had expressed a belief that he would feel completely isolated and depressed, so he did perceive that there was benefit in her approach to his situation:

> *"I think if we weren't as close as we were already then things would have broken down. After Grandad died, Grandma moved in with us so it became the*

> *four of us so we ended up all relying on each other and became very close knit. So, if we weren't as close I think I would have ended up on my own and become even more isolated."* (PwCFS/ME, Sean)

Although Sean felt that having such a close-knit family was his saving-grace, I was curious about whether, had he been able to develop a sense of freedom and independence, he could have found solutions or ways to prevent or tackle this social isolation for himself? There is evidence that Sean suffers from social anxiety and that his social development has been affected by his situation:

> *"I've had a few panic attacks when I was out, just two or three…it was in places that I didn't know, with people I didn't know very well, with my Dad once and talking about stressful things under a lot of pressure."* (PwCFS/ME, Sean)

However, there is also an argument that this could be a side effect of his illness and the social isolation that his symptoms enforce upon him, rather than it being a consequence of family patterns:

> *"Social isolation for so long. Been ill for so long. Been in my head for so long, it's hard to process and vocalise…it makes an impact on my approach to social situations and how I interact with people. It's kind of a side effect of an illness really. All these years of social isolation…erm, before I was ill I was a little quiet, a little shy, but since being ill I feel I have lost all social competence and my confidence has just dropped from the depression and isolation. I can cope one-on-one if it's on a specific topic. I was a little nervous before you arrived. If I have to go to a social event or family event it becomes a bigger deal*

> *than it should be for me. I worry about how people see me, if I'm going to faint or pass out which is highly embarrassing. I have to do a lot more thinking and preparing."* (PwCFS/ME, Sean).

This illustrates how he views his confidence as having slumped as a consequence of six years of social isolation, not having been part of society, or experienced the 'typical' developmental stages that his peers would perhaps have done as they moved from school to college to university or working life. In this way of viewing Sean's situation, it could be considered that his mother offers a lifeline in an otherwise alienating and isolating world. Possibly, the relationship is enmeshed, but perhaps it is also just highly cohesive because of the daily challenges they face in trying to deal with Sean's illness. Either way, this may be more of a consequence of the shrinking world that they inhabit together and the rejection they have experienced from outside others such as healthcare professionals, rather than purely a consequence of psychological and relational dysfunction within the family system that was evolving prior to the onset of illness.

Steve & Helen: Relational patterns. Steve and his mother, Helen, had a similarly complex dynamic. Helen explained that she felt the rest of their family (her husband, Steve's siblings) did not fully understand the situation, but she described maintaining Steve's distance from the family because she felt that this protected Steve:

> *"…they don't know all the details….they know that he is ill you know he gets tired and all that but they don't know all the details of what goes on between me and [Steve] because like some of the things he said I know there'd be a rift between them and Steve and I don't want that…sometimes he says you know he isn't very nice and shout and I've got to do these things and all this and I know they'll think, Mam let him do it himself, they don't fully understand, and my*

> *husband doesn't fully understand and do you know he's great you know, but he doesn't fully understand the illness."*

Helen is making assumptions here about how her family will view her son's illness, and during the interviews I gained a sense that Steve was very isolated within the wider family, that there was a lack of compassion for him. It seemed that his mother was everything to him – carer, friend, confidante, protector – but that this was perhaps limiting him from exploring other possibilities for intimacy with other people. In her attempts to protect him, she was unwittingly preventing him from having any sense of autonomy and casting doubt in his mind when he did make attempts to be more independent, which could again be considered to be an example of enmeshment:

> *"I mean he actually went on to a dating site last year but it's the emotions that that threw up about even just putting a little profile on the website do you know what I mean? And I personally think that he is not ready for that kind of thing but I could not say like 'You can't look after yourself, you're not going to be able to look after somebody else'..."* (SO, Helen)

Although Helen states that she did not verbalise her doubts to Steve, it is likely that, considering that this is the only intimate relationship in Steve's life, he would pick up on her doubts and gain a sense even without realising it that she believes his attempts to move forwards are doomed to fail. Further, her doubts are to a certain extent based on the assumption that an intimate relationship would require Steve to take care of another person, but it occurred to me that there is every possibility that there are other individuals with similar challenges in their life who would cherish companionship. Helen was so concerned about the consequences of things not 'working out' that she was apprehensive about him even trying. It struck me that she did not want him to get his hopes up because she finds it so painful to see when things do not meet his expectations, but also because of the consequences for her and her husband when Steve faces disappointment or his depression takes a turn for the worst:

> *"Well actually he did seem to get a little bit better and he did move into a flat, erm, I didn't really want him to move into the flat, I didn't say that to him, but he did move into a flat and he was there, we decorated it lovely, he had it lovely and everything, and he used to come home for his tea on a night but he wasn't really coping and then the final straw was me and my husband was going away, we was going to Italy and then he was round and he was down… and we didn't go on holiday…we didn't go…"* (SO, Helen)

It is easy to see why Helen takes such a protective stance towards Steve – it seems she is in a bind. If she tries to help him be independent and it goes wrong, then she has to witness how low he becomes, and she is the person who has to pick up the pieces. But if she does not help him to be independent, then she remained trapped as his most significant other.

Too much support: A discussion. It became evident to me that Sean and his mother's relationship had reached a point which in family theory might have been termed 'enmeshed' (Barber & Buehler, 1996). Enmeshment is an aspect of family functioning (Barber & Beuhler, 1996) and has been defined in family research as patterns within families in which members become fused emotionally and psychologically. It is thought that in such fusion, the process of individuation is inhibited and psychological maturity is unlikely to be developed and maintained (Barbarin & Tirado, 1985; Barber, Olsen, & Shagle, 1994; Greenberger & Sørensen, 1974). Individuation is a concept in analytical psychology, describing the process through which an individual develops an integrated sense of self independent and differentiated from others (Jung, Read, Fordham, & Adler, 1953). However, Olson, Sprenkle, & Russell (1979) suggested that high levels of cohesion equate to enmeshment. Olson et al (1979) defined cohesion in families as emotional bonding between family members, with levels of cohesion varying from low cohesion in which family members have emotionally disengaged from one another, to high levels of cohesion in which they are enmeshed emotionally. The ideal level of cohesion for family functioning is the mid-level, whereby family members have balance between separateness and connectedness, whereas being disengaged or enmeshed are seen as problematic in families. Other theorists suggest that cohesion ranges from low to high and that high cohesion indicates healthy functioning in families, for example, Moos (1974) suggested that cohesive families consist of family members who are committed to the family and concerned for another, being helpful and supportive of one another. However, Barber & Beuhler (1996) considered enmeshment and family cohesion as separate constructs, whereby cohesion measures supportive interaction, and enmeshment measures psychological control. They defined enmeshment as follows:

> *"Enmeshment is not an element of supportive relationships but represents controlling and constraining interaction patterns that inhibit psychological autonomy. Such psychological control has been found to be negatively related to healthy functioning…Confounding cohesion and enmeshment has impeded the theoretical and empirical examination of the proposition that humans develop best when they feel close to significant others and yet feel free to explore and create an independent sense of self."* (p. 434)

Barber & Beuhler (1996) also demonstrated that cohesion was associated with lower levels of both internalising and externalising problem behaviours in adolescents, and that enmeshment was associated with increased problems among youths and with the internalisation of problems, thereby supporting their argument that they are separate constructs. Barber et al. (1994) suggested that enmeshment is usually accompanied by an intrusion into a child's psychological development that serves to inhibit the process of individuation and causes anxiety in the child who then lacks the personal skills to be able to manage social interactions. This could be seen to reflect the social anxiety experienced by Sean and the family patterns presented in the above excerpts.

If viewed through the lens of systemic theory (Dallos & Draper, 2010), it is likely that there is a dynamic, interactional, circular relationship between Sean's illness, the high cohesion/ enmeshment that exists in their relationship, and Sean's lack of individuation and social development that has developed into a feedback loop. We have also seen evidence of triangulation in their relationship as Sean's sister Sarah provided her account of the family experience of Sean's illness. The relationship between Sean and Sally has become a closed system, with Sarah sometimes returning into that closed system, but generally living away from it (living away at university). A major stage in the family life cycle – Leaving Home – has been disrupted by CFS/ME for Sarah but clearly also for Sean who has never been able to leave home, and as Sperry (2012) points out, this is likely to lead to negative social development consequences for the child. In spite of his illness, systemic theory would view the family as striving to maintain structure in their system as they attempt to adjust to the illness. As Sean states, he has been ill for so long that they have fallen into habitual patterns; Sean's illness may have now become a stabilising influence on the family system and on levels of family cohesion, preserving homeostasis and serving the function of keeping him and his mother together, thus preventing individuation. If this is the case, then it is possible that no matter how much Sean and his mother want to change, they have developed such habitual ways of relating to each other that change becomes deeply difficult.

I also constructed a view that she and Steve were in a deeply enmeshed relationship and closed, circular system, to the detriment of all other relationships in their lives. But because Helen had physical and social abilities beyond Steve's, the relationship was imbalanced – for Steve, she was his lifeline, whereas she had a life beyond him; she engaged in and was a part of a broader open family system of which Steve did not feel a welcome part. Steve was reluctant to engage with the open system because he felt his siblings viewed him negatively, and she both protected Steve but also maintained the distance between him and his siblings by keeping the truth of Steve's situation and the extent of their enmeshment from them.

Like in Sean's family, in Steve's family the life cycle had been disrupted by Steve's illness in late adolescence so that Steve has never been able achieve the individuation that would allow them to be separate yet connected – the 'healthy' characteristics of balanced family cohesion (Olson et al., 1979).

Helen communicated a genuine fear that he may end his life when at his lowest points, and this was a default position to which Steve returned whenever he feared losing his proximity to his mother, his caregiver. He would say or do whatever it took to bring his mother back to him when he experienced the anxiety associated with abandonment owing to her continuing with her life beyond their relationship. As well as being indicative of enmeshment, which we can recall involves psychological control (Barber & Beuhler, 1996), this is also suggestive of anxious-resistant attachment behaviour (Hazan & Shaver, 1987).

Bowlby's (1907-1990) Theory of Attachment (1969) built upon earlier psychoanalytic ideas around personality and adult relating styles being rooted in childhood experiences. It draws on evolutionary psychology to argue that proximity to the primary caregiver is crucial to survival for an infant, and therefore early patterns of relating aim to maintain proximity to and prevent separation from primary caregivers. Mary Ainsworth developed these ideas by proposing three styles of attachment in infants that develop as a result of the responsiveness of the primary caregiver to the infant's needs (Ainsworth & Bell, 1970). The first, secure attachment, is a pattern of infant-parent relating in which the parent is responsive to the child's needs and the child therefore feels that the parent provides a safe base from which to be able to explore the world beyond the attachment relationship. The other two, anxious-avoidant and anxious-resistant are insecure and dysfunctional styles of attachment whereby the parent is inconsistent in meeting the needs of the infant, and may occasionally or frequently be rejecting of the child. The infant displays conflicting behaviours designed to punish the caregiver, whilst also wanting comfort (Ainsworth & Bell, 1970).

It has been argued that these infant-caregiver attachment patterns form the basis of adult relationships (Hazan & Shaver, 1987). Individuals with a secure attachment style in adult relationships tend to have healthy relationships with successful strategies for communicating and meeting one another's needs. Meanwhile, adults with anxious-resistant attachment patterns tend to be 'needy' and dependent in relationships and can be easily angered if their attachment needs are not met. Those with anxious-avoidant attachment styles tend to appear disinterested in having close relationships and actively avoid dependence on others and others becoming dependent on them.

It is also interesting to note that whilst we are exploring adult relationships in this research, some of these are couple relationships, and some are parent/child relationships, such as Steve and Helen's. In these relationships the nature of CFS/ME can lead to the PwCFS/ME taking on the role of dependent whilst the SO becomes the caregiver, so it can be useful to consider these infant/caregiver attachment styles in relation to patient/caregiver behaviours in the sample. This was visible whether the dyad was a couple or a parent/child dyad.

Final thoughts on the concept of 'Too much support'. Systemic thinking views problems and symptoms as interpersonal rather than individual. Prior to the introduction of systems theory, those who experienced mental health difficulties faced the problem of oppression within the family system and the stigma of the consequences they experienced, because their problems were viewed as being within them as an individual. This new thinking viewed problems that had previously been considered in psychiatry as individual phenomena were presented instead as consequences of interpersonal processes; relationships are central, and communication and interaction are part of the construction of the problem as well as a part of the solution (Dallos & Draper, 2010). These ideas are inherent in my analysis and subsequent recommendations, as are more specific aspects of systemic theory. It is beyond the scope of this document to provide an entire overview of family therapy, and some aspects of systemic theory are of less relevance than others to this work. Therefore, I have cherry-picked the aspects of systemic theory that best fit the data and concepts, outlined below.

Interdependence of each member of the system is stressed in systemic theory, with each individual within the system seen to influence others and vice versa. This is viewed as a continuous process of action-response-action-response between individuals, so that the actions are seen as responses and responses are seen as actions – these patterns are called 'circularities'. Circularity can be seen in each concept within the model I have presented, and we shall see examples of how relationships with SOs can become dysfunctional circular patterns of enmeshment and dis-enabling or withdrawal and isolation in CFS/ME, both of which I argue lead to poorer outcomes. There is also a common pattern in families related to circularity whereby two people experiencing conflict tend to draw in a third person, who then becomes involved in these repeating circular patterns. This is known as ‘triangulation’ and this can be seen in some of the excerpts presented (Dallos & Draper, 2010).

Also prevalent in systemic theory and of relevance here are the concepts of open and closed systems and family homeostasis (Dallos & Draper, 2010). Open systems have fluid boundaries that allow interaction with information from the outside world so that it flows into the system and information from the system flows into the outside world. Closed systems have more rigid boundaries that are not easily crossed, so that the outside world is kept out. Within open systems, feedback tends to lead to escalation, such as conflict which leads to physical conflict or the breakdown of a relationship. Closed systems tend to utilise feedback to stabilise and maintain existing patterns and thus maintain ‘homeostasis’ within the system. Much like the body’s tendency to attempt to maintain systemic balance, Jackson (1957) theorised that family systems do the same. A symptom develops within the system, with one or several members of the family, and the circular process of action and response kicks in. It becomes an integral part of the system, and therefore serves a function maintaining the stability, balance and familiarity within the system. Resistance may be encountered if attempts are made to change the symptom or system more broadly. The system operates as a whole and all participants become engaged in striving to maintain homeostasis, and the resistance to change may be expressed as unconscious emotional responses in one or more members of the system. For relationships to be functional, both patterns must be present; open system functioning allows the system to change and adapt to internal and external changes, provided the relationship did not break down entirely. Completely closed systems are unable to adapt and change in the face of environmental changes. Relationships containing ‘pathology’ tend to function as closed systems, whereby any change in one individual within the system leads to actions by others within the system that actually serve to reduce change rather than enabling it. Family members claim that they want change but symptoms and habitual behaviours relating to them have become integral to the maintenance of current relational dynamics and to the return to or maintenance of homeostasis (Dallos & Draper, 2010). There are multiple examples of closed and open system functioning in my findings, and of expressions of desires for change within family systems alongside evidence of behaviours that serve to reduce change and adaptation.

In relating the examples presented to the concept in question, ‘Too much support’, I have presented two dyads in which the common theme of enmeshment is apparent. Enmeshment emerges as a consequence of circularity in relational processes - both a consequence of and a cause of the SO giving too much support and the PwCFS/ME becoming too dependent upon one individual to meet all of their physical, social and emotional needs. The more a person does for another, the more dependent both individuals become on that relationship, so that the dependence experienced by a PwCFS/ME is deepened thus disenabling them further. But, the SO also becomes dependent on the PwCFS/ME – they structure their reality, roles and routines around the PwCFS/ME so that any change in the PwCFS/ME’s behaviour or well-being becomes a threat to the closed system’s homeostasis. Although these dyads could be communicating high levels of cohesion, it seems more appropriate to view the relationships as enmeshed because of the level of dysfunction and lack of individuation that was also evident within them. Ultimately, the dyads in question here put forward narratives that suggest that the ultimate consequence of enmeshment is deepening social isolation and social anxiety, as well as family patterns that have become entrenched and stagnated. This will be explored further in upcoming concepts.

As mentioned before, it is worth noting that these were both parent/child relationships and in fact mother/son relationships. Future studies may yield useful results through investigating mother/daughter dyads, comparing the ways of relating with mother/son dyads, and similarly with father/son and father/daughter dyads. However, as the purpose of constructivist grounded theory is not to be able to make generalisations based on specific examples, but rather to examine the fabric of specific examples in order to develop an understanding of the meaning in other peoples' constructions of their experiences, it is not a failing to have no mother/daughter comparisons, but rather offers ideas for potentially fruitful future research. There was a further mother/son dyad in the study, Rob and Kath, and Rob's father, David, also participated. There was no evidence of enmeshment in their narratives, rather, they appeared to have been mutually supportive but appropriately separate and in fact, Rob had moved away from home and made a full recovery. This is expanded upon further in Category C. Balance.

8.4.2. Too little support: Emotional disengagement and isolation. Within the present sample, there was one dyad in which the PwCFS/ME communicated clear feelings that the SO did not do enough to support them, and two participants who had CFS/ME and had no SO that they felt appropriate to participate in the research (neither had a romantic partner and both lived alone). They were interviewed in the hope that their stories might offer some clues as to what features of a relationship in CFS/ME might contribute to it breaking down, and I was also interested in what aspects of life with the illness might stand in the way of an individual who considers themselves to have no SO from forming new relationships, or leaning more on existing ones.

What struck me most about these three individuals was how disengaged and isolated they each appeared to have become. However, through the stories they told, I started to develop the feeling that all three were behaving in ways that actually served to put distance between themselves and other people and prevent others from getting close enough to be able to help – that perhaps they were not allowing others to provide support to them. Perhaps this was a consequence of having been let down by people in the past, or feeling misunderstood and therefore not wanting to let anyone get close, or it may have been an effort to protect themselves from the guilt of being a ‘burden’ and to protect others from the burden that the PwCFS/ME believe themselves to potentially cause. In disengaging themselves from others, it seemed that, much like the cycle of enmeshment described above, these individuals had slipped into similar and yet opposite self-perpetuating cycles of disengagement, and, like in enmeshment, the ultimate consequence seemed to be increased social isolation. In this section, I will provide evidence of the consequence of receiving limited support from others.

Disengagement due to SO negative responses. Eileen, a PwCFS/ME who was aged 82, and described herself as having been ill with what she now believes to be CFS/ME since her early 20s following childbirth. Eileen described a relationship with her husband that was disparate and disengaged and lacked physical affection. However, she did not believe that their relationship was like that because of CFS/ME. It seemed instead that she was inferring the issue was in the relationship and CFS/ME just happened to occur at the same time:

Interviewer: *Do you think you can explain to me the impact that you think CFS had on your marriage?*

Eileen: *Can’t blame it – see I didn’t know it was CFS right at the beginning, you see it created – the mercury poisoning created that thing, and so we took it as something else. It might have, it was just illness. I mean it could have been the same if I had rheumatoid arthritis badly or anything like that. It could have just have been, it didn’t have to be something like that. Or somebody that began to develop multiple sclerosis…*

Interviewer: *So, this attitude that you said that [Brian] had was that 'it's not my problem'?*

Eileen: *Yes, 'get on with it'…Yes, 'look after yourself' and you know, and 'sort it'. And so, he's never investigated, as I say, he doesn't want to be part of it.*

Interviewer: *But you wish that he had done?*

Eileen: *Well it would have been a great help, except that I now accept that this is something I've had to experience. And what I've experienced has helped me to understand that other people have been supportive…If I started to talk about something related to the illness and he'd just go 'ah she's at it again' sort of thing (rolls eyes to imitate husband).*

Interviewer: *Is that a response you get quite often, the eyes rolling?*

Eileen: *Well, I, then I try not to talk about it you see…*

In the excerpt above, we can see evidence of responses that might also be classified as negative responses in that they are not perceived by the PwCFS/ME to be supportive or understanding and instead are challenging and critical. We see how these negative responses caused Eileen to become reluctant to communicate how she felt with her husband, and to withdraw. However, it seems unlikely that this was a consequence of her illness and instead was a product of the way in which Eileen and Brian related to one another in general; Eileen is clear in this excerpt that she does not blame her illness for the difficulties in her marriage, but she also stated that she felt that her illness had made her less able to cope with any elements of dissatisfaction in their relationship:

> *"Loving is very different from the marriage thing. You don't have to have a relationship to care, shall I say. And erm, so, I felt it was my job to stay and care, apart from not having the energy to shift. And I made my vows in the beginning, and tried to go on with that in that way… I probably didn't have long enough to build the marriage before I was ill… [In the early days] we seemed to be happy, and yes, we worked alright together. I married him…I think probably if the illness hadn't occurred then I could have coped much better with the fact that there wasn't the friendly, caring relationship about the house, with [Brian] looking after things."* (PwCFS/ME, Eileen)

It is clearly evident in Eileen's description of her husband's behaviour that she perceived that he exhibited a lack of emotional support and engagement, but there is also evidence of hostility and critical comments, and of Eileen feeling that she was being blamed for her illness and criticising the things she did in her attempts to get better:

> *"I say the practical thinking moved in, He would take me here and there, and whatever I had to do there. But the other, would mean an understanding of the illness, and I didn't think he has that in any way. And he said a comment, 'oh, you make yourself ill with taking all those supplements'. Now, I'm taking the supplements as a result of going to a specialist who was a dietician…But he didn't, I would hope that he would have read the odd book you see, and find out why I'm taking it, but he didn't really want to know why I was taking it. So, I think it might have annoyed him being ill."* (PwCFS/ME, Eileen)

Her husband Brian described his wife's lifestyle and in doing so revealed behaviours that suggested that Eileen may have been taking an active approach to maintaining the levels of disengagement between herself and Brian by living very separately despite the fact that they are living in the same house. He reveals his belief that she lives this way because of her illness, and in fact during his interviews I did not get the sense that he had felt as dissatisfied with their marriage as his wife had. He was quite comfortable attributing the difficulties to Eileen's illness:

> *'Well it changed over a period…it wasn't one day it is this and the next days it's that…we still did things, we went to the odd function, you know, and all the rest of it, but over a period of years we have gone right to the other end of the spectrum, somewhere in a year or 10, so that [Eileen's] biggest ask now is if she goes into the garage. She is basically housebound, which is no fun for her, I'm quite sure of that, and her timing is all upside down in that she spends a lot of time in the bedroom of a daytime, comes downs at various times in the evening or through the night because that's the way her system's acclimatised…but over the years there has been less and less simply because she does something and it triggers her.'* (SO, Brian)

However, on Eileen's part, she was clear that he had been unsatisfactory in the way that he had coped, and there was an undertone of resentment for this throughout her interviews. She seemed to have accepted that her husband was not supportive in the way that she wanted him to be, and that this had led her to become more isolated than if she had had an emotionally supportive SO:

Eileen: *If he cared I feel he would have come to find out what other people were like with this illness, and then be able to understand it. So, I think that's what it was, indifference. I think that will be how I would feel it. And so, as I say it's been difficult to talk about the illness because we've got this eye rolling business…so I haven't been able to share the problems in that way which would have helped.*

Interviewer: *You would have liked somebody to share with and talk to about it?*

Eileen: *Yes, yes.*

Interviewer: *Instead of feeling isolated*

Eileen: *That's right, that's right. And you see people who live on their own, that is a great struggle for them, the ones that I talk to…one of the ladies said to me, she loved her husband dearly, but erm, [she] said '[Eileen], sometimes it's such a relief that he isn't here' – this is after he died – because she didn't have to be responsible for him, she could go at her own pace, that seems an awful thing but I know exactly what she means.*

Interviewer: *Do you feel in some ways it would be easier on your own? Because you wouldn't have the concern for somebody else's behaviour? Mood?*

Eileen: *To a certain extent yes… I'm feeling that perhaps if I moved to somewhere that I was being cared for, the lack of worry and anxiety would again improve that, because I would be stronger in myself, so there is that there, yes, better on my own.*

Disengagement due to SO negative responses: A discussion. Eileen asserted that she would be better on her own. SO critical comments and hostility have been found to be associated with increased fatigue and disability in CFS/ME, and negative SO responses are associated with increased depression in the PwCFS/ME (Band et al., 2014; Kerns & Rosenberg, 1995; Romano et al., 2009). Eileen is certainly not alone in finding her the responses of SOs upsetting. In a qualitative study, Dickson, Knussen & Flowers (2007) found that as well as facing the distress caused by delegitimation from medical professionals and outside others, 11 out of the 14 participants they interviewed also experienced delegitimation from their partners because they felt that their partners did not believe that they were ill. Dickson et al. (2007) describe how deligitimising encounters with their SOs are described as the most difficult thing for PwCFS/ME to cope with, particularly because they experienced it as rejection. One particular paragraph within the paper that describes how participants wished things had been mirrors Eileen's excerpt above:

> *"They also reported that understanding and support from their partners would have been both comforting and reassuring in providing the sense that they were not alone in their illness, and many suggested that such support and acceptance would have raised their own quality of life as well as facilitating their coping. There also appeared to be a great need for, and expectance of, a shared truth, a reciprocal acceptance and support between the participant and their partner. When this was not met, participants' accounts were tinged with anger, resentment and bitterness"* (Dickson et al., 2007, p.860)

Eileen's statement about her husband not reading about her condition, not seeking to understand, illustrates her desire for the 'shared truth' to which Dickson et al refer.

Dickson et al. also reported that the consequence of relationship difficulties and communication breakdowns was isolation.

Disengagement through fear of deligitimising experiences. PwCFS/ME, Amy, who was single at the time of the interview, explained how she felt her social circle had decreased through not being in a relationship, but her description also suggests that she holds back from the possibility of a relationship because of her fears about the impact she might have on the life of an SO:

> *"It is quite daunting to be honest, I think it also links in to almost having the expectation of yourself and all the expectations of the other person and also not wanting to be a burden, so yeah it makes it…for me…it's made it a barrier, quite a big barrier I would say. And to be honest, I'm walking much better than I was but it's still a barrier, rightly or wrongly…even if someone said to me come and visit me in a week or in a month I'm already panicking and it's a month away…I have a picture of myself where I should be able to do this and therefore I shouldn't have to expect that, I think that's what would be difficult. I don't see why somebody else should have to compromise on anything."*
> (PwCFS/ME, Amy)

I interpreted this as Amy communicating her fears that in order to be in a relationship she would have to be a certain way – and that she feels she falls short of what would be expected of her at present due to her illness. Therefore, she avoids the possibility of a relationship through fear of not being able to be enough for someone, and of consequently experiencing negative responses from them. This perspective is reflected by Dickson et al. (2007) who described how participants experienced loss of friendships due to a lack of ability to live up to peoples' expectations of reciprocity, or what they perceive other people's expectations of reciprocity to be.

Amy highlighted expectations and compromise, signalling that she feels that in order for another person to be in a relationship with her, she would have to live up to their expectations, which she felt she would be unable to do. She felt that this would put pressure on her, and the other person would have to give more than they take which she found to be unacceptable to her. Her solution at the time of interview was to avoid intimacy completely. I asked Amy whether she was comfortable with the situation:

> *"Mostly, mostly almost too comfortable because I do kind of want to be in a relationship but at the same time I can't bring myself to do it, not that to say marriage isn't going to come around, it's… yeah… I'm kind of, I'm comfortable, but not as happy as I could be…I suppose a relationship would be a difficult one, with age and everything else, yeah, it's something that comes across my mind. Everyone else is in relationships, so your social circle kind of decreases, there is a bit of a negative to the situation."* (PwCFS/ME, Amy)

Given that she had described to me having already been in two serious relationships that had broken down, and that both of these broke down primarily due to her illness, I perceived it as possible that she was trying to protect herself from further pain by avoiding intimacy at this stage.

Amy presented as somebody who tries to make the most of her life and maintain a positive mind set despite her illness, but I imagine that a positive mindset must be difficult to maintain if you are feeling emotionally drained by the breakdown of a relationship. Further, when a relationship breaks down and this can be perceived to be a consequence of the illness, the sense of rejection and depletion may be particularly poignant. Perhaps when every day is a battle just to function, the prospect of further emotional strain through a poorly-functioning relationship, or through the breakdown of a relationship, may just feel like too much of a risk to take.

Disengagement through avoidance of social relationships. For Ally, it seemed she also held the belief that being in an intimate relationship would render her a burden, and she preferred being alone to having another person who was forced into becoming her carer. The fear of not living up to the expectations of another and of not wanting to be a burden were also present in the way she spoke about relationships, as was a recognition that she had become emotionally disengaged from others:

> *"I thank God every day that I never had kids because I've had a lot of voluntary work with kids who risk reoffending… and kids need parents to be emotionally and physically there for them and I'm not. If I'd have stayed with my partner, he'd be the best person to look after somebody because he was so kind. But there is always the added pressure of being the partner because then the other person becomes the carer I supposed. In my own warped way, I'm pleased I'm on my own."* (PwCFS/ME, Ally)

In using the phrase 'In my own warped way', Ally is acknowledging that she knows that it may not be considered a societal norm to be content with being alone. It felt that, much like Amy, Ally was not saying that she would not like a relationship, but that she felt having one would put too much pressure on her because of her situation, and so she felt safer and more able to protect herself without one. I did wonder, however, whether there might be a hint of regret in her statement about her ex-partner. I asked whether or not she had wanted a relationship in the past:

> *"When I was younger maybe. I do like…I know a lot of people suffer from loneliness but I'd quite happily not see people for a few weeks. I'm quite happy on my own…"*

I did not interpret Ally's preference for being alone as being entirely a consequence of her illness; as she spoke of her feelings about being in the company of other people, it became evident that this may be due to her personality rather than her illness. I also wondered whether her illness actually gives her a get-out clause that excuses her from having to interact with others, without having to take responsibility for the fact that she simply would prefer not to have to deal with other people:

> *"I don't mind [the company of others] as long as they are quiet and not over demanding, and if they are slow…When I worked I was on the telephones and always with people and I liked that but I liked coming home to retreat to silence afterwards…so that there was balance…[Silence] just doesn't take anything off you really. Like you coming today, so I've had days before this where I've prepared for it and days after where there will be silence."*
> (PwCFS/ME, Ally)

The idea that illness may offer a get-out clause when it comes to socialisation was a reflection I noted when interviewing Amy. Her statement about panicking whenever she made plans with other people suggested that there was an element of psychological pressure that she was placing upon herself based on her perspective of herself and her ability to cope, that led to social anxiety and a reluctance to make plans with others or actively socialise. This was not restricted to Ally and Amy, it was also present in the descriptions of almost all participants with CFS/ME, but it was particularly poignant for them because of the lack of presence of other people in their day-to-day lives.

Disengagement & attribution. There may be secondary gains socially for PwCFS/ME who find social situations stressful, because they are able to attribute their perceived inability to participate in social interactions to their illness symptoms rather than blaming themselves for failings in their social abilities, or through having to admit that they would prefer not to socialise. To illustrate, an excerpt from Brian describes Eileen's reluctance to participate in social interaction even with her family:

> *"I've got some friends, I was best man at their weddings, I know their wives, and ever since I have known [Eileen] she has known them as well…we have a monthly meeting just for lunch but [Eileen] hasn't been for about 18 months to see them because she can't face it, she loves it when she is there but is knackered when she comes back, she may be in bed for two days when she comes back…it's a nice run out, normally you would say that, a run out to (Seaside town) with a meal provided and a chat with your friends, nice but as [Eileen] will say it just takes too much out of me, even with the family. I mean I went to our daughter's for a meal and [Eileen] said you'll have to excuse me…because she can't face the trip in the car and the chat."* (SO, Brian)

It was frequently described by PwCFS/ME that even having a conversation can feel like too much of a strain for them due to their illness, as stated by Brian above and echoed by Ally:

> *"There are some days where I think I'd like someone to come in with a warm bowl of water and a flannel, and not really speak to me because I can't be doing with the interactions, and just wash my face, make me a cup of tea and just go. I'd be quite happy with that."* (PwCFS/ME, Ally)

However, upon further exploration, it emerged that in Ally's case this may have been symptomatic of her general attitude towards socialising rather than simply symptomatic of her illness. Upon my asking whether she enjoyed socialising prior to her illness, she responded:

> *"Oh, I couldn't stand it much then anyway. Some Christmas dinner one time… people were talking and my head was just…I couldn't stand it…the noise."* (PwCFS/ME, Ally)

So, we see that whilst Ally and Amy are certainly socially isolated, perhaps this is not always entirely a consequence of the illness. Rather, the illness can in some cases become a functional reason for the PwCFS/ME to avoid social activities. This suggestion could, in some cases, incite anger by suggesting this there is an inference of responsibility on the part of the PwCFS/ME, and an inference of them having some degree of control. I believe however that there is sufficient evidence supporting this assertion presented here. I gained a sense that social isolation breeds social isolation – the more time participants spent on their own, the more challenging and tiring any form of socialising became, and the less they felt they needed or wanted to socialise. This is reminiscent of the deconditioning theory of CFS/ME (Wessely et al, 1989), but applied to social and emotional aspects of the illness instead of solely the physical aspects.

Disengagement: A discussion. The concept of 'disengagement' has featured in the CFS/ME literature previously. Moss-Morris & Petrie (2000) offer a brief summary of this concept, describing it as a coping strategy that is regarded as different from activity limitation or avoidance; it refers to a process of giving up completely when faced with something the PwCFS/ME finds stressful or overwhelming. Disengagement has been found to be linked to a habit of focussing on symptoms (Ray, Weir, Stewart, Miller, & Hyde, 1993), and has been consistently found to be related to poorer outcomes in terms of fatigue, disability and psychological distress (Antoni et al., 1994; Heijmans, 1998; Moss-Morris et al., 1996; Ray et al., 1997). Disengagement has also been found to distinguish PwCFS/ME from people with chronic pain and healthy controls, as they were significantly more likely to cope with stressors by disengaging (Blakely et al., 1991). However, Moss-Morris and Petrie (2000) explain that this relates more to disengaging from dealing with CFS/ME, offering examples of participants who disengaged as follows:

> *"[it] is so incapaciting that one can virtually do nothing to fight back. One can only wait until the extreme weakness has lifted"*
>
> *"When I'm in relapse I just can't cope…sometimes I get tired of being ill and wish I could die, because I feel so awful"*

(Moss-Morris & Petrie, 2000, p. 112)

However, in using the term disengagement here, I am referring to the disengagement discussed in the family literature as very low family cohesion (Olson et al., 1979) which in the examples above have led the individuals concerned to isolate themselves completely from SOs, in particular, romantic partners. The examples I have presented suggest that some people may, at least to some extent, make a choice to disengage from others socially and emotionally, partly following previous experiences, especially deligitimising ones, and through subsequent fear of the anticipated consequences of engaging in the behaviour. Therefore, we could view social and emotional disengagement as another form of the activity limitation and avoidance that has previously been described in CFS/ME populations (Band et al., 2017; Heins et al., 2013; Wearden & Emsley, 2013). However, rather than the limitation of social activity being due to symptom experience as suggested by Band et al. (2017), the evidence presented here suggests that limitation of social activity in these examples may be a consequence of cognitions about the expectations and reactions of others as well as about perceived burden and guilt about this burden. However, PwCFS/ME articulate that their avoidance of social activity is in direct relation to their symptom experience, which suggests that they do have an internal narrative in which they excuse themselves from social activities due to their beliefs about their illness. This supports the findings of Knoop et al. (2010) that the behaviours of PwCFS/ME are driven by their beliefs about their symptoms and the best ways to manage them.

Abramson, Seligman & Teasdale (1978) described how an individual's tendencies in explaining the cause of events affects how they react to it. An internal attribution is made when a person attributes the cause of events to an aspect of themselves as a person. They take personal responsibility for it and this has a negative impact on self-esteem. For example, if a person attributes failing an exam to themselves not being clever enough, this would make them feel more negative about themselves.

External attribution is the process of attributing the cause of events to something external, thereby removing personal responsibility preserving self-esteem – for example, if they attributed it to the exam being harder than in previous years, then they would not feel that failure was their own fault.

Responsibility and blame have a role to play here. Oakley (1992) explains that causal attributions and blame are different. With causality, we seek to explain a cause, which may infer responsibility but does not necessary assign blame. If a person believes another to be responsible for something, then they believe them to be blameworthy. When one blames others for events, they are assigning moral or legal responsibility to them for the events and making external attributions. When they blame themselves (self-blame), they are making internal attributions.

In CFS/ME, it is possible that the illness sometimes becomes a part of the story people tell themselves and others about why they behave in the ways they do so that they do not have to admit that they would sometimes prefer not to engage in certain social activities, thus preserving their self-esteem. Even though the illness is largely described in dominant medical explanations as something that is ‘within’ them, if they do not perceive it as something over which they have control, then they view it as something external. There is a possibility that the same individuals might have similar patterns of behaviour in relation to socialising in the absence of illness.

8.4.3 Too much support and too little support: Different paths to the same destination. If we return to the concept of family cohesion and the model presented by Olson et al. (1979) in which disengagement and enmeshment are the extremities on the continuum of cohesion, we can see in the examples presented thus far that both disengagement and enmeshment seem to co-exist with social and emotional dysfunction, and that they are both self-perpetuating cycles. Both disengagement and enmeshment appear to have some form of relationship with PwCFS/ME and SO illness attributions and beliefs about symptoms and management, as well as with fears about how outside others perceive the illness, and with the experience of perceived rejecting attitudes from others.

It is also likely that these patterns of relating have their roots in attachment patterns, and an in-depth psychodynamic study may be able to trace the nature of these adult relationships in the context of CFS/ME to childhood attachment patterns. No such study exists to date in the literature. It does seem clear that having too much support or too little support can be both a symptom and a cause of relationship dysfunction, and can have negative consequences for the PwCFS/ME. Where there is a SO, it can also have negative consequences for them, in particular because they both serve to cause ever-greater social and emotional isolation.

Of course, in this way of looking at social interactions in dyadic relationships, I propose that there is another dimension – introduced earlier as ‘healthy’ levels of family cohesion. Because successful adjustment and balance has positive outcomes, this is not considered as an aspect of the problem pattern and is instead presented as a separate concept. Within this concept, The Problem Pattern, we now move on to take a deeper look at some of the emotional experiences within relationships between PwCFS/ME and their SOs.

8.5. Fear, frustration & anger

We have already seen in a number of excerpts presented that fear, frustration and anger can become prevalent emotions experienced and sometimes expressed within the relationships of PwCFS/ME and their SOs. The PwCFS/ME fear what the future will hold and that they may never make a recovery. They fear and feel frustrated by the rejection they perceive from others and the stigmatisation that accompanies the illness. They fear getting worse which then affects how they manage their illness (activity limitation). They become increasingly frustrated by their condition, their limitations, their failed efforts at improvement, the lack of answers offered by health professionals, and the dissatisfactory interactions they have with others including friends and relatives.

> *'[I felt] scared, really scared as much as anything… er frustrated *long pause* erm…I was frustrated, frustrated like hell. I wouldn't say I was depressed at all, I was determined like hell to kick the damn thing into touch and do something about it…'* (PwCFS/ME participant Richard)

> *"I'm terrified of going back and doing too much like I did at A-levels because that was the darkest part of my depression. I was isolated, and didn't understand all these symptoms I had."* (PwCFS/ME participant, Sean)

The distress of living with fear, frustration and the isolation that has already been illustrated can lead to anger, especially when PwCFS/ME and their SOs find it difficult to communicate what they are feeling. In the current study, it became evident that anger and/or resentment was expressed more frequently by those who were in relationships where enmeshment was a feature, but that it usually emerged following a period in which members of the dyad had struggled to express their emotions. In the following excerpts, we see Steve's Mum's description of the consequences of his anger, the kind of things that triggered it, and her recognition that expression of anger was symptomatic of other emotions that he struggled to communicate. This is followed by Steve's reflections on his anger:

> *"…My daughter was having fireworks on the night time…so I'd done his tea, he wasn't going, erm, and we go and then I'm just driving to my daughters*

> *have my granddaughter with me, and my phone rang so I pulled over and I answered it and there was just a horrible message on my answerphone saying 'I've kicked the fucking door' and he'd thrown his dinner all over the bedroom, so I just took them to my daughters and I just come back home, I mean [my husband] wanted to come back home with me and I said 'No I'll just go' and he was just sat crying and he just couldn't cope any more you see, we'd been out having a good time the night before and then we was going to have the fireworks and then he had a counsellor who he didn't see that often and I just did not know what to do, he wouldn't let me ring the mental health team, I mean he'd kicked the door you know, he'd kicked it upstairs, there was a big hole in it… He didn't know how to tell me what it was, he just showed it with anger."* (SO, Helen)

> *"The worst period of my life was getting the diagnosis. Before that it did get really bad because I wouldn't be able to sleep for days so I just tried getting through the day. But after I got the diagnosis, I got it really through speaking to [Eileen], I then had to accept what she was saying. And then of course because the GP had got it wrong, the anger…I started smashing things up and I've never done that before and I did take it out on my mum…It made me so angry. I was just in the anger for such a long time."* (PwCFS/ME, Steve)

Sean also described tension and anger in family interactions and difficulties for him in communicating how he was feeling. This first extract is one we have seen before, but illustrates the mutual frustration both Sean and Sally experienced:

> *"It's very frustrating conversations, mostly the same questions over and over. My mum wanting to know how to help and me not having an answer. Both of us being frustrated by the situation."* (PwCFS/ME, Sean)

Sean went on to explain how frustrated discussions had become arguments, presumably when anger started to emerge in the communications, but also that these arguments can then lead to disengagement:

> *"We've had recent arguments about this before, so I'm not communicating and I feel I can't communicate enough…"* (PwCFS/ME, Sean)

Sean explained that the anger expressed within the family when they tried to talk and problem solve together was one of the things that made it difficult for them to communicate effectively as a family, and suggested that as a family they could have benefited from an external source of support to help them through these difficulties with communication:

> "*We could have done with someone who understands my illness and has a perspective of carers caring for someone with my illness. Someone for all of us to talk to simultaneously, so we could go to an external place with someone outside the family." (PwCFS/ME, Sean)*

Other participants also expressed anger and frustration at their inability to function to their desired level, but these emotions tended not to be directed at the SO in dyads where enmeshment was not a feature of the relationship, or where there was no SO. Instead, the anger was an emotion that was directed at outside others. Rachel's anger was directed mostly at her previous employer who she blamed for pushing her to work hours that they had previously agreed she would not work because her health was ‘fragile’. In the first interview, she explained how this had happened:

> *“I’d thought I’d put everything in place so I wouldn’t have a relapse but I was totally…it was out of my hands because I was so badly cheated by the line manager who is no longer at the [employer], they were sacked and they were in disgrace and they had to leave soon after I left because they’d done it to a number of staff I think…so you can hear the anger and the emotion in my voice now I’m still really in touch with that, how badly cheated I was”* (PwCFS, Rachel)

In the second interview she explained the long-term consequences of her illness in her life, and the anger she still feels about the loss she has suffered:

Rachel:	*I just can’t be the Mum that I want to be, or the colleague that I want to be, or the wife I want to be, I give as much as I receive (in tears)…*
Interviewer:	*What do you think it is that makes you emotional and upset when you’re talking about this? What’s the emotion you’re feeling at the moment?*
Rachel:	*(Long pause)…angry?*
Interviewer:	*Anger…mmm…What’s the anger about? What’s the anger at? Who are you angry with?*

> Rachel: *That it's not fair. It's not fair that I can't be the mummy that the girls deserve or the wife that [my husband] deserves…I feel a sense of loss, grief.*

Kath also experienced anger; hers was experienced mostly in the process of trying to get a diagnosis, or at least an explanation of what was going on, and she expressed her frustration at healthcare professionals. However, the outcome was more positive for her because she did receive what she perceived to be useful input following her angry outburst, and also felt validated and empowered by her ability to advocate for herself:

> *"I said 'When is somebody going to pull their finger out and tell me what this thing was that I had in 2004 that has triggered all these symptoms?' And, erm, because I got angry, which I shouldn't have had to do, he went running after the head rheumatologist in the next room and he said oh just a minute, and off he went and he said 'oh he shall see you in a minute', and I said 'I'm sorry to have to get angry with your staff but,' I said, 'This has been going on since 2004, I said it's now 2007' and I said 'I feel worse than I ever did', I said 'and I'm getting no answers, and am fed up of just coming here and being fobbed off and not getting anywhere', and I said 'it's about time somebody put all these things that are in my notes together and made some sense of them', you know but I, I, I, I think 'What happens to people who were not like me, who can't stand up for themselves? (*PwCSF/ME, Kath)

Sean's mother Sally experienced anger towards a healthcare system that would not fund treatments for her son, communicating a sense of desperation at the lack of support available to him, and the constant need to fight for the right to access care:

> *"…[I feel] like I'm knocking my head against a brick wall. This is my son, a bright and intelligent lad who has had a really rotten time without hardly any support and he's sitting in front of a committee board and they're saying he isn't an exceptional case but to me he is."* (SO, Sally)

A further example of anger towards the healthcare system, but also of blaming others, can be seen in another excerpt from Helen as she described how her son blames his GP and her for his condition:

> *"I knew something was seriously wrong so I went to the doctor asking about it and the doctor just said it wasn't and that I had to accept it. But I think that put a barrier on our relationship, because even now [Steve] still blames me for not fighting harder…I think it's because I wasn't 100% and the doctor is a doctor, isn't he? I just trusted the doctor.* (SO, Helen)

Helen went on to describe her belief that Steve blamed her for his situation, which may have been pivotal in the development of the resentment that had developed between them:

> *"Well yeah, I might start getting emotional when I think about it really…I think [Steve] does feel I failed him really but I don't think I did fail him because of all the things I have done and he has took [SIC] over my life (pauses, tearful and visibly upset)."* (SO, Helen)

Interestingly, Steve's mother also expressed feelings of anger towards her son about how her son's illness has impacted upon her life, although this seemed to manifest more as resentment. Her anger seemed to be more due to the imbalance in their relationship, and she blamed him for taking over her life:

> *"Inside I feel anger at him and think I'm sick of doing this… At times it's like [Steve] has got blinkers on and it's just self. He just sees it, he wants what he wants and that's the little child…I wouldn't say I show it as anger. I feel angry inside but it's not always come out. I've just moaned and said I'm sick of this… I think because of the way my life has been took over I've got the right to feel that way. But I don't show it to other people…I organise everyone and I like to go out and have fun. I see myself as quite a happy person, but sometimes the situation with [Steve] takes the happiness away from me."* (SO, Helen)

Helen suggests here that she feels her son's behaviour and level of dependence on her is not acceptable, but she does not say anything to him about it. She chooses passivity instead of expressing her feelings and lets him get on with what he wants instead of challenging him. We have already seen in previous excerpts that she fears the consequences of challenging him, and that may be why she chooses not to assert her own wants, needs and feelings. However, in doing so, this gives way to mounting feelings of anger and resentment within the relationship, as well as dissatisfaction and continued dysfunction. Although we have seen that hostility and critical comments from the SO can have negative consequences for the PwCFS/ME (Band et al., 2014), it is important that the needs of the SO are also considered. In such challenging circumstances, an individual cannot be expected to completely ignore their own needs in order to support someone else. This in itself is likely to exacerbate dysfunction in a relationship and therefore give rise to further damaging tensions and frustrations.

Finally, it is worthy of note that anger is something that seemed to depend upon an individual's outlook, personality and coping style; not all participants described feelings of anger:

> *"Some people refer to anger, but I can't say myself I felt particularly angry, because I wouldn't know what to direct the anger at really… other than illness and how on earth do you get angry at an illness, if you are angry what do you do with it?"* (PwCFS/ME, Richard)

However, it is also interesting to note that although Richard did not feel angry or describe any anger in his relationship with his wife. I interpreted in Caroline's narrative a private resentment of the impact upon her life, and a sense of feeling as though she had been taken advantage of, as well as frustration at the self-focus that the illness seemed to have encouraged in her husband, according to her perspective:

Caroline: *People say it's all in their head, they'll get over it.*

Interviewer: *How did you feel about that?*

Caroline: *Well it's all about "me". It's a good title is ME because it's all about me, me, me.*

Interviewer: *What do you mean by that? Do you mean that they are selfish or that they can't look past their own experiences?*

Caroline: *They can't look past their own experiences. When I was ill he had to look past that and focus on me and take the focus away from himself.*

Interviewer: *And did that help him?*

Caroline: *I think so. Either that or the new diet he was on at the time.*

Interviewer: *Did the self-focus side of it bother you?*

Caroline: *It did a bit. It always centred on what he could do.*

Interviewer: *And you felt pushed aside or ignored?*

Caroline: *Not ignored but pushed aside a bit yeah. Like I used to drive him to the Dales for his shooting, and I used to drive all the way there and he said he'd drive back but then he'd only drive a little bit before making me drive the rest of the way back. He wouldn't do housework when I was at work all day, but he does do a bit now.*

So, it seems that even in relationships where overt anger may not be present, it can manifest in other ways, particularly when people do not communicate their own needs. If we consider Helen's description of her private anger which she did not express, alongside Caroline's description of feeling taken advantage of, then we see that it seems difficult for SOs to communicate their own needs. SO participants described behaving in ways that are self-sacrificing and that they do not necessarily speak up when they feel that the PwCFS/ME may be expecting too much of them, instead passively accepting behaviour that they actually find to be unacceptable. This gives rise to feelings of anger which manifests as resentment because it is not expressed, and this has a negative impact on the relationship with the PwCFS/ME as well as on their own well-being.

The description of anger was accompanied by descriptions of depression and, in some cases, suicidal ideation, for example, in the cases of Sean and Steve who we have already seen experienced and expressed anger in their relationships:

> *"I did suffer a lot from depression and social isolation, and err… suicidal thoughts which I don't really like to talk about but there you go."* (PwCFS/ME, Sean)

> *"…They just prescribed him Prozac but then I was going to take [Steve] back because he was just a mess, like nearly suicidal, on Prozac and then they just gave him another one, a different one. But that was it 'Oh alright depression' and that's how I think they just felt."* (SO, Helen)

Fear, frustration and anger: A discussion. The emotional response of frustration occurs when an individual faces opposition in their pursuit of a goal, with a possible outcome being aggression (Dollard, Miller, Doob, Mowrer, & Sears, 1939). The frustration-aggression hypothesis (Dollard et al., 1939) proposed that aggression was the outcome of frustration, although this was re-formulated to account for the fact that frustration does not always lead to aggression (Sears, 1941). However, the hypothesis does suggest that aggression is always the consequence of frustration (Zillmann, 1979). Later research suggested that attributions and negative affect also play a role in determining whether an individual's frustration is expressed as aggression (Berkowitz, 1989, Frustration-aggression hypothesis: examination and reformulation). We see evidence of frustration across a range of excerpts, both PwCFS/ME and SOs, although it seems more prevalent amongst PwCFS/ME, with this frustration giving rise to feelings of anger, not always expressed as aggression.

Anger can be understood to be an intense negative emotional experience for participants. It has been related to other negative emotional experiences including depression and anxiety due to their common characteristics (Carmony & DiGiuseppe, 2003). For example, people tend to have more attentional focus on negative emotions than on positive emotions, so negative emotions tend to lead to greater analysis of events and situations (Schwarz, 1990).

Smith, Haynes, Lazarus & Pope (1993) suggested that negative emotions are differentiated by attributions and appraisals of the impact of an event, and that appraisals mediate between attributions and emotional responses. The attributions alone do not produce emotional responses because they tend to be based on facts and are non-evaluative (Lazarus & Smith, 1988). However, attributions do contribute significantly to the development of negative emotions (Carmony & DiGuiseppe, 2003). The type of emotion experienced tends to depend on whether the attributions are external or internal in nature (Neumann, 2000). Individuals who make external attributions tend to experience anger, whereas those who make internal attributions experience guilt (Averill, 1983; Neumann, 2000). Referring back to the excerpts presented so far in this concept, we can see that in almost all of the examples of anger depicted, the participants are making external attributions, which supports the link between anger and tendency to attribute blame to others found in previous studies.

The emotions of anger emerge in the data also amongst SOs. However, this is typically manifested in resentment. TenHouten (2007) described resentment as present when an individual feels that another person has caused them harm, and consisting of a combination of disappointment, anger and fear. Solomon (1993) proposes that resentment exists on a continuum on which status is a factor in how an individual channels feelings of anger. When a person perceives another to have a higher status than themselves their anger is experienced and is traceable in their communications as feelings of resentment. When they perceive the other person to be of equal status, then the anger is expressed simply as anger. When the individual is perceived as being lower-status, the anger manifests as contempt. People are likely to feel a sense of resentment when they feel that they have been taken advantage of or that another person has used them. This could suggest that SOs place the needs of the PwCFS/ME above their own, which would explain why SOs presented here experienced their anger as resentment; the illness/carer dynamic disrupts the equal status dynamic and places the PwCFS/ME in the higher status position. This leaves the SO feeling like they cannot or dare not 'speak out', perhaps for fear of making their symptoms worse, thus the anger turns to unspoken resentment.

Research has demonstrated significant similarities between anger and depression in terms of the coping strategies people employ, as well as in what treatments are most successful. In particular, high levels of anger and depression were found among adolescents who had made multiple attempts to end their life (Stein, Apter, Ratzoni, Har-Even, & Avidan, 1998), whilst Boerger, Spirito and Donaldson (1998) found that a wish to die was predicted by high levels of anger and depression, reflecting the narratives provided by participants of Sean and Steve’s experiences presented earlier.

Also worthy of comparison to the data presented in this section are the findings of Whitehead (2006). Whitehead undertook a qualitative study applying narrative typologies developed by Arthur Frank (1997) to analyse interviews that had previously been conducted with 17 PwCFS/ME. Frank (1997) proposed that in any illness narrative, three narratives are told, but with one type being the most prominent at any one time: restitution, chaos and quest. A restitution narrative is characterised by a belief in the temporality of illness and a belief that one will get better. The plot of a chaos narrative is that things will never get better and no one has control over the situation, and a quest narrative is characterised by an attitude of acceptance and a belief that there is some good that can come out of it (Whitehead, 2006). Having discovered this work late on in the analytic process, it bears striking resemblance to my own findings and will be discussed in more detail in further concepts. It’s relevance here is in the chaos narrative, in which Whitehead (2006) observed expressions of anger amongst

PwCFS/ME. The chaos narrative in CFS/ME was also characterised by a number of other aspects that have already been presented here, including social isolation, detachment (similar to disengagement), depression and frustration as a consequence of loss:

> *"Chaos was depicted by expressions of anger, depression and isolation. People described social isolation, detachment, an empty present and desolate future contrasted with a past that had promised much until illness destroyed this. Frustrations included the loss of career or plans for this, the loss of income and social contacts."* (Whitehead, 2006, p. 2243)

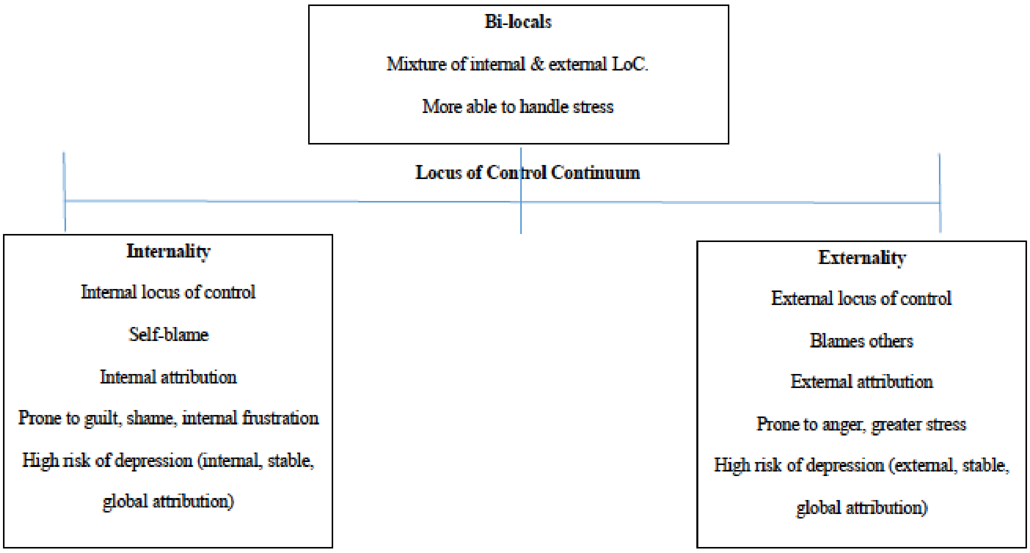
It seems clear that the experiences of participants in this research are similar to those presented in other qualitative research studies. However, in this study I am evidencing that these experiences do not just affect the PwCFS/ME. The SO is also deeply affected emotionally, and this can lead to relational patterns that cause the unhelpful responses presented in previous research. It is also important to consider that the participants volunteered to participate and many came from a CFS/ME group and who were willing to talk to me. The sample may not be representative of the most isolated individuals with CFS/ME and the examples of isolation within the sample may be just the tip of the iceberg.

Anger, attribution and locus of control. In the literature, anger has been related to attribution, and to the concept of blame as well as locus of control. Individuals who have an internal locus of control believe that events occur as a consequence of their own actions, and therefore they tend to either blame themselves when things go wrong or to praise themselves when things go well. They have a higher need for achievement than people who have an external locus of control, because they attribute success to their own efforts. Individuals who have an external locus of control believe control is located externally, and therefore attribute successes or failures to external others or events, feeling that they have less control over what happens to them (Benassi, Sweeney, & Dufour, 1988; Rotter, 1966). Rotter (1975) proposed a continuum of control on which internality would be one extreme and externality would be the other extreme. Those who have a more flexible approach, able to combine the two types of control, are Bi-local. People who are bi-local have been shown to be able to have increased coping capacities, are more able to handle illness, take responsibility for their own actions but are also willing to make use of external resources (Rotter, 1975).

Multiple studies have shown that people who believe they have no control over events have poorer health-related outcomes, including greater stress, experience negative emotions including anger, and find it difficult to perform certain cognitive tasks – for example, they struggle to problem-solve independently (Roth, 1980; Sullivan et al., 2012; Wortman & Brehm, 1975). A feeling of a lack of control has also been linked to depression (Benassi et al., 1988; Peterson, Maier, & Seligman, 1993) and learned helplessness (Peterson, Maier, & Seligman, 1993). Those who have a high level of external locus of control at the same time as low self-efficacy have been shown to have higher levels of illness-related distress (Roddenberry & Renk, 2010).

These theories about anger and control and their links to blaming others are particularly interesting in relation to my findings because I have presented evidence of the existence of anger and blame within the sample, and there is a body of research into the role of external attributions in the perpetuation of CFS/ME (e.g. Michielsen, Van Houdenhove, Leirs, Onghena, & Vandenbroeck, 2006; Powell et al., 1990). Previous research suggests that the tendency to make external attributions is associated with increased fatigue and disability (Chalder, Power, & Wessely, 1996) in this population. The evidence presented here provides insight into the emotional consequences of this tendency for both the PwCFS/ME and their SO. I have attempted to bring the various emotions and concepts discussed in this section together conceptually in Figure 2. *Locus of control continuum.*

Figure 2. *Locus of control continuum*



As with the continuum of family cohesion described (and discussed in more detail in Category C. Balance), we can see that at both extremes people experience negative consequences, whereas in the centre there exists a place of balance, with the flexibility to be able to move between internality and externality based on what best maintains participants' emotional health at a given time. It is also important to note that internal attribution is not synonymous with an internal locus of control. Individuals can make an internal attribution but still feel that they have no power to do anything about it (Berckman & Austin, 1993; White, 1991). This can lead to self-blame, whereby an individual's internal attributions tip over into being self-critical, thus causing emotional distress (Callebaut, Molyneux, & Alexander, 2017). So, whilst previous research in CFS/ME may have suggested that external attributions play a crucial role in perpetuation of symptoms, it may be the case that encouraging internal attributions could have a detrimental effect on the PwCFS/ME. It also seems likely that this would put their relationship with their SO under further strain, as it may increase the likelihood that the SO would blame the PwCFS/ME for their illness and its consequences, or that the PwCFS/ME would perceive the SO as blaming them. They may become yet more frustrated and resentful, whilst the PwCFS/ME may still continue to feel that they do not have control over their situation.

Final thoughts on the concepts of fear, frustration and anger. I have demonstrated how experiences of fear, frustration and loss give rise to feelings of anger, but we have also seen that the tendency to feel and express anger is related to the tendency to make external attributions – not necessarily external attributions for the cause of illness, but external attributions for other factors in the illness experience – for example, blaming others for the build-up of events that gave way to illness; blaming healthcare professionals for a failure to diagnose or treat appropriately and the subsequent distress that is experienced; and blaming family members for not offering what they perceived to be the right support at the right time - for offering too much support, or too little support. The experience and expression of anger can be a part of, or is similar to, depression, and is not limited to the PwCFS/ME; SOs also experience feelings of anger and frustration. The excerpts presented suggest that PwCFS/ME felt comfortable expressing their anger towards their SO. However, whilst these expressions of anger gave rise to feelings of anger in the SOs, they did not always feel that they could express their emotions equally. I have suggested that this may be because they feel that they must prioritise the needs of the PwCFS/ME ahead of their own. Therefore, they remain quiet about their feelings but instead allow resentment to build. We have also considered briefly the findings of Neumann (2000) that external attributions tended to lead to anger, whilst internal attributions lead to guilt. This brings us succinctly onto our final, but most complex, concept within the Problem Pattern: the process of guilt.

8.6. Guilt

Guilt emerged as a concept worthy of theoretical exploration because when it was expressed, it was often accompanied by strong emotional reactions amongst participants. Guilt is constructed and discussed here as an emotional experience but also as an inherently social process that underpins a range of other emotions and behaviours in the dependent-caregiver relationship. As well as proposing that guilt is an interpersonal process between the PwCFS/ME and the SO, I propose that it was constructed in the interview as a part of the interpersonal process between myself and the research participants, perhaps more so than with other concepts highlighted in this work.

The absence of feelings of guilt were mentioned by Helen when speaking about the fact that she had not pushed their GP harder to offer an appropriate diagnosis:

> *"I don't, I don't feel guilty I don't really feel guilty because I thought I did everything I could at that time…I just supported him…"* (SO, Helen)

Following this statement Helen broke down in tears which might suggest that although she does not want to see herself as blameworthy or feel guilty, perhaps she does. As a PwCFS/ME, Rachel also spoke of guilt, saying said that she did have feelings of guilt about the impact of her illness on her husband; she had a similarly emotional response in that she too broke down in tears:

> Interviewer: *And how do you feel when there is the need for him to do that, to step in?*

Rachel: *(voice changes, seems less calm and controlled) I feel guilty about it, I feel really guilty cos, erm, I like doing it, I like looking after the girls…(breaks down)…and I feel sorry for him that I'm not the wife I would like to be.* (PwCFS/ME, Rachel)

In this excerpt, guilt appears to be related to societal roles and expectations. Another example of expressions of guilt about the impact of illness on loved ones is extracted from PwCFS/ME Sean's first interview:

Participant: *My mum has lots of responsibility and we're all aware of it. I have my illness to deal with and the guilt of how my illness has affected me and everyone else.*

This extract made me wonder about the role of attribution in feelings of guilt. It seemed as though he was saying he felt guilty because he felt he ought to due to the impact of his illness on those around him, but that this was because of their expectations of him rather than his own. I did not gain a sense from Sean that he felt responsible for his illness, but I did see in the narratives of his SOs evidence that they may feel sometimes that he bears a degree of responsibility. For example, in an excerpt from one of his SOs, his sister Sarah suggested that he does not do enough to help himself:

> *"Every so often I do go in and tear a strip off him and say 'What are you doing? You can't live like this" and he'll come around for a few days…I'll say like 'I don't want to come back after my three years of uni and see you sat in the same room. You need to do something.' It is a reaction to him because I feel that*

> *you can't always see that he's fighting. Maybe he's just not as vocal as we are…"* (SO, Sarah)

Sarah's excerpt points towards a degree of causal attribution to Sean of which he has been made aware through the ways in which she has spoken to him. These attributions are, to a certain degree, backed up by Sean's Mother, Sally:

> *"…I could leave supplies on the bedside table and leave for work and come back and it would all still be there. So, despite it all being there, he was so brain-foggy and out of it that he didn't take his first lot of meds therefore he didn't improve during the day, and didn't take in any nutrition. So, it's a difficult pattern… how much of it is that he just can't be bothered and how much of it is that he's so exhausted he can't even take that first med?"* (SO, Sally)

Sally makes allowances for her son's lack of self-help by suggesting that a symptom of the illness (brain fog) is the barrier, but she then goes on to tentatively but indirectly suggest that he may bear some responsibility for his situation because of his inaction. However, as stated earlier, during our interviews and the subsequent analysis, I did not interpret Sean as self-blaming for his illness. This is why his expressions of guilt lead me to wondering if they were motivated by his perceived expectations of others, and of what he believed he should be feeling according to their standards.

The concepts of standards and expectations in interpersonal relationships were also raised in Amy's interview. For example, Amy said of her feelings about any future potential intimate relationship:

> *"…it also links in to almost having the expectation of yourself and all the expectations of the other person and also not wanting to be a burden…"*

Here, Amy is drawing a direct link between the expectations of others which she separates from her expectations of herself, and the potential to experience guilt as a consequence of the discrepancy between the two. Interestingly, guilt was absent from Amy's narrative. The expectations of others and the impact of illness upon one's ability to meet them was however evident in Amy's description of difficulties in the intimate relationship she was in during the onset of her illness:

> *"I was struggling to stay awake and I think it was a bit annoying for me because he'd want to stay out and I'd be a bit kind of like, maybe not even say it, but be a bit like 'I just want to go'. Yeah, yeah, I think it started to be a bit…a bit more difficult 'cause I was just tired and wanting to go but then I also didn't want to leave, and I didn't want to be putting pressure on him in certain ways as well…[he was] a little bit defensive I would say, a bit resistant and kind of almost rebelling against the fact, and wanting to stay out longer because he wouldn't really get why I was being quite so…I don't even know what the right word is…kind of wanting him there when he wanted to stay out."* (PwCFS/ME, Amy)

Amy does not mention any feelings of guilt; however, she is clearly describing discrepancies between her expectations and those of her partner, and their behavioural responses to these discrepancies.

This can be linked to issues of self-blame in CFS/ME. In describing events, Amy did not construct a negative or self-blaming narrative about her own behaviour; she simply drew attention to their differing needs and desires, and attributed her behaviour to her tiredness and emotional need to her illness. It felt throughout her interview that the language she used in constructing her narrative was self-accepting, non-critical, she did not self-blame or express feelings of guilt. In fact, throughout the transcript of her second interview, Amy frequently referred to the impact she has had on her SOs (we discussed several SOs that had been in her life since the onset of her illness) but at no point did she overtly state that she felt in any way guilty for the impact her needs and behaviour may have had on others.

I gained a sense that Amy felt she was justified in her behaviours and actions because she had accepted that she was ill, but that others had experienced frustrations with her that she was aware of, because they did not understand her illness in the same way as she did. Even though she did not state specifically that she felt guilty, she alluded to the fact that she recognised she had had a negative impact on her SOs by placing demands on them physically and emotionally. For example, in the following excerpt, Amy described how her illness had impacted her and her ex-partner whilst they were travelling shortly after graduating from university:

> *"Like, he had to drive everywhere because I just wasn't able to drive at that time, and over those seven months it just got worse and worse and worse, erm, physically I was not in good shape and emotionally, I mean it was quite difficult for him because obviously we were away from everything else and normally you kind of mingle with other people but because I couldn't, I wasn't*

> *able to, it kind of restricted him a little bit as well so the relationship just got…yeah…gradually and gradually worse…"*

I perceived Amy to have accepted her own situation, having adjusted her standards according to her perceived needs; she recognised that her behaviour may not always be aligned with social norms or with the expectations of another in a relationship, but she saw her situation as acceptable. She did not feel that she should take responsibility for her behaviour because it was due to her illness – over which she believed she had no control.

Indebtedness. Feelings of indebtedness were also evident in participant excerpts. For example, Rachel spoke of how she had learned to be thankful rather than grateful because being grateful made her feel she was weak:

> *"I had counselling last year as well, so I have learnt to be thankful in a more healthy way rather than grateful, because it's a different thing…being thankful I see as recognising the positive things in my life of which there are loads, and being grateful is for me…it has come in the past from a position of need, and has made me vulnerable to unhealthy relationships."* (PwCFS/ME, Rachel)

This clearly demonstrates the process of cognitive restructuring through which she has gone; I interpreted her perception of being grateful to mean that a grateful person feels indebted, and indebtedness makes her feel more vulnerable. Through reframing how she feels when she receives help and support, she is explaining that she no longer feels indebted and instead feels blessed and lucky in her life. This enables her to feel that her relationships maintain balance even when, on a practical level at least, they may be imbalanced.

In thinking about indebtedness and reflecting on participant narratives, it struck me that relationships that lack reciprocity come to be viewed in terms that are different to those of a healthy adult-adult relationship. Participants in this research often referred to the PwCFS/ME and SO relationship as being more like a parent-child relationship, for example:

> *"...If I was normal I would be... The relationship would be more equal at this point. It's still very mother and dependent child at this point, and I wish it wasn't."* (PwCFS/ME, Sean)

Within this excerpt concepts of social exchange, roles and expectations, and reciprocity are evident. Sean views his status in the dyadic relationship as being like that of a child, illustrating his experience of a sense of powerlessness and inequality which is heightened by his inability to reciprocate the care provided to him by his SO. His regret at being unable to fulfil their expected roles and at depending on his SO is also evident.

Guilt: A discussion. In the literature, guilt is described as a distressing emotion that arises when someone feels concerned that they may have harmed another person. As such, it is viewed as a pro-social emotion that protects social bonds (Nelissen & Zeelenberg, 2009). It has been suggested that in drawing our attention to the effect of our behaviour on others, guilt enables people to identify when a relationship could be at risk of breaking down so that they are able to take action to repair it (Vaish, 2018). Guilt is also a social phenomenon in that as much as being an internal experience, it occurs between people (Baumeister, Stillwell, & Heatherton, 1994) and can potentially be used to influence one's environment. Horney (1937) posited that guilt arises out of a fear of the disapproval of others and that neurotic interpersonal motives can underlie attempts to evoke guilt in others or exaggerated confessions of guilt. Studies suggest that guilt also serves the function of appeasing both victims and bystanders, thereby increasing the likelihood that others will empathise and cooperate with the transgressor (Vaish, 2018). This is an important consideration within a constructivist approach because it draws attention to the need to consider the influence of my presence on the expressed emotions of participants. The fact that participants knew their words were being recorded and would be analysed and heard by others may have influenced the ways in which they expressed themselves.

Guilt has been identified as a key feature of caregiving that is more likely to be experienced where caregivers feel ambivalent, and correlates with increased levels of depression in caregivers (Losada et al., 2018). There is a body of research into caregiver guilt (e.g. Losada, Márquez-González, Peñacoba, & Romero-Moreno, 2010) most of which focus on caregiver guilt in the context of cancer (e.g. Duggleby, Doell, Cooper, Thomas, & Ghosh, 2014; Spillers, Wellisch, Kim, Matthews, & Baker, 2008), dementia (e.g. Roach, Laidlaw, Gillanders, & Quinn, 2013; Springate & Tremont, 2014), ageing (e.g. Gonyea, Paris, & de Saxe Zerden, 2008) and parenting (e.g. Rakow et al., 2009). However, studies that draw particular attention to the guilty feelings experienced by a person with chronic illness in relation to their caregiver are more limited. A concept called 'Self-perceived burden' has been explored in chronic pain; Kowal, Wilson, McWilliams, Péloquin, & Duong (2012) found that 70% of chronic pain participants had clinically-elevated levels of self-perceived burden, and that symptoms of depression, pain self-efficacy and adult attachment were found to be predictors of this.

However, I have been unable to identify previous research identifying guilt or self-perceived burden as potential factors in the perpetuation of CFS/ME. In my research, guilt was embedded in the constructed narratives of participants' day-to-day experience and relational patterns, whether they were PwCFS/ME or SOs, suggesting that the role of self-perceived burden in CFS/ME may warrant further exploration.

The contribution of guilt to self-perceived burden has been highlighted in the case of terminal illness. In an interpretative phenomenological study of the experience of self-perceived burden among participants who were terminally ill with cancer (McPherson, Wilson, & Murray, 2007), participants were found to feel concern for the physical, social and emotional consequences that their illness had for others. Participant extracts bore striking resemblances to those in this study, for example:

> *"That (caregiving) causes her some anxiety too, when she hits things she doesn't know, who does she talk to? Where does she turn? In my case it's reliance on her and in her case that's an awful lot of responsibility".* (Participant extract from McPherson et al. 2007, p.420-421)

Participants in McPherson et al (2007)'s study experienced feelings of guilt and regret. However, they also found participants felt responsible for these consequences and blamed themselves, which was less evident in my sample:

> *"Many participants struggled with conflict around internal and external attributions of responsibility for having developed cancer, and for being limited functionally."* (McPherson et al., 2007, p.421)

As well as being related to experiences of anger and frustration in illness (Averill, 1983; Carmony & DiGiuseppe, 2003; Neumann, 2000; Smith et al., 1993; Tangney, Wagner, Fletcher, & Gramzow, 1992), guilt is associated with causal attribution (Neumann, 2000). Much research in clinical psychology has focused on causal attributions as related to other-blame, self-blame and shame (Callebaut et al., 2017). Whilst guilt and shame may be the emotions experienced by an individual, blame is a social process through which guilt is evoked. An individual blames him or herself for an event if they believe it is their fault and they therefore take responsibility for what occurred (Mantler, Schellenberg, & Page, 2003).When people attribute blame to another person for an occurrence, they do so because they believe that the person to whom they are assigning blame could have caused the event(s) or controlled the causal factors (Mantler et al., 2003; Shaver & Drown, 1986; Shaver, 1985). In the case of chronic illness, people blame themselves when they perceive that they had some control over the causal factors of their illness (Dirksen, 1995; Rich, Smith, & Christensen, 1999). Because PwCFS/ME tend not to feel that they have control over their illness, it seems less likely that they would blame themselves, as I have highlighted with some of the excerpts presented. Therefore, their feelings of guilt may not be due to feelings of self-blame. Instead they may be related to the beliefs they hold about the standards and expectations of others, and their beliefs about how others might think they should feel for the imposition on their lives. McPherson et al (2007) linked self-perceived burden to the experience of dependence and feeling unable to meet expectations based on their social roles. Baumeister et al., (1994) also highlighted how research participants described guilty feelings in relation to the standards of other people. They stated that when there were apparent discrepancies between standards and expectations of two people, guilt was likely to be a consequence for the person who failed to meet the standards and expectations of the other.

These arguments suggest that guilt can be separated from the concept of self-blame in illness. We have already seen that research suggests self-blame occurs when a person believes themselves to have been responsible for their illness (Dirksen, 1995), whereas the arguments presented above suggests guilt can occur when they do not blame themselves but feel that others may have evaluated them as not having met their expectations. If we recall Sean's expressions of guilt but apparent absence of self-blame, and his sister's and mother's attributions, then this is a plausible explanation for the emergence of genuine feelings of guilt for Sean, even in the absence of a belief in his own responsibility for the onset or perpetuation of his illness.

In reviewing the literature around the concept of guilt, I also realised the relevance of social exchange theory (Homans, 1961; Thibaut & Kelley, 1959). This is based on the concept that individuals act on the basis of an expectation that whatever they do will be rewarded with an equitable reaction from another (Blau, 1964). This could be, for example, emotional exchanges of love and encouragement, practical exchanges such as doing something for one another – person A takes person B to the airport in the expectation that if they ever needed a lift to the airport it would be fair to expect person B to be willing to do this - or economic exchanges such as in the case of working to earn money. This process is referred to by Emerson (1976) as 'implicit bargaining'. It follows then that when one person acts and the favour is not returned, it is common for the person who profited from the exchange to feel indebted to the other person (Greenberg, 1980). Indebtedness was of interest to me when considering the caregiving relationships in the sample because during periods of disability, participant narratives illustrated that their relationship with their SO is not balanced or equal – one person was always doing more for the other, and the other may not be able to reciprocate. Greenberg (1980) suggests that a person in receipt of care and support may begin to feel indebted particularly because they become concerned that the person providing the care is prioritising the recipient’s needs ahead of their own, and therefore there are likely to be costs to the caregiver; a feeling of indebtedness is likely to give rise to feelings of obligation to reciprocate, uneasiness and discomfort, and a sense that they must take steps to reduce the indebtedness. As Greenberg states, “The recipient can behaviourally reduce his feeling of indebtedness by *reciprocating* the benefit, and/or he can *cognitively restructure* the situation” (p.14). I presented evidence of this process of cognitive restructuring as a coping mechanism in the sample, illustrating how feeling thankful rather than grateful had helped one participant to feel more positive. This appears to support Greenberg’s theory, and also provides evidence for the process of reflection and reframing as a potential mechanism for alleviating damaging feelings of guilt and indebtedness in PwCFS/ME.

The concept of guilt: Final thoughts. I have presented an argument that feelings of guilt and indebtedness can leave a person feeling powerless. In CFS/ME, I have suggested that the PwCFS/ME does not necessarily feel guilty because they blame themselves for their illness, but because they feel others think of them as in some way responsible and therefore they are acutely aware that they are failing to meet the expectations of others. The PwCFS/ME must learn to live with the guilt they may feel about their inability to uphold what they perceive to be ‘normal levels’ of social exchange, if they are to maintain a balanced and harmonious relationship with their SO. Part of living with this guilt appears to involve the process of cognitively restructuring so that people reduce guilty feelings, otherwise they avoid situations that give rise to it. It is easy to see why guilt could be something that someone with CFS/ME would actively avoid – because of the discomfort caused by guilt, people make attempts to soothe themselves by reducing guilt through the reduction of indebtedness by reciprocity. But in the case of chronic illness, there is no knowing whether the ability to reciprocate will ever improve, and consequently an imbalance in a relationship ends up becoming something to which the dyad must adjust and adapt; the SO also has to come to terms with the emotional consequences of giving so much more than they receive, as is aptly illustrated in the following excerpt:

> *"I can never think 'oh right I'll go do this'; I need to think of him first and then go and then do it, do you know what I mean? [The situation is] challenging, very challenging yeah…it's just part of life now yes, yes it's just part of life."* (SO, Helen)

8.7. The Problem Pattern: Conclusions

I have presented examples of the negative emotional and relational experiences of PwCFS/ME and their SO. We have seen how fatigue and disability present in CFS/ME and how these lead to dependence on others. In exploring the experiences of both the PwCFS/ME and the SO, I have highlighted some dyads in which family cohesion appeared to have become a difficulty. In some, specifically the mother and son relationships, enmeshment appeared to be contributing to a vicious cycle of disability and social anxiety. In others at the other end of the scale, disengagement appeared to have the same consequences. For most PwCFS/ME, anger towards others (SO, health professionals, previous work places) appeared to feature, but the relationships in which the anger was directed at the SO were also the relationships in which enmeshment was a feature. They were also the relationships in which major stages of the family cycle had been disrupted, in particular the 'Leaving home' phase, so that the two male participants concerned had struggled to develop autonomy away from the family, possibly because they had not been able to engage in opportunities for individuation. I have hinted at the development of a new family homeostasis that keeps them locked in unhelpful patterns.

In the disengaged relationships, anger seemed less of a feature. However, avoidance of guilt emerged as a potential reason that some individuals choose not to engage with others at the present time, as did a fear of negative evaluation from others and of being able to live up to their perceptions of the expectations of others.

Loss and grief have been highlighted multiple times throughout this section and emerge as crucial aspects of experience, particularly for the PwCFS/ME. This will be explored in more detail in later concepts. Attribution and locus of control also emerged in the data analysis process as being crucial mechanisms in the emotional experiences of the PwCFS/ME and their SO. In particular, the attributions and perceptions of control that the PwCFS/ME holds about their illness influences their emotional response to their illness which in turn impacts upon their behaviours towards their SO. The SO then experiences an emotional response to these behaviours, and may then behave in the ways that have been found to be unhelpful.

9. Category C: Balance

I have so far presented evidence for the problem pattern in CFS/ME, in which I highlighted the emotional experiences that gave rise to and were a consequence of relational patterns in participant narratives. I suggested that the most extreme emotional responses and dysfunctional relational patterns occurred more frequently in relationships where there was disengagement or enmeshment, the extremities of family cohesion. I presented recent research that supports the view that dysfunctional patterns, sometimes called emotional over-involvement, criticism, negative responses, and solicitous responses, have a negative impact on clinical outcomes for the PwCFS/ME, and linked these terms to the concepts of enmeshment and disengagement. The problem pattern was the most salient concept to emerge during the analytical process. However, its emergence led me to pose a number of further questions in my analysis:

1) Is there a middle ground; a space where the SO can both support and encourage without inadvertently worsening the condition of the PwCFS/ME? If so, what does it look like?
2) Is there evidence in the data that such an approach has positive outcomes?
3) How can this evidence be meaningfully linked to existing theory?

Participant excerpts illustrate some of the ways PwCFS/ME and their SOs were able to find ways forwards. In this place we see that a balance between togetherness and autonomy is necessary. The characteristics of balanced relationships presented here are as follows:

- Love and Togetherness
- Individual autonomy within the relationship
- Holidays and days out

In constructing these concepts, discussions during interviews drew more upon participants' spider diagrams (for examples, see Appendix 6) than was the case in Category B. This may be because the content of these concepts was drawn more from second interviews than first interviews; in the first interviews discussion was less directed by me and there was a tendency among participants to focus upon negative experiences in CFS/ME. In the second interviews, we used the spider diagrams as a means of structuring the conversation. Thus, discussions remained participant-led but were in response to the specific aspects I had asked participants to consider when developing their spider diagrams. In Category B I evidenced a range of psychological and emotional experiences, drawing on the full range of participant narratives. In doing so, I provided contextual information and detail about the cases that gave rise to the construction of those concepts. These were more heavily weighted in the relationships in which greater levels of tension and dysfunction existed. It was not appropriate to provide a contextual overview of participant relationships prior to presenting the evidence because I wanted to tell the story of the participants' experiences as I presented the concepts within the category, and not limit that section to specific participants. However, in this category, narratives feature more prevalently from the participants whose narratives suggested constructive patterns of relating because they reported happier relationships, less conflict and referenced positive qualities in their lives and relationships more frequently than those who featured more prominently in Category B. In order for the extracts in this section to make as much sense as possible contextually, I have provided below a brief contextual overview of the participants that feature more prominently in Category C. Those participants included one triad, one dyad, and one individual participant:

- Rob, Kath and David (Son with CFS/ME, Mother, also with CFS/ME, and Father/Husband)

- Richard (PwCFS/ME) and Caroline (wife)
- Rachel (PwCFS/ME, a wife and mother)

Rob, Kath and David. Rob was one of the first participants recruited to the study. He had made what he believed to be a full recovery from his episode of CFS/ME. During the one-year period of illness that he experienced aged 18 following overtraining as an athlete, he was delayed in going to university, and instead remained living at home with his parents. Therefore, he described his SOs for the purposes of this research as his parents. Rob was not bedbound but was housebound in the early months of his illness. His Mother, Kath, had also had CFS/ME. Kath had not been bedbound or housebound for significant periods as she tried to keep going despite her illness. She considered herself to have made a full recovery at the time of the interview. Kath participated as Rob's SO but in the interview spoke much more in depth about her own experience of the illness. David, Rob's father and Kath's husband, participated as SO to both Rob and Kath, and his narrative tended to focus more upon his relationship with his wife but he also spoke of how he supported Rob. All three participants described positive, supportive and constructive relationships and there was little conflict evident between them in any of their narratives.

Richard and Caroline. Richard had developed CFS/ME whilst working in education and had to retire from work eventually. In the early months of his illness he was bedbound but then attempted to return to work. He had prolonged periods during which he was housebound but in recent years he has experienced considerable improvements which he attributed to nutrition. His wife, Caroline, lived with him and provided care for him. She was also caring for her father at the same time and she described feeling a loss of sense of self, and expressed frustration at her own needs not being met. There appeared in their narratives evidence of marital dissatisfaction for a period of time, but it also seemed they had found ways of coping with the illness and with their competing needs so that they were both now satisfied with the functioning of their relationship and lives, and both seemed positive and constructive in their discussion of how things are for them now.

Rachel. Rachel participated as a PwCFS/ME and described her relationships with her husband and her young children. Rachel had experienced a six-month period of acute illness during which she was bedbound, and following this there were gradual improvements. Several years on at the time of interview, she viewed herself as living within the limitations of CFS/ME and spent a lot of time at home but was not housebound as such. She experienced a deep sense of loss through her illness and had worked through this with the support of a counsellor. Whilst Rachel's narrative was emotive and communicated deep distress at her experiences, she viewed herself as someone who had learned how to live positively with her illness, citing her post-doctoral level of education as a crucial aspect of her being able to cope through reflection and reframing. She spoke of her and her husband's love for one another as being of great value to her in maintaining quality of life despite her illness, and in particular spoke of the importance they each placed on maintaining a sense of self within and beyond the family unit for the health of their relationship.

Presentation of the Results. In the previous section, discussion of each concept followed the presentation of evidence. However, in this section, I will first present evidence of the characteristics of cohesive relationships in the sample as a series of concepts with evidence from the data, followed by a discussion.

9.1. Love and togetherness

Love emerged as a crucial element of coping with the illness and maintaining a balanced relationship. That love was described as being maintained through togetherness and supportive behaviours. For example, when thinking about what helped in their relationship in the context of CFS/ME, David (SO) described the love and togetherness shared between he and his wife Kath (PwCFS/ME), and what this meant for him:

> *"I think it is the love and support you give; if you didn't love the person…I have known couples who are married, but they are married and the husband goes off to the pub every other day and we see couples our age who you see go out together very rarely, we always go together… I know it sounds a bit soppy but I put 'love' (indicating to spider diagram) and put that as a main thing really, if you love somebody and you're happy, it can make a lot of difference."* (SO, David)

What is also interesting here is that David mentioned love, doing things together and being happy in the same paragraph. For him, despite the challenges posed by Kath's health, he viewed them as happy in their relationship and this was seen by him as a positive force in helping Kath cope with her illness. Togetherness was also highlighted as of great importance to David in maintaining their relationship:

> *"It seemed strange because when I was working, we would be working in Scarborough, Whitby, places and I would go for a walk on my own and thought 'Well this isn't much fun on my own'. We have always been close, always done everything together, the worst thing would be if one of us go before the other, I don't know what we would do. But we have always… we have never gone off on our own, I mean [Kath] goes and sees her friend in Beverley once every three or four months and she comes back here. Everything else, even shopping…the weekly shop, I'm not keen on shopping but I always go with her anyway. We went to Bridlington yesterday and walked along the beach for*

> *maybe an hour. We may not say much to each other, but we still hold hands, we still hold hands, we get a walk in, she holds my arm and just be together really."* (SO, David)

This excerpt from David does not directly refer to Kath's illness or how the couple coped with the illness, but does much to illustrate the quality of their relationship, and what togetherness looks like for them. It hints at physical affection that is valued and appreciated by both members of the dyad. Kath's description of their relationship echoed David's. It is worth noting that although Kath and David undertook the first interview together, their second interviews, from which these excerpts were drawn, were conducted separately:

> *"…We do a lot of things together when we are together. We don't live independent lives like some married couples. I look around I think 'Well why are they married? They don't do anything together'. Whereas we are quite a couple and always have been. We spend a lot of time, when we can, doing things together before we're retired, not just interim retired. And I do think that makes a difference."* (PwCFS/ME, Kath)

This suggests that for Kath, doing things together is also crucial in the maintenance of their couple relationship, and that they both prefer doing things together rather than separately but also that they make a conscious effort to ensure they do spend time together. Kath hinted at togetherness as important in building resilience as a couple in the face of significant life challenges, particularly in relation to health. She explained that her and David's marriage was stronger because of their ability to cope with setbacks together, a quality that was established early on in their relationship:

> *"No, [my illness] didn't create any discord, and I could see with some people it would…I think the fact that it didn't create any discord was partly because I did have an illness at the beginning of our marriage, I had an overactive thyroid, which put a lot of pressure on us, and I ended up having it operated on when I was 28. I also had endometriosis and then when that was diagnosed I had an ovarian cyst in my 20s, before I had children, I had my left ovary removed. So, he had to cope with that then, when we were young, you know, really right at the beginning of our marriage. So, the fact that we pulled through all that, I think puts you on a stronger footing than somebody who's been fit and healthy all their life and then suddenly ME hits them, because we've been through that together already once, erm, and at the beginning…"*
> (PwCFS/ME, Kath)

In the phrase *'we've been through that together once'* Kath highlights that it was not just her who had to get through the illness, it was the couple. It is possible that this togetherness served them well in coping with Kath's illness because they felt equipped to deal with the challenges – the relationship was functioning in a way that they were both happy with prior to illness onset, which may have made dealing with illness a more co-operative process than if there had been challenges and dysfunction. This is particularly poignant when compared with the narrative of Eileen presented earlier in Category B. The Problem Pattern, *Disengagement,* who felt that there was a lack of support and togetherness in her marriage. It is interesting to note that Kath did make a full recovery from her illness whereas Eileen did not.

Rachel spoke of the love between herself and her husband as important in the context of her illness. I had previously separated 'practical support' and 'emotional support' by posing them as separate questions and separate aspects for participants to consider when completing the spider diagram, but Rachel was clear that for her, these two could not be separated. Rachel constructed her narrative of her husband's behaviours as both practically and emotionally supportive, demonstrating the meaning these behaviours have for her. She suggests that he does these things because he loves her and they are therefore clear expressions of love, which she takes care to recognise and be thankful for:

> *"The practical stuff works – [he] brings me a cup of tea every single morning, every single morning he makes me a cup of tea and it's a gift, and it's a gift of love, I recognise it as a gift of love, so the practical stuff is really important, but the practical stuff actually speaks of something much more profound – it does speak of love, because he does it with a generous heart – he doesn't bring it in and slam it down on the side in a really despicable way because he's resentful; he just brings me a cup of tea…I feel loved. I feel lucky. I count my lucky stars, it could easily not be like that, you know, couldn't it? So yeah, those two things that are really important, the emotional support and the recognition of that emotional support, so him bringing me a cup of tea or taking the wash out or whatever it is, the practical stuff, it's not just practical is it? That is a loving act. And my recognising it as a loving act is comforting, and I think that's really crucial, is seeing those two things as connected."* (PwCFS/ME, Rachel)

Here, Rachel is reflecting on the fact that she plays a role in the maintenance of their relationship as healthy by choosing to interpret his behaviours as loving. She recognises that not all relationships function in this way. In some relationships, carers may undertake a caring role out of a sense of duty, but Rachel interprets her husband's care as an expression of his love for her, and this helps her to feel positive about the things he has to do for her and about their relationship. This is not to say that she does not feel guilty about the extra load for him, she did express feelings of guilt as presented in Category B. The Problem Pattern. But, she also chooses to see her husband's love as supportive, kind and understanding, and described his responsiveness to her illness as being crucial to her ability to cope:

> *"So, after the diagnosis in 2010, since then, [he] has just carried on just being, you know, marvellous – loving, and supportive, and understanding, and if I have a relapse and a period of debility, he just steps in and picks up what I would normally do in a way that is just incredible really, and I wouldn't be able to cope without him. You know, he picks up all the stuff with the [children], he picks up making the dinner, doing the laundry, he just picks everything up, and he also does emotional labour as well – he's very mindful of my wellbeing and he's very kind. He can be a bit grumpy sometimes, it does get to him sometimes, but he is generally speaking kind and loving, and understanding."* (PwCFS/ME, Rachel)

Rachel also described togetherness as important. She described how her illness and their family life interacted to pose challenges for them in finding the space and time to maintain their couple relationship, which she considered 'grown-up time', but explained that she recognised the importance of their time together as a couple:

> *"We don't have much grown-up time together. And we don't have much time when it's just me and him chatting, because the [children] go to bed, they're*

> *[age] so they go to bed at half 8/9 o'clock and I go to bed at the same time…when they were little, when they went to bed at 7 o'clock, we would have a couple of hours. We're not big TV watchers so we'd sit in here in the kitchen and have a cup of tea and a chat and it was really nice because he's funny and he's clever and I like his company, and I think he likes mine, so we don't get much of that…he doesn't get much of that companionship out of me."* (PwCFS/ME, Rachel)

This highlights again the importance of togetherness, which she terms 'companionship', in maintaining the relationship, and the fact that her illness and their roles as parents do affect their ability to maintain this companionship. This excerpt can also be linked to excerpts presented earlier in which Rachel described feeling guilty about the fact that as a wife she feels she cannot give her husband what he deserves due to her illness, which she spoke of following the above description. Rachel also highlighted however that the scarcity of time spent together made it all the more important and meaningful when they did have time together:

> *"Luckily, when we do have opportunities to have grown-up time, it is really nice, it's really treasured. So both the [children] were away over the weekend on a choir tour with their school…so we went to the movies, and we went out for dinner, and we had little naps in the afternoon together, it was really lovely, it was only for three days, but it was really lovely, it was dead romantic; we didn't go away anywhere, we stayed home, but it was really nice, and I think knowing that when we get the time we can still be companionable and still like each other, that kind of sustains us."* (PwCFS/ME, Rachel)

Rachel's use of the word 'like' in both of the above excerpts is of interest – she suggests here that their love is built on a genuine and long-term enjoyment of one another's company, an ability to treat each other with love and kindness when possible, and to seize opportunities to be together physically. This also suggests that she believes that in some marriages perhaps couples can become dependent or reliant on one another, but not necessarily actually like each other or enjoy one another's company all that much. Rachel also highlighted how important family time was to them but clearly recognises that time alone together is of great importance to their relationship and to her husband specifically:

> *"We really like family life, so, it's our wedding anniversary coming up and we'll go out with the girls, we won't go out on our own, all four of us go out, and when it's a birthday, all four of us go out; we like our four for company, but the grown-up time, you know, time to have a cuddle and a kiss, and relate to each other romantically, that does feel like, it's nice, it is really nice and it is really crucial…and it's really important to [him], it's really important."*
> (PwCFS/ME, Rachel)

This infers that physical affection is something that they both value greatly within their relationship, and Rachel's repeated use of the word 'romantic' suggests that the physical affection is meaningful to her as a form of 'romance'. Rachel also recognised her husband's efforts to ensure they do things together recreationally; even though her illness can sometimes derail their plans, she described how he makes the most of recreational time and opportunity:

> *"[He] is really good at arranging grown up things for us to do so we go to concerts occasionally, maybe once a month, sometimes we buy tickets for concerts and then I'm not well enough to go so he goes with one of the neighbours…so he's really good at making those plans for us to do grown-up things together. I will sometimes say 'Sorry that we're not getting much grown-up time with each other'."* (PwCFS/ME, Rachel)

Rachel went on to describe how different she believes things would feel for her and her husband if they did not make the most of time together and as a family:

> *"Oh God, I'd just be like a third child in the house, sometimes I am that dependent."* (PwCFS/ME, Rachel)

It should be noted that in terms of the family life cycle, Rachel and her husband were at the stage of raising young children, whereas Kath and David had two adult children and were nearing retirement. However, in both cases it seems clear that they recognise the importance of togetherness in maintaining their love.

It is also interesting to note that although in the case of Sean and his mother Sally, enmeshment has been highlighted as a feature, the relationship between Sally and her daughter Sarah, Sean's sister, appeared notably different. They too spoke of the importance of quality time together on their own as mother and daughter:

Interviewer:	*And do you spend time together?*
Sally:	*We do, yeah.*
Interviewer:	*And what sorts of things do you do together?*
Sarah:	**Laughs* Collapse in front of the telly because we're shattered!*

Sally: *With a few galaxy ripples! Or if [Sarah] is back from uni and she doesn't need the car for the week then I'll take her back and we'll go get coffee or cake together and we go to [place] and have a meal there.*

Interviewer: *So, you have a relationship independent of [Sean]?*

Sally: *Yeah, we don't want to exclude him, when we can include him like that meal we had he did stay well.*

Sarah: *Yeah that meal in [place]. I always invite him and if he feels well enough to come then he can come. But he usually says 'You two go have some time on your own.'*

This suggests that Sean also recognises that it is important for his Mum and his sister and their relationship with each other that they have some time together, just the two of them. However, this was not a feature that emerged as present in his relationship with Sally. Sean and Sally spent a lot of time together because they lived together, but taking 'quality' time together to just enjoy recreation and one another's company was not something that either of them spoke of.

I have highlighted in this concept the importance of love and togetherness in the maintenance of the balanced relationship functioning in the context of CFS/ME. This has drawn predominantly on data from participant narratives where there was little or no evidence of relational dysfunction. Kath and David's relationship struck me as 'healthy' in that is was functioning, happy and supportive and yet David described how they '*do everything together'*. Similarly, I interpreted Rachel's depiction of her relationship as an illustration of 'healthy' cohesion and balance, despite the fact that at times her husband had to assist her with most tasks. It is therefore necessary to explore why I viewed this togetherness as an aspect of balanced cohesion as opposed to the enmeshment described earlier.

9.2. Individual autonomy within the relationship

As well as a commitment to togetherness, what also differentiates the cohesive relationships from the enmeshed ones in this research was the degree of autonomy and individuation or 'separateness' within the relationship, and the acknowledgement of the importance of each individual in the relationship maintaining their sense of self in order to also maintain their togetherness and connectedness. I therefore present here evidence for autonomy and individuation within the cohesive relationships.

Despite the degree of togetherness that both Kath and David spoke of, Kath described herself as an independent person who struggled to adjust to being dependent upon her husband during her periods of illness:

> *"The first thing was changes in the relationship before and after the ME. The only things I've got down were, I felt more dependent on [David]…I suppose I am quite independent and do like to do things for myself. I don't like having things done for me. That goes against the grain with me, definitely."* (PwCFS/ME, Kath)

Kath spoke of her attempts to maintain independence despite the illness and forced dependence, but also recognised that she had needed to strike balance between doing things for herself in order to maintain her independence without overdoing it and reducing her independence in the long-term:

> *"I sorted my life out basically so that I could cope on my own because [David], as I say, was at work most – well he was at work all the time. So, to a*

> *certain extent there are things that you make yourself do, you know, whether you can do them or not virtually. But, at the same time you had to sort of think, 'Well no I mustn't over-do it otherwise I'll end up worse and I will be totally dependent'."* (PwCFS/ME, Kath)

It was apparent that maintaining independence within the relationship was important for David too. He described his hobby of running as being important in maintaining his wellbeing. He told me that it was his strategy for coping with challenges such as his wife's and son's illnesses:

> *"I do running, I've being running at work in my lunch break to cut my time at the weekend, so I was doing the exercise I enjoy anyway and that was a stress-buster. I have found running is my thinking time, and my planning time, whilst I go running…I think about different things, things from the past, things I want to do in the future maybe. I do a lot of thinking whilst I'm running. I go on my own, I never liked running with other people, I like to just go off on my own running."* (SO, David)

What was particularly interesting about this excerpt is the fact that he really valued this alone time, his time to think, and he appeared to have a very loving, healthy relationship with his wife in which they very much valued togetherness. Therefore, it is perhaps fair to conclude that this time alone was a necessary part of the maintenance of their relationship, without which the functioning of the relationship may have been altered. Kath and David's son Rob also had a similar relationship with them:

> *"I got on with them OK really never really had any, you know, any issues really, I mean obviously it's like living with anybody, there's odd times when*

> *you're on top of each other and there is the odd argument but you know overall yeah, no, I had a good relationship with them really, so, yeah…"* (PwCFS/ME, Rob)

The mention of 'odd arguments' suggests that conflict was acknowledged as a normal part of relationships and the fact that Rob mentioned this in the same sentence as being 'on top of each other' also suggests that he recognised that too much time together and a lack of separateness can be a source of conflict.

It struck me that in order for dyads to experience emotional health in their relationships, what was really important for both members of the dyad was for the SO to go on living their lives, maintaining their sense of self, and maintaining space for themselves. This appeared to have positive benefits for both members of the dyad. This was evident in the case of Rachel who described her appreciation of her husband's need to maintain his sense of self, and to spend time with friends, and a recognition that time at home could have the potential to feel suffocating:

> *"I wonder actually thinking about it now, I wonder if [he] does find it suffocating. Although he does still travel, he still goes away every now and again, still goes to gigs with his friends, so he maintains his own life in a way that's healthy…"* (PwCFS/ME, Rachel)

I asked Rachel why she felt that this was healthy for him:

> *"I think he has a sense of himself as a man beyond carer-husband-father. Although he loves being a Daddy, and he loves being a husband - it's really important to him…[But] it is healthy for him, and I can see the difference in him*

> *if he doesn't do that, he does get grumpy, and part of him doing that and it being healthy is because that's how he relates to his male friends. That's their medium of communication and their medium to connect, is going to gigs, and a lot of his friends are spread all around the country and so he'll go down to London sometimes, Bristol, Birmingham, so he travels all over and he sees his mates and they go to a gig together, sometimes he'll come home the same night, sometimes the next day, but I think it's really good for him."* (PwCFS/ME, Rachel)

The fact that Rachel viewed this as good for him, rather than communicating any sense of abandonment suggests that she too has a degree of self-sufficiency and autonomy within the relationship. From Rachel's depiction of their life and marriage, there appeared several roles that her husband strives to fill. Rachel seemed to be communicating her husband's need for his sense of self to be formed of an integration of his various parts, or roles. She believes he values his roles as father and husband and that he also comfortably fills the role of carer when necessary but she also sees it as important that he maintains his individuality. This would appear to be formed of remnants of his life prior to their relationship and marriage. Despite being an employee, a father, a husband and a carer, what Rachel was communicating was that it was important to both of them and to the health of their relationship that he retains space in his life to continue being the person he was prior to the marriage even if it is only for one night a month. Rachel also strove to maintain her sense of self despite her illness, but she felt she had to be careful about the energy she put into her own friendships:

> *"I really protect myself, so I'm careful with friends, and I'm careful with what I expose, careful with what I tell people about myself because I don't want to be taken advantage of...I'm so vulnerable that I have to be careful...I don't*

> *really have much emotional reserve for…er…friendships actually, when I think about it…"* (PwCFS/ME, Rachel)

However, Rachel also explained that she very much valued her relationships with other women:

> *"I have a really good friend, and she is a psychotherapist, and she also does like a project and I think I was her project for a few years, and she's a really good, close friend…she sends me really loving emails which are really important to me and there are two of the other Mums who I used to know from the toddler group from when the children were toddlers…we don't see each other very often, maybe go to the pub once a month and have Ginger Beer…going out with women friends, yeah it's really nice. They're both really smart…we have really intense, deep conversations that are really curious and interrogative, which is really good for all of us."* (PwCFS/ME, Rachel)

Rachel demonstrates an awareness that she cannot expect her every need to be met entirely by her familial relationships. This is evident in the way she speaks about her ability to maintain bonds with female friends with whom she has things in common, and how she views these relationships as important to her own wellbeing, and to the wellbeing of her friends. An important element of these relationships for Rachel is their intellectual and emotional quality. In addition to the above excerpt, she also described attending a group related to her interests. What emerges as important to her in these relationships is the fact that everyone there has challenges in their lives:

> *"I haven't mentioned it, but it's really crucial. I go to a [type of] group and we meet once a month, ish, sometimes more, sometimes less…we're all kind of highly educated, interesting women. We've been meeting for 10 years, maybe more, 11 years, and we've all got something going on…it's a really interesting group in that…this group is really important, that we're all…you know, we just recognise the complexity of being…I don't think it's an unusual group apart from the level of education we all have, but everybody's got something going on…"* (PwCFS/ME, Rachel)

Rachel's time with friends and her maintenance of her sense of self beyond her illness also seemed dependent upon her interpretation of how they viewed her. She explained that she is not always open with her friends about her illness. Instead, she keeps this to herself so that they get to know her aside from her illness:

> *"I want to be [Rachel] first, and then they know [Rachel] and the identity of me is very secure and they see me as a person… I can't stand pity. I can't stand it. It's undoing. It's not about them and their intelligence or their empathy; they are compassionate and smart souls, they're amazing women, it's about my wanting to be seen as [Rachel], not the illness...I want my identity to not be tied to my illness…[the illness] shapes my every day, it shapes my every hour of every day…"* (PwCFS/ME, Rachel)

Although Rachel wanted her friends to know her for who she is beyond the illness and she actively behaved in ways that aimed to keep the extent of her day-to-day experience from them, she acknowledged that she was unable to maintain that persona with her husband and was very aware of the impact her illness may have on his perception and experience of her:

Interviewer: *So, in their [friends] eyes, you want your identity to be [Rachel]?*

Rachel: *Yeah, [Rachel] who's interested in philosophy and current affairs and who's a Mummy.*

Interviewer: *But in your eyes you do have the illness as a massive part of your sense of self?*

Rachel: *Yes.*

Interviewer: *In your own perception? What about your husband's perception of you?*

Rachel: *It must be massive for him because I'm so absent from his life, even though we live in the same house.*

However, Rachel very succinctly managed to explain that although her illness forms a large part of her life and sense of self, it does not define her, and does not define her relationships with others. Instead, she described it as being 'an overlay':

> *"I would see more of an overlay. If I was to draw a diagram of myself, so there would be my family, there would be [my husband], the children, there would be my studies, there would be my work, there'd be my interests …and then the illness is like an overlay that goes over the top, but it moves, so sometimes that overlay is casting a…putting a complexion on those activities and relationships and other times, it doesn't…it lifts…so it casts a certain complexion over things. You see I spend quite a lot of time lolling about in bed reflecting on all of this in my endeavour to have a good life within the physical*

> *constraints of this ill health, I don't know whether other people will understand it in that way, so rather than being read as an ill person, I want to be read and understood as [Rachel] first."* (PwCFS/ME, Rachel)

It seems fair to conclude that Rachel's desire for people to get to know her without disclosing her illness to them is partly because she has a strong sense of self aside from her illness and she wants others to see who she is 'as a person' as she says. Whilst she is clear that in her relationship with her husband it is not possible for him to see her for who she is irrespective of her illness, it also emerges that her ability to maintain her sense of self is based on others seeing her as Rachel, and that this is good for her, and good for her relationships.

The development of autonomy and individuation also seemed to be a protective or restorative factor in relationships that were put under strain. Caroline shared in her interview that there were problems in her marriage, particularly once her husband became ill. She had raised their children whilst her husband worked long hours, she had cared for other elderly family members and then cared for her husband for a number of years. But this had required her to sacrifice the things she enjoyed doing:

> *"[Caroline]'s always been a very big rock for the family, when my mother was very ill, you know she stopped working and looked after her and…she's not someone who expresses her emotions on the surface. She's a very pragmatic sort of practical kind of person…instead of doing the things that she would normally have been doing, she was there for me. You know, not sort of spending every hour with me, but you know, just generally going about household tasks and up and down the stairs, and with meals and things like that."* (PwCFS/ME, Richard)

Richard acknowledged that it might have been difficult for Caroline at certain points, and his mention of her pragmatism and lack of emotional expression suggests that he realises that she may have experienced difficult feelings relating to their situation, but that she just got on with what had to be done. Richard stated that they had quite different interests prior to his illness:

> *"Even before the ME we did a fair bit different, you know, we weren't a couple that were sitting in each-others pockets all the time …"* (PwCFS/ME, Richard)

However, he did feel that the illness had caused them to become more separate from one another because the things they did used to do together were no longer possible due to his inability to participate. However, he had not stopped her doing these activities:

> *"It has changed, we kind of virtually diverged a little bit in terms of things at one time that we'd have done together, to doing separately…we both loved walking, erm, which Caroline has been able to continue with, no point in her stopping doing it really. I discovered watercolour painting. And, got quite a lot of satisfaction out of that."* (PwCFS/ME, Richard)

Richard felt it had been helpful for him that Caroline had established her own interests. He had written on his spider diagram 'Needing a life of your own', which I asked him to elaborate on:

> *“Yep, that’s Caroline you see, and the big thing was she joined the ramblers…We never belonged to any walking groups or anything we just walked together, she joined the ramblers so she was you know safe while she was out there, basically…That helped me yeah…because I knew she was doing something she was enjoying.”* (PwCFS/ME, Richard)

Caroline was appreciative of the fact that she had been able to establish something for herself with her walking and other activities and that Richard had ‘allowed’ this. It had enabled her to make the most of her life despite her husband’s illness and the problems in their marriage, thus giving her a sense of independence, and possibly freedom:

> Interviewer: *Did the walking help you cope?*
> Caroline: *Yeah, definitely.*
> Interviewer: *Has it made the process of staying in the relationship easier?*
> Caroline: *Yeah, I think so, because I’m allowed to go when I want to go.*
> Interviewer: *Do you think it affected your identity?*
> Caroline: *I’m more confident now.*

In the context of a marriage where she had had to hold so much together, Caroline seemed to have rediscovered her sense of self and independence when she joined a walking group and went away with them independent of Richard. It seemed that this had enabled them to continue living together as husband and wife despite the fact that the romantic side of their relationship was no longer functioning. Therefore, although their lives had become more separate, this actually served to facilitate cohesion and enabled the marriage to continue working for them both because they were both finding happiness in their own lives:

> "*…Now we can have our separate lives, he can do what he wants and I can do what I want… he's quite happy for me to just go out and do things. He just says 'go' which I think has helped. I'm physically fitter. And have a good social life. [Richard] never had a social life because he was always working! But I'm never in now! *laughs*"* (SO, Caroline)

Similarly, Richard had taken up a new hobby of drawing and painting, and he spoke at length of how much he enjoyed this. He had discovered a new skill and passion and became very animated when talking about this. It seemed to have given him something for himself too, and improved his quality of life very much as he gradually overcame the worst of his illness:

> *"One of my children bought me one of the three 'Teach yourself to' books…so I thought I might as well give it a go, so I went and invested £50 on paper, brushes and paints, and made a start. I could draw, and I was very proud of my drawings and as soon as I'd paint them I'd ruin them you see, but suddenly using good quality water colour paper and all the right stuff, rather than the rubbish you use at school, I thought well this is fantastic and I was hooked, and I've sold about 60-odd originals."* (PwCFS/ME, Richard)

Richard felt that his painting enabled him to re-energise and explore his creativity after experiencing so much frustration from being unable to do the things he could previously do. It was clear that his work had been very important to his sense of self, and his illness had forced him to give this up, and he was saddened by his inability to contribute to the maintenance of the home:

> *"I was so frustrated with all of the things that I used to do that I could no longer do, both in a working context and recreation I suppose. And to some extent, sort of, you know, household tasks and maintain – I used to, I love gardening you see…"* (PwCFS/ME, Richard)

I asked him if his painting had changed how saw himself and he explained *"Yeah, yeah, I was succeeding in something".* Richard had discovered a passion for painting that he was able to do despite his illness and this had helped him to rebuild his self-esteem and confidence. Meanwhile, Caroline had established what I interpreted to be a renewed sense of self through taking a break from her caring roles to join groups and do the things that she enjoyed doing recreationally. They both rediscovered a sense of self independent of one another. This new process of individuation within the relationship seemed to have renewed their enjoyment of life, but had also brought them back together eventually in some ways:

> *"She was doing a lot of craft work, she had an industrial sewing machine and used to do all the craft fairs…and I used to go around with her, and then eventually of course when I started trying to sell some of my stuff, like the greeting cards, we had stalls along-side each other. It's funny, I hadn't even thought about some of these things when I'd put them down on there, because that's one way that this relationship has sort of stayed together…"* (PwCFS/ME, Richard)

Although it seemed that at one stage, they may have been disengaging, Caroline's ability to develop autonomy had enabled them to re-engage to a certain degree. Her determination to build herself a life outside of the marriage whilst she still remained within it had led to positive outcomes for both members of the dyad and Richard had then found something for himself too. This had enabled their relationship to be maintained despite significant challenges. Caroline had developed new social networks in association with this, and her husband was able to join her occasionally. He was able to lean on her newfound sense of self and her new social circles to build his own social world:

> *"I tried, I mean, I would go out with her on days when she was walking and I would spend some of the social time with members of her walking groups erm…I got into the habit of going out with her and taking my sketch pad with me and plonking yourself down on a stone somewhere and doing a sketch and then converting it into a painting. They would be doing a circular loop while I was doing my thing… I just love being outside."* (PwCFS/ME, Richard)

> *"…He can join us whenever he wants to, like we're having a quiz night tomorrow and he'll be joining us for that. And we have a social every first Monday of the month at a pub in [place] and he'll come to that."* (SO, Caroline)

We have seen how pastimes such as running, walking, painting and drawing as well as social groups and friendships have been a positive force in the relationships of some participants. This was also the case for Sean's Mother, Sally, whose relationship had become enmeshed. I have evidenced the heavy burden felt by Sean of his mother's dedication to helping him through his illness. However, even in their relationship, there was evidence that they might be beginning to explore autonomy a little more. Sally explained that she did retain something for herself now and then, and that this was what helped her to cope:

> *"I write things out, I cook and cake decorate, I make things for other people…That and dog walking. Also, if I'm doing a cake for someone, that focuses my mind and I come up with a solution a lot of the time because I stop thinking about the problem for a while."* (SO, Sally)

However, in Sally's narrative this hint at individuation was overshadowed by her immersion in her attempts to help Sean through his illness:

> *"After the divorce she kind of [long pause] I don't know… she just decided that her life was just going to be caring for me and my sister and she didn't have much of a social life then and that was what she chose at the time. When we got older and realised she'd been doing that, isolating herself in ways, we encouraged her to stop that. Ever since I've become ill she has become as isolated as me really, and she has a job she hates at the moment and she's not treated very well but that's her world at the moment and it's horrible."* (PwCFS/ME, Sean)

Although a desire to experience autonomy was also present in Sean's narrative, evidence that he was moving towards it was limited. There was little in the way of declarations of who he sees himself to be beyond his illness, and little evidence of a social life. So, we see that although there are some elements of the kinds of hobbies and interests that others spoke of in Sally's narrative, it does not seem that this has permeated her life enough to protect both her and Sean from enmeshment.

In conclusion, it emerged in the analysis that in the relationships where both members of the dyad kept something for themselves and did not disappear completely into a life defined by illness, this went some way to protecting their emotional well-being through enabling individuation within the relationship, resulting in positive outcomes such as healthily cohesive relationships. It may or may not be a coincidence that in those 'healthier' relationships presented, the PwCFS/ME in each (Rachel, Kath, Rob, and Richard) all described either having recovered from their illness or being on a road to recovery and having reached a state of acceptance and peace with their situation and limitations. This will be explored in more detail in Category D. Acceptance.

9.3. Holidays and days out

In addition to togetherness and autonomy, holidays featured in the data as an issue of importance for all participants, not just those whose relationships seemed healthier; in all of the interviews conducted, the subject of holidays came up. Some spoke of the impact of illness on the dyad's ability to go on holiday, or the ability of a holiday to alter the perception not just of the PwCFS/ME but the dyad and sometimes the wider family. Holidays emerged as a key time when dyads were able to escape the difficulties of their day-to-day existence and enjoy time with one another more fully, and days out featured as opportunities for escapism – this was not a finding that was prompted by questions and instead emerged organically within the data as participants explored and described their experiences. When speaking about the impact of her illness on their relationship, Kath described holidays, days out and enjoyable time together as something they made more of a conscious effort with because of her experience of illness:

> *"It does make you stronger I think, but, I don't know, it depends what your relationship is like to begin with. I think if it's on a bit of a…not on a very strong basis to start with, it could then…it could have the opposite effect. Erm, it made us more proactive, I think I've put down on impact on long term, we tend to sort of, erm, we're busy doing more enjoyable things together, whereas before we sort of focused more on 'well this needs doing, the house…' oh blow that, let's get out and have a day out, you know. You sort of think more like that. You don't, erm, sort of, get rid of it, and sort of think well this time we've been away five times this year by the time we get to the end of this year we have our holidays and we're doing things, getting away and doing things while we can."* (PwCFS/ME, Kath)

But it was not only in the more balanced relationships where holidays were cited as being important. Sally and Sean both spoke of the change in their relationship when they were on holiday:

> *"We've been through an awful time, and at times it seems like we're all pulling in different directions but at the end of it we still all get in the car and go on holiday together. We have a good bond and that's got us through."* (SO, Sally)

> *"We have flashes of the older relationship, mostly when on holiday and out of this environment, which is good."* (PwCFS/ME, Sean)

> *"The best support we've had was me re-registering as a carer and they said there's a carers 'cottage at [place] and for £35 they let me take [Sean] to*

> *[place] Monday to Friday to a lovely cottage and it was a change of scene and it was Easter and we could sit on a bench by the cliffs looking at the sea and then go back in and have a nice hot meal and that was the best care and support we could have."* (SO, Sally)

Holidays appeared refreshing for Sally and Sean, and a time when they could do things they might feel less able to do at home:

Sally:	*…we got a cottage in [place] and it's two hours but we know where to stop. We go up there and went for [Sean's] birthday in January.*
Sarah:	*He always wants to go there as well, he loves it there.*
Interviewer:	*What does being there mean to him?*
Sally:	*It feels like home to him he says. We left there when he was 18 months but he says it feels like home. We have some good friends up there.*
Interviewer:	*Is the outdoors something he gets solace from?*
Sarah:	*Yeah. He likes to walk the dog, there's a track down the side of the cottage and he loves walking up and down there in the fresh air and nice scenery.*
Sally:	*It has nice views, and we had a sprinkling of snow so it was very pretty last time.*
Sarah:	*And you can drive to the coast from there as well.*

Sarah's description of Sean enjoying time walking the dog hints at a sense of autonomy and independence for him in doing so.

The coast, the sea, and water generally, also featured highly in many of participants' descriptions of recreation and holidays as breaks from routine. Time spent near bodies of water seemed to symbolise escapism and an opportunity for quiet contemplation and peace that they perhaps felt unable to achieve in their day-to-day existence. It also seemed that returning to the same place repeatedly was something in which participants found comfort. Rachel spoke of their annual family holiday near a beach on the North Coast, describing how their knowledge of the area and knowing what to expect minimised stress and maximised relaxation:

> *"As a family we do, low-key holiday cottages, in the UK…in the summer we always, always go for at least a week, sometimes 10 days to [place]…a holiday needs to be totally relaxing and refreshing so we go to where we know every year, I know people find it weird, so we stay in the same village, sometimes a different cottage, and we take the [children], and it's just a walk away from the beach…and there's a tea shop on the corner and there's a restaurant, you know, everything we need is within walking distance…and that works really well because I can take the [children] out on my own and [he] gets some time on his own and we can all do things together, and if I feel unwell, I can rest without missing out on the party. That works really well."* (PwCFS/ME, Rachel)

What is particularly interesting about this excerpt is that it pulls together the importance of holidays, escapism, togetherness and autonomy all at the same time. It is clear that for Rachel, all of these aspects combined constitute health in their relationships, and she views holidays as an excellent opportunity to maintain the health of the family relationships. She views holidays and changes of scenery as stimulating and restorative, but for Rachel, the change of scenery between home and the place to which she goes on holiday is what's restorative. She recognises that holidays can have the potential to feel stressful, and so they safeguard against this by going to the same place regularly:

> *"I just feel, you know, Grayson Perry's wife, she's a psychotherapist and she's written a book called 'How to Stay Sane' and it was really affirming because a lot of the things she suggests I actually manage to do. And she talks in this book about when you go away for a weekend, the change of scene is stimulating, and that stimulation is good for your wellbeing…So when we go away, it's stimulating which can be tiring but it's restorative, so it's that old adage isn't it, a change is as good as a rest. And we just really like it, and although we go to a familiar place, it's still a change…So yeah, all the stuff that can be stressful, you know, where to go, where to park, where is a nice place to eat, we've navigated all that beforehand, so it is properly restorative."* (PwCFS/ME, Rachel)

Rachel also found that she could gain some of that restoration from days out as a family, which offered opportunities for her to achieve some of the outcomes in terms of family time and togetherness achieved through taking a holiday but on a more regular basis:

> *"What we would normally do is go for a little drive, go to the beach or the country park or something and it's not much physical activity but it's out of the house and it's a change of scene which is restorative."* (PwCFS/ME, Rachel)

Holidays also featured highly in the narrative of Steve's SO, Helen. In her description of days out with Steve, it sounded as though he perhaps found them restorative too, but for her, she communicated more frustration. For example, she described an instance when she had been unable to go on a planned holiday with her husband because she had been unable to leave Steve:

> *"He [Steve] wasn't really coping and then the final straw was my husband was going away with me, we were going to [place abroad], and then he [Steve] was round and he was down…and then we came home and finished the packing and then he came 'round about 11 o'clock and, and we didn't go on holiday…we didn't go."* (SO, Helen)

This excerpt demonstrates the difficulties in autonomy and individuation in the relationship between Steve and his mother. This description suggests that Steve fears being away from his mother and takes steps to maintain proximity to her, even though this means that his mother sacrifices her autonomy and consequently her own wellbeing. It was clear that for Helen holidays were of importance to her. However, her ability to enjoy her holidays seems to have been hampered by the lack of autonomy in their relationship:

> *"You know, going away on holiday, I mean we didn't go to [place] that year and then so we got…I made Steve get a passport and we said you can come with us and so we did get a passport and so we went, I think it was two years later that we went, he didn't really, he didn't really enjoy it, do you know what I mean? And then because I know he's not enjoying it then that puts it on to me, do you know what I mean? And then I feel guilty but then I think well I would never do anything or go anywhere."* (SO, Helen)

So, it seems for Helen that rather than tackle the issue of her being unable to do these things without her son, and him being able to do them without her, Helen instead handles it by sacrificing her own needs for time alone and time with her husband and other family members so that Steve can be with her. Helen also spoke of how Steve enjoyed days out which she facilitated, but that it would not usually be her preference to spend the day in that way:

> *"I mean in the early days and his illness, at the beginning and we used to just drive to [place] and we used to maybe just walk down the front and then just sit there watching the waves, I mean the other week we just went to [place], erm, and we just sat on some stones on the beach and it was just nice, do you know what I mean? Just nice and relaxing…and he took some pictures, erm, but I didn't really want to go, do you know what I mean, I wanted to do something else at that time but because he wanted to do it I knew that if I didn't then he'd get himself upset and this is where (whispers) it controls my life. And then another time if we've got the grandbains [grandchildren] he'd just go and he'd just look at the sea and things like that but sometimes I don't want to do things but I know I have to do them for a quiet life."* (SO, Helen)

So, it seems in these excerpts that Helen sometimes resents spending her time doing these things when she may have had other desires and wishes that she felt she had to sacrifice or ignore. It seems that she longs for autonomy and she longs for her son to have more autonomy too. For this dyad in particular, holidays and days out seemed to be a further source of complication and challenge, rather than escapism and solace. Had they been able to enjoy days out and holidays separately, perhaps this could have had a positive influence in their relationship through helping them to develop more independence from one another.

The evidence presented here suggests that holidays and days out are an important aspect of normal life and constructive relationship functioning. For some, they offer escapism, restoration and stimulation, even in situations where the relationship may be under considerable strain when at home, the strain can lift when on holiday. Holidays can offer an opportunity for family members to enjoy both autonomy and togetherness at the same time, and build or strengthen these elements of the relationship. However, when individuation has not been achieved within the relationship and holidays and days out are undertaken together out of a sense of duty or limited options, this may have a negative effect, particularly on the SO who may otherwise enjoy spending that time with someone else. This suggests that services that facilitate days out, simple holiday opportunities such as the cottage that Sally spoke of, and opportunities for respite care so that SOs can have greater autonomy, may all be of benefit to PwCFS/ME and their family members in maintaining balance within their relationships. It could foster both togetherness and develop individuation which I have constructed as crucial in balanced cohesive relationships in the context of CFS/ME.

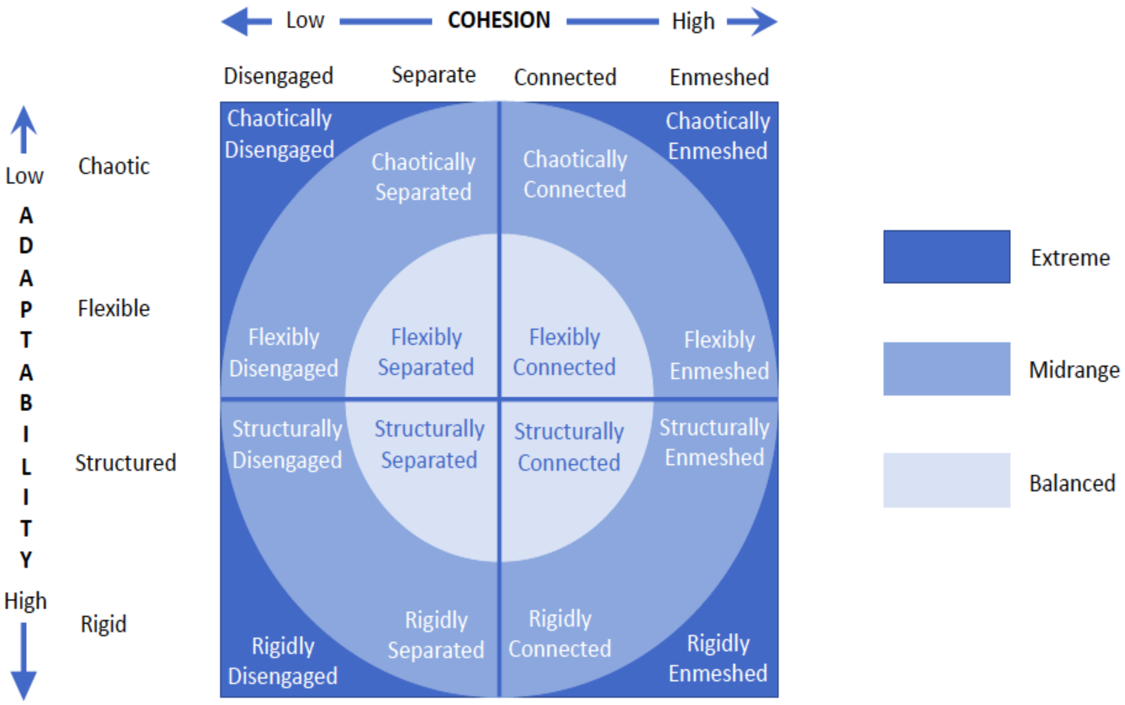
9.4. Balance & cohesion: A discussion

It has been argued that humans who feel close to others such as family members and yet simultaneously feel free to explore and develop their sense of self independently are most likely to develop optimally (Barnhill, 1979; Rogers, 1961). I have presented evidence demonstrating the consistent presence of these aspects and a balance between them in those relationships that appeared healthier. Occasional or growing presence of these aspects in less balanced relationships has also been illustrated, such as through the development of interests and a sense of self, or through breaking out of everyday routines such as on holidays. However, in relationships that were under strain these emerged as issues presenting further challenge to members of the dyad. In the relationships where togetherness and individual sense of self and autonomy were present, there is evidence that this brought comfort and satisfaction to the relationship and to the individuals within it, and love was spoken of explicitly.

In discussing the concept of cohesion and my relating this to enmeshment and disengagement, it is first useful to provide reflections upon how I came to explore these concepts. I had applied the term ‘enmeshment’ very early on in the research process when searching for an appropriate term to describe the relational processes emergent in the data. I selected the term as an independent concept, before having engaged with literature around family cohesion or family functioning, because descriptions of enmeshment fitted well with what I was seeing in the data. I had seen evidence of emotional overinvolvement as described by Band et al., (2014) but felt that there was more to this involvement than was explained by the term ‘emotional overinvolvement’. It felt to me that there was a something interactional - multilayered, social, emotional and behavioural - in these relationships that was keeping the dyads ‘stuck’ within patterns that lack a balance between connectedness and separateness. This was partly explained by theories of family homeostasis in systemic theory, but in terms of describing the quality and nature of these relationships, enmeshment felt like a better fit. I had also seen the effect of isolation and disengagement from relationships among some participants. As I explored the concepts of disengagement and enmeshment and sought to understand how they might be related to one another, I discovered the Circumplex Model (Olson, Sprenkle, & Russell, 1979), referred to in the literature as Olson’s Circumplex Model. This model (see Figure 3) places disengagement (not enough closeness) and enmeshment (too much closeness) at the extremities of family cohesion and views mid-level cohesion as ‘balanced’. It also considers the dimension of adaptability, in which too little change causes rigidity in a family system and too much change causes a chaotic family system.

Whilst there was some evidence of rigidity in some of the dyads, more so than chaos, I did not feel there was enough focus in the data to be able to apply the elements of adaptability in Olson’s model to my data. I decided that it was beyond the scope of this work to explore this in full detail. I began to explore instead the evidence for the presence of connectedness and separateness simultaneously in relationships that might be found in Olson’s view of balanced cohesion and soon discovered that there was evidence that these were both valued characteristics of the healthiest and happiest relationships. Upon reflecting on the codes and categories I had developed, it became apparent that there was evidence in the data that Olson’s model was a good fit for explaining the differences between the relationships that seemed to function in healthier, happier ways and those in which individuals seemed enmeshed or isolated and alone.

Figure 3. *Olson’s Circumplex Model*



Note. Olson’s Circumplex Model, adapted from Burr & Lowe, (1987) p.6

There have since been criticism and revisions of this model; for a review of these, see Burr & Lowe (1987) and Barber & Buehler (1996). In particular, it should be noted that evidence for enmeshment as an extremity of cohesion has been contested. Family functioning depends on the presence of qualities such as affection for one another, helping, caring for and supporting one another (Barbarin & Tirado, 1985; Moos, 1974). Meanwhile, enmeshment has been defined as patterns within families that inhibit the process of individuation, in which psychological and emotional fusing between enmeshed family members has the potential to prevent the development of psychosocial maturity and autonomy (Barbarin & Tirado, 1985; Barber et al., 1994). Multiple models of family functioning have proposed that family cohesion and enmeshment are two important inter-related elements of family systems (Beavers, Lewis, Gossett, & Phillips, 1975; Bloom, 1985; Olson et al., 1979) and some theories do suggest that enmeshment is the resulting extremity of excessively high levels of family cohesion (Epstein, Bishop, & Baldwin, 1982; Olson et al., 1979). However, others have treated enmeshment and family cohesion as separate constructs (e.g. Barber & Beuhler, 1996; Bloom, 1985; Moos, 1974;). Moos (1974) viewed high levels of family cohesion as indicative of healthy functioning, and some theorists suggest that cohesion represents positive patterns of interaction (Farrell & Barnes, 1993; Maccoby & Martin, 1983), whereas it has been argued that enmeshment is not a feature of relationships that are supportive but instead represents dysfunctional patterns that control and constrain family members, thus inhibiting individual autonomy (Barber & Beuhler, 1996).

Another model of family functioning could also be considered relevant, and links to more recent research into family functioning in CFS/ME. The McMaster Model of Family Functioning (Epstein et al, 1982) proposes six levels of family affective involvement which can be mapped onto Olson’s model:

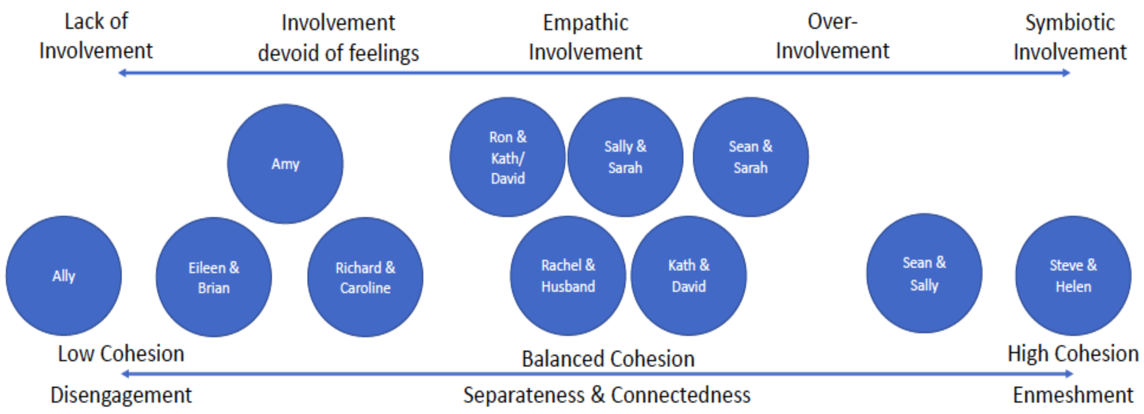
- Lack of involvement (Very low cohesion - Disengagement)
- Involvement devoid of feelings (Low cohesion)
- Empathic involvement (Balanced cohesion, considered healthy)
- Overinvolvement (High cohesion – bordering on enmeshment)
- Symbiotic involvement (Very high cohesion – enmeshment)

At the lower end of cohesion where disengagement features in Olson's model, Epstein et al.'s model suggests lack of involvement and involvement devoid of feelings may characterise relationships. In relationships that may be considered enmeshed in Olson's model, Epstein et al.'s model may recognise this as overinvolvement or symbiotic involvement. In symbiotic involvement, boundaries are either blurred or non-existent. Meanwhile, in Epstein et al.'s (1982) model, empathic involvement is placed centrally and is viewed as optimum for 'healthy' functioning.

Having interviewed a range of individuals in the context of CFS/ME and analysed the relational patterns evident in the data, my position is that there is evidence in the data that cohesion and enmeshment make sense as aspects of the construct of family cohesion, or at the very least as aspects of a model of family cohesion, if not the same construct. This is evidenced by the examples of healthy cohesion presented alongside evidence of relationships where enmeshment or disengagement was a feature, because in some of these relationships there was some evidence of healthier aspects of functioning alongside evidence of dysfunction. In relationships where there was evidence of low cohesion, individuals had disengaged emotionally, physically and socially from SOs. In relationships where enmeshment was identified, SOs attempted to be supportive and caring and there was evidence of other aspects of cohesion such as empathy and connectedness, but elements of balance between individuation, autonomy and togetherness were lacking. Meanwhile, it seemed that these individuals had moved beyond just supporting and caring into relationships that trapped both members of the dyad. The most extreme examples of this were present in Steve and Helen's accounts of their relational patterns, but were also evident in Sean and Sally's relationships. I have plotted the relational patterns that have been presented in this study along a continuum of family cohesion aligned with Olson et al. (1983)'s model of family functioning from low to high cohesion, with Epstein et al.'s model of affective involvement mapped over it (See Figure. 5. *Family cohesion continuum.)*

Figure 4 communicates my conceptualisation of where participants might fall within Olson et al. and Epstein et al.'s models. Whether the terms used to explain the relational patterns are cohesion, enmeshment, disengagement, lack of involvement, overinvolvement, or any other construct or label that might be applied or considered similar, the evidence I have presented suggests that family functioning is related to degrees of involvement with one another. I have presented an argument that disengagement sits at one extreme with boundary-less enmeshment sits at the other extreme. A 'healthy' level of cohesion sits in the middle – a balance of individuality and connectedness, autonomy and togetherness.

Figure 4. *Family cohesion continuum*



What is novel about the work within this thesis is the identification of aspects of balanced cohesion in relationships in the context of CFS/ME. No causal relationship between cohesion and recovery can be inferred, but there is evidence that dyads and families that function in more balanced and cohesive ways maintain more positive attitudes towards one another, and towards the management of their lives and illness.

In Rolland's family systems-illness model (Rolland, 1984; 1987; 1994) the changing demands on the whole family over a period of time in dealing with chronic illness are conceptualised. It considers family development as progressing in stages of varied levels of instability and imbalance, each of which gives way to a new state following adaptation. In typical family development, families alternate between periods requiring high levels of cohesion within the family (centripetal family style), such as when a baby is born, and low levels of cohesion (centrifugal family style), such as when children become adolescents and become more independent. In chronic illness, it is suggested that families go through regular periods of reorganisation as they try to manage the inconsistencies of the illness, periods of improvement and relapse. In Rolland's model, high levels of family cohesion may be required in taking care of the afflicted family member at times that would normally require lower levels of cohesion. This is viewed as potentially very disruptive for the family as it forces them to go against the natural flow of family dynamics in the normal family life cycle. These changes can also affect development of the individuals concerned (Rolland, 1984; 1987; 1994).

Certainly, my findings support Rolland's model, particularly in the cases presented where the period of illness struck at a crucial point in the family life cycle. For example, Rachel was raising young children and her husband had to take over the running of the home, which she found distressing. Sean and Steve were the two cases in which enmeshment has been presented as a core feature; these were also both mother/son relationships. It is therefore likely that the very nature of these relationships as parent/child relationships was a factor in the quality and styles of relating – for example, would one expect to see togetherness in a parent/child relationship in the same way as one might in a healthy adult/adult romantic relationship? In both cases, the sons had become ill at the time in the family life cycle when they might otherwise have started to develop autonomy through the process of individuation, and this had been hampered by their illness and resulting need to remain in the family home. Neither son had been able to develop extensive social relationships or romantic relationships, which supports Rolland’s notion that normal development can be affected by the disruption to the family by the greater need for high levels of cohesion in CFS/ME. Conversely, all of the individuals whose relationships under study here were at a medium level of cohesion, or were disengaged, had already been through a process of individuation beyond their family of origin, and all had established other social, working and couple relationships. This suggests that although I have presented evidence here that the characteristics of the relationship do interact with and influence coping strategies, there may be an argument for treating specific types of dyads (e.g. husband and wife, mother and son/daughter, father and son/daughter) as separate categories in future studies in order to clarify whether coping tends to be distinctly different in different types of relationship, or whether they can be better understood by differentiating them according to other psychosocial theories such as attachment styles.

Balance & Cohesion: Final thoughts. At the beginning of this section, I outlined the questions that were posed during the analytic process, as the problem pattern had become the dominant category within the research, and I felt that there was a need to interrogate the data further with a focus on what helps:

1. Is there a middle ground; a space where the SO can both support and encourage without accidentally and inadvertently worsening the condition of the PwCFS/ME? If so, what does it look like?
2. Is there evidence in the data that this approach has positive outcomes?

3. How can this evidence be meaningfully linked to existing theory?

I have presented evidence that there is indeed a middle ground. It is given the name ‘balance’ which is a ‘healthy’ level of cohesion and is characterised by the coexistence of togetherness and separateness. Love and a commitment to spending quality time together both seemed helpful in the maintenance of togetherness. Meanwhile, autonomy can be achieved if both members of the dyad develop or maintain a sense of self through the nurturing of relationships and/or interests beyond the dyad and beyond their illness.

The evidence I have presented has been considered alongside theories of family functioning. The extremities of disengagement and enmeshment were a good fit with much of the data presented in the problem pattern, and were constructed as concepts prior to engagement with literature around family cohesion. However, after reading about Olson et al.’s (1979) model and posing further questions regarding the data, it became clear that there was evidence of a middle ground, consisting of healthy cohesion. Therefore, despite research that challenges models of cohesion and poses alternative models of family functioning, I felt that my adapted version of Olson et al.’s model offered the best fit for explaining what I saw in my data, and for understanding relational patterns and interactions in the context of CFS/ME.

Individuals presented here seem to have found ways to achieve positive outcomes such as finding joy in their relationships and adapting to their illness so that they feel they can get the most out of life in the context of their illness. Although I have evidenced that families cope better with CFS/ME when both togetherness and separateness coincide, I have presented limited evidence that within this space the SO both supports and encourages without worsening the condition as per the question. To understand whether this can be achieved, we have to look more directly at the ways individuals coped specifically with the illness and at what they believed worked best in terms of attitudes and approaches to coping.

10. Category D: Acceptance

This fourth and final category emerged as an aspect of relationships and of coping with the illness across the dataset. There was a general recognition among participants that acceptance is a necessary part of moving forwards and of finding ways of living a meaningful life in spite of or beyond their illness. Letting go of losses emerged as necessary so that the PwCFS/ME can move into a more positive mindset, thereby helping the dyad to move beyond the guilt, anger, frustration and sadness experienced in the earlier days of illness. The process of acceptance requires reflection which can be a positive force in the relationship between the PwCFS/ME and their SO. The evidence I present also suggests that for the PwCFS/ME the presence of acceptance from others is of importance to the PwCFS/ME in beginning to recognise and nurture the positives in their life and in maintaining healthy attitudes towards themselves and relationships.

10.1. Facing loss

PwCFS/ME and their SOs experience a multitude of losses as their illness progresses. In Category B. The Problem Pattern, I highlighted sources of grief and experiences of the various stages of grief in the context of CFS/ME for both the PwCFS/ME and their SO. This section will focus on the sense of loss experienced by the PwCFS/ME. It was clear that SOs also experienced many losses, and it should be noted that loss featured highly across all of the transcripts. However, full exploration of these is beyond the scope of this research. The main aim here is to illustrate the importance of acceptance in moving forwards, and so I focus here on evidencing the kinds of losses experienced by PwCFS/ME in order to then move on to discuss the ways in which individuals dealt with these losses.

Sean (PwCFS/ME) described his sense of loss as being about the hopes he had previously had for himself for the future. As a teenage boy with a good academic record, he previously had ideas about where he saw himself in the future, what kind of career he might have, and the life experiences he might have along the way:

> *"After I recovered first I thought I might want to do something medical, that's never really left me. It's in the back of my mind now since 14,15. I've always been pretty good at science. I wanted to go to uni and try and do something... whether it be med school or something related, that was the road I saw myself on. Since being ill, like I said it's just in the back of my mind now. I'm not sure if I want to do it anymore. I don't know if it's an option anymore. Not recovering." (PwCFS/ME, Sean)*

It is particularly interesting that Sean seems to have let go of this as an option now. During the interview, I did get the impression that he felt a great deal of regret about these hopes never coming to fruition. However, it is fair to conclude that Sean is beginning to see these as losses and in reflecting on this perhaps he is learning to accept them. But with Sean, this could also be interpreted as a defeat – the statements '*I'm not sure if I want to do it anymore. I don't know if it's an option anymore'* sound more like someone who is giving up than someone who is accepting and finding ways to move on.

A sense of loss and the process of coming to terms with this was clearly demonstrated by Rachel. This included loss of her previous capabilities, which were also contributory to her pre-morbid sense of self, a sense of having lost things she did have in her life prior to illness – things she used to do that she is no longer able to now – so the loss of hobbies, interests and simple pleasures:

> *"I just can't be the Mum that I want to be, or the colleague that I want to be, or the wife I want to be, to give as much as I receive (begins to cry)...it is*

> *difficult…(long pause)…I feel a sense of loss, grief; I used to love being in the garden, I used to love walking the dogs, going for runs, feeling the wind in my face, feeling physical pleasure from exertion…I miss a lot of things, and I feel jealous when I hear of the things that others do…I have a friend up the road…she plays hockey and goes for a run every day and I'm jealous…"*
> (PwCFS/ME, Rachel)

In this excerpt, Rachel is communicating the loss her family members may have experienced as a consequence of her illness – the impact on her identity as woman, her and her husband's identity as a couple, and the functioning of the family unit. These losses give rise to comparison with others and feelings of envy when she sees what others can do that she cannot, which further exacerbates the feelings of loss. It was clear that the disruption to family norms was perceived as a loss and a source of grief for Rachel. As she spoke, Rachel broke down and clearly conveyed the anguish and loss she felt about the most acute phase of her illness because it left her unable to be as present to her two young children as she wanted and needed to be. She expressed distress at the extent of her perceived impairment, and although Rachel was a participant who went on to describe that she has now made peace with her situation, the complex emotions triggered by the experience of a complete loss of her sense of self, of functioning, and of the life she had previously are a direct result of her impairment and physical discomfort.

Rachel's description of this experience of loss fits with the dual process model of coping with grief (Stroebe & Schut, 1992). Although she is now living a meaningful and positive life in which she has learned to cope with her limitations (restoration orientated), she is also able to get in touch with her sense of loss at becoming so impaired and the burden this placed upon her family; she still experiences her deep sense of grief (loss orientated). This process occurs simultaneously for her. She also describes her anger, which is the second stage of grief in Kübler-Ross' model (2009). In 1969, Elizabeth Kübler-Ross (1929-2004) presented her book 'On Death and Dying' (Kübler-Ross, 2009) in which she proposed a five-stage model of grief. The five stages are:

- Denial
- Anger
- Bargaining
- Depression
- Acceptance

Although Rachel is now in a place of acceptance, which is the fifth stage of grief according to this model, the fact that she is still able to feel her anger at such a loss supports Kübler-Ross' notion that people move in and out of the stages of grief often and on an ongoing basis.

The experience of loss was not only felt by the PwCFS/ME; SOs experienced a sense of loss when comparing themselves and their relationship with others. This was described by Helen (SO of Steve), who spoke of the life that her son Steve (PwCFS/ME) could have had but has not been able to have. The loss is highlighted by Helen as being in stark contrast with the life her other children have been able to have:

> *"I think he just wants more, he sees his brother and sees his sister…and now my other son, he's 38 and they've got [name of daughter] but they are expecting a baby but I couldn't say anything to Steve about it, but I mean it's only early days I mean she'd only be like 12 weeks now but I mean my son comes every week anyway they come every Sunday, they come for their dinner, but it hadn't been brought up in conversation and I hadn't actually told*

> *[Steve]…I just wish he could be healthy like my other son, erm, but it's… I think it's going to be a long way away off if ever…"* (SO, Helen)

These are not losses of things that Steve once had; they are things he has never had but may not now have the opportunity to have: so, it is a loss of an imagined future self. They both experience a sense of loss because they are things Helen, and possibly Steve, thought or hoped he might have in his life at some point, and now that opportunity seems to have passed him by. He has lost them as possibilities that never materialised. In the phrase '*he sees his brother and sees his sister'*, Helen is suggesting that Steve feels envy when he sees what his siblings have in their lives - partners, children, jobs. In her comparison of Steve to his brother and sister, Helen imagines that seeing what they have leads Steve to feel more aware of what he does not have and of what he has missed out on. Steve confirmed this sense of loss and of having missed out. When asked about the impact the illness had had on his life, he responded:

> *"Well it's devastated it! (Quietly) I've not been able to work, I've not been able to have relationships, social life, I mean, just enjoying things, you know, my relationships to my family and friends…"* (PwCFS/ME, Steve)

Sean's Mum and sister, Sally and Sarah, also described the process of comparison with other people in relation to loss and grief, and suggested that in order for Sean to move forwards, acceptance was key:

> Sarah: *I think he's beginning to accept it but for a long time he's not wanted to.*
>
> Interviewer: *Do you think in that time he's been angry then?*

Sarah: *Oh definitely. He has been.*

Interviewer: *So, fighting it with anger has become almost… counterproductive?*

Sally: *Yeah, it's become almost like a grieving process, it's highlighted that his peers from school are going off to university, and he has come off Facebook because there was just a constant feed of things reminding him, and that has become difficult but now that he's beginning to accept things he might be able to cope.*

Sarah: *But he can cope with me doing it and he tells me that he's happy that I'm doing it, but he can't cope with friends doing it and I think it's because they're his own age.*

10.2. Reflecting and reframing

The sense of loss is clearly highlighted for PwCFS/ME through seeing what others are doing and realising what they themselves are missing out on. However, whilst it is necessary to face and come to terms with these losses, it is also crucial to begin moving forward. In order to begin the process of letting go of and accepting these losses, individuals appeared to benefit from a process of reflection and reframing so that they were able to move forward with hope. In doing so, they began to release their sadness and anger and developed an appreciation for what they do have in their lives, particularly with regard to relationships. After describing her sense of grief and loss, Rachel explained:

> *"…but I live in a lovely house with a husband who, he adores me, he really, really loves me and I really love him, with two healthy children with whom I've got really healthy relationships, you know, they confide in us and they*

> *trust us and they love us, so I've an awful lot to be thankful for, and that thankfulness actually is important because without being thankful and recognising the blessings then it would be a really bleak place to be…that thankfulness, it is really important, it does keep me more…it is good for my wellbeing."* (PwCFS/ME, Rachel)

This excerpt provides evidence of how someone who has experienced such a sense of loss and grief can begin the process of making peace with this by reframing and reminding herself of the things for which she is thankful. Rachel was not claiming here that she has completely let go of the losses she has experienced; she spoke about her grief and her feelings of jealousy in the present tense and became visibly emotional during the interview. But she does counter-balance her sadness with her recognition of and appreciation for the positive things in her life. It is clear she has reached these reframed interpretations through a process of reflection, and that her relationships with her family members and with friends are what enable her to feel positive and thankful.

Not all individuals with CFS/ME or indeed chronic illness may be able to reflect in such a way. Rachel explained that she sees herself as very reflective and as 'lucky' because she views her ability to reflect as being largely a result of her education, which she said was 'inherently reflective':

> *"…but you see I'm really highly, highly educated and my discipline is inherently reflective, so my ability to analyse and look at and try and respond to the situation I find myself in is possibly not common."* (PwCFS/ME, Rachel)

Certainly, her reflection and reframing were the most pronounced across the dataset, but they give us clues as to how someone with CFS/ME can begin to move forward with their life in a positive way. Rachel had also had counselling which she viewed as very useful. She felt that many in her situation who haven't had such high levels of training in reflective thinking may not know how to reach beyond their current suffering and start to see their situation differently. Rachel also attributed her time alone thinking as important to her ability to reflect, and her ability to reflect as crucial in maintaining her relationships:

> *"You know, it is easy to take each other for granted isn't it? But I do have, I work two days a week, and the rest of the week I'm resting; I do have a lot of time alone in which to reflect and think about how to have a good life within the confines of really limited energy, and I do want to have a good life and it's really important to me that the girls see that I'm…you know, that the girls have a nice life in a house with somebody that is chronically unwell…it is difficult…it is difficult but I'm really lucky that I'm able to reflect and I'm able to find ways to make the most of it and create a good life."* (PwCFS/ME, Rachel)

What is also particularly poignant here is Rachel's emphasis on the fact that she actively has to find ways to make the most of her life and enjoy her relationships with those she loves in the context of her illness, which she views as beneficial to the whole family.

Sean's mother, Sally, also spent time reflecting during counselling sessions, and found this to be conducive to improving her interactions with her son. She felt it may have helped them reframe their relationship in a more positive light:

Interviewer: *With the counselling do you feel it helps?*

Sally: *Yes, definitely. It helps you get things into perspective and maybe tackle things in different ways. When you get yourself in a state, in a low mood, you can't necessarily see ways to deal with other people so you don't know how to lift someone else.*

Interviewer: *Do you think in turn that has helped Sean?*

Sally: *I hope so. I hope that because I've offloaded some of the weight I've had, I can then help him in a better, more positive way.*

The phrase '*putting things into perspective*' suggests that Sally recognises that things could be worse and that the way she thinks about things does have an impact of Sean and on their relationship. She appears to recognise that the situation at home may be having a negative impact on her mood which in turn negatively impacts upon Sean and her interactions with him. Through having the opportunity to reflect and reframe things in counselling sessions, she had been able to take a step back from their relationship and rethink their interactions, which she believed had helped her to help him.

Kath also recognised her ability to reflect as crucial in her having overcome her illness and returned to her normal levels of functioning. She considered her reflective nature and intellect as reasons why she was able to do this:

> *"If you can't get your head around it, I think I'm very much a thinking person and I think around problems and think 'Well, how can I get around this?' and if there's something I can't do, is there a way I can get around it or do I give up and you know, not bother?"* (PwCFS/ME, Kath)

Kath believed that her reflection enabled her to problem-solve in a positive and pro-active way. She believed she and her son were both ‘positive people’ and that this had an influence on how they managed their illness and the fact that they both eventually managed to get through it:

Kath: *On good days as I say I had it at the same time for different reasons…I was feeling good, he was feeling good and I’d say ‘Come on shall we go out? Do you feel like getting out?’ and we’d go and have a walk somewhere on days when we were okay…he didn’t talk a lot about it, if he felt bad he had emotional support from me but we were the sort of, we’re both made from the same cloth in that we tend to get on with life, you know…we’re both positive people, you tend to be a negative person (towards husband), [Rob] and myself are quite positive…*

Interviewer: *…Do you think that impacted on the way that you dealt with things at the time?*

Kath: *Yes, I think at some point you think you’re going to get through it but I think again probably both very much the same book with him being positive and me being positive you do think there’ll be a way out of it.*

Kath’s son, Rob, also considered his mother’s positivity and ability to reflect on things in a positive light to be crucial to his proactive approach. In his interview, which was conducted separately from his parents’ interviews, he said:

> *"I was sort of looking for stuff on the internet and I was also talking to this doctor, and [Mum] just sort of said 'oh' you know, she was more positive and said 'Yeah you know, it's definitely...you can try that'. When I said 'suggestions' maybe I said the wrong word there really, it was more like reaffirming, you know, keeping positive about, saying 'Oh, you can try that, it definitely might help, er, yeah, and yeah, so…"* (PwCFS/ME, Rob)

Rob appears to have found it helpful that he felt supported and accepted when he took a proactive approach in finding solutions. His mother supporting him in this process was meaningful to him in maintaining his own positivity and effort, and in maintaining their relationship.

Through reflection and reframing, it becomes possible for the PwCFS/ME to begin to accept the situation as it is. This may mean accepting limitations, accepting pain, accepting energy levels that are lower than most people's and accepting the changes that have occurred in many aspects of their life due to their illness. Acceptance does not mean giving up on attempts at recovery or becoming resigned to illness. Rather, it means approaching the situation with greater calmness, less stress and decreased negative emotion so that new ways of living can be adopted. Amy explained that initially she believed a proactive, positive attitude was needed, but over time acceptance becomes necessary:

> *"Some of the people say 'Well some people say you'll never recover from this' and that's not good I don't think, not until you accept the fact you have to live differently, you're okay then, and it's okay to say 'Well maybe, maybe not', but before you reach that point, at the beginning when you're still quite confused and your life's still a muddle because you're in a different mindset, a*

> *different place, I think at that point it is best to stay positive more than anything else…"* (PwCFS, Amy)

Steve had also started to focus on acceptance. Following a visit to a specialist clinic, he had been introduced to mindfulness, an approach to acceptance that is gaining pace in Western medicine (Kabat-Zinn, 2001), and he found it particularly helpful, especially in learning to manage his emotions:

> *"I'm part of a meditation group, which I do online…that's the thing I probably found the most helpful, psychologically, especially the mindfulness…I think first of all it gives you the awareness of what is going on, you know, the thought pattern during and the behaviour during, and it helps you to step back from it, to see what's going on, you know to come back to the present to ground, it's like if you're feeling stressed you can just ground yourself and reverse it…I mean that's the thing about mindfulness, really what you're doing is, you know if you go into your head you're kind of disconnecting from your emotions so when you actually do become more present you are then more in touch with what's going on…"* (PwCFS/ME, Steve)

Steve explained how this approach had enabled him to deal with emotions that he had previously struggled with, and that it had become a useful strategy for accepting the negative emotions he experiences:

> *"It was certainly more difficult at the beginning because you're getting in touch with things that you probably haven't wanted to feel for a long, long time so then you've got that to process…I've done a few different things [to learn to manage*

> *that], erm… I mean just allowing it to be there and breathing with it…If I can accept it and breathe with it, it just kind of eases, it can pass through, erm, I mean sometimes you can cry, which is great, and the clinic you know, they also talk about digesting your emotions… because feelings are information, there's a reason for them to be there, that's what I've learnt, so they're not actually bad, even if it feels bad, it is telling you something."* (PwCFS/ME, Steve)

Having presented many excerpts demonstrating frustration, anger, and other difficult emotions that were experienced by Steve and his Mum, Helen, in Category B. The Problem Pattern, this excerpt provides evidence of how far Steve seemed to have come. During our interview, I gained a sense that Steve was accepting of his situation now, and that he was also accepting of himself. The narrative of his mother which focused on negative experiences may have drawn on events that happened some time ago. She did not appear to have accepted or moved on from her own suffering in relation to her relationship with her son or his illness. Steve was working hard on developing a calmer, more positive attitude, and it is possible that there was disparity between Steve's perception of himself now having worked to develop acceptance, and his mother's perception of him. His mother was adamant that counselling and similar opportunities to reflect would not be of use to her, but this could be having a negative impact on her emotionally and on her perception of her son and her ability to let him move forward:

> Helen: *There's been times he's been really down, I've always been there to support him, but this one time I said 'Steve I can't go through all this again', and I've said that a few times but this one particular time he just turned around and said "just get counselling then".*

Interviewer: *What would be your thoughts on doing something like that?*

Helen: *No, I wouldn't go, I don't know how a counsellor would help me. I work through it in my own way.*

Interviewer: *Mm. So, you wouldn't give it a try if it was an option?*

Helen: *No, no. So, I'm a bit negative there, but I don't feel like I need to see a counsellor, because what I'm doing now is I'm talking to you and that brings up emotions and I've worked through things how I feel I should do things. Like, I know [Steve] and I'm the parent, and I do things my own way at the end of the day.*

In choosing not to engage with outside others in order to reflect and think about different ways of coping, it is possible that Helen keeps both herself and Steve stuck in potentially unhelpful patterns of relating I have presented.

10.3. Acceptance from others

An important part of the process of moving forward is for others around the PwCFS/ME to demonstrate acceptance. This involves removing their resistance, challenge or other negative responses towards the PwCFS/ME, as these responses can be a source of anger and frustration for them. Having presented evidence of the importance of positivity from others, it is also important to highlight the fact that most participants experienced negativity from others and that this was perceived as unhelpful. This negativity was experienced by some from family members, friends, and in general social interactions, but for many it was experienced during interactions with health professionals.

Ally explained how she perceived the response of older relatives, and the feeling of shame that these responses caused for her:

> *"I think people think you're just lazy. Like oh you've had plenty of rest you should be alright now. You can rest a year and still not be alright. In my family, my elderly relatives, erm… I think it was very difficult for them to see their young niece who worked and was in the thick of it… I got comments like 'If only she tried a bit harder', and one uncle, very elderly, thought it would be a good idea if I got married and had kids and that would take my mind of the tiredness. That is a different generation and I do accept that. But there is a stigma…Nobody would choose this life, it's an existence not a life. [I feel] Ashamed. Yes, ashamed really. I can't afford to have that, that's quite a negative feeling."* (PwCFS/ME, Ally)

In reading this excerpt, it becomes clearer why Ally may have become so isolated and disengaged – experiencing these feelings of shame because of relatives' lack of acceptance of her situation led her to need to protect herself by withdrawing.

Amy's second interview also illustrated the importance of acceptance from others. Family members can attempt to be supportive but when the PwCFS/ME feels that they, their illness, and their situation is not fully accepted by their family, the support is limited in its usefulness. Amy described the variation in the support she received from different family members, and how challenging it could sometimes be not to have their full acceptance:

> *"…financially they helped, which is a huge thing…my Mum and my Gran were always sending me articles. My Gran for ages would send me articles about ME and things like that and my Mum would try to get hold of vitamins and specialist serums…but then Dad thought 'It's not good to have vitamins' blah de blah so it kind of…although they wanted to help it was with*

> *some negativity I think…I suppose sometimes the emotional support from Dad was there but sometimes not, it was a doubled-edged thing, sometimes it was a very good thing, sometimes not…"* (PwCFS/ME, Amy)

Amy highlights here the internal conflict caused by hearing different advice and perspectives from various people. When health professionals are unable to offer answers, people seek alternative approaches to managing their difficulties. Amy's well-meaning family members attempted to demonstrate their support by seeking out relevant literature and sources of information. This may have felt validating for Amy in some ways, but could also have been a source of much confusion, because of the lack of evidence base and the plethora of unsolicited and sometimes unhelpful advice published online and in magazines. It also seemed that it was a challenge for family members to show their support without Amy feeling that they were forcing perspectives upon her, and that they were therefore not accepting of her current situation, instead pushing for change. She described sometimes feeling that her closest family members did not believe her:

> *"I suppose not forced help, but offered help [was most useful]…I would say when I needed it, because it wasn't all of the time, some of the time I would get the support, like if I wasn't really okay, there would some support behind me to go to the doctors, but then when the tests kept coming back negative, they'd be like 'Oh, the doctors know what they are doing, there must not be anything wrong'…it would flip, and that would be horrific for me, because I would be like 'No, but it's still the same, I told you before, there is definitely still something'*

> *so it went from being totally supportive to totally alienated."* (PwCFS/ME, Amy)

The true cost of not feeling that her illness and her experience were accepted and understood by those closest to her appears to be the alienation, which she describes as 'horrific'. I argued in Category B. The Problem Pattern that Amy became isolated and disengaged from those around her. Her narrative suggests she had to plead with her loved ones to believe her and that her self-reported subjective experience of feeling ill was discounted because of a lack of medical evidence of the existence of her illness, even by her closest family members.

Having experienced such alienation, Amy was clear about how important feeling accepted by family members was for her in moving forward and reducing her psychological suffering. In response to a question about what could be most helpful in the way family members could support her, she responded:

> *"I think the support thing, whilst it is good, it's really good if you can say to somebody 'I'm telling you, I'm not okay, I'm going to go back,' and I think accepting that is really good and I think trying different things, and support if it did fail being along the lines of, 'Well if it doesn't work', being 'Oh well, next!', like brush it off, no negative impact with that…it's trial and error, it's all a process and [they need to] understand that every day is different, or every minute is different, and the non-judgement I think…"* (PwCFS/ME, Amy)

This illustrates Amy's desire for the freedom to handle her illness in her own way whilst also receiving unwavering and unquestioning support from family members. This may be a difficult balance for anyone to achieve, particularly in an illness that is so controversial and where its very existence is in question, but Amy's excerpts illustrate that without achieving the fine balance of this type of support and acceptance, the individual with CFS/ME can end up feeling isolated and alienated from family members.

I have already presented evidence that both Kath and Rob felt that they were positive and both felt supported by one another, and by David (Husband and Dad). David acknowledged in his interview that he had been accepting in his approach to coping with his son's and his wife's illness:

David: *Well, changes in [Rob], he couldn't – he would run a lot and he couldn't do that, erm, he deferred going to university because of that.*

Interviewer: *How did it make you feel, to see that happening?*

David: *I just accepted it, I didn't sort of, you know, I presume I was a bit, not upset about it but a bit worried about it but I didn't, I just thought that's life, and if he goes back another year he doesn't, you know, it doesn't matter really…*

Interviewer: *Do you think he appreciated the acceptance?*

David: *Well I'm sure he did, I never spoke to him about it but I'm sure he did, yes. Because we looked after him and we didn't charge him any rent or well, you know, he didn't have to pay us for anything at that time because he wasn't working…he had no problems, he had no financial problems at all, I mean I didn't know, he had* *some savings obviously but I didn't know what he had, but we never said to him 'Oh you've gotta [SIC] pay for food' or anything, just accepted it really. Yeah.*

Interviewer: *And for [Kath] then, when she became ill…how did it effect the relationship?*

David: *Erm, again I accepted it really, obviously I put down that I'm a planner so I was unable to – I always think of what we're going to do at Christmas or what are we going to do for holidays next year…And then of course, we had to cancel a holiday to [place], we were going – that was a bit upsetting for me because you know I, and her of course, but we were fully insured…*

It is interesting that this attitude of acceptance was present in the relationship in which I have argued that cohesion was at a healthy level. Meanwhile, acceptance was not always present in the relationships where there was dysfunction. I would like to return to two sections of the earlier excerpt presented from Helen in which she talked about her other son having a baby and her feelings about telling her son Steve about this:

> *"…they are expecting a baby but I couldn't say anything to Steve about it…I just wish he could be healthy like my other son, erm, but it's… I think it's going to be a long way away off if ever…"* (SO, Helen)

Helen is expressing here her own anxieties about how she imagines Steve might feel, and her own desires for his life. She is also communicating something about what she values as evidence of a successful life, and that she feels he has lost the opportunity to live that type of life. In wishing he could be healthy, she is not accepting him as he is, in his current state and situation. Instead, she is fighting against it and viewing him as not strong enough, which serves to fuel her sense of grief and loss, as well as being a source of frustration and limitation for Steve. This is in stark contrast to the excerpt from David, although it should be noted that Rob was ill for one year and had made a full recovery several years before our interview, whereas Steve had been ill for nearly 20 years and still considered himself to be ill at the time of the interview.

Although Steve's narrative was initially dominated by his anger about feeling let down by his GP and isolated from his family, he then focused more on describing the skills he had learned that are helping him cope with his illness. He communicated a sense of feeling limited by others' expectations of him. For example, his mother's desire to protect him appeared to be experienced as limiting for him when he attempted online dating:

> "*So, I did kind of decide to have a go at internet dating. And erm, I started doing that end of last year, November time, and I did get talking…We went on a couple of dates, I did like her but I didn't fancy her, she wasn't the one…I talked to a few people about it and only got two negative reactions and one of those was my mother. She wasn't so sure about it…being ill does make you more vulnerable. Again, I did feel… it wasn't a massive reaction to it, maybe disappointment, but then I kind of expected it really. Whereas a few others were really positive but she wasn't… I've learnt to be a lot more proactive. Erm, really learning the psychology stuff has helped that."* (PwCFS/ME, Steve)

Although his mother's perspective seems more shaped by the belief that the opportunity for a relationship has passed him by, Steve's narrative suggests that a relationship is something he is still working towards. He communicates having reached a place of acceptance of his situation and actively trying to live his life in spite of it. His mother's narrative on the other hand seemed dominated by loss, grief and enmeshment. Her anxieties and inability to let go of losses, and perhaps to let go of Steve a little too, appeared limiting for him. Her focus was on wishing he could be something he is not instead of accepting him for who and what he is and supporting him in his attempts to move forward.

The perceived lack of recognition from GPs in particular also emerged as a barrier to PwCFS/ME feeling accepted, as well as potentially have consequences in family relationships of PwCFS/ME. There were examples in every participant narrative that evidence this point, but I will present just a small selection here. PwCFS/ME often described seeing multiple GPs who they felt had been unsupportive before they finally found one whom they felt offered them validation and acceptance. Rob (PwCFS/ME) explained his negative experiences with GPs, before later moving on to describe a GP he saw as accepting and supportive:

> *"The GP I saw, erm, she kept just going on about antidepressants and 'Are you depressed?' and I got to the point where I was just getting so frustrated, you know, I think at one point I did get a bit ratty with her…but this is what I found over years, going to GPs in the past, they don't look at it as a holistic view, they just look at 'Oh, you're depressed', they don't look at why, why the problem is… [my current GP], he suffers with ME himself…because he's had a problem where he's been ill himself, he has more empathy…he's a bit more of a sort of openminded doctor…"* (PwCFS/ME, Rob)

This idea of doctors having to be openminded in order to be the type of doctor who will support someone with CFS/ME effectively appeared in a number of interviews. Another example was that of Ally. When describing her feelings about her own GP, who she identified as being the most significant other in her life and her main source of support, she said she felt believed, understood, accepted and that he was openminded and proactive:

> *"He's a really together chap, first of all he believes what you say. He understands that there are multiple aspects to M.E., so not everyone has the same range of symptoms. I probably speak to him once a fortnight and we discuss pain relief. I don't know how he's trained but he comes across as very person-centred. He never forces anything on you… he is not dogmatic… what I particularly like about him is that he's really open for me to try various things…He thinks outside of the box. If I say to him what do you think about trying this or whatever, and yeah, he's open to trying, for me to try various things. I think a lot of doctors, if you don't fit into a category then you're a bit bonkers… and you read that over and over again if you read online, where people have seen their doctor and not been believed."* (PwCFS/ME, Ally)

Similar to Amy's description of how she preferred family interactions to play out, Ally expresses here the value she finds in her GP not being forceful with her, instead allowing her to find her own ways of coping, being accepting of her feelings and supportive of her attempts to find ways forwards. We also see again in this excerpt, as in Amy's, that the concept of not being believed is of great importance to PwCFS/ME, whether that is by loved ones, acquaintances or professionals. The period when they felt they had not been believed was typically a period of frustration and anger, but once they found that acceptance from others, it offered them relief and a sense of freedom to explore new ways of coping in a supported way.

Another excerpt I found particularly interesting in thinking about acceptance from others was one from Rob' narrative in which he described a treatment recommended by a doctor which he believed to have been helpful. Rob described the treatment but at the same time spoke of how the doctor had been positive and that he had then felt positive. Rob did not draw a link between the attitude of the doctor, his own subsequent attitude, and his perceived response to the treatment, but the three did seem to coincide:

> *"He said like, what I've found…it's very difficult to grow yeast so he said the likelihood is that you have got an overgrowth of yeast candida probably as well he said, 'cause you've got such low levels of good bacteria so he told me to take caprylic acid erm, and I took that and the first three or four days I felt absolutely dreadful, I thought oh you know, I felt really terrible but then he made me feel you know quite positive 'cause he said well, that's because it's killing everything off so even though I felt quite unwell, I felt quite positive and after a week I started feeling a lot better and that did help a lot as well, so…"*
> (PwCFS/ME, Rob)

The behaviour of GPs towards PwCFS/ME also has the capacity to influence the relationship between the PwCFS/ME and their family members and social contacts. Eileen succinctly described this link:

> *"So, it's various things that cause this illness and this is what isn't easy for the partners and the friends and the family to understand. If the doctors can't*

> *find out that you're ill perhaps you're not ill and you're just putting it on, and it's annoying for them and it gets in the way and, erm, it's very…it can make people very, very unhappy, very unhappy, if you find a partner that is good then that's splendid."* (PwCFS/ME, Eileen)

It can also be liberating for PwCFS/ME to learn to accept the stigma of their illness; the challenges they face in accessing treatment via the NHS can bring about such negative emotions that it serves to worsen the wellbeing of the PwCFS/ME. Accepting the fact that there is a lack of adequate support available from many GPs can also be a step forward for some, enabling them to then seek more helpful treatment, because if they do not accept it, it has the potential to keep them held in a place of frustration and unpleasant emotion which takes up valuable energy and resources that could be better used elsewhere. For example, Steve (PwCFS/ME) shared his theory on why people in his situation do not get the support they feel they need, but how he had given up fighting it:

> *"It's a waste of time. It's a complete waste of time, going to the NHS for stuff like this. It all flies under the radar…unless you are like so ill you are like hospitalised, because that's what doctors see in hospital, so you see people like me, we are like in the middle between being so ill that you can't get out of bed and the other end… They just don't deal with stuff like this so almost all the tests I had done have come back as normal… I think I've just got to a point of acceptance with it, that's the way it is, you know, I don't, I don't think about it, I don't bother, I just don't bother going."* (PwCFS/ME, Steve)

Whilst Steve's description suggests exasperation and frustration which he has been dealing with for many years, he copes now by having learned to accept it. However, this did not mean that Steve had resigned himself to being ill forever. He was actively seeking alternative solutions and had tried a range of alternative therapies, but he had reached a point of acceptance of his own situation and acceptance of the fact that he could not get his needs met within the NHS.

The excerpts presented suggest that a positive and accepting attitude and a willingness to be supportive is better for the PwCFS/ME, and for the relationship between the PwCFS/ME and their SO, than the experience of negativity and rejection. The process of accepting life following the changes caused by CFS/ME and rebuilding a sense of self is challenging and can be a solitary and isolating process if they do not have the support of others around them. That support often comes in the form of practical help, but a more implicit level of support that demonstrates acceptance is also crucial to the PwCFS/ME in moving forwards and rebuilding their emotional strength. For someone who is trying to cope with the challenges of disability and physical illness as well as the loss and grief these bring, a lack of acceptance from others can be stigmatising and makes an already difficult process even more challenging. Whether this is experienced at the hands of family members, friends, colleagues, health professionals, or the wider society, it only serves to isolate an already isolated individual even further. However, the final excerpt presented also illustrates how fighting with others to be believed or to get help can be an unhealthy process for PwCSF/ME. What may be better for them, and for their relationships, is to accept things as they are, and instead put energy into moving forwards in their own way.

10.4. Acceptance of the situation

Finally, and ultimately, my research suggested that PwCFS/ME may find the most comfort in developing acceptance of themselves and their situation. Richard explained that his drawing and painting had helped him to come to terms with his illness, but also highlighted why acceptance could be better than ‘fighting’ the illness and aspects of the illness experience:

> *“Yeah, it sorts of helped with the whole acceptance thing, really, and I think that’s a massive thing for people with ME, is accepting it, because at that point you’re no longer fighting against it quite so much. You accept this is what you’re like, and there are lots and lots of signposts out there towards what you can do and how you might be able to manage the condition…they’re all out there saying, you know, this might work, there are possibilities.”* (PwCFS/ME, Richard)

Sally also acknowledged that accepting and learning to live with his illness was perhaps the most important aspect of Sean being able to live a more fulfilling life moving forward. Despite the evidence I have presented that Sean’s hope seemed to be waning, Sally still had hopes for him for the future:

> *“I hope that he does accept that he has to live with this, as I feel he is starting to do, and I hope he can go onto university and I’ve tried help him with his options. I’d like to see him doing psychology or biomed, something he can manage to work at the end of but tailor at the end to cope with his illness. That’s what I’d hope, and I’ve seen a glimmer of it when he did the phlebotomy course but he really struggled with that. The day before I said no matter how rubbish you feel just do it no matter what because then you’ve got it under your belt and no one can take it away from you. And I had a real good talk with him because I thought he was going to back out, but he went and did it and I thought that was*

> *a real turning point for him because I feel now that he can think I felt horrendous when I did it but now I can do this."* (SO, Sally)

Sally is recognising here that it is possible for Sean to feel discomfort but to accept it and push through it in order to achieve the things he wants to, rather than continuing to avoid things that he feels will be too difficult or painful. This is yet another kind of acceptance, and one which Eileen also seemed to be adopting:

> *"I've learned to dowse, do you know about dowsing?...I said to this friend I just don't know what to do, I don't know what to eat, I've got to watch this, the PH balance is wrong, I mustn't eat that and I mustn't have this…and she said 'Oh I'll get you a book about dowsing and you can find out for yourself,' and it's been a Godsend, absolute Godsend. I have a pendulum…and you ask, put it over your prominent knee, which is the right knee and I asked for, it's like tuning into your sixth sense, as a child, a very small child sometimes children eat coal or sand or soil and they usually need what's in the coal or sand or soil…you get in tune with that and then I ask for a yes answer, (demonstrates) and that's my yes answer, and no answer (demonstrates) and that's my no answer, so as long as I ask a question that can have one of those answers so then I find out, so now I find out which food I can eat, which food I can put to combine, and that's been an enormous help…that made a huge difference and now that has made me, as I call it 'I'm [Eileen], I'm calm, useful and serene because of the help I get', now whether I'm tuning into my spirit, my guardian angel, whatever it might be, it's what I call my friend there, and we have quite an amusing time…so it's really, it's very warming and, it has made my life so much easier, and this is how I can be more content with my limitations and, like, when*

> *I did strain the muscles, erm, I sometimes feel as if I can't accept it, I sometimes feel 'What was the purpose of this?' and I might get a yes and I possibly would get a yes and then sometimes I think well yes but I'm suffering you know it's really painful this put up with it Eileen put up with it Eileen, you know get on with it"* (PwCFS/ME, Eileen)

Eileen had found comfort and an ability to accept through the practice of 'dowsing'. It helped her to make decisions about which she had previously felt paralysed and lost in her ability to make the right decision and lacking in any authorative guidance in what to do for the best. It removed the need to deliberate and make decisions for herself, and in doing so it enabled her to be more accepting of herself, her needs and her situation. It also enabled her to accept pain and discomfort in doing things, rather than engaging in avoidance behaviours.

10.5. Acceptance: A discussion

We see here that acceptance really can be crucial to being able to find more positive ways of coping with the illness, increasing activity, and improving the relationship with oneself and life. Acceptance, and tasks that lead to eventual acceptance, feature in all of the dominant theories of coping with loss and grief (Bowlby, 1969; Kübler-Ross, 2009; Parkes, 1972; Stroebe & Schut, 1992; Worden, 2003). I have already introduced Kübler-Ross' theory in which acceptance is defined as the final stage of grieving and adjusting to loss. In Parkes' phases of grief (1972) the four stages (Shock and Numbness; Yearning and Searching; Disorientation and Disorganisation; Reorganisation and Resolution) do not explicitly include acceptance, but the process of reorganisation and resolution is a phase during which there is light at the end of the tunnel. The individual finds new focus, energy and confidence, and although they may never get over the loss, they do manage to rebuild a meaningful and fulfilling life having moved through the worst of the grieving process. Worden's tasks of grieving (2003) also relate strongly to many of the processes discussed here and related to a global acceptance:

- Acceptance (Recognising and accepting the loss)
- Experiencing the Pain (Mourning and expressing grief)
- Adjusting (Performing new tasks that must be taken on to move forwards)
- Letting Go (Developing a new view of the future)

The task of Acceptance requires acceptance of the loss – in the context of death, accepting that the person has died. During this stage, individuals may be reluctant to let go of things belonging to the deceased individual in the vain hope that the loss might not be real. This stage varies in length according to the specific features of the relationship and the nature or circumstances surrounding the loss. Experiencing the pain requires that the individual allows himself or herself to explore the loss, talking about it, making sense of their emotional responses, and recognising the pain they are experiencing. Maladaptive coping mechanisms which aim to avoid experiencing this pain, such as use of drugs, alcohol, or over-working, can lead to the individual not moving through this stage properly, prolonging the grief and preventing them from being able to move forwards and fully process their grief. The task of Adjusting involves a process of making necessary changes in life in order to adjust to the loss of the deceased, and may require major changes such as a house move, financial rearranging, but also, and crucially to this research, the loss of a sense of identity and role, both of which are particularly relevant to the experience of PwCFS/ME and their SO. Finally, the task of 'Letting go' involves a reinvestment of energy by focusing on the growth and development of other relationships. This suggests that there may be an interactional effect between the processes of grieving, acceptance and rebuilding and finding balance in relationships.

Stroebe & Schut's (1992) Dual Process model of grief is perhaps the most current and up-to-date model of grieving and suggests that individuals experience two processes of grieving simultaneously, Loss Orientation and Restoration Orientation. During Loss Orientation, the individual may engage in expressions of grief such as anger, crying, and talking about their grief in therapy – I have already presented ample evidence in Category B. The Problem Pattern of these processes taking place. Meanwhile, the process of Restoration Orientation involves adjustment such as taking on new roles, fostering new independence and skills in order to manage the new demands in life forced upon them by their loss. So again, within this model of grief we see that there is interaction between concepts presented in Category C. Balance, and the process of acceptance, such as developing greater autonomy and learning to live meaningfully within the limitations of illness. Stroebe & Schut propose that individuals move between these two processes in daily life and that healthy grieving involves allowing oneself permission to grieve and live simultaneously, suggesting that the process of acceptance is not necessarily an end point in grief, but a work-in-progress.

I have presented a range of dimensions of acceptance evident in the data, including acceptance of the situation, acceptance from others and, perhaps most importantly, self-acceptance. In doing so, I have constructed acceptance as a key factor highlighted by participants in managing illness and feeling supported/offering appropriate support. I have illustrated how acceptance was particularly present in some relationships, and how it was lacking in some others. It may not be coincidence that those in which it was sometimes or often lacking were those that I placed at the more extreme ends of cohesion. It is therefore fair to conclude through increasing levels of acceptance in PwCFS/ME and their SOs, it may be possible to enable relationships to become more balanced. Through developing acceptance, individuals may be able to live more meaningful and contented lives, whether this is within the context of CFS/ME and the limitations it brings, or in recovery from the illness.

However, I have also noted that development of acceptance should not be entirely the responsibility of PwCFS/ME and their family members. NHS professionals have an important role to play, and their negative communications are clearly damaging for PwCFS/ME. Even in situations where individuals cannot be ‘cured’ or where treatment is limited, a positive, constructive and helpful attitude that makes the PwCFS/ME feel believed and validated is crucial in them being able to move on from fighting towards acceptance. An atmosphere of support and acceptance should surround them.

Acceptance was a concept that emerged in the data, and I did not engage with any literature around acceptance prior to or during data collection in relation to acceptance as a construct, or in CFS/ME. It is worth noting however that I have been engaging with Buddhist teachings, practising meditation and teaching sessions on mindfulness throughout the data collection and analysis period. It is therefore likely that my personal belief in the usefulness of acceptance in life generally and in mental health services greatly influenced the fact that I raised this concept up to being a core theoretical concept worthy of an entire section in this thesis, and forming a part of my main conclusions about relationships in CFS/ME. Someone without these interests may not have paid so much attention to the frequent mentions of acceptance and related concepts (perception, reflection, reframing, mindfulness).

Upon exploring acceptance as a concept of importance during the data analysis period, I of course engaged with literature and discovered that the concept of acceptance as a broad construct worthy of consideration in the context of chronic illness is not new. The role of acceptance in the management of illness has been explored in a range of conditions including depression (e.g. Dougher & Hackbert, 1994), obsessive compulsive disorder and panic disorder (e.g. Hayes, Wilson, Gifford, Follette, & Strosahl, 1996), anxiety (e.g. Roemer & Orsillo, 2006), chronic pain (e.g. McCracken, Carson, Eccleston, & Keefe, 2004), and indeed in recent years, CFS/ME (e.g. Brooks, Rimes, & Chalder, 2011). In presenting some of the most relevant debate around acceptance in CFS/ME, I will first discuss acceptance as an approach that a PwCFS/ME may take towards themselves, their illness and their condition. In some research, this might be regarded as 'self-acceptance'. I will discuss acceptance in research and self-acceptance as a construct under the heading 'Acceptance in the PwCFS/ME'. I will then move on to discuss acceptance in terms of how others think and behave towards PwCFS/ME, under the heading 'Acceptance in others'.

Acceptance in the PwCFS/ME. Rothbaum, Weisz, & Snyder (1982) posited that when control over an event becomes unattainable, attempts to accept and understand the event become the focus of attention. When acceptance and understanding are achieved, experiences of negative emotions are reduced (Rothbaum et al., 1982). McCracken et al., (2004) defined acceptance in relation to chronic pain as follows:

> *"(Acceptance is) an active willingness to engage in meaningful activities in life regardless of pain-related sensations, thoughts, and other related feelings that might otherwise hinder that engagement. It is about not engaging in unnecessary struggles with private experiences, struggles that often intensify the aversiveness of those experiences and enhance their life disrupting influences" (p. 4)*

Studies have shown that people with chronic pain experience more distress and frustration when they have a tendency to attempt to control pain that is uncontrollable (which is a lack of acceptance) (Aldrich, Eccleston, & Crombez, 2000; McCracken & Eccleston, 2003). Lack of acceptance has been found to be associated with reduced physical functioning in chronic pain (McCracken et al., 2004). It has also been found to have a negative impact on the ability of an individual to take positive action and make long-lasting changes to improve their health and wellbeing (Afrell, Biguet, & Rudebeck, 2007). Meanwhile, increased acceptance in chronic pain has been shown to be associated with improved functioning (Yu, Norton, & McCracken, 2017), psychological wellbeing and adjustment, and lower levels of psychological distress (McCracken, 1998; McCracken et al., 2004).

Self-acceptance and acceptance of others have been identified as separate concepts but have been found to be positively associated (Sheerer, 1949). Self-acceptance is a more specific aspect of acceptance and as a construct is regarded in theology (Buddhism, Christianity) to have the potential to alleviate distress (Bernard, 2013). These ideas found their way into psychological theories developed by Maslow (e.g. 1943), Rogers (e.g. 1951) and Ellis (e.g. 1962) and have since been incorporated into psychological models proposed in cognitive behaviour therapy (CBT) and third-wave therapies such as acceptance and commitment therapy (ACT), dialectical behaviour therapy (DBT) and compassion focused therapy (CFT) – for an in-depth discussion of this process, see Hoffman, Lopez, & Moats (2013).

Self-acceptance has been defined by Bernard (2013) succinctly as ‘an affirmation or acceptance of self in spite of weaknesses or deficiencies’ (p. xiv). In order to explore the concept of self-acceptance beyond this definition, it is first necessary to deconstruct the component parts of the term. We have already considered a definition of acceptance from the chronic pain literature, but it has greater meaning philosophically. Williams & Lynn (2010, pp. 8-10) defined acceptance as consisting of five dimensions:

Non-attachment:	letting objects come and go naturally without attempting to control or hold on to them.
Non-avoidance:	Remaining present and not trying to run away if there is no real physical threat
Non-judgment:	Attempting consciously to abstain from the categorisation of situations as good or bad, it just is.
Tolerance:	Staying in the present moment with full awareness even when the situation may be undesirable or unpleasant
Willingness:	Recognising the degree of choice involved in experience

Scientific consensus about what constitutes the self is lacking, but there is agreement that self is a fluid construct of thoughts, feelings, memories, characteristic traits, behaviours and body sensations (Bernard, 2013, p. xiv). Popper & Eccles, (1981) proposed that self is a theory we hold about our own existence and of who we are, whilst Baumeister & Bushman (2011) proposed self as composed of self-knowledge (self-awareness, self-concept, self-esteem, self-deception; the social self (relationships with others, social roles, group membership), and the agent self/executive function (decision-making, self-management). There is agreement that self-acceptance also refers to the tendency of humans to self-evaluate on a continuum from good to bad (Bernard, 2013).

The skill or practice of self-acceptance requires that a person works on being able to see themselves, their strengths and weaknesses, in a realistic way whilst also recognising the subjectivity of their perception of themselves. In order to truly accept the self, self-criticism must be suspended; perceived flaws or faults must also be viewed with tolerance and acceptance, and any pre-occupation with these must be halted (Shepard, 1979). This resonates with Rogers' (1951) concept of positive self-regard and was built upon further by Neff’s (2003) promotion of taking an attitude of kindness and compassion towards the self.

Gregg (2013) offers a comprehensive review of the literature around self-acceptance in chronic illness, highlighting two ways in which previous studies have approached it. The first views self-acceptance as the goal, whereby the individual works on accepting themselves as an individual with their illness, with the outcome being they have successfully accepted themselves (self-as-content). The second approach to self-acceptance seen in the literature is to consider it as a process of adjustment, whereby ongoing work towards self-acceptance offers an opportunity for improvement (self-as-process). Gregg, however, argues that there is another way: self-acceptance is not viewed as an outcome or as a process, but rather as ‘a by-product of living a vital, meaningful life’ (p.251), and in which one becomes the observer of the self, also termed ‘perspective taking’ (self-as-context).

The aim of the self-as-context approach is for the observing self to ‘notice the noticing’. An individual must be able to recognise their mind, body and experience as something observable. They must also become able to observe these aspects of themselves without judgement which is what might be achieved in a self-as-content approach, but in a self-as-context approach, one becomes able to detach the self from experience. In doing so, it becomes possible to change behaviour without requiring the alteration of thoughts and feelings, for example, recognising negative self-evaluations non-judgementally and separating the self from the self-evaluations reduces the need to avoid them. This is because by noticing experience non-judgementally, one becomes aware of one’s own perceptions, emotions, perspectives and of the possibility of multiple perspectives, so that a skill called ‘psychological flexibility’ is fostered (Gregg, 2013). Psychological flexibility involves an individual being able to be present and to acknowledge feelings, thoughts and experiences, but to act based on longer term values rather than short term impulses, and it is posited by Kashdan & Rottenberg (2010) that it is a fundamental aspect of health. In the case of chronic pain, a change in 'self-as-context' has been shown to be the mechanism through which functioning may improve in following Acceptance and Commitment Therapy (Yu et al., 2017).

Research into acceptance in CFS/ME has been limited but had steadily been gaining pace over the last 12 years. In 2006, the first study to investigate the role of acceptance in CFS/ME specifically Van Damme, Crombez, Van Houdenhove, Mariman, & Michielsen (2006) echoed findings from the chronic pain literature, demonstrating that higher levels of acceptance in PwCFS/ME was associated with increased wellbeing, decreased distress, and had a positive impact on emotional stability and levels of fatigue. A 2007 study used an imagery paradigm to investigate acceptance versus hostile resistance and found that acceptance imagery was associated with less hyperventilation in PwCFS/ME than imagery of hostile resistance (Bogaerts et al., 2007). In their qualitative study of the experience of living with CFS/ME from the perspective of the individuals themselves, Dickson, Knussen & Flowers highlighted acceptance as central to adjustment in life with CFS/ME (2008). Brooks et al. (2011) deepened the exploration further by investigating a possible relationship between acceptance and perfectionism in CFS/ME, suggesting that PwCFS/ME with higher levels of perfectionism may have greater intolerance for symptoms, thus being less accepting. Neuroticism and perfectionism had previously received attention in the CFS/ME literature (e.g. Besharat, Behpajooh, Poursharifi, & Zarani, 2011; Kempke et al., 2013; Sáez-Francàs et al., 2014) , and research into other conditions had previously demonstrated links between perfectionism and acceptance. For example Flett, Besser, Davis, & Hewitt (2003) found that the three trait dimensions of perfectionism, defined as self-oriented, other-oriented, and socially prescribed perfectionism, were negatively associated with unconditional self-acceptance, and that depression was also linked to low levels of unconditional self-acceptance. The researchers concluded that "perfectionists evaluate themselves in terms of a contingent sense of self-worth, and as such, they are vulnerable to psychological distress when they experience negative events that do not affirm their self-worth" (P.119). Further, Scott (2007) found that perfectionism leads to low levels of unconditional self-acceptance and therefore to depression.

Perfectionism can be viewed as consisting of two factors, 'Personal standards', whereby one strives to achieve high standards, and 'Evaluative concerns', which refers to the tendency to be self-doubting and self-critical (Dunkley, Blankstein, Masheb, & Grilo, 2006). It has been proposed that there are two types of perfectionism: positive perfectionism which is considered normal and healthy, and negative perfectionism which is pathological (Slade & Owens, 1998). Studies have linked CFS/ME to negative perfectionism (Deary & Chalder, 2010; Kempke et al., 2011; White & Schweitzer, 2000), and self-critical perfectionism has been shown to be associated with sensitivity to stress and depression in CFS/ME (Luyten et al., 2011) and increased fatigue and pain symptoms (Kempke et al., 2013). These studies led to recommendations that interventions for CFS/ME should aim to target self-critical perfectionism. The findings of Brooks et al. (2011) suggested that lack of acceptance was associated with perfectionism in CFS/ME, as well as with poorer levels of physical functioning, work and social adjustment, and fatigue. Following treatment with CBT targeting unhelpful perfectionist beliefs, acceptance levels were found to have increased, correlating with improved work and social adjustment. The authors concluded that acceptance may be an important factor worthy of further investigation in CFS/ME.

Poppe, Petrovic, Vogelaers, & Crombez (2013) investigated whether pre-treatment levels of acceptance and neuroticism in CFS/ME were related to any changes in quality of life following CBT intervention in a group setting. They found that high neuroticism or low acceptance pre-treatment correlated with greater improvement in quality of life following group CBT treatment (although effect sizes were low). Therefore, they recommended that individuals should engage in treatment that targets acceptance and neuroticism prior to CBT in order to maximise the effectiveness of CBT interventions in CFS/ME. A recent case series study trialing a six-week self-help Acceptance and Commitment Therapy intervention for six PwCFS/ME yielded promising results. Individuals' pursuit of values and activity levels were improved, but it was concluded that effects were likely to be short-term unless further intervention was available (Roche, Dawson, Moghaddam, Abey, & Gresswell, 2017).

Given the recent move in CFS/ME research towards a focus on acceptance, it is useful to note that none of the participants in this study had engaged in ACT. Some had engaged with CBT and counselling, and some with mindfulness and Buddhist teachings. Yet most of them recognised the central role of acceptance in their ability to adjust to their illness. If these individuals rate highly in traits of perfectionism, it is likely that their default position in regard to unconditional self-acceptance was one of very low levels, and so self-acceptance must be learned and practised. Self-acceptance emerged as a crucial factor for participants moving from a place of suffering and fighting against their illness into a more peaceful place of accepting their illness so that they could begin to move forwards. Through developing acceptance, individuals with CFS/ME may be able to come to terms with their losses, feel less distressed by negative responses from others, and be less vulnerable to negative emotions and subsequent behavioural cycles.

Yet acceptance also has the capacity to help people challenge themselves more by accepting pain and discomfort, thereby reducing behavioural avoidance and potentially increasing activity levels. McCracken (1998)found that when greater acceptance of pain was reported, pain intensity was reported to be lower, and pain-related anxiety, avoidance, depression, physical and psychosocial disability were all reported to be lower. It was also associated with improved outcomes related to daily activity and work life. Pain acceptance also predicted improved adjustment on all other functional measures, regardless of perceived pain intensity. Therefore, acceptance is not the same as resignation and does not mean that an individual gives up trying. Instead, it facilitates improved coping with discomfort, rather than avoiding it altogether, and could therefore be key in breaking free from the cycle of avoidance and deconditioning that can occur in CFS/ME, as discussed in Category B. The Problem Pattern.

Acceptance in others. As we have already seen, previous research suggests that spousal perspectives of illness have influence over the way a person responds to illness (Moss-Morris & Petrie, 2000). The excerpts presented in this research provide supporting evidence that this is indeed the case in CFS/ME, and that acceptance may also be a crucial factor in improving relationships and responses. This research offers insights into aspects of relationships, approaches to and responses to the illness and to one another that PwCFS/ME find unhelpful and helpful. Responses from others that are perceived by PwCFS/ME as helpful are clearly linked in the data to improved perceptions of their ability to cope with illness, to move forwards, and to find joy and meaning in life and relationships.

However, the rhetoric in previous research in CFS/ME has been somewhat lacking in terms of defining what helps where spouses and families are concerned, instead focusing predominantly on what is not helpful. Based on findings that PwCFS/ME who report greater relationship satisfaction also report lower activity and higher fatigue levels (Schmaling & DiClementi, 1995), Moss-Morris and Petrie (2000) argued it possible that a supportive partner in CFS/ME ‘unintentionally colludes with patients in maintaining their disability’ (p.104). Moss-Morris and Petrie further supported their argument by citing research findings that PwCFS/ME who reported differing illness beliefs to their spouses also reported less marital satisfaction (Heijmans, Ridder, & Bensing, 1999), concluding that relationship quality may be mediated by concordance of illness beliefs (Moss-Morris & Petrie, 2000). This argument was supported by findings suggesting that partners' solicitous responses and illness perceptions were associated with poorer fatigue and disability outcomes following CBT for PwCFS/ME, and that relationship dissatisfaction was negatively associated with improved outcomes in fatigue (Verspaandonk et al., 2015).

Whilst previous research suggests that a spouse who accepts or agrees with a person's illness beliefs is validating those beliefs, my findings in this research leave me concerned about the direction of previous research in terms of SO relationships in CFS/ME. Previous research findings seem to indirectly suggest that in managing and recovering from their illness, PwCFS/ME may benefit more from an SO who does not agree with their perception of their illness and instead challenges them. However, I argue that if SOs help PwCFS/ME to feel accepted and supported, they may have a more fulfilling relationship, and this in turn is likely to have a positive impact on their ability to cope with their illness and on their subjective wellbeing. A tendency of many studies in CFS/ME historically has been to evaluate the impact of interventions or responses from others based on measures of fatigue and other physical symptoms. However, more attention needs to be paid to the concept of living well with the illness, and to the subjective perspectives of PwCFS/ME and their SOs in terms of what they feel improves their lives and helps them to cope. Dickson et al. (2007) usefully highlighted two inter-related themes following interviews with 14 PwCFS/ME regarding their experience of living with CFS/ME: "Negotiating a diagnosis" and "Negotiating CFS with loved ones". This work highlighted the fact that although PwCFS/ME found their experiences around obtaining diagnosis and treatment from their GPs to be negative (GPs are cited as being 'sceptical, disrespectful, and to be lacking in knowledge and interpersonal skills' (p.851)), PwCFS/ME found their relationships and encounters with partners were often deligitimising and that this was harder to cope with than the negative experiences with healthcare professionals. Delegitimation by those closest to them was experienced by PwCFS/ME as a hurtful form of personal rejection. All of the evidence I have presented in this research suggests that having an appropriately supportive, positive and accepting relationship with an SO is of great value and meaning to a PwCFS/ME and that this positively impacts upon their wellbeing. Excerpts suggested that individuals who felt pushed by others, or made to feel that others do not support them, felt more resistance towards others and tended to disengage from them. This further isolated already socially isolated people. In order for an individual to feel they are capable of regaining physical strength, taking small steps towards increasing their activity, thereby improving their quality of life, they need emotional strength and they need to feel supported and understood by those around them. This approach of support and acceptance must also be afforded to PwCFS/ME by health professionals. The evidence is clear: PwCFS/ME encounter a great deal of stigmatising negative responses from many health professionals with whom they interact, and it seems difficult for them to make positive steps forward when under the care of a GP they perceive to be lacking in understanding. An entirely new holistic approach towards working with PwCFS/ME that is at once empathic and supportive, is needed. PwCFS/ME must be afforded the same sympathy and support as those with any other chronic physical or mental health problems. PwCFS/ME do appear to benefit from psychosocial interventions, but unless they have a firm foundation of perceived social support on which to rebuild their strength, it is less likely they will engage in or adhere to treatment. My findings suggest that PwCFS/ME benefit from feeling believed and accepted so that they can begin to move towards behaviour change and gradual alterations to their perception of illness with positivity. Negativity from those around them is not conducive to their being able to regain a sense of control over their illness and therefore serves to maintain their levels of disability, weaken their relationships and increase their sense of isolation and social anxieties.

Acceptance: Conclusion. I have presented participant excerpts that highlight acceptance as crucial in the process of rebuilding life following a period of illness with CFS/ME. This acceptance is something that the PwCFS/ME needs to nurture within himself or herself via a process of reflection in order to come to terms with loss. Acceptance has also been highlighted as a helpful characteristic of relationships with others, including partners, parents and health professionals. Recent research in CFS/ME has started to investigate acceptance as a potential mechanism for change in CFS/ME. Although research suggests that quality of life can be improved by reducing perfectionism and increasing acceptance in CFS/ME through treatment with CBT, recommendations in recent research have suggested that interventions that target acceptance specifically may be beneficial. I would argue that these interventions could usefully be offered to both the PwCFS/ME and their SO(s). There may be a possibility that through the development of acceptance, relationships can be improved, and quality of life enhanced. It is when living in a more positive state of mind and with purpose in their lives that PwCFS/ME can find ways to live meaningfully within the limitations of this illness.

11. Final Discussion

11.1. Research question and aims

This study sought to investigate how PwCFS/ME and their SOs experience their relationship in the context of CFS/ME. Data was gathered and analysed in line with grounded theory methodology, and a grounded theory was built and has been presented and explained here. I will first address the overarching research question, followed by each of the three aims that were set out.

11.1.1. Research question

How do people with CFS/ME and their significant others experience their relationship in the context of illness?

This thesis offers an in-depth account of relations and emotions in the context of CFS/ME for a range of relationship types, comparing and contrasting them with one another to offer an overarching theory of what helps and what does not in terms of family processes and responses to CFS/ME. In brief, what helps is acceptance and a balance of togetherness and autonomy. What does not help is enmeshment (or overinvolvement), and disengagement as a consequence of negative responses.

11.1.2. Research aims 1 & 2

1. To develop a more nuanced understanding and explanation of the difficulties in relationships between PwCFS/ME and their SO in the context of CFS/ME than currently offered in the literature.

2. To describe in detail the emotional processes that may be underlying previous findings in research regarding SO responses.

This study explored narratives of PwCFS/ME and their SO and in doing so, it is a novel piece of work. Further, because the sample was broad, offering a range of ages and relationship types, the findings are of relevance to a broad population. The grounded theory approach yielded rich data, as well as flexibility and theoretical sensitivity in the analytic process. This has enabled an in-depth exploration of relationships in terms of the emotions, behaviours and dynamic interactions between PwCFS/ME and their SOs in the context of CFS/ME.

Through a process of reflexivity, I recognised that the concepts I was constructing tended to focus on the more negative and difficult aspects of experience, despite the fact that I had included questions about what worked and what helped in relationships in the interview topic guide. This also mirrored the tendency of participants to focus on the negative aspects. I consequently posed further questions during the analytic process that required me to interrogate the data further in order to identify aspects of broader experiences within the context of CFS/ME.

11.1.3. Research aim 3

To develop a model that explains the characteristics, processes and consequences of relationships between PwCFS/ME and their SO in the context of CFS/ME.

The model presented as the overarching theory behind this thesis (Figure 1, p.88) meets this research aim by offering explanations of causes in Category A. Onset and within several concepts of Category B. Core Category: The Problem Pattern. Across the Problem Pattern and within Category C. Balance, I have presented a detailed description and analysis of the characteristics of relationships. This research builds on existing theory and research findings. For example, similar excerpts and interpretations were found post-analysis in studies investigating self-perceived burden in terminal cancer (McPherson et al., 2007) identity crisis, loss and adjustment in CFS/ME (Dickson et al., 2008) and the family life-cycle in CFS/ME (Sperry, 2012). The results of this study also fit well with previous findings of Band et al. (2014), Ax (1999) and Ax et al. (2002), extending the work previously undertaken by using an inductive and in-depth approach to obtaining rich data from a variety of sources, both PwCFS/ME and their SOs, with a range of relationship types and focus on building theory.

The process of theory-building by relating concepts to one another was pivotal in the construction of the third category, Balance. Through comparison of the concepts that eventually formed the category of Balance to concepts in the core category, The Problem Pattern, I was able to relate these to the relational qualities that had been identified – enmeshment and disengagement (which started out as ‘withdrawal and social isolation’). In doing so, and at the same time as engaging with the theories to which my data had directed me, I identified the relevance of the concepts that had been evolving – e.g. togetherness, love, sense of self – to the theoretical construct of cohesion and finding balance in that cohesion. Through the application of a simplified version of Olson’s model (Olson et al., 1979), I also saw how these were related to enmeshment, disengagement, and the difficult emotional experiences that these engendered. Although this aspect of my theory is not novel as it applies a previously developed theory, it does offer a new way of looking at relationships in the context of CFS/ME. It also helped me to recognise how this could be seen to overlay aspects of my broader model (Figure 1. page 88) which offers balance in relationships and acceptance as exits from the vicious cycle of the pattern described in the core category, The Problem Pattern.

11.2. Locating the research contextually

In Chapter 1, I have explained the approach taken during the research towards literature reviews, which was predominantly guided by the constructivist approach to grounded theory (Charmaz, 2014). This approach involved undertaking a literature review to understand the historical context of CFS/ME, exploring issues such as the development of our understanding of the illness, the label, and a description of some of the most prominent past and present theories around causes and treatments. It also involved undertaking a scoping literature review in order to understand where the knowledge gaps existed and to refine the research question and aims prior to data collection. Following this, the research data was prioritised, and further engagement with the literature was guided by conceptualisation and theory-building. In reporting the findings, I have presented in-depth discussion of each concept, relating the findings to literature. However, what has not yet been provided is a discussion of the overarching findings in relation to the broader CFS/ME literature or the most recently published research in CFS/ME.

It is recognised within the tradition of constructivist grounded theory that any theory has been co-created by the researcher and the research participants bears the 'imprint' of the interests and ideas of the author whilst also reflecting the historical context and development of research and ideas (Charmaz, 2014). This piece of work is located in British society where healthcare is publicly funded and is free at the point of need. An ageing and growing population places the current system in England under immense financial pressure as demand increases and funding is not aligned with need, leading to estimates in 2013 of a £20 billion funding gap by 2020; attempts to close this gap led to government demands for 'efficiency' and cuts to services across the country (Full Fact, 2018). Mental health services are currently separate organisations to acute care and primary care services (GPs) and as such this could be seen to communicate to the public an arbitrary separation of mind and body, mental and physical. Within this system, I have witnessed illnesses which cannot be 'pinned down' to specific biomarkers frequently labelled 'functional' or 'medically unexplained', and viewed as psychosomatic. This places such illnesses in the domain of mental illness, with CFS/ME being one such illness.

To attempt to separate CFS/ME from the perception of this illness in the medical professions would be to ignore the influence and impact of these perceptions on those who have this illness. It is equally difficult to study any aspect of CFS/ME without regard for the current medicalised view of illness in general, and CFS/ME specifically. Further, the medicalised view cannot be separated from the political climate that has led to the current cash-strapped NHS in which parity of esteem between mental and physical healthcare is an ideal, not a reality. In the research process, I had to work hard to remain focused on my research question because a dominant concept across the data was the difficulties individuals had experienced with health services and health professionals. Indeed, there is an entire thesis-worth of data relating to this issue. Although this data did not provide answers to the research question, it must be noted that the experience of the relationship between PwCFS/ME and their SO is deeply affected by the context within which it exists. The difficulties faced by couples in their interactions with outside others have already been highlighted by Brooks, King and Wearden (2014). I have presented further evidence that these challenges can serve to isolate them as an individual, dyad and family, can give rise to anger and frustration, and can even contribute to family members questioning the PwCFS/ME, to their detriment. Participant extracts suggest that challenging relationships with health professionals or difficulties with accessibility of treatment can leave individuals feeling reluctant to engage with any services that might be offered. This, in turn, can lead to feelings that there is nothing that can be done, and to a gradual loss of hope and feelings of helplessness for the dyad. I have also demonstrated that in this situation, the responses of family members become particularly important in whether the PwCFS/ME is able to maintain a positive attitude towards making improvements.

These perspectives are supported by the findings of a report presented to the Chief Medical Officer (CMO) in 2002 (A Report of the CFS/ME Working Group, 2002). In 1998, the CMO commissioned a working group whose task it was to conduct research with the aim of producing a report that would advise on best practice guidelines for the care and treatment of CFS/ME. My decision to include this discussion of the CMO report at this point rather than in Chapter 3 is guided by the fact that I came to this report after completion of the analysis and writing-up – I did not come across this document in my earlier literature reviews. It is therefore fascinating that many of the findings are strikingly similar to mine. Published in 2002, the report draws on quantitative and qualitative data, including the perspectives of PwCFS/ME and their carers, to tackle a number of issues including diagnosis, treatments and future research directions. The report clearly highlights the need amongst PwCFS/ME for recognition, a clear diagnosis, acceptance and acknowledgement and for carers to receive greater recognition, support and respite. It emphasises the importance of early diagnosis, information and advice, and that PwCFS/ME should be the key decision-maker regarding their own care. The working group noted the problems faced by PwCFS/ME due to a lack of understanding and acceptance from healthcare professionals and the general public and the experience of being disbelieved, suggesting that education and training for medical professionals in relation to CFS/ME was insufficient. It also highlighted that this disbelief makes the situation even more challenging for carers who find the ignorance of GPs particularly distressing. It makes clear that GPs failing to take action due to ignorance or denial of CFS/ME is not acceptable, and stresses the importance of a positive therapeutic relationship and an understanding attitude, which are more likely to lead to more successful outcomes.

The report has been criticised by Straus (2002) for being too heavily influenced by patient perspectives, which some consider ‘anecdote’, and not enough by quantitative data and experimental research. Straus also criticised the lack of a detailed research plan, but commended the articulation of issues of belief and trust – it states that PwCFS/ME do not feel believed and they do not trust the medical establishment to meet their needs adequately. Sharpe (2002) stated that whilst the report addressed the key issues and highlighted the needs of PwCFS/ME and a wider population with similar symptoms, it failed to offer resolutions and gave rise to more questions that it had successfully answered.

Had I read this report prior to or during data collection, it would have been a natural conclusion to assume that my findings were largely influenced by the contents of this report. The fact that I had not read this report and still the findings are so similar to my findings suggests a number of things. Firstly, that my findings do reflect the sentiments expressed by a much broader population of PwCFS/ME and their SOs; secondly, that my conclusions echo the conclusions reached by other researchers in response to such data; and thirdly, as the CMO report was published in 2002, there is a very real possibility that things really have not moved on much for PwCFS/ME and their SOs since then in terms of the quality of care received – certainly my findings seem to suggest so. However, it is interesting to note that since 2002 there has been a growing body of research exploring significant other perspectives and responses (e.g. Dickson et al., 2007; Dickson et al, 2008; Drachler et al., 2009; Band et al., 2014; Brooks et al., 2014), of which this thesis now becomes a part.

The argument that my findings are aligned with the perspectives of a much broader population of PwCFS/ME and the conclusions of other researchers' engagement with such data is further supported by the findings of a recent qualitative study. Williams, Christopher & Jenkinson (2019) explored the psychological impact of dependency among adults with CFS/ME which generated six themes: *loss of independence and self-identity, an invisible illness, anxieties of today and the future, catch-22, internalised anger*, and *acceptance of the condition.* These themes echo several concepts and categories highlighted within my study, and it is particularly interesting that dependence, anger, anxiety and acceptance feature so prominently in both Williams et al. and my study. Much like my own conclusions, Williams et al. suggest improved education for families, friends and professionals and future interventions aimed at developing acceptance which they believed would be likely to improve psychological wellbeing.

Another important development over recent years that adds weight to the argument for the consideration of ACT as a possible treatment in CFS/ME is the controversy surrounding the PACE trial (White et al, 2011). There have been many papers published criticising the methodology, data management, findings and even the integrity of the authors. For example, McGrath (2015) called into question the use of a causal mediation model for this data set and the amount of missing data for data groups within the trial. McGrath claimed that the design of the trial meant that only associations, not causal links, could be established, and therefore that the claims about causal mediators and the associated theoretical treatment model are not sufficiently supported. Meanwhile, Courtney (2015) criticised the objective outcome measures and subjective self-report measures used in the trial and suggested that the results of these outcomes do not support the fear avoidance model or the deconditioning theory that underpin the use of CBT to treat CFS/ME. Courtney also highlighted the risk that results may have been due to bias or placebo effects because it was an open-label trial, which is likely to have affected the expectations of the groups. The authors responded with a statement (Chalder, Goldsmith, White, Sharpe & Pickles, 2015) defending their approach, methodology and findings, claiming that there is an ‘apparent campaign to bring the robust findings of the trial into question’, and stating that the criticisms of the report simply repeat limitations highlighted by the authors themselves in the original research, and fail to acknowledge the value of subjective outcome measures when dealing with a subjective illness experience.

Yet, the controversy persisted. Following a five-year campaign, an independent tribunal ordered the lead author’s institution to honour a Freedom of Information request from a PwCFS/ME by releasing the data from the PACE trial. When re-analysed

(Wilshire, Kindlon, Courtney, Matthees, Tuller, et al., 2018), improvement levels and recovery reported in the original paper (White et al., 2011), were called into question, with effectiveness of CBT and GET falling by almost two-thirds when compared with standard medical care and APT. Particularly damning was Kirby's (2015) statement that there is 'growing recognition that results from the PACE trial have been presented in a way which has exaggerated the benefits of CBT and GET to patients' (e.10) and the suggestion that this may have been due to the authors' concerns about their professional and international status. Had this data not been re-analysed, PwCFS/ME and healthcare professionals would have had to work on the basis that CBT and GET offer the most effective treatments for CFS/ME. Yet, the authors remain steadfast, arguing recently that despite the criticisms and re-analysis, there is no convincing reason to discount the conclusions of the PACE trial (Sharpe, Goldsmith & Chalder, 2019).

The controversies surrounding the PACE trial make clear that further research into effective treatments for CFS/ME are necessary. Recent research has sought to generate further evidence around the use of CBT for CFS/ME in community mental health settings (e.g. Janse, Bleijenberg & Knoop, 2019; Janse, van Dam, Pijpers, Wiborg, Bleijenberg et al., 2019) and has explored the measurement and definition of recovery in CFS/ME (Devendorf, Jackson, Sunnquist, & Jason, 2019). I have suggested that ACT should be considered for PwCFS/ME and their significant others. A recent feasibility study offered promising results for ACT; Jonsjö, Wicksell, Holmström, Andreasson & Olsson (2019) trialled ACT for 40 adults with a diagnosis of CFS/ME who engaged in 13 sessions of ACT treatment. Data was collected pre-, mid-, and post-treatment, and at follow-ups at three and six months. Results showed an improvement in disability and psychological flexibility between pre and post-treatment, and this was maintained at follow ups. However, this research, along with the PACE trial, fails to acknowledge the role of significant others and wider family systems. Whether or not the findings of the PACE trial are indeed robust, there is significant evidence now available of the role of significant others in CFS/ME, and my research supports the argument that interventions should consider the role and needs of significant others for maximum impact. Psychosocial interventions that target only the PwCFS/ME are likely to yield only limited results. What is needed is a systemic approach that adapts ACT for use with both PwCFS/ME and their families.

Of course, as a researcher and a Grounded Theorist, I must recognise my own biases here. As a psychologist, I have trained to think and analyse from a biopsychosocial perspective. I have outlined my leaning towards systemic theory, constructivism, interpretivism and my regard for positivism as useful but not able to tell the whole story. These factors influence my decisions to highlight my interpretation of the current treatment of CFS/ME in healthcare, and even my use of the term ‘medicalisation’. I am influenced by the work I have undertaken in mental health services and by the individuals with whom I have worked in my clinical role as an Assistant Psychologist and in my role as a researcher, and by my belief in family systems theory. I do not believe that an individual can be separated from the systems within which they function, and this belief has been pivotal in shaping my research questions, aims, interview questions and my interpretation of the data.

11.3. Limitations

PwCFS/ME recruited for this study had a self-reported diagnosis of CFS/ME. Because of the subjective nature of CFS/ME, no further diagnostic checks were required prior to participation, and no checks were made as to which diagnostic criteria participants believed they had been diagnosed against. Although all participants with CFS/ME stated that they had been diagnosed by a health professional, there remains the possibility that they may have self-diagnosed.

Although the intention was to employ the purposive sampling method of maximum variation and the study sample is broad with a range of ages and relationships types, the sample is a convenience sample recruited on the basis of availability within the timeframe of the PhD. The sample was self-selected, and many came from a local CFS/ME support group that I discovered, following data collection, may be particularly hostile to the NHS – few of them had engaged successfully with NHS services, although perhaps this was due to services repeatedly failing to meet their needs. I attempted to balance this by engaging with the local CFS service, but was unsuccessful in doing so. Consequently, the sample shared similar views regarding the origins of their illness and the adequacy of healthcare services, and for this reason the sample may have a degree of bias. One individual within the group had a particular influence over who was contacted to participate in the study acting in a gatekeeping role at times, and so it is possible that she may have selected individuals whom she believed would present an image or attitude that she felt was appropriate for the research.

Although the range of dyadic relationships explored in the study was broad, no mother/daughter relationship was included, and similarly no dyad where the PwCFS/ME was a parent and the SO was their son or daughter. Inclusion of individuals who had divorced partners in the context of CFS/ME may also have offered further interesting comparisons, as would further examples of dyads where the PwCFS/ME had made a full recovery. However, as theoretical saturation had been reached, it was no longer necessary to continue collecting data. With 15 participants, it is not a large sample, although in grounded theory there is no stipulation regarding sample size as a researcher is to be guided by theoretical development and saturation rather than by numbers of participants (Charmaz, 2014). Because each participant completed two interviews, the volume of data meant that pushing for many more participants may have made the workload unmanageable within the scope of this project.

A further limitation of this study is the lack of ethnic diversity in the area in which the research took place, which is reflected in the fact that all participants were white British. This means that the findings may only be of relevance to white British populations, as the literature search I conducted did not reveal any studies regarding cultural factors in patterns of relationships and social norms in CFS/ME, although this is an avenue worthy of further research.

A real challenge for me to overcome in writing this thesis about human attitudes and behaviours was the need to write a coherent linear document and describe discreet elements of processes as if these are how the phenomena exist. I view human experience and the way individuals attempt to understand and describe it as inherently messy, inconsistent and vague. I believe that as humans, we apply meaning to our experiences that can change depending on factors such as our mood, audience, recent readings or experiences, and whether we have slept well and/or eaten properly. My research participants may have analysed the past based on how they were feeling in the present rather than based on what actually happened, and when they were communicating their experience to me, present affect and knowledge likely influenced the way they explained their past experiences. For the purpose of this thesis, I have attempted to define elements of the experience of the research participants, to put them into boxes, and to assign meaning that is rooted in participant experiences and narratives but can be linked to theory. And yet life, experience, meaning, theory and our reflections are co-constructed and changeable. What we have in this thesis is not a description of how PwCFS/ME and their SOs experience their relationships in the context of CFS/ME, but my description of how I have ordered and made sense of my interpretations of their experience. Together with the participants, we co-constructed their description of their current views of their experiences. Those views are likely to change over time; certainly, those with a tendency to be more self-reflective or who engage in psychological therapies may be likely to change their view of their experience with the passage of time. If I was to return to them in five years and ask exactly the same questions, the responses may be similar but would not be exactly the same, and my responses would be similar, but not exactly the same. Therefore, the findings of this research should be viewed as subjective explanations and interpretations constructed within a specific context, at a specific time, by specific individuals.

11.4. Implications & recommendations

Clinical implications. I have highlighted existing research that is already beginning to explore the influence of SOs and the role of acceptance in CFS/ME. My research confirms that these are lines of enquiry worthy of further research and exploration within clinical settings, but it also lends credibility to the idea of tying the concepts of acceptance and SO responses together: acceptance emerges as a crucial factor in learning to live meaningfully with CFS/ME for both the PwCFS/ME and their SO, and as having the potential to improve the ways in which they relate to one another. Viewing the PwCFS/ME as purely an individual to be treated independently is to ignore years of systemic research and theory, and is likely to severely limit the potential for positive outcomes. I have argued that the key to re-establishing quality of life in the context of CFS/ME is finding balance in relationships through the development of autonomy and togetherness, and acceptance – acceptance of the self, of others, from others, and of the situation. Acceptance can be fostered through cognitive behavioural treatment that has a particular focus on acceptance, and as such, third wave cognitive behavioural therapies such as ACT (Hayes, Strosahl, & Wilson, 2003) may be better placed to achieve this than more traditional models of CBT such as those described by Beck (1993). I have presented literature which demonstrates that ACT is starting to gain pace as a viable treatment for chronic pain, and for a range of other chronic illnesses, and is also beginning to emerge as an avenue worthy of consideration in CFS/ME; in recent years it has been trialed for use with people with CFS/ME (for example, Roche et al., 2017). However, the findings of my research suggest that treating only the individual is likely to have limited results, whether the intervention is CBT, ACT, or any other therapeutic model. A more systemic and relationship-centred approach to care that involves SOs and possibly other family members may be more beneficial. In this way, acceptance as a broader approach to the self and to life can be fostered in the family home, and could lead to improved outcomes for the individual, for the dyad, and for the wider family system.

Focusing on symptom improvement as the main goal/outcome measures in CFS/ME treatment and research may also be limiting; rather than seeking to reduce symptoms, a better avenue for both treatment and research at this stage in our understanding of CFS/ME may be to seek to improve subjective wellbeing. Supporting PwCFS/ME to fully experience and actively seek social support, love, togetherness, enjoyment, joy, and ultimately to do all of this whilst accepting the reality of their experience of illness may have the potential to yield more positive results than simply aiming to reduce fatigue and other physical symptoms.

Recommendations for future research. Future research should consider the potential efficacy and positive impact of ACT for both PwCFS/ME and their SO. A further avenue worthy of exploration is the potential for acceptance training for GPs in supporting PwCFS/ME to have a positive impact on doctor-patient relationships and therefore on clinical outcomes. ACT offered to the PwCFS/ME and their SO by the NHS is only likely to be accepted by PwCFS/ME if it is offered by an NHS professional who has also demonstrated an attitude of acceptance towards the individual. My research supports previous findings that a lack of acceptance and appropriate support from NHS professionals can be extremely damaging for PwCFS, making it less likely that they will engage with interventions that are subsequently offered by the NHS.

11.5. Conclusion

Following recent findings that PwCFS/ME are affected by the responses of their SO, I set about exploring these relationships and experiences further. My aim was to understand the processes and emotions that may be underlying recent findings, such as the negative impact of solicitous responses and/or emotional overinvolvement, negative responses and/or critical comments. I have presented a model for understanding how PwCFS/ME and their SO experience their relationship in the context of CFS/ME; the challenges of managing the condition and the relationship. Doing too much is constructed as being related to enmeshment in relationships, whilst doing too little is related to disengagement – social isolation and withdrawal. I argue here that, based on Olson et al.'s model (1979), these are the extremities of cohesion, and more positive outcomes can be achieved through a shift into the middle-ground of cohesion. I have argued that this requires a balance of togetherness and autonomy, and that reframing through opportunities for reflection, such as through counselling, can be helpful in developing a more positive approach to illness and relationships, so that the positive elements of life can be nurtured. However, the most useful approach for the PwCFS/ME and their SO emerges as acceptance. My findings suggest that where the PwCFS/ME and their SO are able to come to terms with illness, to accept its impact on life and experience, and yet to make attempts to move forwards, more positive outcomes may be achieved.

12. References

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13. Appendices

Appendix 1. Informed Consent

UNIVERSITY OF Hull

CONSENT FORM **Date of Issue:**

Title: How do patients and their significant other experience their relationship in response to a diagnosis of chronic fatigue syndrome/myalgic encephalomyelitis?

Investigators: Katie Oxtoby
Department of Psychology, University of Hull, Cottingham Road, Hull HU6 7RX
Email: k.j.oxtoby@2014.hull.ac.uk Tel: 07872 388844

The participant should complete the whole of this sheet himself/herself. Please cross out or delete as necessary:

☐ Have you read and understood the participant information sheet?	YES/NO
☐ Have you had the opportunity to ask questions and discuss the study?	YES/NO
☐ Have all the questions been answered satisfactorily?	YES/NO
☐ Have you received enough information about the study?	YES/NO
☐ Do you understand that you are free to withdraw from the study at any time until 24 hours following the final interview without having to give a reason?	YES/NO
☐ Do you agree to take part in the study?	YES/NO

This study has been explained to me to my satisfaction, and I agree to take part. I understand that I am free to withdraw at any time.

Signature of the Participant. . . . .

Date. . . . .

Name (in block capitals) . . . .

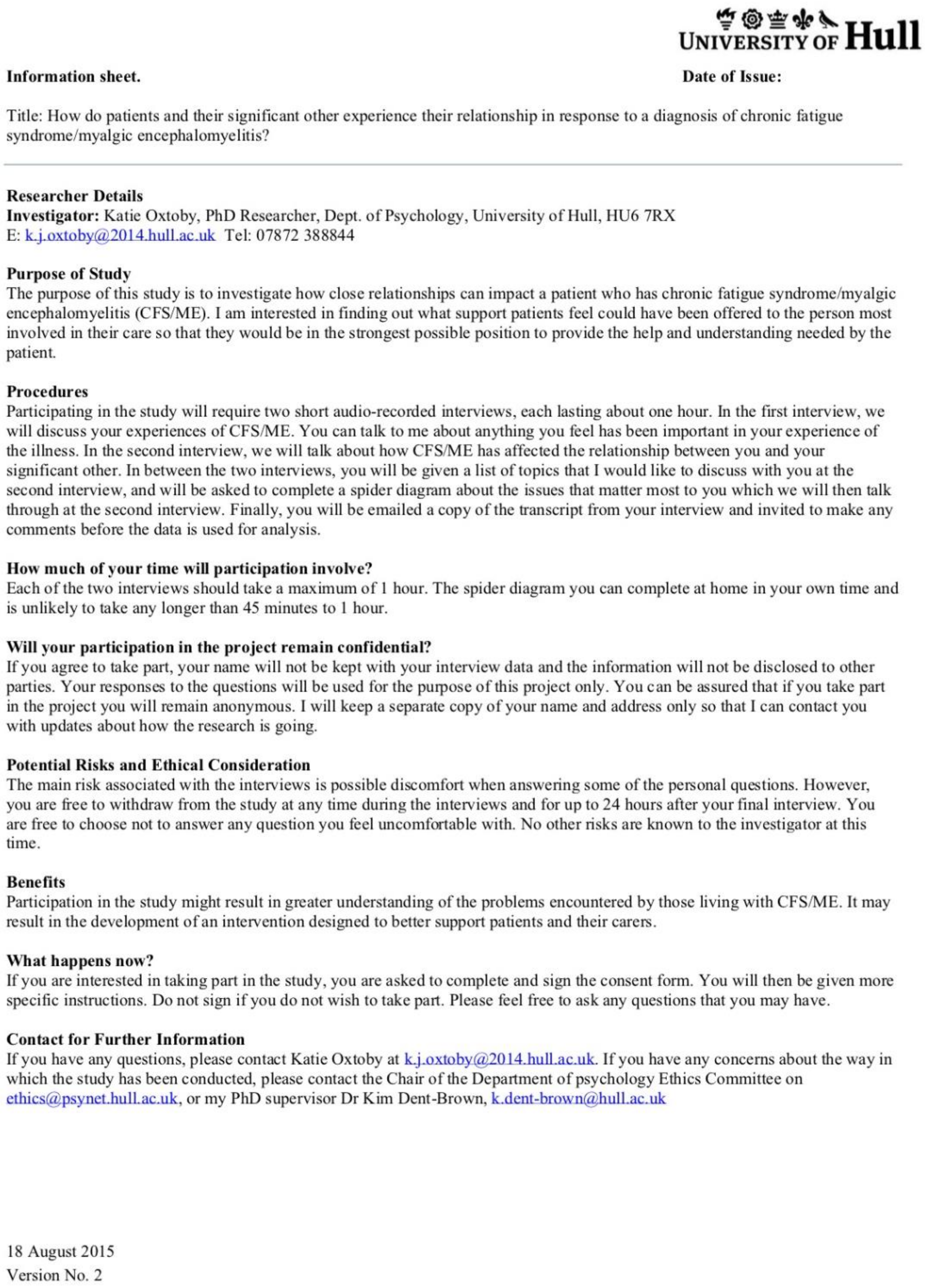
I have explained the study to the above participant and he/she has agreed to take part.

Signature of researcher . . . .

Date. . . . .

18 August 2015
Version No. 1

Appendix 2. Participant Information & Debrief Sheets



UNIVERSITY OF Hull

Information sheet. **Date of Issue:**

Title: How do patients and their significant other experience their relationship in response to a diagnosis of chronic fatigue syndrome/myalgic encephalomyelitis?

Researcher Details
Investigator: Katie Oxtoby, PhD Researcher, Dept. of Psychology, University of Hull, HU6 7RX
E: k.j.oxtoby@2014.hull.ac.uk Tel: 07872 388844

Purpose of Study
The purpose of this study is to investigate how close relationships can impact a patient who has chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME). I am interested in finding out what support patients feel could have been offered to the person most involved in their care so that they would be in the strongest possible position to provide the help and understanding needed by the patient.

Procedures
Participating in the study will require two short audio-recorded interviews, each lasting about one hour. In the first interview, we will discuss your experiences of CFS/ME. You can talk to me about anything you feel has been important in your experience of the illness. In the second interview, we will talk about how CFS/ME has affected the relationship between you and your significant other. In between the two interviews, you will be given a list of topics that I would like to discuss with you at the second interview, and will be asked to complete a spider diagram about the issues that matter most to you which we will then talk through at the second interview. Finally, you will be emailed a copy of the transcript from your interview and invited to make any comments before the data is used for analysis.

How much of your time will participation involve?
Each of the two interviews should take a maximum of 1 hour. The spider diagram you can complete at home in your own time and is unlikely to take any longer than 45 minutes to 1 hour.

Will your participation in the project remain confidential?
If you agree to take part, your name will not be kept with your interview data and the information will not be disclosed to other parties. Your responses to the questions will be used for the purpose of this project only. You can be assured that if you take part in the project you will remain anonymous. I will keep a separate copy of your name and address only so that I can contact you with updates about how the research is going.

Potential Risks and Ethical Consideration
The main risk associated with the interviews is possible discomfort when answering some of the personal questions. However, you are free to withdraw from the study at any time during the interviews and for up to 24 hours after your final interview. You are free to choose not to answer any question you feel uncomfortable with. No other risks are known to the investigator at this time.

Benefits
Participation in the study might result in greater understanding of the problems encountered by those living with CFS/ME. It may result in the development of an intervention designed to better support patients and their carers.

What happens now?
If you are interested in taking part in the study, you are asked to complete and sign the consent form. You will then be given more specific instructions. Do not sign if you do not wish to take part. Please feel free to ask any questions that you may have.

Contact for Further Information
If you have any questions, please contact Katie Oxtoby at k.j.oxtoby@2014.hull.ac.uk. If you have any concerns about the way in which the study has been conducted, please contact the Chair of the Department of psychology Ethics Committee on ethics@psynet.hull.ac.uk, or my PhD supervisor Dr Kim Dent-Brown, k.dent-brown@hull.ac.uk

18 August 2015
Version No. 2

DEBRIEF FORM **Date of Issue:**

Title: How do patients and their significant other experience their relationship in response to a diagnosis of chronic fatigue syndrome/myalgic encephalomyelitis?

Name of Principal Investigator: Katie Oxtoby
Department of Psychology, University of Hull, Cottingham Road, Hull HU6 7RX
Email: k.j.oxtoby@2014.hull.ac.uk Tel: 07872 388844

Background and Research Question:
Previous research has found that relationships between people with chronic fatigue syndrome (CFS/ME) and the person most involved in their care can have an impact on whether the patient's condition improves. Studies of this relationship in the context of other illnesses have found that intervention materials such as leaflets, videos or online courses can be effective in improving this relationship. The aim of this research is therefore to establish what information patients and their carers wish they had received sooner that would have better supported their home relationships following the CFS/ME diagnosis. It is intended that an intervention can then be developed to help future patients and their families, friends or carers. This phase of the research is designed to answer the following question:

- How do patients and their significant others experience their relationship in response and relation to CFS/ME?

Anticipated Findings:
Because this is an exploratory study, there are no anticipated findings. The information collected from participants will direct and shape the study and the subsequent intervention.

Further Information:
If you would like to know more about this study, please feel free to ask any questions by emailing me, Katie Oxtoby – k.j.oxtoby@2014.hull.ac.uk If you have any complaints, concerns, or questions about this research, please feel free to contact, Dr. Kim Dent-Brown, k.dent-brown@hull.ac.uk

Further Support:
It is not expected that participation in this study caused any harm. However, should you feel that it has raised issues for you or your significant other, then you may be able to access more support by contacting one of the following:

Let's Talk Hull
Counselling and psychotherapy services for people with depression and anxiety, by GP or self-referral
01482 247111 (Option 1)

Relate:
Relationship counselling and psychotherapy
Tel: 01482 329621 Website: www.relate.org.uk

Samaritans:
Listening service, email and text support
Tel: 0845 790 9090 Email: jo@samaritans.org

Carers Uk:
Support for people caring for others
Tel: 0808 808 7777 Email: advice@carersuk.org

Action for M.E.:
M.E. support organisation that organizes local support groups and publishes information pamphlets
www.actionforme.org.uk

18 August 2015
Version No. 1

Appendix 3. Topic Guide Interview 1

Katie Oxtoby BA, MSc
PhD Researcher
Department of Psychology
University of Hull
Cottingham Road
HU6 7RX
Tel: 07872 388844
Email: k.j.oxtoby@2014.hull.ac.uk

Topic Guide (DRAFT)

INTERVIEW 1

Demographics
Name

Assigned Participant Number

Date of Birth

Gender

Ethnic Origin

Religion

Place of Birth

Current Hometown

Occupation

Education Level

Marital Status

Semi-structured Topic Guide
Living situation
Length of time since diagnosis
Experience of symptoms
Experience of diagnosis
Current situation in relation to illness

16 April 2015
Version No. 1

Appendix 4. Topic Guide Interview 2

Katie Oxtoby BA, MSc
PhD Researcher
Department of Psychology
University of Hull
Cottingham Road
HU6 7RX
Tel: 07872 388844
Email: k.j.oxtoby@2014.hull.ac.uk

Topic Guide (DRAFT)

INTERVIEW 2

Changes in the relationship before and after onset of CFS/ME
The impact of CFS/ME on the relationship in the long term
The impact of CFS/ME on the lives of the dyad
Emotional and practical support for one another
How members of the household coped
Emotional support received from external sources
Practical support received from external sources
Support networks within the family
Reflections on what worked and what didn't
What support could be in place in an ideal world

16 April 2015
Version No. 1

Appendix 5. Spider Diagram Instructions

Instructions for the person with CFS/ME

UNIVERSITY OF Hull

Katie Oxtoby BA, MSc
PhD Researcher
Department of Psychology
University of Hull
Cottingham Road
HU6 7RX
Tel: 07872 388844
Email: k.j.oxtoby@2014.hull.ac.uk

Topic Guide

INTERVIEW 2

Changes in the relationship before and after onset of CFS/ME
The impact of CFS/ME on the relationship in the long term
The impact of CFS/ME on the lives of the dyad
Emotional and practical support for one another
How members of the household coped
Emotional support received from external sources
Practical support received from external sources
Support networks within the family
Reflections on what worked and what didn't
What support could be in place in an ideal world

Instructions for Spider Diagram

Please generate a spider diagram (also sometimes called a 'Mindmap' or 'Brainstorm') around the topics listed above, focused on your relationship with the person who was closest to you during your experience of CFS. You may do this in whatever way you see fit. You may choose to just do one, or to split it over a couple of diagrams depending on how many notes you wish to make around the topics. The purpose of the task is to facilitate your thinking about the topics over the next week or two so that you have developed your ideas around these topics when we talk again. We can use the completed diagram(s) as an aid in our conversation.

Should you have any questions, please do not hesitate to get in touch.

16 April 2015
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Instructions for the significant other

Katie Oxtoby BA, MSc
PhD Researcher
Department of Psychology
University of Hull
Cottingham Road
HU6 7RX
Tel: 07872 388844
Email: k.j.oxtoby@2014.hull.ac.uk

Topic Guide

INTERVIEW 2

Changes in the relationship before and after onset of CFS/ME
The impact of CFS/ME on the relationship in the long term
The impact of CFS/ME on the lives of the dyad
Emotional and practical support for one another
How members of the household coped
Emotional support received from external sources
Practical support received from external sources
Support networks within the family
Reflections on what worked and what didn't
What support could be in place in an ideal world

Instructions for Spider Diagram

Please generate a spider diagram (also sometimes called a 'Mindmap' or 'Brainstorm') around the topics listed above, focused on your relationship with the person to whom you were close who was struggling with CFS. You may do this in whatever way you see fit. You may choose to just do one, or to split it over a couple of diagrams depending on how many notes you wish to make around the topics. The purpose of the task is to facilitate your thinking about the topics over the next week or two so that you have developed your ideas around these topics when we talk again. We can use the completed diagram(s) as an aid in our conversation.

Should you have any questions, please do not hesitate to get in touch.

16 April 2015
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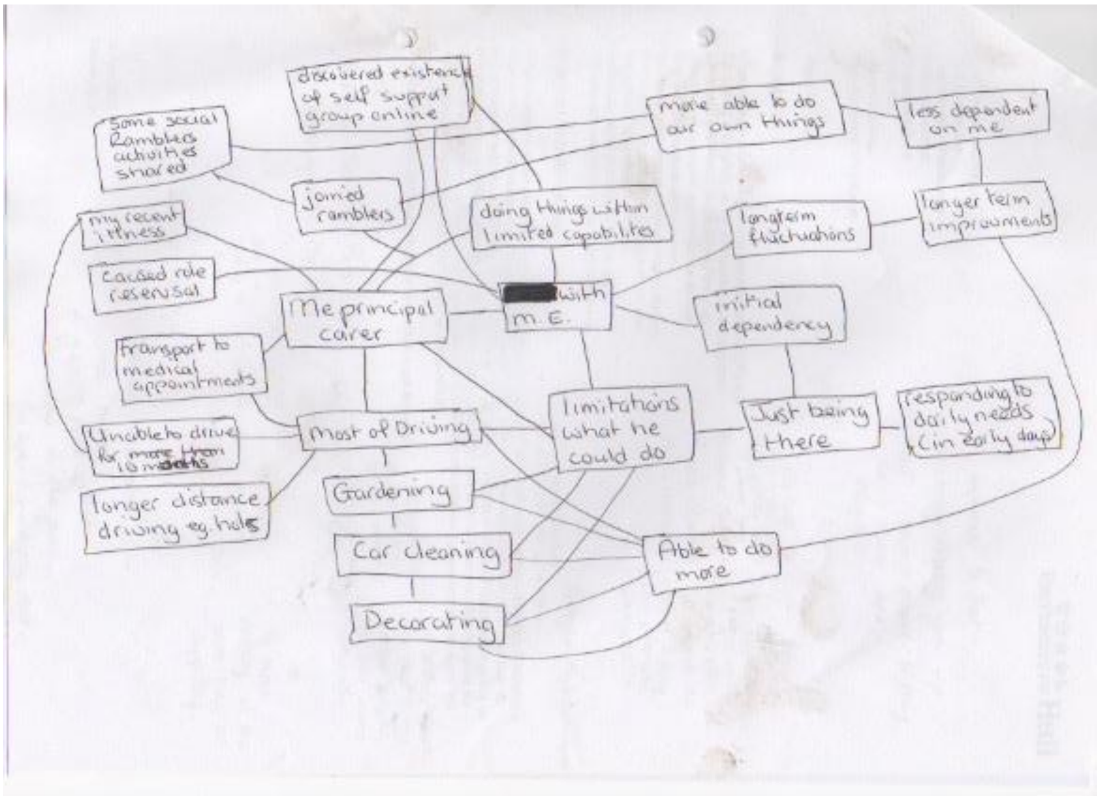
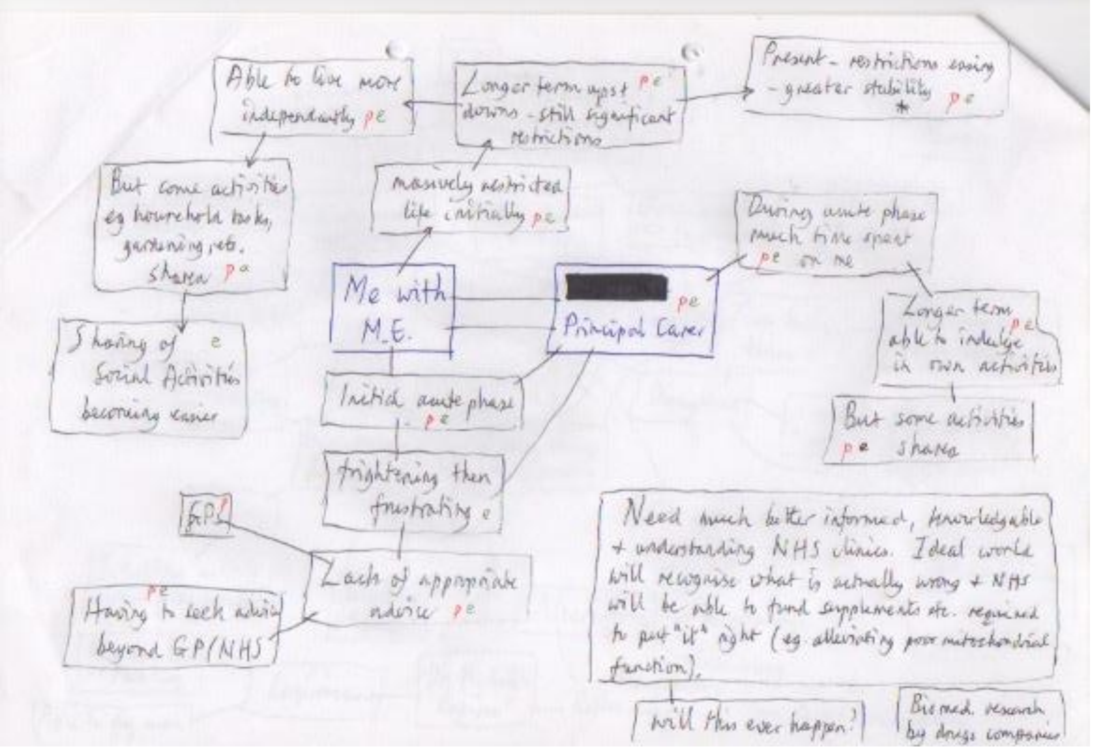
Appendix 6. Spider Diagram Examples

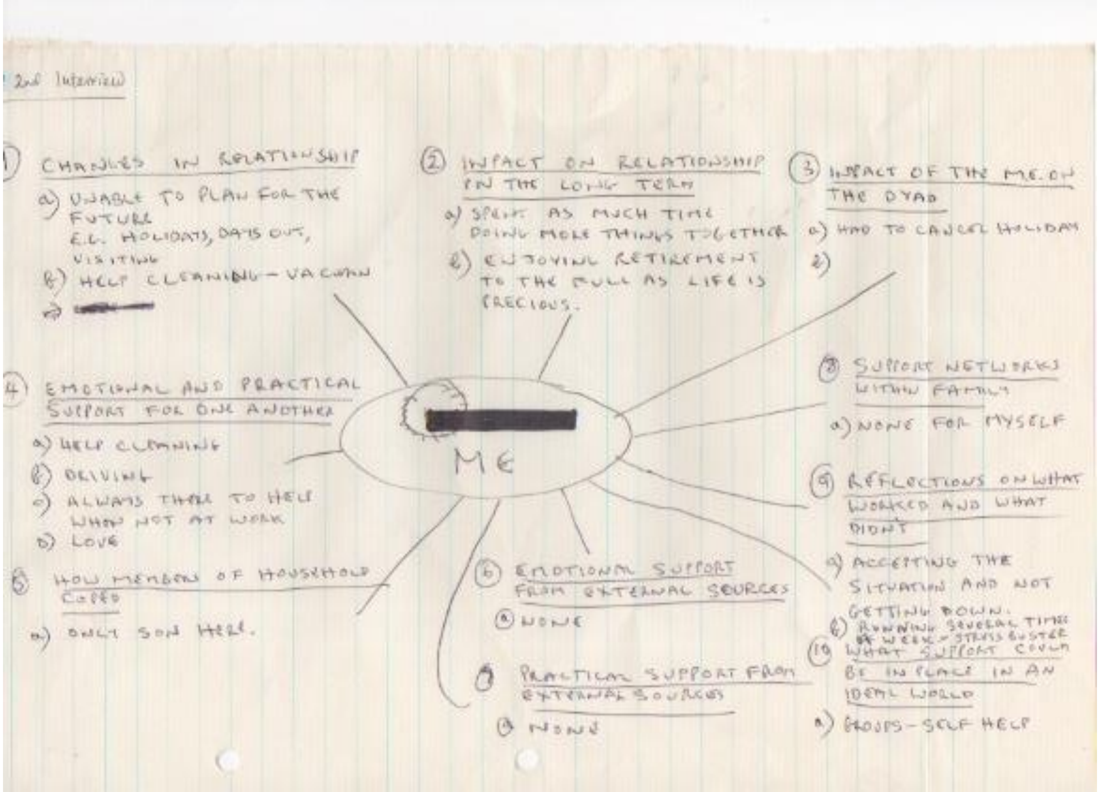
Top: Caroline; Bottom: Richard





Need much better informed, knowledgable + understanding NHS clinics. Ideal world will recognise what is actually wrong + NHS will be able to fund supplements etc. required to put "it" right (eg alleviating poor mitochondrial function).

Top: Eileen; Bottom: Kath



2nd Interview

ME [redacted]

1) CHANGES IN RELATIONSHIP
a) UNABLE TO PLAN FOR THE FUTURE E.G. HOLIDAYS, DAYS OUT, VISITING
b) HELP CLEANING – VACUUM
c) [redacted]

2) IMPACT ON RELATIONSHIP IN THE LONG TERM
a) SPENT AS MUCH TIME DOING MORE THINGS TOGETHER
b) ENJOYING RETIREMENT TO THE FULL AS LIFE IS PRECIOUS.

3) IMPACT OF THE M.E. ON THE DYAD
a) HAD TO CANCEL HOLIDAY
b)

4) EMOTIONAL AND PRACTICAL SUPPORT FOR ONE ANOTHER
a) HELP CLEANING
b) DRIVING
c) ALWAYS THERE TO HELP WHEN NOT AT WORK
d) LOVE

5) HOW MEMBERS OF HOUSEHOLD COPED
a) ONLY SON HERE.

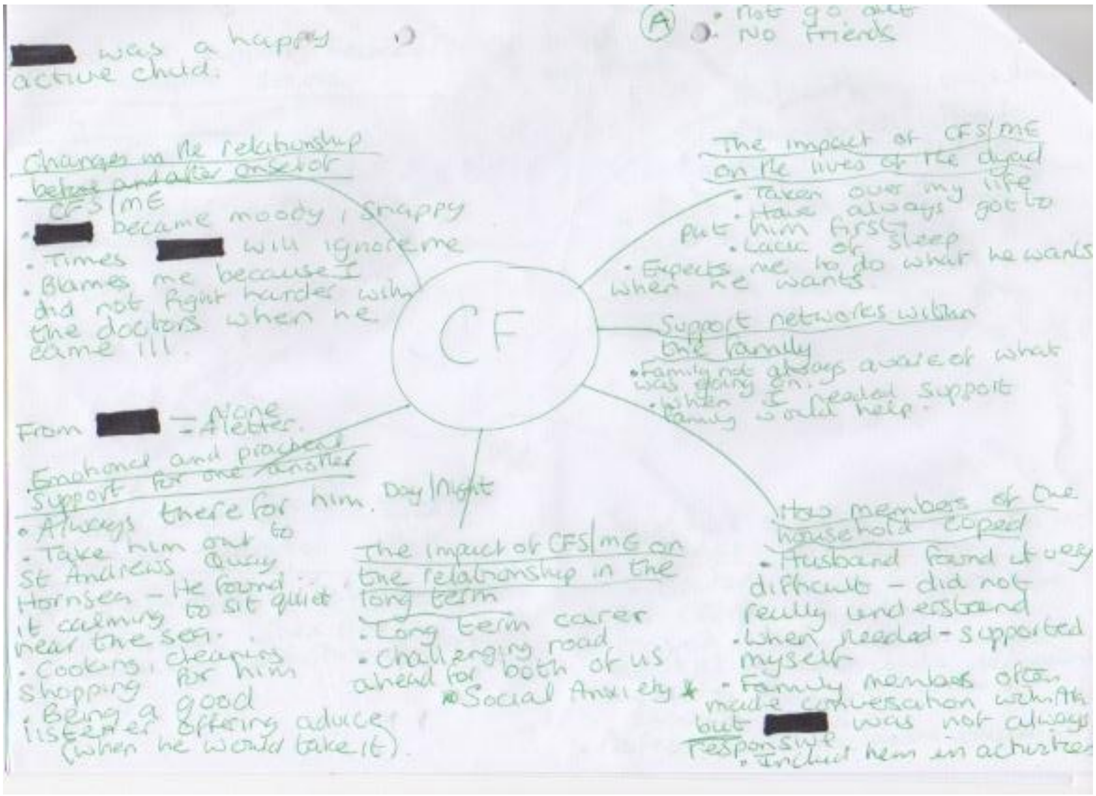
6) EMOTIONAL SUPPORT FROM EXTERNAL SOURCES
a) NONE

7) PRACTICAL SUPPORT FROM EXTERNAL SOURCES
a) NONE

8) SUPPORT NETWORKS WITHIN FAMILY
a) NONE FOR MYSELF

9) REFLECTIONS ON WHAT WORKED AND WHAT DIDN'T
a) ACCEPTING THE SITUATION AND NOT GETTING DOWN.
b) RUNNING SEVERAL TIMES A WEEK – STRESS BUSTER

10) WHAT SUPPORT COULD BE IN PLACE IN AN IDEAL WORLD
a) GROUPS – SELF HELP



[redacted] was a happy active child.

(A) • not go out
• No friends

CF

Changes in the relationship before and after onset of CFS/ME
• [redacted] became moody, snappy
• Times [redacted] will ignore me
• Blames me because I did not fight harder with the doctors when he came ill.

From [redacted] – None
– A letter.

Emotional and practical support for one another
• Always there for him. Day/Night
• Take him out to St Andrews Quay, Hornsea – He found it calming to sit quiet near the sea.
• Cooking, cleaning, shopping for him
• Being a good listener offering advice (when he would take it).

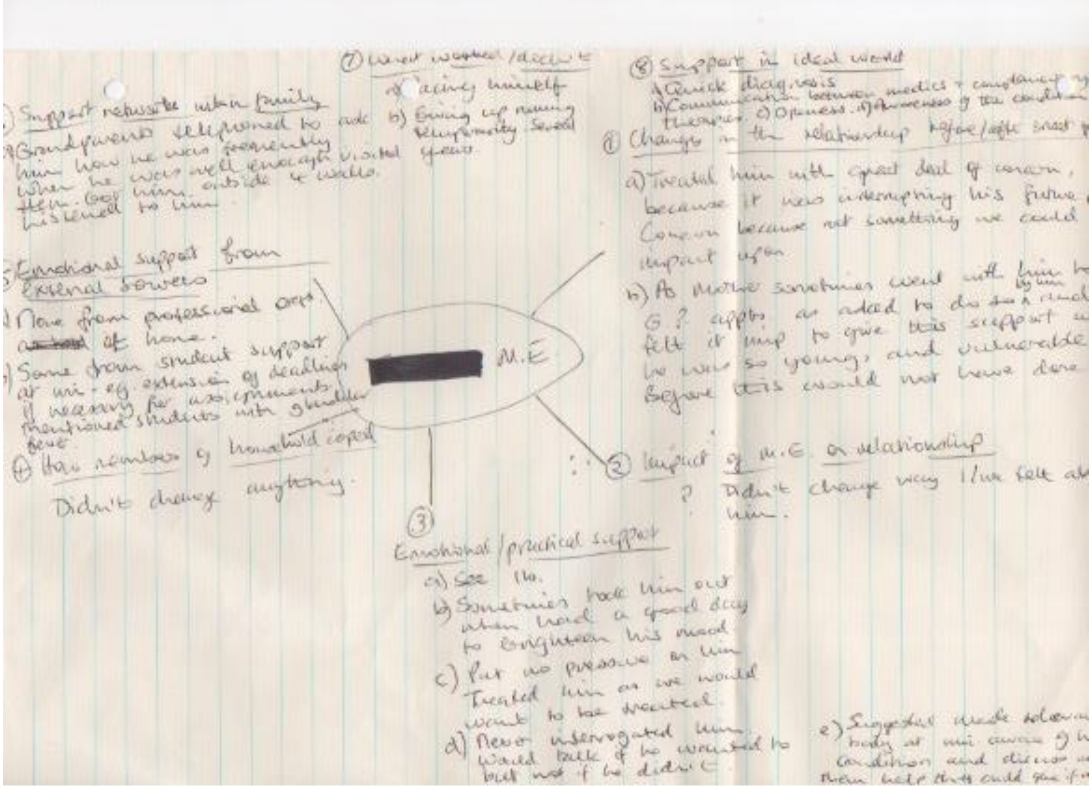
The impact of CFS/ME on the relationship in the long term
• Long term carer
• Challenging road ahead for both of us
* Social Anxiety *

The impact of CFS/ME on the lives of the dyad
• Taken over my life
• Have always got to put him first
• Lack of sleep
• Expects me to do what he wants when he wants.

Support networks within the family
• Family not always aware of what was going on.
• When I needed support family would help.

How members of the household coped
• Husband found it very difficult – did not really understand
• When needed – supported myself
• Family members often made conversation with [redacted] but [redacted] was not always responsive
• Include him in activities

Top: Helen; Bottom: Sean



[redacted] M.E

① Support network within family
a) Grandparents telephoned to ask him how he was frequently. When he was well enough visited them. Got him outside & walks. Listened to him.

⑤ Emotional support from External sources
a) None from professional orgs. [illegible] of none.
b) Some from student support at uni - eg. extension of deadlines if necessary for assignments. Mentioned students with glandular fever.

④ How members of household coped
Didn't change anything.

⑦ What worked / didn't
a) Pacing himself
b) Giving up running temporarily. Several years.

⑧ Support in ideal world
a) Quick diagnosis
b) Communication between medics + complementary therapies c) Openness. d) Awareness of the condition

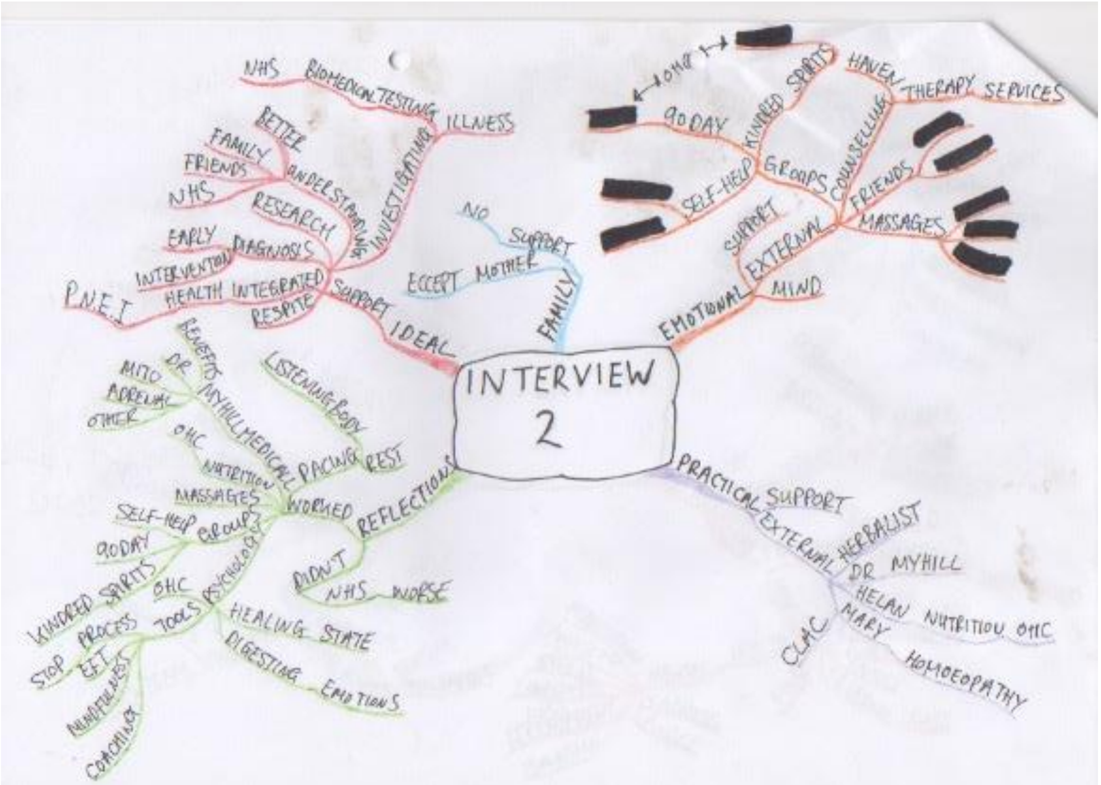
① Changes in the relationship before/after onset

a) Treated him with great deal of concern, because it was interrupting his future, Concern because not something we could impact upon
b) As mother sometimes went with him to G.P. appts as asked to do so by him. Felt it imp to give this support as he was so young, and vulnerable. Before this would not have done.

② Impact of M.E on relationship
? Didn't change way I/we felt about him.

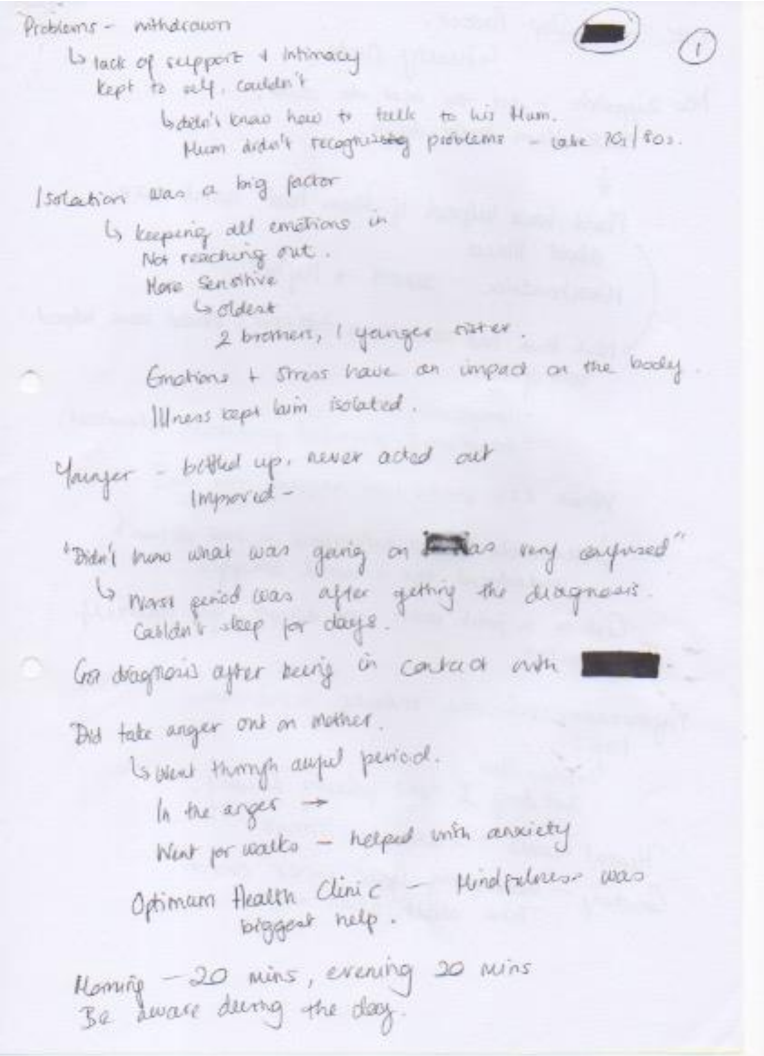
③ Emotional/practical support
a) See 1b.
b) Sometimes took him out when had a good day to brighten his mood
c) Put no pressure on him. Treated him as we would want to be treated.
d) Never interrogated him. Would talk if he wanted to but not if he didn't.

e) Suggested made welcome body at uni aware of his condition and discuss with them help that could give



Appendix 7. Examples of Field Notes

From Steve's First Interview



[redacted] ①

Problems – withdrawn
↳ lack of support + intimacy
Kept to self, couldn't
↳ didn't know how to talk to his Mum.
Mum didn't recognising problems – late 70s/80s.

Isolation was a big factor
↳ keeping all emotions in
Not reaching out.
More sensitive
↳ oldest
2 brothers, 1 younger sister.
Emotions + stress have an impact on the body.
Illness kept him isolated.

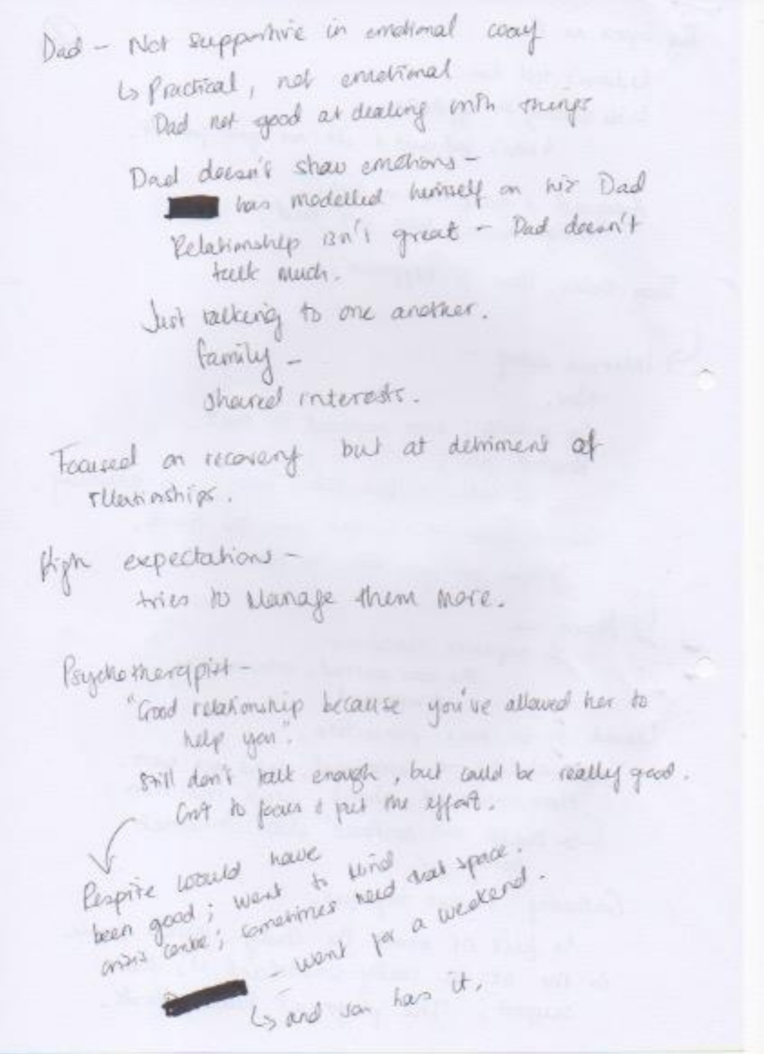
Younger – bottled up, never acted out
Improved –

"Didn't know what was going on [redacted]as very confused"
↳ Worst period was after getting the diagnosis.
Couldn't sleep for days.

Got diagnosis after being in contact with [redacted].

Did take anger out on mother.
↳ Went through awful period.
In the anger →
Went for walks – helped with anxiety
Optimum Health Clinic – Mindfulness was biggest help.

Morning – 20 mins, evening 20 mins
Be aware during the day.



Dad – Not supportive in emotional way
↳ Practical, not emotional
Dad not good at dealing with things

Dad doesn't show emotions –
[redacted] has modelled himself on his Dad
Relationship isn't great – Dad doesn't talk much.

Just talking to one another.
family –
shared interests.

Focused on recovery but at detriment of relationships.

High expectations –
tries to manage them more.

Psychotherapist –
"Good relationship because you've allowed her to help you".
Still don't talk enough, but could be really good.
Got to focus & put the effort.

Respite would have been good; went to Mind crisis centre; sometimes need that space.
[redacted] – went for a weekend.
↳ and son has it.

Big impact on Mum ②
↳ doesn't tell him
↳ like walking on eggshells
↳ didn't feel good + it's not good for her.

Apologised + gave her a hug.
things haven't been as bad. [redacted]

Been stable, Mum is happier.

↳ Internet dating –
Nov.
Got talking; were supposed to meet.
Reacted fine;
2 dates – but didn't amount to anything.
Would rather be honest from the start.
↳ Now, just goes with the flow.

↳ Mum →
2 negative reactions
She was worried, vulnerability.
disappointed.
Learnt to be more proactive.
Vulnerability → emotional, could get hurt.
More concerned about your reaction.
↳ Would still continue with internet dating.
Gathering things together.
↳ feels OK about the study. Wants people in the NHS to really understand it; More support; The physical hea[redacted]tests.

OHC [redacted] Stop Process.
↳ healing state.
Not supportive – did the best she could.
2009 – Mum broke down.
⇓
Would have helped if Mum had learnt more about illness
Mitochondria – 2009 – Dr. Myhill
↳ Wish Mum had shown more interest; Would have helped both of them.
- Complexity.
- Emotions, physical problems, behaviour.

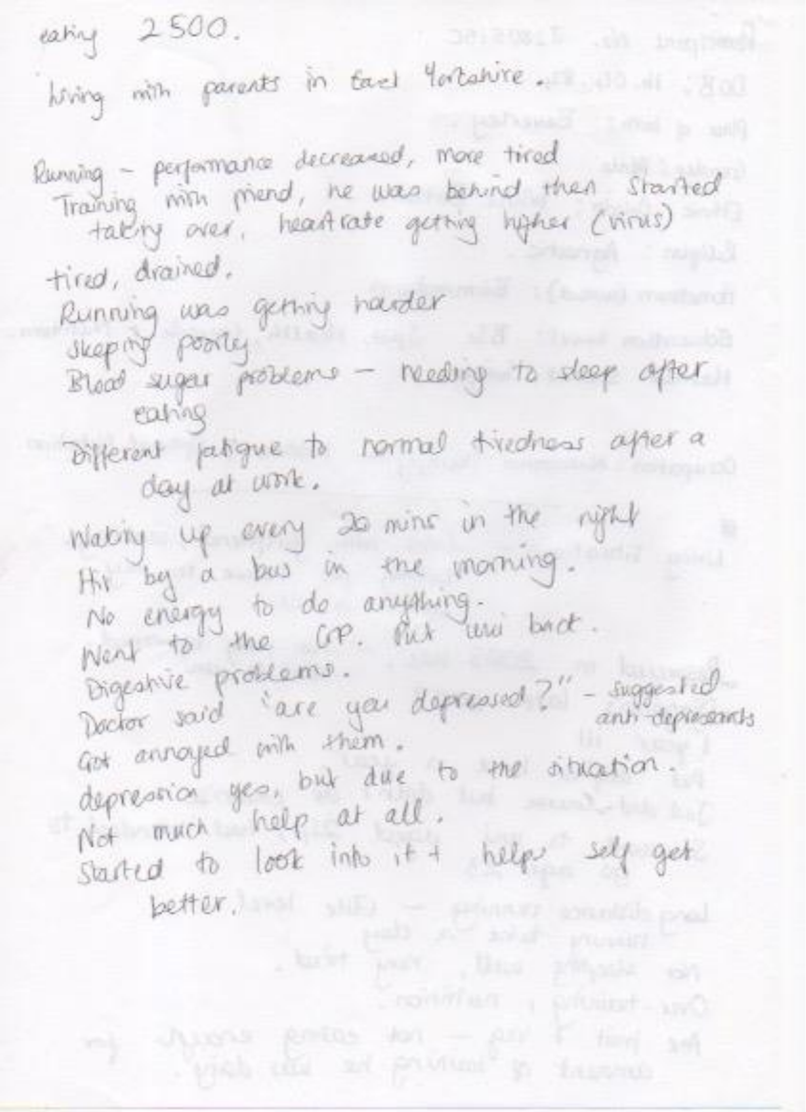
What can people who support you do?

Understands [redacted]'s behaviour – but doesn't understand the internal struggle.

Get to a point where you accept, get yourself [redacted] better.

Psychoneuroendocrine immune syndrome.
PNEI
↳ Epstein Barr
Just doing 1 test polarity dowsing.
"Mental tennis" → anxiety – stress.
Coaching – "developing your inner coach"
Think about what –

From Rob's First Interview



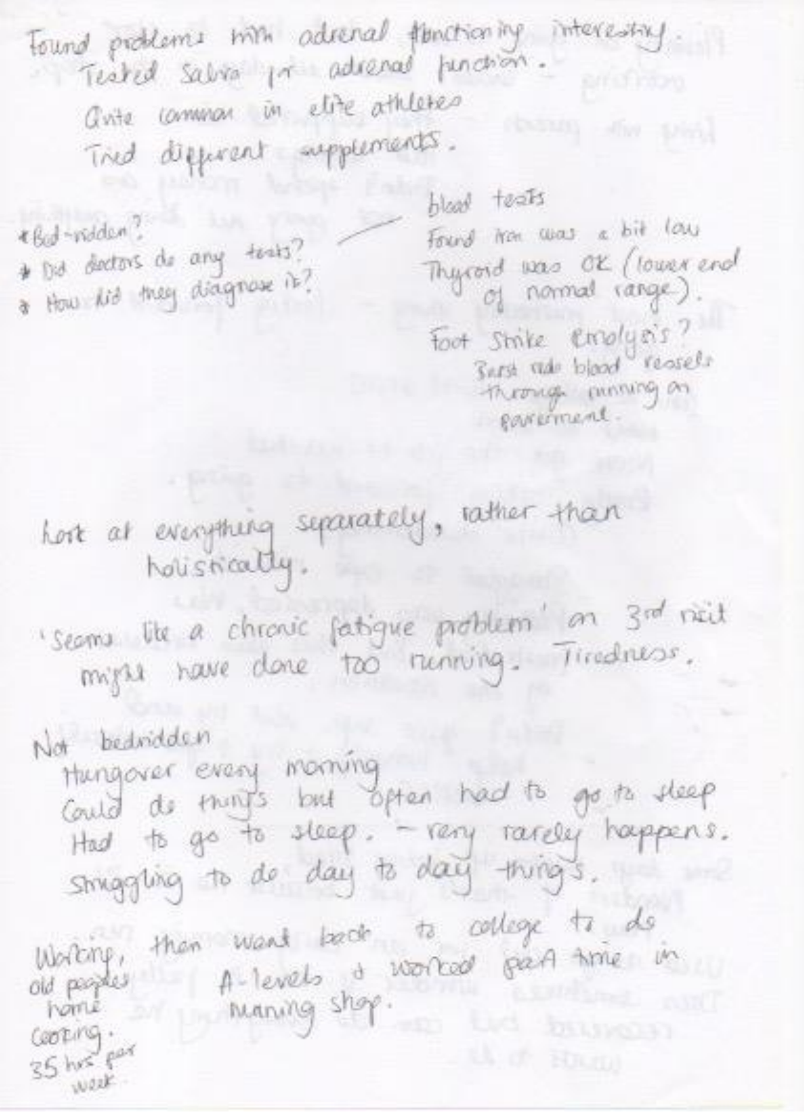
eating 2500.

living with parents in East Yorkshire.

Running – performance decreased, more tired
Training with friend, he was behind then started taking over. heartrate getting higher (virus)

tired, drained.
Running was getting harder
sleeping poorly
Blood sugar problems – needing to sleep after eating
Different fatigue to normal tiredness after a day at work.

Waking up every 20 mins in the night
Hit by a bus in the morning.
No energy to do anything.
Went to the GP. Put uni back.
Digestive problems.
Doctor said 'are you depressed?" – suggested anti-depressants
Got annoyed with them.
depression yes, but due to the situation.
Not much help at all.
Started to look into it + help self get better.



Found problems with adrenal functioning interesting.
Tested Saliva for adrenal function.
Quite common in elite athletes
Tried different supplements.

* Bed-ridden?
* Did doctors do any tests?
* How did they diagnose it?

blood tests
Found iron was a bit low
Thyroid was OK (lower end of normal range).
Foot Strike emolysis?
Burst red blood vessels through running on pavement.

Look at everything separately, rather than holistically.

'Seems like a chronic fatigue problem' on 3rd visit
might have done too running. Tiredness.

Not bedridden
Hungover every morning
Could do things but often had to go to sleep
Had to go to sleep. – very rarely happens.
Struggling to do day to day things.

Working, then went back to college to do A-levels + worked part time in running shop.

old peoples home
cooking.
35 hrs per week.

Planning on going to uni, but had to stop
working - couldn't stand all day in the shop.

Living with parents - they supported him
Had savings
Didn't spend money as
not going out doing anything.

The most frustrating thing - looking forward to
going.

Gone to college 16-18 GNVQ
Went to work
Never got the job he wanted
Really looking forward to going.
Quite devastating.
Struggled to cope with it.
Probably was depressed, Was
frustrated. But this was because
of the situation.
Didn't give up, did try and
help himself & try & get himself
better.

Some days wakes up feeling tired,
Wonders if that's just because he is 31
now.
Used to go out for an early morning run.
Does sometimes wonder if he is fully
recovered but can do everything he
wants to do.

Was single at the time - Mum had been ill
herself - Mum understood - had IBS etc.

Dad has been generally quite well.
↳ He was understanding - but.

Send through & then we can arrange to
talk.

GF used to worked at Hull Uni
↳
Caffeine + effects on 5k running
performance.
↓
Improved performance
but only 8
participants
Proplus + orange
drink.

Mum had
fibromyalgia
+ had problems
with chronic
fatigue.
↓
Mum & Dad happy
to help.

Nafferton - retired.

Giving parents a
ring at the weekend.

Appendix 8. Journal Excerpts

Sample 1.

5th July 2015

Notes: In this entry I use the term ME in place of CFS or CFS/ME because that is the terminology used by the relevant participants.

One of the most interesting things to come out of the two interviews so far was that both participants seemed to believe that for someone to really understand illness, they have to have experienced disabling illness themselves. [Participant] talked about his mother who had fibromyalgia and his mothers' doctor, who had ME, and explained that these were the two people who expressed the most empathy and understanding and who knew the best what was needed by them. He also talked about his father who had struggled with understanding but explained that this had improved since a recent bout of illness leading to an operation. It seemed the participant felt this experience had given him more insight into what he and his mother had been going through.

Similarly, [Participant] talked about a friend who also has ME and is very supportive and understanding because she completely understands what it is like. The participant talked about acceptance being vital.

Both participants highlighted that their fathers had tried to help by 'doing' – such as things around the house, picking up meds etc. but that both had struggled to 'understand' because they didn't have any experience of illness themselves. It seemed clear as well that both felt there was an element of emotional distance from the illness from their fathers.

Both participants also talked about dissatisfaction with the medical profession, in particular GPs – a lack of understanding and empathy seemed to be the major issue here too, although [Participant] made clear that he also felt this was partially down to the pressure placed upon doctors to fix things within a 10-minute consultation.

The overarching theme definitely seems at present to be one of feeling that those who have not experienced illness do not ‘understand’. I’d like to try to draw more out of what participants actual mean by the word ‘understand’, and the need to be and to feel understood. So, acceptance and validation also seemed to be important for these participants.

Sample 2.

27TH August 2015

Grounded Theory Interviews

I have just arranged the first interview with [Participant]'s parents to take place next Tuesday. On the phone, [Participant] told me she had also had ME herself and asserted some very strong opinions about the way that it is treated in mental health services. I found myself feeling a little frustrated, in that she was misunderstanding the rationale behind decision-making in medicine, as well as the focus of my study.

She was very keen to tell me all about what did work for her too, she did not find anything that the doctors recommended useful, she thinks the use of antidepressants is appalling because they make people more fatigued, not less so. She told me she has been well since 2011 and that it was an alternative medicine that helped for her.

She is frustrated by what she perceives as 'them' treating it 'like a type of depression'.

I am really interested in this issue of people with the illness feeling failed by the medical profession, but then this is not the focus of my research. It is difficult because Grounded Theory would suggest that I should let the participants lead the study, rather than my preconceived ideas and currently existing theories, but then Kim has recommended that I try to remain focused on my research question.

I suppose part of the problem is that it is easy for people to misunderstand the nature of the research – many hear that I am researching CFS and think I'm looking for a cure. But I'm not. I'm specifically interested in the relationships between PwCFS/ME and their significant others and how their relationship is affected by the illness and vice versa.

During the telephone conversation I really had to restrain myself from entering into a debate about the nature of evidence-based treatment in medicine. I need to ask questions about this idea – who does she perceive ‘them’ to be? What does she think might be the reason that it is treated in mental health services? What had she expected the GPs to do?

But then I also need to remain focused on the research question, and remember that I am interviewing her as [Participant]’s significant other. So, in some way, I do have to lead the research and the interview, even though it seems to go against everything that my grounded theory bible tells me to do!

Remember: “Explore participants’ terms and locate what they tell you within a social process.” “Whether participants recount their concerns without interruption or researchers request specific information, the result is a construction – or reconstruction – of a reality.”

Don’t: “correct the research participant about his or her views, experiences, or feelings; interrogate or confront; ignore or gloss over what the participant wishes to talk about.”

“Try to frame questions in neutral terms.”

It is important to consider the ‘researcher and participants relative power to control the interview. Participants may have stories they want to tell and tales that they wish to sidestep or tread on softly. Thus, they may exert control over the interview – and the situation – by avoiding areas that might elicit probing questions.’ (p.93) (All from Charmaz, 2014)

Sample 3.

15TH September 2015

Comparison of [Participant] & [Participant]

I am currently transcribing interviews with [Participant] & his SO, and [Participant]'s SOs and there are some interesting comparisons arising between the two.

Both participants fell ill at around the same age – 18 or 19. However, one seemed to know immediately what was wrong and treat it and behave accordingly (his mother was well-versed in CFS and actually has it herself). He recovered within a year.

Other similarities include the fact that both boys experience uprooting and being moved away from friends – although this wasn't far away it appears to affected both significantly and has been raised in both interviews by parents.

In both interviews, issues were raised about parenting skills which both parents vehemently refuted. On both occasions the issues had been raised as a result of psychotherapy sessions. (Although in [Participant]'s case it was his sister who raised the issue with his parents, not him).

Both experienced separation and withdrawal from family members around that time – losing touch with siblings, no longer showing an interest in one another's lives.

In both families there was talk by the parents of not fully disclosing family situations to siblings, or letting siblings support parents over the behaviour of an ill person within the family because the parent doesn't want it to cause problems between the siblings.

Sample 4.

17TH September 2015

Reflections on 2nd interviews thus far

Yesterday's interview posed a real challenge. Having just transcribed the data from this particular participant, I became very aware of how much I had allowed her to shape the direction of the interview, and how much it resulted in anecdotal storytelling that actually wouldn't be useful in terms of direct relations to my research question.

In the first interview, that was fine because I was trying to be a 'good grounded theorist', but the interview did sprawl on for a long time, covering a lot of ground that wasn't relevant, but that was precisely why I set up the first interview/second interview system. The first interview was intended as an opportunity for them to do all of this unfocused talking and tell me their story. It has in some case brought out a great deal of 'rich' data, which is why I have taken the decision to start transcribing the data, but it has also meant that some participants have slightly taken advantage of the opportunity to talk, and literally told me every tiny detail. This presented great conflict to me as a researcher as during the first interviews, I didn't mind letting them talk, but once I have the transcription task at the back of my mind, and the realisation of the size of this task, I became acutely aware of my own intentions and needs in the research, and almost a little resentful of what I perceived to be the 'hijacking' of the interview, and a slight reluctance to actually answer my questions directly. This did improve somewhat towards the end of the interview when I became more focused, direct and assertive, but the first 45 minutes of the interview consisted of only one question and then non-stop talking, most of which was anecdotal. Unfortunately, something went wrong with the recording and the first 45 minutes are inaudible. However, I did recap on the main points and will have those covered in the next hour of data that is recorded.

I think there is a need to be mindful of the conflicts between being an orthodox grounded theorist and conducting interviews accordingly, and the practicalities of being a PhD researcher working within tight time constraints. I have to be realistic about the time I have, and also be mindful of the fact that the interviewees do want the opportunity to talk, and it might not be about the things I want to talk about. I will speak with Kim about this issue – it is very difficult to manage the balance between the purpose of the research and the topics that participants want to talk about, such as their issues with the NHS. In the first interview, I am inviting them to do this because otherwise I knew they would do it in the second interview. However, I then have to alter my role in the second interview and be a bit more assertive in order to get them to focus on the questions asked.

Sample 5.

Wednesday 11th November 2015

I am struggling to make sense of [Participant]'s transcript. There is so much belief in there about homeopathic remedies. I have heard numerous participants talk of similar things; in response to the apparent inability of the NHS to offer appropriate support or symptom relief, many patients have no option but to go in search of other treatment and other support. Many find this in homeopathic remedies, although it would seem anecdotally this is of more interest amongst my female participants than the males.

To come back to [Participant], at times it sounds like she has really struggled but I can't help but have this gut reaction that she has just learned to be ill and that is just a way of life for her now, to be the patient, to be exempt from day-to-day life, from socialising and leaving the house. That springs to mind in a number of cases but then it brings me to wondering about why this becomes the case. It does not seem that these patients consciously want to live this life. They are miserable living in this way. Some have other people who care for them, particularly in the case of men, but the women tend to be having to manage more independently, with limited help.

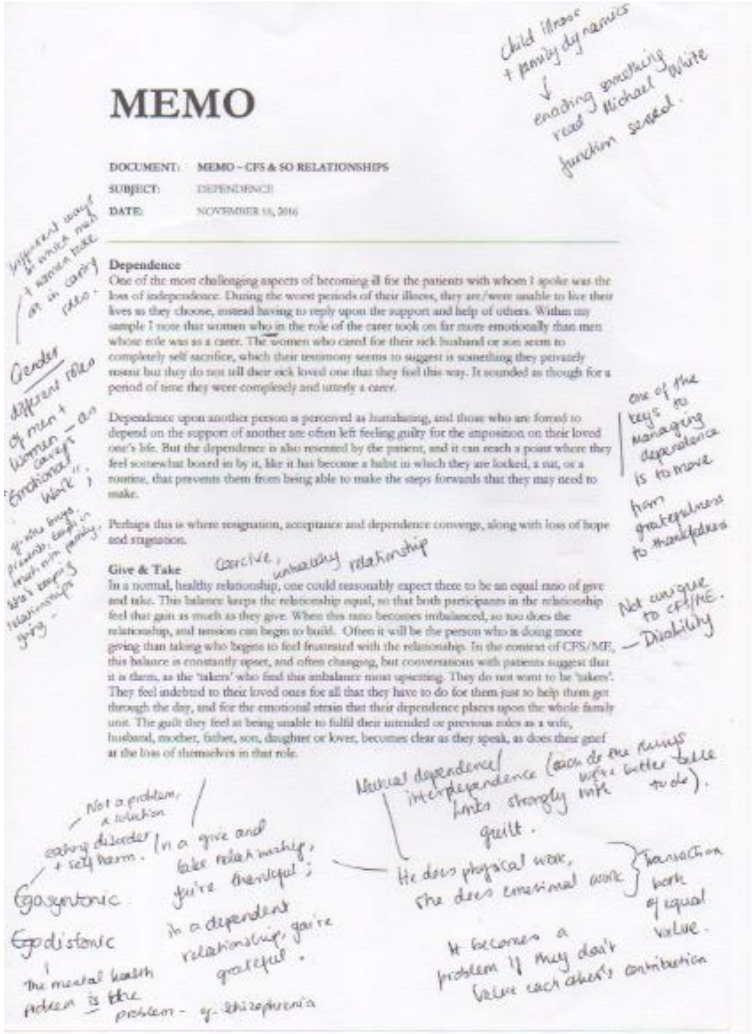
There seems to be a longstanding discontentment in her marriage, a lack of affection and romantic love; she told me that their physical relationship ended around the time that she became ill. It occurs to me that being ill has become something to focus on, a way of coping with the dissatisfaction married life – an extreme way of burying her head in the sand whilst simultaneously escaping from or hiding from the disappointment of the relationship and the pressures of life as a wife.

Meanwhile, I am feeling guilty for reaching these conclusions because I feel like I am betraying the participants. They feel like negative evaluations that they would not appreciate and that is something that doesn’t sit well with me personally because of my need to please and not wanting to upset people. However, I have to be careful not to allow that to bias my thinking and my analysis. I do also empathise with my participants; I do see their pain and their struggles. I don’t view them as ‘malingerers’ – I don’t think, although maybe I need to go back and think a bit about that term ‘malingerer’ before I make that claim.

There does seem to be a thread running through with many of them; they are very in touch with their bodies and believe themselves to be highly sensitive to external environmental factors. Although most of them do also seem to recognise the impact of stress and emotional difficulties.

In terms of the relationship with the SO, there appears to be a lot of tension in these relationships. They are relationships that are under a great deal of strain, whereby the ill person has high expectations of the SO and the SO becomes trapped in a world of enforced caring, resentment towards the ill person, concern for them, regret, disappointment at the loss of their own freedoms, but frequently also unwilling to confront the fact that they are missing out at the hands of their partner’s illness, instead just accepting. In some cases, they find another interest that keeps them going such as gardening, walking, running, socialising or work but in other cases they appear to forgo everything in favour of being the carer. This does not always have the intended positive impact on the person with CFS. It seems that the SO needs to strike a fine balance between caring and supporting, being there but also having their own life. They need to find ways of ensuring they don’t feel that they are missing out, because that breeds yet more resentment, but at the same time ensuring that the person with CFS doesn’t feel left behind because again resentment then develops, and resentment leads to anger and a withdrawal from communication. Perhaps here, communication is key.

Appendix 9. Memos



Child illness
+ family dynamics
↓
enacting something
read Michael White
function served.

MEMO

DOCUMENT: MEMO – CFS & SO RELATIONSHIPS
SUBJECT: DEPENDENCE
DATE: NOVEMBER 16, 2016

Different ways in which men + women take on in carer roles +

Gender
different roles of men + women – as carers
"Emotional Work";
e.g. who buys presents, keeps in touch with family, who's keeping relationships going –

Dependence
One of the most challenging aspects of becoming ill for the patients with whom I spoke was the loss of independence. During the worst periods of their illness, they are/were unable to live their lives as they choose, instead having to reply upon the support and help of others. Within my sample I note that women who in the role of the carer took on far more emotionally than men whose role was as a carer. The women who cared for their sick husband or son seem to completely self sacrifice, which their testimony seems to suggest is something they privately resent but they do not tell their sick loved one that they feel this way. It sounded as though for a period of time they were completely and utterly a carer.

Dependence upon another person is perceived as humiliating, and those who are forced to depend on the support of another are often left feeling guilty for the imposition on their loved one's life. But the dependence is also resented by the patient, and it can reach a point where they feel somewhat boxed in by it, like it has become a habit in which they are locked, a rut, or a routine, that prevents them from being able to make the steps forwards that they may need to make.

One of the keys to managing dependence is to move from gratefulness to thankfulness

Perhaps this is where resignation, acceptance and dependence converge, along with loss of hope and stagnation.

Give & Take coercive, unhealthy relationship
In a normal, healthy relationship, one could reasonably expect there to be an equal ratio of give and take. This balance keeps the relationship equal, so that both participants in the relationship feel that gain as much as they give. When this ratio becomes imbalanced, so too does the relationship, and tension can begin to build. Often it will be the person who is doing more giving than taking who begins to feel frustrated with the relationship. In the context of CFS/ME, this balance is constantly upset, and often changing, but conversations with patients suggest that it is them, as the 'takers' who find this imbalance most upsetting. They do not want to be 'takers'. They feel indebted to their loved ones for all that they have to do for them just to help them get through the day, and for the emotional strain that their dependence places upon the whole family unit. The guilt they feel at being unable to fulfil their intended or previous roles as a wife, husband, mother, father, son, daughter or lover, becomes clear as they speak, as does their grief at the loss of themselves in that role.

Not unique to CFS/ME. – Disability

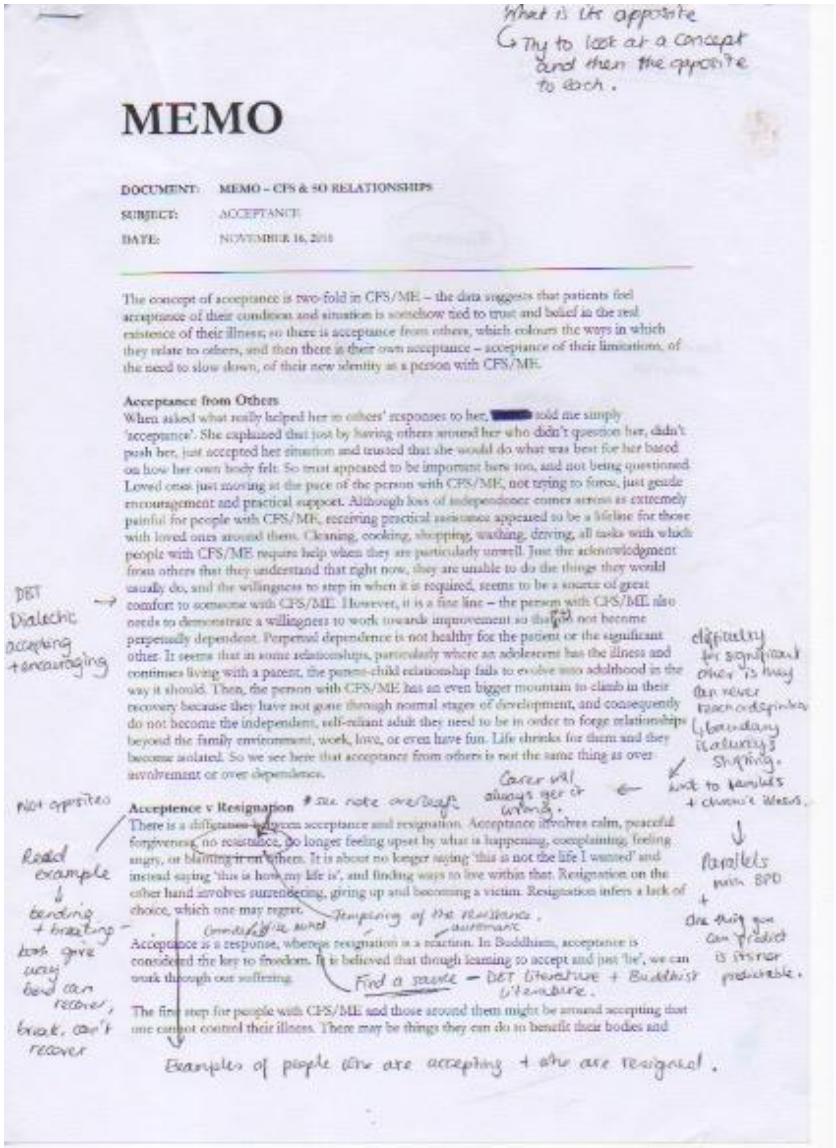
Not a problem, a solution
eating disorder + self harm.
Egosyntonic
Egodistonic
The mental health problem is the problem – eg. schizophrenia

In a give and take relationship, you're thankful;
in a dependent relationship, you're grateful.

Mutual dependence/ interdependence (each do the things we're better able to do).
links strongly with guilt.

He does physical work,
She does emotional work
Transaction both of equal value.

It becomes a problem if they don't value each other's contribution



What is the opposite
↳ Try to look at a concept and then the opposite to each.

MEMO

DOCUMENT: MEMO – CFS & SO RELATIONSHIPS
SUBJECT: ACCEPTANCE
DATE: NOVEMBER 16, 2016

The concept of acceptance is two-fold in CFS/ME – the data suggests that patients feel acceptance of their condition and situation is somehow tied to trust and belief in the real existence of their illness; so there is acceptance from others, which colours the ways in which they relate to others, and then there is their own acceptance – acceptance of their limitations, of the need to slow down, of their new identity as a person with CFS/ME.

Acceptance from Others
When asked what really helped her in others' responses to her, [redacted] told me simply 'acceptance'. She explained that just by having others around her who didn't question her, didn't push her, just accepted her situation and trusted that she would do what was best for her based on how her own body felt. So trust appeared to be important here too, and not being questioned. Loved ones just moving at the pace of the person with CFS/ME, not trying to force, just gentle encouragement and practical support. Although loss of independence comes across as extremely painful for people with CFS/ME, receiving practical assistance appeared to be a lifeline for those with loved ones around them. Cleaning, cooking, shopping, washing, driving, all tasks with which people with CFS/ME require help when they are particularly unwell. Just the acknowledgment from others that they understand that right now, they are unable to do the things they would usually do, and the willingness to step in when it is required, seems to be a source of great comfort to someone with CFS/ME. However, it is a fine line – the person with CFS/ME also needs to demonstrate a willingness to work towards improvement so that ~~they~~ do not become perpetually dependent. Perpetual dependence is not healthy for the patient or the significant other. It seems that in some relationships, particularly where an adolescent has the illness and continues living with a parent, the parent-child relationship fails to evolve into adulthood in the way it should. Then, the person with CFS/ME has an even bigger mountain to climb in their recovery because they have not gone through normal stages of development, and consequently do not become the independent, self-reliant adult they need to be in order to forge relationships beyond the family environment, work, love, or even have fun. Life shrinks for them and they become isolated. So we see here that acceptance from others is not the same thing as over involvement or over dependence.

DBT
Dialectic
accepting + encouraging →

difficulty for significant other is they then never reach independence
↳ boundary is always shifting.

↙ link to families + chronic illness.
↓
Parallels with BPD
+
One thing you can predict is its not predictable.

Carer will always get it wrong. ←

Not opposites

Acceptence v Resignation # see note overleaf.
There is a difference between acceptance and resignation. Acceptance involves calm, peaceful forgiveness, no resistance, no longer feeling upset by what is happening, complaining, feeling angry, or blaming it on others. It is about no longer saying 'this is not the life I wanted' and instead saying 'this is how my life is', and finding ways to live within that. Resignation on the other hand involves surrendering, giving up and becoming a victim. Resignation infers a lack of choice, which one may regret.

Tempering of the resistance.
Considering acceptance
autonomic

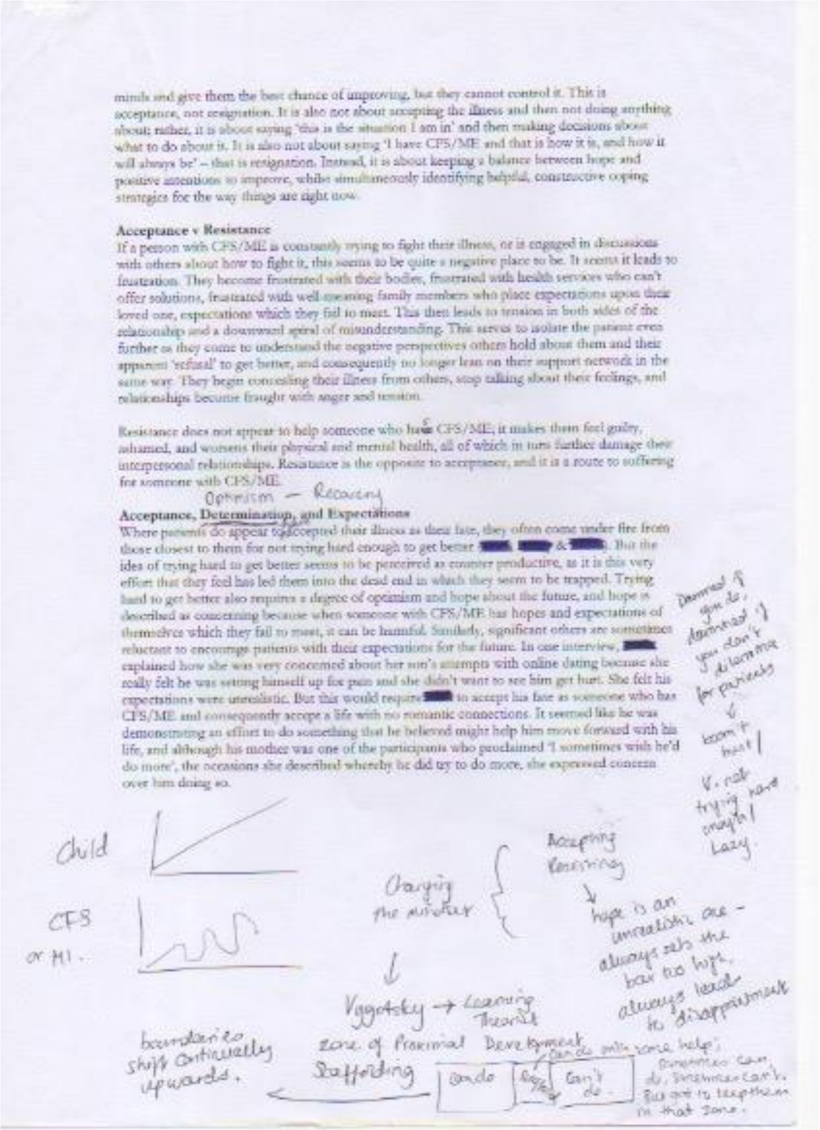
Acceptance is a response, whereas resignation is a reaction. In Buddhism, acceptance is considered the key to freedom. It is believed that though learning to accept and just 'be', we can work through our suffering.

Find a source – DBT literature + Buddhist literature.

Read example
↓
bending + breaking – both give way
bend can recover, break, can't recover

The first step for people with CFS/ME and those around them might be around accepting that one cannot control their illness. There may be things they can do to benefit their bodies and

Examples of people who are accepting + who are resigned.



minds and give them the best chance of improving, but they cannot control it. This is acceptance, not resignation. It is also not about accepting the illness and then not doing anything about; rather, it is about saying 'this is the situation I am in' and then making decisions about what to do about it. It is also not about saying 'I have CFS/ME and that is how it is, and how it will always be' – that is resignation. Instead, it is about keeping a balance between hope and positive intentions to improve, whilst simultaneously identifying helpful, constructive coping strategies for the way things are right now.

Acceptance v Resistance
If a person with CFS/ME is constantly trying to fight their illness, or is engaged in discussions with others about how to fight it, this seems to be quite a negative place to be. It seems it leads to frustration. They become frustrated with their bodies, frustrated with health services who can't offer solutions, frustrated with well-meaning family members who place expectations upon their loved one, expectations which they fail to meet. This then leads to tension in both sides of the relationship and a downward spiral of misunderstanding. This serves to isolate the patient even further as they come to understand the negative perspectives others hold about them and their apparent 'refusal' to get better, and consequently no longer lean on their support network in the same way. They begin concealing their illness from others, stop talking about their feelings, and relationships become fraught with anger and tension.

Resistance does not appear to help someone who has CFS/ME; it makes them feel guilty, ashamed, and worsens their physical and mental health, all of which in turn further damage their interpersonal relationships. Resistance is the opposite to acceptance, and it is a route to suffering for someone with CFS/ME.

Optimism – Recovery

Acceptance, Determination, and Expectations
Where patients do appear to have accepted their illness as their fate, they often come under fire from those closest to them for not trying hard enough to get better ([redacted], [redacted] & [redacted]). But the idea of trying hard to get better seems to be perceived as counter productive, as it is this very effort that they feel has led them into the dead end in which they seem to be trapped. Trying hard to get better also requires a degree of optimism and hope about the future, and hope is described as concerning because when someone with CFS/ME has hopes and expectations of themselves which they fail to meet, it can be harmful. Similarly, significant others are sometimes reluctant to encourage patients with their expectations for the future. In one interview, [redacted] explained how she was very concerned about her son's attempts with online dating because she really felt he was setting himself up for pain and she didn't want to see him get hurt. She felt his expectations were unrealistic. But this would require [redacted] to accept his fate as someone who has CFS/ME and consequently accept a life with no romantic connections. It seemed like he was demonstrating an effort to do something that he believed might help him move forward with his life, and although his mother was one of the participants who proclaimed 'I sometimes wish he'd do more', the occasions she described whereby he did try to do more, she expressed concern over him doing so.

Damned if you do, damned if you don't
dilemma for patients
↓
Don't hurt / V. not trying hard enough / Lazy.

Child

CFS or ME.

Changing the mindset
Accepting
Resisting
↓
hope is an unrealistic one – always sets the bar too high, always leads to disappointment

↓
Vygotsky → Learning Theorist
Zone of Proximal Development
Scaffolding
Can do | Can do with some help | Can't do.
Sometimes can do, sometimes can't. But got to keep them in that zone.

boundaries shift continually upwards.

Phases of mourning – worth mapping onto data? – what would bereavement look like normally?

Is it harder to get through to bargaining + acceptance – What does it look like in the context of chronic fatigue. (No closure) ↳ More like an abduction rather than death.

MEMO

DOCUMENT: MEMO – CFS & SO RELATIONSHIPS
SUBJECT: DARK EMOTIONS
DATE: NOVEMBER 18, 2016

Loss & Grief

Stolen, rather than died at other people.

Relationships in the context of CFS/ME are coloured by loss and grief. Both members of the dyad grieve for the loss of the person they and their loved one was before the illness, and also for the loss of their life before the illness. They grieve for the loss of everything they wanted to be, and everything they were hoping for and working towards. Life grinds to a halt and is filled with a sense of decline, or worse, being stationary. Being stationary is conceived of as worse than decline because at least with decline, there is a hope that at some point the bottom will be reached and then there may be a progression towards improvement – at least things continue to unravel, develop and change during a period of decline, so there is movement. But when things plateau, seeming to be stationary and stagnant, it becomes a rut, a frustrating circle which casts a dark, unmoving cloud over everyone involved. Progress and improvement has virtually ground to a halt and belief in improvement is almost non-existent. This is a dark place for any family to be in. Memories of how they used to be become painful for all of them, as they remember how full of life they once were, it highlights their decline. Thoughts about the future, and the **loss of hope** that they will ever be able to live the life they wanted to live, and do normal things like work, have friendships and relationships, become sad and painful – the loss of something that you once thought you might have.

Loss of past + loss of future.

Where you've been and where you're going.
↓
Eriksonians 20-30s.

Anger

Directional relational – in relation to another.

The dyadic relationship in many cases becomes tainted by anger. The patient is overwhelmed with anger at the unfairness of what they have lost, what they are missing out on, and for the lack of answers offered by health services. They wonder 'why me?' and feel jealous of those who don't have the illness. They feel jealous of those able to live a normal life, able to do the things that they once did or would have liked to do, and this comparison of their disability with another's ability leaves them feeling angry. Often, their anger and frustration is expressed in their interactions with significant others, either through tears or arguments.

Resentment

This anger can give way to resentment, both on the part of the patient and the significant other. As explained, the patient is not only the patient who experiences loss. The significant other also loses a great deal. They lose the functioning version of the person they love, which often goes hand-in-hand with the loss of support for them – they become the carer in many cases, and the person for whom they are caring does not have the energy to give very much back. Over time, the pressure on the significant other's life can give way to the development of a deep feeling of resentment, but its one which they appear to feel so guilty about feeling that they push it back, repress it, and dare never to express it. But it did come through in some of the interviews. Some of the significant others, particularly women, ([redacted]) clearly resented the way their lives had been suffocated by their husband/son's illness. The fact that they had become carers, nursing a dependent, clearly took away so much from them, it constrained them and their time, so that they felt unable to be themselves, unable to have the freedom to do what they needed or wanted to do. Their responsibility towards the patient left them feeling tied and often silently frustrated, afraid to show their feelings for fear of making matters worse. They appeared to feel walked over, walking on eggshells and severely limited by their SO's illness.

There was also resentment apparent in the patients; many of them resented periods where their significant other was unable to care for them or give them the attention they needed, for example when the SO was ill, or needed to go away for work, or had responsibilities to other family members. The patient, often already experiencing a sense of complete loss of control or power is then hit by a feeling of abandonment and they resent the SO and anyone else involved in the apparent abandonment, such as other family members, for this.

If they can do this and not feel guilty, we reach regression.

The antidote to resentment for the SO appears to be making time for oneself, doing something that is just theirs. One participant described how his running time is his headspace, and that was what kept him going through the most difficult times. Another described how she had joined a walking club and regularly goes away with them for weekends. This seemed to allow her to regain her sense of self and to build a new social life away from the marriage, and in turn this is what enabled the marriage to continue. Another patient explained how her husband takes time away to attend gigs with his friends, enabling him to just be himself for a while, temporarily free of the stresses of family life – this relationship appeared to be a particularly healthy one. Another liked to play golf, and although in this relationship there appeared to be resentment on the part of the patient, there was very little resentment apparent in the account of the SO. The highest levels of resentment were apparent among those, both patients and significant others, whose lives were engulfed by the illness, and who had limited opportunities to escape its clutches.

Bargaining/acceptance for the SO. – It's a deal we make to make it bearable.

Guilt

If they can't do this without the side effects of guilt,

Often this loss, grief and resentment is accompanied by a deep sense of guilt. On the part of the patient, they feel guilty for the impact they know they are having on their family. They feel deep guilt at being unable to fulfil the roles they feel should have been filling more fully if they were physically able, for example as a parent, a lover, a husband/wife, a colleague or as a son or daughter caring for an elderly parent. They feel guilty that their loved ones have experienced loss as a consequence of their illness, guilty about the worry their families carry for them, and about the limitations their illness have placed on the lives of their loved ones.

Tom wants her to do this to alleviate his guilt.

Guilt gets parcelled out between them.

Significant others are also often consumed by guilt. They feel guilty for being well and able bodied; they feel the need to go out and get on with life – they have to hold the family together by working, caring for family members, and often they have to hold themselves together by pursuing their own hobbies simultaneously, but this is all accompanied by a sense of guilt because in doing all of this, they are both absent from their needy loved one, and are doing the things that they know their loved one so wishes that they could do.

Ben's guilt over + patient feels guilty

More accommodating

Shame & Humiliation

Similar to anyone who has a long-term illness. It would be easier to accept a blue badge for physical.

Many people with CFS/ME feel useless, completely unable to contribute to society, and this negatively impacts their self esteem. They feel ashamed of their status as a dependent. Many have to accept support offered for people with disabilities, such as blue badges, the use of walking sticks or wheelchairs, and receiving Disability Living Allowance. However, most seem to struggle with the concept of themselves as disabled. They find the acceptance of this label humiliating, and despite being severely limited by their illness, they don't necessarily identify with being disabled. They find this label disempowering somehow, and perhaps also more final, like it is an acceptance of more permanent limitations. Whilst many CFS/ME patients do appear fairly static and stuck where they are, the reality is that most experience highs and lows, periods of improvement and relapse, and for this reason, many are unwilling to accept a label that they find so limiting and final.

grateful = guilty
hateful = less guilty.

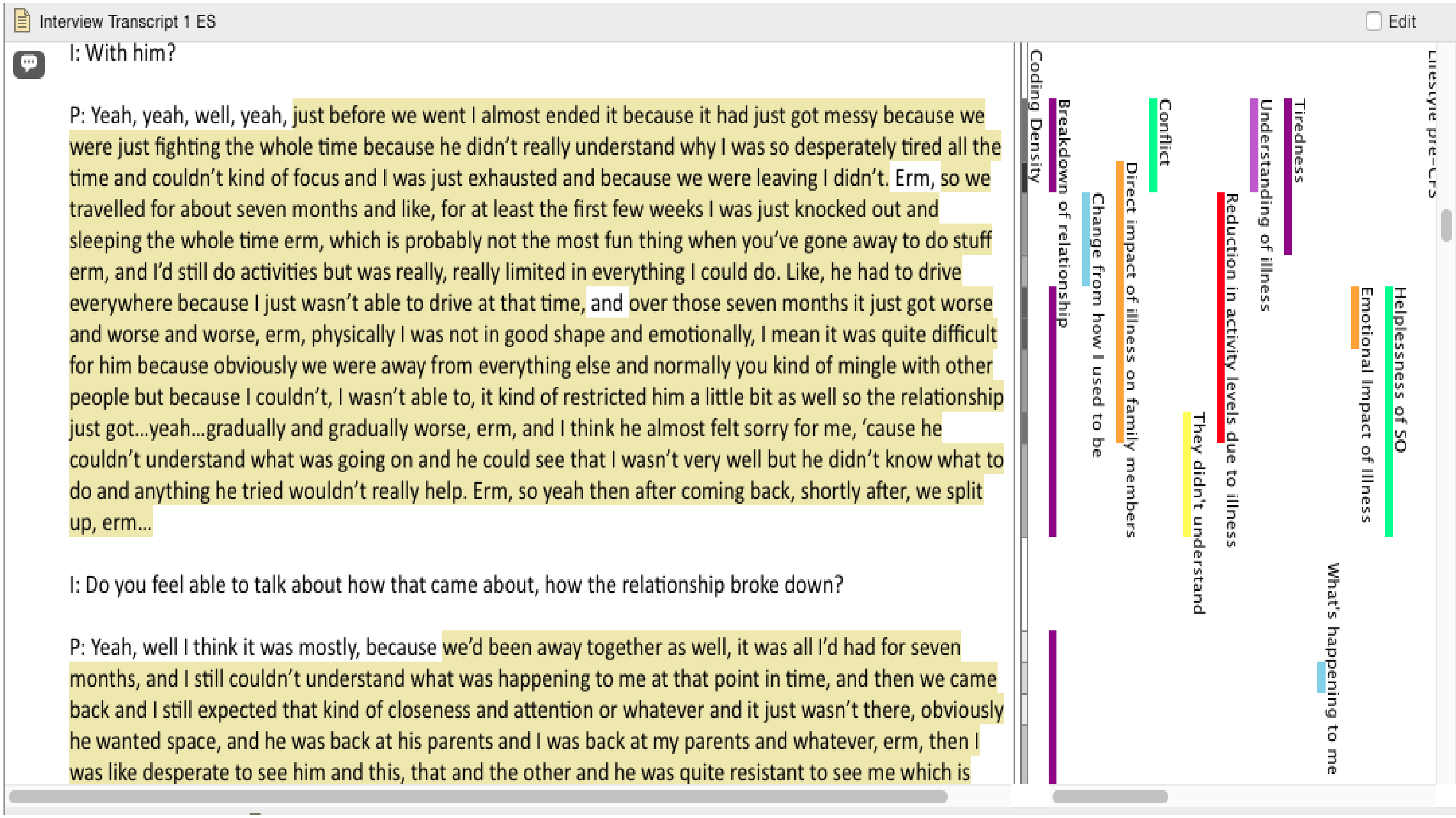
Moved away from guilt, guilt has moved out of the dyad.

Their periods of complete dependence on others can become a source of shame and humiliation for them too. One patient described how it pained her that her husband had to help her wash her hair and brush her teeth at times, and that it was like they had another child in the house. There was pain evident in her voice as she spoke of this, as well as deep shame at becoming so dependent and the humiliation of her regression from wife and mother to dependent child. Another participant,

Comes from a remaining hope that they will recover
↳ unwilling to accept reality. but not able to lift themselves out of it.

Appendix 10. Line-by-Line Coding Examples

NVivo Screen Shots

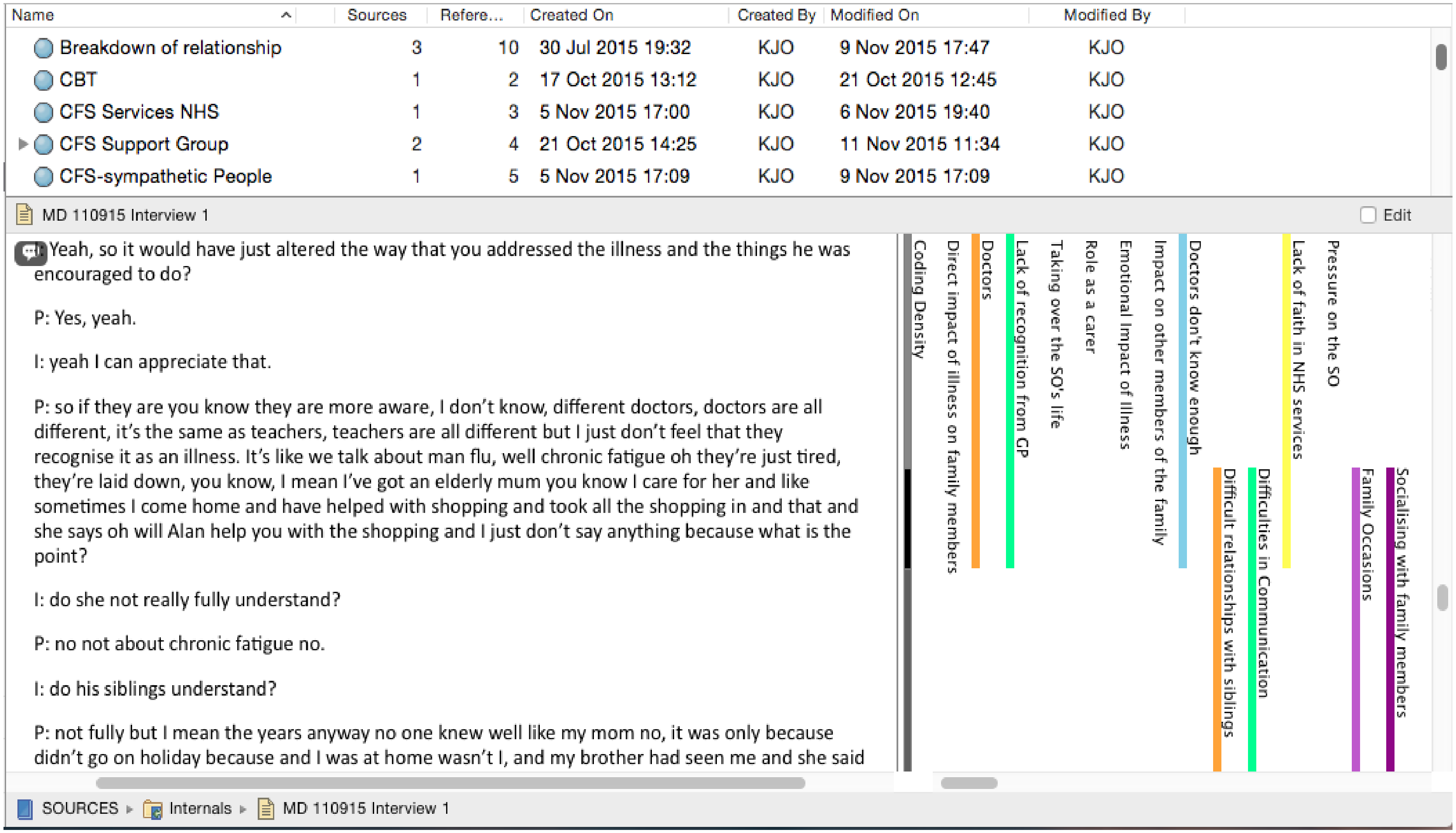


I: With him?

P: Yeah, yeah, well, yeah, just before we went I almost ended it because it had just got messy because we were just fighting the whole time because he didn't really understand why I was so desperately tired all the time and couldn't kind of focus and I was just exhausted and because we were leaving I didn't. Erm, so we travelled for about seven months and like, for at least the first few weeks I was just knocked out and sleeping the whole time erm, which is probably not the most fun thing when you've gone away to do stuff erm, and I'd still do activities but was really, really limited in everything I could do. Like, he had to drive everywhere because I just wasn't able to drive at that time, and over those seven months it just got worse and worse and worse, erm, physically I was not in good shape and emotionally, I mean it was quite difficult for him because obviously we were away from everything else and normally you kind of mingle with other people but because I couldn't, I wasn't able to, it kind of restricted him a little bit as well so the relationship just got...yeah...gradually and gradually worse, erm, and I think he almost felt sorry for me, 'cause he couldn't understand what was going on and he could see that I wasn't very well but he didn't know what to do and anything he tried wouldn't really help. Erm, so yeah then after coming back, shortly after, we split up, erm...

I: Do you feel able to talk about how that came about, how the relationship broke down?

P: Yeah, well I think it was mostly, because we'd been away together as well, it was all I'd had for seven months, and I still couldn't understand what was happening to me at that point in time, and then we came back and I still expected that kind of closeness and attention or whatever and it just wasn't there, obviously he wanted space, and he was back at his parents and I was back at my parents and whatever, erm, then I was like desperate to see him and this, that and the other and he was quite resistant to see me which is



Name	Sources	Refere…	Created On	Created By	Modified On	Modified By
Breakdown of relationship	3	10	30 Jul 2015 19:32	KJO	9 Nov 2015 17:47	KJO
CBT	1	2	17 Oct 2015 13:12	KJO	21 Oct 2015 12:45	KJO
CFS Services NHS	1	3	5 Nov 2015 17:00	KJO	6 Nov 2015 19:40	KJO
▶ CFS Support Group	2	4	21 Oct 2015 14:25	KJO	11 Nov 2015 11:34	KJO
CFS-sympathetic People	1	5	5 Nov 2015 17:09	KJO	9 Nov 2015 17:09	KJO

MD 110915 Interview 1

Yeah, so it would have just altered the way that you addressed the illness and the things he was encouraged to do?

P: Yes, yeah.

I: yeah I can appreciate that.

P: so if they are you know they are more aware, I don't know, different doctors, doctors are all different, it's the same as teachers, teachers are all different but I just don't feel that they recognise it as an illness. It's like we talk about man flu, well chronic fatigue oh they're just tired, they're laid down, you know, I mean I've got an elderly mum you know I care for her and like sometimes I come home and have helped with shopping and took all the shopping in and that and she says oh will Alan help you with the shopping and I just don't say anything because what is the point?

I: do she not really fully understand?

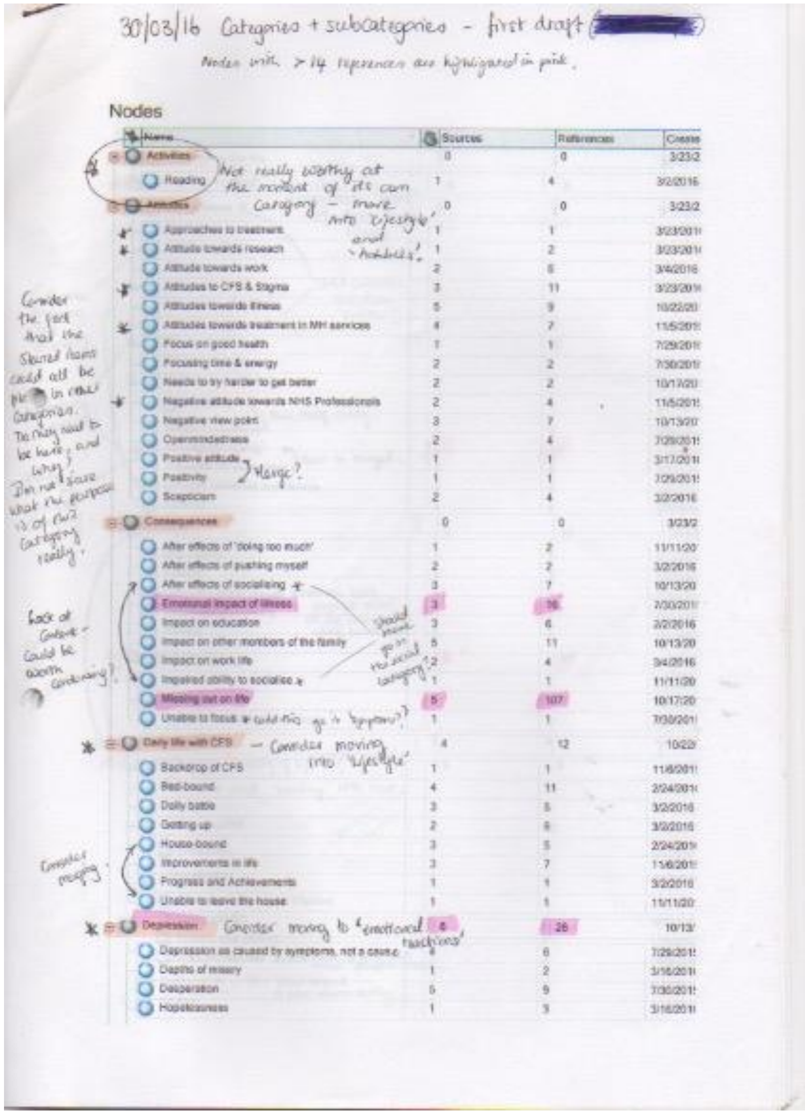
P: no not about chronic fatigue no.

I: do his siblings understand?

P: not fully but I mean the years anyway no one knew well like my mom no, it was only because didn't go on holiday because and I was at home wasn't I, and my brother had seen me and she said

Appendix 11. Early Coding Structures

Example consists of 4 pages out of 13 from an NVivo print-out



30/03/16 Categories + subcategories - first draft [redacted]

Nodes with >14 references are highlighted in pink.

Nodes

Name	Sources	References	Create
Activities	0	0	3/23/2
Reading	1	4	3/2/2016
Attitudes	0	0	3/23/2
Approaches to treatment	1	1	3/23/201
Attitude towards reseach	1	2	3/23/201
Attitude towards work	2	6	3/4/2016
Attitudes to CFS & Stigma	3	11	3/23/201
Attitudes towards illness	5	9	10/22/20
Attitudes towards treatment in MH services	4	7	11/5/201
Focus on good health	1	1	7/29/201
Focusing time & energy	2	2	7/30/201
Needs to try harder to get better	2	2	10/17/20
Negative attitude towards NHS Professionals	2	4	11/5/201
Negative view point	3	7	10/13/20
Openmindedness	2	4	7/29/201
Positive attitude	1	1	3/17/201
Positivity	1	1	7/29/201
Scepticism	2	4	3/2/2016
Consequences	0	0	3/23/2
After effects of 'doing too much'	1	2	11/11/20
After effects of pushing myself	2	2	3/2/2016
After effects of socialising	3	7	10/13/20
Emotional impact of illness	3	16	7/30/201
Impact on education	3	6	3/2/2016
Impact on other members of the family	5	11	10/13/20
Impact on work life	2	4	3/4/2016
Impaired ability to socialise	1	1	11/11/20
Missing out on life	5	107	10/17/20
Unable to focus	1	1	7/30/201
Daily life with CFS	4	12	10/22/
Backdrop of CFS	1	1	11/6/201
Bed-bound	4	11	2/24/201
Daily battle	3	5	3/2/2016
Getting up	2	6	3/2/2016
House-bound	3	5	2/24/201
Improvements in life	3	7	11/6/201
Progress and Achievements	1	1	3/2/2016
Unable to leave the house	1	1	11/11/20
Depression	6	26	10/13/
Depression as caused by symptoms, not a cause	4	6	7/29/201
Depths of misery	1	2	3/16/201
Desperation	5	9	7/30/201
Hopelessness	1	3	3/16/201

Not really worthy at the moment of its own category – more into 'lifestyle' and 'hobbies'.

Consider the fact that the starred items could all be [illegible] in other categories. They may need to be here, and why? I'm not sure what the purpose is of this category really.

Merge?

Should these go in the social category?

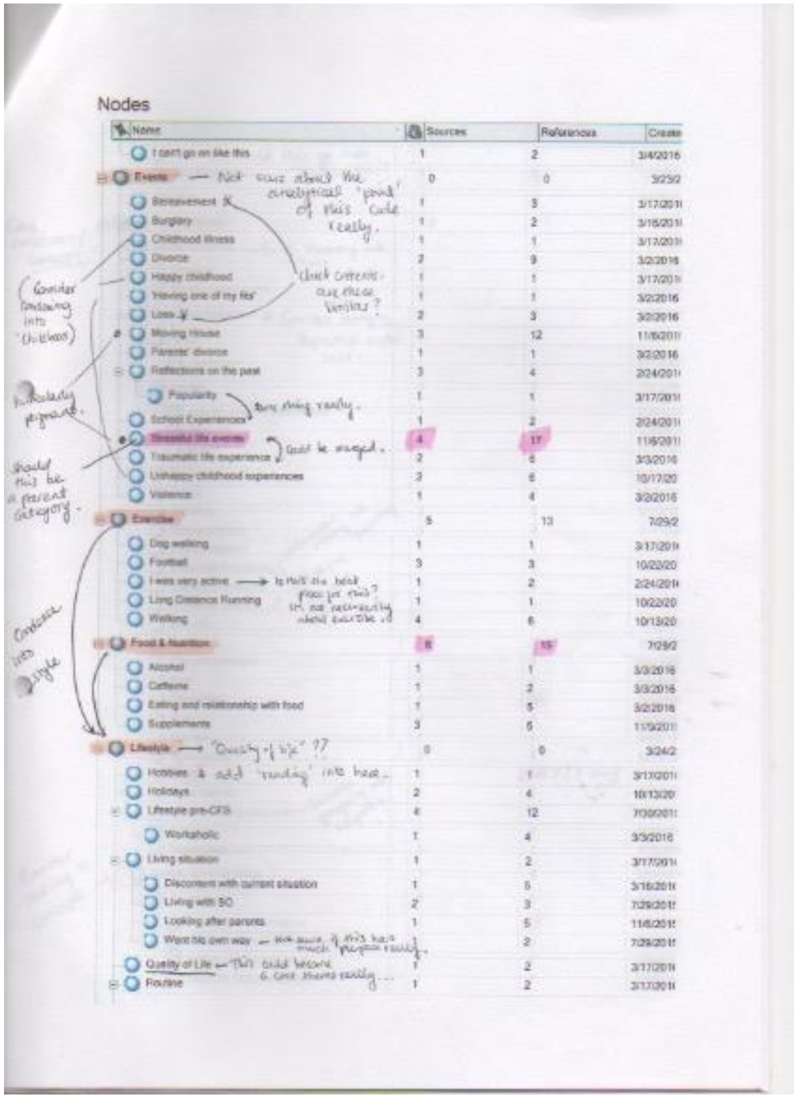
Look at Codebook – could be worth condensing?

could this go to 'symptoms'?

– Consider moving into 'lifestyle'

Consider merging.

Consider moving to 'emotional reactions'



Nodes

Name	Sources	References	Create
I can't go on like this	1	2	3/4/2016
Events	0	0	3/23/2
Bereavement	1	3	3/17/201
Burglary	1	2	3/16/201
Childhood illness	1	1	3/17/201
Divorce	2	9	3/2/2016
Happy childhood	1	1	3/17/201
'Having one of my fits'	1	1	3/2/2016
Loss	2	3	3/2/2016
Moving House	3	12	11/6/201
Parents' divorce	1	1	3/2/2016
Reflections on the past	3	4	2/24/201
Popularity	1	1	3/17/201
School Experiences	1	2	2/24/201
Stressful life events	4	17	11/6/201
Traumatic life experience	2	6	3/3/2016
Unhappy childhood experiences	2	6	10/17/20
Violence	1	4	3/3/2016
Exercise	5	13	7/29/2
Dog walking	1	1	3/17/201
Football	3	3	10/22/20
I was very active	1	2	2/24/201
Long Distance Running	1	1	10/22/20
Walking	4	6	10/13/20
Food & Nutrition	6	15	7/29/2
Alcohol	1	1	3/3/2016
Caffeine	1	2	3/3/2016
Eating and relationship with food	1	5	3/2/2016
Supplements	3	6	11/9/201
Lifestyle	0	0	3/24/2
Hobbies	1	1	3/17/201
Holidays	2	4	10/13/20
Lifestyle pre-CFS	4	12	7/30/201
Workaholic	1	4	3/3/2016
Living situation	1	2	3/17/201
Discontent with current situation	1	5	3/16/201
Living with SO	2	3	7/29/201
Looking after parents	1	5	11/6/201
Want his own way	1	2	7/29/201
Quality of Life	1	2	3/17/201
Routine	1	2	3/17/201

– Not sure about the analytical 'point' of this code really.

Check criteria – are these similar?

(Consider condensing into 'childhood')

Particularly poignant.

Same thing really.

Could be merged.

Should this be a parent category.

Is this the best place for this? It's not necessarily about exercise...

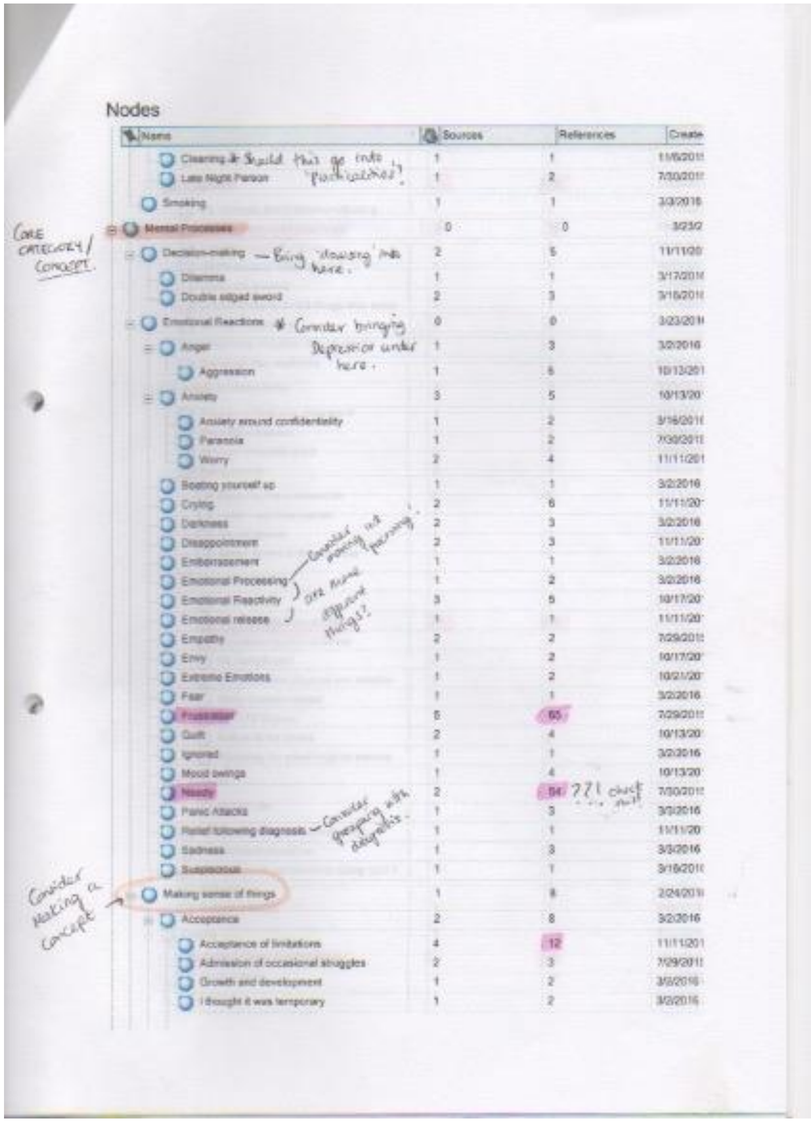
Condense into lifestyle

→ 'Quality of life' ??

& add 'reading' into here.

– Not sure if this has much purpose really.

– This could become 6 core themes really...



Nodes

Name	Sources	References	Create
Cleaning	1	1	11/6/201
Late Night Person	1	2	7/30/201
Smoking	1	1	3/3/2016
Mental Processes	0	0	3/23/2
Decision-making	2	5	11/11/20
Dilemma	1	1	3/17/201
Double edged sword	2	3	3/16/201
Emotional Reactions	0	0	3/23/201
Anger	1	3	3/2/2016
Aggression	1	6	10/13/201
Anxiety	3	5	10/13/20
Anxiety around confidentiality	1	2	3/16/201
Paranoia	1	2	7/30/201
Worry	2	4	11/11/201
Beating yourself up	1	1	3/2/2016
Crying	2	6	11/11/20
Darkness	2	3	3/2/2016
Disappointment	2	3	11/11/20
Embarrassment	1	1	3/2/2016
Emotional Processing	1	2	3/2/2016
Emotional Reactivity	3	5	10/17/20
Emotional release	1	1	11/11/20
Empathy	2	2	7/29/201
Envy	1	2	10/17/20
Extreme Emotions	1	2	10/21/20
Fear	1	1	3/2/2016
Frustration	5	65	7/29/201
Guilt	2	4	10/13/20
Ignored	1	1	3/2/2016
Mood swings	1	4	10/13/20
Needy	2	94	7/30/201
Panic Attacks	1	3	3/3/2016
Relief following diagnosis	1	1	11/11/20
Sadness	1	3	3/3/2016
Suspicious	1	1	3/16/201
Making sense of things	1	8	2/24/201
Acceptance	2	8	3/2/2016
Acceptance of limitations	4	12	11/11/201
Admission of occasional struggles	2	3	7/29/201
Growth and development	1	2	3/3/2016
I thought it was temporary	1	2	3/2/2016

* Should this go into 'particulars'?

Core category / concept

— Bring 'slowing' into here.

* Consider bringing Depression under here.

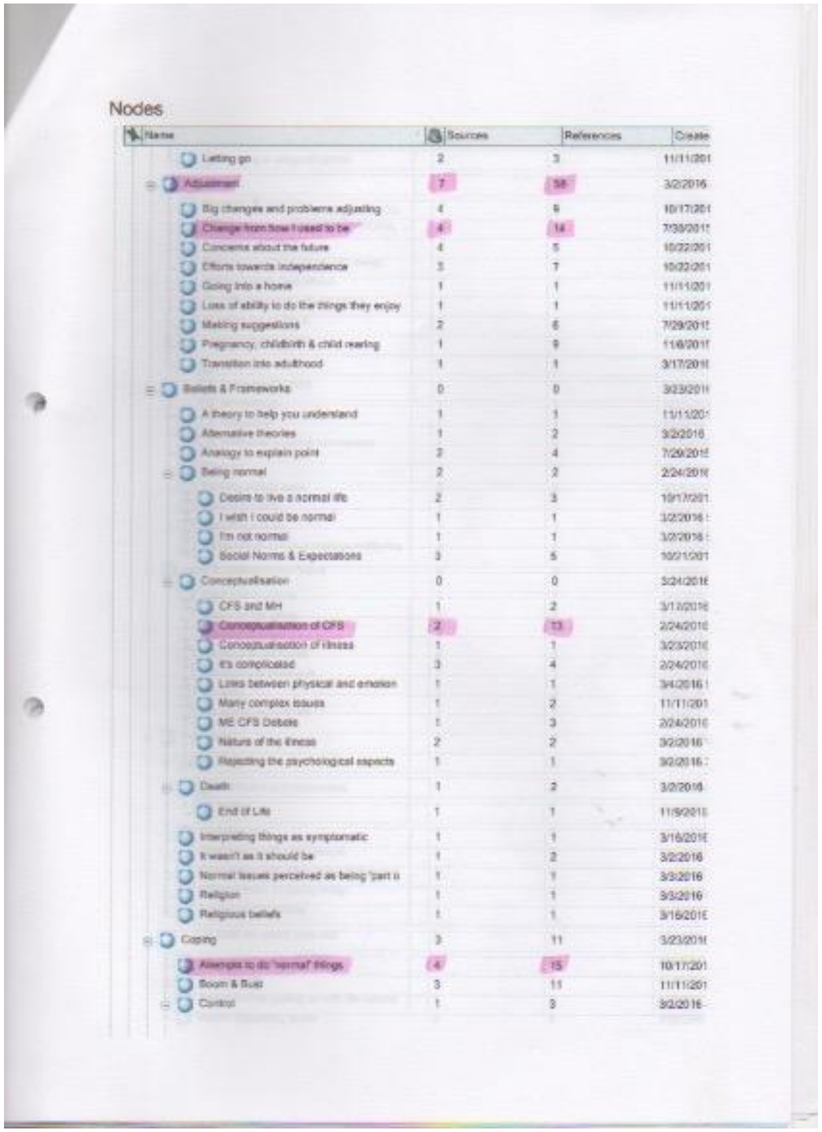
Consider moving into 'processing'?

are these opposite things?

??! check this

— Consider grouping with diagnosis.

Consider making a concept

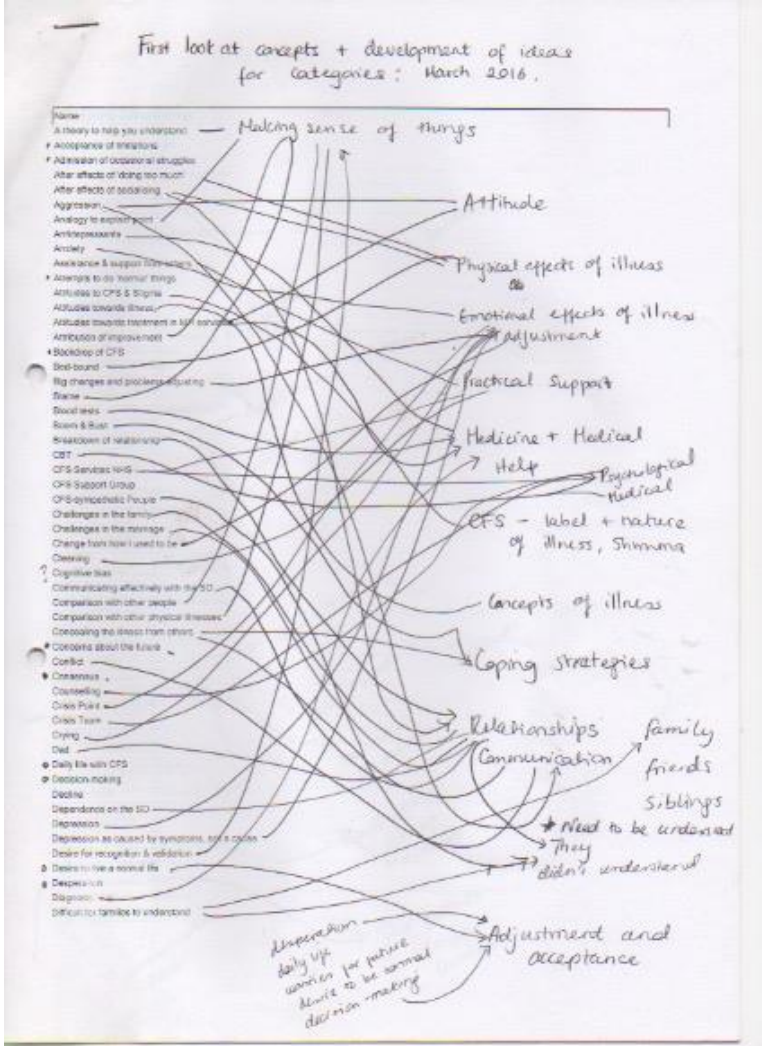


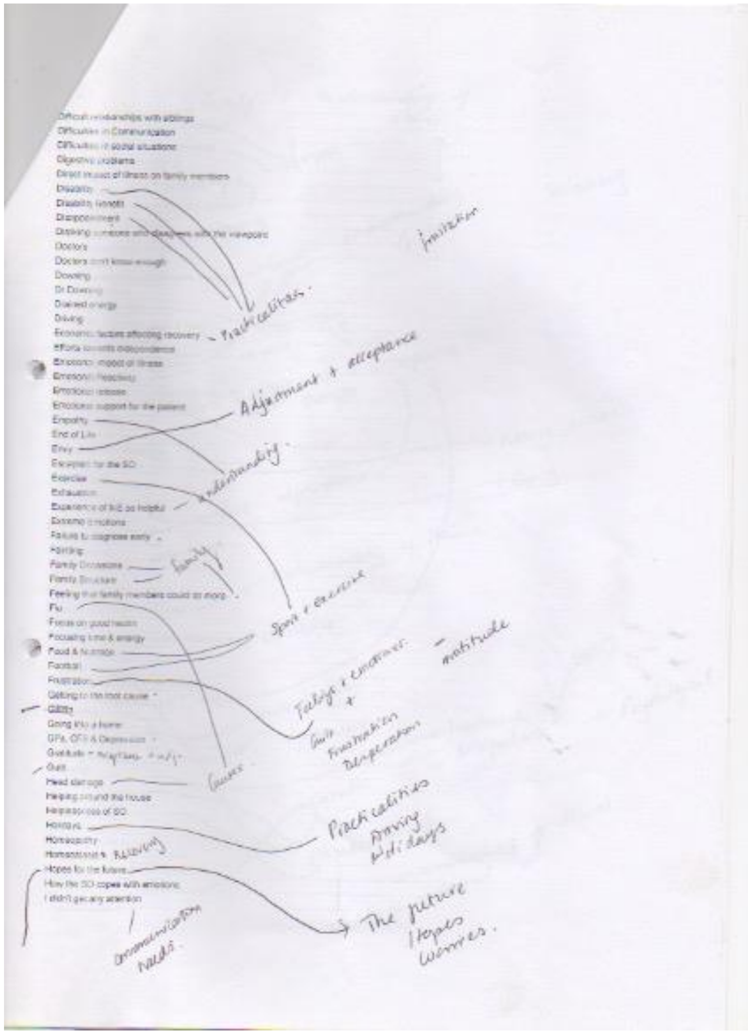
Nodes

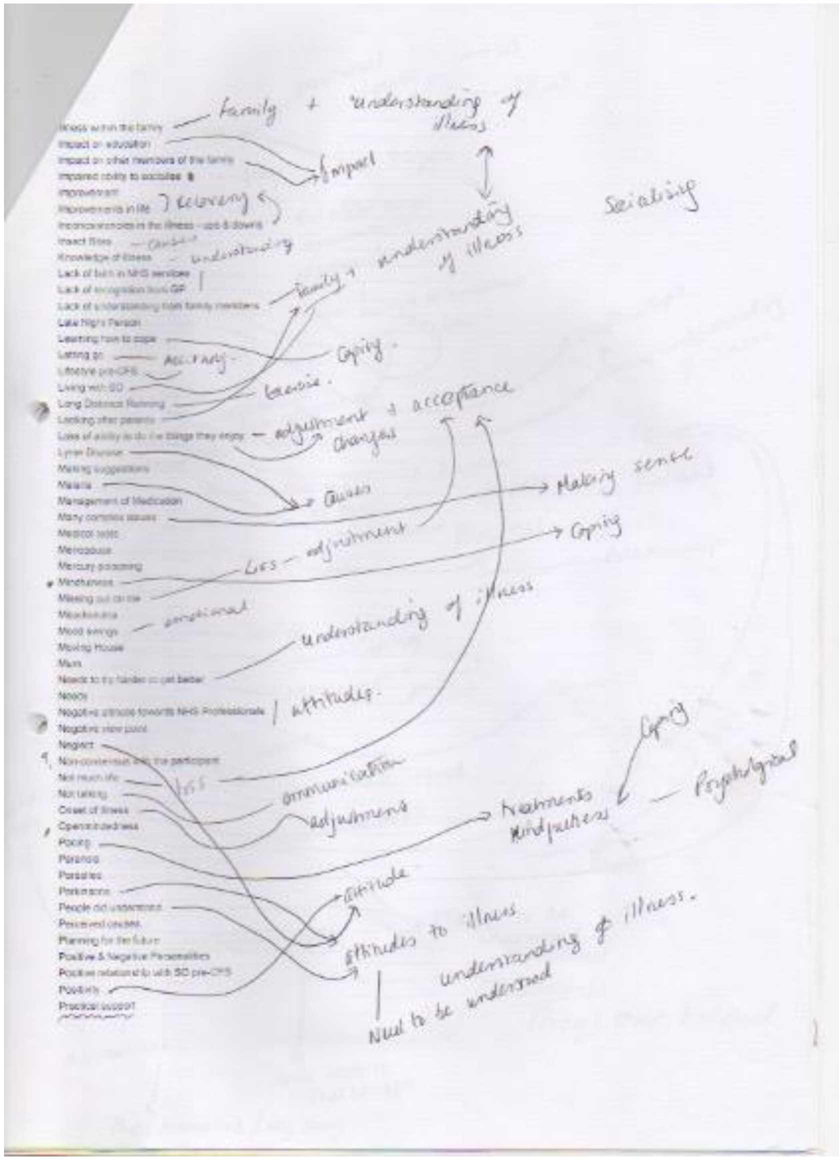
Name	Sources	References	Create
Letting go	2	3	11/11/201
Adjustment	7	58	3/2/2016
Big changes and problems adjusting	4	9	10/17/201
Change from how I used to be	4	14	7/30/201
Concerns about the future	4	5	10/22/201
Efforts towards independence	3	7	10/22/201
Going into a home	1	1	11/11/201
Loss of ability to do the things they enjoy	1	1	11/11/201
Making suggestions	2	6	7/29/201
Pregnancy, childbirth & child rearing	1	9	11/6/201
Transition into adulthood	1	1	3/17/201
Beliefs & Frameworks	0	0	3/23/201
A theory to help you understand	1	1	11/11/20
Alternative theories	1	2	3/2/2016
Analogy to explain point	2	4	7/29/201
Being normal	2	2	2/24/201
Desire to live a normal life	2	3	10/17/201
I wish I could be normal	1	1	3/2/2016
I'm not normal	1	1	3/2/2016
Social Norms & Expectations	3	5	10/21/201
Conceptualisation	0	0	3/24/201
CFS and MH	1	2	3/12/201
Conceptualisation of CFS	2	13	2/24/201
Conceptualisation of illness	1	1	3/23/201
It's complicated	3	4	2/24/201
Links between physical and emotion	1	1	3/4/2016
Many complex issues	1	2	11/11/201
ME CFS Debate	1	3	2/24/201
Nature of the illness	2	2	3/2/2016
Rejecting the psychological aspects	1	1	3/2/2016
Death	1	2	3/2/2016
End of Life	1	1	11/9/201
Interpreting things as symptomatic	1	1	3/16/201
It wasn't as it should be	1	2	3/2/2016
Normal issues perceived as being 'part o	1	1	3/3/2016
Religion	1	1	3/3/2016
Religious beliefs	1	1	3/16/201
Coping	3	11	3/23/201
Attempts to do "normal" things	4	15	10/17/201
Boom & Bust	3	11	11/11/201
Control	1	3	3/2/2016

Appendix 12. Selective Coding

Re-thinking and refining codes – Examples Phase 1

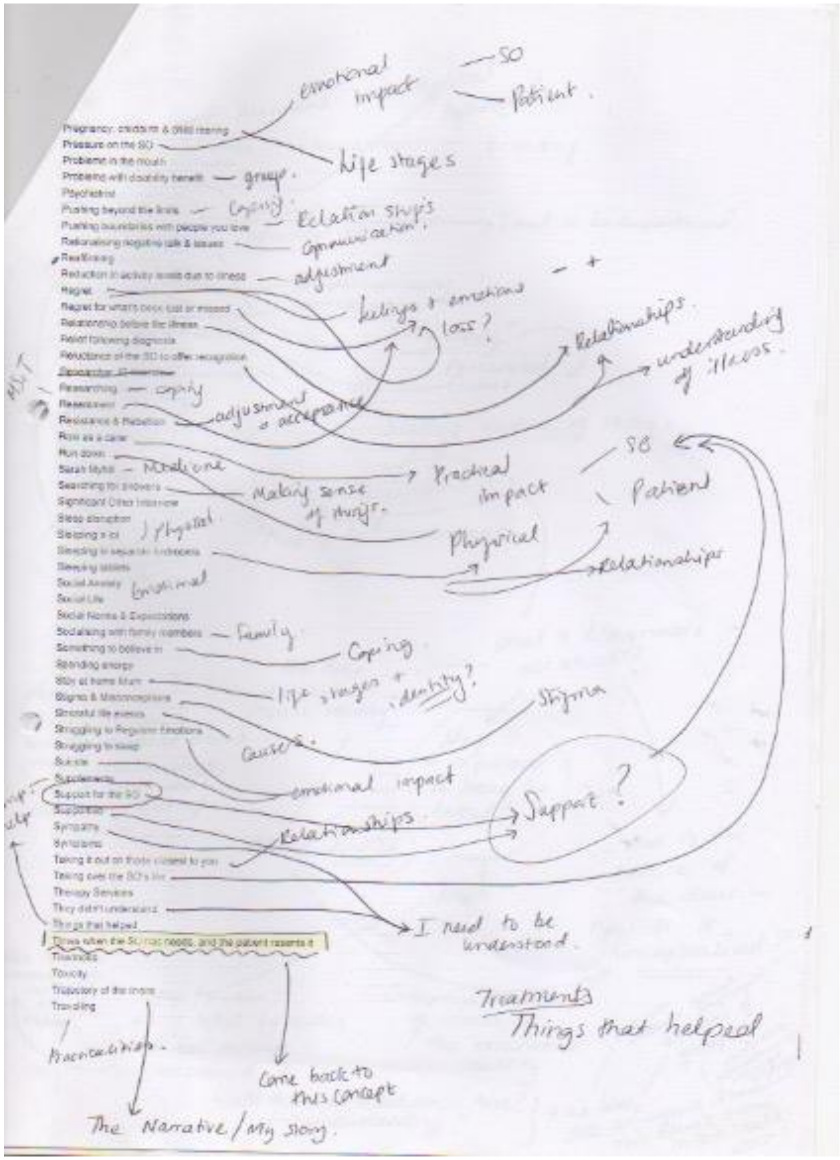






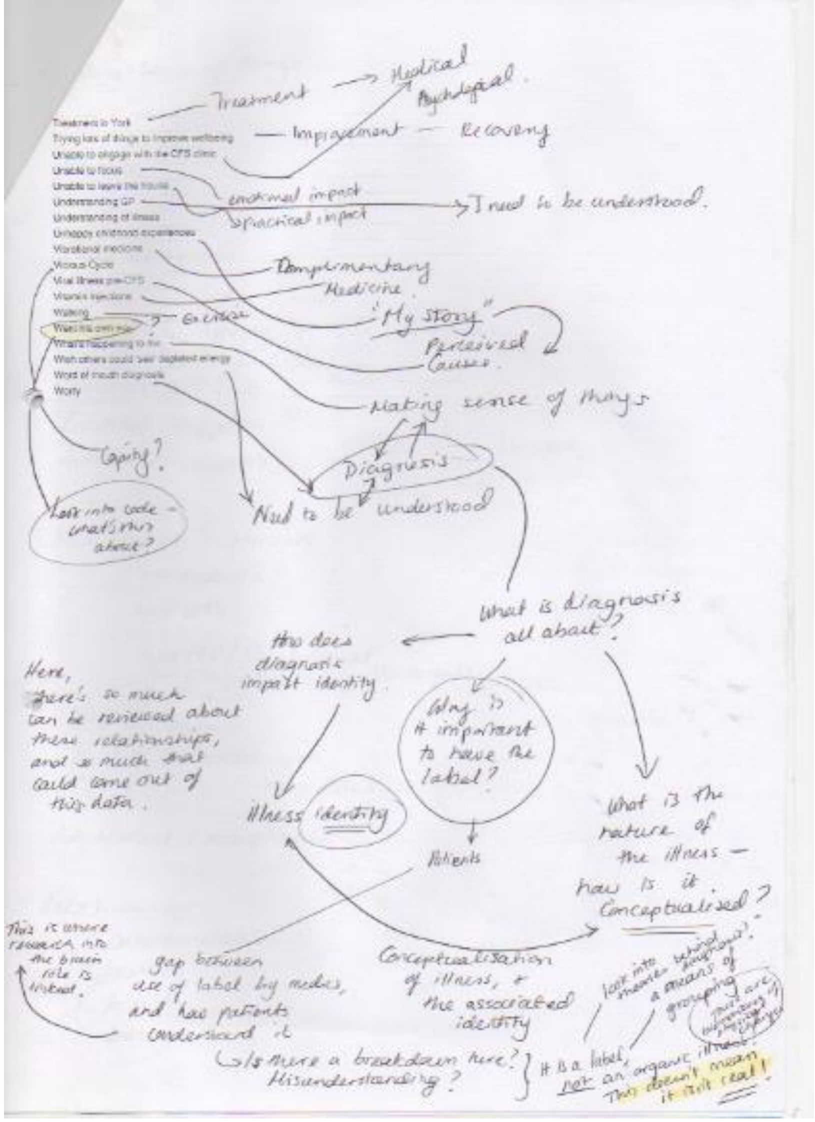
Illness within the family — family + understanding of illness
Impact on education
Impact on other members of the family
Impaired ability to socialise
Improvement
Improvements in life] Recovery
Inconsistencies in the illness - ups & downs
Insect Bites — Causes
Knowledge of illness — understanding
Lack of faith in NHS services
Lack of recognition from GP
Lack of understanding from family members
Late Night Person
Learning how to cope
Letting go — Acc + Adj
Lifestyle pre-CFS
Living with SO
Long Distance Running
Looking after parents
Loss of ability to do the things they enjoy — adjustment + acceptance
Lyme Disease
Making suggestions
Malaria
Management of Medication
Many complex issues
Medical tests
Menopause
Mercury poisoning
Mindfulness
Missing out on life
Misdiagnosis
Mood swings — emotional
Moving House
Mum
Needs to try harder to get better
Needs
Negative attitude towards NHS Professionals / attitudes.
Negative view point
Neglect
Non-consensus with the participant
Not much life
Not talking
Onset of illness
Openmindedness
Pacing
Paranoia
Parasites
Parkinsons
People did understand
Perceived causes
Planning for the future
Positive & Negative Personalities
Positive relationship with SO pre-CFS
Positivity
Practical support

impact
Socialising
family + understanding of illness
Coping.
Exercise.
changes
Causes
Making sense
Coping
LFS — adjustment
understanding of illness
Coping
Psychological
LFS
communication
adjustment
treatments
Mindfulness
attitude
attitudes to illness
understanding of illness.
Need to be understood

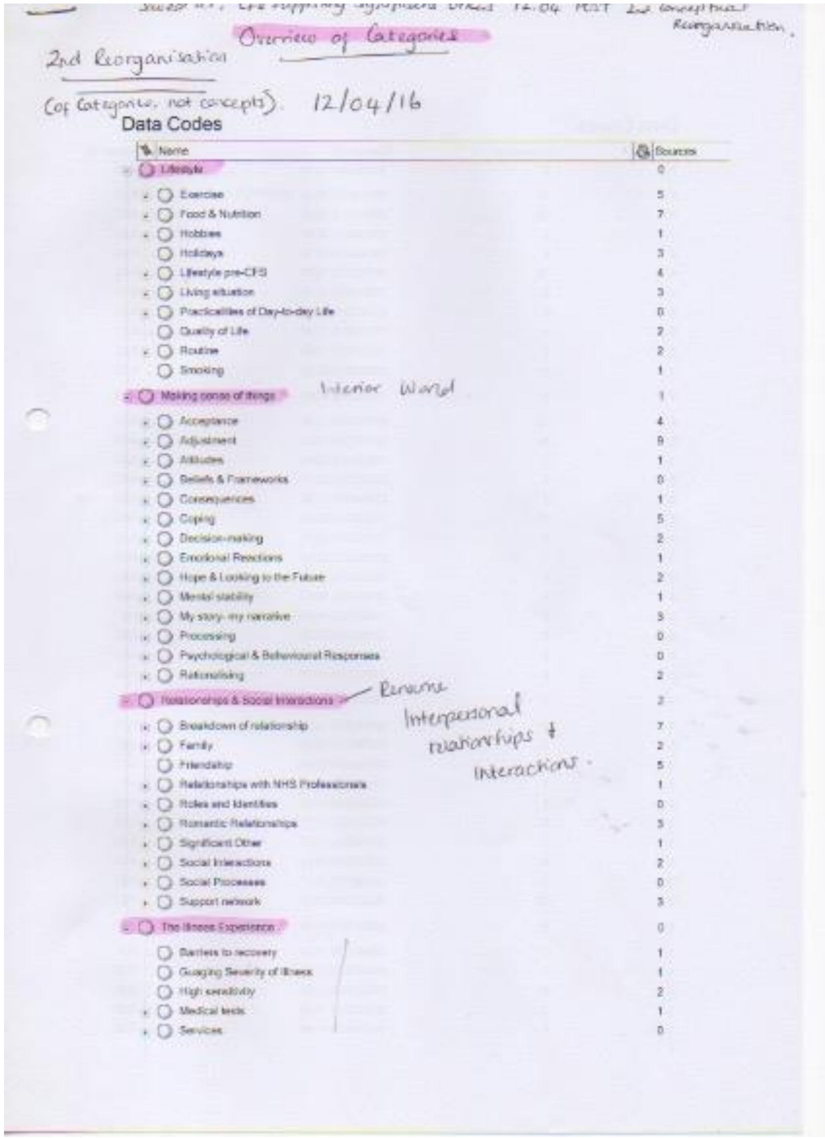


Pregnancy, childbirth & child rearing
Pressure on the SO
Problems in the mouth
Problems with disability benefit — group.
Psychiatrist
Pushing beyond the limits — Coping
Pushing boundaries with people you love — Relationships
Rationalising negative talk & issues — Communication.
Realising
Reduction in activity levels due to illness — adjustment
Regret
Regret for what's been lost or missed
Relationship before the illness
Relief following diagnosis
Reluctance of the SO to offer recognition
~~Researcher SO interview~~
Researching — Coping
Resentment
Resistance & Rebellion
Role as a carer
Run down
Sarah Myhill — Medicine
Searching for answers
Significant Other Interview
Sleep disruption
Sleeping a lot } Physical
Sleeping in separate bedrooms
Sleeping tablets
Social Anxiety Emotional
Social Life
Social Norms & Expectations
Socialising with family members — family
Something to believe in
Spending energy
Stay at home Mum
Stigma & Misconceptions
Stressful life events
Struggling to Regulate Emotions
Struggling to sleep
Suicide
Supplements
Support for the SO
Supporter
Sympathy
Symptoms
Taking it out on those closest to you
Taking over the SO's life
Therapy Services
They didn't understand
Things that helped
Times when the SO has needs, and the patient resents it
Treatments
Toxicity
Trajectory of the illness
Travelling

emotional impact — SO
— Patient.
Life stages
— +
feelings + emotions
loss?
Relationships.
understanding of illness.
adjustment + acceptance
Practical impact
— SO
Patient
Making sense of things.
Physical
Relationships
Coping.
life stages + identity?
Stigma
Causes.
emotional impact
Relationships
Support?
I need to be understood.
Treatments
Things that helped
Procrastination.
Come back to this concept
The Narrative / My story.


Treatment in York
Trying lots of things to improve wellbeing
Unable to engage with the CFS clinic
Unable to focus
Unable to leave the house
Understanding GP
Understanding of illness
Unhappy childhood experiences
Vibrational medicine
Vicious Cycle
Viral illness pre-CFS
Vitamin injections
Walking
Want my own life
What's happening to me
Wish others could 'see' depleted energy
Word of mouth diagnosis
Worry
Treatment
Medical
Psychological.
Improvement
Recovery
emotional impact
practical impact
I need to be understood.
Complementary
Medicine
Exercise
?
"My story"
Perceived
Causes.
Making sense of things
Coping?
Look into code - what's this about?
Diagnosis
Need to be understood
What is diagnosis all about?
How does diagnosis impact identity.
Why is it important to have the label?
Illness identity
Patients
What is the nature of the illness - how is it Conceptualised?
Here, there's so much can be reviewed about these relationships, and so much that could come out of this data.
This is where research into the brain role is instead.
gap between use of label by medics, and how patients understand it
Conceptualisation of illness, + the associated identity
Is there a breakdown here? Misunderstanding?
It is a label, not an organic illness.
This doesn't mean it isn't real!
look into theories behind diagnosis?
a means of grouping
[illegible]

Rethinking & Refining Codes Examples from Phase 2 (Example is 6 of 18 pages printed from NVivo)



[illegible] 12.04 Post 2nd conceptual Reorganisation.

Overview of Categories

2nd Reorganisation

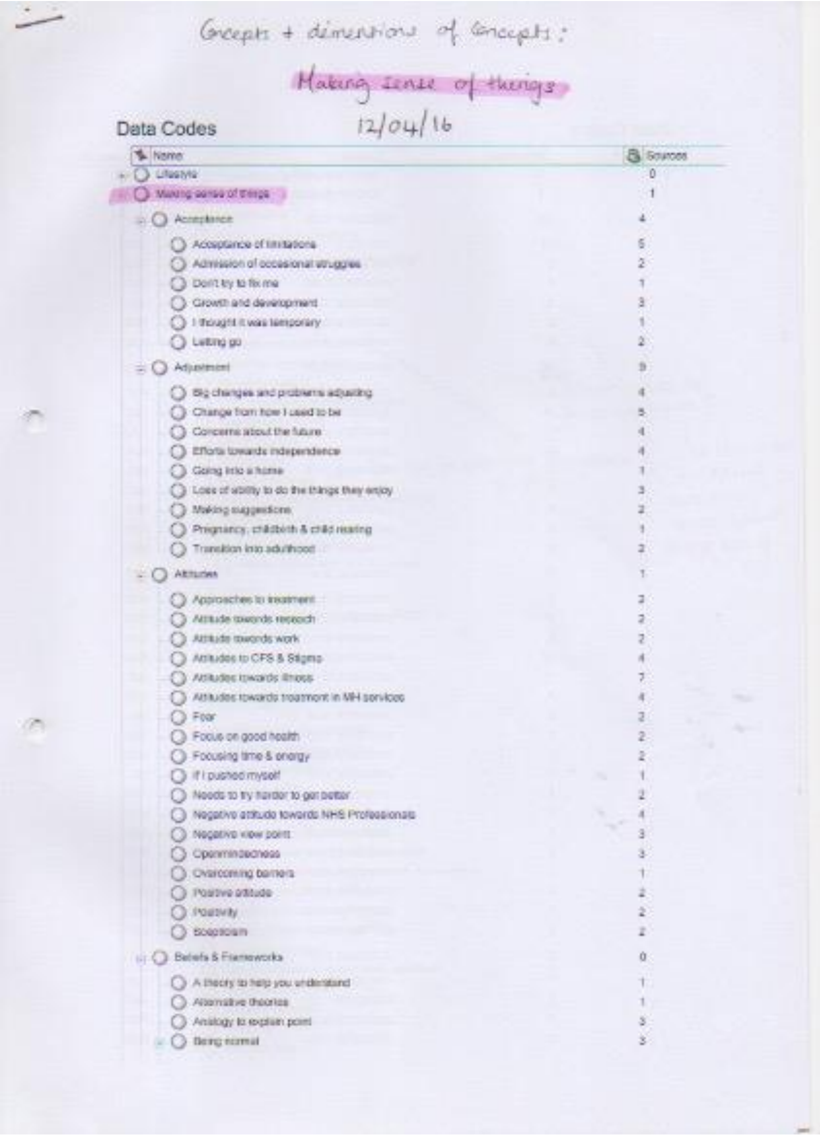
(of Categories, not concepts). 12/04/16

Data Codes

Name	Sources
Lifestyle	0
Exercise	5
Food & Nutrition	7
Hobbies	1
Holidays	3
Lifestyle pre-CFS	4
Living situation	3
Practicalities of Day-to-day Life	0
Quality of Life	2
Routine	2
Smoking	1
Making sense of things	1
Acceptance	4
Adjustment	9
Attitudes	1
Beliefs & Frameworks	0
Consequences	1
Coping	5
Decision-making	2
Emotional Reactions	1
Hope & Looking to the Future	2
Mental stability	1
My story- my narrative	3
Processing	0
Psychological & Behavioural Responses	0
Rationalising	2
Relationships & Social Interactions	2
Breakdown of relationship	7
Family	2
Friendship	5
Relationships with NHS Professionals	1
Roles and Identities	0
Romantic Relationships	3
Significant Other	1
Social Interactions	2
Social Processes	0
Support network	3
The Illness Experience	0
Barriers to recovery	1
Guaging Severity of Illness	1
High sensitivity	2
Medical tests	1
Services	0

Interior World

Rename Interpersonal relationships & interactions.



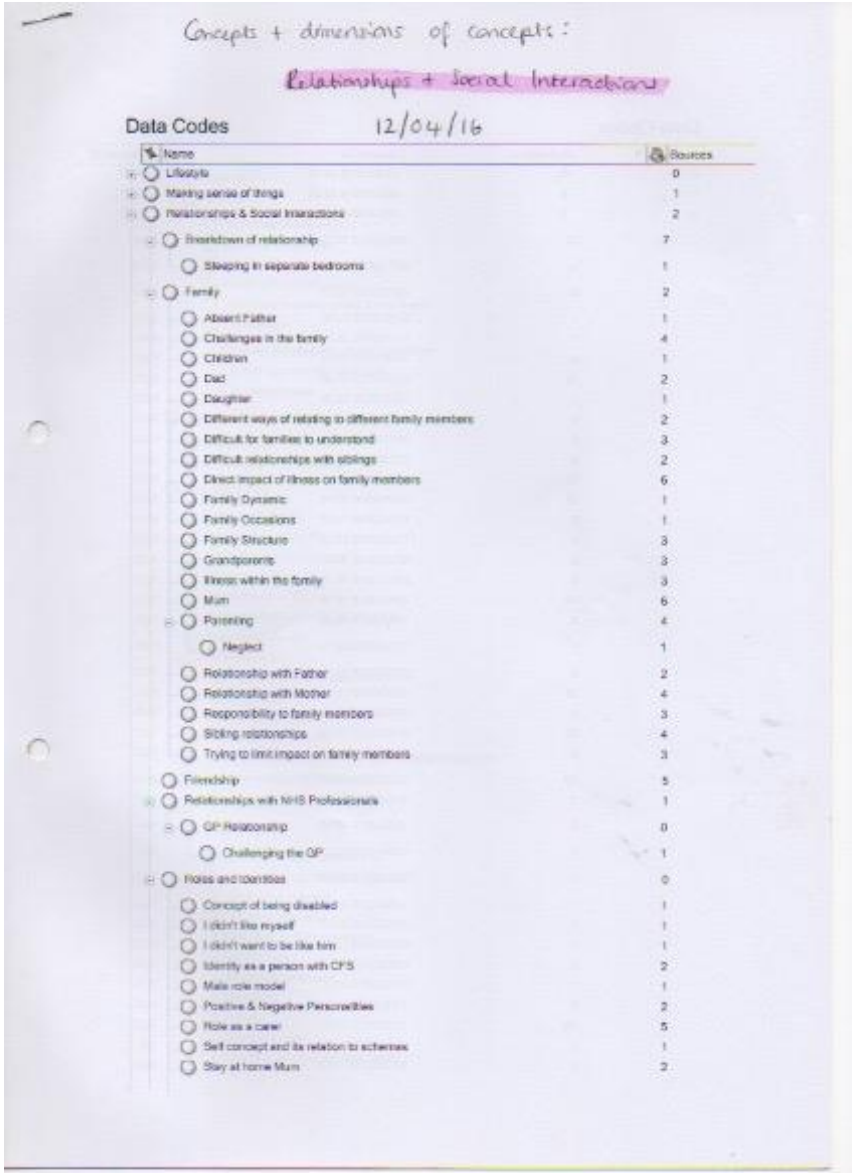
Concepts + dimensions of Concepts:

Making sense of things

12/04/16

Data Codes

Name	Sources
Lifestyle	0
Making sense of things	1
Acceptance	4
Acceptance of limitations	5
Admission of occasional struggles	2
Don't try to fix me	1
Growth and development	3
I thought it was temporary	1
Letting go	2
Adjustment	9
Big changes and problems adjusting	4
Change from how I used to be	5
Concerns about the future	4
Efforts towards independence	4
Going into a home	1
Loss of ability to do the things they enjoy	3
Making suggestions	2
Pregnancy, childbirth & child rearing	1
Transition into adulthood	2
Attitudes	1
Approaches to treatment	2
Attitude towards research	2
Attitude towards work	2
Attitudes to CFS & Stigma	4
Attitudes towards illness	7
Attitudes towards treatment in MH services	4
Fear	2
Focus on good health	2
Focusing time & energy	2
If I pushed myself	1
Needs to try harder to get better	2
Negative attitude towards NHS Professionals	4
Negative view point	3
Openmindedness	3
Overcoming barriers	1
Positive attitude	2
Positivity	2
Scepticism	2
Beliefs & Frameworks	0
A theory to help you understand	1
Alternative theories	1
Analogy to explain point	3
Being normal	3



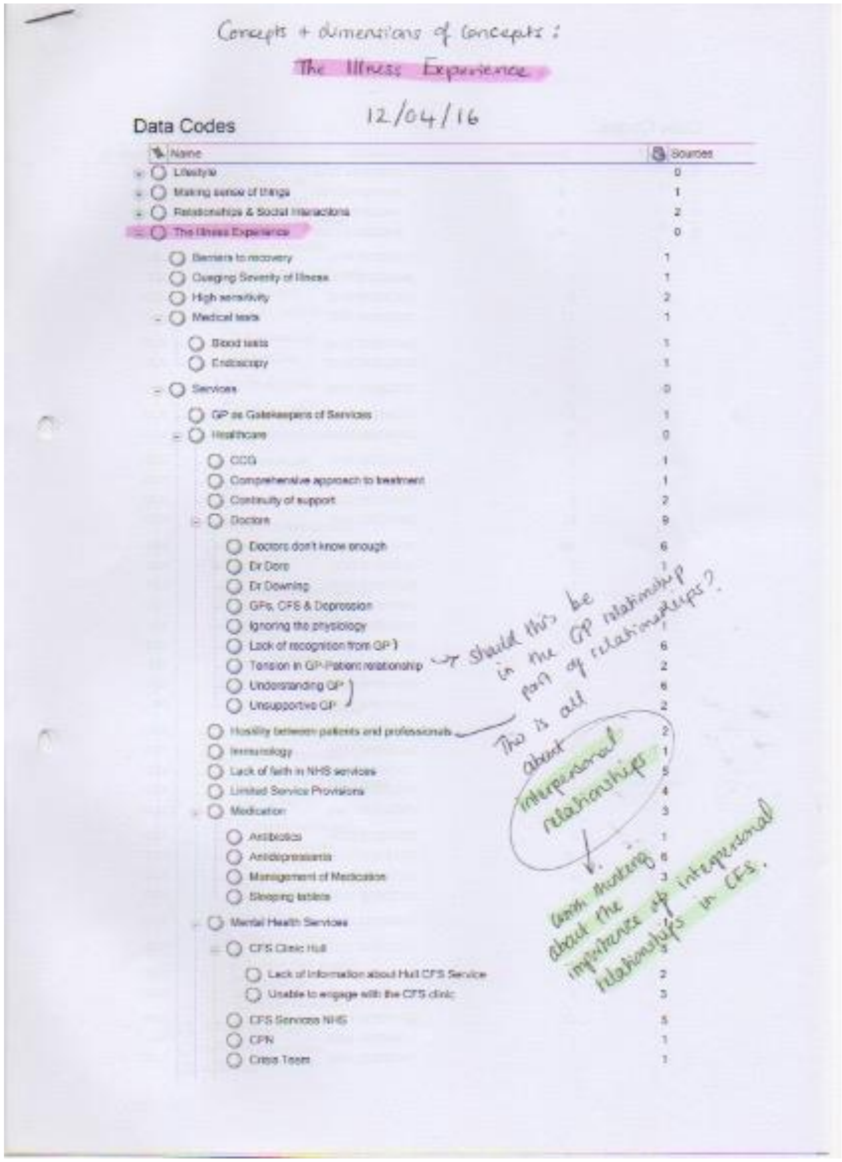
Concepts + dimensions of concepts:

Relationships + Social Interactions

Data Codes

12/04/16

Name	Sources
Lifestyle	0
Making sense of things	1
Relationships & Social Interactions	2
Breakdown of relationship	7
Sleeping in separate bedrooms	1
Family	2
Absent Father	1
Challenges in the family	4
Children	1
Dad	2
Daughter	1
Different ways of relating to different family members	2
Difficult for families to understand	3
Difficult relationships with siblings	2
Direct impact of illness on family members	6
Family Dynamic	1
Family Occasions	1
Family Structure	3
Grandparents	3
Illness within the family	3
Mum	6
Parenting	4
Neglect	1
Relationship with Father	2
Relationship with Mother	4
Responsibility to family members	3
Sibling relationships	4
Trying to limit impact on family members	3
Friendship	5
Relationships with NHS Professionals	1
GP Relationship	0
Challenging the GP	1
Roles and Identities	0
Concept of being disabled	1
I didn't like myself	1
I didn't want to be like him	1
Identity as a person with CFS	2
Male role model	1
Positive & Negative Personalities	2
Role as a carer	5
Self concept and its relation to schemas	1
Stay at home Mum	2



Concepts + dimensions of concepts:

The Illness Experience

Data Codes

12/04/16

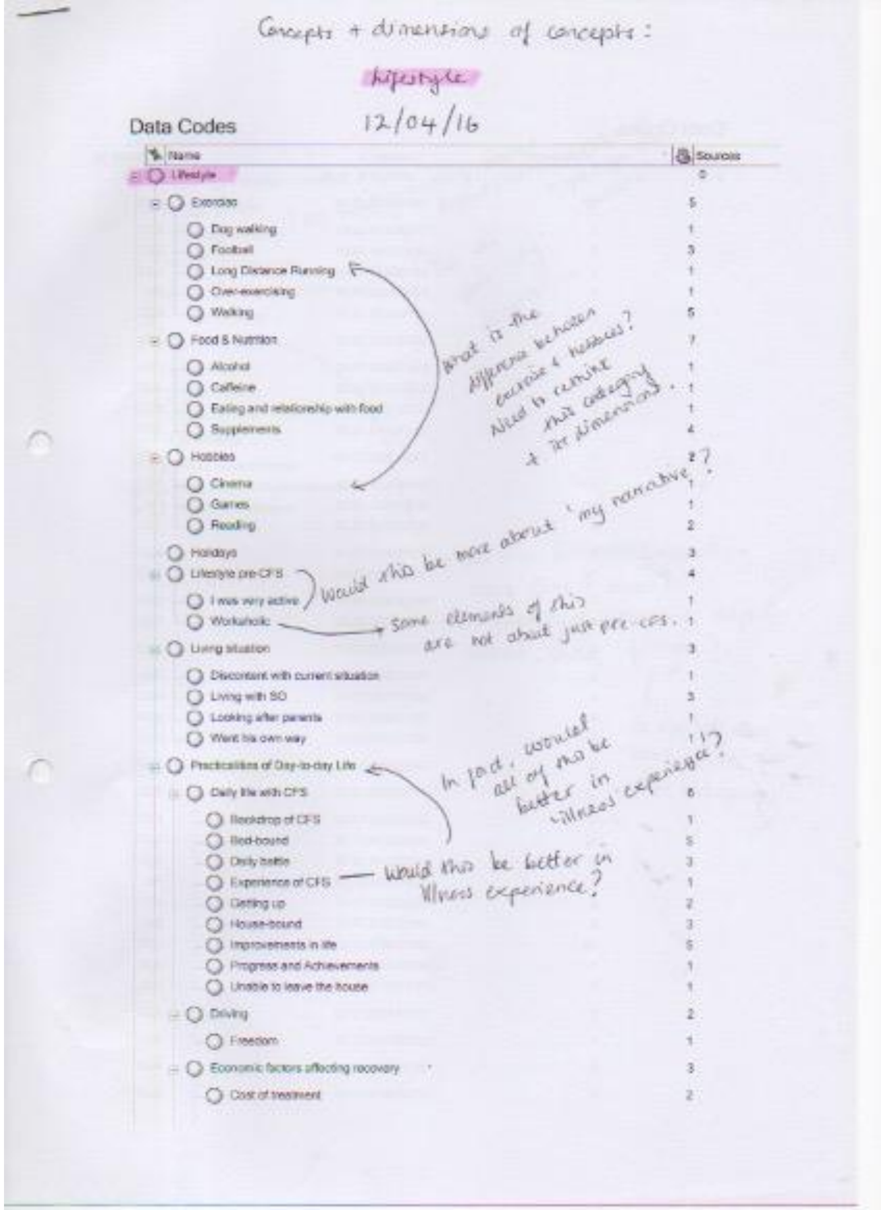
Name	Sources
Lifestyle	0
Making sense of things	1
Relationships & Social Interactions	2
The Illness Experience	0
Barriers to recovery	1
Gauging Severity of Illness	1
High sensitivity	2
Medical tests	1
Blood tests	1
Endoscopy	1
Services	0
GP as Gatekeepers of Services	1
Healthcare	0
CCG	1
Comprehensive approach to treatment	1
Continuity of support	2
Doctors	9
Doctors don't know enough	6
Dr Doro	1
Dr Downing	[illegible]
GPs, CFS & Depression	[illegible]
Ignoring the physiology	1
Lack of recognition from GP	6
Tension in GP-Patient relationship	2
Understanding GP	6
Unsupportive GP	2
Hostility between patients and professionals	2
Immunology	1
Lack of faith in NHS services	5
Limited Service Provisions	4
Medication	3
Antibiotics	1
Antidepressants	6
Management of Medication	3
Sleeping tablets	[illegible]
Mental Health Services	1
CFS Clinic Hull	3
Lack of information about Hull CFS Service	2
Unable to engage with the CFS clinic	3
CFS Services NHS	5
CPN	1
Crisis Team	1

→ should this be in the GP relationship part of relationships?

This is all about interpersonal relationships

↓

Worth thinking about the importance of interpersonal relationships in CFS.



Concepts + dimensions of concepts:

Lifestyle

12/04/16

Data Codes

Name	Sources
Lifestyle	0
Exercise	5
Dog walking	1
Football	3
Long Distance Running	1
Over-exercising	1
Walking	5
Food & Nutrition	7
Alcohol	1
Caffeine	1
Eating and relationship with food	1
Supplements	4
Hobbies	2
Cinema	1
Games	1
Reading	2
Holidays	3
Lifestyle pre-CFS	4
I was very active	1
Workaholic	1
Living situation	3
Discontent with current situation	1
Living with SO	3
Looking after parents	1
Went his own way	1
Practicalities of Day-to-day Life	
Daily life with CFS	6
Backdrop of CFS	1
Bed-bound	5
Daily battle	3
Experience of CFS	1
Getting up	2
House-bound	3
Improvements in life	5
Progress and Achievements	1
Unable to leave the house	1
Driving	2
Freedom	1
Economic factors affecting recovery	3
Cost of treatment	2

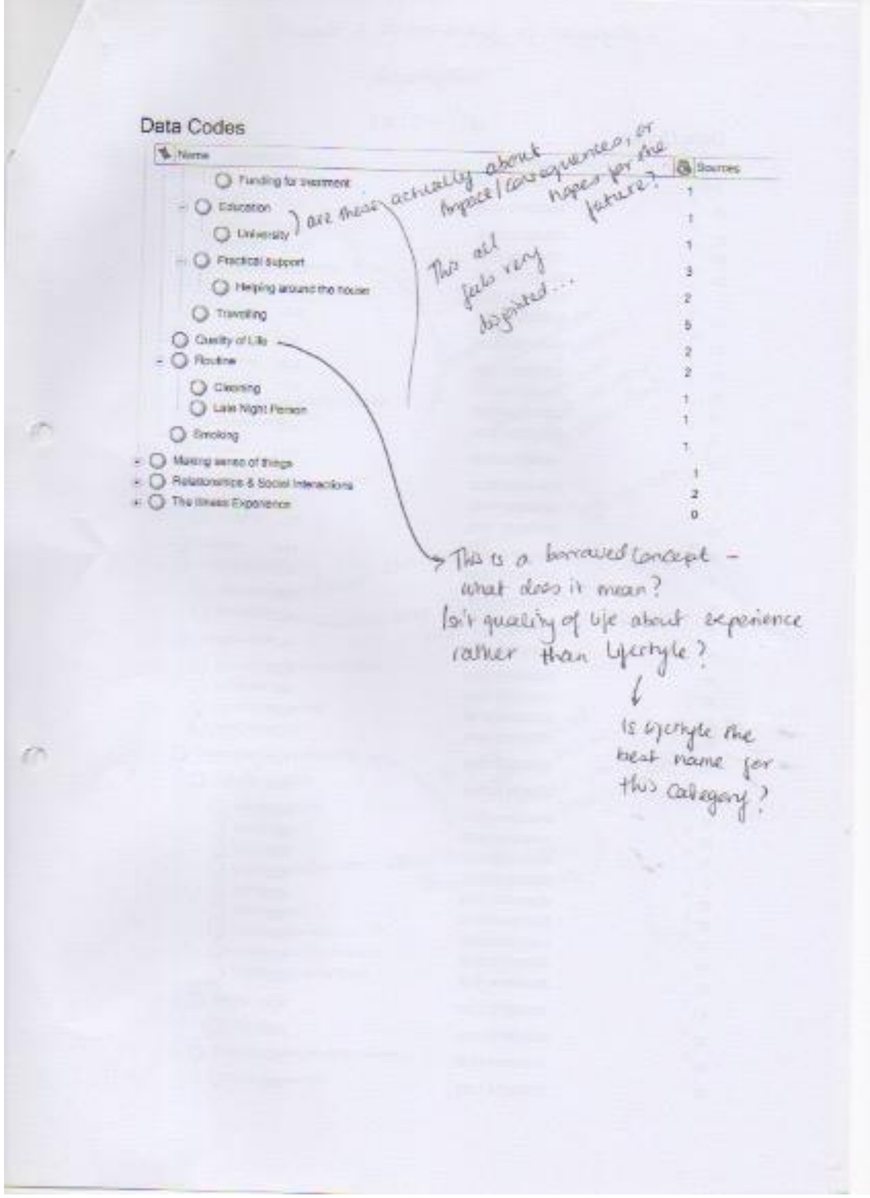
What is the difference between exercise & hobbies? Need to examine this category + its dimensions.

Would this be more about 'my narrative'?

Some elements of this are not about just pre-CFS.

In fact, would all of this be better in illness experience?

Would this be better in Illness experience?



Data Codes

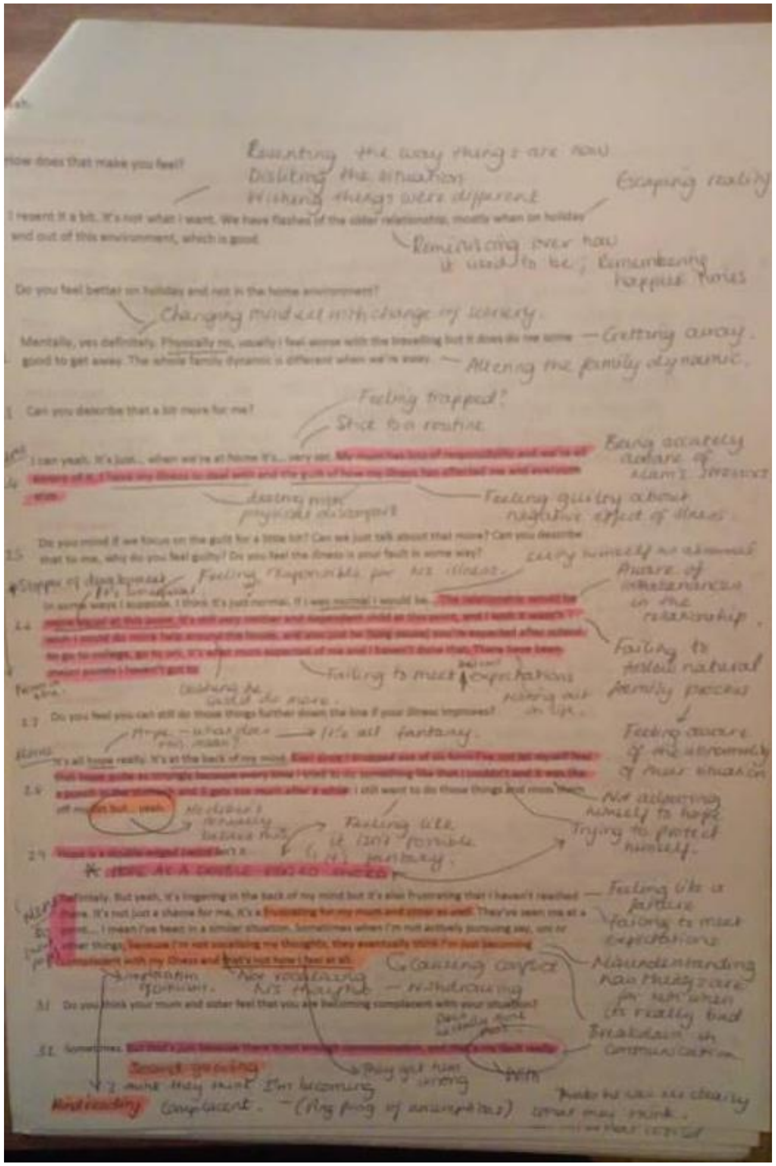
Name	Sources
Funding for treatment	1
Education	1
University	1
Practical support	3
Helping around the house	2
Travelling	5
Quality of Life	2
Routine	2
Cleaning	1
Late Night Person	1
Smoking	1
Making sense of things	1
Relationships & Social Interactions	2
The Illness Experience	0

are these actually about impact/consequences, or hopes for the future?

This all feels very disjointed...

This is a borrowed concept – what does it mean? Isn't quality of life about experience rather than lifestyle?

Is lifestyle the best name for this category?



How does that make you feel?

Resenting the way things are now
Disliking the situation
Wishing things were different

I resent it a bit. It's not what I want. We have flashes of the older relationship, mostly when on holiday and out of this environment, which is good

Escaping reality

Reminiscing over how it used to be; Remembering happier times

Do you feel better on holiday and not in the home environment?

Changing mindset with change of scenery.

Mentally, yes definitely. Physically no, usually I feel worse with the travelling but it does do me some good to get away. The whole family dynamic is different when we're away.

— Getting away.

— Altering the family dynamic.

Can you describe that a bit more for me?

Feeling trapped?
Stuck to a routine

I can yeah. It's just... when we're at home it's... very set. [illegible]

Being acutely aware of [illegible]

[illegible] physical discomfort

Feeling guilty about negative effect of illness

Do you mind if we focus on the guilt for a little bit? Can we just talk about that more? Can you describe that to me, why do you feel guilty? Do you feel the illness is your fault in some way?

Feeling responsible for his illness.

In some ways I suppose. I think it's just normal. If I was normal I would be. [illegible]

Aware of [illegible] in the relationship.

Failing to follow natural family process

Failing to meet expectations

Wishing he could do more.

Missing out on life.

Do you feel you can still do those things further down the line if your illness improves?

Hope — what does this mean? → It's all fantasy.

Feeling aware of the [illegible] of their situation

It's all hope really. It's at the back of my mind. [illegible] I still want to do those things and more [illegible]

Not allowing himself to hope

Trying to protect himself.

Feeling like it isn't possible. (It's fantasy.)

Hope is a double edged sword isn't it.

* HOPE: A DOUBLE EDGED SWORD

[illegible] But yeah, it's lingering in the back of my mind but it's also frustrating that I haven't reached there. It's not just a shame for me, it's a frustrating for my mum and sister as well. They've seen me at a point... I mean I've been in a similar situation. Sometimes when I'm not actively pursuing say, uni or other things, because I'm not vocalising my thoughts, they eventually think I'm just becoming complacent with my illness and that's not how I feel at all.

— Feeling like a failure

Failing to meet expectations

Causing conflict

Misunderstanding

Not vocalising his thoughts — withdrawing

Do you think your mum and sister feel that you are becoming complacent with your situation?

Breakdown in communication

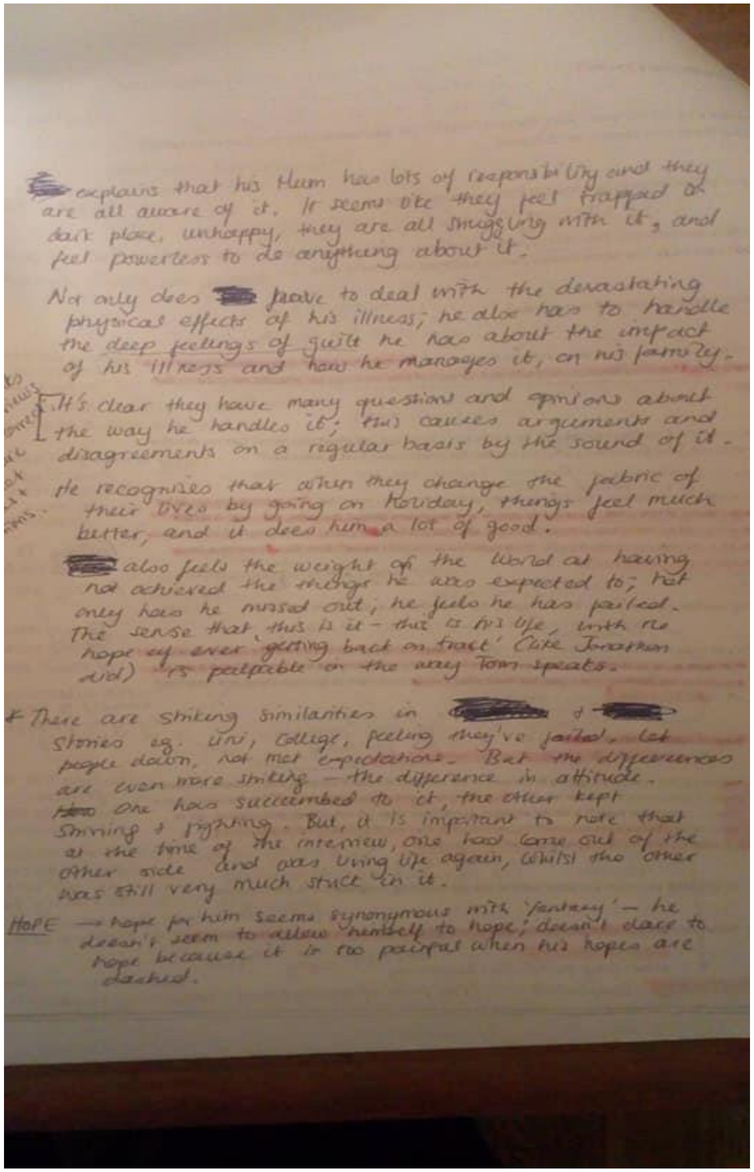
Sometimes. [illegible]

Second guessing

They got him wrong

Mindreading

I think they think I'm becoming complacent. — (Ping pong of assumptions)



[redacted] explains that his Mum has lots of responsibility and they are all aware of it. It seems like they feel trapped & dark place, unhappy, they are all struggling with it, and feel powerless to do anything about it.

Not only does [redacted] have to deal with the devastating physical effects of his illness; he also has to handle the deep feelings of guilt he has about the impact of his illness and how he manages it, on his family.

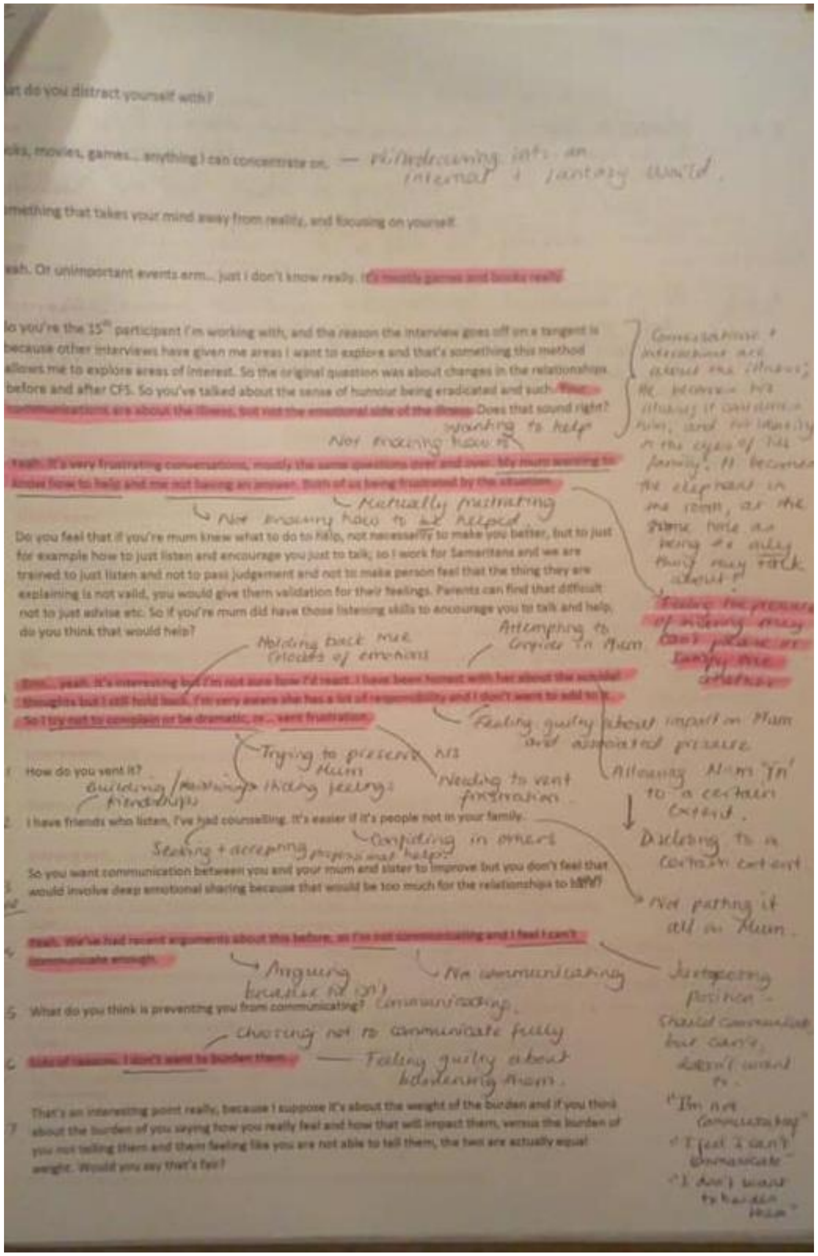
It's clear they have many questions and opinions about the way he handles it; this causes arguments and disagreements on a regular basis by the sound of it.

He recognises that when they change the fabric of their lives by going on holiday, things feel much better, and it does him a lot of good.

[redacted] also feels the weight of the world at having not achieved the things he was expected to; not only has he missed out, he feels he has failed. The sense that this is it – this is his life, with no hope of ever 'getting back on track' (like Jonathan did) is palpable in the way Tom speaks.

* There are striking similarities in [redacted] & [redacted] stories eg. uni, college, feeling they've failed, let people down, not met expectations. But the differences are even more striking – the difference in attitude. ~~Hm~~ One has succumbed to it, the other kept swimming + fighting. But, it is important to note that at the time of the interview, one had come out of the other side and was living life again, whilst the other was still very much stuck in it.

HOPE → hope for him seems synonymous with 'fantasy' – he doesn't seem to allow himself to hope; doesn't dare to hope because it is too painful when his hopes are dashed.



at do you distract yourself with?

oks, movies, games... anything I can concentrate on. — Withdrawing into an internal + fantasy world.

mething that takes your mind away from reality, and focusing on yourself.

eah. Or unimportant events erm... just I don't know really. It's mostly games and books really.

o you're the 15th participant I'm working with, and the reason the interview goes off on a tangent is because other interviews have given me areas I want to explore and that's something this method allows me to explore areas of interest. So the original question was about changes in the relationships before and after CFS. So you've talked about the sense of humour being eradicated and such. Your communications are about the illness, but not the emotional side of the illness. Does that sound right?

Conversations + interactions are about the illness; he becomes his illness if considered him, and his identity in the eyes of his family. It becomes the elephant in the room, at the same time as being the only thing they talk about?

Wanting to help

Not knowing how to

Yeah. It's very frustrating conversations, mostly the same questions over and over. My mum wanting to know how to help and me not having an answer. Both of us being frustrated by the situation.

Mutually frustrating

Not knowing how to be helped.

Feeling the pressure of knowing they can't please or satisfy the other

Do you feel that if you're mum knew what to do to help, not necessarily to make you better, but to just for example how to just listen and encourage you just to talk; so I work for Samaritans and we are trained to just listen and not to pass judgement and not to make person feel that the thing they are explaining is not valid, you would give them validation for their feelings. Parents can find that difficult not to just advise etc. So if you're mum did have those listening skills to encourage you to talk and help, do you think that would help?

Holding back the floods of emotions

Attempting to confide in Mum

Erm... yeah. It's interesting but I'm not sure how I'd react. I have been honest with her about the suicidal thoughts but I still hold back. I'm very aware she has a lot of responsibility and I don't want to add to it. So I try not to complain or be dramatic, or... vent frustration.

Feeling guilty about impact on Mum and associated pressure

Trying to preserve his Mum

Needing to vent frustration.

Allowing Mum 'in' to a certain extent.

↓

Disclosing to a certain extent

1 How do you vent it?

Building/maintaining friendships

Sharing feelings

2 I have friends who listen, I've had counselling. It's easier if it's people not in your family.

Seeking + accepting professional help?

Confiding in others

Not putting it all on Mum.

3 So you want communication between you and your mum and sister to improve but you don't feel that would involve deep emotional sharing because that would be too much for the relationships to have?

4 Yeah. We've had recent arguments about this before, so I'm not communicating and I feel I can't communicate enough.

Arguing because he isn't communicating.

Not communicating

Juxtaposing position – should communicate but can't, doesn't want to.

5 What do you think is preventing you from communicating?

Choosing not to communicate fully

6 Lots of reasons. I don't want to burden them.

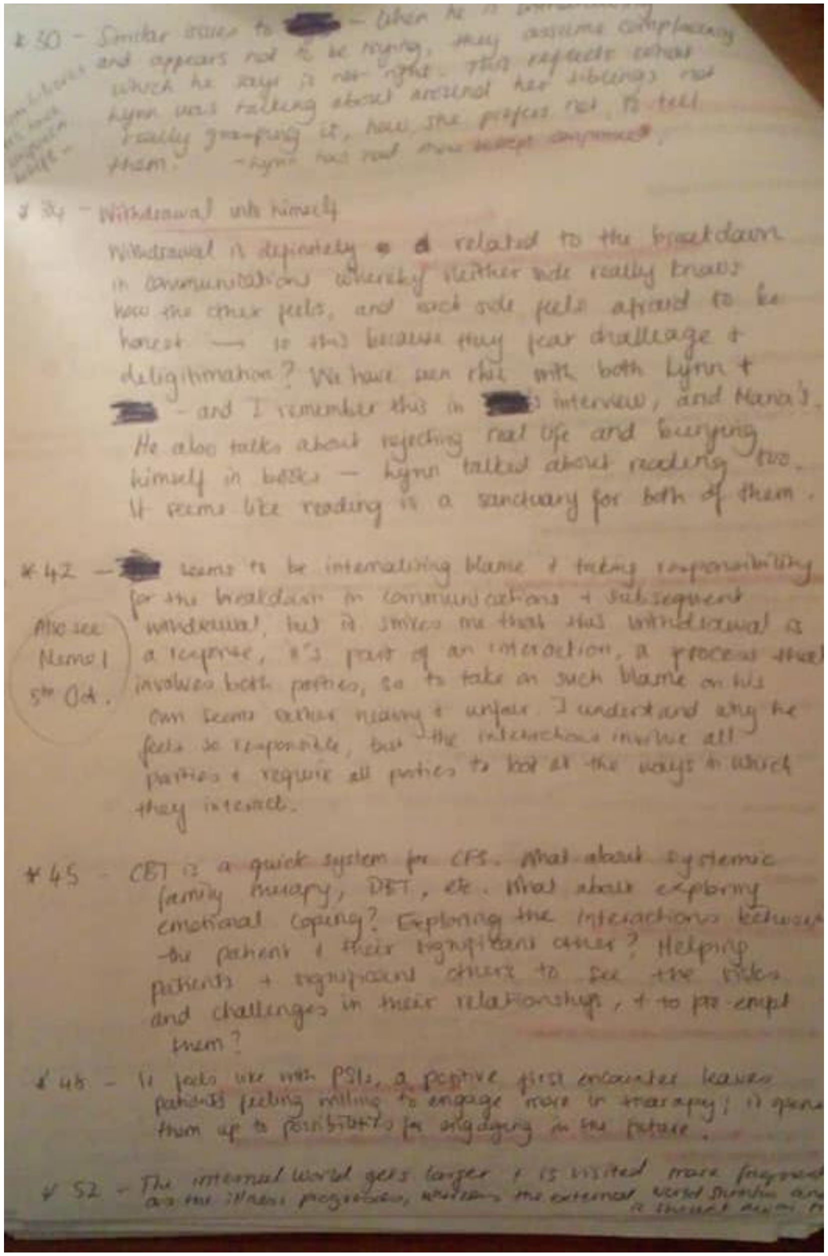
Feeling guilty about burdening them.

"I'm not communicating"

"I feel I can't communicate"

"I don't want to burden them"

7 That's an interesting point really, because I suppose it's about the weight of the burden and if you think about the burden of you saying how you really feel and how that will impact them, versus the burden of you not telling them and them feeling like you are not able to tell them, the two are actually equal weight. Would you say that's fair?



\# 30 – Similar issues to [redacted] – when he is [illegible] they assume complacency and appears not to be trying, they expects which he says is not right. This reflects what Lynn was talking about around her siblings not really grasping it, how she prefers not to tell them. – Lynn has not [illegible]

\# 34 – Withdrawal into himself

Withdrawal is definitely related to the breakdown in communications whereby neither side really knows how the other feels, and each side feels afraid to be honest → is this because they fear challenge + delegitimation? We have seen this with both Lynn + [redacted] – and I remember this in [redacted] interview, and Maria's. He also talks about rejecting real life and burying himself in books – Lynn talked about reading too. It seems like reading is a sanctuary for both of them.

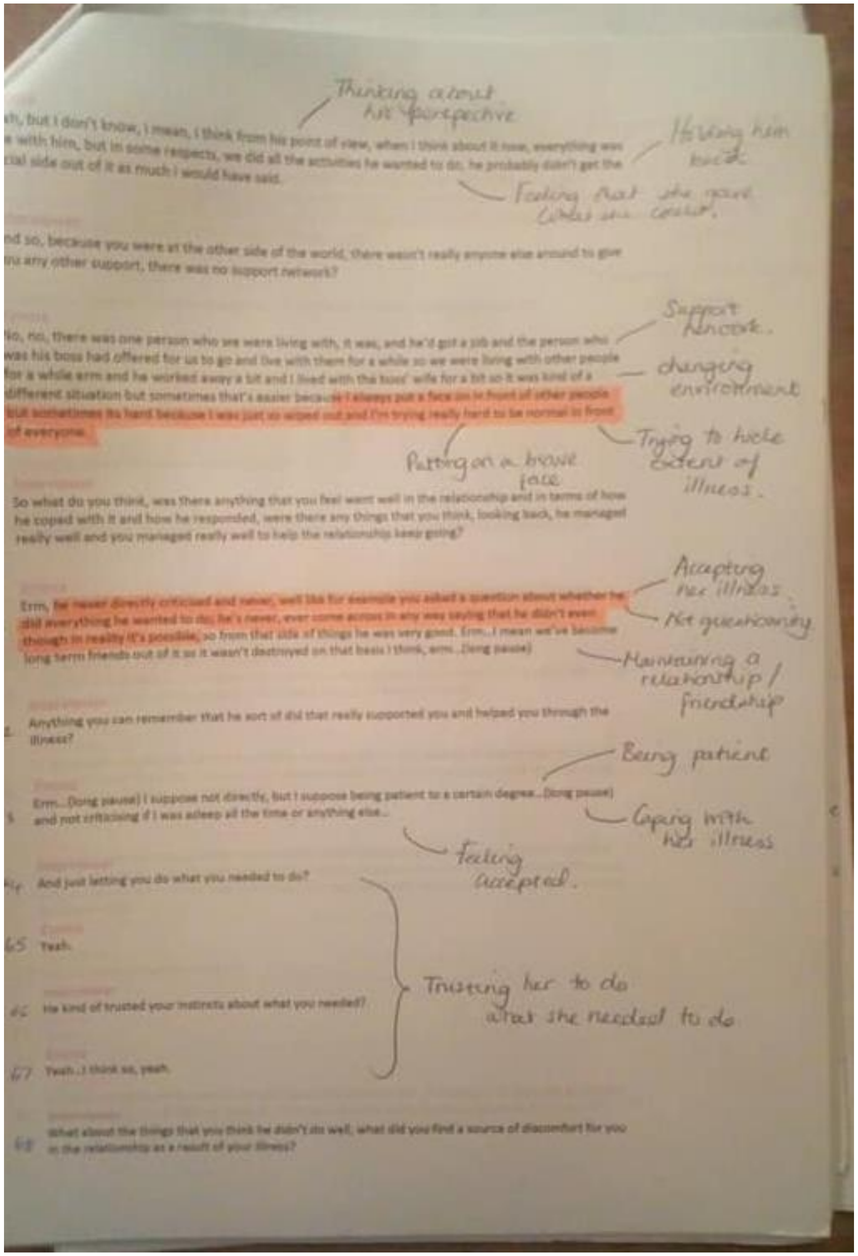
* 42 – [redacted] seems to be internalising blame + taking responsibility for the breakdown in communications + subsequent withdrawal, but it strikes me that this withdrawal is a response, it's part of an interaction, a process that involves both parties, so to take on such blame on his own seems rather heavy + unfair. I understand why he feels so responsible, but the interactions involve all parties + require all parties to look at the ways in which they interact.

Also see Memo 1 5th Oct.

* 45 – CBT is a quick system for CFS. What about systemic family therapy, DBT, etc. What about exploring emotional coping? Exploring the interactions between the patient + their significant other? Helping patients + significant others to see the risks and challenges in their relationships, + to pre-empt them?

\# 48 – It feels like with PSIs, a positive first encounter leaves patients feeling willing to engage more in therapy; it opens them up to possibilities for engaging in the future.

\# 52 – The internal world gets larger + is visited more frequently as the illness progresses, whereas the external world shrinks and is [illegible]



Thinking about his perspective

h, but I don't know, I mean, I think from his point of view, when I think about it now, everything was
with him, but in some respects, we did all the activities he wanted to do, he probably didn't get the
cial side out of it as much I would have said.

Holding him back

Feeling that she gave what she could.

nd so, because you were at the other side of the world, there wasn't really anyone else around to give
ou any other support, there was no support network?

o, no, there was one person who we were living with, it was, and he'd got a job and the person who
was his boss had offered for us to go and live with them for a while so we were living with other people
for a while erm and he worked away a bit and I lived with the boss' wife for a bit so it was kind of a
different situation but sometimes that's easier because I always put a face on in front of other people
but sometimes its hard because I was just so wiped out and I'm trying really hard to be normal in front
of everyone.

Support Network.

changing environment

Trying to hide extent of illness.

Putting on a brave face

So what do you think, was there anything that you feel went well in the relationship and in terms of how he coped with it and how he responded, were there any things that you think, looking back, he managed really well and you managed really well to help the relationship keep going?

Erm, he never directly criticised and never, well like for example you asked a question about whether he did everything he wanted to do; he's never, ever come across in any way saying that he didn't even though in reality it's possible, so from that side of things he was very good. Erm…I mean we've become long term friends out of it so it wasn't destroyed on that basis I think, erm…(long pause)

Accepting her illness

Not questioning

Maintaining a relationship / friendship

2 Anything you can remember that he sort of did that really supported you and helped you through the illness?

3 Erm…(long pause) I suppose not directly, but I suppose being patient to a certain degree…(long pause) and not criticising if I was asleep all the time or anything else…

Being patient

Coping with her illness

Feeling accepted.

64 And just letting you do what you needed to do?

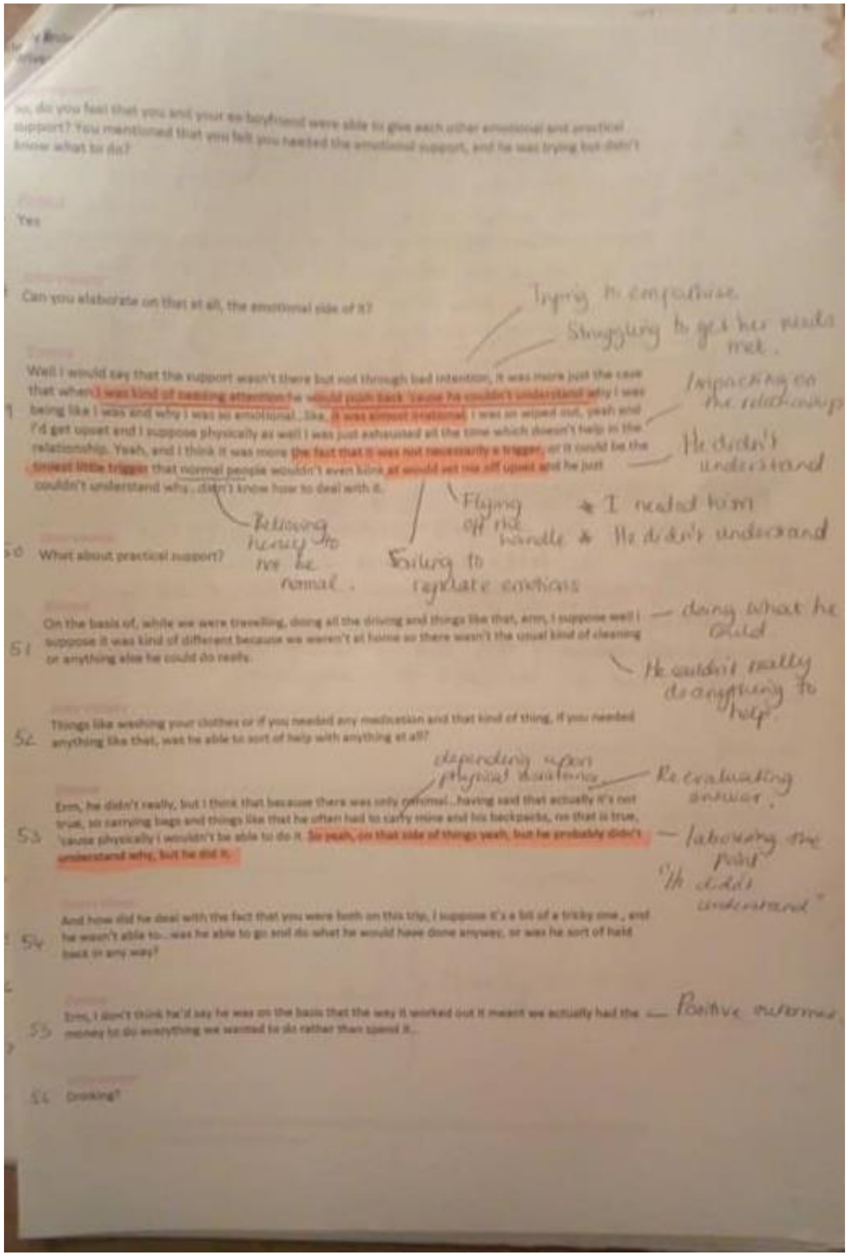
65 Yeah.

66 He kind of trusted your instincts about what you needed?

67 Yeah…I think so, yeah.

Trusting her to do what she needed to do

68 What about the things that you think he didn't do well, what did you find a source of discomfort for you in the relationship as a result of your illness?



so, do you feel that you and your ex-boyfriend were able to give each other emotional and practical support? You mentioned that you felt you needed the emotional support, and he was trying but didn't know what to do?

Yes

Can you elaborate on that at all, the emotional side of it?

Well I would say that the support wasn't there but not through bad intention, it was more just the case that when I was kind of needing attention he would push back 'cause he couldn't understand why I was being like I was and why I was so emotional…like, it was almost irrational. I was so wiped out, yeah and I'd get upset and I suppose physically as well I was just exhausted all the time which doesn't help in the relationship. Yeah, and I think it was more the fact that it was not necessarily a trigger, or it could be the tiniest little trigger that normal people wouldn't even blink at would set me off upset and he just couldn't understand why…didn't know how to deal with it.

Trying to empathise

Struggling to get her needs met.

Impacting on the relationship

He didn't understand

Believing herself to not be normal.

Failing to regulate emotions

Flying off the handle

★ I needed him

★ He didn't understand

50 What about practical support?

51 On the basis of, while we were travelling, doing all the driving and things like that, erm, I suppose well I suppose it was kind of different because we weren't at home so there wasn't the usual kind of cleaning or anything else he could do really.

— doing what he could

He couldn't really do anything to help.

52 Things like washing your clothes or if you needed any medication and that kind of thing, if you needed anything like that, was he able to sort of help with anything at all?

53 Erm, he didn't really, but I think that because there was only minimal…having said that actually it's not true, so carrying bags and things like that he often had to carry mine and his backpacks, no that is true, 'cause physically I wouldn't be able to do it. So yeah, on that side of things yeah, but he probably didn't understand why, but he did it.

depending upon physical assistance

Re-evaluating answer.

— labouring the point "he didn't understand"

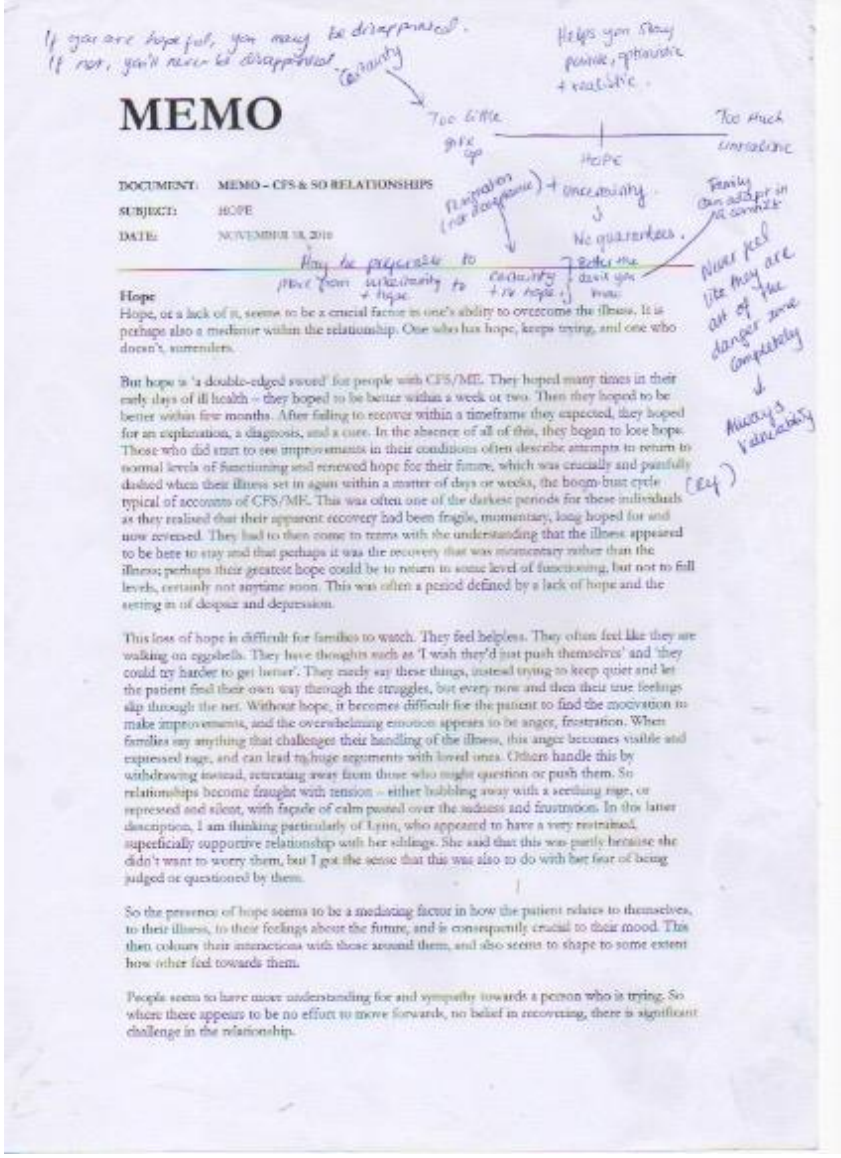
54 And how did he deal with the fact that you were both on this trip, I suppose it's a bit of a tricky one, and he wasn't able to…was he able to go and do what he would have done anyway, or was he sort of held back in any way?

55 Erm, I don't think he'd say he was on the basis that the way it worked out it meant we actually had the money to do everything we wanted to do rather than spend it…

— Positive outcome

56 Drinking?

Appendix 13. Theoretical Memos



If you are hopeful, you may be disappointed.
If not, you'll never be disappointed. Certainty

Helps you stay
positive, optimistic
+ realistic.

Too little
give up

Hope

Too much
unrealistic

MEMO

DOCUMENT: MEMO – CFS & SO RELATIONSHIPS

SUBJECT: HOPE

DATE: NOVEMBER 18, 2010

(Frustration
(not diagnosis)) + uncertainty.
↓
No guarantees.

Family
can adapt in
the context

May be preferable to
move from uncertainty to certainty
+ hope + no hope. } Better the
devil you
know

Never feel
like they are
out of the
danger zone
completely
↓
Always
vulnerability

Hope

Hope, or a lack of it, seems to be a crucial factor in one's ability to overcome the illness. It is perhaps also a mediator within the relationship. One who has hope, keeps trying, and one who doesn't, surrenders.

But hope is 'a double-edged sword' for people with CFS/ME. They hoped many times in their early days of ill health – they hoped to be better within a week or two. Then they hoped to be better within few months. After failing to recover within a timeframe they expected, they hoped for an explanation, a diagnosis, and a cure. In the absence of all of this, they began to lose hope. Those who did start to see improvements in their conditions often describe attempts to return to normal levels of functioning and renewed hope for their future, which was crucially and painfully dashed when their illness set in again within a matter of days or weeks, the boom-bust cycle typical of accounts of CFS/ME. This was often one of the darkest periods for these individuals as they realised that their apparent recovery had been fragile, momentary, long hoped for and now reversed. They had to then come to terms with the understanding that the illness appeared to be here to stay and that perhaps it was the recovery that was momentary rather than the illness; perhaps their greatest hope could be to return to some level of functioning, but not to full levels, certainly not anytime soon. This was often a period defined by a lack of hope and the setting in of despair and depression. (Ref)

This loss of hope is difficult for families to watch. They feel helpless. They often feel like they are walking on eggshells. They have thoughts such as 'I wish they'd just push themselves' and 'they could try harder to get better'. They rarely say these things, instead trying to keep quiet and let the patient find their own way through the struggles, but every now and then their true feelings slip through the net. Without hope, it becomes difficult for the patient to find the motivation to make improvements, and the overwhelming emotion appears to be anger, frustration. When families say anything that challenges their handling of the illness, this anger becomes visible and expressed rage, and can lead to huge arguments with loved ones. Others handle this by withdrawing instead, retreating away from those who might question or push them. So relationships become fraught with tension – either bubbling away with a seething rage, or repressed and silent, with façade of calm pasted over the sadness and frustration. In this latter description, I am thinking particularly of Lynn, who appeared to have a very restrained, superficially supportive relationship with her siblings. She said that this was partly because she didn't want to worry them, but I got the sense that this was also to do with her fear of being judged or questioned by them.

So the presence of hope seems to be a mediating factor in how the patient relates to themselves, to their illness, to their feelings about the future, and is consequently crucial to their mood. This then colours their interactions with those around them, and also seems to shape to some extent how other feel towards them.

People seem to have more understanding for and sympathy towards a person who is trying. So where there appears to be no effort to move forwards, no belief in recovering, there is significant challenge in the relationship.

- Write in the first person
- Write clearly + more straight forward.

- Trudie Chalder
* Alison Wearden

MEMO

DOCUMENT: MEMO – CFS & SIGNIFICANT OTHER RELATIONSHIPS

SUBJECT: INTRODUCING THE CORE CONCEPTS AND THEIR RELATIONSHIP TO EACH OTHER

DATE: JANUARY 17 2017

I identified

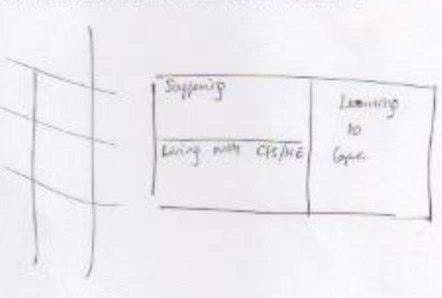
Through the analytic process, 3 core concepts have been identified. These are defined as 'Suffering with CFS/ME', 'Learning to Cope' and 'Living with CFS/ME'. Across these 3 core concepts, 10 subcategories have emerged. Whilst it will become clear that there is a degree of cross over between the elements of these core categories, these can initially be conceptualised as follows:

Use I.

Suffering with CFS/ME
Dark Emotions
Dependence
Resignation
Resistance

Living with CFS/ME
Acceptance
Hope
Light Emotions

Learning to Cope
Making Sense of Things
Family Processes
Identity



Brief Overview of the Core Categories

this

It became clear in the research that the quality and experience of the relationship between the person with CFS/ME and their significant other is mediated by many factors, but importantly in the context of CFS/ME, by factors relating to how they and their family make sense of their illness and the emotional consequences of this process. Not only was consequential for their relationship, but also for the family, for their quality of life, and potentially for their recovery journey.

At the simplest level, people with CFS/ME and their families can be understood to be either 'suffering' with CFS/ME or 'living' with CFS/ME. Those families who suffer with CFS/ME experience dark and difficult emotions in relation to their situation on an ongoing basis. These include despair, frustration, guilt, and grief. The person with CFS/ME often struggles with loss of independence and either resist and 'fight' their illness, which can serve to worsen their condition, or they resign themselves to their illness and their suffering. Both of these mindsets have devastating consequences for their recovery. Meanwhile, their family members and in particular their significant other, struggle with a similar cocktail of negative and distressing emotions, for example, loss of their own autonomy, difficulty with adjusting to being so relied upon, and in cases where they become entangled with emotional over-involvement or enmeshment, a loss of their independence can give rise to challenging combination of resentment and guilt.

More measured

Conversely, 'living' with CFS/ME is a state of being within the context of CFS/ME that requires the presence of acceptance, hope and understanding. Moving gradually to a place whereby the person with CFS/ME, their significant other, the wider family and sometimes their network beyond the family environment, accepts the situations and makes adjustments to adapt to their limitations. This enables them to find their way through their experience with less resistance; it helps them to begin letting go of their losses (such as of their prior self, lifestyle, hopes and expectations), and instead to focus on enhancing their quality of life within the limitations that their illness impresses upon the family. It becomes possible for them to rediscover their positive emotional experiences more fully than when they are trapped in a place of suffering. Hope and looking to the future begin to emerge, as does the potential for growth. The family re-engage with forward-motion and rekindle enjoyment, finding ways to make the most of their life, interests, passions and love for one another in spite of the cloud of illness that continues to hang over them. The sun begins to shine again and through this, they begin to move towards a place of recovery.

It is important to note however, that for many people with CFS/ME, and within this sample, recovery does not spell a return to their pre-CFS/ME state or lifestyle. Firstly, it is likely that it was that lifestyle that contributed to their illness. In the absence of medical explanations, the majority of people who experience CFS/ME spend enough time searching for answers to develop an understanding of how their own lifestyle choices could have contributed to their illness. Having gone through such a challenging illness experience, often coupled with dark emotional times, most develop a very sturdy determination never to return to their darkest times or most difficult experiences of the illness. They develop a set of rules for themselves about what constitutes acceptable levels of stress, activity, socialising and expectations, usually benchmarked much lower than their pre-CFS/ME levels. These become their 'limitations' – consequences of having experienced CFS/ME. They often report that on occasions where they may exceed these levels and push their own boundaries, they have experienced a period of relapse – either a return to or worsening of symptoms. Consequently, for the vast majority of people who have experienced CFS/ME, recovery means a return to an acceptable level of functioning and quality of life, not a return to their prior self.

Across the two states – 'Suffering with CFS/ME' and 'Living with CFS/ME', and integral to their persistence, is the concept of 'Learning to cope'. In learning to cope, the ability to make sense of what is happening ('Making sense of things') and to understand, hold on to or re-establish a sense of identity are important factors in the development of one's mental state in relation to coping with their illness. Across both of these processes, family processes also interweave and have an influence, such as developing a shared sense of identity, reaching a shared understanding, collective conceptualisations and behaviours, and working together to improve quality of life. Where the family, and particular the patient significant other dyad, faces challenges in these areas, the family is more likely to become stuck in a place of suffering rather than living with CFS/ME.

Because of the complex nature of CFS/ME and the way in which it develops, is (or is not) diagnosed, and the lack of promise around treatment protocols, it seems fair to say that most people with CFS/ME and their families experience a period of 'suffering' with CFS/ME at least for a while at the beginning of their illness. For some, it can take years to move to a place of 'living' with CFS/ME, and for others they may never get there. Fortunately, there are also those who make a full recovery, and this is discussed here to an extent. However, the sample for this research consisted predominantly of individuals who had never recovered – people who were either continuing to 'suffer' or who had learned to 'live' within the context of CFS/ME.

MEMO

DOCUMENT: MEMO – CFS & SO RELATIONSHIPS
SUBJECT: MAKING SENSE OF THINGS
DATE: NOVEMBER 28, 2016

Related Codes:
What's Happening to Me?
Acceptance
Adjustment
Who I used to be
Attribution
Label, Diagnosis, Roadmap
Daily Life with CFS/ME
Acceptance
Support Group
Beliefs and Frameworks – Conceptualisation of CFS/ME

Making Sense of Things

The overwhelming emotion communicated by both participants and SOs throughout the interviews is one of frustration. This frustration is a consequence of their confusion around their illness. As the illness takes hold, it permeates every aspect of their life. Both the patient and SO are forced to completely re-evaluate their identity. They grapple with the concept of who they are – their identity before the illness becomes 'Who I used to be' and is most frequently conceptualised by both patients and SOs as someone who was active, worked hard, 'a grafter', often enjoying sports, certainly not someone who was 'lazy'. The stark reality that they will not recover overnight begins to dawn on them, and they begin to realise that the more they push or force, the less control they appear to have over their body or their situation. They often experience a period of depression as a consequence of their sense of confusion - 'What's happening to me?', loss - 'Who I used to be', a 'Growing awareness of own limitations', before entering a difficult period of acceptance and adjustment in which their entire map of themselves and their lives must be redrawn, including their expectations of life and of themselves.

*I would like to know more about this period. How did it feel? How did the significant other tackle this both in themselves and with their loved one?

Label, Diagnosis, Road Map

Receiving a diagnosis can become almost an obsession at this point, and is problematic because of the patients' understanding of what a diagnosis is and what it isn't. A diagnosis of CFS/ME is, in many ways, like a diagnosis of a mental health condition: a term applied flexibly for the purposes of clustering patients to reach a shared understanding. In many ways, a formulation, rather than a diagnosis, would be more appropriate and would help patients to find more effective ways forwards. Unfortunately, it seems as though patients seek the diagnosis for reasons of attribution (being able to attribute to a physical causes alleviates blame, and the CFS/ME label, although contested, does still have a large question mark of possibility over the physical health/biological mechanism models of causation) Once the patient receives the diagnosis of CFS/ME, in many ways it acts like a label applied to people with personality disorder: there is almost an attitude of 'We can't help you'. The medical model suggests there is no pharmaceutical treatment for CFS/ME, other than offering antidepressants, so patients are referred for CBT or further support via a CFS Clinic. This is a bone of contention among patients because they then perceive this to mean that the medical establishment view it as 'a mental health' or 'psychological' problem, AKA 'it's all in the mind'. My understanding of this is actually just that the only evidence-based treatments available are only available within mental health services because that's where those specialists sit. But patients see it as being fobbed off with a psychiatrist. Many are open to meeting a psychiatrist, and often they are told by them that there is 'nothing wrong with them' in mental health terms. They take this as further confirmation that illness must 'be physical' and that there is a cause that has not yet been established. They often then struggle on for years with patchy appointments with counsellors, OTs or CBT therapists, making small steps in recovery if they are lucky. They become trapped in a new identity of 'a person with CFS/ME'; they are forced by their circumstances to try to access social support such as the Disability Living Allowance (which they cannot receive without a diagnosis, and even then the diagnosis and severity of symptoms is often questioned by a panel of 'experts'. They become embroiled in a messy state of hypochondriasis whereby everything becomes a symptom. They continue with gruelling assessments with a range of specialists, desperately seeking answers and understanding, and often face rejection after rejection. This must be extremely demoralising for someone who is trying so hard to make sense of things. They begin to form their own ideas about what is going on for them and can become attached to these ideas, defensive when anyone challenges their newfound beliefs and understandings, and at this point it becomes easy for them and their families to get caught up in rejecting the medical establishments whilst simultaneously refusing any responsibility for what they continue to go through. The lack of medical answers and input at this point can lead them to place blame for their entire experience on NHS services, and their anger and frustration becomes directed at those services. The truth is, there is a gap between what health professionals understand the diagnosis of CFS/ME to mean, and what patients understand it to mean for them and for the services to which they have access. In addition, there is misunderstanding amongst professionals, GPs included, about what the diagnosis is, what it means, and how these patients can be supported (parallels with BPD). Finally, there is also then a hole in service provisions suited to these individuals. They are being failed by the health system. They subsequently completely reject services and are forced to go private – for example visiting the Optimum Health Clinic, contacting Sarah Myhill, seeing osteopaths, homeopaths, nutritionists, all of whom willingly develop a therapeutic relationship of positive regard, belief in positive outcomes…the patients' illness begins to shift, and they become convinced that the treatment 'is working' – a placebo effect, but one which cannot be achieved by a health service that takes an attitude of 'We can't do any more for you'. All that does is foster frustration, resentment and anger, which are never going to be conducive to helping a patient overcome their their difficulties.

Something about
patient + Sig O. is mirrored by patient + Healthcare
/ Psychotherapy team
Parallel process with worse cases.
↳ Not being believed (GP rel.)
Skepticism
feeling fobbed off
Being told there's nothing wrong with you.
Labelled as a hypochondriac.

Sig O.
Can be similar

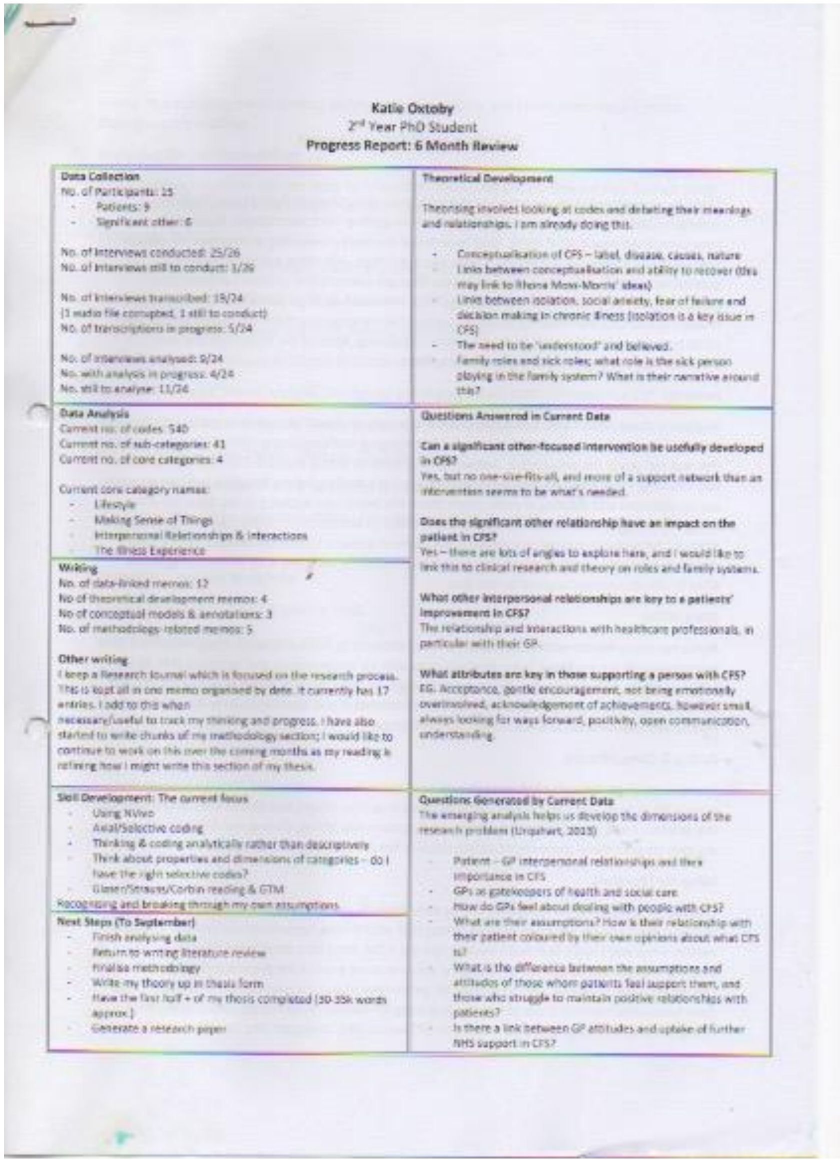
oppositional
distrusting.

↓
leads to desperate measures.
Oste. / Sarah Myhill
OH

Sig O.
Also a parallel.

Solicitors.

Appendix 14. Supervision Record



Katie Oxtoby
2nd Year PhD Student
Progress Report: 6 Month Review

Data Collection
No. of Participants: 15
- Patients: 9
- Significant other: 6

No. of interviews conducted: 25/26
No. of interviews still to conduct: 1/26

No. of interviews transcribed: 19/24
(1 audio file corrupted, 1 still to conduct)
No. of transcriptions in progress: 5/24

No. of interviews analysed: 9/24
No. with analysis in progress: 4/24
No. still to analyse: 11/24

Theoretical Development

Theorising involves looking at codes and debating their meanings and relationships. I am already doing this.

- Conceptualisation of CFS – label, disease, causes, nature
- Links between conceptualisation and ability to recover (this may link to Rhona Moss-Morris' ideas)
- Links between isolation, social anxiety, fear of failure and decision making in chronic illness (isolation is a key issue in CFS)
- The need to be 'understood' and believed.
- Family roles and sick roles; what role is the sick person playing in the family system? What is their narrative around this?

Data Analysis
Current no. of codes: 540
Current no. of sub-categories: 41
Current no. of core categories: 4

Current core category names:
- Lifestyle
- Making Sense of Things
- Interpersonal Relationships & Interactions
- The Illness Experience

Writing
No. of data-linked memos: 12
No of theoretical development memos: 4
No of conceptual models & annotations: 3
No. of methodology-related memos: 5

Other writing
I keep a Research Journal which is focused on the research process. This is kept all in one memo organised by date. It currently has 17 entries. I add to this when necessary/useful to track my thinking and progress. I have also started to write chunks of my methodology section; I would like to continue to work on this over the coming months as my reading is refining how I might write this section of my thesis.

Questions Answered in Current Data

Can a significant other-focused intervention be usefully developed in CFS?
Yes, but no one-size-fits-all, and more of a support network than an intervention seems to be what's needed.

Does the significant other relationship have an impact on the patient in CFS?
Yes – there are lots of angles to explore here, and I would like to link this to clinical research and theory on roles and family systems.

What other interpersonal relationships are key to a patients' improvement in CFS?
The relationship and interactions with healthcare professionals, in particular with their GP.

What attributes are key in those supporting a person with CFS?
EG. Acceptance, gentle encouragement, not being emotionally overinvolved, acknowledgement of achievements, however small, always looking for ways forward, positivity, open communication, understanding.

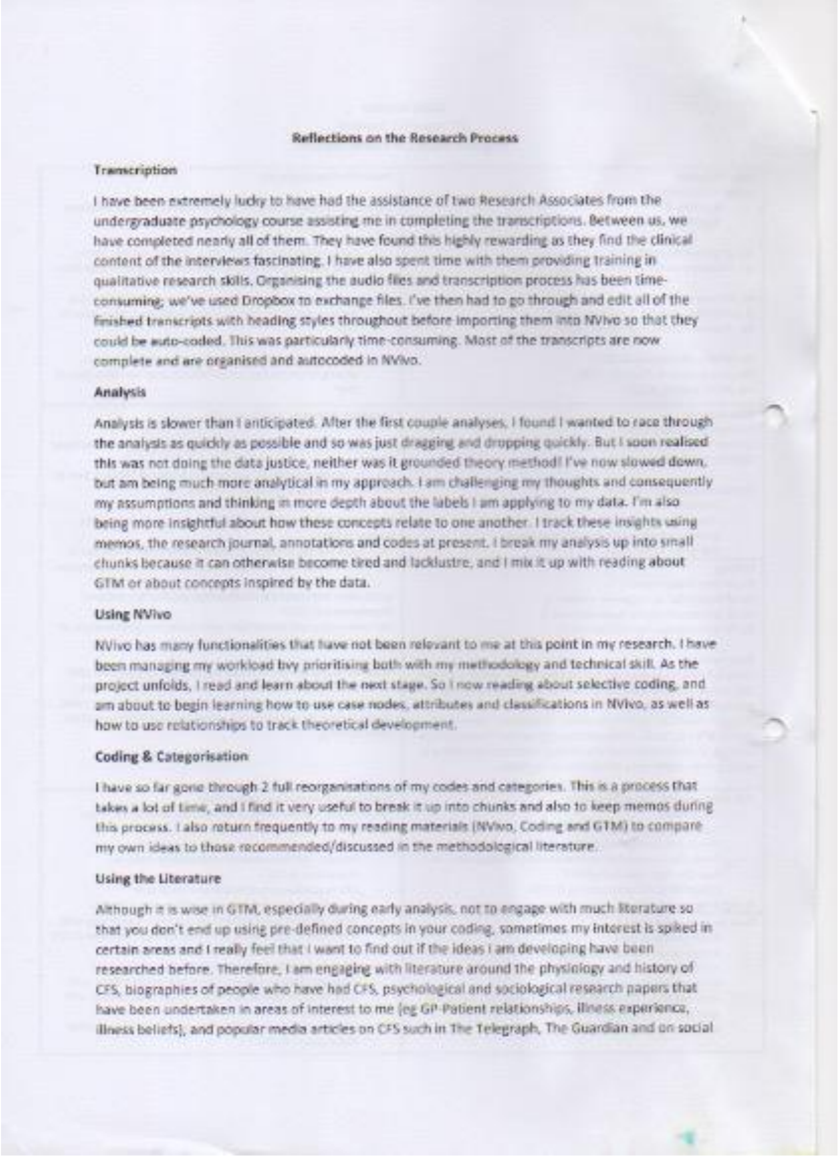
Skill Development: The current focus
- Using NVivo
- Axial/Selective coding
- Thinking & coding analytically rather than descriptively
- Think about properties and dimensions of categories – do I have the right selective codes?
- Glaser/Strauss/Corbin reading & GTM

Recognising and breaking through my own assumptions

Next Steps (To September)
- Finish analysing data
- Return to writing literature review
- Finalise methodology
- Write my theory up in thesis form
- Have the first half + of my thesis completed (30-35k words approx.)
- Generate a research paper

Questions Generated by Current Data
The emerging analysis helps us develop the dimensions of the research problem (Urquhart, 2013)

- Patient – GP interpersonal relationships and their importance in CFS
- GPs as gatekeepers of health and social care
- How do GPs feel about dealing with people with CFS?
- What are their assumptions? How is their relationship with their patient coloured by their own opinions about what CFS is?
- What is the difference between the assumptions and attitudes of those whom patients feel support them, and those who struggle to maintain positive relationships with patients?
- Is there a link between GP attitudes and uptake of further NHS support in CFS?



Reflections on the Research Process

Transcription

I have been extremely lucky to have had the assistance of two Research Associates from the undergraduate psychology course assisting me in completing the transcriptions. Between us, we have completed nearly all of them. They have found this highly rewarding as they find the clinical content of the interviews fascinating. I have also spent time with them providing training in qualitative research skills. Organising the audio files and transcription process has been time-consuming; we've used Dropbox to exchange files. I've then had to go through and edit all of the finished transcripts with heading styles throughout before importing them into NVivo so that they could be auto-coded. This was particularly time-consuming. Most of the transcripts are now complete and are organised and autocoded in NVivo.

Analysis

Analysis is slower than I anticipated. After the first couple analyses, I found I wanted to race through the analysis as quickly as possible and so was just dragging and dropping quickly. But I soon realised this was not doing the data justice, neither was it grounded theory method! I've now slowed down, but am being much more analytical in my approach. I am challenging my thoughts and consequently my assumptions and thinking in more depth about the labels I am applying to my data. I'm also being more insightful about how these concepts relate to one another. I track these insights using memos, the research journal, annotations and codes at present. I break my analysis up into small chunks because it can otherwise become tired and lacklustre, and I mix it up with reading about GTM or about concepts inspired by the data.

Using NVivo

NVivo has many functionalities that have not been relevant to me at this point in my research. I have been managing my workload bvy prioritising both with my methodology and technical skill. As the project unfolds, I read and learn about the next stage. So I now reading about selective coding, and am about to begin learning how to use case nodes, attributes and classifications in NVivo, as well as how to use relationships to track theoretical development.

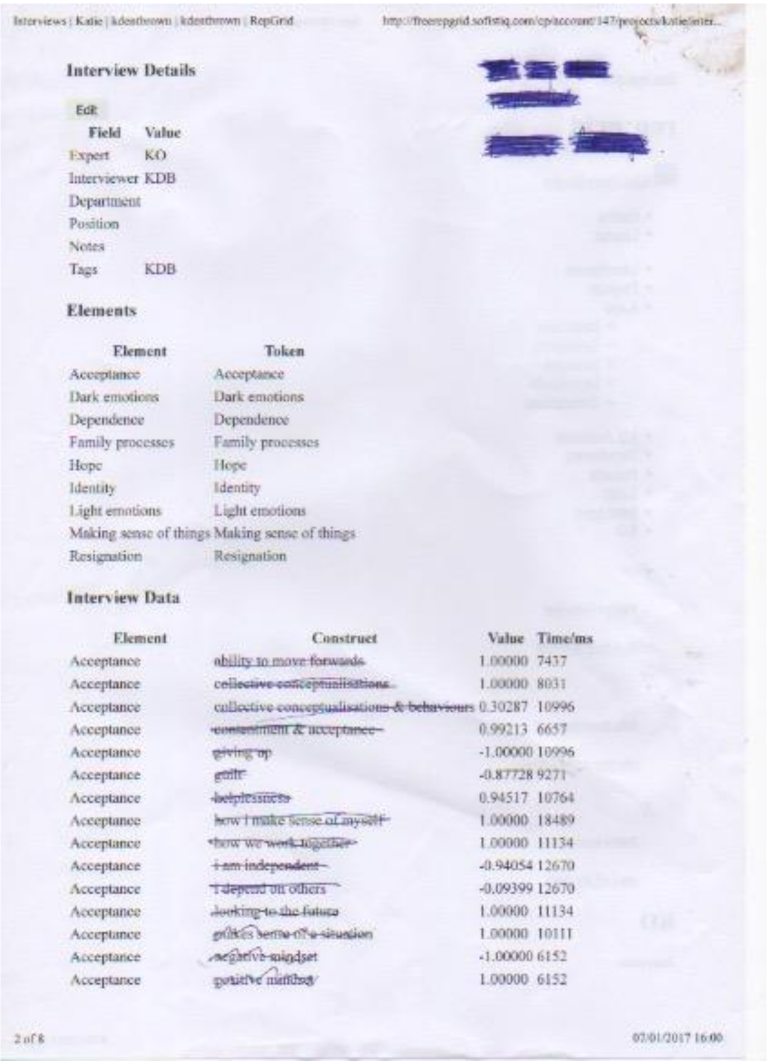
Coding & Categorisation

I have so far gone through 2 full reorganisations of my codes and categories. This is a process that takes a lot of time, and I find it very useful to break it up into chunks and also to keep memos during this process. I also return frequently to my reading materials (NVivo, Coding and GTM) to compare my own ideas to those recommended/discussed in the methodological literature.

Using the Literature

Although it is wise in GTM, especially during early analysis, not to engage with much literature so that you don't end up using pre-defined concepts in your coding, sometimes my interest is spiked in certain areas and I really feel that I want to find out if the ideas I am developing have been researched before. Therefore, I am engaging with literature around the physiology and history of CFS, biographies of people who have had CFS, psychological and sociological research papers that have been undertaken in areas of interest to me (eg GP-Patient relationships, illness experience, illness beliefs), and popular media articles on CFS such in The Telegraph, The Guardian and on social

Appendix 15. RepGrid Example



Interviews | Katie | kdentbrown | kdentbrown | RepGrid http://freerepgrid.sofistiq.com/cp/account/147/projects/katie/inter...

[redacted]

Interview Details

Edit

Field	Value
Expert	KO
Interviewer	KDB
Department	
Position	
Notes	
Tags	KDB

Elements

Element	Token
Acceptance	Acceptance
Dark emotions	Dark emotions
Dependence	Dependence
Family processes	Family processes
Hope	Hope
Identity	Identity
Light emotions	Light emotions
Making sense of things	Making sense of things
Resignation	Resignation

Interview Data

Element	Construct	Value	Time/ms
Acceptance	~~ability to move forwards~~	1.00000	7437
Acceptance	~~collective conceptualisations~~	1.00000	8031
Acceptance	~~collective conceptualisations & behaviours~~	0.30287	10996
Acceptance	~~contentment & acceptance~~	0.99213	6657
Acceptance	~~giving up~~	-1.00000	10996
Acceptance	~~guilt~~	-0.87728	9271
Acceptance	~~helplessness~~	0.94517	10764
Acceptance	~~how i make sense of myself~~	1.00000	18489
Acceptance	~~how we work together~~	1.00000	11134
Acceptance	~~i am independent~~	-0.94054	12670
Acceptance	~~i depend on others~~	-0.09399	12670
Acceptance	~~looking to the future~~	1.00000	11134
Acceptance	~~makes sense of a situation~~	1.00000	10111
Acceptance	~~negative mindset~~	-1.00000	6152
Acceptance	~~positive mindset~~	1.00000	6152

2 of 8 07/01/2017 16:00

Interviews | Katie | kdentbrown | kdentbrown | RepGrid http://freerepgrid.sofistiq.com/cp/account/147/projects/katie/inter...

Element	Construct	Value	Time/ms
Identity	surrendering to loss of control	0.95822	8016
Identity	understanding and adjustment	0.15666	10513
Identity	who i used to be	-0.00288	6072
Light emotions	ability to move forwards	0.46475	7048
Light emotions	collective conceptualisations	0.97128	9290
Light emotions	collective conceptualisations & behaviours	0.65796	7348
Light emotions	contentment & acceptance	0.94974	7297
Light emotions	giving up	-0.89316	7348
Light emotions	guilt	-0.85379	5816
Light emotions	helplessness	-0.90862	7395
Light emotions	how i make sense of myself	0.54462	11126
Light emotions	how we work together	0.97218	8708
Light emotions	i am independent	0.08689	10631
Light emotions	i depend on others	0.14360	10631
Light emotions	looking to the future	0.98710	8708
Light emotions	makes sense of a situation	0.56919	9001
Light emotions	negative mindset	-1.00000	6010
Light emotions	positive mindset	0.99217	6010
Light emotions	resisting a situation	-0.94802	9001
Light emotions	sadness & guilt	-0.85901	7297
Light emotions	scepticism and refusal to adapt	-1.00000	7861
Light emotions	self efficacious	0.63053	7395
Light emotions	stuck in a rut	-0.94802	7048
Light emotions	surrendering to loss of control	-0.57441	11126
Light emotions	understanding and adjustment	0.89295	7861
Light emotions	who i used to be	-0.29715	5816
Making sense of things	ability to move forwards	0.88773	9531
Making sense of things	collective conceptualisations	0.99478	7653
Making sense of things	collective conceptualisations & behaviours	0.73107	9656
Making sense of things	contentment & acceptance	0.70535	6336
Making sense of things	giving up	-0.36199	9656
Making sense of things	guilt	0.04178	6402
Making sense of things	helplessness	0.44909	9873
Making sense of things	how i make sense of myself	0.94829	9839
Making sense of things	how we work together	0.97966	11867
Making sense of things	i am independent	0.81008	12503
Making sense of things	i depend on others	0.83551	12503
Making sense of things	looking to the future	0.13073	11867
Making sense of things	makes sense of a situation	0.95300	9677
Making sense of things	negative mindset	-0.46174	14742

6 of 8 07/01/2017 16:00

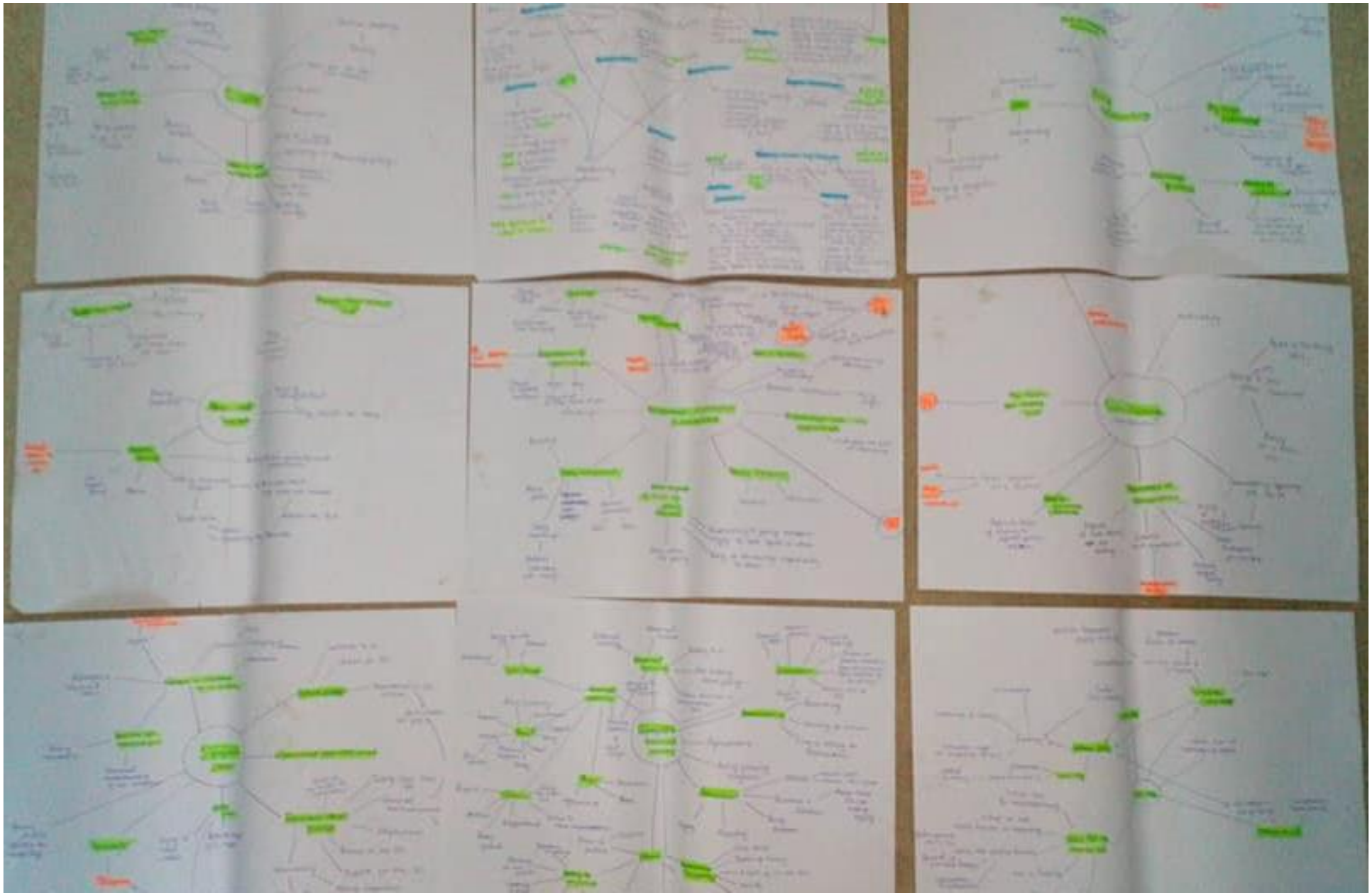
Element	Construct	Value	Time/ms
Dependence	i am independent	-1.00000	7380
Dependence	i depend on others	0.99478	7380
Dependence	looking to the future	-0.96968	8335
Dependence	makes sense of a situation	0.04439	22365
Dependence	negative mindset	0.64051	9315
Dependence	positive mindset	-0.20888	9315
Dependence	resisting a situation	-0.01036	22365
Dependence	sadness & guilt	0.86684	9847
Dependence	scepticism and refusal to adapt	-0.42682	8981
Dependence	self efficacious	-0.92059	8146
Dependence	stuck in a rut	-0.19490	9510
Dependence	surrendering to loss of control	0.96084	8482
Dependence	understanding and adjustment	-0.25065	8981
Dependence	who i used to be	-0.82084	8507
Family processes	ability to move forwards	1.00000	7502
Family processes	collective conceptualisations	0.95561	6582
Family processes	collective conceptualisations & behaviours	1.00000	8729
Family processes	contentment & acceptance	0.83003	8822
Family processes	giving up	-0.54154	8729
Family processes	guilt	0.86945	6596
Family processes	helplessness	0.60052	9194
Family processes	how i make sense of myself	0.78195	12761
Family processes	how we work together	1.00000	8144
Family processes	i am independent	0.90734	10074
Family processes	i depend on others	0.91906	10074
Family processes	looking to the future	1.00000	12761
Family processes	makes sense of a situation	0.04961	7146
Family processes	negative mindset	1.00000	6025
Family processes	positive mindset	1.00000	6025
Family processes	resisting a situation	-0.00787	7146
Family processes	sadness & guilt	0.84073	8822
Family processes	scepticism and refusal to adapt	1.00000	8591
Family processes	self efficacious	0.64300	9194
Family processes	stuck in a rut	1.00000	7502
Family processes	surrendering to loss of control	0.45953	11891
Family processes	understanding and adjustment	1.00000	8591
Family processes	who i used to be	0.03452	6596
Hope	ability to move forwards	0.81201	6989
Hope	collective conceptualisations	0.94778	6337
Hope	collective conceptualisations & behaviours	0.67363	8263

Element	Construct	Value	Time/ms
Making sense of things	positive mindset	0.49347	14742
Making sense of things	resisting a situation	0.96220	9677
Making sense of things	sadness & guilt	0.21932	6336
Making sense of things	scepticism and refusal to adapt	1.00000	9256
Making sense of things	self efficacious	0.49088	9873
Making sense of things	stuck in a rut	-0.44927	9531
Making sense of things	surrendering to loss of control	0.64230	9839
Making sense of things	understanding and adjustment	0.99739	9256
Making sense of things	who i used to be	0.14425	6402
Resignation	ability to move forwards	-0.84856	8721
Resignation	collective conceptualisations	0.98695	14329
Resignation	collective conceptualisations & behaviours	0.99217	8902
Resignation	contentment & acceptance	-0.96298	5489
Resignation	giving up	1.00000	8902
Resignation	guilt	0.03916	6774
Resignation	helplessness	0.97650	9029
Resignation	how i make sense of myself	-0.09457	14329
Resignation	how we work together	0.24151	8800
Resignation	i am independent	-0.83331	7476
Resignation	i depend on others	0.92950	7476
Resignation	looking to the future	-0.99396	8800
Resignation	makes sense of a situation	0.11488	17619
Resignation	negative mindset	0.63801	7563
Resignation	positive mindset	-1.00000	7563
Resignation	resisting a situation	-1.00000	17619
Resignation	sadness & guilt	0.94778	5489
Resignation	scepticism and refusal to adapt	-0.44677	17629
Resignation	self efficacious	-1.00000	9029
Resignation	stuck in a rut	1.00000	8721
Resignation	surrendering to loss of control	1.00000	8510
Resignation	understanding and adjustment	0.57963	17629
Resignation	who i used to be	-0.13755	6774

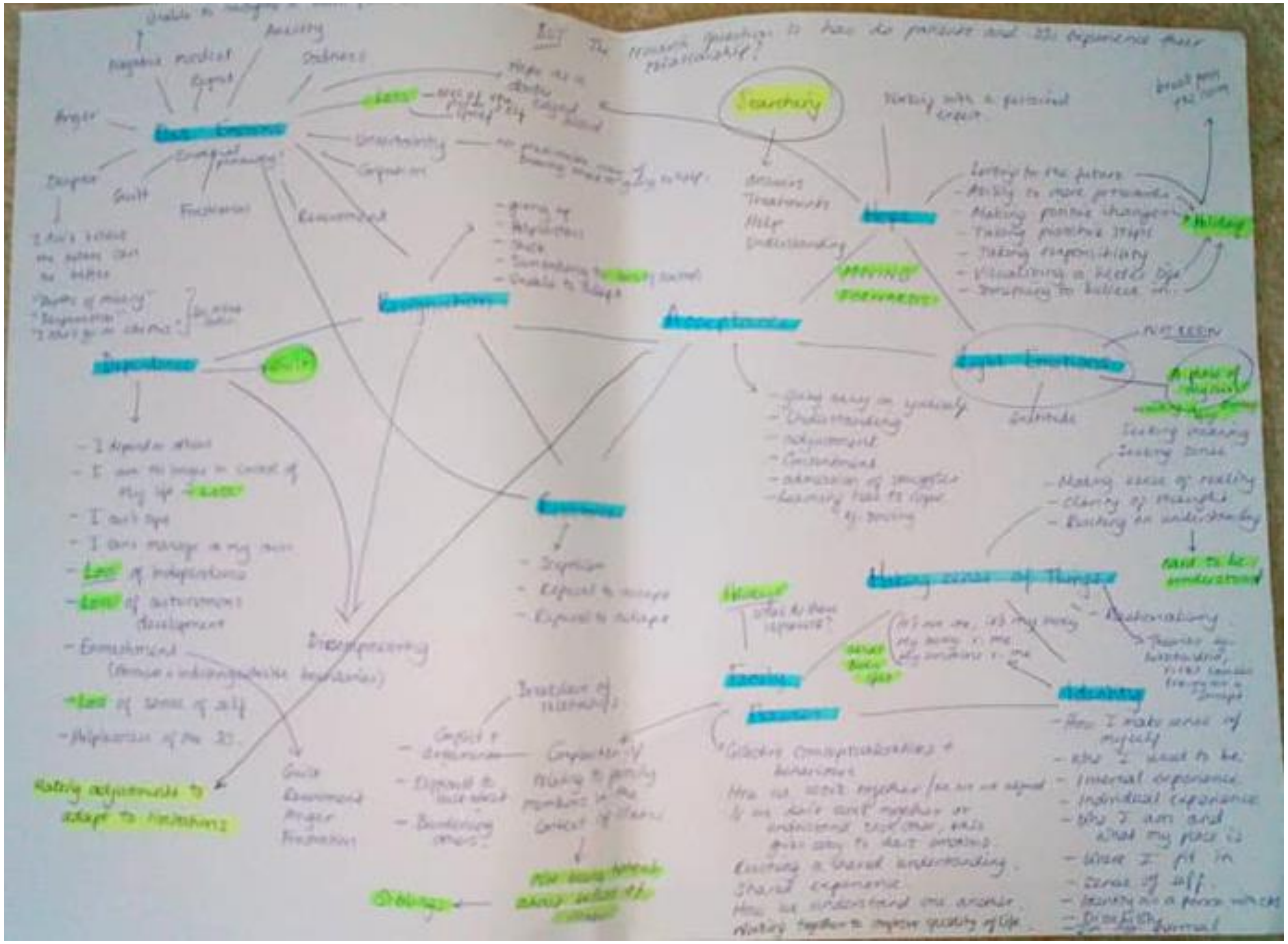
Default Poles

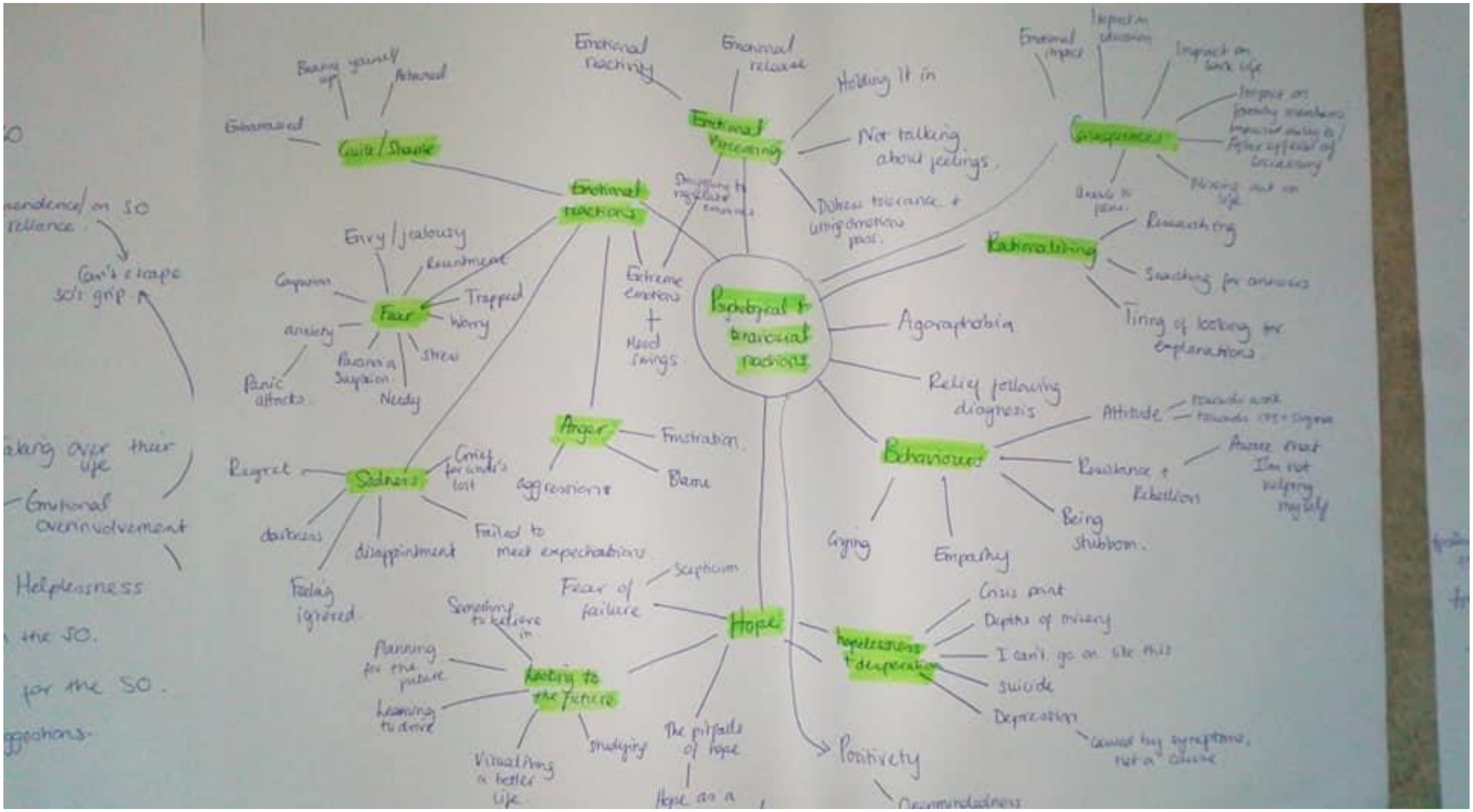
Construct	Construct
Guilt	Who I used to be
I depend on others	I am independent
~~Makes sense of a situation~~	~~Resisting a situation~~
Understanding and adjustment	Scepticism and refusal to adapt

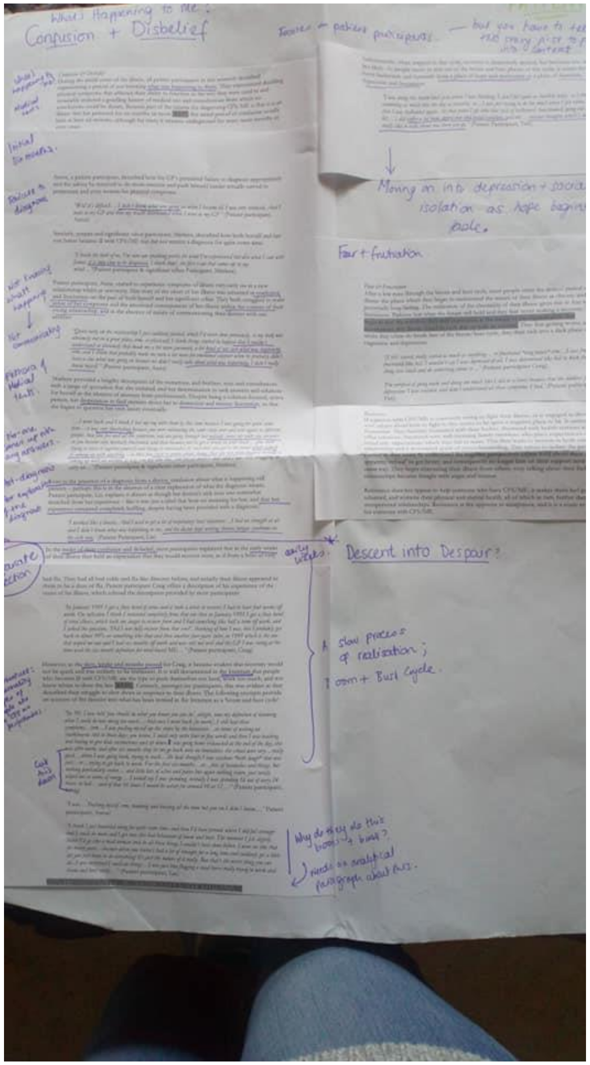
Appendix 16. Conceptual Maps



Sample close up




Psychological + behavioural reactions
Emotional reactions
Guilt / Shame
Beating yourself up
Ashamed
Embarrassed
Fear
Envy / jealousy
Resentment
Comparison
Trapped
Worry
anxiety
Stress
Paranoia
Suspicion
Needy
Panic attacks
Anger
Frustration
Blame
aggression
Sadness
Regret
Grief for what's lost
darkness
disappointment
Failed to meet expectations
Feeling ignored
Extreme emotions
+
Mood swings
Emotional processing
Emotional reactivity
Emotional release
Holding it in
Not talking about feelings
Struggling to regulate emotions
Distress tolerance + unprocessed emotions
Consequences
Emotional impact
Impact on education
Impact on work life
Impact on family members
Impaired ability to
After effects of
Missing out on life
Unable to plan
Rationalising
Researching
Searching for answers
Tiring of looking for explanations
Agoraphobia
Relief following diagnosis
Behaviours
Attitude
towards work
towards CFS + Stigma
Resistance + Rebellion
Aware that I'm not helping myself
Being stubborn
Crying
Empathy
Hope
Fear of failure
Scepticism
Looking to the future
Something to believe in
Planning for the future
Learning to drive
Visualising a better life
Studying
The pitfalls of hope
Hope as a
Hopelessness + desperation
Crisis point
Depths of misery
I can't go on like this
Suicide
Depression
caused by symptoms, not a cause
Positivity
Open mindedness
dependence / reliance on SO
Can't escape SO's grip
taking over their life
Emotional overinvolvement
Helplessness
the SO.
for the SO.
suggestions.



What's Happening to me?
Confusion + Disbelief

— patient participants — but you have to tell the story first to put into context

Initial Dismissals.

Failure to diagnose

Not knowing what's happening

Not communicating

Persona of Medical tests

Moving on into depression + social isolation as hope begins to fade.

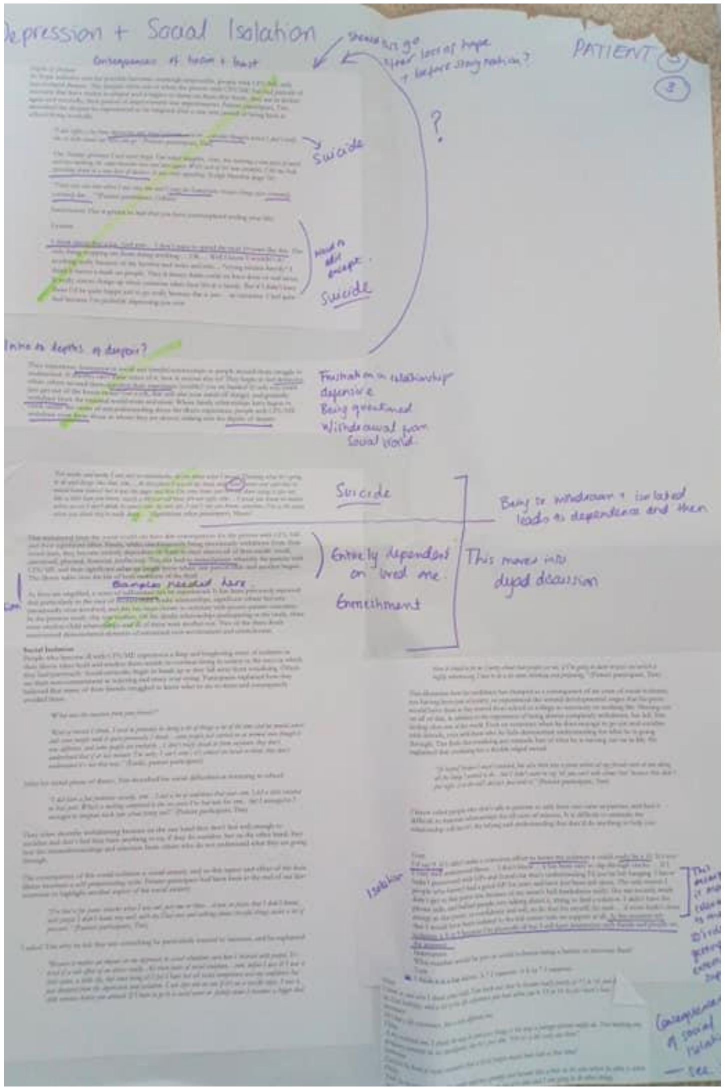
Fear + frustration

Descent into Despair?

early weeks.

slow process of realisation;
Boom + Bust Cycle

Why do they do this boom + bust?
needs an analytical paragraph about this.



Depression + Social Isolation

Consequences of boom + bust

should this go after loss of hope + before stagnation?

PATIENT

Suicide

Need to still escape.

Suicide

Into the depths of despair?

Frustration in relationship
defensive
Being questioned
Withdrawal from Social World.

Suicide

Being so withdrawn + isolated leads to dependence and then

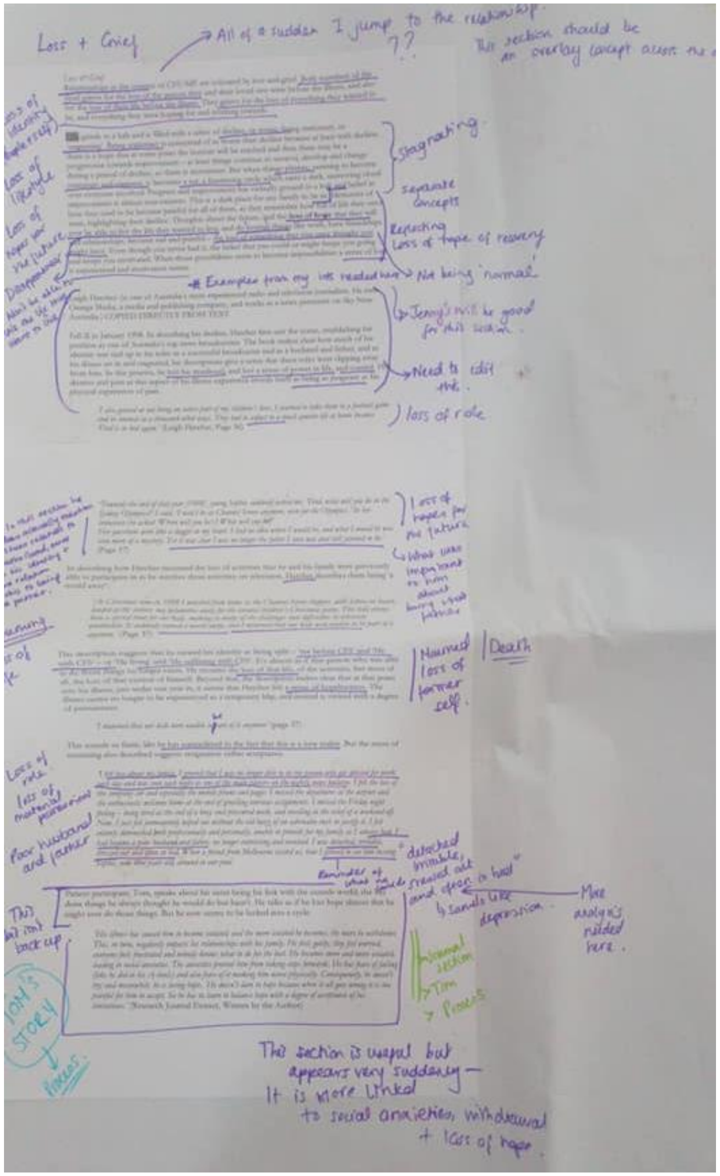
Entirely dependent on loved one.

Enmeshment

This moves into dyad discussion

Examples needed here

Consequences of social isolation



Loss + Grief

All of a sudden I jump to the relationship.
??

This section should be an overlay concept across the

loss of identity

loss of lifestyle

Loss of hopes for the future

Disappointment

Stagnating

Separate concepts

Reflecting
Loss of hope of recovery

Examples from my info needed here

Not being 'normal'

Jenny's will be good for this section.

Need to edit this.

loss of role

loss of hopes for the future

What was important to him

Mourning

Mourned loss of former self.

Death

Loss of role

loss of material possessions

Poor husband and father

Reminder of what he could

detached irritable, stressed out and open to hurt

Sounds like depression.

More analysis needed here.

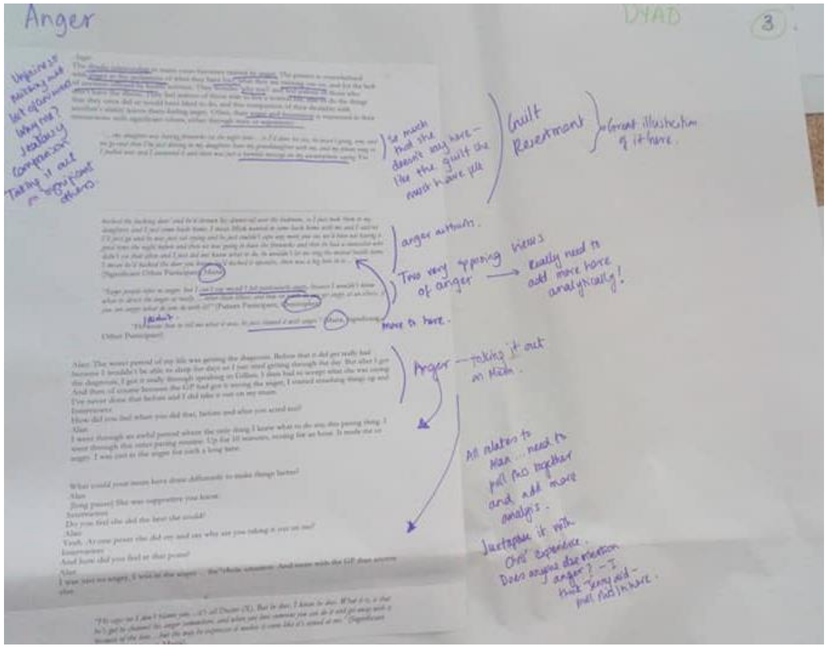
This does not back up

Tom's story

Process.

Personal section
Tom
Process

This section is useful but appears very suddenly —
It is more linked to social anxieties, withdrawal + loss of hope.



Anger

DYAD

3

Unfairness
Why me?
Jealousy
Comparison
Taking it out on significant others.

So much that she doesn't say here — like the guilt she must have felt

Guilt
Resentment

Great illustration of it here.

anger outbursts.

Two very opposing views of anger

Really need to add more here analytically!

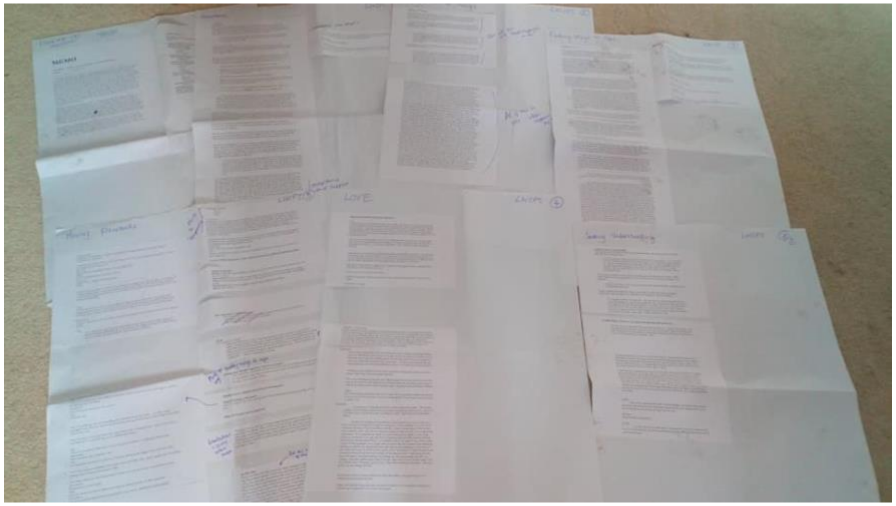
more to here.

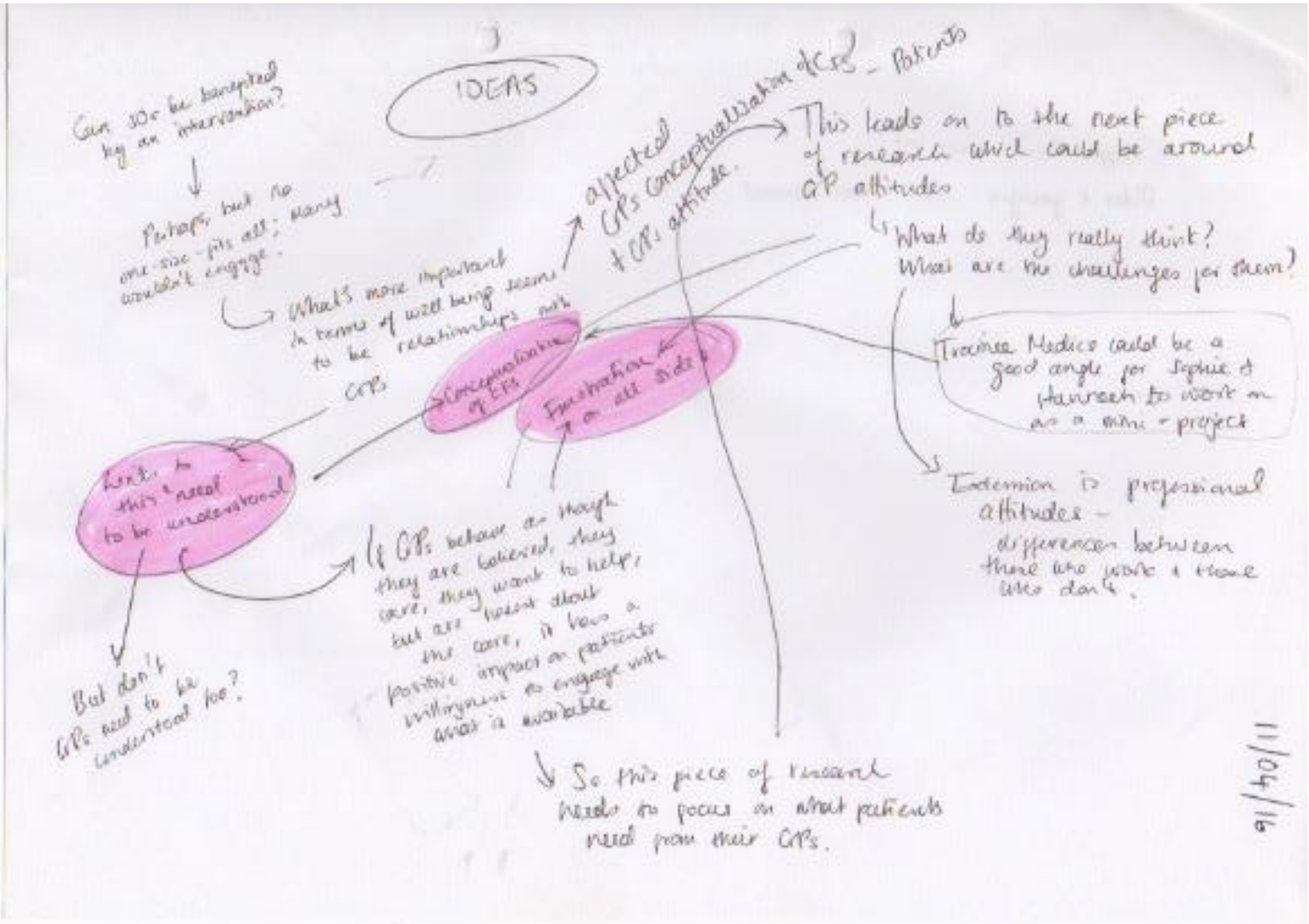
Anger — taking it out on Mum.

All relates to Mum... need to pull this together and add more analysis.

Juxtapose it with Chris' experience.

Does anyone else mention anger? — I think Jenny did — pull this in here.





IDEAS

Can 30s be benefited by an intervention?

Perhaps but no one-size-fits all; many wouldn't engage.

What's more important in terms of well being seems to be relationships with GPs

affected GPs conceptualisation of CSS – Patients
& GPs attitude.

This leads on to the next piece of research which could be around GP attitudes

What do they really think? What are the challenges for them?

Trainee Medics could be a good angle for Sophie & Harpreet to work on as a mini-project

Extension is professional attitudes – differences between those who work & those who don't.

Conceptualisation of EPs

Frustration on all sides

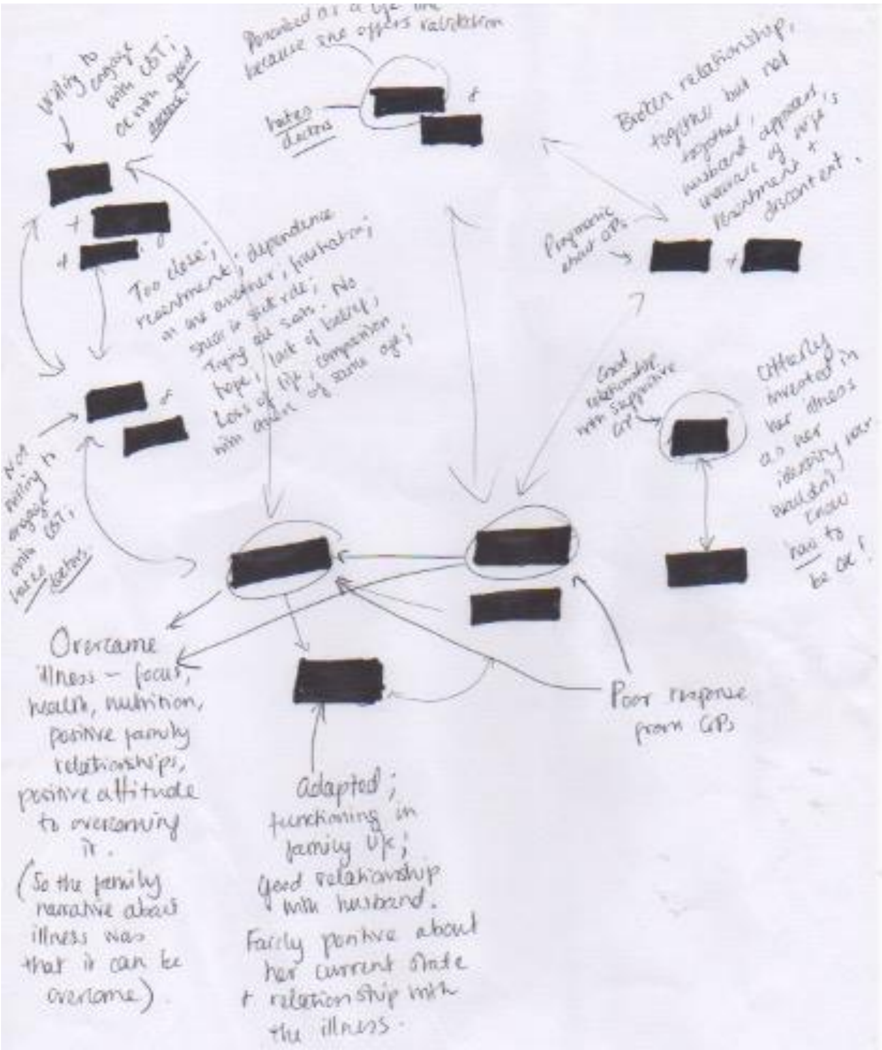
Link to this – need to be understood

If GPs behave as though they are believed, they care, they want to help, but are honest about the care, it has a positive impact on patients willingness to engage with what is available

But don't GPs need to be understood too?

So this piece of research needs to focus on what patients need from their GPs.

11/04/16


Perceived as a life line because she offers validation
[redacted]
+
[redacted]
hates doctors
Willing to engage with CBT; as with good doctors.
[redacted]
+
[redacted]
+
[redacted]
Too close; resentment; dependence on one another; frustration; stuck in sick role; Trying all sorts. No hope; lack of belief; Loss of life; companion with others of same age;
[redacted]
[redacted]
Not willing to engage with CBT; hates doctors
Broken relationship; together but not together; husband appears unaware of wife's resentment + discontent.
Pragmatic about GPs
[redacted]
+
[redacted]
Good relationship with supportive GP
[redacted]
Utterly invested in her illness as her identity was wouldn't know how to be ok!
[redacted]
[redacted]
[redacted]
[redacted]
[redacted]
Overcome illness – focus, health, nutrition, positive family relationships, positive attitude to overcoming it. (So the family narrative about illness was that it can be overcome).
Adapted; functioning in family life; good relationship with husband. Fairly positive about her current state + relationship with the illness.
Poor response from GPs

Appendix 17. Data Management Plan

University of Hull

Data Management Plan

(NB: This form should be completed <u>at the start</u> of all projects where data management is not dealt with otherwise). Shaded areas are considered essential, particularly when a data management plan is required for a grant application.

Date	21/10/2019
Researcher(s)	Researcher: Katie Cunnah (nee Oxtoby), PhD Student & Postdoctoral Researcher, Department of Psychology PhD Research Supervisor: Dr Kim Dent-Brown, Department of Psychology Line Manager in Postdoctoral Role: Dr Fiona Earle, Centre for Human Factors
Project title	Relational and psychological aspects of the experience of chronic fatigue syndrome for patients and their significant others: A grounded theory study
Brief description	Social processes have been investigated as a maintaining factor in chronic fatigue syndrome, also known as myalgic encephamyelitis (CFS/ME), and the responses of the significant others (SOs) of people with CFS/ME (PwCFS/ME) have been shown to be associated with illness outcomes. Experiences of PwCFS/ME and their significant others have been explored, but with limited depth and/or breadth. This study aimed to develop a nuanced understanding of relationships between PwCFS/ME and their SOs, describing emotional and relational characteristics, processes and consequences. 15 participants consisting of PwCFS/ME (*n*=9) and SOs of PwCFS/ME (*n*=6) completed a total of 26 audio-recorded, semi-structured interviews which were analysed according to grounded theory methodology. This study is a PhD study being undertaken by the Researcher, under the supervision of Dr Kim Dent-Brown.

For detailed, updated explanations of the various parts of the document that require completion, please refer to the accompanying Appendices.

This University of Hull History Data Management Plan (HDMP) applies the DCC Checklist for Data Management (v3.0 17 March 2011).

Contents

Section 1: Project Information

A summary of the project details and associated data management requirements

1.1 Project title: Relational and psychological aspects of the experience of chronic fatigue syndrome for patients and their significant others: A grounded theory study

1.2 Project duration 29/09/2014-30/11/2019

1.3 Partners (if applicable)
N/A

1.4 Brief description

Social processes have been investigated as a maintaining factor in chronic fatigue syndrome, also known as myalgic encephamyelitis (CFS/ME), and the responses of the significant others (SOs) of people with CFS/ME (PwCFS/ME) have been shown to be associated with illness outcomes. Experiences of PwCFS/ME and their significant others have been explored, but with limited depth and/or breadth. This study aimed to develop a nuanced understanding of relationships between PwCFS/ME and their SOs, describing emotional and relational characteristics, processes and consequences. 15 participants consisting of PwCFS/ME (*n*=9) and SOs of PwCFS/ME (*n*=6) completed a total of 26 audio-recorded, semi-structured interviews which were analysed according to grounded theory methodology.

This study is a PhD study being undertaken by the Researcher, under the supervision of Dr Kim Dent-Brown.

1.5 Faculty or University requirements for data management

Complete a data management plan with the advice of the University's Researcher Officer.

1.6 Funding body(ies)

PhD Studentship

1.7 Budget (estimate if necessary)

Not Applicable

1.8 Funding body requirements for data management

Not Applicable

Section 2: Data, Materials, Resource Collection Information

This section is used to more fully describe the data

2.1 Brief description of data being created or compiled

Audio recorded interview data
Interview transcripts as Word documents and NVivo files
Hard copies of interview transcripts
Hard copies of signed consent forms and records of personal data
Field notes (paper form) and memos

2.2 Data collection process

26 Interviews were undertaken with 15 participants consisting of people with CFS/ME and their significant other. Interviews took place in the homes of participants and were audio-recorded a MacBook Pro using Logic Pro software. These were converted into MP3 format and the Logic files were destroyed. The interviews were transcribed using Microsoft Word and analysed in Nvivo and in paper format.

2.3 Are there existing forms of the data that will be used within this research project, or which will be used as the basis for the research? If so, provide a brief description and citation

No.

2.3 Will data be available in electronic format (if so then state format(s))?

Audio data will stored in MP3 format until the completion of the PhD at which point these files will be destroyed by being deleted.

Transcribed data are saved as Microsoft Word Documents and as NVivo files.

2.4 Will the data be available in non-digital form (if so then state format(s))?

Field notes have been kept in hand-written format.
Consent forms are kept as hard copies.
Printed transcripts are stored in paper format.

2.5 Will the data stand alone and be comprehensible to a third party or be accompanied by explanatory documentation (e.g., a data dictionary)?

No.

2.6 Describe the quality assurance process for data management

Data Management plans have been discussed with the Research Supervisor throughout the project, and checked with the external and internal thesis examiners. Amendments to the long-term data storage plans have been made according to the recommendations of the thesis examiners and are in-line with more recent GDPR requirements.

This Researcher meets regularly with the research team in her current role to review data management for all current and past projects. This data management plan has been designed to bring the plans in-line with data management for data in the current role so that quality assurance can be maintained beyond the end of the PhD and the Researcher can be supported in doing so.

Section 3: Ethics, Intellectual Property

This section is used to address issues surrounding relevant ethical and intellectual property issues the research will encounter

3.1 How will the ethical aspects of data storage and subsequent access be addressed?

During the Project

Hard Data

- Personal data and participant consent forms were stored in paper form only, in a locked drawer, in a locked filing cabinet, in a locked office at the University of Hull.
- Copies of transcripts were printed out and annotated as part of the research process. These were stored in an office in the Researcher's home during the data analysis phase. Upon completion of the data analysis, these were taken to the Researcher's locked office at the University of Hull where they are stored in a locked filing cabinet.

Digital Audio Data

- Audio recordings were made on a MacBook Computer with Firewall and FileVault enabled. These were immediately transferred onto an encrypted hard drive belonging to the Researcher, and all other copies were deleted.
- Audio recordings of interviews were then stored on this encrypted hard drive.
- Audio recordings will remain on the hard drive until the thesis has been submitted and accepted by the examiners as complete, at which point the audio recordings will be permanently deleted.

Digital Written Data

- Anonymised research data (transcripts) and NVivo files were stored in password-protected files, on a password hard drive, accessible only to the researcher, the research supervisor, and two university-approved research associates. The Research Associates received training from the Researcher in safe data management and confidentiality in line with the procedures outlined in this data management plan.
- When documents (Word documents and NVivo files) were in use, this took place on a MacBook Pro belonging to the Researcher with FileVault, Firewall, and antivirus software enabled, or on University Desktop PCs on the University Campus on the Researcher's personal University drive.
- When documents were in use by the research associates, they were transferred via the University Box file storage system and were briefly stored on password-protected personal computers belonging to the research associates. Once completed and returned to the researcher via Box, they were deleted from the research associates' computers, and downloaded onto the researcher's encrypted hard drive and deleted from Box.

- Memos and analytical writing was stored as password-protected files, on an encrypted hard drive belonging to the researcher.

Back-up

- All of the above documents were backed up on a secure, access-restricted personal drive on the University of Hull network.

Long-term Data Storage Plan

Hard Data

- All hard data (consent forms, personal data, field notes, copies of memos) will be stored in a locked filing cabinet, in a locked office belonging to the Researcher in the Department of Psychology at the University of Hull.
- Should the Researcher leave the University, this data will be handed over to the Department of Psychology for safe storage for six years after the end of the PhD.

Digital Data

- All audio data will be destroyed by being deleted from the encrypted hard drive and university server upon completion of the PhD.
- Following acceptance by the examiners of this thesis as complete, the anonymised data (transcripts) and NVivo files will be uploaded onto a private folder as encrypted, confidential documents on the cloud storage system, Figshare. The Researcher will have responsibility for ensuring this data is destroyed six years after the end of the PhD.
- Once stored on Figshare, all data stored on the hard drive and backed up on the Researcher's personal university drive will be fully deleted.

3.2 Will the data comply with relevant legislation such as Data Protection Act, Copyright, Design and Patents Act, Freedom of Information Act, etc.?

The Researcher has completed the relevant training to ensure that all work complies with legislative requirements.

3.3 If several partners are involved how will compliance with 3.1 and 3.2 be assured?

Not Applicable

Section 4: Access and Use of Information

This section is used to consider if and how you will share the data once it has been created/compiled

4.1 Are you required, or do you intend, to share the data, and with whom? If so, when?

Audio data was shared with two University-approved Research Associates who transcribed the data. The Research Associates received training from the Researcher in safe data management and confidentiality in line with the procedures outlined in this data management plan.

Sections of the transcript data was shared with the Research Supervisor during the data analysis phase.

4.2 If 'yes' to 4.1, in what format will data be shared?

Audio data was shared in MP3 format via the University Box drive.

Written data was shared in digital typed form and print format as part of Microsoft Word documents, and within NVivo files.

4.3 Will the data have to be stored and/or made accessible for a specific period (if so, how long)?

The data will be stored for six years after the end of the PhD which is in line with statutory limitation on legal action (recommended by Stuart Bentley, Research Officer, University of Hull).

The Researcher will take responsibility for the deletion of this data six years after the end of the project.

Should the Researcher leave the university, arrangements will be made for paper data to be held within the Department of Psychology until the date at which it must be destroyed.

4.4 Who may need or wish to have access to the data?

No one other than the Researcher will have a need to access the data after the end of the project. For work on publications, the Researcher will be the lead author on such work, and it will not be necessary for other contributors to access data other than to review excerpts presented in Word format by the Researcher.

4.5 How do you anticipate the data being used subsequent to the project?

It is likely that the transcript data may be used by the Researcher to develop publications after the conclusion of the project. In this case, the data will only be accessible to the Researcher who may then share excerpts of data with a small selected team of writers. The team will only have access to sections of data selected by the Researcher.

Section 5: Storage and Backup of Data

This section is used to clarify details of how the data will be stored

5.1 Where and how will the data be stored **<u>during the lifespan of the project</u>**?

Hard Data

Personal data and participant consent forms were stored in paper form only, in a locked drawer, in a locked filing cabinet, in a locked office at the University of Hull. Copies of transcripts were printed out and annotated as part of the research process. These were stored in an office in the Researcher's home during the data analysis phase. Upon completion of the data analysis, these were taken to the Researcher's locked office at the University of Hull where they are stored in a locked filing cabinet.

Digital Audio Data

Audio recordings were made on a MacBook Computer with Firewall and FileVault enabled. These were immediately transferred onto an encrypted hard drive belonging to the Researcher, and all other copies were deleted. Audio recordings of interviews were then stored on this encrypted hard drive. Audio recordings will remain on the hard drive until the thesis has been submitted and accepted by the examiners as complete, at which point the audio recordings will be permanently deleted.

Digital Written Data

Anonymised research data (transcripts) and NVivo files were stored in password-protected files, on a password hard drive, accessible only to the researcher, the research supervisor, and two university-approved research associates. The Research Associates received training from the Researcher in safe data management and confidentiality in line with the procedures outlined in this data management plan. When documents were in use by the research associates, they were transferred via the University Box file storage system and were briefly stored on password-protected personal computers belonging to the research associates. Once completed and returned to the researcher via Box, they were deleted from the research associates' computers, and downloaded onto the researcher's encrypted hard drive and deleted from Box. Memos and analytical writing was stored as password-protected files, on an encrypted hard drive belonging to the researcher.

Back-up

All of the above documents were backed up on a secure, access-restricted personal drive on the University of Hull network.

5.2 Where and how will the data be stored **<u>on completion of the project</u>**?

Hard Data

All hard data (consent forms, personal data, field notes, copies of memos) will be stored in a locked filing cabinet, in a locked office belonging to the Researcher in the Department of Psychology at the University of Hull. Should the Researcher leave the University, this data will be handed over to the Department of Psychology for safe storage for six years after the end of the PhD.

Digital Data All audio data will be destroyed by being deleted from the encrypted hard drive and university server upon completion of the PhD. Following acceptance by the examiners of this thesis as complete, the anonymised data (transcripts) and NVivo files will be uploaded onto a private folder as encrypted, confidential documents on the cloud storage system, Figshare. The Researcher will have responsibility for ensuring this data is destroyed six years after the end of the PhD. Once stored on Figshare, all data stored on the hard drive and backed up on the Researcher's personal university drive will be fully deleted.
5.3 What provision is being made for backup of the data? Digital data is stored on Researcher's University of Hull server throughout the project. This will be destroyed upon completion of the project.
5.4 Will different versions of the data be stored? If so, what frequency of versioning will be appropriate? File names reflect the date and version, and have been updated at each save.

Section 6: Archiving and Future Proofing of Information

This section is used to describe long-term, post-project aspects of managing the data

6.1 What is the long-term strategy for future proofing of the data? Transcript data will be stored as Microsoft Word documents and NVivo files. These will be uploaded onto the password protected cloud storage Figshare in encrypted files and kept as private documents at the conclusion of the project for a period of six years from the end of the project.
6.2 How will the data be managed after the life of the project, for how long and in what format (NB this section refers to the detail of preservation and archiving actions, not just how it will be stored – this is addressed in section 5.2)? All data will be stored in encrypted files at all times. Transcript data will be stored as Microsoft Word documents and NVivo files. These will be uploaded onto password protected cloud storage Figshare and kept as private documents at the conclusion of the PhD and stored for a period of six years from the end of the project.
6.3 If the data include confidential or sensitive information, how will these data be managed to prevent possible future breaches? Data will not be stored with confidential labels attached. Transcript data has been anonymised and does not contain personal data, but it may contain confidential or sensitive information. Files will be stored in encrypted files, in password protected storage systems and files will be only accessible to the Researcher.
6.4 If metadata or explanatory information is to be archived, how will this be linked to the data? No metadata or explanatory information needs to be saved in relation this project data.
6.5 How will the data be cited? Not applicable, as it is not likely anyone would cite the data directly.

Section 7: Resourcing of Data Management

This section is used to outline the staffing and financial details of the data management

7.1 List the specific staff who will have access to the data and denote who will have the responsibility for data management. Katie Cunnah (nee Oxtoby) – Responsible for data management
7.2 How will the data management described in this document be funded? N/A
7.3 How will data storage be funded? N/A

Section 8: Review of Data Management process

This section is used to clarify how data management will be an embedded part of the research project

8.1 How will the data management plan be adhered to?

This data management plan has been designed to align with data management for all current projects being undertaken by the Researcher at the University of Hull, in order to reduce the risk of error. Regular review meetings will be organised between Katie Oxtoby and her current research team to review adherence to and suitability of all data management plans relating to research undertaken at the University of Hull.

8.2 Who will review the data management plan? What is the schedule for this review?

Katie Cunnah (nee Oxtoby) will seek approval of the data management plan from the PhD Internal Examiner. Upon approval, ongoing reviews will commence throughout the Researcher's career.

Section 9: Statements and Personnel Details

9.1 Statement of agreement

I agree to the specific elements of the plan as outlined:

Researcher

Title	Mrs
Designation	Researcher
Name	Katie Cunnah (nee Oxtoby)
Date	22/10/19
Signature	

Researcher

Title	Dr
Designation	PhD Supervisor & Senior Lecturer
Name	Kim Dent-Brown
Date	22/10/19
Signature	

Researcher

Title	Dr
Designation	Line Manager (Postdoctoral Role), Senior Lecturer & Director of the Centre for Human Factors
Name	Fiona Earle
Date	22/10/19
Signature	

9.2 Expertise of Researchers

Title	Dr
Name	Katie Cunnah nee Oxtoby
Contact Details	**Centre for Human Factors & Department of Psychology** T: 01482 463364 E: k.cunnah@hull.ac.uk
Expertise	NVivo Qualitative Research Grounded Theory Psychology Work-related stress Enterprise

Title	Dr
Name	Kim Dent-Brown
Contact Details	**Department of Psychology** T: 01482 462021 E: k.dent-brown@hull.ac.uk
Expertise	Qualitative Research Psychology Occupational Therapist

	Dramatherapist Cognitive Analytic Therapist Mental Health Evaluation of psychological services

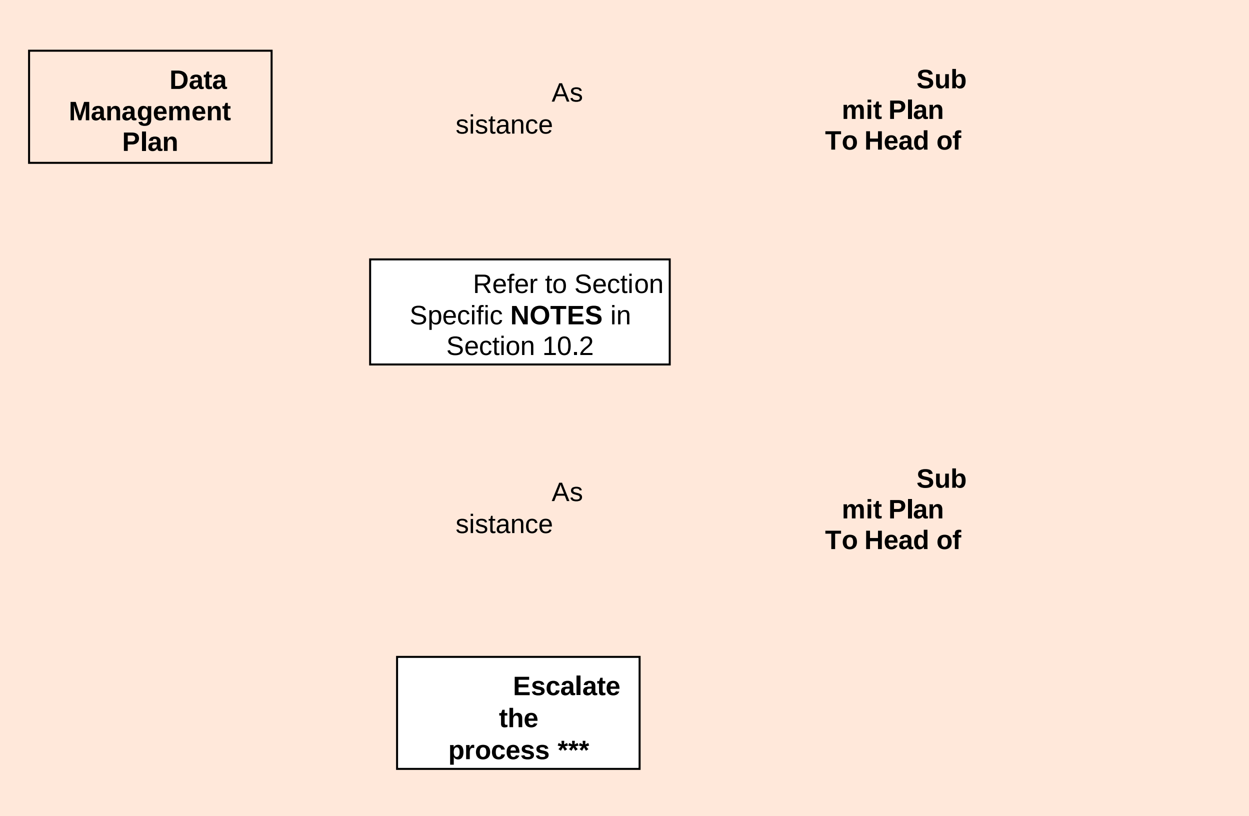
Title	Dr
Name	Fiona Earle
Contact Details	Centre for Human Factors & Department of Psychology T: +44(0)1482 465552 E: f.earle@hull.ac.uk
Expertise	Occupational Psychology Human Factors SPSS Quantitative data analysis Research Design Enterprise

** More than one Researcher may be involved. Continue on a separate page if necessary.

Section 10: Appendices

10.1 Specific Help with completing the Plan

In certain instances, specific guidance may be required in order to complete this Data Management Plan. Assistance should be sought by following the flow chart below:



Escalate the process by requesting assistance from the Departmental Head of Research. Typically this will entail contacting Departmental or Faculty data managers and/or Library and Learning Innovation and ICTD. Specific assistance may be available through the Research Funding Office as well, particularly in relation to funder requirements for data management.

10.2 Notes

These notes refer to the specified sections and subsections in this document. Any areas not addressed may be referred to the project lead, supervisor, or the Head of Research. Technical issues may be addressed to the HDMP development team in the first instance.

Front Cover

Details are required to ensure the correct future referencing, storage and archiving of the Data Management Plan. There will be strict adherence to applicable law, including the Data Protection Act; this information will not be made available outside of the specific remit of the Faculty of Health and Social Care of the University of Hull.

Section 1: Project Information

1.1 No specific guidance available

1.2 No specific guidance available

1.3 Required for funded projects – this refers to organisations other than the University of Hull

1.4 If necessary, further information may be provided on an attached, clearly labelled **typed** or **printed** sheet. For online forms, the space will automatically be increased to accommodate extra text.

1.5 State what local requirements are in place – details from Head of Research

1.6 Details may be requested from the project Supervisor, or the Head of Research.

1.7 Applies specifically to funded projects. If necessary, further information may be provided on an attached, clearly labelled **typed** or **printed** sheet. For online forms, the space will automatically be increased to accommodate extra text.

1.8 Applies specifically to funded projects. If necessary, further information may be provided on an attached, clearly labelled **typed** or **printed** sheet. For online forms, the space will automatically be increased to accommodate extra text. Details may be requested from the project Supervisor, or the Head of Research.

Section 2: Data, Materials, Resource Collection Information

2.1 If necessary, further information may be provided on an attached, clearly labelled **typed** or **printed** sheet. For online forms, the space will automatically be increased to accommodate extra text. NOTE: details may change as the project evolves; provide a best estimate.

2.2 If necessary, further information may be provided on an attached, clearly labelled **typed** or **printed** sheet. For online forms, the space will automatically be increased to accommodate extra text.

2.3 It is vital that there is a clear understanding of exactly which data types are being discussed in order to plan for future storage, accessibility and integrity. Example data types and formats are available at http://en.wikipedia.org/wiki/List_of_file_formats.

2.4 A great deal of non-digital data may need to be stored securely and/or archived. Various examples of this type of data are:

- Documents: Printed digital, Original artefact, , etc.
- Images: Photographs (size, print type, age), posters, etc.
- Artefacts: Physical model (scale/non-scale, size, availability), archaeological, etc.
- Film: 8/16/32mm, Video, microfilm, negative, etc.
- Other: Live performance, logical model, etc.

2.5 "Standalone" implies a provided information resource that requires no further explanation and may be used "as is" without additional resource. Accompanied implies information that is informed by accompanying documentation or resource(s) which help to understand the resource. For example, a database may need to be accompanied by a "metadata" informative document which explains the purpose, use of specific fields, and instructions for utilisation. Details may be requested from the project Supervisor, or the Head of Research.

2.6 Quality Assurance/Management in this context refers to the concise provision of a breakdown of what will be done to ensure that the project's progress will be monitored for accuracy, quality of work or research, and timely delivery at regular intervals. Typically, this would be the remit of the Research Supervisor, the Project Lead, or the Head of Department. Details may be requested from the project Supervisor, or the Head of Research.

Section 3: Ethics, Intellectual Property, Citation

3.1 If your research has an impact on the welfare, confidentiality or economic status of any individual or corporate group, this should be clearly stated. If necessary, further information may be provided on an attached, clearly labelled **typed** or **printed** sheet. For online forms, the space will automatically be increased to accommodate extra text. **NOTE**: details may change as the project evolves; provide a best estimate.

3.2 It is vital to comply with applicable law. Provide a brief outline of how relevant legislation and regulations will be complied with where appropriate. Where there is any doubt, the first line of contact is the project Supervisor, or the Head of Research.

3.3 See note 3.2 above. Partners in the project must be held to the same legal and regulatory standards. Partners are also protected by applicable law and may avail themselves of the prospect of legal recourse in the event of any perceived illegality or infringement by any party. This applies to all participants effecting or affected by the research project. Where there is any doubt, the first line of contact is the project Supervisor, or the Head of Research.

Section 4: Access and Use of Information

4.1 Sharing data, i.e. making it publically available, may be a requirement of a funding bid, or of a University research project (e.g. Doctoral thesis or research project). Details may be requested from the project Supervisor, or the Head of Research.

4.2 Provide details of how you intend to share your data (if relevant). This may include several options, such as an online accessible dataset or database, or online images. It could also be in the form of a paper based document or set of documents. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

4.3 If your data are sensitive (e.g. not suitable for general access until you have completed, or contains personal data or information) you may need to keep the data secure until you are ready to publish – if at all. Similarly, if the project funder requires "mile-stone" releases, this should be indicated. If in doubt, check this with the project Supervisor, or the Head of Research.

4.4 It is vital that you have a clear perspective of who the outcome of your research is intended to reach. Funding bodies may stipulate specific outcomes – e.g. public access, etc.

4.5 Funding bodies will typically require an explanation of the usefulness of your research once completed, and you should be able to provide a clear idea of what will be done with your data once published or released. Certain obvious options should not be overlooked, such as: paper presented at conference for history community, or book chapter published for community and public research/interest, etc.

Section 5: Storage and Backup of Data

5.1 It is vital that the research materials and data are kept *safely at every stage* of the research process lifespan. There may be help available from IT Services, the Library or the Department. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

5.2 As for 5.1 above, it is vital that you have a clear understanding of how, where and when the research materials and data will be maintained after research process lifespan. This is particularly true where funding bodies have specific outcome criteria (e.g. making a public website available, etc.). There may be help available from IT Services, the Library or the Department. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

5.3 Similarly to 5.1 and 5.2 above, it is vital that you have a clear understanding of how, where and when the research materials and data will be backed up and kept safely, both during and after the after the research process lifespan. This is particularly true where funding bodies have specific outcome criteria (e.g. ensuring that online datasets are maintained for a specific period after the end of a project, etc.). There may be help available from IT Services, the Library or the Department. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

5.4 Very often work is added to, revised or altered and older versions are either overwritten, left as they were, or deleted. It may be wise to maintain a clearly labelled and stored set of older versions of current work in order to backtrack if necessary. Itis imperative that a logical and sequenced filing system is used. On computer systems this may be attained by uniquely numbering each version. A useful means of achieving this is by using the current date and time as the unique numbering reference – e.g. "yyyymmdd FHSC Data Management Plan".

Section 6: Archiving and Future Proofing of Information

6.1 Provide information about how you intend for the project outcome(s) or deliverable(s) to be maintained after the end of the project. For example, a dataset may be perpetually maintained by the University's online provision. However, this will need to be confirmed. There may be help available from IT Services, the Library or the Department. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

6.2 Any information that is kept after the lifespan of a project will still need to be stored safely, maintained and be provided in a useable format. If specific file formats are used, they may become unusable after a few years as new software replaces the old. Also, media such as DVDs, CDs and diskettes may become unusable after a while. There may be help available from IT Services, the Library or the Department. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

6.3 It is vital that any confidential data (e.g. personal information about any individual who is protected under the terms of the Data Protection Act, or information that may infringe copyright if released, etc.) must be kept and maintained in a secure environment. All reasonable steps should be taken to ensure the safety of such information. This applies to any information that is kept after the lifespan of a project as well. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

6.4 Datasets, databases, standalone documents, and even artefacts may prove useless without explanatory notes (metadata) accompanying them. These materials need to be clearly linked to the materials so that they can adequately inform any future user about the material. For example, a published dataset will typically be accompanied by a metadata document that explains the various fields, their usefulness and summarises the purpose of the dataset in general. These documents will be stored along with the dataset and are accessible in the same manner as the dataset (e.g. online, or download). Examples of such accompanying documentation are available for download. If you wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

6.5 Typically, any stored data, materials, artefacts, etc. will need to be cited when accessed and referenced by other researchers. It is useful to provide clear and concise citation information for researchers to access. This can be done via the accompanying documentation (metadata) indicated in 6.4 above. If you wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

Section 7: Resourcing of Data Management

7.1 In the event that this is an individual project or piece of research, your own name should be listed. Include any other staff or assistants are to be involved in the project as well. It may be necessary to include staff from other departments of the University. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

7.2 Funding strategies are often outlined by funders and will include a data management aspect. The costs of any materials, equipment and specialist knowledge will need to be factored to arrive at a reasonable estimate. Include any materials or equipment that will be funded by the University and/or you. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

7.3 As in 7.2 above, funding strategies are often outlined by funders and will include a data management aspect. Typically the University will support on-going research projects, and assist in facilitating post project maintenance and/or presence of outputs. However, this needs to be confirmed to ensure that the service will be available in the form that is required. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

Section 8: Review of Data Management process

8.1 Funders will need to be informed about how the data management process will be implemented. Provide specific information about how you intend to follow through with the commitments and processes that have been discussed in the rest of this document. Typically, regular reviews, reports and assessments of progress will suffice, but some funders may require specific means of identifying adherence to the plan. If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

8.2 Based on 8.1 above, list those who will be carrying out the reviews and subsequent reports or processes necessary to ensure the successful implementation and completion of the data management plan. Typically, in the event of smaller research projects or individual research, the project Supervisor will fill this role. In the event of PhD research, this role will be carried out by the PhD Supervisor(s). If you are uncertain, or wish to explore this avenue further, the first line of contact is the project Supervisor, or the Head of Research.

Section 9: Statements and Personnel Details

9.1 The Statement of Agreement is necessary to clarify the areas of responsibility and work that will be carried out by the various researchers engaged in the project. This information is vital for funding bodies that will require these details.

9.2 As in 9.1 above, the Expertise of Researchers is necessary to clarify the areas of responsibility and work that will be carried out by the various researchers engaged in the project. This information is vital for funding bodies that will require these details in the form of a brief résumé for each researcher.

Section 10: Appendices

10.1 Assistance with completing the Plan; follow the instructions to obtain help specific to each section.

10.2 Follow the guidance for each specific section as necessary.

10.3 This list of Relevant Contacts will be reviewed and altered regularly.

10.3 Relevant Contacts

The following list of contacts should be regularly revised as appropriate for the purposes of your research:

Head of Research	
Library and Learning Innovation	Chris Awre Head of Information Management Phone: +44 (0) 1482 465441 Email: c.awre@hull.ac.uk
ICT Directorate	IT Helpdesk Phone: +44 (0)1482 462010 E-mail: help@hull.ac.uk
Head of Department	
Document Author	Chris Awre Details as above