

Long-term Experiences of Managing Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME): A Qualitative Study

Deborah Samantha Williams

(née Baldwin)

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Faculty of Health and Applied Sciences, University of the West of England, Bristol

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Abstract

Background: Frustrations are noted in the management of CFS/ME as it's a complex and individual condition with no known cure. Despite being a Long-term Condition (LTC) limited research has focused on long-term experiences. This study aims to extend the knowledge of long-term experiences of CFS/ME specifically focusing on management of the condition. Thinking about therapeutic moderators and mechanisms of change, whether management changes throughout the course of the illness and what support people might benefit from.

Methods: A qualitative research design, using semi-structured interviews was adopted. Nine participants' were recruited from a specialist CFS/ME Service in the UK who were over 18 years old, had a diagnosis of CFS/ME and reported experiencing fatigue related symptoms for over 5 years. Interviews were audio recorded, transcribed verbatim and analysed using Thematic Analysis. **Findings:** Three themes; Awareness, Acceptance, Connection and two subthemes; connection with self and connection with others were constructed from the data. An overarching theme of Awareness appeared to facilitate the ability to accept and connect with what was important for people with CFS/ME, enabling people to adapt to living with the condition and achieving a standard of living. Commonalities occurred across all themes of development over time, individuality and ongoing balance or monitoring. **Discussion:** Findings suggest supporting adults with CFS/ME to become more self-aware of their illness experience and identifying their values will be beneficial at any stage of the illness duration, but particularly important for long-term management. These findings provide further support for tailored treatment plans (NICE, 2007) with some individuals' needing more, or occasional contact with understanding professionals to achieve. The results support the use of acceptance-based interventions in LTC management; specifically Acceptance and Commitment Therapy (ACT) and Focused ACT. Further research into outcomes and experiences of ACT in CFS/ME, and interdisciplinary approaches is advocated.

1 Introduction

1.1 CFS/ME

1.1.1 Terminology

Myalgic Encephalomyelitis or Encephalopathy (ME) translates to pain and an inflammation of the brain and spinal cord (Campling and Sharpe, 2008). However, to date there is limited biological evidence of any inflammation or conclusive biological markers (Hamilton et al, 2005), so the Centers for Disease Control and Prevention (CDC) suggested a more accurately named Chronic Fatigue Syndrome (CFS) in the late 1980's (Arroll and Senior, 2008). Chronic denotes the significant length of time people are affected (unlike acute illnesses), which is proposed as an excess of 6 months; Fatigue being the overriding symptom and Syndrome to convey a collection of other symptoms experienced (Fukuda et al, 1994). Although it has been suggested there is a difference between CFS and ME (Twisk, 2014) it continues to be disputed whether they are in fact different illnesses (Hamilton et al, 2005). Many patients report preferring the ME label as it sounds more medicalised. Similarly a preference of the term ME can also be noted by the two leading UK charities; Action for ME (AfME) and the ME Association (Arroll and Senior, 2008; Brooks, King and Wearden, 2014; Campling and Sharpe, 2008). This thesis will use the abbreviation CFS/ME throughout. Firstly due to the absence of clear evidence that ME and CFS are separate conditions, secondly to be inclusive of people using the different terms to describe their illness and thirdly to reflect the descriptions adopted in the literature (Krzeczkowska, Karatzias and Dickson, 2015).

1.1.2 Associated terms

An associated term with CFS/ME is Medically Unexplained Symptoms (MUSs) which is a broader label than can include various heterogeneous conditions, like CFS/ME, that cause distress but have no organic cause (Brown, 2006). Similarly other labels that appear in the fatigue literature are Burnout (often related to work or study), exhaustion, Chronic Fatigue and Immune Dysfunction Syndrome (CFIDS) (often used in America) and Neurathenia (a very historical term from the late 1880's meaning 'weak nerves') (Campling & Sharpe, 2008).

1.2 Presentation

Common symptoms that accompany the overriding symptom of fatigue include: post exertion malaise, poor, disturbed and un-refreshing sleep, myalgia, poor concentration, memory difficulties, headaches, swollen glands, persistent sore throats and sensitivity to light and noise (Fukuda et al, 1994; Sharpe et al, 1991; Söderlund, Skoge and Malterud, 2000). The CFS/ME picture is complex, as it is a heterogeneous condition that affects people differently, not

everyone with CFS/ME will experience the same symptoms and similarly symptoms can change in severity as well as over time, often referred to as an individual and fluctuating condition (Anderson et al 2012: Friedberg and Jason, 1998: Jason, 1999: Larun and Malterud, 2007: Lombaard and Mouton, 2005: NICE, 2007: Pinxsterhuis, Strand and Sveen, 2015: Söderlund, Skoge and Malterud, 2000).

Due to the changes in symptom severity and frequency many people with CFS/ME report feeling out of control (Horton-Salway, 2004: Jason, 1999: Tucker, 2004). Unsurprisingly the symptoms experienced negatively impact people's lives specifically the ability to carry out normal daily routines, such as self-care activities, caring for others and engaging in work-related activities (Attree et al, 2014: Söderlund, Skoge and Malterud, 2000). Similarly the ability to interact, communicate and sustain relationships is impeded. On this basis '*CFS/ME intrudes into more life domains (i.e. work, recreation and health), and to a greater degree, than other chronic illnesses*' (p.198, Stenhoff et al, 2015). Health status prior to illness is described as being a high achiever and operating at high speed (Pemberton and Cox, 2014: Söderlund, Skoge and Malterud, 2000: Ware, 1999), resulting in a striking difference observed after illness onset (McInnis et al, 2015). As a result people with CFS/ME can experience low mood and in some cases clinical depression and, or anxiety (Castell, Kazantzis and Moss-Morris, 2011: Hadlandsmyth and Vowles, 2009). This supports many findings that report people with CFS/ME often indicate significantly higher levels of anxiety and depression than a 'healthy' control groups (Deary and Chalder, 2010: Oldershaw et al, 2011).

1.3 Aetiology

To date there is no definitive explanation for CFS/ME aetiology, with only theoretical involvement of immunological, genetic, viral, neuroendocrine and psychological factors, either singularly or in combination (Deary and Chalder, 2010: NICE, 2007). Like most conceptualisations of health and illness, there is an assumption that our experiences are based on multiple factors that influence our experiences; which is largely uncontested therefore theories of causation of CFS/ME are largely multifactorial (Deary and Chalder, 2010: Harvey and Wessely, 2009: NICE, 2007: Söderlund, Skoge and Malterud, 2000).

Commonly an initial physical episode (viral infection or immunological disorder) is recalled before becoming significantly fatigued, sometimes labelled as Post Viral Fatigue Syndrome (PVFS). Related to this is the Epstein-Barr virus (also known as Glandular Fever) often occurring prior to developing CFS/ME (Ax, Gregg and Jones, 2001). This is not to say all people that have Glandular Fever go on to develop PVFS or CFS/ME, but the link between this virus and CFS/ME is frequently reported (Whiting et al, 2001), with some thought PVFS

is as a subclass of CFS/ME (Campling and Sharpe, 2008; White et al, 1998). In addition to viral infections, many people experience significant stress (emotional distress, significant life changes, bereavement etc) prior to CFS/ME onset (Anderson et al, 2012; Deale et al, 1997).

Some gender differences have also been noted in theories of causation as demonstrated in a qualitative study by Clarke (1999) where men attributed CFS/ME to working environments, whilst women attributed to stress. However, there is such limited research for men with CFS/ME in particular; it remains unclear if significant gender differences occur in causal attributions or illness experience.

1.3.1 Predisposing, Precipitating and Perpetuating model

A widely used biopsychosocial conceptualisation of CFS/ME that draws on many different studies and belief systems, details three phases that frequently occur in the complex CFS/ME picture (Friedberg and Jason, 1998). The first phase includes Predispositions such as inherited vulnerability (Campling and Sharpe, 2008), personality qualities (namely perfectionism and conscientiousness), early childhood illness and over-activity (Deary and Chalder, 2010; Harvey et al, 2008; Kato et al, 2006; Viner and Hotopf, 2004; Vos-Vromans et al, 2013; Ware, 1999). These factors exist prior to a triggering event which marks the second phase of this model, also referred to as Precipitating factors. Most frequently an infection or viral episode and/or significant stress are reported as precipitating factors, resulting in an initial fatigue episode (Anderson et al, 2012; Campling and Sharpe, 2008; Harvey and Wessely, 2009). The third phase encompasses a variety of aspects which maintain the initial fatigue causing CFS/ME. Previous studies have highlighted that these maintaining, or Perpetuating factors can include biological (endocrine function, sleep and body mass index), psychological (illness perceptions and cognitions, attributions, high expectations, stress, psychiatric disorders and self-esteem), behavioural (activity levels and sleep) and social elements (poor social relationships, relationships and roles with parents) (Coremans and Vause, 2013; Gotts et al, 2015; Gray and Rutter, 2007; Harvey and Wessely, 2009; Lievesley, Rimes and Chalder, 2014; Oldershaw et al, 2011).

There is also an acknowledgement that some aspects are not limited to one phase, for instance prolonged stress can be thought to predispose someone developing CFS/ME and be a potential precipitating trigger for onset, as well as a perpetuating factor if this if the stress continues. Equally an '*over-active lifestyle*' defined as high achievers, multi-tasking with busy lives has been identified as both a potential perpetuating factor and a predisposition (Lievesley, Rimes and Chalder, 2014; Vos-Vromans et al, 2013). That said, the sheer contrast in personality that is observed following CFS/ME, presenting the person as less extroverted

than the 'normal' population, has actually been described as an '*illness artefact*' (p.471 Deary and Chalder, 2010) rather than a CFS/ME susceptibility.

This thesis will periodically refer to this 3P's model to situate relevant theories and study findings firstly because as it is so widely adopted in attempting to understand CFS/ME literature and secondly it can account for individual and discipline variance in causal attributions. This is particularly relevant at a time when concrete conclusions cannot be drawn regarding aetiology.

1.4 Prevalence

The National Institute for Health and Care Excellence (NICE) estimated 0.2 - 0.4% of the population are affected by CFS/ME. It has also been proposed approximately 250,000 people are affected in Britain alone (Macrae, 2015).

Gender differences are noted in the prevalence of the condition, with four times more women reported to be affected (NICE, 2007). This is also reflected in the demographic information reported in CFS/ME literature (Ax, Gregg and Jones, 2001; Baldwin, 2013; Whiting et al, 2001). This could indicate women are more susceptible to CFS/ME relating to hormonal or genetic factors or could indicate the figures include a potential bias for more females getting diagnosed or perhaps a combination of both. The latter argument might link to comparatively lower numbers of men seeking medical help and therefore male CFS/ME is under-reported (Yousaf, Grunfeld and Hunter, 2015).

A higher prevalence within white ethnic backgrounds has also been observed in documented populations (Ax, Gregg and Jones, 2001; Baldwin, 2013; Williams, Johnson and Smyth, 2015). However, there is some research to challenge this as one of six health care professionals (HCPs) reported that in their experience, the common presentation of a white, middle class female was not supported (Horton et al, 2010). Similarly more recent research proposes CFS/ME could be more prevalent in Black Minority Ethnic (BME) groups, but due to biases in diagnosis and referral pathways and health seeking behaviour lower rates of CFS/ME are reported (Bayliss et al, 2014a; Bhui et al, 2011; Luthra and Wessely, 2004).

Obtaining accurate prevalence figures is challenging given the difficulties relating to achieving a diagnosis (further discussion to follow) but can include practitioner scepticism and access to GP services. As with many conditions there is an assumption the scale of the problem is under-reported.

1.5 Diagnosis

CFS/ME was recognised as a chronic and treatable condition in the UK in 2004 and was likened to other long-term health conditions (LTCs) such as Multiple Sclerosis and Rheumatoid Arthritis (McCue, 2004; NICE, 2007). The World Health Organisation classifies CFS/ME as a neurological disorder, although this classification has been rejected by some that it is biologically inaccurate (Campling and Sharpe, 2008; Dayes, 2010), similarly to the ME label.

1.5.1 Diagnostic criteria

Two main diagnostic criteria are commonly used to diagnose CFS/ME (Flo and Chalder, 2014); CDC Criteria (Fukuda et al, 1994) and Oxford Criteria (Sharpe et al, 1991). The CDC Criteria specify minimum of 4 specified symptoms; sore throat, lymph node pain, muscle pain, joint pain, post-exertional malaise headaches, memory and concentration difficulties and unrefreshing sleep. In addition to these symptoms the fatigue must be a new onset of fatigue that is not alleviated by rest. The fatigue must impact occupational, social or educational activities, which has lasted longer than 6 months, where no organic cause can be found. The Oxford criteria is similar but specifically includes mental fatigue within its symptomatic check list (Sharpe et al, 1991).

Both CDC and Oxford criteria have been criticised to be too inclusive, particularly being unable to discriminate between mental health conditions and CFS/ME. Therefore an alternative; the Canadian criteria (Carruthers et al, 2003) is preferred by some. The Canadian criteria places emphasis on cardiovascular and neuroendocrine disturbances, rather than psychiatric features (Jason et al, 2004). It is worth noting however, despite these proposals the Canadian criteria appears more frequently in American and Canadian research papers, with the CDC criteria most commonly used in the UK (Densham et al, under review; McDermott et al, 2014).

1.5.2 Diagnosis procedure

Diagnosis relies on patient reported symptoms and negative blood screens to exclude other medical reasons for fatigue (Chambers et al, 2006; Fukuda et al, 1994), for example anaemia, pregnancy or thyroid dysfunction. Further red flags questioning a CFS/ME diagnosis could be; active eating disorders, sleep disorders and mental health conditions. Illness duration was revised from 6 months (Fukuda et al, 1994; Sharpe et al, 1991) to 4 months (NICE: 2007) to enable people to access support earlier. This is particularly important as a diagnosis of CFS/ME is often reported to provide validation and legitimisation of illness (Anderson et al, 2012; Drachler et al, 2009; Horton et al, 2010; Larun and Malterud). Whilst a diagnosis is often given by medical practitioners most commonly a GP, patients and GPs have expressed that

referral to specialist services is important to confirm the diagnosis (McDermott, Lynch and Leydon, 2011).

1.6 Difficulties diagnosing CFS/ME

1.6.1 Comorbidities

It has been acknowledged in various qualitative studies there are difficulties for both practitioners and patients regarding CFS/ME diagnosis (Anderson et al, 2012: Larun and Malterud, 2007). One of the many complications with diagnosing CFS/ME already alluded to is the overlap of other conditions in addition to the known impact of aging on health outcomes (Jason et al, 2011: Joyce, Hotopf and Wessely, 1997). Co-associated disorders that are common with CFS/ME are depression, Irritable Bowel Syndrome (IBS) and Fibromyalgia (FMS) (Blazquez, Alegre and Ruiz, 2009: Campling and Sharpe, 2008: Jason, Taylor and Kennedy, 2000: Maes 2011: Whitehead, Palsson and Jones, 2002). Chronic fatigue could be present because of, in conjunction with or could be masking other health conditions. CFS/ME in nature is heterogeneous and can contribute to GP's confusion and reluctance to diagnose, based on a varied symptom presentation (Chew-Graham et al, 2010). This is also illustrated by the considerable overlap between FMS and CFS/ME symptomology, and both are contested conditions (Campling and Sharpe, 2008). Similarly there is debate whether the red flags mentioned earlier, such as mental health co-morbidities are grounds for refusing a fatigue diagnosis. This is because the treatment options currently available for CFS/ME are centred around management approaches which would still be applicable, regardless if fatigue is the primary health condition. Some clarity around this has been suggested by Anderson et al (2014) indicating if the mental health condition has been present for longer than 5 years, it should not be treated as exclusion to obtaining a CFS/ME diagnosis.

1.6.2 GP access and HCP beliefs

Another barrier in obtaining a diagnosis was reported by Bayliss et al (2014) which links to access to GPs. A further factor highlighted in this qualitative study was that patients tended to make GP appointments in relation to physical symptoms such as pain, rather than the non-descript symptom of fatigue. As CFS/ME is variable in the symptoms experienced, the perception of the symptoms could prevent access to GP's and subsequently delay or prevent an accurate diagnosis and treatment.

A further reluctance in seeking support for CFS/ME has been reported due to a perceived stigma compared to other conditions (including mental health conditions) and perceived inferior treatment by HCPs (McInnis et al, 2015: Tucker, 2004). Lack of understanding of

others has been widely reported by those with CFS/ME (Anderson et al, 2012: Larun and Malterud, 2007) and there is a well-documented history of practitioner scepticism of CFS/ME, subsequently de-legitimising the condition for patients (Asbring and Narvanen, 2003: Raine et al, 2004). Another difficulty in achieving an accurate diagnosis can be affected by medical gender stereotypes. McCue (2004) found when presenting with the same symptoms GPs are more likely to diagnose women with a mental health condition rather than CFS/ME compared to men. These factors can act as a significant barrier for people seeking support if they do not feel they are believed or taken seriously (Larun and Malterud, 2007: Stenhoff et al, 2015: Tucker, 2004).

That said, Chew-Graham et al (2010) indicated GPs are aware of the stigmatisation and as a consequence, GPs are cautious to diagnose CFS/ME because of the negative impact of the condition label. In some instances this hesitance could be helpful to enable confidence in the diagnosis if and when provided, but in other cases it delays diagnosis and can potentially damage the patient-doctor relationship. Interestingly Clements et al (1997) concluded patients and doctors may have more shared beliefs about CFS/ME than expected. This research indicates the importance of fostering open communication to discover such shared beliefs. Echoed in more recent research also emphasises the importance of supportive communication in CFS/ME rehabilitation (Gladwell et al, 2014). Heins et al (2013) similarly advocates a shared understanding about cause of the condition between patient and practitioner can assist with building a good therapeutic alliance. This would also facilitate working collaboratively towards aligning perspectives which was a need expressed by people with CFS/ME (Drachler et al, 2009).

1.7 Treatment

In addition to the personal burden of CFS/ME there is an estimated annual cost of £75-129 million in the UK related to loss of productivity and burden of disease on healthcare systems (Collin et al, 2011). Therefore the need to adopt effective treatment is vital for patients and the wider community to improve quality of life as well as consider cost efficiency.

1.7.1 Treatment Setting

Approximately 49 specialist CFS/ME Services exist in the NHS across the UK that autonomously implement the NICE (2007) guidelines (AfME, 2015: McDermott et al, 2014). Patients' have reported they need diagnosis, guidance, support and hope from specialist services (Gladwell et al, 2014: McDermott, Lynch and Leydon, 2011: Pinxsterhuis et al, 2015) and without the support and hope from specialist services people can feel abandoned with

serious detriment to health status (Arroll and Senior, 2008: Larun and Malterud, 2007: McDermott, Lynch and Leydon, 2011).

Most commonly specialist services operate at a tertiary care level requiring referrals from GPs, because of this there can be a delay in accessing support if a diagnosis has not been given. Frustrations around this have been reported by patients requesting more information earlier on in their fatigue journey to promote self-management (Drachler et al, 2009: Haig-Ferguson, 2014: McDermott, Lynch and Leydon, 2011). This is echoed in research stating education and advocacy as particularly important for effective coping (McKenzie et al, 1995: Pinxsterhuis et al, 2015) however there is considerable variation in needs and availability to support (Bayliss et al, 2014).

Therapists within specialist services can also vary depending on location and can include; GPs, nurses, occupational therapists, psychologists and physiotherapists. Houlton et al (2015) highlighted that CFS/ME service provision is inconsistent in the UK, with a variation of treatment options and delivery style being offered. Similarly AfME have reported 65 per cent of people surveyed had not been offered any treatment for their CFS/ME (Bayliss et al, 2014b). Equally support for those severely affected who are often bed or house-bound is variable; with 16/49 NHS CFS/ME Services not providing a service for severely affected individuals (McDermott et al, 2014). It is also frequently reported this sub-group are not represented in CFS/ME research, often a self-reported limitation of research studies.

1.7.2 Treatment options

Whilst it's ideal to receive a diagnosis and treatment as soon as possible to minimise the impact of CFS/ME, it's particularly encouraging to know illness duration is not a moderating factor for outcomes (Castell, Kazantzis and Moss-Morris, 2011: Malouff et al, 2008: Poppe et al, 2013: Wilson et al, 1994), especially given the historical issues with achieving a diagnosis, delaying support and subsequently negatively impacting mood.

For some, because there is currently no cure for CFS/ME this has translated into GP's lack of hope for patient improvement (Chew-Graham et al, 2010: McDermott, Lynch and Leydon, 2011: Vercoulen et al, 1998). That said, improvement can be possible without fully understanding the cause of the illness (Deale et al, 1997). As the comparison offered by Fernie and Murphy (2009) illustrates; a broken leg can be treated regardless if it was caused by a football injury or falling due to an uneven pavement. It is with this mind-set CFS/ME treatment tends to target the Perpetuating factors or symptoms that can exacerbate the condition (Harvey and Wessely, 2009: Lievesley, Rimes and Chalder, 2014: Vos-Vromans et al, 2013).

Various approaches have been used to treat CFS/ME including; pharmacological, complementary therapies, psychological therapies, dietary and nutritional with varying degrees of success (Malouff et al, 2008: Whiting et al, 2001). It has also been noted that whilst some people have found different therapies to be helpful, NHS practitioners are unable to make recommendations for therapies without evidence base for them (Horton et al, 2010). Similarly to diagnosis, successful management of the condition can also be '*hampered by insufficient understanding of the condition, and further complicated by practitioner scepticism*' (Söderlund, Skoge and Malterud, 2000 p.165).

1.8 NICE guidelines

Chambers et al (2006) conducted a Systematic Review (SR) which informed the NICE guidelines (2007) on the treatment and management of CFS/ME. Reviewing the available evidence they concluded that behavioural interventions, specifically Cognitive Behaviour Therapy (CBT) and Graded Exercise Therapy (GET), combined with GP symptomatic treatment were the most effective interventions for CFS/ME. CBT and GET fall under the umbrella of behavioural interventions and were classified in this way to allow for the inclusion of other non-pharmacological treatments such as, non-directive counselling and other activity management approaches.

1.8.1 Updated review

Since the NICE (2007) guidelines were published, (data collection performed before August 2005), there have been many more studies conducted. In response to this an updated SR (Baldwin, 2013) was performed for behavioural interventions for CFS/ME in adults (See Appendix A). The main findings from this updated SR were consistent with Chambers et al (2006) review, providing further evidence that behavioural interventions are beneficial for adults with CFS/ME. Similarly to the first review, the specific interventions most effective in improving fatigue, functioning, depression and anxiety outcomes were cognitive interventions: (Cognitive Therapy (CT) and CBT) and GET. Also supporting the earlier recommendations for more research to investigate the long-term effects of interventions (long-term follow up defined as ≥ 12 months). That said the number of studies assessing outcomes 12 months or over had increased to 55% of studies compared to the initial review. Further conclusions of this updated review highlighted the need to understand what specific elements of therapies are most effective and whether the use of combination interventions can harness the optimal elements of treatment for the best outcomes.

1.9 Why are behavioural interventions effective?

Given the limited control over the predispositions or precipitating elements that precede individuals developing CFS/ME and the initial trigger of the illness (such as a virus), it seems reasonable to focus on perpetuating factors that maintain or exacerbate CFS/ME symptoms. Behavioural interventions have been described as a way to equip people with ‘*assessable tools*’ to respond to the profound changes caused by the condition such as; the effect on relationships, financial implications and impact on mood (Blazquez, Alegre and Ruiz, 2009). These tools can assist in bringing about a sense of control and empowerment for people which is often lacking as a result of living with a fluctuating and misunderstood condition (Horton-Salway, 2004: Jason et al, 1999: Pinxsterhuis et al, 2015: Pinxsterhuis, Strand and Sveen, 2015: Vercoulen et al, 1998). Wilson et al (1994), an Australian study investigated predictors of long-term outcome in CFS/ME found whilst functional impairment remained present over time, illness beliefs and coping style affected long-term outcomes compared to immunological or demographic variables. These findings may explain why behavioural interventions, demonstrate more significant improvements for people by targeting unhelpful illness beliefs and coping styles, and/or facilitating more helpful illness beliefs and coping.

1.10 Activity interventions

Reducing activity has been described as one way to achieve control over CFS/ME with many studies have reported effective coping by reducing activities as a way to accommodate the fatigue and related symptoms to reduce the occurrence of set-back/relapses for a more stable illness trajectory (Anderson et al, 2012: Clements et al, 1997). However, the reduction in physical activity can subsequently lead to deconditioning and fears about activity fostering an unhealthy relationship with physical activity (Marques et al, 2015: Vercoulen et al, 1998). Similarly Lombaard and Mouton (2005) propose restricting activity or excessive activity is not helpful, but to find a balance to prevent physiological deconditioning and promote a sense of achievement for psychological wellbeing. This is sometimes referred to as staying within the energy envelope or baseline, which is considered an individual quantity (Jason, 1999).

1.10.1 GET

GET is a collaborative and supervised behavioural intervention for people with mild or moderate CFS/ME to gradually increase aerobic exercise appropriate for the individual, based on their abilities, goals and preferences. It is also recommended GET should include sleep and relaxation components as well as set-back planning as the intervention can result in an increase in symptoms (Gladwell et al, 2014: Marques et al, 2015: NICE, 2007: NICE, 2014). Walking is often a common exercise chosen as part of GET programmes to increase fitness gradually (White et al, 2011).

1.11 Psychological interventions

It is proposed that psychological interventions can address how the patient appraises and reacts to both internal and external experiences (Scott and Freeman, 2010), with a view to change the relationship with unpleasant thoughts, emotions and bodily sensations experienced reducing distress (Lopez et al, 2011). This is particularly relevant for CFS/ME as unpleasant and distressing thoughts, feelings and bodily sensations are frequent and persistent, which negatively impacting on the person's life (Pinxsterhuis, Strand and Sveen, 2015). Emotion-focused coping fits this description whereby emotions are regulated following stressful events as a way to respond to an event that cannot be changed. Psychological approaches can also assist people to move away from excessive symptom monitoring which can perpetuate unhelpful coping, in some cases creating health anxiety and draining energy levels further. People that cope with CFS/ME by focus on their symptoms have been found to have poorer outcomes (Gray and Rutter, 2007), which can also increase feelings of helplessness and not being in control, subsequently affecting mood.

1.11.1 CBT

CBT originated from the Cognitive-Behaviour approach which includes problem solving, changing focus and close attention to cognitive processes that are used to monitor and control behaviour. The focus of CT and CBT aims to identify and challenge unhelpful thoughts that reinforce problematic behaviours (Price et al, 2009). CBT itself is a broad term that refers to the work by Ellis in the 1960's and Beck in the 1970s (McLeod, 2003). CBT interventions have been widely used and demonstrating beneficial results for various health conditions (Stiles et al, 2006) including depression, anxiety, chronic pain, MUS and CFS/ME (Eccleston et al, 2002: Malouff et al, 2008: NICE, 2007, 2009, 2011, 2014: Price et al, 2009: van Dessel et al, 2014: White, 2000). A meta-analysis conducted by Malouff et al (2008) for CBT in CFS/ME reported a respectable effect size of 0.48 (using Cohen's criteria, just shy of a medium effect).

The CBT model of CFS/ME links with the 3 P's model, targeting some of the perpetuating factors of CFS/ME such as; over activity, unhelpful thinking styles and preoccupation with bodily sensations (Deary and Chalder, 2010). Various descriptions and manuals have been used but consistently CBT in CFS/ME involves therapists encouraging gradual activity increases, goal setting, examining the relationship between thought patterns, emotions, body sensations (symptoms) and behaviours to be able to increase awareness and challenge unhelpful responses (Malouff et al, 2008: Meng, Friedberg and Castora-Binkley, 2014: NICE, 2007: NICE, 2014: White et al, 2011). CBT addresses factors that can enable a person to become vulnerable, specifically conscientiousness and perfectionism characteristics, beliefs that certain emotions are unacceptable, fear avoidance and health or symptom anxiety (Flo

and Chalder, 2014). Detailed CBT study protocols for CFS/ME can be found elsewhere (see www.pacetrail.org, Jason et al, 2007).

There are many variations of CBT delivery (Malouff et al, 2008) which is a positive feature of the intervention; face to face, self-guided instructions, telephone, online and delivered individually or in groups. However, most commonly CBT in CFS/ME research studies have adopted an individual and face to face delivery (Baldwin, 2013). Interestingly Castell, Kazantzis and Moss-Morris (2011) found no differences between individual or group treatment and there have been promising patient and economic outcomes for brief CBT interventions in CFS/ME (Knoop, van der Meer and Bleijenberg, 2008; Meng, Friedberg and Castora-Binkley, 2014; Tummers et al, 2012). This is of interest given NICE (2014) advocate CBT is offered on a one to one basis if possible. In addition, for people with difficulties in accessing services, for geographic or illness severity constraints it is encouraging that there might not be a significant difference in outcomes for different delivery methods, which may also prove to be a more cost effective (Arroll and Senior, 2008; Houlton et al, 2015).

Interestingly illness duration has not been found to affect outcomes of CBT for those with CFS/ME (Pope et al, 2013). However, predictably CBT seems to be more effective for people with CFS/ME that also have depression or anxiety comorbidities (Castell, Kazantzis and Moss-Morris, 2011; Song and Jason, 2005). This also links to research that found people with CFS/ME indicate higher levels of neuroticism which suggests interventions such as CBT that target anxiety, loneliness, fear and frustration would be advantageous as these are the common features of neuroticism (Deary and Chalder, 2010). Equally unsurprisingly the dose of CBT suggests, the more sessions relates to better results (Castell, Kazantzis and Moss-Morris, 2011). Baldwin (2013) reported average number of sessions as 14, which is much higher than clinical practice. Potentially this links to concerns clinicians have regarding generalisation from research studies, time pressure and funding availability (Cranney et al, 2001).

1.12 Criticisms of behavioural therapies

1.12.1 Perceptions and de-legitimisation

CBT is widely researched in CFS/ME populations and therefore contributes a wealth of information to CFS/ME treatment evidence, which may account for the dominance in CFS/ME treatment. Although CBT is a psychological therapy and has been found to be effective in reducing CFS/ME symptoms, there is no firm evidence to suggest CFS/ME is a psychological condition (Hadlandsmyth and Vowles, 2009; Song and Jason, 2005). Furthermore, it has been reported that conceptualisation of CFS/ME as a psychological condition is damaging for

patients, resulting in stigmatisation and isolation, as well as preventing access to treatment and support (Anderson et al, 2012: Brooks, King and Wearden, 2014: Dayes, 2010: Larun and Malterud, 2007: McDermott, Lynch and Leydon, 2011: Raine et al, 2004: Whitehead, 2006a). Twisk and Maes (2009) also question current thinking around the dominance of psychological factors in CFS/ME and subsequently state treatment options based on this model of CFS/ME as incorrect.

Whilst the majority of people acknowledge the mind and body interaction, as illustrated by a participant quote *'I firmly believe it's [CFS/ME] physiological...I think psychology plays an important part but I think that's true with any illness'* (p.575, McDermott, Lynch and Leydon, 2011), some people with MUSs like CFS/ME report a reluctance in participating in psychological therapies because of the perception psychological support is only required for psychological illness (van Ravesteijn et al, 2014). Despite some concerns of delegitimising CFS/ME by the use of psychological interventions McDermott, Lynch and Leydon (2011) reported that people with CFS/ME are open to exploring the interactions of physical, emotional, social and environmental factors in order to understand their illness experience. Similarly participants of Tucker's (2004) study attributed ignorance to those that consider CFS/ME to be only psychological, which could be seen as an effective coping strategy by avoiding any concerns about scepticism, accusations of laziness or inferior treatment.

1.12.2 Adverse effects

Behavioural therapies have been considered preferable as beneficial results have been observed without the harmful side-effects, namely immunological and anti-viral treatments (Chambers et al, 2006). Yet this has been challenged with claims adverse effects following CBT/GET, resulting in a considerable deterioration which may explain the high drop-out rates (Gladwell et al, 2014: Malouff et al, 2008: O'Dowd et al, 2006: Stubhaug et al, 2008: Twisk and Maes, 2009). That said using review data the average drop-out rate for CBT was 19 per cent which related to low mood or heightened anxiety (Whiting et al, 2001). Gladwell et al (2014) highlighted several accounts of CFS/ME patients experiencing increased symptoms during GET treatments and subsequently discontinued them. This has been consistently acknowledged that a flexible and individualised approach is needed for exercise interventions to allow for CFS/ME symptom fluctuation and rest, avoiding extreme symptom increases that may deter patients (Marques et al, 2015: Pinxsterhuis, Strand and Sveen, 2015).

1.12.3 Fidelity of interventions

There is a clinical debate that clarity is needed for exercise and activity related interventions. This is because GET, Graded Activity Therapy (GAT), Adaptive Pacing Therapy (APT), Pacing

and Activity Management (AM) all share similar features. With multiple definitions of these interventions used there is a great deal of overlap of theoretical perspective and the language used interchangeably (Twisk and Maes, 2009: NICE, 2007: NICE, 2014: White and Naish: 2001, White et al, 2011). This understandably causes confusion for patients', HCPs and researchers. Tensions have occurred in clinical practice because over exertion or exercising at maximal levels can be detrimental for people with CFS/ME, often resulting in relapse or severe set-backs (Gladwell et al, 2014: Lapp: 1997, Sheppard: 2001: Twisk and Maes, 2009).

Research by Godfrey et al (2007) suggest that therapy outcomes may show limited differences between interventions but the method and application of treatment may indicate the 'active ingredients'. This debate highlights the importance of understanding the treatment available, and whether other factors need to be considered before recommendations are made; such as age, comorbidities, patient goals, therapists and intervention type. This also maps onto the NICE (2007) guidelines advocating tailored and individual treatment for people with CFS/ME.

1.12.4 Long-term efficacy

There are concerns about the long-term efficacy of behavioural interventions, purely due to the limited data or inclusion of long-term follow in studies (Baldwin, 2013: Chambers et al, 2006). Baldwin (2013) reported in the updated review of behavioural interventions that significant improvements in fatigue were found for CT and GET at long-term follow up (29 per cent) although this was much less in comparison to short-term effects (71 per cent). This differed to mood and general functioning which suggested significant result in the short-term and either remained the same, or improved further at long-term follow up. Conversely Leone et al (2006) conducted a 4 year follow up study for an RCT investigating a GP delivered CBT intervention in the Netherlands and reported the intervention was not effective at follow up. Specifically the study demonstrated significantly higher levels of fatigue, lower functioning and more participants were receiving financial benefits due to inability to work compared to the control group. Potentially these findings could be explained as no significant results were found in the original trial at 4, 8 or 12 month follow up points. Equally the CBT delivery (5-7, 30 minute minimum sessions) is much less intense than other studies (Baldwin, 2013: Chambers et al, 2006: Malouff et al, 2008: Price et al, 2009). In addition it could be a result of the study's broad definition of fatigue as no diagnostic tool for CFS/ME was used, which consequently the study was excluded from the updated review.

1.13 Evolution of Cognitive Therapies

More recently variations of Cognitive Therapies (CT) such as Mindfulness-based Stress Reduction (MBSR: Kabat-Zinn, 1990), Mindfulness-Based Cognitive Therapies (MBCT: Segal

et al, 2002), Acceptance and Commitment Therapy (ACT: Hayes, Strosahl and Wilson, 1999) and Focused Acceptance and Commitment Therapy (FACT: Strosahl, Robinson and Gustavsson, 2012) have appeared in research and practice in health care (Hacker, Stone and MacBeth, 2016). Broadly these approaches aim to increase awareness to the present moment, to observe and accept non-judgementally, to prevent further struggle and discomfort in trying to change or avoid thoughts, feelings, memories or sensations (Bohlmeijer, 2010: Hayes, 2004: Price et al, 2009). These types of intervention move away from deficit models of illness and towards wellbeing, which is particularly relevant in chronic illness (Stanley, 2013) ultimately decreasing disability even though symptoms may be unaffected.

Whilst there is considerable application of these approaches in chronic pain (Dahl et al, 2005: Hayes, Strosahl and Wilson, 1999: McCracken and Eccleston, 2003: Vowles et al, 2014), depression and anxiety (Fledderus et al, 2013: Hacker, Stone and MacBeth, 2016: Hayes, 2004), there is limited published research in CFS/ME. Of the available research in MUSs (such as FMS, CFS/ME and Cancer-related Fatigue) promising results have been reported (Brooks, Rimes and Chalder, 2011: Densham et al, under review: Krzeczowska, Karatzias and Dickson, 2015: Hulbert-Williams et al, 2015: Luciano et al, 2014: van Ravesteijn et al, 2014: Rimes and Wingrove, 2011: Surawy, Roberts and Silver, 2005: Wicksell et al, 2013) suggesting a utility warranting further investigation, particularly given the comparable condition presentation of CFS/ME with other chronic illness populations (Blakely et al, 1991).

1.14 Challenges of living with CFS/ME

1.14.1 Lack of understanding

Lack of understanding has also been present in the general public as well as HCPs (Anderson et al, 2012: Larun and Malterud, 2007: Söderlund, Skoge and Malterud, 2000: Stenhoff et al, 2015). Stigmatisation stems from several elements; one being a historical lack of understanding of the condition where unjustified stereotypes were imposed (Söderlund, Skoge and Malterud, 2000), for instance 'Yuppie Flu' is a documented derogatory term used in the 1980's because those affected were professionals aged 20-40 (Macrae, 2015). The media have also been blamed by misrepresented research and depicting CFS/ME '*in simplistic and stereotypic ways*' (p. 278, Song and Jason, 2005). Further complications arise as the condition does not fit the dominant medical model of illness and there are few visible features of CFS/ME like wounds or swelling. This invisibility can lead others to questioning the existence or extent of the symptoms experienced (Edwards, Thompson and Blair, 2007).

This lack of knowledge about the condition can undoubtedly feed into a sense of helplessness and frustration for those living with the illness (Drachler et al, 2009), which can in turn

exacerbate fatigue and related symptoms. As a result people are reluctant to disclose they have the condition due to perceived stigmatisation and lack of understanding, consequently creating isolation and disconnected with others (McInnis et al, 2015: Horton et al, 2010: Larun and Malterud, 2007: Tucker, 2004: Ware, 1999).

1.14.2 Identity

Expectedly the impact on a person's sense of self is affected and so the recognition that identity is important to acknowledge in CFS/ME is widely supported (Anderson et al, 2012: Larun and Malterud, 2007). This is a result of the vast negative impact of CFS/ME on various aspects of life such as; physical and mental function, work status, mood and relationships, which have also been reported in other chronic and mental health conditions (Arroll and Senior, 2008). Identity can also be affected due to the uncertainty surrounding aetiology; the loss of the person they were, doubt about who they are now and concerns of who they will be/unable to be in the future due to the imposed limitations of the condition (Anderson et al, 2012: Larun and Malterud, 2007: Whitehead, 2006a). This was also reflected in Drachler et al's (2009) review into the self-expressed needs of those with CFS/ME that support is needed to help people make sense of their illness.

The application of occupational perspectives of health can assist our understanding of the disruption experienced during CFS/ME. In particular the four core concepts of the occupation; *doing*, *being*, *becoming* and *belonging* which encompass both the visible, performance based elements as well as the invisible, relationship components. It is the holistic view of practical actions or performance of tasks '*doing*', the reasons for these actions, linked to who we are as a person or how we feel about what we do '*being*', the process of development or change of these over time '*becoming*' and the purpose or values that gives life meaning which also includes a social connection '*belonging*' (Hitch, Pépin and Stagnitti, 2014: Lyons et al, 2002).

1.15 Illness trajectory

Similarly to aetiology, there is no consensus regarding the prognosis of CFS/ME (AfME, 2014: Asbring, 2001: Jason et al, 2012). Furthermore the condition is reported to fluctuate in symptom severity and frequency where periods of remission and relapse can be experienced as feature of the condition (NICE, 2007). Whilst the condition name 'Chronic' indicates a significant period of time, there isn't much clarity on what this can mean for patients. This uncertainty is a significant source of concern about the future for people, particularly earlier phases of the CFS/ME journey (Anderson et al, 2012: Drachler et al, 2009: Haig-Ferguson, 2014: Larun and Malterud, 2009: Whitehead, 2006b). Also, these concerns regarding illness

trajectory are not limited to the individual with CFS/ME but their families as well (Haig-Ferguson, 2014; Drachler et al, 2009). This is also supported by Leone et al (2006) that demonstrated less-advanced fatigued participants had poorer longer-term prognosis, which illustrates the need for appropriate support for longer-term management.

One view point expressed by Ax, Gregg and Jones (2001) that goes against many conceptualisations of CFS/ME is that it is not a lifelong condition, as the only deaths associated with CFS/ME are through suicide. Yet many studies have reported CFS/ME symptomology is persistent over time, reporting this through illness duration demographics or longitudinal methodology (Bell, Jordan and Robinson, 2001; Brown et al, 2012; Cairns and Hotopf, 2005; Hickie et al, 1999; Horton et al, 2010; Jason et al, 2011; McKenzie et al, 1995; Wilson et al, 1994). The latter, more prevalent thinking that CFS/ME can remain present over time also links with the proposal that the diagnosis of CFS/ME '*may only be the first chapter of a much longer chronical*' (p.456 Arroll and Senior, 2008). Concluding that CFS/ME may not result in death but it can impact a person's life indefinitely.

NICE (2007) state '*there are difference stages in the natural course of CFS/ME: acute illness, maintenance or stabilisation, and recovery.*' (p.14) but no timeframes or further details are offered for these stages. Likewise Whitehead (2006b) investigated experiences of identity in CFS/ME using a longitudinal qualitative study and grouped CFS/ME experiences into three themes; acute phase, medium term and longer term. As with the NICE (2007) stages they were not time-bound but marked by experiences and coping strategies. Whilst there is a common sense of caution and difficulty in predicting CFS/ME prognosis (AfME, 2014; Brown et al, 2012), it is also worth noting there can be a danger of having timeframes of illness trajectories, as with diagnoses themselves. In some cases they can be unhelpful due to the inflexibility and one size fits all approach. With the available research proposing different periods of the condition exist there appears to be commonality of variation in experiences within the condition presentation (ie severity, frequency, physical, cognitive and emotional) and different coping responses. The prevalence of individual and flexible approaches as effective ways to manage these factors are reflected in the research and best practice guidelines (Drachler et al, 2009; McKenzie et al, 1995; NICE, 2007; Whitehead, 2006b; Wilson et al, 1994).

1.15.1 Recovery

Estimated recovery rates in CFS/ME provide vast ranges from between 8 and 63 per cent of people showing improvement and very low incidences of full recovery of between 5 and 10 per cent (Brown et al, 2012; Cairns and Hotopf, 2005; NICE, 2007). Similarly, Wilson et al (1994) reported only 6 patients of their longitudinal study of outcomes in CFS/ME reported no

current symptoms, equating to 9 per cent of their overall sample. Bell, Jordan and Robinson, 2001 undertook a 13 year follow up study for children and adolescents with CFS/ME and reported whilst 80 per cent expressed improvements, within that group mild to moderate symptoms continued to be experienced. The remaining 20 per cent of participants reported experiencing significant symptoms that greatly impacting their activity.

1.15.2 Difficulties with recovery rates

It is acknowledged when considering recovery in chronic illness it's not clear cut (Pope et al, 2013) and within CFS/ME *'recovery is more than just a decrease or disappearance in symptoms'* (van Damme et al, 2006). Wilson et al (1994) argue that people who have recovered will not be engaging with specialist services or support organisations so accurate information using their records or study recruitment methods may underestimate those who have actually recovered. Difficulties in reporting recovery rates can also come from the variable meanings of the term 'recovery'. For example no longer meeting the diagnostic criteria would suggest recovery (Flo and Chalder, 2014) yet some studies have reported whilst diagnostic criteria is not fulfilled at follow up, participants remain functionally impaired unable to achieve premorbid levels (Bell, Jordan and Robinson, 2001: Brown et al, 2012). Some have therefore proposed a broader view of recovery that considers levels of fatigue and function as well as patient perception of what recovery means for them (Adamowicz, Caikauskaite and Friedberg 2014). However, this method has been criticised calling for objective measures only to be used; before, during and after interventions (Twisk, 2014). This is a really interesting debate as with many objective measures, they have psychometric flaws and limitations for real-life situations (Flo and Chalder, 2014). Equally unless premorbid levels were assessed prior to developing CFS/ME itself, rather than intervention it would be difficult to know what objective premorbid outcomes were met or not following other assessments for that individual.

1.15.3 Definitions of long-term

When drawing on information outside healthcare definitions, timeframes of what equates to 'long-term' are much shorter than the 4-6 months proposed for a CFS/ME diagnosis. For example UK government financial systems (such as benefits) adopt a 4 week or more period to signify long-term sickness (Gov.uk, 2014). Similarly turning to another health condition Complex Regional Pain Syndrome (CRPS) the term 'chronic' is applied when CRPS has been present for over 6-8 weeks (Bailey and Nelson, 2014). Concluding other systems and conditions, 'long-term' is applied when an illness episode turns from weeks into months. With some sources of CFS/ME literature report that symptom experiences occur for just over a year (AfME, 2014: Ax, Gregg and Jones, 2001) increasing up to 59 years in some cases (Arroll and Senior, 2008: Ax, Gregg and Jones, 2001: Deary and Chalder, 2010: McKenzie et al, 1995)

the idea of 'long-term' equating from weeks to months, doesn't seem to reflect the considerable months and years people with CFS/ME experience debilitating symptoms.

Friedberg et al (2000) labelled CFS/ME in the short-term as 3 years and Wilson et al (1994) similarly stating 3.2 years. Yet these figures are slightly higher than other researchers defining 2 years or less symptom onset as a short illness duration (Brown, Brown and Jason, 2010: Nisenbaum et al, 2003: van der Werf et al, 2002). Similarly Friedberg et al (2000) reported long illness duration as 18 years, which again is higher than 10 years McKenzie et al (1995) used in their study. Unfortunately no rationale was provided by McKenzie et al (1995) for their inclusion criteria of 10 years and so it is unknown what significance this bracket has. Although interestingly McKenzie et al (1995) found no significant differences in coping methods adopted by people with CFS/ME for less than 5 year and people with CFS/ME for 10 years.

1.15.4 'Long-term' focused research

Long-term follow up (defined as 12 months or more) is rare and there is considerable variation of follow up periods between CFS/ME studies (Baldwin, 2013: Chambers et al, 2006: Houlton et al, 2015: Whiting et al, 2001). Of the available Randomised Controlled Trials (RCTs) that assessed participants over 12 months, long-term follow up was reported at 1.4 (Wearden et al, 2010), 3.2 (Wilson et al, 1994) and 3.8 (Leone et al, 2006) years. It seems counterintuitive that studies investigating interventions for CFS/ME are only reporting shorter-term outcomes given the longitudinal nature of the condition and the cost of the condition on an individual and societal level.

Similarly there is very limited research specifically addressing long-term experiences, despite illness duration commonly reported as part of demographic variables. This is particularly interesting given the proposal that symptoms and coping can change over time (Ax, Gregg and Jones, 2001: Brown et al, 2012: Jason et al, 2000: Reynolds, Brown and Jason, 2009: Whitehead, 2006a: Whitehead, 2006b). To date few studies have been published where research questions draw on long-term experiences. Of the available research it seems outcomes and coping over time have been investigated quantitatively (Bell, Jordan and Robinson, 2001: Brown et al, 2012: McKenzie et al, 1995: Wilson et al, 1994), with general CFS/ME experiences, identity reconstruction and mental health and recovery explored qualitatively (Arroll and Senior, 2008: McCue, 2004: Whitehead, 2006b).

Clements et al (1997) also stated '*Most patients (76%; 50 of 66) reported they could control their symptoms to some degree but distinguished between short-term relief of symptoms and the longer-term course of the illness*' (p 619). Therefore it is reasonable to suggest along with the complex and longitudinal nature of CFS/ME different interventions, coping strategies and

support might also be needed at different points of the illness experience. This has been illustrated by McKenzie et al (1995) exploring long-term coping in CFS/ME participants by concluded that *'coping in CFS may lessen the emotional distress of patients; however, coping may not influence the course of the illness'* (p. 67).

1.16 Project Rationale

Synthesising the findings from the available studies suggests a longevity of fatigue and related symptoms, resulting in impairment over time for people with CFS/ME. Factors that appear to be influential in successful management relate to the individual's illness beliefs, coping strategies and support received from others (Leone et al, 2006: McKenzie et al, 1995: Wilson et al, 1995: Whitehead, 2006b). However, more research is needed to explore longer term experiences of CFS/ME. Particularly in relation to the illness pattern over time and effectively ways to respond to symptom experienced and the significant impact on a person's life for those who are unable to 'recover' or reduce symptoms comparable to premorbid levels.

The argument presented by this thesis is suggesting more knowledge is needed to provide clarity of timeframes or phases of CFS/ME, if they exist supporting or rejecting current models of CFS/ME illness trajectory. Considering whether different support and treatment is needed depending on illness duration, given the number of years the illness can dominate a person's life. This theory is supported by research into identity reconstruction following illness and injury indicating time is a factor in adapting to illness (Arroll and Howard, 2013: Dickson, Knussen and Flowers, 2008: Pinxsterhuis, Strand and Sveen, 2015: Whitehead, 2006b: Yoshida, 1993). Equally this thesis will draw on therapeutic approaches and theoretical conceptualisations of CFS/ME treatment and management, specifically thinking about therapeutic mediators and mechanisms of change (Castell, Kazantis and Moss-Morris, 2011).

As highlighted, the research question aims to generate more knowledge about how people with CFS/ME manage their condition drawing on comparable therapy approaches if relevant and whether coping methods vary dependant on other factors, such as illness duration. Specifically this study will comment on experiences and attributions of management which also links to the suggestion by Anderson et al (2012) that more research is needed regarding self-perceptions of those with CFS/ME using qualitative research methods.

2 Methodology

2.1 Research Planning

2.1.1 Patient and public involvement

Input was sought to inform the research planning (recruitment and interviews) and dissemination of the study findings. Three patient research advisors (referred to as Advisors hereon in) accepted this role; two female and one male. The rationale to have more than one Advisor was to distribute research input preventing over-burdening one Advisor, particularly important if the Advisors are still actively managing their fatigue related symptoms. It is of note that one of the Advisors considered they were recovered from CFS/ME whilst the other two were actively self-managing their fatigue but were not involved in any ongoing treatment with the researcher.

2.1.2 Design

Effectiveness of interventions is based largely on quantitative outcome measures, but it is also important to provide some context to individualise people's experiences living with CFS/ME. As noted by Jason et al (2000) the subjective experience of people living with CFS/ME is really important as contradictory results can be found from standardised outcome measures depending on when they are completed. This was echoed by Gotts et al (2015) stating the '*complex experience of living with it [CFS/ME] is often lost*' (p. 2-3) and so their research utilised qualitative methods to gather detailed information about the experiences of sleep.

Quantitative patient reported outcomes can only comment on *what* has changed for patients', in terms of fatigue, depression, anxiety, function/ disability, and valued action etc. Whilst these are important to evaluate the effectiveness of the treatment, using this data (certainly alone) fails to answer questions about *what* specifically is helpful, *when*, and *why* (Godfrey et al, 2007). These unanswered questions also link to the findings and gaps in knowledge identified by the qualitative reviews (Anderson et al, 2012; Larun and Malterud, 2007) and SRs (Baldwin, 2013; Chambers et al, 2006). Investigating long-term management of CFS/ME and addressing individual needs, also fits with NHS outcome frameworks for effective management of LTCs (DoH, 2013a).

Given the research topic is concerned with *what* specific aspects of strategies, *when* and *why* changes have occurred for individuals, a qualitative approach was deemed be best suited to answer these types of questions. A qualitative research design was adopted to understand and interpret rich meanings (Braun and Clarke, 2013; Clements et al, 1997) of long-term experiences of managing CFS/ME. As stated by Anderson et al (2014) '*using qualitative*

methods to study contested illness provides us with a deep understanding of the interaction of multiple systems involved in chronic illness' (p.22). A semi-structured interviewing approach was chosen to square up a) the research question, b) the Advisor's ideas of what is valuable to know, c) existing theoretical knowledge and d) providing a space for participants to express their views and experiences not captured by the interview questions. The flexibility of the semi-structured interview format also allowed for rapport building, and was personally preferred by the researcher, because it is more akin to the her clinical experience, skills and values.

2.1.3 Ethical approval

Ethical approval was sought using the NHS Health Research Authority online ethics system (Integrated Research Application System) in July 2014 allocated to the next available committee for proportionate review. Following clarification NRES Committee Yorkshire & The Humber – Bradford Leeds approved the study on 11th August 2014. Local NHS ethics approval was sought from the Royal National Hospital for Rheumatic Diseases (RNHRD) Research and Development Ethical committee and was approved in September 2014 subject to minor wording changes on participant documentation. The University of the West of England's faculty of Health and Applied Sciences Research Ethics Committee provided study approval in November 2014 (See Appendix B).

2.2 Rationale for analysis

Various research methods have been used in CFS/ME qualitative research, such as grounded theory and Thematic Analysis (TA) to explore GP and HCPs perceptions (See Raine et al, 2004: Horton et al, 2010) and Discourse Analysis (DA) for the construction of legitimising CFS/ME avoiding stigmatisation (See Tucker, 2004). Interestingly most papers adopt an Interpretative Phenomenological Analysis (IPA) and interpretive theoretical perspective. These studies have focused on general experiences of living with CFS/ME, often relating to symptoms and diagnosis journey. Perhaps an IPA approach has been favoured because it's particularly suited to exploring lived experiences, gathering rich personal understandings of individuals' meanings. The research question suited a variety of qualitative research methods and analysis however, TA was deemed most appropriate. Below outlines the rationale for this decision.

2.2.1 IPA Vs. TA

Although personal experiences and meanings were being explored the focus of the research was not idiographic. The aim was to create a more generalised view of long-term experiences of CFS/ME therefore a pattern-based analysis of TA allowed for this, to create transferability of the study findings to similar groups. Similarly only one interview was appropriate to capture long-term management experience up to that moment in time, it was not expected to look at changes over time between several interviews. Therefore conducting multiple interviews per participant to create a biographical image of the participant's experience (which would suit an IPA approach) was not necessary, nor the focus of the research question. In addition TA allowed for theoretical flexibility, accommodating the contextualist perspective adopted (further detail to follow regarding epistemology).

2.2.2 DA Vs TA

The researcher accepts the need for language as a means of communication to convey the participant's reality and takes this more on face value. The use of narratives as a communication function suggests that through language the participants' cognitions can be accessed, which contradicts the assumptions of DA in that '*language creates meaning and reality, rather than reflecting it*' (pg. 330 Braun and Clarke, 2013).

2.2.3 Selection of TA

Given the points outlined TA was deemed most appropriate for the current study. (Clements et al, 1997: Hareide, Finset and Wyller, 2011: Horton et al, 2010) The 6-phase TA outlined by Braun and Clarke (2006) was used to analyse the data (1. Familiarisation with the data 2. Generating initial codes 3. Searching for themes 4. Reviewing themes 5. Defining and naming themes 6. Producing the report). A deductive TA was used to analyse the data by the researcher only, removing any issues regarding discrepancies and differences of opinion when analysis is performed by multiple researchers. Some steps were taken to mitigate the bias of one researcher analysing the data resulting in initial themes being discussed with the Director of Studies (DoS) prior to refinement. Although this procedure does invite subjective criticism about the findings produced, arguably this will exist despite the number of researchers involved in the analysis or attempts made to control for sources of bias (Braun and Clarke, 2013). Similarly as Tucker (2004) wrote '*It is not possible to stand back and objectively report on the different versions of CFS in the world, without creating a particular version in the process*' (p. 162) the aim of the research is to capture an interpretation of an interpretation of long-term management of CFS/ME. Fully accepting the process and

conclusions will be different for all parties (participants, researcher and reader) at different points in time.

2.2.4 The Researcher's position

This qualitative project was underpinned by a critical realist ontology and contextualism epistemological perspective. The researcher rejected the realist position that there is one truth to be discovered. Instead believing human experience emerges from subjectivity and contextual factors, and that knowledge of these can be accessed either partially or completely through language (Madill, Jordan and Shirley, 2000). The use of reflexivity within contextualism also sits well with the researchers understanding and professional practice. Although reflexivity is employed in most qualitative research methods generally as well as more specific schools of thought, such as hermeneutics. Within a contextualist conceptual framework the dual interpretative process can be acknowledged, whereby the researcher is interpreting the participants' interpretations (Braun and Clarke, 2013: Madill, Jordan and Shirley, 2000).

The deductive element relates to the conceptualisation of treatment from a cognitive behavioural model, although CFS/ME is not a psychological problem. This highlights the researcher's assumption that CFS/ME as a LTC and that in the most cases will require continual monitoring and management by the individual. Adjustment to the limitations of the condition is complex and as research suggests this occurs over time (Clarke and James, 2003: Dickson, Knussen and Flowers, 2008: Gray and Rutter, 2007: McKenzie et al, 1995 Whitehead, 2006b). The condition also has a huge impact on mood and social interactions (Lombaard and Mouton, 2005). The researcher rejects the idea CFS/ME is a psychosomatic or mental health condition, but acknowledges the psychological impact of this invisible and contested condition (Clarke and James, 2003: McKenzie et al, 1995: Tucker, 2004). To an extent there is also a rejection for the need for a biomedical model to 'prove' the existence by chemical or biological markers. It is the researcher's belief that the experience is real regardless of label or medical tests and so in a time when medicine cannot provide a cure, the way in which symptoms are responded to remains a realistic treatment approach.

Building on the previous point it is important to acknowledge the researcher has a clinical role as an Assistant Psychologist and Macmillan Professional at the BCFS using various psychological theory to inform practice. Her main models for interventions employ; health behaviour change models, motivational interviewing, relaxation within a biological stress model, goal setting, CBT, mindfulness and ACT. These approaches are used by other clinical members of the inter-disciplinary team at the BCFS and an awareness of her clinical thinking was reflected upon throughout the research process.

It was considered what impact a 28 year old, female, white researcher might have on the research process and data produced, specifically when CFS/ME reports higher incidence in females. This factor could have been advantageous when interviewing female participants as a shared sense of being a woman may have created an implicit open non-judgemental environment. On the other hand when interviewing men the opposite may have occurred, that the men may have thought the researcher couldn't understand about their experiences as a man with CFS/ME, potentially inhibiting what they said, and as a result limiting the data collected. The Advisors did not indicate any issues with the researcher conducting the interviews, although it is of course possible they may have not wanted to voice any doubts directly to the researcher. An awareness of these potential influences was held by the researcher so attempts were made to voice the researcher's stand point; promoting a non-judgemental inquisitive view. It was stated that the participant was the expert and the purpose of the researcher was to listen to their experiences. In addition the researcher commented upon any instances of this within personal reflections throughout the research process in a reflective journal. Interestingly one male participant in his interview did talk about perceived sexism during a CFS/ME group treatment programme at a specialist CFS/ME service but described for him, having a male or female doctor is irrelevant and it is the person's profession that's important and not gender. The participant spoke about the ability to relate to what the professional is saying rather than the physical characteristics of the professional.

An etic perspective (Lapan, Quartaroli and Riemer, 2012) was also adopted for this project, as the researcher was an outsider to CFS/ME community with no personal experience of living with CFS/ME. The impact of this was also considered, however given the individual variation of CFS/ME (Anderson et al 2012: Friedberg and Jason, 1998: Jason et al, 1999: Larun and Malterud, 2007: NICE, 2007: Pinxsterhuis, Strand and Sveen, 2015: Söderlund, Skoge and Malterud, 2000) it was thought to be beneficial to the research that the researcher did not have personal experience of CFS/ME. None of the participants asked whether the researcher had or has had CFS/ME and so this was not disclosed during the interview, suggesting it did not have an impact on the data collected.

2.2.5 Reflective journal

As mentioned a reflective journal was kept during the research process capturing personal reflections and thoughts about the research. Extracts of the journal can be found in Appendix D iv; however two critical incidences were captured using this journal worth noting. The first was an automatic initial analytical view during the transcription process where coding and analytical thoughts were occurring very early on in process and seemed to be with ease and flowing freely. Second was a powerful sense of being overwhelmed and confused by the data

during the refinement of the themes, when there was a real struggle to see the meaning and purpose of the data. An assumption the researcher was being confused by her clinical experiences and unable to identify what codes were related and what were distinct from each other. This was also attributed to experiencing stress and perhaps having specific research expectations, which were abstract and created a barrier in achieving an undefined outcome.

2.3 Study specifics

2.3.1 Research setting

The research was undertaken alongside a specialist NHS outpatient service offering interdisciplinary treatment for adults with chronic fatigue in the South West of England. The service operates under the 2007 NICE guidelines for the treatment and management of CFS/ME and provides tailored holistic treatment using CBT principles and activity management. The service was established in 2005 and the current team is made up of occupational therapy, physiotherapy and psychology professionals, with both individual and group treatment options available. The service provided recruitment opportunities for the current study and a location for the interviews to take place.

Prior to and during the data collection phase of the study the service was known as The Adult Fatigue Management Service (AFMS) at the RNHRD Foundation Trust. During the write up phase of the project the Trust was acquired by another hospital. Therefore the Service is now known as the Bath Centre for Fatigue Services (BCFS) at the RNHRD site, part of the Royal United Hospitals (RUH) Bath. This change can be seen in the study documentation.

2.3.2 Recruitment

Participants were recruited using the ethically approved AFMS Clinical and Research database (Ethics registration number 08/H0101/58) based on the inclusion criteria. Recruitment took place between October and November 2014. Purposeful sampling was used in an attempt to achieve a varied sample, with the demographics used to perform this purposeful sampling as; age, ethnicity, marital status, length of time experiencing fatigue related symptoms and time since diagnosis.

2.3.3 Inclusion Criteria

Inclusion criteria required participants to be 18 years of age or over, had experienced of fatigue related symptoms for 5 or more years (symptom onset) and provided informed consent to take part.

Symptom onset was deemed the most appropriate criterion compared to diagnosis because the person begins to manage their symptoms as soon as they occur, not when they receive a diagnosis. Another complicating factor of using diagnosis as criterion is the variability experienced between symptom onset and receiving a diagnosis (Anderson et al, 2012). This approach of using patient reported symptom experience has also been used in other research papers (Hareide, Finset and Bruun Wyller, 2011; Whitehead, 2006b) where the important factor is whether the individual believes they have CFS/ME when investigating their lived experience, rather than having a medical diagnosis to legitimise the illness. Although this decision may invite potential criticism, a diagnostic measure of the illness was implicitly used because all the patients that are registered on the AFMS database have obtained a diagnosis from their GP (CDC/ Fukuda criteria). This potentially satisfies both arguments by emphasising patient perceptions are important but also using a recognised diagnostic tool to be able to differentiate the CFS/ME diagnosis from chronic fatigue more generally.

In the absence of clear guidance or reasonable suggestions for what long-term management might relate to for adults with CFS/ME, this study has provided its own definition. Long-term management is defined as 5 years or longer (≥ 60 months) based on the calculated mean illness duration observed in an updated systematic review (Baldwin, 2013). It also takes into account a relevant long-term study by McKenzie and colleagues (1995) that used an illness duration of between 1 and 10 years and reported no difference in coping between people with a 5 years illness duration and 10 years. In addition to the related thinking that mental health co-morbidities are not regarded as exclusionary for a CFS/ME diagnosis after 5 years (Anderson et al, 2014). Comment may be provided following the project on the appropriateness of this selected timeframe, ideally with the views of the participants reflected in this conclusion.

2.3.4 Exclusion Criteria

As suggested by the UWE Research Degrees Committee (RD1 project registration approval Aug 2013) and adopted in other applied research contexts (Pemberton and Cox, 2014), participants were excluded if they had previously received treatment from the CI in her clinical role at the specialist fatigue Service, to avoid influencing participants. This would not exclude those currently patients of the AFMS unless the treatment was provided by the Researcher.

Two further exclusions were adopted relating to practical aspects of taking part: If participants were unable to attend the hospital for the interview (approximately 45-60 minutes) in Nov 2014 - Jan 2015 and if they were unable to speak and understand English as informed consent would not have been achieved.

2.3.5 Procedure

Following NHS ethical approvals (National Research Ethics Service and the local RNHRD R&D committee), recruitment was sought using the AFMS Clinical and Research Database to extract details of potential participants. Patients were extracted from the database if they have experienced fatigue related symptoms for 5 or more years. After potential participants were identified, a SPINE trace was conducted to obtain current address and GP details. Using this search it was also checked whether the person was alive in order to avoid distressing family members by sending out information to their deceased relative.

The potential participants were sent by post a study pack containing; an invitation letter, Participant Information Sheet (PIS), consent form (for reference only), a response form and a pre-paid self-addressed envelope. They were also given the CI and DoS contact details should they want to discuss anything further. The response form was intended to capture either a) interest in taking part and their preferred method of contact or b) to decline participation and a brief reason for decline to help future researchers understand why this might be.

Preferred communication was requested, was because often people with CFS/ME find telephone conversations very difficult due to common cognitive difficulties of the condition. To remove this as a potential barrier for people to take part in research, email, letter and Skype communication were offered prior to the interview to answer questions and arrange the interview date and time. Participants were also made aware in the research invitation letter, that although the CI was employed by the AFMS, the research project is being undertaken as part of a Doctoral Programme and the CI was not acting as clinician for the project. The distinction was important to avoid influencing the participants' views of the treatment received from the AFMS and to make it clear the project was for research and not a service evaluation. The participants were also made aware that participation or declining to participate would not affect the treatment they receive from the FT in the future.

When the CI had received a participant's response that they were interested in taking part, the CI contacted the participant to answer any additional questions through their preferred method of contact. All but one participant requested to be contacted by phone. If all was satisfactory at this point, the interview date and time was arranged with the participant. Meeting at a convenient time for the participant was essential to ensure attending the interview is as accessible as possible (Arroll and Senior, 2008; Houlton et al, 2015) and can be fitted into their other demands. This was particularly important if they were still actively managing fatigue related symptoms, and need to travel to the hospital as well as taking part in the interview.

A brief outline of the interview topics and questions were given to the participant prior to the interview. This was to compensate for any common cognitive difficulties experienced with CFS/ME (poor concentration, memory and word finding difficulties) (Fukuda et al 1994). This step is also an attempt to reduce potential anxiety about the interview procedure and was supported by the Advisors.

Understanding the fluctuating nature of the condition, cancellation of the interviews was made available up to any point, as suggested by Arroll and Senior (2008) noting particular concern by participants that they would be unwell on the day. One participant cancelled on the morning of the interview due to a virus and rescheduled the interview for two weeks later and another participant cancelled a day before as was unwell related to their CFS/ME, rearranging the interview a week later.

The semi-structured interviews all took place at the RNHRD in an AFMS clinic room. Despite being a clinical room there was no medical equipment in the room. The room contained an office desk, small table, computer, several chairs and a hand basin. It was reassuring that the Advisors didn't believe having the interviews at the Hospital would be a detrimental or inhibiting environment, in fact one Advisor thought it would encourage more openness in an environment usually used to share personal details. Hot and cold drinks and biscuits were provided throughout the interview. There was an attempt to create a more relaxed space, by setting up the chairs and table to a friendly set up rather than a medical consultation format where the desk can act as a barrier between the two parties.

The researcher checked the recording equipment before the interview started and also ensured the participant understood the project requirements, answering any questions to obtain informed consent. Pseudo names were chosen by the participant and the participant was referred to this false name for the entirety of the recorded interview for anonymity reasons.

The interview audio was transcribed verbatim by the researcher as soon as possible after the interview as suggested by Braun and Clarke (2013) to. The transcription process was aided by Olympus DSS Player and typed onto a Microsoft Word document. Any names of family members, healthcare professionals or place names were anonymised with the general content captured. For example if BCFS or another Service was mentioned [Specialist NHS Fatigue Service] was indicated on the transcript, similarly [WIFE] or [HUSBAND] was typed when a participant was referring to their spouse.

Although conversational analysis was not used the researcher included any mistake words phrases to remain as true to the data as possible. It was considered particularly relevant to the study population as word finding difficulties and poor memory can be common symptoms therefore reporting on this may have been useful, so therefore was included. Several participants lost their train of thought or checked what the researcher's question was after talking for some time with only one participant, Diane frequently displayed these mistake words and phrases. Interestingly Diane had come with a prepared typed word document to the interview and as expected more mistakes were made when talking with the researcher unaided by the paper notes.

TA was performed across the dataset by the CI only. The 6-phase TA outlined by Braun and Clarke (2006) was used to analyse the data (1. Familiarisation with the data 2. Generating initial codes 3. Searching for themes 4. Reviewing themes 5. Defining and naming themes 6. Producing the report). As suggested a thematic map was also used to review and refine themes, to achieve an organised description and using theory that go beyond the data items collected creating a coherent story of peoples' experiences. The researcher attempted to systematically and thoroughly review the data with flexibility to construct a coherent pattern-based analysis at a latent level (Braun and Clarke, 2013).

2.4 Ethical considerations

2.4.1 Data storage

All *physical*, Patient Identifiable Data (PID) (such as consent forms) was kept in the AFMS office at the RNHRD in a locked filing cabinet, with only authorised access. All *electronic* PID (such as interview audio) was saved in the AFMS research folder, within the RNHRD central electronic storage space (Data1) with only authorised access.

All *physical or electronic* anonymous data (such as interview transcripts, analysis and written reports) was handled with care by the CI. Physical documents were locked away in the AFMS office or personal office at the researcher's home. Electronic documents were only accessed on password protected computers (RNHRD computers, UWE computers and CI personal laptop) by authorised members of the team.

2.4.2 Informed consent

Multiple opportunities were provided for the potential participants to ask questions about the research prior to the interview. Informed consent was taken in writing by the CI at the start of the interview with a copy provided for the participant. (See Appendix C vii)

2.4.3 Emotional distress

There was a moderate risk that participants would become emotionally distressed when talking about the condition and how it has impacted their lives. The CI was experienced in managing such sensitive issues from her clinical role and a procedure was in place to follow, however this was not required as no participants became distressed during the interview.

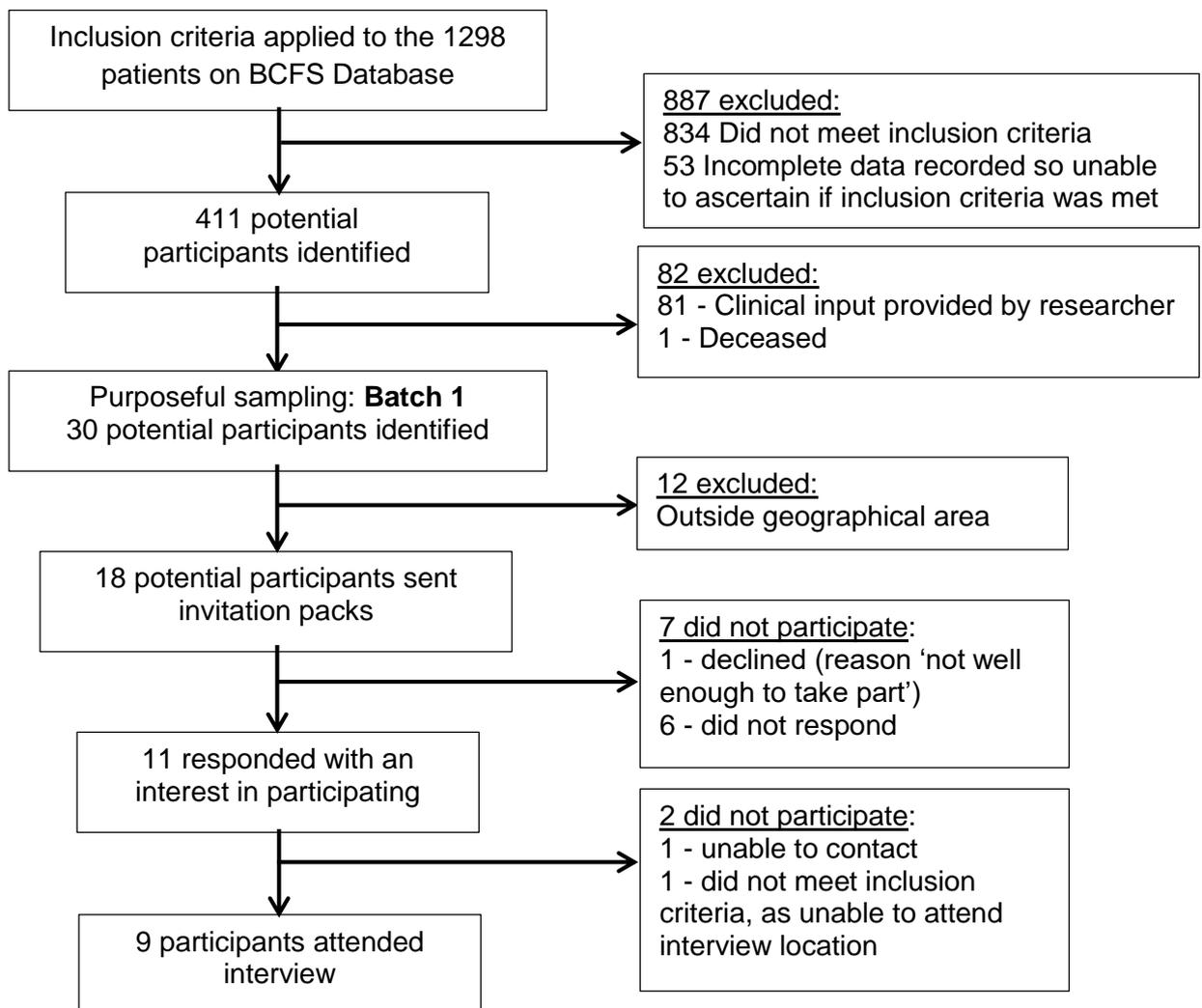
3 Results

3.1 Participants

3.1.1 Response rate

After applying the inclusion/exclusion criteria to the BCFS database, a batch of 18 potential participants were identified and sent study invitation packs by post. Of these, 11 responded and 9 participants took part in the study. Single interviews lasting on average for 48 minutes were carried out at the RNHRD site during October 2014 and February 2015.

Figure 1: Recruitment flow chart



3.1.2 Demographics

Of the 9 participants; there was a 2:1 female to male ratio with all participants reporting their ethnicity as White British, the majority of participants were married (44%), a median age of 55 years (range 23 - 64), median illness duration of 11.3 years (range 9 – 17.5) and median time between illness onset and diagnosis was 2 years (range 0 – 10). Three general groups of participants existed; those diagnosed as children (n=3), men (n=3) and those in employment (n= 6).

Table 1: Demographic information

Pseudo name	Gender	Marital Status	Age*	Illness onset*	Time between illness onset and diagnosis*
Colin	Male	Married	61	11.1	4.25
Stuart ^a	Male	Divorced	62	11.3	1
Jane	Female	Married	31	17	0
Kate	Female	Single	28	17.3	0.5
Maria	Female	Co-habiting	57	13	4.8
Alison ^a	Female	Married	64	17.5	10
Oliver	Male	Married	55	11.25	4.5
Diane ^a	Female	Divorced	47	9	2
Penny	Female	Single	23	10.5	0.2

* presented in years

^a Pseudo name chosen by participants were personally associated names and so were changed by researcher to protect confidentiality

3.2 Themes

Following Braun and Clarke's (2006) six phases of TA, 3 main themes and 2 sub-themes were 'identified'. When organised into categories, some data code was relevant to more than one theme and so was included for the relevant themes. This was a similar approach taken by Anderson et al (2014) who described code as '*not necessarily mutually exclusive [...] Thus one code may also contribute to other codes and corresponding themes.*' (p.8).

Figure 2: Thematic Map

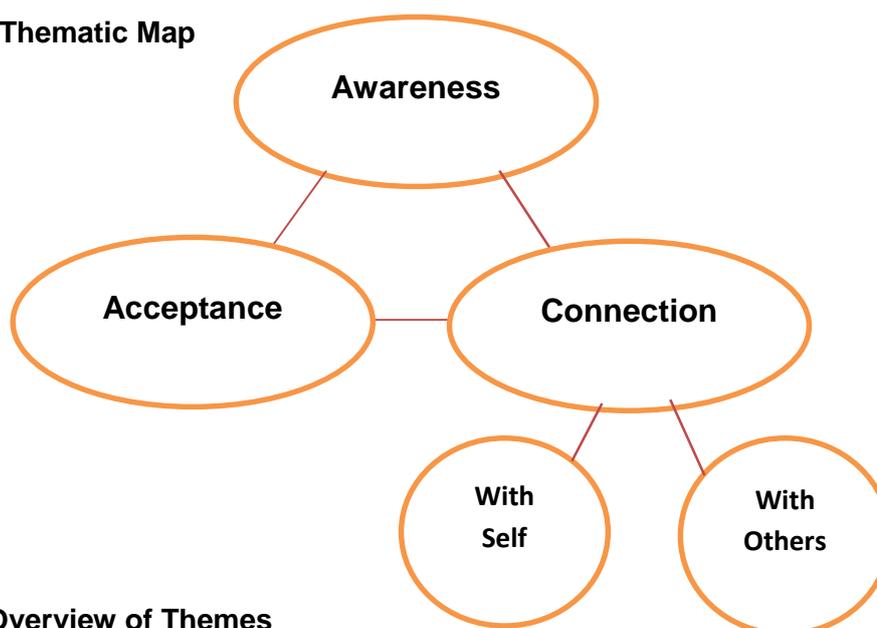


Table 2: Overview of Themes

Theme	Synopsis	Related Code
1. Awareness	Awareness as the foundation for managing CFS/ME. Participants developed a deeper awareness and understanding about their experience. Listening to their body and mind, not ignoring experiences. Including the awareness of others as an asset and a challenge.	Listening to body/mind, Symptoms, Urges/ desires, Pushing beyond limits, Quality of sleep/rest, Impact of routines, Identification of warning signs/triggers, Monitoring, Awareness of others, Invisible illness, Validating, Stress, Urges, Environment, Pacing, Past experiences, Choice, Control
2. Acceptance	Accepting the diagnosis and that CFS/ME as a life-long illness, not fighting the condition or condition symptoms. Subsequently reducing the impact of the illness by acknowledging it and adapting around limitations.	Accept, Fighting, Denial, Openness, Closed, Rejecting, Challenges, Balance, Internal battles, Perspective, Adjustment, Flexible, Validation, Understanding, Individuality, life challenges, Employment Comorbidities, Life-long illness, Scepticism of others' recovery, Pacing, Fluctuations and uncertainty. Past experiences, Being different, Normality, Medication
3. Connection	Connecting with what's important, including having roles and responsibilities. Adapting and changes to roles and responsibilities to still connect with what's important. In addition to conceptualising gradual and consistent progress as the goal.	Values, Roles and Responsibilities, Gradual and consistent, Balance, Adapting, Making changes, Employment, Sense of self, Choice, Pets, Compromise, Talking and Sharing, Identity, Achievements, Goals
3i With Self	Connection and disconnect with the sense of self, addressing psychological impact and the ability to weigh up costs and gains.	Balance, Costs and gains, Sense of self, Humour, Loss, Abstract help, Emotional impact, Confidence, Making changes, Compassion, Compromise, Control, Identity,
3ii With Others	Connecting with others for concrete or practical support. Attempts to communicate effectively with others, using metaphor and analogies.	Practical help, unhelpful people, GP, HCP, family, friends, Metaphors, Analogies, Sayings, Disclosing, Hiding, Pets, Normality, Social interaction, Talking & Sharing

3.3 Theme: Awareness ‘to listen to your body and your mind and, not denying that’ (Diane)

Participant narratives described various instances of being aware, making up an overarching theme of awareness. This theme related to participants’ understanding their experiences in much greater depth, which was beneficial for long-term management.

‘being much more aware of, my body and if it’s telling me something I have to listen’ (Kate)

When participants had low awareness, or actively ignored their experiences they conceptualised this as poor long-term management. Interestingly judgements of poor long-term management were not just limited to the participants themselves, and were expressed towards others with CFS/ME if they stayed in bed or continually pushed themselves beyond their limits. This response was described as ‘giving into’ rather than managing the illness.

Heightened awareness was talked about in many different ways. One instance was in relation to rest and sleep, with participants describing an ability to differentiate the quality of sleep and rest.

‘And also when I went to [HOSPITAL NAME] I was taught and learnt that what I thought was rest isn’t necessarily rest (laughs) [...] so you know watching EastEnders apparently that’s too stimulating (laughs) and actually it is, erm and I do just sit and listen to music and stuff a lot more now like if it’s just peaceful umm, yeah things I, put as, rest were actually low level activity so I just never stopped and now I do try’ (Kate)

All participants mentioned the experience and role of sleep. Several participants described a distinct pattern in their initial illness onset, stating they slept more than they do now. None of these participants offered any theories as to whether this was due to features of CFS/ME but explicitly mentioned this was an initial coping method for them. In simplistic terms, the awareness of their symptoms (pain, tiredness, difficulty concentrating and sleeping) suggested a reasonable conclusion was to sleep more. It was only later when this strategy was not effective at reducing symptoms; participants questioned the effectiveness and tried alternative approaches. This experience was also present in Maria’s narrative where she described being judgemental about sleeping as a short-term coping strategy:

‘I feel like a failure if I go to sleep [in the day] so I want to sleep, desperate to sleep but then it’s often disrupted the dog barks, the door goes or something [...] I have managed quite poorly because I’ve just gone to sleep’ (Maria)

The challenges of figuring out when sleep was necessary were also present within participant narratives. Similarly quality sleep was favoured by participants rather than quantity. For most participants this increased sense of awareness of sleep quality had developed over time.

'getting quality sleep [...] you can sleep and sleep and sleep and sort of feel really exhausted' (Stuart).

In negotiating the role of sleep some participants found having a sleep routine really helpful for their long-term management, which also reportedly reduced the extreme fluctuations in symptoms as well as increasing a sense of control for participants.

'um to go and have a routine in my case it's like being going to bed by nine and getting up at half past seven and that helps a lot 'cause if I lay in in the morning even just for half an hour I'll get a migraine or I'll be all over the place all day' (Alison)

Another instance when participants were acutely self-aware, was related to their environment, for example the effect of extreme temperatures or changing temperatures for their body.

'I just don't like hot weather it's just too demanding-physically demanding hot, you know, going on a hot holiday I find it's just too much stress on my body' (Stuart)

Increased self-awareness was also helpful in recognising warning signs or triggers that they had over done it. There were also many participants that explained learning from their past experiences also assisted them in responding appropriately to a set-back or relapse.

'if I'm feeling myself getting ill I might, like take a day off work even though I'm not actually ill but it might stop me from getting really ill so I'll take the day off and then, carry on if it doesn't develop and take longer if it does develop into something which is a difficult thing to explain to manager (laughs) 'are you ill?' 'I'm not but I might be so I'm not coming in' (laughs)' (Penny)

'I now recognise when I'm heading towards a crisis I know when to pull the plug so it's experience too' (Oliver)

Participants' described achieving this deeper awareness of their experiences as something that had developed over 'a good few years'. Highlighting it did not come naturally to participants and was a learnt way of coping with their fatigue and related symptoms. Participants talked about learning to monitor their energy and activities, which was understood to be something they would always need to do for successful long-term management.

'I've got to watch what I'm doing um and know that tomorrow I've-I need to take things steady [...] yeah y-you have to monitor you have to watch what you're doing [...] you have to keep a continual eye on it' (Colin)

'I think I-you know I will always have to, watch what I'm doing' (Jane)

Most participants recalled after a while, monitoring their energy had become second nature. However, participants also spoke about instances of not getting things right, suggesting relying on second nature or past experiences alone was not always helpful. This fits with Diane's experience; who attempts to be curious in each moment, allowing experiences to be new. In order to achieve this curiosity she needed awareness of her body and mind to connect with her experience to mitigate the risk of making the wrong decision.

'looking at each situation umm in its own space as it were not as umm everything's the same 'cause it's not each day each situation each each interaction with someone they're all different as you have to, erm (tut), you have to treat each one as a new experience as it were. [...] when I was younger I'd just carry on I'd take lots of things for granted an-and just go for it you know in terms of it-the expenditure of energy umm, no it really has only something that that's I've learnt to do, since, not since I've had that condition but since I've learnt to live with it (laughs)' (Diane)

Many participant stories presented contradictions about what was successful long-term management; in one instance something would help and in another it wouldn't. These contradictions and caveats highlighted the challenges of managing their fluctuating condition. Participants with a deeper level of awareness about the energy activities required from them (mentally, physically and emotionally), reported being able to pace their energy appropriately. This was achieved by making decisions about when and how they would do activities to suit their energy levels and symptoms. Subsequently 'Pacing' that was based on this deeper understanding of energy expenditure, was referred to by the majority of participants as a way to gain more control over the fluctuating symptoms of CFS/ME.

'years long of trial and error not oh when I first got it [...] just working out how things affect you, and balancing out as well, what, you can get away with and what you can't on what level of suffering you get from, whatever activity and almost working it out at that well I can I can do activity A and activity B, but that means I won't be able to do activity C or I'll suffer, um, you know symptom A symptom B you know you get different it's all balancing and all trial and error working out the combinations you can get away with working out what you're prepared to get away with (laughs) erm and yeah sometimes you get it right sometimes you get it wrong and just learning from it' (Jane)

'you know a lot of managing the condition is about being self-aware of what you can do, or being aware what repercussions might be if you do over do it so you plan um but obviously (laughs) we are not in control of everything are only in control of ourselves' (Diane)

Challenges of developing heightened awareness also existed in participant narratives. A barrier in achieving awareness that was commonly reported was the urge to overdo it when

feeling well. That said, most participants reported this urge was only a short-term measure and that this approach did not work for long-term management. Many participant narratives included being aware of the urge to push on, but by having a deeper sense of bodily awareness enabled some participants to 'stop before' (Kate) they had gone beyond their limits.

Mentioned by several participants was how the awareness of others could be an asset but also a challenge. Interestingly Kate was able to empathise with others for finding the condition hard to understand as she struggled to understand it too, but made a distinction between those in her life trying to understand to those not willing to do so. The latter existed in participant stories when the other person had limited awareness about CFS/ME. Which was further complicated because participants did not look ill, so incorrect assumptions could be made based on their appearance.

'I think my wife has just about got used to it now and-as someone who hasn't got ME you know after seven years she's just about getting used to it but I still think in the back of her mind she's thinking 'oh god I wish he'd get better soon' mm' (Oliver)

'she [fellow student] looked at me and she said 'oh you don't look disabled' (laughs) you know it's things like that' (Diane)

However when people spent time with the participants, they realised they were not well. Colin recounted when a friend could visibly see he was struggling and getting emotionally upset during a sailing trip. He explained it was validating that somebody could visibly see things weren't right for him.

3.4 Theme: Acceptance 'it's always going to be with you you'll-the illness might not get better but you'll deal with it better so therefore you will be better' (Penny)

This theme links to participants' openness to their experiences. This included reducing the struggle of not wanting CFS/ME, reducing the internal battles experienced and coming to terms with what the illness meant for them. The word acceptance was explicitly used by the majority of participants in relation to the diagnosis of CFS/ME. As with the previous theme, participants also explained acceptance became stronger over-time.

'there's a hell of a lot of denial and fighting against it for many years [...] so yeah accepting that this is how it is and it's ok to do whatever I can do. (Smiles) Focus on what you can do not what you can't do' (Kate)

Some participant's explained further how their doubts can get in the way of accepting the condition and subsequently from using management strategies that can help them. Kate's narrative included this concept of pushing herself to prove she was not ill, but concluded

through her experiences she was. Furthermore, despite the relapses she experiences she still considers making progress by remaining mentally well and not giving herself a hard time. As with many other participant stories, although participants may have felt resistance to accepting the illness initially, participants later found by accepting and accommodating the illness was helpful. With many participants reporting, as Kate did '*fighting against what is happening*' used more energy and prevented doing things that were important.

'I spent maybe the first three years thinking' No I can actually get over this I can fight this' and the last sort of three years I've been thinking 'So ok I tried fighting it didn't win so now I've got to learn to live with it and manage it [...] after you've been through sort of like three years of booming and busting as in sort of really trying to fight and then falling down again I think you kind of need to realise (laughs) it's an idea to try something else (laughs) [...] I mean with the outcome is survivable you know it's it's not going to totally, ruin your life [...] but there is a way through it it may not be what you want but there's definitely a way through it you have to survive' (Oliver)

It was key for participants not to deny they had the condition, as this was in opposition to acceptance and seemed to have limited long-term efficiency. It also fed into unhelpful management strategies of '*proving*' to themselves and others that they were well and creating the boom and bust cycle of activity identified in the first theme (Awareness).

'It's about public, my public view me as opposed to the private me, because I often think when I'm at work they must think I'm a right laugh and I'm always joking about and being silly but when I am at home I'm a different person, you know if they saw me they wouldn't believe I was the same person [...] Because I don't portray that sort of dreariness that I often feel (becomes tearful) [...] um you know I put on an act, you know, it's putting on an act [...] be normal, look normal' (Maria)

Many participants spoke about the need for validation and to be understood because of the significant emotional impact of CFS/ME on their life. Most participants that received validation reported it was central in their acceptance of the condition, and often found this support from healthcare professionals.

'when somebody told me that I did actually have something wrong I was kind of a relief to think 'oh there is something going on here' 'cause I would have to take a lot of days off work 'cause I couldn't go to work and then as soon as the doctor I saw be printed it off his computer explaining everything about it and it all fitted in and it all clicked together and it was exactly how I was. That was a help just to know that I'm just wasn't being lazy or something like that you know. It was useful. (Alison)

Although participants talked about management being second nature after so long, there were also examples of when seeking support from others was helpful in accepting they needed to make changes. An example of this was when Jane sought support from a specialist service when she was were aware of doing too much but struggling to accept it;

'I suppose it just was, it was good on the one hand just to come and, kind of get a reality check and for myself 'cause I knew I was doing too much but having somebody tell you that, makes you listen a little bit more (smiles) I kind of ignore the voice in my head quite well but having somebody else say 'oh you need to slow down' it's a bit different isn't it? slightly'. (Jane)

The notion of accommodating the illness as a beneficial long-term strategy fitted with all participant narratives. Adapting to the illness was explicitly spoken about within a reduced capacity rather than stopping activities completely. Maria and Oliver thought having a significant amount of time off work and then attempting to return is not a beneficial strategy. Maria drew on her own employment experiences *'I was off work for a long period of time, up to a year and I think that wasn't a good idea at all being off work for so long'*. This was echoed by Oliver who suggested reducing energy expenditure to a manageable level in order to stay in work instead of getting out of employment is more helpful way to cope in the longer-term.

Adapting to illness was identified as a long-term coping strategy to achieve some level of quality of life, even though compromises were made in the process. Participant's stories demonstrated choices were still possible, although at times it was between a rock and a hard place. Jane explained how going out was difficult and more so with her baby, but she had choices available. For example taking the pram or carrier and she could take the bus or walk depending on her symptoms and energy levels. A level of flexibility was inherent within participant stories that participants were able to adapt to the condition fluctuations and life stressors.

'[Goal to achieve] some kind of standard of living, not the standard of living I had before but it [motivation to get out of bed to do something] enables me to do things' (Colin).

Participants recognised making choices could also use up emotional energy, by worrying about which decision is the right option or feeling frustrated about the choices available. Yet within participant stories, there was recognition that there was a degree of control related to additional pressures participants put on themselves which subsequently used more emotional energy. For many participants by reflecting on past experiences and giving themselves permission to let go of the emotional struggles of making choices helped considerably. When

participants were able to accept the frustrations of the condition and making difficult decisions, it didn't change their circumstances or outcome but it relieved some of the additional pressures and emotional energy usage.

'If I've got a really bad day then I'll just rest all day 'cause if I try to do anything I would just get irritable and I'll cr-burst into tears 'cause I there's can't physically do it so I find rather than get myself too stressed that I, I just won't do it on those days 'cause it's not worth it really' (Alison)

'so like Sunday when I cancelled [...] I didn't spent ages stressing over whether I was going to or not I was just like 'right this is what I need to do' whereas before, I would have spent ages going-and that would have made it worse' (Kate).

Similarly many participants acknowledged the role of stress for them and their condition. Many participants had experienced severe relapses due to stressful life events. In some stories, participants described that CFS/ME is always in the background so when stressful event happens their symptoms are exacerbated.

'I went through difficult sort of periods with um a divorce and-an we had a still birth and we had a divorce, I lost my parents and I-I think that those triggers sort of re-energised the ME for want of a bad pun (laughs) other expression' (Stuart)

All participants' accepted there was no cure for CFS/ME and subsequently discussed the use of medication in relation to accommodating the fatigue into their lifestyle, to treat specific symptoms experienced. Penny reported taking melatonin for *'a good couple of years [...] I never used to think if it was making a difference but I'm guessing 'cause I was sleeping it probably was (laughs)'* Several participants also reported benefiting from using antidepressants. For the most part, participants described their low emotional state impacting their ability to cope, with their GP suggesting antidepressants:

'when you sort of just in a mess emotionally then nothing, it can affect you physically as well ah you feel better about yourself [by taking anti-depressants] without being you know way-hey crazy high like that was a lose does and uh (breath in) I have tried to come off it, I took myself off it it you know and naively very quickly that was really bad, I had to go straight back on (laughs) and I have talked to the doctor about wanting to come off it 'cause I don't like the idea of taking anything, and at the same time I realise that it may be something I need to continue to take.' (Stuart)

That said, some participants used very little medication or reported infrequently medication use related to short-term relief of an extreme or acute symptom.

'I get sort of general pain throughout um I had that before I had my back problem um so I've been on medication to help with that but um other than taking um the odd sleeping tablet which I don't take on a regular basis but I take them [...] where I just need something (clicks fingers) to switch me. Off. You know' (Colin).

'I've never really taken anything other than you know the usual Ibuprofen cream for muscle aches and stuff erm I've never, never really, taken anything [...] There weren't any (sigh) when I was younger there weren't really anything-any sort of symptoms where I could take anything' (Jane)

All participants accepted that there would be fluctuations and uncertainties with their condition symptoms. With some participants describing finding it hard to deal with an unknown future at the beginning of the illness and overtime found this became easier to manage:

'it is very much this rollercoaster you're on all the time' (Colin)

'you get knocked back you go back to the beginning and you start again' (Stuart)

Oliver described the extreme energy highs and lows in the earlier phase of his illness experience did level out overtime, reducing the fluctuations and uncertainty. The price of this however was life seemed less fun. He also noticed a reduction in physical fitness including the ability to push on, he accepted this way of coping was no longer possible and potentially aged related.

'because that sort of vaguely sort of relaxed slower pace of life it seems to even out the extremes the bad and the good extremes [...] I mean you have no choice but otherwise you would be constantly booming and busting all the time I mean y-you get to the stage where, I mean now I couldn't push myself the way I could push myself when I first had ME you know physically I couldn't do it but when I first had ME you know I could really sort of go for it and I'd pay for it by crashing and burning afterwards but at least I could do that now I don't have that choice, now I must plan now I must. To tell you the truth yeah I had more fun then (laughs) a lot more fun then uh but uh I suppose the way I am now it's probably more suitable to my age and stuff' (Oliver)

Linked to fluctuations was the idea of cycles, raised by some participants when talking about recovery, in that recovery related to coming out of relapse cycles. It was these relapse cycles that contributed to the pattern of CFS/ME as a fluctuating illness. From the participant narratives by accepting relapses would happen, reduced the sense of personal responsibility and as a result alleviated additional distress.

'I've realise like I absolutely accept it's not going away (laughs) it's going to keep it might keep coming back so if I can maintain it and keep at a level, that I know I'm ok at and not do too much' (Kate).

Accepting that keeping a routine and achieving 'balance' was present in many participant stories as effective long-term management. Many participants also stated by visually seeing their commitments in a planner they could balance their commitments more easily. Although a helpful management strategy, routine and achieving balance was also a challenge; as Oliver described *'it's very boring (laughs) it's really fucking boring (laughs)'* and Kate said her friend laughed at her for having a paper diary instead of using her phone to manage her commitments. This strategy of planning a routine was present in several participant narratives as a way of adopting effective long-term management. It was also reported to help manage the extreme fluctuations of the condition; creating a more consistent and stable energy expenditure. Many participants also stated that accepting getting up in the morning even if they were not feeling well was important. Staying in bed was not useful physically, mentally or emotionally and so enabled participants to accept that despite the challenges, sticking to a routine was particularly helpful for their long-term management.

'when I was at my best is when I had a routine' (Jane)

'and there' no-no question I-a-I-a if anyone said to me 'I-I've got it what should I do?' I would say 'Keep a routine don't-don't break a routine you know just stay in bed if depending on how severe they are try to keep in the routine of getting up in the morning' (Stuart).

All of the participants had a conceptualisation of CFS/ME as life-long illness. Which in the most part enabled participants to adjust to living well with the illness instead of battling against it. Jane described *'going with the flow'* which helped with accepting the potential life-long impact of the condition, as she had no expectations about the length of time she would be unwell. She reported being diagnosed as a teenager at a time when little was known about the condition and had to get on with it. This proactive attitude was also found in other participant narratives as a positive long-term strategy:

'I think you know you're looking at something that you may have to realise is going to be with you forever and I mean certainly the way I've started to look at it now that I don't see it going and I don't see any way I can, make it you know [...] and if you don't, accept it (laughs) [...] it's going to make you mad it'll just send you over the edge I mean I've for long period I've been really quite depressed about it and figured myself it's just not worth it and uh, I've managed to (laughs) plateau out' [...] I don't think it's going to go uh unless they come up with a recognised something that there is a drug for basically suppose what I'm saying I don't see myself as ever being free of it believe me I'd love to be free of it (laughs)' (Oliver)

However getting to a place of acceptance with the longevity of the condition was harder for some participants:

'I have always done I know it might sound management speak but one hundred and ten per cent all the time [...] so to suddenly go round turn round and say I'm not doing that anymore that's a real life changing thing to try to deal with you know.' (Colin)

'maybe when I got a diagnosis and was referred to [Hospital name] having some sort of hope but then having that hope dashed in a way because I thought real-the realisation that I'm probably stuck with this if not for the whole of my life then at least for quite a long time amm, so, it basically comes down to how you manage it' (Diane)

Some scepticism about other people recovering from CFS/ME was also evident in participant narratives, but those that raised this suspicion concluded for them, CFS/ME would always be present:

'I don't thi-I honestly believe it ever goes I know some people say they've recovered from it, maybe they have but, I, don't think that I will ever truly be rid of it I think I-you know I will always have to, watch what I'm doing I believe it can-you know g-it goes into remission as it were I think that' (Jane)

'I suppose my strongest thought on that is did they actually have what I've got [...] I don't know-I-I I don't really think about it erm, I just focus on myself really and making sure I keep going forward rather, than back' (Stuart)

This scepticism was also linked with a curiosity for two participants, to see if other people they'd met with CFS/ME in the past still had symptoms or disclosed an interest in seeing them in the future to check if the condition had *reared 'it's ugly head again'*.

A sense of normality was discussed by some participants, in both their illness experience and management techniques where they had become normal or second nature.

'it's difficult to explain really especially when you've had it for so long you sort of forget it's second nature to me now' (JANE) 'in the early days I-I, it hit me, a few times you know I-I remember being quite upset one time thinking 'oh I just don't remember the last time I felt well' (tut) and then obviously when it got to thirteen years it sort of fourteen years I sort of thought 'oh well you know I've had it longer than I haven't had it now'' (Jane)

'just normal that I call it normal 'cause I'm so used to it' (Alison)

There were also instances within participant narratives where it was normal within the household to have a slower paced life or a shared illness experiences. These instances appeared to make it easier for participants to accept not being able to do desired activities due to CFS/ME symptoms. Alison's husband has Multiple Sclerosis and so within their home there

was a shared illness experience as she explained; *'I would like to go walking if I could walk further 'cause we're both restricted in that way'* and also Oliver's experience:

'I'd like ah you know [...] go out with the wife and stuff and uh mind you she's too knackered being an-staff nurse (laughs) to so much so, that's not much of a problem' (Oliver).

Normality was also raised by Colin, that his inability to work resulted in a dramatic change to family lifestyle and this adjustment was hard in the earlier period of being ill. Whilst the adjustment was ongoing Colin did not indicate this was still a struggle compared to the initial illness onset. Accepting the limitations of CFS/ME was also identified by many participants as a normal experience, that people regardless of health cannot do all they want to do. Jane explained after her baby was born she struggled with all the demands as many new parents do, and she went through a process of accepting her limitations and accepting help from her husband:

'the real turning point for me was realising I couldn't do the four o'clock night feeds and that he [husband] would have to do that so and from that point when I admitted that actually I wasn't wonder woman and I, couldn't do, every single feed erm things got easier' (Jane).

Acknowledging everyone, regardless of illness has difficulties managing demands was comforting for participants and enabled them to achieve a broader perspective that did not just focus on their illness. Conversely some participants also acknowledged they were different from others, but that these differences existed prior to the illness. Stuart described noticing he was different from his peers in his early twenties and thirties; needed longer recovery periods following excessive exercise when he just wanted to sleep and noticed feeling very low emotionally. This was before his diagnosis of glandular fever and so he attributed his experiences as just the way he was.

Acknowledgement of the individuality of people's experience of CFS/ME was present in all participant narratives, and was reflected in the individualised language participants used. For example; *'for me'* *'personally'* or *'I find'*. This notion of individuality was explicitly raised regarding the role of individual complicating factors (such as age, fitness and comorbidities). Comorbidities shared included Fibromyalgia Syndrome, Coeliac disease, heart attack, back pain, arthritis and a hernia.

'as I became older umm you know you had you know you you become old and knackered anyway (laughs)' (Stuart)

'I have other things wrong with me too which doesn't help [...] but the ME by far is the biggest, one of the worst things but I think once you get past a certain age I would say 45, your chances of recovering from ME fully are reduced, as and that's you combined that with having other physical problems' (Oliver)

Some female participants also reported the involvement of hormonal changes adding further complications; during puberty and menopause. These individual complicating factors were a way for some participants to make sense of how they developed CFS/ME, offering theories of causation. Alison wondered about the involvement of hormones as she experienced early menopause, putting her illness down to that initially. Following the bereavement of losing her mother some years later her CFS/ME was diagnosed that same year. So she began to question what had caused the fatigue, she also wondered if her job in a school may have exposed her to viruses. Likewise Colin conceptualised his CFS/ME to be a combination of other health issues (heart attack, hernia, spinal issues, hip problems) and his job (working long hours, travelling and stressful role). Some participants who had a theory of causation seemed to accept having the condition with less struggles than those that couldn't make sense of getting the diagnosis. A rare theory of causation was offered by Oliver implying that through karma or consequences of his actions earlier in life he developed CFS/ME:

'I was quite a hooligan for quite a long time and uh, I kind of think that somehow may be this is (laughs) is what I deserve in the long run (laughs), it's what I deserve life kicking you in the ass' (Oliver)

3.5 Theme: Connection at least I feel that I've got areas of responsibility and control [...] and I achieve things' (Oliver)

This theme encompasses the experience of participants connecting with themselves and what is important to them, as well as connecting with other people in their life. Connection with the self and others appeared to increase understanding about the condition impact, enabling participants to respond according to personal goals and values. Understanding and acknowledging experiences in more detail as described in the previous themes (Awareness and Acceptance) seemed to precede effective decision making allowing participants to tailor goals and direct their focus on something important.

Connection was also discussed in relation to talking and sharing experiences; as a beneficial process in connecting with the self and others. Interestingly some participants stated for them, the interview itself was helpful to talk about and reflect on their journey:

'It [the interview] made me realise how far I've come (I wouldn't have got through it without tears in the past!) so was useful to me, thank you' (Kate, email following interview)

That said, Jane explained she doesn't tell people she has CFS/ME anymore as she is managing ok, therefore doesn't feel the need to tell people. She also described the condition not being central to her identity, as did Maria which also contributed to not disclosing to others their diagnosis.

Jane: ...and until I could sort of manage it and keep it under my hat...

Researcher: uh huh

Jane: ...and things like that, an-and I just I just hide it now to be honest...

Researcher: yeah and that wor-that's a helpful thing for you to be doing?

Jane: Umm, some ways it is some ways not, erm, but it does leave question marks sometimes people will you know people I think some people wondered why I only worked part time or why I get the bus for free or you know things like that and some people ask, some people don't...

Researcher: mm hmm

Jane: ...erm but yeah it's just I suppose, when I was younger it was sort of almost used to define me 'oh that's JANE she's the one that's got ME' so new people that I met...

Researcher: mmm

Jane: ...I didn't want to, sort of know me as 'oh that's Jane she's got ME'...

Researcher: mmm

Jane: ...or whatever or she doesn't work because or you know things like that...

Researcher: mm hmm

Jane: ...so I think that's why just as I've got older I've sort of thought no there's more to me than...

Researcher: mm hmm

Jane: ...this, you know

Kate found that having significant time off school and some time in a pupil referral unit that by building up her energy levels slowly she was able to return to school. The importance for long-term management to gradually build up to tasks or increasing energy was shared by my participants in achieving their aims. It was the opinion of most of the participants that gradually and consistently working towards personal goals was central to successful long-term management:

'but not setting your goals too high, ah too-too , to-to-to-to build up gradually to a point' (Stuart)

Equally participants also spoke about connecting with things that were important in terms of their roles and responsibilities. Having a purpose or reason to be doing certain tasks was helpful although some participants still wanted to be able to do more of the things they enjoy:

I didn't have anywhere to b-well I did have somewhere to walk to if I'd wanted to but I didn't have a purpose it's like not having to work-walk to work I didn't have to do anything um so not having an incentive to do anything was really bad I think' [...] not having a role and not feeling useful and not having a purpose' (Maria)

'I'll never be able to work full time again I know that I'm not-not a chance in hell but I might be able to work part time I might be able to work from home [...] I think you need that, um I mean before I'd keep myself occupied by doing loads of DIY and stuff and I was going out and helping people and that, my father who's ninety three and I was going down and looking after Beryl [mother-in-law] she's ninety three, one each with each family ah but that's not, I'm not doing that so much, so you're invalidated by not having work' (Oliver)

Maria also explained responsibilities could be both; unhelpful because it was an added pressure, as well as helpful being a positive motivation to do things. For example looking after her children was daunting as she questioned whether she could manage some tasks, but reported it was helpful to get her out of the house and out of the headspace that was telling her she couldn't do it:

'sense of wallowing in, 'uh god I feel so tired I could just stay in' you know and not go anywhere not do anything. I found if I made an effort, and went to-o the cinema say, I found that that was an achievement and I felt better for it rather than just giving in and feeling 'oh I'm incapable of doing anything so I think , making-it's almost like making the effort to go to work and making the effort to go out I felt better having gone out but it's a massive effort to do it-sometimes I couldn't you know it was a b-my children were still at sc-little then, and I had to go and collect them from school and I remember just then just think 'oh god, I can't go and collect them' and it was a huge effort to go um, yeah so sometimes making the effort helped and other times not' (Maria)

This extract really illustrates the complexity of managing CFS/ME that roles and responsibilities can give a purpose and in some ways force people to carry on when feeling unwell, where the body and mind is saying they can't do something. Despite it being challenging, connecting with those roles and responsibilities was a useful long-term strategy participants adopted. It also illustrated the dichotomy of having roles and responsibilities as they can create a sense of pressure and obligation which is not seen as helpful. This pressure could also feed into difficulties or confusion in prioritising what was important for participants to use their energy doing:

'we used to be in a skittles team and it ran throughout the winter so it would go from September right through to May, so that was a commitment once a week you know as part of the team and my daughter took on the captaincy of the team as well and I helped her so that was a-a commitment that sometimes you know when you get home from work and you think 'I really can't handle that tonight' but knowing you have to so putting you're-I know it was fun and it's good being with other people and playing skittles but at the same time if you know it was um, (tut) something you always have to do when you didn't always feel like it so stopping that' (Diane)

Other stories also recognised the importance of connecting with family for motivation and despite the challenges; family membership was viewed as a helpful management strategy in providing a role and purpose. Several participants also explicitly mentioned family pets as beneficial to them with regards to having roles and companionship;

'she's [dog] been there for me since the day I was-well a month after we got her I was I was signed off [work] so we've been together literally all the time you know she's my little shadow [...] she's just nice you know [...] and lovely placid erm mannerisms about her um you know if you're on your own my wife's at work you know she's there so so I talk to her.' (Colin)

'Get a cat or dog, dogs are probably better because you can walk dogs umm but um something so you're not spending a lot of time on your own. Uh I personally feel incredibly sorry for people who've got ME who are on their own, yeah it's dr-it must be dreadful I mean the one thing that is motivating is I've got my wife and I want to do what I can for her and that's a great motivation and so that really makes you press ahead and uh, yeah having responsibilities like aging parents and things although it's a pain and it does bring on crises 'cause it's stressful because you can't manage the time you've got to do all the things in it's useful because at least you-you feel that you've accomplished something and you know.' (Oliver)

As with changes reported in valued activities, participants also spoke about the changes they've made to their role and responsibilities. These changes were in light of living with and accommodating CFS/ME, which provided a purpose within the condition limits.

'you can make long-term plans as long as you build lots of things into them and what I've done with my wife I've sort of said 'listen, I'll look after the house, I'll do all the cooking, the cleaning I'll do everything with the house I'll do all the DIY but if-the way I can do it' and so try not to push myself too much so I am going to get really bad [...] and I manage to just about keep all the sort of household stressors off my wife, she doesn't have to do anything so she can concentrate on being a nurse an at least I feel that I've got areas of responsibility and control [...] and I achieve things' (Oliver).

Some participants' changed their job and found this had a big improvement as their roles before were too energy demanding or not suitable since developing the illness.

'I have improved an awful lot and I think a lot of that is to do with not having the stressful job, 'cause that was really hard work [...] physically and emotionally and cognitively in every way really to continue and then I made the decision to leave and that was a huge relief' (Maria)

'it's got better for me since I've I've changed jobs and I've altered my existing job role o-or given up my degree studies so in despite about not being particularly happy about that I know that, that's what I had to do' (Diane)

This theme also included the need for support and there was clear difference presented within participant stories of needing 'concrete' support and 'abstract' support. The former related to practical and physical tasks that participants were unable to do when symptoms were increased, often sought from others. With the latter relating to less defined concepts such as addressing mental and emotional wellbeing which participants achieved themselves, or with the help of others.

3.5.1 Sub-theme: Connection with self *'I'm letting myself down and making myself ill if I'm not, erm, doing what's best for me and my body' (Kate)*

Linking again to the previous themes, being aware and accepting of CFS/ME left participants feeling disconnected with their sense of self at times. Several participant narratives included the importance of connecting with their sense of self during the illness experience, which for many also required working on their psychological and emotional health. Diane described working on the self an 'abstract' concept but fundamental in her long-term management. This was a similar thread for several participants, with some reporting being able to address this themselves in a very natural way whilst others benefited from professional support to achieve it. Kate found working with a psychologist at a specialist service as really helpful:

'and-yeah 'cause it was always a-there always seems to be a dip mentally when I dip physically so can-being able to stop the mental dip so much helps the physical if that makes sense? [...] I think I'd be in a heap if I hadn't come to [hospital]' (Kate)

Two participants described finding their 'own way' in managing the fatigue and related symptoms with minimal input from specialist services, but found it useful to have support from professionals if needed. A few participants also highlighted timing and appropriateness of professional support available could be important.

Kate believed if the same psychological support was available to her when she was first ill and diagnosed as a child it wouldn't have been as helpful. This was because she didn't think she would have been able to articulate how she felt as a child and therefore not made the progress she experienced.

Relevance and timing of professional support was also raised in negative scenarios by a few participants:

'two three years into it [...] I thought I needed practical advice not psychobabble (laughs) learning to be self-aware is important but that in my mind isn't the same (laughs) as CBT or Mindfulness the way they [hospital] um what's the word, uh, gave it to you' (Diane)

Colin also shared his view that the NHS courses were *'run solely for women'*. This was based on examples in the course he attended related to household chores specifically; ironing and washing. These examples were considered to be female orientated with men as the *'main bread winner'* in most cases and he explained the examples used on the courses did not relate to him. Colin explained he needed help with managing his work commute rather than household chores and so the course was not personally relevant to him. This potential gender preference was mentioned again during Colin's interview *'you could relate to the male [counsellor]'* however when explored further this was not a strong perspective in seeking out helpful professional support:

Researcher: I was just wondering is there something you know saying about your GP it's difficult to see a male GP um and you know your experience with more female typical tasks in kind...

Colin: yeah

Researcher: of the PLACE area I just wondered if that actually that could be an element w-that you find personally helpful?

Colin: To be honest with you I mean (sigh) if you've had y-had a hip joint replaced dignity and all the rest of it is out the window (smiles)

Researcher: (laughs)

Colin: I'm big enough and ugly enough to (laughs) just get on with it now, 'you're a doctor just do what you've gotta do' and-um...

Researcher: mm

Colin: ...so u-e-a, you know I mean, , had a p-prostate examination well you know what that entails...

Researcher: mm

Colin: ...and the doctor says 'well would you like me to get a male colleague in?' I said 'well just get on with it' you know and that's it, it doesn't bother me...

Researcher: ok,

Colin: ...it doesn't bother me...

Researcher: Yeah

Colin: ...um what does irritate me though is when you get the stereotyping

Researcher: Yeah

Colin: you know pace yourself don't do too much washing don't do so much ironing g-get off (laughs)

Researcher: yeah

Colin: you know (laughs)

Researcher: yeah so more maybe kind of assumptions w-s-whoever you are seeing brings some assumptions about tasks and things...

Colin: yep

Researcher: ...would that be right?

Colin: That's, yeah, yeah.

Relevance and connection with the self drew heavily on what mattered most to participants; core values. Within all participant narratives were various examples of how participants were able to still connect with their values despite the fatigue and related symptoms. Many participants also described changes in this area, through re-evaluating their values or adapting activities they did before illness to remain consistent with what was important to them.

'I kind of figured out a way where I can function and I've got aims and objectives which I hope to I'm getting some electric bike as well low impact mobile. Yeah yeah you've got to think on your feet and try and work your way around it' (Oliver)

Colin described enjoying photography, although going out to take photo was not as easy for him since developing CFS/ME, he particularly enjoyed editing photos. The latter task suited his energy levels as he was able to use existing photos he had, could do it at home and take regular rest breaks as he needed. Diane also explained she was able to do enjoyable activities that were also beneficial for her health:

'I love reading and just lying in bed reading is is relaxing although I am using mental energy reading it's still you know it-it's good and enjoyable and at the same time I am resting my body' (Diane)

Competing values were spoken about by many participants as compromises or choices they had to make, which were often uncomfortable. Diane likened this approach to losing occasional battles to give her more chance of *'winning the war'*. This example of having an overall picture or direction of life based on personal values was also included in many stories and included *'costs and gains'*. Many instances of this in participant stories included the process of weighting up whether activities were worth it, in terms of energy expenditure and the gain they got emotionally, physically or mentally.

'I'd quite like to get out I don't go out in the evenings ever because I'm (laughs) screwed by about seven or eight o'clock I would be just gone [...] but uh I was it's no don't go out in the evenings don't have late nights no have parties it's very boring (laughs) it's really fucking boring (laughs) [...] probably drink too much I gave up all the really interesting things, you know sort of drugs and motor bikes and stuff ummm because I couldn't really handle them any longer you this sort of it it wasn't feasible anymore the returns were diminishing yeah' (Oliver)

'I if-if something that I enjoy doing maybe, going to visit family or or playing tennis with my son it makes me feel good psychologically and physically for a while but then has it's repercussions you know' (Diane)

Whilst enjoyable activities were given up if the balance of costs and gains was not right, participants also identified lost values since developing CFS/ME. Kate talked about the loss of exercising since being ill, but now her health is a priority so she described not risking exercising at the cost of her health, even if there was a chance it could help.

'it's so frustrating (laughs) I don't do anything [exercise] apart from walking, which to me isn't enough like I used to love playing badminton but that's not an-and like going dancing but no (laughs) erm so and I'm not willing to take the risk now 'cause I know that some people do recommend it but I'm because of stuff that's happened before I'm like no I'm-pacing works' (Kate)

Several participant stories drew upon recognising and valuing self-care and self-compassion. This existed in participant narratives as having the confidence to follow what action was needed at the time to be in line with what participant's valued. Stuart described being more confident to say no if he is unable to do something, this was something he had to develop over time like many other participants;

'I'm, more confident in saying 'no' to people than I used to be so yeah in that sense I'm being kinder to myself some people might call it selfish other people might say no actually you know huh you're actually helping yourself you know in a healthy way' (Diane)

'learning to say no has been a big one um because I don't like to let people down umm but actually, I'm letting myself down and making myself ill if I'm not, erm, doing what's best for me and my body' (Kate)

Another aspect of self that was acknowledged by participants as being important to armour against the impact of living with CFS/ME was humour.

'mentally you've got to have a certain mental strength to cope with it preferably a fairly dark sense of humour (laughs) Yeah. If you don't have a sense of humour you are, gone it's not going to work and-and that does help a lot' (Oliver)

'we have a huge laugh there [at work] we laugh a lot and really nice people [...] I look forward to going into because it's not demanding and

it's very sociable and we laugh a lot and I think laughter is really important (laughs) you know having a good time' (Maria)

Whereas, for others the use of humour enabled an expression of their sense of self: When talking about multiple health problems, Colin stated *'is quite, depressing quite frankly you know'*. He then took a laser light out his pocket and began to shine it during the interview:

'so then you do this (brings out a red light from pocket), takes your mind off things [...] I-I often act a fool [...] I just find it quite funny you know [...] I've always been that way' (Colin)

3.5.2 Sub-theme: Connection with others *'support from family is-is a must you can't do it on your own' (Jane)*

Many participants spoke about having *'concrete support'* which included more *'practical'* and physical tasks. Generally participants described this form of support received from others, however information was also highlighted by Stuart as something concrete which gave him a sense of validation and relief.

'my husband helps me do the heavy things like hoovering and I can find ironing very tiring so he do that for me and um we get someone in to do the windows and the oven which are the two jobs I just wouldn't even attempt because I know they wouldn't get finished so that-those are the things that help me through getting through it u-i-it's practical help' (Alison)

Further practical support detailed in the majority of participant narrative included; financial support, blue badges, cooking and housework. Many participants described the need for this practical support but recognised the desire of not wanting it. During a relapse with six months off work, Kate was unable to leave the house and to look after herself:

'I couldn't-I wasn't cooking so I ended up moving back to my parents for a month um, which was, amazing you know but-an obviously they would have me but I didn't want to do that-that was a massive loss of independence' (Kate)

Kate also highlighted the relapse was hard for her financially so getting those systems in place was a real help for her. However, other participants found this difficult, finding the process of accessing the benefits system exhausting and demoralising. Despite her partner suggesting it, Maria didn't explore financial support as she didn't think she was severe enough to qualify and recognised she was struggling to accept the illness:

'I don't want to be on DLA' that's implying that I'm ill and I don't think I'd get it anyway because I'm doing a lot you know I'm not-not crawling around being at home not doing anything I'm actually f-I'm here I'm going to work I'm doing all sorts of things I'm not disabled you know so I, I am managing but on a reduced lifestyle if you know what I mean yeah' (Maria)

Several participants shared examples of interacting with unsupportive people which created a barrier for them and in worst cases negatively impacted their long-term management. For instance Kate's GP asked her when she was going to get better as if it was a choice, which she found particularly unhelpful. This prompted her to change GP's so she felt supported and as a result was referred to a specialist service enabling her to manage her CFS/ME more effectively:

'that's just not helpful so I've changed [GP] She-she knew about [hospital] I was like 'wow' (laughs) he didn't and I-I don't expect them to know everything I don't like the-no but, she'd been really good and actually been alright since I've changed to her so haven't really-but I know that, because of her reaction when I first went to her and explained why I'd changed surgery's erm I think she would be fine so that's having people onside is massive' (Kate)

Similarly when a teacher at school suggested to Jane she needed to build up stamina so told her to work through her fatigue as she wasn't going to be allowed home, she believed it *'doesn't work like that if you need to rest you need to rest [...] such idiots (laughs)' (Jane)*. Common responses mentioned in participant stories when managing unsupportive people was to remove them from their life:

'I haven't had that many negative, things happen I've been quite lucky in that sense guess the lack of understanding is very detrimental especially with working and umm university was a difficult one for the first year, but so they people not really getting why you're doing the things you do or in some cases not doing certain things is probably the hardest [...] I did a lot of ignoring people (laughs) Umm, explaining which is not easy if people have never heard of it' (Penny)

One participant, Stuart also presented caution in talking to others about his condition because of the lack of understanding:

'I think you have to be really really careful about who you tell and 'cause it can sometimes it can make-other people don't understand it, it can almost belittle th-the you know the position you are in 'cause they say 'ah ay-y-yeah I'm I get tired, I get run down' they don't they don't understand this sort of deep fatigue that you have you don't see them when you're like that 'cause you don't go, you stay at home don't you? So um I think it's very important to find someone that you do really that is a soul buddy someone that you do trust and you do trust you can confide in' (Stuart)

Alternatively some participants tried to find ways of explaining the condition to important people in their life, highlighting the importance of communication for long-term CFS/ME management. Colin and his wife benefited from formal counselling which reframed their conversations to remove the conflict that was occurring due to the lack of understanding, on both sides.

'very first off we nearly got divorced [...] um I'm at home all the time, and, uh, I'm-she'd used to being there doing her thing now I'm in the way and it just-it was spiralling out-out-out of control' (Colin)

Participants also spoke about attempting to communicate with others about their illness using as little energy as possible. Participant narratives that included examples of communication also indicated emotional energy could be used by becoming upset or frustrated when people did not understand.

'my sister's not that helpful because she doesn't understand th-t debilitating side to it but she thinks you're just tired you know go have a sleep and you'll be better but it's not like that at all so I don't tend to talk to her a lot [...] you think 'if only it was that easy, it's not' (laughs) Don't talk about it just makes me cross really (laughs)' (Alison)

Several metaphors and analogies were used by participants to explain their condition to others as well as to the researcher during the interview. Many participants used a battery analogy to specifically describe energy levels, with Oliver also describing energy in a quantifiable unit form; *'you learn to sort of parcel out energy'*. Jane used a common illness comparison to explain to others what her general experience was like:

'I've always explained it is-is when people have got full on flu, and they've got rid of the cold snotty bit and they're left with that horrible, tired, drowned sort of ti-you know that kind of horrible feeling [...] it's like tired I just, can't move' (Jane)

Through effective communication participants described procuring the benefits of talking and sharing their experiences with others. Participants reported that although it could be difficult, talking to others was useful in both informal and formal situations for long-term management.

The impact of the condition on the family was also touched on in participant narratives. The perception of good long-term management in participant stories was assisted by supportive family and partners that had shared beliefs and ideas. This was similar for Colin who also mentioned substantial impact of health problems on marital relationships and how counselling was really helpful in facilitating his wife to understand his experiences.

'I've got a very good supportive husband knowing he helps me through it a lot 'cause it affects him as well as me really you know he's good with me and that help me. Um I talk things over with him he'll give me a hug when I need a hug and things like that [...] an that he's not very well himself but we sort of help each other (laugh) at the end of the day we've got to (laughs)' (Alison)

Meeting other people with CFS/ME was talked about by participants as both good and bad. In one respect it was found to be a useful process; gaining reassurance, reducing isolation and highlighting what not to do, but also unhelpfully as it could be emotionally draining:

'they were wallowing and that was not something I was prepared to do' (Jane)

'a lot of the people [at the hospital group treatment] were very similar to me in terms of were working or some people doing incredibly difficult jobs u-um very high powered jobs and so I was reassured to see that people who other people had chronic fatigue weren't dreary old boring people you know drooping around all the time that they could be interesting and lively and whatever so that was useful' (Maria)

Alison explained that different people need different levels of support and for her getting professional support was more helpful than peer support from other people with CFS/ME.

'I think people vary really some people find going to support groups help them discussing things there, well I've never gone down that road I've just sort of dealt with it in my way with the GP and I did come to [Hospital name] to see a lady I've forgotten her name now, um, can't remember her name it's gone but she was really helpful as well'
(Alison)

A further comment regarding professional support was discussed with several participants feeling alone to manage their condition. That said several participants also spoke about empathising with healthcare providers recognising the complexity and limitations of support. It seemed to be the act of sharing their experiences in a relatively informal way but having the opportunity to talk to HCPs that would be really valuable and was lacking in participant's day to day interactions with others. This notion also fitted with many participants describing the interview itself as a useful way to reflect and talk to someone who seemed interested in their experiences:

'as far as I'm concerned no um my GP doesn't really want to have any sort of regular contact and to a large extent I can understand there's not a lot they can do but i-t-h it's just nice I guess from my point of view to know that somebody gives a damn (laughs) [...] I mean daft as it might seem and I used to go and see the GP every-every three-four months something like that 'cos I needed to (sigh) but that in itself was quite, dare I say therapeutic if I can-it enabled-enables, a bit like what we're doing now really although you're not saying a great deal I'm offloading onto you sort of thing and I think 'well ok somebody's listening' um three people listening really (nods to recorder) with ah (laughs) [...] um and for somebody just to for the GP to turn round and say 'well w-a-actually that's quite natural and that's quite normal perhaps you could try this?' (Colin)

3.6 Comment on population differences

Whilst there weren't many striking differences between participant stories there were two potential gender difference observed: Firstly the three male participants explicitly mentioned temperature awareness in relation to their long-term management. Although on closer inspection, these differences may not have been gender related; Colin reported liked hot weather and not the winter considering this a particular management strategy he had some control of but equally a female participant Alison also commented on the benefit of warmth, although this was specifically mentioned in the context of a comorbidity (FMS). Stuart and Oliver however were unable to tolerate extreme temperatures and extreme differences. This awareness with regards to temperature perhaps was more of an individual awareness for participants about recognising triggers for Stuart and Oliver compared to Colin and Alison understanding what temperature environment they prefer. The second gender difference related to the management strategy of monitoring, again within the awareness theme. The three male participants likened monitoring energy to being on a diet, yet this analogy was not used by any of the female participants.

3.7 Instances of embodiment

Additional interactions occurred during the interviews which is noteworthy and so included in the study findings. Whilst the data was based on thematically analysing the interview transcripts there were various instances where participants demonstrated their experiences beyond the words they used. As the data was not coded for discourse analysis, nor the selected analytical method it could not be ignored there was a performance element of the interview interaction. Whilst these instances of embodiment did influence the generation of themes, the process in which they were reported was more akin to content analysis. It wasn't until the most obvious instance of embodiment took place, which was Colin and his laser light were the other participant transcripts searched for instances of embodiment. Table 3 includes the instances of embodiment found using the transcripts and an email from Kate following the interview which included a photo (Figure 3) that was shared during her interview.

Table 3: Instances of embodiment

	Talked of:	Related theme:	Interpretation of function:	Instance of embodiment:
Colin	Humour	*Connection with self	Lighten mood, distract mind, be person he is/ wants to be	Using laser light – laughter with the researcher
Kate	Pacing	*Awareness *Acceptance	Spreading out activities and interspersing with rest	Cancelled and re-arranged interview
	Self-assurance	*Connection with self *Acceptance	Mantra, inner-voice to reassure herself, but used an external prompt to reinforce this	'Wall of Wisdom' (Emailed researcher photo)
	Reflecting on experiences	*Awareness *Connection with self	Value of talking and sharing to reflect on management and experiences as a process	Email to researcher, following interview
Stuart	Information seeking	*Acceptance	Actively trying anything to help, taking any opportunity to gather information to help	Asking the research for new knowledge and experience
Diane	Problematic symptoms	*Awareness	Different symptoms could require different strategies/approaches	Contradicting accounts and seeming muddled at times

Figure 3: Kate's 'Wall of Wisdom'



4 Discussion

4.1 Study review

4.1.1 Aims

As there are so many facets of CFS/ME unknown (aetiology and illness trajectory) and with high individuality in presentation, this study aimed to explore in greater depth how people manage their condition over time. Whilst CFS/ME is classified as a long-term condition very little research has specifically sought to gather information from those that have had the condition for a long time. Therefore this study specifically drew on the participant's wealth of experience in managing CFS/ME over many years. The research aims were to think about effective and non-effective ways of coping appraised by the participants, which could be useful in thinking about appropriate and relevant support for those with CFS/ME. Equally adding to the vast amount of quantitative research suggesting effective interventions this research hoped to comment on the therapeutic mediators and mechanisms of change that can facilitate better outcomes and maintain improvements. It was supposed that those who had longer experiences of living with CFS/ME would have encountered various treatment settings and adopted various coping methods and therefore of significant value to discuss. There was also an agenda to explore participant's experiences of illness trajectory, to support or contradict the suggestion of distinct stages or models of illness.

4.1.2 Procedure

Potential participants were identified using the BCFS patient database and eligible patients were invited to take part by post. Those interested in taking part were then invited to attend a face to face interview with the researcher at the RNHRD site at a convenient time for them. Interviews were audio taped and transcribed following the interview. Data was analysed using TA as outlined by Braun and Clarke (2006) by the researcher only. However discussions took place with the researcher's Director of Studies (JBD) during the refinement of themes and Advisors to gather expert reflections during the writing phase.

Whilst the interview transcripts were the main source of data for the analysis, this can only be a representation of the interview, which differs from the interview experience itself (Braun and Clarke, 2013). Various unexpected interactions occurred during the interview; where participants demonstrated what they were saying in more than just words. For example Colin was talking about lightening the load of the emotional the impact of CFS/ME and his other health conditions through humour. During the interview he began using a laser light, shining it around the room and also spoke about his sense of self during; as a *'happy-go-lucky person'*.

By playing with the laser light during the interview he demonstrated this characteristic beyond his words alone. Instances of embodiment occurred for other participants too and although the method of analysis was less concerned with the performance of language (unlike Discourse Analysis), there was an element of literal performance in these instances of embodiment of participants showing or proving what they were telling the researcher. The instances of embodiment played a central role in the researcher's reflection with regards to their clinical practice, but also gave more power in influencing the TA findings. This was because the instances of embodiment provided confirmation that the words used were providing access to participant cognitions and therefore the content of the shared experienced seemed more credible.

Another deviation in the planned data collection was an unanticipated email from a participant sent following the interview. The content of the email was included in the research findings because it contained similar code found in other participant transcripts. The code referred to the benefits in talking and sharing experiences with several participants highlighting this function was achieved by the interview itself, demonstrating the importance of reflecting on their experiences. This concept has been found elsewhere in health research, as suggested by Kralik, van Loon and Visentin (2006) reflecting on illness experiences enables adapting and developing resilience to chronic illness.

4.1.3 Findings

Themes created from the data demonstrated the complexities and individual nature of managing CFS/ME. Gaining a deeper awareness, accepting and taking the time to reflect on what is important has enabled participants to manage their CFS/ME symptoms more effectively and reduce the impact on their life.

The participants indicated that information gathering through awareness is an ongoing process, which has developed over time. Awareness was the foundation of participant's long-term management, which included achieving a heightened level of awareness in relation to symptom experience, personal values and external circumstances.

Acceptance based on the participant's experience was conceptualised as being different to giving up, it was described as an active process in helping participants live with CFS/ME. Again this process was achieved over time and was reported to reduce the impact of the condition on their lives. This view of acceptance was the polar opposite to denial, fighting the illness and giving into it which were considered unhelpful and short-term responses.

Participants also acknowledged being aware and accepting allowed them to connect with what was important in their lives in terms of the sense of self, but also the activities they wanted to do for a purposeful life. Meaning was still achievable despite the condition limitations and general life struggles. Many participants adopted a gradual and consistent approach to long-term management and fully acknowledged there would be uncertainty and fluctuations. Using softness and flexibility, participants were able to learn from their experience to reduce the negative impact of symptoms or relapses, for example remaining mentally well during these times.

Addressing mental and emotional needs was also important for long-term management and for some required additional support from others to achieve. All participants spoke about the need to accept support from others and highlighted the importance to communicate effectively with those around them. Some participants did so using various strategies such as metaphor and analogies attempting to create a shared understanding. As well as the use of discretion in some instances, as the need to disclose they had CFS/ME was not always necessary, particularly when the illness was not a central part of participants' identities.

4.2 Connection between Themes

As mentioned, the themes produced from the data were not mutually exclusive. Similar threads ran through all three themes which is not uncommon in qualitative research. Haig-Ferguson (2014) investigating the experiences of paediatric CFS/ME on the family experience, likened this joining together of themes as a '*jigsaw*' creating a complex but connected CFS/ME picture. It is important to acknowledge this process as it can be used as a strength for understanding CFS/ME; a heterogeneous chronic illness with so many individual facets that cannot be predicted or related to causation. It is also this appreciation of CFS/ME complexity that should be adopted in healthcare to be able to deliver relevant person-centred support throughout people's experience of living with CFS/ME.

A common thread among the data was the development of knowledge, skills and learning over time. This is of particular relevance to the current study aims that focused on exploring long-term management experiences. The discovery that participants specifically noted differences in their experience as time went on is of extreme relevance, and is discussed in more depth throughout.

Another thread was the expression and conceptualisation of individuality within CFS/ME. Participants used language to represent this expression explaining for them it was a particular way and acknowledging for others it might be different. Equally there were similar topics discussed across the dataset yet, participants had individual needs and views about them. This highlights the need to listen and support this individuality of experience and need for support. This thread is not particularly surprising and fits with wider health agendas to promote individual care and patient choice (DoH, 2004, 2012, 2013b), as well as treatment and management guidelines for CFS/ME specifically (NICE, 2007, 2014). It has also been a previously reported failing of health services that can cause detriment to those managing CFS/ME if it is absent from the professional relationship (McDermott et al, 2013).

The third common thread was the belief that long-term management required continual monitoring and balance. Also mapping onto other research findings advocating balance of activities and monitoring activities to assist in managing frustrations and help adjust expectations for a positive response to the impact of CFS/ME on activity (Drachler et al, 2009: Pemberton and Cox, 2014). Self-awareness supported participants in the present study to continually monitor their internal experiences which informed their decision making in relation to management appropriate to the situation. In addition to awareness, acceptance also appeared to facilitate participants' ability to prevent over or under activity facilitating adapting around the illness limitations, which is again consistent with other CFS/ME studies (Njoku, Jason and Torres-Harding, 2005). Commonalities can be drawn with applied research studies and service evaluations of CFS/ME that report patients positively live with CFS/ME through developing self-awareness and activity management strategies in combination (Coremans and Vause, 2013; Borseti, 2015; Densham et al, under review; Wearden et al, 2010).

4.3 Illness trajectory

The present study was unable to provide clarity as to whether timeframes or distinct phases of the illness occurred. That said participants did identify relapses (as an increase in CFS/ME symptoms) were naturally occurring fluctuations of the condition and understood to be the stages of CFS/ME. Relapse management was discussed in two ways; the first as preventative measures which fitted with a long-term strategy of condition management. Primarily this involved activity management approaches and continual monitoring to remain aware of experiences, linking to one of the common threads present within the study findings. The second element of relapse management was coping during a relapse, which was considered as short-term management. This involved targeting specific symptoms (like pain or fatigue) and remaining as physically, mentally and emotionally well throughout the relapse episode. This conceptualisation of CFS/ME fits with previous research and condition theories which

postulate management of CFS/ME through effective coping can lessen emotional distress whilst illness course may be unaffected (Garrett, 2001: McKenzie et al, 1995).

Reflecting on their long-term experiences participants in the present study identified other distinct short and long-term management strategies they used which are described below. In addition, the notion of short and long-term was also applicable for coping adopted that was more relevant at the start of the fatigue experience compared to later in their illness experience.

4.3.1 Short-term coping

Compared to the initial illness onset some aspects were less prominent as time went on, for example the extremes of energy usage. Many participants described a short-term way of coping with CFS/ME as following urges to do more when feeling well but then noticing a crash. This approach was in opposition to the long-term strategy of keeping a consistent routine and regulating energy and activity levels. Participants found when they followed urges to do more when feeling well or pushed on regardless, this fed into the boom and bust cycle of activity which ultimately reduced function. This has also been found in other studies commenting on general illness experiences in CFS/ME (Lombaard and Mouton, 2005: Jason, 1999 Pinxsterhuis et al, 2015:). In particular, McKenzie et al (1995) investigated coping and long-term CFS/ME and found few participants (3.9%) adopted this method of coping of pushing through symptoms and refusing to accommodate the illness. Interestingly of those that did, the majority of the participants reported their condition to be progressively worsening over time.

This fits with Maria's account of poorly managing her CFS/ME in this boom and bust pattern and ultimately she thought she had '*no life*' by trying to ignore her symptoms and just go to sleep. This process is comparable to denial, the opposite of acceptance, and has been linked to less adaptive outcomes in chronic fatigue and other health problems (Njoku, Jason and Torres-Harding, 2005). That said, denial has been thought to have a protective function to enable people to deal with health stressors (Kassebaum and Baumann 1965: Telford, Kralik and Koch, 2006) and stigmatisation of an invisible illness from others (Njoku, Jason and Torres-Harding, 2005), which is particularly relevant to CFS/ME. The present findings however suggest the idea of denial involving ignoring experiences was understood to be a short-term way of coping and largely ineffective. This is supported by Njoku, Jason and Torres-Harding (2005) who also suggest acceptance can facilitate help seeking behaviours, and therefore urge therapists to '*reduce denial and promote alternative strategies for managing life events*'

(p.263). Perhaps suggesting that focusing on acceptance-based models of health would be most advantageous to newly diagnosed groups.

Emotion-focused coping, whereby the emphasis is on regulating emotions following stressful events (Pinxsterhuis, Strand and Sveen, 2015) has been suggested to be more prominent in chronic fatigue (Blakely et al, 1991) and is present within current treatment approaches for CFS/ME, specifically CBT. The present study proposes that during a relapse emotion-focused coping can be advantageous, which was illustrated particularly by Kate stating being mentally well during a relapse was progress for her. That said, the present study also highlighted that this method of coping might be more of a short-term strategy most relevant to managing relapse. This idea is complimented by Krzeczowska, Karatzias and Dickson (2015) suggesting emotion-focused coping can be unhelpful, resulting in maladaptive responses to illness. Extending this argument, is the proposal that if emotion-focused coping is used repeatedly as long-term strategy it could direct focus away from other forms of coping. Njoku, Jason and Torres-Harding, (2005) claim that frequency and circumstance denotes coping as adaptive or maladaptive rather than the type of coping alone. Therefore indicating the need for various strategies and coping options for people with chronic and fluctuating conditions. This flexible approach has high face validity in managing the variable CFS/ME symptom experience and fit with the anecdotal accounts of the present study.

4.3.2 Long-term coping

As noted by Arroll and Howard (2013) a common experience for those with CFS/ME is to compare their previous life with their life after developing CFS/ME. Whilst references of this existed in the present study, only one participant described making attempts to keep up with the same level of activities they did prior to illness. The majority of participants' reported they'd slowed down and re-evaluated their priorities since having the condition. Pemberton and Cox (2014) also noted a similar shift; with one of their CFS/ME participants initially expressing a desire to return to a fast paced life however, during their second interview a year later, indicated a more meaningful slower paced life. This could indicate movement from wanting to resume the same pattern before illness to a more 'realistic' objective over time is a helpful attitude change in living with CFS/ME. Some of the participants in the present study also attributed these changes as being more age appropriate and many acknowledged the role of complicating factors like other health problems. This fits with previous qualitative studies suggesting comorbidities and age contribute to poorer outcomes in CFS/ME (Joyce, Hotopf and Wessely, 1997). That said, it seemed from the participants' accounts adopting a standpoint of recognising life changes overtime combined with the challenges of chronic illness, acceptance of these facts made it easier to adjust. This shift over time suggests the

difference between short and long-term management where participants explained how they strive to achieve a '*standard of living*' despite the CFS/ME symptoms. This process of adapting through gaining a broader perspective has been observed in other CFS/ME qualitative research; describing accepting, rebuilding identities and lives as psychological processes linked to adopting adequate coping strategies (Pinxsterhuis et al: 2015: Whitehead, 2006b).

The advantage of having CFS/ME for a longer period of time can enable self-reflection and re-evaluation of values and life choices (MacKian, 2000: Pinxsterhuis et al, 2015). This idea was supported by the participants in the current study reporting their acceptance and connection with what is important to them developed over time and was still possible despite the condition limitations. Specifically, learning from past experiences and drawing on self-awareness allowed this process to occur. In particular it was achieving a 'purpose' in life despite CFS/ME symptoms that gave participants a different perspective. This idea also fits with increasing prevalence of the four core concepts of occupation; *doing, being, becoming* and *belonging* (Hitch, Pépin and Stagnitti, 2014). These concepts are of extreme relevance to the processes described in the present study, as well as in other research findings illustrating how people understand and negotiate life with CFS/ME in relation to change or rebuilding their sense of self and connection with purpose (Dickson, Knussen and Flowers, 2008: Drachler et al, 2009: Wilcock, 1999: Wilcock, 2006: Whitehead, 2006b). It makes sense for individuals with CFS/ME experiencing 'chaos' (Anderson et al, 2012: Larun and Malterud, 2007: Whitehead, 2006a, 2006b) as the ability to 'do' activities is prevented, subsequently leaving people feeling lost and disconnected with the life they want or had. This was emphasised in several parts of the present study within Themes; Acceptance and Connection. Specifically thinking about how participants' viewed themselves in light of having CFS/ME on their ability to be true to their sense of self.

Changing the focus away from uncontrollable aspects such as the individual CFS/ME symptoms and towards what can be changed also fitted with the participants' accounts of effective long-term management. Accepting what could be changed and what couldn't was helpful in directing change efforts. Accommodated CFS/ME limitations but still providing meaningful opportunities was described in the context of employment and roles within relationships. As well as occupational links, this approach also fits with acceptance models of change (Hayes, Strosahl and Wilson, 1999: Segal et al, 2002: Strosahl, Robinson and Gustavsson, 2012). As reported by Krzeczowska, Karatzias and Dickson (2015) moving away from personal control towards improving functional ability in line with values is advantageous. This way of long-term coping is comparable to problem-focused coping. Whilst the initial stressful event of CFS/ME cannot be changed (which is often the objective of

problem-focused coping), there are other aspects identified within the challenges of CFS/ME that can be changed as a form of adaptive coping (Pinxsterhuis, Strand and Sveen, 2015). Examples of how participants employed such changes related to removing themselves from unsupportive friends, family and HCPs, changing jobs to suit their condition restrictions as well as trying out new activities.

Another theoretical proposal for the effective long-term coping discussed within participants' narratives was to remove the personal ownership of being ill within the participants' sense of self. This method is often used by people with chronic conditions to remove personal blame or ownership of the condition on identity. This is underpinned by self-as-context component with an ACT model whereby people are able to view themselves as separate from unpleasant symptoms, thoughts or feelings (Flaxman, Blackledge and Bond, 2011; Harris, 2008). Some clear discourse and body language was used by participants in the present study demonstrated the self-as-context such as Alison referred to her FMS as '*it loves warmth*' rather than saying alternative self-as-content phrases like '*I feel better when I'm warm*'. In a similar way of lessening or removing guilt or blame people feel for their illness, receiving a diagnosis of CFS/ME and recognising medical conditions has been reported as advantageous (Huibers and Wessely, 2006).

4.4 Comment on therapeutic mediators and mechanisms of change

Typically mediators, moderators and mechanisms of change are present in quantitative research that can statistically comment on the effects of defined variables following treatment. Yet often empirically tested concepts are derived from self-reported questionnaires attempting to access complex constructs such as mood, self-efficacy, personality traits (neuroticism), coping styles and so on. It's therefore argued that qualitative research can gather the same level of participant perception of such constructs.

When investigating mediators, moderators and predictors of CBT for chronic pain Turner, Holtzman and Mancl (2007) measured self-efficacy using unstandardised questions. They asked how certain participants can decrease their pain and control it from interfering with sleep and desired activities, as well as their abilities to improve their mood in the presence of pain. It is also argued that whilst no measure of effectiveness was objectively tested in the present study it did elicit the participants' appraisal of effectiveness and provided rich and contextual information that questionnaires and outcome measures simply cannot achieve. It is acknowledged the present study did not focus on a specific single treatment but had a more global view of treatments experienced. This may not be a negative point based on some suggestions that prescribed techniques in difference interventions operate using similar

processes and it is in fact the theoretical language that masks the similarities (Godfrey et al, 2007).

Using Wilt's (2012) definition 'A mechanism is defined as a step or series of steps in a process through which change is produced (p. 53) with each step in the process can be viewed as a mediator. With this in mind several potential therapeutic mediators and mechanisms of change were reflected in the present study's findings and arguably implicitly in the previous sections discussing short and long-term coping strategies.

4.4.1 Awareness as a foundation

There is no doubt a person has to negotiate life differently when living with a chronic illness (Kralik, van Loon and Visentin, 2006). The awareness theme appeared to be the foundation in facilitating participants to accept and connect with themselves and others. In turn, this level of awareness enabled participants to make changes that were beneficial for them in adapting and living with CFS/ME. This point is echoed by Strosahl and Robinson (2015) who state that change cannot occur without awareness. Equally findings are consistent with van Ravesteijn and colleagues (2014) who reported the role of MBCT in patients with MUSs assisted to increase self-care and self-compassion embodying awareness and acceptance of their chronic condition. Yet paying attention to experience is different to simply focusing on condition symptoms, which creates a narrow focus on symptoms alone and has been described as a maladaptive coping method, particularly unhelpful in CFS/ME when symptoms are unavoidable (Gray and Rutter, 2007; Njoku, Jason and Torres-Harding, 2005). This is further supported by researchers in the Netherlands reanalysing RCT data using CBT interventions for CFS/ME. The authors Wiborg et al (2012) concluded reduced focus on symptoms and perceived control of activities mediated outcome, rather than actual activity levels. Suggesting interventions that empower people with CFS/ME to be in control, compared to the condition viewed as being in control facilitates change. There is also an argument that focusing only on symptoms distracts the mind away from actions to help deal with the illness, as well as feeding into a pessimistic outlook and reducing self-efficacy becoming a vicious downward cycle (Njoku, Jason and Torres-Harding, 2005).

A caution could be expressed in facilitating self-awareness or specifically present-moment interventions (such as mindfulness, MBRS and MBCT) that by increasing people's awareness of their experience, specifically bodily sensations (like pain) could facilitate health anxiety. Whilst this concern has high face validity the opposite seems to occur; once present-moment awareness is developed people tend not use it as an experiential avoidance control strategy that it is sometimes viewed as (to turn off the mind/body), but as it is intended to undermine

experiential or fear avoidance (Flaxman, Blackledge and Bond, 2011; Njts et al, 2013). In addition, research has demonstrated such interventions are effective in treating health anxiety itself as illustrated by a recent Danish pilot study (Eilenberg et al, 2013). That said a recent systematic review investigating mind body interventions for FMS reported limited studies of low quality were available to comment of the effectiveness of mindfulness interventions for FMS, calling for more high quality research to be done (Theadom, et al, 2015).

Equally despite the immense challenges of living with CFS/ME, positive changes in identity were found by Whitehead (2006a) that were generated through the participants' reflections of their journey so far with CFS/ME and their experience before illness. Similarly some research has suggested that the traumatic event of CFS/ME can enable positive experiences to achieve a new 'true' self which is a useful (Arroll and Howard, 2013). Also notable shift in focus from 'doing' to 'being' was reported by Pemberton and Cox (2014) where a sense of appreciation of the lived experience was gained, such as interacting with others focusing on relationships and being present with nature. This appreciation seemed to be facilitated by focusing on the present moment which was not present prior to developing CFS/ME. Furthermore, outcomes have improved following present-moment interventions (Densham et al, under review), and application of these interventions are reinforced by high levels of patient satisfaction (AFMS, 2014; Dayes, 2010; BCFS, 2015, 2016). The present study therefore provides further support of present-moment interventions with people with CFS/ME as a potential mechanism of change.

4.4.2 Self-efficacy

Self-efficacy has previously been offered as a mediator for successful outcomes in CFS/ME management (Coremans and Vause, 2013; Findley et al, 1998; van Damme et al, 2006). Equally when considering common treatment approaches for CFS/ME, CBT specifically harnesses the use of patient's self-efficacy. This is in relation to building confidence in interrupting, challenging and controlling problematic thoughts and feelings. A strong link between self-efficacy and CBT would therefore be expected, and has been observed in CFS populations (Godfrey et al, 2007) and chronic pain (Turner, Holtzman and Mancl, 2007) describing self-efficacy as a key ingredient for change. Perhaps given the similarities in presentation of chronic pain patients and CFS/ME patients (Blakely et al, 1991; Densham et al, under review) it is not surprising that self-efficacy was also present in the current study narratives alluding to its role in promoting effective change. In particular self-efficacy was illustrated when participants' talked about having confidence to follow management strategies that were beneficial to them (including similar CBT assumptions or strategies to manage CFS/ME symptoms). For example saying no to commitments even though enjoyable if this

compromised their activity management (pacing, routine etc) and could be conflicting with effective long-term management. Arguably ACT interventions have an advantage over CBT interventions in this regard as ACT emphasises personal values to guide action despite unpleasant thoughts or feelings. Individuals who are able to be confident in *why* they are doing, or not doing certain activities because it is in line with what is important to them seems to have more potency, compared to CBT challenging or changing problematic thoughts or feelings as rigid personal or social beliefs.

4.4.3 Validation and understanding

As already presented there have been consistent reports within the CFS/ME literature illustrating the detrimental impact of others not understanding the condition leading to stigmatization and de-legitimation (Anderson et al, 2012; Asbring and Narvanen, 2003; Larun and Malterud, 2007; Raine et al, 2004). Interestingly one finding of the present study highlighted the participants had a great deal of empathy for others not understanding given the complexities and invisible features. This ability to empathise seemed to come from participants' own challenges of understanding and accepting the condition.

Yet it seemed that in order for the participants to understand and accept themselves, they required validation and understanding from others, and so based on the study findings a tentative relationship can be drawn. Validation and understanding appeared to be central in achieving acceptance of the condition restrictions and adapting to life with CFS/ME, in a way that worked for individuals. The role of acceptance (Brooks, Rimes and Chalder, 2001; Hadlandsymth and Vowles, 2009) has previously been offered as a mediator for successful outcomes in CFS/ME management and has been reported to help patients positively live with CFS/ME (Borseti, 2015; Chew-Graham et al, 2011; Densham et al, under review). It is thought that even when interventions are not explicitly using acceptance models (such as CBT), being able to facilitate acceptance has the potential to improve physical and social function and reduce fatigue (Brooks, Rimes and Chalder, 2011).

As already discussed denial appears to be a useful short-term strategy but used repeatedly does not contribute to effective long-term management for CFS/ME. Therefore facilitating acceptance of CFS/ME is advantageous in directing efforts towards change to improve function and wellbeing. Yet Telford, Kralik and Koch (2006) describe caution in using acceptance and denial labels proposing there can be negative associations of such labels that can interfere with adjusting to illness. Instead the authors urge HCPs to listen to individuals' stories instead of *'interpreting behaviour as denial and promoting acceptance as desirable end point'* (p. 463). This links to the findings of Ring et al (2005) who found that GP consultations

with individuals presenting with MUSs all gave cues to require emotional or psychological support yet more commonly GP's offered physical interventions such as medication, with a very low number (16%) of encounters found GP's to be verbally empathetic to the individual's symptomatic experience. The present study advocates the value of being able to share experiences in a professional setting, without the need of an in-depth therapy or intervention. This point gathers further evidence as participants explicitly stated that the interview itself was a helpful process to reflect on their experiences in a way not available to them in day to day life. This was depicted as an important part of long-term management to be able to receive have occasional contact with professionals (GP and specialist) to provide a space for validation as well as reminding them of helpful management approaches if appropriate. Therefore the current study extends this notion that reflection, validation and understanding is particularly important for effective long-term management (DoH, 2012: Gauntlett-Gilbert, 2015: Kralik, van Loon and Visentin, 2006), spotlighting the opportunity for HCPs to support this process in CFS/ME interactions.

When thinking about the client-centred approach to therapy advocated by Carl Rogers stating empathy and unconditional acceptance towards the patients helps form a collaborative and trusting relationship for the basis for change (Horvath and Luborsky, 1993). It is also this client-centred approach consistently encouraged in the delivery of health care, including CFS/ME (NICE, 2007). That said, when looking more closely at the role of working alliance and therapeutic relationship in CFS/ME research is limited. Findings of Heins, Knoop and Bleijenberg (2013) explored therapeutic alliance in CBT for CFS/ME and concluded the active ingredients were patient expectations of therapy and agreement of the content of therapy, influencing outcome. This is interesting when some research advocated the importance is placed more on the therapeutic bond, or relationship (Gauntlett-Gilbert, 2015). Heins, Knoop and Bleijenberg (2013) also reported outcomes were unaffected by illness duration therefore potentially relevant to those with CFS/ME for short and longer durations. This could indicate that it is less important who the HCP is, (or indeed anyone supporting the person with CFS/ME), but the more crucial factor is the agreement and shared focus. This has been reported in other studies in CFS/ME, and chronic illness more broadly if patients and therapists have a shared model of illness it can positively impact patient outcomes (Brooks, King and Wearden, 2014: Chew-Graham et al, 2011: Daniels and Wearden, 2011, Gauntlett-Gilbert, 2015). Together the idea of negotiated content or process of therapy and validation would be advocated by the present study. Without supportive environments people with CFS/ME can feel inhibited from disclosing their needs, as Stuart found reporting caution in disclosing his CFS/ME with others. This use of discretion conforms to other thinking that inhibition occurs

through fears of disbelief and stigmatisation (McInnis et al, 2015: Horton et al, 2010, Larun and Malterud, 2007: Tucker, 2004: Ware, 1999).

This value of HCPs listening to individuals' stories links with the present study when thinking about the importance of relevance and personal identification in treatment. Illustrated by Colin sharing his irritation with HCPs aiming CFS/ME treatment at women and found the therapist to be employing incorrect assumptions. Again this has been supported elsewhere (see Bayliss et al, 2014b: Drachler et al, 2009: Song and Jason, 2005) reporting the value and good practice of professionals that demonstrate a genuine interest in listening to people's experiences treating them as individuals.

Encouragingly none of the participants reported not being believed by others, although several reported some HCPs (specifically GPs) were unable to help them, leaving participants feeling alone to manage their condition. This was a particular barrier of long-term management identified in the present study, with regards to professional support. Whilst many participants acknowledged there may not be a solution available, they needed validation and understanding throughout their fatigue journey.

4.4.4 Uncertainty and acceptance

The issue of uncertainty within CFS/ME is rife. The findings of the present study found that ways participants overcame the uncertainty of CFS/ME symptoms and unknown prognosis, was to accept these aspects as simply that; 'unknown'. The threads within themes of Acceptance and Connection particularly fit with this varied and flexible response to the uncertainty and fluctuation.

Participants in the current study had accepted the diagnosis and through their experiences conceptualised the illness for them would be life-long. Alison did express a hope for a cure to be found, however this was directed towards others being able to get better in the future. This finding differs from McKenzie et al (1995) who reported 14 (5%) of their long-term CFS/ME participants stated they were hopeful to be cured. For the majority of the participants hoping to be cured subjectively reported their condition to be worsening overtime and only two reporting their illness course was improving.

There is an argument that it is not a continuum with acceptance of CFS/ME on one side and hope to be cured on the other, but that adapting and accepting is a continual process, which was an aspect highlighted by many participants in the present study as central to long-term management. This has been described by Garrett (2001) *Chronic illness is a form of suffering from which cure is unlikely but healing is possible* (p.99). When discussing the process of

healing in chronic illness she explains moving away from fear, anxiety, guilt and shame towards peace, joy and love in a process that is not achieved *at a single time, once and forever* (p.100).

Conversely it might be suggested for those who are resigned or fixed to one of these options (accepting or hopeful for cure) it may hamper physical or psychological progress. Hope to be cured could distract from effective management in the present (Njoku, Jason and Torres-Harding, 2005), and being totally accepting of the condition might prevent people from making beneficial changes. The former idea fits with the present study as Maria explained she was waiting to get better and she didn't. Suggesting this perception is a temporary coping method, unsuitable for long-term management. The latter proposal of being too accepting could be comparable to Jane's experience where she described getting into 'bad habits' and subsequently found it helpful getting specialist input. This finding highlights the importance of fostering a flexible attitude, in addition for HCPs and service provision to support individuals with CFS/ME throughout their fatigue journey, not just in earlier stages of diagnosis (Arroll and Senior, 2008).

4.5 Findings in relation to CFS/ME treatment approaches

4.5.1 Supporting existing guidance

The participants viewed stages of CFS/ME relating to relapses, with management including short or long-term strategies. This complements current treatment recommendations of including relapse planning in various types of interventions for CFS/ME. Specifically including the identification of triggering factors, temporarily reducing energy expenditure, increasing rest and relaxation and gradually building back energy levels (NICE, 2007). All of these concepts existed in the participant narratives, however compared to this current thinking, more emphasis was placed on the role of self-awareness in identifying (triggering factors and indication a relapse is occurring) as well as managing during the relapse itself.

Behavioural interventions have been shown to significantly improve outcomes for adults with CFS/ME (Baldwin, 2013; Chambers et al, 2006; NICE, 2007), which include reducing fatigue, functional disability, depression and anxiety. The interventions reported to be most effective are CBT, MBCT and GET. Findings of Wilson et al (1994), an Australian study investigated predictors of long-term outcome in CFS/ME and can help explain why behavioural interventions demonstrate more significant improvements. This is because the authors found whilst functional impairment remained present over time, illness beliefs and coping style

affected long-term outcomes compared to immunological or demographic variables. It is therefore proposed that behavioural based therapy targets unhelpful illness beliefs and coping styles, and/or facilitate more helpful illness beliefs and coping. This is echoed by the present study's findings; as all participants viewed CFS/ME as life-long for them, but by developing self-efficacy and broadening awareness helpful change was possible.

4.5.2 Interdisciplinary approaches

Throughout the interpretation of the present study findings a clear message of holistic treatment approaches are needed, where treatment approaches are aligned with patient priorities (NICE, 2007). As already discussed at length, the importance of people with CFS/ME to feel heard is vital for wellbeing and access to support, and is widely advocated (Anderson et al, 2012; Larun and Malterud, 2007; Coyne, 2016; NICE, 2007). Interdisciplinary working compared to multi-disciplinary working is where specialists with training and competence operate outside the usual discipline realms. This allows HCPs to respond to the complex experience of living with CFS/ME treating patients as a whole rather than whether it fits into the discipline's knowledge base. The call for this style of working fits with NICE (2007) guidelines and the self-expressed needs of people with CFS/ME voicing support to understand and manage their symptoms through control, collaboration and support (Drachler et al, 2009). It also attempts to overcome where *'a crude dualism pits biological against psychological models and the complex experience of living with it [CFS/ME] is often lost'* (p.2-3, Gotts et al, 2015). This also promotes flexibility in treatment, a key message highlighted by Horton et al (2010) capturing viewpoints of HCPs that were nominated by CFS/ME patients as clinicians providing helpful treatment or support. High levels of patient satisfaction and outcomes in a group setting have also been observed using these styles of support (Borseti, 2015; Densham et al, under review; Houlton et al, 2015; McDermott, Lynch and Leydon, 2011; Pinxsterhuis et al, 2015). However similar questions remain; what 'parts' of treatment are most effective, and for whom (Baldwin, 2013). The present study suggests more research is undertaken using this approach of combination of interventions often used in practice to be able to offer effective, individualised treatment and better understand mechanisms of change.

4.5.3 Application of acceptance-based models

The present study has also periodically referred to increasing theoretical utility of acceptance-based approaches in CFS/ME treatment and management, namely mindfulness and ACT. Mindfulness directly relates to the awareness theme participants expressed throughout the dataset as essential in CFS/ME long-term management. Mindfulness promotes observation of experiences and through this awareness comes the ability to look at thoughts as well as

from them (Hayes, 2016). The benefits of mindfulness on wellbeing lie in enabling individuals to disengage *'from automatic thoughts, habits, and unhealthy behaviour patterns'* (p. 823, Brown and Ryan, 2003). Present-moment techniques also clearly linked to the development of acceptance in the present study. Acceptance was also strongly constructed from the data in the present study allowing people with CFS/ME not to fight against the condition, as this was a short-term strategy that provided limited relief. That said, some caution has been noted in the use of mindfulness techniques, specifically that there are religious links that might conflict other religious or non-religious beliefs. There is occasional resistance in adopting mindfulness based approaches due to the strong link with Buddhism. However mindfulness frequently used in healthcare settings differs to meditation in Buddhism through the absence of ethical reflection (Stanley, 2013). There is also some thought suggesting similar practices to mindfulness such as emptying the mind actually pre-dates Buddhism (Manocha, 2014) and that other religions, spirituality and philosophies also adopt comparable present-moment contact or processes (Shapiro and Carlson, 2009). Flaxman, Blackledge and Bond (2011) indicate psychoeducation prior to doing experiential exercise can be useful in setting up expectations and purpose for exercises. It could also be suggested whether the mediation or present-moment exercises do or do not have any religious links, it may be advantageous to disclose or discuss this before and during therapy if it arises. Yet another line of argument that comes back to the idea of fostering an open, honest and collaborative therapeutic relationship.

The specific utility of acceptance-based models in CFS/ME can add to existing models of effective interventions (CBT/GET/Activity management). As demonstrated by Rimes and Wingrove (2011) improvements were found in their pilot study for individuals with CFS/ME whom previously had CBT treatment. This potentially suggests acceptance-based interventions could offer additional benefits to those who have had previous evidence-based treatments for CFS/ME but are still struggling. Equally the more evidence-based interventions available might assist with meeting the demand of tailored interventions that patients can chose for themselves (DoH, 2004: NICE, 2007). These proposals are complimented by the present study findings with regards to HCP support being valued throughout the fatigue experience.

There are some studies in other illness populations that suggest ACT may provide equally effective not superior outcomes; comparing CT and ACT in anxious or depressed participants. Forman et al (2012) concluded similar outcomes post intervention, but at long-term follow up 18 months later CT was more effective than ACT. Hunot et al (2013) reported limited differences between CBT and ACT interventions for depressed participants, however noted the *'quantity, quality and breath of available studies'* (p 2) limits the conclusions that can be

drawn, urging for more research look at effectiveness. It is also worth noting that traditionally outcomes assessed in quantitative studies are expected to improve; reduction in fatigue, depression, anxiety and disability. However this could be a potential flaw when comparing to acceptance-based interventions as the model fundamentally does not seek to change symptom severity or frequency but to facilitate valued action in the presence of symptoms (Hayes, Strosahl and Wilson, 1999; Luciano et al, 2014; Harris, 2008). It is therefore important to use appropriate outcome measures or assessments to correctly comment on the value of these approaches.

Despite questions over superiority of interventions, working on the assumption acceptance-based models are equally effective as current treatment approaches, it is of interest to consider that acceptance-based models in theory can overcome some of the criticisms of current behavioural CFS/ME interventions. For instance the pre-judgement of what thoughts or feelings are negative or problematic in CT and CBT, within ACT individuals define what is problematic for them and are encouraged to be less judgemental towards themselves (Harris, 2008; Hayes, Strosahl and Wilson, 1999).

Likewise typical CT approaches fail to attend to individual stories (van Houdenhove, 2002) considering more manualised treatment plans that focus on changing maladaptive thoughts to reduce frequency. Moving away from a 'control' strategy of CT can be advantageous for fluctuating chronic illness for which there is no cure, as there isn't a method to control of symptoms other than treatments targeting perpetuating factors (Coremans and Vause, 2013; Gray and Rutter, 2007; Harvey and Wessely, 2009; Lievesley, Rimes and Chalder, 2014). That said Forman et al (2012) propose that ACT is a counterintuitive approach for patients compared to CT, which is simple to understand with higher face validity in helping them reduce symptoms. This might be more fitting to their research population of depression and anxiety however, compared to chronic illness, where there is no cure and symptom reduction is not necessarily an achievable goal.

Mapping onto this, is the suggestion that tackling self-doubt and self-criticism for individuals with CFS/ME is a more potent clinical priority than 'high standard' personality types (Deary and Chalder, 2010). ACT offers an explicit self-compassion element towards thoughts and feelings unlike other behavioural interventions, where these are treated as maladaptive thought patterns requiring modification. Self-compassion was mentioned within several participant narratives when connecting with their sense of self which allowed for confidence and clarity amongst the chaos of living with CFS/ME. ACT harnesses the ability to translate what individuals already know to empower them to live full lives despite the challenges of

illness (Hayes, 2016) directly emphasising self-efficacy to address self-doubt and self-criticism.

There is also an argument that because a central part of CBT is to challenge negative thought patterns and emotion regulation, implementation requires cognitive and emotion energy. Perhaps this explains why CBT studies have reported higher drop-out rates or adverse effects in CFS/ME groups as the condition features makes this process even more challenging (Gladwell et al, 2014: Malouff et al, 2008: O'Dowd et al, 2006: Stubhaug et al, 2008: Twisk and Maes, 2009). Working on this assumption, energy needed to implement CBT techniques could be 'saved', or redirected towards more effective strategies that are more flexible and assessable from a behavioural component.

On the line of energy efficient interventions it is hard to ignore the similarities of the main themes reported in the present study and the recent model of Focused Acceptance and Commitment Therapy (FACT) (See Appendix D vi). FACT is a condensed model of ACT comprising of three pillars; open, aware and engaged which has been effective at promoting change in various healthcare settings (Strosahl, Robinson and Gustavsson, 2012: Strosahl and Robinson, 2015). Arguably FACT could be a more suitable intervention, more 'practical' help as patients want (Drachler et al, 2009) also fitting with some individuals not wanting purely psychological treatments. This is best illustrated by Diane in present study verbalising she didn't want '*psychobabble*' but practical help. Another practicality of FACT working well in fatigued populations is the intervention lends itself to be shorter and less intensive. Cognitive difficulties are common symptoms in CFS/ME (Fukuda et al, 1994: Sharpe et al, 1991 Söderlund, Skoge and Malterud, 2000) therefore participating in therapy that's shorter is not only is more efficient for individuals but healthcare providers. If this is possible using FACT, not only will patients receive a holistic, individually focused treatment that is able to work around the inherent barriers of CFS/ME (no cure, unknown aetiology and prognosis and condition symptoms), but it could provide a cost effective intervention that is economically viable (Strosahl and Robinson, 2015).

Yet a repetitive caveat is that the present study can only make tentative links with acceptance-based models, as there has been virtually no attention in CFS/ME populations, with no published research studies to date investigating FACT and MUSs let alone specifically CFS/ME. Promising results have been shown however in research looking at psychological flexibility like processes; mindfulness or present-moment focus, acceptance or openness to experiences and valued behavioural action (Densham et al, under review: Krzeczowska, Karatzias and Dickson, 2015: Luciano et al, 2014: Rimes and Wingrove, 2011: Shapiro and Carlson, 2009: Wetherell et al, 2011). However, more understanding about acceptance-based

interventions is needed, including long-term efficacy (Eilenberg, Kronstrand and Frosthalm 2013: Hayes et al, 2006: Hunot et al, 2013: Shapiro and Carlson, 2009).

4.6 Study strengths and limitations

4.6.1 Validity of findings

The researcher's clinical experience in fatigue management presented both strength and limitation, namely a subject bias when coding and creating themes. However, the content is deemed important to facilitate understanding of long-term experiences of managing CFS/ME which is an under researched area and study methodology is similar to other researchers working in this way (See McInnis et al, 2015).

Themes were created according to the sole researcher's knowledge, skills and experiences and as highlighted in the methodology chapter, invites criticism. No member checking was sought to mitigate this bias as objective analysis was considered impossible (Braun and Clarke, 2006: Tucker, 2004). However, the DoS provided counsel during the data analysis process and reflections were sought from one of the research Advisors. As a result, wording was changed on a poster created for an Educational Session for the BCFS team based on an Advisors feedback, the illustrative Acceptance Theme quote was shortened removing the preceding words *'it's always going to be with you you'll-'* this change allowed the content to remain true to the data, but for the purpose of the poster audience 'softened' the idea that people never recover from CFS/ME, causing any misinformation or upset. Feedback was also sought from eight members of an expert fatigue management team (BCFS) in the latter stages of theme interpretation in an attempt to broaden the researcher's analytical thinking. In addition, the researcher used guidelines outlined in Koch (1994) and Braun and Clarke (2013) as frameworks to challenge the integrity of the methodology and findings to subsequently strengthen conclusions drawn.

Additional attempts were made to ensure quality of the current study guided by Koch (1994). Credibility was actioned using a reflective diary. Transferability of the findings has also been highlighted indicating the specific participant population characteristics and only tentative links have been proposed to others with CFS/ME. Finally, Dependability was evidenced through keeping a coherent and systematic audit trail of the analytical process that could be audited by others.

4.6.2 Subjective appraisal of coping

Similarly to other small qualitative studies (See Pemberton and Cox, 2014) it is fully acknowledged no causal link can be attributed to the findings nor were there any objective measures looking a effectiveness of long-term management. As stated by Hareide et al (2011) *'Our interpretation is also based solely on the information conveyed in the interviews combined with our clinical impression, as no other assessments were used.'* (p. 2262). Yet the importance of this study lies in the relevance placed on the content of participant stories which were meaningful to them. From these accounts it is useful to draw comparisons to understand CFS/ME through those living with the condition. As discussed in the methodology, objective measures within quantitative research removes context and crucially personal meanings.

Despite the lack of objective measures, it was possible through participants' own admission and the researcher's clinical impression to suggest an appraisal of how successfully they were managing their condition using various strategies. This was broadly; those not adopting awareness, acceptance or connection with themselves and/or others, which conformed to poorer management and lifestyle quality. Using energy on fighting with the diagnosis and attempting to control aspects that were not able to be control bringing about a sense of low mood and disconnect with the life they wanted.

4.6.3 Population

All participants were recruited from a specialist NHS Service. Although none were active patients at time of the study it could be argued the findings are limited to a population that seek or have access to health care. This is also of relevance as help seeking behaviours have been associated with higher levels of acceptance in fatigue populations (Njoku, Jason and Torres-Harding, 2005) and so it might explain why acceptance-related code came across so strongly in the analysis. That said, simply because this association is feasible having research findings to support this theory is still of merit. In addition, the sample bias can be challenged by research from Anderson, Jason and Hlavaty (2014), who specifically investigated CFS/ME in a community sample due to the dominance of research using health care populations. Their findings were similar to those found in populations recruited from and research based on treatment delivered in specialist services. This is convincing that there might not be differences in these CFS/ME groups and subsequently reduces the weight of this particular bias. Furthermore, this study contributed to the calls for clinically representable populations to be the focus of research (Stiles et al, 2006).

As with many research studies investigating CFS/ME, severely affected individuals who are often bed or house bound were unable to participate in the current study. This limits the transferability of the study findings to other subgroups, because they were not represented in the current study. This issue is also reflected within practice as reported by a recent scoping survey of NHS Service provision for severely affected CFS/ME patients found limited and variable support for those with severe CFS/ME (McDermott et al, 2014). The issue prompts innovation and equality in the provision of care and access to participate in research, perhaps through alternative media. This is supported by a potential participant of the current study; who returned their invitation pack indicating they were interested in taking part but were unable to attend the hospital. They suggested a telephone interview instead. This was declined by the researcher based on the conditions of the ethical approval given and an attempt to keep data collection method consistent (face to face interview). This face to face method enabled unexpected interactions to be captured, as already discussed (the instances of embodiment during the interviews). If the interviews had been via the phone not only would this element have been impossible, but also the additional dimensions of body language would have been omitted. Research does however need to ensure people are able to take part, perhaps by travelling to participants, video calling instead of face to face interviews and the use of alternative data collection methods can assist with this aim of being more inclusive without being at the expense of data quality.

An attempt was made to promote a varied sample. However, ethnicity was incorrectly recorded on the database used for participant recruitment, resulting in all nine participants being white British. On reflection, this may have been an unavoidable issue as the geographic area where the research took place has a lack of ethnic diversity. According to the 2011 CENSUS 90.1 per cent of the local residents were recorded as white British, which is 10 per cent higher than the national average (BANES Council, 2016). The consequences of this sample bias means experiences of those who have had CFS/ME for a long time from different ethnic backgrounds were not represented in this study. The current study therefore suggests future studies conducting research in different geographic locations and using wider inclusion criteria to investigate where there are differences in experiences of managing long-term CFS/ME for different ethnic groups.

Similarly there were only three men in the present study and there were no participants that described themselves as transgender. A study flaw was that only 40 per cent of those asked to participate were men, a preventative measure could have been to invite 50 per cent or more men during the recruitment phase of the study. Men are a particularly under-represented group in CFS/ME research commonly justified by reported prevalence rates higher in women (Ax,

Gregg and Jones, 2001; Baldwin, 2013; NICE, 2007; Whiting et al, 2001). The current study did not report any significant gender differences within the participant narratives other than one interesting observation of language used by all three men. This was the explicit comparison of explaining how energy was continually monitored for health reasons, akin to being on a diet. It is of interest as the women in the study did not mention this analogy, nor did the women use a singularly common way of discussing their experiences. Whether the analogy of a diet was used because it represents a particularly meaningful concept for men or whether it performed a particular function to convey meaning with a female researcher, it is unclear. The study did not adopt discourse analysis nor a critical epistemology yet this difference in the expression of language would be worth expanding on by further research. In general, more research is required to represent men's experiences of CFS/ME to ascertain what similarities and differences exist (in use of language or otherwise), with a view to provide more appropriate support for men with CFS/ME, mapping onto the relevance of treatment threads found in the current study.

Diversity was however sought for marital status, recruiting married, divorced, cohabiting and single participants. This is another under researched area focusing on intimacy and relationships for those with CFS/ME, which undoubtedly is affected but perhaps gets less focus in research and potentially those with CFS/ME conceptualised as a less significant issue compared to traditional ideas of 'function'. Although marital status was recorded, participants were not asked about their sexual orientation and so it is unknown whether experiences of managing CFS/ME differ for lesbian, gay, bisexual or heterosexual people. Further exploration is needed to ascertain if similarities and differences exist and whether subsequent services or support is needed.

4.7 Implications of study findings

When considering adjustment to living with a chronic illness such as CFS/ME which is a fluctuating and poorly understood illness, there is immense value in providing a therapeutic space to be able to re-evaluate values and goals that are important to individuals. Considering practical support as well as more abstract emotional and psychological support as and when required was particularly relevant to effective long-term management of CFS/ME.

Relevance and timing of support was mentioned by several participants and so it is important to recognise the widespread fluctuations a person with CFS/ME will experience, not just in symptom experiences but in life experiences more widely. Having specialist support to manage throughout the illness experience is important to support people with CFS/ME,

specifically to enhance self-awareness, adopt an open and accepting attitude and strengthen connection with the self and others. This would also assist in individually tailored treatment accounting for personal objectives, social circumstances and comorbidities.

The results demonstrated a 'quilting' of interconnected threads among themes, so too is the recommendation for treatment approaches which can respond flexibly to individual need. It is proposed that drawing on an interdisciplinary model founded on genuine interest about the person with CFS/ME in relation to their experience and goals.

The value of talking and sharing experiences with healthcare professionals should not be overlooked. Other studies have suggested that over time effective treatment outcomes can diminish, particularly after a year (Forman et al, 2012) and calls for the effects of 'refresher' sessions to be investigated (Baldwin, 2013). Therefore, perhaps an intensive intervention may not be required, particularly after successful self-management have been adopted for some time. There could however, be some real mileage in having a space to receive compassion, validation and advice from professionals and this therapeutic space might provide a mediating factor for maintaining outcomes and personally defined '*standards of living*'.

4.7.1 Future research

Whilst this study has been able to share the valuable experiences of those managing CFS/ME over several years, it would be interesting to expand on the findings by investigating experiences of those with fatigue in the short-term as well as those that consider themselves to be recovered. Looking at commonalities and differences of these groups may enable more appropriate support to be made available, and whether any particular markers could be used to triage or signpost specific needs.

The findings clearly point to the use of acceptance-based models, in particular the role of Mindfulness interventions, ACT and FACT that promote change in a person-centred way which is unfazed by incurable and fluctuating conditions. However, unanswered questions remain on the effectiveness of these interventions in CFS/ME, the long-term effects of the approaches and patient acceptability (Hunot et al, 2013).

Fitting with the idea of combination treatment and holistic approach, investigating the role of interdisciplinary or combination intervention both quantitatively and qualitatively could help answer questions about short and long-term outcomes, combined with contextual information regarding patient experiences. Equally based on the present study, the updated SR (Baldwin, 2013) preceding the present study and other CFS/ME research recommendations the implications of having 'refresher' sessions following interventions needs further attention.

Tentatively it is suggested that long-term management the need for intensive interventions is not necessary, and whether 'lighter' or brief interventions would be more appropriate warrants further investigation.

5 Conclusion

Living well with chronic illness is firmly rooted in the field of health psychology, with behaviour and management being central to preventing and managing chronic health conditions (Matarazzo, 1982). The value of this research was to expose the currently unheard experiences of people living with CFS/ME for a considerable number of years'. An attempt to draw out transferable patterns of experiences that can help understanding of what people go through living with CFS/ME and how healthcare professionals can use this knowledge to best support people with CFS/ME particularly when '*for many CFS patients, long-term adaptation may require therapeutic attention*' (p 497, van Houdenhove, 2002).

The present study supports the known challenges of managing CFS/ME, in terms of variable symptoms, unknown cause and illness trajectory and the lack of understanding of others. That said, different accounts regarding others was reported; where participants empathised with others for not understanding and reported that others could visibly see their condition. These findings contradict some previous research that conceptualises others conceptualising CFS/ME as an invisible illness met with stigmatisation and de-legitimisation.

No clear stages of CFS/ME were found based on the long-term experiences of those with CFS/ME, however the fluctuating nature of relapses was conceptualised as stages requiring short-term management to overcome the relapse and long-term management to reduce the likelihood of occurrence.

The findings compliment the use of current behavioural treatments for the management of the condition to reduce the impact on a person's life by targeting the perpetuating factors of the condition. The study also echoes the theoretical value of using alternative treatment approaches based on acceptance-based models of change. These model aims appear to align with the effective long-term management strategies employed by people who have lived with CFS/ME for several years and so therapeutic use could make a difference to people sooner and throughout their illness experience, improving their quality of life in response to condition symptoms.

The overarching themes identified the chaos that is experienced with CFS/ME which significantly impacts on a persons' life resulting in complete lifestyle re-evaluation. It is also important to foster a sense of purpose, contribution and valued living despite CFS/ME symptoms, working within the condition limitations rather than against it. However, it is noted as an ongoing process of monitoring, flexibility and softness.

The findings map directly onto the three pillars of FACT, suggesting a potential cost effective treatment warranting further exploration in CFS/ME. As well as further research to determine more concrete conclusions regarding mediators and mechanisms of change within an interdisciplinary approach to CFS/ME treatment. It is these flexible and holistic approaches that can theoretically best serve the needs of CFS/ME patients (Drachler et al, 2009), facilitating the known moderators and mechanisms of change; self-awareness, self-efficacy, re-evaluating priorities and working on personal goals or objectives in a way that patients are listened to, understood and treated in a person-centred approach.

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Appendices

Appendix A - Systematic review (Baldwin, 2013)

Behavioural interventions for the treatment and management of CFS/ME in adults (Review)

Baldwin, DS

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[Intervention review]

Behavioural interventions for the treatment and management of CFS/ME in adults

Deborah S Baldwin¹

¹ School of Psychology, University of the West of England, Bristol, UK.

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ABSTRACT

Background

Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME) is a disabling long-term condition, characterised by extreme fatigue not alleviated by rest and accompanied by other symptoms such as myalgia, poor sleep and concentration problems (Fukuda 1994: Sharpe 1991). A systematic review appraising all available treatments for CFS/ME was performed by Bagnall and colleagues in 2007. Based upon the publication of those findings and the recommendations of the National Institute for Health and Clinical Excellence (NICE) (2007) guideline it was concluded behavioural interventions are beneficial for treatment of CFS/ME. Since that time, many more studies have been conducted, therefore an update to review the available evidence for behavioural interventions is needed and specifically evaluating the long-term effects of behavioural treatments.

Objectives

To review available evidence to update the knowledge on the effectiveness of behavioural interventions for CFS/ME in adults compared to usual care, and to draw conclusions on the long-term effectiveness of these interventions.

Search Methods

Ten electronic databases were searched (AMED, BNI, CINAHL, Cochrane, Embase, Index to Thesis, Medline, PsychINFO, Pubmed and Science Direct) in April 2012 using a comprehensive search strategy. Citation searches were conducted using reference lists of relevant studies and reviews returned from the search.

Selection criteria

Randomised controlled trials (RCTs) were included in the review if they were published after Aug 2005, investigated the effects of behavioural interventions for adults (≥ 18 years) with CFS/ME as their primary diagnosis.

Data collection and analysis

After abstracts and full papers were independently reviewed by two reviewers, data extraction, quality assessments and risk of bias were rated by the author and checked by an independent reviewer. Included studies were analysed narratively given the heterogeneous nature of the included studies.

Main results

This review identified a total of 11 trials (1958 participants) for inclusion. Behavioural interventions reported statistically improved fatigue, functioning, depression and anxiety for adults with CFS/ME. Specifically cognitive interventions and GET were effective at improving fatigue, functioning depression and anxiety, supportive (non-directive) interventions effective at improving depression but had significantly worse effect on functioning, Pragmatic Rehabilitation (PR) significantly improved fatigue and relaxation had beneficial effects on anxiety. Of the significant effects found, 29% of fatigue, 60% of functioning, 50% of depression and 75% of anxiety were found at long-term follow up.

Author's conclusions

The majority of papers were of good quality; with some issues surrounding feasibility of blinding therapists and participants, limited adverse event reporting and information why eligible participants' refused to take part. The findings of this updated review have provided additional support that behavioural interventions are beneficial for CFS/ME. With long-term effects reported for functioning, depression and anxiety. Fatigue however reported more frequently short-term effects only. Further research is needed to draw firm conclusions on what type and aspects of behavioural interventions are effective, the effects for different subgroups of CFS/ME, namely severity and illness duration and the potential impact of refresher sessions on long-term effects of treatment.

PLAIN LANGUAGE SUMMARY

The effect of behavioural treatments for CFS/ME

Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME) is a long-term condition when individuals experience extreme fatigue and other symptoms for longer than 6 months,

for which no cause can be found. This review aimed to update knowledge on effective treatments for CFS/ME, looking at behavioural treatments compared to usual care. Behavioural treatments included various cognitive, exercise, pacing, supportive, relaxation and combined therapies commonly used in the treatment and management of the condition. Specifically fatigue, functioning, depression and anxiety were investigated. This review included 11 studies, with a total of 1958 CFS/ME participants. The review found that participants receiving behavioural therapies experienced improvements in fatigue, functioning, depression and anxiety compared to participants receiving usual care. Long-term benefits were found over 12 months for functioning, depression and anxiety for those attending behavioural therapies. For fatigue, short-term benefits were more common than long-term benefits after behavioural therapies but still reported better results than the usual care participants. Cognitive and exercise therapies showed greatest improvements for all four domains which was similar to previous review findings. Most studies were of good quality but few failed to report some details like why eligible participants did not want to take part and if any adverse effects happened during or after the behavioural treatments. Few studies looked at the effects of combining different behavioural therapies, which could provide greatest improvements both in the short and the long-term. More studies should investigate the effects of re-fresher sessions for long-term effects, the combination of behavioural therapies and to look at different CFS/ME severities and illness duration.

BACKGROUND

Description of the condition

Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME) is a long-term condition whereby disabling fatigue (both physical and mental) occurs without presence of disease lasting over 6 months (Fukuda 1994; Sharpe 1991). Common symptoms that accompany the overriding symptom of fatigue include: post exertion malaise, poor, disturbed and un-refreshing sleep, myalgia, concentration and memory difficulties, headaches, swollen glands and persistent sore throats (Fukuda 1994). Co-associated disorders that are common with CFS/ME are depression, Irritable Bowel Syndrome (IBS) and Fibromyalgia (Maes 2011; Whitehead 2002). Diagnosis relies on patient reported symptoms and negative blood screens to exclude other reasons for fatigue (Chambers 2006; Fukuda 1994). Recovery is poor if treatment is not sought and so effective treatment is essential to improve prognosis (Cairns 2005).

Description of the intervention

Following the recommendations of the NICE (2007) guidelines Cognitive Behavioural Therapy (CBT) or Graded Exercise Therapy (GET) are effective treatments for CFS/ME (Chambers 2006: Price 2009: Whiting 2001). Behavioural interventions have shown most beneficial effects without harmful side-effects (Chambers 2006). Documented behavioural interventions for CFS/ME include: cognitive therapy, CBT, mindfulness-based cognitive therapy (MBCT), GET, Adaptive Pacing Therapy (APT) and modified CBT (including counselling or occupational therapy) (Bagnall 2007: Malouff 2008: Price 2009: Whiting 2001).

How it might work

Cognitive therapies and cognitive behavioural therapies aim to identify and challenge unhelpful thoughts that reinforce problematic behaviours (Price 2009). More recent 'third wave' of cognitive therapies such as Mindfulness-based Stress Reduction (MBSR) (Kabat-Zinn 1990) and Mindfulness-Based Cognitive Therapies (MBCT) (Segal 2002) aim to increase awareness to the present moment, to observe and accept non-judgementally rather than replacing thoughts, feelings and bodily sensations, to prevent further struggle and discomfort in trying to change them (Bohlmeijer 2010: Hayes 2004: Price 2009).

Exercised focused interventions aim to systematically increase a specified amount or time spent exercising over time to improve strength, cardiovascular function, exercise tolerance and psychological status (Edmonds 2010: Fulcher 1997).

Other behavioural therapies that have shown to be helpful in CFS/ME are pacing (activity management), relaxation and counselling. These use a variety of techniques to improve quality of life (QoL) such as activity diaries, sleep hygiene, breathing techniques, guided imagery and active listening (Bagnall 2007: Goudsmit 2012: NICE 2007: Thomas 2006).

Why is it important to do this review

Little is known about the aetiology of CFS/ME and currently the only available evidence base for effective management of the condition is through pharmacological and behavioural treatments (NICE 2007). Effective treatment is essential to improve prognosis, provide long-term benefits, improve QoL, aid return to employment (reducing the need to seek government financial support) and to look at reducing the cost for the health service. Since the publication of the NICE guidelines over 5 years ago many more studies have been conducted, therefore an update to review the available evidence for behavioural interventions is needed and to evaluate the long-term effects of such treatments.

OBJECTIVES

To evaluate the effects of behavioural treatments specifically focusing on fatigue, functioning, depression and anxiety outcomes, and based on findings of the previous review to comment on the long-term effects of behavioural interventions for adults with CFS/ME.

METHOD

Types of Study

Randomised controlled trials (RCTs) published or unpublished in any language after August 2005.

Types of participants

Adults (≥ 18 years) with a confirmed primary diagnosis of CFS/ME. Studies involving comorbid physical or mental disorders were eligible for inclusion providing this was secondary to the CFS/ME diagnosis. Psychiatric diagnosis of substance-related disorder, schizophrenia or psychotic disorder were excluded.

Types of interventions

Intervention

Included studies need to evaluate and report the effect of one or multiple behavioural interventions. Intervention inclusion was not specific to a particular setting, length, intensity or delivery. Pharmacological, immunological, complementary therapies and supplement treatments were excluded.

Control

Control groups were defined as care as usual, which included GP care or waiting list conditions.

Types of outcome measure

The patient reported outcomes of interest were:

1. Measures of fatigue
2. Measures of daily functioning (including physical, mental, social functioning)
3. Measures of depressions
4. Measures of anxiety

Search methods for identification of studies

Electronic searches

The search strategy was applied to 10 databases (AMED, BNI, CINAHL, Cochrane, Embase, Index to Thesis, Medline, PsychINFO, Pubmed and Science Direct) in April 2012.

Condition terms:

- CFS
- Fatigue Syndrome, Chronic
- Chronic Fatigue Syndrome
- Myalgic Encephalomyelitis
- CFS/ME
- ME/CFS

Intervention terms:

- Behavior therap*
- Behaviour therap*
- Psychological therap*
- third wave
- Adaptive pacing therap*
- APT
- Pacing
- Pacing therap*
- Cognitive behavior therap*
- Cognitive behavioural therap*
- CBT
- Graded exercise therap*
- GET
- Acceptance and commitment therap*
- ACT
- Mindfulness
- Goal setting
- intervention*

Searching other resources

Citation checking of relevant studies and reviews returned from the search for potentially relevant RCTs.

Data collection and analysis

Studies were retrieved in full if the inclusion criteria were met in the abstract, or if it was unclear the full paper was required to confirm. Two reviewers (DB & DR) independently screened the returns using the inclusion criteria at abstract and full paper stages. All disparities were resolved between the two reviewers without the need of a third reviewer. Author DB extracted data using a standardised sheet specifically for this review which included: study, participant and intervention characteristics, outcomes used, results, potential sources of bias, dropout rates and adverse events. Author DB assessed the quality of the included RCTs using the JADAD appraisal tool (Jadad 1996).

Assessing risk of bias

The following were assessed using criteria from the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011):

- Random sequence generation
- Allocation concealment
- Blinding
- Incomplete data
- Selective reporting

Data extraction, quality appraisals and assessment of risk were checked for accuracy and consistency by the second reviewer (DR).

Measures of treatment effects

Determined by the study characteristics (namely type of behavioural intervention and outcome measures used) returned from the search. If heterogeneity of studies is low, meta-analysis will be performed for behavioural interventions compared to control groups for each of the outcomes of interest. Conversely if the studies differ considerably the included studies will be analysed narratively. Treatments will be considered effective if over 50 per cent of conditions showed significant improvement in fatigue, functioning, depression and/ or anxiety.

RESULTS

Description of studies

Table 1: Characteristics of included studies

Study	Setting	Sample Size & characteristics	CFS/ME criteria used	Conditions				Outcomes	Baseline differences	Results	Drop-out rates	Jadad score
				Intervention group(s)	Length of intervention	Control condition	Length of follow up					
(1) Knoop 2008	Tertiary care (Netherlands)	n = 166	CDC/ Fukuda et al 1994	Self-guided CBT instruction booklet	> 16 weeks (email/ Tel contact fortnightly)	Yes - WL	CBT: 10.5 months (93%), WL: 9.7 months (95%). No sig difference in time of f/u assessment.	CIS-F, MOS-SF-36 & SIP-8	None reported	CBT 'sig' less fatigued than WL (38.9 vs. 46.4 mean scores). CBT reported fewer disabilities than WL (1079 vs. 1319 mean scores). CBT 'sig' higher for physical functioning than WL (65.9 vs. 60.2 mean scores). CBT showed clinically sig improvement in fatigue at f/u (27% vs. 7%). No p values given.	CBT: 7% (n = 3), WL: 5% (n = 4) Total: 4.2%.	1

(2) Burgess 2011	NHS Research & Treatment Units (UK - London)	n = 80 Mean age of 37.42 years, 79% female, 40% were married, 90% of participants were white & 3.9 years mean disease duration.	CDC/ Fukuda et al 1994 & Oxford/ Sharp et al 1991	F2F CBT	15 sessions (first 2 sessions 1.5 hours, subsequent sessions 50-60 minutes)	No	3, 6 and 12 months.	MOS-SF-36, ChFS, WSAS, HADS & GI	Anxiety scores statistically sig for F2F CBT group (p = 0.046)	No sig increase in fatigue scores over time for either CBT condition (all p values > 0.05). No sig difference in fatigue between conditions (p = 0.19). sig reduction in impairment (p = 0.013) and increase in physical functioning (p = 0.043) at 12 months f/u. Baseline physical functioning predicted later physical functioning scores (p < 0.001).	F2F: 34% (n = 12) Tel: 56% (n = 25). Total: 46.3%. No sig difference in drop outs between groups (p > 0.05). Participants that were employed were more likely to dropout a post treatment and follow up assessments (p=0.003).	2.5
				Tel CBT	14 sessions (first session F2F for < 3 hours followed by 13 subsequent 30 minute telephone calls).							

(3) Jason 2007	Nurse delivered (in a hospital setting?) (USA - Chicago, IL)	n = 114, mean age of 43.8 years, 83% female, 88% Caucasian, 4% African- American, 4% Latino and 4% Asian- American, 49% were married or cohabiting.	CDC Fukuda etal 1994	CBT	13 sessions (although varied), lasting 45 minutes	No	12 months	MOS- SF-36, FSS, BDI-II, BAI, QoL & GI	None reported	No sig improvements in fatigue scores were observed (p > 0.5). Sig reduction in fatigue for COG (p =0.01). Sig improved depression for COG. Sig improved anxiety at 12 months for Relax.	Total: 25%, not sig different per condition (p value not given).	2
				COG	13 sessions (although varied), lasting 45 minutes							
				ACT	13 sessions (although varied), lasting 45 minutes							
				Relax	13 sessions (although varied), lasting 45 minutes							

(4) Nunez 2011	Chronic Fatigue Unit - Hospital setting (Spain)	n = 120 Intervention group :mean age of 42.65, 91% female & 32 months mean disease duration. Control group: mean age of 44.27, 84% female &f 33 months mean disease duration	CDC/ Fuku da et al 1994	Intervention group: MD group (CBT+ GET+ GP)	CBT: 9, 90 min sessions over 2.5-3 month & GET: three times a week, 1 hour sessions carried out for periods of 10 mins for 3 months.	Yes - CaU & exerc ise couns elling (3 daily 10- min sessi ons in separ ate perio ds with adapt ed aerob ic exerc ises & stretc hes)	12 months	MOS- SF-36, HAQ, HRQoL, HADS & FIQ.	None reported.	No statistically sig improvements in fatigue scores (p > 0.5). Intervention sig lower physical functioning (p = 0.004) (due to increased pain and weakness). Sig difference in physical functioning scores at 12 months between conditions (p = 0.040).	Intervent ion: 3% (n = 2) Control: 5% (n = 3) Total: 4.2%	3
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(5) O'Dowd 2006	Health psychology department for the management of chronic illnesses in a general hospital (UK - Bristol)	n = 153, mean ages: CBT:41.6, EAS: 38.8 and StMC 42.9 years, 67% female (N=102) & 47% had symptoms >60 months	CDC/ Fukuda et al 1994	CBT (including graded activity scheduling and stretches taught) EAS	8, 2 hour sessions, fortnightly (over 16 weeks) 8, 2 hour sessions, fortnightly (over 16 weeks)	Yes – GP/ StMC	6 and 12 months	MOS-SF-36, GHQ, ChFS, HADS & a cognitive battery	More men in the CBT condition. Fewer patients in EAS group had baseline score within the normal range for physical and mental functioning scores.	Sig improvement in fatigue scores for the StMC group between 6 and 12 month f/u (p = 0.024). CBT group had better fatigue scores than EAS (p = 0.011) and StMC (p = 0.027). sig improvement for anxiety in CBT group (p = 0.045). sig increased mental functioning for CBT (p = 0.019). No sig difference in physical functioning between 6 and 12 months (p = 0.038) or between groups (p = 0.36). No sig changes in depression (p < 0.05). sig improvement in anxiety for CBT (p = 0.45).	CBT: 30% (n = 13) EAS: 8% (n=2) SMC: 14% (n=7) N=27/153 (17.6%)	3.5
(6) Rimes 2011	Specialist NHS CFS Unit (UK-London)	n = 35, 83% female (N=29), Mean age - intervention: 41.4 years control:	CDC/ Fukuda et al 1994 & Oxford/ Sharp et	MCBT	9 sessions lasting 2.25 hours	Yes - WL	2 and 6 months (6 month - MBCT data only)	ChFS, WSAS, PF-10 & HADS	None reported	MBCT had sig improved fatigue (p = 0.014), improved impairment (p = 0.040), lower depression scores (p = 0.038) at post treatment. MBCT had sig improvement in fatigue (p = 0.033) at 2 months and sig improved impairment (p = 0.004) at 6 months.	MBCT: 12.5% (n=2) WL: 0% (n = 0) Total: 5.4%	3

		45.2 & illness duration - intervention: 8.5 years control: 6.1 years.	al 1991									
(7) Stubhaug 2008	Specialist clinic at Study Hospital (Norway-Bergen)	n = 72, mean age: 46.32, 82% female (N=59)	CFS/ME criteria: CDC/Fukuda et al 1994 & Oxford/Sharpe et al 1991 Neur	Drug+CCBT	Mirtazapine (15-45mg) for 24 weeks, after 12 weeks of starting the drug a CCBT programme started consisted of 2, 1.5 hour sessions a week for 12 weeks.	Yes - Placebo+CCBT	12 weeks (mid-point) and 24 weeks (post treatment).	ChFS, SHCI, MOS-SF-36, CGI & HRSD.	None reported	sig improvement in fatigue for CCBT compared to medication (drug/placebo) at 12 weeks (p = 0.014). sig improvement in fatigue for Drug+CCBT at 24 weeks compared to Placebo+CCBT (p < 0.001). sig improvement in mental functioning for Drug+CCBT at 24 weeks (p = 0.045). sig CGI for CCBT compared with medication at 12 weeks (p = 0.001). Significant CGI improvements for Drug + CCBT at 24 weeks (p < 0.001). sig subjective improvements at post treatment (p = 0.001) but not between group.	Drug+CCBT: 24% (n = 6), Placebo+CCBT: 8%(n = 2), CCBT+drug: 27% (n = 3) CCBT+Placebo: 8% (n = 1) Total: 16.6%	5

			asthenia criteria: ICD-10 research criteria.	CCBT+ Drug	CCBT programme started consisting of 2, 1.5 hour sessions a week for 12 weeks followed by 12 weeks of Mirtzapine (15-45mg).	Yes - CCBT +Plac ebo						
(8) Tummers 2010	Tertiary treatment facility for CFS/ME (Netherlands - Nijmegen)	n = 169, CBT: Mean age 37.6 years, 82% female (N=69) & illness duration: 72 months. CaU: Mean age: 38.5 years, 75% female	CDC/Fukuda criteria et al 1994	Self-guided CBT instruction booklet & additional CBT if needed	Booklet: > 16 weeks (email/tel contact fortnightly) Additional CBT: 14 sessions over 6 months	Yes - CaU	6 months	CIS-F, SIP & MOS-SF-36 (Physical functioning subscale).	sig difference in SIP score for CBT group.	No sig differences in fatigue scores found between CBT and CaU ($p > 0.5$). No sig differences for disability and physical functioning found between CBT and CaU ($p = 0.77$ and $p = 0.72$).	Stepped care: 14% (n = 12) and CaU: 7% (n = 6)	2

		(N=64), & illness duration: 96 months.										
(9) Tummers 2012	Regional community-based mental health centre (Netherlands - south west)	n = 123 CBT: mean age 36.3, 74% female (n = 46) & 48 months illness duration. Control: mean age 36.4, 82% female (n = 50) & 60 months illness duration.	CDC/Fukuda et al 1994	Self-guided CBT instruction booklet	Booklet: > 16 weeks/20 weeks (email/ Tel contact fortnightly)	Yes - WL	> 6 months follow up (patient delay in returning questionnaires)	CIS-F, SIP & MOS-SF-36 (physical & social subscales) & BSI.	None reported	Sig improvement in fatigue for CBT at f/u (p < 0.01). No sig improvements in physical (p = 0.08), social (p = 0.16) functioning or psychological distress (p = 0.30) between conditions.	Intervention 11% (N=7) and control 8% (N=5). Total: 9.8%	2.5

(10) Wearden 2010	GP surgeries/ Home visits (UK - north west England)	n = 283, 78% female, mean age: 44.6 & 7 years median illness duration.	Oxford criteria/ Sharp et al 1991 and London ME criteria	PR	10 sessions over an 18 week period, consisting of 5 home visits lasting 60- 90 minutes and 5 telephone calls lasting 30 mins.	Yes - GP treat ment	20 and 70 weeks	MOS- SF-36, ChFS & HADS	None reported	sig improvement in fatigue scores for PR compared to GP at 20 weeks (p = 0.021) but not maintained at 70 weeks (p = 0.076). SL sig worse physical functioning compared to the GP at 20 weeks (p = 0.005). No sig differences physical functioning found at 70 weeks (p > 0.05). sig improvements in depression for SL compared to GP at 20 weeks (p = 0.018), but not maintained at 70 weeks (p = 0.629).	PR: 15% (n = 14), SL: 11% (n = 11), GP: 14% (n = 14) Total 13%	3
				SL	10 sessions over an 18 week period, consisting of 5 home visits lasting 60- 90 minutes and 5 telephone calls lasting 30 mins.							

(11) White 2011	NHS Centres (UK)	n = 641, mean age: 38 years, 77% were female (N=495), 93% white ethnicity (N=595) & 32 months mean illness duration.	Oxford criteria/ Sharpe et al 1991 and London ME criteria	APT	< 14 sessions offered during the first 23 weeks the first 4 were weekly the subsequent sessions were fortnightly . An additional booster session offered at 36 weeks. Mostly face-to- face sessions although some were by phone.	Yes - SpMC > 3 sessions during 12 months	52 weeks	ChFS, MOS- SF-36 (Physical subscale), GI, WSAS & HADS.	None reported	CBT and GET sig improved on fatigue compared to APT (p = 0.0130 & p = 0.0294) and the SpMC (p = 0.006 & p = 0.0013). CBT & GET sig improved physical functioning compared than SpMC (p = 0.0342 & p = 0.0025) and decreased impairment (p = 0.001 & p = 0.0006) compared than SpMC. CBT & GET sig improved physical functioning compared than APT (p = 0.0012 & p = 0.0002) and decreased impairment (p = 0.0001 & p = 0.0004) compared than APT. The majority of participants reported 'minimal' GI over time for all conditions. CBT & GET sig improved for depression compared to SpMC (p = 0.0003 & p = 0.0035). CBT sig improved for depression compared to APT (p = 0.00382). CBT & GET sig improved for anxiety compared to SpMC (p = 0.003 & p = 0.0142).	SpMC: 2% (n =3) APT: 1% (n = 1) CBT: 4% (n = 6) GET: 1% (n =1). Total 1.7% Drop-outs did not sig differ between groups (p = 0.5)	2
				CBT								
				GET								

Table 1 Abbreviations' Key:

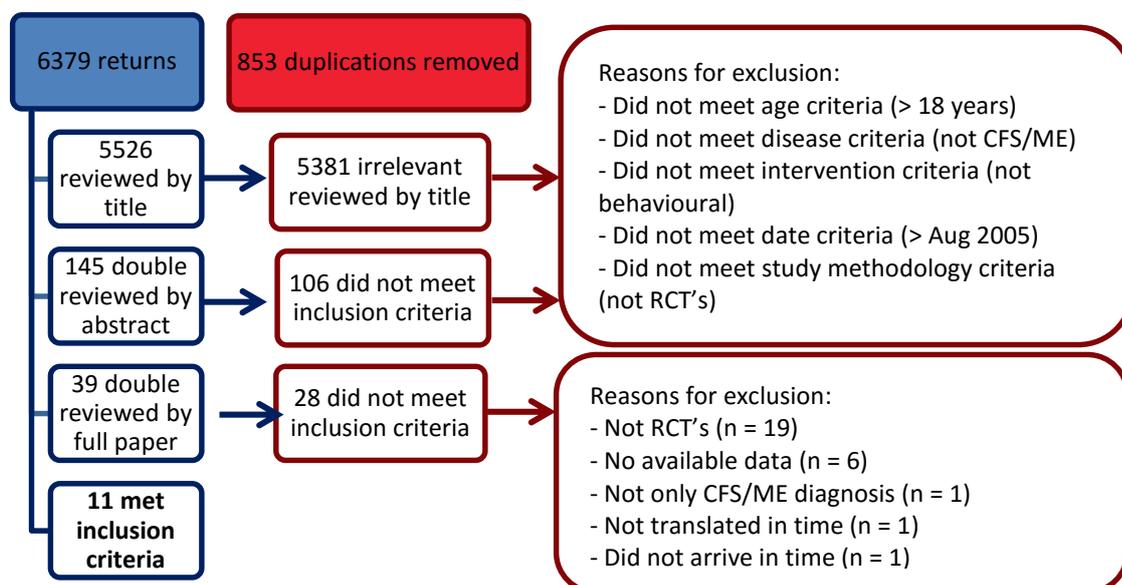
ACT	Anaerobic Activity Therapy
APT	Adaptive Pacing Therapy
CaU	Care as Usual
CBT	Cognitive Behavioural Therapy
CCBT	Comprehensive Cognitive
	Behavioural Therapy
COG	Cognitive Therapy Treatment
EAS	Education and Support
f/u	Follow up
F2F	Face to Face
GP	General Practitioner
MBCT	Mindfulness-based
	Cognitive Therapy
MD	Multidisciplinary
Mins	Minutes
PR	Pragmatic Rehabilitation
Relax	Relaxation Training
Sig	Significantly
SL	Supportive Listening
SpMC	Specialist Medical Care
StMC	Standard Medical Care
Tel	Telephone
WL	Waiting list

Table 1 Outcomes Key:

BAI	Beck Anxiety Inventory
BDI-II	Beck Depression Inventory II
BSI	Brief Symptom Inventory
ChFS	Chalder Fatigue Scale
CIS-F	Checklist Individual Strength: Fatigue subscale
FIQ	Fatigue Impact Scale Questionnaire
FSS	Fatigue Severity Scale
GHQ	General Health Questionnaire
GI	Global Improvement
HADS	Hospital Anxiety and Depression Scale
HAQ	Stanford Health Assessment Questionnaire
HRQoL	Health related Quality of Life
HRSD	Hamilton Rating Scale for Depression
MOS-SF-36	Medical Outcomes Survey: Short Form
PF-10	Physical Functioning
QoL	Quality of Life
SHCI	Subjective Health Complaint Inventory
SIP-8	Sickness Impact Profile: version 8
WSAS	Work and Social Adjustment Scale

Result of the search

Figure 1: results following application of search strategy.



Included studies

After screening, eleven met the inclusion criteria and were analysed narratively. One study (7) using drug (Noradrenergic and Specific Serotonergic Anti-depressant) and placebo interventions combined with a CBT intervention was included in the review as the drug used within the study would not be dissimilar to GP prescribed medication for symptomatic management of depression which is a common co-associated disorder for CFS/ME patients (Maes 2011) and the effects of CBT for CFS using outcomes of interest to this review were measured by the trial and therefore included.

Design

All trials were described as randomised (see Appendix 2).

Duration of the interventions ranged from >9 weeks (6) to 26 weeks/6.5 months (3), with a median duration of 18.9 weeks/4.4 months. Follow up assessments ranged from 2 months (6) to 16 months (10) with the most common follow up length of 12 months (median = 9.6 months). One trial (7) measured final outcomes at post-treatment, immediately after intervention delivered.

Ten studies were granted ethical approval, and the majority provided the committee or board that gave approval and the trial reference numbers. The one study (8) that did not indicate whether ethical approval was sought or granted, but did provide details of taking participant consent. Nine trials detailed how participant consent was sought.

Comparison groups

The included trials used between two and four arms to conduct comparisons between conditions (See table 1 study details).

Setting

Five of the studies (2, 5, 6, 10 & 11) were conducted in the UK, three (1, 8 & 9) in The Netherlands, one (3) in the USA, one (4) in Spain and one (7) in Norway. 82 per cent (n = 9) of trials took place in specialist CFS/ME units or tertiary treatment facilities (See table 1 for full details).

Participants

Five studies (4, 6, 9, 10 & 11) detailed reasons why participants were excluded from taking part, in most cases these were broken down figures relating to specific exclusion criteria already outlined. Nine studies (1, 2, 4, 6, 7, 8, 9, 10 & 11) reported figures for the total number of eligible participants. Few studies (n = 2) reported the reasons why eligible participants declining to take part, other than providing reasons related to the study exclusion criteria or stating 'refusal'.

Sample size

A combined sample size of 1958 participants across the eleven included studies provided a mean of 178 participants (median = 123) and ranged from a pilot randomised study (6) of 35 participants to 641 recruited by a multicentre trial (11).

64 per cent (n = 7) of trials conducted power calculations, but only 29 per cent of those matched or exceeded the minimum sample size required at the follow-up assessment (7 & 11). One study (5) exceeded the calculated sample size for two of its three conditions.

Demographics

91 per cent (n = 10) of included trials reported demographic information, a mean age of 40.9 years was calculated across the studies. A total of 80 per cent were female (range: 67% – 91%). Ethnicity was reported in 27 per cent (n = 3) of studies which created a mean of 90 per cent of participants reported as White/Caucasian. Two studies (2 & 3) also provided marital status information which indicated 45 per cent of participants were married or cohabiting.

Duration of illness

Illness duration was reported for 73 per cent (n = 8) of studies and a mean of 61.7 months was calculated from available data. One trial (5) report illness duration as over 60 months for 47 per cent of their sample. Of the three studies (1, 3 & 7) failed to report illness duration it can be assumed an illness duration of over 6 months following the CFS/ME diagnostic criteria used.

Inclusion criteria

All eleven trials used recognised diagnostic criteria to ensure a CFS/ME diagnosis for study participation. Six studies (1, 3, 4, 5, 8 & 9) used The Centres for Disease Control and Prevention (CDC)/ Fukuda 1994 criteria alone, two studies (10 & 11) used Oxford/ Sharpe 1990 criteria alone and three studies (2, 6 & 7) used both CDC and Oxford criteria. Two studies (10 & 11) included the London ME criteria and one study (7) included the ICD-10 research criteria for Neurasthenia. In addition, one study (9) stated 4 or more of the possible 8 Fukuda 1994 criteria were met for study participation.

Six studies also included other CFS/ME criteria using scores on validated scales: ≥ 35 CIS-F (1, 8 & 9), < 700 SIP8 (1&8), ≥ 4 ChFS (6 & 10) ≥ 6 ChFS (11), ≤ 70 MOS-SF-36 (9, 10) and ≤ 65 MOS-SF-36 (11).

One trial (7) reported '*positive screening of fatigue by questionnaire in the waiting rooms*' no further information of this questionnaire was provided. Participation was not based on the responses to this unknown questionnaire, but provided an initial screen for the structured interviews and clinical evaluation to assess eligibility for study participation.

Five studies (1, 3, 5, 8 & 10) mentioned participants were required to read and speak the language in which the study was conducted in.

Two trials (9 & 10) explicitly stated GP referrals as part of their inclusion criteria.

One trial (6) stated as part of their inclusion criteria that participants must have already under taken CBT previously with the NHS service conducting the trial.

Exclusion criteria

82 per cent (n = 9) of the included trials stated exclusion criteria. Which included psychiatric, mental illness, depression, somatic/somatoform disorders, antisocial/boarder line/paranoid personality disorders or drug abuse .The Hospital Anxiety and Depression Scale (HADS) was used in one study (4) to screen participants for significant psychiatric disease.

Six studies (2, 3, 4, 5, 6 & 11) explicitly mentioned participants were excluded if they were unable to participate with study requirements such as attending appointments.

One study (6) excluded participants if they had any '*interpersonal difficulties*' that would make group participation inappropriate for them and other participants.

Three studies (2, 4 & 9) excluded participants if they did not meet the required blood screen, had a medical condition or were taking any medication that could account for their fatigue. Another trial (5) excluded participants if they were currently undergoing any physical investigations.

Three studies (1, 4 & 8) stated participants were excluded if they were engaged in any legal conflicts, including occupational conflicts and seeking financial support.

Two trials (5 & 10) excluded participants if they were engaged in any planned, concurrent rehabilitation or systematic psychological therapies. In addition, two trials excluded participants if they had engaged in the trial therapy before (11) or had received the trial therapy within the last year (10).

Other exclusion criteria were: pregnancy (2 & 3) and medication change within the last three months (2).

Assessment of participants for recruitment

Four studies employed set interview formats to assess for participation, these were: protocol for CFS management in Catalonia (4), schedules for clinical (SCAN) structured interviews (7), structured questionnaires (3) and structured clinical interview from the Diagnostic and Statistical Manual of Mental Disorders IV (11).

Study eligibility was assessed by a psychiatrist (2), a psychiatric nurse (9) and research assistant later confirmed by a qualified member of staff, all following a GP referral (10).

36 per cent (n = 4) of included trials did not state the process in which participants were assessed for recruitment following referral to the centre(s), in most cases this was via GPs. One of those trials (8) did state '*application of CFS criteria*' as the process of recruitment assessment but no other information was given as to who conducted this or how it was carried out.

Behavioural interventions

Behavioural conditions used by the included studies were: Cognitive Therapy Treatment (COG), Cognitive Behavioural Therapy (CBT), Mindfulness-Based Cognitive Therapies (MBCT), Graded Exercise Therapy (GET), Adaptive Pacing Therapy (APT), Relaxation training (Relax), Educational and Support (EAS), Supportive Listening (SL) and Pragmatic Rehabilitation (PR).

Comparison groups

82 per cent (n = 9) of studies compared at least one intervention group with a control group. With 18 per cent (n = 2) of trials comparing at least one behavioural treatment with another, without using a control group.

Manualisation

Eight trials (2, 3, 4, 6, 8, 9, 10 & 11) made reference to following manuals or treatment protocols.

Two trials (3 & 10) tested fidelity to manuals using taped sessions which were then rated. One of these trials (3) was also able to ensure each treatment condition was distinct from each other using fidelity testing.

Seven studies (2, 3, 6, 8, 9, 10 & 11) reported the therapists delivering the interventions received regular supervision, often fortnightly and included individual and/ or group supervision.

Cognitive therapies [see Appendix 1]

91 per cent (n = 10) of trials used cognitive therapies within the study: Nine studies had one (1, 3, 5, 6, 7, 9 & 11) or two (2 & 8) CBT conditions, one study (4) combined CBT with GET in a multidisciplinary group condition and one study (7) used a 12 week CBT intervention in each of the four conditions using drug and placebo medication. One trial (3) also included a Cognitive Therapy Treatment (COG) condition in addition to a CBT condition.

Model used

Six trials (1, 2, 3, 8, 9 & 11) referenced previous protocols or manuals for their CBT interventions, with an additional trial (6) noting Segal 2002 manual for MBCT. Although a referenced manual was not used for the COG condition (3) it was described as a '*broad-based cognitive approach*' formulated by a clinical psychologist and details of the session plans were provided.

Various components were included in the cognitive conditions. The most common features of the cognitive interventions were: modifying negative thought patterns and/ or challenging avoidant behaviours, challenging responses to symptoms (one study (6) achieved this by increased awareness of self while two studies (8 & 9) advocated disengagement from bodily sensations), incorporation of setback planning or relapse prevention and goal setting.

Delivery of intervention

Mode of delivery varied between studies but generally delivered face to face for individual sessions.

Therapist characteristics

Cognitive therapies were delivered by multidisciplinary professionals including clinical psychologists, psychiatrists, cognitive behavioural therapists, physiotherapists, occupational therapists and nurses.

Intensity of intervention

While there were differences of intensity of cognitive interventions between the included studies, the most common were 14 sessions delivered fortnightly. With 64 per cent (n = 7) of trials reporting the number and frequency of sessions. For one trial (9) it was unclear if the CFS booklet was at least 16 weeks based on a previous trial referenced or at least 20 weeks based on a model also referenced.

Exercise focused interventions

27 per cent (n = 3) of trials used an exercise condition; one trial used GET alone (11), one trial (4) used GET combined with CBT for a multidisciplinary group condition and the final trial (3) used Anaerobic Activity Therapy (ACT).

Model used

Two trials (4 & 11) made reference to using protocols or manuals from previous trials and one study did not, did provided detailed session plans for their ACT condition.

All three trials included gradually increasing exercise and physical activities over time, in a safe and monitored format, two of these trials also stated maximum heart rate (11) and

maximum time exercising at 40 minutes a day (4). Two trials (4 & 11) explicitly mentioned establishing a baseline and increasing incrementally from that level. Two studies (3 & 4) included flexibility exercises and relaxation. One study (3) stated flexibility and grip strength tests were conducted before the intervention started to guide therapy goals. Set back planning and daily exercise dairies were also mentioned as part of the ACT condition in one trial (3).

Delivery of intervention

Two trials (3 & 11) conducted individual face to face sessions (although one trial (11) acknowledged some were via telephone) and one study (4) used face to face group GET.

Therapist characteristics

One study (3) had trained nurses supervised by an exercise physiologist deliver the ACT condition, in one study (4) the GET condition was supervised by a physiotherapist and trained nurse with 20 years' experience (8 years in CFS) and the last trial (11) had 10 physiotherapists and 1 exercise physiologist to conduct the GET condition, with a median of 5 years' experience and regular supervision.

Intensity of intervention

One trial (3) conducted 13, 45 minute sessions fortnightly, one trial (4) had 1 hour sessions, three times a week for 3 months and another trial (11) reported up to 14 sessions, the first 4 were weekly and the subsequent sessions were fortnightly with an optional follow up session at 36 weeks.

Supportive (non-directive) therapies

18 per cent (n = 2) of trials used a supportive therapy condition: an Education and Support (EAS) group (5) and a Supportive Listening (SL) group (10).

Model used

Neither studies made reference to using a previously published manual or protocol but provided details of session aims/plans.

Both trials included the following components: sharing of experiences of CFS/ME, (including symptoms and previous treatments) and talking about the past experience of stigmatism and others in relation to their illness. One trial (5) also included the following features: CFS/ME education, exploring active and passive rest, stress, relaxation and breathing exercises and a discussion around anaerobic exercise. While the other (10) described as 'non-directive counselling approach' gave no explanations for symptoms and therapists were discouraged from giving advice or leading participants.

Delivery of intervention

One study (5) was conducted face to face in groups of between 8 – 10 participants and the other study (10) delivered the intervention individually using home visits and telephone consultations.

Therapist characteristics

One study (5) had a multidisciplinary team delivering the condition including a specialist physiotherapist, consultant clinical psychologist, clinical psychologist and senior occupational therapist. The other trial (10) had three nurses, provided with 4 months of training but had no previous experience of CFS/ME. The nurses also engaged in regular supervision with qualified counsellors.

Intensity of intervention

One trial (5) had 8 group sessions, lasting 2 hours run fortnightly, while the other trial (10) had 10 individual sessions over 18 weeks, using a mixture of 60 minute home visits (90 minutes for the first appointment) and 30 minute telephone calls.

Other behavioural treatments

27 per cent (n = 3) of trials used other behavioural conditions, one study (3) had a relaxation condition, one study (10) used a 'Pragmatic Rehabilitation' (PR) condition (consisting of elements of CBT and increasing activity collaboratively with the participant) and the other study (11) had an Adaptive Pacing Therapy (APT) condition.

Model used

The relaxation condition (3) was stated to be based on prior studies yet no references were provided in the condition description. The PR condition (10) did not mention a manual but signposted an earlier study by the same authors. However for the APT condition (11) the authors designed a pilot manual with the collaboration with experienced clinicians and Action for ME (AfME) as a manual was unavailable.

Features of the relaxation condition (3) were: to improvement of coping skills, reduce sleep disturbances and anxiety and improve wellbeing and tolerance to illness with the use of stress/fatigue diaries, relaxation exercises, breathing exercises and thematic imagery.

The PR condition (10) included graded return to activity, to regularise sleep patterns, addressing concentration and memory problems with the use of relaxation exercises, goal setting and relapse prevention.

Components of the APT condition (11) included planning, prioritising and pacing activity, to reduce or avoid fatigue and over exertion, identifying early warning signs, and not to exceed

more the 70% of perceived energy using daily diaries and planning regular daily rest and different types of activities.

Delivery of intervention

All three trials delivered conditions on an individual basis, with one trial providing some telephone consultations (11) and another study (10) delivered the intervention using home visits and telephone consultations.

Therapist characteristics

Two trials (3 & 10) reported trained nurses with regular supervision delivered the conditions and the other trial (11) was delivered by 9 occupational therapists with a median of 7 years' experience, who also received regular supervision.

Intensity of intervention

One study (3) reported 13, 45 minute sessions run fortnightly, one study (10) had 10 individual sessions over 18 weeks, using a mixture of 60 minute home visits (90 minutes for the first appointment) and 30 minute telephone calls and the last trial (11) reported up to 14 sessions, the first 4 were weekly and the subsequent sessions were fortnightly with an optional follow up session at 36 weeks.

Outcomes

100 per cent (n = 11) of included trials measured fatigue using: Chalder Fatigue Scale (ChFS) (Chalder 1993), Fatigue Severity Subscale of the Checklist Individual Strength (CIS-F) (Vercoulen 1994), Fatigue Severity Scale (FSS) (Krupp 1989) and Fatigue Impact Scale Questionnaire (FIQ) (Meads 2009).

100 per cent (n = 11) included trials employed at least one measure of functioning, however there was variation in the description for these measures, some stated they were assessing functioning, disabilities, impairment, quality of life (QoL) or general health. Some trials used total scores while others only used specific subscales, most often a physical functioning subscale alone. The measures used by the included studies were: Medical Outcomes Survey Short Form (MOS-SF-36) (Stewart 1988), The Sickness Impact Profile (SIP) (Bergner 1981: Jacobs 1990), Work and Social Adjustment Scale (WSAS) (Mundt 2002), Physical Functioning (PF-10) (McHorney 1994), Stanford Health Assessment Questionnaire (HAQ) (Esteve-Vives 1993) the General Health Questionnaire (GHQ) (Goldberg 1988), Subjective Health Complaint Inventory (SHQI) (Eriksen 1999), and the QoL Scale (Burckhardt 2003). 36 per cent (n = 4) of included trials also included a perceived change in overall health measure (Clinical Global Improvement CGI) (Sharpe 1996), which were mostly individual to each trial rather than a standardised assessment tool.

64 per cent (n = 7) of included trials assessed depression using: the Hospital Anxiety and Depression Scale (HADS) (Zigmond 1983), the Beck Depression Inventory (BDI) (Beck 1996) and the Hamilton Rating Scale for Depression (HRSD) (Hamilton 1960).

55 per cent (n = 6) of included trials assessed depression using: HADS and Beck Anxiety Inventory (BAI) (Beck 1990). One study (9) stated assessing psychological distress using the Brief Symptom Inventory (BSI) (Derogatis 1983).

One study (5) also included an assessment of mood (alertness, hedonic tone and anxiety) as part of a cognitive battery (Smith 1996).

Excluded studies

After scrutinising the 39 full papers most studies were excluded on methodological basis because they were not RCTs (n = 19) and others such as reviews were excluded as there was no data available to extract (n = 6). One did not fulfil the review disease terms, one study was not translated in time (Martin 2011) and one study (Bazelmans 2005) was requested in full but did not arrive in time to make this review.

Quality assessment

Trial quality was assessed using the JADAD appraisal tool (Jadad 1996). A slight scoring modification was applied as double blinding is difficult for behavioural trial conditions; as a result 0.5 points were awarded to studies that made every attempt to blind wherever possible, this also increase the sensitivity of this tool when comparing quality across all studies included (See Appendix 2).

Randomisation sequence generation

All trials were described as randomised; 82 per cent (n = 9) presented as low risk and for 18 per cent (n = 2) the risk was unclear.

Allocation concealment

64 per cent (n = 7) of the included trials presented as low risk and for 36 per cent (n = 4) the risk was unclear.

Blinding of outcome assessment

Only 9 per cent (n = 1) of trials included conducted double blinding, which was appropriate to the methodology of the RCT (24 weeks medication and 12 weeks CBT or 12 weeks CBT then 12 weeks medication, medication was either drug or placebo identical in size (15mg) and appearance with no smell). This left 91 per cent (n = 10) where blinding therapists and participants was inappropriate and so not done.

Incomplete data

82 per cent of the eleven included studies (n = 9) reported addressing missing data for their analysis resulting in low risk of bias and for 18 per cent (n = 2) the risk was unclear.

Selective reporting

91 per cent (n = 10) of the included trials reported all pre-specified outcomes with 9 per cent (n = 1) that only reported these as baseline characteristics (2) resulting in high risk of bias.

Other risks of bias

Differences in baseline characteristics were reported for three of the included trials (2, 5 & 8) see table 1.

One of the included trials reported payment for participation (3).

Description of withdrawals/dropouts

Criteria used to describe dropouts for 91 per cent (n = 10) of included trials was if participants didn't complete post treatment and/or follow up assessments, however for one study (3) dropouts were described as attending four or less sessions prior to post treatment assessment.

Ten trials described dropout rates by condition and only one trial (3) gave an overall dropout percentage of 25%. The mean aggregate reported dropout rate of all eleven studies was 9.2%, ranging from 1.7% (11) to 46.3% (2), with only two trials (2 & 3) reporting rates over 20%.

The mean reported dropout rate by condition at post treatment or first follow up was 11.3% (median = 9.5%), ranging from 0% (control conditions 6 & 10) to 36% (Telephone CBT 2). The mean reported dropout rate by condition at study end point was 14.4% (median = 10%), ranging from 0% (control condition 6) to 56% (Telephone CBT 2).

Five trials (5, 6, 7, 9 & 10) provided reasons for dropouts (See Appendix 3).

Effects of Interventions

Meta-analysis (MA) is inappropriate for trials that address different hypotheses, and when the level of generalization is inappropriate for the question being asked (Greenlaugh 1998). Due to the differences of included components, delivery, intensity, therapist characteristics, outcomes and follow up assessment time points between the included studies meta-analysis was deemed inappropriate. Furthermore with varying quality of included studies if an MA had been conducted this would be a considerable criticism of the results (Wolf 1986). With this in mind a narrative synthesis was adopted for this review.

Fatigue

Of the eleven included studies that measured fatigue, 55 per cent (n = 6) reported an improvement in mean fatigue scores, with 64 per cent (n = 7) finding a statistically significant improvement in fatigue scores for the behavioural interventions compared to control groups at post treatment (1), 2 months (6), 2.8 months (7), 4.6 months (10), 5.6 months (7), 8.2 months (9), 12 months (5 & 11) and 16.3 months (10) follow up.

Specifically, of the ten trials that used cognitive therapy conditions, 55 per cent (n = 6) reported a statistically significant improvement in fatigue scores at post treatment (1), 2 months (6), 2.8 months (7), 5.6 months (7), 6 months (6), 8.2 months (9) and 12 months (6 & 11) follow up. 30 per cent (n = 1) of trials that used exercise focused conditions reported a statistically significant improvement in fatigue scores at 12 months (11) follow up. 33 per cent (n = 1) of trials that used other behavioural conditions, found a significant improvement in fatigue at 4.7 months follow up (PR condition).

Functioning

Of the eleven included trials, 82 per cent (n = 9) reported an improvement in mean functioning scores, with 73 per cent (n = 8) observing a statistically significant improvement in functioning on at least one measure of functioning outcomes compared to control groups at post treatment (1 & 6), 2.8 months (7), 5.6 months (7), 6 months (6) and 12 months (2, 3, 4, 5 & 11) follow up. In addition, 9 per cent (n = 1) reported a statistically significant decrease in functioning at 4.7 months follow up after receiving the supportive listening condition (10).

By condition type, of the ten trials that used cognitive therapy interventions, 80 per cent (n = 8) reported a statistically significant improvement in functioning on at least one measure at post treatment (1), 2.8 months (7), 5.6 months (7), 6 months (6) and 12 months (2, 3, 4, 5 & 11) follow up. Of the three trials that used exercised focused interventions, 67 per cent (n = 2) reported a statistically significant improvement in functioning on at least one measure at 12 months follow up (4 & 11).

Depression

Of the seven included trials that measured depression, 86 per cent (n = 6) reported an improvement in mean depression scores, with 71 per cent (n = 5) finding a statistically significant improvement in depression scores compare to control groups at post treatment (6), 4.7 months (10), 5.6 months (7) and 12 months (3 & 11) follow ups. Interventions producing significant improvements were: CBT (n = 2), COG (n = 1), MBCT (n = 1) supportive listening conditions (n = 1) and GET (n = 1).

Anxiety

Of the six included trials that measured anxiety, 11 per cent (n = 5) reported an improvement in mean anxiety scores, with 50 per cent (n = 3) observing a statistically significant improvement compared to control groups at 6 months (6) and 12 months (3 & 11) follow up. The conditions that produced the significant results were: CBT (n = 2), Relax (n = 1) and GET (n = 1).

Adverse events

Of the eleven included studies 27 per cent (n = 3) reported adverse events (See Appendix 4).

DISCUSSION

The Bagnall (2007) review identified and reported thirteen studies using behavioural interventions, this review has found an additional eleven trials adding to the evidence base for behavioural interventions in CFS/ME.

This review has found behavioural interventions have beneficial effects in improving fatigue, functioning, depression and anxiety for adults with CFS/ME. Specifically finding cognitive interventions and GET were effective at improving fatigue, functioning depression and anxiety, supportive (non-directive) interventions effective at improving depression but had significantly worse effect on functioning, PR significantly improved fatigue and relaxation had beneficial effects on anxiety.

CBT was the most common form of treatment used in the included studies, this is assumed to reflect treatments currently used in clinical practice following the NICE (2007) guideline and the growing evidence base for CBT in CFS/ME (Malouff 2008, Price 2009) and long-term condition management. This was consistent with the previous review which reported 69 per cent of included trials using CBT or modified CBT (Bagnall 2007).

Consistent with conclusions from Price (2009) this review found a limited evidence base for combination interventions, with two included studies using a combined rehabilitation approach (4 & 10) In which, one of the trials found significant worsening of physical functioning due to increased pain and weakness compared to the control group (4) and the other only found significant improvements in fatigue at the 20 week follow up that were not sustained at the 70 weeks follow up. Given the complexity, unpredictability and individual nature of CFS/ME presentations (AfME 2013a), to achieve optimal improvements combination treatment might be most appropriate (Price 2009). This is supported by the findings of Stabhaug (2008) where participants taking mirtazapine (noradrenergic and specific serotonergic anti-depressant) for

12 weeks after CBT significantly improved in fatigue, functioning and depression compared to the placebo group. This initial evidence suggests the combination of pharmacological and behavioural treatments might provide optimal improvements for patients with CFS/ME, and the order in which these treatments are given. This finding also supports the NICE (2007) recommendations for CFS/ME treatment and management.

Given the popularity of 'third wave' behavioural treatments in professional practice for long-term conditions, it was surprising only one included study (Rimes 2011) used such interventions. It is predicted future reviews will be able to provide more conclusive findings for the effectiveness of third wave interventions, and specifically mindfulness in the treatment and management of CFS/ME.

One of the SL conditions found participants were significantly worse in functioning at 4.7 month follow up. Although this trial conducted by Wearden (2010) scored moderately on the JADAD quality assessment, there were only two trials that used a supportive condition not led by any therapeutic style (described as non-directive counselling). Therefore, it is difficult to draw conclusions based on this evidence alone.

An unexpected result was found by one trial (5) when a control group indicated statistically significant improvements in fatigue between 6 and 12 months assessments. This could have occurred as part of natural fluctuation of CFS/ME (Jason 1999) or that standard medical care provides limited short-term beneficial effects, as it was not maintained at the 12 months follow up. As no other included studies reported a statistically improved result for their control groups, it may be likely this is not a typical finding.

Long-term effects

Although the fluctuating nature of the illness with regards to symptom severity and activity levels (Jason 1999) does complicate matters when attempting to identify the long-term effects of treatment, Chambers (2006) suggests long-term follow up is needed due to the chronic nature of CFS/ME. For the purposes of this review long-term is defined as 12 months or over. Using this definition it is promising over half the included studies 55 per cent (n = 6) assessed outcomes at or over 12 months.

Fatigue

Significant improvements in fatigue scores were more frequently noted at assessments under 12 months rather than at long-term follow up (71 per cent \geq 12 months: 29 per cent $<$ 12 months). These review observations would suggest improvements in fatigue are not sustainable in the long-term and reasons for this need to be investigated and addressed to ensure patient benefit and cost-effectiveness of treatment.

Functioning

Significant improvements in functioning were more frequently found at long-term follow up compared to assessments under 12 months (60 per cent \geq 12 months: 40 per cent $<$ 12 months). One of the included trials that found significant improvements at 12 months offered a 'booster' session at approximately at 3 months (10). The results of this review could indicate behavioural treatments (specifically cognitive treatments) are effective at improving functioning levels in the long-term, without the requirement of a refresher session. A caveat to this might be, that if refreshers session were employed, the effect of the intervention might be strengthened and subsequently result more significant improvements reported.

Depression

Significant improvements in depression were found at both short and long-term follow ups (50 per cent \geq 12 months: 50 per cent $<$ 12 months). This evidence would suggest behavioural interventions (specifically cognitive therapy) provide stable beneficial effects on reducing depression over time.

Anxiety

Significant improvements in anxiety were more frequently found at long-term follow up (75 per cent \geq 12 months: 25 per cent $<$ 12 months). Furthermore, one of the trials that reported significant improvements in anxiety at 6 months but attributed this significance to high testing of data rather than a genuine result (5). The review findings might indicate individual behavioural treatments are not adequate to improve anxiety in the short-term (6 months) but could positively affect anxiety in the long-term (12 months). Conversely as the conditions that provided significant findings were three distinct interventions (CBT, GET and Relaxation), it would be wise to make more accurate conclusions when more data is available.

Quality of the evidence

The overall attribution rate was reasonably low, with only two of included trials reported drop-out rates over 20%. In contrast, few trials reported reasons for withdrawals so it is unclear if these are similar across trials.

It is noted there was variable quality between the included studies (Median 2.5/5 JADAD score ranging from 1-5). Although it is encouraging that the majority of included studies provided details of randomisation processes and employed strategies to manage missing data.

O'Dowd (2006) justified changing participant condition allocation to protect condition integrity, however this does weaken their randomised trial status.

Few studies provided adverse effect reporting. It is suggested behavioural interventions produce fewer adverse effects compared to pharmacological interventions (Bagnall 2007),

however this review cannot support this finding based on the few included studies that reported this information.

Few studies reported on reasons why eligible participants did not want to participate other than study exclusion criteria or stating 'declined to participate'. Ideally this should be reported to ensure a representable sample is included and to be able to generalise the findings.

Over half of the included trials limited participation to those physically able to attend sessions. Four of the included trials used self-guided CBT instructions and email/ telephone contact or telephone CBT but still required an initial face to face session at the trial centre. There was only one included study able to include more severely affected CFS/ME patients by conducting home visits as part of the intervention modality (10). This finding is consistent with other reviews (Chambers 2006: Price 2009) concluding more severely affected CFS/ME patients are not being represented in the available data.

Although one included study (5) recruited a slightly higher proportion of men compared to the other included studies (33% vs. 18%) this figure remained consistent with CFS/ME gender prevalence rates, which estimate twice as many women are affected with CFS/ME (AfME 2013b: Afari 2003).

One study specified participants must have completed CBT within the NHS unit to be eligible to participate (6) while another excluded patients if they have received the intervention treatment in the last year (10). It may indicate some therapies required a certain level of treatment experience, or none whatsoever before engaging in treatment for optimal outcomes. Or this could simply be due to the RCT methodology to ensure effects are a result of the trial interventions.

Combined with increased monitoring of symptoms by participating in studies that would not usually happen outside research, it has been suggested that intervention groups have increased contact with healthcare and/ or research staff compared to control group, which may influence findings (Cramp 2012). Only one of the included studies (4) matched their control condition in terms of intensity and length with their intervention conditions. However to achieve this, an additional component was added of 'exercise counselling' was included in the control condition. It could be questioned that this control condition is not comparable to the other usual care or waiting list conditions the other studies used, or that in fact it is an intervention in its own right.

Limitations of this review

Two studies were excluded from this review as one was unable to be translated in time and subsequently the included papers were all published in the English language, creating a

potential language bias. The other excluded study Bazelmans (2005) did not arrive in time to make this review. At this time it is unknown if either of these studies fulfilled the review's inclusion criteria.

Although contact was made to authors obtained from the search returns for further information, no unpublished data was used in this review. While this ensures that all the included studies were peer reviewed it also increases the chance of publication bias, whereby significant results are more likely to be reported.

It is acknowledged there is a potential risk of multiple publication bias, as WL data from one trial was used in another included trial (1 & 8). The two trials had different intervention data and one of the trials (1) contributed very little to the review findings as little data was provided in the 'short report' publication (despite contacted the authors).

Although high heterogeneity of studies and study conditions was described throughout this review, no heterogeneous tests were performed to support the author's judgement not to perform meta-analysis.

Cognitive therapy conclusions were drawn from various types of therapies: Self instructions CBT, telephone CBT, face to face CBT, cognitive therapy and MBCT. This was done because similarities were found between components included, providing merit in combining them to comment on overall cognitive treatment effects. However, it is acknowledged they have different theoretical underpinning and techniques used, in addition to modality and delivery differences than might warrant evaluating separately when more research is available. This might also help to determine what particular elements of cognitive treatments are most beneficial, and for whom. Similarly the difference between group and individual therapy effects should be addressed when more data is available.

Functioning was a broad term in this review to encompass parallel constructs measured by similar outcome measures. Alternatively functioning could be specified by types, for example physical and mental to gain more insight between different types of functioning.

AUTHOR'S CONCLUSIONS

Planning is required for future studies to address appropriate methods of blinding therapists and participants to increase the quality of research and the confidence in findings produced.

Although it was not an objective of this review to compare between behavioural conditions four of the included trials did compare more than one type of intervention condition (3, 5, 10 & 11). It was reported by O'Dowd (2006) the CBT group significantly improved in fatigue and

functioning compared to the EAS group. Similarly it was observed by White (2011) that both CBT & GET conditions significantly improved in fatigue, functioning and depression compared to APT and Wearden (2010) noted improved fatigue for PR and improve depression but worse functioning for SL. Further research needs to establish whether specific combination interventions provide beneficial effects compared to behavioural interventions alone and other available treatment options.

Similarly, while it was not the scope of this review to comment on the effects of behavioural interventions for subgroups of CFS, due to the complexity of the condition, it is apparent this could be a co-variate to the effectiveness of treatment and should be considered in future trials. This finding is consistent with other reviews (Chambers 2006: Malouff 2008: Price 2009).

In addition the evidence of the effects of booster sessions for maintaining long-term effects of interventions needs to be investigated and further research needs to determine effective treatments for the varying severities of CFS/ME

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Included review studies:

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APPENDIX 1

Characteristics of cognitive therapies

Table 1.1 Components of cognitive therapies:

Components included in cognitive interventions	1	2	3	4	5	6	7	8	9	11
Modifying negative thought patterns and/or challenging avoidant behaviours		√	√ ^{ab}	√	√	√	√	√	√	√
Challenging responses to symptoms				√	√	√		√	√	√
Setback planning/ relapse prevention		√	√ ^{ab}	√	√			√	√	
Goal setting	√	√	√ ^a		√			√	√	
Establishing and gradually increasing baseline energy levels		√	√ ^a		√				√	√
Promoting healthy sleep patterns		√	√ ^a	√					√	√
Relaxation, breathing exercises and/ or stress management			√ ^b	√	√	√	√			
Exercise programmes				√			√	√		
Communication and/or assertiveness				√	√				√	

Increasing awareness of self						√	√			
Fostering a more accepting, non-judgemental and compassionate self			√ ^b			√				
Increasing attention and memory					√					
Sensorial focalisation on sexual inhibition					√					
Addressing triggers and maintaining factors									√	
Planning return to work									√	
Addressing expectations of social environment									√	
Achieving healthy balance of activity, rest and leisure without using structural schedules			√ ^b							
Use of cognitive coping statements			√ ^b							
Imagery exercised used			√ ^b							
Use of memory compensation and cognitive retraining techniques			√ ^b							
Use of CDs							√			

^a CBT condition ^b COG condition

Table 1.2: Mode of delivery of cognitive therapies

ID	Study first author and Date	Individual	Group	Instructions	Face to Face	Telephone	Notes/ Group Sizes (GS)
1	Knoop 2008	1		1			
2	Burgess 2011	1			1	1	2 CBT conditions
3	Jason 2007	1			1		
4	Nunez 2011		1		1		GS = 16
5	O'Dowd 2006		1		1		GS = 8-12
6	Rimes 2011		1		1		GS = 7 and 11
7	Stubhaug 2008		1		1		GS = unknown
8	Tummers 2010	1		1			Plus additional F2F CBT after booklet if needed
9	Tummers 2012	1		1			
11	White 2011	1			1		
	Total	6	4	3	7	1	

Table 1.3: Intensity of cognitive therapies:

Study:	Number of sessions	Length of sessions (minutes)	Frequency of sessions	Length of condition
1	16		Weekly	> 16 weeks
2	(1) 14 (2) <15			
3	13	45	Fortnightly	
4 §	9	90	Biweekly	2.5-3 months
5	8	120	Fortnightly	
6	9	135		
7				12 weeks
8	(3) >16 (4) 14		(3) Weekly (4) Fortnightly*	(3) >16 weeks (4) 6 months
9	>16/ >20		Weekly	> 16 weeks/ > 20 weeks
11	<15		Sessions 1-4 weekly, 5-14 fortnightly and optional follow up (session15) at 36 weeks	23 weeks & follow up at 36 weeks

(1) Telephone CBT condition (2) Face to Face CBT condition (3) CBT booklet (4) additional CBT sessions after CBT booklet *calculated from information provided § CBT component of a multidisciplinary group condition.

Table 1.4: Therapists delivering cognitive therapies

	Delivered by
1. Knoop 2008	Nurses
2. Burgess 2011	Nurses
3. Jason 2007	Nurses
4. Nunez 2011	Clinical psychologist and Physiotherapist (also nurse)
5. O'Dowd 2006	Consultant clinical psychologist, clinical psychologist, specialist physiotherapist and senior occupational therapist
6. Rimes 2011	Clinical psychologist
7. Stubhaug 2008	Psychiatrist, physiotherapist and psychiatric nurse
8. Tummers 2010	Cognitive behavioural therapists
9. Tummers 2012	Psychiatric nurses
11. White 2011	Clinical psychologists, nurses and counselling psychologists

APPENDIX 2

Risk of Bias and Quality Assessments

Table 2.1 Risk of Bias and JADAD appraisal

	Random sequence generation	Allocation concealment	Blinding of outcome assessment	Incomplete outcome data	Selective reporting	Randomised	Randomisation described and appropriate	Double blind	Blinding described and appropriate	Description of withdrawals	JADAD score
1. Knoop 2008	Green	Green	Red	Green	Green	1					1
2. Burgess 2011	Green	Green	Red	Green	Red	1	1			0.5	2.5
3. Jason 2007	Green	Blue	Red	Green	Green	1	1				2
4. Nunez 2011	Green	Green	Red	Blue	Red	1	1		0.5	0.5	3
5. O'Dowd 2006	Green	Green	Red	Green	Green	1	1		0.5	1	3.5
6. Rimes 2011	Green	Green	Red	Blue	Green	1	1			1	3
7. Stubhaug 2008	Green	Blue	Green	Green	Green	1	1	1	1	1	5
8. Tummers 2010	Green	Green	Red	Green	Green	1	1				2
9. Tummers 2012	Green	Green	Red	Green	Green	1	1			0.5	2.5
10. Wearden 2010	Green	Blue	Red	Green	Green	1	1			1	3
11. White 2011	Blue	Green	Red	Green	Green	1	1				2

APPENDIX 3

Withdrawal reasons

Table 3.1 Reasons reported for patient withdrawal

	Reasons for drop-out
1. Knoop 2008	Not reported
2. Burgess 2011	Not reported
3. Jason 2007	Not reported
4. Nunez 2011	Not reported
5. O'Dowd 2006	SMC @ 6m 3 withdrew (no reasons given) (84/123/143), 1 on holiday (66) and 1 UTA (99) @ 12m 3 withdrew (no reasons given) (84/123/143), 2 Unable to contact (99/152), 1 transport problems (98) and 1 unknown (7).
	EAS ' 6m 1 moved away (153), 1 CFS too severe (140), 1 didn't find approach helpful (17) and 2 unknown (56/73) @ 12m 1 CFS too severe (140), 1 too ill to attend (138), 1 Unable to contact (96) and 1 unknown (56)
	CBT @ 6m 2 moved away (20/76), 1 dropped out following an argument (31), 1 work pressures (154) 1 transport problems (137), 2 UTA bereavement (132/142) and 2 unknown (23/119) @ 12m 4 moved away (14/20/28/76), 1 dropped out following an argument (31), 1 work pressures (65), 1 'did not see the point' (132), 1 too ill to attend (150), 1 Unable to contact (145), 1 declined to make further appointment (154) 3 unknown (34/37/137)
6. Rimes 2011	MCBT Family illness (n = 1) Didn't like 'group nature' (n = 1)
7. Stubhaug 2008	Drug+CBT Protocol violation (n = 5) unknown (n = 1)
	Placebo+CBT Protocol violation (n = 2)
	CBT+Drug Protocol violation (n = 3))
	CBT+Placebo Protocol violation (n = 1
8. Tummers 2010	Not reported
9. Tummers 2012	CBT Did not want to complete second assessment (n = 7)
	WL Did not want to complete second assessment (n = 5)
10. Wearden 2010	PR 'Unhappy with randomisation' (n = 4) 'Too busy' (n = 3) 'no benefit or felt worse' (n = 2) 'Lost contact or no reason' (n = 2) 'Nurse therapist safety concern' (n = 1) Lost to follow up (n = 3).
	SL 'no benefit or felt worse' (n = 3) 'Nurse therapist safety concern' (n = 1) 'Too busy' (n = 1) 'Misdiagnosis' (n = 1) 'Received different treatment' (n = 1) Lost to follow up (n = 4).
	GP Lost to follow up: Declined (n = 8) No response (n = 6)
11. White 2011	Not reported

APPENDIX 4

Adverse events

Table 4.1 Description of adverse events reported

	Adverse events reported
1. Knoop 2008	Not reported
2. Burgess 2011	Not reported
3. Jason 2007	Not reported
4. Nunez 2011	Not reported
5. O'Dowd 2006	Not reported
	Not reported
	Not reported
6. Rimes 2011	No substantive adverse events
7. Stubhaug 2008	Not reported
	Not reported
	Not reported
	Not reported
8. Tummers 2010	Not reported
9. Tummers 2012	Not reported
	Not reported
10. Wearden 2010	Development of Herpes Simplex (n =1), Suicide attempt (n = 1) Bleeding peptic ulcer (n = 1) Recurrence of cancer (n =1) *Independent data monitoring committee considered all were unrelated to trial treatments.
11. White 2011	APT Serious adverse events (n = 16) Serious adverse reactions (n = 2) Suicidal thoughts: Worsened depression
	CBT Serious adverse events (n = 8) Serious adverse reactions (n = 4) Episode of self-harm: Low mood and episode of self-harm: Worsened mood and CFS symptoms: Threatened self-harm
	GET Serious adverse events (n = 17) Serious adverse reactions (n = 2) Deterioration in mobility and self-care: Worse CFS symptoms and function
	SpMC Serious adverse events (n = 7) Serious adverse reactions (n = 2) Worse CFS symptoms and function: Increased depression and incapacity

Appendix B - Ethical Approval:

- NHS NRES approval


Health Research Authority
NRES Committee Yorkshire & The Humber - Bradford Leeds
Room 002
TEDCO Business Centre
Viking Industrial Park
Rolling Mill Road
Jarrow
NE32 3DT
Telephone: 0191 4283548

01 August 2014

Miss Deborah Baldwin
[Redacted]

Dear Miss Baldwin

Study title: Long-term experiences of managing CFS/ME: A qualitative study
REC reference: 14/YH/1135
IRAS project ID: 143726

Thank you for your application for ethical review, which was received on 01 August 2014. I can confirm that the application is valid and will be reviewed by the Proportionate Review Sub-Committee in correspondence. To enable the Proportionate Review Sub-Committee to provide you with a final opinion within 10 working days your application documentation will be sent by email to Committee members.

One of the REC members is appointed as the lead reviewer for each application reviewed by the Sub-Committee. I may contact you by email (or phone) between 4th August and 6th August to clarify any points that might be raised by members and assist the Sub-Committee in reaching a decision.

If you will not be available between these dates, you are welcome to nominate another key investigator or a representative of the study sponsor who would be able to respond to the lead reviewer's queries on your behalf. If this is your preferred option, please identify this person to us and ensure we have their contact details.

You are not required to attend a meeting of the Proportionate Review Sub-Committee.

Please do not send any further documentation or revised documentation prior to the review unless requested.

Documents received

The documents to be reviewed are as follows:

Document	Version	Date
Evidence of Sponsor Insurance or Indemnity (non NHS Sponsors only) [UWE Employers and Public Liability]	n/a	22 June 2014
Interview schedules or topic guides for participants [Participants Interview Topics]	1.1	16 July 2014

IRAS Checklist XML [Checklist_01082014]		01 August 2014
Letters of invitation to participant [invitation letter]	1.2	13 July 2014
Other [Researcher Interview Topics]	1.1	16 July 2014
Other [Response Form]	1.2	13 July 2014
Other [UWE Professional Liability]	n/a	15 July 2013
Other [J. Byron-Daniel CV]		
Participant consent form [Consent Form]	1.2	13 July 2014
Participant information sheet (PIS) [PIS]	1.2	13 July 2014
REC Application Form [REC_Form_01082014]		01 August 2014
Research protocol or project proposal [Protocol]	1.1	30 July 2014
Summary CV for Chief Investigator (CI) [Research CV CI]	July 2014	16 July 2014

No changes may be made to the application before the meeting. If you envisage that changes might be required, you are advised to withdraw the application and re-submit it.

Notification of the Sub-Committee's decision

We aim to notify the outcome of the Sub-Committee review to you in writing within 10 working days from the date of receipt of a valid application.

If the Sub-Committee is unable to give an opinion because the application raises material ethical issues requiring further discussion at a full meeting of a Research Ethics Committee, your application will be referred for review to the next available meeting. We will contact you to explain the arrangements for further review and check they are convenient for you. You will be notified of the final decision within 60 days of the date on which we originally received your application. If the first available meeting date offered to you is not suitable, you may request review by another REC. In this case the 60 day clock would be stopped and restarted from the closing date for applications submitted to that REC.

R&D approval

All researchers and local research collaborators who intend to participate in this study at sites in the National Health Service (NHS) or Health and Social Care (HSC) in Northern Ireland should apply to the R&D office for the relevant care organisation. A copy of the Site-Specific Information (SSI) Form should be included with the application for R&D approval. You should advise researchers and local collaborators accordingly.

The R&D approval process may take place at the same time as the ethical review. Final R&D approval will not be confirmed until after a favourable ethical opinion has been given by this Committee.

For guidance on applying for R&D approval, please contact the NHS R&D office at the lead site in the first instance. Further guidance resources for planning, setting up and conducting research in the NHS are listed at <http://www.rdforum.nhs.uk>. There is no requirement for separate Site-Specific Assessment as part of the ethical review of this research.

Communication with other bodies

All correspondence from the REC about the application will be copied to the research sponsor and to the R&D office for RNHRD Foundation Trust. It will be your responsibility to ensure that other investigators, research collaborators and NHS care organisation(s) involved in the study are kept informed of the progress of the review, as necessary.

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

14/YH/1135	Please quote this number on all correspondence
------------	--

Yours sincerely



Miss Christie Ord
Research Ethics Committee Assistant

Email: nrescommittee.yorkandhumber-bradfordleeds@nhs.net

Copy to: *Ms Leigh Taylor, University of West England*

Ms Jane Carter, RNHRD Foundation Trust

• RNHRD approval

Royal National Hospital 
for Rheumatic Diseases
NHS Foundation Trust

Upper Borough Walls
Bath, BA1 1RL
Telephone: 01225 465941
Safe Haven fax: 01225 473461

Debbie Baldwin
CFS Adults

18th September 2014

Dear Debbie

RBB 438 – Long-term experiences of living with CFS. A qualitative study

Thank you for your application for approval of the above project, which was considered at the R&D Committee meeting on the 16th September 2014. I am pleased to inform you that the R&D Committee approved the study subject to sign off from Hayley Sewell re: data protection. UWE will act as sponsor.

The following documents were approved

- Consent Form Version 1.4
- Response Form Version 1.3
- Invitation Letter Version 1.3
- PIS version 1.3
- Protocol Version 1.2

Details of this project will be entered onto the RNHRD database. The reference number for your project is **RBB 438** and this should be used in all correspondence. A short progress report will be required annually or at the end of the project, whichever occurs first.

All research approved by the R&D Committee should follow good clinical practice and adhere to the systems in place for Research Governance. All Principal Investigators must undertake Good Clinical Practice training and are responsible for ensuring that their research staff have received appropriate training.

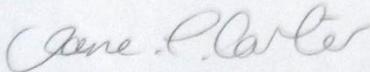
You are responsible for ensuring that, all participants sign informed consent (whenever applicable) and that the protocol agreed by the local research ethics committee is adhered to by yourself and any co-workers.

You are required to provide us with information about any amendments to the protocol, changes in funding, personnel or end date and any research-related adverse events. Any staff working on this study at this site must be issued with a contract with RNHRD (honorary or substantive) or Letter of Access before they commence work on the study at this site. Please make sure that the RNHRD is acknowledged on all academic papers which may be written as a result of this research.

In addition, other information may be requested from time to time and a lay summary of the results will be requested from you at the end of the study. This study may be subject to audit by the R&D Office.

We wish you well with this research.

Yours sincerely



Jane Carter
R&D Manager

• UWE approval



Faculty of Health &
Applied Sciences
Glenside Campus
Blackberry Hill
Stapleton
Bristol BS16 1DD

Tel: 0117 328 1170

Our ref: JW/lt

14 November 2014

Miss Deborah Baldwin

Dear Deborah

Application number: HAS/14/11/38

Application title: Long-term experiences of managing CFS/ME: A qualitative study

NHS Application Number: 14/YH/1135

Your NHS Ethics application and approval conditions have been considered by the Faculty Research Ethics Committee on behalf of the University. It has been given ethical approval to proceed with the following conditions:

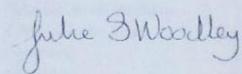
- You comply with the conditions of the NHS Ethics approval.
- You notify the Faculty Research Ethics Committee of any further correspondence with the NHS Ethics Committee.
- You must notify the Faculty Research Ethics Committee in advance if you wish to make any significant amendments to the original application.
- If you have to terminate your research before completion, please inform the Faculty Research Ethics Committee within 14 days, indicating the reasons.
- Please notify the Faculty Research Ethics Committee if there are any serious events or developments in the research that have an ethical dimension.
- Any changes to the study protocol, which have an ethical dimension, will need to be approved by the Faculty Research Ethics Committee. You should send details of any such amendments to the committee with an explanation of the reason for the proposed changes. Any changes approved by an external research ethics committee must also be communicated to the relevant UWE committee.
- Please note that any information sheets and consent forms should have the UWE logo. Further guidance is available on the web:
<http://www1.uwe.ac.uk/aboutus/departmentsandservices/professionalservices/marketandcommunications/resources.aspx>

- Please note that the University Research Ethics Committee (UREC) is required to monitor and audit the ethical conduct of research involving human participants, data and tissue conducted by academic staff, students and researchers. Your project may be selected for audit from the research projects submitted to and approved by the UREC and its committees.

Please note that your study should not commence at any NHS site until you have obtained final management approval from the R&D department for the relevant NHS care organisation. A copy of the approval letter(s) must be forwarded to Leigh Taylor in line with Research Governance requirements.

We wish you well with your research.

Yours sincerely



Dr Julie Woodley
Chair
Faculty Research Ethics Committee

c.c. *James Byron Daniel*

Appendix C - Study Documents:

Adult Fatigue Management

Specialist help for people with enduring fatigue

T 01225 473456 F 01225 473411
E info@rnhrd.nhs.uk W www.rnhrd.nhs.uk

Royal National Hospital for Rheumatic Diseases
NHS Foundation Trust, Upper Borough Walls,
Bath, BA1 1RL



University of the
West of England

Invitation letter to participate in research

Participant's Full Name

Address line 1

Address line 2

Address line 3

Post code

DDMMYY

Dear **NAME**

Re: Long-term experiences of managing Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME): A qualitative study

I am a final year Health Psychologist Trainee at the University of the West of England (UWE), in Bristol. For my doctorate research project I am looking at exploring long-term experiences of managing CFS/ME, to improve knowledge and understanding of the condition and how to provide better management care for the future.

You have been sent this research invitation letter because you have been, or are currently a patient at the Adult Fatigue Management Service (formerly Bath and Wiltshire CFS/ME Service) at the Royal National Hospital for Rheumatic Diseases, Foundation Trust (RNHRD FT), in Bath, and previously consented to be approached for any appropriate research studies. It is highly recommended you read the enclosed Study Participant Information Sheet before deciding.

This project is sponsored by UWE and is supported by the RNHRD FT. It has received ethical approval by the NHS National Research Ethical Service (NRES), in **MONTH** 2014 (REC & ref number).

Please find enclosed the following:

- Study Invitation Letter
- Participant Information Sheet
- Study Consent Form (* only for your information, this will be discussed at a later date if you are interested in participating)
- Response Form
- Pre-paid self-addressed envelope

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Version number 1.3 Sep 2014
NHS NRES ref number: 14/YH/1135

Royal National Hospital
for Rheumatic Diseases



NHS Foundation Trust

Thank you for reading this letter and considering participating in this study.

Please feel free to contact a member of the research team if you have any questions about this study. Contact details are available on the reverse of this letter and on the enclosed PIS.

Yours sincerely

Deborah Baldwin MSc, BSc (Hons), MBPsS
Trainee Health Psychologist

Further information and contact details

UWE

Deborah Baldwin – Chief Investigator/Lead researcher

Email: Deborah.baldwin@live.uwe.ac.uk or Deborah.baldwin@rnhrd.nhs.uk

Tel: 01225 473456 (Tuesdays, Thursdays or Fridays, answer phone service available)

Study Mobile: 07779 943 899

Dr James Byron-Daniel – Academic supervisor

Email: James.Bryon-Daniel@uwe.ac.uk

Tel: 0117 32 8153

RNHRD FT

Jane Carter – RNHRD Research and Development Manager

Email: jane.carter@rnhrd.nhs.uk

Tel: 01225 465941(x224)

Laura Davies – RNHRD Patient Advice and Liaison Service (PALS)

Email: PALS@rnhrd.nhs.uk

Tel: 01225 473424

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NHS Foundation Trust, Upper Borough Walls,
Bath, BA1 1RL



University of the
West of England

Study Response Form

Study title: Long-term experiences of managing Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME): A qualitative study

Full Name (Print) **Date**

Please tick the appropriate box below to indicate your response:

1. I am interested in participating in the above study.

Please indicate below which is your preferred method of contact:

a) Postal address (unless specified we will use the postal address used for this invitation pack)

b) Telephone
if so, when are the best times to contact you?

.....
.....

c) Email

.....

d) Skype

2. I am NOT interested in participating in the above study

So we can improve our research processes, if you are happy to, can you give a brief reason for declining to participate in this study in the space below. Many thanks.

.....
.....

***Thank you for taking the time to read the study information and
to respond to the research team.***

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Adult Fatigue Management

Adult CFS/ME Service

Specialist help for people with CFS/ME

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Further information and contact details

UWE

Deborah Baldwin – Chief Investigator/Lead researcher

Email: Deborah.baldwin@live.uwe.ac.uk or Deborah.baldwin@rnhrd.nhs.uk

Tel: 01225 473456 (Tuesdays, Thursdays or Fridays, answer phone service available)

Study Mobile: 07779 943 899

Dr James Byron-Daniel – Academic supervisor

Email: James.Byron-Daniel@uwe.ac.uk

Tel: 0117 32 8153

RNHRD FT

Jane Carter – RNHRD Research and Development Manager

Email: jane.carter@rnhrd.nhs.uk

Tel: 01225 465941(x224)

Laura Davies – RNHRD Patient Advice and Liaison Service (PALS)

Email: PALS@rnhrd.nhs.uk

Tel: 01225 473424



Study title: Long-term experiences of living with Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME): A qualitative study

We would like to invite you to take part in this research study. Before you decide, we would like you to understand why the research is being done and what it would involve for you. One of our team will go through the information sheet with you and answer any questions you have. We suggest this should take about 30 minutes. We also would encourage you to talk to others about participating in this study so you can make an informed decision to take part or not.

What is the purpose of the study?

CFS/ME is considered a long-term condition, yet there is limited research looking specifically at its management in the longer-term. This project is interested in learning more about how people have managed when their fatigue related symptoms have lasted 5 years or more. This project looks to find out **what** support or interventions are helpful or unhelpful, and **if** there are different stages of the condition that require differing levels of support.

Why have I been invited?

You have been or are currently a patient at the Adult Fatigue Management Service (formerly Bath and Wiltshire CFS/ME Service) at the Royal National Hospital for Rheumatic Diseases, Foundation Trust (RNHRD FT) and have received a diagnosis of CFS/ME. You have also indicated on your clinical questionnaire, collected by the service, that you have experienced fatigue related symptoms for 5 years or more.

Do I have to take part?

Participation is completely voluntary. If you agree to take part we will ask you to sign a consent form. You are also free to withdraw at any time if you change your mind. The care you are receiving, or may receive in the future at the RNHRD FT, will not be affected in any way if you choose to participate or not in the study.

What will happen to me if I take part?

After reading this Information Sheet and talking to others about participating in this study (family, friends, your GP etc) you can complete the enclosed response form. Once the research team has received this form, they will contact you on your preferred method of contact **if** you wish to participate. Any questions you may have about the research can be answered and if you are still happy to participate, a date and time for an interview will be arranged to suit you.

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Royal National Hospital for Rheumatic Diseases
NHS Foundation Trust, Upper Borough Walls,
Bath, BA1 1RL

Confirmation of the interview details will be sent to you. If you are unwell, or change your mind about participating, you are able to cancel at any time. The researcher will go through this Information Sheet and Consent Form with you at the start of the interview. The interview will last between 30 and 60 minutes, largely guided by your current energy levels. The interview will also be audio-taped for accuracy and transcription by the researcher, at a later date. All identifiable information will be locked away at the RNHRD FT, with only authorised access and all study data will be anonymised. This means any personal details will be removed so you cannot be identified personally. You will be given a pseudonym during the interview, so your real name will not be used.

What will I have to do?

You will need to be able to attend the RNHRD **once** for an interview, for approximately 30-60 minutes. You don't need to stop any treatments or therapies you are currently taking or engaged in. It is common with CFS/ME to experience cognitive difficulties (brain fog, word finding difficulties and memory problems) so you will be sent the interview topics before the interview to assist with this. If you find it helpful, feel free to make notes about the topics and bring them with you to the interview. A Consent Form will be provided at the interview to sign if you are still happy to participate and a copy will be provided for your own records.

What are the possible disadvantages of taking part?

You will need to attend the RNHRD FT for the interview, which means we are asking you to give your time and travel. Unfortunately we are not able to reimburse your travel costs to attend the interview as there are no funds available. However, refreshments will be provided throughout the interview.

There is also a possibility during the interview you may become upset when talking about the condition and how it has impacted your life, which is something to be aware of. This is perfectly normal and the researcher is experienced in discussing sensitive issues. However, it is important to clarify that the nature of the interview is for research purposes only and not for therapeutic work. If you become significantly distressed the interview will be stopped and your GP will be notified to enable you to access appropriate support. You will be fully informed about any action taken by the researcher and additional information on available services can also be provided at the interview.

What are the possible benefits of taking part?

There is no direct participant benefit anticipated for this study. However participants' often express feeling valued through the opportunity of qualitative research to tell their stories, which in turn may help others. By taking part you can contribute to new knowledge about long-term management of CFS/ME, which can assist health care professionals to support people with CFS/ME.

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Bath, BA1 1RL

What if there is a problem?

If you have a concern about any aspect of this study, you can ask to speak to the researcher who will do their best to answer your questions (Deborah Baldwin, contact details on page 4) If you remain unhappy and wish to complain formally you can do this through the NHS Complaints Procedure. Details can be obtained from Laura Davies (Contact details on page 4).

Will taking part in the study be kept confidential?

Yes, we follow strict ethical and legal practice and all information about you will be handled in confidence. Any data which may personally identify you (name, postcode) will be kept locked in a filing cabinet at the RNHRD FT and will be accessible only to authorised people. No personal information will be taken out of the hospital, and any data which leaves the hospital will have your name and address removed so you cannot be recognised.

Only in extreme circumstances (your safety) will confidentiality be broken to contact your GP or relevant health care professionals, but you will be informed that this will happen.

All personal information will be destroyed at the end of data collection and verification. Only anonymous written transcripts of the interviews will be kept for a period of 5 years for data checking or re-analysis purposes by authorised people.

Will my General Practitioner (GP) be informed?

Your GP will not be notified about participation in this study, unless you become significantly distressed during the interview and require additional support. If the researcher identifies that you are at risk, you will be fully informed before any action is taken.

What will happen if I don't want to carry on with the study?

It is ok for you change your mind. If you let the principal researcher (Deborah Baldwin) know you no longer want to take part your contact details will be destroyed and you will not be contacted again for this study.

If you have already taken part in the interview and change your mind before January 2015 your data can be destroyed and not included in the analysis.

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After data analysis has begun your interview transcript will have been analysed together with the other participants and so unfortunately withdrawing your data is not possible.

What will happen to the results of the research study?

Analysis of the study data will be published anonymously in various different formats such as: A written Research Summary and Presentation in public, academic and professional settings. A Full Research Report will be written up as the researcher's Thesis for the Professional Doctorate in Health Psychology programme and held in the University of the West of England (UWE) Library. In addition, a paper will be prepared for submission to a peer-reviewed Journal, for publication in the public domain to influence policy and clinical practice.

To reassure you, no personal information will be used in any publications of the study results, only pseudonyms so individual participants' cannot be identified.

Who is organising the research?

Deborah Baldwin is the lead researcher for this study. UWE is sponsoring this research study as part of Deborah's Professional Doctorate in Health Psychology. UWE is an internationally respected Research University looking to lead the way forward in IMPACT research.

Further information and contact details

Research Team:

Deborah Baldwin – Chief Investigator/Lead researcher

Email: Deborah.baldwin@live.uwe.ac.uk or Deborah.baldwin@rnhrd.nhs.uk

Tel: 01225 473456 (Tuesdays, Thursdays or Fridays, answer phone service available)

Study Mobile: 07779

Dr James Byron-Daniel – Academic supervisor

Email: James.Byron-Daniel@uwe.ac.uk

Tel: 0117 32 8153

Alternative Contact details, independent of the research team:

Jane Carter – RNHRD Research and Development Manager

Email: jane.carter@rnhrd.nhs.uk

Tel: 01225 465941(x224)

Laura Davies – RNHRD Patient Advice and Liaison Service (PALS)

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Bath, BA1 1RL

Research Summary Request

Yes No

I would like to receive a copy of the research findings at the end of the study

<input type="checkbox"/>	<input type="checkbox"/>
--------------------------	--------------------------

If you have selected 'Yes', please provide your email or postal address in the space below:

.....
.....
.....

Further information and contact details

UWE

Deborah Baldwin – Chief Investigator/Lead researcher

Email: Deborah.baldwin@live.uwe.ac.uk or Deborah.baldwin@rnhrd.nhs.uk

Tel: 01225 473456 (Tuesdays, Thursdays or Fridays, answer phone service available)

Study Mobile: 07779 943 899

Dr James Byron-Daniel – Academic supervisor

Email: James.Bryon-Daniel@uwe.ac.uk

Tel: 0117 32 8153

RNHRD FT

Jane Carter – RNHRD Research and Development Manager

Email: jane.carter@rnhrd.nhs.uk

Tel: 01225 465941(x224)

Laura Davies – RNHRD Patient Advice and Liaison Service (PALS)

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Version number 1.3 Aug 2014
NHS NRES ref number: 14/YH/1135

Royal National Hospital 
for Rheumatic Diseases
NHS Foundation Trust

Outline of Interview Topics

for Participants

Meet the researcher, discuss any questions about the research and interview process. The consent form will be discussed, and if you are happy to proceed this will be signed by you and the researcher.

Fake name decided upon for confidentiality reasons you will be referred to this name throughout the interview. The equipment to audio record the interview will be tested. Then the interview will start.

Interview questions/topics:

1. Can you tell me about your personal experiences of managing CFS/ME?
2. Based on your experiences what strategies, techniques and/or support has been detrimental to managing the condition...
3. Based on your experiences what strategies, techniques and/or support has been beneficial to managing the condition...
4. If you have noted different stages, or time points in living with CFS/ME, could you briefly mention what coping mechanisms you used for those different stages
 - (prompt) did the coping mechanisms differ, or were the same mechanisms used throughout managing your CFS/ME?
5. What are your views on 'long-term' management and 'acute/short-term' management?

If time allows:

6. When thinking about management for this condition, often words like 'recovery' and 'prognosis' arise. Based on your experiences, I am interested in your understanding and meaning of the term 'recovery'.

Outline of Interview Topics

Broad opening topic:

- Participant's personal experiences of managing CFS/ME

If it has not been mentioned explicitly, the following questions will be asked, or more detail requested (order of question may vary):

- Strategies, techniques and/or support that has been detrimental to managing the condition...
 - How have particular interventions or professional input not helped you?
- Strategies, techniques and/or support that have been beneficial to managing the condition...
 - How have particular interventions or professional input helped you?

Other questions, the order may vary in response to participants' comments if naturally occurring:

- If you have noted different stages, or time points in living with CFS/ME, could you briefly mention the coping mechanisms you used for those different stages.
 - Were there different coping mechanisms used, at different stages? – Expand.
 - Were the coping mechanisms the same throughout CFS/ME journey? – Expand.
- What are your views on 'long-term' management and 'acute/short-term' management
 - have you experienced this difference, if so can you expand
 - does this idea make sense base on your understanding and experience of CFS/ME: if so, how? if not, why doesn't it fit?
 - How would you bracket, or define these stages/periods

If time allows:

- When thinking about management for this condition, often words like 'recovery' and 'prognosis' arise. Based on your experiences, I am interested in your understanding and meaning of the term 'recovery'.

Adult Fatigue Management

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E info@mhrd.nhs.uk W www.mhrd.nhs.uk

Royal National Hospital for Rheumatic Diseases
NHS Foundation Trust, Upper Borough Walls,
Bath, BA1 1RL

Participant Check List

Name:

Telephone number:

Email:

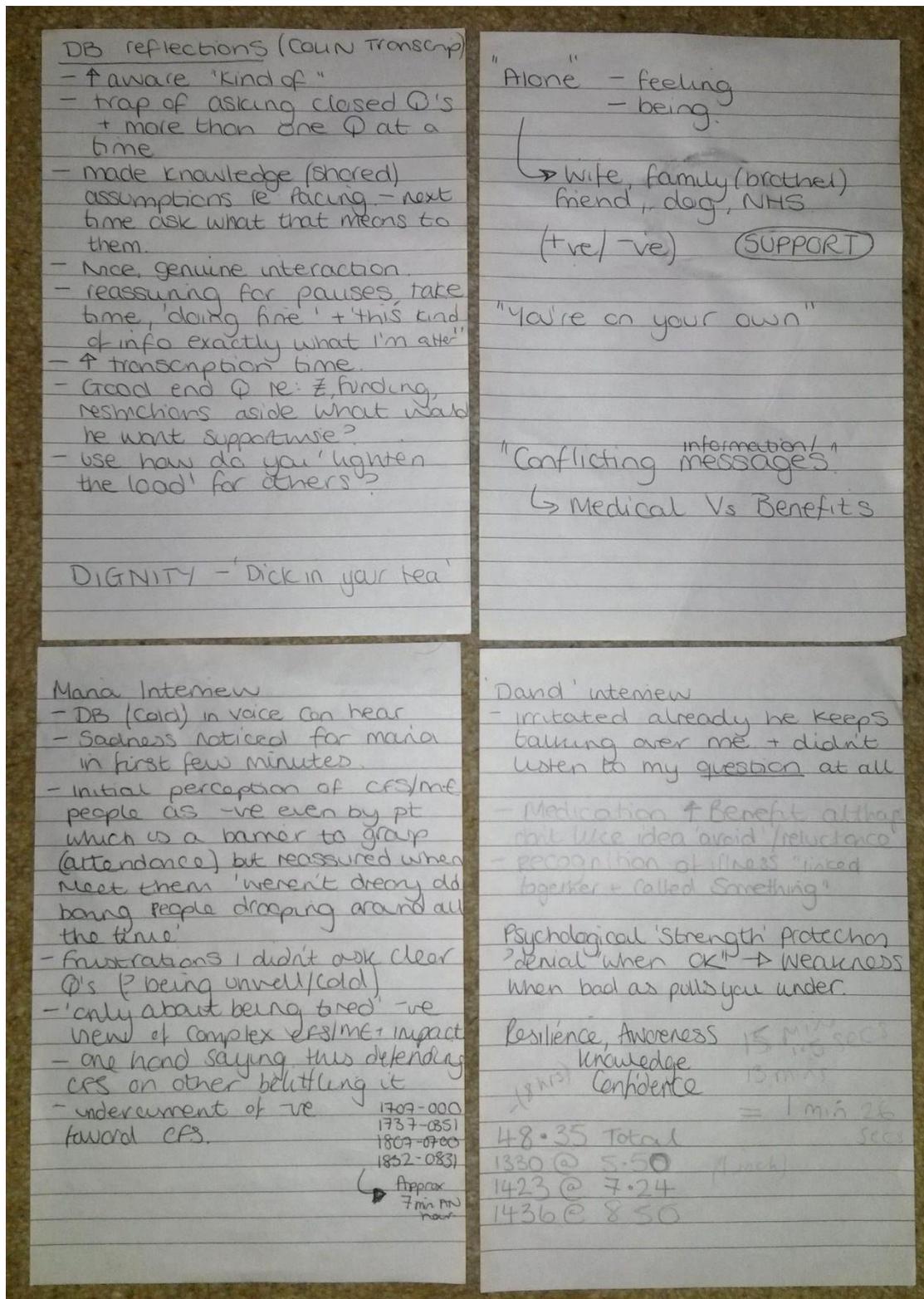
Address:

Task	Done	Date
Sent invitation		
Received response		
Opportunity to answer questions (1)		
Arranged interview		
Sent confirmation		
Sent interview topics		
Opportunity to answer questions (2)		
Talked through PIS		
Opportunity to answer questions (3)		
Taken consent (x2 copy for participant)		
Debrief after interview		
Opportunity to answer questions (4)		
Send study summary		

Notes:

Appendix D - Analysis:

- Analysis notes during transcription



? General theme re-evaluate lifestyle (job in particular) to assist with mental (pressure + wellbeing) and physical demands.

uncertainty documented previous but kind of says X would be useful such as different work options to change jobs - but at that point "too early" "you don't know how long you going to be ill" mona

- Get to 'd level - accept, - aware of energy/achieve/limits, some small variations but pleasure "things can only improve" mona

↓
getting to this is a 'stage'
Milestone

How language is used to describe experiences, view points and information can't be ignored in qualitative research methods - as this is the data. Equally the researchers language in interpreting participants' data is also important - yet when does this become discourse analysis?

- ① Self awareness + learning
 - ② Guided by values + 'limitations'
 - ③ Flexibility, adaption + balance
- overlapping

* learning + awareness

* NO one size fits all for symptom, problem, person all context/situation + time specific

initial? "for however long
'ADJUSTMENT' = you
- wife
- roles
- physical

(↳ values, goals etc)

Purpose - Contribution

"listened to" - "that
Somebody gives a damn"

④ 1507 @ 9.32

adrenaline - "not real energy"
→ parallels to stress response
↳ links to "deep, deep relaxation" + "good quality sleep"

impaired at first talking over me - warmth feeling towards him - pride when I comforted him / commended him - his genuine thanks Nice example of human empathy interaction + understanding

↓
I knew separate from therapy but still exchange + interaction important to (P + T)

- Table to organise grouped code

Initial groupings	Summary of grouping/ Interpretations	No of code
Treatment experiences	Variable accounts of Rx type, delivery and X. Negative and positive experiences and beliefs shared yet two potent themes: Availability and timing of Rx of interest - Links to RQ is there any Rx relevant for different points - YES, and research would suggest these 'times' aren't illness duration but more closely related to acceptance and existence of committed action. (also links to CONTROL) *Adjustment, Illness beliefs, acceptance, psychological state. Concrete and abstract practical support like housework/cooking/finances is concrete and abstract is mental wellbeing and acceptance. Some talk about the recognition health care is limited practically and financially. GP experiences & beliefs: Mostly unhelpful experiences shared including beliefs GP/medical Rx limited or unhelpful	47
Difficulties implementing	knowing what to do, but sticking to it hard and pushing on tempting. Also idea pacing/life is boring & frustrating	8
Fluctuating & Uncertainty	Nothing new here - difficulties as lots unknown including when relapse/set backs will happen.	14
Unknown/ Uncertainty	Nothing new here - Stress has unknown effect unlike over doing it.	6
Triggers/ What is known	Past experiences useful, triggers known over time and through trial and error - takes some figuring out	23
Illness trajectory/experience	(individual) commonality of extreme variability at start often responded to with increased sleeping as illness progresses energy levels off and can become more predictable/controlled through behavioural and lifestyle changes and support.	33
Prognosis & Recovery ideas	Strong belief coming through believe condition is long-term largely through own experiences 'had it longer than I haven't'. With this a sense of 'normality' has been achieved. Expressions of doubt of other recovering - either thinking misdiagnosed or it will return in the future. Recovery mostly often conceptualised as symptom free and living a life not limited by symptoms. (perhaps this is why better adjusted/effective management strategies targets achieving a full life within the condition limitations)	(13+27) 40
Long-term belief	As above a really strong sense of life long condition (n = 4). Also talk of acceptance and adapting as a long process, practical examples given as changing jobs.	14
Causal ideas	Virus+stress and one participant talks about personality as a family predisposition - fits with idea of the recipe for CFS/ME and/or Precipitating, Perpetuating and Predispositional	6
Change in coping overtime	Life and coping: developed and was learnt over time often using past experiences. Earlier in illness experience responses different. (N=5) talked about these changes overtime and mostly related to learning about self & illness overtime to be able to make changes.	14

Choice?	Recognising personal control: that choices and decision making is possible, although at times the options available aren't ideal.	11
Acceptance	Unanimous talk about the need for acceptance for better quality of life.	23
Acceptance (Acceptance Vs Fighting + Denial)	Conceptualisation constructs are different and the dichotomy 'you can't have one without the other'. Shared belief fighting was carrying on regardless of illness and parallels made with denial. Largely assumptions (beliefs and past experiences) fighting it doesn't work.	7+7 (14)
Adapting (for the best possible experience/outcome/life)	Being flexible, open and adapting to illness and self, essential to 'achieve best standard of living'	23+12 (35)
Analogies, Metaphors and Sayings	Almost all (n=8) used A, M and/or S. P7 particularly and was directly asked the function of this vital to explain something so 'alien' - attempt to find shared understanding to convey meaning to others (DISCOURSE)	19+
Pets	Having company and purpose as dog owners.	2
Awareness	The need for awareness/familiarity/noticing was an important foundation for change/life/adjustment. Interestingly a participant spoke about being able to differentiate normal sensations and ME sensations - perhaps this awareness enables more positive adjustment/acceptance/LIVING and the difficulties of knowing and 'being honest with yourself'	13
Knowing limits/Awareness	Learning limitations through past experience. Difficulties raised about ignoring inner voice (/natural) to push beyond limitations.	9
Judgements about management	An interpretation was made on several occasions that participants voiced judgements about various management/coping approaches. Either claimed as their belief (through experiences) or what from information they found/were told. Some of these judgements were implied personally and some to how others' with CFS/ME cope (exercise, not helping selves). Some map onto social constructionist view (chores first, rest later and selfish to focus on self. Both these were from P5) but largely fit with learning/behavioural assumptions.	17
Self portrayed to others	Fuels 'hidden' invisible nature of CFS/ME when participants spoke of Proving they are ok/attempts to be normal to themselves and others. *LINKS to fighting/not accepting/denial but perhaps more 'healthy' spins on this could be increase committed action/valued living/behavioural engagement and not withdrawing.	4
Being different	Impact of CFS/ME on social and development meant that feeling different from others was thought. One participant also linked identity with being different as thought ME was severe and therefore she couldn't have ME as this contradicted her thoughts. *LINKS with normality/identity	9
Normality	LT experience of CFS/ME becomes 'normal' for participants and this process assists with coping (not searching, fighting, explaining to others, working around limits). There was interestingly one participant that spoke about chronic illness as the norm in the household . Largely this theme fits with Identity and Acceptance. There was also a significant role for normalising and reassurance which participants reported being important for LT management.	22

Valued activities	Enjoyment important so engaging in VA important sometimes this requires low energy VA options (ADAPTING). Value placed on process not outcome and with this a sense of appreciation - this appreciation may have been lacking before unwell or/and has been developed OVERTIME. An acknowledgement was also made by one participant that changes in VA could be a result of other factors (age) not CFS/ME.	13
Natural coping	New idea of natural and un-natural coping. Differs for indies but seemingly Rx options/approaches that come naturally to people enables quicker adjustment to life with CFS/ME and also requires minimal input from professionals - whereas if Rx options/approaches are not natural = more struggle and more input/support needed to facilitate. The latter seems to be the more commonly reported stating that techniques are developed or learnt over time. Also LINKS to personality, adapt, changes overtime, normality.	38
Complicating factors	Many participants described the complexities of comorbidities, environment and x on effective management. Complicating factors also related to receiving the right support at the right time to suit the individuals' needs.	22
Comparisons	Comparisons frequently made with previous self/abilities and others' abilities. Only one participant reported not making any comparisons as this was not helpful. Comparisons were also made to explain physical symptoms and energy using metaphors, analogies and saying , ie Flu like symptoms, energy as mobile phone battery.	9
Compromise	Compromise occurs and the loss and disappointment involved is difficult, but if an acceptable compromise is reached then balance can occur. Links with Choices, Changes, Valued action, learning, cost/gains and consequences	12
Costs and gains/ Consequences	Decision making based on costs and gains was evident. Often participants talked about whether activities were 'worth it' in relation to 'payback'. It was also acknowledged the emotional impact of not doing a high cost activity - life is boring and stressful which feeds negative emotions	26
Confidence	Self-confidence was identified by participants as helpful for long-term management. Defined as 'strong mind/mental strength' to be able to act confidently, for example saying no, remove any unsupportive friends, change job overcome fear of symptoms	17
Control	Control over self: mind-set and activities. CBT model applied well to sense of control and supports effective interventions - figuring out what element is in control BS, T or F. When one of these dominates action sense of out of control is experienced which doesn't help long-term management. Structure highlights as way to control and empower. LINKS to acceptance, identity, Valued action, adapting, choice.	31
Battle	Internal battles raised as difficulties not to give in or fight against it. This fighting and pushing on was considered bad management and required strength in mind to overcome	10
Empathy for others	Interesting idea empathy for others' lack of understanding when difficult for person to understand themselves	4
Perceptions of others	Nothing new here. Concerns and stress experienced due to stereotyping and negative perceptions others will have. Based on limited activity, invisible illness and fluctuating nature. Assumptions and judgements based on physical appearance that aren't true due to illness (male looks tall and strong - unable to lift objects). Some participants also acknowledged assumptions of other people with CFS/ME - they are worse off.	18

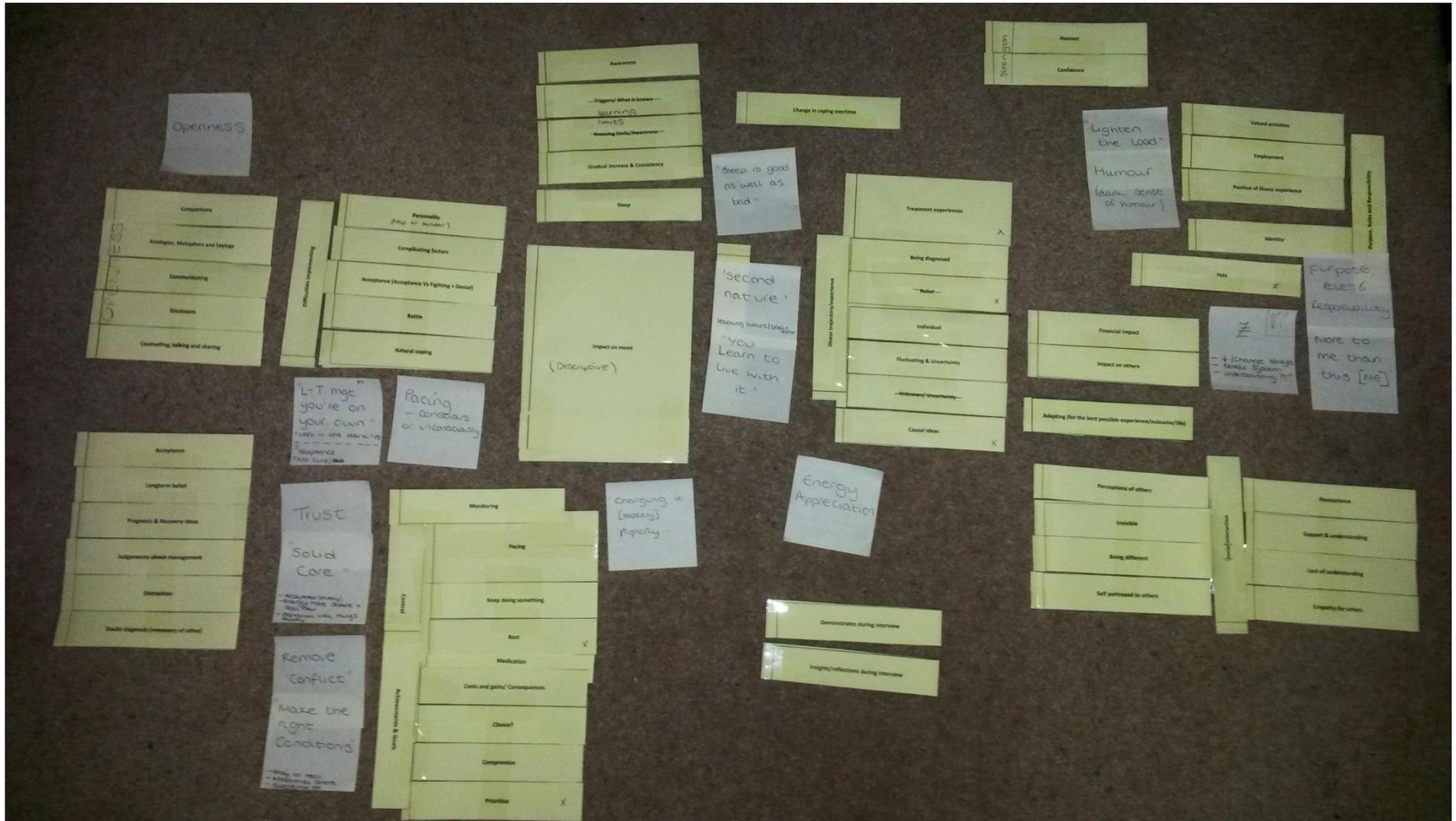
Lack of understanding	Nothing new here. Not understanding the condition prompted participants to feel empathy with others not understanding it and also was a barrier to getting on with their life. (denial, control, acceptance etc). Protective behaviour of avoiding or removing those people who didn't understand if possible (not direct family member) was described as a positive step in effective management	26
Social interaction	Social interaction was conceptualise as stressful but was valued by all participants to combat sense of isolation (common in population). interaction with others with CFS/ME can be reassuring and a way to share information. Withdrawing from others tended to occur if they were unable to relate to them or did not have the energy required to interact.	20
Communicating	Communicating difficult and impacts relationships. Helpful if able to communicate experience and ask for help, although not disclosing can occur.	9
Counselling, talking and sharing	Positive impact of sharing experience by talking out-loud or to someone else (friends or HCP) sense of offloading useful in long-term management. Relevant to interview process as this was reflected on as reasons for taking part and reflection during interview as helpful to talk about experience. Openness/accepting/non-judgemental + practical problem solving.	10
Disclosure	Divided approaches to disclosure reported that varies based on situation. Honesty and openness was favoured for reduced emotional consequences. Some functional aspects to not disclosing such as reducing unpleasant interactions/environments and being selective about what is shared and with whom.	5
Employment	Participants that raised employment spoke of the role and purpose achieved through employment and the negative impact of not working (finances, lifestyle, marriage). It was also conceptualised the job should suit the person not the other way around, one participant identified job prior to CFS/ME was not possible so changed to alleviate pressure (self and others).	20
Financial impact	Difficulties noted from the benefit system. Impact of inability to work and the reduction of money had a dramatic effect for some participants resulting in significant lifestyle changes and effect on mood (relationship, marriage breakdowns and suicide attempt).	9
Information seeking/ avoidance	Some variance in information seeking and avoidant behaviour - both scenarios reflected participants deciding for themselves what was most helpful. More participants chose not to engage in information seeking and followed a more individualised approach taking on relevant information to them and figuring out what works/doesn't work for them. (trial and error, acceptance, awareness, confidence)	14
Being diagnosed	Experiences of being diagnosed with CFS/ME relate to known experiences that diagnosis is difficult with feelings of upset, doubt and un-satisfaction. One participant expressed feeling of validation having diagnosis (anecdotally short-lived relief after diagnosis).	7
Distraction	Function of distraction conceptualised as bad management but required for some form of life	1

Doubt diagnosis (+recovery of others)	Doubt of diagnosis often result of experience not fitting with belief - unable to understand, for instance not fitting pattern, medical model of illness. Largely this doubt occurred earlier in fatigue experience when making sense (links to quest, chaos and restitution phases). Interestingly doubt was present towards others who say they have recovered.	11+2
Purpose, Roles and Responsibility	Purpose, roles and responsibilities are significantly disrupted having CFS/ME. Pushing to fulfil commitments is tempting but at the expense of condition management. Balance was encouraged to have purpose, roles and responsibilities but at the right energy level so this didn't become detrimental longer-term: One participant unable to work but desire to have purpose redefined role at home with spouse to suit energy levels and desire to contribute.	27
Humour	Humour was described as a positive coping strategy to combat low mood and functional limitations of condition. It was considered essential by one participant 'you need a pretty dark sense of humour' to make light of CFS/ME a taboo subject.	4
Identity	Participants described a shared experience of creating a sense of identity over time that either including CFS/ME as a part but not the whole, or that placed lesser focus on CFS/ME identity. The latter of these approaches included participants not meeting with others with CFS/ME or engaged in information/research seeking/health care support. A consensus was implied that CFS/ME despite the great impact on people's lives was not the only focus.	13
Personality	Personality factors had been identified by participants as helpful or hindrances in long-term management of CFS. The former includes being able to be independent and motivated to look after themselves and the latter included things like being impatient, putting others before themselves and having high standards/perfectionism	21
Support & understanding	High value was placed on received support and understanding from partners, family, friends, HCP and wider society. Support mostly related to emotional support but financial support was experienced from wife. Stress was experienced if support and understanding was not sought and resulted in removing unsupportive people as protective factor.	34
Impact on mood	CFS/ME significantly impacts on mood, at times participants describe being overwhelmed and resentful of the impact the condition has. Many participants believe there is mental and physical link, but reject psychological disorder as the cause for illness. Overwhelming and powerful were ways of talking about the condition impacting mood - one participant attempted suicide. Giving into the mind or symptoms was considered a non-effective solution and long-term management was developing strength of mind, humour, getting psychological support, optimism and doing enjoyable activities as long-term strategies to tackle low mood.	26
	Frustrations were experienced towards others with CFS/ME that didn't help themselves and HCP arranging Rx they were unable to attend. Wider frustrations talked about related to resenting CFS/ME for the stealing precious moments (with grandchildren etc) and that life is boring (no longer limitless)	5
	Stress was unanimously talked about having a negative impact and that emotional stress has an unknown impact unlike overdoing which the effects were more predictable and short-lived.	6

	Emotional stress was mostly self-directed, felling guilty, blame self and disappointment. Some talk of emotional stress related to others, treatment by the benefit system, friends who didn't understand and resentment towards CFS/ME for impact on family.	17
	Disappointment was raised specifically when cancelling plans and letting people down. More general disappointment related to loss of independence and impact on life - changes in relationships, financial and lifestyle. One participant also shared feelings of being let down by HCP being 'dumped' and not supported more.	16
	Self-blame was offered as explanation for having CFS/ME or relapses. One participant spoke of causation as 'karma' for the things previously done.	4
Individual	There was a shared belief that CFS/ME is an individual condition and many participants made reference to their experiences by saying 'for me' 'for others it might be' etc. As a result of viewing the condition individually one participant did not relate to treatment as it was views as female-focused.	21
Invisible	The majority of participants mentioned on numerous occasions the difficulties experienced due to the limited physical 'symptoms' that others could recognise including being in bed not seeing anyone when at the worst. One participant described having only one visible symptom of uncontrollable sweating which was embarrassing but useful as people can visible see an abnormality.	8
Insights/reflections during interview	Reflections offered by participant by taking part in the research illustrated reflective space for participants to experience benefits of sharing their experience and allowed for new insights into their experiences.	7
Demonstrates during interview	Instances of embodiment noticed by the researcher as additional demonstrations of what participants were saying, such as using the interview for information gathering, seeking reassurance, using humour, contradicting self (complex management, individual) and becoming muddled (symptom)	6
Reassurance	Comfort/compassion, validating and normalising identified import generated by self and sought from others. Having strong mind, discipline and optimism for themselves and from others to have understanding were important for long-term management	16
Medication	Medication was reported as helpful (n=6) with the most commonly reported anti-depressants and pain killers. All the males participants found medication to be helpful with only half the women reporting using medication. *medication use was not asked so could be under reported - but it was assumed to come up in discussing useful/not useful management.	9
Monitoring	Continual need to notice/watch/monitor energy. Highlighting stability and sticking to level (baseline) and only increasing steadily so there wasn't a detrimental increase and risk of setback. Physical prompts were used to assist with this: logging activity and using 'wall of wisdom' for reminding	16
Impact on others	All participants mentioned the impact of their condition on others. Often talking about the emotions felt (guilt, resentment, depression) and mostly the impact on their partners and close family. Through re-negotiation of roles, communication & sharing (through counselling) participants had found their way through this difficulty. Impact is long-term therefore long-term management would need to address this.	17

Keep doing something	Majority of participants held the view it is not good to do nothing 'lying around is no good' nor is it useful to do too much as this increases symptoms. Many participants directly or indirectly spoke about pacing their activities. Motivation to keep going and identifying lower energy activities they enjoy is useful for long-term management *links to balance	14
Gradual increase & Consistency	Almost all (n=8) explained how important consistency is for long-term management. Promoting maintenance as the goal OR gradual/steady increases of activities. Pushing beyond limits although tempting is not helpful. Consistency was also talked about in external ways such as routine and temperature consistency.	24
Sleep	Nothing new here, but perhaps noting more sleep during earlier months/years of diagnosis as either common coping/response AND-OR natural disease progression. Role of sleep. Sleep for one participant was described as 'enemy' or 'best friend'. One participant reported melatonin was helpful in regulating sleep but only during interview reflection made the connection it was helpful.	10
Rest	Quality of rest emphasised 'actual rest' including relaxation vital for long-term management. [surprising only 3 participants mentioned rest]	7
Pacing	Almost all (n=8) mentioned Pacing experiences and beliefs. Participants described pacing as spacing out activities with rest. Somewhere using pacing principles as defined by the other participants but didn't name it. It was considered a good strategy but not practical, difficult to stick to and was boring.	21
	Planning was discussed in combination with pacing and managing the fluctuating/uncertainty of the condition.	7
Prioritise	Prioritising each when choices weren't ideal was highlighted by some participants as useful. Thinking about what can and can't be controlled	4
Achievements & Goals	Setting and reaching goals acted as a way to evaluate success (monitor), gain control and build. It was emphasised goals should be small and one participant discounted far away goals. Goals and achievements were motivating and promoted a sense of control for participants. Although providing an opportunity to build goals were also relating to maintenance.	17
Positive of illness experience	Positive experiences shared from having illness included a shared sense of appreciation/valuing/enjoyment which wasn't present before. One participant also viewed the outcome as survivable so a positive of the condition.	5
Relief	Relief related to diagnosis when participants felt validated as unwell and relieved not life threatening nor psychological condition	5

- Sorting grouped code to create themes



- Reflective journal entries:

30.12.14 Research Activity: Transcribing

Transcription is taking for ever, I feel so slow at typing. It's really hard to make out some words too. I find myself getting irritated at listening to my pattern of speech – at times I'm so unclear this is annoying JUST SAY WHAT YOU MEAN?!?! Learning point for sure – take your time and be clear when speaking in life, in general. Also finding myself getting annoyed at a participant for talking over me, I'm barely saying anything, anyway (not like normal! actually that's quite hard to bite my tongue I really want to say loads, as I would do in Clinic, guess that's the point I'm not in clinic I am not trying to connect in that way I want a more one sided conversation). But anyway being interrupted when I haven't even asked the question is annoying! Another learning point, let others finish before butting in, or talking over them. Perhaps when I think about myself, as I am guilty of talking over, or interrupting people I worry I will forget what I am going to say...with CFS/ME memory can be an issue, so perhaps that's what was happening, that makes me feel more calm, and seeing another perspective is important.

10.03.15 Research Activity: Transcribing

I'm feeling really motivated (despite transcription taking ages, as I knew it would) I am jotting down notes and seemingly getting ideas all the time. I can appreciate why transcription and listening repeatedly to interviews is an important part of the data analysis.

04.05.15 Research Activity: Transcribing/ Analysis

I have a slight concern that I am not finding anything that surprises me, all the data is known to me from my clinical role and I'm worried my findings will not be novel and exciting. Yes that might be the case, but you are undertaking research with a group of people that haven't been the focus of research. Just because it's not surprising to you doesn't mean it's not important to report, others may not know this – or take for granted this. Publication of the findings can help this group of people's stories to be heard. And it's a good thing that your ideas are being confirmed, backs up your clinical work. Yes but watch out too, you might be 'seeing' what you know anyway and missing information that doesn't fit. Keep coming back to this, and use Advisors, AFMS team and JBD to help minimise any bias – if at all possible.

02.06.15 Research Activity: Analysis

There is so much data, I'm struggling to find my way. Serious doubts as a researcher, I should be better at this. What if I don't report my participants stories correctly and do them justice. Feeling inadequate and useless, perhaps time to step away – I also acknowledge I'm stressed with wedding planning and when I'm analysing I'm giving myself a hard time I'm 'wasting' my time not doing quality work and not 'doing enough' wedding planning either! I can't seem to win.

08.06.15 Research Activity: Analysis

Reflecting on previous week – what is 'correct' anyway. I will 'see' what I 'see', this is the whole point of qualitative research, I am guided by my experiences and knowledge

and the interaction in the interview. On a different day the two of us could have come up with completely different things, so put down this 'perfect, correct' way of doing things as it's not a helpful benchmark. Acknowledge you are stressed, there is a lot to do, but the reason you are stress is because it's important to you! Enjoy the process. As you have done talk to JBD and colleagues to get perspective.

10.08.15 Research Activity: Analysis

I'm just getting so confused! Everything relates to everything I can't seem to get distinct groupings or Themes. I've tried physically sorting the grouped code but getting no where. I'm concerned I won't finish in time and be able to present at the DHP conference, or meet my deadline. I feel pressured and yet again feeling stupid, why can't I figure this out.

I have a plan to talk through with my friend who has no knowledge of CFS/ME to try to get me to refocus on the key points – lay explanation, how does that differ to these complex webs I seem to have.

I will also go back to the literature to look for guidance on what is known already in terms of my data code – even though I don't want this to influence me greatly I need some help to separate.

20.09.15 Research Activity: Dissemination of findings (BPS DHP conference)

The instances of embodiment that was observed during my interviews was presented within a professional arena at the Division of Health Psychology Annual Conference in London on 17th September 2015. After talking with my clinical supervisor who found it really interesting I chose this as my topic for my presentation. Originally billed was my study findings, but I'm just not there and so talking about the embodiment, that probably won't feature too heavily in my write up seemed a good subject to talk about – and fun! I was concerned a standard INTRO, METHODS, RESULTS, DISCUSSION talk would be really dull – so many of them happening, I'd like to be different. I suppose my first DHP conference I want to make an impact too, if I can.

I wanted to share this part of my research as I knew it would not take the foreground in my research write up but I think it was important to share as finding this unexpectedly has really made a difference to my clinical work. I now seek out opportunities to use any instances of embodiment in my clinical work to highlight the work we are doing in 1:1 and group work. Using real examples and specifics to work with is useful I find.

I was really pleased that my talk had sparked the interest of several people, I had fears that only a handful were going to attend – and seeing some talk sizes were quite small I was pleased with the turn out (about 25-30 people). I noticed a few familiar faces from the days preceding but I hadn't met them, and I'm really relieved in one case I hadn't met them yet, as it turned out at least one audience member has an established career in CFS/ME practice and research so I would have been even more nervous if I'd have known who they were before-hand.

I was harsh on myself appraising the success of the talk as I skipped through the slides towards the end in fear of running out of time yet I had a full minute left in the end. I also felt my answers to questions were not answered to the best of my ability, yet feedback from two of the audience members (one of which had asked a question) told me the following day they thought I answered them well. This was a boost and I was really excited to share this more unusual study finding with peers. It sparked a conversation about the use of alternative media in qualitative research to capture such instances to build the depth and quality of the findings. I definitely think my research, presenting this element at the conference and being able to discuss my research in the way with my peers was immensely valuable. I certainly think my research abilities are progressing and the quality of my research in the future will undoubtedly benefit.

12.10.15 Research Activity: Analysis (Supervision JBD)

I am encouraged that when I explain, JBD said it made sense...but it doesn't make sense to me – all these connections between each code. Be cut-throat, organise what connects with each other, remember you are telling a story and it's ok if there are links, if that's what the data shows.

I think it was good I didn't read too much of the literature, as there was a concern it might influence me more than the data.

12.10.15 Research Activity: Analysis

I don't know if it's being in London, and enjoying the FACT workshop but I can see my data maps onto this model so clearly. This was really unexpected. After so many months of grappling with the data and fitting into broad themes, I can see a therapeutic model is workable, and fits with the literature. This is new and exciting as very little research has used ACT in CFS/ME and even less research using FACT. I'm feeling so motivated and excited to write up and talk to others about this. It quiets the voices in my mind of doubting the usefulness of my research, as it is an under researched area and it points to a very useful therapy model not explicitly used in CFS/ME. Really exciting. This is what is about! Yay! It's such a roller coaster of a ride doing this Doctorate/Thesis.

• Table 4: Overview of themes and sub-themes with illustrative quotes

Theme	Interpretation	Illustrative quotes
THEME 1 AWARENESS	Listening to body and mind, not to ignore experiences, which has developed over time.	<p><i>'Well its-right at the top of the triangle is self-awareness um, learning, to listen to your body and your mind and, not denying that, that might be too much to do or that's ok and you know it's-it's about learning to to to read yourself' (Diane)</i></p> <p><i>'one of the things I've done is to learn to put myself in the situations or avoid those situations where I can't pace or put myself in a situation where I've got a recovery period so there-that's sort of a longer-term strategy' (Stuart)</i></p> <p><i>'it's something I've learnt I mean I've had it for I don't know eight nine years and it's only really this last three or four I suppose that I've started to become, more aware or, myself, aa-and how this condition affects me and others yeah. It's taken a long time (smiles)' (Diane)</i></p>
	Developing heightened self-awareness useful in relation to quality of sleep, rest, impact of routines and identification of triggers or warning signs.	<p><i>'this last couple of weeks is a c-primary example actually (smiles) I did too much and at the weekend I had to just cancel everything for three days and that's not managing it actually that's boom and bust' (Kate)</i></p> <p><i>as much as sometimes you feel sleep is your enemy, it's actually your very best friend [...] you do have to know, when to rest, and when not to rest um, so yeah I think, thinking that you have to be awake is, is not right that's probably the worst thing you can do I think you just have to realise that, sleep is good as well as bad.' (Jane)</i></p> <p><i>great temperature differentials [...] Bang like that and even fairly moderate physical exercise [...] then I'll start within five minutes and I'm drenched [in sweat]' (Oliver)</i></p> <p><i>And also when I went to [HOSPITAL NAME] I was taught and learnt that what I thought was rest isn't necessarily rest (laughs) [...] so you know watching EastEnders apparently that's too stimulating (laughs) and actually it is, erm and I do lis-just sit and listen to music and stuff a lot more now like if it's just peaceful umm, yeah things I, put as, rest were actually low level activity so I just never stopped and now I do try' (Kate)</i></p> <p><i>'I don't know whether it is adrenaline or not but something happens and it's like there's a high before a low [...] but noticing, I now know that if that happens [...] to prepare' (Kate)</i></p>
	Awareness of constant monitoring of experiences to make decisions, also including the ability to notice the urge to push beyond limits when feeling well.	<p><i>'there's been a battle between psychological and physical aspects of management um one doesn't necessarily help the other um so for example when I've been physically resting my body because it's absolutely necessary it can render me sort of feeling depressed because I have feelings of helplessness and uselessness and inadequacy and also sort of wasting time I know my life is slipping away (cough) er but on the other hand if I'm doing that's benefit psychologically then, it can tired (laughs) me out physically (laughs). So there's a two edged sword at sometimes and I have to try and manage one with the other' (Diane)</i></p> <p><i>'I'm really careful about not doing too many other things in the week so I look at my diary at the beginning of the week and think 'oh I'm working on that day that day and that day, so therefore I'm not to do anything social like meeting people for coffee or something during the day I can't do that because I need those days off' [...] but then I'm not very good at managing on the other hand because when-I've got two kids and dog and all that, house, um a partner and when I've got the day off I tend to do loads of jobs I do the washing I do the shopping I do the, faffing around sorting things out and I'm really really bad at stopping and that's why when I do stop I fall asleep in the afternoon so I think I'm pretty poor at managing my symptoms [...] I can't pace to save my life my way of coping is to do everything you've got to do and then you collapse [...] maybe it's helpful but I don't have much of a life' (Maria)</i></p>

	<p>The impact of the awareness of others, asset or a challenge.</p> <p>Although an invisible illness it could be 'seen' by others which was validating.</p>	<p><i>'difficulty in reconciling what I know of my abilities and limitations and convincing my employer-so [...] my own colleagues at work sort of now understand that I have this thing and they are quite understanding but maybe the upper echelons of management and HR who have this policy to operate over the whole of the organisation and because they don't know me personally, I-I I feel, sometimes it's a struggle to to get them to see how difficult things can be for me' (Diane)</i></p> <p><i>'My dad found it very difficult 'cause he was a teacher so he found it very difficult when he came home from work every day and saw that I'd not been to school and, but he got it know he-he spent some time with me and he-he got it quite quickly, um understood it um (tut) yeah.'</i> (Jane).</p> <p><i>people look at you and I'm [height] tall and quite broad and my you know I don't look too awful (laughs), uh and people don't see it as debilitating as it is. [...] I mean the thing is as soon as once people get to know you once they see you regularly they can see it 'cause it's pretty damn obvious (laughs) you know you-I I go grey and I'm literally drenched in sweat' (Oliver)</i></p> <p><i>'I looked perfectly fine 'cause I-I'd-I've never really looked terribly ill which is part of the probably nobody um and if I did look ill I just wouldn't have gone out so people go 'oh you look really well' and I was like 'well doesn't mean I feel it' you know it's not my fault I don't have spots or a bandage or anything' (Jane)</i></p>
<p>2. Acceptance</p>	<p>Openness to experiences that included not fighting with the symptoms, diagnosis in an internal battle. Also reported to develop over-time.</p>	<p><i>'you have to, have to accept you've got it, which is not easy because it's only diagnosed by default [...] people who have had it for a long time then I think that all they can really expect is to, w-m-manage it as best they can, and you know have a very gradual um prevent flare ups' (Stuart).</i></p> <p><i>I can imagine it's very easy to be completely consumed and, erm finding cures and things like that but, I mean at the end of the day if it's not going to kill me I'll just live with it' (Jane)</i></p> <p><i>'Accepting it that was a massive one that I worked on at [Hospital name] erm, but it's myself and others but for me it was massive. Wasn't sure I believed it myself so how could I expect other people to believe me' (Kate)</i></p> <p><i>'it really has only something that that's I've learnt to do, since, not since I've had the condition but since I've learnt to live with it (laughs) [...] umm I suppose at the point, when I realised that boom and bust situation was, not giving me a quality of life that, I need ed or-or my family you know my children and also it wasn't doing my work ethic much good and as I say I really love my job and I don't (laughs) wanna get the sack (laughs)so when all those things came together and-and I started to realise that, um, I shouldn't be so tough on myself and-and, start, being kinder and one of the ways of doing that was becoming aware of what I could do and what I couldn't do' (Diane)</i></p> <p><i>'I was waiting to get better and I didn't get better [...] because if you wait to get better you will never get better, you know' (Maria)</i></p> <p><i>'and sometimes you can't do everything you need to do (laughs) and I try not to get upset about it I'll just kind of put that on and spread it out, and also I try not to be too specific I don't say 'oh, yeah I'll get that done tomorrow' I try and plan it a bit more globally than that so (laughs) I can actually say 'it's the end of the week oh look what I've managed to this, this and this'. (Oliver)</i></p> <p><i>'Umm the major thing is me and my acceptance that I can't do everything' (Kate)</i></p>
	<p>Achieving a broader perspective that included not stopping completely but</p>	<p><i>'On a practical level changing my work pattern so at the time I was doing different days and being aware that, I can only, I-I-I find morning really hard so I've changed my working pattern so I start later in the morning I've got a day in between each day at work so those are the practical things so you need to be have the confidence to, approach your employer and and find a compromise' (Diane)</i></p> <p><i>'people need to be told fighting it is not a good idea, you need to manage it quickly preferably before it fucks you up so much you don't necessarily lose you job maybe-you could manage it and still keep your job even if it means cutting yourself to sort of fifteen hours a week [...] I mean it's-cause it's a I mean it's it's hard to get back into things once you're out'. (Oliver)</i></p>

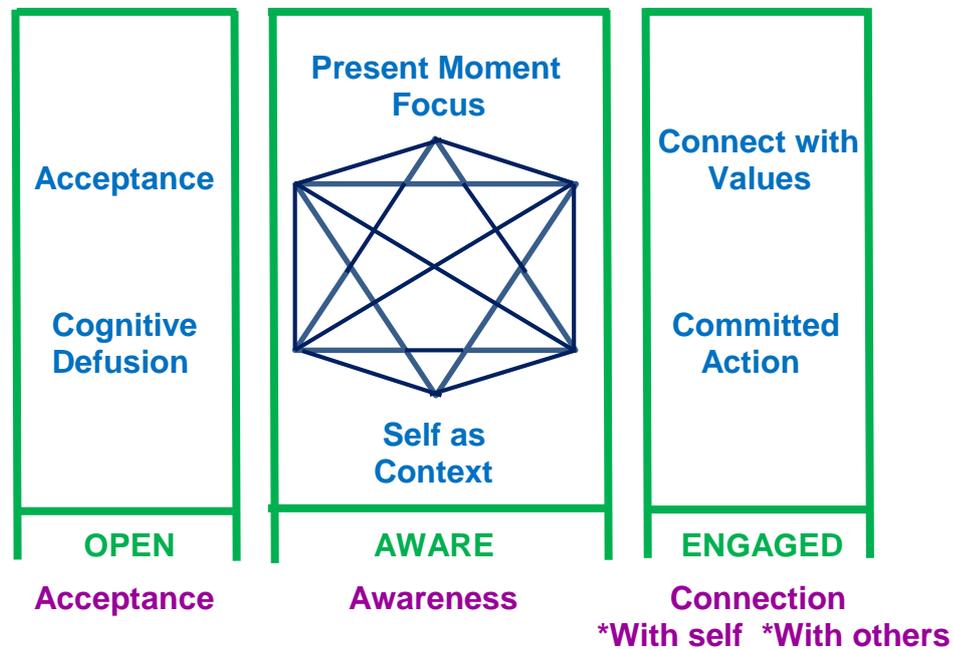
<p>accepting and adjusting to reduce impact on life by being flexible. Also including need and benefits of validation and understanding</p>	<p><i>'I don't work full time I only work part time but then that means I get a bit of a social life as well so, 'cause when I worked full time there was just no social life at all so it's just a balance' (Kate)</i></p> <p><i>'I didn't feel alone anymore there's somebody that gets it well a team of people that get it' (Kate)</i></p> <p><i>'I-I found that um understanding what it was gave me some kind of relief so , y-you know i-i-it was a relief to know it is something that exists and you haven't made it up.' (Stuart)</i></p>
<p>Recognition of the individuality, similarities and differences with others to accept life challenges and condition challenges.</p>	<p><i>'I didn't realise that anything was really wrong and then I lost my mum and it all kicked off then and I thought 'well maybe there is something not right' (Alison)</i></p> <p><i>'have a routine in my case it's like being going to bed by nine and getting up at half past seven and that helps a lot 'cause if I lay in in the morning even just for half an hour I'll get a migraine or I'll be all over the place all day' (Alison)</i></p> <p><i>'each person has got their own sort of symptoms with it and their own way of dealing with it' (Alison)</i></p> <p><i>'I think it's such a, well for me that's what I've experienced it's such an independent thing to go through and not one way is going to work for everyone 'cause people have different ways of coping slash different lifestyles and, different expectations of what they want to do and how fast they want to get better so I think it's as long as you get the right course that's right for you which obviously is, half, personal and half the help you get. You can have as many doctors telling you to so as many different things and not necessarily to them' (Penny)</i></p> <p><i>'I'm getting to the point now where I've got to slow down I've got to have me, some discs removed in my back as well in a couple of weeks' time I've had my hip joint replaced, so I'm gradually wearing me-self out, so you know, I'm having to slow down. But it's not something I like. No, th-that's that is the problem' (Colin)</i></p> <p><i>'then it was the ME that come out of that [...] because I had that much pressure it's taken me-it's like a balloon going down, with a tiny hole in it-it's taken all that time for the sort of pressure to equalise to a point (breath) to enable me to get back to some sort of normality (laughs).' (Colin)</i></p>
<p>Conceptualised as a life-long illness with some scepticism of complete recovery but experience of significant improvements due to management.</p>	<p><i>'it doesn't bother me being on them [antidepressants] it's not a big deal don't mind at all [...] I was crying all the time saying I'm so tired I can't cope I'm so tired I'm not at work and she [GP] said 'look, w-what about antidepressants?' and I said 'well I'm not depressed I'm not depressed I'm just fed up with my circumstances and I'm tired all the time' and um she said 'well try it and see how you feel' and it was a marked difference it was a real difference. So you know I just stayed on them I haven't even questioned coming off it to be honest' (Maria)</i></p> <p><i>'I think it's too open ended in terms of you can't tell what's going to happen in terms of chronic fatigue. Actually it I felt-if I, w-if was told when I was first diagnosed that this will be my prognosis I'd be very happy you know what I mean, that I would be going back to work and I'd be functioning quite normally I'd be happy if that had been my prognosis, um (tut) although I don't feel back to normal completely but um I am functioning' (Maria)</i></p> <p><i>'for me I think there isn't a full recovery um I used to, and that's not me being negative and pessimistic I just, it's been a long time like there been several relapses and I just don't think there will be a full recovery I think it can be managed to a point [...] I know people say that they do [recover] brilliant, amazing umm, but I'm not one of them by the looks of it so (laughs) Until they find what causes it, I don't think you can find a cure, and I'm not convinced there will be a cure, so, you've just got to make the best of (laughs) umm, how you are with it' (Kate)</i></p>

		<p><i>'just thinking back to the people that I met when I was younger and how they dealt with it and how they, and that was only a few people literally only a few people so, be interesting for me to see if-I know one-I'm still in contact with one of them and he-he's recovered um, but it would be interesting to see those girls again-see how they are and what has happened to them down the line' (Jane) 'I'd be interested in research on ME if it comes back again I've never seen any research that so people get it more than once, so if you do recover from it does it come back? because I-I've don't know I-I kind of because of the way it's hit me I sort of think 'oh shit does it actually ever go away?' are you still carrying whatever you are carrying around with you? and some people manage to suppress it and when they get older do-does it come back hit them? I wonder if some of the kids that I was with on the group thing, I'd be interested to see what in twenty years' time if it had reared it's ugly head again I don't know I don't know' (Oliver)</i></p>
3. Connection	<p>Talking and sharing experiences was beneficial. That said CFS/ME was not central to identity, as a result the need to disclose was not always necessary.</p>	<p>NEED TO ORGANISE TO CORRECT SUBTHEME/CODE GROUPING</p> <p><i>'I think it's very important to find someone that you do really that is a soul buddy someone that you do trust and you do trust you can you can confide in' (Stuart)</i></p> <p><i>'It [the interview] made me realise how far I've come (I wouldn't have got through it without tears in the past!) so was useful to me, thank you' (Kate, email following interview)</i></p> <p><i>I suppose, when I was younger it was sort of almost used to define me 'oh that's JANE she's the one that's got ME' so new people that I met I didn't want to, sort of know me as 'oh that's Jane she's got ME' or whatever or she doesn't work because or you know things like that so I think that's why just as I've got older I've sort of thought no there's more to me than this, you know' (Jane)</i></p> <p><i>'you know I don't even mention it to people unless, it comes up for some weird reason that they're talking' (Maria)</i></p>
	<p>Having roles, purpose and responsibilities despite symptoms, which meant adapting or changing aspects. There was also an acknowledgement that responsibilities was unhelpful too.</p>	<p><i>'actually working does give me a sense of self-esteem that I'm doing something useful and I actually do really like my job so although it it's demanding and can be quite debilitating (laughs) at times you know it's it's healthy (laughs)' (Diane)</i></p> <p><i>'I managed to get a part time job which was a real confidence booster and a real you know an-and so that took my concentration off things and that you know I-I sort of erm, worked that [inaudible] quite successfully at it' (Jane)</i></p> <p><i>'when I've got bad days I to try to get up and make myself like I take the dog around the block and I do some dusting and washing or something just to get through that day and to make myself do it really. yeah 'cause I think you could easily say 'oh I'll stay in bed today I'm not going to get up today' and but I don't want to do that. [...] she's good [dog] 'cause she's got to be looked after she can't look after herself so I've got to brush her in the morning and feed her take her round the block in the afternoons (laughs) so [...] It's like Bill [husband] can look after himself if he's got to but the poor little dog can't (laughs) and she' so nice to cuddle up to and I chatter away to her as well (laughs)' (Alison)</i></p> <p><i>'Yeah it's all ticks in the plus box you know against the constant sort of you sort of start off with the sort of fifty minus points and you try and work your way up to get fifty plus points and least you can balance it out.' (Oliver)</i></p> <p><i>'we used to be in a skittles team and it ran throughout the winter so it would go from September right through to May, so that was a commitment once a week you know as part of the team and my daughter took on the captaincy of the team as well and I helped her so that was a-a commitment that sometimes you know when you get home from work and you think 'I really can't handle that tonight' but knowing you have to so putting you're-I know it was fun and it's good being with other people and playing skittles but at the same time if you know it was um, (tut) something you always have to do when you didn't always feel like it so stopping that' (Diane)</i></p>
	<p>Participants reported that</p>	<p><i>"to build up a level of fitness [for walking] that is better than I have at the moment. I'm then hoping that, if I can manage than in sort of like the middle-the-middle of next year I'm going to really have a go at getting a part-time job doesn't really matter don't care which-what sort of job it's going to have to be about you know three or four hours a day preferably in the morning as in the morning as possible uh and</i></p>

	gradually and consistency was particularly important working towards goals, even small steps.	<p><i>that's what I'd be looking for if I can manage to do that-that that is as far as I'd be able to go I don't see anything getting much better than that" (Oliver)</i></p> <p><i>'but not setting your goals too high, ah too-too , to-to-to-to build up gradually to a point' (Stuart)</i></p> <p><i>'when I was first, diagnosed I was really severe I was wheelchair um year off school um obviously I couldn't really do anything to help myself then and as I've steadily come back, it's just been picking it up along the way' (Penny)</i></p>
3i Connection with self	Establishing a sense of self despite illness, which linked to mood and valued activities or personal values. Humour was spoken about as helpful and a way to express sense of self.	<p><i>'learning to say no has been a big one um because I don't like to let people down umm but actually, I'm letting myself down and making myself ill if I'm not, erm, doing what's best for me and my body' (Kate)</i></p> <p><i>'I was really struggling to cope I remember my father coming up and my father's 83 and we went for a walk and I didn't find it too bad at the time but the following days I was just talking and I was just burst out crying because of the exertion of this' (Stuart)</i></p> <p><i>'I was crying all the time saying I'm so tired I can't cope I'm so tired I'm not at work and she [GP] said 'look, w-what about antidepressants?' and I said 'well I'm not depressed I'm not depressed I'm just fed up with my circumstances and I'm tired all the time' and um she said 'well try it and see how you feel' and it was marked' (Maria)</i></p> <p><i>'I was in a situation [financial], two years ago where things were so bad and I felt so, useless, um, that I took an over dose' (Diane)</i></p> <p><i>"cause I-my one of my worries when I went to HOSPITAL NAME was is it all in my head? I've been I was sent to a psychologist therefore it's all in my head-but <u>no</u>. There's a r-reac-psychological reaction to everything that's going on' (Kate)</i></p> <p><i>'Just I-I'm normally quite a sort of happy-go-lucky person I just find it quite funny you know, just sort of sit down there and uh you know these-these silly things happen you know' (Colin)</i></p>
	Relevance and timing of support was raised as a barrier or facilitator for change.	<p><i>'I think the courses that are run by the NHS are run solely for women They do not consider um what the impact it has on a man 'cos no disrespect the majority of cases or a lot of cases the man is the main bread winner-winner. If the man does stop working, then it has large impact on the whole household. (breath) and um, you know the course is so female orientated so you know 'pace yourself don't do so much ironing' (slaps hands on thighs) but I don't do ironing. (laughs) you know.'</i> Colin</p> <p><i>'when I was an adult amazing so much support and, I think I'd be in a heap if I hadn't have come to HOSPITAL NAME, umm and I get-I guess actually as the d-it's completely different as a teenager and effectively I was still a child at eleven...how can a twelve year old talk about what's going on when they don't know what's going on so (pause) you know there was support' (Kate)</i></p>
	Re-evaluating activities and values to enable efforts in important areas. This processed also involved weighing up the costs and gains.	<p><i>'I love knitting I've got a little granddaughter I can knit for now' (Alison)</i></p> <p><i>'but on the other hand if I'm doing that's benefit psychologically then, it can tired (laughs) me out physically (laughs) So, there's a two edged sword at sometimes and I have to try and manage one with the other' (Diane)</i></p> <p><i>'I went through that really really terrible period of feeling absolutely awful, and then because of the changes I made to my life I'm able to cope' (Maria)</i></p> <p><i>'to live with it' 'work your way around it' (Oliver)</i></p> <p><i>'I do like to get outside um I do like to try and do a little bit of walking although I am limited with walking' (Colin)</i></p>
	Importance of self-care and self-compassion in	<p><i>'I started to realise that, um, I shouldn't be so tough on myself an-and, start, being kinder' (Diane)</i></p> <p><i>'I blamed myself for a lot of things it was like well I should be able to get myself out of this if I got myself into it, but actually there was a pre-existing condition that was exacerbated-exacerbated whatever the word is' (Kate)</i></p>

	long-term management	<i>'Yeah (pause) yeah definitely (pause) Yeah I'm not you know there's this (pause) this underlying thing that goes right back to my childhood about you know um gaining approval and, being the best, and, becoming aware only the last few years that actually to hell with it (laughs) Why can't I just be me?' (Diane)</i>
3ii Connection with others	Unsupportive people were not helpful, although there was empathy for lack of understanding. Attempts for different communication methods including metaphors and analogies.	<i>'Wasn't sure I believed it myself so how could I expect other people to believe me' (Kate)</i> <i>'they help you as best they can I'm sure I'm not criticising them [HCPs]' (Stuart)</i> <i>'I guess one of the most unhelpful things is peoples' lack of understanding. and that includes my family as well. and my brother in particular (smiles), who goes to church every Sunday nd helps his neighbours and all the rest of it, you know, can't understand ME (sighs) you know, doesn't want to try and understand ME, you know um even though I've-I've uh tried I've been given hand-outs and they've suggested you give it to your family to read and I've sent it off to them can't understand it.'</i> (Colin) <i>'you're on the floor' not a chance in hell' 'reared its ugly head' 'icing on the cake' 'pull the plug' 'crashing and burning' 'anything that is as alien to people as ME it's useful-any you have to use metaphors 'cause otherwise you'd just be repeating your symptoms to them constantly (laughs) and it's like this-but it's like this <u>every</u> fucking day (laughs) y-yeah they're thinking 'yeah that must be bad until you get over it' 'NO you don't get over it' (laughs) you know 'it's it keeps on coming back'' (Oliver)</i> <i>'losing each individual battle I have a lesser chance of losing the war' 'logger heads' 'two edged sword' 'recharge batteries' 'one hand, on the other hand' 'straw that broke the camel's back' (Diane)</i> <i>'throw a massive spanner in the works' [re: impact of sick days on colleagues] (Penny)</i>
	Normality occurred after having the illness so long it was understood as normal for them, and in the household for some.	<i>'I didn't do anything before I went to work and people would come in (laughs) to work and say they've changes their beds and oh they did this and they did that I used to think 'how can they do that and go to work as well'' (Alison)</i> <i>'when it got to thirteen years it sort of fourteen years I sort of thought 'oh-well you know I've had it longer than I haven't had it now' (Jane)</i> <i>'I call it normal 'cause I'm so use to it' (Alison)</i> <i>'if somebody said recovery I would imagine going back to completely normal, "normal" (signals inverted commas in the air) what every normal is' (Maria)</i> <i>'fairly fit and I used to do regular exercise but I did find that that time even you know in my early twenties and early thirties that I had much longer recovery periods for, ah from ex-excessive exercise. Compared to my peers' (Stuart)</i>
	Feeling alone to manage was common, although some found this useful.	<i>'the-it's that bottom line is that you are on your own with it to a very much to a large extent' (Colin)</i> <i>Y-you just completely (pause) you know they come err th-they they they see you, they talk to you they don't really help you an awful lot-they help you as best they can I'm sure I'm not criticising them but erm erm the y-your left on your own and and you think 'well there no point in going back anyway' I mean my doctor said to me do you want to go back there again some while ago I said 'well no, they don't know anything, they can't help you' there's no point you just feel left in the dark' (Stuart)</i> <i>'that-that's partly me, I-I don't keep going back there because I <u>know</u> there's nothing they can do so I just try and manage it myself' (Diane)</i>

• Themes mapping onto ACT and FACT models.
 Figure 4: Model of Psychological Flexibility in ACT (blue), three pillars in FACT (green) and study themes (purple).



(adapted from McCracken, 2010; Strosahl and Robinson, 2015)

Appendix E - Dissemination of findings:

- British Psychological Society (BPS) Division of Health Psychology Annual Conference 2015 Presentation (17.19.15)

CFS/ME

- Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME) is a long-term condition whereby disabling fatigue (both physical and mental) occurs without presence of disease lasting over 6 months (Fukuda et al, 1994; Sharpe et al, 1991).
- Common symptoms include: post exertion malaise, poor, disturbed and un-refreshing sleep, myalgia, concentration and memory difficulties, headaches, swollen glands and persistent sore throats (Fukuda et al, 1994).

d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

Royal United Hospitals Bath 
NHS Foundation Trust

CFS/ME

- Co-associated disorders can include: depression, Irritable Bowel Syndrome and Fibromyalgia (Maes 2011; Whitehead 2002).
- Diagnosis relies on patient reported symptoms and negative blood screens to exclude other explanations for fatigue (Chambers 2006; Fukuda 1994).
- No firm conclusions have been drawn regarding the illness trajectory, prognosis and recovery of CFS/ME (Action for ME, 2014; Asbring, 2001; Jason et al, 2012).

d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

Royal United Hospitals Bath 
NHS Foundation Trust

Background

- Treatment guidelines (NICE, 2007) reported Cognitive Behavioural Therapy (CBT) and Graded Exercise Therapy (GET) as most effective based on the evidence available. Both under the umbrella term 'Behavioural interventions.'
- Updated Systematic Review (Baldwin, 2013) for 'Behavioural Interventions' provided further support for NICE guidance. Specifically noting Cognitive Therapies (CBT and Mindfulness-based Cognitive Therapy (MBCT)) and GET effective at significantly improving fatigue, functioning, depression and anxiety.

d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

Royal United Hospitals Bath 
NHS Foundation Trust

Rationale

- Limited research focusing on long-term experiences of CFS/ME.
- To date only three published studies (McKenzie et al, 1995; Whitehead, 2006; Wilson et al, 1994) have specifically focused on longer term outcomes and experiences of adults with CFS/ME.
- Qualitative study conducted to gather valuable subjective data exploring patients' long-term experiences of managing CFS/ME.

d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

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Methodology

Design & Procedure

- Qualitative research
- Critical realist ontology and contextualism epistemology
- Semi-structured interviews
- Audio recorded and transcribed verbatim
- Analysed using deductive Thematic Analysis (Braun and Clarke, 2006)

Participants

- Adults (≥ 18 years)
- Fatigue experienced ≥ 5 years
- Diagnosed with CFS/ME by GP
- Provided informed consent

Exclusion criteria

- Direct patient of researcher
- Unable to attend interview
- Unable to speak and understand English

d.williams16@nhs.net
deborah.s.williams@live.uwe.ac.uk

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Demographics



	Age (years)	Symptom onset (years)	Time between onset and diagnosis (years)
Mean	48	13.1	3
Median	55	11.3	2
Range	23 - 64	9 - 17.5	0 - 10

d.williams16@nhs.net
deborah.s.williams@live.uwe.ac.uk

Royal United Hospitals Bath 
NHS Foundation Trust

The Interview



d.williams16@nhs.net
deborah.s.williams@live.uwe.ac.uk

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Instances of Embodiment

Colin – during the interview

- Spoke of using humour to '*lighten the load*' of the serious impact of CFS/ME on his life.
- Demonstrated using laser light.
- For Colin it acts as a distraction '*So you do this [...] takes your mind off things*' and enables him to be the comedian he is '*I'm normally quite a sort of happy-go-lucky person [...] I just find it quite funny you know [...] I've always been that way*'.

d.williams16@nhs.net
deborah.s.williams@live.uwe.ac.uk

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Instances of Embodiment

Kate – after the interview

- Spoke of self-reassurance and using Mantra/inner voice.
- Demonstrated emailing a picture of her 'Wall of Wisdom'
- Kate described her wall as '*it's just a big frame of lots of quotes in [...] it's got things like "if you say yes to someone else make sure you're not saying no to you" and "stop comparing the way you think you should be to where you are" all sort of little quotes that sort of actually for me work really well*'

d.williams16@nhs.net
deborah.s.williams@live.uwe.ac.uk

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Instances of Embodiment

Kate – before the interview

- Spoke of Pacing as beneficial for her, not doing too much activity during the day/week/month.
- Demonstrated cancelling interview rescheduling a week later.
- Kate '*I have to pace like pacing is, key for me, this last couple of weeks is primary example actually (smiles) I did too much and at the weekend I had to just cancel everything for three days, and that's not managing it actually that's boom and bust.*'

d.williams16@nhs.net
deborah.s.williams@live.uwe.ac.uk

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Instances of Embodiment



d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

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Instances of Embodiment

	Talked of:	Related theme:	Interpretation of function:	Instance of embodiment:
Colin	Humour	*Connection with self	Lighten mood, distract mind, be person he is/ wants to be	Using laser light – laughter with the researcher
Kate	Pacing	*Awareness *Acceptance	Spreading out activities and interspersing with rest	Cancelled and re-arranged interview
	Self-Reassurance	*Connection with self *Acceptance	Mantra, inner-voice to reassure herself, but used an external prompt to reinforce this	*Wall of Wisdom' (Emailed researcher photo)
	Reflecting on experiences	*Awareness *Connection with self	Value of talking and sharing to reflect on management and experiences as a process	Email to researcher, following interview
Stuart	Information seeking	*Acceptance	Actively trying anything to help, taking any opportunity to gather information to help	Asking the research for new knowledge and experience
Diane	Problematic symptoms	*Awareness	Different symptoms could require different strategies/approaches	Contradicting accounts and seeming muddled at times

d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

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Research Implications

- Research focusing on long-term experiences of managing CFS/ME, not only were the participants SPEAKING about what is beneficial or detrimental, they were also DEMONSTRATING it.
- Some more explicitly than others, perhaps this is because these strategies have become a more natural way of life.
- Discussion point also maps onto wider research findings thinking about therapeutic moderators and mechanisms of change.

d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

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Wider Implications

- Going beyond words - not only paying attention to WHAT is said.
- Unexpected instances such as these were able to be captured by the qualitative research methods.
- Are there any embodiments of what is said (or contradictions) in the room and HOW can we use them?

Research Communication Clinical

d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

Royal United Hospitals Bath
 NHS Foundation Trust

Questions



d.williams16@nhs.net
deborahs.williams@liverpool.ac.uk

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- Bath Centre for Fatigue Services, Education Session (over leaf)

Long-term experiences of managing CFS/ME: a qualitative study

Background

Chronic Fatigue Syndrome/ Myalgic Encephalomyelitis (CFS/ME): a long-term condition (LTC) whereby disabling fatigue and other symptoms (ie pain, poor & unrefreshing sleep and cognitive difficulties), occur without presence of disease, lasting over 6 months, with no medical cause (Fukuda et al, 1994; Sharpe et al, 1991). No cure exists however, behavioural interventions have significantly improved patient outcomes demonstrated by quantitative research (Baldwin, 2013; Chambers et al, 2006; NICE, 2007).

So many facets of CFS/ME are unknown (aetiology and prognosis) complicated further by high individuality in presentation and experience (Action for ME, 2014; Asbring, 2001; Jason et al, 2012).

Whilst CFS/ME is classified as a LTC, very little research has specifically sought to gather information from those that have had the condition for a long period of time. This study therefore aimed to explore in greater depth how people with CFS/ME manage their condition over time to consider; illness experience overtime, whether different support is needed at different stages and potential therapeutic mediators and mechanisms for change to maintain improvements or outcomes.

Methodology

Design

- Qualitative research
- Critical realist ontology and contextualism epistemology
- Semi-structured interviews
- Analysed using deductive Thematic Analysis (Braun and Clarke, 2006)

Participants

- Adults (≥ 18 years)
- Fatigue experienced ≥ 5 years
- Diagnosed with CFS/ME by GP
- Provided informed consent

Exclusion Criteria

- Direct patient of researcher
- Unable to attend interview
- Unable to speak and understand English

Results

Demographics:

A total of 9 participants took part. Gender ratio of 2:1, females to males, all were white British, majority were married (n=4) and majority were employed (n=6).

	Age (years)	Symptom onset (years)
Mean	48	13.1
Median	55	11.3
Range	23 - 64	9 - 17.5

Themes and sub-themes:

1. **Awareness** *'to listen to your body and your mind and, not denying that' (Diane)* This theme included a heightened sense of self-awareness of experiences, that developed over time and the awareness of others.

2. **Acceptance** *'the illness might not get better but you'll deal with it better so therefore you will be better' (Penny)* This theme included accepting CFS/ME as a life-long illness - not fighting against it, subsequently impact of the condition reduced.

3. **Connection** *at least I feel that I've got areas of responsibility and control [...] and I achieve things' (Oliver)* This theme included connecting with what was important (core values), having roles and responsibilities despite illness that adapted or changed and conceptualisation gradual and consistent progression was the goal.

3i with the self *I'm letting myself down and making myself ill if I'm not, erm, doing what's best for me and my body' (Kate)* This sub-theme included connection and disconnect with sense of self, addressing psychological impact and weighing up costs & gains.

3ii with others *'support from family is-is a must you can't do it on your own' (Jane)* This sub-theme included connecting with others for concrete or practical support and attempts to communicate with others using metaphor and analogies.

Discussion

Themes related closely to each other and some themes contained similar threads/code such as individuality, adapting and development or overtime.

Themes fit with previous research;

- highlighting the complexity of managing a fluctuating condition
- echo adopting an individualised treatment approach using behavioural therapies to manage the impact of CFS/ME

Findings also suggest the merit of adopting and researching contextualised cognitive therapies (Acceptance and Commitment Therapy (ACT) and Focused ACT) for CFS/ME.

Limitations:

- Interpretations based on interview data and clinical impression of 1 researcher
- No objective comment can be made regarding effectiveness of management participants used other than their subjective appraisal
- Limited transferability of findings (specifically to other ethnic groups and severely affected individuals)