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Running head: CFS and health anxiety

A novel approach to treating CFS and co-morbid health anxiety: a case study

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Key Practitioner Message

- CFS is a debilitating condition which is difficult to treat successfully; first line recommended treatments achieve only moderate effect sizes
- Anxiety, particularly about health, is reported to be common in CFS. However, anxiety is not specifically targeted within treatment and may negatively influence outcome due to the potentially mutually maintaining nature of these complex conditions
- The present study demonstrates that an integrated treatment approach designed to encompass physical and psychological symptoms yields reliable and clinically significant outcomes in 50% of time recommend for first line treatments.
- Results reflected non-case level status for both CFS and health anxiety at end of treatment, in addition to reductions across all clinical measures.
- This study demonstrates the fundamental importance of an individualised, rather than generic treatment approach to complex cases; the ‘meaning’ of experience is a central tenet within a cognitive approach which should be reflected in treatment.

Abstract

Objectives. CFS is a debilitating condition which affects 0.2-0.4% of the population. First line treatments are Cognitive Behaviour Therapy or Graded Exercise Therapy; however these treatments yield only moderate effect sizes. Emerging research suggests that anxiety about health may be common in CFS. Health anxiety treatment models demonstrate good therapeutic outcomes however these models have yet to be applied to CFS. This paper describes the application of a novel cognitive behavioural approach to the treatment of both physical and anxiety related symptoms in a patient with CFS/ME, and furthermore presents a conceptual hypothesis regarding the mutually maintaining relationship between these two co-occurring conditions.

Design. A single-case design was used, with pre, post and follow up data. The cognitive behavioural model of health anxiety was adapted and delivered as an eight session intervention. The intervention was driven by an individualised formulation developed collaboratively with the patient.

Results. The application of this approach generated reliable and clinically significant reductions in physical and psychological symptoms, which were maintained at 12 month follow-up. The participant no longer fulfilled the criteria for CFS or health anxiety following eight treatment sessions. The treatment approach was found to be agreeable to the patient. All treatment hypotheses were supported.

Conclusions. An adapted cognitive behavioural approach to treating CFS and health anxiety yields positive results and shows promise for application to the broader CFS population.

Introduction

Chronic Fatigue Syndrome (CFS) is a debilitating condition which is characterised by unexplained, ongoing and excessive fatigue that is not alleviated by rest. Other common symptoms including joint pain, sleep disturbances and cognitive difficulties (NICE, 2007). CFS can lead to significant disability and contribute to poor quality of life. Estimates suggest that the economic impact of CFS on employment and productivity is over £102 million a year in the UK (Collin, Crawley, May, Sterne & Hollingworth, 2011). The aetiology of CFS remains poorly understood, with prevalence rates of CFS in the region of 0.2-0.4% of the population (NICE, 2007). Despite high quality CFS treatment trials, a recent meta-analysis (Castell, Kazantzis & Moss-Morris, 2011) indicated that the gold standard treatment of Cognitive Behaviour Therapy (CBT) for this condition (NICE, 2007) resulted in only moderate improvements overall ($g=0.33$) with small to moderate improvements across the domains of fatigue, functional impairment, depression and anxiety. Furthermore, when CBT for CFS is delivered in routine practice settings, levels of fatigue, depression and anxiety have been shown to decrease, but not to the same extent as in clinical trial settings (Fernie, Murphy, Wells, Nikčević, & Spada, 2016). The small to moderate effect sizes found contrasts with large effect sizes found for CBT in other conditions such as anxiety (Olatunji, Cisler, & Deacon, 2010).

Current approaches to CBT for CFS are based on an original model developed by Wessley (1991). This model suggests that a heightened focus on disease or signs of disease, serves to reinforce beliefs that there is an acute and on-going disease process and activity is reduced accordingly. Surawy and colleagues (1995) subsequently extended Wessley's model further to include a number of personality related factors in the development and maintenance of CFS (Surawy, Hackmann, Hawton, & Sharpe, 1995). However, findings related to the importance of personality factors are relatively inconclusive (van Geelen, Sinnema, Hermans, & Kuis, 2007). Surawy's elaborated model was further adapted and tested for clinical utility, forming the basis for CBT interventions delivered across specialist CFS services (Deale, Chalder, Marks & Wessley, 1997). The PACE trial (White et al., 2011) reported the largest multi-centre trial for the application of the CFS model of CBT, however results reflected only moderate outcomes. The CBT model for CFS adopts a primarily behavioural approach: focussing on physical symptoms and working towards behavioural change through an activity management programme; there has been little emphasis on the interpretation and 'meaning'

of symptoms for the patient, which feature as a central tenet to the theory and clinical application of cognitive behaviour therapy (Beck, Rush, Shaw, & Emery, 1979, Beck, 1995; Kelly, 1963; Leventhal, Safer & Panagis, 1983). However, more recently researchers have reported an association between cognitive elements, such as conceptual thinking about symptoms and symptom focus, and symptom severity (Ferne, Maher-Edwards, Murphy, Nikcevic, & Spada, 2015; Maher-Edwards, Fernie, Murphy, Nikcevic, & Spada, 2012; Maher-Edwards, Fernie, Murphy, Wells, & Spada, 2011).

There is a high level of psychiatric co-morbidity in CFS patients. For example, the PACE trial (White et al., 2011) identified that 56% of the N=147 CFS sample had co-morbid psychiatric diagnoses, two thirds of which were identified as anxiety disorders (Cella, White, Sharpe & Chalder., 2013). Although health anxiety was not reported in the PACE trial, clinical levels of anxiety related to health concerns were reported to range from 17.5% to 24.7% across medical disorders (Tyrer et al. 2011). Preliminary data suggests that prevalence of health anxiety in CFS could be as high as 69% (Daniels, Kacorova, & Brigden, in submission). It is unsurprising, given the complex nature of CFS/ME, the often protracted process of diagnosis by exclusion, and the lack of clear medical explanation for the aetiology of CFS/ME, that rates of health anxiety may be higher in this population.

Current research into anxiety disorders and CFS suggests that these conditions share similar maintaining factors which can be understood from the perspective of cognitive-behavioural theories; such research also supports the hypothesis that cognitive and behavioural factors maintain both anxiety and CFS (Surawy et al., 1995; Salkovskis, 1996; Deale et al., 1997; Tyrer et al., 2011).

The cognitive behavioural model of health anxiety (Salkovskis & Warwick, 1986) is based on the notion that ambiguous health-relevant stimuli (including bodily variations and sensations, health information, medical consultations and so on) are subject to misinterpretation which informs behavioural responses such as hypervigilance to stimuli bodily sensations, avoidance, bodily monitoring and reassurance seeking in order to address health concerns. These responses, also known as Safety Seeking Behaviours (SSB), then negatively impact on the perception of bodily function and related illness concerns as part of a set of vicious circles which fuel and maintain psychological distress. SSB are considered to be a key component in the maintenance of the anxiety cycle through the mechanism of

prevention of disconfirmation: i.e. the patient is unable to test or critically appraise their health beliefs as they employ strategies that they perceive ensures ‘safety’, rather than confronting fears or consequences. The tendency to appraise ambiguous health-relevant stimuli negatively and catastrophically is driven by underlying self-referent beliefs about health and illness, which give such stimuli particular meaning for an individual (Salkovskis, 1996). Similarly, CFS is characterised by the way an individual interprets the cause and meaning of bodily sensations and symptoms experienced (Afari & Buchwald, 2003).

Early cognitive behavioural models of CFS suggested that illness beliefs and coping strategies are also key factors in the development and perpetuation of CFS. More specifically, Wessely (1991) defined the trajectory of this condition as commencing with a virus or illness of organic/biological origin which precipitates coping strategies shaped by illness beliefs. Wessely’s model postulates that a heightened focus on disease or signs of disease, serves to reinforce beliefs that there is an acute and ongoing disease process and activity is reduced accordingly. This was borne out empirically in the PACE trial, in which fear avoidance beliefs (i.e. fears that activity will make the symptoms worse) mediated change in both CBT and GET (Chalder, Goldsmith, White, Sharpe & Pickles, 2015). These beliefs are paradoxically unhelpful, as in chronic illness, sustained activity reduction leads to muscle deconditioning which is likely to exacerbate symptoms such as pain and fatigue, creating a vicious cycle of chronic fatigue and disability (Browne & Chalder, 2006). Coping strategies such as activity reduction serve to mediate between the acute initial illness and the chronic syndrome (Moss-Morris, 2005). In both health anxiety and CFS, increasing chronicity is accompanied by preoccupation with symptoms, a perceived lack of control and demoralisation (Moss-Morris, 2005). Furthermore, in both CFS and health anxiety, coping strategies employed to either maintain health status or prevent worsening of health, serve to maintain the difficulties.

Despite the overlap in processes maintaining CFS and health anxiety, the health anxiety model has not previously been evaluated in CFS patients. However, a recent large scale RCT tested the efficacy of Cognitive behaviour therapy for Health Anxiety in Medical Patients, (known as the CHAMP trial), using a CBT formulation driven approach to treatment (Tyrer et al., 2014). In this trial, N=444 health anxious patients were recruited from cardiac, neurological, respiratory, endocrine and gastroenterological clinics. They were randomly assigned to 5 to 10 sessions of CBT for health anxiety (CBT-HA) or standard care. The CBT

protocol emphasised the patient's interpretation of their fears about their symptoms and formulating their worst fears, before considering alternative explanations and gathering evidence for these, whilst reducing SSBs (Tyrrer et al., 2011). Outcomes reflected successful application of CBT-HA across all patient groups over a 2 year period and cost neutral interventions.

Given the theoretical overlap between underpinning mechanisms in CFS and health anxiety, it is plausible to suggest that an integrated treatment approach drawing on the CHAMP trial with medical patients, will yield positive results for a sub-group of CFS patients who present with co-morbid health anxiety. It is postulated that the mechanisms by which CFS and health anxiety are both similarly maintained present as a complex and unique clinical challenge in treatment; the striking overlap in theoretical underpinnings may dictate a formidable mutual maintenance of CFS and health anxiety, particularly when undetected. It is suggested that while all CBT models are more similar than they are different, the health anxiety model has tested utility across health conditions and features 'meaning' of the symptoms (i.e. self-referent beliefs) which is the essential component within the CBT-HA model and absent in the CBT model of CFS/ME, therefore the former model is considered the most appropriate application to treat the complex presentation of CFS and co-morbid health anxiety.

The aim of the present study was firstly to pilot the CBT health anxiety model in CFS with co-morbid health anxiety using a single case study design; secondly, to achieve reliable and clinically significant improvements across physical and psychological health dimensions, achieving non-case level status for the patient in both CFS and health anxiety.

Method

An AB case study design was utilised to explore the effectiveness of using an adapted cognitive behavioural treatment approach for CFS with health anxiety. Five time points for measurement were used: t1=baseline/pre-treatment; t2=mid treatment/wk6; t3=end of treatment/wk8; t4=follow-up/wk 12, t5=12 month follow up.

The treatment was based on empirically grounded cognitive-behavioural models of health anxiety (Salkovskis, 1996) and CFS (Surawy et al., 1995, Deale et al., 1997), using the formulation driven CBT model for health anxiety as described in Salkovskis, Warwick and

Deale (2003), and drawing on the work of Tyrer and colleagues (2014). Treatment was delivered by a clinical psychologist (JD) with extensive clinical experience of CFS and accreditation in using the CBT approach; specialist supervision was given by experts in health anxiety (PS) and CFS (HoD).

Treatment was contracted to be eight 60 minute sessions in addition to two assessment sessions. A treatment rationale was given in the first session, and explicit consent was obtained for using CBT for health anxiety to treat CFS and co-morbid health anxiety. The initial two sessions were used to complete an assessment and develop a formulation which integrated CFS associated symptoms and health anxiety related symptoms. This was done by eliciting a recent episode where symptoms were particularly problematic, incorporating significant amounts of detail to illustrate the links between the cognitive, behavioural, emotional and physiological domains. The formulation formed the basis for treatment, where standard CBT techniques and interventions were used to test the ‘hypothesis A/hypothesis B’ approach (Salkovskis & Bass, 1997) which underpinned the patient led testing of safety behaviours and inter-session work.

Measures

M.M completed the following routine in-service questionnaire measures, the majority of which were used nationally across CFS services:

Chalder Fatigue Scale

The Chalder Fatigue Scale (Chalder et al., 1993) is an 11-item questionnaire covering both physical and mental fatigue with four response options per question (“Less than usual”, “no more than usual”, “more than usual” and “much more than usual”). Extensive research into the validity of this measure has been carried out (Deale, et al., 1997) and overall it is considered reliable (Chalder et al., 1993). This binary scoring method was used in the present study (0, 0, 1, 1).

Visual Analogue Scale (VAS)

The Visual Analogue Scale is a self-report measure of pain. The scale is scored out of 100 and the distance in mm across the line is measured and forms the participants' score (no pain 0-4mm, mild pain 5-44mm, moderate pain 45-74mm, severe pain 75-100mm).

Visual Analogue Scales have been considered to be a reliable way to quantify levels of pain (Price, McGrath, Rafii & Buckingham, 1983).

Self-Efficacy Questionnaire

This six item questionnaire rates participants' confidence on a Likert style scale from 0 (Not at all confident) to 10 (Completely confident) that they are able to manage various aspects of their long term condition to reduce the impact on their everyday life (Lorig, Sobel, Ritter, Laurent & Hobbs, 2000). The original authors note that the measure is suitably reliable. Higher scores indicate higher self-efficacy.

Hospital Anxiety and Depression Scale (HADS)

The Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983) is a 14 item questionnaire designed to separately measure both anxiety and depression with seven questions each. Cronbach's Alpha for both measures combined in a non-clinical sample being 0.86 (Crawford, Henry, Crombie & Taylor, 2001). Scores range from 0-3 on each question (0-7 normal, 8-11 mild, 12-14 moderate, 15+ severe).

SF-36 Measure of functioning

The short form-36 is a quality of life measure consisting of 36 questions which measure eight subscales, Physical Function, Bodily Pain, General Health, Vitality, Social Functioning and Role Emotional and Role Physical, two subscales which specifically measure restrictions as a result of physical or emotional difficulty (Jenkinson, Coulter & Wright, 1993). The SF-36 is considered to be reliable with a good test-retest reliability score (>0.75) (Brazier et al., 1992). Higher scores indicate better functioning.

Epworth sleepiness scale

The Epworth Sleepiness Scale (Johns, 1991) requires participants to rate how likely they are to fall asleep in certain situations, from 0 (Would never doze) to 3 (High chance of dozing).

A score of over 16 suggests the person is unusually sleepy by day, 10–24 range indicates that expert medical advice should be sought. Test-retest reliability by Johns (1992), using the same participants as the original research, was good ($r=0.82$) and Cronbach's Alpha score was good for patients ($\alpha = 0.88$).

Health anxiety Inventory (HAI)

The Health Anxiety Inventory (Salkovskis, Rimes, Warwick & Clark, 2002) is an eighteen item measure in which participants select one of four statements for each question. The measure is reported to have suitable test-retest reliability (0.90) and good overall internal consistency ($\alpha = .89$). Further research has also confirmed reliability (Rabiei, Kalantari, Asgari & Bahrami, 2013). Clinical cut off for health anxiety is ≥ 15 , with severe health anxiety scoring ≥ 18 .

Case study: background and assessment

M.M. was a 40 year old married white female with two young children. At the time of referral, she was not working due to her health problems. Prior to the onset of her CFS, M.M had worked at a senior level within a large organisation. She had experienced a number of difficult life events over the previous years, and was unable to identify a specific trigger for the onset of her fatigue. However, she believed that stress had contributed to her developing CFS. M.M. presented with overwhelming fatigue, muscle pain, un-refreshing sleep, impaired memory and concentration, and joint pain. She met the Centre for Disease Control Fukuda diagnostic criteria (Fukuda et al., 1994), and was accepted into the specialist service for assessment.

At assessment, M.M disclosed having experienced anxiety and depression in the past, which had been significantly ameliorated with CBT. She described herself as a worrier, with a recent history of panic attacks. To cope with her CFS symptoms, she employed a number of strategies which she perceived to be helpful. These included alternating rest, activity and sleep throughout the day within a highly detailed and rigid daily routine that she had developed, with the motivation of ensuring that she was able to meet the demands of her family and prevent a worsening of her symptoms. When M.M began to feel unwell, or

emotionally distressed by her symptoms, she would absent herself from the situation and rest/sleep or isolate herself for fear of behaving in a way she considered ‘horrible’. She would also look for ‘warning signs’ that her health was worsening (e.g. higher perceived need for sleep, ‘snappy’ responses, increased fatigue) so that she could take preventative action.

M.M reported a strong loss of role and identity due to the pervasive and substantial reduction in her activities of daily living; resigning from her high level job due to health difficulties was particularly challenging. She reported feeling very restricted and trapped, that she was not living a normal life. M.M. reported unfavourable comparisons to others and her own life prior to the CFS. In addition to the disabling physical symptoms she experienced, M.M. reported significant changes to her lifestyle, including inability to sustain a social life, reduced activities with her children, and significant concerns relating to her inability to engage more fully in her relationship with her husband.

Based on clinical material and empirical findings, the following hypotheses were developed for testing within treatment:

Table 1: Treatment hypotheses

<i>Treatment Hypotheses</i>
1. Cognitive beliefs relating to CFS and CFS symptoms inform behavioural strategies which maintain CFS. Targeted cognitive restructuring (and associated behavioural experiments) will lead to a reduction in the 100% conviction that specific behaviours (i.e. SSB) are necessary to prevent a worsening of CFS/ME, and that a worsening will lead to collapse.
2. Symptoms of CFS (specifically fatigue and pain) are maintained by behavioural strategies which are designed to prevent worsening of (CFS/ME) symptoms. Extinction of specific strategies identified in the formulation will lead to an improvement in the primary physical symptoms of fatigue (Chalder Fatigue Scale) and pain (VAS).

3. The health focused anxiety serves to exacerbate the CFS symptoms and vice versa; therefore, addressing both within an integrated approach will lead to reductions in both CFS symptoms (Chalder Fatigue Scale) and health anxiety (health anxiety inventory) as well as improvements in functioning (SF-36) and self-efficacy (self-efficacy scale).

Formulation

As illustrated in the diagrammatic representation of the formulation, M.M.'s episodes of fatigue and associated symptoms were usually triggered by a perception of feeling 'tired'. Once this appraisal of her physical state had been made, she experienced negative automatic thoughts related to perceived restrictions; M.M. believed that she possessed finite resources and therefore, perceived signs of tiredness would activate her assumptions that she would be unable to engage in further activity and that she 'can't cope/will collapse' (100% conviction): M.M. believed that if she continued to engage in physical activity beyond her perceived capacity, this would both worsen her condition and ultimately lead to the revelation (to others) that she was an unpleasant and 'horrible person'. When operationalized, M.M. described her feared 'horrible' side as unreasonably angry behaviour that was unpleasant and would lead to rejection by others.

Fear relating to her perceived inability to cope and the prospect of exposing this 'horrible' side was intolerable for M.M., and thus fuelled behavioural strategies designed to avoid further worsening of symptoms for these reasons. However, these strategies were counterproductive and not only prevented disconfirmation of her ability to cope, but also led to emotional and physical symptoms which paradoxically generated evidence consistent with her belief that she was unable to cope, e.g. further physical symptoms detected through hypervigilance were more likely to reinforce her belief that she was unable to cope and is at risk of collapse. This consequently led to an increase in triggering episodes due to the hypervigilance to the perceived sensation of tiredness. This further perpetuated one of several cycles which ultimately resulted in a restricted existence with entrenched behaviours maintained by the meaning given to the perceived sensation of tiredness (I can't cope/ I'm going to be horrible/collapse).

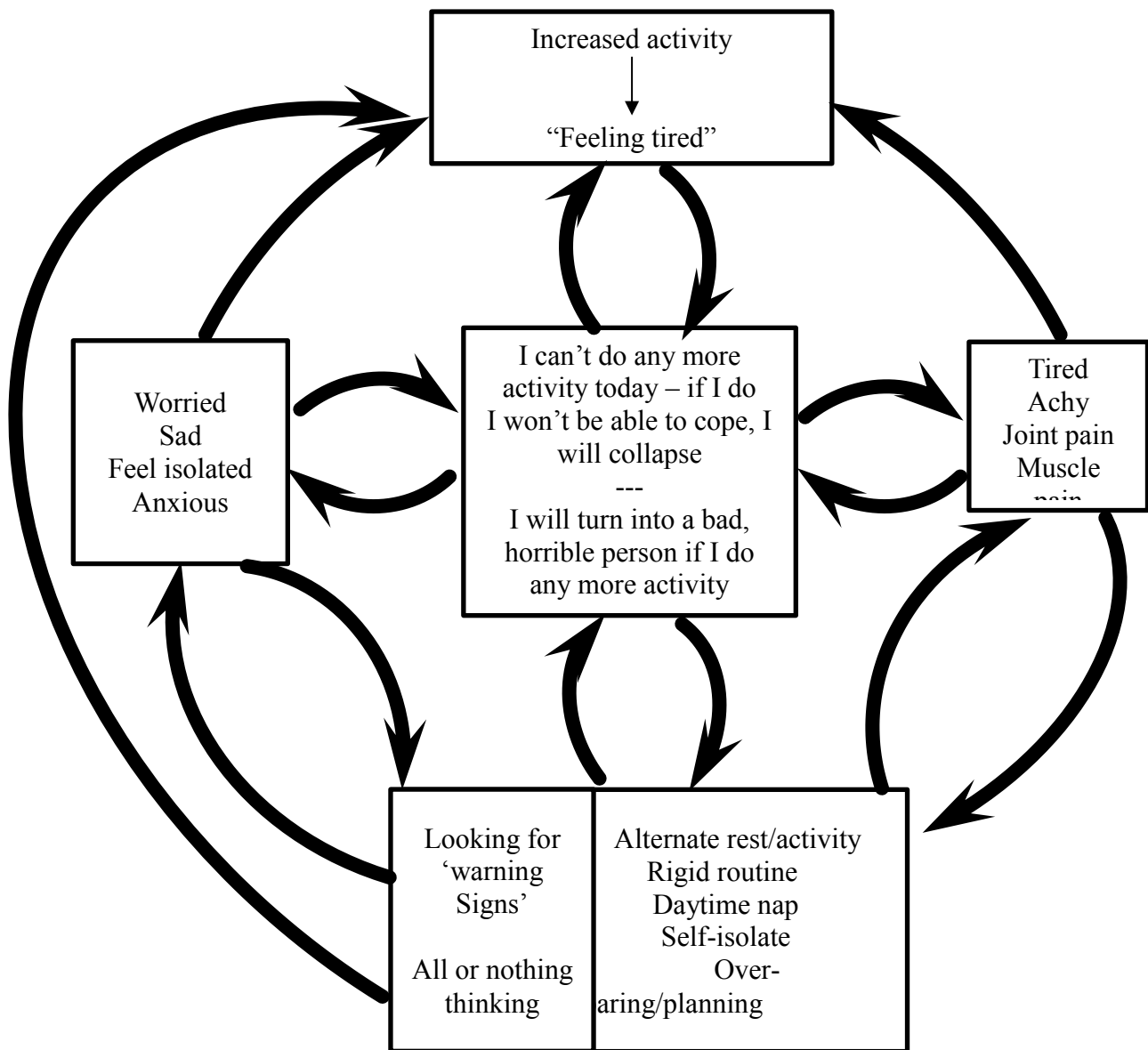


Figure 1: Formulation of presenting difficulties.

Overview of treatment

Following the development of the formulation, early treatment sessions focussed on operationalising the meanings and appraisals of specific beliefs, for example: “I can’t cope/collapse” and the hidden “horrible” person. The ‘worst case scenario’ for the beliefs were elicited, and belief ratings were established to ensure that change could be measured across treatment sessions. Psychoeducation and discussion around the mind-body link served to highlight the overlap between the physical sensations of anxiety and physical symptoms

associated with CFS/ME, and how normal bodily variations can be interpreted as signs of something worrying happening in the body. This also served to highlight the counter-productive role of symptom hypervigilance and was demonstrated through the use of body-focus tasks, illustrating how increased attention can lead to symptoms or the intensifying of symptoms (Wells, 1999).

Goals for treatment were established and activity monitoring was set up to give a clearer insight into the frequency and full repertoire of all behaviours employed in response to the interpretation of physical symptoms and prevention of the 'worst case scenario'. The 'Hypothesis A, Hypothesis B' (Salkovskis & Bass, 1997) concept was introduced as an underpinning approach to treatment, with the collaborative agreement that in order to ascertain whether M.M.'s current 'coping' behaviours were helping /preventing the worst case scenario from happening, we would need to test them out. The 'builder's apprentice' metaphor was used to facilitate this discussion of the scientist's approach (Stott, Mansell, Salkovskis, Lavender, Cartwright-Hatton, 2010). Simply put, the metaphor describes the long suffering builder's apprentice who holds up his newly built wall, for fear that it may fall or collapse if he does not support it. The question is: how will he discover whether he needs to continue to hold up the wall? He must test it. This relates to evidence gathering within the scientist approach, specifically the testing of SSB.

In M.M.'s case, hypothesis A/hypothesis B was as follows:

- A. I need to take measures to prevent a worsening of my CFS/ME, and if I do not prevent a worsening then I will be unable to cope and/or will collapse (belief rating 100%).
- B. I am taking measures to prevent a worsening of my CFS/ME, however these measures may be contributing to or maintaining the problem, rather than helping (belief rating 0%).

Cognitive strategies were employed to evaluate appraisals such as "I can't cope" and challenge underlying beliefs that "being angry = I am a horrible person". For example, on examination, M.M. revealed evidence for 'snappiness' and other interpersonal behaviours commonly associated with tiredness or lack of sleep (e.g. irritability), however even at her worst she found no evidence of unreasonable behaviour that would constitute her definition of 'horrible', despite the agreement that 'horrible behaviour' does not necessarily equate to a horrible, unloveable person.

Common thinking errors were identified and considered in relation to how these might inform coping behaviours, such as ‘all or nothing’ thinking. Through discussion, review of the formulation and an invited critical review of her own coping strategies, M.M quickly concluded that ‘looking for trouble’ i.e. symptom hypervigilance was contributing to her symptom experience rather than preventing a feared catastrophe. The Socratic method was used to elicit how these behaviours prevented disconfirmation that she would ‘cope’ and would be unlikely to ‘collapse’ and the unintended consequences of increase anxiety and

	Chalder	VAS	Self- efficacy	HADS-A	HADS-D	sf36	Epworth	HAI
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worry. Within the hypothesis A/B framework,

behavioural experiments were employed to test beliefs relating to the need to ‘sleep/rest’ and the perceived reliance on a rigid routine and the resultant effect on mood, physical sensations and beliefs. These experiments were patient led and set as homework by the patient.

Final sessions focussed on reviewing the formulation and evidence in favour of hypothesis A/B: all behaviours previously considered necessary to prevent a worsening of symptoms were abandoned, which facilitated M.M in reaching all of her therapy goals. All maintaining cycles were judged to have been broken, and she no longer believed she was unable to cope, or on the verge of unleashing a ‘horribleness’ each time she perceived an increase in symptoms. A therapy blueprint was produce by the patient and based on the treatment notes.

Results

At commencement of therapy, M.M. scored within the clinical range on the Chalder Fatigue Scale, HAI, and VAS, with mildly elevated clinical range scores on the HADS anxiety scale. Scores across all measures were below the clinical thresholds at session eight and at follow up. Scores on the Chalder Fatigue Scale reduced from full complement (11/11) to no fatigue related problems (0/11); this infers a complete absence of fatigue. Pain

scores reflected no pain where pain was ‘moderate’ at baseline. Anxiety and health anxiety reduced to sub-clinical levels.

Table 2: Outcome data baseline to 12 month follow up

Baseline	11	45	26	9	6	17	4	15
T2 - S6	8	20	40	9	1	22	4	11
T3 – S8	7	15	50	5	2	25	4	7
T4 – 4 week FU	0	0	54	6	2	30	3	3
12 month follow up	2	0	50	6	1	32	3	

Table 3: Reliable and clinical significant change

Measure	Points change required for Reliable Change Index	Points change required for Clinically significant change	Patient's change in scores from baseline to post-treatment	Reliable change?	Clinically significant change?
*Chalder Fatigue Scale	3 (Chilcot, Norton, Kelly & Moss-Morris, 2015)	4 (Sabes-Figuera et al., 2012)	4	Y	Y
Hospital Anxiety and Depression Scale – Anxiety	5 (Morley, Williams & Hussain., 2008)	8 (Morley et al., 2008)	4	N	N
Hospital Anxiety and Depression Scale –	5 (Morley et al., 2008)	8 (Morley et al., 2008)	4	N	N

Depression					
*Health Anxiety Inventory	4 (calculated based on info from Salkovskis et al., 2002)	7.58 (Tyrrer et al., 2014)	8	Y	Y
SF-36	8.4 (Hays, Brodsky, Johnston, Spritzer & Hui, 2005)	-	8	N	-
Self-efficacy Scale			24		
Epworth Sleepiness Scale	6 (Smith & Sullivan, 2007)	-	4	N	-

Change in scores on the outcome measures were compared to published data on reliable and clinically significant change on these measures. Changes in the fatigue, health anxiety and pain measures were reliable and clinically significant (* denotes significance).

The course of treatment was completed following eight treatment sessions as agreed; treatment success was attained using half of the recommended 16 sessions of CBT (NICE, 2007). M.M. no longer met the Fukuda criteria for CFS at the completion of treatment.

As part of the 12 month follow-up within the CFS service, M.M. completed a global impressions scale (Busner & Targum, 2007) which indicated optimum outcome of 'very much better.' M.M. reported that gains had been maintained within the 12 month period and that she had secured employment within that time.

Discussion

The present study piloted the use of an integrated treatment based on a health anxiety model in a patient with CFS and comorbid health anxiety. The success of the treatment as reflected in the measures and patient self-report provides preliminary evidence for the acceptability of this approach. Outcome data reflects non-case level scores at end of therapy and 12 month follow up, including both physical and anxiety/mood related symptoms. Furthermore this treatment approach yielded reliable and clinically significant outcomes across target measures following eight treatment sessions. M.M. reported significant functional improvements, reporting that she felt able to return to live a normal life. This was further demonstrated by her ability to secure employment and report optimum outcome in the global impressions scale at 12 month follow up. Evidence generated and reported supports all treatment hypotheses outlined.

The key aspect of cognitive behaviour therapy specifically employed and elaborated within this novel CFS intervention was the interpretation and meaning of symptoms. Although a cognitive shift can result from behaviour change (Beck, 1979), cognition is the cornerstone of any high integrity CBT intervention. M.M.'s interpretation of physical sensations were pivotal in shaping her responses to managing her CFS/ME; her 'safety seeking behaviours' were directed at avoiding a CFS related 'collapse' however they were ineffective, fuelled by anxiety and likely to be maintaining her CFS symptoms. M.M. had not previously taken a critical approach to her strategies: as illustrated in the builders apprentice metaphor (Stott, et al. 2010); she had not tested whether her strategies were actually holding up the wall (i.e. preventing her feared outcome), she assumed they were working because she had not reached her worst case scenario. Cognitive restructuring with the aid of behavioural experiments resulted in M.M. tentatively yet successfully withdrawing her hands from the wall to find that neither she nor the wall collapsed. M.M.'s absolute belief conviction in hypothesis A shifted to absolute belief conviction in hypothesis B through the course of therapy, supporting treatment hypothesis one.

Through the course of treatment M.M. was able to experiment with her SSB, which enabled her to challenge her anxiety related beliefs and generate evidence to disconfirm her fears, i.e. that she was able to 'cope' with increased symptoms and that her hidden 'horribleness' was in fact normal responses to tiredness and feeling under resourced: M.M. shifted to an acceptance that frustration, anger and exasperation were not emotional experiences unique to her, and that irritation and snappiness did not affirm her status as a 'horrible' person. M.M.,

was able to test the necessity of her rigid routine by gradually expanding her bandwidth: she was surprised to find that she was able to engage in significantly more activities than she had anticipated, without the feared consequence of becoming *unreasonably* angry and hostile.

M.M.'s abandonment of her SSB allowed her to engage more actively in life, breaking the cycles that were maintaining her symptoms: appropriately graded activity akin to Graded Exercise Therapy (albeit more informally conducted) is likely to have contributed to an amelioration of symptoms due to a reversal of deconditioning symptoms commonly found in CFS (Wearden et al., 2010). This supports treatment hypothesis two.

The present treatment approach for CFS was based on the underlying supposition that CFS and health anxiety share similar maintaining mechanisms and theoretical roots and where presenting together, an integrated approach would yield amelioration of symptoms associated with both. This was achieved within 50% of the time recommended for current first line treatment (NICE, 2007) and maintained at 12 month follow up. We therefore conclude that this case study was a successful application of the model and that hypothesis three is supported.

The 'scientist' approach was meaningfully utilised within treatment: using the 'theory A/ theory B' technique (Salkovskis & Bass, 1997) therapy progressed on a patient-led basis, enabling M.M. to explore her own beliefs at her own pace, taking ownership of the therapeutic process. M.M. reported that the scientific/critical approach to cognitive beliefs and assumptions was superlatively useful in the treatment approach.

The Socratic, collaborative relationship that underpinned the course of treatment was palpably fundamental to the success of treatment; M.M. was highly engaged as an active participant in therapy, affirming the agreeability and accuracy of the treatment model throughout the therapy process through positive engagement. The ease of arriving at a shared formulation and continued understanding and agreement of the aims of therapy and how to achieve those aims reflected not only the acceptability of the approach, but also a high degree of socialization to the treatment model (Roos & Wearden, 2009), which may have acted as an active component in the therapy (Daniels & Wearden, 2011).

Agreement and acceptance of a psychologically based model has been an area of contention in CFS where rejection of treatment rationale or psychologically based treatments has been cited as barriers to treatment (Chew-Graham, Brooks, Wearden, Dowrick & Peters, 2008; Butler, Chalder, Ron & Wessely, 1991; Wearden, Riste, Chew-Graham & Peters, 2008). This has not been the case here; M.M. responded to the treatment approach with enthusiasm and rigour. It is difficult to ascertain whether M.M. was unusual in her response to the treatment model, however a larger trial could test acceptability of the approach more fully.

It is suggested that there are key differences between current routine treatment models and the present approach which may have contributed to the positive outcome. Firstly, the underpinning model used in the present treatment is empirically grounded and has been developed and tested across patient groups, specifically developed for formulation-based application and therefore more suitable for patients with co-existing anxiety. Secondly, the current approach places an emphasis on the ‘meaning’ derived from patient symptoms; this meaning informs and precipitates the use of behavioural strategies and does not feature as a key component in standard CBT for CFS. It was evident from the intervention that core beliefs and feared outcomes were directly related to M.M.’s strategies and the potency of this as a driver in CFS may be underestimated.

There are inherent limitations using a single-case design, specifically that the findings cannot be generalized to the broader CFS population. However, a single-case study provides a legitimate trial of an experimental intervention and a basis for further treatment development (MRC, 2000) which looks promising. This study would have benefitted from further baseline monitoring to establish a stable presentation prior to treatment and reliability of the treatment outcome. This does not negate the merit of the study, but the findings must be interpreted cautiously: M.M. reported a history of anxiety however this was not evidenced objectively over multiple baseline time points, which would have been ideal.

It is noted that while scoring the highest possible score on the Chalder Fatigue Scale and moderate levels of pain, M.M. did not represent a severe case of health anxiety. Future testing

of this approach for CFS should assume a controlled approach to recruitment to test utility of the treatment for those affected across a range of severity.

Given the findings of the present study, future research should focus on testing the success of the intervention within a small trial or case series, specifically with CFS patients with higher levels of health anxiety. With recently emerging phenotype based research identifying sub-groups within CFS population, further testing of the model should be considered within this context.

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