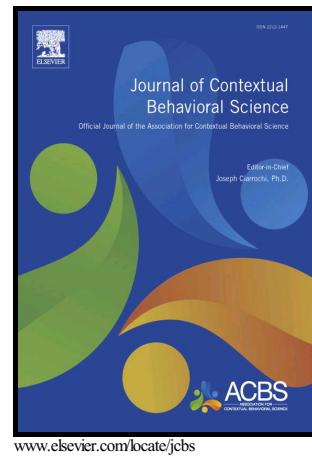


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An Acceptance and Commitment Therapy (ACT) intervention for Chronic Fatigue**Syndrome (CFS): A case series approach**

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Chronic Fatigue Syndrome (CFS) is characterised as medically unexplained fatigue, alongside a range of neurological and rheumatologic symptoms, persisting for at least six months (Fukuda et al., 1994). CFS is not yet well understood, and there is ongoing debate about its nomenclature, particularly in relation to aetiology, and underlying biological and psychological mechanisms (Nicholson, Brown, Jason, Ohanian & O'Connor, 2016). It is recognised, however, that CFS can be significantly disabling, and those diagnosed with the condition often report poorer quality of life than individuals with other long-term conditions (Anderson & Ferrans, 1997).

In line with literature recognising various psycho-social determinants of health (Martikainen, Bartley, & Lahelmac, 2002), a range of psychosocial factors appear to contribute to the maintenance of CFS. Cognitive-Behavioral models of CFS suggest that unhelpful beliefs and thinking patterns, in response to initial symptom experiences, may lead to a reduction in activity, which in turn can exacerbate symptoms, creating a 'vicious circle' of impairment and distress. Similarly, previous research has suggested that individuals experiencing CFS often use avoidance-based coping strategies in an attempt to manage their condition, which can in turn lead to increased impairment and distress (Heins, Knoop, Burk, & Bleijenbergh, 2013). Also, tendencies towards cognitive styles such as 'maladaptive perfectionism' have been associated with these patterns (Sirois & Molnar, 2014; Kempke et al., 2013).

Cognitive Behavioral Therapy (CBT) and Graded Exercise Therapy (GET), focused specifically on increasing physical activity, are recommended evidenced-based interventions for CFS (National Institute of Clinical Excellence (NICE), 2007; White et al., 2011), with recent research reporting beneficial effects such as less fatigue and better physical function that can be maintained in the longer term (Sharpe et al., 2015). However, these findings have received significant criticism from the CFS community; CFS clients and advocates have raised concerns regarding the methodological underpinnings of this research, as well as concerns that understanding CFS within such models may serve to trivialise and invalidate the very real experiences associated with the condition (Shepherd, 2015; Tuller, 2015).

A significant body of research has investigated psychological factors and adaptive mechanisms that appear to facilitate greater coping with CFS specifically (e.g. Moss-Morris, 2005) and chronic illnesses more broadly. One of the most promising of these factors in terms of the latter is 'acceptance', an active psychological strategy that aims to disrupt the negative cycle between avoidance and reduced quality of life. Acceptance is commonly portrayed as a willingness to live with an illness without reactance, disapproval, or attempts to reduce or avoid it (Bogaerts et al., 2007), in contrast to passive resignation or submission, or attempts to control the uncontrollable (Brooks, Rimes, & Chalder, 2011). Among individuals with chronic pain, acceptance has been associated with positive physical and psychosocial outcomes, including increased activity engagement, lower distress, and improved quality of life (Rankin & Holttum, 2003). Similar associations have been found in initial empirical work specifically examining individuals with CFS (Brooks et al., 2011; Van Damme, Crombez, Van Houdenhove, Mariman & Michielsens, 2006), implicating acceptance in lower fatigue and distress, and greater physical functioning and adjustment.

Acceptance and Commitment Therapy (ACT) is a trans-diagnostic third-wave psychotherapeutic approach that incorporates acceptance-based processes to facilitate

behavioral change. In contrast to therapeutic techniques that aim to change the frequency, form, or meaning of thoughts, ACT proposes that psychological suffering primarily arises as a consequence of attempts to avoid these unwanted private experiences (*experiential avoidance*), which in turn functions to reduce the frequency of personally-meaningful pursuits an individual engages in (*values-inconsistent behavior*; Hayes, Strosahl, Bunting, Twohig & Wilson, 2004). ACT aims to reduce experiential avoidance (in the service of increasing values-consistent behavior) by fostering greater *psychological flexibility* – “the ability to contact the present moment more fully as a conscious human being, and to change or persist in behavior when doing so serves valued ends” (Hayes, Luoma, Bond, Masuda & Lillis, 2006, p.7).

There is strong research support for the efficacy of ACT for long-term health conditions; ACT has been found to be beneficial for chronic pain populations (eg. Ghomian & Shairi, 2014; Hann & McCracken, 2014), in cancer (Feros, Lane, Ciarrochi, & Blackledge, 2013), epilepsy (Lundgren, Dahl, & Hayes, 2008), diabetes (Gregg, Callaghan, Hayes, & Glenn-Lawson, 2007), fibromyalgia (Wicksell et al., 2013), and tinnitus (Westin et al., 2011).

Many of the challenges associated with living with CFS could be understood within an ACT framework – for example, strategies to avoid fatigue, both behaviorally and cognitively, serve to increase distress, particularly when they compromise valued action. Similarly, changes to physical ability and stigma associated with the diagnosis challenge a rigid sense of self. The functional goal of an ACT intervention with CFS would be movement in the processes of the model, so an increase in flexible ways of relating to fatigue, and ability to change behavior when it is not allowing the achievement of desired goals in a specific context. Balancing acceptance processes with behavioral commitment, clients would be encouraged to reduce experiential avoidance of distressing thoughts and evaluations about

self and fatigue as well as feared bodily sensations, and to find ways to move towards valued life directions even when unpleasant fatigue thoughts and symptoms are present.

Chronic illness models place emphasis on the need for self-management, to maximise self-efficacy and control and to minimise dependency on health services (Bodenheimer, Lorig, Holman, & Grumbach, 2002; Nodturf et al., 2000). Behavior change is also more likely and more sustainable if it is personally meaningful and freely chosen; ‘activating’ the patient in their own treatment as part of a patient centred approach is consistently associated with improved physical health outcomes (Anderson, Funnel, & Arnold, 2002). This paradigm of chronic illness is compatible with both the application of self-help interventions and the broader idiographic treatment model of ACT. ACT appears suited to a range of delivery formats, including self-help (Ljótsson et al., 2014) and bibliotherapy (Johnston, Foster, Shennan, Starkey & Johnson, 2010) which are important treatment pathways to consider for those individuals whose physical and cognitive symptoms may make regular travel and attendance at clinics difficult.

Considering the findings from other physical health populations, as well as the issues associated with currently recommended treatments for CFS, the potential of acceptance-based approaches for individuals experiencing CFS warrants further examination. This project sought to investigate a guided bibliotherapy self-help intervention based on ACT with six participants from a specialist CFS service. A multiple single-case design with repeated and comprehensive mixed method measures was implemented in order to facilitate initial analysis of the potential change processes involved. Our specific aims were:

1. To examine the effects of an ACT self-help intervention on self-report and behavioral measures of change in people with CFS, specifically in relation to core processes outlined by the model underpinning ACT.

2. To explore via qualitative data whether this intervention might be feasible and acceptable for this population.

Method

This research was approved by the University of Lincoln Research and Ethics Committee, the NHS Trust Ethics Committee, and the East Midlands National Research Ethics Service Committee.

Design

A mixed-method multiple single-case series was implemented. The timings of the research were agreed pragmatically with each participant.

Participants

Six participants (meeting minimum standards for replication in single case designs: Kratochwill et al., 2013) were recruited from an established CFS service in the UK via written and verbal advertisement from the primary researcher and clinicians working within the service. Eight people expressed interest in the study; two were unable to take part within the required timescale.

Written informed consent was obtained from each participant prior to the study commencing.

Five participants were female, one was male; ages ranged between 19 years and 62 years (mean = 38.5 years, SD 15.22). CFS is diagnosed more frequently in women, so the gender imbalance in this sample is reflective of that documented within the broader CFS population (Afari & Buchwald, 2003). Participants were recruited after having received the standard service treatment of a 10 week group programme, all within the last 12 months: this programme was based on psychoeducation, socialising participants to a neurobiological model whereby different lifestyle factors are individually explored, then conceptualised as part of a 'jigsaw' of CFS (this previous experience was helpful to orientating the participants

towards the ACT model, whereby behavioral and cognitive components dynamically interact). All participants were considered to have a diagnosis of CFS in line with CDC criteria (Fukuda et al., 1994): the reported duration of CFS symptoms varied between participants (2-29 years) as did time since diagnosis (1-27 years).

Inclusion criteria

Participants were required to be aged 18 years and above, to have received treatment as normal, to have a good understanding of English and to be able to give informed consent throughout the research in order to take part. There were no further exclusion criteria.

Measures

Quantitative Measures.

A comprehensive battery of standardised self-report measures was implemented once a week (see below); four ACT process measures and two CFS measures were compiled into one online questionnaire totalling 79 items.

A condensed questionnaire, comprising 12 items, was developed to be administered a further two times each week, in consideration of participant fatigue and burden. For this, key items were selected from each of the ACT process measures based on highest factor loadings of each item (indicating construct validity), and through expert consensus between three Clinical Psychologist ACT practitioners (indicating face/content validity), with further reference to key texts (for example, Hayes, Strosahl, & Wilson, 2011). Given that items were picked to address key components of ACT theory, responses to this measure were taken as an indication of overall psychological flexibility, with responses adjusted so that higher scores indicated increasing psychological flexibility across all items.

ACT process measures.

The Philadelphia Mindfulness Scale (PHLMS; (Cardaciotto, Herbert, Foreman, Moitra & Farrow, 2008)

This questionnaire is designed to measure two ACT processes: acceptance and present moment awareness (mindfulness). We included another measure of mindfulness (the MAAS: see below) and so the PHLMS was largely used to monitor acceptance. Participants rate 20 items addressing experience with thoughts and emotions (e.g., ‘There are aspects of myself I don’t want to think about’) on a five point Likert scale (1 = never and 5 = very often). Odd items are summed for the awareness score, and even items are reversed and summed for the acceptance score. Scores can range from 10 to 50 on each scale: higher scores indicate higher levels of the components being measured. The two subscales were not correlated in previous studies, suggesting that they can be examined independently; Cronbach’s alpha was found to be .85 and .81 respectively, suggesting good internal consistency for both and the scale has also been shown to be able to distinguish between clinical and non-clinical samples of psychiatric outpatients and patients with eating disorders (Cardaciotto et al., 2008).

The Cognitive Fusion Questionnaire (CFQ; Gillanders et al., 2014).

This is a seven item self-report questionnaire assessing cognitive fusion. Items (e.g., ‘I got so caught up in my thoughts that I was unable to do the things that I most wanted to do’) are rated on a seven point Likert scale (1 = never true and 7 = always true), with higher scores indicating higher cognitive fusion. Elevated levels of fusion have been reliably identified amongst clinical samples in contrast to nonclinical samples, and the scale has demonstrated good internal consistency and test-retest reliability (alphas $>.80$; Gillanders et al., 2014); this scale has been considered valid and reliable in a chronic pain sample (see McCracken, DaSilva, Skillicorn, & Doherty, 2014).

The Mindfulness Attention and Awareness Scale (MAAS: Brown & Ryan, 2003).

The MAAS is a 15-item self-report inventory designed to measure attention to and awareness of what is occurring in the present moment. Items (e.g., ‘I found myself doing things without paying attention’) are rated on a six point Likert scale (1 = almost always and

6 = almost never), and the mean of these items is calculated to give a score from 1-6, with higher scores indicating greater mindfulness. The MAAS has been found to have good internal consistency, with Cronbach's alphas of .82 and .87 in clinical and non-clinical samples; clinical samples have been found to obtain lower scores (Brown & Ryan, 2003). This scale is considered to be reliable and valid within a physical health sample of cancer patients (Carlson & Brown, 2005). Preliminary analyses supported reliability and validity of the MAAS for patients with chronic pain (McCracken & Thompson, 2009).

The Engaged Living Scale (ELS; Trompetter et al., 2013).

Designed to assess an engaged response style, this questionnaire relates to the values and committed action aspect of the ACT model. Participants are asked to rate 16 statements (e.g., 'I have values that give my life meaning') on a five point Likert scale from 1 = not at all to 5 = totally agree. Total scores range between 16 and 80, with higher scores indicating a more engaged response style. The scale has been demonstrated in a normative sample to have good internal consistency with Cronbach's alpha .9, and it was thought to have incremental validity in explaining relationships between ACT theory and behavioural outcomes in a chronic pain sample (Trompetter et al., 2013).

CFS Measures.

SF-12 Physical Function subscale (Ware Jr, Kosinski, & Keller, 1996).

The SF-12 is itself a short version of the SF-36, a generic and widely used health-related-Quality of Life measure evaluated for use within general population samples as well as across a number of health diagnoses. Participants are asked to indicate on a three point Likert scale (0 = not limited at all, 5 = limited a little and 10 = limited a lot) the extent to which their health limits their ability to participate in 10 activities, such as 'Lifting or carrying groceries'. Scores range between 0 and 100, with higher scores indicating greater limitations.

Chalder Fatigue Questionnaire (Chalder et al., 1993).

This 11 item scale asks patients to rate their fatigue symptoms (e.g., ‘Do you need to rest more?’) on a four point Likert scale (0 = less than usual and 3 = much more than usual). Total scores range between 0 and 33, with a score of 11 indicating no change from usual, and higher scores indicating higher levels of fatigue; scores have been shown to reliably discriminate between clinically fatigued and non-clinical samples (Matteo & Chalder, 2010).

Behavioral Measure.

Participants wore a ‘Fitbit Flex’ Activity Tracker, a portable bracelet device monitoring physical activity in the form of ‘steps’. This information is uploaded to a computer via a USB device and accessed via the Fitbit website. Data were uploaded by the primary researcher with participants receiving no feedback until completion of the study (to limit any confounding effects of self-monitoring). There was no feedback function on the device itself. This outcome measure recognises the importance of functional improvement in evaluating CFS treatments and was included to improve ecological validity and to support self-report data.

Change Interview.

Following intervention (see below), participants took part in a change interview based on guidelines by Elliott, Slatick & Urman (2001) and Elliott (2002), to provide further context for the above self-report measures, and to ascertain which aspects of the intervention, if any, had promoted change and/or were useful from the participants’ perspectives (e.g., whether any negative impact of the intervention was noted; the importance of the therapy; the language used within the self-help book; individual descriptors of change; important contextual factors that may have impacted on the intervention). The interview was conducted by telephone by an individual external to the research team in order to limit demand

characteristics and to encourage open self-reflection. It was recorded and transcribed by the first author.

Procedure

Baseline.

Baseline measures were administered until at least five data points were recorded and trend lines appeared either stable or declining (approximately 2 weeks per participant), after which the intervention commenced. These individual baselines acted as control periods.

Intervention.

A section of text from the book '*Get out of your mind and into your life*' (Hayes & Smith, 2005) was sent weekly via email or post to participants. Each section was chosen and ordered by its relation to underlying ACT processes (see Table 1 below); participants did not read the book sequentially. Participants were encouraged to read the material each week and complete the exercises therein. The primary researcher checked comprehension and adherence via a phone call each week (approximately 10 minutes in length per participant).

Table 1

An outline of the intervention, including book chapters read each week, correlating ACT processes targeted, and the measures used to evaluate change in each of these.

Intervention Stage	ACT Process	Chapters	Content	Measures monitoring target process
Overview of ACT (Orientation phase)	N/A	1/2/3	<ul style="list-style-type: none"> • Introduction • Human suffering • Why language leads to suffering 	N/A
Week 1	Willingness (Acceptance)	4/9/10	<ul style="list-style-type: none"> • Avoidance • Willingness 	PHLMS
Week 2	Cognitive Defusion	5/6	<ul style="list-style-type: none"> • Trouble with thoughts • Having a thought vs. buying a thought 	CFQ

Week 3	Self as Context	7	• Three senses of self	MAAS
Week 4	Present Moment Awareness	8	• Mindfulness	MAAS
Week 5	Values	11/12	• What are values? • Choosing your values	ELS
Week 6	Committed Action	13	• Commitment / barriers	ELS

Follow up.

All participants agreed to complete the follow up three months after completion of the intervention.

The three phases of the study are graphically represented in Figure 1.

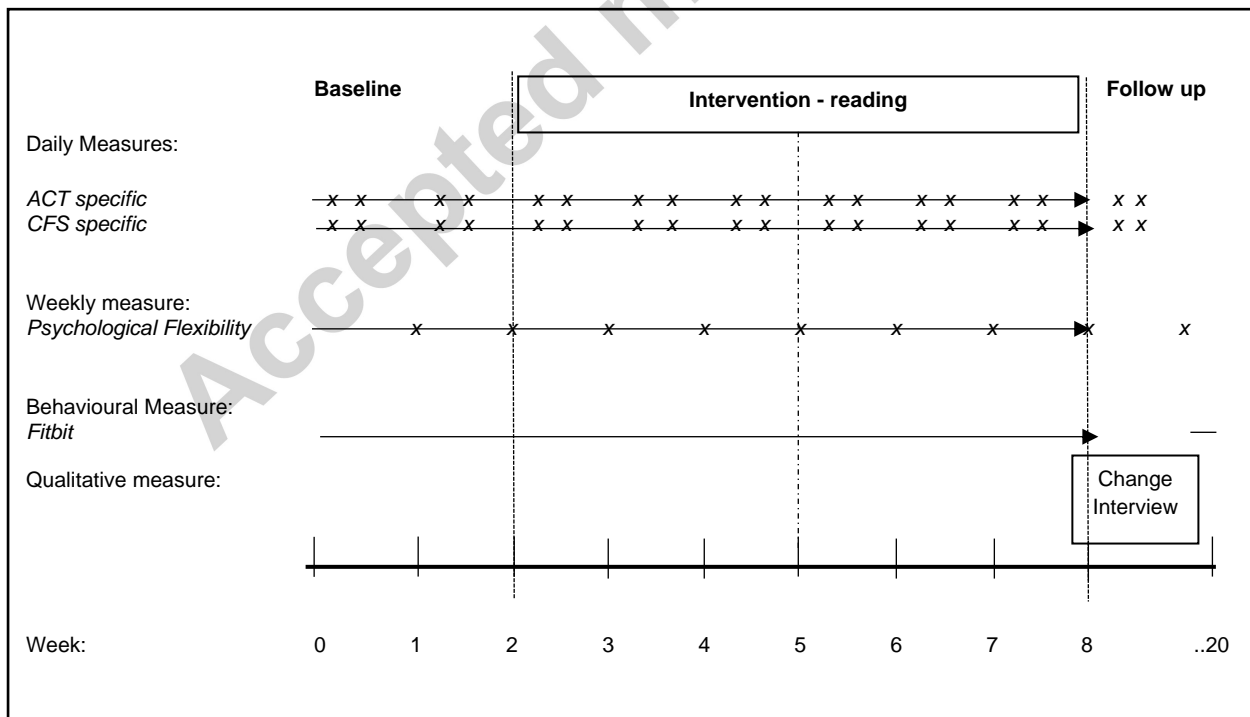


Figure 1. Diagram outlining procedure and measure collection throughout the study.

Throughout the baseline and intervention phase participants received an email link to the

online questionnaires three times each week - these are marked by an x in the figure. The Fitbit was worn throughout. Post-intervention participants completed the change interview. The complete battery of measures was completed at follow up.

Analysis

The condensed questionnaire items were summed to produce an overall score of psychological flexibility. These results were analysed using systematic visual analysis procedures, both within and across the two conditions of this study (Lane & Gast, 2014); analysis examined trend, defined as progress and direction over time; level, defined as magnitude of the data; and stability, defined as the variability or 'bounce' of data (Lane & Gast, 2014).

Full questionnaires were scored according to the original measures (the CFQ, and any items included in the condensed questionnaire, was reversed so that higher scores reflect positive attributes as in all other included ACT measures). These results were analysed through the calculation of reliable and clinically significant change indices (Jacobson & Truax, 1991). The reliable change index estimates whether an observed change represents a statistically reliable effect (i.e., whether it is greater than might be expected by chance or simple measurement error): this is calculated using a function of the standard deviation of the measure used, and uses a confidence interval of 95%. If change is deemed reliable, it can then be further evaluated as to whether it is clinically significant: that is, whether the individual has moved from being within a range of scores typically obtained by a clinical sample to a range obtained by a non-clinical, healthy sample. Given that data was available for clinical and non-clinical means for the measures used, Jacobson's second and third criteria for calculating clinical significant change were used (see https://medhealth.leeds.ac.uk/info/618/clinical_psychology_dclinpsychol/797/leeds_reliable_change_index).

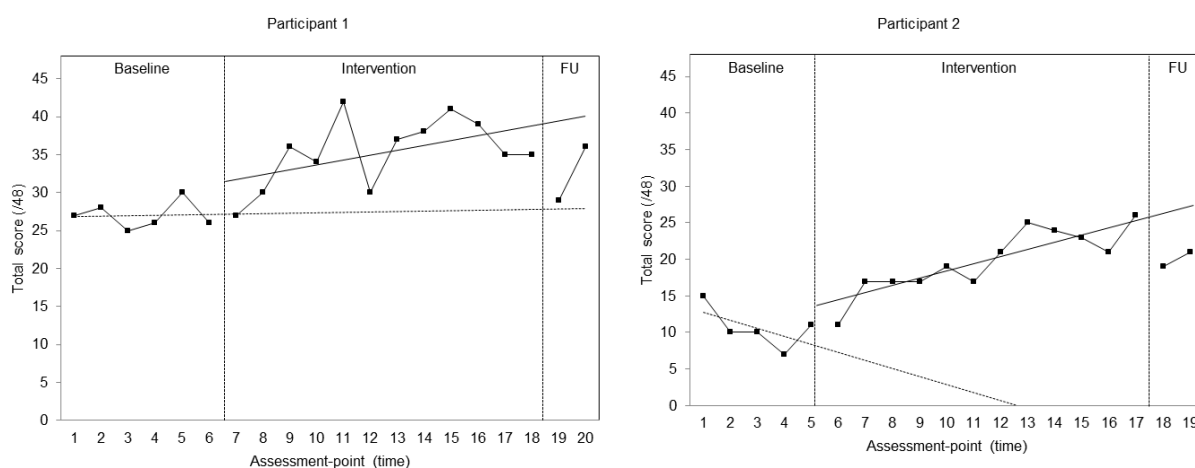
Quantitative data analysis was considered in conjunction with qualitative data from the change interviews: for example, participants were asked which aspect of the intervention they thought had been most useful, or to describe any particular changes resulting from the intervention, which was then considered as largely consistent or inconsistent with change scores on other outcome measures.

Data regarding completion rates, difficulties encountered, and data from change interviews were considered by the researchers to explore the feasibility and acceptability of the intervention.

Results

Overall psychological flexibility

Overall, psychological flexibility appears to have increased upon introduction of the intervention and maintained at follow up beyond what might have been expected from baseline trends in three of six participants. The data were highly individual for each participant, however, and variable in terms of the visual analysis: as such they need to be interpreted with caution.



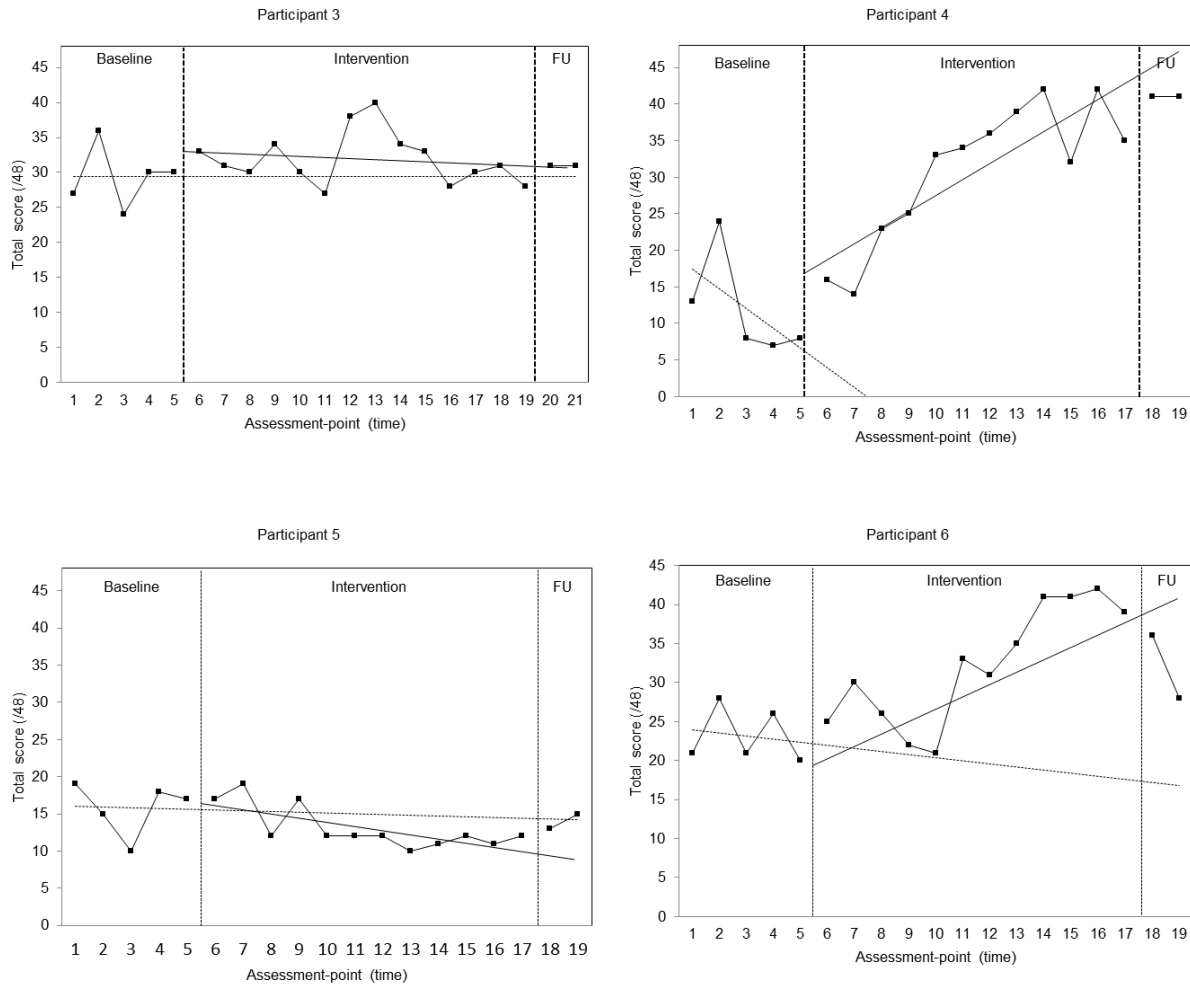
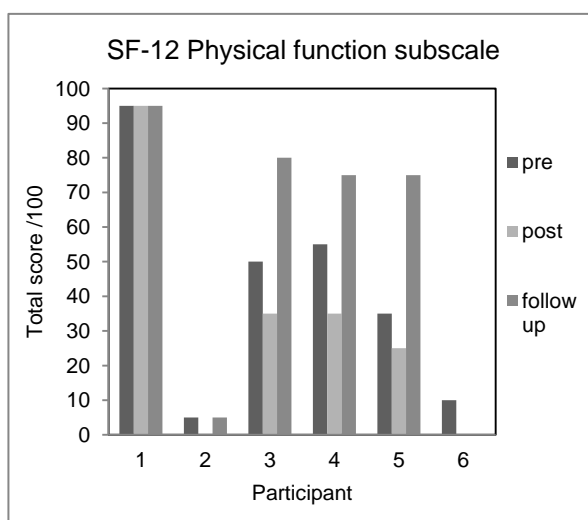
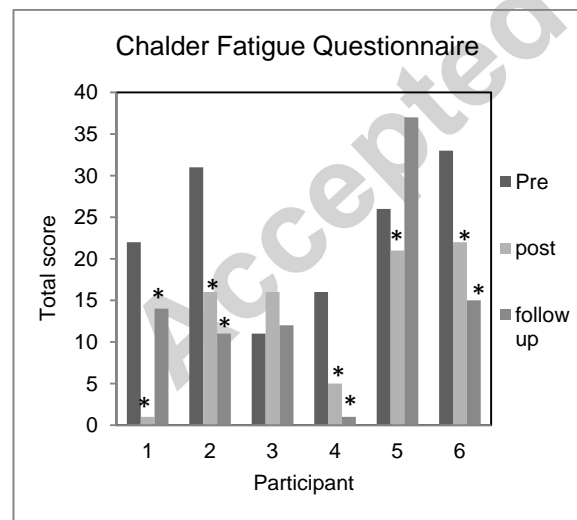
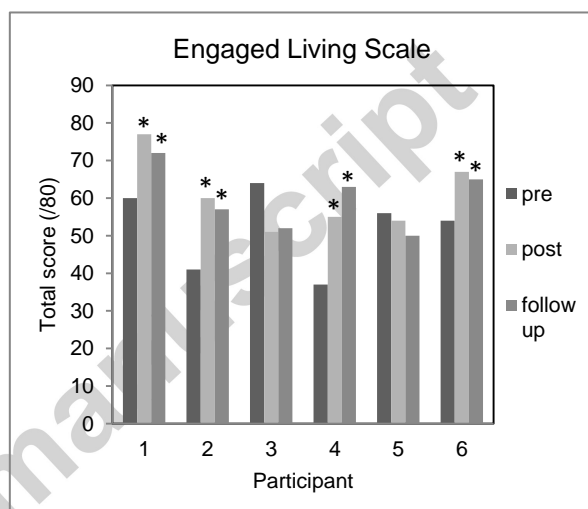
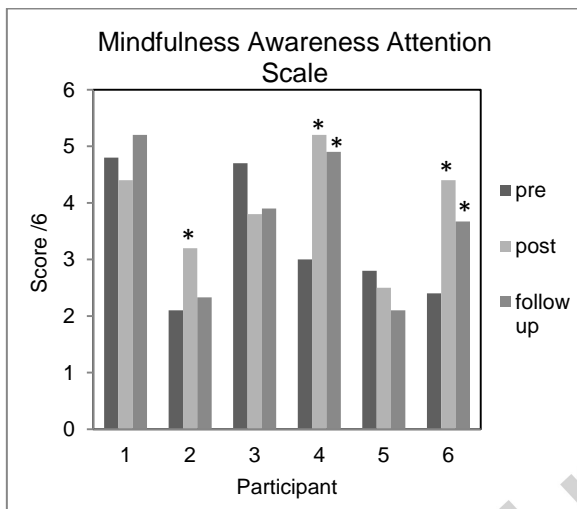
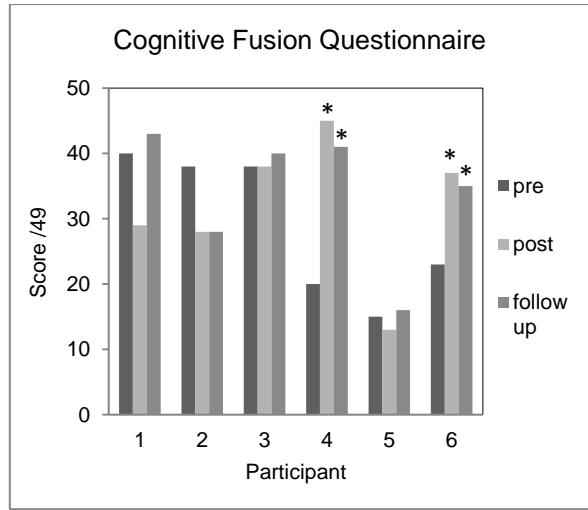
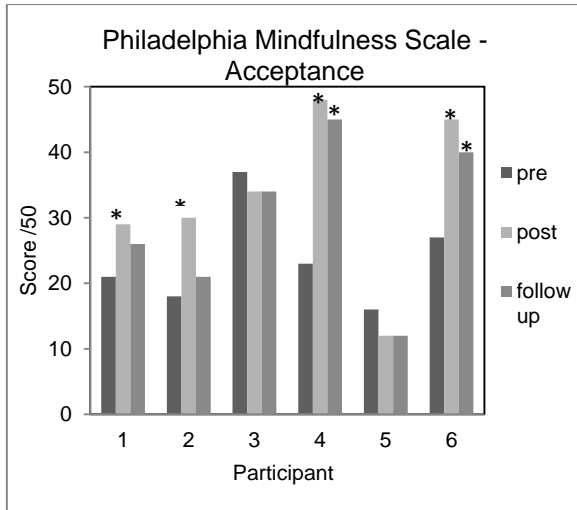


Figure 2. Graphs depicting overall psychological flexibility scores for each participant – dotted lines indicate trend lines of baseline data, whilst solid lines indicate trend lines of intervention data. FU = Follow-Up period.

ACT Process Measures

As demonstrated by the graphs below, scores on other outcome measures were also highly variable.



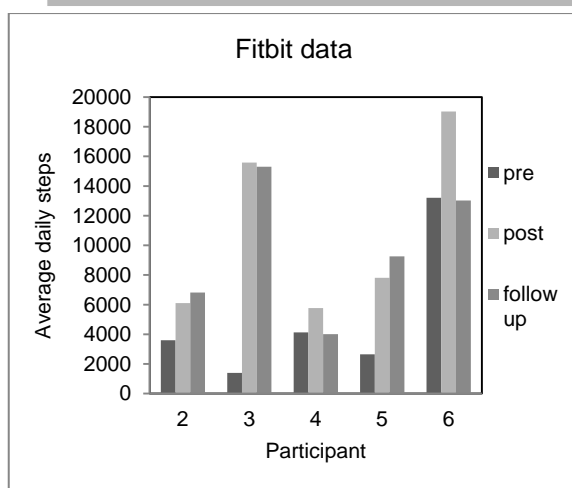


Figure 3: Scores on key outcome measures pre and post self-help ACT intervention, and at follow up. Asterisks on the ACT process measures and the Chalder Fatigue Questionnaire indicate change from baseline that was calculated to be clinically significant.

Five participants were below the average score on the ELS at baseline (60.8): four of these showed clinically significant improvements that were maintained at follow up. On the acceptance scale of the PHLMS, five participants were below the normative mean at baseline (30.19) and four were below the clinical mean (24.62). Four participants indicated clinically significant change from baseline to post-intervention; two participants maintained this at follow up.

Participants indicated variable fatigue levels; five participants demonstrated clinically significant improvement at post-intervention, four of which indicated that this was maintained or further improved at follow up. Scores on the SF-12 subscale indicate varying levels of functional impairment, which appeared to decrease at post intervention in five participants but was not maintained at follow up.

There was an observed increase in recorded physical activity at post-intervention in all five participants; this appeared maintained at follow up in three participants. There was

insufficient data to analyse for participant 1 due to technical and set-up difficulties with the monitor.

Change Interviews

In line with Elliott (2002), data from the change interviews were evaluated in terms of their congruence with data from the quantitative measures. During the supportive telephone calls throughout the intervention, participants reported completing all reading and evidenced this in their ability to talk convincingly about the content.

Overall, participants' reports appeared to be largely congruent with quantitative measures in all but one participant, participant five, who spoke about positive changes that were not evident in the ACT process data. Where change was otherwise noted, participants were able to give detailed examples of applying strategies and real life change, including job changes and direct efforts to increase valued activity (physical activities and activities with family), indicating good engagement with and comprehension of the material, adding support to the validity of the results. Three participants (participants two, four and five) attributed important changes directly to the intervention: a further two (participants three and six) thought improvements would have happened naturally but that the intervention facilitated it more quickly. Half of the participants noted that the timescale of the intervention was demanding: two participants had a week mid-intervention where data collection was ongoing but no additional reading was introduced. These difficulties were notably exacerbated by the participant's cognitive symptoms of CFS including poorer memory, attentional ability, and increased mental fatigue. Despite these difficulties, participants spoke about revisiting the text in their own time, indicating that they saw some value in it for managing their CFS in the longer term, but that the format of the intervention, in terms of cognitive demand and time, may be difficult – the implications of this are considered further in the discussion. Four

participants recommended this text to others with CFS, indicating good acceptability of the content of the intervention.

Discussion

We investigated the relationship between a self-help ACT intervention and a number of outcome measures in six participants with CFS. The findings suggest that this intervention is acceptable for the population – no adverse effects were noted. Three of six participants demonstrated an improvement beyond what would have been expected from baseline trends in the primary process measure of psychological flexibility. There was clinically significant change on two processes: the values component indicated improvements for four participants that were also maintained at follow up and supported by qualitative data - this is furthermore a distinct addition to current therapeutic components. Acceptance scores were initially low, in line with previous literature; although this improved for four participants, it was not maintained. All participants wearing the Fitbit monitor evinced increased activity.

Psychological flexibility

Measures of overall psychological flexibility were complex. We hypothesised that psychological flexibility would increase during the intervention, which was the case in three participants: this is enough to satisfy recommended replicability criteria (Kratohwill et al., 2010), allowing some ability to conclude that these processes are demonstrable across cases. However, with only half of the participants improving there are clearly questions remaining about this differential responsiveness – these are discussed in further detail below.

Valued living

It was notable that clinically significant, positive change occurred and was maintained in four out of six participants on the Engaged Living Scale. This was further supported by qualitative data, with similar numbers of participants reporting that the values aspect of the intervention was most useful. ACT allows a highly idiographic approach to facilitating

behavior change aligned with personally meaningful activities, which may contrast with a more structured CBT approach; explicit values work is not a focus of either CBT or GET and so this finding indicates an important area of potential added value.

Physical Activity

Another noteworthy finding was the increase in physical activity observed in all participants wearing the Fitbit activity monitor. Previous interventions have included increased activity as a target for intervention: physical activity has beneficial effects on general health and mood (Penedo & Dahn, 2005), and the more specific processes of building up physical strength and resilience are thought to be important for a positive prognosis with CFS (White et al., 2011). Nonetheless, interventions specifically targeting activity levels are controversial, with detrimental effects noted by a proportion of the CFS community. Increased activity was not an explicit focus of this intervention, and there was a limited 'dose' of intervention in terms of time (6 weeks) and format (guided bibliotherapy), yet there was a notable increase in physical activity for some participants. Within the ACT model this change in activity could be understood to arise from increased commitment to engage in valued behaviors, reflecting scores on the ELS. From this perspective, an increase in engaged living is a key outcome in its own right, which adds further promise to this approach.

There are, however, some limitations with the Fitbit data – for example, data were necessarily condensed into a weekly average due to variable data collection. The baseline therefore comprises a single measure, limiting the ability to reliably assess change following the introduction of the intervention. Although the role of the activity monitor itself as a mechanism of change is possible (Fritz, Huang, Murphy, & Zimmermann, 2014), participants did not have access to feedback about the data during the intervention and so the role of reinforcement (via self-monitoring feedback) was minimised. Similarly, the maintenance of change in three participants over three months without the continual presence of the Fitbit

indicates that there are other underlying change processes operating. None of the participants cited the Fitbit as an important change factor.

It is important to note, in consideration of literature noting unhelpful thinking styles and detrimental patterns of activity characteristic of CFS (eg. Sirois & Molnar, 2014; Kempke et al., 2013), that self-monitoring may be powerful motivator for this population, although not necessarily a helpful one. Within the timeframe of the study, no adverse effects were noted by any participants. In line with other considerations as to the strength of the ACT model, it was considered that an idiographic and values based approach may be safer as well as more effective.

Acceptance

Clinically significant change replicated across at least three participants was also noted on the Acceptance scale of the PHLMS. Participant's baseline scores were lower than a comparable clinical mean, which is in line with previous literature suggesting that acceptance is an important consideration for this sample (Van Damme et al., 2006) and a key candidate for intervention. Clinically significant increases in levels of acceptance in four participants was promising, but the lack of maintenance perhaps again raises questions about the 'dose' of this intervention. Future research should look to determine whether additional clinician input or a longer time period could maintain this positive effect.

Self-reported functional impairment and symptoms

Focusing instead on second order change, symptom reduction is not a primary goal of ACT, although it is commonly noted as a secondary benefit. The overall trend within this study was one of decreasing fatigue throughout the intervention, largely maintained at follow up (with participant five being the notable exception). The change in fatigue scores mirrors the increase in activity, and although there isn't a clear pattern across participants about which preceded the other, it seems probable that they might be interlinked, with increased

activity likely to have a positive effect on other systems such as sleep, mood and energy (Penedo & Dahn, 2005).

Reporting of physical impairment did not correlate with fatigue, with a trend of reduced impairment post-intervention reverting to baseline levels at follow up. This may indicate that change on other measures, such as valued living, occurred even in the context of limited change in symptoms, in line with an ACT model. Furthermore, this reflects the pattern of change in acceptance scores, and although directionality is unclear from the observed data, this finding would be compatible with previous literature and ACT theory indicating that increased acceptance can have a positive impact on symptom experience (Van Damme et al., 2006).

Strengths

Although the text used was not tailored to be specific to CFS, neither was it specific to other or incompatible understandings of well-being or distress, and it was considered a strength that ACT is a transdiagnostic model - whilst it was possible that this approach could have felt invalidating to participants, recognising the importance of addressing the idea that is not simply 'all in your head', the previous group programme of which all participants had previously been a part of would have begun to address this idea. It may be precisely the holistic and flexible nature of the intervention that is particularly suited to the heterogeneity of CFS presentations because it encourages individuals to ideographically identify values and challenges, be that physical or cognitive, and tailor the approach themselves. This was further supported in this project by the regular researcher contacts, helping to clarify the rationale and personal applicability of these processes. The acceptability of this model in this intervention was evidenced by the lack of dropout and disengagement.

Limitations

The use of a multiple single case design was suitable for the exploratory nature of the study and the population of focus, in order to capture complexity that is often missed in group-based designs (e.g. the PACE RCT outlined above), and to facilitate a detailed analysis and consequent discussion of potential change processes. However, there are also limitations with this design. There is ongoing debate about and development of standardised analysis methods for single case designs that seek to maximise reliability and validity of such results. The analysis chosen here did indicate variability across the data and it is acknowledged that high levels of experimental control were somewhat compromised in order to facilitate participant engagement and maximise ecological validity. Participants represented the complexity associated with CFS, reporting other health concerns and important external events. Although steps were taken to collect detailed information through frequent and mixed measures, ultimately the results gave a multifarious picture, a recognised characteristic of such designs (Elliott, 2002). Similarly the self-help method was practical as well as in line with chronic illness literature, but also limited experimenter control and may have had further clinical disadvantages that could be explored with different formats of this intervention in the future.

We did consider randomising the start-points of intervention as literature would indicate is the most robust approach to such designs, however we decided against this due to the burdensomeness of assessment and considerations of unnecessary response-fatigue in this population. Whilst random assignment to different length baseline-phases would help to bolster internal validity, we made an ethical and pragmatic decision to minimise the baseline phase. We also considered randomising the order of the components within the ACT workbook, but we felt that the book had a broadly linear structure, such that it was unclear whether randomisation of components would undermine the coherence of the intervention (or in fact whether the 'components'/sections were meaningfully discrete). Finally, it is likely

that due to the low number of observations, again pragmatically minimised for purposes of limiting response burden, phases (only AB given that it would not have been possible to return to A phase having introduced intervention/learning), and participants, we would not have sufficient power to detect even large effects using randomisation tests (for example, discussed by Heyvaert & Onghena, 2014).

Other limitations of the study design include the potential for selection bias given that individuals volunteered themselves to the study; however, there is little evidence to suggest that this sample was systematically atypical in comparison to the wider client group, although it is not known how individual personality traits might have impacted on the complex results. Data from the change interviews indicated that all participants were actively engaged with the material provided; given the high levels of perfectionism noted in this population and the recruitment procedure, it is possible that these participants were highly motivated to achieve target outcomes. Further exploration of individual factors within the context of this therapeutic approach would be beneficial in facilitating an understanding of who could benefit most from such an intervention and to identify potential barriers to change.

The psychological flexibility measure used in this study was developed from validated questionnaires drawn together in a novel format; the psychometric properties should therefore be further evaluated with regards to the underlying model and the results of this study interpreted with these limitations in mind. Daily full-length measures would have given a more detailed understanding of individual change processes, but this design would have increased participant burden and have a potentially negative impact on study engagement, particularly in view of feedback suggesting that participants found the applied procedures to be demanding. Also, the less frequent use of measures may have helped to balance any threats to validity resulting from instrumentation effects of frequent assessment

over time. Similarly, the measures used to record the specific ACT processes have well-documented psychometric properties.

Conclusion

In summary, this study aimed to explore the effect of a self-help ACT intervention for six participants with CFS. It was largely exploratory, building upon a body of research suggesting that increased acceptance in chronic health conditions can lead to positive psychosocial and physical outcomes. Despite some limitations and a complex picture of results, there are some important implications for further research. ACT might be a more acceptable therapy for individuals experiencing CFS, given the idiographic emphasis on personally meaningful values rather than increased activity per se. It would be clinically useful to establish whether an adapted format could promote stable increases in acceptance and engaged living over time. A longer follow-up period would be useful to determine whether valued behavior change was maintained. Finally a more detailed understanding of who would benefit from this intervention could improve client outcomes and service provision.

Conflict of interest

There is no conflict of interest to declare by the authors.

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Highlights

- Acceptance and Commitment Therapy can improve wellbeing in chronic health conditions
- Chronic Fatigue Syndrome is a debilitating and challenging chronic syndrome
- We examine guided ACT self-help in a case-series of individuals with CFS
- ACT was linked with increased physical activity and valued behaviour
- Across cases, evidence for effects on psychological flexibility was mixed