

A pilot case series of a brief acceptance and commitment therapy (ACT)-based guided self-help intervention for improving quality of life and mood in muscle disorders

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Abstract. This study aimed to demonstrate proof of concept and acceptability of a brief acceptance and commitment therapy (ACT)-based guided self-help intervention for improving quality of life (QoL) and mood for people with muscle disorders (MD). A case-series with an AB design was used to assess changes in primary (QoL) and secondary (depression and anxiety) outcome variables across the period of study. Change in the psychological process targeted by ACT – psychological flexibility – was also investigated, to allow insight into possible treatment mechanisms. Post-intervention, participants also completed a brief free-text evaluation. Relative to pre-intervention scores, four (of seven) participants showed varying degrees of improvement in all primary and secondary outcome variables and were thus considered responders. However, consistent concomitant improvements in psychological flexibility were not apparent. Participants reported a mostly positive experience of the intervention; all appeared to complete the intervention, and no adverse events were reported. Nonetheless, there was evidence that those with compromised concentration or who report good initial QoL and low levels of distress may derive less benefit. Although several methodological weaknesses limit the strength of our conclusions, this ACT-based guided

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self-help intervention shows encouraging utility for improving QoL and mood in MD.

Key words: acceptance and commitment therapy, psychological flexibility, mood, quality of life, muscle disorders, muscular dystrophy

Introduction

The category of muscle disorders (MD) comprises genetic and acquired conditions such as facioscapulohumeral muscular dystrophy, inclusion body myositis and limb-girdle muscular dystrophy. Implicated muscle pathology generally leads to a progressive decline in mobility – often resulting in considerable functional impairment. Some MDs may also cause varying degrees of respiratory and cardiac symptoms, dysphagia, dysarthria and ophthalmoparesis (Merrison and Hanna, 2009). Currently, no disease modifying treatments are available for most MDs. Thus management is usually symptomatic, involving physiotherapy and pain and fatigue management, alongside cardiorespiratory monitoring (Merrison and Hanna, 2009).

Qualitative studies suggest that the functional impairment caused by MD can elicit difficult thoughts and emotions (Natterlund *et al.*, 2001), comprising challenged identity and fears about the future (Natterlund *et al.*, 2001; Graham, 2012). Given this demanding context it is unsurprising that average quality of life (QoL; Graham *et al.*, 2011; Burns *et al.*, 2012) and mood (Kalkman *et al.*, 2007; Blumen *et al.*, 2009) are adversely affected by MD.

Nonetheless, there is considerable variation in QoL and mood between people with MD (Graham *et al.*, 2013), and some do experience excellent QoL and mood, even with high levels of disease severity. Empirical studies suggest that differential ways of responding to the challenges presented by MD helps to explain this variation. In addition to the functional impairment caused by the condition, psychological/behavioural factors, including illness perceptions, psychological flexibility and coping methods, explain significant proportions of the variance in QoL and mood (Sadjadi *et al.*, 2010; Graham *et al.*, 2011, Rose *et al.*, 2012; Graham *et al.*, 2014, 2015, 2016a). Thus an intervention that targets psychological/behavioural factors might improve outcomes in MD.

To date there has been just one published study of a psychological intervention in MD (Voet *et al.*, 2014). Here, a traditional cognitive behavioural therapy (CBT) intervention was designed to improve fatigue in those with facioscapulohumeral MD. This intervention elicited a significant improvement in the primary outcome, fatigue. Encouragingly, it also appeared to improve secondary outcomes which overlap with mood and QoL, such as physical activity, social participation and sleep. However, there have been no trials of psychological interventions designed to primarily target broader outcomes, such as QoL and mood. Therefore we developed a psychological intervention to improve QoL and mood in MD.

Acceptance and commitment therapy

Acceptance and commitment therapy (ACT; Hayes *et al.*, 2006) is a newer model from within the CBT family. ACT focuses on improving psychological flexibility, which is defined as: being open, aware and in contact with the present moment, flexibly engaging in behaviours which facilitate overarching life goals. Psychological flexibility includes six overlapping components: *experiential acceptance*, taking an open stance towards private experiences (e.g.

pain, fear, embarrassment, excitement); *contact with the present moment*, attending to the here-and-now; *cognitive defusion*, seeing thoughts as separate from the person doing the thinking; *self-as-context*, flexible perspective-taking; *values*, knowing with clarity what is personally meaningful; and *committed action*, choosing to take actions that are consistent with one's values. ACT uses several methods to engender psychological flexibility. For example, aspects of mindfulness may be used to increase present moment focus; perspective taking to develop a flexible view of one's self; 'defusion' methods to undermine the literality of thoughts; and variations of behavioural activation can be used to bring behaviour in line with personally held values (Hayes, 2005; Harris, 2009).

There is evidence that both traditional CBT (Hofmann *et al.*, 2012) and ACT (Graham *et al.*, 2016b) are helpful for improving outcomes, such as QoL, mood and functioning, in chronic diseases. Traditional CBT may be most suitable for targeting illness perceptions, which have been shown to correlate with QoL and mood in MD (Graham *et al.*, 2014). ACT instead targets psychological flexibility, which was found to predict QoL and mood in a previous longitudinal study with an MD population (Graham *et al.*, 2016a). ACT has not yet been investigated in the context of MD. We thus trialled ACT as a novel approach to improving QoL and mood in MD. To improve uptake, the intervention was kept brief (three sessions). To reduce the burden of travelling for therapy, we delivered the guided self-help materials via e-mail with telephone support from a therapist.

Aims

In this study we wanted to: (1) investigate proof of concept by observing whether primary (QoL) and secondary (depression and anxiety) outcomes improved across the period of the intervention; (2) test the acceptability of the intervention via reports of adverse events, evaluation of deterioration in outcomes and feedback from patient experiences of the intervention; and (3) assess whether the intervention might affect changes in outcomes via the presumed treatment mechanism – improvement in psychological flexibility. We thus undertook a case series evaluation of a brief ACT-based guided self-help intervention, with seven participants. As ACT takes a salutogenic approach to well-being, in this initial study we did not select participants on the basis of pre-existing poor QoL or distress.

Method

Design

An AB case series design was utilized, with outcome and process variables measured at multiple time points pre-, during and post-intervention. A post-intervention free-text participant evaluation was also included.

Participants

Participants were recruited from throughout the United Kingdom via an advertisement placed on the Muscular Dystrophy UK website. This advertised for participants to trial a psychological skills manual – to investigate whether changing some ways of living with MD could improve QoL and mood. Participants initially self-assessed themselves against the inclusion/exclusion criteria. These criteria were kept broad, in keeping with the idea that the

intervention was primarily designed to enhance QoL, with a secondary impact on distress via improved psychological flexibility. Inclusion criteria were: a diagnosis of MD; duration of MD greater than six months; and over the age of 18 years. Exclusion criteria included: a diagnosis of myotonic dystrophy, which is associated with cognitive impairment (Minnerop *et al.*, 2011); major active co-morbidities unrelated to MD; major diagnosed active mental health co-morbidities (e.g. psychosis, obsessive compulsive disorder); and current participation in other treatment intervention studies. The first seven participants who met the inclusion/exclusion criteria were recruited to the study.

Procedure

NHS ethical approval was gained for the study (National Research Ethics Service Committee London-Brent; 12/LO/0487). Participants self-assessed as meeting the inclusion/exclusion criteria contacted a researcher via telephone. Following a brief review of the inclusion/exclusion criteria with the researcher, they were then sent study materials – including a consent form. (The presence of mental health diagnoses was not formally assessed.) Upon receipt of a completed consent form, participants were sent an e-mail containing a link to the first weekly online questionnaire battery (time point 1). This questionnaire battery was then sent on seven more occasions: at weekly intervals for six further weeks (time points 2–7), with a follow-up questionnaire sent at week 10 (4 weeks post-intervention; time point 8). The intervention began immediately following the completion of the third group of questionnaires, and continued for three weeks (between time points 3 and 6). At post-intervention, participants gave a brief evaluation of their experience of the intervention

Intervention

The intervention involved three weekly modules of guided self-help (Table 1). Each module comprised a PDF containing written self-help material, audio tasks (MP3 files) and homework tasks. We estimated that each module would take between 60 and 90 minutes to complete, excluding homework tasks. Each module was followed by a 15 to 30 minute telephone call with a therapist.

The written and MP3 content of the intervention was informed by several sources (Hayes *et al.*, 1999; Hayes, 2005; Harris, 2009). The booklets were assessed and edited by a clinical psychologist with expertise in ACT. The therapist was a PhD student in clinical health psychology, with training in CBT, and with limited clinical experience of ACT. The therapist's telephone calls were supervised by a CBT therapist, with experience of delivering ACT. The content of the telephone call was mostly participant-directed. Here the therapist sought to reinforce responding that was in line with psychological flexibility, as presented in the modules. Neither the modules nor the therapist behaviours were formally assessed for ACT treatment fidelity.

Measures

Primary outcome

Visual Analogue Scale for Quality of Life (VAS QoL; de Boer *et al.*, 2004)

The VAS QoL is a single-item QoL measure. Here participants indicate their QoL on a single-dimensional scale, ranging from 0 ('worst possible quality of life') to 100 ('best possible

Table 1. Description of the intervention techniques used, by module, modality (booklet, MP3, homework task), and the targeted aspects of psychological inflexibility

Module	Psychological flexibility process	Content: booklets	Content: MP3	Content: homework tasks
1	Acceptance	<p><i>Introduction</i></p> <p>Describing common emotional responses to MD</p> <p>Noticing current behaviours, e.g. ‘what do you normally do with unwanted thoughts and feelings?’</p> <p>Willingness exercises, involving experiential tasks e.g. ‘don’t think of the white bear’</p> <p><i>Intervention</i></p> <p>Acceptance and recognizing/observing emotions and thoughts that occur in relation to MD. Use of metaphors: relaxing during a volcano, or when facing a tidal wave; tug-of-war with a monster</p>		
	Defusion	<p><i>Introduction</i></p> <p>Noticing when we struggle with thoughts, framed in relation to common emotional responses to MD. The mind conceptualized as a salesman, selling junk thoughts that can take us off-track</p> <p><i>Intervention</i></p> <p>Noticing where/when we get taken when hooked by thoughts and where this takes us; ‘milk, milk, milk’; ‘I am having the thought that ...’</p>		Noticing when you have been hooked in by a thought. Using a variation of ‘I am having the thought that ...’ to unhook from that thought
	Contact with the present moment	<p><i>Introduction</i></p> <p>Where are <i>you</i> when you are struggling with thoughts?</p> <p><i>Intervention</i></p> <p>Introduction to present moment awareness tasks. Noticing that (understandable) illness-related worrying/rumination can take us away from here-and-now</p>	Mindfulness tasks: ‘turning the struggle switch off’; anchoring attention in the now	Five minutes mindfulness practice per day – present moment awareness and/or turning the struggle switch off

Table 1. *Continued*

Module	Psychological flexibility process	Content: booklets	Content: MP3	Content: homework tasks
2	Values	<i>Introduction</i> Description/contrast of ‘values’ and ‘goals’; values as a direction of travel. Values in the context of declining mobility		
	Committed action	<i>Intervention</i> Ninetieth birthday task; values compass <i>Introduction/intervention</i> Following on from the values compass, what is the smallest possible step that you can take to get back on track with your values?		Smallest possible step: consistently choosing small steps towards values
	Acceptance	<i>Introduction/intervention</i> Passengers on the bus metaphor: uncomfortable thoughts and feelings as passengers on a bus		
	Self-as-context		Notice who is doing the noticing task: observing that thoughts/feelings can be experienced as separate from one’s self	
	Contact with the present moment			Five minutes mindfulness practice per day – present moment awareness and/or turning the struggle switch off

Table 1. *Continued*

Module	Psychological flexibility process	Content: booklets	Content: MP3	Content: homework tasks
3	<p>General psychological flexibility</p> <p>Contact with the present moment</p>	<p><i>Introduction</i></p> <p>Unwelcome party guest metaphor. Encouraging the noticing of experiences/events that occurred/got in the way of the ‘smallest possible step’ homework exercise</p> <p><i>Intervention</i></p> <p>Recap of the defusion and mindfulness exercises that can be applied to enable values-consistent-action with MD and in the presence of difficult thoughts and feelings</p>	<p>Flexible attention task: shifting attention to various aspects of the present moment</p>	

quality of life'). Previous psychometric assessment of the VAS QoL with chronic disease populations has shown good reliability, validity and responsiveness (de Boer *et al.*, 2004).

Secondary outcomes

The Hospital Anxiety and Depression Scale – depression and anxiety sub-scales (HADS; Zigmond and Snaith, 1983)

The HADS is a validated (Mykletun *et al.*, 2001; Bjelland *et al.*, 2002), 14-item measure of anxiety and depression. There are two scales (depression, anxiety), which were used as independent measures of depression and anxiety. Scores range from 0 to 21 for each sub-scale, and higher scores indicate greater mood disturbance. Scores above 11 on either scale are considered to indicate the possible presence of a 'mood disorder' (Zigmond and Snaith, 1983).

Measure of treatment mechanism

The Acceptance and Action Questionnaire (AAQ-II; Bond *et al.*, 2011)

The AAQ-II (Bond *et al.*, 2011) was used to measure psychological flexibility, the process targeted by the intervention. It consists of seven items; scores range from 7 to 49, with higher scores indicating greater inflexibility. It is the most widely applied measure of psychological flexibility and several studies suggest that it has good validity and reliability (Bond *et al.*, 2011; Fledderus *et al.*, 2012). Nonetheless, others have noticed problems with the measure: that it largely captures distress (Wolgast, 2014), and measures only some aspects of psychological flexibility – mostly experiential avoidance (Francis *et al.*, 2016).

Brief patient evaluation/acceptability measure

A brief measure of patient experience of the intervention was developed and used to allow further insight into the acceptability of the intervention. This measure consisted of one item, which asked participants to indicate whether the intervention had been helpful: 'Did the intervention help you?'. Responses were made on a scale of 1 (no, it made things worse) to 6 (yes, it was extremely helpful). They were then asked to support their selection using an open-ended free text response.

Other measures

The Stanford Health Assessment Questionnaire Disability Index (HAQ-DI; Bruce and Fries, 2003b)

The HAQ-DI was used to measure each participant's level of physical disability (excluding psychosocial or emotional functioning). We used the alternative scoring method (Bruce and Fries, 2003b), which has demonstrated sound psychometric properties (Bruce and Fries, 2003a). Here scores can range from 0 to 3, with higher scores indicating greater levels of disability. The HAQ-DI was used to describe the sample, but was not used as an outcome measure.

Analysis

Changes in measures of the primary (VAS QoL) and secondary (HADS depression and anxiety sub-scales) outcome variables, and the process variable (AAQ-II), were investigated using visual analysis. We chose this method as it allows detailed understanding of change in variables across the period of study. In visual analysis, scores across pre- and post-intervention time points are analysed to take into account the level (i.e. the mid-point across a series of data, considered horizontally), the variability in the data, and the trend (i.e. the direction of the data path, increasing or decreasing; Harrington and Velicer, 2015). Inference to an intervention is made if the level and/or trend of scores in the intervention phase visually differs from the level and/or trend of the scores in the pre-intervention phase (Kazdin, 1978).

Our three pre-intervention time points allowed for an indication of the baseline level and variability of scores, which was compared with post-intervention scores. Trend across the period of the intervention was also considered. Consequently, we used the following criteria to infer an intervention-related improvement in a given variable: (a) improvement in score over the intervention period (i.e. a trend demonstrating improvement); with (b) improved level at post-intervention, taking into consideration; (c) the variability of pre- compared with post-intervention scores, with the majority of post-intervention scores (i.e. at least two out of three) outside of the range of pre-intervention scores.

Participants were classified as responders if there was an improvement (as per the aforementioned criteria) in all three primary and secondary outcome variables (QoL, anxiety and depression). As described, change in psychological flexibility (AAQ-II) was assessed to investigate whether the intervention affected the processes it was designed to target; however, this variable was not used to categorize responders and non-responders.

Aspects of acceptability were assessed via the reporting of adverse events and observation of change in outcome variables (i.e. clear evidence of deterioration via the aforementioned criteria). To give insight into participants' experiences of the intervention an item (described previously, regarding utility of the intervention) and associated free-text evaluation were included. Completion of the intervention was considered to have occurred where a participant attended all telephone calls with the therapist.

Results

There were seven participants, five men and two women, who were aged in their thirties to sixties. They had a range of MD diagnoses, including facioscapulohumeral muscular dystrophy, limb-girdle muscular dystrophy and vacuolar myopathy. They had lived with a MD diagnosis for between 1 and 34 years.

Over the course of the intervention four participants showed improvements in all primary (QoL) and secondary (depression and anxiety) outcomes; these participants were considered to be responders (Figs 1–4). Three participants did not show improvement in all three outcome variables; these participants were considered non-responders (Figs 5–8). The following sections will describe each participant's progress across the period of study.

Responders

Participant 3, a man in his forties, had been recently diagnosed with a MD and had a moderate level of functional impairment (HAQ-DI = 0.55). His QoL was initially quite low, but

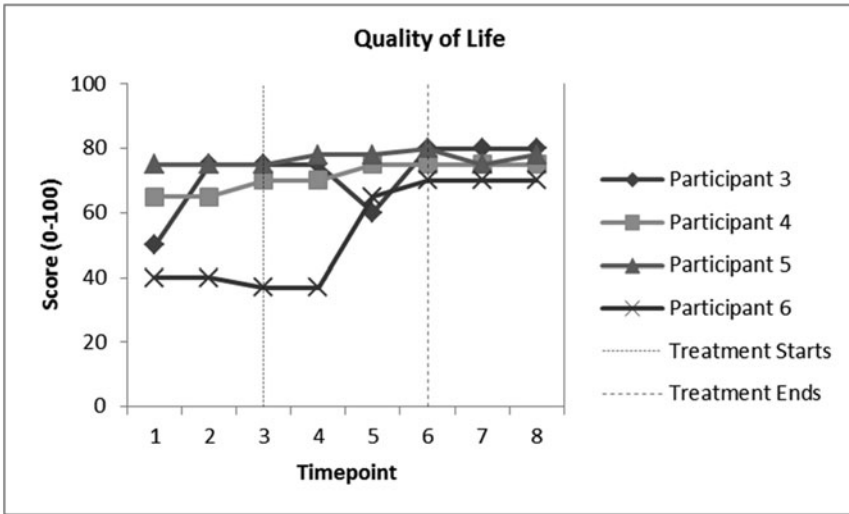


Fig. 1. Responders' VAS QoL scores across measured time points

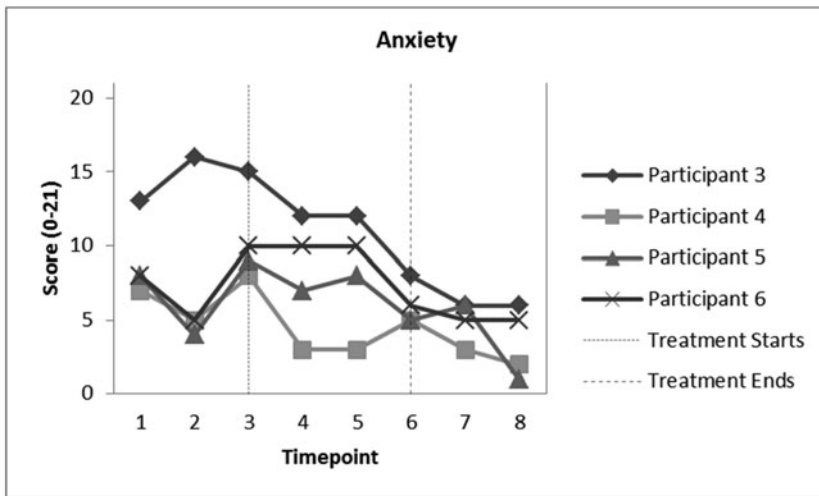


Fig. 2. Responders' anxiety (HADS) scores across measured time points

recovered to a stable level over two further pre-intervention measurement points (Fig. 1). He also reported difficulty coming to terms with his diagnosis, and presented with high anxiety and depression scores (Figs 2 and 3).

QoL increased pre- to post-intervention and was stable at all post-intervention time points (Fig. 1). There was a steep improvement in depression and anxiety scores across the period of the intervention (Figs 2 and 3). However, there was little evidence of a change in psychological

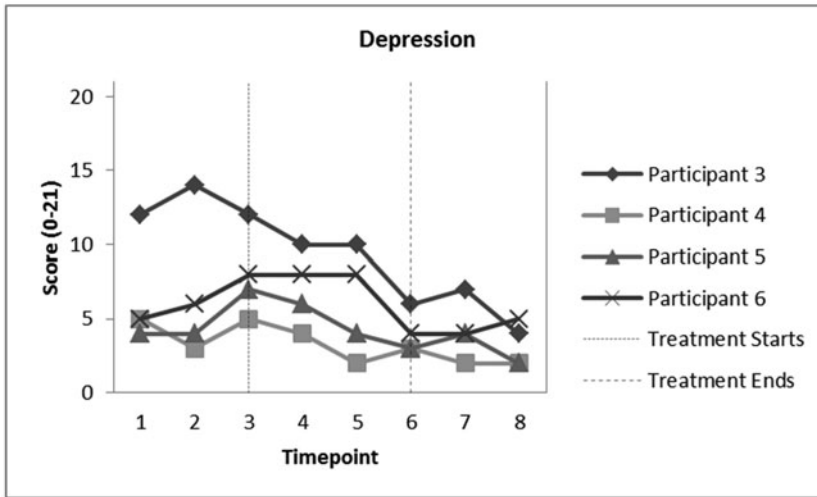


Fig. 3. Responders' depression (HADS) scores across measured time points

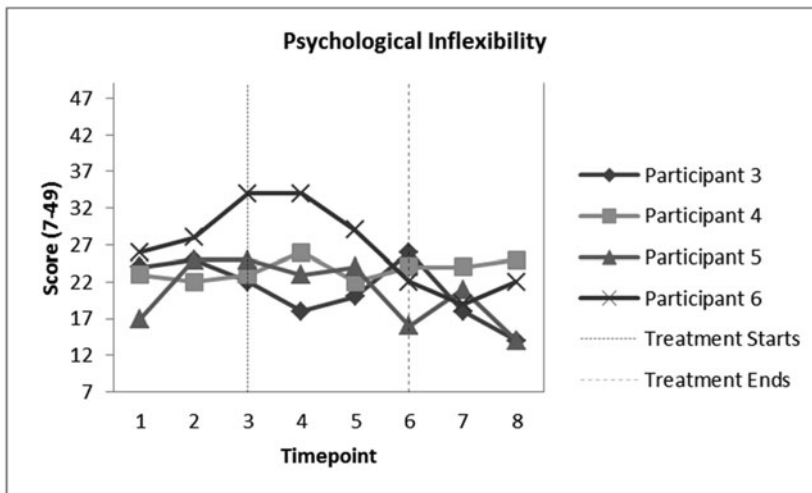


Fig. 4. Responders' psychological inflexibility (AAQ-II) scores across measured time points

flexibility (Fig. 4). In his post-intervention evaluation, participant 3 rated the intervention as 6/6 ('yes, it was extremely helpful'); his free text comment supported this selection:

'... The exercises can be used in everyday life and I find myself using the tools given now more and more. They do take practice but are effective. It also helped me realize that you can have a life with this illness.'

Participant 4 was a professional man in his thirties. He had been diagnosed with MD in his teenage years, but had been minimally affected by symptoms (HAQ-DI = 0.30). At pre-intervention time points he reported reduced QoL and his mood scores stayed below the HADS clinical cut-off (Zigmond and Snaith, 1983) (Figs 1–3). His initial psychological flexibility scores were at a good level (Fig. 4).

There was an improvement in QoL during the intervention. This improvement was maintained post-intervention (Fig. 1). Mood scores also improved across the intervention period (Figs 2 and 3). However, psychological flexibility remained constant across measurement points (Fig. 4). In his post-intervention evaluation, participant 4 rated the intervention as 5/6 ('yes, it was very helpful'), and he commented:

'It hasn't flicked a switch and made me super happy but it has made me confident that I can live a fuller life if I make the effort. By constructively focusing on my life I have been able to make small changes for the better...'

Participant 5 was retired and in his sixties. He had advanced MD which caused a high level of disability (HAQ-DI = 1.65). While he reported good QoL at all pre-intervention time points, there was also a slight improvement in QoL across the period of the intervention (Fig. 1). Similarly, while his initial mood scores appeared healthy, these also appeared to improve steadily across the intervention (Figs 2 and 3). The improvement in outcomes was mirrored by improved psychological flexibility (Fig. 4). In this post-intervention evaluation he indicated that the intervention was a 5/6 ('yes, it was very helpful'):

'The intervention came at a time of kitchen refurbishment and major dental treatment and I think that it may have helped me to be more "grounded". The intervention helped to keep me on the right track and encouraged a progression in a good direction...'

Participant 6 was in her forties. She had been living with MD for 17 years and reported quite a high level of disability (HAQ-DI = 2.1). She reported struggling to know her purpose in life as her life circumstances had recently changed. At baseline, her QoL was low and stable, and her mood scores appeared moderately affected (Figs 1–3). Pre-intervention, her baseline psychological flexibility was low and worsening (Fig. 4). She appeared to experience a great improvement in QoL during the intervention (Fig. 1), alongside a slight improvement in mood (Figs 2 and 3). These gains were maintained at follow-up. Improvements in QoL and mood mirrored improvements in psychological flexibility (Fig. 4). Participant 6 rated the intervention as 6/6 ('yes, the intervention was extremely helpful'), and commented:

'It helped me look more clearly at my values, and what is important in life, and how to take small steps to achieve them [values]...'

Non-responders

Participant 1 was in her fifties. She had experienced MD symptoms for 16 years, which, at the time of study, were causing significant disability (HAQ-DI = 1.45). She reported frustration at the restrictions MD imposed upon her life. As shown in Figs 5–7, she was experiencing very low QoL alongside significant low mood and anxiety at the beginning of the intervention. This was coupled with a good psychological flexibility score (Fig. 8). Overall, there appeared to be

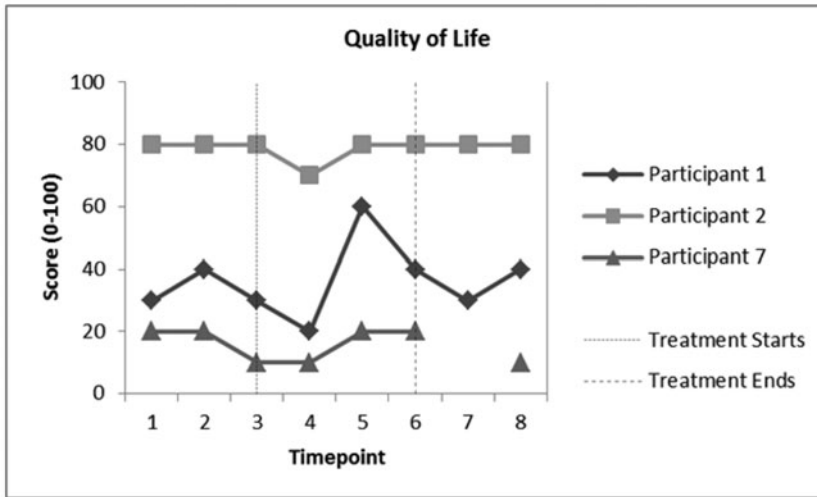


Fig. 5. Non-responders' VAS QoL scores across measured time points

no improvement in QoL, mood or psychological flexibility after starting the intervention. In reviewing the intervention, which she rated as 'it was a little bit helpful' (3/6), she commented:

'I think it implied that it's my thoughts that are holding me back, which is not necessarily the case... Tried using some of the tips and challenged myself by doing things I thought I couldn't do (tiny things though which really don't count in the whole scheme of things)... I know what matters to me but I can't do what matters however hard I think about it...'

Participant 2 had been diagnosed with MD at a young age and was now in his forties. He reported similar levels of physical disability to participant 1 (HAQ-DI = 1.45). He had a very active work and social life, and reported excellent initial QoL with little evidence of anxiety or depression. Similar to participant 1, his pre-intervention psychological flexibility scores were at a healthy level (see Fig. 8). There were no clear changes in outcome or process variables across the period of the intervention (Figs 5–8). In his post-intervention evaluation, where the intervention was rated as being '... a little bit helpful' (3/6), this participant commented:

'... It was helpful in confirming that I seem to be in a fairly positive place at the moment. I found some bits very useful although it was reassuring that some of the techniques designed to 'de-stress' were not really required by me.'

Participant 7 was in his fifties and had been diagnosed with a MD 11 years previously. He reported significant functional impairment (HAQ-DI = 2.1) and he rated his initial QoL as very low (Fig. 5). He reported poor mood scores that were worsening over the three pre-intervention time points (Figs 6 and 7). Similarly, his initial psychological flexibility was the lowest of any included participant (Fig. 8).

Across the period of the intervention there was a slight recovery in QoL score; however, QoL reduced to follow-up (Fig. 5). Depression and anxiety, which were worsening over the

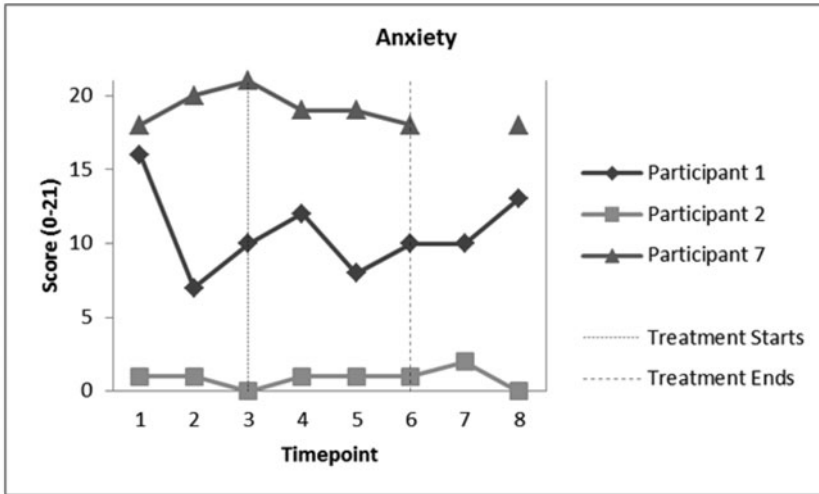


Fig. 6. Non-responders' anxiety (HADS) scores across measured time points

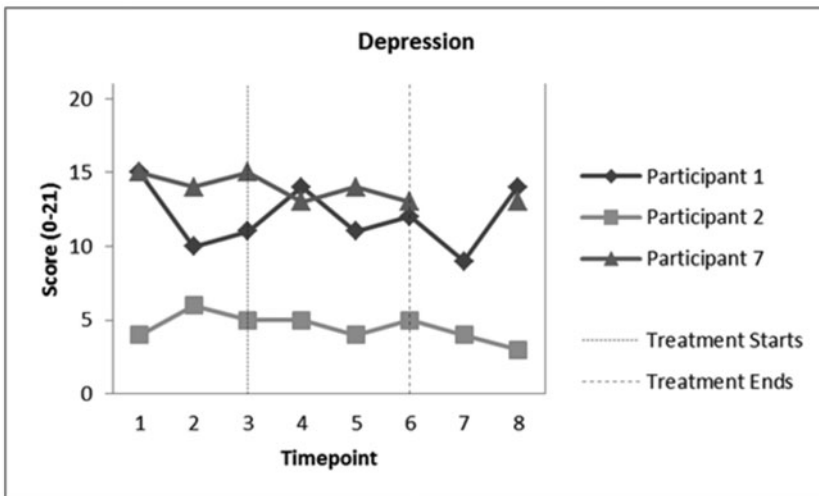


Fig. 7. Non-responders' depression (HADS) scores across measured time points

three pre-intervention time points, improved slightly over the period of the intervention and remained stable to follow-up. Psychological flexibility continued to improve throughout the intervention (Fig. 8). This participant reported that high levels of pain and fatigue impeded his engagement with the intervention. Similarly he rated the intervention as '...moderately helpful', and commented:

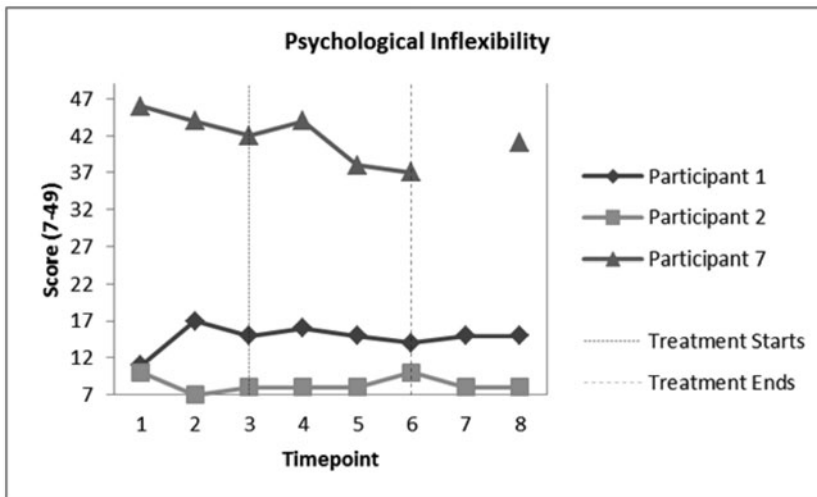


Fig. 8. Non-responders' psychological inflexibility (AAQ-II) scores across measured time points

'[the intervention was] Very good, but a lot of information to take in at one time. When your pain is high your levels and duration of concentration are greatly reduced... the lessons taught a new and different approach, which hopefully will improve things in the coming weeks and months...'

General acceptability

All participants completed all telephone calls, and appeared to have engaged with the modules. No adverse consequences were reported, and there were no clear deteriorations in QoL or mood during the period of the intervention. On the brief patient evaluation/acceptability measure, when asked 'did the intervention help you?', no participant perceived any damaging consequences from the intervention. Here scores ranged from 3 ('it was little bit helpful') to 6 ('yes, it was extremely helpful'), with an average score of 4.57. With the possible exception of participant 1's response, all supporting statements were positive.

Discussion

This evaluation of a brief guided self-help ACT intervention for improving QoL and mood in MD returned some encouraging results. While many of the improvements appeared small in magnitude, four out of seven participants appeared to experience an improvement across all three primary (QoL) and secondary (depression and anxiety) outcome measures, relative to their respective pre-intervention scores. For half of responders there was also an improvement in the presumed ACT treatment mechanism, psychological flexibility. Nonetheless, two responders' improvements in outcomes were not mirrored by improvements in our measure of psychological flexibility. This suggests that improvement in outcomes may have occurred via processes other than psychological flexibility. However, it is also possible that improvement occurred via an aspect of psychological flexibility not captured by the AAQ-II. Indeed, the

seven-item AAQ-II may not be of sufficient scope to comprehensively capture all aspects of psychological flexibility (Francis *et al.*, 2016). Thus more comprehensive measurement of psychological flexibility is indicated for future evaluations.

The intervention appeared generally acceptable to participants. There was no clear deterioration in outcome measures for any participant. Indeed, even for non-responders, the feedback, delivered online after exiting the study, was mostly positive. The possible exception was participant 1, whose feedback indicated that the intervention was perceived as ineffective, or perhaps even invalidating. Clearer conceptualization of the intervention's purpose (i.e. acting in line with values insofar as is possible given the limitations caused by MD symptoms) within the intervention materials might improve engagement.

Three participants did not appear to experience a benefit from the intervention. Non-response may have been due to different reasons for each participant. Participant 1 described difficulties with engagement. Participant 2 reported high QoL, and no indication of either depression or anxiety, arguably leaving very little scope for the intervention to have a beneficial effect. Participant 7 reported that high levels of pain, and fatigue levels diminished his ability to concentrate on the intervention materials. This suggests that those with extremely high levels of distress, pain or fatigue may struggle to engage with this intervention in its current format.

Limitations

First, while the visual analysis method that we used has several benefits (idiographic analysis, the inclusion of multiple baseline measurements), it also has drawbacks. For example, there can be disagreement between raters on whether change is present; therefore, inter-rater agreement is often low in visual analysis (Harrington and Velicer, 2015). Also, in this study, only three pre- and three post-intervention time points were recorded. This meant that only a limited understanding of fluctuation/stability in outcome variable scores at pre- and post-intervention periods was possible. Thus while results are encouraging, due to our pragmatic assessment methods our confidence in the findings must be limited. We did not assess reliable change (Jacobson and Truax, 1991), because, due to our inclusive entry criteria, this would have necessitated post-intervention scores at or above ceiling level for several participants.

In addition, for many reasons (e.g. placebo response, regression to the mean) these data cannot be taken as evidence of efficacy (Chambless and Hollon, 1998; Öst, 2008; Graham *et al.*, 2016b). We were also uncertain about therapist or intervention treatment fidelity as this was not assessed in any formal way, and given that we had just one therapist, there was a condition/therapist confound (Öst, 2008). Finally, limiting our understanding of the acceptability of the recruitment procedures, we did not formally record number of participants approached or excluded.

Conclusion

Although several caveats must be considered with these results, and psychological flexibility did not consistently improve, four of seven participants appeared to experience an improvement in the primary (QoL) and secondary (anxiety and depression) outcomes across the period of study. Regarding acceptability, all appeared to complete the intervention, no participants experienced a clear worsening in outcome measures, and feedback was generally

positive. Thus these results provide an encouraging initial evaluation of a brief guided self-help ACT intervention for people with MD. A future study will evaluate efficacy of the intervention in a randomized controlled trial.

Main points

- (1) Muscle disorders often cause significant functional impairment. Yet there is much variation in quality of life (QoL), and many do live well with these conditions.
- (2) Several studies suggest that psychological processes, such as illness perceptions and psychological flexibility, can help explain some of this variation in QoL. Therefore, psychological interventions might be helpful for some people with muscle disorders.
- (3) In the present study we piloted a brief guided self-help ACT intervention which aimed to improve QoL via manipulation of psychological flexibility.
- (4) Several participants (4 of 7) appeared to have varying degrees of response to the intervention, with improvements noted in QoL and mood across the period of study. Participants generally found the intervention acceptable.
- (5) While this was a promising initial evaluation of the intervention, several methodological weaknesses limit the conclusions that should be drawn from this study. Also, consistent change in psychological flexibility was not apparent.

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Ethical statement

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committees on human experimentation and with the Helsinki Declaration of 1975, as revised in 2008.

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Conflicts of interest

The authors report no conflicts of interest.

Recommended follow-up reading

Graham CD, Gouick J, Krahe C, Gillanders D (2016). A systematic review of the use of Acceptance and Commitment Therapy (ACT) in chronic disease and long-term conditions. *Clinical Psychology Review* **46**, 46–58. doi: <https://doi.org/10.1016/j.cpr.2016.04.009>

Voet N, Bleijenberg G, Hendriks J, de Groot I, Padberg G, van Engelen B, Geurts A (2014). Both aerobic exercise and cognitive-behavioral therapy reduce chronic fatigue in FSHD: an RCT. *Neurology* **83**, 1914–1922. doi: 10.1212/wnl.0000000000001008

References

- Bjelland I, Dahl AA, Haug TT, Neckelmann D** (2002). The validity of the Hospital Anxiety and Depression Scale: An updated literature review. *Journal of Psychosomatic Research* **52**, 69–77. doi: [https://doi.org/10.1016/S0022-3999\(01\)00296-3](https://doi.org/10.1016/S0022-3999(01)00296-3)
- Blumen SC, Bouchard JP, Brais B, Carasso RL, Paleacu D, Drory VE et al.** (2009). Cognitive impairment and reduced life span of oculopharyngeal muscular dystrophy homozygotes. *Neurology* **73**, 596–601. doi: 10.1212/WNL.0b013e3181b388a3
- Bond FW, Hayes SC, Baer RA, Carpenter KM, Guenole N, Orcutt HK et al.** (2011). Preliminary psychometric properties of the Acceptance and Action Questionnaire-II: a revised measure of psychological inflexibility and experiential avoidance. *Behavior Therapy* **42**, 676–688.
- Bruce B, Fries JF** (2003a). The Stanford Health Assessment Questionnaire: a review of its history, issues, progress, and documentation. *Journal of Rheumatology* **30**, 167–178.
- Bruce B, Fries JF** (2003b). The Stanford Health Assessment Questionnaire: dimensions and practical applications. *Health and Quality of Life Outcomes* **1**, 1.
- Burns TM, Graham CD, Rose MR, Simmons Z** (2012). Quality of life and measures of quality of life in patients with neuromuscular disorders. *Muscle and Nerve* **46**, 9–25. doi: 10.1002/mus.23245
- Chambless DL, Hollon SD** (1998). Defining empirically supported therapies. *Journal of Consulting and Clinical Psychology* **66**, 7–18.
- de Boer AG, van Lanschot JJ, Stalmeier PF, van Sandick JW, Hulscher JB, de Haes JC, Sprangers MA** (2004). Is a single-item visual analogue scale as valid, reliable and responsive as multi-item scales in measuring quality of life? *Quality of Life Research* **13**, 311–320.
- Fledderus M, Oude Voshaar MA, ten Klooster PM, Bohlmeijer ET** (2012). Further evaluation of the psychometric properties of the Acceptance and Action Questionnaire-II. *Psychological Assessment* **24**, 925.
- Francis AW, Dawson DL, Golijani-Moghaddam N** (2016). The development and validation of the Comprehensive assessment of Acceptance and Commitment Therapy processes (CompACT). *Journal of Contextual Behavioral Science* **5**, 134–145. doi: <https://doi.org/10.1016/j.jcbs.2016.05.003>
- Graham CD** (2012). Explaining and changing adverse illness perceptions in people with muscle disease. King's College London.
- Graham CD, Gouick J, Ferreira N, Gillanders D** (2016a). The influence of psychological flexibility on life satisfaction and mood in muscle disorders. *Rehabilitation Psychology* **61**, 210–217. doi: 10.1037/rep0000092
- Graham CD, Gouick J, Krahe C, Gillanders D** (2016b). A systematic review of the use of Acceptance and Commitment Therapy (ACT) in chronic disease and long-term conditions. *Clinical Psychology Review* **46**, 46–58. doi: <https://doi.org/10.1016/j.cpr.2016.04.009>
- Graham CD, Rose MR, Grunfeld EA, Kyle SD, Weinman J** (2011). A systematic review of quality of life in adults with muscle disease. *Journal of Neurology* **258**, 1581–1592. doi: 10.1007/s00415-011-6062-5
- Graham CD, Rose MR, Hankins M, Chalder T, Weinman J** (2013). Separating emotions from consequences in muscle disease: Comparing beneficial and unhelpful illness schemata to inform intervention development. *Journal of Psychosomatic Research* **74**, 320–326. doi: <https://doi.org/10.1016/j.jpsychores.2012.09.012>
- Graham CD, Simmons Z, Stuart SR, Rose MR** (2015). The potential of psychological interventions to improve quality of life and mood in muscle disorders. *Muscle and Nerve* **52**, 131–136.

- Graham CD, Weinman J, Sadjadi R, Chalder T, Petty R, Hanna MG et al.** (2014). A multicentre postal survey investigating the contribution of illness perceptions, coping and optimism to quality of life and mood in adults with muscle disease. *Clinical Rehabilitation* **28**, 508–519. doi: 10.1177/0269215513511340
- Harrington M, Velicer WF** (2015). Comparing visual and statistical analysis in single-case studies using published studies. *Multivariate Behavioral Research* **50**, 162–183. doi: 10.1080/00273171.2014.973989
- Harris R** (2009). *ACT Made Simple: An Easy-to-Read Primer on Acceptance and Commitment Therapy*. New Harbinger Publications.
- Hayes SC** (2005). *Get Out of Your Mind and Into Your Life: The New Acceptance and Commitment Therapy*. New Harbinger Publications.
- Hayes SC, Luoma JB, Bond FW, Masuda A, Lillis J** (2006). Acceptance and commitment therapy: Model, processes and outcomes. *Behaviour Research and Therapy* **44**, 1–25.
- Hayes SC, Strosahl KD, Wilson KG** (1999). *Acceptance and Commitment Therapy: An Experiential Approach to Behavior Change*. Guilford Press.
- Hofmann SG, Asnaani A, Vonk IJJ, Sawyer AT, Fang A** (2012). The efficacy of cognitive behavioral therapy: a review of meta-analyses. *Cognitive Therapy and Research* **36**, 427–440. doi: 10.1007/s10608-012-9476-1
- Jacobson NS, Truax P** (1991). Clinical significance: a statistical approach to defining meaningful change in psychotherapy research. *Journal of Consulting and Clinical Psychology* **59**, 12.
- Kalkman JS, Schillings ML, Zwarts MJ, Van Engelen BGM, Bleijenberg G** (2007). Psychiatric disorders appear equally in patients with myotonic dystrophy, facioscapulohumeral dystrophy, and hereditary motor and sensory neuropathy type I. *Acta Neurologica Scandinavica* **115**, 265–270. doi: 10.1111/j.1600-0404.2006.00737.x
- Kazdin AE** (1978). Methodological and interpretive problems of single-case experimental designs. *Journal of Consulting and Clinical Psychology* **46**, 629–642. doi: 10.1037/0022-006X.46.4.629
- Merrison AFA, Hanna MG** (2009). The bare essentials: muscle disease. *Practical Neurology* **9**, 54–65. doi: 10.1136/jnnp.2008.167171
- Minnerop M, Weber B, Schoene-Bake J-C, Roeske S, Mirbach S, Anspach C et al.** (2011). The brain in myotonic dystrophy 1 and 2: evidence for a predominant white matter disease. *Brain* **134**, 3530–3546. doi: 10.1093/brain/awr299
- Mykletun A, Stordal E, Dahl AA** (2001). Hospital Anxiety and Depression (HAD) scale: factor structure, item analyses and internal consistency in a large population. *British Journal of Psychiatry* **179**, 540–544.
- Natterlund B, Sjoden PO, Ahlstrom G** (2001). The illness experience of adult persons with muscular dystrophy. *Disability Rehabilitation* **23**, 788–798.
- Öst LG** (2008). Efficacy of the third wave of behavioral therapies: a systematic review and meta-analysis. *Behaviour Research and Therapy* **46**, 296–321. doi: 10.1016/j.brat.2007.12.005
- Rose MR, Sadjadi R, Weinman J, Akhtar T, Pandya S, Kissel JT et al.** (2012). Role of disease severity, illness perceptions, and mood on quality of life in muscle disease. *Muscle and Nerve* **46**, 351–359. doi: 10.1002/mus.23320
- Sadjadi R, Rose MR, Muscle Study Group** (2010). What determines quality of life in inclusion body myositis? *Journal of Neurology, Neurosurgery and Psychiatry* **81**, 1164–1166. doi: 10.1136/jnnp.2009.183863
- Voet N, Bleijenberg G, Hendriks J, de Groot I, Padberg G, van Engelen B, Geurts A** (2014). Both aerobic exercise and cognitive-behavioral therapy reduce chronic fatigue in FSHD: an RCT. *Neurology* **83**, 1914–1922. doi: 10.1212/wnl.0000000000001008
- Wolgast M** (2014). What does the Acceptance and Action Questionnaire (AAQ-II) really measure? *Behavior Therapy* **45**, 831–839. doi: 10.1016/j.beth.2014.07.002

Zigmond AS, Snaith RP (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica* **67**, 361–370.

Learning objectives

- To learn about psychological research in muscle disorders.
- To learn about the participant experience of a brief guided self-help ACT intervention.
- To learn about the implementation of remote guided self-help interventions in chronic disease contexts.