

Guided graded exercise self-help for chronic fatigue syndrome: Patient experiences and perceptions

Abstract:

Purpose: This study explored patient experiences of Guided graded Exercise Self-help delivered as part of a randomised controlled trial for people with chronic fatigue syndrome/myalgic encephalomyelitis, which found that Guided graded Exercise Self-help was better than specialist medical care at reducing fatigue and improving physical functioning.

Methods: Semi-structured interviews were conducted with patients reporting improvement (n=9) and deteriorated (n = 10), and analysed using a thematic “constant comparison”.

Results: The improved group described more facilitators to doing Guided graded Exercise Self-help, and were more likely to describe high levels of self-motivation, whilst the deteriorated group described more barriers to Guided graded Exercise Self-help (including worse exacerbation of symptoms after Guided graded Exercise Self-help, greater interference from comorbid conditions and obstacles to Guided graded Exercise Self-help in their lives), and had been ill for longer. Having the capacity to do Guided graded Exercise Self-help was important; of note, those with relatively lower levels of functioning sometimes had more time and space in their lives to support their Guided graded Exercise Self-help engagement. We identified an important ‘indeterminate phase’ early on, in which participants did not initially improve.

Conclusions: Guided graded Exercise Self-help may be improved by targeting those most likely to improve, and education about the indeterminate phase.

Keywords: chronic fatigue syndrome; myalgic encephalomyelitis; graded exercise therapy; randomised controlled trial; qualitative.

Introduction

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) is a long-term health condition, the causes of which – and specific disease processes – are poorly understood [National Institute of Health and Care Excellence (NICE), 1]. There is no consensus as to whether CFS and ME are the same or different conditions [2], but since this study used the NICE criteria for CFS/ME we employ this hybrid term. CFS/ME causes a range of symptoms, primarily fatigue, particularly after minimal exertion. Other common symptoms include difficulty sleeping, headaches, cognitive dysfunction and painful lymph nodes [1]. People with CFS/ME report coping with reductions in function, which can lead to a loss in their personal “identity” as their role in life changes due to illness (e.g. no longer able to work), often resulting in loss of confidence and self-esteem. Additionally, they may experience scepticism about - and lack of knowledge of – CFS/ME among members of the medical community [3]. Recovery from CFS/ME is uncommon without treatment [4]. It is estimated CFS/ME affects one quarter of a million people in the UK (around 0.4% of the population), and costs the UK economy £102 million a year in lost earnings alone [5, 6].

Treatments used by health professionals, as recommended by the National Institute of Health Care Excellence (NICE), include graded exercise therapy (GET) and cognitive behaviour therapy (CBT) [1]. Evidence from randomised controlled trials (RCTs) support the use of GET and CBT in people with mild to moderate CFS/ME [7]. A recent systematic review of eight RCTs concluded there is moderate-quality evidence that GET is helpful for the management of CFS/ME, suggesting that GET provides moderate improvements in fatigue and functioning [8]. Reports of serious adverse reactions in trial were rare, although many did not report such data [8]. There is, however, controversy around the use of GET, with some ME patient charities raising concerns about the emphasis placed on GET by the NICE

guidelines [9], and about its safety. In lay surveys of ME charity members many respondents considered GET unacceptable or harmful [6, 9]. For example in the 2014 Action for ME survey, 47% of respondents who reported using GET said it made them ‘a bit or much worse’, 18% reported ‘no change’ and 35% said it was ‘a little or very helpful’. Other management approaches, including pacing, are instead advocated by some leading charities as more effective at managing CFS/ME [6]. Nevertheless, charities have highlighted the need for further investigation as to why some people find GET helpful and others do not, and how this can be addressed [6, 9]. Findings from an online qualitative survey of a self-selected sample of 76 ME charity members regarding their experiences of rehabilitation therapies showed a range of positive and negative experiences [10]. The therapies included GET, graded activity therapy and exercise on prescription, and were provided by professionals in a range of settings. The authors reported that supportive communication with the therapist was key to participants’ positive experiences, and that negative experiences included a conflict in beliefs between therapist and patient about the nature of their condition and the role of rehabilitation, with some participants reporting pressure to comply with treatment.

NICE describe GET as an “approach to CFS/ME that involves physical assessment, mutually negotiated goal-setting and education” [1]. When commencing GET, first a sustainable baseline of physical activity is agreed between patient and health professional/therapist. Once achieved, the time spent being physically active is then gradually increased over time, followed by a gradual increase in intensity. Physical activity is tailored to the individual to fit with their current functioning, lifestyle and preferences. NICE recommend that GET is delivered by a therapist trained in GET with experience in CFS/ME, under appropriate clinical supervision.

Guided graded Exercise Self-help (GES) is based on graded exercise therapy (GET) principles but it is not a therapy, it is guidance intended for self-management. GES comprised a booklet describing a six-step graded exercise self-management programme, which patients were encouraged follow independently over approximately 12 weeks (<http://www.wolfson.qmul.ac.uk/images/pdfs/getset/GET%20guide%20booklet%20version%201%2022062010.pdf>). The booklet was based on the GET approach developed for the PACE trial [11] (a RCT comparing GET, CBT, pacing and specialist medical care, SMC). It was based on the GET booklet written by the PACE trial GET therapists, based on NICE recommendations [1], and piloted with patients. Alongside the booklet, participants were offered up to four support sessions with a physiotherapist (maximum of 90 minutes for all four sessions) over 8 weeks. The first session was delivered face-to-face, by telephone or Skype and the remaining three sessions were delivered by telephone or Skype. Two physiotherapists, experienced in delivering GET as a treatment for CFS/ME, were trained to support and guide the participants through the GES booklet, but were not allowed to provide therapy as an active intervention. This is in contrast to a GET programme which is therapist-delivered and comprises a larger number of therapy sessions delivered, usually face-to-face in a clinic, over a longer time period [8].

Reports suggest that exercise programmes may not always be delivered according to NICE guideline recommendations [6, 10]. GET delivered under the controlled conditions of a RCT can better ensure therapy is delivered as intended. The use of manual-defined treatments provided by suitably-qualified, trained, competent and closely supervised therapists, has been shown to result in high rates of participant satisfaction and good therapeutic alliances [11]. While differences in delivery may account for some of the discrepancies between RCT findings and patient experiences, even optimal GET delivery produces a range of outcomes

i.e. some patients improve while others do not [8]. This suggests that more needs to be done to understand participants' experiences of GET, to maximise its acceptability and effect in the future.

Factors which influence patients' experiences of, and outcomes following, GET are likely to be complex and are, as yet, poorly understood. Previous work has shown a range of challenges, coping behaviours, illness experiences and illness management practices employed by people with CFS/ME [3] that may influence their outcomes [12, 13]. Work from previous studies on patient change attributions in self-management interventions, suggests that specific behaviour changes, stress, life events, self-efficacy and locus of control may be important factors in the amount of change reported after these interventions [14, 15]. The range of conflicting views about GET as a treatment/intervention can also be unhelpful for patients attempting to understand, manage and recover their health [16]. Thus, more needs to be done to understand the circumstances under which GET works and does not work [10]. The research question for this study was: "What are the differences and similarities in treatment perceptions and experiences of GES among CFS/ME participants reporting an improvement compared to those reporting a deterioration in their condition?"

Methods

This qualitative, one-to-one interview study was situated within a pragmatic RCT of GES; which found that GES was moderately better than SMC at reducing fatigue and improving functioning [17]. This design was chosen because narratives from interviews provide an overarching 'code' for the way people construct – and then communicate - meanings about themselves [18]. This compels us to face the existential, moral and contextual issues entailed in experiences of treatments [19, 20]. As Frank points out, 'the truth of stories is not only

what *was* experienced, but equally what *becomes* experience in the telling and its reception' [21].

The Graded Exercise Therapy guided Self-hElp Trial (GETSET)

Participants were recruited to the GETSET trial (n=211) at the time of being placed on the waiting list for treatment at one of two NHS specialist CFS/ME secondary care clinics in the south of England [22]. All participants had at least one SMC consultation, delivered by doctors with specialist experience in CFS/ME. SMC could involve prescriptions or advice regarding medication, as indicated for symptoms or comorbid conditions such as insomnia, pain, or depressive illness. Although not routinely scheduled during the trial, further SMC sessions were available after randomisation for patients who required it, but it was not a standardised intervention. Study inclusion criteria included a diagnosis of NICE-defined CFS/ME [1], in order to ensure generalisability to secondary care in the UK. Patients were excluded if they: could not speak and/or read English adequately; had current suicidal thoughts; had read the GES guide previously; had had previous GET at one of the trial clinics; had physical contraindications to exercise; or were under 18 years old [22].

Participants received either SMC (n=104) or SMC plus GES (n=107). At 12 weeks post-randomisation those receiving GES had reduced fatigue (measured by the Chalder fatigue questionnaire [23]) and improved physical function (measured by the short form-36 physical function subscale score [24]), compared to those receiving SMC alone. All but one secondary outcome showed similar differences. No serious adverse reactions were recorded and other safety measures did not differ between groups, once missing data was included GES [17].

Sample and Recruitment

The aim was to recruit a sample of 20 patients who had participated in the GES arm of the GETSET trial [22] and had completed their follow-up questionnaire 12 weeks after randomisation. We used the Clinical Global Impression change scale [25] to identify suitable patients. The scale asked ‘overall how much do you think your CFS/ME has changed since the start of the study?’ and provides seven possible responses: ‘very much better’, ‘much better’, ‘a little better’, ‘no change’, ‘a little worse’, ‘much worse’ and ‘very much worse’. Those who reported their CFS/ME as being improved (“much better” or “very much better”) or worse (“much worse” or “very much worse”) at 12-weeks on the Clinical Global Impression change scale were eligible. However, because no single participant rated themselves as “much worse” or “very much worse”, we expanded the inclusion criteria to include participants who rated themselves as a “little worse”.

Participants meeting the inclusion criteria were posted an invitation pack including an invitation letter, patient information sheet, consent form and prepaid envelope to return the consent form. The first author followed them up with a telephone call, and discussed their potential participation; they subsequently agreed or declined to be interviewed. The intention was to recruit 10 participants to each group for comparison. Thirty-two participants were identified (14 “much better” (none rated themselves as “very much better”); 18 “a little worse”), and were invited to be interviewed according to how recently they had finished GES. Our quotas were filled after we had invited 27 patients. Of these, eight declined [too unwell to do the interview (n=2), felt they had not used GES enough to be interviewed about it (n=1) and no reason given (n=5)]. Therefore nine participants who reported feeling “much better” and 10 who reported feeling “a little worse” provided consent and were interviewed.

Interviews

Interviews were arranged at a time and place most convenient for the participant. Eleven opted to be interviewed by telephone, six at home, one at their place of work and one at the University. Interviews were conducted three to 26 months after participants completed GES, median 22 months (median 14 = “a little worse”, 22 = “much better”). Interviews used a semi-structured approach [26]. The interview topics were developed in consultation with the wider research group and patient representatives, and included before and after trial wellbeing, expectations of GES, the meaning of exercise, experiences of GES, barriers and facilitators to GES, and any outside influences on trial or GES participation. Interviews lasted between 13 and 80 minutes (mean = 45 minutes). All interviews were audio-recorded, with permission from the participant, transcribed verbatim and returned to the participant for checking after anonymisation. The point of data saturation [27] – where no new themes were emerging – was reached for the “much better” group, and approached saturation for the “a little worse” group.

Data analysis

Data were analysed using thematic analysis [28]. Researchers independent of the implementation of the GETSET trial conducted the analysis. For the initial seven transcripts one researcher immersed herself in the data by repeatedly reading and annotating transcripts. For each transcript, key words and phrases were highlighted and underlined, and themes and ideas were written in the margins. Key themes were then listed in a separate document and examined: similar themes were grouped together, duplicate themes deleted and higher order concepts were debated among the first two authors to produce a final list of themes for each transcript. Themes from each of the seven transcripts were then compared and amalgamated into one draft list of themes. The remaining 12 transcripts were searched for new themes to produce a final list. The data were then coded and explored in the qualitative data analysis

software environment, NVivo [29]. NVivo was used to generate reports for the “much better” and “a little worse” groups for each theme, enabling systematic comparison of the groups for each relevant topic. The initial analysis was iteratively debated at length with the second author, and then sent to other authors to critique our findings. The qualitative analysis and drafts of the manuscript were then written by the first two authors, which were then also critiqued and contributed to by other authors, independent researchers, and the patient representatives.

The first author’s (and interviewer) position: The researcher is an experienced qualitative researcher in the field of health psychology. She predominantly conducts research into complex health interventions for people with long-term health conditions using mixed methods, and qualitatively explores the patient perspective of living with long-term health conditions. She has had a chronic disabling illness for 25 years.

Results

Participants were predominantly female (n=17) and of Caucasian ethnicity (n=17). Mean age was 43 years (IQR 28-66) for the “a little worse” group and 39 years (IQR 21 to 54) for the “much better” group. The median length of time since onset of CFS/ME symptoms was 13 years (IQR 8 to 21) for the “a little worse” group and 4 years (IQR 3 to 5) for the “much better” group.

This results section makes references throughout regarding similarities and differences between the “much better” and “a little worse” groups. It is worth noting, however, that two participants who had rated their condition as “a little worse” on the 12 week follow-up questionnaire for the trial, subsequently reported at interview that, on later reflection, they

had experienced a modest improvement from GES. These participants were still counted in the “a little worse” group for the purposes of this study, as this was the actual trial outcome at the time. Table 1 shows key data themes and subtheme; results are presented around these themes.

Table 1- key themes and subthemes

Theme	Sub-theme
<i>Getting started and false starts</i>	
<i>The “indeterminate phase” of GES</i>	
<i>Competing commitments</i>	
<i>Interfering symptoms and comorbid conditions</i>	
<i>Maintaining motivation</i>	<i>The importance of guidance</i> <i>Participant beliefs and understanding of GES</i> <i>Support from other people and therapies</i>

Getting started and false starts

Participants moved through the self-management programme by attempting to stabilise their routine, choosing their specific physical activity, and setting their “baseline” level of activity.

Most participants reported finding these tasks relatively straightforward:

Just looking at what my general activity is on a day to day basis and keeping it quite similar which to be honest it was anyway. ... I feel like that was almost done and we agreed what it was from the first session. P2, “a little worse”

As participants commenced their GES activity (which involved completing an agreed additional physical activity), two participants in the “a little worse” (and none in the “much better”) group described “false starts”. That is, they reported not feeling physically/emotionally well enough to engage with GES. One participant described a physical reaction, which she believed was due to a pre-existing hip condition, and was given medical advice to discontinue GES. Another reported a number of major life events occurring after she had consented to the trial, which left her feeling too preoccupied to engage with GES. All other participants reported being able to start their programmes.

I did do it sporadically but at the time it wasn't something that I could commit to fully ... I think for things like that, I have to be really dedicated to the cause and be in the right frame of mind to be able to do it with that dedication that is required. And I didn't have that at that point, there was too much else on my mind. P27, “a little worse”

What strongly emerged from the data was that regardless of whether participants reported feeling that GES improved or worsened their CFS/ME, following the GES programme was considered “hard work” by the majority of participants. The following sections discuss participants’ specific experiences associated with following the GES programme.

The “indeterminate phase” of GES

Some participants focused their discussions on their body’s reaction to GES. Two participants (one = “a little worse”; one = “much better”) who were relatively highly functioning (e.g. were able to work) reported that they felt better immediately after exercise. Such immediate positive feedback encouraged them to continue with the programme:

10, 15 minutes [walk], I just always remember feeling that much more energised, like when I got to work, as opposed to, feeling like really sluggish. P17, “a little worse”

However, during the first phase of the GES programme the majority of participants in both groups noticed no immediate difference in their symptoms, or else an exacerbation. For some, the exacerbation of symptoms interfered with their everyday lives (discussed further below). For those who did begin to feel better, improvement was often reported as remarkably incremental. For example, a ten minute walk could take weeks or months to achieve for a participant who started with one minute of walking each day. Even for participants who were already reasonably active, their goals (e.g. being able to play a game of badminton) could also seem extraordinarily far away:

I used to do a lot of walking, I was in the scouts I used to do that sort of walking, climbing and doing things like that. ... I don't think I was getting any nearer [to] sort of goals of going on walks and things like that. P29, “a little worse”

Understandably, when participants experienced a setback to their incremental progress, it could be experienced as particularly demoralising.

*What it really did was make them [legs] more achy, that's what it did. I mean then, then, you see that answer to that is, the therapist I spoke to said well then you just cut back and do much less. But that, that's going nowhere. **Right okay so you would be increasing it [GES walking activity] even more slowly?** Yeah. P24, “a little worse”*

In practice, this meant that many GES participants had delayed gains and little or no short-term benefit, which resulted in them not knowing if GES was actually helping or hindering their condition. Thus, an “indeterminate phase” was established which, unsurprisingly, participants in both groups reported that maintaining motivation through this phase was

challenging, particularly when experiencing an exacerbation of symptoms or when finding their programme hard work and/or boring.

Cos it is hard work, it really is, and even now, it's still hard work to go out and go for a walk round the block, just, you just want to sit down and watch the telly, like normal people. ... It can take years, and, that can be quite discouraging. P11, "much better"

There was a point at which I was oh I'm just sick of this routine and I just want to do whatever I feel like doing. So actually the base-lining wasn't so frustrating as about a month in where I just felt like I need some bloody variation. P14, "much better"

Nevertheless, participants who avoided false starts were generally able to stick to their GES programmes at least to some extent through the indeterminate phase and beyond.

Competing commitments

It was important for participants to have time and space in their lives to follow the GES programme. The flexibility and patient-centeredness of the GES programme supported participants to develop programmes that (at least in theory) fitted into their lifestyle. For example, one mother just parked her car a little further away from her child's school to increase her walking. Perhaps more significantly, however, participants described needing enough "capacity" in their lives to experience an exacerbation of symptoms and for this not to interfere with essential life activities. Thus, GES seemed to work best for participants who had fewer commitments that interfered with GES (e.g. life responsibilities such as work, looking after children, housework, food shopping; lifestyle changes participants were making; or other activities which supported them emotionally). If a supportive partner or workplace could relieve the participant of other commitments, then they seemed to be better placed to benefit from GES.

I haven't got children so, married but I haven't got children, and so I think that makes a big difference too because I haven't got to be running around after them. I think it would've been a lot, lot harder to, balance my days and be systematic about the increases, if, if I'd had to, look after children or if I'd had a particularly demanding husband, who was expecting dinner on the table. P11, "much better"

I was very fortunate because my company were very understanding. They didn't put any sort of pressure on me to return to work. P8, "much better"

Severely affected patients were not included in this trial; participants had to be able to attend an outpatient clinic for diagnosis and assessment. For some (but not all) participants who were more physically disabled, but well enough to be included in the trial, having lower levels of functioning could create time and space to do GES. Firstly, fitting in GES was easier for these participants who only needed to find a small amount of time each day, e.g. a few minutes to go for a short walk. Higher functioning participants had more to do in their lives, and reported more challenges fitting GES into busier lifestyles. This sometimes resulted in them either not finding time to fit in the activity, or doing additional activity than recommended by their particular programme because they had things they needed to do.

I mean I do struggle to keep to exercise routines just because my life is quite busy and I struggle to add things in. P2, "a little worse"

Secondly, because lower functioning participants could only do limited household chores, childcare, or work, they were sometimes already in a situation where they had few other commitments. Thus they were able to focus on GES more fully. One participant who started GES when she was quite ill, with few other commitments, illustrated this complex situation: she improved over the 3-month trial period, and was then able to start doing more household chores/childcare. However, this meant that although she was able to substitute GES for activities of daily living, she reported a limit on the improvements she had already made.

I would put everybody in front of me and I think that Graded Exercise Therapy [GES] made me focus on me. ... So because I slowly got to a stage where I felt more comfortable about being out and physically able to do stuff. Then I suppose I just carried on doing stuff. ... Because of everybody's pull on my time, I know I can't ever recover to the extent that I want because I just don't get time to myself to do that, not at the moment. P15, "much better"

Interfering symptoms and comorbid conditions

Additional challenges arose for both "much better" and "a little worse" participants if they experienced an exacerbation of their symptoms (particularly fatigue) whilst following the GES programme. At this point, the extent of the exacerbation and level of their other commitments could influence whether a participant felt able to continue doing GES activities. These exacerbations were reported as more debilitating for participants in the "a little worse" group, who had had CFS/ME for longer. Half of participants in this group reported discontinuing their GES activities during, or at the end of, their GES programme for this reason (although they may not have informed the therapist they were discontinuing).

I started the stretching at home and then I introduced just a couple of minutes further walking. I'd just found, it was just enough to tip me over the edge really. ... Towards the end I sort of said I can't do this and you know it was impacting on me and obviously my family in turn quite massively. So, I think I gave it a fair go but I was happy to stop. P5, "a little worse"

Interview participants in the "a little worse" group also reported more comorbid conditions including joint hypermobility, fibromyalgia, irritable bowel syndrome, endometriosis, depression, arthritis, sciatica, and asthma ("a little worse" = six participants with one to four

comorbid conditions, median 2; “much better” = four participants with one to two conditions, median 0) and greater interference from these conditions when doing GES.

*The first sort of, few weeks I didn't do much at all because it was too painful just to, put your foot down really. **So was the pain from the arthritis or from the ME? Or a bit of both?** Both the arthritis and then I suppose, because, I think, I find that if you add anything else to the ME it makes things almost impossible. P29, “a little worse”*

One participant reported that her CFS/ME included memory problems, which impacted on her ability to undertake GES.

I have a lot of problems with my memory. So, even remembering to do these things, it is the hardest part really. P13, “a little worse”

Maintaining motivation

Maintaining motivation to continue GES through the challenges described above was discussed by participants in both groups. A number of participants, particularly in the “much better” group, gave accounts revealing high levels of motivation to continue with GES “For the first 1 or 2 years it was difficult.” P21, “much better”. A number of factors were mentioned by participants as influencing both their motivation and their ability to undertake GES, these are described in turn below.

The importance of guidance

Many participants praised their physiotherapist’s attitude and approach as enthusiastic, gentle and understanding. The physiotherapists’ patient-centred (rather than prescriptive) approach generally facilitated participants’ engagement with them and the GES programme.

The physiotherapist, she was just amazing, she was so encouraging and understanding. I mean I just, yeah she was phenomenal. There was a part of me that

was, I know this is working but you really need to clone this particular woman to make it work. ...The thing also, once it got started, because it was like every fortnight and someone was taking interest in what I'd been recording because I felt so isolated, it was almost like I wanted to do a good job for my teacher! P14, "much better"

Features of the GES programme

When some participants were recruited to the study at their secondary care appointment they had been seen by a psychiatrist [22] (some CFS/ME specialists are psychiatrists). Two participants raised objections to this as they felt this suggested their condition was mental rather than physical. For one, her anger was enough for her to question her commitment to GES.

And although I realised that a lot of physical illness can also, how your thoughts [are] et cetera, can also affect you physically, I just felt by just being totally under the psychiatric department, was the wrong move. ... I felt a bit angry. I felt almost as if I'd been betrayed by being put under that department, so it wasn't a very good start ... If I'm being honest I didn't want it to work because I didn't like being under the psychiatric department, because I felt it's not a psychiatric illness. P12, "a little worse"

Whilst the GES booklet was helpful for some participants two described it as patronising, having the feel of marketing material or seemingly designed for participants with a higher level of functioning than their own. They noted in particular that the statement suggesting that there should be no ill effects from GES was not accurate in their experience.

I really felt like it was written for an entirely different group of people, because all the 'example' people, were people who in my opinion are miles ahead of me [in ability]. ... it said research shows that, there will be no ill effects, and I already knew at that

point that yes there can be ill effects and, yes there was, was ill effects for me. p23, “a little worse”

Participants reported that being allowed to choose their own activities (e.g. walking, gardening) helped “keep it interesting” and supported motivation. Some felt that the remit of GES was too narrow and that it needed a broader approach, e.g. one that included CBT, or took into account mental activity.

Making it enjoyable really. Keeping it interesting, because if it’s hard, I mean it’s hard enough as it is, but without, I mean if I was doing housework put music on, put nice loud upbeat music so it keeps you motivated. If it’s sunny go outside, anything that gets you moving. P10, “much better”

The graded exercise is good, but if you have mental activity, that’s just as straining as exercise isn’t it? P29, “a little worse”

Participant beliefs and understanding of GES

A key factor for maintaining motivation to do GES was participants’ belief that it could actually help their CFS/ME, to some extent. For many participants this was the first time they had been offered an NHS treatment for their CFS/ME and they had few other treatment options, and this increased their motivation to try GES:

I was thinking that, this is me, this is awful, this is something that could be with me forever. This was quite frightening if I’m honest. ... I think having that approach [GES] was really important, cos for me I couldn’t find anything anywhere that was particularly encouraging in the sense that nobody was saying look this could work for you over time. P19, “much better”

An understanding of the theory behind GES helped participants understand and therefore engage in GES. For many participants this understanding was established when GES was explained to them at the beginning of the trial, or from a previous experience of using GET. Participants who had previously unsuccessfully tried GET, or attempted to increase their levels of activity without support, found it useful to have an explanation for the possible failure of previous attempts (e.g. baseline set too high, tried to increase level of activity too quickly), this explanation could motivate them to stick to their GES programme and do it “correctly”.

Every time I tried to do it myself, just from what other people had said to me I just always overdid it and ended up having to stop so. ... I thought right the only way I'm going to do this is by sticking exactly to this plan of like increasing it slowly. P6, “much better”

Where participants had engaged in exercise pre-illness, a belief that exercise equated to health, and a knowledge that steadily increasing physical activity could lead to improved ability and fitness, may have been established prior to GES. Thus, GES already fitted their prior health beliefs model, albeit at a much slower pace, and from a lower baseline.

I used to do triathlon. And I knew about sort of pushing myself a little bit extra each time I trained to accomplish a little bit more. ... It sort of made perfect sense to me that there would be similar approach with the therapy. P8, “much better”

Other factors that participants from both groups reported as being important motivators included: personal attributes (stubbornness, determined, or positive), life philosophies (e.g. taking personal responsibility for their own destinies, preferring not to be on medication), or overcoming fears/scepticism about GES.

To be fair I had quite a sceptical mind to begin with because I have tried many things to improve my symptoms over the years and actually doing more, even if it is just a

little bit more, has never ever benefited me. But I was at a stage where I would do anything to see if it worked. P5, “a little worse”

Support from other people and therapies

Participants described how their partner, family and friends also helped them to maintain their motivation. These significant others could provide practical and emotional encouragement and support.

Definitely the support of my wife. Just you know, supporting me and encouraging me when I'd managed to do a little bit more. And I think again, if you're living alone and you're on your own I think that could be very difficult. P8, “much better”

A number of participants in the “much better” group reported their use of GES being supported by complementary therapies, counselling, CBT, self-help or peer support (that they either undertook concurrently or after taking part in the GES programme). For example, two participants had used complementary therapy approaches during the trial, which they felt supported their recovery and gave them more energy, making it easier for them to engage with GES.

Even if I was getting better from another method [complementary therapy] it's still important to gradually start that walking because otherwise I might end up going backwards again. P6, “much better”

Discussion

Key findings

This study sought to understand the experiences of guided graded exercise self-help delivered as part of an RCT. In particular, we compared experiences, perspectives and approaches of those who were “much better” or “a little worse” after the 12 week programme [17]. Our

findings revealed similarities between the “much better” and “a little worse” groups in their experiences and approaches to GES: many found doing GES challenging, and personal motivation played a key role in participants sticking with their GES programmes. We also discovered a commonly experienced “indeterminate phase”, where participants had to wait a considerable time (from weeks to months) before they experienced any benefits of GES, whilst concurrently experiencing various challenges associated with increasing their activity. However, both the “a little worse” and “much better” groups reported being motivated to undertake GES, and the majority were able to undertake at least some of their GES programme. Participants felt their engagement was supported by skilled therapists who demonstrated understanding about what it was like to cope with CFS/ME. Other things happening in participants’ lives, bodily responses to GES (e.g. effect on CFS/ME symptoms) and illness duration influenced subjective assessments of the success of GES. Both groups reported similar barriers and facilitators to GES, although the “much better” group reported more facilitators to doing GES (e.g. support from significant others, using complementary therapies as support), and reported high levels of motivation to undertake and persevere with GES. The “a little worse” group were more likely to report barriers, including greater symptom exacerbation in response to GES, which interfered with their life commitments (e.g. chores, childcare). Greater interference from comorbid conditions and things happening in their wider lives interfering with GES were also reported in the “a little worse” group. This group had also had CFS/ME for considerably longer than the “much better” group.

Discussion of key findings

The GETSET trial included a structured booklet and guided support from trained physiotherapists. Our results suggest that the booklet alone is unlikely to be sufficient to support patients through GES successfully; participants considered the additional assistance

from the trained physiotherapists a key to success. However, the results also suggest that individuals who have been ill with CFS/ME for a relatively longer period of time and have additional comorbid conditions may benefit from more intensive and bespoke therapy, additional or substitute treatments (e.g. CBT), or a broader graded activity approach which also considered mental energy expenditure [30, 31]. Participants in this, and other studies [32], noted the helpful attitudes (e.g. gentle) and supportive approaches of their GES physiotherapists, regardless of whether GES helped them or not. This contrasts with findings that people with CFS/ME using rehabilitation therapies in a range of settings report communication challenges with therapists and pressures to comply with treatment [10]. Previous research has highlighted a gulf of understanding between health professionals and people with CFS/ME, which markedly contrasts with our findings [33, 34, 35]. Taken together our findings highlight that having knowledgeable, patient-centred and empathic health professionals providing support can make a difference in patient-perceived successes of CFS/ME interventions. This approach is certainly important for positive patient experiences of interventions and support, and may help patients better deal with any challenges they experience.

Our findings also show that the “a little worse” group had been ill substantially longer than the “much better” group. It is beyond the scope of this study to establish how this could influence results. It may be that shorter illness duration reflects lower illness severity and/or is linked to exacerbation of symptoms, which was reported as worse in the “a little worse” group, and cited as a reason for discontinuing GES activities. It may also be that illness duration influences self-efficacy (confidence that one’s actions will lead to the desired goal): one participant in the “a little worse” group felt the length of time she had been ill made it difficult for her to change her behaviour to help improve her condition. Thus, it may be that

an issue such as kinesiophobia (fear of movement and activity), which is known to occur in CFS/ME [36] is more established in those with longer illness duration. However, studies into patient change attributions for self-management interventions found that those who did not improve often attributed this unchanged behaviour due to lack of time or motivation, or a more general sentiment that everything was the same or nothing will help [14, 15]. Our findings correspond with other studies that have found recovery is more likely in those who have been ill for shorter durations [37, 38], and that delaying treatment is associated with reduced treatment efficacy [30]. Our results also suggest the importance of early diagnosis of CFS/ME [6, 9] and targeted GES for those newly diagnosed.

It was important for participants to have time in their lives to undertake GES. This included time to do GES activities and, perhaps more significantly, spare capacity remaining to accommodate any exacerbation of symptoms and for such setbacks not to interfere with essential life activities. Thus, GES was reported to work best for participants who had fewer commitments that interfered with GES, and/or for those who were able to organise their lives around the activity, where gaining additional support from partners, family and employers was also helpful. Those with more severely disabling CFS/ME may, paradoxically, find engagement easier because GES activities were a primary focus of their day, and were necessarily of shorter durations (and therefore it was easier to find time to do them). The exclusion of the most severely disabled patients from this trial precludes us generalising this observation to the most disabled patients found in clinical practice.

Clinical implications

Our findings suggest a number of clinical implications for the way exercise interventions are delivered. Motivation appeared vital in participants adhering to their GES programme,

although engagement did not guarantee improvement. More participants in the “much better” group articulated exceptionally high motivation. One might speculate that such motivation is linked to an internal locus of control, defined as the expectancy that one can influence outcomes directly as a result of one’s behaviour [39]. Participants in a CBT programme for CFS/ME suggested that their motivation to participate required ‘open-mindedness’ and an acceptance of the biopsychosocial model of illness for CFS/ME [32]. Our results suggest that it is useful to help patients internalise a convincing theory of how GES works to support motivation, although this may be challenging given that the exact mechanisms underpinning any improvement following GET are unclear [40]. We know that GET does not work by improving physical fitness, although it may improve exercise tolerance; it may be that it also works as a graded exposure therapy by reducing fear avoidance [41]. Participants in this study developed an understanding of how GES worked in relation to pre-illness knowledge/experiences of exercise e.g. gradually building strength and functioning to reach exercise goals. In addition, debriefing about past use of exercise interventions (including providing an explanation if it was previously unsuccessful) was important. Our findings also suggest that patients should be encouraged to enlist the support of significant others and find an activity they enjoy as part of their GES programme. Additionally, working with patients’ existing knowledge of GES and exercise could help them develop a more useful understanding of GES. Being seen by a psychiatrist in a psychiatric department had negative connotations for two participants, which may affect engagement in treatment [42]. Thus psychiatrists’ involvement in assessment and treatment needs to be explained to patients as not implying that CFS/ME is a psychiatric condition, e.g. that psychiatrists have a role in both assessment (to help rule out psychiatric diagnosis) and in treatment should the patient also have a comorbid condition such as depression.

A number of participants from both groups reported an exacerbation of symptoms during their GES programme (particularly pain and fatigue). This contrasts with the usual perceived response to physical activity by healthy people, who frequently report feeling good after physical activity [43]. However, it is not uncommon for healthy people to report pain after exercise in the form of delayed onset muscle soreness, which is more common if unaccustomed to exercise [44]. CFS/ME patients hope that physical activity will also make them feel good relatively quickly, and may become fearful or demotivated when not only do they not feel better/progress is slow, but also experience pain and fatigue after exertion, which is a characteristic feature of the illness [1, 45]. It is an apparent paradox that graded exercise programme are prescribed for patients with CFS/ME, when post-exertional malaise is a feature, which requires explanation.

What can also be confusing is that the booklet used for GES suggests that GES will not cause them any “ill effects”. This is specifically in reference to the lack of serious adverse reactions and lack of other increases in adverse outcomes reported in GET trials, and not the exercise-induced symptom exacerbations described later in the booklet. Nevertheless, this may cause patient confusion regarding the usual effect of exercise they might expect and any post-exertional malaise they may experience, and careful editing of the booklet is recommended here.

Finally, our findings have clinical implications for GES delivery, emphasising that GES activities should prioritise the incorporation of personalised and enjoyable exercise into the patient’s daily routines. As in other areas of health promotion research and theory (e.g. stroke rehabilitation, physical activity) [46, 47, 48], this was found to be key to adherence and sustainability.

Limitations

Our sample of 19 participants were invited from a potential list of 32 participants. It is possible that patients who did not take part had views and experiences of GES that we were unable to document in this study (for example, two participants felt too ill to be interviewed). We used the Clinical Global Impression change scale [25] to identify suitable patients, those who reported being improved (“much better” or “very much better”) or deteriorated (“much worse” or “very much worse”) post-trial, but because no participant rated themselves as “much worse” or “very much worse”, we also included those who rated themselves as “a little worse” [17]. This approach therefore unbalanced our groups, although at the same time, the lack of worsening is reassuring. Data saturation was reached for the ‘much better’ group and was approaching saturation for ‘a little worse’ group. The researcher was not blinded to group during the study and analysis, as this would not have been possible. Thus we do not make claims of generalisability to wider populations, but rather hope to provide insights which may be useful for future research and delivery of graded exercise approaches for patients with ME/CFS. The “a little worse” group had been ill substantially longer than the “much better” group (see above section for discussion on this point), this finding would be important for future studies to examine further, as findings on illness duration and response to treatment are mixed [49].

Participants had completed GES an average of 22 months (range 3 to 26) earlier at the time of interview. Including diverse time points and allowing time for some patients to consolidate their accounts about the treatment and trial, is consistent with aiming to include diverse sampling in qualitative research [50]. However, occasionally some participants had challenges remembering the finer details of their GES programmes. Additionally, whilst it

was the aim of this study to ascertain subjective accounts, some of these accounts may have been subject to recall bias. For example, peak-end theory [51] suggests people often remember an experience based on how they felt at ‘peak’ moments (which might be good or bad) and the end, rather than the experience as a whole. Alternatively, current mood states have been shown to influence how positively or negatively events are recalled [52], which may account for more positive reviews of GES from those who were feeling better at the time of the interview, and vice versa. Finally, this is a study of GES and not therapist delivered GET. A similar qualitative study for a GET trial may uncover different participant experiences, which would add further to our knowledge of exercise interventions for CFS/ME.

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Informed consent: Written informed consent was obtained from all individual participants included in this study.

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