

Fear of movement and avoidance behaviour toward physical activity in chronic-fatigue syndrome and fibromyalgia: state of the art and implications for clinical practice

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Abstract Severe exacerbation of symptoms following physical activity is characteristic for chronic-fatigue syndrome (CFS) and fibromyalgia (FM). These exacerbations make it understandable for people with CFS and FM to develop fear of performing body movement or physical activity and consequently avoidance behaviour toward physical activity. The aims of this article were to review what measures are available

for measuring fear of movement and avoidance behaviour, the prevalence fear of movement and avoidance behaviour toward physical activity and the therapeutic options with fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM. The review revealed that fear of movement and avoidance behaviour toward physical activity is highly prevalent in both the CFS and FM population, and it is related to various clinical characteristics of CFS and FM, including symptom severity and self-reported quality of life and disability. It appears to be crucial for treatment (success) to identify CFS and FM patients displaying fear of movement and avoidance behaviour toward physical activity. Individually tailored cognitive behavioural therapy plus exercise training, depending on the patient's classification as avoiding or persisting, appears to be the most promising strategy for treating fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM.

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Abbreviations

FM Fibromyalgia
CFS Chronic-fatigue syndrome
TSK Tampa scale of kinesiophobia

Introduction

Chronic-fatigue syndrome (CFS) and fibromyalgia (FM) are severely disabling and largely overlapping [1, 2] disorders, both characterised by chronic fatigue, chronic widespread pain, concentration difficulties and physical inactivity [3, 4]. Both disorders are associated with high direct and indirect

medical costs [5, 6]. CFS and FM share many clinical features, and the diagnostic criteria are partly overlapping as well [1, 2]. Both disorders are characterised by the same underlying mechanism: signs and symptoms result from the hyperexcitability of the central nervous system (or central sensitisation) [7–9]. Hence, it comes as no surprise that evidence-based treatments (i.e. exercise therapy and cognitive behaviour therapy [10–13]) are identical in CFS and FM.

Despite of the partly overlapping diagnostic criteria, disease mechanism and treatments, CFS and FM are not identical disorders. CFS and FM are diagnostically different entities. For instance, while pain is the hallmark symptom in FM, between 6 and 16 % of CFS patients do not suffer from pain [14]. Indeed, patients with chronic fatigue but without widespread pain can comply with the 1994 Center for Disease Control and Prevention criteria for the diagnosis of CFS [4]. Likewise, the 1990 [3] and the 2010 [15] American College of Rheumatology criteria for the diagnosis of FM enable that patients without debilitating chronic fatigue can fulfill the diagnostic criteria for FM. Taken together, although the similarities between CFS and FM have inspired us to combine both disorders in one review paper, study findings will not be pooled and will be discussed separately throughout.

In patients with CFS and FM, vigorous exercise [16–18] or a sudden increase in physical activity [19, 20] frequently triggers a symptom exacerbation. Physical activity can be defined as any bodily movement produced by skeletal muscles that result in energy expenditure, while exercise is a subset of physical activity (i.e. one that is planned, structured, repetitive and targets improvement or maintenance of physical fitness). This severe exacerbation of symptoms following physical activity, as seen in CFS and FM patients, is one of the core features of the illnesses [20–22]. Thus, it seems understandable for people with CFS and FM to develop fear of performing physical activity and consequently avoidance behaviour toward physical activity.

Vlaeyen et al. [23] developed the cognitive behavioural model for fear of movement. This model is based on the assumption that for some patients, a pain experience will lead to fear of movement and consequently to avoidance behaviour as a maladaptive way of coping with their pain. In this chain of events, the fact whether or not a patient exaggerates his pain experience level (i.e. pain magnification as part of the catastrophising construct), plays a key role in determining whether avoidance of physical activity or confrontation will be used as a coping strategy. In the long term, avoidance of movement and physical activity results in invalidating and psychological changes (e.g. depression, disuse and disability) that contribute to the symptom complex of chronic pain patients. In addition, the fear-avoidance model implies that noncatastrophising patients will more likely use confrontation as an active and adaptive coping

strategy, which increases the odds favouring functional recovery [23]. Ever since its introduction in 1995, a series of studies have examined the applicability of this cognitive behavioural model in people with CFS and FM, reviewed in the present paper.

Fear of performing physical activity or body movement has been described with a variety of conceptual definitions among which pain-related fear, fear-avoidance beliefs, fear of movement and kinesiophobia are the most commonly used [24]. Kinesiophobia is defined as ‘an excessive, irrational and debilitating fear of physical movement and activity resulting from a feeling of vulnerability to painful injury or reinjury’ [25]. Following its definition, kinesiophobia is one end of the spectrum of fear of movement, and kinesiophobia does not comply with the requirements of a phobia, as stipulated by the Fourth Edition of the Diagnostic and Statistical Manual of Mental Disorders [26]. Therefore, the term kinesiophobia will not be used here. For closely related constructs like ‘fear-avoidance beliefs’ and ‘pain-related fear-avoidance beliefs’, a conceptual definition is unavailable in the scientific literature [24]. Still, fear-avoidance beliefs is a more general concept referring to fear and hence avoidance of all kinds of activities, not specifically focusing on movement or physical activity (e.g. fear that a cognitive task like reading a book will worsen symptoms). Pain-related fear is not a valuable alternative either, as it has been broadly defined as fear that incorporates fear of pain, fear of injury, fear of physical activity and so forth [27]. The present paper focuses on fear of worsening symptoms due to physical activity/body movement and avoidance of such physical activities/body movements by patients with CFS and FM. Consequently, and for clarity reasons, fear of movement and avoidance behaviour toward physical activity are used throughout the paper.

The aims of this article are to review what measures are available for measuring fear of movement and avoidance behaviour, the prevalence of fear of movement and avoidance behaviour toward physical activity, the relation of fear of movement and avoidance behaviour toward physical activity with symptom severity and disability, and the therapeutic options with fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM.

Methods

The answers to the above-presented aims are based on relevant scientific literature up to September 2012. Two scientific, electronic search engines were used, namely PubMed (<http://www.ncbi.nlm.nih.gov/pubmed/>) and EBSCO (<http://www.ebscohost.com/>). For the latter search engine, the two following databases were used: Medline and Academic Search Premier. Relevant papers were retrieved

by combining the following keywords: FM, CFS, kinesiophobia, fear of movement, fear avoidance, avoidance behaviour, Tampa scale for kinesiophobia (TSK), fear-avoidance beliefs questionnaire, fear-avoidance questionnaire, treatment, therapy, rehabilitation, prognostic factors, perpetuating factors and mediating factors. Relevant papers were included only if they reported original data of humans suffering from either CFS or FM, addressed at least one of aims of this review, were published as full papers and were written in English, Dutch or Spanish. Papers reporting animal studies, case reports, review articles, congress proceedings, abstracts, letters to the editor or editorials were not included for further appraisal. Initially, a systematic literature review with methodological quality appraisal was initiated. However, the heterogeneity in study design, patient characteristics (e.g. diagnostic criteria used, age, and gender), study aims and assessment methods precludes such a systematic approach to the literature review. Hence, a more narrative approach is used.

Assessments of fear of movement and avoidance behaviour toward physical activity in CFS and FM

Only 10 out of 20 CFS patients were able to achieve 85 % of the age-predicted target heart rate during an incremental exercise stress test, compared to 11 out of 14 healthy controls [28]. It seems rational to use the non-achievement of 85 % of the age-predicted target heart rate during an incremental exercise stress test for the ‘diagnosis’ of avoidance behaviour toward physical activity, as done in this study [28]. However, the data in the control group indicate that this is an inappropriate way of ‘diagnosing’ avoidance behaviour toward physical activity, a notion which is further supported by the lack of association between fear of movement and peak exercise capacity testing data of CFS patients [29, 30].

For measuring beliefs concerning fear of movement and avoidance behaviour toward physical activity among CFS patients, Deale et al. used four statements about exercise and activity reductions, to rate on a 4-point scale from strongly agree to strongly disagree [31]. They found that 41 out of 60 (68 %) CFS patients agreed that they should avoid exercise when tired, 37 (62 %) agreed that doing less resulted in less fatigue, 38 (63 %) agreed that exercise was harmful, but only 5 (8 %) agreed that they should avoid physical activity [31].

In FM research, little attempts have been made to adopt the existing scales for measuring fear of movement and avoidance behaviour toward physical activity. van Koulil et al. used the Pain-Coping Inventory for identifying pain-avoidance patterns in patients with FM [32]. The TSK [23] is often used for studying fear of movement in FM patients [20, 33, 34]. Scores greater than 37 on the TSK are used to classify FM patients as having high levels of fear of

movement, but this cut-off was derived from a group of low back pain patients, questioning its validity in a widespread pain condition like FM.

In CFS research, several attempts were made to make the TSK more appropriate for studying fatigue-related fear of movement. The *TSK version CFS (TSK-CFS)* has been constructed for the assessment of fear of movement in patients with CFS. The TSK-CFS is a modification of the TSK, a self-assessment tool for the measurement of pain-related fear of movement [23], in which the word ‘pain’ has been replaced by ‘my symptoms’ [29]. Data supportive of the internal consistency, congruent and convergent validity of the Dutch version of the questionnaire have been provided [29]. The French and Dutch version of the TSK-CFS displayed good test-retest reliability (intraclass correlation coefficients=0.91 and 0.83 respectively), and the Cronbach’s α coefficient of the items included in the French version was 0.74 [35].

Fatigue-related fear of movement can be measured with the *TS fatigue* [36, 37]. The TS fatigue is a scale based on the TSK in which the word ‘pain’ has been replaced with the word ‘fatigue’ [36]. The English version of the TS fatigue has been shown to have a good test-retest reliability (correlation coefficient (r)=0.89), fair internal consistency (Cronbach’s α =0.76) and two identified subscales: illness beliefs (α =0.78) and beliefs about activity (α =0.70) [36]. The scale has been translated into Dutch [37] and has been used in a large population of CFS patients, and patients with other conditions. Based on a varimax rotated factor-analysis, a shortened scale was developed consisting of eight items (item 1, 3, 6, 7, 10, 11, 14 and 17 of the original TS fatigue) with a total score ranging from 8 to 36. Reliability of the new scale was assessed in 195 patients with a diagnosis of CFS and was found to be sufficient (Cronbach’s α =0.80) [38].

When comparing these two different measures for assessing fear of movement in people with CFS, it becomes clear that the TS fatigue measures fatigue-related fear of movement and the TSK-CFS measures fear of worsening the overall symptom complex (including fatigue, pain, concentration difficulties etc.) due to physical activity. One study compared some of their psychometric characteristics, but the study findings are inconclusive regarding the utility or validity of one measure over the other [38]. Data in support of the validity (concurrent and criterion validity) of both measures was provided [38].

In conclusion, advances have been made in self-reported measures for assessing fear of movement and avoidance behaviour toward physical activity in CFS and FM patients [39]. While pain-related fear of movement can be assessed using the original TSK, as typically done in FM studies, the TS fatigue and TSK-CFS provide valuable options for measuring fatigue- and symptoms-related fear of movement respectively in patients with CFS. A gold standard for

assessing fear of movement and avoidance behaviour toward physical activity in patients with CFS or FM is currently lacking, neither are well-validated cut-off scores for interpreting results from the TSK (fatigue) in either populations.

Prevalence of fear of movement and avoidance behaviour toward physical activity in CFS and FM

Chronic-fatigue syndrome

When examining the study findings addressing fear-avoidance beliefs in CFS patients, several compelling findings appear. Although similar to non-CFS-fatigued controls, patients with CFS ($n=43$) displayed more escape-avoiding behaviour than healthy controls [41]. However, escape-avoiding behaviour addresses all kinds of activities such as administrative work and social activities, and thus is not limited to physical activity or exercise.

The lack of a difference in daily physical activity levels in anticipation to an exercise test, both on the day of and the day before the test [42, 43], has been interpreted as absence of ‘exercise phobia’ in CFS patients without a comorbid psychiatric disorder [42]. This contradicts to several studies that reported fear of movement to be a common feature in patients with CFS [29, 30, 35, 36] and to the study by Wallman et al. [44] who concluded from an exercise physiology study that differences in exercise tolerance and exercise capacity between CFS patients and healthy sedentary controls are due to avoidance behaviour toward physical activity [44]. More specifically, they suggested that patients with CFS are reluctant to exercise to the target heart rate due to an underlying, yet unexpressed, fear of relapse [44].

Fibromyalgia

Turk et al. studied a large group of FM patients ($n=233$), and found that nearly 39 % ($n=90$) displayed high levels of fear of movement [34]. In another study, fear of movement and avoidance behaviour toward physical activity was identified in 145 (40 %) out of 359 FM patients [32].

The above study findings suggest that not all patients with FM have fear-avoidance beliefs. In fact, many patients with FM display persistence behaviour (214 of 359 FM patients) [32], which often results from self-discrepancies. Patients with persistence behaviour try to ignore pain sensations and the (physical) boundaries of their body, suppress pain-related thoughts and persist in daily activities (including physical activities), resulting in over activity. Compared with fear-avoidant FM patients, persisting FM patients experience less pain and fatigue, functional disability, negative mood, worrying about, hypervigilance, helplessness, and

perform better on physical fitness tests (e.g. they walk longer during a shuttle walking test) [32, 40].

Summary

The scientific literature suggests that approximately 40 % of patients with FM display high levels of fear of movement and avoidance behaviour toward physical activity, but the CFS literature - although consistently indicating that fear of movement is prevalent in the CFS population - is less clear in reporting prevalence data.

Fear of movement and avoidance behaviour toward physical activity in CFS and FM in relation to symptom severity or disability

Chronic-fatigue syndrome

Little data addressing the possible etiological role of fear of movement and avoidance behaviour toward physical activity in CFS or FM are currently available. In a large British national birth cohort study ($n=4,779$), continuing to be active despite increasing fatigue was identified as a possible crucial step in the development of CFS [45]. This is an important finding, as it provides evidence that patients with CFS are certainly not lazy, even though this finding does not preclude the development of fear of movement and avoidance behaviour toward physical activity once the illness is established.

In people with CFS, escape-avoiding behaviour is significantly associated with bodily pain, reduced activity, low energy level, but not with muscle fatigue [41]. These findings lead the authors to speculate that escape-avoiding behaviour is of prime clinical importance to patients with CFS, and that interventional strategies for CFS should address this maladaptive way of coping with their illness [41]. They feel that discouraging avoiding activity should be included in behavioural interventions for CFS [41].

As is the case with escape-avoiding behaviour, evidence supportive of the clinical importance of fear of movement to CFS patients has been provided [29, 35, 36]. Indeed, fear of movement is associated with self-reported activity limitations and participation restrictions [29]. In addition, in a Flemish population of CFS sufferers, the TSK-CFS total scores correlated significantly with the general health perception subscale of the short-form 36 items health status survey but not with the physical-functioning subscale scores or any of the remaining subscale scores [35]. Among a population of French-speaking patients with CFS, associations between the TSK-CFS total scores and various domains of quality of life (physical functioning, role limitations due to emotional problems and social functioning) were identified [35]. However, the total scores of the TSK-CFS were unrelated to symptom severity [35].

When CFS patients were asked to ride a stationary bike for as long as they felt able to, the beliefs of activity subscale and total score of the TSK fatigue correlated highly with distance travelled and were identified as an important predictor of behavioural persistence [36]. The same study revealed that in patients with CFS, beliefs about activity are more important than physical symptoms, physical disability, mood or other illness perceptions [36].

Anticipated symptoms before physical activity (i.e. the subject's predicted symptoms during a future physical activity) might influence a patient's physical performance, daily physical activity level, as well as beliefs [18]. When patients strongly anticipate symptom exacerbations in response to a particular physical demanding task, before the initiation of that task, they may avoid the task or perform it less vigorously. As a consequence, these patients might even become less physically active. There is some evidence suggesting that fatigue severity is related to actual physical activity level and high fatigue expectations are related to low physical activity levels in people with CFS [46]. This suggests that people with CFS try to prevent symptoms by avoiding physical activity. This view is further supported by the observation that high levels of fear of movement are related to low daily physical activity and high disability levels [29].

Unfortunately, several studies examining fear of movement in CFS patients used exercise stress tests [29, 30, 36, 42], which are unlikely to have high ecological validity. Previously, we reported that in Dutch people with CFS, performance during stair climbing is related to higher momentary and expected fatigue levels, and focusing on bodily symptoms and fatigue catastrophising influence expected fatigue levels [37]. Applying the same experiment to a Belgian sample of CFS patients revealed that fear of movement and fatigue catastrophising, but not symptom expectancies, are strongly related to stair climbing performance in Belgian people with CFS [38]. These data underscore the importance of fear of movement for performing threatening and physical-demanding tasks in everyday life of people with CFS [38].

Importantly, data from one CFS study indicate that fear of movement is of relevance for threatening physical activities like stair climbing but not for physical activity in general [38]. Fear of movement was not associated with daily physical activity in patients with CFS [38]. This is important for treatments aiming at increasing daily physical activity level in patients with CFS (as typically done in graded activity programs). Diminishing fear of movement appears only relevant for targeting physical activities that are perceived as threatening and not for increasing work-related or social (physical) activities. This suggests that clinicians should first identify physical activities that are perceived as threatening, and then restructure the patients' beliefs in relation to this threatening task specifically. Furthermore, treatments aiming at increasing physical activity level in people with

CFS should focus on activities that individual patients find pleasant to do and thus are not perceived as threatening.

Fibromyalgia

Likewise, in a study on repetition-induced summation of activity-related pain, patients with FM were asked to lift a series of 18 weighted canisters and to rate their pain after each lift [20]. The repetition-induced summation of activity-related pain was significantly correlated with fear of movement and not with pain catastrophising or depressive thoughts [20]. Whether fear of movement is a consequence of previous experiences of activity-related pain like post-exertional malaise, or alternatively aggravates (expectations for) activity-induced symptom increases, requires further study.

In patients with FM, fear of movement was unrelated to changes in psychological distress over a 9 month period [33]. However, other study findings provide evidence favouring the clinical importance of fear of movement in patients with FM. In one study, the total scores on the TSK showed small but statistically significant associations with disability, pain severity and treadmill performance (r between 0.20 and 0.25) [34]. Smaller but still statistically significant correlations were found with the TSK scores and grip strength and trigger point severity ($r=-0.16$ and 0.14, respectively) [34]. When comparing the high fear of pain and activity FM patients with the low fear group, the former had greater disability, depressed mood, pain severity and lower treadmill performance [34].

Summary

Taken together, these findings stress the clinical importance of fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM. This implies that once identified, clinicians should address fear of movement and avoidance behaviour toward physical activity during their treatment provided to patients with CFS and FM.

Treatment of fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM

A randomised controlled clinical trial examining the effectiveness of pain physiology education versus pacing activity self-management education in patients with CFS used the TSK-CFS as a secondary outcome measure [47]. The pain physiology education comprised educating the patients about hyperexcitability of the central nervous system in CFS and the implications for daily practice, including physical activity. Although the decrease in mean total score on the TSK-CFS was larger in the pain physiology education group (i.e. from 39 at baseline to 33 post-treatment), there

was no statistical significant difference between groups [47]. Hence, it was concluded that neither pain physiology education, nor pacing activity self-management education was able to decrease fear of movement in people with CFS.

Cognitive behaviour therapy is an effective treatment for patients with CFS [10, 11], but it does not result in a decrease in avoidance behaviour toward physical activity [48]. Moreover, the decrease in the focus on fatigue rather than changes in avoidance toward physical activity mediates the effects of cognitive behaviour therapy in CFS [48]. Partly contrasting these findings are the results from a randomised controlled clinical trial showing that illness beliefs about avoidance of physical activity in CFS patients improved in response to cognitive behaviour therapy but not following relaxation therapy [31]. In addition, the same study showed that good outcome following cognitive behaviour therapy in CFS is associated with changes in avoidance behaviour and related beliefs, rather than causal attributions [31]. Differences in treatment protocols, type of measures used, and the focus on fatigue versus the general symptom profile of CFS patients may account for the observed differences. By using a fatigue-modified version of the Pain Coping Inventory, Wiborg et al. focused on fatigue-related avoidance behaviour toward physical activity and they did not find an effect of cognitive behaviour therapy on this fatigue-related avoidance behaviour of physical activity [48]. The contrasting findings in the study of Deale et al., is based on the rating (strongly agree to strongly disagree) of four statements about exercise and activity reductions [31]. In general, it is concluded that cognitive behaviour therapy for people with CFS might result in decreased avoidance behaviour toward physical activity, but this depends on the type of cognitive behaviour therapy applied and is certainly not restricted to the fear of aggravating fatigue following physical activity.

In FM, a multidisciplinary treatment comprising 7 sessions of education and 25 sessions of physiotherapy (including relaxation, exercise and graded activity), did not alter the level of fear of movement in 65 patients [49]. However, the multidisciplinary treatment resulted in improved fatigue, quality of life and functionality [49].

As outlined above, not all patients with CFS or FM have fear of movement and avoidance behaviour toward physical activity: many patients with CFS and FM display persistence behaviour. Compared with fear-avoidant CFS or FM patients, persisting CFS and FM patients require cognitive restructuring, pacing activity self-management and acceptance-based approaches rather than operant behavioural interventions like graded exposure or graded exercise therapy [32]. This view has been validated in a randomised trial in patients with FM, who received different cognitive behavioural therapy plus exercise training depending their classification as pain avoidant or pain persistent [40].

The pain persistence treatment focused on regulating and diminishing persistence behaviour by teaching patients to pace their activities and to alternate between activity and inactivity, followed by gradually increasing their daily activities [40, 50]. Such a tailored approach was found to be effective in improving physical and psychological functioning [50], and changing avoidance behaviour toward physical activity and activity pacing in the pain-avoidance and pain-persistence groups, respectively [40]. Importantly, the mediation analyses revealed that changes in physical functioning were mediated by decreases in avoidance behaviour toward physical activity in the pain-avoidance treatment group, and by improvements in activity pacing in the pain-persistence group [40]. To the best of our knowledge, we are unaware of similar studies in patients with CFS. However, pacing activity self-management for addressing persistent behaviour in CFS patients has been described in detail [51], and preliminary findings from pilot studies supports its use [51–53].

In summary, progress has been made with regard to identifying effective and ineffective treatment strategies for tackling fear of movement and avoidance behaviour toward physical activity in people with CFS and FM. A crucial aspect for treatment appears to be the identification of CFS and FM patients displaying fear of movement and avoidance behaviour toward physical activity. If present, specific cognitive behaviour therapy plus exercise therapy appears warranted.

Discussion including a research agenda

Patients suffering from CFS or FM often apply avoidance as a strategy of dealing with issues, including avoidance of emotions [54]. The present paper focused on fear of worsening symptoms due to physical activity/body movement and avoidance of such physical activities/body movements by patients with CFS and FM. From the available literature, it is concluded that fear of movement and avoidance behaviour toward physical activity are highly prevalent in the CFS and FM population, and once present they are related to various clinical characteristics of CFS and FM, including symptom severity and self-reported quality of life and disability. Individually tailored cognitive behavioural therapy plus exercise training depending on the patient's classification as avoiding or persisting appears to be the most promising strategy for treating fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM (Table 1).

At current, a gold standard for assessing fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM is unavailable, neither are well-validated cut-off scores for interpreting results from the TSK

Table 1 Overview of treatment approaches for diminishing fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM

Reference	Population	Treatment	Outcome	Results
[47]	CFS	Pain physiology education↔pacing activity self-management education	Fear of movement	=
[48]	CFS	Cognitive behavioural therapy	Avoidance behaviour toward physical activity	=
[31]	CFS	Cognitive behavioural therapy	Illness beliefs about avoidance of physical activity	+
[40, 50]	FM	Tailored cognitive behavioural therapy plus exercise and training depending their classification as pain avoidant or pain persistent	Avoidance behaviour toward physical activity	+
[49]	FM	Multidisciplinary treatment: education+relaxation+exercise+graded activity	Fear of movement	=

(fatigue). Clinicians and researchers can use the original TSK for assessing pain-related fear of movement, and the TS fatigue and TSK-CFS provide valuable options for measuring fatigue- and symptoms-related fear of movement respectively. Besides listing the search for a gold standard as a research priority for clinimetric studies (Table 2), future studies should address the issue of objectively measuring avoidance behaviour of physical activity, including avoidance of fear-eliciting activities as well as more subtle safety-seeking behaviours [39].

Several studies have examined and confirmed the clinical importance of fear of movement and avoidance behaviour toward physical activity in people with CFS and FM [20, 29, 34–36, 38], especially with self-reported quality of life and disability measures. When using ecological valid assessments like lifting tasks and stair climbing protocols, fear of movement and avoidance behaviour toward physical

activity appear to be determinants of physical performance as well [20, 37, 38]. It seems rational to study the role of fear of movement and avoidance behaviour toward physical activity in CFS and FM within the framework of the extended fear avoidance model of chronic pain [55]. Such an extended or next generation fear avoidance model might include a motivational perspective on goals and self-regulation, as typically applied in pain management programs [55]. These are important avenues for further work in this area.

For the treatment of fear of movement and avoidance behaviour toward physical activity in people with CFS, cognitive behaviour therapy for people with CFS might result in decrease avoidance behaviour toward physical activity, but this depends on the type of cognitive behaviour therapy applied and is certainly not restricted to the fear of aggravating fatigue following physical activity. Likewise,

Table 2 Research agenda for studying fear of movement and avoidance behaviour toward physical activity in CFS and FM

Domain	Population	Content
Measurement quality	CFS and FM	Search of a gold standard for assessing fear of movement and avoidance behaviour toward physical activity
Measurement quality	CFS and FM	Objectively measuring fear of movement and avoidance behaviour toward physical activity, including avoidance of fear-eliciting activities as well as more subtle safety-seeking behaviours [39]
Role as perpetuating factor	CFS and FM	Study of the role of fear of movement and avoidance behaviour toward physical activity in FM and CFS within the framework of the extended fear avoidance model of chronic pain, including a motivational perspective on goals and self-regulation [55]
Role as perpetuating factor	CFS and FM	Examining whether fear of movement is a consequence of previous experiences of activity-related pain like post-exertional malaise or alternatively aggravates (expectations for) activity-induced symptom increases
Treatment	CFS and FM	Examining the feasibility and effectiveness of exposure in vivo for ‘treating’ fear of movement and avoidance behaviour toward physical activity
Treatment	CFS	Examining the effectiveness of individually tailored cognitive behavioural therapy plus exercise training depending on the patient’s classification as avoiding or persisting

individually tailored cognitive behavioural therapy plus exercise training depending on the patient's classification as pain avoidant or pain persistent was found to be effective in diminishing pain-avoidance behaviour in the pain avoidant group [40, 50]. Such an individually tailored approach to cognitive behavioural therapy plus exercise training holds promise and requires testing in a population of CFS sufferers. Pain physiology education, pacing activity self-management education, relaxation, or multidisciplinary treatment comprising of education, relaxation, exercise and graded activity are ineffective for diminishing fear of movement in people with CFS and FM [47, 49].

For patients with more localised chronic musculoskeletal pain disorders, specific treatment strategies to reduce fear of movement and consequent avoidance behaviour toward physical activity, like exposure in vivo to a set of individually tailored and fear eliciting physical movements [56], have been developed. Exposure in vivo holds the capacity of being one of the most powerful cognitive behavioural treatments for reducing disabling fear of movement [39], but this treatment has yet to be tested in patients with CFS or FM. One study addressing fear of movement and avoidance behaviour toward physical activity in CFS supports such a targeted approach of threatening physical tasks, rather than a general approach toward increasing physical activity [38].

In conclusion, much work has been done for examining the prevalence and clinical importance of fear of movement and avoidance behaviour toward physical activity in people with CFS and FM. Likewise, several high-quality studies have examined various treatment strategies for diminishing fear of movement and avoidance behaviour toward physical activity in patients with CFS and FM. It is concluded that fear of movement and avoidance behaviour toward physical activity are highly prevalent in both the CFS and FM population, and they are related to various clinical characteristics of CFS and FM. Still, many issues remain, including the development and testing of treatment strategies that specifically tackle fear of movement and avoidance behaviour toward physical activity in people with CFS and FM, as well as other issues listed in the research agenda.

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