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Measuring the physical activity level and pattern in daily life in persons with chronic fatigue syndrome/myalgic encephalomyelitis: a systematic review

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AQ1

AQ2

AQ3

Background: A lower activity level and imbalanced activity pattern are frequently observed in persons with chronic fatigue syndrome (CFS)/myalgic encephalomyelitis (ME) due to debilitating fatigue and post-exertional malaise (PEM). To provide an optimal treatment strategy, insight into a patient's current physical activity level and pattern is necessary and identification of reliable and valid measures or scales measuring physical activity level and pattern in this population is warranted.

Objective: To identify measures or scales used to evaluate activity level and/or pattern in patients with CFS/ME and review their psychometric properties.

Methods: A systematic literature search was performed in the electronic databases PubMed and Web of Science until 12 October 2016. First, articles including relevant measures were identified. Secondly, psychometric properties of relevant measurement instruments were extracted and rated based on the COSMIN checklist.

Results: The review was performed and reported according to PRISMA statement. A total of 51 articles and 15 unique measurement instruments were found, but only three instruments have been evaluated in patients with CFS: the Chronic Fatigue Syndrome-Activity Questionnaire (CFS-AQ), Activity Pattern Interview (API) and International Physical Activity Questionnaire-Short Form (IPAQ-SF), all self-report instruments measuring physical activity level.

Conclusions: The IPAQ-SF, CFS-AQ and API are all equally capable of evaluating the physical activity level, but none of these are optimal to use. Although often used as gold standard to capture physical activity patterns, activity monitors have not yet been evaluated in these patients. More research is needed to evaluate the psychometric properties of existing instruments, including activity monitors.

Keywords: Chronic fatigue syndrome, Data collection, Physical activity, Psychometrics, Review

Introduction

Chronic fatigue syndrome (CFS)/myalgic encephalomyelitis (ME) comprises a complex of symptoms characterized by clinically defined debilitating fatigue that cannot be explained by other medical or psychiatric conditions and is not sufficiently reduced by resting.[1,2] The experience of fatigue causes substantial reductions in previous levels of occupational, educational, social or personal activities, resulting in limitations in meaningful areas of life.[1,2] Scientific evidence indeed shows that the activity levels of patients with CFS/ME are significantly lower than those of healthy subjects and a large variation exists in activity levels between patients.[3–9] Additionally, the performance of mild physical or mental activities can lead to the exacerbation of symptoms, also known as PEM.

PEM is one of the primary characteristics of CFS/ME and a main reason why patients with CFS/ME are unable

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DOI 10.1080/10833196.2017.1300624

to continue their daily routine.[4,8,10,11] The presence of these exacerbations may result in avoidance of activities and prolonged periods of rest, expecting that this strategy will cause improvement.[11,13,14] In contrast to this perception, this strategy instead results in decreased exercise tolerance and reduced ability to perform activities of daily living (ADL).[3,9,11–14]

Additionally, although most patients perform fewer, and mostly sedentary activities, bursts of exertion are sometimes observed in periods in which patients try to perform at pre-morbid level.[6,14,15] This deviant activity pattern observed in some patients with CFS/ME may cause a negative feedback cycle where PEM, an overall lower activity level and imbalanced activity pattern are often observed.[3,6,15,16]

Therefore, the focus of rehabilitation treatment lies in enabling patients to participate in meaningful life activities, depending on a patient's physical, social, cultural and spiritual context and beliefs that promotes or maintains their health, well-being, participation and autonomy.[17–19] Treatment strategies for CFS/ME are focused on activity self-management preventing both PEM and avoidance behaviour.[3,12–14,20,21] Frequently used therapeutic interventions are activity pacing, graded exercise therapy and cognitive behavioural therapy.[15,21–25,27]

To maintain an optimal activity level and balanced pattern over a longer period of time, insight into daily activity performance of a patient is necessary.[6,9,20] Thus, the activity level and pattern need to be established using reliable and valid measures or scales before a clinical practitioner can assess and evaluate a patient's health status, provide information, a suitable treatment strategy and evaluate a patient's course of recovery after treatment.[3–5,9,28]

The aim of the review was twofold. The first aim was to systematically review the literature for measures or scales capable of evaluating the activity level and/or pattern that were used in patients with CFS/ME; second, to critically appraise the psychometric properties of identified measures or scales in patients with CFS/ME.

Method

PRISMA guidelines (Preferred Reporting Items for Systematic reviews and Meta-Analyses) were used to structure the review methods[30] and the eligibility criteria, search strategy, methods for study selection, data-extraction and rating were specified in advance.

Eligibility criteria

To be included in the first part of the present systematic review, studies had to report the use of measures or scales evaluating (physical) activity level or pattern and the study had to be undertaken with adult patients with CFS/ME. The second part of the systematic review only included studies that evaluated the psychometric properties of identified measures or scales during the literature search.

Information sources and search strategy

The electronic databases PubMed and Web of Science were used to execute the literature search (Table 1). Both databases were searched until 12 October 2016 for relevant articles. No limits were set for the date of publication.

Study selection

Study selection was based on two screening phases. Both screening phases were performed by two independent reviewers. The initial literature search was performed until February 2014 and studies were screened by two of the authors (JVR and AD). An update of the systematic literature search was performed from March 2014 until 12 October 2016 and the studies were screened by two other authors (IH and KV). During both literature searches, a third reviewer (MM) was only involved in the screening process if consensus could not be reached between the two reviewers.

The first selection was based on title and abstract. Articles that met the first two inclusion criteria and could not be excluded based on the criteria mentioned below were included for full text reading. The third inclusion criterion was only applied during full-text reading, because not all articles mentioned the used measure or scale in their abstract. All articles that used a relevant measure or scale evaluating the activity level or activity pattern were included, unless exclusion criteria were identified during full-text reading. References of all included articles were checked to identify other articles measuring the psychometric properties of relevant measures or scales.

Inclusion criteria:

- The study included adult humans with CFS/ME;
- was written in Dutch or English;
- included a measure or scale that evaluates (physical) activity level or pattern.

Exclusion criteria:

- Studies regarding measures or scales evaluating limitations in activities, quality of life or any other construct than the activity level or pattern;
- studies measuring body functions including biomarkers, sleep, spirometry or participation;
- laboratory research or *in vitro* research;
- use of a model or theory as intervention;
- random non-further specified or dichotomous questions or instrument measuring fatigue;
- abstract, guideline, congress report, review, meta-analyses, study protocol or case study.

Data extraction and rating

First, all relevant measures or scales evaluating the physical activity level and/or pattern in patients with CFS/ME were extracted from the articles and compiled (Table 2).

Second, as recommended by Mokkink et al., the research methodologies of articles evaluating the psychometric properties of measures or scales assessing the physical activity level or pattern of patients with CFS/ME were rated using

Table 1 Search strategy

Population	Intervention		Outcome	Exclusion
Chronic fatigue syndrome (MeSH)	Measurement(s)	Activities of daily living [MeSH]	Activity	Depression [MeSH]
(benign) myalgic encephalo(mye)litis (ME)	Outcome/health impact/ outcome and process/ risk/process/symptom/ self-) assessment(s) [mesh]	Activity level	Exercise [MeSH]	(depressive/mental) disorder(s) [MeSH]
CFS/ME	Self-reporting questionnaire(s)	Activity pattern(s)	(leisure/human) activities [MeSH]	Psychiatric status rating scales [MeSH]
Post-viral/infectious fatigue syndrome	Interview [mesh]	(treatment) outcome(s)	Physical endurance [MeSH]	Neurasthenia
Yuppie flu	Evaluation(s)	Metabolic equivalent [MeSH]	(physical) movement	
Chronic Epstein-Barr virus syndrome	Health surveys [mesh]		(an)aerobic,	
Myalgia syndrome	Accelerometry [mesh]		Daily functionality	
Myalgic encephalopathy	Actigraphy [mesh]		Psychological adaptation [MeSH]	
	Exercise test [mesh]		Activities of daily living [MeSH]	
	Monitoring		Physical/mental exertion(s)	
	Data collection [mesh]		Physical exertion [MeSH]	
	Evaluation studies as topic [mesh]		Motor activity [MeSH]	
	Instrument(s)		Movement [MeSH]	
	Self-evaluation programs [mesh]			
	Diagnostic self-evaluation [mesh]			
	Health care evaluation mechanisms [mesh]			
	Psychometric characteristics			
	Clinimetric properties			
	Treatment outcome [mesh]			
	Test			
	Interview as topic [mesh]			
	Assessment(s)			
	Questionnaires [mesh]			
	Outcome(s)			

the COSMIN checklist (Consensus-based Standards for the selection of health Measurement Instruments).[31]

The COSMIN checklist was developed in 2010 according to a Delphi study by international experts in health-related measurement instruments. The COSMIN checklist evaluates ten psychometric properties and consists of four possible answers: 'excellent', 'good', 'fair' and 'poor'. The 'Interpretability' box was filled in for every article and scored based on the number of questions that could be answered with 'yes' (1 or 2 = poor; 3 or 4 = fair; 5 or 6 = good; 7 = excellent). A general score for the methodological quality was provided for every individual psychometric property for every measure or scale by taking the lowest score from every box[32] (Table 4). General information for every study and measure or scale was extracted with the help of the 'Generalizability' box of the COSMIN checklist and compiled in Table 3.[32]

Results

Identification of measures or scales evaluating the physical activity level or activity pattern

The systematic literature search identified 919 articles. After exclusion of 717 articles based on the criteria mentioned above, 202 articles were included for full text reading. Full-text reading led to the exclusion of another 151 articles (Figure 1).

During full-text reading, 15 unique, relevant measures or scales evaluating the physical activity level or activity pattern of patients with CFS/ME were identified (Table 2), but the psychometric properties of only three instruments were evaluated (Tables 3 and 4).

Critical appraisal of psychometric properties of included measures or scales

Chronic fatigue syndrome – activity questionnaire

The Chronic fatigue syndrome – activity questionnaire (CFS-AQ) was used by Scheeres et al. to measure activities performed in the previous two weeks by patients with CFS.[33] The measure consists of four subscales: physical activity (four items), rest (four items), using aids (one item) and social activity (one item). The 10 items are scored on a four-point Likert scale. The time to complete the questionnaire ranged from five to seven minutes. Scheeres et al. described that this newly developed questionnaire has good *internal consistency* (Cronbach's alpha = 0.73) and *test–retest reliability* (Spearman's rho = 0.72). Although

Table 2 Characteristics of included measures or scales

Measures or scales	Goal	Refs.
Activity monitor	To measure physical activity	[6,15,16,26,35–37,40,43,49–66,80–82,86]
Activity Record (ACTRE)	To measure physical activity	[43,44]
Activity Pattern Inventory (API)	To measure the usual activities performed on a typical day	[33]
Atherosclerosis Risk in Communities (ARIC) Baecke Physical Activity Questionnaire	To assess habitual leisure and occupational physical activities	[45–48]
Checklist Individual Strength (CIS)	To measure physical activity	[6,36,37,49–51,57,60–62,64,65,67–69,81,82,86]
Chronic Fatigue Syndrome-Activities Questionnaire (CFS-AQ)	To measure physical activity	[33]
Dartmouth Primary Care Cooperative Research Network functional health assessment charts/World Organization of General Practice/Family Physicians (COOP/WONCA Charts)	To measure physical activity	[70]
Diary and Self Observation List	To measure physical activity	[33,35,43,53,71–74]
Godin Leisure-Time Exercise Questionnaire	To measure physical activity	[75]
Intelligent Device for Energy Expenditure and Activity	To measure physical activity	[71]
International Physical Activity Questionnaire-Short Form (IPAQ-SF)	To measure physical activity	[33,35,76]
Older Adult Exercise Status Inventory (OA-ESI)	To measure physical activity	[77]
Paffenbarger Physical Activity Questionnaire	To measure physical activity	[78]
Physical Activity Index of College Alumnus Health Questionnaire	To measure physical activity	[79]
Visual Analogue Scale (VAS) Daily Physical Activity Level	To measure physical activity	[6,83]

Table 3 Characteristics and COSMIN rating of included studies

Study	Population and pathology	Measures or scales	Psychometric qualities and methodological quality	
Meeus et al. (2011)	CFS (*n* = 56) 41.09 years SD 9.51 Range 20–62 years ♀: *n* = 56 (100%) *Disease duration* 93.61 months SD 78.41 months Range 6–360 months	IPAQ-SF	Internal consistency	Fair
			Criterion validity	Good
Scheeres et al. (2009)	CFS (*n* = 226) 37 years SD 11.3 Range 15–68 years ♀: 167 (74%) *Disease duration* 5 years Range 2–32 years	CFS-AQ	Internal consistency	Poor
			Test–retest reliability	Poor
			Criterion validity	Fair
		API	Criterion validity	Fair
		IPAQ-SF	Criterion validity	Fair

the internal consistency and test–retest reliability appear to be adequate, insufficient information about the research methodology was provided. Methodological quality of the study by Scheeres et al. for evaluating the internal consistency and test–retest reliability is therefore rated as poor by the COSMIN checklist.[33]

Criterion validity was evaluated by calculating the correlations between the three measures or scales CFS-AQ, Activity Pattern Interview (API) and International Physical Activity Questionnaire – Short Form (IPAQ-SF), and continuous scores of the activity monitor, a frequently used measure to objectively evaluate daily physical activity.[6,15,16,26,35,36,40,43,49–66,80–82]

The mean daily physical activity score of CFS patients was calculated based on 12 days actography to define an activity monitor typology (passive/fluctuating active). Patients scoring zero or one days of the 12 measured days above a reference score were defined as 'passive'. Patients scoring two or more days above a reference score were defined as 'fluctuating active'.

Logistic regression analyses were performed with the CFS-AQ and IPAQ-SF and activity monitor typology as dependent variable to predict the probability that a person with CFS is active, according to the activity monitor typology. The obtained predicted probability scores led to the development of a dichotomous outcome scale of activity level (active/passive) for the CFS-AQ and IPAQ-SF. Receiver operating characteristics (ROC) curve was calculated to identify the CFS-AQ's sensitivity and specificity. *Sensitivity* is the number of passive patients identified as being passive, while *specificity* is the number of active patients identified as being active. The best cut-off point

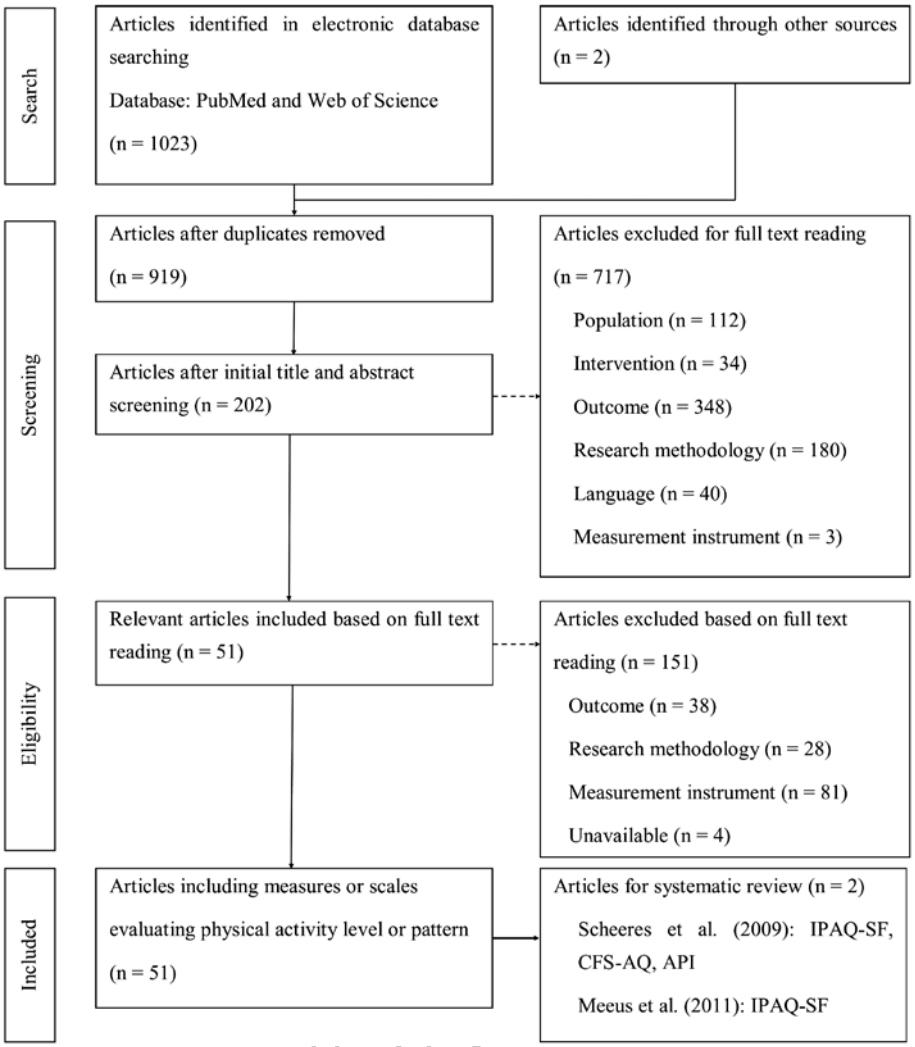

Figure 1 Flowchart of search strategy
Notes: CFS-AQ = Chronic Fatigue Syndrome – Activity Questionnaire; API = Activity Pattern Interview; IPAQ-SF = International Physical Activity Questionnaire – Short Form

for the CFS-AQ is 0.73 with a sensitivity of 64.6% and specificity of 65.2%. Area under the curve (AUC) calculated the CFS-AQ's validity. The AUC was 0.710, which means that the validity of the CFS-AQ is higher than the API, but lower than the IPAQ-SF. The CFS-AQ correlated moderately with the continuous scores of the activity monitor (Spearman's rho = 0.41).[33]

Methodological quality of the study for evaluating the criterion validity was found to be fair by the COSMIN checklist.[33] Scheeres et al. concluded that the CFS-AQ has no added value compared to the IPAQ-SF or the API.[33] No other studies evaluating the CFS-AQ were found.

Activity pattern interview

The API is an interview which identifies the usual activities performed on a typical day. During the interview, three relevant topics are questioned: routine pattern of activities, amount of time laying or sitting the day before, the number of times leaving the house during a day and practising an (un)paid job or not. Based on the answers on these three topics, the interviewer classified the person as 'active' or 'passive'. The routine pattern was investigated by questioning the day of yesterday as detailed as possible. When the day of yesterday was not a typical day, another day of the past week was used to minimize recall bias. The average time to complete the interview was 10 minutes. To produce valid results, experience in CFS and training in using the interview is recommended.[33]

The psychometric properties of the API were evaluated by Scheeres et al. and, as described earlier, the instrument was compared to the CFS-AQ and IPAQ-SF to identify the most suited measure or scale to evaluate the daily physical activity level of patients with CFS. More specifically, all instruments were evaluated on their capability to correctly classify a patient as (fluctuating) active based on activity monitor typology.[33]

Criterion validity was evaluated by calculating correlations between the API and continuous activity monitor scores, but only weak correlations were found (Spearman's rho = 0.27). ROC curve was calculated to identify the API's sensitivity and specificity. The sensitivity of the API was 52.3% and specificity was 75.8%. The API had an AUC of

Table 4 Methodological quality for every psychometric property for every measure or scale based on COSMIN checklist

Measure or scale	Internal consistency	Reliability: relative measures	Measurement error: absolute measures	Contentvalidity	Structural validity	Hypotheses testing	Cross-cultural validity	Criterion validity *(including AUC, sensitivity and specificity)*	Responsiveness	Interpretability
CFS-AQ Scheeres et al. (2009)	Poor	Poor						Fair		Poor
API Scheeres et al. (2009)								Fair		Poor
IPAQ-SF Scheeres et al. (2009)								Fair		Poor
Meeus et al. (2011)	Fair							Good		Poor

0.643, which was smaller than the validity of the CFS-AQ (0.710) and the IPAQ-SF (0.711). Methodological quality of the study by Scheeres et al. for evaluating the criterion validity was found to be fair by the COSMIN checklist[33] No other studies evaluating the API in patients with CFS/ME were found.

International physical activity questionnaire-short form

The International Physical Activity Questionnaire-Short Form (IPAQ-SF) is a self- or telephone-administered measure which evaluates health-related physical activity. The instrument was developed in 1996 by the 'International Consensus Group of Physical Activity Management' and validated in twelve countries by Craig et al.[34] Four long and four short versions of the instrument are available. The use of a short self-administered version with persons with CFS was suggested, because these patients often experience cognitive impairments.[35]

The IPAQ-SF consists of nine items and gathers information on the time spent walking, the performance of moderate and vigorous physical activity and the minutes spent sitting on weekdays during the past seven days.[33,35] Patients also have to rate how many days and how many minutes they spent per specific activity category. The amount of Metabolic Equivalents (METs)-minutes is calculated for all categories by multiplying the amount of minutes with 1.3 (sitting), 3.3 (walking), 4 (moderate physical activity) or 8 (vigorous physical activity).[35] Four subscale scores and one total score can be calculated by adding the METs-minutes of the last three categories together.[33,35] The time to complete the questionnaire ranged from five to seven minutes.[33]

The *internal consistency* was evaluated in a study of Meeus et al. in a population of patients with CFS. Cronbach's alpha was calculated for the categories walking, moderate and vigorous activities and was 0.337.[35] Methodological quality of this study was found to be fair by the COSMIN checklist.[35]

The *criterion validity* was evaluated by two studies. Scheeres et al. calculated correlations between the CFS-AQ, API and IPAQ-SF and the continuous activity monitor scores, as mentioned earlier.[33] The logistic regression analysis and calculation of predicted probability scores were also performed with the IPAQ-SF. ROC calculated the sensitivity and specificity. The best cut-off point for the IPAQ-SF is 0.67 with a sensitivity of 70.1% and specificity of 62.7%. The AUC was 0.711 and the IPAQ-SF had a greater validity than the API and CFS-AQ. The IPAQ-SF and continuous activity monitor scores had a weak correlation (Spearman's rho = 0.33).[33] Methodological quality of the study by Scheeres et al. for evaluating the criterion validity was found to be fair by the COSMIN checklist.[33]

Meeus et al. evaluated the *criterion validity* by comparing the IPAQ-SF with an activity monitor and an activity

diary.[35] METs-minutes spent per activity category (sedentary, moderate and vigorous activity) were the outcomes that were compared between the three measures. Spearman's rho varied between 0.282 and 0.426 ($p = 0.05$) indicating only weak correlations. Furthermore, the weak significant correlations were especially found in the moderate and vigorous activities. These were found to be irrelevant, because CFS patients rarely perform such activities. No correlations were found in the sedentary activities, which are the ones CFS patients perform the most.[35] Methodological quality of the study by Meeus et al. for evaluating the criterion validity was found to be good by the COSMIN checklist.[35]

Discussion

The aim of this review was twofold. First, scientific literature was systematically reviewed for currently used measures or scales evaluating the physical activity level or pattern in patients with CFS/ME. The systematic literature search identified 51 studies and a total of 15 different unique measures or scales. Second, the methodologies of studies evaluating the psychometric properties of identified measures or scales in a population with CFS/ME were critically appraised by use of the COSMIN checklist.[31,32] It was remarkable that, despite the high number of available instruments, only two studies evaluated the psychometric properties of three different measures in patients with CFS: the CFS-AQ, API and IPAQ-SF.[33,35] When listing all measures or scales identified by the literature search, activity monitors were found to be the most frequently used ($n = 29$) and are often seen as the gold standard to compare other measures or scales evaluating the perceived physical activity level or activity pattern to, such as self-report measurements.[6,15,16,26,35,36,39,40,43,49–66,80–82]

Based on the critical appraisal of the two studies evaluating the psychometric properties of the CFS-AQ, IPAQ-SF and API, both studies used an activity monitor to evaluate the criterion validity of the CFS-AQ, API and IPAQ-SF.[33,35] The research methodologies of the studies of Scheeres et al. and Meeus et al. were rated 'fair' and 'good', respectively, on the COSMIN checklist for evaluating the criterion validity and it can be concluded that these three measures are equally valid or equally invalid, given the lack of studies evaluating the psychometric properties of these activity monitors in patients with CFS/ME.

All three instruments can be used to measure the perceived physical activity level in daily life in CFS patients but have a low correlation with the actual activity level measured by an activity monitor.[33] The validity, tested with the area under the curve, of the CFS-AQ and IPAQ-SF (0.710 and 0.711) was slightly higher than the API (0.643). Some experience with CFS and training in performing the interview is enough to produce equally valid results to the self-reporting questionnaires IPAQ-SF and CFS-AQ.[33]

If a patient's activity pattern needs to be determined, the API could be more practical to use in the work field, because it has a dichotomous outcome (active/passive). The CFS-AQ and IPAQ-SF on the other hand, solely measure the activity level and the results have to be transformed to a dichotomous outcome by use of complicated formulas. However, a high number of false predictions by all three measures were found when compared to an activity monitor. If patients are incorrectly identified as being active (scoring two or more days above a reference score) or passive (scoring zero or one days of the twelve measured days above a reference score) according to an activity monitor typology in clinical practice, they could receive inappropriate treatment which could lead to more functional and participation restrictions.[85] Future research addressing this problem is recommended.

Considerations

The CFS-AQ, API and IPAQ-SF are all self-reported measures and consequently assess a patient's perception of daily performed physical activities.[4] Self-reports might not be highly related to the actual, objectively measured, daily life activity level as measured with activity monitors.[4,9,35,36,42] A previous study by Vos-Vromans et al. found discrepancies between perceived daily activities and objectively measured daily activities in patients with CFS; however, the cause of this discrepancy in patients with CFS needs to be further investigated.[37] In patients with chronic low back pain (CLBP), this discrepancy was associated with the presence of depressive symptoms influencing a patient's perception of their activity level,[87] that could lead to the assumption that mood changes in patients with chronic conditions influence the perceived activity level. Activity monitors on the other hand are known to be reliable and valid measures or scales to objectively evaluate a patient's activity level in the general population,[38] but their psychometric properties have not yet been evaluated with patients with CFS/ME.[4,8,35–38] The reliability and validity depend on the device, population and the studied activity behaviour.[29,35,39] First of all, the optimal place of attachment has not been established. The place of an activity monitor on the body influences its output and activity monitors worn on the lower body tend to underestimate activities of the upper body and vice versa.[36,38,39] Since patients with CFS/ME perform mostly sedentary activities, the place of attachment that provides the most accurate results of their performed physical activities needs to be determined.[40] Secondly, it is unknown when and for how long the activity monitor needs to be worn to obtain sufficient valid information for an accurate representation of a patient's activity level. In patients with chronic pain, it is recommended to include more than three days, because they have large between-day variations in physical activity and need periods of rest between activities.[84] Patients with CFS/ME also often have a fluctuating activity level, therefore inclusion of more than three assessment days and at least one weekend day can be useful to have an accurate representation of a patient's activity level. Third,

the influence of an activity monitor on the behaviour of CFS/ME patients is also unknown. Some patients engage in reactive behaviour, which means changing their normal physical activity pattern when consciously wearing an activity monitor.[40] Because information about the actual daily activity level in patients with CFS/ME is useful, evaluation of the psychometric properties of activity monitors and development of a protocol encompassing clear instructions on the place of attachment, duration of measurement etc. are necessary in order to obtain high quality results.

The discrepancy between the objective measurement and subjective perception of a patient's performed physical activities is an important topic for further investigation. Patients with CFS/ME, classified as active based on self-report measurement, may have a tendency to overestimate themselves due to the presence of PEM after performing many or intensive activities. On the other hand, patients classified as being passive are hypothesized to have a tendency for underestimation. They avoid most activities causing PEM, but the performance of other sedentary or light activities, such as cleaning, cooking, walking during household activities, washing and doing laundry,[84,88] will be performed, resulting in a similar activity level as active patients,[15] as found by Huijnen et al.[41] Measurement of the objectively measured physical activity level indicated that there were no significant differences between the avoidant group and persistent group with CLBP.[41] Vos-Vromans et al. also found no discrepancies between the actual activity level of passive and relatively active patients with CFS established by an activity monitor, but discrepancies were found between the perceived and actual physical activity level[37] and clinical practice should take this discrepancy into account when working with patients with CFS/ME.

Implications

Based on the evaluation of all measures or scales, their psychometric properties and further remarks, none of the three measures or scales should be used in isolation and training in performing the API is necessary to evaluate the activity level and pattern of activity in a population with CFS.

Future research is needed to further evaluate the reliability and validity of the IPAQ-SF, CFS-AQ and API and activity monitors. The systematic literature search identified fifteen unique measures evaluating physical activity in patients with CFS/ME of which the psychometric properties are not or insufficiently known. It is therefore recommended to first evaluate the psychometric properties of these measures, because they could potentially be appropriate for patients with CFS/ME. If psychometric properties are insufficiently robust, then perhaps new measures or scales to assess the activity level in a population with CFS/ME should be developed. Such measurements would need to have good psychometric properties, be short and easy to administer. Recall over a long period of time should be avoided, due to the possible presence of cognitive impairments. The questions and answers ought to be simple without the possibility of subjective interpretation. Since patients with CFS/ME mostly perform sedentary and light activities, these should be the instrument's focus.[35] Because ambulatory monitoring assesses the physical activity pattern more accurately than a measure using retrospective self-report, Meeus et al. suggest the development of a kind of activity diary with daily registration, which minimizes recall bias as previously discussed.[16,35] According to Wickel et al., self-report measures where the type, amount and intensity of physical activity can be recorded are the most used to measure physical activity levels.[36] The more details available on performed daily activities, the more accurate the physical activity level or pattern can be determined and false predictions can be prevented.

Moreover, Jason et al. state that solely looking at the total daily activity might not be enough to differentiate between patients with CFS/ME and healthy controls, but examination of the variability of their activity pattern over time is necessary.[22] The ability to map activity patterns would be a useful improvement for clinical practice, because patients with CFS/ME often have an imbalance between rest and activity and do not spread their activities equally during the day.[4,6,15] Mapping of a patient's activity pattern could lead to better understanding their problems and origin of their complaints, which would ultimately lead to better management and rehabilitation.[35]

Limitations

This systematic review has several limitations. First, although the research methodology was specified in advance, the protocol was not published.

Second, both screening phases of the systematic literature search were performed by two independent reviewers and a third if consensus could not be reached between the first two. However, an update of the systematic literature search was performed from March 2014 until October 2016 by two different reviewers than the initial literature search, which could have led to a slightly different selection. Nevertheless, the final supervision was continuously performed by the last author.

The literature search was performed in two electronic databases. Searches in additional databases could have generated additional relevant studies. Restricting the inclusion criteria to English- and Dutch-language publications could also have limited the results.

The quality of the research methodology of the studies varied. One patient population was smaller than 100 participants which, according to the COSMIN-checklist, is insufficient for evaluating the psychometric properties of measures or scales. The other publication provided insufficient information about its research methodology and is therefore automatically assigned with the lowest score. However, if the research methodology was performed accurately but reported poorly, this could have led to the underestimation of the measurement's qualities.

Conclusion

This systematic review identified 15 unique and relevant measures or scales used in patients with CFS/ME to evaluate the physical activity level and pattern, but the psychometric properties of only three measures or scales were evaluated in patients with CFS/ME: the CFS-AQ, API and IPAQ-SF. Based on the critical appraisal of their psychometric properties, it can be concluded that none of the three unique measures or scales are optimal to evaluate the activity level or pattern in patients with CFS/ME. Their psychometric properties have been insufficiently evaluated; therefore, their results should be interpreted with caution when used. The results of this systematic review clearly indicate that more research is necessary to further evaluate the psychometric properties of existing measures or scales and it is recommended to evaluate the validity and use of activity monitors for the population of patients with CFS/ME.

Acronyms

ADL	Activities of Daily Living
API	Activity Pattern Interview
AUC	Area Under The Curve
CBT	Cognitive Behavioural Therapy
CFS	Chronic Fatigue Syndrome
CFS-AQ	Chronic Fatigue Syndrome – Activity Questionnaire
CLBP	Chronic Low Back Pain
COSMIN	Consensus-based Standards for the selection of health Measurement Instruments
EE	Estimated Energy Expenditure
GET	Graded Exercise Therapy
IPAQ-SF	International Physical Activity Questionnaire-Short Form
ME	Myalgic Encephalomyelitis
MET	Metabolic Equivalent
PEM	Post-Exertional Malaise
PRISMA	Preferred Reporting Items for Systematic reviews and Meta-Analyses
ROC	Receiver Operator Characteristics
TEE	Total Energy Expenditure
QOL	Quality Of Life

Device status

The manuscript submitted does not contain information about medical device(s).

Disclosure statement

No potential conflict of interest was reported by the authors.

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