



Short communication

Paediatric short fatigue questionnaire, a 4-item fatigue questionnaire for children

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ABSTRACT

Objective: To investigate whether a paediatric Short Fatigue Questionnaire (pSFQ) assesses a valid construct for subjective fatigue, to assess its psychometric properties and provide a cut-off score for severe fatigue in children. **Methods:** The pSFQ consists of 4 items from the Checklist Individual Strength-8 (CIS-8). Data of previous studies using the CIS-8 were used to assess the pSFQ in healthy children ($n = 316$), children with chronic fatigue syndrome ($n = 173$), and children with a chronic disease ($n = 442$). All children were 12–18 years old. Confirmatory factor analysis (CFA) was performed, followed by Cronbach alpha's to investigate internal consistency, and Spearman's correlations to assess construct validity. With ROC analysis, we determined a cut-off score for severe fatigue and provide normative data on the pSFQ for children with and without a chronic disease. **Results:** CFA confirmed a one-factor model in the pSFQ representing subjective fatigue. Cronbach's alpha ranged from good to excellent (0.84–0.94), as did construct validity (-0.76 and -0.87 for correlation with two other fatigue measurements). ROC analysis delivered a cut-off score of ≥ 21 , with a sensitivity of 93.9% and specificity of 96.2% for severe fatigue. Normative data for children with and without a chronic disease showed similar patterns compared to other fatigue questionnaires. **Conclusion:** The pSFQ is a practical and reliable screening instrument for severe fatigue in children with and without a chronic disease, and effectively reduces the questionnaire length with 50% compared to the conventional CIS-8.

1. Introduction

Severe fatigue is a debilitating symptom, leading to decreased daily life participation in children with and without a chronic disease [1,2]. Fatigue is defined by Thomas (2018) as 'extreme tiredness resulting from physical or mental exertion or illness', and as a subjective or perceived symptom [3]. Therefore, it is important to be able to screen for severe fatigue using patient-reported outcomes. Several assessment measurements exist for fatigue, but for screening purposes in paediatric clinical practice, it is important to keep the burden of questionnaires to a minimum and questionnaires as short as reasonably possible. For this purpose, the checklist individual strength (CIS-8) is a valid and reliable instrument with the additional advantage that, using a cut-off, it is able

to distinguish between 'normal' levels of adolescent fatigue and severe, debilitating fatigue [4–6]. For adults, this CIS-8 (containing eight questions) has been shortened into the Short Fatigue Questionnaire (SFQ), which contains only four of the eight items [7]. The SFQ proves to be a reliable and valid way to measure subjective fatigue in adults and also uses a cut-off. However, as adolescents are generally more fatigued than adults, influenced by hormonal changes during puberty, and new psychological, social and academic challenges, a different cut-off is needed for adolescents than for adults [8–11]. If the paediatric SFQ (pSFQ) shows similar psychometric properties and validity in children as the SFQ in adults, its use will be preferred over the CIS-8 and it will provide a very short screening instrument for severe fatigue. Only the children who report severe fatigue may then be offered additional

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questionnaires in order to gain more insight into this fatigue and its dimensions, for example using the PedsQL multidimensional fatigue scale [12].

In this study, we aim to investigate whether the pSFQ shows one construct for subjective fatigue, and to assess its psychometric properties. We also aim to provide normative data for the pSFQ with and without a chronic disease and to determine a cut-off score for the presence or absence of severe fatigue.

2. Methods

2.1. Study population

In order to answer our research questions, we used all data samples of children (12–18 years old) from previous studies on chronic fatigue. From the studies by van de Putte et al. [13], Nijhof et al. [2], and Vroegindewij et al. [14], we derived a norm population (healthy sample; $n = 316$) and a sample with Chronic Fatigue Syndrome (CFS; $n = 173$). From the PROactive cohort study [15], a sample with various chronic conditions was derived ($n = 442$; descriptive data available via Dataverse NL: <https://doi.org/10.34894/FXUGHW>). All studies were IRB approved and all participants (and parents of children aged 12–15) provided consent for study participation and the use of their data for research purposes. For descriptives on all study samples, please see Supplementary Table S1.

2.2. Measures

2.2.1. SFQ

All children filled out the CIS-8, which also contains the four items of the proposed pSFQ. These four items are similar to the ones used in the adult SFQ: “I feel tired”, “I tire easily”, “I feel fit”, and “I feel physically exhausted”. All items are scored on a seven-point Likert scale, ranging from “yes, that is true” (1 point) to “no, that is not true” (7 points). Scores of the first, second and fourth item are reversed, so that higher scores reflect more fatigue here as well. The total score ranges from four to 28.

2.2.2. Other fatigue measures

Except for the studies by van de Putte et al. [13] and Nijhof et al. [2], all children filled out the PedsQL multidimensional fatigue scale (PedsQL MFS; $n = 467$) [12]. Children with a chronic disease also completed a five-point Likert scale on fatigue derived from the Health Behaviour in School aged Children (HBSC) study ($n = 441$) [16]. From the PedsQL MFS, we used the general fatigue subscale, containing six items and yielding a total score from 0 to 100, with lower scores indicating more fatigue. The HBSC question on fatigue ranges from “never” to “very often”.

2.3. Data analyses

Descriptive statistics were used to summarize the clinical characteristics of the subsamples (i.e., the healthy sample, CFS sample, and chronic disease sample). In line with Penson et al. [7] the item correlation matrix (ICM), Kaiser-meyer Oklin (KMO) test, and Bartlett’s test of sphericity were used to test the adequacy of the healthy sample data to perform confirmatory factor analysis (CFA). CFA was performed on the data to test our hypothesis of a one-factor model representing subjective fatigue. Good model fit was defined by a Root Means Square Error of Approximation (RMSEA) of <0.06 [7]. Normative data for the pSFQ was provided using descriptive characteristics for the CFS and chronic disease samples. Internal consistency of the pSFQ was determined by Cronbach’s alpha in the subsamples. Although there is no clear consensus on how to label Cronbach’s alpha values, we follow the terminology used in studies from leading science education journals that denoted values of 0.71 to 0.91 as good, 0.91 to 0.93 as strong, and 0.93

to 0.94 as excellent [17]. Construct validity of the SFQ was tested by Spearman correlations between the pSFQ, the PedsQL MFS general fatigue scale, and the HBSC question on fatigue. To derive a cut-off score for the pSFQ indicating severe fatigue, all children were first classified as (not) severely fatigued, with CIS-8 total scores of >39 indicating severe fatigue [8–10].

Next, Receiver Operating Characteristic (ROC) analysis were used. Area Under the Curve (AUC) values were evaluated, with values of <0.5 indicating poor classification performance. Higher AUC values are preferred, with values of 1 implying perfect classification performance [18]. Besides sensitivity and specificity, the Positive Predictive Value (PPV) and Negative Predictive Value (NPV) were computed, indicating the probability of correctly identifying a child as (not) severely fatigued in the total sample, and specifically in the CFS and chronic disease subsample. In the event of multiple cut-off scores performing equally, the final cut-off score was determined by computing the measure of accuracy (i.e., how correct the pSFQ identifies and excludes severe fatigue) for each cut-off, by dividing the number of correct assessments by the number of total assessments [19]. The cut-off score with an accuracy value closest to 1 would be preferred [19]. CFA was performed with the “lavaan” package in Rstudio version 1.3.1093, all other analyses were performed in SPSS version 26.0.0.1.

3. Results

3.1. Participants

We included 931 children between 12 and 18 years old (66% girls; mean age 15.4 ± 1.6 years; details per subsample in Supplementary Table S1). Of the 442 children with a chronic disease, there were 269 with an autoimmune disease, 60 with cystic fibrosis, 86 with a congenital heart disease, and 27 with a chronic kidney disease.

3.2. Factor analysis

The ICM, KMO test, and Bartlett’s test of sphericity all confirmed the adequacy of the healthy sample data for CFA. With CFA, the one-factor model was confirmed with excellent fit (Eigenvalue = 2.482; RMSEA = 0.000). All pSFQ items had factor loadings >0.75 , and explained 83.58% of the total variance. For the CFA scree plot, see Fig. 1.

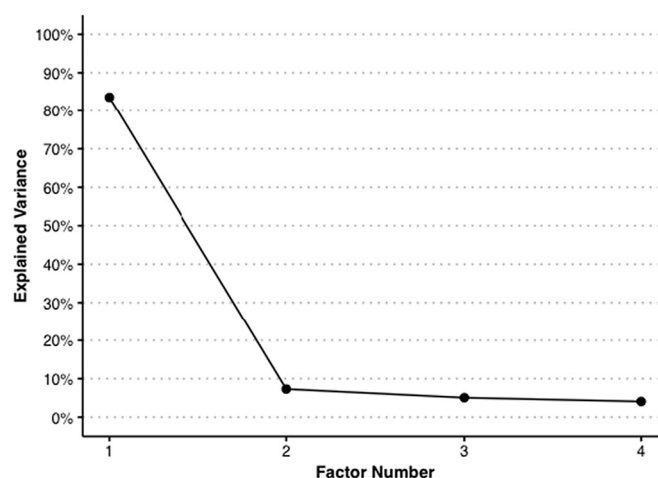


Fig. 1. Scree plot of the Confirmatory Factor Analysis.

Note. As the pSFQ consists of four items, four factors would theoretically be possible. However, Fig. 1 shows that one factor would be the most fitting solution, as this would explain 83.58% of the total variance.

3.3. Psychometric properties of the SFQ

In Table 1 we present normative data and Cronbach's alpha for all subsamples, which were all good to excellent (0.84–0.94). Spearman's correlation was -0.86 for the pSFQ and the PedsQL MFS general fatigue score ($p < 0.01$; $n = 467$), and -0.77 for the pSFQ and HBSC question on fatigue ($p < 0.01$; $n = 441$).

3.4. Cut-off score

ROC analysis yielded an AUC of 0.992, indicating that the pSFQ can distinguish excellently between children with ($n = 294$) and without severe fatigue ($n = 637$). The ROC curve is presented in Fig. 2. In Table 2 we present the sensitivity, specificity, PPV and NPV for the total sample, as well as the PPV and NPV values for the CFS and chronic disease subsamples. Two cut-off scores performed almost equally, namely cut-off scores ≥ 20 (sensitivity 96.6%, specificity 93.9%) and ≥ 21 (sensitivity 93.9%, specificity 96.2%). The accuracy values were respectively 0.948 and 0.955. This results in a slight preference for ≥ 21 as pSFQ cut-off score. With cut-off score ≥ 21 , the PPV and NPV values in the total sample indicated that 92% of the children identified as 'severely fatigued' were truly severely fatigued, and 97% of the children identified as 'not severely fatigued' were truly not severely fatigued.

4. Discussion

The 4-item pSFQ showed good psychometric properties in children, with good to excellent Cronbach alpha's. CFA confirmed that the pSFQ represents one underlying factor. Furthermore, we present a cut-off for severe fatigue of ≥ 21 with excellent sensitivity, specificity, and positive and negative predictive values.

Our results are in line with the results of Penson et al., showing good psychometric properties for the SFQ in adults [7]. These results were confirmed in childhood cancer survivors, of which 67 were under the age of 20 (3% of the total cohort) [20]. Besides those results, this is the first study to test the SFQ in children. As it is known that a different cut-off should be used for children than for adults to classify severe fatigue using the CIS-8, this stressed the need to redefine a cut-off for the pSFQ [8–10]. The excellent sensitivity and specificity found in our study provide a reliable cut-off, supported by predictive values that indicate that the probability of misclassifying a child as (not) severely fatigued are below 10% in the present study. It is important to minimize the risk of misclassification to avoid patient burden due to unnecessary medical care or delayed or even absent treatment.

A strength of this study is the inclusion of healthy samples, children

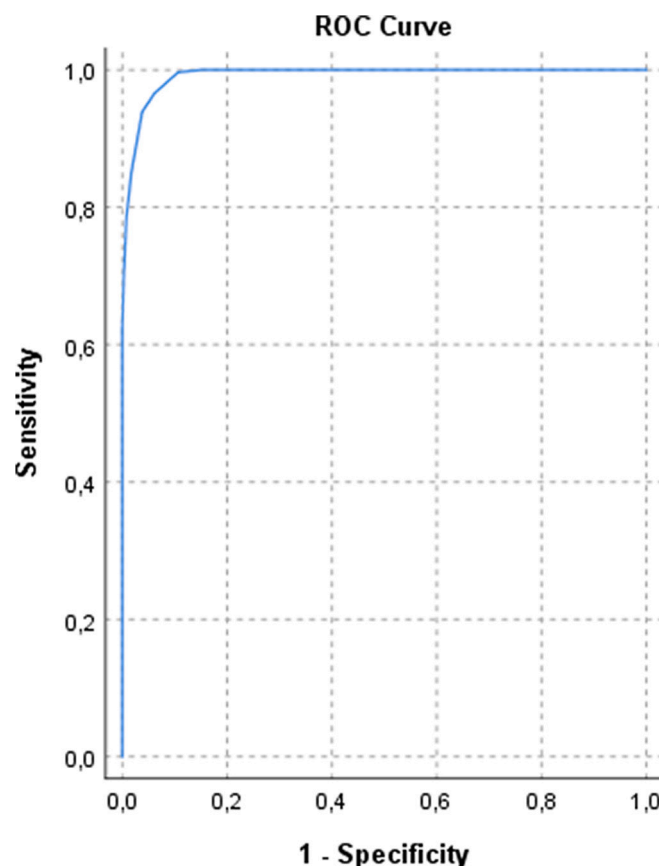


Fig. 2. ROC curve for pSFQ total score.

with CFS, and children with a chronic disease. This increases the generalizability of our results and the possibility of the use of the pSFQ in all these groups. The normative data of the pSFQ shows that, in line with earlier studies, children with a chronic disease are generally more fatigued than healthy children, but less fatigued than children with CFS [1,21,22]. We therefore believe our results are generalizable for children with and without a chronic disease, and powerful enough to sample in populations with various levels of fatigue. The sample size of the subsamples of children with a chronic disease prevents further analysis, which is a limitation. Children with chronic kidney disease however did show a trend to report lower scores than the healthy population, but this study was underpowered to determine significant differences between patient groups. Another limitation is that we do not know the test-retest error of this cut-off value. This would be of interest for future research, as fatigue is known to vary over time.

In clinical practice, it is important to distinguish between normal fatigue and severe, debilitating fatigue that impacts daily life participation. The latter should timely be diagnosed and treated [23,24]. Therefore, the pSFQ is an excellent choice as a short screening instrument. If severe fatigue is present, additional measurement tools, such as the PedsQL MFS, can be used to gain more insight in the different dimensions of fatigue [25].

5. Conclusions

The 4-item pSFQ is a useful and reliable screening instrument for severe fatigue for children with and without a chronic disease.

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jpsychores.2022.111130>.

Table 1

Population norms for the pSFQ in healthy children, children with CFS and children with a chronic disease.

Subsample	N	Mean \pm SD	25th	50th	75th	α
Healthy population	316	11.6 \pm 5.9	6.0	10.0	16.0	0.89
Children with CFS	173	24.9 \pm 4.0	23.5	26.0	28.0	0.84
Children with a chronic disease	442	14.5 \pm 7.6	7.0	14.0	21.0	0.92
Children with an autoimmune disease	269	15.1 \pm 7.6	8.0	15.0	22.0	0.91
Children with cystic fibrosis	60	15.6 \pm 8.0	7.0	16.0	23.0	0.93
Children with a congenital heart disease	86	12.8 \pm 6.4	7.0	17.3	23.3	0.88
Children with a chronic kidney disease	27	11.4 \pm 8.2	4.0	8.0	18.0	0.94

CFS = chronic fatigue syndrome; SD = standard deviation; α = Cronbach's alpha.

Table 2

Overview of pSFQ cut-off scores with corresponding sensitivity, specificity, PPV and NPV values.

Cut-off score \geq	Total sample (N = 931) ^a				CFS sample (n = 173) ^b		Chronic disease sample (n = 442) ^c	
	Sensitivity	Specificity	PPV	NPV	PPV	NPV	PPV	NPV
18	100.00%	84.80%	75.19%	100%	98.09%	100%	68.94%	100%
19	99.70%	89.30%	81.16%	99.82%	98.71%	94.44%	75.34%	99.66%
20	96.60%	93.90%	87.93%	98.36%	99.33%	75.00%	84.25%	98.73%
21	93.90%	96.20%	92.00%	97.15%	99.31%	64.29%	88.89%	97.85%
22	85.00%	98.30%	95.79%	93.43%	99.24%	42.86%	94.00%	95.03%
23	78.60%	99.20%	97.47%	90.92%	99.18%	35.29%	96.67%	93.06%

CFS = Chronic Fatigue Syndrome; PPV = Positive Predictive Value; NPV = Negative Predictive Value. Percentage that is severely fatigued according to the CIS-8 total score = ^a 32% of the total sample; ^b 89% of the CFS sample; ^c 25% of the chronic conditions sample.

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Declaration of Competing Interest

The authors have no conflicts of interest relevant to this article to disclose.

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