

The PedsQL™ Multidimensional Fatigue Scale in Pediatric Rheumatology: Reliability and Validity

JAMES W. VARNI, TASHA M. BURWINKLE, and ILONA S. SZER

ABSTRACT. *Objective.* The PedsQL™ (Pediatric Quality of Life Inventory™) is a modular instrument designed to measure health related quality of life (HRQOL) in children and adolescents ages 2–18 years. The recently developed 18-item PedsQL Multidimensional Fatigue Scale was designed to measure fatigue in pediatric patients and comprises the General Fatigue Scale (6 items), Sleep/Rest Fatigue Scale (6 items), and Cognitive Fatigue Scale (6 items). The PedsQL 4.0 Generic Core Scales were developed as the generic core measure to be integrated with the PedsQL Disease-Specific Modules. The PedsQL 3.0 Rheumatology Module was designed to measure pediatric rheumatology-specific HRQOL.

Methods. The PedsQL Multidimensional Fatigue Scale, Generic Core Scales, and Rheumatology Module were administered to 163 children and 154 parents (183 families accrued overall) recruited from a pediatric rheumatology clinic.

Results. Internal consistency reliability for the PedsQL Multidimensional Fatigue Scale Total Score ($\alpha = 0.95$ child, 0.95 parent report), General Fatigue Scale ($\alpha = 0.93$ child, 0.92 parent), Sleep/Rest Fatigue Scale ($\alpha = 0.88$ child, 0.90 parent), and Cognitive Fatigue Scale ($\alpha = 0.93$ child, 0.96 parent) were excellent for group and individual comparisons. The validity of the PedsQL Multidimensional Fatigue Scale was confirmed through hypothesized intercorrelations with dimensions of generic and rheumatology-specific HRQOL. The PedsQL Multidimensional Fatigue Scale distinguished between healthy children and children with rheumatic diseases as a group, and was associated with greater disease severity. Children with fibromyalgia manifested greater fatigue than children with other rheumatic diseases.

Conclusion. The results confirm the initial reliability and validity of the PedsQL Multidimensional Fatigue Scale in pediatric rheumatology. (J Rheumatol 2004;31:2494–500)

Key Indexing Terms:

HEALTH RELATED QUALITY OF LIFE
RHEUMATOLOGY

PEDIATRICS

FATIGUE
CHILDREN

Health related quality of life (HRQOL) measurement has progressively been integrated into clinical trials, clinical practice improvement initiatives, and healthcare services research^{1–3}. Although health status, functional status, and HRQOL are terms often used interchangeably, a metaanalysis suggests that a conceptual distinction between these terms is warranted⁴. Health status and functional status refer

to the physical functioning dimensions of the broader HRQOL construct. HRQOL additionally includes the psychosocial dimensions of emotional, social, and role functioning, as well as related constructs. A pediatric HRQOL instrument must be multidimensional, consisting at the minimum of the physical, mental, and social health dimensions delineated by the World Health Organization⁵.

The importance of measuring pain⁶ and functional status^{7–11} in pediatric rheumatology is now well accepted. However, HRQOL has emerged more recently as an essential health outcome in pediatric rheumatology^{12,13}. While the assessment of fatigue in adult rheumatology patients has received empirical attention^{14–17}, the multidimensional fatigue measurement construct in pediatric rheumatology has not been empirically reported, and is a more contemporary conceptualization in pediatric chronic health conditions¹⁸.

Pediatric HRQOL measurement instruments must be sensitive to cognitive development and include child self-report and parent proxy-report to reflect their potentially unique perspectives. Imperfect agreement between self and proxy report, termed cross-informant variance¹⁹, has been consistently documented in the HRQOL assessment of children with chronic health conditions and healthy children^{20,21},

From the Department of Landscape Architecture and Urban Planning, College of Architecture, Department of Pediatrics, College of Medicine, Texas A&M University, College Station, Texas; Department of Anesthesiology, University of Washington, Seattle, Washington; and Division of Pediatric Rheumatology, Children's Hospital and Health Center, Department of Pediatrics, University of California School of Medicine, San Diego, California, USA.

Supported by research grants from The Arthritis Foundation.

J.W. Varni, PhD, Department of Landscape Architecture and Urban Planning, College of Architecture, Department of Pediatrics, College of Medicine, Texas A&M University; T.M. Burwinkle, PhD, Department of Anesthesiology, University of Washington; I.S. Szer, MD, Division of Pediatric Rheumatology, Children's Hospital and Health Center, Department of Pediatrics, University of California School of Medicine.

Address reprint requests to Dr. J.W. Varni, Department of Landscape Architecture and Urban Planning, College of Architecture, Texas A&M University, 3137 TAMU, College Station, TX 77843-3137.

E-mail: jvarni@archone.tamu.edu.

Submitted November 17, 2003; revision accepted July 12, 2004.

including in rheumatology¹². Agreement has been found to be lower for internalizing problems (e.g., pain, depression) than for externalizing problems (e.g., walking, hyperactivity)^{20,21}. The demonstration of cross-informant variance and the general acceptance that HRQOL derives from an individual's perceptions¹ indicates an essential need in pediatric HRQOL measurement for reliable and valid child self-report instruments for the broadest age range possible. However, while self-report is considered the standard for measuring perceived HRQOL, it is typically parents' perceptions of their children's HRQOL that influences healthcare utilization²²⁻²⁴. Thus the imperfect agreement observed between child self-report and parent proxy-report supports the need to measure the perspectives of both the child and parent in evaluating pediatric HRQOL.

The PedsQL™ (Pediatric Quality of Life Inventory™) Measurement Model²⁵ was designed as a modular approach to measuring pediatric HRQOL, developed to integrate the relative merits of generic and disease-specific approaches. The PedsQL builds on a programmatic measurement instrument development effort by Varni and colleagues in pediatric chronic health conditions^{12,18,26-28}, including rheumatology^{8,29,30}, during the past 15 years. The PedsQL Measurement Model²⁵ emphasizes the child's perceptions. The items chosen for inclusion were derived from the measurement properties of the child self-report scales, while the parent proxy-report scales were constructed to directly parallel the child self-report items.

Given that the PedsQL Measurement Model integrates generic core scales and disease-specific modules into one measurement system, the PedsQL 4.0 Generic Core Scales were specifically designed for application in both healthy and patient populations^{31,32}. The PedsQL 3.0 Rheumatology Module was designed to measure HRQOL dimensions specifically tailored for pediatric rheumatology¹². The PedsQL Multidimensional Fatigue Scale is a more recently developed instrument, designed to measure child and parent perceptions of fatigue in pediatric patients¹⁸. In the development of the PedsQL Multidimensional Fatigue Scale, multidimensional constructs were derived from reviews of both the adult and pediatric fatigue literature, and integrated into the PedsQL Measurement Model. The acute version (7-day recall period) of the PedsQL Multidimensional Fatigue Scale was first field tested in pediatric cancer patients¹⁸. The standard version (one-month recall period) has not been previously field tested.

We describe the measurement properties of the standard version of the PedsQL Multidimensional Fatigue Scale in pediatric rheumatology, reporting on initial reliability and validity in a diverse sample of children with rheumatic diseases.

MATERIALS AND METHODS

Rheumatology sample. Participants were children ages 6 to 18 years (n =

163) and parents of children ages 2 to 18 years (n = 154), with 183 families accrued overall. For 134 children ages 6 to 18 years, both child self-report and parent proxy-report were available. Families were recruited from the pediatric rheumatology clinic at Children's Hospital and Health Center, San Diego. The PedsQL was self-administered for parents and for children ages 8 to 18 and interviewer-administered for children ages 6 and 7. The measures were administered in 2 languages, English (n = 173, 94.53%) and Spanish (n = 3, 1.64%; missing = 7, 3.83%).

For all forms combined, the average age of the 144 girls (78.7%) and 39 boys (21.3%) was 12.02 years (SD 4.15) with a range of 2 to 18 years. For child self-report, the average age of the 131 girls (80.4%) and 32 boys (19.6%) was 13.05 years (SD 3.20) with a range of 6 to 18. The sample was heterogeneous with respect to race/ethnicity, with 75 (41.0%) White non-Hispanic, 44 (24.0%) Hispanic, 6 (3.3%) Black non-Hispanic, 9 (4.9%) Asian/Pacific Islander, 2 (1.1%) American Indian or Alaskan Native, 4 (2.2%) other, and 43 (23.5%) missing. Mean socioeconomic status (SES) was unavailable for this sample. The sample included children with dermatomyositis (n = 11, 6.01%), juvenile rheumatoid arthritis [JRA; n = 54 (29.51%); 11 (6.0%) pauciarticular, 29 (15.8%) polyarticular, 14 (7.7%) systemic], systemic lupus erythematosus (SLE; n = 16, 8.74%), juvenile fibromyalgia (FM; n = 29, 15.85%), spondyloarthritis (n = 18, 9.84%), and other rheumatic diseases (n = 50, 27.32%; missing = 5, 2.73%). Patients and parents completed the PedsQL during rheumatology clinic visits.

Healthy sample: Multidimensional Fatigue Scale. Subjects were 102 families of healthy children ages 2 to 18 as described¹⁸. Healthy children ages 5 to 18 (n = 52) and parents of healthy children ages 2 to 18 (n = 102) were administered the PedsQL Multidimensional Fatigue Scale Acute Version by telephone. This sample was accrued from a list of patients who had attended an orthopedic clinic for broken bones or fractures 6 months prior to assessment with the PedsQL, and who had been identified by the clinic nurse as having "returned to health" (e.g., no current problems due to their orthopedic injury). The average age of the 69 boys (67.7%) and 30 girls (29.4%; missing = 3, 2.9%) was 8.88 years (SD 10.98). For child self-report, the average age of the 40 boys (76.9%) and 11 girls (21.2%; missing = 1, 1.9%) was 10.40 years (SD 14.45). The sample was heterogeneous with respect to race/ethnicity, with 28 (27.45%) White non-Hispanic, 47 (46.08%) Hispanic, 6 (5.88%) Black non-Hispanic, 2 (1.96%) Asian/Pacific Islander, 1 (0.98%) American Indian or Alaskan Native, 14 (13.73%) other, and 4 (3.92%) missing. Mean SES was unavailable for this sample. Chi-squares and t tests between this healthy sample and the rheumatology sample indicate that there were more subjects who were White non-Hispanic in the rheumatology sample than there were in the healthy sample, more males in the healthy sample and more females in the rheumatology sample, and the rheumatology sample was significantly older than the healthy sample.

Measures

PedsQL Multidimensional Fatigue Scale. The 18-item PedsQL Multidimensional Fatigue Scale comprises 3 subscales: (1) General Fatigue (6 items, e.g., "I feel tired"; "I feel too tired to do things that I like to do"), (2) Sleep/Rest Fatigue (6 items, e.g., "I feel tired when I wake up in the morning"; "I rest a lot"), and (3) Cognitive Fatigue (6 items, e.g., "It is hard for me to keep my attention on things"; "It is hard for me to think quickly")¹⁸. The PedsQL Multidimensional Fatigue Scale was developed based on our research and clinical experiences in pediatric chronic health conditions, and the instrument development literature³³⁻³⁵, which consisted of a review of the extant literature on fatigue in adult and pediatric patients, patient and parent focus groups and individual focus interviews, item generation, cognitive interviewing, pretesting, and subsequent field testing of the new measurement instrument.

The format, instructions, Likert response scale, and scoring method are identical to the PedsQL 4.0 Generic Core Scales, with higher scores indicating better HRQOL (fewer problems or symptoms). The instructions for the standard version ask how much of a problem each item has been during the past one month. The standard version was utilized for this investigation.

The PedsQL Multidimensional Fatigue Scale comprises parallel child self-report and parent proxy-report formats. Child self-report includes ages 5–7 (young child), ages 8–12 (child), and ages 13–18 years (adolescent). Parent proxy-report includes ages 2–4 (toddler), 5–7 (young child), 8–12 (child), and 13–18 (adolescent). The parent proxy-report forms are parallel to the child self-report forms, and are designed to assess the parent's perceptions of their child's fatigue. The items for each of the forms are essentially identical, differing in developmentally appropriate language, or first or third-person tense. A 5-point Likert scale is utilized across child self-report for ages 8–18 and parent proxy-report (0 = never a problem; 1 = almost never a problem; 2 = sometimes a problem; 3 = often a problem; 4 = almost always a problem). To further increase the ease of use for the young child self-report (ages 5–7), the Likert scale is reworded and simplified to a 3-point scale (0 = not at all a problem; 2 = sometimes a problem; 4 = a lot of a problem), with each response choice anchored to a happy to sad faces scale^{29,30}. Parent proxy-report also includes the toddler age range (ages 2–4), which does not include a self-report form given developmental limitations on self-report for children younger than 5 years of age^{30,36}.

Items are reverse-scored and linearly transformed to a 0–100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0), so that higher PedsQL Multidimensional Fatigue Scale scores indicate better HRQOL (fewer symptoms of fatigue). This direct linear transformation does not affect the measurement properties of the scales, and is computed for ease of interpretation so that scores near 0 indicate poorer HRQOL (more fatigue) and scores near 100 indicate better HRQOL (less fatigue). Scale scores are computed as the sum of the items divided by the number of items answered (this accounts for missing data). If more than 50% of the items in the scale are missing, the Scale score is not computed³⁷. For this study, over 99.2% of child and parent respondents were included in the Scale score analyses for all instruments.

The PedsQL 4.0 Generic Core Scales. The 23-item multidimensional PedsQL 4.0 Generic Core Scales encompass: (1) Physical Functioning (8 items), (2) Emotional Functioning (5 items), (3) Social Functioning (5 items), and (4) School Functioning (5 items), and have been field tested in pediatric rheumatology¹². To create the Psychosocial Health Summary Score (15 items), the mean is computed as the sum of the items divided by the number of items answered in the Emotional, Social, and School Functioning Subscales.

PedsQL 3.0 Rheumatology Module. The 22-item multidimensional PedsQL 3.0 Rheumatology Module Scales encompass: (1) Pain and Hurt (4 items), (2) Daily Activities (5 items), (3) Treatment (7 items), Worry (3 items), and (4) Communication (3 items)¹². The format, instructions, Likert scale, and scoring method are identical to the PedsQL 4.0 Generic Core Scales, with higher scores indicating better HRQOL (fewer problems or symptoms). For the purposes of this investigation, only the Pain and Hurt and Daily Activities Scales were included.

Physician global assessment of disease severity. The physician global assessment of disease severity was provided by the director of pediatric rheumatology at the time of assessment with the PedsQL. The pediatric rheumatologist indicated her assessment of the patient's disease severity on a 100 mm visual analog scale, anchored by 0 (remission, no indication of disease) to 100 (severe disease activity).

Procedure

Potential subjects were identified through the clinic appointment schedule. The parents of the children identified as possible study participants were informed of the study after checking in for their appointment, but before being seen by the pediatric rheumatologist. Written parental informed consent and child assent were obtained. Parents and children completed the PedsQL instruments separately. One parent (65.57% mothers; 8.74% fathers; 1.64% other, 24.05% missing) completed the proxy-report version. A research assistant or specifically trained clinic personnel were available to answer questions regarding the parent self-administered instruments. A research assistant or specifically trained clinic personnel administered the PedsQL for the young child (ages 5–7), and was available to assist the self-

administered instrument for the child (ages 8–12) and adolescent (ages 13–18) after the instructions had been given and clarified. The average time needed to complete the PedsQL Multidimensional Fatigue Scale is estimated to be less than 5 minutes based on research that indicated that it takes roughly 4 minutes to administer the 23-item PedsQL 4.0 Generic Core Scales³¹. The average time needed to complete the PedsQL 4.0 Generic Core Scales and the PedsQL 3.0 Rheumatology Module together is estimated at 15 minutes for child self-report and 10 minutes for parent proxy-report, based on our experience administering these measures in the rheumatology clinic. This research protocol was approved by the Institutional Review Board at Children's Hospital and Health Center, San Diego.

Statistical analysis. Feasibility or practicality was determined from the percentage of missing values³⁸. Scale internal consistency reliability was determined by calculating Cronbach's coefficient alpha³⁹. Scales with reliabilities ≥ 0.70 are recommended for comparing patient groups, while a reliability criterion of 0.90 is recommended for analyzing individual patient scale scores^{40,41}.

Construct validity was primarily examined through an analysis of the intercorrelations among the PedsQL Multidimensional Fatigue Scale scores with the PedsQL 4.0 Generic Core Scales and the PedsQL 3.0 Rheumatology Module Pain and Daily Activities Scales. It was hypothesized that higher PedsQL Multidimensional Fatigue Scale scores (fewer problems or symptoms) would be correlated with higher Generic Core Total Scale scores (better overall HRQOL), based on the conceptualization of disease-specific symptoms as causal indicators of HRQOL¹. Correlation effect sizes are designated as small (0.10–0.29), medium (0.30–0.49), and large (≥ 0.50)⁴². Intercorrelations were expected to show medium to large effect sizes¹. It was further hypothesized that higher PedsQL Multidimensional Fatigue Scale scores (fewer problems or symptoms) would be correlated with higher PedsQL 3.0 Rheumatology Module Pain and Hurt Scale scores (fewer problems or symptoms) based on findings from the adult rheumatology empirical literature¹⁵. It was hypothesized that the intercorrelations between fatigue and pain would show large effects sizes. We further explored other intercorrelations for heuristic and hypothesis-generating purposes.

Construct validity was additionally explored utilizing the known-groups method. The known-groups method compares scale scores across groups known to differ in the health construct being investigated. In this study, PedsQL Multidimensional Fatigue Scale scores in groups differing in known health condition (healthy children and children with rheumatic diseases as a group) were computed^{43,44} using t tests. The data for the healthy group of children were derived from the initial field trial of the PedsQL Multidimensional Fatigue Scale¹⁸. We hypothesized that healthy children would report higher PedsQL Multidimensional Fatigue Scale scores (fewer fatigue symptoms) than pediatric patients with rheumatic diseases as a group. Finally, we explored whether there were different levels of fatigue among the different rheumatic diseases in the sample, and whether greater disease severity as assessed by the pediatric rheumatologist was associated with greater patient and parent reported fatigue.

Parent/child intercorrelations for the PedsQL Multidimensional Fatigue Scale were computed to examine cross-informant variance. Pearson product moment correlation coefficient effect sizes are designated as small (0.10–0.29), medium (0.30–0.49), and large (≥ 0.50)⁴². Intraclass correlations (ICC) are designated as ≤ 0.40 poor to fair agreement, 0.41–0.60 moderate agreement, 0.61–0.80 good agreement, and 0.81–1.00 excellent agreement⁴⁵. Parent/child concordance for the total score and the same subscale were expected to show medium to large effect sizes (agreement), but not so large that child and parent reports would be redundant.

Statistical analyses were conducted using SPSS for Windows⁴⁶. Response equivalence has been demonstrated across languages (English vs Spanish) for the PedsQL by examining the percentage missing data, floor and ceiling effects, and scale internal consistency across languages³¹. Therefore, responses were pooled across languages. Responses were also pooled across the age ranges for both self-report and proxy-report.

RESULTS

Missing item responses. To assess the feasibility or practicality of administration for the PedsQL Multidimensional Fatigue Scale, the percentage of missing values was calculated. For child self-report and parent proxy-report, the percentage of missing item responses was 0.4% and 0.8%, respectively.

Means and standard deviations. Table 1 presents the means and standard deviations of the PedsQL Multidimensional Fatigue Scale for children with rheumatic diseases as a group and the healthy children population group from our previous field trial¹⁸.

Internal consistency reliability. Internal consistency reliability alpha coefficients for the PedsQL Multidimensional Fatigue Scale across all ages are presented in Table 2. All the child self-report scales and parent proxy-report scales exceeded the minimum reliability standard of 0.70 for group

comparisons⁴⁰. Most scales approached or exceeded the reliability criterion of 0.90 recommended for analyzing individual patient scores^{40,41}.

Construct validity. The intercorrelations among the PedsQL Multidimensional Fatigue Scale, the PedsQL 4.0 Generic Core Scales Total Score, and the PedsQL 3.0 Rheumatology Module Pain and Daily Activities Scales are shown in Table 3. As anticipated, the correlations are in the medium to large effect-size range. Specifically, for child self-report, the Multidimensional Fatigue Scale Total Score correlated 0.87 with the Generic Core Scales Total Score (0.86 for parent proxy-report). As hypothesized, the Multidimensional Fatigue Scale Total Score correlated 0.68 and 0.58 with the Pain and the Daily Activities Scales for child self-report, respectively (0.70 and 0.52, respectively, for parent proxy-report). The individual scales also correlated in magnitude and direction consistent with the constructs being measured.

Table 1. Scale descriptives for PedsQL Multidimensional Fatigue Scale child self-report and parent proxy-report and comparisons with healthy children's scores. The standard version of the PedsQL Multidimensional Fatigue Scale has a one-month recall interval in the directions for both child self-report and parent proxy-report. The acute version of the PedsQL Multidimensional Fatigue Scale has a 7-day recall interval in the directions for both child self-report and parent proxy-report. Higher values equal fewer symptoms or problems. Effect sizes are designated as small (0.20), medium (0.50), and large (0.80).

Scale	No. of items	Rheumatology Sample			Healthy Sample			Difference	Effect Size	t
		N	Mean	SD	N	Mean	SD			
Child self-report										
Total fatigue	18	163	73.82	21.93	52	80.49	13.33	6.67	0.38	−2.07*
General fatigue	6	163	72.76	25.48	52	85.34	14.95	12.58	0.62	−3.38**
Sleep/rest fatigue	6	163	68.70	24.77	52	75.00	18.76	6.30	0.29	−1.67
Cognitive fatigue	6	163	79.84	22.65	52	81.14	17.43	1.30	0.06	−0.38
Parent proxy-report										
Total fatigue	18	152	72.53	21.11	102	89.63	11.38	17.09	1.05	−7.75***
General fatigue	6	153	69.10	24.45	102	89.30	13.33	21.20	1.12	−7.32***
Sleep/rest fatigue	6	153	68.55	24.47	102	88.86	14.72	20.32	1.04	−7.52***
Cognitive fatigue	6	152	79.71	21.60	102	90.72	15.15	11.01	0.60	−4.46***

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$.

Table 2. PedsQL Multidimensional Fatigue Scale internal consistency reliability for child self-report and parent proxy-report by age and summary score/subscale.

Scale	Age Group				Total Sample
	Toddler (2–4)	Young Child (6–7)	Child (8–12)	Adolescent (13–18)	
Child self-report					
Total fatigue	NA	0.89	0.95	0.96	0.95
General fatigue	NA	*	0.94	0.93	0.93
Sleep/rest fatigue	NA	*	0.84	0.89	0.88
Cognitive fatigue	NA	*	0.93	0.93	0.93
Parent proxy-report					
Total fatigue	0.88	0.88	0.95	0.97	0.95
General fatigue	0.79	0.85	0.92	0.93	0.92
Sleep/rest fatigue	0.86	0.84	0.89	0.92	0.90
Cognitive fatigue	0.79	0.90	0.95	0.97	0.96

* For the age group 6-7 years, there were only 2 children who completed the PedsQL, so alpha coefficients are presented for total score only.

Table 3. Association between PedsQL Multidimensional Fatigue Scale scores and physician global assessment of disease severity. N = 175; lower PedsQL Multidimensional Fatigue Scale scores indicate more fatigue symptoms or problems.

PedsQL Fatigue Scales	Disease Severity Correlations*
Child self-report	
Total fatigue	-0.39
General fatigue	-0.38
Sleep/rest fatigue	-0.30
Cognitive fatigue	-0.36
Parent proxy-report	
Total fatigue	-0.29
General fatigue	-0.30
Sleep/rest fatigue	-0.29
Cognitive fatigue	-0.18

* All values significant at $p < 0.05$. Effect sizes are designated as small (0.10), medium (0.30), and large (0.50) for Pearson product moment correlations.

For example, the Cognitive Fatigue Scale correlated highest (0.77) with the School Functioning Scale for child self-report (0.77 for parent proxy-report).

Table 1 gives the comparisons between the PedsQL Multidimensional Fatigue Scale for the healthy children group and children with rheumatic diseases as a group. For every parent proxy-report comparison, there was a statistically significant difference between healthy children and children with rheumatic diseases. For child self-report, the Total Scale Score and the General Fatigue Scale showed a statistically significant difference between healthy children and children with rheumatic diseases. The hypothesis was generally confirmed that children with rheumatic diseases as a group would manifest greater fatigue symptoms than healthy children as a group.

In exploratory analyses of different levels of fatigue among the rheumatic diseases studied, the only significant differences to emerge revealed a significantly greater degree of fatigue in patients with FM than most patients with other rheumatic diseases ($p < 0.05$). For the Total Fatigue Scale Score, for instance, patients with FM self-reported more fatigue (mean 57.51) than children with dermatomyositis (78.43), JRA [pauciarticular (90.90), polyarticular (77.38), systemic (72.06)], SLE (84.03), and spondyloarthritis (81.97). Similarly for the Total Fatigue Scale score, parents proxy-reported that their children with FM manifested more fatigue (mean 54.98) than children with dermatomyositis (73.24), JRA [pauciarticular (84.72), polyarticular (80.32), systemic (81.81)], SLE (82.81), and spondyloarthritis (82.81). However, because of the small sample sizes across the various rheumatic diseases in the study, these analyses may not have sufficient statistical power to differentiate further among the groups under investigation.

Finally, for the majority of children ($n = 175$), the physician global assessment of disease severity was available. For all the PedsQL Multidimensional Fatigue Scales there was a

significant relationship, in that physician ratings of greater disease severity were associated with greater levels of self-reported and proxy-reported fatigue (Table 3). The majority of the correlations are in the medium effect-size range.

Parent/child concordance. The parent/child concordance intercorrelations matrix is shown in Table 4. Consistent with the literature, child self-report and parent proxy-report correlations are in the medium to large effect-size range.

DISCUSSION

This study presents the measurement properties for the PedsQL Multidimensional Fatigue Scale standard version in pediatric rheumatology. The analyses support the initial reliability and validity of the PedsQL Multidimensional Fatigue Scale as a child self-report and parent proxy-report multidimensional fatigue measurement instrument for pediatric rheumatology. The PedsQL is the only empirically validated, generic, rheumatology-specific and fatigue-specific HRQOL measurement instrument that we are aware of to span this broad age range for child self-report and parent proxy-report while maintaining item and scale construct consistency.

Items on the PedsQL Multidimensional Fatigue Scale had minimal missing responses, suggesting that children and parents are willing and able to provide good quality data regarding the child's fatigue. All the PedsQL Multidimensional Fatigue Scale self-report and proxy-report internal consistency reliabilities exceeded the recommended minimum alpha coefficient standard of 0.70 for group comparisons, with most scales approaching or exceeding an alpha of 0.90, recommended for individual patient analysis⁴⁰.

The intercorrelations among the PedsQL Multidimensional Fatigue Scale and the PedsQL 4.0 Generic Core Scales and PedsQL 3.0 Rheumatology Module Pain and Daily Activities Scales were consistent with the conceptualization of disease-specific symptoms as causal indicators of HRQOL¹ and the literature on fatigue in adult rheumatology. The PedsQL Multidimensional Fatigue Scale generally performed as hypothesized utilizing the known-groups method, differentiating fatigue in healthy children in comparison to children with rheumatic diseases as a group, and was associated with physician global assessment of disease severity. Children with FM as a group manifested significantly more fatigue than children with other rheumatic diseases, suggesting an important area in need of treatment research.

While other pediatric HRQOL instruments exist, including generic measures and rheumatology-specific measures, it has been an explicit goal of the PedsQL Measurement Model to develop and test brief measures for the broadest age group empirically feasible, specifically including child self-report for the youngest children possible. This goal was originally determined in previous empirical efforts to meas-

Table 4. Intercorrelations among PedsQL scales: child self-report above the diagonal, parent proxy-report below the diagonal, child/parent concordance on the diagonal. Concordance (intraclass correlation and Pearson product moment) between child self-report and parent proxy-report are underlined. Average measure intraclass correlation coefficients (ICC) are given in italics below Pearson product moment correlation values for child and parent concordance. ICC values were derived using a 2-way fixed-effects model (with consistency type rather than absolute agreement).

Scale	TF	GF	SF	CF	Tot	Ph	Psy	Em	Soc	Sch	Pain	DA
Total fatigue (TF)	0.87 <u>0.93</u>	0.95	0.91	0.84	0.87	0.80	0.84	0.74	0.63	0.81	0.68	0.58
General fatigue (GF)	0.94	0.83 <u>0.91</u>	0.82	0.73	0.87	0.84	0.80	0.71	0.60	0.77	0.72	0.58
Sleep fatigue (SF)	0.92	0.83	0.81 <u>0.89</u>	0.60	0.91	0.71	0.73	0.64	0.55	0.68	0.60	0.51
Cognitive fatigue (CF)	0.80	0.65	0.58	0.74 <u>0.85</u>	0.71	0.58	0.74	0.63	0.53	0.77	0.49	0.48
Total score generic core (Tot)	0.86	0.85	0.76	0.69								
Physical health (Ph)	0.79	0.81	0.72	0.55								
Psychosocial health (Psy)	0.82	0.80	0.71	0.71								
Emotional functioning (Em)	0.75	0.73	0.69	0.59								
Social functioning (Soc)	0.62	0.60	0.51	0.54								
School functioning (Sch)	0.80	0.76	0.66	0.77								
Pain	0.70	0.69	0.67	0.46								
Daily activities (DA)	0.52	0.50	0.46	0.43								

All correlations are significant at $p < 0.001$. Effect sizes are designated as small (0.10), medium (0.30), and large (0.50) for Pearson product moment correlations. Intraclass correlations (ICC) are designated as ≤ 0.40 poor to fair agreement, 0.41–0.60 moderate agreement, 0.61–0.80 good agreement, and 0.81–1.00 excellent agreement.

ure pain in pediatric rheumatology through the development and testing of the Pediatric Pain Questionnaire²⁹. Thus, the development and testing of the PedsQL Measurement Model emphasizes the child's perceptions, including the youngest children empirically possible.

These findings have several potential limitations. Information on nonparticipants was not available, and the field test was conducted in one children's hospital, which may limit the generalizability of the findings. The healthy sample comparison group data were derived from a previous report utilizing the acute version (7-day recall period) of the PedsQL Multidimensional Fatigue Scale¹⁸. The standard version (one-month recall period) used in our study may not be directly comparable in findings to the acute version. However, in the previous report in which the acute and standard versions of the PedsQL 4.0 Generic Core Scales in healthy samples were compared, no significant differences were observed¹⁸. Further, there were sample differences between the healthy and rheumatology samples, which may limit the generalizability of the findings. Thus, the known-groups validity comparisons in this study should be viewed with caution until a more closely matched healthy sample using the standard version is generated. Although physician global assessment of disease severity was available for the majority of patients, additional clinical data and external measures such as healthcare utilization would further strengthen the instrument's construct validity. Finally, there was limited child self-report data for ages 5–7, although our previous report supported the reliability of the instrument for child self-report for ages 5–7 with a Total Fatigue Scale Score Cronbach coefficient alpha of 0.86¹⁸, similar to the

Total Score alpha of 0.89 in the present study. Nevertheless, instrument validation is an iterative process and consistent with this paradigm, these PedsQL Scales are currently being field tested nationally and internationally in pediatric rheumatology.

The results demonstrate the initial reliability and validity of the PedsQL™ Multidimensional Fatigue Scale in pediatric rheumatology. The PedsQL measurement instruments may be utilized as outcome measures in pediatric rheumatology clinical trials, research, and clinical practice for HRQOL outcome assessment.

The PedsQL™ is available at <http://www.pedsqol.org>.

REFERENCES

1. Fayers PM, Machin D. Quality of life: Assessment, analysis, and interpretation. New York: Wiley; 2000.
2. Spilker B. Quality of life and pharmacoeconomics in clinical trials. Philadelphia: Lippincott-Raven; 1996.
3. Varni JW, Seid M, Kurtin PS. Pediatric health-related quality of life measurement technology: A guide for health care decision makers. J Clin Outcomes Manag 1999;6:33-40.
4. Smith KW, Avis NE, Assmann SF. Distinguishing between quality of life and health status in quality of life research: A meta-analysis. Qual Life Res 1999;8:447-59.
5. World Health Organization. Constitution of the World Health Organization basic document. Geneva: World Health Organization; 1948.
6. Varni JW, Bernstein BH. Evaluation and management of pain in children with rheumatic diseases. Rheum Dis Clin North Am 1991;17:985-1000.
7. Murray KJ, Passo MH. Functional measures in children with rheumatic diseases. Ped Clin North Am 1995;42:1127-53.
8. Varni JW, Wilcox KT, Hanson V, Brik R. Chronic musculoskeletal pain and functional status in juvenile rheumatoid arthritis: An empirical model. Pain 1988;32:1-7.

9. Lovell DJ, Howe S, Shear E, et al. Development of a disability measurement tool for juvenile rheumatoid arthritis: The Juvenile Arthritis Functional Assessment Scale. *Arthritis Rheum* 1989;32:1390-5.
10. Howe S, Levinson J, Shear E, et al. Development of a disability measure tool for juvenile rheumatoid arthritis: The Juvenile Arthritis Functional Assessment Report for Children and Their Parents. *Arthritis Rheum* 1991;34:873-80.
11. Singh G, Athreya BH, Fries JF, Goldsmith DP. Measurement of health status in children with juvenile rheumatoid arthritis. *Arthritis Rheum* 1994;37:1761-9.
12. Varni JW, Seid M, Knight TS, Burwinkle TM, Brown J, Szer IS. The PedsQL™ in pediatric rheumatology: Reliability, validity, and responsiveness of the Pediatric Quality of Life Inventory™ Generic Core Scales and Rheumatology Module. *Arthritis Rheum* 2002;46:714-25.
13. Tucker LB. Whose life is it anyway? Understanding quality of life in children with rheumatic diseases. *J Rheumatol* 2000;27:8-11.
14. Wolfe F, Hawley DJ, Wilson K. The prevalence and meaning of fatigue in rheumatic disease. *J Rheumatol* 1996;23:1407-17.
15. Stone AA, Broderick JE, Porter LS, Kaell AT. The experience of rheumatoid arthritis pain and fatigue: Examining momentary reports and correlates over one week. *Arthritis Care Res* 1997;10:185-93.
16. Belza BL. Comparison of self-reported fatigue in rheumatoid arthritis and controls. *J Rheumatol* 1995;22:639-43.
17. Tench CM, McCarthy J, McCurdie I, White PD, D'Cruz DP. Fatigue in systemic lupus erythematosus: A randomized controlled trial of exercise. *Rheumatology* 2003;42:1050-4.
18. Varni JW, Burwinkle TM, Katz ER, Meeske K, Dickinson P. The PedsQL™ in pediatric cancer: Reliability and validity of the Pediatric Quality of Life Inventory™ Generic Core Scales, Multidimensional Fatigue Scale, and Cancer Module. *Cancer* 2002;94:2090-106.
19. Varni JW, Katz ER, Colegrove R, Dolgin M. Adjustment of children with newly diagnosed cancer: Cross-informant variance. *J Psychosoc Oncol* 1995;13:23-38.
20. Drotar D, editor. Measuring health-related quality of life in children and adolescents. Mahwah, NJ: Lawrence Erlbaum Associates; 1998.
21. Koot HM, Wallander JL, editors. Quality of life in child and adolescent illness: Concepts, methods and findings. East Sussex, UK: Brunner-Routledge; 2001.
22. Varni JW, Setoguchi Y. Screening for behavioral and emotional problems in children and adolescents with congenital or acquired limb deficiencies. *Am J Dis Child* 1992;146:103-7.
23. Janicke DM, Finney JW, Riley AW. Children's health care use: A prospective investigation of factors related to care-seeking. *Med Care* 2001;39:990-1001.
24. Campo JV, Comer DM, Jansen-McWilliams L, Gardner W, Kelleher KJ. Recurrent pain, emotional distress, and health service use in childhood. *J Pediatr* 2002;141:76-83.
25. Varni JW, Seid M, Rode CA. The PedsQL™: Measurement model for the Pediatric Quality of Life Inventory. *Med Care* 1999;37:126-39.
26. Varni JW, Burwinkle TM, Jacobs JR, Gottschalk M, Kaufman F, Jones KL. The PedsQL™ in Type 1 and Type 2 diabetes: Reliability and validity of the Pediatric Quality of Life Inventory™ Generic Core Scales and Type 1 Diabetes Module. *Diabetes Care* 2003;26:631-7.
27. Varni JW, Burwinkle TM, Rapoff MA, Kamps JL, Olson N. The PedsQL™ in pediatric asthma: Reliability and validity of the Pediatric Quality of Life Inventory™ Generic Core Scales and Asthma Module. *J Behav Med* 2004;27:297-318.
28. Uzark K, Jones K, Burwinkle TM, Varni JW. The Pediatric Quality of Life Inventory™ in children with heart disease. *Progr Pediatr Cardiol* 2003;18:141-8.
29. Varni JW, Thompson KL, Hanson V. The Varni/Thompson Pediatric Pain Questionnaire: I. Chronic musculoskeletal pain in juvenile rheumatoid arthritis. *Pain* 1987;28:27-38.
30. Varni JW, Waldron SA, Gragg RA, et al. Development of the Waldron/Varni Pediatric Pain Coping Inventory. *Pain* 1996;67:141-50.
31. Varni JW, Seid M, Kurtin PS. The PedsQL™ 4.0: Reliability and validity of the Pediatric Quality of Life Inventory™ Version 4.0 Generic Core Scales in healthy and patient populations. *Med Care* 2001;39:800-12.
32. Varni JW, Burwinkle TM, Seid M, Skarr D. The PedsQL™ 4.0 as a pediatric population health measure: Feasibility, reliability, and validity. *Ambul Pediatr* 2003;3:329-41.
33. Aday LA. Designing and conducting health surveys: A comprehensive guide. San Francisco: Jossey-Bass; 1996.
34. Fowler FJ Jr. Improving survey questions: Design and evaluation. Thousand Oaks, CA: Sage Publication; 1995.
35. Schwarz N, Sudman N. Answering questions: Methodology for determining cognitive and communicative processes in survey research. San Francisco: Jossey-Bass; 1996.
36. Thompson KL, Varni JW. A developmental cognitive-biobehavioral approach to pediatric pain assessment. *Pain* 1986;25:282-96.
37. Fairclough DL. Design and analysis of quality of life studies in clinical trials: Interdisciplinary statistics. New York: Chapman & Hall/CRC; 2002.
38. McHorney CA, Ware JE, Lu JFR, Sherbourne CD. The MOS 36-item short-form health survey (SF-36): III. Tests of data quality, scaling assumptions, and reliability across diverse patient groups. *Med Care* 1994;32:40-66.
39. Cronbach LJ. Coefficient alpha and the internal structure of tests. *Psychometrika* 1951;16:297-334.
40. Nunnally JC, Bernstein IR. Psychometric theory. 3rd ed. New York: McGraw-Hill; 1994.
41. Pedhazur EJ, Schmelkin LP. Measurement, design, and analysis: An integrated approach. Hillsdale, NJ: Lawrence Erlbaum Associates; 1991.
42. Cohen J. Statistical power analysis for the behavioral sciences. 2nd ed. Hillsdale, NJ: Lawrence Erlbaum Associates; 1988.
43. McHorney CA, Ware JE, Raczek AE. The MOS 36-item short-form health survey (SF-36): II. Psychometric and clinical tests of validity in measuring physical and mental health constructs. *Med Care* 1993;31:247-63.
44. McHorney CA, Ware JE, Rogers W, Raczek AE, Lu JFR. The validity and relative precision of MOS short- and long-form health status scales and Dartmouth COOP charts: Results from the Medical Outcomes Study. *Med Care* 1992;30:MS253-65.
45. Bartko JJ. The intraclass correlation coefficient as a measure of reliability. *Psychol Rep* 1966;19:3-11.
46. SPSS. SPSS 8.0 for Windows. Chicago: SPSS Inc.; 1998.