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Measures of Juvenile Fibromyalgia: Pain and Symptom Assessment Tool (PSAT), PROMIS® Pain Interference, Anxiety and Depression scales, Functional Disability Inventory (FDI) and Pediatric Quality of Life (PedsQL) 3.0 Rheumatology Module

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Abstract

Juvenile fibromyalgia (JFM) is a chronic and debilitating noninflammatory musculoskeletal pain syndrome that is typically diagnosed in adolescence. There are no specific medical tests or disease markers to diagnose the condition, and classification is based on patient report of pain and other associated symptoms after ruling out other underlying medical causes. JFM can be disabling in multiple life domains and therefore, a multidimensional assessment of JFM is recommended to gain a full picture of the extent of JFM symptoms along with their impact on physical and emotional functioning and quality of life. The following updated review outlines evidence-based measures useful in the assessment of school-age children and adolescents with JFM. New measures include 1) the Pain and Symptom Assessment Tool (PSAT) that offers a standardized tool for the classification of fibromyalgia in pediatric patients and 2) the Patient-Reported Outcomes Measurement Information System (PROMIS®) Pediatric Pain Interference, Anxiety, and Depression Scales. Updated information is presented on previously established measures that assess the impact of JFM on functioning and quality of life - the Functional Disability Inventory (FDI) and the Pediatric Quality of Life Inventory (PedsQL) 3.0 Rheumatology Module Pain and Hurt Scale, are also discussed. In general, there are increasing options for validated patient-reported outcome measures available to measure the spectrum of symptoms in JFM and assess impact on daily life. Greater consistency in identification of JFM and use of standardized assessment tools will undoubtedly lead to higher quality research much needed in this relatively understudied musculoskeletal pain condition.

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Conflict of Interest Statement

M. Daffin declares that she has no conflict of interest.

R.C. Gibler declares that he has no conflict of interest.

S. Kashikar-Zuck declares that she has no conflict of interest.

Introduction

Juvenile fibromyalgia (JFM) is a chronic and debilitating noninflammatory musculoskeletal pain condition that is typically diagnosed in adolescence. JFM is characterized by diffuse widespread pain, sleep difficulty, fatigue, and other associated symptoms.[1] This condition often involves impairment in physical, social, and emotional functioning.[2–9] Currently, there are no specific medical tests or disease markers to diagnose the condition. Diagnosis is based on subjective patient-report of pain and other symptoms, after ruling out underlying inflammatory disorders or other medical conditions. A multidimensional assessment of fibromyalgia syndrome, including measures of pain, fatigue, sleep, overall functioning, and quality of life is recommended to gain a complete picture of JFM symptoms and their impact on the patients' ability to function in their daily lives.[10]

Information presented in this review reflects several recent and important developments since the last review of measures used in JFM[11] that have enhanced evidence-based assessment in youth with JFM. In the following sections, we discuss a new measure for the proper classification of fibromyalgia in pediatric patients derived from the 2010 American College of Rheumatology criteria for fibromyalgia – the Pain and Symptom Assessment Tool (PSAT) for JFM. Specifically, we present results from the initial validation of the PSAT, a measure developed to reliably identify youth with JFM and distinguish JFM from other chronic localized pain conditions. Next, we review the psychometric properties and utility of the recently developed pediatric Patient-Reported Outcomes Measurement Information System (PROMIS®) scales – the Pain Interference Scale, the Anxiety Scale, and Depression Scale. Finally, we present updated information on widely-used instruments discussed in our previous review for measuring difficulties associated with JFM, including the Functional Disability Inventory (FDI) and the PedsQL® – Rheumatology Module.

PAIN AND SYMPTOM ASSESSMENT TOOL (PSAT)

Description

Purpose.—Since Yunus and Masi[1] proposed their classification criteria for JFM in 1985, there have been no updates to the classification guidelines in pediatrics, despite multiple iterations of suggested classification criteria for adults with fibromyalgia.[12,13] The Pain and Symptom Assessment Tool (PSAT) was developed to enhance consistent classification of adolescents with juvenile fibromyalgia (JFM) based on the 2010 American College of Rheumatology (ACR) criteria for adult fibromyalgia.

Content.—The PSAT consists of two subscales – the Widespread Pain Index (WPI) which assesses the number of pain locations and the Symptom Severity (SS) scale which assesses severity of cardinal symptoms of FM, including fatigue, tiredness, and concentration/ memory difficulties and the presence of other somatic symptoms that commonly co-occur with FM, such as symptoms of dysautonomia, irritable bowel, numbness, tenderness, migraine etc.

Number of items.—The PSAT consists of a total of 72 items across the WPI and the SS checklist. The WPI consists of 19 pain locations and the SS scale is comprised of the remaining items

Response options/scale.—For the WPI (Part A), respondents simply endorse their location(s) of pain from a list of pain locations. The SS scale is comprised of a section (Part B) in which respondents rate the severity of their symptoms on a Likert scale from 0 (“no problem”) to 3 (“severe, pervasive problem”) for the 3 cardinal symptoms - fatigue, tiredness, and concentration/memory difficulties, and complete a checklist (Part C) of additional somatic symptoms they may experience.

Recall period for items.—Respondents indicate the location(s) of pain and symptom severity experienced in the past week. When using the PSAT for classification of JFM (which requires symptom duration of at least 3 months), respondents are asked to report on their pain and somatic symptoms experienced daily or almost daily for the past 3 months.

Cost to use.—The PSAT can be accessed and used for no charge.

How to obtain.—Copies can be obtained directly from Dr. Kashikar-Zuck.

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Practical application

Method of administration.—The PSAT is a brief questionnaire that can be administered as a patient self-report measure with or without clinician assistance (based on the respondent’s age and comprehension of the items).

Scoring.—The total number of endorsed pain locations is summed to yield a WPI score (Part A) score. The Symptom Severity (SS) score consists of the sum of their ratings on Part B and a total somatic symptoms score (0–3) derived from Part C. Based on the number of items endorsed in Part C, a score of 0 (“no symptoms”) to 3 (“a great deal of symptoms”) is assigned. The total SS score is computed by summing the Part B and Part C scores.

Score interpretation.—A classification of JFM is met if the WPI score is ≥ 7 and SS is ≥ 5; or WPI score is 3–6 and SS is ≥ 9.[13] A total score on the PSAT (WPI + SS scores) can be used as an indicator of JFM symptom severity.

Respondent time to complete.—The PSAT takes approximately 5 minutes for respondents to complete. It may take up to 10–15 minutes for younger children if clinician assistance is needed to ensure they understand the items.

Administrative burden.—Administration and scoring take between 5–10 minutes each. Some training and familiarity with the measure is required due to the need to derive scores for Part C of the measure.

Translations/adaptations.—The current PSAT is a pediatric adaptation of the WPI and SS index used for classification of fibromyalgia in adults using ACR 2010 criteria.[13] There are no adaptations or translations of the pediatric measure at present.

Psychometric information

Floor and ceiling effects.—Floor and ceiling effects have not been detected in patients who have chronic pain (localized or widespread pain as in JFM). It is only appropriate for use in youth with pain symptoms (not healthy adolescents)

Reliability.—The PSAT is a new measure for which psychometric properties are still being tested. Currently, there are no psychometric data available to document the reliability of the PSAT in JFM.

Validity.

Evidence of content validity.: In a preliminary validation study, the 2010 ACR criteria for FM demonstrated high sensitivity and specificity (89.4% and 87.5%, respectively) in correctly identifying youth with JFM and differentiating these adolescents from those with localized pain conditions.[14]

Responsiveness.—Information about responsiveness to change is not yet available.

Minimally important differences.—Not available.

Generalizability.—Studies are underway to evaluate whether the PSAT can be used to identify secondary JFM/widespread pain in youth with rheumatic diseases such as juvenile arthritis.

Use in clinical trials.—The PSAT has not yet been used in any published clinical trials.

Critical appraisal of overall value to the rheumatology community

Strengths.—The PSAT is a potentially useful tool for proper identification of patients with JFM, which will enhance consistency in classification for clinical and research purposes. The PSAT appears to be sufficient for correctly classifying youth with JFM and an additional tender point examination does not affect the accuracy of the measure.[14,15] It is a brief measure that can be easily incorporated into clinical care or electronic medical records.

Caveats and cautions.—Although the items on this tool have been adapted from the adult WPI and SS scales which have much stronger evidence from validation studies, only a single published paper has been published on the pediatric version of the measure. Larger validation studies with more diverse samples of youth with chronic pain are underway. Additional work is also needed to determine if the PSAT is appropriate for use among boys with JFM. Boys form a very small proportion of JFM patients and the validity of this measure has not yet been tested for males.

Clinical usability.—The PSAT can be administered easily as a screening measure in clinical settings and appears to be useful in distinguishing youth with JFM from those with localized chronic pain conditions. It has potential use for monitoring the severity of JFM symptoms over the course of treatment.

Research usability.—The PSAT is easily incorporated as a self-report measure to screen for JFM and also as an outcome measure in research settings.

PROMIS-Pediatric Pain Interference, Anxiety, and Depression

Description

Purpose.—The Patient Reported Outcomes Measurement Information Systems (PROMIS®; www.nihpromis.org) initiative was established by the National Institutes of Health (NIH) to develop patient-reported assessment measures for various aspects of health among individuals with a range of chronic conditions. The intent was to create a set of common set of brief and precise tools with strong psychometric properties that are applicable across disease conditions to be used in clinical research, clinical trials, and eventually, clinical settings. Initial work focused on PROMIS® measures for adults, but in recent years, a number of pediatric-specific measures have been developed. For the current review, we present information about PROMIS® instruments that are particularly relevant for use in JFM - the PROMIS® Pain Interference (PROMIS®-PPI), PROMIS® Anxiety, and PROMIS® Depression scales.

All PROMIS® pediatric measures were developed using modern test theory methods and a systematic process to ensure rigorous test development. The process began with a review of all currently available measures for each specific domain, expert review of items, focus groups and individual interviews with patients and content experts to develop large item banks for each domain of interest. This was followed by analyses based on item response theory methods and selection of the most informative items for the final versions of each scale.[16–20] The item banks derived from field testing have been subsequently examined and validated for use in several pediatric populations, including among youth with chronic pain.[21]

Content.—Items on the PROMIS®- PPI assess the extent to which pain interferes with social, cognitive, emotional, physical, and recreational domains of daily living. The PROMIS® Anxiety scale assesses feelings of fear, worry, and somatic symptoms (e.g., racing heart, dizziness) that occur in multiple contexts. The PROMIS® Depression scale measures common depressive symptoms, including negative mood, decreased positive affect, anhedonia (e.g., loss of interest), and negative social cognition (e.g., loneliness).

Number of items.—Short-form version of the PROMIS® instruments generally include 8 items for each item with no subscales. The most recent version of the parent proxy report of the PROMIS®- Depression contains 6 items. PROMIS® scales are also available in computerized adaptive testing (CAT) form which require administration of fewer items to arrive at a score.

Response options/scale.—The child, adolescent, or parent responds to each item using a five-point Likert scale ranging from 1 (“never”) to 5 (“almost always”).

Recall period for items.—Respondents are asked to report how often each item applied to them in the last 7 days.

Cost to use.—There is no cost to use PROMIS® in research or clinical settings.

How to obtain.—All PROMIS® instruments can be accessed at www.nihpromis.org. Options for fixed-length, customized short forms and administration through CAT can be found at www.assessmentcenter.net

Practical Application

Method of administration.—The PROMIS®- PPI, PROMIS®- Anxiety, and PROMIS®- Depression scales are self-report instruments completed by children and adolescents. Parent proxy report versions are also available. Each instrument can be administered in paper format or via Computerized Adaptive Testing, which uses item response theory to select subsequent items from an item pool based on patient’s previous responses. This may reduce patient burden and improve the precision of the measure.

Scoring.—Item scores on the child and parent proxy reports are summed to yield a total score ranging from 8 to 40, with higher scores reflecting more difficulties in a particular domain. Note that scores for the parent proxy version of the PROMIS®- Depression range from 6 to 30.

Score interpretation.—The total scores for the PROMIS® instruments can be converted to *T*-scores. Scores on the *T* distribution have a mean of 50 and a standard deviation of 10. Raw to *T*-score conversion tables for each of the PROMIS® instruments are located at www.healthmeasures.net/promis-scoring-manuals.

Respondent time to complete.—Completion of each of the short-form measures generally takes less than 10 minutes; however, the instruments may take longer for younger children to complete and may require administration in an interview format.

Administrative burden.—The time required to complete each measure is approximately 5 minutes, and time required to score is less than 5 minutes. No special training is necessary to administer or score the measure.

Translations/adaptations.—Each of the reviewed PROMIS® instruments has been translated into Spanish, Dutch, and German and other languages will be added. See http://www.healthmeasures.net/explore-measurement-systems/promis/intro-to-promis/available-translations for currently available translations.

Psychometric Information

Floor and ceiling effects.—Based on the extensive psychometric examination of the items included in each of the scales and validation across multiple disease groups (including pediatric pain), floor and ceiling effects are not expected to be a concern.

Reliability.—PROMIS® scales are highly precise and reliable based on the modern test theory methods used to develop the measures. Some papers have published traditional metrics such as Cronbach's alpha (internal consistency) coefficients in a range of pediatric populations. These studies reported PROMIS®-PPI coefficients as ranging from acceptable to high (α = .68−.90)[16,17] and the PROMIS® Anxiety and Depression coefficients as high (α range = .85 – .91 for PROMIS®-Anxiety; α range = .85 – .94 for PROMIS®-Depression).[18,19]

Validity.

Evidence of content validity.: The appropriateness of the PROMIS® pain domain framework was confirmed using a rigorous, iterative qualitative methodology that considered perspectives from multiple stakeholders, including patients, parents, and content experts.[20] Although there are no published studies in JFM at the present time, studies in other painful conditions show evidence that PROMIS®-PPI scores differentiate between children with rheumatic diseases who demonstrate higher degrees of school-related impairment.[22] Also, the PROMIS®-Anxiety scale differentiates between children with sickle cell disease who had and had not received home treatment for pain.[22]

Evidence of construct validity.: The PROMIS®-PPI demonstrated a significant and positive correlation with pain intensity on the visual analog scale (VAS), and daily functioning on the Functional Disability Inventory (FDI) in an 8 week clinical trial for youth with JFM.[17] The PROMIS®-PPI and the FDI were moderately and significantly correlated across baseline and post-treatment in a large randomized clinical trial of youth with JFM (T1 r =.51, T2 r =.53; $p < .05$).[17]

Responsiveness.—The PROMIS®-PPI, PROMIS®-Anxiety, and PROMIS®-Depression scales have demonstrated responsiveness to change similar to commonly-used legacy measures (i.e., Children's Depression Inventory – Second Edition [CDI-2], the PedsQL QOL subscales, and the Functional Disability Inventory (FDI) among youth receiving treatment in outpatient pain clinic and intensive day treatment settings[21]. One study reported PROMIS®-PPI scores to be a sensitive indicator of clinical improvement in patients enrolled in a small-scale trial of cognitive-behavioral and exercise treatments for JFM.[17]

Minimally important differences.—Minimally important differences for the PROMIS®-PPI have been established for use in juvenile idiopathic arthritis (JIA)[23], but have not yet been established in JFM. No data is available for PROMIS®-Anxiety, and PROMIS®-Depression scales.

Generalizability.—The pediatric PROMIS® instruments have been specifically developed to be highly generalizable across clinical populations and settings and are appropriate for use

in children as young as 8 years of age. The language difficulty of the measure is adapted to the typical reading level of children and adolescents.

Use in clinical trials.—The Pediatric Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (PedIMMPACT) guidelines recommend measurement of several domains of functioning for pediatric chronic pain trials[24] for which the PROMIS® measures are ideally suited. The PROMIS® instruments have begun to be used in clinical research[21,25] and to our knowledge has only been used in one published small-scale trial in JFM.[17]

Critical Appraisal of Overall Value to the Rheumatology Community

Strengths.—The PROMIS® instruments were developed using rigorous, mixed-method techniques.[16,26–28] The pediatric PROMIS® measures discussed in this review are reliable, valid, and assess the physical and psychosocial impact of pain commonly experienced by youth with JFM.

Caveats and cautions.—To date, there are no published clinical reference points for the interpretation of *T*-scores based on established norms. Currently, higher scores on the PROMIS® measures are representative of more difficulties in a respective domain but clinical cutoffs have yet to be established in JFM.

Clinical usability.—The PROMIS® pediatric measures are brief, valid and reliable measures that are easy to incorporate into clinical settings. The PROMIS® PPI scale performs similarly to the FDI (a well-established measure of functioning discussed below) and may be a briefer substitute for the FDI. The clinical utility of the PROMIS® Anxiety and Depression scales are more likely to be in the realm of brief screening/tracking for mood symptoms. In contrast, other established measures of clinical anxiety (e.g., the SCARED, The Screen for Child Anxiety Related Disorders;[29] and depression (e.g., Children's Depression Inventory[30,31]) that are more detailed and multidimensional will still be needed for diagnostic purposes. More information about the clinical utility of PROMIS® measures is required before these tools are more widely deployed in pediatric pain.

Research usability.—PROMIS® measures are brief, require minimal instructions, and are simple to administer in short-form or CAT versions. Scoring is also straightforward. PROMIS® measures are particularly well-suited for research because they are precise tools that have been validated in various pediatric conditions and normed and validated in the US. Use of PROMIS® measures enables comparison of results across studies and the next several years will yield results of several ongoing studies using these tools.

FUNCTIONAL DISABILITY INVENTORY (FDI)

No additional psychometric validation studies have been published using the FDI since the previous review of measures for JFM.[32] However, additional information strengthening the evidence for the use of the FDI in clinical settings and utility in clinical trials continues to accumulate.

Description

Purpose.—The FDI is a brief 15-item measure of impairment in daily functioning in children and adolescents with chronic pain. The FDI was originally created for children ages 8–18 years with chronic abdominal pain[33,34] but has been used extensively in research with other pediatric pain conditions including JFM.[35–37] Since being developed in 1991, the FDI has not updates or revisions. An FDI parent-report version also is available.

Content.—The FDI assesses difficulty with completing activities in a number of domains including home, school, recreational, and social settings. The child or adolescent rates the degree of difficulty completing each activity (e.g., “being at school all day”, “completing chores” or “doing something with a friend”).

Number of items.—The FDI includes a total of 15 items with no subscales.

Response options/scale.—Responses are rated on a 5-point Likert scale (0=no trouble, 1=a little trouble, 2= some trouble, 3=a lot of trouble, or 4=impossible), regarding amount of difficulty performing each activity.

Recall period of items.—Respondents are asked to report the level of difficulty completing each activity “in the last few days.”

Cost to use.—The FDI can be accessed and used for no charge.

How to obtain.—Copies and permissions can be obtained directly from the following website at no charge: https://pediatrics.mc.vanderbilt.edu/interior.php?mid=5679.

Practical Application

Method of administration.—The FDI is a child/adolescent self-report measure. It can be administered by paper format or in interview format for younger children. The instrument can be completed in person, by mail, or by phone. The FDI was developed to monitor patient progress providing follow-up assessment via phone interview. A parent-report version of the measure also is available.

Scoring.—Each items’ score ranges from 0–4 on a Likert scale and the total score is a sum of endorsed items. The total FDI score can be easily hand scored and does not require computer scoring.

Score interpretation.—FDI total scores range from 0–60 with higher scores indicating greater functional disability. Clinical reference points have been developed to identify 3 categories of disability in pediatric chronic pain ranging from no/minimal disability (0–12), moderate disability (13–29), and severe disability (≥30).[38] Clinic-based studies reveal that youth with chronic pain generally endorse scores in the moderate range of disability (13–29).[38] Community based studies indicated that healthy-school aged children report overall FDI scores in the range of 3–8.[39,40]

Respondent time to complete.—It generally takes <10 minutes to complete the FDI. However, the FDI may take longer for younger children with reading difficulties. The interview format can be administered as needed. The FDI has a Flesch reading ease of 89.7 and a Flesch-Kincaid grade level of 3.2.

Administrative burden.—Administration of the FDI is 5–10 minutes while scoring time is <5 minutes. Special training is unnecessary to administer or score the FDI.

Translation/adaptions.—The FDI is available in English and 32 other languages (see website above for a full list).

Psychometric Information

The FDI is an established measure with strong evidence for reliability and validity.[17,33,34] The FDI items were originally created from reviewing and adapting existing items from adult measures of physical and psychosocial functioning, i.e. the Sickness Impact Profile[41] and the Duke-UNC Health Profile[42]. Selected items underwent pilot testing with children and their parents in a pediatric outpatient clinic, following which items were removed and other items reworded[34] to reach the final set of items.

Floor and ceiling effects.—Clinic-based studies reveal that youth with chronic pain generally endorse scores in the moderate range of disability (13–29)[38]. Community studies indicated that healthy school-aged children report overall FDI scores in the range of 3–8[39]. The FDI has no known floor or ceiling effects, as individuals rarely score either 0 or 60.

Reliability.

Evidence for internal consistency.: Cronbach's alpha reliability coefficients for the FDI are high (α = .79−.92)[17,33,34]. The mean interim correlation is .38 (12)[34].

Evidence of stability.: Test-retest correlations are high at 2-week (r=0.80, P < 0.001), 6-week (r=0.70, P <0.001), and 6-month follow-up (r= 0.63, P < 0.001)[34].

Validity.

Evidence of content validity.: Concurrent validity was assessed by calculating correlation on the FDI with school absences, a common proxy for child disability[41]. Scores on the FDI significantly correlated (r = 0.52, P <0.001) with the number of school days missed in the previous 3 months. Discriminant validity was assessed by examining whether the FDI could discriminate between diagnostic groups (i.e. abdominal pain with organic etiology, recurrent abdominal pain, and health controls). The FDI discriminated between the 3 groups (F[2,97] = 26.40, P < 0.001). Post hoc analyses indicated significantly higher FDI scores for adolescents with abdominal pain conditions when compared to healthy controls[34].

Evidence of construct validity.: Construct validity has been assessed by comparing the association between the FDI and other measures of child well-being. Studies have shown support for construct validity of the FDI with significant positive correlations with measures

of depression (r=0.43–0.45, P < 0.01), pain (r = 0.37–0.41, P < 0.05)[38], and pain interference (r = 0.51, P <.01) in youth with chronic widespread pain[17,38].

Evidence of criterion validity.: Predictive validity was examined in an abdominal pain population by correlating FDI scores and illness-related school absences over the course of 3 months following their initial clinic appointment. (r=0.44, P < 0.001). In the same study, initial FDI scores were highly correlated with medication use (r=0.26, P < 0.05) and somatic symptoms (r= 0.45, P < 0.001) at 3 month follow up[34]. Currently, no studies examine criterion validity explicitly in juvenile FM.

Responsiveness.—Treatment studies examining the efficacy of non-pharmacological interventions in juvenile fibromyalgia consistently find significant decreases in FDI scores from pre- to post-treatment.[2,17,43]

Minimally important differences.—Sil and colleagues[44] published a study in which the Reliable Change Index of 7.8-points on the FDI. This RCI distinguished treatment responders from non-responders in the context of a clinical trial of cognitive-behavioral therapy for JFM. A reduction of ≥ 7.8 points in FDI scores from pre-to-post treatment reflected an average 40% reduction in disability - considered a clinically meaningful change.

Generalizability.—The FDI is generalizable across many chronic pain conditions including chronic abdominal pain,[33] recurrent pain,[35] and headache.[45]

Use in clinical trials.—The FDI has been recommended for the assessment of physical functioning outcomes in clinical trials of pediatric chronic pain by the Pediatric Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (PedIMMPACT) guidelines[24]. The FDI has been used in a number of clinical trials examining the efficacy of non-pharmacological interventions in juvenile fibromyalgia consistently find significant decreases in FDI scores from pre- to post-treatment.[2,17,43]

Critical Appraisal of Overall Value to the Rheumatology Community

Strengths.—The FDI is a well-established measure widely used to assess functional impairment in school-age children and adolescents with chronic pain. The PedIMMPACT guidelines recommend the measure for use in pediatric pain clinical trials.[24] The FDI has been used in several clinical trials for pediatric pain conditions, including JFM.[2,17,43,46]

Caveats and cautions.—Items on the FDI tend to focus more on difficulties with physical function. Social and emotional areas of functioning that may be impacted by pain are not as well-captured by this measure.

Clinical usability.—The FDI is a reliable, valid measure for assessing functional impairment in children and adolescents with juvenile fibromyalgia in a clinic setting. It is an efficient and user-friendly tool for tracking patient outcomes throughout treatment and successfully has been integrated into an outpatient clinic setting.[47] The FDI can be utilized in treatment with patients and their parents when developing treatment goals aimed at decreasing disability. The FDI requires minimal administrative/respondent burden.

Research usability.—The FDI has been successfully used in clinical research and clinical trials in JFM.[2,6,7,17,43] The measure is easy to administer, involves minimal administrative/respondent burden, and is a proven outcome indicator of treatment efficacy.

PEDIATRIC QUALITY OF LIFE INVENTORY (PedsQL) 3.0 RHEUMATOLOGY MODULE

Description

Purpose.—The PedsQL 3.0 Rheumatology Module assesses disease-specific quality of life (QOL) among children and adolescents with rheumatologic conditions, including juvenile idiopathic arthritis, systemic lupus erythematosus, and juvenile fibromyalgia. There are different versions of this measure for children and adolescents, and a parent-proxy report version is also available.

Content.—The pain and hurt subscale assesses stiffness and disrupted sleep due to pain ("I have trouble sleeping because of pain or aching in my joints and/or muscles"). The daily activities subscale assesses the extent to which pain has interfered with tasks such as writing or drawing with pencils and turning door handles. The treatment subscale measures the physical and emotional impact of receiving treatment for pain ("My physical therapy or daily exercise hurts"). The worry subscale captures fear and anxiety surrounding illness and medical treatment ("I worry about whether or not my medicines are working"). The communication subscale assesses difficulties with discussing illness with others, such as medical staff ("It is hard for me to explain my illness to other people").

Number of items.—The Rheumatology Module contains 22 items and five subscales: pain and hurt (4 items), daily activities (5 items), treatment (7 items), worry (3 items), and communication (3 items).

Response options/scale.—For each of the subscales, respondents rate items using a 5-point Likert scale ("never," "almost never," "sometimes," "often," "almost always") with higher ratings indicating more difficulty with the item.

Recall period for items.—Respondents are asked how much of a problem each item has been within the last month.

Cost to use.—The PedsQL is free to use for certain types of non-funded academic research. An annual license fee is required for funded academic research, large non-commercial organization research, and commercial use. See www.pedsql.org/PedsQL-CostStructure.pdf for more information.

How to obtain.—James W. Varni, PhD, Professor Emeritus of Architecture and Medicine, Department of Landscape Architecture and Urban Planning, College of Architecture, Texas A&M University, 3136 TAMU, College Station, TX 77843–3137; jvarni@tamu.edu. Copies can also be ordered at the following website: http://www.pedsql.org.

Practical application

Method of administration.—The measure is a self-report measure for children (ages 8–12 years), adolescents (ages 13–18) and a proxy-report version for their caregivers.

Scoring.—Responses are reversed scored and transformed to a 0–100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0) such that higher scores indicate better functioning or fewer problems in an area. Subscale scores are computed by summing items and dividing by the total number of items answered.

Score interpretation.—Scores on each of the subscales range from 0–100, with higher scores indicating better disease-related QOL in a respective domain. Several studies suggest that youth with JFM report worse QOL across all domains relative to youth with rheumatic diseases[48–50].

Respondent time to complete.—The measure takes approximately 10 minutes for youth and caregivers to complete.

Administrative burden.—The entire Rheumatology Module can be administered in approximately 10 minutes. Scoring also takes approximately 10 minutes and requires minimal training.

Translations/adaptations.—The PedsQL 3.0 Rheumatology Module is available in English, French, German, Italian, Russian, Spanish, and Slovenian. There are cultural adaptations for English for the US and Spanish for the US.

Psychometric information

Floor and ceiling effects.—There are no known floor or ceiling effects for the Rheumatology Module but there is a high degree of variability in patients' responses on each of the subscales[48–50].

Reliability.

Internal consistency.: All scales on the Rheumatology Module demonstrate high internal consistencies among youth with rheumatologic diseases[50]. A more recent study of 114 adolescents with JFM revealed adequate to strong internal consistencies for each of the subscales, with Cronbach's α ranging from 0.68 – 0.86[48].

Evidence of interrater reliability.: Correlations between parent-proxy and child reports on the Rheumatology Module are moderate (Spearman *r* range 0.33–0.45).[48]

Validity.

Evidence of content validity.: Medical experts, patients, and families of patients were included in the development of the PedsQL.

Evidence of convergent validity.: One study reported moderate negative correlations between the daily activities scale scores and the FDI on both the child and parent proxy

report (Pearson's *r*s = −0.44 and −0.42, respectively). The parent proxy report of the pain and hurt scale demonstrates a moderate negative correlation with Visual Analog Scale pain reports ($r = -0.52$), though this correlation is smaller for the child/adolescent report ($r = -0.28$;[48]).

Evidence of discriminant validity.: This same study showed that the FDI moderately negatively correlated only with the child report pain and hurt ($r = -0.41$), daily activities ($r = -.44$) and worry ($r = -0.34$) scales. Similarly, the VAS showed only small, non-significant correlations with all of the Rheumatology Module scales except the pain and hurt scale.[48]

Responsiveness.—The PedsQL Rheumatology Module has been shown to be responsive to change, showing improvements after treatment (see use in clinical trials below). However, improvements seem to reflect generic improvements in well-being that may not be specific to type of treatment.

Minimally important differences.—Currently, there are no available data on the Rheumatology Module that provide information about minimally important differences in JFM treatment.

Generalizability.—The PedsQL Rheumatology Module appears to be generalizable for use in most pediatric rheumatic diseases but several of the scales do not appear to be clearly applicable in JFM.

Use in clinical trials.—To date, the Rheumatology Module has been used as an outcome measure in one clinical trial of cognitive-behavioral therapy (CBT) or fibromyalgia education (FE) for 114 youth with JFM. Both the CBT and FE groups demonstrated improvements (i.e., increased scores) on the pain and hurt, worry, treatment, and communication scales at the end of treatment and over two follow up periods. However, there were no main or group interaction effects for time for the daily activities scale.[48]

Critical appraisal of overall value to the rheumatology community

Strengths.—The PedsQL 3.0 Rheumatology Module is a brief and easily administered self-report measure that assesses quality of life in multiple domains, including the pain and hurt scale relevant to JFM. The subscales discriminate between groups of youth with various rheumatologic conditions, and four of the five scales demonstrate sensitivity to change.

Caveats and cautions.—The items on the daily activities subscale likely do not specifically capture the functional limitations of youth with JFM (e.g., turning knobs, using utensils), which may explain why this subscale failed to demonstrate sensitivity to change in the context of a clinical trial. Researchers have recommended using an alternative measure, such as the FDI discussed previously, in order to capture impairment in daily functioning among patients with JFM.[48]

Clinical usability.—The measure and its subscales are simple to administer and score, though the clinical utility of the measure has not been examined. It has been recommended that both the child and parent proxy reports be used in clinical settings, as youth and their

parents may have different perspectives particularly on items related to communicating about the child's diagnosis with healthcare professionals.[48]

Research usability.—The Rheumatology Module demonstrates strong psychometric properties for use in pediatric rheumatology populations. It likely is best suited for use as a supplemental measure of disease-related QOL as it provides useful information about specific difficulties experienced by youth and their families (e.g., worries and difficulties with communication about their conditions). Other instruments, such as the FDI and the new PROMIS® measures may provide more specific and relevant measures of the impact of pain and symptoms among youth with JFM.[48]

Summary and Recommendations

The present review summarizes recent and important developments that have enhanced evidence-based assessment among youth with JFM. The PSAT, which has demonstrated initial evidence for its sensitivity and specificity, shows promise as an instrument for classifying children and adolescents with JFM, characterizing the severity of their symptoms, and differentiating these youth from those with more localized pain conditions. Similarly, the PROMIS® instruments allow for a valid and reliable assessment of the range of physical and emotional difficulties experienced by this population. The PROMIS® measures also provide opportunities for comparison across pediatric medical conditions. Finally, the FDI continues to be a gold-standard measure in outcomes research, as it demonstrates excellent reliability, validity, and responsiveness to change among youth with JFM.

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Table 1.

Practical Applications for Juvenile Fibromyalgia Measures

Measure	Number of Items	Content/Domains	Method of Administration	Recall period	Response format	Range of scores	Score interpretation	Availability of normative data	Cross-cultural validation
PSAT	72	Widespread Pain Index (WPI) and Symptom Severity (SS) scale.	Patient self-report (with or without clinician support)	Past 7 days Modified to – Past 3 months for JFM classification	Paper and pencil	WPI – 0–10 SS scale – 0–12	A classification of FM is made if the WPI ≥ 7 and SS ≥ 5 or WPI 3–6 and SS ≥ 9	Only applicable in patients with pain	None available
PROMIS – PPI, Anxiety, Depression	8 for child and parent self-report short forms; 6 for depression parent proxy report	Assessment of pain-related impairment in various domains of functioning; assessment of anxiety and depressive symptoms	Patient and parent proxy report	Last 7 days	Paper and pencil; computer assisted testing (CAT) available	0–40 (0–32 for parent proxy report of depression)	Raw scores can be converted to *T* scores with a mean of 50 and standard deviation of 10	Yes	None available
FDI	15	Assesses difficulty with completing daily activities in multiple domains	Patient self-report; parent report version available	Last few days	Paper and pencil	0–60	0–12 = mild disability; 13–29 = moderate disability; >30 = severe disability	Yes	None available
PedsQL	22	Assesses disease-related QOL across five domains: pain and hurt, daily activities, treatment, worry, and communication	Patient and parent proxy-report	Last month	Paper and pencil	0–100 (items are reverse scored and transformed)	Higher scores indicate better functioning or fewer problems in an area	Yes	None available

Measure	Floor, ceiling effects	Reliability	Validity	Responsiveness	Minimally important differences	Use in RCTs
PSAT	None reported	Not yet established	Excellent sensitivity and specificity	Not yet examined in JFM	Not yet established in JFM	No information available at this time
PROMIS – PPI, Anxiety, Depression	None	Good-to-excellent internal consistency reliability	Excellent evidence for content, construct, and criterion validity	Appears to perform comparably to the FDI in treatment studies	Not yet established in JFM	PPI Scale found to be useful
FDI	None	Good-to-excellent internal consistency reliability	Excellent evidence for content, construct, and criterion validity	Excellent	Decrease of ≥7.8 points distinguishes treatment responders from non-responders in CBT trial for JFM	Well-established measure
PedsQL 3.0 Rheumatology Module	None	Excellent internal consistency reliability	Excellent evidence for content and convergent validity	Shows improvement following treatment	Not yet established in JFM	Responsive to overall well-being changes. May not be sensitive to treatment specific changes

*PSAT = Pain and Symptom Assessment Tool; FDI = Functional Disability Inventory; PedsQL = Pediatric Quality of Life Inventory – Rheumatology Module