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Using Patient Reported Outcome Measures to Capture the Patient's Voice in Research and Care of Juvenile Idiopathic Arthritis

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Synopsis

Patient reported outcome (PRO) measures provide a valuable window into how patients with juvenile idiopathic arthritis and their parents perceive their functioning, quality of life, and medication side effects in the context of their disease and treatment. Momentum behind adoption of PRO measures is increasing as these patient-relevant tools capture information pertinent to taking a patient-centered approach to health care and research. This article reviews the clinical and research utility of obtaining PROs across domains applicable to the experience of juvenile idiopathic arthritis, and summarizes available patient report and parent-proxy PRO measures. Current challenges and limitations of PRO usage are also discussed.

Keywords

juvenile arthritis; health outcomes; patient-centered care; self report; quality of life; pediatric rheumatology; chronic disease; comparative effectiveness research

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Introduction

Juvenile idiopathic arthritis (JIA) is the most common cause of acquired disability in the United States and the fifth most common chronic childhood disease.[1] Children with JIA experience an unpredictable disease course, with periods of improved disease control intermixed with episodes of flare.[2–4] Over the past decade, assessing patient reported outcomes (PROs) has become increasingly important in the context of clinical care and research centered on JIA. [5–7]

Several factors are driving the evolution of PRO measures and their adoption within pediatric health care and research efforts. Studies in the general population indicate that PROs are predictive of future healthcare utilization for adults and children.[8–10] Measures of health behaviors and mental health status – domains suitable to PRO assessment — can predict future disease activity, as may levels of stress, adequacy of sleep, and availability of coping supports. [11] These issues are often reported by patients, however they are not captured by routine clinical measures. At a system level, capturing a broad range of information about symptoms, side effects, and treatment outcomes is thought to contribute to more patient-centered care, improved patient experience, and potentially better treatments, particularly when this information is used to assess comparative effectiveness. Recognizing this, infrastructure has been developed to support patient-centered outcomes research. In addition, the development and validation of PROs has been prioritized through federal initiatives and investment. [12, 13] Capturing the knowledge and voice of the patient through high quality, standardized and validated measures may provide information to guide interventions and improve disease trajectories.

In this review we provide an overview of the major domains of PROs assessment in JIA – pain, health related quality of life, physical functioning and medication side effects and commonly used measures in these domains – and the rationale for incorporating PROs into JIA clinical care and research.

Juvenile idiopathic arthritis

Currently there are seven categories of JIA as defined by the International League of Associations Rheumatology (ILAR) classification criteria. [14] The JIA subtypes vary with regard to associated clinical features, laboratory studies and severity of disease, however the common clinical feature across the categories of JIA is arthritis in one or more joints presenting prior to sixteen years of age. Like adult patients with arthritis, pediatric arthritis patients experience inflammation of their joints leading to pain, swelling, stiffness and loss of range of motion. Unique to pediatrics is the impact these symptoms can have on physical development (e.g., learning to walk) and social development (e.g., attending school, playing sports). Appropriate assessment of the pediatric patient's experience with arthritis is crucial to understanding the impact of the disease on the patient, their family, disease course and outcomes.

Domains of patient reported outcome measures

Examples of generic and JIA specific PROs by measurement domain are provided in Table 1. When selecting a particular PRO measure for clinical or research use, in addition to considering whether a given domain is represented by a generic or disease-specific measure, factors such as length, target age for administration, response format and recall period may be relevant.

Pain

Assessment of pain has been identified by patients and their parents as one of the most important outcome measures in JIA. [15] For youth with JIA, near daily pain is experienced by the majority of patients, and reports of severe pain are found in some one in four patients. [16] Daily pain is significantly associated with increased functional disability and increased anxiety and other daily symptoms,[16] so understanding factors that contribute to pain and addressing pain may have high clinical benefits. Several psychological factors appear to influence the report of pain among JIA patients with high pain but low disease activity, such as cognitive health beliefs regarding disability and physical harm. [17] Variability in pain among youth with JIA is highly associated with the ability to effectively cope with stress, however it is only minimally related to changes in disease activity.[18, 19] Pain in youth with JIA is a primary determinant of the extent of difficulties these children have with vital socio-emotional functioning including in areas related to physical ability, social life, and academic performance. [18, 19]

Because of the complexity of measuring pain, its assessment often includes evaluation of pain intensity and the extent to which the experience of pain interferes with life activities (pain interference). Common pain measures are shown in Table 1. Measures of pain intensity include the Wong-Baker FACES ®Pain Rating Scale, which can be used to assess pain in patients three years of age and older. Also frequently used is a numerical rating scale or a visual analogue scale response to a simple question such as: “How much pain has your child had because of his/her rheumatic condition in the past week?” Respondents are asked to provide an ordinal numerical rating on a scale of 0 to 10 where 0 is “no pain” and 10 is “very severe pain.” The Pediatric Quality of Life Inventory (PedsQL™), a quality of life measure, includes a Pediatric Pain Questionnaire™ [20–22].

The results of pain assessment in JIA should be interpreted with some caution. Several studies have demonstrated that patients with JIA have a lower pain tolerance or threshold than healthy children or their peers. [23] It is not uncommon for patients whose arthritis is in remission on medications to continue to report pain. [24] Because of the challenge posed by subjectivity for evaluating pain, a trend has developed toward evaluating pain interference with measures such as the Child Activity Limitations Interview-21 (CALI-21) [25]. Another is the National Institutes of Health Patient Reported Outcomes Measurement Information System (NIH PROMIS) Pain Interference measure. This measure ascertains the extent to which the experience of pain interfered with daily activities and tasks in the past week [26]. Providing this type of context around the experience of pain is thought to help clinicians and researchers interpret reports about pain as these measures are grounded in the functional role and impact on activity.

Functional status

The assessment of functional ability and mobility is important for a condition like JIA which can directly impact activities of daily living. The most widely used measure for assessing functional status in JIA is the Childhood Health Assessment Questionnaire (C-HAQ), which assesses functional health status for pediatric patients one to 19 years of age with a chronic rheumatic disease. [27] This measure was originally validated for patients with JIA in 1994. [27] The C-HAQ assesses disability in eight domains including dressing and grooming, arising, eating, walking, hygiene, reach, grip, and activities with respondents selecting the amount of difficulty the child may have with a particular task with 0 being “without any difficulty” and 3 being “unable to do.” Although the C-HAQ is still commonly used in clinical studies and trials for assessing functional impairment in JIA, its utility has been questioned due to its significant ceiling effect, particularly for patients with well-controlled JIA. Revised versions of the C-HAQ have been developed with only modest improvement in the discriminative validity between patients with JIA with minimal disease activity and healthy controls. [28, 29]

Other measures of functional ability used in JIA include the Juvenile Arthritis Functional Assessment Report (JAFAR), Juvenile Arthritis Self-Report Index (JASI), Physical Activity Questionnaire (PAQ), NIH PROMIS Physical Function Instruments, Pediatric Outcomes Data Collection Instrument (PODCI), and Activities Scale for Kids (ASK) (see Table 1). [30] The Children’s Assessment of Participation and Enjoyment (CAPE) has been used to assess participation in leisure activities among JIA patients. [31]

Quality of Life

Health related quality of life (HRQoL) is a multidimensional concept which generally includes the self-assessment of a person’s physical and mental health. Children with JIA report poorer HRQoL than their healthy peers, with decreased HRQoL, even in the setting of low disease activity and treatment with biologic agents. [32–34] The impact of JIA is not confined to childhood as longitudinal, observational cohorts have reported nearly one-half of children with JIA have recurrent or ongoing disease activity on entry into adulthood, with active arthritis, progressive joint destruction, and decreased quality of life.[33, 35–38] Early intervention to improve HRQoL may lessen morbidity and during childhood, possibly increasing the odds of successful participation in age-appropriate activities (e.g., school, independence from parents, employment) with compounding benefit over the life course. [39] Both JIA specific and non-disease specific measures have been used to assess quality of life among JIA patients (Table 1).

Medication side effects

Many patients with JIA require treatment with systemic medications including non-steroidal anti-inflammatories, disease modifying antirheumatic drugs (DMARDs), such as methotrexate, and biologic therapies. Depending on the severity of the illness the medication burden can be high, which can impact adherence to the treatment regimen, HRQoL and disease outcomes. [40] The patient and family experience with medication administration and potential side effects may impact adherence. This is particularly important to understand as it relates to comparative effectiveness studies, where it needs to be determined if a group

of patients are not responding optimally to a medication due to inadequate response versus intolerance. Utilizing the self-administered Gastrointestinal Symptom Scale for Kids (GISSK) tool, Brunner *et al.* found that over half of JIA patients on second-line medications have gastrointestinal symptoms. When compared to clinically similar patients without GI symptoms, patients with GI symptoms had a lower HRQoL. [41] Methotrexate intolerance also occurs frequently among JIA patients. [42] A validated questionnaire called the Methotrexate Intolerance Severity Score (MISS) has been developed to assess this common clinical issue (see Table 1). [42]

Composite Patient Reported Outcome Measures Specific to JIA

The majority of the PROs previously discussed are “disease generic” and have been used to measure outcomes across rheumatic diseases. In contrast, the Juvenile Arthritis Multidimensional Assessment Report (JAMAR) relies solely on parent or patient reports to assess PROs in JIA. The JAMAR includes 15 parent- or patient-reported measures/items that assess functional status (utilizing the JAFAR), pain, HRQOL (utilizing the PRQL), well-being, joint symptoms including stiffness, pain and swelling, assessment of extraarticular symptoms (fever and rash), level of disease activity, rating of disease status (remission, continued activity, relapse), rating of disease course as compared to the prior visit, listing of medications, description of medication side effects, difficulties with medication administration, school problems related to JIA, and satisfaction with the illness outcome. [43] The JAMAR can be completed by parent-proxy report or by patient report for patients 7–18 years of age. In a study of 940 JIA patients, the JAMAR was found to be feasible and to have face and content validity. Parent and patient reports were concordant and the measure performed well across age group and JIA categories. Although the authors could not identify any studies describing the use of the JAMAR in routine clinical settings it is being studied in a novel multinational collaborative effort to study JIA outcomes. [44]

Use of the PROMIS Pediatric Measures in JIA

The recently developed NIH PROMIS Pediatric measures include several psychosocial and behavioral domains with relevance across a range of chronic diseases and conditions. [45] Pediatric measures with item banks and scoring algorithms are available in formats suitable for paper or computer-assisted administration so that patients can enter PROs electronically. Importantly, the PROMIS measures capture a range of domains relevant to JIA including measures that assess physical, mental and social health. [46] Not all PROMIS measures have been validated for JIA, but the system is growing and considerable attention continues to be paid to validation of new measures against clinical anchors (to establish responsiveness) and other similar measures (to establish convergent validity). Investment in standardization and validation of PRO measures and integration of high quality item banks for clinical epidemiology and post-marketing surveillance enables both scientific rigor and patient voice/annotation.[5, 6] The available PROMIS measures can be obtained at: <http://www.nihpromis.org/measures/availableinstruments>.

Potential Challenges with Pediatric PROs for JIA

There are several potential challenges to utilizing PROs for JIA. One of the challenges is that JIA patients can be affected across a wide age range. For example, oligoarticular JIA, which

is the most common subtype of JIA, has a peak age of onset in the toddler and preschool years. In contrast, rheumatoid factor positive polyarticular JIA has its peak incidence in adolescence. The use of various PRO measures in JIA may vary depending on the patient age and development and the appropriateness of patient versus parent/proxy report measures.

Rheumatology providers also need to be aware that the explanatory power and levels of PROs vary across the categories of JIA. A recent study by Taxter *et al.* demonstrated that patients with enthesitis-related arthritis (ERA) and undifferentiated JIA reported higher pain and poorer quality of life than patients with other types of JIA, including polyarticular rheumatoid factor positive JIA, which is similar to adult rheumatoid arthritis (RA) [47]. Poorer physical function was reported among patients with polyarticular JIA, ERA, and undifferentiated JIA. The variability of PROs based on JIA categories suggests that even within JIA, no one size fits all.

Another potential issue is that patients' experience of their disease state and HRQoL may be quite different from the perceptions of external observers.[48] Hence, care is needed when collecting and interpreting PROs. Some measures, including select PROs developed under the NIH PROMIS, provide versions that are calibrated and validated for patient- versus parent proxy-report. While dyadic reports may track each other, they may differ with regard to intensity, severity, or clinical implications of a problem. [49] Pediatric patients may provide a more favorable evaluation of their wellbeing than their parents, due to the inherent resilience and optimism represented in a child's point of view. [50] Conversely, parents' worry, anxiety and burden of healthcare decision making may lead them to describe their child's wellbeing including disease state, symptoms and experience of medications, in worse terms than their child reports. For treating clinicians, this can be a confusing picture. Similarly, investigators undertaking longitudinal cohort research may need to consider the potential for subtle reporting bias due to a change in the respondent, as when a child starts self-reporting on a measure that heretofore had been completed by a parent. A related issue is that patients/families may attend to and prioritize aspects of their illness and treatment experiences differently than their clinicians. Discordance between parent/patient and physician reported ratings of functional ability, pain, and assessment of inactive disease in JIA have been well described, with the physician tending to have a more optimistic view of these measures than the patient or family. [51–53] At this point it is not clear how to address these potential issues of discordance.

PROs for the management and study of JIA

Clinicians who want to integrate PROs into practice, and researchers aiming to include PROs in cohort studies and clinical trials, may need to consider nuanced issues related to measurement focus, burden (length), periodicity, point of view (parent and/or child report), developmental relevance, and clinical validity. Although the field is still evolving, an emerging body of literature offers insight into these issues.

Use of PROs in Clinical Care

As work to clinically validate PRO measures matures, opportunities are increasing to utilize PROs as part of the routine clinical care of JIA patients. Clinical validity has not been established for all PROs, however. The development of more robust electronic health records (EHRs) should allow for increased utilization of PROs in the clinical setting and for making “real-time” decisions regarding patient response to treatments and interventions. Similarly, opportunities are growing to add the voice of the patient to clinically reported measures through mHealth tools and Web-enabled information technologies that may interoperate with EHRs or complement them. [54] Interestingly, Dijkstra *et al.* recently described the use of patient reported joint counts in JIA; there was only moderate correlation between patient and physician-reported joint counts as patients with JIA tended to overestimate the number of active joints. [55] The authors suggest that based on these results, self-reported joint counts should not be used in lieu of clinical assessment.

Use of PROs in Various Research Settings

In pediatric rheumatology, clinical treatment trials and comparative effectiveness research (CER) using observational cohorts are frequently utilized to study medication efficacy and safety. Because the voice of the patient is largely missing in these data sources, key efficacy endpoints, including aspects of HRQoL, may not factor into clinical decision making and policy recommendations regarding treatments. For JIA, psychosocial stress, functional status, and even experience of problems/side effects from medications, and adverse events, including those that fall below the threshold for a typical healthcare encounter, may influence adherence and treatment outcomes. Hence measuring these factors may greatly enhance knowledge of treatment efficacy, safety, and disease progression.

PROs are also important in clinical research, particularly for assessing long-term outcomes when a patient may not be able to be seen in a clinical setting and the assessment relies on patient report. An example is a study by Swarup *et al.* which examined implant survival and patient-reported outcomes after total hip arthroplasty (THA) in JIA patients. [56] The patients were assessed a mean of 12 years after hip replacement using a survey which included a patient-reported hip disability and osteoarthritis outcomes score (HOOS). In multivariate analysis the HOOS scores were associated with important patient characteristics (such as implant type and history of a THA revision) and outcomes.

While inclusion of the patient perspective regarding response to therapy and symptoms through measuring PROs has gained increasing recognition by clinicians and researchers who strive to understand treatment outcomes and the comparative effectiveness of therapies, researchers still need to choose among the broad range of available PROs to measure these factors. Choice will be governed by the treatment model being explored, time available to collect data from a patient (which may require parsimony), and the reporting period for various measures. For example, intervening to address depression, anxiety and coping skills that contribute to the experience of pain may require a somewhat long-term time horizon for assessing outcomes since changes in these factors may take time to manifest. In contrast, detection of change in pain interference or inflammation may take a relatively short time to investigate with regards to effects of medications.

Conclusions

Significant momentum exists for adopting PRO measures within JIA clinical care and research. Use of these measures may help advance the patient experience by ensuring that the voice and knowledge of the patient and parent proxy are incorporated into problem and progress appraisal and healthcare decision making. Including PROs in studies of comparativeness effectiveness can capture the acceptability and impacts of therapies from the patient's point of view, and thus lead to more meaningful patient outcomes. While progress has been made, much remains to be learned about PRO usage, particularly in the areas of interpretation and clinical response in JIA. Not all PROs have been clinically validated and responsiveness of a given measure to treatment changes and shifts in disease course may not be established. The choice of measures can be confusing where multiple measures for assessing pain, quality of life and other domains exist. Nuance, sensitivity and consideration of the developmental status of the patient are needed when choosing between patient and parent-proxy report and when mounting longitudinal studies in which a shift from parent-proxy to patient report over the course of a study can confound observations and introduce reporting bias. Although the use of PROs is a relatively new phenomenon, considerable investment in high-quality validation work, including testing against known clinical anchors and in relation to "gold standard" measures, offers the potential for greatly advancing meaningful integration of the patient voice and experience within health care and research settings. This may be particularly on point for a chronic disease like JIA, where lifelong patient engagement, adherence and shared decision making are prerequisites for favorable outcomes.

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Key Points

- Incorporating patient reported outcome (PRO) measures into routine clinical care of patients with juvenile idiopathic arthritis can help facilitate movement from physician-centered to patient-centered care.
- PRO measures relevant to juvenile idiopathic arthritis provide information germane to evaluating treatment outcomes and comparative effectiveness of therapies.
- Valid and reliable PRO measures are available that capture the experience of juvenile idiopathic arthritis from the perspective of the patient and parent.
- Length, age, potential discordance between parent and child responses, and clinical validity are among the issues that need to be considered when selecting PRO measures.

Table 1

Pediatric patient reported outcome measures for JIA

Domain	Measure	Number of items	Ages of administration		Time frame
			Self-report	Parent-proxy report	
Pain interference	Child Activity Limitations Interview-21 (CALI-21) [25]	21	8-18	8-18	Past month
	PROMIS® Short Form v1.0 – Pain Interference 8a [26, 46]	8	8-17	5-17	Past week
Pain intensity	Faces scales (e.g., Faces Pain Scale-Revised [57]) [58]	1	>5	n/a	Current
	Numerical rating scales (NRS) [59]	1	>8	Any	Varies
	PedsQL™ Pediatric Pain Questionnaire (PPQ)™ [20–22]	3	8–18	5–18	Past week, current
	Visual analogue scales (VAS) [60]	1	>7	Any	Varies
Quality of life	Child Health Questionnaire (CHQ) [61, 62]	87 (self-report version), 28 or 50 (parent versions)	10–18	5–18	Varies by question
	Children's Assessment of Participation and Enjoyment (CAPE) [31, 63]	55	6-21	n/a	Past 4 months
	Juvenile Arthritis Quality of Life Questionnaire (JAQQ) [64]	74	>9	2–18	Past 2 weeks
	Pediatric Quality of Life (PedsQL)™4.0 Generic Core Scales [65–67]	21–23, depending on age	5–18	2–18	Past month
	PedsQL™3.0 Rheumatology Module [66]	14–22, depending on age	5–18	2–18	Past month (standard version), past week (acute version)
	Pediatric Rheumatology Quality of Life Scale	10	7–18	2–18	Past month

Domain	Measure	Number of items	Ages of administration		Time frame
			Self-report	Parent-proxy report	
	(PQQL) [68, 69]				
Physical function or activity	Activities Scale for Kids (ASK) [70]	30	5–15	n/a	Past week
	Childhood Health Assessment Questionnaire (C-HAQ) [27, 71, 72]	30 (simplified version assessing disability without aids/devices and help items [73]) 2 (VAS of pain and overall well-being)	9–19	1–8	Past week
	Juvenile Arthritis Functional Assessment Report (JAFAR) [72, 74]	23	7–18	7–18	Past week
	Juvenile Arthritis Self-Report Index (JASI) [72, 75, 76]	100	8–18	n/a	Current
	Physical Activity Questionnaire (PAQ) [77, 78]	9–10, depending on age	Grades 4–12 (approx. ages 8–20)	n/a	Past week
	Pediatric Outcomes Data Collection Instrument (PODCI) [79, 80]	83–86, depending on age	11–18	2–18	Varies by question
	PROMIS® Short Form v1.0 Physical Function Upper Extremity and Mobility instruments) [26]	8	8–17	5–17	Current
	Gastrointestinal Symptom Scale for Kids (GISSK) [41]	9 (GI symptoms) 1 (VAS of symptom severity)	8–18	2–18	Past week
	Methotrexate Intolerance Severity Score (MISS) questionnaire [42, 81]	12 (modified version)	12–18	1–18	Past month