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

Aimee O. Hersh, MDa,*, Parissa K. Salimian, BAb, Elissa R. Weitzman, ScD, MSc,c,d,e

KEYWORDS

- Juvenile arthritis • Health outcomes • Patient-centered care • Self-report
- Quality of life • Pediatric rheumatology • Chronic disease
- Comparative effectiveness research

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 patients and parents.
- 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.

Research reported in this publication was supported by the National Library of Medicine and by the National Institute of Arthritis and Musculoskeletal and Skin Diseases of the National Institutes of Health under award numbers R01LM011185 and U19AR069522, respectively, to Dr E.R. Weitzman and the National Institute of Arthritis and Musculoskeletal and Skin Diseases of the National Institutes of Health under award number K23AR066064 to Dr A. Hersh. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health.

a Pediatric Rheumatology, University of Utah, 81 Mario Capecchi Way, 4th Floor, Salt Lake City, UT 84113, USA; b Division of Developmental Medicine, Boston Children’s Hospital, 300 Longwood Avenue BCH3185, Boston, MA 02115, USA; c Division of Adolescent/Young Adult Medicine, Boston Children’s Hospital, 300 Longwood Avenue BCH3187, Boston, MA 02115, USA; d Department of Pediatrics, Harvard Medical School, 300 Longwood Avenue BCH3187, Boston, MA 02115, USA; e Computational Health Informatics Program, Boston Children’s Hospital, 300 Longwood Avenue BCH3187, Boston, MA 02115, USA

* Corresponding author.

E-mail address: aimee.hersh@hsc.utah.edu

http://dx.doi.org/10.1016/j.rdc.2016.01.004
rheumatic.theclinics.com

0889-857X/16/$ – see front matter © 2016 Elsevier Inc. All rights reserved.
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. Children with JIA experience an unpredictable disease course, with periods of improved disease control intermixed with episodes of flare. Over the past decade, assessing patient-reported outcomes (PROs) has become increasingly important in the context of clinical care and research centered on JIA.

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 health care utilization for adults and children. 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. 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. Capturing the knowledge and voice of patients through high-quality, standardized, and validated measures may provide information to guide interventions and improve disease trajectories.

In this review, the authors provide an overview of the major domains of PRO 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 7 categories of JIA as defined by the International League of Associations for Rheumatology’s classification criteria. 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 before 16 years of age. Like adult patients with arthritis, pediatric patients with arthritis 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 (eg, learning to walk) and social development (eg, attending school, playing sports). Appropriate assessment of pediatric patients’ experience with arthritis is crucial to understanding the impact of the disease on patients, their families, 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.
Assessment of pain has been identified by patients and their parents as one of the most important outcome measures in JIA.\textsuperscript{15} For youth with JIA, most patients experience near-daily pain and reports of severe pain are found in one in 4 patients.\textsuperscript{16} Daily pain is significantly associated with increased functional disability and anxiety and other daily symptoms,\textsuperscript{16} so understanding factors that contribute to pain and addressing pain may have high clinical benefits. Several psychological factors seem to influence the report of pain among patients with JIA with high pain but low disease activity, such as cognitive health beliefs regarding disability and physical harm.\textsuperscript{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.\textsuperscript{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.\textsuperscript{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 3 years of age and older. Also frequently used is a numerical rating scale or a visual analog scale response to a simple question such as: How much pain has your child had because of his or 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, a quality-of-life measure, includes a Pediatric Pain Questionnaire.\textsuperscript{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.\textsuperscript{23} It is not uncommon for patients whose arthritis is in remission on medications to continue to report pain.\textsuperscript{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.\textsuperscript{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.\textsuperscript{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 1 to 19 years of age with a chronic rheumatic disease.\textsuperscript{27} This measure was originally validated for patients with JIA in 1994.\textsuperscript{27} The C-HAQ assesses disability in 8 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 because of its significant ceiling
<table>
<thead>
<tr>
<th>Domain</th>
<th>Measure</th>
<th>Number of Items</th>
<th>Ages of Administration</th>
<th>Self-Report</th>
<th>Parent-Proxy Report</th>
<th>Time Frame</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pain interference</td>
<td>Child Activity Limitations Interview-21</td>
<td>21</td>
<td></td>
<td>8–18 y</td>
<td>8–18 y</td>
<td>Past month</td>
</tr>
<tr>
<td></td>
<td>PROMIS Short Form v1.0 – Pain Interference 8a</td>
<td>8</td>
<td></td>
<td>8–17 y</td>
<td>5–17 y</td>
<td>Past week</td>
</tr>
<tr>
<td>Pain intensity</td>
<td>Faces scales (eg, Faces Pain Scale-Revised)</td>
<td>1</td>
<td></td>
<td>&gt;5 y</td>
<td>not applicable</td>
<td>Current</td>
</tr>
<tr>
<td></td>
<td>Numeral rating scales</td>
<td>1</td>
<td></td>
<td>&gt;8 y</td>
<td>Any</td>
<td>Varies</td>
</tr>
<tr>
<td></td>
<td>PedsQL Pediatric Pain Questionnaire</td>
<td>3</td>
<td></td>
<td>8–18 y</td>
<td>5–18 y</td>
<td>Past week, current</td>
</tr>
<tr>
<td></td>
<td>VAS</td>
<td>1</td>
<td></td>
<td>&gt;7 y</td>
<td>Any</td>
<td>Varies</td>
</tr>
<tr>
<td>Quality of life</td>
<td>Child Health Questionnaire</td>
<td>87 (self-report version), 28 or 50 (parent versions)</td>
<td></td>
<td>10–18 y</td>
<td>5–18 y</td>
<td>Varies by question</td>
</tr>
<tr>
<td></td>
<td>Children’s Assessment of Participation and Enjoyment</td>
<td>55</td>
<td></td>
<td>6–21 y</td>
<td>not applicable</td>
<td>Past 4 mo</td>
</tr>
<tr>
<td></td>
<td>Juvenile Arthritis Quality of Life Questionnaire</td>
<td>74</td>
<td></td>
<td>&gt;9 y</td>
<td>2–18 y</td>
<td>Past 2 wk</td>
</tr>
<tr>
<td></td>
<td>PedsQL 4.0 Generic Core Scales</td>
<td>21–23, depending on age</td>
<td></td>
<td>5–18 y</td>
<td>2–18 y</td>
<td>Past month</td>
</tr>
<tr>
<td></td>
<td>PedsQL 3.0 Rheumatology Module</td>
<td>14–22, depending on age</td>
<td></td>
<td>5–18 y</td>
<td>2–18 y</td>
<td>Past month (standard version), past week (acute version)</td>
</tr>
<tr>
<td></td>
<td>Pediatric Rheumatology Quality of Life Scale</td>
<td>10</td>
<td></td>
<td>7–18 y</td>
<td>2–18 y</td>
<td>Past month</td>
</tr>
<tr>
<td>Physical function or activity</td>
<td>Activities Scale for Kids\textsuperscript{27}</td>
<td>30</td>
<td>5–15 y</td>
<td>not applicable</td>
<td>Past week</td>
<td></td>
</tr>
<tr>
<td>------------------------------</td>
<td>-----------------------------------------------</td>
<td>----</td>
<td>--------</td>
<td>--------------</td>
<td>----------</td>
<td></td>
</tr>
<tr>
<td></td>
<td>C-HAQ\textsuperscript{27,71,72}</td>
<td>30 (simplified version assessing disability without aids/devices and help items\textsuperscript{27})</td>
<td>9–19 y</td>
<td>1–8 y</td>
<td>Past week</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>2 (VAS of pain and overall well-being)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>7–18 y</td>
<td>7–18 y</td>
<td>Past week</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Juvenile Arthritis Functional Assessment Report\textsuperscript{72,74}</td>
<td>23</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Juvenile Arthritis Self-Report Index\textsuperscript{72,75,76}</td>
<td>100</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Physical Activity Questionnaire\textsuperscript{77,78}</td>
<td>9–10, depending on age</td>
<td>Grades 4–12 (approx 8–20 y)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pediatric Outcomes Data Collection Instrument\textsuperscript{79,80}</td>
<td>83–86, depending on age</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>PROMIS Short Form v1.0 Physical Function (Upper Extremity and Mobility instruments)\textsuperscript{26}</td>
<td>8</td>
<td>8–17 y</td>
<td>5–17 y</td>
<td>Current</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Medication side effects</td>
<td>Gastrointestinal Symptom Scale for Kids\textsuperscript{41}</td>
<td>9 (GI symptoms)</td>
<td>8–18 y</td>
<td>2–18 y</td>
<td>Past week</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Methotrexate Intolerance Severity Score questionnaire\textsuperscript{42,81}</td>
<td>1 (VAS of symptom severity)</td>
<td>12 (modified version)</td>
<td>12–18 y</td>
<td>1–18 y</td>
<td>Past month</td>
</tr>
</tbody>
</table>

Abbreviations: approx, approximately; C-HAQ, Childhood Health Assessment Questionnaire; PedsQL, Pediatric Quality of Life; PROMIS, Patient Reported Outcomes Measurement Information System; VAS, visual analog scale.
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, Physical Activity Questionnaire, NIH PROMIS Physical Function Instruments, Pediatric Outcomes Data Collection Instrument, and Activities Scale for Kids (see Table 1).30 The Children’s Assessment of Participation and Enjoyment has been used to assess participation in leisure activities among patients with JIA.31

**Quality of Life**

Health-related quality of life (HRQoL) is a multidimensional concept that 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 biological 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 (eg, 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 patients with JIA (see Table 1).

**Medication Side Effects**

Many patients with JIA require treatment with systemic medications, including nonsteroidal antiinflammatories, disease-modifying antirheumatic drugs, such as methotrexate, and biological 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 patients’ and families’ experience with medication administration and potential side effects may impact adherence. This point is particularly important to understand as it relates to comparative effectiveness studies when it needs to be determined if a group of patients are not responding optimally to a medication because of inadequate response versus intolerance. Using the self-administered Gastrointestinal Symptom Scale for Kids tool, Brunner and colleagues found that more than half of patients with JIA on second-line medications have gastrointestinal (GI) 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 patients with JIA.42 A validated questionnaire called the Methotrexate Intolerance Severity Score has been developed to assess this common clinical issue (see Table 1).42

**Composite Patient-Reported Outcome Measures Specific to Juvenile Idiopathic Arthritis**

Most 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 (using the JAFAR); pain; HRQoL (using the Pediatric Rheumatology Quality of Life scale); well-being; joint symptoms, including stiffness, pain, and
swelling; assessment of extra-articular symptoms (fever and rash); level of disease activity; rating of disease status (remission, continued activity, relapse); rating of disease course as compared with 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. The JAMAR can be completed by parent-proxy report or by patient report for patients 7 to 18 years of age. In a study of 940 patients with JIA, 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 investigators 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.

Use of the Patient Reported Outcomes Measurement Information System Pediatric Measures in Juvenile Idiopathic Arthritis

The recently developed NIH PROMIS pediatric measures include several psychosocial and behavioral domains with relevance across a range of chronic diseases and conditions. 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. 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 postmarketing 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 Patient-Reported Outcomes for Juvenile Idiopathic Arthritis

There are several potential challenges to using PROs for JIA. One of the challenges is that patients with JIA 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 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 and colleagues 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. 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. 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. Although dyadic reports may track each
other, they may differ with regard to intensity, severity, or clinical implications of a problem.\textsuperscript{49} Pediatric patients may provide a more favorable evaluation of their well-being than their parents because of the inherent resilience and optimism represented in a child’s point of view.\textsuperscript{50} Conversely, parents’ worry, anxiety, and burden of health care decision making may lead them to describe their child’s well-being, 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 patients or families.\textsuperscript{51–53} At this point it is not clear how to address these potential issues of discordance.

**PATIENT-REPORTED OUTCOMES FOR THE MANAGEMENT AND STUDY OF JUVENILE IDIOPATHIC ARTHRITIS**

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 Patient-Reported Outcomes in Clinical Care*

As work to clinically validate PRO measures matures, opportunities are increasing to use PROs as part of the routine clinical care of patients with JIA. 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 patients to clinically reported measures through mHealth (mobile health) tools and Web-enabled information technologies that may interoperate with EHRs or complement them.\textsuperscript{54} Interestingly, Dijkstra and colleagues\textsuperscript{55} 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. The investigators suggest that based on these results, self-reported joint counts should not be used in lieu of clinical assessment.

*Use of Patient-Reported Outcomes in Various Research Settings*

In pediatric rheumatology, clinical treatment trials and comparative effectiveness research using observational cohorts are frequently used to study medication efficacy and safety. Because the voices of patients are largely missing in these data sources, key efficacy end points, including aspects of HRQoL, may not factor into clinical decision making and policy recommendations regarding treatments. For JIA, psycho-social stress, functional status, and even experience of problems/side effects from medications and adverse events, including those that decrease below the threshold...
for a typical health care encounter, may influence adherence and treatment outcomes. Hence, measuring these factors may greatly enhance the 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 a patient report. An example is a study by Swarup and colleagues, which examined implant survival and patient-reported outcomes after total hip arthroplasty (THA) in patients with JIA. The patients were assessed a mean of 12 years after hip replacement using a survey that 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.

Although inclusion of patients’ 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 patients (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 because 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 regard to effects of medications.

**SUMMARY**

Significant momentum exists for adopting PRO measures within JIA clinical care and research. Use of these measures may help advance patients’ experiences by ensuring that the voice and knowledge of the patient and parent proxy are incorporated into problem and progress appraisal and health care decision making. Including PROs in studies of comparative effectiveness can capture the acceptability and impacts of therapies from the patients’ point of view and, thus, lead to more meaningful patient outcomes. Although 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 patients 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 patients’ voices and experience within health care and research settings. This idea may be particularly on point for a chronic disease like JIA, whereby lifelong patient engagement, adherence, and shared decision making are prerequisites for favorable outcomes.
REFERENCES


