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ORIGINAL ARTICLE

Facilitating patient-centered care: the development of illustrated multidimensional patient-reported outcome measures for children/adolescents with juvenile idiopathic arthritis

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Abstract

Objectives To assess the validity, reliability, comprehensibility, and responsiveness to change of an illustrated child/parent multidimensional patient-reported outcome measures (PROMs) questionnaire which can assess construct outcome measures of children with juvenile inflammatory arthritis.

Methods A total of 122 children with juvenile idiopathic arthritis were included in this work in a multicenter study. The questionnaire included seven categories: (1) functional ability; (2) health-related quality of life; (3) disease activity measures: pain, global assessment, fatigue, and morning stiffness; (4) self-reported joint tenderness; (5) current medication, side effects, and adherence to therapy; (6) comorbidities; and (7) patient motivation. All the items were supported by illustrations to provide children with a visual impression of what was meant by the questions. The questionnaire has parent and patient versions. The disease activity status was assessed using JADAS-27.

Results The questionnaire was reliable as demonstrated by a high-standardized alpha (0.890–0.978). The questionnaire items correlated significantly ($p < 0.01$) with clinical parameters of disease activity. The patient-reported tender joints correlated significantly with the physician's scores (0.842). Changes in functional disability, quality of life, and the motivation score showed significant variation ($p < 0.01$) with disease activity status in response to therapy. The illustrated PROMs questionnaire showed also a high degree of comprehensibility (9.6).

Conclusions Integrating PROMs into standard clinical practice is feasible and applicable. The illustrated questionnaire was valid and reliable. It provides an informative, quantitative measure for the disease activity score set data, and in the meantime, facilitates the assessment of the children's adherence to therapy, comorbidities, and motivation on an individual basis.

Keywords c-PROMs · Functional ability · Health-related quality of life · JADAS

Introduction

Juvenile idiopathic arthritis (JIA) is a chronic disease characterized by persistent inflammatory changes in the synovial joints with consequent structural joint damage [1]. Nonreversible pathological changes may also occur in extra-articular organs, such as the eye or kidney, or may result from adverse effects of drug therapies [2]. This morbidity may impair the children's functional ability as well as the quality of life and has a negative impact on their willingness to continue taking the medical management [3].

The main targets of JIA management are to halt the inflammatory arthritic joint disease, achieve a state of remission, minimize the comorbidity risk, prevent organ damage, and improve the child's functional ability as well as the quality

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of life. These goals can be attained by a regular monitoring of disease activity status as well as the child's health status. This has been facilitated through the use of patient-reported outcomes [4, 5]. The incorporation of these assessments in daily care requires the use of simple and feasible tools that are patient friendly and easy to use in busy clinics.

In the past few years, numerous outcome measures have been developed and validated for use in JIA, including methods for scoring disease activity and joint damage, therapeutic response criteria, and questionnaires for the estimation of physical functioning and HRQL [6, 7]. Recently, there has been an increased focus on parent- and child-reported outcomes [6, 8–10]. These instruments are considered valuable as they capture the parent and child perception of disease state and management outcomes [11].

Traditionally, patient-reported outcomes (PROs) have been used at a group level in research and quality improvement projects. Challenges to implementing PROs in standard clinical practice, particularly for children, included the tool's comprehensibility, length, and items. Furthermore, several available tools are unidimensional. The ideal patient-reported outcome measures tool should be children friendly and easy to be completed by the parent and/or the child in a short time [12]. The ideal tool should not only focus on the disease activity assessment and possible side effects of the medical therapy but also measure the health-related quality of life measures and psychological, school, social, and behavior issues. It should also be able to assess comorbidity risk and patient's motivation. In addition, the tool should be valid, reliable, and responsive to change [13].

It was assumed that an illustrated version of the PROMs questionnaire would be more comprehensive and motivating for the child or parents to provide the useful information desired to prescribe the most convenient management.

The aim of this study was to assess validity, reliability, and responsiveness to change of an illustrated and comprehensive child/parent multidimensional patient-reported outcome measures (c-PROMs) questionnaire which can assess construct outcome measures of children with JIA and to assess the cognitive value of the illustrated questionnaire, in comparison to the non-illustrated version of the tool, as well as its value in improving the quality of the data collected.

Methods

Patients Eligible patients were male or female children, aged 3–16 years diagnosed within the last 6 months with any type of JIA as per the International League of Associations for Rheumatology (ILAR) [14], irrespective of the specific JIA category. Patients who carried the diagnosis of a medical condition that would otherwise severely impair their health-related quality of life (HRQoL) (e.g., cerebral palsy, spina bifida, severe mental retardation, fibromyalgia) were excluded from study participation.

Study design The work was a multicenter, prospective study. The patients' questionnaire was incorporated into the routine clinical practice. Consecutive patients eligible for the study were approached as soon as a diagnosis of JIA was established. The patients were treated according to the national guidelines [15].

Local ethical and methodological protocols for approval of the study were followed. All the patients who participated in the study signed an informed consent according to the Declaration of Helsinki (at the General Assembly in October 2008) and ensured that their data is kept confidential after explaining the benefit and potential null risk of the study.

The illustrated multidimensional PROMs questionnaire

The questionnaire was constructed to assess the patient-reported outcomes and was presented in a self-administered format. An explanatory cartoon was selected by a panel of four (a rheumatologist, nurse, occupational therapist, and 2 patients) for each of the items, with the aim of visually supporting the comprehensibility and making the tool patient friendly. All the patients were asked to fill out the illustrated multidimensional PROMs questionnaire, while sitting in the waiting area prior to their assessment in the rheumatology clinic. The illustrations were aimed at providing children with a visual impression of what was meant by the questions they had to answer. The questions were phrased as self-report statements and children's approval of the statements was assessed using a 4-point rating scale or a visual analogue scale 0–100. The questionnaire included the following:

1. Illustrated functional disability: physical function was assessed by using illustrated Childhood Health Assessment questionnaire [16]. The CHAQ comprises two indices: disability and discomfort [7]. The total score ranges from 0 to 3. Discomfort is determined by the presence of pain as well as child/parent global assessment. Both were measured using a numeric visual analogue scale (VAS) on a 21-numbered circle VAS [11].
2. Assessment of HRQoL: this reflects the individual's perceptions of their physical, psychosocial, and role functioning with respect to their health [17]. The questionnaire included 10 questions, five items to assess physical health, whereas another five items assessed the child's psychosocial health. Assessment of the child ability was carried out using the 4-point Likert response, referring to the prior month, as "never" (score = 0), "sometimes"

(score = 1), "most of the time" (score = 2), and "all the time" (score = 3). A "not assessable" column was included in the parent version of the questionnaire to designate questions that cannot be answered because of developmental immaturity, and this item was not included in the final score calculation. The total HRQoL score ranges from 0 to 30 with the higher scores indicating worse HRQoL. A separate score for physical health as well as psychological health (range 0–15) can be calculated [12–14].

3. Assessment of the level of disease activity VAS using a 21-numbered circle VAS.
4. Global rating of disease status at the time of the visit assessed using a categorical scale.
5. Global evaluation of the disease course in contrast to the last visits using a categorical scale.
6. Assessment of morning stiffness (present/absent) and duration if the patient feels stiff in the morning.
7. Patient self-reported joint tenderness.
8. Report of school/university/work problems caused by the disease (list of items).
9. Current *arthritis medications*: checklist of the medications the patient is taking (list of DMARDs, biologic therapy, anti-inflammatory, and pain killers).
10. *Side effects*: checklist of side effects of medications for the patient to tick.
11. Medication adherence: the child/parent is asked if the treating doctor has asked the patient to stop certain medications, if yes, what was the reason. The patient is then asked if there were any reasons for not taking the medicine and lastly, the patient is asked to rate how regular he is taking his DMARDs medication. This is carried out using a numeric VAS using a 21-numbered circle VAS where "0" stands for not taking the medications and "10" stands for taking the medication regularly as prescribed [11].
12. Report of difficulties with medication administration: this was in the form of a list of items to tick including the following: I do not want to take my medications, difficult to take the medicine in the school, I take too many medications, afraid of the side effects, other reasons (to be mentioned by the patient).
13. *Comorbidity assessment*: a list of possible comorbidities and extra-articular manifestations: a list of items for the child/parent to tick any relevant item.
14. *Patient motivation questionnaire*: 10-items evaluated using a numeric VAS using a 5-point Likert scale. These include the following: (1) I am aware of the nature of my condition, the reasons for the symptoms, the course it runs, and the consequences if left untreated; (2) I have knowledge of my personal care plan, the treatment options available to me, understand how each treatment I am taking works, and the possible side effects I might get; (3) Regardless of whether I feel down, tired, or anxious, I am still able to cope, and not to let my illness stop me from doing things I want to do; (4) I am aware of my role in my own care, feel/able to share in the treatment decision-making, and know when to seek medical advice; (5) I would not be worried about discussing any question I may have regarding my condition or treatment with my doctor/nurse; (6) I am confident I can take any tablet and/or administer any injection prescribed for me; (7) I can self-manage my condition, ease the symptoms, and overcome some of the difficulties which might develop in association with my illness between doctors/hospital visits; (8) I am happy that I can find a way around any new problem related to my condition, and how to solve it to prevent it from interfering with my everyday activities; (9) I am confident that I will be able to continue with any changes I make to my lifestyle such as diet, work, exercise, drinking, or smoking, even if it gets tough; (10) I am confident I can find reliable and useful sources of information to educate myself about my condition, treatment, and health choices [18].

Clinical evaluation

Full history, including disease duration, assessment for articular as well as extra-articular manifestations, revision of the current medications, and assessment for possible comorbidities, was carried out for every patient. Each patient was then subjected to a full clinical examination to assess the parameters of disease activity including the number of tender and swollen joints, the physician overall assessment score, and the presence of extra-articular signs.

Each patient had a blood check for erythrocyte sedimentation rate (ESR) and CRP levels, rheumatoid factor. The ESR was measured using Westergren's method and CRP using the ELISA technique.

Disease activity assessment: This was carried out by

(a) Assessment of the tender joint count: by scoring tenderness to pressure and joint manipulation on physical examination; the types of tenderness are collapsed into a single tender versus non-tender dichotomy for each point. The scores for each patient were summed over 27 joints.
(b) Swollen joint count: analogous to tender joint count, the scores of 27 joints for each patient were summed.
(c) The physician's global health assessment of disease activity measured on a continuous 0–10 VAS.
(d) Disease activity score (JADAS-27). The JADAS [19] includes the following four measures: the physician's global assessment of disease activity, measured on a 0–10 VAS where 0, no activity and 10, maximum activity;

the parent global assessment of well-being, measured on a 0–10 VAS where 0, very well and 10, very poor; the ESR normalized to a 0 to 10 scale; and a count of joints with active disease. Cutoff points for oligoarthritis, < 1 (inactive); 1.1–2 (low disease activity); 2.1–4.2 (moderate disease activity); and > 4.2 (high disease activity). For polyarthritis, < 1 (inactive); 1.1–3.8 (low disease activity); 3.9–8.5 (moderate disease activity); and > 8.5 (high disease activity) [20]. The JADAS ranges 0–57.

Validation

The routine clinic was used as the setting for the questionnaire evaluation. All patients/parents were asked to complete the PROMs questionnaire while sitting in the waiting area before being examined by the treating physician. A supervising nurse was present to provide help, if needed.

Construct validity Construct validity was assessed by comparing the response of the questionnaire administration and five established measures of disease activity: (1) disease activity score: JADAS-27, (2) disease activity status (VAS), (3) child's/parent global assessment score, (4) physician global assessment score, and (5) patient motivation score [21].

Internal consistency Internal consistency was measured by Cronbach's alpha, the intraclass correlation (the correlation between two scales should be lower than their reliability coefficients, as measured by Cronbach's alpha).

Reliability and comprehensibility Test–retest reliability (reproducibility) was assessed by asking the patients to complete a second copy of the questionnaire 1 week after the initial visit when they completed the first copy. If the patient was in need of a fast working therapy, e.g., local injections, this was scheduled to be carried out on the seventh day after completing their second copy of the questionnaire.

Responsiveness has been described as the ability of an instrument to measure clinically important change over time with the change at present [7]. Sensitivity to change of the PROMs questionnaire was assessed in 112 patients who were treated with DMARDs and anti-TNF therapy. The patients completed the questionnaire before and 6 months after the treatment. Changes in the questionnaire scores were compared to the changes of other disease activity parameters including JADAS-27.

Psychometric properties of the illustrated questionnaire

A case–control study was implemented to assess the impact of the illustrations on the c-PROMs, psychometric properties, and acceptability for the children. This was carried out by taking the following steps: First, the mean scores for the child's functional ability and HRQoL with and without the illustrations were computed, and correlation coefficients were obtained; second, the number of missed items on each questionnaire (mean deficit percent); and third, to study the acceptability for children/parents, the mean comprehensibility, and user-friendliness scores. The active cohort included 122 children–parents/adolescents with JIA, whereas the control group included 123 JIA patients. The control group patients were asked to complete an unillustrated copy of the questionnaire, which had exactly the same format but without the illustrations. Randomized allocation was done by the hospital chart number (odd/even).

Statistical analysis

Statistical analysis was performed using the 20th version of SPSS. Variables are summarized in the form of mean and standard deviation if continuous and frequency distribution if categorical. Median and interquartile range (IQR) were calculated for skewed data. The Pearson correlation coefficient was used to figure out the correlation between quantitative variables. Error bars and scatter diagram were used to illustrate deviations and correlation of different variables respectively. Changes in the PROMs questionnaire were calculated by subtracting the second record score from the first record. Intraclass correlation coefficient for agreement (reliability) and consistency was calculated, and alpha statistic was calculated as an additional measure of reliability. Validation was tested by calculation of Spearman's correlation coefficient with the tested questionnaire and the selected confirmatory tests. *P* value is significant if less than 0.05.

Results

Demographics A total of 245 JIA patients were assessed for the validation study. No significant differences were found in demographic data between the patients who answered the illustrated version and those who completed the non-illustrated c-PROMs questionnaire (Table 1).

The JIA categories were 20.0% with systemic JIA, 10.0% with oligoarthritis, 18.0% with rheumatoid factor–negative polyarthritis, 20.0% with rheumatoid factor–positive polyarthritis, 12.0% with psoriatic arthritis, 12.0% with enthesitis-related arthritis, and 8.0% with undifferentiated arthritis. A total of 60/122 (49.2%) completed the parent version of the c-PROMs questionnaire, whereas the children version was completed by 62/122 (50.8%) of children aged 8 years or older in the study group. Similarly, in the control group, 61/123 (49.6%) of the c-PROMs questionnaire were completed

Table 1 Basic characteristics of the study versus the control group. There was no significant difference between the study and control groups in terms of baseline characteristics

	Study group (no., 122)	Control group (no., 123)
Age in years, mean ± SD	11.2 ± 0.9	11.4 ± 0.7
Female gender, no. (%)	85 (69.7)	86 (69.9)
JIA subtype, *N* (%)		
Oligoarticular arthritis	13 (10.7%)	14 (11.4%)
Polyarticular rheumatoid factor positive	24 (19.7%)	25 (20.3%)
Polyarticular rheumatoid factor negative	15 (12.3%)	16 (13%)
Psoriatic arthritis	14 (11.5%)	15 (12.2%)
Systemic	24 (19.7%)	24 (19.5%)
Enthesitis-related arthritis	15 (12.3%)	16 (13%)
Undifferentiated	11 (9%)	13 (10.6%)
JADAS-27 score	9.2 ± 0.9	9.4 ± 0.8

by parents, whereas 62/123 (50.4%) were completed by children aged 8 years or older.

Cronbach's alpha internal consistency was 0.89 for functional ability, 0.91 for child's motivation, 0.88 for HRQoL physical health, and 0.86 for HRQoL psychological health.

Convergent validity To assess the validity of the c-PROMs questionnaire, items were compared to the parameters of disease activity. Table 2 shows the correlation of the c-PROMs items with the disease activity parameters assessed by the physician as well as the inflammatory markers (ESR and CRP) as validating tools. Results revealed significant correlations with the disease activity parameter tests.

Test–retest reliability Instrument test–retest reliability was assessed using the intraclass correlation coefficient. The patients' population for determining test–retest reliability was 82 patients in each of the active group and control group. The median period between the first and second tests was 7.6 ± 1.3 days (mean ± standard deviation). The intraclass correlation coefficients for the functional ability was 0.993 (95% confidence interval (CI) 0.931 to 0.952), whereas it was 0.986 (95% CI 0.915 to 0.943) for HRQoL. The intraclass correlation coefficient for the illustrated c-PROMs indicated sufficient reproducibility. As far as the control group, the interclass correlation coefficients for functional ability and HRQoL were 0.891 and 0.868 respectively.

An average rate of missing items for each question was defined as mean deficit item percent, that is, the number of missing items from the total number of the questionnaire items in a percent figure. They were evaluated and compared between the study and control groups (Table 3). There was no specific pattern or specific items on studying the missed questions. Statistical analysis showed that the illustrated c-PROMs had significantly better completion of items than the non-illustrated group ($p < 0.01$).

Responsiveness to change (discriminant validity): Median disease duration was 2 years (4 months–3.6 years). After 6 months of management, there were significant improvements noted in all disease activity parameters and all patient-reported outcomes. Table 4 shows the changes of the different c-PROMs after 6 months of treatment in the illustrated group. The changes in the patient-reported outcomes were significantly correlated with the changes in all the disease activity parameters as well as inflammatory markers, namely ESR and CRP.

Table 2 Convergent validity: correlation of the PROMs items with the disease activity parameters assessed by the physician as well as the inflammatory markers (ESR and CRP) as validating tools

Items of the c-PROMsQ	TJC (physician assessed)	SJC (physician assessed)	Physician global assessment	Adherence to therapy	ESR	CRP	JADAS-27
Functional ability	0.572**	0.461**	0.693**	0.961**	0.479**	0.378**	0.652**
Physical health	0.795**	0.696**	0.671**	0.571**	0.691**	0.562**	0.786**
Psychological health	0.538**	0.425**	0.367*	0.592**	0.584**	0.467**	0.775**
Pain score	0.466**	0.063	0.496**	0.523**	0.642**	0.391*	0.668**
Patient global assessment	0.427**	0.521**	0.562**	0.461**	0.422**	0.464**	0.692**
Morning stiffness	0.331**	0.315**	0.498**	0.448**	0.017	0.396*	0.475**
Patient-reported TJC assessment	0.841**	0.526**	0.553**	0.563**	0.322*	0.471**	0.559**
Motivation	0.652**	0.685*	0.563**	0.686**	0.438**	0.462**	0.767**

*$p < 0.05$, **$p < 0.01$

Table 3 Mean deficit percent items in the study group versus the control group

	Study group	Control group	P value
Functional ability (mean ± standard deviation)	2.5 ± 0.4	2.1 ± 0.7	0.05*
Health-related quality of life (mean ± standard deviation)	2.6 ± 0.3	2.2 ± 0.6	0.05*
Mean deficit percent first test	2.8 ± 0.2%	6.9 ± 0.6%	0.002**
Mean deficit percent retest	0.3 ± 0.3%	3.8 ± 0.8%	0.001**
Comprehensibility (mean ± standard deviation)	9.7 ± 0.2	8.8 ± 0.5	0.01**
User-friendliness (mean ± standard deviation)	9.9 ± 0.1	7.8 ± 0.7	0.01**
Time to complete (min) (mean ± standard deviation)	14.3 ± 1.8	18.9 ± 2.6	0.05*

*$p < 0.05$, **$p < 0.01$

Discussion

Patient-reported outcomes provide an insight into knowledge only known to the patient. In recent years, increasing attention has been paid to c-PROMs in JIA [8]. Incorporation of these measures in patient management is deemed important as they reflect the parents' and children's perception of the disease course and effectiveness of therapeutic interventions. Information obtained with parent- or child-centered measures may contribute significantly to medical decision-making and increase the probabilities of success in patient care. Furthermore, as physician's therapeutic decisions are of primary importance to parents and patients, integration of their perspective in clinical evaluation may facilitate concordance with physician's choices and compliance with the prescribed medication [11, 22, 23]. Results of the work revealed that the developed c-PROMs-illustrated questionnaire was valid, reliable, and sensitive to change of the disease activity. The focus of the developed c-PROMs was widened to cover other important outcomes such as school attendance, adherence to therapy, comorbidity risk, as well as the child's motivation. In the chronic phase of the disease, patients or parents might report high perceived disease activity, high pain levels, high fatigue, and HRQoL irrespective of current active disease. Identification of these challenges to the patient's life should help in tailoring a management plan aiming at meeting the child's expectations. Therefore, these measures should help in supporting the child in his/her perceived impaired health status and could help the physician to recognize the patients at risk for impaired perceived health in its broadest way.

Measures of pain, quality of life, and physical function were used traditionally in research. However, over the past years, there has been a growing interest in the use of these parameters in the clinical management of pediatric rheumatologic conditions [24]. This has been emphasized by the Food and Drug Administration recommendation to use PROs for medical product labelling by the US Affordable Care Act to create the Patient-Centered Outcomes Research Institute (PCORI) and by the National Institute of Health development of the Patient-Reported Outcomes Measurement Information System (PROMIS). The results of this work revealed significant correlations between the patient-reported outcomes assessed and the disease activity as well as physician global assessment. Though, it is not uncommon for discordance between patient- and physician-reported outcomes [7, 22, 25, 26]. This is not surprising as physicians and patients and their families conceptualize disease differently and value somewhat different outcomes. Thus, full evaluation of disease activity and treatment goals should include both physician assessment and parent or child reports.

Using illustrations attached to a self-administered questionnaire for children offered three major advantages. First, the explanatory illustrations appear to raise acceptability and shorten the completion time of the questionnaire. Second, only the illustrated c-PROMs met recommended reproducibility standards for tracking individual patients and minimized the possibility of missing items while completing the questionnaire. Third, as shown in this study, using the illustrations helped in enhancing the questionnaire's comprehensibility and user-friendliness. Theoretically, including illustrations may have several advantages. Visual information improves text comprehension [27]. The illustrations may thus help subjects visualize and report their health status accurately. In addition, patients with low literacy particularly benefit from illustrations [28]. Moreover, the pictures appear to make participation more enjoyable for patients, especially children [29]. Only a few illustrated questionnaires have been developed to measure health status [4, 5]. In the study carried out by Kurimoto and his colleagues [30], the authors investigated the potential advantages of attaching illustrations to the "Hand10" which is a self-administered questionnaire for upper extremity disorders. A series of 106 elderly patients with upper extremity disorders, whose symptoms had been stable, were randomly allocated to receive either the illustrated Hand10 or the unillustrated Hand10. The use of explanatory illustrations attached to the Hand10 contributed to raise acceptability for elderly patients and suggested to improve the reproducibility.

There are seven categories of JIA defined by the International League of Associations for Rheumatology (ILAR): oligoarticular, systemic, polyarticular rheumatoid

Table 4 Changes of the different c-PROMs measures after 6 months of treatment in the illustrated group

	Baseline (mean ± SD)	After 6 months (mean ± SD)	Standardized alpha	ICC (95% CI)
Functional ability	2.53 ± 0.4	1.1 ± 0.5*	0.984	0.913 (0.884–0.942)
Health-related quality of life	2.6 ± 0.3	1.2 ± 0.4*	0.965	0.942 (0.922–0.957)
Pain score	8.8 ± 0.1.1	4.8 ± 1.4*	0.894	0.852 (0.831–0.845)
Patient global assessment	8.7 ± 1.2	4.6 ± 1.2*	0.941	0.864 (0.83–0.87)
Rate of school problems	36%	7%*	0.912	0.956 (0.938–0.962)
Duration of morning stiffness	134.9 ± 46.2	37.2 ± 22.6*	0.975	0.913 (0.884–0.942)
Medication adherence	6.4 ± 2.1	8.9 ± 0.9*	0.934	0.931 (0.903–0.936)
Motivation score	6.1 ± 2.8	9.1 ± 0.4*	0.928	0.871 (0.842–0.865)
Patient-reported tender joint count	14.1 ± 3.4	4.3 ± 1.5*	0.896	0.882 (0.84–0.862)
Physician-reported tender joint count	12.8 ± 3.1	3.9 ± 1.2*	0.927	0.945 (0.948–0.953)
Swollen joint count	5.6 ± 2.4	0.9 ± 0.9*	0.951	0.863 (0.831–0.854)
JADAS-27	9.2 ± 0.9	1.4 ± 0.5	0.946	
Physician global assessment	8.2 ± 1.4	3.9 ± 1.3*	0.887	0.954 (0.846–0.868)

$^*p < 0.05$

factor (RF) positive, polyarticular RF negative, psoriatic, enthesitis-related, and undifferentiated arthritis [14]. Although JIA is a group of heterogeneous conditions, many of the categories are combined for both clinical treatment guidelines [31] and clinical trials [32, 33]. In particular, those patients with polyarticular disease are often considered as a group regardless of distinguishing features such as the presence of rheumatoid factor, psoriasis, enthesitis, or sacroiliitis. The results of this work revealed that the c-PROMs tool was valid for use among the different JIA categories both to assess disease activity and monitor response to therapy. Previous studies leveraging national cross-sectional registry data from the Childhood Arthritis and Rheumatology Research Alliance (CARRA) as well as several small single-center studies demonstrated that children with enthesitis-related arthritis have more pain and lower physical function than other JIA subtypes [34, 35]. However, all of these studies included limited patient-level factors.

In conclusion, the developed c-PROMs tool has expanded the health domains that can be evaluated quantitatively. The results of this work revealed that the illustrated c-PROMs questionnaire was valid, reliable, and sensitive to change. The explanatory illustrations made the tool easy to understand and led to better rates of response and fewer missing data. This progress may promote the regular administration of parent/child questionnaires in standard clinical care. The questionnaire can be completed in the waiting area before the patient is called into the examining room, and the physician should spend only a few seconds reviewing and scoring the data.

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Contribution All authors contributed in the study methodology, analysis, and interpretation of the data and outcomes as well as the manuscript writing, reading, and approval of the final version. Prof. EG carried out the statistical analysis and reviewed the "Methods" and "Results" sections.

Compliance with ethical standards

Local ethical and methodological protocols for approval of the study were followed. All the patients who participated in the study signed an informed consent according to the Declaration of Helsinki (at the General Assembly in October 2008) and ensured that their data is kept confidential after explaining the benefit and potential null risk of the study.

Disclosures None.

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