Information technology in paediatric rheumatology

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ABSTRACT

Information technology in paediatric rheumatology has seen several exciting developments in recent years. The new multidimensional questionnaires for juvenile idiopathic arthritis, juvenile dermatomyositis, and juvenile autoinflammatory diseases integrate all major parent- and child-reported outcomes (PCROs) used in these diseases into a single tool, and provide an effective guide to manage, document change in health, assess effectiveness of therapeutic interventions, and verify the parent and child satisfaction with illness outcome. The Pharmachild registry is aimed to gain information concerning the long-term effectiveness and safety of the medications currently used in juvenile idiopathic arthritis, particularly biologic agents, through collection of prospective data in a large, multinational sample of patients. Children and their parents are directly involved in the data collection by means of the regular completion of a digital version of a multidimensional questionnaire. The Patient-Reported Outcomes Measurement Information System (PROMIS) employs modern measurement science to advance assessment of PCROs, particularly HRQL, and offers multidimensional profile measures. The conceptual link of paediatric PROMIS with adult instruments facilitates harmonisation of assessments made in children and adolescents with those carried out in young adults in the process of transition of medical care. Development of electronic versions of questionnaires that permit their completion through smartphones or touch-screen devices will revolutionise information collection from parents and children, foster the regular collection of PCROs in routine care, and ultimately improve the quality of self-reported health data, and patient outcomes.

Introduction

Paediatric rheumatic and autoinflammatory diseases encompass a broad range of disorders characterised by either chronic or periodic systemic inflammation, which may have a serious impact on child health and well-being and may cause nonreversible damage to several organs and systems (1-4). The objectives of management in these illnesses are to relieve subjective patient complaints and to control objective signs of inflammation with the aim of enhancing health-related quality of life (HRQL) and prevent or minimise organ damage. Achievement of these objectives is facilitated by the regular monitoring of child health status and disease course, through periodic completion of reliable and standardised outcome measures (5, 6). To enable their incorporation in daily practice, these tools should be simple and feasible, and easily utilisable in a busy clinical setting (7).

Over the past two decades, there has been a growing interest in the use of parent- and child-reported outcomes (PCROs) to assess the health status of children with rheumatic and autoinflammatory diseases. Incorporation of these tools in daily care is important as they provide information concerning parent and child perceptions of disease burden, as well as effectiveness and adverse effects of therapeutic interventions. Because parents and children (when mature enough to understand the clinical and therapeutic issues related to their disease) are asked with increasing frequency to participate actively in shared decision-making, integration of their perspectives into clinical assessment may facilitate concordance with physician's choices and improve adherence to treatment (6, 8, 9). Thus, information obtained from the parent or the child may contribute to the success of patient care. It is now recognised that the use of PCROs in routine practice may improve the quality of care (10).

In recent years, a great deal of innovation has been developed in PCRO assessment, including electronic versions of health questionnaires, which enable

their completion on smartphones or touch-screen devices. The aim of this review article is to provide a summary of the recent achievements in this field of research.

Multidimensional questionnaires

In analogy with multidimensional questionnaires for adults (11), the rationale that led to the development of the multidimensional questionnaires for children was based on the consideration that there are several PCROs not addressed by traditional instruments, such as morning stiffness and overall level of disease activity, rating of disease status and course, proxy- or self-assessment of joint involvement and extra-articular symptoms, side effects of medications, and assessment of therapeutic compliance and satisfaction with illness outcome, which may provide valuable insights into the impact of disease and its treatment on child's health (7). Incorporation of these PCROs, together with the traditional outcomes of physical function, health-related quality of life (HRQL), overall well-being, and pain, in the clinical evaluation enables a more adequate appraisal of the impact of the illness. The multidimensional questionnaires integrate all main PCROs used in paediatric rheumatic and autoinflammatory diseases and are intended primarily for regular administration in routine clinical practice. To ensure feasibility without compromising completeness, all individual measures included in the questionnaires are short and simple.

Juvenile Arthritis Multidimensional Assessment Report (JAMAR)

The JAMAR includes the following 15 PCROs, of which 10 have been previously validated and 5 are descriptive: 1) assessment of physical function; 2) rating of the intensity of child's pain on a 21-numbered circle VAS (12, 13); (3) assessment of HRQL; 4) rating of child's overall well-being on a 21-numbered circle VAS; 5) assessment of the presence of pain or swelling in the following joints or joint groups: cervical spine, lumbo-sacral spine, shoulders, elbows, wrists, small hand joints, hips, knees, ankles, and small foot joints; 6) assessment of morning stiffness. 7) assessment of extraarticular symptoms (fever and rash); 8) rating of the level of disease activity on a 21-numbered circle VAS; 9) rating of disease status at the time of the visit as remission, continued activity or relapse; 10) rating of disease course from previous visit as much improved; slightly improved; stable; slightly worsened; or much worsened; 11) checklist of the medications the child is taking; 12) checklist of side effects of medications; 13) report of difficulties with medication administration; 14) report of school problems caused by the disease; 15) a question about satisfaction with status and the outcome of the illness (14).

A parent proxy-reported version for ages 2–18 (14) years and a child self-report version of the JAMAR are available. Its format proved to be simple, easily understood, and readily answered by parents and children. Compilation of the questionnaire requires <15 min and scoring takes a few seconds. The JAMAR was tested in 618 Italian children with juvenile idiopathic arthritis (JIA) and was found to perform well in capturing differences in the degrees of disease activity and severity.

Data obtained from the JAMAR enables calculation of 2 novel composite scores entirely based on PRCOs: the Juvenile Arthritis Parent Assessment Index (JAPAI) and the Juvenile Arthritis Child Assessment Index (JACAI) (15). Furthermore, it allows the determination of so-called Parent- and Child-Acceptable Symptom States (16).

The JAMAR has been selected for the assessment of PCROs in a multinational study aimed to investigate the EPidemiology, treatment and Outcome of Childhood Arthritis throughout the world (EPOCA Study) (17). For the purposes of this study, the questionnaire has been so far translated and cross-culturally adapted and validated in 40 national languages.

Juvenile Dermatomyositis Multidimensional Assessment Report (JDMAR)

The multidimensional questionnaire for JDM has been modelled on the JAMAR (7). Its format is, therefore, similar, and several items are identical. However, the JDMAR includes a different physical function tool, which is specific for JDM and is in progress of separate validation (18). In addition, it contains, beside the 3 VAS for rating of pain, disease activity and overall well-being, a fourth VAS for assessment of fatigue. Evaluation of morning stiffness is not included and the section devoted to JIA manifestations is replaced with a checklist of the most common symptoms of JDM, with specific focus on those related to skin manifestations and muscle weakness. Likewise, the registration of treatment side effects is focused on the medications most frequently prescribed to children with JDM, namely systemic corticosteroids and immunosuppressants. The JDMAR is currently being validated in the context of a multinational collaborative effort.

Juvenile Autoinflammatory Disease Multidimensional Assessment Report (JAIMAR)

The evaluation of functional and social skills, school problems, emotional status, and difficulties of living with a chronic illness in patients with autoinflammatory diseases are frequently neglected. Recently, a multidimensional questionnaire has been developed for four autoinflammatory diseases, including familial Mediterranean fever (FMF), mevalonate kinase deficiency (MKD), tumour necrosis factor receptor-associated periodic syndrome (TRAPS), and PFAPA by the FAVOR (FMF Arthritis Vasculitis and Orphan disease Research in Paediatric Rheumatology, www.favor.org.tr) through an international collaborative effort. The new tool, which was called JAIMAR, includes 16 PCROs and four dimensions that evaluate functional status, pain, treatment compliance and HRQL (physical, social, school, and emotional status). The first validation study of JAIMAR was conducted in 250 children with FMF (19, 20). This analysis showed that the questionnaire is suitable for use in daily routine as well as clinical studies for the assessment of different aspects of the disease in periodic fever syndromes. After the

validation in FMF, the instrument will be tested in other autoinflammatory diseases (MKD, TRAPS, PFAPA) under the umbrella of the Eurofever Project (21).

Administration of multidimensional questionnaires in daily practice

The multidimensional questionnaires have been specifically designed for regular administration in a busy clinical setting, with particular attention to feasibility and acceptability in daily care. They are completed in the waiting area before the patient is called into an examining room. Almost all the work is done by the parent and/or the patient, not the physician or the staff, and the physician should spend only a few seconds reviewing and scoring the data. The best strategy to assure completion of questionnaires at each visit is for the receptionist or a nurse to distribute a questionnaire at the time of patient registration for the visit. Importantly, by involving the nurses in the help activity, they can be motivated to gain a more important role within the team. However, it should be emphasised that the best form of "help" is not to try to respond to the question, but to tell the child or parent that the response should be "whatever you think or feel, as there are no right or wrong answers (other than about facts of patient history such as illnesses and medications used)".

The use of questionnaires requires a change in clinic procedure, which may be seen as added complexity. However, their administration not only supports management decisions and improves documentations, but may also save time (after a brief "learning curve," as required with any new activity) (22).

Advantages of multidimensional questionnaires.

The creation of the multidimensional questionnaires has introduced a new approach to management of children with inflammatory and autoinflammatory diseases. Through their administration, information pertaining to patient history may be collected in a standardised way. Regular use of the questionnaires in daily practice allows to construct a flow sheet of patient's course over time.

Table I. Outcome assessments included in paediatric multidimensional questionnaires.

	JAMAR	JDMAR	JAIMAR
Physical function	✓	✓	✓
Health-related quality of life	✓	✓	✓
Pain	✓	✓	
Fatigue		✓	✓
Well-being	✓	✓	
Morning stiffness	✓	✓	✓
Symptoms self-assessment	✓	✓	✓
Symptoms recurrence frequency			✓
Disease activity	✓	✓	✓
Disease status	✓	✓	
Disease course	✓	✓	
Checklist of therapies	✓	✓	✓
Therapy side effects	✓	✓	✓
Compliance to therapy	✓	✓	✓
School related problems	✓	✓	✓
Satisfaction with illness outcome	✓	✓	✓

A flow sheet may aid in the detection of variations in clinical symptoms, functional capacity, pain, overall well-being, fatigue, and psychological status over time. This manner of recording clinical data is potentially advantageous in the management of chronic diseases, as it allows the physician to store serial parent/patient data, together with physician-reported outcomes, laboratory results, drug therapies, and other information (23). Composite scores derived with this approach can be depicted graphically to provide a quick overview of the diseases course over several years.

Completion of the questionnaires helps the parent and the patient to focus on information needed for care and enhances their capacity to describe concerns in the limited time allotted for a clinical encounter. Use of questionnaires is important because they address the primary concerns of children and their parents. Availability of the data to the physician at the time of the visit is helpful, particularly in view of the importance of the information provided by parents and patients in clinical decisions. Notably, patient self-report questionnaires provide information in considerably less time with more completeness than the traditional interview process – saving time for the doctor (if used properly), rather than adding to the doctor's burden.

The PCROs included in the multidimensional questionnaires are listed in Table I.

Information technology and disease registries: the example of Pharmachild

In 2011, the Paediatric Rheumatology INternational Trials Organisation (PRINTO) and the Paediatric Rheumatology European Society (PRES) created an international observational registry aimed to enroll children with JIA treated with methotrexate or biologic medications in any available formulations. This registry is named "Pharmacovigilance in JIA patients treated with biologic agents and/or methotrexate - Pharmachild" (European Union grant 260353) and is aimed primarily to evaluate both the long-term effectiveness and safety of these therapeutic agents (24). Paediatric rheumatology centers in more than 50 countries that are part to the two networks have joined this effort to estimate frequency of response and safety over time.

To facilitate the inclusion of data of a large number of patients by many participating centers, a simple and user-friendly online data collection system was set up. After the inclusion of patient information, the system provides a detailed report of the efficacy and safety data (including American College of Rheumatology, ACR, Paediatric response, occurrence of disease flare, achievement of clinical remission, Juvenile Arthritis Disease Activity Score, JADAS, and a safety summary) in a few minutes. The investigator also may obtain a graphical depiction of the course over time of quantitative

measures of treatment efficacy, such as the JADAS.

All families that possess an access to the internet and consent to provide their personal contact information are involved directly in data collection. Before the scheduled clinic visit, a parent or the child (as appropriate) is asked to complete the JAMAR in order to provide in advance to the caring physician the key information regarding the child's health status and disease course. Each participating family receives an email two days before the scheduled visit, which asks a parent or the child to complete their respective digital version of the questionnaire.

Both physicians and families are regularly updated (*e.g.* at least yearly) about safety and efficacy issues concerning drug administration with appropriate electronic newsletters. Periodic reminders also are sent to each Pharmachild user registered in the PRINTO website to remind completion of physicians' and family case report forms.

The Patient-Reported Outcomes Measurement Information System (PROMIS)

The PROMIS arose out of an initiative of the US National Institutes of Health (NIH) to employ modern measurement scientific approaches to advance assessment of patient-reported outcomes (PROs) across the life course (paediatric and adult), in order to enhance performance of clinical outcomes research (25;26). The programme was funded from 2004 for over 10 years. A portfolio of universal measures to assess health concepts, or "domains", from the patient or parent perspective for use across chronic conditions were created within PROMIS. This universal system facilitates studies of comparative effectiveness and population health.

PROMIS measures are developed with a mixed-methods approach. A high degree of rigor and input from patients affected by the conditions goes into their development and/or validation. Standards for development and validation of PROMIS measures have been codified into a maturity model (27). Item response theory (IRT) methods are applied in development of unidimensional PROMIS item banks (*i.e.* individual item banks for unique HRQL concepts), with items calibrated on a single scale according to difficulty. This enables efficient, precise measurement with low respondent burden as a score can be generated by using a subset of items, or even a single item, from an item bank depending on level of precision in scoring desired. This can either be done by selecting short forms of measures (fixed, 4–8 item short forms), or using computerised adaptive testing which is an algorithm that selects items based on response to items previously administered.

PROMIS offers measures assessing multiple aspects of HRQL that conceptually map onto the World Health Organisation (WHO) tripartite conceptualisation of physical, mental and social health (28). Paediatric measures are available for self-report for 8-17 years of age. Proxy-report versions are available for 5–17 years of age. At 18 years of age, a PROMIS measurement system for adults is available built on the same conceptual framework. A current limitation for paediatric care is the absence of PRO measures for children younger than age 5. A list of available paediatric self-and proxy measures and their characteristics is available on www.healthmeasures.net (29). An international initiative has promoted language and cross-cultural translations of PROMIS measures which in addition to English are available in Spanish, Dutch, and a subset in German and simplified Chinese. The global health measure is also available in French and Italian (30, 31).

With respect to the concept of multidimensional assessment, a 7-item PROMIS Paediatric Global Health (PGH-7) measure is designed to be brief (1-2 minutes to complete), comprehensive, amenable for conceptual linkage with an adult measure, practical and applicable for use in research, population health, and quality improvement, in addition to routine clinical care settings. This contains single items assessing the following health concepts: general health, quality of life, physical health, mental health, sadness, having fun with friends, and parents listening to the child. Four of the items overlap with the adult PROMIS global health measure for linkage across the life course (32, 33). There is also a 9 item global health measure which includes the core 7 items plus a single item on pain interference and fatigue. PROMIS measures are also available as profile measures, which combine items from six unidimensional domains (mobility, pain interference, fatigue, anxiety, depressive symptoms, peer relationships) plus a pain intensity item and are available as 25, 37 or 49 item versions (29). Measures are available as PDFs by user request at http://www.assessmentcenter.net/. The PROMIS measures are publicly available without licensing fees.

In the field of paediatric rheumatology, a number of initial validation studies of PROMIS measures have been conducted. The content validity of domains represented in the PROMIS profile measures was examined through interviews with children with JIA patients and chronic non-inflammatory pain (34). PROMIS paediatric measures have been examined across paediatric chronic conditions, including paediatric rheumatic disease, and shown to discriminate across subgroups (35). Validation studies have been conducted in JIA (36), systemic lupus erythematosus (37) and chronic musculoskeletal pain/juvenile fibromyalgia (38), In addition, results of longitudinal studies to assess responsiveness of measures over time are now being reported. Validation is an ongoing and continual process, and there is a new NIH funded initiative to validate PROMIS measures across multiple chronic conditions including in paediatric rheumatic disease, and this work is in progress through the Paediatric Patient-Reported Outcomes in Chronic Diseases (PEPR) Consortium (39).

Regarding integration into electronic databases and clinical health care information systems, PROMIS measures were developed with common data element standards and definitions in order to facilitate such integration, including Logical Observation Identifiers Names and Codes (LOINC) and Systematised Nomenclature of Medicine-ClinicalTerms (SNOMED CT). PROMIS

measures are available in large electronic health record systems used in the US, such as Epic. Measures may be administered by electronic device at point of care, with data scored and available seamlessly in the health record, or alternatively emailed to the patient in advance of the visit or triggered by clinical events (40, 41). PROMIS measures can also be administered through REDcap, a web-based application for building and managing online research surveys and databases (https://redcap-vanderbilt.edu/). Additionally, the US Patient Centered Outcomes Research Institute (PCORI) sponsored clinical research network, PCORnet, is combining health systems data on a large scale to enable comparative effectiveness research. PCORnet has incorporated select PROMIS items into the recommended Common Data Model (PRO Common Measures) (42) for collection by registries which will support broader uptake of PROMIS measures.

Electronic collection of self-reported data

At this time, computer-based utilities, such as office-based touch-screen computers, telephone-based interactive voice response systems, handheld computers, and smartphones, are emerging to address workflow challenges in obtaining, aggregating, calculating, and displaying data in real time, as well as minimising response errors. A computer-based touch-screen questionnaire process offers advantages over paper-based process in facilitating the collection of PCROs, ensuring reliability and validity (*e.g.* error checking) of the data capture, and simplifying the effective use of the data (43). By means of this technology, the questionnaires are automatically scored and results are stored in a database. Information can be presented in real time electronically with summary scores from previous encounter. Development of electronic versions of questionnaires on touch-screen handheld devices is in progress at authors' unit. However, although electronic versions of diaries and questionnaires are likely to be generally more accurate then paper versions, *e.g.* avoiding the transcription in clinical research databases, still errors may be present, and some error-checking steps are desirable.

Recent studies of self-reported pain and disease symptoms in children with JIA have shown that smartphones not only improve the quality of self-reported data, but also afford ease in obtaining repeated measures over a short time frame. These experiences have validated the use of technologically advanced data collection methods for capturing self-reported data via electronic diaries (e-diaries) (43-47). Thus, smartphones offer a promising method for collecting a variety of information about patient health status, disease course and treatment outcome, which can be assessed in the child's typical environment.

Conclusion

In recent years, there have been major advances in information technology in paediatric rheumatology. The new multidimensional questionnaires represent an innovative and feasible method for quantitative measurement in paediatric rheumatology care. These tools allow for greater focus on issues important to parents or patients, and may be used effectively to guide management, document change in health, assess the effectiveness of therapeutic interventions, and verify the parent or child satisfaction with illness outcome.

While different versions of multi dimensional assessment may be desirable for research concerning juvenile arthritis, dermatomyositis and auto-inflammatory diseases, etc., to gain further insight into mechanisms and outcomes, the development of a single questionnaire for paediatric rheumatic conditions could be considered in the near future.

The Pharmachild registry is collecting prospective data concerning long-term effectiveness and safety of treatment with methotrexate and biologic agents, which will facilitate gaining information in a very large, multinational sample of patients with JIA. Children and their parents are directly involved in the data collection by means of the regular completion of a digital version of the JAMAR.

The PROMIS is another important initiative aimed to assess PCROs, particularly HRQL, uniformly across diseases in which unidimensional measures can be used in combination as multidimensional global or health profile measures. Paediatric PROMIS measures have the advantage of being linked conceptually with adult instruments, which permits the harmonisation of the assessments made in children and adolescents with those carried out in young adults in the process of transition of medical care.

The availability of electronic versions of questionnaires that enable their completion through smartphones or touch-screen devices will revolutionise the way information is obtained from parents and children, and will ultimately improve the quality of self-reported health data, and facilitate and foster the regular collection of PCROs in routine clinical care.

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