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EDITORIAL

Preface

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The assessment of parent- and child-reported outcomes (PCROs) is gaining increasing importance in the management of children with rheumatic disorders. Parents and children (when mature enough to understand the clinical and therapeutic issues related to their disease) are asked with increasing frequency to actively participate in shared decision-making and the integration of their perspective in clinical assessment may facilitate concordance with physician's choices and improve adherence to treatment. In addition, the use of PCROs may help the physician to identify with greater accuracy the salient issues for each patient and to focus the attention on the relevant matters. It is now agreed that the inclusion of PCROs in clinical practice may lead to improve the quality of care. In keeping with these principles, regulatory bodies such as the Food and Drug Administration and the European Medicine Agency recommend the use of PCROs in paediatric clinical trials.

A number of tools for the assessment of PCROs in paediatric rheumatic diseases are available, including visual analogue scales for rating of child's overall well-being and intensity of pain, and questionnaires for the estimation of functional ability and health-related quality of life (HRQL). These clinical measures have been included in several observational studies, therapeutic trials, and long-term outcome surveys and some of them are included in standardized core sets of outcome measures, disease activity state definitions, or composite disease activity scores for juvenile idiopathic arthritis (JIA). However, in spite of their popularity and large-scale adoption, most of the instruments used to assess PCROs have remained fundamentally research tools and are not routinely administered in most paediatric rheumatology centres. One of the reasons that may explain why these evaluations are uncommonly performed in daily clinical care is the length and complexity of some questionnaires, particularly those used for the assessment of physical function and HRQL. There is the concern that their regular administration may interfere with routine clinical activity, with consequent increased costs and time.

On the other hand, the heterogeneous and multidimensional nature of JIA implies that numerous disease domains should be evaluated simultaneously to appraise the full impact of the illness. In this respect, there are several PCROs not addressed by conventional instruments, such as morning stiffness and overall level of disease activity, disease status and course, proxy- or self-assessment of joint involvement and extra-articular symptoms, side effects of medications, therapeutic compliance, and satisfaction with the outcome of the illness, which may provide important insights into the influence of the disease and its treatment on child's health.

Information related to PCROs is usually obtained from parents and children in form of interview at the time of the visit and is recorded in clinical charts or computer-based records as written notes. However, collecting this data in a standardized form would provide a physician with a thorough and systematic overview of the patient status to be scanned quickly at the beginning of the visit. Pursuing this objective through the administration of multiple lengthy questionnaires is clearly not feasible in a busy clinic. Assessment tools for use in routine clinical care must be at the same time comprehensive and practical.

These considerations have provided the rationale for the development in 2011 of the Juvenile Arthritis Multidimensional Assessment Report (JAMAR), a multidimensional questionnaire for the assessment of patients with JIA in standard clinical care that incorporates all main PCROs. The JAMAR addresses all domains included in the WHO International Classification of Functioning and Health. The

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JAMAR is proposed for use as both proxy-report and patient self-report, with the suggested age range of 7–18 years for use as self-report. The questionnaire format has been found very user-friendly, easy to understand, and readily responded to by parents and children. It is quick, taking less than 15 minutes to complete and can be scanned by a health professional for a clinical overview in a few seconds. Scoring of its components can be performed in less than 5 min.

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). The study is primarily aimed to obtain information on the frequency of JIA categories in different geographic areas, the therapeutic interventions made by paediatric rheumatologists practicing in diverse countries or continents, and the current disease and health status of children with JIA followed worldwide. Additional aims are to investigate the availability of biologic medications in developing countries and to foster the regular quantitative clinical assessment of children with JIA in standard clinical care.

To obtain figures generalizable on a worldwide basis, the involvement of a large number of countries was sought for. To reach this goal, participation in the study was first proposed to the national coordinating centre of all countries belonging to the Paediatric Rheumatology International Trials Organisation (PRINTO at http://www.printo.it), and at least to one qualified paediatric rheumatology centre in the US and Canada. For the purposes of this study, the JAMAR needed to be translated and cross-culturally adapted and validated in the national language of each participating country as per international guidelines.

PRINTO supported the EPOCA project by fostering the active involvement of its worldwide membership. PRINTO is a non-governmental international network founded by Alberto Martini and Nicolino Ruperto in 1996 and based at the Istituto Giannina Gaslini of Genoa Italy. The PRINTO main goal is to foster, facilitate and co-ordinate the development, conduct, analysis, and reporting of multi-centre, international clinical trials and/or outcome standardization studies in children with rheumatic diseases. PRINTO is composed of four main structures: the Advisory Council (which acts as the steering committee), the international coordinating centre located in Genoa, Italy (which coordinates the international projects), one national coordinator for each country (who coordinates the work among the individual paediatric rheumatology centres in his/her country and was in charge of the national implementation of the EPOCA project), and more than 600 centres (hospitals and/or universities) distributed in 88 countries around the world. As of today, PRINTO has collected, for academic studies, data of over 37,500 children in 300 centres in 67 countries, and, in collaboration with the Paediatric Rheumatology Collaborative Study Group (PRCSG at http://www.prcsg.org) and pharmaceutical companies, over 3500 children for clinical trials in more than 250 centres in 40 countries worldwide.

This supplement represents one of the main results of this cooperation among the different centres and countries belonging to PRINTO. Its objective is to make available to the paediatric rheumatology community a standardized tool aimed to foster a multidimensional approach in outcome assessment of JIA.

The supplement begins with an introductive review article that summarizes the study methodology and the sample of JIA patients and healthy controls collected for the study. It is followed by one article for each of the 49 countries. Each paper presents the results of the cross-cultural adaptation and psychometric evaluation of the JAMAR in that particular country, plus a table which compares the demographic, disability, physical and psychological data of JIA patients and their healthy peers. The EPOCA effort involved the participation of more than 300 clinical researchers from 125 centres in 52 countries.

The cross-cultural adaptation and validation of the JAMAR could not be accomplished without the support of PRINTO and the cooperation of its members in their local paediatric rheumatology centres as well as of the families of the JIA patients and healthy controls. This effort led to the involvement of more than 10,000 children all over the world.

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Compliance with ethical standards

Conflict of interest Prof. Ravelli has received speaker's bureaus and consulting fees from AbbVie, BMS, Pfizer, Hoffman LaRoche, Novartis, Centocor. Dr. Consolaro, has nothing to disclose in relation to this manuscript. Prof. Martini does not have any conflict of interest to declare since 1 March 2016, when he became the Scientific Director of the G. Gaslini Hospital: this role does not allow him to render private consultancies resulting in personal income. Prof. Martini per-

forms consultancy activities on behalf of the Gaslini Institute for Abbvie, Boehringer, Novartis, R-Pharm, but the money received for these activities were directly transferred to the Gaslini Institute's bank account. Before March 2016, when AM was the head of Pediatric Rheumatology department at the G. Gaslini Hospital, he received speaker bureaus and consultancies fees from Abbvie, Astrazeneca, Novartis, Pfizer, Roche, R-Pharm, Sanofi, UCB. Dr. Ruperto has received grants from BMS, Hoffman-La Roche, Janssen, Novartis, Pfizer, Sobi, during the conduct of the study and personal fees and speaker honorarium from Abbvie, Ablynx, Amgen, AstraZeneca, Baxalta Biosimilars, Biogen Idec, Boehringer, Bristol Myers Squibb, Celgene, Eli-Lilly, EMD Serono, Gilead Sciences, Janssen, Medimmune, Novartis, Pfizer, Rpharm, Roche, Sanofi, Servier and Takeda.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study as per the requirement of the local ethical committee.