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Rheumatology International (2018) 38 (Suppl 1):S291–S298 https://doi.org/10.1007/s00296-018-3963-y

Rheumatology

VALIDATION STUDIES



The Norwegian version of the Juvenile Arthritis Multidimensional Assessment Report (JAMAR)

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Received: 22 December 2017 / Accepted: 11 January 2018 © The Author(s) 2018. This article is an open access publication

Abstract

The Juvenile Arthritis Multidimensional Assessment Report (JAMAR) is a new parent/patient-reported outcome measure that enables a thorough assessment of the disease status in children with juvenile idiopathic arthritis (JIA). We report the results of the cross-cultural adaptation and validation of the parent and patient versions of the JAMAR in the Norwegian language. The reading comprehension of the questionnaire was tested in 10 JIA parents and patients. Each participating centre was asked to collect demographic and clinical data and the JAMAR in 100 consecutive JIA patients or all consecutive patients seen in a 6-month period and to administer the JAMAR to 100 healthy children and their parents. The statistical validation phase explored descriptive statistics and the psychometric issues of the JAMAR: the 3 Likert assumptions, floor/ceiling effects, internal consistency, Cronbach's alpha, interscale correlations, test–retest reliability, and construct validity (convergent and discriminant validity). A total of 301 JIA patients (3.3% systemic, 41.2% oligoarticular, 25.9% RF negative polyarthritis, and 29.6% other categories) and 74 healthy children were enrolled in three centres. The JAMAR components discriminated well healthy subjects from JIA patients. All JAMAR components revealed good psychometric performances. In conclusion, the Norwegian version of the JAMAR is a valid tool for the assessment of children with JIA and is suitable for use both in routine clinical practice and clinical research.

Keywords Juvenile idiopathic arthritis · Disease status · Functional ability · Health-related quality of life · JAMAR

Introduction

The aim of the present study was to cross-culturally adapt and validate the Norwegian parent, child/adult version of the Juvenile Arthritis Multidimensional Assessment Report (JAMAR) [1] in patients with juvenile idiopathic arthritis

The local members of the Paediatric Rheumatology International Trials Organisation (PRINTO) participating in the project are listed in dedicated tables of "https://doi.org/10.1007/s00296-018-3944-1 / Cross-cultural adaptation and psychometric evaluation of the Juvenile Arthritis Multidimensional Assessment Report (JAMAR) in 54 languages across 52 countries: review of the general methodology".

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(JIA). The JAMAR assesses the most relevant parent/patient-reported outcomes in JIA, including overall well-being, functional status, health-related quality of life (HRQoL), pain, morning stiffness, disease activity/status/course, articular and extra-articular involvement, drug-related side effects/compliance, and satisfaction with illness outcome.

This project was part of a larger multinational study conducted by the Paediatric Rheumatology International Trials Organisation (PRINTO) [2] aimed to evaluate the Epidemiology, Outcome and Treatment of Childhood Arthritis (EPOCA) in different geographic areas [3].

We report herein the results of the cross-cultural adaptation and validation of the parent and patient versions of the JAMAR in the Norwegian language.



Materials and methods

The methodology employed has been described in detail in the introductory paper of the supplement [4]. In brief, it was a cross-sectional study of JIA children, classified according to the ILAR criteria [5, 6], and enrolled from March 2012 to September 2013. Children were recruited after Ethics Committee approval and consent from at least one parent.

The JAMAR

The JAMAR [1] includes the following 15 sections:

- 1. Assessment of physical function (PF) using 15 items in which the ability of the child to perform each task is scored as follows: 0 = without difficulty, 1 = with some difficulty, 2 = with much difficulty, 3 = unable to do, and not applicable if it was not possible to answer the question or the child was unable to perform the task due to their young age or to reasons other than JIA. The total PF score ranges from 0 to 45 and has three components: PF-lower limbs (PF-LL); PF-hand and wrist (PF-HW), and PF-upper segment (PF-US) each scoring from 0 to 15 [7]. Higher scores indicating higher degree of disability [8–10];
- 2. rating of the intensity of the patient's pain on a 21-numbered circle visual analogue scale (VAS) [11];
- assessment of the presence of joint pain or swelling (present/absent for each joint);
- 4. assessment of morning stiffness (present/absent);
- 5. assessment of extra-articular symptoms (fever and rash) (present/absent);
- 6. rating of the level of disease activity on a 21-circle VAS;
- 7. rating of disease status at the time of the visit (categorical scale);
- 8. rating of disease course from previous visit (categorical scale);
- 9. checklist of the medications the patient is taking (list of choices);
- 10. checklist of side effects of medications;
- 11. report of difficulties with medication administration (list of items);
- report of school/university/work problems caused by the disease (list of items);
- 13. assessment of HRQoL, through the Physical Health (PhH), and Psychosocial Health (PsH) subscales (5 items each) and a total score. The four-point Likert response, referring to the prior month, are '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. The total HRQoL score ranges from 0 to 30, with higher scores indicating worse HRQoL. A separate score for PhH and PsH (range 0–15) can be calculated [12–14];
- 14. rating of the patient's overall well-being on a 21-numbered circle VAS;
- 15. a question about satisfaction with the outcome of the illness (yes/no) [15].

The JAMAR is available in three versions, one for parent proxy-report (child's age 2–18), one for child self-report, with the suggested age range of 7–18 years, and one for adults.

Cross-cultural adaptation and validation

The process of cross-cultural adaptation was conducted according to international guidelines with 2–3 forward and backward translations. In those countries for which the translation of JAMAR had been already cross-cultural adapted in a similar language (i.e. Spanish in South American countries), only the probe technique was performed. Reading comprehension and understanding of the translated questionnaires were tested in a probe sample of 10 JIA parents and 10 patients.

Each participating centre was asked to collect demographic and clinical data and the JAMAR in 100 consecutive JIA patients or all consecutive patients seen in a 6-month period and to administer the JAMAR to 100 healthy children and their parents.

The statistical validation phase explored the descriptive statistics and the psychometric issues [16]. In particular, we evaluated the following validity components: the first Likert assumption [mean and standard deviation (SD) equivalence]; the second Likert assumption or equal item-scale correlations (Pearson r: all items within a scale should contribute equally to the total score); the third Likert assumption (item internal consistency or linearity for which each item of a scale should be linearly related to the total score that is 90% of the items should have Pearson $r \ge 0.4$); floor/ceiling effects (frequency of items at lower and higher extremes of the scales, respectively); internal consistency, measured by the Cronbach's alpha, interscale correlation (the correlation between two scales should be lower than their reliability coefficients, as measured by Cronbach's alpha); test-retest reliability or intraclass correlation coefficient (reproducibility of the JAMAR repeated after 1 or 2 weeks); and construct validity in its two components: the convergent or external validity which examines the correlation of the JAMAR subscales with the 6 JIA core set variables, with the addition of the parent assessment of disease activity and pain by the



Spearman's correlation coefficients (*r*) [17] and the discriminant validity, which assesses whether the JAMAR discriminates between the different JIA categories and healthy children [18].

Quantitative data were reported as medians with first and third quartiles and categorical data as absolute frequencies and percentages.

The complete Norwegian parent and patient versions of the JAMAR are available upon request to PRINTO.

Results

Cross-cultural adaptation

The Norwegian JAMAR was fully cross-culturally adapted from the standard English version with two forward and two backward translations with a concordance for 112/123 translations lines (91.1%) for the parent version and 109/120 lines (90.8%) for the child version.

Of the 123 lines in the parent version of the JAMAR, 116 (94.3%) lines were understood by at least 80% of the 10 parents tested (median = 100%; range: 50–100%); 107/120 (89.1%) lines in the patient version of the JAMAR were understood by at least 80% of the children (median = 100%; range: 20–100%). Lines 1, 6, 66, 73, 97, 114, and 118 of the parent version of the JAMAR were modified considering parents' suggestions. Lines 1, 23, 62, 64, 76, 77, 78, 79, 80, 94, 114, 116, and 119 of the patient version of the JAMAR were modified considering patients suggestions.

Demographic and clinical characteristics of the subjects

A total of 301 JIA patients and 74 healthy children (total of 375 subjects) were enrolled at three paediatric rheumatology centres.

In the 301 JIA subjects, the JIA categories were 3.3% with systemic arthritis, 41.2% with oligoarthritis, 25.9% with RF negative polyarthritis, 3.3% with RF positive polyarthritis, 4.0% with psoriatic arthritis, 9.0% with enthesitis-related arthritis, and 13.3% with undifferentiated arthritis (Table 1).

A total of 357/375 (95.2%) subjects had the parent version of the JAMAR completed by a parent (285 from parents of JIA patients and 72 from parents of healthy children). The JAMAR was completed by 289/357 (81.0%) mothers and 68/357 (19.0%) fathers. The child version of the JAMAR was completed by 298/375 (79.5%) children age 6.5 or older. In addition, patients younger than 7 years old, capable to assess their personal condition and able to read and write, were asked to fill in the patient version of the questionnaire.

Discriminant validity

The JAMAR results are presented in Table 1, including the scores [median (first-third quartiles)] obtained for the PF, the PhH, the PsH subscales and total score of the HRQoL scales. The JAMAR components discriminated well between healthy subjects and JIA patients.

In summary, the JAMAR revealed that JIA patients had a greater level of disability and pain, as well as a lower overall well-being and HRQoL than their healthy peers.

Psychometric issues

The main psychometric properties of both parent and child versions of the JAMAR are reported in Table 2. The following "Results" section refers mainly to the parent's version findings, unless otherwise specified.

Descriptive statistics (first Likert assumption)

For all the JAMAR items, the median number of missing responses was 0.4 (0.4–0.4).

The response pattern for both PF and HRQoL was positively skewed toward normal functional ability and normal HRQoL. All response choices were used for the different HRQoL items, whereas a reduced number of response choices were used for PF items 9, 11, 14 and 15.

The mean and SD of the items within a scale were roughly equivalent for the PF and for the HRQoL items (data not shown). The median number of items marked as not applicable was 1% (1–2%) for the PF and 8% (6–9%) for the HRQoL.

Floor and ceiling effect

The median floor effect was 83.5% (75.4–92.3%) for the PF items, 52.6% (38.6–59.3%) for the HRQoL PhH items, and 58.2% (54.4–68.4%) for the HRQoL PsH items. The median ceiling effect was 0.4% (0–0.7%) for the PF items, 2.8% (2.1–7.4%) for the HRQoL PhH items, and 1.4% (1.1–1.4%) for the HRQoL PsH items. The median floor effect was 26.7% for the pain VAS, 22.8% for the disease activity VAS, and 27.7% for the well-being VAS. The median ceiling effect was 0.3% for the pain VAS, 0.7% for the disease activity VAS, and 0.3% for the well-being VAS.

Equal item-scale correlations (second Likert assumption)

Pearson item-scale correlations corrected for overlap were roughly equivalent for items within a scale for 87% of the PF



Table 1 Descriptive statistics (medians, first-third quartiles or absolute frequencies, and %) for the 301 JIA patients

	Systemic N=10	Oligoarthritis $N=124$	RF- polyar-thritis $N=78$	RF+ polyar- thritis $N=10$	Psoriatic arthritis N=12	Enthesitis- related arthritis N=27	Undifferentiated arthritis $N=40$	All JIA patients N=301	Healthy $N=74$
Female	7 (70%)	96 (77.4%)	53 (67.9%)	8 (80%)	7 (58.3%)	10 (37%)	28 (70%)	209 (69.4%)*	35 (47.3%)**
Age at visit	12.6 (8–16.1)	11.2 (7.1–14.2)	11.5 (8.7–15.4)	15.4 (11.6– 16.6)	12.7 (11.4– 16.2)	15.2 (12.4– 17.2)	14.7 (10.5– 16.3)	12 (8.3– 15.4)**	12.9 (9.8–15.9)
Age at onset	4 (1.9–6.6)	2.8 (1.8–5.7)	2.5 (1.6–8.3)	10.4 (5.1–14.2)	8.6 (5.2–11.4)	8.7 (6.2– 11.9)	6.4 (2.2– 11.3)	3.7 (1.9–8.8)#	
Disease duration	9.3 (1.7– 12.5)	6 (2.3–9.2)	5.9 (3.2–9.7)	2.2 (1.2–7.3)	3.9 (1.6–7.3)	4.9 (3.7–8.4)	5 (2.4–9)	5.8 (2.5–9)	
ESR	6 (2–12)	7 (4–12)	7 (3–11)	6 (2–12)	6 (3–8)	6 (4–10)	7.5 (5–12)	7 (4–12)	
MD VAS	1 (0-2)	1 (0-2)	1 (0-2)	1.5 (0.5–3)	2.5 (2-3)	0 (0–1)	1 (0-2)	1 (0-2)	
No. swollen joints	0 (0-0)	0 (0-1)	0 (0-1)	1 (0-8)	1 (0-1.5)	0 (0-0)	0 (0-1)	0 (0-1)*	
No. joints with pain	0 (0–1)	0 (0–1)	0 (0–1)	0 (0–1)	1.5 (0–2)	0 (0–1)	0 (0–1)	0 (0–1)	
No. joints with LOM	0 (0–2)	0 (0–1)	0 (0–2)	0 (0–2)	1 (0–4.5)	0 (0–1)	0 (0–1)	0 (0–1)	
No. active joints	0 (0-1)	0 (0-1)	0 (0-1)	1 (0-8)	1.5 (0-2.5)	0 (0-1)	0 (0–1)	0 (0–1)	
Active systemic features	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	
ANA status	0 (0%)	27 (21.8%)	17 (21.8%)	3 (30%)	2 (16.7%)	2 (7.4%)	7 (17.5%)	58 (19.3%)	
Uveitis	1 (10%)	32/123 (26%)	18 (23.1%)	1 (10%)	2 (16.7%)	7 (25.9%)	15/39 (38.5%)	76/299 (25.4%)	
PF total score	2.5 (0-4)	1 (0-3)	2 (0-6)	1 (0-1)	4 (0–20)	0 (0-3.5)	1 (0-4)	1 (0-4)	0 (0-0)#
Pain VAS	1.5 (0-2.5)	1.5 (0-3.5)	1.5 (0.5–4.5)	2 (0.5–2.5)	4.5 (4–7)	1 (0-4)	1.5 (0-4)	2 (0-4)*	0 (0-0) #
Disease activity VAS	2.3 (0-5)	1.5 (0.5–3.5)	2 (0.5–5)	1 (0-3.5)	4 (2.5–7)	1.3 (0-3.3)	1.5 (0.5–4)	2 (0.5–4)	
Well-being VAS	1.3 (0-2)	1 (0-3)	2 (0.5–3)	1 (0-3)	4.5 (1-7)	0.8 (0-2.5)	1 (0-3.5)	1 (0-3)	
HRQoL PhH	3.5 (2-6)	3 (1–5)	3 (1–6)	3 (1–3)	7 (3–10)	1 (0-6)	3 (1–5)	3 (1–5)*	0 (0-1)#
HRQoL PsH	2 (1–3)	2 (0-4)	2 (0-4)	1 (0-2)	2 (1-4)	1 (0-3)	1 (0-3)	2 (0-4)	0 (0-1)#
HRQoL Total Score	5 (4–10)	5 (1–9)	5 (2–10)	4 (1–6)	9 (5–16)	2.5 (0.5–9.5)	5 (2–7)	5 (2–9)	0 (0-1)#
Pain/swell. in > 1 joint	4 (40%)	88/123 (71.5%)	48/74 (64.9%)	6/9 (66.7%)	10/11 (90.9%)	11/24 (45.8%)	22/34 (64.7%)	189/285 (66.3%)	6/72 (8.3%)#
Morning stiff- ness > 15 min	2 (20%)	41/122 (33.6%)	35/73 (47.9%)	3/9 (33.3%)	8/11 (72.7%)	7/24 (29.2%)	13/34 (38.2%)	109/283 (38.5%)	1/72 (1.4%)#
Subjective remission	4 (40%)	79/123 (64.2%)	46/73 (63%)	6/9 (66.7%)	10/11 (90.9%)	13/24 (54.2%)	21/34 (61.8%)	179/284 (63%)	
In treatment	9 (90%)	84/123 (68.3%)	63/73 (86.3%)	9/9 (100%)	11/11 (100%)	21/24 (87.5%)	28/34 (82.4%)	225/284 (79.2%)*	
Reporting side effects	5/9 (55.6%)	39/84 (46.4%)	33/63 (52.4%)	1/9 (11.1%)	8/11 (72.7%)	9/21 (42.9%)	15/28 (53.6%)	110/225 (48.9%)	
Taking medica- tion regularly	9/9 (100%)	76/84 (90.5%)	57/63 (90.5%)	7/9 (77.8%)	11/11 (100%)	20/21 (95.2%)	25/28 (89.3%)	205/225 (91.1%)	
With problems attending school	1/4 (25%)	20/74 (27%)	18/45 (40%)	0 (0%)	5/7 (71.4%)	5/19 (26.3%)	5/19 (26.3%)	54/172 (31.4%)	0 (0%)#
Satisfied with disease outcome	9 (90%)	85/123 (69.1%)	44/73 (60.3%)	6/9 (66.7%)	4/11 (36.4%)	16/24 (66.7%)	28/34 (82.4%)	192/284 (67.6%)	

Data related to the JAMAR refer to the 285 JIA patients and to the 72 healthy subjects for whom the questionnaire has been completed by the parents

JAMAR Juvenile Arthritis Multidimensional Assessment Report, ESR erythrocyte sedimentation rate, MD Medical Doctor, VAS visual analogue scale (score 0–10; 0=no activity, 10=maximum activity), LOM limitation of motion, ANA anti-nuclear antibodies, PF physical function (total score ranges from 0 to 45), HRQoL health-related quality of life (total score ranges from 0 to 30), PhH physical health (total score ranges from 0 to 15), PsH psychosocial health (total score ranges from 0 to 15)

p values refers to the comparison of the different JIA categories or to JIA versus healthy. *p < 0.001, *p < 0.001, *p < 0.0001



Table 2 Main psychometric characteristics between the parent and child version of the JAMAR

	Parent $N = 285/357$	Child $N = 228/298$	
Missing values (1st–3rd quartiles)	0.4 (0.4–0.4)	0.9 (0.4–0.9)	
Response pattern	PF and HRQoL positively skewed	PF and HRQoL positively skewed	
Floor effect, median ^a			
PF	83.5%	81.1%	
HRQoL PhH	52.6%	53.9%	
HRQoL PsH	58.2%	59.6%	
Pain VAS	26.7%	25.9%	
Disease activity VAS	22.8%	26.3%	
Well-being VAS	27.7%	35.5%	
Ceiling effect, median ^a			
PF	0.4%	0.4%	
HRQoL PhH	2.8%	2.2%	
HRQoL PsH	1.4%	1.8%	
Pain VAS	0.3%	0.4%	
Disease activity VAS	0.7%	0.0%	
Well-being VAS	0.3%	1.3%	
Items with equivalent item-scale correlation	87% for PF, 90% for HRQoL	73% for PF, 90% for HRQoL	
Items with item-scale correlation ≥ 0.4	87% for PF, 100% for HRQoL	87% for PF, 100% for HRQoL	
Cronbach's alpha			
PF-LL	0.86	0.86	
PF-HW	0.85	0.80	
PF-US	0.69	0.74	
HRQoL PhH	0.87	0.86	
HRQoL PsH	0.83	0.83	
Items with item-scale correlation lower than the Cronbach alpha	100% for PF, 100% for HRQoL	100% for PF, 100% for HRQoL	
Test-retest intraclass correlation			
PF total score	0.66	0.63	
HRQoL PhH	0.72	0.80	
HRQoL PsH	0.55	0.70	
Spearman correlation with JIA core-set variables, median			
PF	0.5	0.4	
HRQoL PhH	0.5	0.4	
HRQoL PsH	0.3	0.3	
Pain VAS	0.3	0.3	
Disease activity VAS	0.2	0.3	
Well-being VAS	0.4	0.4	

JAMAR Juvenile Arthritis Multidimensional Assessment Report, JIA juvenile idiopathic arthritis, VAS visual analogue scale, PF physical function, HRQoL health-related quality of life, PhH physical health, PsH psychosocial health, PF-LL PF-lower limbs, PF-HW PF-hand and wrist, PF-US PF-upper segment

items, with the exception of PF items 11 and 15, and for 90% of the HRQoL items, with the exception of item 1.

Item internal consistency (third Likert assumption)

Pearson item-scale correlations were \geq 0.4 for 87% of items of the PF (except for PF items 11 and 15) and 100% of items of the HRQoL.



^aFrequency of items at lower and higher extremes of the scale, respectively

Cronbach's alpha internal consistency

Cronbach's alpha was 0.86 for PF-LL, 0.85 for PF-HW, and 0.69 for PF-US. Cronbach's alpha was 0.87 for HRQoL PhH and 0.83 for HRQoL PsH.

Interscale correlation

The Pearson correlation of each item of the PF and the HRQoL with all items included in the remaining scales of the questionnaires was lower than the Cronbach's alpha.

Test-retest reliability

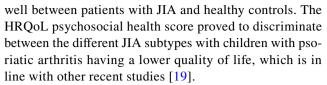
Reliability was assessed in 9 JIA patients, by re-administering both versions (parent and child) of the JAMAR after a median of 7 days (7–17 days). The intraclass correlation coefficients (ICC) for the PF total score showed a substantial reproducibility (ICC = 0.66). The ICC for the HRQoL PhH showed a substantial reproducibility (ICC = 0.72), while the ICC for the HRQoL PsH showed a moderate reproducibility (ICC = 0.55).

Convergent validity

The Spearman correlation of the PF total score with the JIA core set of outcome variables ranged from 0.3 to 0.6 (median = 0.5). The PF total score best correlation was observed with the parent disease activity VAS (r=0.6, p<0.001). For the HRQoL, the median correlation of the PhH with the JIA core set of outcome variables ranged from 0.2 to 0.7 (median = 0.5), whereas that for the PsH ranged from 0.1 to 0.6 (median = 0.3). The PhH showed the best correlation with the parent's assessment of pain (r=0.8, p<0.001) and the PsH with the parent global assessment of well-being (r=0.6, p<0.001). The median correlations between the pain VAS, the well-being VAS, and the parent disease activity VAS and the physician-centred and laboratory measures were 0.3 (0.2–0.5), 0.2 (0.2–0.4), and 0.4 (0.3–0.6), respectively.

Discussion

In this study, the Norwegian version of the JAMAR was cross-culturally adapted from the original standard English version with two forward and two backward translations. According to the results of the validation analysis, the Norwegian parent and patient versions of the JAMAR show satisfactory psychometric properties. The disease-specific components of the questionnaire discriminated



Psychometric performances were good for all domains of the JAMAR with few exceptions: two PF items (stretch arms and bite a sandwich or an apple) showed a lower item's internal consistency. However, the overall internal consistency was good for all the domains.

In the external validity evaluation, the Spearman's correlations of the PF and HRQoL scores with JIA core-set parameters ranged from weak to strong.

The results obtained for the parent version of the JAMAR are very similar to those obtained for the child version, which demonstrates that children are equally reliable proxy reporters of their disease and health status as their parents. The JAMAR is aimed to evaluate the side effects of medications and school attendance, which are other dimensions of daily life that were not previously considered by other HRQoL tools. This may provide useful information for intervention and follow-up in health care.

In conclusion, the Norwegian version of the JAMAR was found to have satisfactory psychometric properties, and it is, thus, a reliable and valid tool for the multidimensional assessment of children with JIA.

Acknowledgements We thank all families who participated in the project, the team that prepared and reviewed the forward and backward translations, in particular Dr. Anne Marit Selvaag, and all members of PRINTO in Norway, including the study nurses Ragnhild Sele and Cecilie Høines Reinsertsen in Trondheim, Brita Lena Hansen and Solfrid Kindlihagen in Tromsø. We thank the staff of the PRINTO International Coordinating Centre in Genoa (Italy) and in particular Marco Garrone for the overall coordination of the translation process, Silvia Scala and Elisa Patrone for data collection and quality assurance, Luca Villa, Giuseppe Silvestri, and Mariangela Rinaldi for the database development and management, and the remaining PRINTO team for data entry. The Principal Investigator of the study was Prof. Angelo Ravelli, MD. The scientific coordinator and study methodologist was Nicolino Ruperto, MD, MPH. The project coordinators were Alessandro Consolaro, MD, PhD, Francesca Bovis, BsA. We also thank Prof. Alberto Martini, PRINTO Chairman. Funding was provided by the Istituto G. Gaslini, Genoa (Italy) and a grant was given from The Medicines for Children Network, Norway. Permission for use of JAMAR and its translations must be obtained in writing from PRINTO, Genoa, Italy. All JAMAR-related inquiries should be directed to at printo@ gaslini.org. Permission for use of CHAQ and CHQ derived-material is granted through the scientific cooperation of the copyright holder ICORE of Woodside CA and HealthActCHQ Inc. of Boston, Massachusetts USA. All CHQ-related inquiries should be directed to licensing@healthactchq.com. All CHAQ-related inquiries should be directed to gsingh@stanford.edu.

Funding This study was funded and coordinated by Istituto Giannina Gaslini, Genoa, Italy.



Compliance with ethical standards

Conflict of interest Dr. Nordal, Dr. Ødegård, Dr. Røisland, Dr. Rygg, Dr. Flatø, and Dr. Songstad report funding support from Istituto Giannina Gaslini, Genoa, Italy, for the translation and data collection performed at their sites within the EPOCA project. 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. Dr. Consolaro, Dr. Bovis, and Dr. Hoftun have nothing to disclose.

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.

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