

REVIEW ARTICLE



Measuring health-related quality of life in young children: how far have we come?

Nicola Germain ^a, Samuel Aballéa^a and Mondher Toumi^b

^aHealth Economics and Outcomes Research, Creativ-Ceutical, Paris, France; ^bPublic Health Research Unit, Aix-Marseille University, Marseille, France

ABSTRACT

The importance of understanding the impact of disease and treatment on children's Health-Related Quality of Life (HRQoL) has given rise to an increasing use of child self-report and observer or proxy instruments. In this article, we review the status quo and challenges of HRQoL measurement specific to children under five. A number of HRQoL questionnaires exist for use with children and/or proxies, and both guidelines and reviews have been published on paediatric HRQoL. However, none address the challenges of measurement for children under five, for whom proxy measures should be used. In reality, there is significant heterogeneity in the cut-off age for self-report questionnaires. Recommendations are that proxies should be used for observable concepts, but not for concepts that require interpretation. Some research has been undertaken on dimensions/concepts in paediatric HRQoL questionnaires. However, no HRQoL models have been developed specifically for children, and heterogeneity in questionnaire dimensions underlines that there is no clear grasp of what HRQoL means in paediatric populations. There is a need to carry out research in order to develop theoretical models of HRQoL that are specific to children at different developmental stages, in order to evaluate and support new and existing measures for paediatric HRQoL and their use in clinical practice as well as clinical trials.

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Introduction



As the role of the patient has evolved over the past decades, so has that of the child as a patient. Children's perception of their disease and their opinions about their treatment have increasingly been solicited and given consideration in clinical practice [1,2]; the importance of understanding the impact of disease and treatment on the Health-Related Quality of Life (HRQoL) of children is now recognised. HRQoL is a complex, multidimensional concept, including social, emotional and physical functioning or well-being, related to the patient's health state [3]. This increased recognition has given rise to a growing use of child self-report and proxy-report instruments in paediatric clinical trials, and a change in the clinician-child-parent dynamic in clinical practice. Moreover, recent advances have rendered previously untreatable conditions treatable (e.g. Cystic fibrosis, Spinal muscular atrophy, Cancer). This has resulted in increased survival rates and life expectancy and the subsequent need to understand what the impact of this is on the paediatric patients' HRQoL. It is now generally accepted that children under the age of five cannot

reliably self-report [4], and that proxy reports should be used. For babies and infants who are unable to self-report, proxy reports are unavoidable. However, numerous studies have demonstrated inconsistencies between child and proxy reports [5–7] and the validity of proxy reports continues to be discussed.

In this article, we will discuss the status quo of HRQoL measurement in children under the age of five, as well as summarizing the main challenges involved in measuring HRQoL in this population that is unable to self-report. This article adds to a previous systematic review carried out in 2007 [8] which focused on the quality of generic HRQoL instruments for children under five years old.

Growing interest in paediatric HRQoL measures

There now exist a number of well-documented and validated generic HRQoL questionnaires for use with children and proxies, such as the Paediatric Quality of Life Inventory (PedsQL) [9], the Child Health Questionnaire (CHQ) [10], or the Quality of Life Scale for Children (QOL-C) [11]. Initially, few disease-specific questionnaires were developed, but these have now become more common,

CONTACT Nicola Germain  nge@creativ-ceutical.com  Health Economics and Outcomes Research Creativ-Ceutical, 215 rue du Faubourg St Honoré, Paris 75008, France

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and are available in a number of paediatric conditions such as asthma, diabetes, cancer and cystic fibrosis. Guidelines including recommendations on HRQoL evaluation in children have been published by ISPOR and the FDA [4,12]. The ISPOR Taskforce recommendations are specific to paediatric patient-reported outcomes (PRO) instruments, and present five good research practices: 1) 'Consider developmental differences and determine age-based criteria for PRO administration'. Four age groups are presented (zero to five years, five to seven years, eight to eleven years, and twelve to eighteen years) but it is recommended that age ranges should be adapted to each population. 2) 'Establish content validity of paediatric PRO instruments', where recommendations are made for interviews with children. 3) 'Determine whether an informant-reported outcome instrument is necessary'. This section presents differences between proxy and observer reports, and provides recommendations concerning these. 4) 'Ensure that the instrument is designed and formatted appropriately for the target age group', in which issues such as format, wording and representation through images are discussed. 5) 'Consider cross-cultural issues'. The FDA's paper refers only briefly to paediatric instruments, recommending that the same approach as for instruments for adults be employed. The importance of adapting vocabulary, comprehension, and recall periods is underlined, and the authors recommend 'fairly narrow age groupings' in order to take developmental differences into account, and clear determination of the youngest age at which children can self-report.

A large number of reviews [8,13–18] have been published documenting or evaluating the measures available for paediatric HRQoL. These reviews discuss the challenges involved in accurately capturing HRQoL in children. However, they have focused mostly on instrument properties, number of items and dimensions, the age range the instrument was developed for, the psychometric validation status, the languages in which it is available, and the disease areas in which it has been used.

More recently, authors have discussed and evaluated the conceptual frameworks and theoretic models underpinning these questionnaires. Work has also been undertaken to compare the concepts included in dimensions of the same name [19–21], notably by Fayed et al. [22] in a study mapping generic HRQoL paediatric measures to the International Classification of Functioning, Disability and Health (ICF). This work is discussed in more detail in the section entitled 'HRQoL definition and conceptualisation in children'.

Some reviews are also specific to HRQoL measurement in the paediatric population for clinical trials [23,24]. Raat [23] reviews recent HRQoL instruments for paediatrics and underlines that a combination of

these questionnaires – considered to be of good quality – should be used in clinical trials. The author specifies that the best approach for clinical trials is to use a combination of generic, disease-specific and preference-based measures, as well as both self- and parent-reports. Clarke [24] concludes that HRQoL measures are insufficiently used and reviews the quality and availability of specific instruments so as to be of help to clinical trial developers. The author highlights barriers to the inclusion of these measures, such as the availability of disease-specific measures, and the higher costs and longer timelines involved in the implementation of such measures.

A few publications exist on the use of paediatric HRQoL measures in clinical practice, but these are scarce in comparison to reviews of instruments and of their performance in the clinical trial setting. Varni [25] reviews the use of HRQoL tools in clinical practice, and underlines the paucity of data on the clinical utility of HRQoL measurement in paediatric clinical practice. Varni concludes that evidence must be generated about how the use of these measures can change clinical outcomes before clinicians will accept to incorporate HRQoL measurement into clinical practice. This subject is discussed in more detail by Haverman [26] who underlines that using HRQoL questionnaires in clinical practice can improve the discussion and monitoring of children's HRQoL. However, no examples of studies that demonstrate an improvement in clinical outcomes are cited by Haverman. The author highlights that online systems implementing HRQoL measures for adults into clinical care are available, but that only one (the KLIK project) was identified in the paediatric population.

Morris [27] published a scoping report regarding the incorporation of child and adult patient-reported outcomes measures into clinical practice in the UK, and concludes that the routine collection of child and parent-reported outcomes measures is feasible. However, major clinical guidelines for paediatric conditions, although recognizing that the HRQoL of children is important, do not give practical recommendations for the use of measures in clinical practice. One can commonly find quality of life referenced only in passing, for example 'The roles of specialized nursing, pharmacy, rehabilitation, and paramedical personnel and access to increasingly complex equipment and facilities are critical to improving long-term survival and quality of life.' – the only mention of HRQoL in the American Academy of Paediatrics' Guidelines for Paediatric Cancer [28].

Amongst the existing reviews and guidelines that focus on paediatric HRQoL measures, none of them address the particular challenges associated with measurement and data collection for children under the age of five. While a review on measures for this age range has

previously been undertaken [8], the aim was an evaluation of the quality of generic measures. The authors addressed practical implications specifically in the context of nursing practice.

Table 1 lists some HRQoL questionnaires for which a proxy report and/or child-report is currently available for children under the age of five. This is not an exhaustive list drawn up from a systematic review, but aims to illustrate the type of measures that are now available. Whereas some generic questionnaires exist for a number of age groups (e.g. The PedsQL, the KINDL, the FSII-R), providing versions from birth onwards in four or five versions, other disease-specific measures provide as little as one single version to be used as a parent report from birth to age 18 (The Paediatric Oncology Quality of Life Scale (POQOLS)) or from age one to age 18 (The Miami Paediatric Quality of Life Questionnaire). However, it should be noted that more recent disease-specific instruments such as the Haemophilia Quality of Life Questionnaire (Haemo-QoL) and the Cystic Fibrosis Questionnaire – Revised provide several age-adapted versions.

Whereas in adult clinical trials, the use of both generic and disease-specific instruments in parallel is now standard, this is not always the case in paediatric clinical trials. In any population, disease-specific instruments are more sensitive to change. Although the number of disease-specific instruments for children and/or proxies is growing, it lags far behind the number available for the adult population. Moreover, there is a need for easy-to-use shorter instruments for use in observational studies, registries and clinical practice. A major barrier to ease of use in these settings is the length of the questionnaires. The majority of currently available instruments for children aged under five contain a large number of items, notably the generic scales; as many as 74 for the Nordic Quality of Life Questionnaire, 43 for the Functional Status R (II), the Preschool Children Quality of Life questionnaire and the Infant Toddler Quality of Life questionnaire (ITQOL), and the shortest generic questionnaire being the Warwick Child Health and Morbidity profile with 16 items. However, a few disease-specific questionnaires are shorter, with the Haemo-QoL available in an eight-items version and the Quality of Life for Children with Otitis Media Questionnaire comprising six items. A well-validated generic HRQoL questionnaire for children under five (and/or their proxies) that is short and simple to use seems to be currently unavailable. Indeed, HRQoL questionnaires tend to be developed for the clinical trial setting and not for clinical practice. This

has resulted in questionnaires that may be valid and reliable tools for clinical trials, but that are not adapted to supporting decision-making in practice.

Proxy reports – the state of the art

There is a consensus that self-report should always be used where possible. Proxy reports are considered to be a valuable way of obtaining information about children whose age or cognitive/health status prevents them from reliably self-reporting. Self-reports are often supplemented with proxy reports for older children, and replaced by proxy reports for children who are too young or too ill to self-report. The issues surrounding proxy reports have been recognised for some time and have been addressed by numerous authors [16,20,29,30] who point out that conclusions from individual studies are contradictory. For example, some studies conclude that there is a greater agreement between child and proxy reports for older children [31], and others conclude the opposite [32] or that there is no effect of age on agreement [33]. Some studies show that parents under-estimate HRQoL [34] and others show that parents over-estimate [35].

Indeed, many authors have examined and discussed the reliability of proxy reports, by comparing child self-report to the proxy (parent) reports on the same questionnaires or concepts [34–38]. Most do so by examining the correlations between the child self-reports and the proxy reports using the same instrument, or a version of the instrument adapted for proxy reports. However, it has been suggested [20] that correlations are inappropriate for studying proxy-child agreement because they are not in themselves an indicator of agreement, and do not explain why proxies are over or under-estimating certain concepts. A comparison of the difference in mean scores, intraclass correlation coefficient (ICC) for continuous data and the Kappa statistic for categorical data could be more relevant indicators of consensus when comparing child and proxy reports [39]. It should also be noted that ICC can demonstrate consistency rather than absolute agreement.

Although some authors report that proxy reports seem reliable, studies evaluating consistency between child and proxy reports have failed to identify instruments where proxy reports can be substituted for the child reports in all dimensions [35]. Current recommendations are that proxy reports should be used for observable concepts, but are often unreliable when it comes to concepts that require interpretation, such as social functioning and emotional well-being. In addition, proxies' own QoL should be collected in order to evaluate the extent to which their own QoL impacts the perceived burden on their child(ren) [12].

Table 1. Generic and disease-specific HRQoL questionnaires for use with children <5 years old.

QUESTIONNAIRE	Generic/Disease-specific instrument	Age group versions	Self or proxy report	Questionnaire dimensions & Items
Pediatric quality of Life inventory (PedsQL)	Generic core module and Disease-Specific Modules for asthma, arthritis, cancer, cardiac disease, duchenne muscular dystrophy, cerebral palsy, cognitive function, diabetes, end stage renal disease, eosinophilic esophagitis, epilepsy	2–4 years 5–7 years 8–12 years 13–18 years (5–7 year old survey is interviewer administered; all others are self-administered)	Parent report only ages 2–4. All other ages self and parent report	23 items for generic core scale Physical Functioning, Emotional Functioning, Social Functioning, School Functioning.
Infant Toddler Quality of Life questionnaire (ITQOL)	Generic	2 months–5 years	Parent report	43 items. Physical functioning, Growth and development, Bodily pain, General Temperament /moods, General behaviour, Getting along, General health, Parental-emotional, Parental-time, Family activities, Family cohesion, Change in health
Warwick Child Health and Morbidity profile (WCHMP)	Generic	0–5 years	Parent report	16 items. General health status, acute minor illness status, behavioural status, accident status, acute significant illness status, immunization status, chronic illness status, functional health status, health related quality of life
Functional status II (R)	Generic	0–9 months 10 months – 2 years 2–5 years 5–11 years	Parent report	43 items. All age groups include total functional status and general health status factor. Stage-specific factors included in long form by age group are as follows: 0–2 Years: Responsiveness 2–3 Years: Activity 4 Years and Older: Interpersonal Functioning
TNO-AZL Preschool Children Quality of Life questionnaire (TAPQOL)	Generic	1–4 years	Parent report	43 items. Physical, social, cognitive, and emotional functioning
Nordic Quality of Life questionnaire for children	Generic	2–6 years 7–12 years 13–17 years	Parent report from age 2 and self-report from age 12	74 items. Global, external, interpersonal, and personal: objective and subjective. Included within these spheres are the following dimensions: physical, mental, spiritual, social, and economic
DISABKIDS	Generic core module for chronic diseases	4–7 years 4–16 years	Self-report with help from interviewer/nurse or parent and proxy report	12/37 items. Mental, social, and physical subscales
KINDL (KiddyKINDL, KidKINDL, KiddoKINDL)	Generic core module and extension for extended hospitalisation. Disease-specific modules for Adiposity, Asthma, Diabetes, Epilepsy, Neurodermatitis, Oncology, Spina bifida,	KiddyKINDL age 4–6 self KiddyKINDL age 3–6 parents KidKINDL age 7–13 self Kid_KiddoKINDL age 7–17 parents KiddoKINDL 14–17 self	Parent and self-report	12/24 items. Physical. Emotional. Self-esteem, family, friends, school
Behavioural, Affective and Somatic experiences scale (BASES)	Bone marrow transplant	<1 year – 20 years	Parent, nurse and self-report	38 items. Somatic Distress, Compliance, Mood/Behaviour, Interactions, Activity, and a group of miscellaneous items that were subsequently labelled Requests (6 items).
The Pediatric Oncology Quality of life Scale (POQOLS)	Cancer	0–18 years	Parent report	21 items. Physical function and role restriction, emotional distress, and reaction to current medical treatment

(Continued)

Table 1. (Continued).

QUESTIONNAIRE	Generic/Disease-specific instrument	Age group versions	Self or proxy report	Questionnaire dimensions & Items
The Miami Pediatric Quality of life Questionnaire	Cancer	1–18 years	Parent report	56 items. Self-Competence, Emotional Stability and Social Competence
Juvenile Arthritis Quality of Life Questionnaire (JAQQ)	Juvenile arthritis	2–8 years 9 – 18 years	Parent and self-report	74 items. gross motor function, fine motor function, psychosocial function, and general symptoms
Quality of Life for Children with Otitis Media (OM-6)	Otitis media	6 months – 12 years	Parent report	6 items. Physical suffering, hearing loss, speech impairment, emotional distress, activity limitations, and caregiver concerns
Haemophilia Quality of Life Questionnaire (Haemo-QoL)	Haemophilia	4–7 years 8–12 years 13–16 years	Parent and self-report	Long version 21–77 items according to age range. Short version 8 items. Physical health, feeling, attitude, family, friends, other persons, sport and school, coping, treatment, future
Cystic Fibrosis questionnaire Revised	Cystic fibrosis	3–6 years (currently being validated) 6–11 years 12–13 years 14+ years	Self-report proxy-report. Interview administered for young children	35–50 according to age range. Physical, role/school, vitality, emotion, social, body image, eating, treatment burden, health perceptions. 3 symptom scales: Weight, respiratory, and digestion

A key challenge inherent in the use of proxy reports is that observable signs, symptoms and concepts are principally measured as frequency; severity can only be reported when it requires observation and no interpretation. When a child can self-report, they can more reliably report on severity of the sign, symptom or experience also.

Authors discuss the discrepancies between child and proxy reports, the different approaches for analysing these, and propose explanations for weaker agreement between child and proxy reports in certain dimensions [16,20,33]. However, no discussion was found of the unique challenge of having to rely only on proxy reports and yet reliably and accurately measure HRQoL in children who cannot self-report. Indeed, it could be argued that proxy reports for HRQoL are themselves inconsistent with the notion of subjective perception of health states that is central to the definition of HRQoL. Moreover, parent proxies are to report only on observable behaviours or concepts [4]. One could, therefore, conclude that measuring HRQoL in children too young to self-report is not possible, and that only certain distinct dimensions of HRQoL can be measured in this population.

Another issue is that of response shift, where individuals' experience between two or more data collection points results in a change in their perception of their health-state and HRQoL. Response shift can explain why individuals whose health state has declined over time do not report an expected decrease in HRQoL. Response shift is an issue in longitudinal studies that fail to demonstrate a positive or negative impact on HRQoL in line with hypotheses. Authors have reported on response shift with proxy questionnaires, and evaluated this bias by various means such as structural equation modelling [40] and comparing responses to proxy HRQoL measures before surgical intervention and retrospectively after intervention [41]. In studies where both child self-reported and parent proxy-reported HRQoL is elicited, response shift may go some way to explaining differences between reports. Some studies demonstrate a larger response shift in the proxy reports [41], and some report the contrary [42]. In studies where only proxy-reports are elicited, and a response shift bias is present, it may be that the children – who have not self-reported – did not experience this change in perspective and perception, and that no response shift has taken place. Notably, Timmerman reported that response shift occurred in proxy reports even in observable dimensions such as physical suffering [41].

Cut-off and context for self-reporting

The ISPOR Task Force report [4] states that ‘This task force group does not think that it is possible to provide age cut-offs that will apply in all situations.’. However, it does recommend that children under the age 5 are not able to provide reliable self-reports, and that proxy measures should be used.

Several HRQoL questionnaires exist in versions for different age groups (for example, for the KINDL and the PedsQL), in order to take into account different developmental stages. The age groups for which a questionnaire is designed should be selected not simply on language and reading ability, but also on children’s ability to grasp concepts surrounding their health state and HRQoL. However, as shown in Table 1, there is significant heterogeneity in the cut-off age for self-report questionnaires and in the age up to which only proxy reports are used. For example, self-report starts age five for the PedsQL, age four for the DISABKIDS and the KINDL, whereas for the FS-R (II) only a parent report version is available up to age 16. Moreover, a search for completed phase three and four trials in the population under 18 on clinicaltrials.gov revealed that children are being asked to self-report pain from three years old, using ‘The Self-reported Wong-Baker Faces Pain Score’ (NCT01351298).

It is possible to carry out face-to-face interviews with children of a young age in order to elicit reports of their HRQoL. These interviews can be used to elicit data in a way that allows children to report impact on their HRQoL without being able to read. Although we were not able to find published evidence of such practices, it would be possible to carry out such interviews within clinical trials for rare conditions, or as part of exit interviews when the protocol does not allow for face-to-face interviews during the trial. However, this may not be an appropriate data-collection method for observational trials and registries due to the logistical and cost constraints for this type of study.

HRQoL definition and conceptualisation in children

A recent review [43] underlines the problems inherent in the fact that QoL and HRQoL have historically been used interchangeably, and identifies at least four different definitions of HRQoL that can be found in the literature. Briefly, these can be summarized as follows: HRQoL as functioning, HRQoL as QoL factors related only to health, HRQoL as all QoL aspects that are affected by health, and finally, the value of health. The author concludes that most questionnaires purporting

to be HRQoL measures, in fact, describe health using functioning and well-being, but that this is at odds with the definitions of QoL, and that a return to distinctions between health status measures and QoL measures may be necessary. This is supported by work carried out by Fayed et al. [22] in which WHO definitions of Functioning, Disability and Health (FDH) and QoL were applied to all items of the 15 most common generic measures used with children to evaluate HRQoL/QoL and functioning. The ICF was then used to describe the content of these measures. The results show that instruments such as the PedsQL, the KINDL and the TNO-AZL contain a majority of FDH content rather than HRQoL content. This conclusion overturns previous acceptance of well-known questionnaires as HRQoL measures because they are labelled as such.

In this paper, we recognize HRQoL as a complex, multi-dimensional concept, and measures should include the core dimensions of social, emotional and physical functioning or well-being, related to the patient’s health state [3]. However, these dimensions and even the underlying concepts are different in the paediatric population compared to the adult population, notably in the first few years of life that are characterised by rapid growth and development. Moreover, there is the need to include in a paediatric HRQoL questionnaire, concepts that evaluate not just functioning but address the child’s development and self-perception.

Table 1 shows that HRQoL instruments for children under five years old cover a wide variety of concepts, but do not consistently include the same core concepts of HRQoL, such as social, emotional and physical well-being. Individual authors add or substitute dimensions that they consider to be relevant for the given population, but these vary greatly across instruments. For example, dimensions have names such as sport and school, self-competence or behavioural status.

De Civita underlines that ‘Rarely have researchers acknowledged the notion of developmental change in their definition’ [20]. Indeed, the dimensions that constitute HRQoL are likely to evolve through different age groups; perhaps to a greater extent than reflected by the age group versions currently available in questionnaires, especially for infants and toddlers. For example, the FSII(R) has one version designed for children from two months to five years of age; the JAQQ has one version designed for children from two to eight years old. It is, however, unlikely that the same concepts are relevant for a very young infant and a school child. Current age group versions – although clearly a step forward compared to a ‘one questionnaire fits all’ approach – may not sufficiently address the developmental changes that occur over a period of several years in a child’s life.

Some research has been undertaken to evaluate the HRQoL dimensions and concepts in paediatric HRQoL questionnaires. Davis [19] evaluates the concepts and conceptual frameworks underpinning a number of paediatric questionnaires, and concludes that – although all instruments reviewed have been psychometrically validated – very few are constructed on a solid basis involving both a definition and a theoretical model of (HR) QoL. A number of (HR)QoL models have been developed, notably those by Wilson and Cleary [44] and the updated version by Ferrans in 2005 [45], but these were not developed for the paediatric population. This was identified as an issue when conceptualizing paediatric HRQoL by Taylor in 2008 [46]. In 2008, Valderas and Alonso propose a classification system of measures, linked to a conceptual model [47] that is also based on Wilson and Cleary's model, integrated into the ICF. Villalonga-Olives [48] tested Wilson and Cleary's model in the paediatric population in 2014 and found a conceptual gap between adults' and children's HRQoL, again encouraging researchers to concentrate on working towards conceptualizing HRQoL in the paediatric population.

Ravens-Sieberger [21] reviews a number of instruments and examined the dimensions and concepts contained in common generic and disease-specific HRQoL questionnaires for children. Many of these did not contain the dimensions recommended by the World Health Organization to be central to the definition of QoL. Kenzik et al. undertook a comparison of the KINDL, the KIDSCREEN-52, the PedsQL and the Child Health and Illness Profile [49]. The authors administered these questionnaires in an observational study and investigated structural, convergent/discriminant, and known-group validities. Correlation coefficients between dimensions purportedly measuring the same domain (physical well-being of the Kiddy KINDL and physical functioning of the PedsQL; psychological well-being of the Kiddy KINDL and emotional functioning of the PedsQL) were weaker than those measuring different domains (physical well-being of the Kiddy KINDL and emotional functioning of the PedsQL; psychological well-being of the Kiddy KINDL and physical functioning of the PedsQL). The authors also suggest that the ICF framework should be used to support comparisons in content. Huang [50] reviews this study and concludes with a key question, that is to say, what are the concepts being captured by these instruments? It seems that there is no clear notion of what HRQoL means in paediatric populations [16,19,20]. The work carried out by Fayed et al. [22] is a robust comparison that

illustrates the difficulties faced when selecting a HRQoL measure for use in children. It highlights the need to review the conceptual content of these questionnaires rather than relying on how they are reported in the literature.

Conclusions

A considerable amount of work remains to be done in the field of HRQoL measurement in children under five years old. There is a lack of user-friendly and short HRQoL instruments adapted for use in observational studies, registries and clinical practice. Reports of proxy measures for HRQoL in young children in the literature are almost entirely restricted to the clinical trial setting. However, there are a number of pharmacological/surgical interventions and childhood chronic diseases for which the collection of HRQoL data is highly relevant in infants and young children, not only in the clinical trial setting but also in clinical practice. Moreover, recent breakthrough treatments for severe childhood conditions require evidence for reimbursement and evidence of impact on HRQoL to aid decision-making.

A key issue is that, in the case of children under five who should not be asked to self-report, the ideal conceptual content of HRQoL questionnaires is at odds with the recommendations that proxies should only report on observable concepts. A continued review of the conceptual content of disease-specific questionnaires would be useful. This would allow comparison of concepts included in self-report questionnaires for each age group and support the validity of questionnaires as regards their conceptual framework. We must also reflect on whether proxy (parent) measures for children who cannot self-report should only include observable concepts, or whether a proxy report on other concepts such as social and emotional functioning is better than no assessment at all.

There is a need to carry out research in order to develop theoretical models of HRQoL that are specific to children at different developmental stages, in order to evaluate and support new and existing measures for paediatric HRQoL. It seems to be the case that instruments that purport to measure HRQoL are often – in fact – measures of functioning (ability to do things) rather than of well-being. The work undertaken by Fayed et al. to map content of generic questionnaires onto ICF definitions should also be undertaken for the most commonly used disease-specific HRQoL measures. There is also a need to carry out an updated systematic review of paediatric

HRQoL questionnaires for children under five years old, including disease-specific instruments.

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ORCID

Nicola Germain  <http://orcid.org/0000-0002-0559-3760>

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