VALUE IN HEALTH 21 (2018) 1419–1427

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Patient-Reported Outcomes

A Rights-Based Approach for Service Providers to Measure the Quality of Life of Children with a Disability

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ABSTRACT

Background: This paper identifies the best instruments for service providers to measure the quality of life (QoL) of children with a disability, with a focus on their alignment with the Convention on the Rights of Persons with a Disability (CRPD). **Methods:** This study reviewed systematic reviews to identify generic QoL instruments for children and adolescents, followed by an appraisal process using newly developed criteria. QoL instruments with a health status, functioning, and condition-specific focus were excluded. **Results:** Twenty generic QoL instruments for children were identified from existing systematic reviews to undergo further review. Only 2 of the 20 instruments were recommended for service providers to measure the QoL of children with a disability (KIDSCREEN and KINDL). Many pediatric QoL instruments (N = 9) focus on functioning and are not consistent with the CRPD, confounding a child's functioning with their feelings about their life. KIDSCREEN and KINDL have self-report and parent report versions, are applicable for childhood and adolescence, demonstrate adequate reliability and validity, involved children in their development, focus on wellbeing, are likely to be able to be completed by a child with a disability, and are low in cost. **Conclusions:** Many instruments focus on functioning rather than wellbeing and thus may not capture the QoL of children with a disability. A child's functional limitations may not be consistent with their feelings about life. Two instruments that assess wellbeing and meet the criteria important for service providers now require further testing to explore their usefulness and validity for children with varying abilities.

Keywords: child disability, Convention on the Rights of Persons with a Disability, disability service provider, generic instruments, quality of life, wellbeing

Introduction

Quality of life (QoL) is defined by the World Health Organization as "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" [1]. QoL encompasses multiple domains of life including physical, social, emotional, and environmental. It is arguably one of the best outcomes of childhood wellbeing because it emphasizes an individual's subjective experience. For children with disabilities, it is particularly important not to restrict QoL to health-related issues, but to include a person's holistic wellbeing [2]. In a mother's words, "The most important thing to remember is that when a child is born, that child is a child first to his or her family. It is his wellbeing, not his disability, which affects all of our everyday lives" [3].

There are no theories of pediatric QoL; however, researchers have considered how QoL fits with the widely accepted International Classification of Functioning, Disability and Health (ICF) [4]. The ICF classifies health and health-related domains and proposes a dynamic system of interrelationships between environmental factors, personal factors, body structure and function, activity, participation, and health. McDougall et al. [5] suggest that QoL encompasses the whole ICF model and is broader than health and functioning. An alternative approach to the ICF, which may appeal more to clinicians and consumers, is the simpler "F-words"

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1098-3015/$36.00 - see front matter Copyright © 2018, ISPOR—The Professional Society for Health Economics and Outcomes Research. Published by Elsevier Inc.
https://doi.org/10.1016/j.jval.2018.05.009

of *fitness, function, friends, family, future,* and *fun* [6]. As with the ICF, it is argued that QoL encompasses all six F-words and is perhaps more clearly articulated with this approach.

In clinical practice, health-related quality of life (HRQoL) instruments are useful in identifying and prioritizing health problems, particularly hidden problems, facilitating communication between staff and the patient or family, and monitoring change over time [7]. Measuring the QoL of children is complex, and it can be challenging to capture the child's perspective. A child's ability to self-report depends on her or his age, developmental status, intellectual ability, and communication skills. While QoL is theoretically an individual's perspective, for some children, it is not possible with current technologies to capture their self-reported QoL. In these cases, parent report may be useful. Even for children who can self-report, there is evidence that child self-reports and parent reports are not highly correlated on existing QoL tools [8–10]. Qualitative research suggests that this is partly children and parents considering different events in evaluating their QoL [11]. This suggests that a parent report can provide a different perspective and additional insights into a child's life. Article 7 of the Convention on the Rights of Persons with a Disability (CRPD) states: "Children with disabilities have the right to express their views freely on all matters affecting them, their views being given due weight in accordance with their age and maturity, on an equal basis with other children, and to be provided with disability and age-appropriate assistance to realize that right" [12]. To be consistent with Article 7, QoL instruments must be able to be completed by children with a disability (where feasible), and measure areas of life perceived to be meaningful by them [13].

The number of instruments developed to measure the QoL of children has grown steadily in the last 15 years with multiple systematic reviews identifying and assessing their content and usability in reporting children's health [14–17]. Most of these reviews describe the number of items, age range, reporter (self or proxy), completion time, and psychometric properties. Recognizing that these basic descriptive criteria are limited when instruments vary greatly in conceptual approach, Waters and colleagues produced a set of more detailed conceptual criteria to aid clarity [18,19]. These include the original purpose and focus of the instrument, origin of domains and clarity of items, and threat of negative wording to self-esteem. Although these conceptual criteria are useful in selecting an instrument for research, additional information is needed to select a QoL instrument for a service provision setting. It is particularly important to consider whether the instrument suits the purpose of the investigation, if the dimensions covered are relevant to the context and ensure that the instrument has good psychometric properties with demonstrated reliability and validity, and that it is sensitive to change if intended to monitor health and wellbeing over time [14]. Condition-specific instruments are designed to capture issues specific to a certain condition as well as generic domains of QoL. Generic instruments are designed to include domains of importance to all children and are useful for comparing children with different conditions—as experienced in a service provision context. Given the sheer number of instruments available and the different population groups they have been designed for, it can be difficult for service providers to choose the most appropriate instrument that measures the QoL of children with a disability and captures their holistic wellbeing.

Aim

The aim of this paper is to identify the best instruments for service providers to measure the QoL of children with a disability, with a focus on examining how the instruments align with the CRPD and pragmatic considerations in a service provision context.

Methods

This study consisted of a review of systematic reviews to identify generic QoL instruments for children and adolescents followed by an appraisal using newly developed criteria.

Search Strategy

Phase I: systematic review search

Search strategy: To identify existing systematic reviews of QoL measures for children, an initial literature search was conducted in January 2015. To identify available reviews, the search strategy included the following five groups of terms: "systematic review," "quality of life and QoL," "child*," "instrument and tool* and questionnaire," and "generic." The following databases of online peer-reviewed journal articles were searched: EBSCO databases, PubMed, Web of Science, Cochrane library, and Google Scholar linked with the University of Melbourne library database.

Inclusion criteria: Systematic reviews were included if they contained generic QoL or health-related QoL instruments for children or adolescents; reported their psychometric properties regarding the reliability or validity of the instrument; were written in English; and were published between 2005 and 2015. This 10-year timeframe was chosen to be confident all existing generic QoL measures published within the literature could be critiqued.

Exclusion criteria: Reviews were excluded if the included measurement instruments were designed for specific or chronic medical conditions, reviewed measurement instruments also designed for adults, and/or were conducted more than 10 years ago.

Phase II: instrument inclusion and exclusion

Instruments from the included systematic reviews were categorized by three reviewers (E.D., D.Y., K.M.G.) based on how they assessed QoL. Any discrepancies regarding the inclusion of an instrument was resolved through discussion among the reviewers. Each instrument was assessed and categorized on its primary focus: wellbeing, functioning, or health status. Instruments that were deemed to have a predominant wellbeing focus were then assessed against the appraisal criteria.

Phase III: review of included instruments against selected criteria

The co-authors of this review combined their collective expertise in clinical, academic, home-, and community-based service provision for children with disabilities to identify key criteria for service providers selecting QOL instruments. First, it was determined that the instrument should be generic because service providers are generally supporting large numbers of children with varying impairments and need to be able to aggregate results for the entire service. In theory, all generic instruments should be applicable; however, some instruments contain items or scoring that may be too complex to be completed by younger children or children with a mild intellectual disability. Second, the instrument should be relevant across childhood and adolescence, with age-appropriate versions so that there is the potential to track children over time. Third, the instrument needs to be reliable, valid, sensitive to change, low cost, and quick to complete to facilitate regular completion (e.g., twice a year). Fourth, although it is well accepted that human and legal rights are essential components to QoL for adults [20], a rights-based approach has not been applied to the measurement of QoL of children. As service providers need to ensure that their practice aligns with the CRPD, examining whether pediatric QoL instruments are consistent with the CRPD is necessary. Finally, aligned with the rights-based approach, it is important that children with a disability can report high QoL if it exists for them, as evidence shows that

functioning is unrelated to wellbeing [21]. It is important therefore that a QoL measure can capture the child's subjective wellbeing.

In summary, the service provider appraisal criteria for QoL instruments for children with a disability are outlined in Table 1, including standard and additional appraisal criteria aligned with the CRPD. Data were extracted from the included instruments regarding reporter (self or proxy), age range, psychometric properties (reliability, validity, and sensitivity), who was involved in the development of the instrument (i.e., children, parents, clinicians), focus of the items (i.e., wellbeing, functioning), instrument cost, length and time taken to complete, and the applicability for children with a disability.

Results

Systematic Reviews

Eighteen systematic reviews were identified in the search (Fig. 1). Sixteen were excluded because they were conducted before 2005, focused on condition-specific instruments, or were designed for adults. Two reviews reviewed QoL instruments for children and adolescents and fit the inclusion criteria [14–16].

QoL Instruments

The two systematic reviews included a combined total of 42 QoL and HRQoL instruments. All of these instruments were categorized by the authors to identify those that were designed to measure QoL with regard to wellbeing (N = 20).

Excluded QoL instruments

Of the 42 instruments, 22 were excluded from this review as they were designed to measure health status or functioning (including CHQ, CHIP, CHRIs, CHRS, CHSCS, CLQI, COOP, CHRS, DHP, FSII(R), HP, HSCS-PS, PIE, VSP-A, WCHMP), or were preference-based QoL measures used for economic evaluations of quality-adjusted life years and to describe the health status of an individual (including 16D/17D, AQoL-6D, CHSCS-PS, CHU-9D, EQ-5D-Y, Health Utilities Index Mark II/III, SF-6D). Supplemental Material lists the instrument names referred to by acronym.

Table 1 – Criteria important for service providers to choose a QoL instrument for children with a disability

Criteria for service providers	Applying to QoL measurement
Does the instrument measure QoL?	Is it reliable, valid, and sensitive to change?
Is the instrument applicable for children with a disability?	Has it been used by children with a disability previously?
	Is the questionnaire able to be completed by a child with a disability? (short, single items, easy response scale)
Cost and length	Is the instrument freely available or of low cost?
	Is the instrument short in length?
Alignment with Article 7—Children with disabilities have the right to express their views freely on all matters affecting them.	Was the questionnaire developed in consultation with children/parents?
	Is there a self-report version available?
	Are there age-appropriate forms available for children and adolescents?
Can children report high QoL if it exists for them?	Do the items focus on wellbeing and feelings rather than functioning?

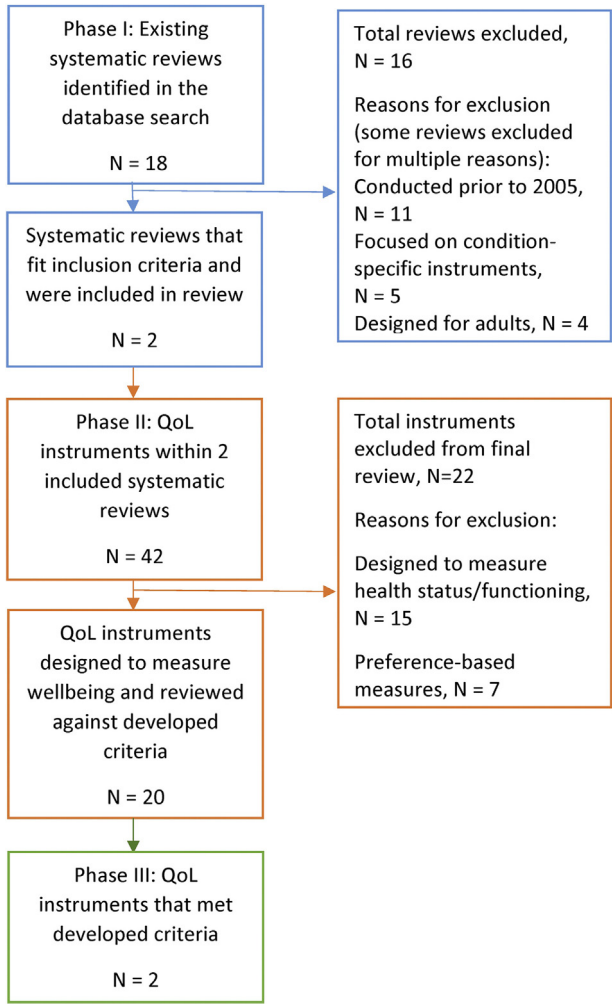

Fig. 1 – Flowchart of search results and selection process.

Included QoL instruments

Table 2 presents information on the instruments deemed by the authors to measure QoL (N = 20), including age range, psychometric properties, development of the instrument, focus of the items (i.e., wellbeing, functioning, or health status), cost, length, and applicability for children with a disability.

Only 2 of the 20 instruments met all of the criteria (KIDSCREEN and KINDL). These two instruments have self-report and parent report versions, are applicable for childhood and adolescence, demonstrate adequate reliability and validity, involve children in their development, focus on wellbeing, are likely to be able to be completed by a child with a disability, and are low cost. Additional details are provided below.

Reporter

The majority of instruments were self-report only (N = 9) or parent proxy and self-report (N = 9). Only two instruments were

Table 2 – QoL instruments for children and adolescents assessed against appraisal criteria for service providers

	QoL instrument name	Child self-report version?	Age range (year)	Reliability*	Validity†	Sensitivity‡	Was the instrument developed in consultation with children/ parents?	Do the items measure wellbeing or functioning?	Able to be completed by children with a disability (i.e., short, simple)	Cost	Length	Reviewers comments
1	AUQUEI [25]	Self	4–12	0	+	0	No	Wellbeing e.g., How do you feel?	Yes	No	27 items	Use of sad and smiley faces useful for children. Assessment of child satisfaction with several QoL domains (family life, social life, children's activities [school and leisure], health)
2	QUALIN [23]	Parent	3 months – 3 years	+	+	0	Yes, parents only	Wellbeing e.g., This child looks well.	N/A	UA	34 items	Instrument designed to assess infant's quality of life (between 3 months and 3 years of age) and is completed by the parent or carer due to child's age.
3	CQoL [33]	Self & parent	9–15	+	+	0	Yes	Functioning e.g., Over the past month how well have you been in these ways (see scale)?	No	UA	15 items	Initial interviews with children during development focused on areas of QoL that were impaired. The questions are considered complex and based on functioning and the child's emotional reactions to their level of functioning (i.e., satisfied or upset).
4	Exqol [27]	Interviewer administered	6–12	–	+	0	No	Wellbeing? e.g., This is Daniel. He is feeling poorly. He knows he gets sick more often than his friends—"How much are you like Daniel?"	Interviewer assisted	UA	24 items	The instrument's theoretical model is based on an assumption that poorer quality of life is the result of discrepancies between an individual's actual and ideal self. Questions may provide too much information and be too complex for children with a disability.
5	GCQ [34]	Self	6–16	+	+	0	Yes	Wellbeing e.g., How much of the time do you feel happy with your life?	Yes	£118	25 items	The discrepancy between the actual and the desired viewpoints are used to establish the quality of life, with the size of the discrepancy that counts. Discrepancy-based measure might affect self-esteem in children with a disability and therefore deemed inappropriate for use with children with a disability.
6	HAY [35,36]	Self Self & parents	6–12 7–13	+ +	+ +	0 0	Yes	Functioning e.g., How well are you able to ride your bike?	Yes	UA	44 items 80- items	Developed for children with a chronic illness, the questionnaire consists of a generic section and a specific section for children with asthma. Explores daily functioning of the children and their associated feelings with the limitations they experience.
7	ITQoL [22]	Parent	2 months to 5 years	++	+	0	No	Functioning e.g., Has he/she been limited in any of the following? Sleeping, grasping, rolling over	N/A	US $400	47 items and 97 items	Limited to assessing infants. The questionnaire is lengthy and has possible ceiling effects. Focused concepts include limitations experienced in the health and functioning of the child. Cost applicable to funded research, higher for ongoing use for patient care.
8	KIDSCREEN [37,38]	Self & parent	8–18	+	+	+s	Yes	Wellbeing	Yes	€40	10-, 27- and 52-item options	Good psychometric properties. Child-friendly computer version. Allows for cross-cultural comparison. Deemed useful particularly if collecting follow-up data and in longitudinal studies. Content for the questionnaire was developed using a literature review, expert opinions, and focus groups with children, adolescents, and parents.
9	KINDL [26]	Self Parent	4–6 7–13 14–17 3–6 7–17	+ + + +	0 + 0 0	0 +s 0 0	Yes	Wellbeing e.g., I had fun and laughed a lot.	Yes N/A	No	24 items	Strengths include use of age-appropriate scales. Child-friendly computer version with clear questions.

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Table 2 – *continued*

	QoL instrument name	Child self-report version?	Age range (year)	Reliability*	Validity†	Sensitivity‡	Was the instrument developed in consultation with children/ parents?	Do the items measure wellbeing or functioning?	Able to be completed by children with a disability (i.e., short, simple)	Cost	Length	Reviewers comments
10	Nordic QoLQ for Children [39]	Self Parent	12–18 2–18	0	+	0	UA	UA	No N/A	UA	74 items	Designed for the family to complete together. Long and complex as questions are also collected of the family context and demographic factors (i.e., income, employment status, and profession)
11	PedsQL 4.0 [40,41]	Parent Self & parent	2–18 5–18	+	++	+s	Yes	Functioning e.g., It's hard for me to walk more than one block.	N/A Yes	US$ 6647	23 items	Focused on functioning. Very costly, which would be prohibitive for a service to utilize.
12	PQ-LES-Q [42]	Self	6–11 12–17	++	0	0	No	Functioning e.g., Over the past week how have things been with your health?	No	No	15 items	Focus of the instrument is on physical and psychological functioning. Has not involved children or families in its development.
13	PWI [43]	Parent Self	Up to 5 5–18	+	++	0	No	Wellbeing e.g., How happy are you with your health?	Yes	No	7 items	A valid and reliable tool developed to measure the subjective wellbeing dimension of QoL, although appears to lack sensitivity to service provider's intervention. The PWI is intended to represent satisfaction with life as a whole, uses a simple 10-point scale. Possible difficulties in use by people with communication difficulties. Predefined domains were not informed by parents.
14	QoLP-AV [29]	Self	14–20	+	+	0	Yes	Wellbeing e.g., How important/ satisfied am I with how I feel about myself?	No	CA$45	54 items	Limited to assessing adolescents. Aligns with our approach to defining QoL; underpinned by the conceptual model of quality of life "How good is your life for you?" Quite lengthy and therefore may be difficult for a child with a disability to complete.
15	QoLQA [30]	Self	10–15	+	0	0	Yes, parents only	Wellbeing and functioning e.g., Difficulty in doing everyday life activities, such as eating or changing your clothes.	No	UA	70 items	Limited to assessing adolescents. Lengthy scale with 70 items. Specifically developed QoL assessment instrument for adolescents in Asian countries that can be used for international comparison.
16	QoLQC [28]	Self & parent	8–12	+	+	0	No	Functioning e.g., I can participate in any physical activity I want to participate in.	No	UA	118 items	Assessment of three broad domains of functioning: physical, psychological, and social functioning. Author concluded that instrument required further revision; psychometric properties were promising but further research was needed. Limited literature on use of scale.
17	TAPQoL [44] TACQoL [45]	Parent Self & parent	1–5 6–15	+ +	+ ++	0 0	Yes, parents only No	Functioning e.g., Did you have difficulty with walking?	N/A No	No	43 items 108 items	Assessment of functioning and difficulties with tasks. The moderately complex scale may be difficult for a child with a disability to complete.
18	TedQL [24]	Self & parent	3–8	–	+	0	No	Functioning e.g., Are you good at running or not good at running?	Yes	UA	22 items	Self-report instrument designed for use by young children involving a child-friendly format. Contains items that assess if the child is good at an activity (i.e., running).
19	TQoLQA [31]	Self	13–15	+	0	0	No	Wellbeing and functioning e.g., Do you have any difficulty in performing your daily activities?	No	UA	38 items	Limited respondent age range of 13–15 years. Considered culturally inappropriate for use in Australia as designed specifically for Taiwanese adolescents. Deemed inappropriate for children with a disability as the instrument consists of negatively worded items that could threaten the

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Table 2 – *continued*

	QoL instrument name	Child self-report version?	Age range (year)	Reliability*	Validity†	Sensitivity‡	Was the instrument developed in consultation with children/ parents?	Do the items measure wellbeing or functioning?	Able to be completed by children with a disability (i.e., short, simple)	Cost	Length	Reviewers comments
												self-esteem of the child. For example, "Do you feel inferior because of your appearance?"
20	YQoL [32]	Self	11–18	++	++	0	Yes, adolescents only	Wellbeing e.g., I feel good about myself.	Yes	US$25	15 items	Strong theoretical foundations, development employed a qualitative grounded theory approach. Limited to assessing adolescents, although the scale is deemed appropriate and useful

N/A, not applicable; UA, information unavailable to assess against criteria.
* 0, not reported; −, reliability is not acceptable in terms of one or both aspects (internal consistency and/or test–retest <0.70 in 40% or more of the dimensions); +, only one type of reliability (internal consistency or test–retest) has been tested, with acceptable results; ++, reliability is acceptable in both aspects (internal consistency and test–retest stability >0.70 in 70% or more dimensions).
† 0, not reported; −, validity is not acceptable in one or more aspects (structural, construct, and/or criterion); +, only one type of validity has been tested, with acceptable results; ++, two types of validity tested with acceptable results; +++, all three types of validity tested with acceptable results.
‡ 0, not reported; −s, sensitivity to change has been assessed with negative results; +s, sensitivity to change has been assessed with acceptable levels.

parent proxy report only and these were for very young children (ITQoL [22] and QUALIN [23]).

Age Range

QoL instruments are available for children aged 2 months to 18 years. Self-report versions are available for children as young as age 3 (TedQL [24]) or 4 (AUQUEI [25], KINDL [26]). Six instruments focused on children only (N = 5; AUQUEI, QUALIN, ExQoL [27], ITQoL, QoLQC [28], TedQL) and four focused on adolescents only (QoLP-AV [29], QoLQA [30], TQoLQA [31], YQoL [32]). The remaining 10 were designed to apply across childhood and adolescence (CQoL [33], GCQ [34], HAY [35,36], KIDSCREEN [37,38], KINDL, Nordic QoL Questionnaire for children [39], PedsQL 4.0 [40,41], PQ-LES-Q [42], PWI [43], and TAPQoL [44]/TACQoL [45]).

Psychometric Properties

The majority of instruments have acceptable reliability and validity (N = 16). Only four instruments have not been tested for reliability (AUQUEI, Exqol, Nordic QoL for Children, TedQL) or validity (PQ-LES-Q, QoLQA, TQoLQA). However, most instruments have not been tested for sensitivity to change (N = 17). The exceptions are KINDL, KIDSCREEN, and PedsQL 4.0, which demonstrate acceptable results. Sensitivity to change was demonstrated in the Spanish KIDSCREEN follow-up study with a stratified sample (N = 454) demonstrating moderate effect sizes across 8 out of 10 dimensions, ranging from −0.10 to −0.34 [46]. KINDL has been completed by 5781 healthy and chronically ill children and adolescents and has been shown to be sensitive to change in patients under treatment, with moderate to high effect size estimates related to treatment (d > 0.6 in modules) [47].

Involvement of Children in Instrument Development

Eleven of the instruments were based on consultations with children or parents. The remainder were based on researcher or clinician knowledge or previous instruments. For example, KIDSCREEN items were derived from literature review, expert consultations, and focus groups with children and parents [48]. KINDL items were also derived from focus groups with children.

Can Children Report High QoL If It Exists?

Eleven of the instruments focus on wellbeing; the remaining 9 focus more on functioning. Examples of items measuring wellbeing include, "How do you feel about …" (AUQUEI), "How much of the time do you feel happy?" (GCQ), "I had fun and laughed a lot" (KINDL), and "I feel good about myself" (YQoL). Items measuring functioning include, "How well are you able to ride your bike?" (HAY), "It's hard for me to walk more than one block" (PedsQL 4.0), and "Did you have difficulty with walking?" (TACQoL).

Able to Be Completed by a Child with a Disability

Nine of the instruments were deemed to be suitable to be completed by a child with a disability. These were short in length and had simple questions and rating scales. For example, KIDSCREEN employs simple, five-point Likert-type rating scales, and has been used in a large study with children with cerebral palsy in which 61% of the children were able to self-report [21]. Instruments that were deemed to be less appropriate for children with a disability were those that included moderately complex scales and item wording; for example, the TAPQoL and TACQoL questions are designed to first assess the occurrence of a functional problem or limitation and then how the child feels about it: "Did you have difficulty with walking? At the time I felt … ."

Cost

Instruments were deemed to be suitable for use by service providers if they are free or low cost. Six instruments were free for use (AUQUEI, KINDL, PQ-LES-Q, PWI, TAPQoL, TACQoL) and four were deemed low cost (GCQ, KIDSCREEN, QoLP-AV, YQoL), ranging up to AU$200. PedsQL 4.0 is costly with access ranging from $1000 to $20,000, which would be very prohibitive for use by a not-for-profit disability service [49]. Cost information cost was unavailable for nine of the instruments (QUALIN, CQoL, Exqol, HAY, Nordic QoLQC, QoLQA, TedQL, TQoLQA).

Discussion

This review highlights that of 42 generic QoL instruments for children, only 2 (KIDSCREEN and KINDL) met criteria incorporating both a rights-based approach and practical considerations for use in a service provision setting, and were also sensitive to change. This review identified two important issues. First, although a generic QoL instrument is necessary for service providers supporting children with varied disabilities, it is important that the instrument captures the issues that have been identified to be important to children with a disability by seeking their perspective. In many cases this does not differ significantly from what all children perceive to be important for QoL, particularly around family, social, and wellbeing domains. Differences may arise around impacts on daily living from pain and discomfort and adequate access to therapy and other services [13,48,50]. Second, many of the generic QoL instruments included within recent systematic reviews focus on functioning rather than wellbeing and thus may not capture the QoL of children with a disability given that functional limitations do not always correlate with lower QoL. This review highlights a number of issues with current QoL instruments that warrant further discussion and examination. These include whether generic instruments are useful to capture the QoL of children with a disability; the lack of information on QoL instruments' sensitivity to change; the inclusion of functioning items to measure QoL; and the absence of developmentally appropriate methods to identify domains of QoL of importance to children. These issues are also supported by Solans et al. [14], who recommended that developers of new instruments include children from the beginning of the development process and test instruments' sensitivity to change.

Using a Generic Instrument for Children with a Disability

Although a generic QoL instrument is necessary for service providers supporting children with a range of disabilities, the instrument needs to capture issues of importance to children with disabilities. Some items on the recommended instruments may not be applicable to some children with disabilities, such as KIDSCREEN items on schooling and free time. Further testing is necessary to examine whether generic instruments capture domains of QoL of importance to children with a disability and to establish psychometric properties of instruments for this cohort. Insight into this issue could come from the development of DISABKIDS—a QoL instrument for children with chronic illnesses and disabilities. DISABKIDS employed a "bottom-up" (patient-derived) method of questionnaire construction involving children and adolescents with a chronic condition [51]. DISABKIDS measures the mental, social, and physical domains of HRQOL. Within each domain are two dimensions: independence and emotion (mental domain), social inclusion and social exclusion (social domain), and limitation and treatment (physical domain) [49]. DISABKIDS was designed for comparisons across children with various chronic conditions but is not a generic instrument able to assess healthy children and thus was not included in this review [52]. DISABKIDS is closely linked with KIDSCREEN. However, a criticism of DISABKIDS is that it may threaten self-esteem by making assumptions about illness and therefore not measuring wellbeing [19].

Although the current review considers the simplicity of response scales within instruments, there is limited research on how accessible any QoL tools are for children using augmentative or alternative communication, such as those with vision impairments. As far as the authors are aware, existing instruments are not available in alternate formats that may be easier for these children to complete. In these cases, a self-report instrument is generally interviewer administered by either a parent or health professional. It is unknown how this may influence children's responses. Future work is required in adapting existing or new QoL measures for children using alternate communication to ensure that they can self-report their QoL.

Sensitivity to Change

Sensitivity to change refers to an instrument's ability to detect clinically important differences over time. It is concerning that many QoL instruments have not yet been assessed for sensitivity to change or what constitutes a meaningful change, especially as these instruments are often used to evaluate interventions. This is a serious issue that requires further examination given that there is much discussion in the literature as to whether QoL does in fact change over time. The empirically supported Homeostatic Theory of Subjective Wellbeing [53] argues that subjective QoL is determined by internal dispositional factors and general human resilience, such that individuals maintain wellbeing within a very limited range over time regardless of changing circumstances. Nevertheless, Cummins promotes the use of measures of QoL in service provision [54], on the basis that scores outside the normative range indicate concerning circumstances temporarily defeating a person's ability to maintain normal homeostatic wellbeing. Without information on sensitivity to change of QoL instruments, it is difficult to understand what is a meaningful change. Even if individual instruments provide normative information, without knowing what constitutes minimal clinically important difference in QoL, there is limited utility for service providers to measure it.

Using Items on Functioning to Measure QoL

This review highlights the problem with relying predominantly on items measuring function to assess QoL for children with a disability. This is discriminatory and inconsistent with the CRPD framework as there is now substantial evidence that functioning is unrelated to wellbeing [2,55,56]. It is important that children with a disability can report high QoL if it exists for them. This becomes difficult if QoL items measure functioning rather than wellbeing. For example, the physical domain of Pediatric QoL Inventory (PedsQL 4.0) [41] is measured by items such as, "It is hard for me to walk more than one block," making it difficult for a child with a severe physical disability and impossible for a child in a wheelchair to score highly. Similarly, in the domain related to school, items such as "I miss school to go to the Doctors or hospital" reflect functioning more than wellbeing.

A seminal publication by the European group SPARCLE showed that the overall QoL of children with cerebral palsy did not differ from that of their able-bodied peers, though some domains showed differences [21]. This was repeated with adolescents more recently [57]. Furthermore, despite large differences in functioning among those with cerebral palsy, there was minimal QoL difference between those with most and least impairment in both studies. This is consistent with research for people with other disabilities who often report good QoL despite impaired functioning [2]. This highlights the requirement for items to focus on wellbeing rather than functional or health limitations and to capture the same opportunity, goals, and QoL needs the child has, similar to all children.

Developmentally Appropriate Methods

Although many of the instruments were based on the perspective of children and parents, there have recently been many advances in understanding age-appropriate ways to engage children that need to be incorporated into QoL research. From a rights-based perspective, it is imperative that children can contribute their views to research about them. Qualitative researchers have

designed novel methods, using drawing, photography, play-based activities, and new technologies to adapt to children's abilities across developmental ages and physical or other impairments [58]. Adapting research to the abilities of participants not only results in richer data, but encourages participation from marginalized groups—including children and young people with disabilities [59]. Without appropriate methods to engage all children in conceptual development, QoL instruments may not capture the key issues affecting subjective QoL for those children. It is worth noting that none of the instruments included here reported using specific child-centered methods in their development.

As Rosenbaum and Gorter [6] discussed, childhood disability has traditionally been viewed through a biomedical lens, with measurement tools aligning with this approach to health and QoL. An instrument aligned with the F-words (*fitness, function, friends, family, future,* and *fun*) approach would enable children with a disability to report strengths in their life and achieve high QoL if it exists for them. Further work needs to address these issues.

Limitations

This study reviewed generic QoL instruments for children and adolescents to examine their usefulness for service providers. Identifying instruments from existing systematic reviews instead of conducting an independent search is a limitation of this study. The appraisal criteria are newly developed for this review based on the CRPD articles deemed most important for promoting children's rights and based on the authors' expertise in this field of research; we acknowledge its subjectivity. Lastly, reviews of generic instruments were limited to QoL tools in English only and do not consider cross-cultural validation for any instruments. For some service providers, this may be an additional consideration in choosing an instrument. Cross-cultural validation of existing QoL instruments for children with disabilities is complex and would be a worthwhile focus for future research [60].

Conclusions

This study identified available instruments for service providers to measure the QoL of children with disabilities and assessed their alignment with the CRPD and pragmatic considerations for service providers. Although a total of 20 generic QoL instruments for children were identified, the appraisal showed that only two instruments followed a rights-based approach and were likely to be feasible for regular use by service providers as part of their evaluation and quality assurance and to inform clinical practice. KIDSCREEN and KINDL measure the areas of life identified through focus groups and interviews as important to children and their parents, are simple and quick to complete, are low cost, and have emerging evidence sensitivity to change. These instruments assess physical and psychological wellbeing, including self-esteem, moods and emotions, family relationships and autonomy, and social environments, including schooling. KINDL also takes into account developmental differences between children and adolescents. The instruments have been used mostly in Europe; further testing is needed to examine cross-cultural validity.

Acknowledgments

This research was supported by a National Health and Medical Research Council of Australia partnership project grant.

Supplemental Materials

Supplementary data associated with this article can be found in the online version at https://doi.org/10.1016/j.jval.2018.05.009.

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