CONTENT ISSUES IN CHILD HEALTH STATUS AND QUALITY OF LIFE INSTRUMENTS: ADDRESSING THE CHALLENGES WITH NEW METHODS.

CONTENT ISSUES IN CHILD HEALTH STATUS AND QUALITY OF LIFE INSTRUMENTS: ADDRESSING THE CHALLENGES WITH NEW METHODS

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Thesis Abstract

Background: Health status and quality of life (QOL) instruments developed for children were created with many different conceptual perspectives. These perspectives were based on contextual understandings of the terms health and QOL at different points in time. As a result, there is a wide variety in the perspectives, health and life domains that are measured with these instruments.

Purpose: This thesis is dedicated to resolving conceptual inconsistencies that arise from various instruments using content analysis techniques.

Method: A method for analyzing the content of self-report instruments has been created and validated for adult measures. This method uses the International Classification of Functioning, Disability and Health (ICF) as the conceptual framework and the standard terminology to code each item from each instrument. Content analysis using this method will be reviewed, revised and validated for child health status and QOL instruments.

Results: The content analysis method was revised to resolve issues regarding the perspective of instruments (e.g. health status versus QOL). Once applied to child self-report instruments, many inconsistencies between measures and their application were discovered.

Discussion: The field of child health and QOL measurement applies and interprets the use of health status and QOL instruments inconsistently and this impacts upon content validity. Additionally, the repercussions of conceptual inconsistencies have an important effect on consequential validity in child health.

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When I remember this time in my life, I will always think of my parents: Salwa (Sally) Abou Omar and (the late) Muhammad El-Sawi Fayed, to whom I dedicate this thesis.

Thesis layout and organization

This thesis is organized in two parts, bracketed by an introduction and a discussion. In Part I, a new method for dealing with content issues is examined and revised for the purposes of this thesis. In Part II, the method developed in Part I is applied to address the problems with content in child health measurement

Introduction

The introduction presents a problem statement regarding current limitations in the field of patient-reported outcomes and child health measurement. A brief literature review outlining the history of child health measurement and the theoretical approach to the thesis is described.

Part I: Method Development

In Chapter One, a systematic review examines previous use of the content analysis method. Challenges and strengths of that method are described. In Chapters Two and Three revisions to the existing method are proposed in general, and then specifically for child-and-youth-focused instruments.

Part II: Method Application

Chapter Four applies the revisions proposed in Part I, specifically in the field of childhood cancer measurement. Chapters Five and Six apply these methods in their most refined form to the most popular health and QOL patient-reported (PRO) instruments used in the child health literature in an attempt to make their content explicit and clear to instrument users. The work here is intended to provide a basis for future quantitative work: comparing data from items deemed to have content equivalence according to the content analysis method.

Discussion

Finally, the discussion and conclusions of the thesis summarize the conceptual and methodological issues gleaned from this work. Future applications of applying the ICF to child health measurement will also be discussed.

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Introduction

Background

The work for this thesis began because, like many others, I needed to find the best quality of life (QOL) instrument for my study. The number of instruments that claimed to measure aspects of health status or QOL in the child health literature was overwhelming. Some instruments were clearly more popular than others. As a novice health status/QOL instrument user, I was expecting the popularity of the instruments to be related to their quality. Digging deeper into validation studies from developers of measures such as the PedsQL, and contrasting this information with a rudimentary knowledge of outcome measures, I just became more confused.

It seemed that with the exception of certain key health domains (e.g., emotions and walking), the instruments asked children about very different things. The PedsQL asked children about their *difficulties* with selected activities of daily living while the Child Health Questionnaire (CHQ) asked children about *lying and stealing*. Despite their differences, these instruments were often treated in the literature as if they measured the same things. Both the PedsQL and the CHQ were being used to measure QOL.

I wondered whether this lack of consistency in the health domains that were assessed between instruments was a problem. Health status and QOL instruments provide a particular kind of assessment, one that differs from others. They require children to form their own assessments of themselves, in what is called a patientreported outcome (PRO). According to Norman and Streiner, in health status and QOL instruments, the items define what is actually being measured; these items are called *causal indicators* (1). This means that what one asks children about is what is actually assessed through using these instruments. If one asks about happiness or walking, one would be measuring happiness and walking. Take those items away and one removes the concepts those items represent from the instrument.

Health status and QOL instruments are therefore conceptually defined by the questions that are asked in the questionnaire. This seemed like an extraordinary advantage relative to clinical assessments which could almost never be taken at face value based on the questions. However, the implication of this realization is not so positive. If the instrument is defined by the questions and the questions are very different from instrument to instrument, then the results obtained through the use of these instruments are not comparable.

This problem I encountered was the basis for wanting to explore the content of the child health status and QOL instruments further, but there was in addition another glaring issue: the same instruments were being used to measure different things. For example, the PedsQL could be mentioned in one study as a health status instrument, in the next as an HRQOL instrument, and in the next as a QOL instrument. The concepts that instrument users claimed to be measuring through the use of these PROs were endless. Researchers could not truly be measuring different things through the use of the same instruments. It did seem likely that researchers were applying their own definitions, ideas or even assumptions to decide what they thought was being measured from study to study. It is now apparent that this is a situation where: 1) the same instruments are being used to measure different concepts, and 2) within an apparently similar concept (e.g., QOL) different instruments are assessing different things.

What can be done about these problems arising from poor articulation by developers about what their instruments measure or lack of consensus among instrument users about what concepts these questionnaires represent? First, some exploration is necessary concerning conceptual definitions of health, HRQOL and QOL. Second, it is important to discuss the impact of conceptual inconsistencies on measurement so that solutions to the problem can be sought. Finally, revising and applying methods for dealing with conceptual inconsistencies should be explored.

This thesis is devoted to exploring, and applying methods to deal with, conceptual inconsistencies in child health status and QOL instruments. As mentioned, some discussion about conceptual definitions and the impact of conceptual problems on measurement will set the stage for the detailed methodological work to follow.

Evolving health concepts and measurement

In 1948, the World Health Organization (WHO) defined health as "a state of complete physical, mental, and social well-being and not merely the absence of disease or infirmity"(2). The concept of health is viewed internationally as a resource for well-being and positive living (2,3). The productivity and potential of entire nations is determined at least in part by the health of their children (4). Furthermore, the efforts of parents, communities and medical service providers are judged on the basis of improvements to child health.

The importance of child health as a priority for families, communities and nations is much less debatable than its definition. Although necessary for expanding

horizons about health, the 1948 WHO definition is hardly sufficient as it is. For any definition to be useful, it must be clear, with identifiable boundaries for inclusion and exclusion (5). While the oft-quoted 1948 definition describes that health is much more than not being ill, on its own it fails to offer clarity about what aspects of a person's life should be included in the concept of health and what aspects should not. Such clarity is needed in order to measure, quantify and eventually compare results of measures of health between children, families, communities and nations. A brief historical overview about health is appropriate for understanding how definitions and measures of health have advanced or delayed progress in health measurement.

Since the advent of germ theory, health has been expressed according to absence-of-disease thinking useful in public health contexts. As early as the 1700s, public health proponents were interested in measuring the pattern of outbreaks and, subsequently, the effects of clean water and community hygiene practices (3,6). At that time health indicators such as the incidence and prevalence of diseases like cholera and dysentery were useful because they measured the proportion of individuals with infirmity in the population. In parallel, data expressed as infant mortality rates indicated the success of communities and nations in preserving child health (4). Since the development of antibiotics and vaccination as well as the proliferation of medical services the incidence and prevalence of infectious disease has declined in the developed world (3) and so have infant mortality rates. Health indicators have shifted from a focus on prevalence and incidence to include symptoms for describing children and adults who were living with disabilities or morbidity as a result of extended life in the context of a chronic condition (7). Symptom checklists were developed to measure pain or discomfort from the perspective of both the child and parent (7,8).

While mortality in childhood declined, chronicity rose and the experiences of childhood chronic health conditions or disability became relevant indicators of health status. A measurement approach to child health status was developed based on child and parent report (also known as PROs). PROs are any reports of the status of a patient's condition that come directly from the child or child proxy without interpretation by anyone else (9). The emergence of PROs presented the opportunity to measure more sophisticated aspects of health than just mortality and symptoms, including health status, health-related quality of life (HRQOL), and quality of life (QOL).

These shifts in the manner in which health can be measured have occurred in tandem with ideas about what constitutes child health. Understanding of health,

definitions of health, and measurement of health are linked. A lack of clarity of the definition of health creates confusion about how to decide what it means for a child to be healthy, how child health should be measured, and consequently which children are healthy and which are not. Before scientists, policy-makers, epidemiologists or health services researchers pursue the collection of data using advanced techniques in child health measurement (such as PROs, computer adaptive testing or item banks) explicit consensus-based health definitions should be applied to health instruments. Unfortunately, use of PROs has exploded in health services research without a great deal of consensus about the concepts of health that serve as the basis for measurement(10). It is essential to apply agreed-upon definitions to PROs to advance our knowledge of child health.

Contemporary definitions of health

Today when one says a child is healthy, what does that mean? The WHO has an updated definition of health in the International Classification of Functioning, Disability and Health (ICF) and in particular for children with the children and youth version (ICF-CY). According to the ICF-CY, health is defined according to a person's functioning, which refers to an individual's performance and potential for daily living (11). The word 'health' is typically used in the context of a health condition (i.e., diagnosis) to describe how a disease, disorder or impairment, in combination with the environment, can impact upon and affect what a person does or can do (11). The ICF framework (Figure 1) and specifically the classification system are intended to capture a biopsychosocial perspective of health using components called 'body functions', 'body structures', 'activities' and 'participation', interacting with contextual factors including the 'environment' and 'personal factors'. Personal factors are part of the conceptual framework of the ICF but they are not yet represented in the ICF classification. At this stage quality of life is not part of the ICF framework.

Health conditions are synonymous with diagnosis (as captured with the International Classification of Diseases (ICD) systems) and are not part of the ICF classification system. The *body functions* component classifies aspects of health pertaining to both psychological and physical functioning of the individual; *body structures* classify the physical structures of the body; *activities and participation* classify the things people do with their bodies and minds (e.g., self-care, socializing with friends); and *environment* classifies the 'with whom', 'with what' and 'where' the activities and participations take place (11). When there is a breakdown among the

components of the *body functions/structures* and *activities and participation*, the negative outcome is disability. Thus, using the ICF disability and functioning are negative and positive outcomes along the continuum of health.

Children with health conditions can experience disabilities that relate to their capability and performance at school and at home (12-14). One of the most significant areas of functioning commonly affected by health problems in children are their relationships with their friends and families as well as social interactions in general (15). Difficulties in social functioning in children with health conditions are complex and multidimensional. Attitudes of others (15,16), learned apprehension in relationships (15,17), health condition-related cognitive problems (18) and other developmental issues can all contribute to functioning problems in social situations. Similarly, school performance is a challenge for children with health conditions (13) due to various factors depending on their condition, including but not limited to: difficulties with attention or memory (18); ability to perform activities supporting school performance (e.g., note-taking or managing homework); attitudes of teachers and parents regarding particular learning needs; and the ability to attend school regularly (15).

As described in both social and school milieus, functioning involves the interaction between a child's internal psychological and physical worlds, ability and potential to perform their daily activities, as well as the social, physical and political aspects of their environment (19). The relationship between children's internal world and their environment has been described as the pathway to functioning for a myriad of clinical populations and this idea is now assumed to be common wisdom among rehabilitation researchers (12,14,19,20). This biopsychosocial view is well expressed by the ICF/ICF-CY.

Conceptual definitions of QOL

Many life domains are covered by a biopsychosocial approach to health. The question then remains: what is QOL? It is unclear from the literature exactly what people mean when they use the term QOL. Various authors have written about the meanings implied in the definition of health or QOL (7,10,21,22), what constitutes the 'true' definition of health versus QOL (10), and how this lack of consensus over the term or its conceptual basis has limited its measurement and hence scientific knowledge in the area (23).

As with health, the way QOL is measured should depend on the conceptual basis or definition used. QOL has been defined as a child's perception of his or her well-being (22) or the congruence or discrepancy between what one expects from their life and what they achieve (24,25). The WHO defines QOL as "the individual's perceptions in the context of their culture and value systems, in relation to their personal goals, standards and concerns" (26). What unites these definitions is the focus on the wishes and expectations of children. *The child's perception of health or life* is essential to measurement of QOL in order to represent this construct accurately and effectively.

Health-related QOL captures similar domains of life as health status. When describing a child's HRQOL, it is as important to investigate his or her internal world, daily activities, the social and physical environment as it is when measuring health (22). HRQOL instruments express a focus on QOL in health-based literature although definitions of the concept are rare. Guyatt et al. describe HRQOL as those aspects of life that are important to people with health and chronic conditions (21). According to this definition, in order to measure HRQOL, an instrument must assess those aspects of life that are typically affected by a child's health condition. A criticism of the HRQOL approach is that it is difficult to partition that portion of a child's life that is affected by illness and distinguish what other factors in a child's life influence their self-assessment. This challenge is exacerbated in childhood measures of ongoing, episodic or chronic conditions where the experience of illness plays a developmental role in the child's experience of life.

In contrast to HRQOL, theories of QOL, such as resource theory or discrepancy theory, are more generalizable approaches across groups. Resource theory targets a child's happiness or satisfaction with predetermined theoretical and empirically derived life conditions (27,28). Applying a resource theory approach to measurement involves first determining the aspects of life that are the most valued by many individuals or most emphasized in the scientific literature, and second measuring the presence or extent of those aspects of life. A resource approach has been applied in health economic instruments where developers of an instrument first determine the most important elements of life of according to the perspectives of people in the general population, then obtain people's valuations of the various combinations of health and functional states, and finally determine a child's QOL based on the extent to which those elements are present in that child's life. The ethical implications of such practices will be discussed in later chapters of this thesis. Discrepancy theory highlights the gap or overlap between what one has and what one wants or wishes for (25,28). As mentioned, the WHO supports a theory of QOL that can be defined as: 'an individuals' 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' (26). In the WHOQOL manual there is no description of whether the WHO definition is based on any one particular theory. The portion of the definition that emphasizes that one's perception with life domains is important *relative* to their personal goals is consistent with discrepancy theory.

The challenge with measurement from a discrepancy theory perspective is that the wishes and expectations of children must be incorporated explicitly when evaluating their life domains. Although this approach is often addressed in the questionnaire item development stage, capturing the individual priorities of children in QOL measurement is often lost. Using individualized approaches to outcome measurement, one can place a weighting on a life domain based on the value given to it by each individual child. Good QOL is represented only based on whether the child has experienced what they value, not based on the extent to which they experience life domains that are deemed by others to be important. In this way, individualized outcome measures are useful for determining QOL using a discrepancy theory approach.

In summary, a child's perceptions, expectations, standards and concerns about their health or life domains should be the targets of HRQOL/QOL measurement irrespective of the actual health state. **This distinction is key to deciphering a health status instrument from an HRQOL or QOL instrument**. The emphasis in HRQOL or QOL differs from health in that it describes what is important or significant in the life of children and their families *from their own point of view*. The distinction between health and HRQOL/QOL concerns a difference not solely of content (what one measures) but essentially of perspective (how one measures). This distinction is described further and applied throughout this thesis.

Impact of conceptual ambiguity on child PRO measurement

Psychosocial elements of child's life have only recently begun to be thought of as health issues and studied. As a consequence, child health researchers and clinicians do not yet apply agreed upon methods in order to appraise consistently or compare PRO measures that probe health disability and QOL (10). Hence, research or clinical findings obtained using these instruments are often not comparable. This lack of consistency makes it difficult to relate one study to another, let alone to combine studies into metaanalyses. A scenario is presented to illustrate this issue further.

Imagine two young children with spastic diplegic cerebral palsy GMFCS level II, who have received Botox injections in their lower extremities at the age of seven. One child lives in Canada, the other in China. Following Botox intervention, a researcher wishes to evaluate the impact of the treatment on health. To do so, the following health indicators are chosen: range of motion, speed of walking five metres, improvements in functional walking ability, and changes in QOL. Measurement error issues aside and assuming reliable assessments, using the two indicators range of motion and speed of five metre walk, the children's therapists can easily compare the measures obtained between cases because the indicators 'functional walking' and 'QOL' present a comparability problem on a conceptual level. These indicators require elaboration about their definitions as well as the components of the definitions before they can be compared.

In the illustration above, our limited understanding of the impact of health conditions in childhood on daily life is related to a lack of consensus on what constitutes 'functioning' or 'disability' versus 'health' or 'QOL'. As a result, the conclusions about the effectiveness of Botox or other interventions using such terms cannot easily be compared. Researchers and clinicians who wish to capture the impact of interventions on daily life require a common conceptual foundation on which to base measurement in order to progress systematically or generalize conclusions from research (29,30).

Comparability issues arising from the application of poor conceptual clarity in child health measurement can also be described as a problem with content validity. The match between an instrument's content and its intended purpose, as well as the representativeness of items relative to the concept it represents, constitute its content validity (1). Historically this challenge has been one of the least explored aspects of validity, especially with respect to PROs. Instead, content development has been reported minimally in papers on instrument development in lieu of quantitative methods that highlight other psychometric properties such as reliability and construct and criterion validity. The result is a vast array of instruments that conceivably meet adequate standards of internal consistency, reliability, and construct validity, etc. but are poorly defined conceptually, so that the results they provide are not comparable. Plainly put by Hobart et al, "[w]e do not really know which variables most rating scales

measure"(31). This problem with content validity may lead to a failure to detect change due to state of the art clinical treatments, in essence falsely dismissing promising therapies or vice versa.

There are many reviews of health and QOL measures the features of which are shown in Appendix A. To date, none offer concrete solutions for dealing with conceptual inconsistency between instruments created with unclear definitions.

Using the ICF/ICF-CY to clarify conceptual content of PRO instruments

A potential solution to address problems with inconsistency of concepts in child health measurement is to apply clear definitions to instruments using standard classifications of health. Classification evokes consensus about diverse terms or ideas among researchers and clinicians, between settings and countries (30). In this way, classifications are tools for creating a collective base on which knowledge is created. The ICF-CY was discussed in the section on concepts of health as a framework to unify the definition of health endorsed by the WHO. The ICF-CY is also an internationally developed system of terminology that classifies functioning, disability and health for the purposes of description and comparison created with input from multiple stakeholders and nations.

In PRO measurement, classifications can code items from functioning and QOL measures for children (29,32-34). At present, the ICF-CY is a tool that can code items using more specific definitions than those found in the framework. On its own, the classification is a neutral set of alpha-numeric codes or categories describing health, and the environment in which functioning takes place.

Although some scientists and clinicians use the ICF to classify individuals, with over 3500 categories this process is at best inefficient and at worst potentially unethical (35). The ICF is most efficiently used when describing and comparing data obtained through existing health status and QOL PROs. The appropriate use of classification in this case is to describe, appraise, compare and contrast similarities (redundancies) and gaps (needs) in the measurement of health and QOL in children for the purpose eventually of creating standards for instruments.

One of the primary applications of the ICF/ICF-CY thus far has been to describe measurement content for the purpose of making PRO contents clearer. In the adult literature, appraisals of measurement content using the ICF have been performed for

stroke measures (34), arthritis measures (36,37) and generic health status and QOL measures (35). Those studies have focused first on describing the content of measures using the ICF framework and categories, then appraising their appropriateness for measuring certain concepts based on the content contained therein. Classified instrument content provided systematic information on which to base selection of measures in order that components of functioning can be matched suitably to their intended purpose. The classification of the content of measures has thus made an important contribution to the field of health measurement in adults.

Linking information about functioning, disability and QOL to the ICF

Once the decision is made to classify information using the ICF, the methods by which this is done become increasingly important. Diagnostic classifications, e.g., DSM or ICD-10, have strict rubrics (criteria and instructions) telling users how to arrive at a certain category in the classification. Clinical assessments clearly outline administration procedures for clinicians. These practices are important to ensure the consistency and reliability of the process of how content is extracted or data are obtained. No such method is officially endorsed in the ICF manual.

In 2002 a group of researchers devoted to ICF methodology published an approach to content analysis, and updated the method in 2005 (32). The method was originally intended to standardize the manner in which researchers and clinicians link the content of health-status and related measures to the ICF. The 2005 update expanded the method so that content from clinical assessments and qualitative sources could be included.

The 2005 method carried with it certain limitations. Although the neutral nature of ICF categories captured health domains quite well, the neutral categories imposed a limitation on additional features of an instrument. The presence or absence of health content could be linked but the perspective found in the instrument (health versus QOL) was lost (33). It is not enough to have codes that have similar *definitions* between languages – the codes must be arrived at according to a similar *perspective*. Two instrument items that are represented by the same ICF category might have the same content but do not necessarily extract or express the same kinds of health or health-related information. This issue is discussed and illustrated in more detail in Chapters 2-6.

Items from PROs should be represented according to *perspective* in addition to health domains. This process is necessary to distinguish between health and QOL. For example, if one does not take perspective into account, a questionnaire item phrased "How much difficulty do you have with walking?" would be linked to the same ICF category as "How important is it for you to be able to walk without difficulty?" The problem is that the former item is about walking from the perspective of functioning while the latter item probes walking from the perspective of HRQOL/QOL (i.e., the value of that walking to the child responding). This distinction is not captured under any known published method. Comparing data scaled from the former walking item with the latter item as if they were equivalent would be incorrect because a child could rate his or her walking as highly impaired while simultaneously placing little or no importance on the activity of walking.

Cieza et al. (33) identified the issue of capturing perspective as a problem in the last published version of the linking rules, yet to our knowledge no critical comments or solutions have been found in the literature. The Cieza et al. (2005) linking method has been shown to be useful for making the health content of PROs explicit (38). However, it remains unclear how to parse out whether a PRO is a health status or QOL instrument.

Future revisions to the linking procedure should first determine the uses and problem areas for those who link content to the ICF. These methods should also determine if any alternatives to this methodology are used, as well as their advantages and disadvantages.

Another issue pertaining to the application of the method of Cieza et al. (2005) is that it was developed on adult content but is used to link childhood content, despite not yet having been validated for that purpose. Linking childhood content to the ICF poses special challenges. Content pertaining to development and to proxy parent versus childhood reports is not captured using the earlier linking method. Thus the method should be expanded and tested for children before it can be rigorously applied to PROs intended for children.

This brief report on literature linking health content to the ICF has focused on its application to measurement content. It should be noted that as more methods related to classification develop more specific uses for classification will emerge. These new methods will be based on different sources of data collected for different purposes. In measurement, classification could in theory be used to verify cross-cultural content of translated questionnaires. In program evaluation, classification can show what areas are

addressed between programs as well as what areas are priorities for patients versus clinicians (for example, by contrasting program data with qualitative information from patients). In epidemiological studies, classification can be used as the content on which to extract or measure data from different sources in different countries and languages (39). For example, if population surveys are properly classified, their findings can be compared between countries. Despite the promising uses and applications of the ICF, many methodological stones remain unturned. These applications and uses for the ICF have been proposed but are as yet under-developed in the literature.

Summary

In summary, health research involving children has begun to place importance on both functioning and QOL in order to capture the impact of health conditions on daily life. Building upon empirical research in health and QOL has been difficult due to a lack of understanding or equivalence of these concepts. Classification using the ICF can be useful to describe, appraise, compare and contrast the content of various sources of data pertaining to functioning and QOL in a standardized manner. Although these applications of the ICF are often stated as useful, methods to achieve such goals are under-developed. The methods that do exist cannot distinguish between content concerning health status versus QOL and have not been validated for children.

Despite the stated limitations, the ICF/ICF-CY framework can provide a rigorous standard for a broad conception of child health. In tandem, the ICF/ICF-CY classification has shown tremendous potential for expanding biopsychosocial thinking in the measurement process. This thesis will build upon the Cieza et al. (2005) linking rules to address the conceptual inconsistency problems found among health status and QOL instruments for children.

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Article Name	Year / Authors	Review Method	Findings
Quality of life instruments: a review of the impact of the conceptual framework on outcomes. (40)	2006 Davis, Waters, Mackinnon, et al.	A systematic review was conducted to identify QOL instruments for children aged 0 to 12 years Both generic and condition-specific measures were reviewed. Medline and PsychLit electronic databases to identify papers published between 1990 and 2004, using the terms 'children' and 'quality of life' (encompassing health-related quality of life) or 'QOL' or 'HRQOL' as keywords. 332 abstracts were identified. The abstracts were reviewed to identify QOL/HRQOL instruments. The title of each instrument was then entered into Medline and PsychLit to obtain further articles about its construction and psychometric properties. Instruments that were used to measure QOL or HRQOL but were actually designed to measure health status or functioning were excluded	Fourteen generic and 25 condition-specific QOL instruments were identified. Eleven types of definition of QOL and health-related QOL and three theories of QOL were identified. QOL was measured by a variety of domains including emotional, social and physical health, and well-being. Items commonly assessed difficulties, or intensity/frequency of feelings/symptoms, in contrast to positive aspects of life and happiness. Lack of empirical evidence for many of the fundamental assumptions of QOL. Three Theories: 1) QOL model developed by Lindstrom with four spheres of human existence: global (ecological, societal, and political resources), external (social and economic resources), inter-personal (social relationships and supports), and personal (physical, mental, and spiritual aspects of the individual). 2) discrepancy theory

Appendix A: Child Quality of Life Reviews Completed to Date

Evaluating Health- Related Quality of Life Studies in Paediatric Populations: Some Conceptual, Methodological and Developmental Considerations and Recent Applications (41)	2005 De Civita, Regier- Evaluat, Alamgir, et al.	(such as the Child Health Questionnaire, the Child Health and Illness Profile and the Warwick Child Health and Morbidity Profile, as well as those designed to assess caregiver difficulties. Health status, functioning, and caregiver difficulties are theoretically different from QOL and the conceptual background of these instruments is expected to be different from QOL instruments. No systematic review Instruments selected by authors	3) utility theory Conceptual issues of mismatch a problem with no consensus. Few researchers combat response shift
Health-related quality-of-life assessment in children and adolescents:	2006 Ravens-Sieberer, Erhart , Wille, et al.	Literature search Medline & PsychInfo from 1980 to 2006 German Institute for Medical Documentation and Information	There are measures available to represent the scope of constructs to represent QOL Intercultural assessment is possible Self-reporting from children is important

methodological considerations.(42)		In a literature search of MEDLINE andPsycINFO incorporating over 5358 and 1647 articles on HR-QOL from 1980 to 21 February 2006 totals of 584 and 213 articles relating to children and adolescents, respectively, were found	
The measurement of health-related quality of life (QOL) in pediatric clinical trials: a systematic review. (43)	2004 Clarke & Eiser	Included paediatric clinical trials published in English between 1994 and 2003 involving children and adolescents up to the age of 20 years, and use of a standardised QOL measure. MEDLINE, CINAHL, EMB Reviews, AMED, BNI, PSYCHINFO, the Cochrane Library, internet, and reference lists from review articles were included. 917 initial screened with 18 articles included	 14 measures were identified but only two met their criteria for quality: 1) Pediatric Cancer Quality of life Varni, et al (1999) 2) Persistent Otitis Media TNO-AZL Quality of life questionnaires Verrips, et al (1999) No mention of conceptual grounds or content of the measures
A review of measures of quality of life for children with chronic illness(44)	2001 Eiser & Morse	1980 to July 1999 Medline, BIDS ISI Science and Social Science Citation Index, PsychLit, Cochrane Controlled Trials Register, metaRegister of Contriolled Trials 255 abstracts identified and sent to	19 generic and 24 disease specific measures found 30 described by authors as QOL, 8 as health status, 2 as functional status, 1 perception of illness, 1 life satisfaction, 1 quality of life 9 measures with both child and parent
		ref manager for screening 137 articles screened	versions, 2 for parents only, 12 for children only Current measures are limited in their scope relative to a proper definition of QOL
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Children's quality of life assessments: a review of generic and health related quality of life measures completed by children and adolescents(45).	2001 Harding L.	PsychLit computer search was made for the years 1990–98 Generic and generic health-related QOL measures for children and/or adolescents were included	Environmental aspects of QOL and qualitative aspects are often missing Within the medical context QOL has been equated with functional status with emphasis on the individual's level of functioning and capacity to fulfil basic self care tasks There is a great diversity in the measures discussed, which probably reflects the theoretical basis of each of the measures, as well as the importance of the qualitative dimension as assessed by the authors.
Assessment of health-related quality of life in children: a review of conceptual, methodological, and regulatory issues(46)	2004 Matza, Swensen, Flood, et al.	Measures were selected and there was no systematic review	 HRQL should be assessed from the patient's perspective. HRQL should incorporate a broad range of concepts Acknowledging context and environment is vital to childhood HRQL assessment

Llaalth Dalatad	2009	Pastristed to OOL and not UPOL	20 generic and 64 disease specific
Health-Related Quality of Life Measurement in Children and Adolescents: A Systematic Review of Generic and Disease-Specific Instruments(47)	Solans, Pane,	Used previous reviews to identify	instruments were identified, 51 of which were published between 2001 and 2005.
	Estrada, Sutton, Berra, Herdman, Alonso, Rajmil,	systematic search from 2001 to 2006 MEDLINE, ISI Science, Citation Index, HealthSTAR and PsycLit Hand-searched references from eligible articles, congress abstract books, and the gray literature, as well as contacting experts working in the	Many generic measures cover a core set of basic concepts related to physical, mental and social health, although the number and name of dimensions varies substantially 26% of the disease-specific questionnaires Were exclusively addressed to proxy- respondents
		field and consulting virtual libraries of PRO instruments (ProQolid and Bibliopro)	
		Documents included for further analysis were those reporting the development, psychometric assessment and/or use of instruments measuring QOL, health status or well- being and intended specifically for children and adolescents up to the age of 19 years.	
		Articles or other documents reporting the use of functional scales and symptom checklists, the results of clinical applications or population studies using HRQOL instruments, and	

		•	
		articles reporting on the cultural adaptation of instruments were also excluded from further analysis.	
		They extracted the reliability, validity and sensitivity to change of the measures. No report of the amount of articles screened was given	
Generic Health- related Quality of Life Instruments in Children and Adolescents: A Qualitative Analysis of Content(48)	2004 Rajmil, Herdman, Fernandez De Sanmame, Detmar, Bruil, Ravens- Sieberer, Bullinger, Simeoni, & Auquier	No systematic review Ten questionnaires were analyzed A descriptive and explanatory qualitative approach was used to analyze the content of the measures Questionnaires were segmented and their content assigned to one of three domains: physical, psychological, or social, on the basis of the World Health Organization concept of health Once dimensions had been assigned to one of the Three major domains, the second stage of the analysis involved reviewing all items in each domain. In this stage of the analysis, categories were allowed to	A reasonably coherent notion of HRQOL underlies instruments available for children and adolescents. HRQOL measurement in young people is still in its developmental step. All of the 10 instruments included items referring to physical, psychological, and social aspects of health.
		"emerge" from the data on the basis	

of careful coding, classification, and constant comparison.	
As the last stage in the analysis, an explanatory framework was developed to integrate the facts emerging from the data	

Chapter 1

Systematic Review of methods for linking health and health-related information to the ICF from 2001-2008

Fayed N, Cieza A, & Bickenbach J.

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The contributions of each author to the manuscript are as follows:

NF conducted the systematic review, wrote the syntax, was one of the abstract screeners, extracted the data from the included articles and was the primary analyst of the qualitative portion of the study. Fayed wrote the manuscript and conceived of the structure of the manuscript (85% contribution).

AC provided scientific feedback on the rationale for the study, supervised the systematic review and functioned as a co-analyst of the qualitative data. AC suggested the presentation of the quantitative portion of the analysis (10%).

JEB provided specialized information on worldwide disability issues as well as the scientific clarification and regarding structure and ontology of the ICF. JB edited the entire paper and provided feedback on the scientific information contained in the introduction and discussion.

Abstract

Introduction

In 1976, the World Health Organization (WHO) estimated worldwide disability prevalence at 10%; recent evidence suggests the prevalence is even higher. Given the extent of disability around the world, it is essential for researchers and policy makers to have a uniform language for describing and discussing disability. The International Classification of Functioning, Disability and Health (ICF) is WHO's attempt to provide that standard language. Linking rules were published in 2002 suggesting a method for standardizing the *process* of connecting outcome measures to the ICF classification. The objective of this paper is to study the extent to which the linking rules published in 2002 and 2005 have been used by researchers to link health and health-related information to the ICF and collect the feedback about the current practices, applications and areas to improve the linking method.

Method

Using a systematic review of health-based literature between 2001 and February 2008 we (1) determined research areas where the linking method is applied (2) examined the characteristics of studies that linked information to the ICF; and (3) described current practices and issues related to the process of linking health and health-related information to the ICF both quantitatively and qualitatively.

Results

The systematic review yielded 109 articles from 58 journals that linked health information to the ICF and 58 of the articles employed published linking rules. The majority of articles was descriptive in nature, used linking for connecting content of health instruments to the ICF and linked English health content. Quality controls such as reliability checks, multiple raters and iterative linking processes were found frequently among users of the linking rules.

Qualitative analysis created themes about: preparing units of information, who links to the ICF, reliability, matching or translating concepts from text to ICF categories, information unable or difficult to capture, quantitative reporting standards and overall linking process.

Discussion

This review also shows that the linking process is a useful way to apply the ICF classification in research. With over 100 articles published in 58 peer-reviewed journals across 50 focus areas, linking health and health-related information to the ICF has been shown to be a useful tool for describing, comparing and contrasting information from

outcome measures used to collect quantitative data, qualitative research results, and clinical patient reports across diagnoses, settings, languages and countries.

Introduction

In 1976, the World Health Organization (WHO) estimated worldwide disability prevalence at 10%; recent evidence suggests the prevalence is even higher¹. Given the extent of disability around the world, it is essential for researchers and policy makers to have a uniform language for describing and discussing disability. The International Classification of Functioning, Disability and Health (ICF)² is WHO's attempt to provide that standard language. In the ICF, disability is a universal human experience representing the complex interaction between a person's health condition and contextual factors: personal factors and environmental factors, resulting in functioning at various levels: body structures and functions, activities and participation². To further understand the impact of disability on individual lives and society as a whole, scientists and health researchers have begun large-scale projects to collect data using this standard language and bio-psycho-social approach. The Measuring Health and Disability in Europe (MHADIE) group provided such an example by collecting data based on the ICF uniform terminology for the purpose of supporting evidence-based policy development^{3,4}.

Another approach to standardizing health data is to study already existing frameworks and sources of information and analyze their similarities and differences in the health domains. In the USA, the Patient Reported Outcomes Measurement Information Systems (PROMIS) gathered items from patient-reported outcomes in the last decade, grouped the concepts that underlie these items into a domain framework and used this framework as the basis for standardized health and disability assessment⁵.

Linking rules were published in 2002 that suggested a method for standardizing the *process* of connecting outcome measures to the ICF classification⁶. Originally in 2002 use of the linking rules helped researchers select outcomes that overlapped with the health domains they wished to measure, thus facilitating content validity. These rules were updated in 2005 to extend their use to link technical and clinical assessments, diagnostic instruments, and health interventions to the ICF⁷. To our knowledge, there has not been any systematic appraisal published about the extent to which these linking rules or alternative processes of ICF linking have been empirically applied.

The objective of this paper is to study the extent to which the linking rules published in 2002 and 2005 have been used by researchers to link health and healthrelated information to the ICF and collect the feedback about the current practices, applications and areas to improve the linking method. Using a systematic review, we sought to (1) determine research areas where the linking method is applied; (2) examine the characteristics of studies that linked information to the ICF; and (3) describe current practices and issues related to the process of linking health and health-related information to the ICF both quantitatively and qualitatively in order to generate feedback to potentially improve subsequent linking rules.

Methods

Study design

A systematic literature review was performed to identify publications that have linked health and health-related information to the ICF. The procedures followed three steps: electronic literature search, publication selection and data extraction. Abstract and full-text article screenings were documented and managed electronically using two independent reviewers and a Microsoft Access database.

Electronic literature search

The electronic searches were conducted in the literature databases MEDLINE, Cumulative Index to Nursing and Allied Health Literature (CINAHL), EMBASE and PsycINFO. Standardized search terms were used. The search strategies varied by database as the specific thesaurus vocabulary of the given database was used. The search strategy used for the MEDLINE database is included in appendix A. We limited the searches to articles published between January 1st 2001 and the 1st week July 2008 in the English language. Study design restrictions were not imposed.

Publication selection

The selection of publications was performed according to a step-wise process. In the first step, literature mentioning relevant search terms was identified. The abstract was then screened for research involving the ICF. Only publications with abstracts, keywords or titles explicitly mentioning the ICF were included, and their full text was retrieved.

Finally, only publications linking text-based health and health-related information to the ICF at the first level of classification or higher were included in the study. Studies that merely grouped information according to the component levels of *body functions and structures, activities, participation, environmental factors* or *personal factors* were excluded. If the information was linked to first level of the ICF (chapter level) for example, activity and participation chapters such as d1 Learning and Applying *Knowledge, d2 General tasks and Demands, d3 Communication, d4 Mobility, d5 Selfcare, d6 Domestic Life, d7 Interpersonal Interactions and Relationships, d8 Major life areas* or d9 Community Social and Civic Life, or more specifically, it would be included in the analysis. Furthermore we disregarded information from studies pertaining to qualifiers because there is typically insufficient information in text-based sources to apply the qualifiers ratings; this is in contrast to the rating of an individual patient's functioning with an ICF code. Two reviewers (NF & AG) performed these steps independently. The reviewers later met to compare their results and resolve inconsistencies. If the reviewers disagreed about the inclusion or exclusion of a publication, they reviewed the inclusion and exclusion criteria. In the event they could not reach consensus, a third reviewer (AC) was consulted in order to settle disagreements.

Data extraction

Research areas where the linking method is applied

The number and names of journals publishing articles that linked health and health-related information to the ICF was extracted from the included articles. The primary focus of the article (i.e., setting or diagnosis) was also extracted.

Article Characteristics

The following information was collected to describe article characteristics: study type, (experimental, analytical or descriptive), article type (e.g., cohort, systematic review, qualitative study), type of text-based information linked to the ICF (e.g., patient records, outcome measures or qualitative interviews), language of the text linked, and the lifespan focus of the article (child, adult or both).

Practices and issues related to ICF linking - quantitative

We extracted the frequency with which quality control methods were employed for the ICF-linked data. The indicators of quality control included: reliability checks, multiple raters, and iteration or stepwise processes. The level of specificity of the ICF classification, i.e., first, second, third or fourth level of classification, was extracted from the articles. Any other quality controls used were classified under 'other'.

Practices and issues related to ICF linking - qualitative

Since we were seeking to *explore* all types of practices and issues related to linking information to the ICF, an inductive content analysis was employed⁸. Information pertaining to the linking process was extracted from the text of each article and analyzed. Each time an issue about linking to the ICF was mentioned, the issue was given a label. Occasionally, one statement generated more than one label if it included information about two ICF linking issues. Each subsequent statement was labeled with an existing label if its content corresponded to previous statements. A new label was created if the content of the statement represented a new topic. The labeling scheme was discussed periodically throughout the labeling process by the first and last authors of the study. This process continued until all written statements pertaining to ICF linking

were labeled from the included articles. The text labels gave rise to sub-themes that were further categorized into major themes.

Results

Electronic literature search

The electronic searches of the three databases combined yielded 2281 citations not including duplicates, 576 of which were screened in full-text form. From those articles, 109 linked health and health-related information to the ICF. Fifty-eight of the articles used the linking rules from 2002 or 2005 and 51 did not. A complete list of the included articles is available in electronic supplement E1.

Research areas where the linking method is applied

Fifty-eight different journals published studies that link information to the ICF in as many as 50 different settings or clinical populations. The details of the journals and settings covered are contained in electronic supplements E2 and E3.

ICF article characteristics

Characteristics of the studies that met inclusion criteria are listed in table 13.

Practices and issues related to ICF linking - quantitative

Table 2 shows the incidence of quality control for each ICF linking method: 2002 linking rules, 2005 linking rules or no linking rules.

Practices and issues related to ICF linking - qualitative

A summary of themes and sub-themes, with example quotes, can be found in table 3. The themes are described in the following sections in further detail.

Preparing units of information

Authors of the included studies described how they partitioned text for linking for the purpose of an inductive analysis (i.e., based on the concepts emerging from or dictated by the text) *or* they selected <u>a priori</u> what was needed to prepare the text for analysis based on a pre-determined scheme, theory or area of interest.

The inductive method for preparing text was achieved in various ways. Some studies partitioned text each time a 'shift in meaning' was detected⁹⁻¹¹. This form of partitioning involves creating a possibility for linking to the ICF each time a new concept was found in the text. For example, in a study by Stamm et al.¹²:

"[a] concept was defined as one separate meaningful entity, such as a body structure, a body function, an activity or a contextual factor. An example [was] the following item of the SACRAH questionnaire: 'Please, assess the pain you had during the last 48 hours caused by your finger joint conditions. How severe was your pain during regular daily work?'. In this item, the following two concepts were identified: 'pain in finger joints' and 'regular daily work'," (p.3).

One research study added another level of interpretation to the unit of information prior to linking the unit to the ICF¹³. Drummond et al.¹³, for example, created a column next to each item in the Disabilities of Arm, Shoulder, and Hand (DASH) questionnaire to describe what raters interpreted each item to be about before it was linked, for the purpose of clarifying the focus of the item between raters prior to translating it to the ICF.

Using an opposite (predetermined scheme) approach, text was prepared by deciding *a priori* the content that was of interest. For example, in one study the authors decided that they would only link the information from qualitative interviews about "the participation domains and environmental factors of the ICF"¹⁵, (p 247).

The only exception to the inductive versus the predetermined scheme dichotomy of data preparation was found in a study of computerized linking to the ICF. In those studies, the authors used a computerized term/semantic alignment system¹⁴. Here a computer program was used to match terms coming from large volumes of text and terms coming from the ICF together instead of using trained humans to find the relationships between the two sources. Text that did not align with ICF terms was eventually matched by a person¹⁴.

Raters: Who links to the ICF?

Two types of information about the raters or linkers who translated text to the ICF were described. Studies described these raters/linkers in terms of their content expertise¹⁶ (e.g., 10 years of clinical practice in stroke rehabilitation), expertise with the ICF itself^{10, 17} (completed an ICF coding training course), as well as professional or research backgrounds^{10, 18} (e.g., physiotherapists vs psychologist). The following quote demonstrates awareness of the reporting about raters or linkers to the process of ICF linking:

"It should be studied whether raters trained similarly in the ICF but coming from different professional backgrounds apply the linkage rules differently"⁹, (p.1286).

Reporting information about raters/linkers in terms of their detailed conceptual background, point of view, or conceptual lens of interpretation was not found.

Reliability

Two sub-themes arose from the statements in the articles pertaining to reliability: the type of statistic used to represent reliability in ICF-linked data and threats to reliability. Agreement statistics that were used in the studies included percentage agreement, kappa statistics, and kappa statistics with bootstrapped confidence intervals. All agreement statistics were performed <u>between</u> raters/linkers. Agreement statistics calculated using repeated measurement such as might typically be found in an intra-rater reliability study were not reported.

Threats to reliability between raters/linkers were related to unclear understanding of the text or a lack of agreement of interpretation between linkers/raters regarding what ICF category best represented the idea in the text. The following quote illustrates how differences in interpretation between users linking content to the ICF can arise:

"The interpretation of [patient care tools] by nurses and others who used the ICF or ICF-derived tools could always be resolved, but led to more discussion. Whereas the content of the [patient care tools] was always clear from the original project materials, the strength of a comment or the evaluation attached to the comment were sometimes open to interpretation."¹⁶

Another threat to reliability was a lack of consensus among linkers as to how much the contextual information in the text should be taken into account during the linking. For example, Soburg et al.¹⁹ described uncertainty about the extent to which various linkers interpreted the meaning of text resulting in divergent ICF categories for the same text: "explanation for disagreements in coding might be related to the fact that words get their meaning from the context in which they are used" and the context was not represented in the responses to the open-ended questions used as linking material in that study.

Matching or translating concepts from text to ICF categories

The ICF captured most of the concepts in functioning, disability, health and even quality of life in the studies that reported coverage. Coverage of the ICF to the text it represents can be indirectly assessed by the percentage of not-covered categories linked. For example, the study performed by Geyh et al.²⁰ about the content of outcome measures used in stroke reported:

"...a total of 11 283 concepts were extracted from the outcome measures. 10 299 or 91% of concepts could be linked to the ICF, 698 or 6% of concepts were considered not to be sufficiently specified for an assignment to the ICF ("not

definable option"), and 286 or 3% of concepts were considered to be not covered by the ICF.", (p58).

Thus we know from the quote that the ICF was able to represent 91% of the concepts found in the outcome measures.

Another issue pertaining to matching concepts from text to the ICF is equivalency. Nonequivalent matches of text to ICF categories were reported as more information in the text than the ICF category that best represented it:

"Several project groups commented on the lack of detail in 'emotional functions' (b152), where more detail was needed to differentiate between different types of emotions, such as happiness, anger and fear."¹⁶, (pg. 438).

At other times there is more detail in the text than in the text category. This problem gives rise to difficulties assigning ICF codes about <u>general</u> functioning, disability and health concepts because they do not link to <u>specific</u> ICF categories.

Information unable or difficult to capture

Certain information was reported to be difficult to capture using the ICF. This information included problems with matching information about general concepts not part of functioning disability or health according to ICF definitions. For example in one study it was reported that "having let people down"²¹, was not linked to the ICF. Information focused on a client's or a patient's point of view such as life-satisfaction or prioritizing goals²¹, was also not linked to the ICF. vanAchterberg et al.¹⁶ also observed this problem:

"the developers in our projects identified insufficient terms and codes to cover patients' sensations, experiences and coping styles.", pg 438

Difficulties capturing activities and participation for ICF linked content was an issue reported in some of the studies. The following quote demonstrates how the issue was dealt with in one study:

"In order to differentiate between activity and participation, it was examined for each concept linked to component (d) (activity and participation component) whether the linked ICF category refers to a task (capacity–activity) or to lifeinvolvement (performance–participation) according to the ICF model. Capacity refers to an individual's ability to execute a task or an action in a 'standardized environment' and performance describes what an individual does in his or her current environment", ¹² (p. 15). Few studies provided the definitions on which they made the distinction between *activities or participation*, reporting instead why they chose to make the distinction:

"In collecting the data with regard to actual use of the ICF and its components, we chose to differentiate between activities and participation... We chose to use differentiation because of its potential relevance to patient care."¹⁶ (p. 437)

Another issue pertaining to information not captured and ICF linking was the relationships between concepts once they were translated into ICF categories. Once linked, the conceptual tie between two or more categories was lost^{14, 22, 23}. For example, in the phrase: 'I am absent from work d850 as a result of my parenting demands d660', the conceptual tie between the categories d850 and d660 can be lost once the phrase is linked. A qualitative study dealt with the problem by creating a matrix in which they preserved the link between activities and participation as they related to the environment:

"This process [of coding] took a number of steps and a coding scheme began to appear where relations between codes were determined as being 'the cause of', 'related to' or 'part of' etc."¹⁴, (p. 247)

Quantitative Reporting Standards

Four techniques for reporting ICF linked findings quantitatively were found, not including reliability, which was so frequently discussed, it generated its own separate theme. These techniques were reports of: 1) what was linked (e.g., 10% of statements from a qualitative interview were randomly selected), 2) frequency and specificity of ICF categories (first to fifth level categories), 3) bandwidth (the extent to which the ICF was covered by the text or vice versa). The following text contains all three of these listed reporting standards:

"...more than one-third of the three-digit codes describing environmental factors were selected to capture circumstances relevant to patients' functioning. Finally, nearly 30% of the three-digit codes in body structures were used, mainly to describe the etiology of functional problems in nursing diagnoses. Body structures was the only component where a substantial number of four digit codes were used 75%. For all other components of the Classification, less than 15% of these more specific codes were use." ¹⁶ (pg. 438).

The final reporting standard technique found was content density (the ratio of ICF categories to a piece of text:

"content density means the average number of concepts per item. A value of one indicates that one concept was contained in each item. A value exceeding one indicates that more than one concept is identified in some items." ¹² (p. 3)

Overall linking process

The overall process by which text was analyzed and linked to the ICF by individuals was reported in the included studies as a stepwise/iterative process or a one-step cooperative/consensus or majority rule process. A typical example of the iterative process involved raters linking to the ICF independently, comparing their results, discussing the areas of disagreement and consulting a third party if necessary to come to a final agreement²⁰. The cooperative or one-step process involved consensus panels or group discussions to decide on ICF categories at one time point²⁴⁻²⁶. Comments about the helpfulness or disadvantages of the iterative process versus linking in a group at one time were not discussed in the studies.

Discussion

The results of this study show that the ICF has had an impact in the peerreviewed literature of functioning, disability and health over the last decade. More than 550 articles addressing issues related to the ICF were identified in the databases reviewed. This review also shows that the linking process is a useful way to apply the ICF classification in research. With over 100 articles published in 58 peer-reviewed journals across 50 focus areas, linking health and health-related information to the ICF has been shown to be a useful tool for describing, comparing and contrasting information from outcome measures used to collect quantitative data, qualitative research results, and clinical patient reports across diagnoses, settings, languages and countries.

Information from 10 languages was found that linked to the ICF, indicating its potential for universal use. Also, the characteristics of the articles included in this review were very broad, showing the wide range of use of the linking process. However, it is important to mention that the study designs of the identified literature were mainly observational and not experimental. The number of experimental studies performed in the field of rehabilitation sciences remains more scarce than in areas such as biomedical research²⁷. These study types are likely reflective of the disciplines in which the ICF is currently being used as opposed to the possible uses of the linking rules.

Quality controls are frequently used in the identified literature linking health and health related information to the ICF. Users of linking rules used reliability checks, linked to specific ICF categories, employed multiple raters and reported the details of their linking with greater incidence frequently relative to non-users of linking rules. Application of the linking rules for researchers who are not expert ICF linkers is therefore recommended as a guide to support a consistent process with built-in quality controls. Expert linkers with needs beyond the current linking rules should report their processes and procedures in greater detail to improve the transparency of their methods and results and contribute refinements in linking methods to a community of ICF users. Reliability was an issue that was emphasized in the ICF linking literature especially with regard to the use of agreement statistics between raters/linkers. The current authors' experience with linking, and the results of this study, indicate that although agreement statistics are valuable indicators of linking consistency, the quality of linking cannot be interpreted solely with statistics. An iterative process that integrates potentially competing views from various linkers with different perspectives seems to strengthen the final product of linking. However, this hypothesis requires further testing and can serve as the basis for further method-based research in the area.

Our analysis of the studies included provided an understanding of the practices and challenges related to linking text based information to the ICF. The qualitative analysis showed that preparing units of information for linking was important to the rigour of the linking process and mirrored the issues of data preparation in qualitative methodology. Users of the ICF for text-based information must therefore decide whether their analysis will be inductive or based on a previous theory *prior* to linking. For example, will one approach the text inductively or according to a predetermined scheme? Is the text to be interpreted almost literally or through a certain lens or point of view? How much contextual information should be considered?

The themes of *matching concepts from the text to ICF categories and information unable or difficult to capture* underscore the challenges of linking accurately. Although many authors reported that there were some concepts that were difficult to capture, they rarely mentioned these concepts specifically. Publishing data that can't be linked to the ICF has theoretical implications for the breadth and limits of the classification. If for example, many of the concepts that cannot be linked represent personal factors, they could be used as possible content for classifying this component. Regardless of what content cannot be classified, presenting these concepts transparently in the peer-reviewed literature creates an empirical basis for further discussion about concepts that could or should fit within future revisions of the ICF.

The relationships between concepts from information linked to the ICF were also lost in the linking process and should be addressed in future revisions of the linking rules. There are recommendations in previous versions of linking rules^{6, 7} for dealing with some of these issues, however it has become clear that future revisions of linking guidelines will be necessary to provide illustrations for preparing data, preserving relationships among ICF linked information and to deal with concepts that are not easily translated to the ICF.

The findings of this study are limited to the English publication literature and literature published in health-based databases. The most dominant source of included articles came from the research program of the authors or their affiliates which might create bias in the analysis. As uses of the ICF spread beyond health milieus, future reviews on this topic might benefit from scanning multi-lingual literature and the literature in education and the social sciences. Finally, researchers not associated with the authors are encouraged to test the linking rules to increase the objectivity of the process.

Conclusion

This study provides an overview of the use of the linking rules published in 2002 and 2005, as well as alternative methods found in peer-reviewed literature of functioning, disability and health. It also sheds light on the kinds of issues users of the ICF will face when linking text-based information including data preparation issues, the procedures or steps involved in linking, as well as issues of relationships and data that are difficult to capture. The information obtained from this study provides hints regarding application of the linking rules in addition to the purposes for which they were originally developed. Regardless of the method chosen, this review points out the need for researchers to be transparent about the process of linking so that users of the ICF can move towards rigorous consensus based methods of *how* to link.

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	models	
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Lomax,-Claire-L; Brown,-Richard-G; Howard,-Robert-J	Measuring disability in patients with neurodegenerative disease using the 'Yesterday Interview'.	International Journal of Geriatric Psychiatry. Vol 19(11) Nov 2004, 1058- 1064.
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Morita-E; Weigl-M; Schuh-A; Stucki-G	Identification of relevant ICF categories for indication, intervention planning and evaluation of health resort programs: a Delphi exercise.	International Journal of Biometeorology. 2006 Jan; 50(3): 183- 91
Mudge, S.; Stott, N. S.	Outcome measures to assess walking ability following stroke: a systematic review of the literature	Physiotherapy. 2007; 93(3): 189
Mullis-R; Barber-J; Lewis-M; Hay-E	ICF core sets for low back pain: Do they include what matters to patients?.	Journal of Rehabilitation Medicine. 2007; 39(5): 353-357
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Stamm-TA; Bauernfeind-B; Coenen-M; Feierl-E; Mathis-M; Stucki-G; Smolen-JS; Machold-KP; Aringer-M	Concepts important to persons with systemic lupus erythematosus and their coverage by standard measures of disease activity and health status.	Arthritis Care and Research. 2007; 57(7): 1287-1295
Stamm-TA; Cieza-A; Coenen-M; Machold-KP; Nell-VPK; Smolen-JS; Stucki-G	Validating the International Classification of Functioning, Disability and Health Comprehensive Core Set for Rheumatoid Arthritis from the patient perspective: A qualitative study.	Arthritis Care and Research. 2005; 53(3): 431-439
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Stamm-TA; Nell-V; Mathis-M; Coenen- M; Aletaha-D; Cieza-A; Stucki-G; Taylor- W; Smolen-JS; Machold-KP	Concepts important to patients with psoriatic arthritis are not adequately covered by standard measures of functioning.	Arthritis Care Research. 2007; 57(3): 487-494
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Partinen-M	obstructive sleep apnea.	
Stucki-A; Stucki-G; Cieza-A; Schuurmans-MM; Kostanjsek-N; Ruof-J	Content comparison of health-related quality of life instruments for COPD.	Respiratory Medicine. 2007; 101(6): 1113-1122
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vanAchterberg-T; Frederiks-C; Thien-N; Coenen-C; Persoon-A	Using ICIDH-2 in the classification of nursing diagnoses: results from two pilot studies.	Journal Advanced Nursing. 2002 Jan; 37(2): 135-44
Van-Baar-ME; Essink-Bot-ML; Oen- IMMH; Dokter-J; Boxma-H; Van-Beeck- EF	Functional outcome after burns: A review.	Burns. 2006; 32(1): 1-9
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Weigl-M; Cieza-A; Harder-M; Geyh-S; Amann-E; Kostanjsek-N; Stucki-G	Linking osteoarthritis-specific health- status measures to the International Classification of Functioning, Disability, and Health (ICF).	Osteoarthritis Cartilage. 2003 Jul; 11(7): 519-23
Weigl-M; Cieza-A; Kostanjsek-N; Kirschneck-M; Stucki-G	The ICF comprehensively covers the spectrum of health problems encountered by health professionals in patients with musculoskeletal conditions.	Rheumatology (UK). Rheumatology 2006; 45(10): 1247-1254
Wessels, R. D.; de Witte, L. P.; Jedeloo, S.; van den Heuvel, W. P. M.; van den Heuvel, W. J. A.	Effectiveness of provision of outdoor mobility services and devices in the Netherlands	Clinical Rehabilitation. 2004; 18(4): 371
Wolff-B; Cieza-A; Parentin-A; Rauch-A; Sigl-T; Brockow-T; Stucki-A	Identifying the concepts contained in outcome measures of clinical trials on four internal disorders using the International Classification of Functioning, Disability and Health as a reference.	Journal of Rehabilitation Medicine. 2004 Jul; (44 Suppl): 37-42
Xie-F; Lo-N-N; Lee-H-P; Cieza-A; Li-S-C	Validation of the comprehensive ICF core set for osteoarthritis (OA) in patients with knee OA: A Singaporean perspective.	Journal of Rheumatology. 2007; 34(11): 2301-2307

Xie-F; Thumboo-J; Fong-K-Y; Lo-N-N; Yeo-S-J; Yang-K-Y; Li-S-C	Are they relevant? A critical evaluation of the international classification of functioning, disability, and health core sets for osteoarthritis from the perspective of patients with knee osteoarthritis in Singapore.	Annals of the Rheumatic Diseases. 2006; 65(8): 1067-1073
Zochling-J; Bonjean-M; Grill-E; Scheuringer-M; Stucki-G; Braun-J	Systematic review of measures and their concepts used in published studies focusing on the treatment of acute inflammatory arthritis.	Clinical Rheumatology. 2006; 25(6): 807-813
Zochling-J; Grill-E; Alten-R; Ernst-J; Stucki-G; Braun-J	Identification of relevant functional issues for the care of patients with acute arthritis by health professionals, using the ICF framework and a multi- disciplinary focus group approach.	Clin-Exp-Rheumatol. Clinical-and- Experimental-Rheumatology. 2007; 25(3): 354-360

Journal title	# of articles
Advances in Speech Language Pathology	1
American Journal of Physical Medicine and Rehabilitation	1
Annals of Rheumatic Diseases	3
American Medical Informatics Association - Annual Symposium Proceedings	1
Archives of Physical Medicine and Rehabilitation	1
Arthritis Care and Research	3
Arthritis Research and Therapy	1
Arthritis and Rheumatism	1
Australian Occupational Therapy Journal	1
BMC Medical Informatics and Decision-Making	1
BMC Musculoskeletal Disorders	1
Burns	1
Canadian Journal of Occupational Therapy	4
Child: Care, Health & Development	1
Clinical Journal of Pain	2
Clinical Rheumatology	1
Current Opinion in Rheumatology	1
Developmental Medicine and Child Neurology	1
Disability and Rehabilitation	18
Disability and Society	1
Health Care Financing Review	1

Electronic Supplement 2: Journals that Published Articles using ICF Linking (58)

Health and Quality of Life Outcomes	1
International Journal of Geriatric Psychiatry	1
International Journal of Obesity	1
International Journal of Audiology	1
International Journal of Biometeorology	1
International Journal of Medical Informatics	1
International Journal of Rehabilitation Research	4
International Nursing Review	1
Journal of Advanced Nursing	2
Journal of the American Medical Informatics Association	2
Journal of Clinical Epidemiology	1
Journal of Hand Therapy	1
Journal of Occupational Rehabilitation	1
Journal of Policy and Practice in Intellectual Disabilities	1
Journal of Visual Impairment and Blindness	1
Journal of Rehabilitation Medicine	10
Journal of Rheumatology	1
Medical Care	1
Mental Retardation and Developmental Disabilities Research Reviews	1
Occupational Therapy International	1
Osteoarthritis and Cartilage	1
Palliative Medicine	1

Perspectives in Health Informatics Management	2
Physiotherapy	1
Physical Therapy	3
Quality of Life Research	3
Rehabilitation	1
Rehabilitation Psychology	2
Respiratory Medicine	1
Rheumatology	2
Nursing	1
Seminars in Speech and Language	1
Sleep Medicine	1
Spinal Cord	1
Spine	1
Work	1
Value in Health	1
Total	109

Amputation	1
Arthritis	18
Attention Deficit Disorder	2
Autism	2
Back impairment/	4
Blindness	1
Burns	1
Cancer/oncology	4
Cerebral palsy	2
Childhood disabilities general	8
Chronic fatigue syndrome	2
Chronic ischaemic heart disease	1
Chronic pain/ fibromayalgia	1
Cognitive disabilities	3
Communication	10
Community members	2
Chronic obstructive pulmonary disorder	3
Cystic fibrosis	1
Depression	1
Diabetes	1
Epilepsy	1
Head injury	1
Health resort patients	1
Hearing impairment	3
Heart disease	1
Idiopathic inflammatory myopathy	2

Electronic Supplement 3: Clinical Focus (Setting/Diagnosis) of the Included Articles*

Internal medicine	1
Irritable bowel syndrome	2
Mental health	3
Migraine headaches	1
Mobility impairments	1
Multiple sclerosis	2
Multiple/complex injuries	1
Muscular dystrophy	1
Musculoskeletal conditions	7
Neurological/neurosurgical	1
Nursing	3
Obesity	2
Osteoporosis	1
Palliative care	1
Posttraumatic stress disorder	1
Prader-Willi	1
Rehabilitation patients	1
Return to Work	3
Rheumatoid arthritis	9
Sleep apnea	3
Spina bifida	1
Spinal chord injury	1
Stroke	1
Upper extremity	9
Vision impairment/blindness	2

*more than one clinical practice area or setting could be counted per article

	Using linking rules 2002	Using linking rules 2005	Not using linking rules	Total
Study type				
Experimental	1	0	0	1
Analytic	6	4	9	19
Descriptive	29	18	42	89
Article Type*				
Qualitative Study (interviews/focus group)	0	3	5	8
Literature/systematic review	10	6	5	21
Appraisal or Teaching	20	9	21	50
Retrospective	0	1	0	1
Cross-sectional (eg: survey)	9	6	19	34
Cohort	3	0	1	4
Controlled Trial	1	0	0	1
Data Type Linked to the ICF*				
Articles	2	0	3	5
Case study	0	0	5	5
Conceptual model/theory/framework	1	0	2	3
Intervention aims	0	1	1	2
Other classification	0	0	5	5
Outcome measures (clinical assessments, questionnaires, health indices)	27	10	21	57
Patient records (eg charts & discharge summaries)	0	3	7	10

Table 1: Characteristics of articles linking text to the ICF

Transcribed data	1	5	5	11
Written responses to open- ended questions	5	3	7	15
Education reports	0	0	1	1
Language*				
Chinese [‡]	2	0	0	2
Dutch	0	0	7	7
English	32	17	40	89
Finnish	1	0	1	2
French	1	2	1	4
German	4	7	2	13
Indian‡	1	0	0	1
IsiXhosa	0	0	1	1
Italian	1	2	1	4
Malay	1	0	0	1
Norwegian	1	0	2	3
Russian	1	0	0	1
Swedish	0	0	4	4
Unknown	0	0	2	2
Age				
Adult	30	19	29	78
Child	3	0	6	9
Both	1	1	5	7
Unknown	2	0	8	10

*Articles can be counted in more than one category

[‡] Specific language not described

Quality Control	Using 2002 linking rules	Using 2005 linking rules	Not using linking rules
Reliability Statistic(s)*	50%	71%	17%
Multiple Raters	83%	81%	28%
Iteration	64%	76%	27%
Most Specific Codes Used	94%	95%	73%
Other Quality Controls	22%	29%	38%

Table 2: Incidence of Quality Control Methods Found in the Included Articles Based onLinking Process Type

Table 3: Qualitative Analysis Themes with example texts

Theme	Sub theme	Example text extracted from article
Preparing units of information	Preparing information - inductive	The text was divided where the researcher discerned a shift in meaning ¹⁰
		A concept was defined as a separate meaningful entity distinct from other concepts ²⁸
	Preparing information according to predetermined scheme	The information was categorized with respect to whether or not it was relevant for classification according to the ICIDH-2, thus info that was not ICF linkable was not linked ²⁹
Raters: Who links to the ICF?	Rater's knowledge of the content being linked or the ICF	Each relevant measure was linked separately by two health professionals who were experts in the ICF ³⁰
	Background of raters	[E]ncoding was carried out by three independent coders, a health care professional, a specialist for Physical Medicine and Rehabilitation, and an occupational therapist ²¹
Reliability	Agreement statistics	The kappa statistic for agreement in the linking of the concepts to the ICF between the 2 investigators was 0.74 ¹²
	Threats to reliability	Items classified differently most often concerned pain related to specific movements or tests. In some cases these items were classified differently because of non-specific descriptions from the physiotherapists and patients, but in several cases non-agreement was caused by lack of clarity in the definitions of the detailed version of the [ICF] ²⁹
Matching or	Ratio of information from	[A]n item suggested by an expert may not be

	-			
translating concepts from the text to ICF	text to ICF category equivalency	equivalent to a respective ICF category, and some information or meaning may be lost ¹⁷		
categories		If a meaning unit contains more than one concept, it was linked to more than one ICF category ¹²		
	General functioning, disability or health concepts with no ICF categories	Coding units that did not fit with the ICF comprised health information such as "somatic symptoms", "global well-being", "feeling sick", "physical activities" or "clinical chemistry ²¹		
		"[T]here were 22 instances where the construct was more general than the corresponding ICF category and could only be linked to an "unspecified" ICF category. ³¹		
	General concepts not covered by the ICF	[C]oding units, that were judged not to be covered by the ICF referred to diagnosis or personal factors like "enjoyment of life" or "having let people down. ²¹		
Information unable or difficult to capture by linking to the ICF	Information about Client/patient perspective not captured	Items assessing patients' satisfaction with, or perceived importance of, physical ability were excluded ³²		
	Distinction between activity and participation not captured	[T]he dimensions of activities and participation was not always clear. This was especially true for activities of daily living (ADL). The exact difference between ADL-activities and ADL as participation was perceived as abstract and confusing ²⁵		
	Relationships	[We] also developed a coding scheme to capture the relationships between the codes such as 'the cause of', 'related to' or 'part of' ¹⁴		
		[T]here is no representation of context thus we have no idea how different concepts are arranged ³³		

Reporting standards	What information was linked	Used 80% frequency of reporting an issue between responders as the cutoff for reporting the categories ³⁴
	Bandwidth	[B]andwidth shows the frequency or extent to which categories were represented across the ICF and was employed ¹⁸
	Content density	[C]ontent density was reported as categories per item: >1, <1, or 1 ²⁰
	Frequencies and specificity of ICF categories	ICF categories with a frequency equal or greater than 10% are shown ²⁰
Overall linking process	Stepwise linking process	Next, the research team, comprised of the primary researcher (an academic occupational therapist), the research assistant (RA), and a clinical occupational therapist (who is also a doctoral candidate in Rehabilitation Science), independently reviewed the coding of the qualitative data and then met as a group to confirm that the barriers and facilitators fit with definitions and that the mapping of these factors to the ICF was congruent with the definitions of the categories. Discrepancies were addressed through open dialogue until agreement on the code and the congruence or lack of fit with the ICF was reached. ³⁵
	One step cooperative linking	Each problem statement was cooperatively classified by a panel of three individuals ²⁵

Appendix A: ICF Search strategy syntax for OVID Medline

#23	#22 and #20
#22	#21 and #19
#21	#17 and #18
#20	(2001 in PY) or (2002 in PY) or (2003 in PY) or (2004 in PY) or (2005 in PY) or (2006 in PY) or (2007 in PY) or (2008 in PY)
#19	ENGLISH in LA
#18	ABSTRACT in XREC
#17	#14 or #15 or #16
#16	#9 or #10 or #11 or #12 or #13
#15	#3 or #4 or #5 or #6 or #7 or #8
#14	#1 or #2
#13	((classification* near3 health) in AB)or((classification* near3 health) in TI)or((classification* near3 health) in MESH)
#12	((classification* near5 handicap*) in AB)or((classification* near5 handicap*) in TI)or((classification* near5 handicap*) in MESH)
#11	((classification* near5 impairment*) in AB)or((classification* near5 impairment*) in TI)or((classification* near5 impairment*) in MESH)
#10	((classification* near5 disabilit*) in AB)or((classification* near5 disabilit*) in TI)or((classification* near5 disabilit*) in MESH)
#9	((classification* near5 functioning) in AB)or((classification* near5 functioning) in TI)or((classification* near5 functioning) in MESH)
#8	international classification impairment*
#7	international classification impairment* disabilit*
#6	international classification impairment* disabilit* handicap*
#5	((ICIDH-2) in AB)or((ICIDH-2) in TI)
#4	((ICIDH 2) in TI)or((ICIDH 2) in AB)
#3	((ICIDH2) in TI)or((ICIDH2) in AB)
#2	international classification functioning disabilit* health
#1	((ICF) in AB)or((ICF) in TI)

Chapter 2

Using the ICF Linking Rules to analyze assessments: A method for demonstrating content equivalency

Cieza A, Fayed N, & Bickenbach JE.

This chapter is not yet published.

The contributions of each author to the manuscript are as follows:

AC drafted the original version of the ICF linking method. She provided scientific feedback on each subsequent draft of the proposed method, including the version presented here. AC conceived of the connection between the response options and the perspectives that is unique to this version of the linking rules (45%).

NF revised numerous versions of this method over two years using philosophical, linguistic and qualitative-traditions techniques. She created the interpretations of ICF and WHO definitions for the purposed conceptual content analysis. NF drafted this paper and the structure of communicating this method for the purpose of content equivalence. (45%)

JB provided specialized refinements about philosophical interpretations of World Health Organization perspectives and ensured the internal consistency of the method by testing the method with the previous two authors (10%).

Introduction

The International Classification of Functioning, Disability and Health (ICF) is a standard vocabulary and uniform language devoted to classifying information about the impact of health conditions on individual lives(1,2). The classification enables the description, comparison and contrast of health and health-related information, thereby facilitating the work of clinicians, epidemiologists, researchers, and policy makers. Uniformity of language is a requirement for the advancement of scientific knowledge(3) and information sharing in the field of functioning, disability and health(4).

As health researchers and policy makers increase the amount, scope and implications of the data they collect, they are increasingly aware of the need to connect the information they obtain from one setting, country or system to another(3,5,6). Linking rules were developed by Cieza et al. in 2002(7) and updated in 2005(8) to connect health and health related information to the standard language of the ICF, using the classification as a common basis for comparison. These linking rules have been used to compare the content of several health assessment tools and have been referenced in over 50 peer-reviewed studies(9).

Systematic review(9) and experiential use have confirmed the limitations stated in the Cieza et al. 2005 linking paper. Firstly, items addressing the same health domain that were linked to the same ICF categories might represent different perspectives. For example, a questionnaire item such as 'how often do you experience difficulty walking?' would have been linked to the same ICF category (d450 walking) as the item: 'how important is it to you to be able to walk?' The former question probes walking from a disability perspective while the latter probes walking from a QoL perspective, thus they are not as equivalent as the ICF category that represents them would imply.

Secondly, the manner by which health domains are quantified or measured through words (operationalized) was not considered. For example, the item '*how often do you experience difficulty walking?*' quantifies the perspective of disability by means of frequency whereas the question: '*to what extent do you experience difficulty walking?*' quantifies disability through intensity or severity. Again, both these questions would be linked to 'd450 walking' even though they quantify the activity of walking differently.

Lastly, the identification of concepts to be linked to the ICF was often mechanical without appreciation of the uses to which an item was to be put' or "the applications

proposed for its use. For example, a response option from the Health Utilities Index III reads as follows: *Able to see well enough to read ordinary newsprint and recognize a friend on the other side of the street, without glasses or contact lenses*. Literal interpretation of the concepts here might result in linking to ICF categories such as *b21002 Binocular acuity of near vision, b21000 Binocular acuity of distant vision, d166 reading,* as well as *e1251 Assistive products and technology for communication*. This complex list of ICF categories masks the true purpose of the response options, which is to assess *b210 Vision*. The concepts 'reading the newspaper', 'identifying the friend on the other side of the street', and 'the use of glasses' are merely probes used to decipher the degree of vision that a respondent might experience; linking them to the ICF in a literal manner did not convey what was being assessed.

The goal of this current linking update is to address the limitations above and propose refinements to the linking method. This will be achieved through data extraction using illustrative items from the following assessments: the World Health Organization Disability Assessment Schedule II (WHODAS II)(10), the World Health Organization QoL 100 Assessment (WHOQOL 100)(11), the Health Utilities Index III (HUI III)(12) and the manual muscle test (MMT)(13). A decision-support tool for linking difficult concepts to the ICF will also be presented.

Preliminary considerations

Before preparing the data for linking one must first consider the types of health information that will be analysed. Although linking can apply to many types of qualitative health information, the focus here will be on assessment instruments (Figure 1). Assessment instruments include patient (and proxy) reported measures, informal clinical assessment, and technical assessments. Patient and proxy reported assessments include health-status and disability questionnaires, quality of life (QOL) questionnaires, and patient and caregiver reports of health domains such as pain, sleep, irritability or shortness of breath. Clinical assessments are the evaluations clinicians use in order to generate a clinical judgement. Measurement of factors such as strength, coordination, reaction time and wound healing are examples of informal clinical assessments. Technical assessments include elements such as laboratory and radiographic assessments as well as tests such as standardized neuropsychiatric assessments.

Prior to analyzing assessment content, the development, instructions and/or manual for completion of the assessment instrument should be consulted as contextual reference. General knowledge about the assessment in clinical practice or scientific

literature is also important to complete an adequate analysis. This information is necessary to appreciate fully the context of the assessment and how respondents to, or administrators of, the measures might reasonably interpret the assessment content.

Preparation of information

Linking an assessment to the ICF involves many considerations that should be addressed. We propose use of a data extraction table to organize content-relevant information about an assessment (Table 1). Selected items, questions or cues from the WHODAS II, WHOQOL 100, HUI III, and MMT will be used as examples to illustrate the procedure. Each section below corresponds to a column in the data extraction table.

Using the data extraction table:

- i. <u>Item</u>: Each question should be copied into this column exactly as it appears in the assessment instrument. No additional information should be added or taken away.
- ii. <u>Response options</u>: The text from the response options should be copied into this column exactly as it appears in the assessment instrument. No additional information should be added or taken away.
- iii. <u>How the response option quantifies (operationalizes) the health-domain</u>: The manner by which the response options quantify the health domain should be listed explicitly. The selected items found in Table 1 from the WHODAS II are operationalized as "intensity (of problems)". QoL is more likely to have operationalizations such as the ones found from the WHOQOL-BREF item #21, where the domain of "sex life" is quantified as "extent of satisfaction".
- iv. <u>Perspective:</u> Health perspectives are the different approaches to, or points of view for, describing, assessing, valuating, explaining, and measuring health. Perspective represents the lens with which an assessment item/question/cue was posed to the respondent. For example, item D4.5 from the WHODAS II and item 21 from the WHOQOL-BREF are both about the health domain of *d72202 Sexual relationships* (Table 1). They differ in perspectives because the WHODAS item represents functioning and disability and the WHOQOL item represents QoL.

A working list of perspectives is provided, developed using definitions found in the ICF, the WHOQOL BREF manual, and thorough testing the data extraction table with various instruments (Appendix A). These perspectives include: functioning/disability (with sub-perspectives of needs/assistance, impact, and norms/development), and QoL (of which health-related QOL is a sub-perspective). If users find additional perspectives through the proposed data extraction procedure, they are encouraged to contact the authors for feedback.

v. <u>What is it about?</u> This column challenges one to determine what the content of the item/question/cue is about. When making decisions about what should be placed in this column, ask the following questions: 'What piece of health information is being extracted from a patient or client when they are responding or reacting to this question or cue?' 'What would the patient, clinical, or research community reasonably expect this question to be about given the context in which it is presented and used?'

In our experience, and in contrast to clinical assessments, health status, disability and QoL self-report questionnaires articulate questions in a relatively direct and transparent way to elicit information from patients and their proxies. Therefore, one can often remain close to the language of the item/question (i.e. refrain from overextrapolating meaning) when making the judgement of what to place in this column.

Clinical assessments such as the MMT or technical assessments require advanced knowledge of the question, cue, or patient instruction in order to appraise the health domain intended by the item with confidence. Advanced understanding of the true purpose of a clinical or technical assessment often requires extrapolation or interpretation to determine 'What it is about?' Thus, data extraction of clinical and technical assessments requires some professional knowledge to fill in the column 'vi. what is it about?'.

Utility instruments and other assessments with health information in the response options often complicate the answer to the question: 'what is it about?' For example, the attribute "Pain" from the HUI-III has information in the response option about intensity of pain along with the extent of activity limitation (Table 1). Pain intensity and activity limitations are used to quantify the stem concept being assessed: pain. Therefore the response options contain useful supplementary information for operationalizing the item but linking the response options to the ICF masks the true purpose of the item.

vi. <u>ICF category</u>: The information entered into this column involves linking to the ICF. See the section on linking to the ICF and Figure 2 for guidelines.

vii. <u>Annotations</u>: Any additional information that is relevant to help a user understand an item, question or assessment cue should be listed here. Examples that help anchor one's understanding of: "What it is about?" should be listed here along with any other relevant information not captured by any of the previous columns. Finally, if an *8 (not specified)* ICF category is listed in column vii, the unit of health information that was not specified should be made explicit here.

Summary on the Preparation of Assessment Data

Regardless of the type of assessment instrument, the core procedure for using the data extraction table always involves determining *what an item/question or cue is about,* whether the answer is easily obtained or needs to be obtained with specific expert knowledge of the instrument. Data extraction of assessment content also involves determining the perspective found in the item and the operationalization of the response options, the quantification of the health domain from response options, as well as any information not captured by the previous steps.

Linking to the ICF

A guide has been devised in the form of a decision tree to facilitate linking for new ICF users and to assist expert linkers with challenging instances of linking (Figure 2). The tree takes advantage of the existing structure of the ICF while resolving problems with linking general concepts. Finally the tree should clarify what to do with concepts not easily captured by the ICF.

In order to begin using the tree, a thorough understanding of the concepts, definitions and structure of the ICF is necessary. It will be difficult to decide whether a concept is about a component of functioning, disability or health without understanding the definitions contained in the ICF.

- <u>Use of the linking tree</u>: When using the tree, it is important to decide on the level of specificity of the ICF category in a step-wise manner. In the WHODAS-II D6.3 example, it was decided, based on the data preparation phase, that the item was about: 1. living with dignity, 2. the attitudes of others, 3. the actions of others (Table 1). We will use the linking tree to link the first two health domains to the ICF:
 - 1. Living with dignity:

- Q: Does it belong to the universe of the ICF?
- A: Yes
- Q: Can it be assigned to a component of the ICF?
- A: Yes, it is a personal factor
- Q: Can it be assigned to a 1st level category (chapter) or higher?
- A: No, there are no 1stnd level categories (chapters or codes) for personal factors.

Thus the unit is assigned the component *pf* which represents the component *personal factors* and "living with dignity" is documented in the annotation column.

- 2. The attitudes of others:
 - Q: Does it belong to the universe of the ICF?
 - A: Yes
 - Q: Can it be assigned to a component of the ICF?
 - A: Yes, it is part of the environment component
 - Q: Can it be assigned to a 1st level category of the ICF?
 - A: Yes, it is part of chapter e4 Attitudes
 - Q: Can it be assigned to a 2nd level category or higher?
 - A: No, those categories are too specific to represent the general concept of the attitudes of others.

The linking tree also supports linking information that is very general or broad in scope. Question H1 from the WHODAS-II is about overall health (Table 1). Use the tree to link it:

- 1. Overall health
 - Q: Does it belong to the universe of the ICF?
 - A: Yes, health is a part of the ICF and it can encompass all of the components
 - Q: Can it be assigned to a component of the ICF?

A: No, choosing any one component will be too specific. Assign to the not defined-general health *nd-gh* code.

Finally, the tree applies to linking very specific information to the ICF. For example, Question D4.5 from the WHODAS-II is about sexual activities (Table 1):

- 1. Sexual activities:
 - Q: Does it belong to the universe of the ICF?
 - A: Yes
 - Q: Can it be assigned to a component of the ICF?
 - A: Yes, it is part of the activities & participation component
 - Q: Can it be assigned to a 1st level category of the ICF?
 - A: Yes, it is part of chapter d7 Interpersonal interactions and relationships
 - Q: Can it be assigned to a 2nd level category or higher?
 - A: Yes, it can be assigned to the 3rd level category *d7702 Sexual relationships*

iv. Special 8 and 9 categories:

There are units of information that are conceptually compatible with the ICF at various levels of the classification, however, they do not map as easily the previous examples. Use of the *other specified* (otherwise known as the 8 categories) or the *unspecified* (otherwise known as the 9 categories) assists in dealing with this issue.

The other specified (8) categories are used when the concept fits very well at a certain health domain or level of the classification but cannot be found as an existing ICF category. The item about pain in the HUI III has a response option that is about 1. pain and 2. discomfort. The concept pain can be easily linked to the ICF using the linking tree to arrive at the category *b280 Sensation of pain* found in the chapter *b2 Sensory Functions and Pain*. The concept of discomfort is defined as a feeling of disturbance or a deprivation of ease(14). It is a body function that is clearly a sensation and can be related to pain, but is distinct from pain. There <u>could</u> be a second level category called discomfort but one does not exist. In this case, the best option is to select *b298 Sensory functions and pain other specified* and then note the concept of discomfort in the annotation section of the table. The unspecified 9 categories are used when the concept clearly fits one or more concepts found in the ICF, yet one cannot be certain about which category it represents. The item D4.3 from the WHODAS II is about "getting along with people who are close to you". This concept is represented by the ICF under the group of categories called *Particular interpersonal relationships* and could be represented by the category *d760 Family relationships* or *d770 Intimate relationships*. It has not been specified from the item whether it is either or both of these categories. Hence we assign the category *d779 Particular interpersonal relationships*.

Use of the tree as well as 8 and 9 categories is encouraged for new users of the linking rules or users who are facing difficult problems in linking items and concepts to the ICF. Expert users should briefly review the linking tree as well as guidelines regarding the otherwise and unspecified categories.

Summary of linking update:

- 1) Understand the concepts, definitions and structure of the ICF
- Extract information about perspectives, item quantifications (operationalizations), what the item is about, and annotations from content analysis separately. Using the data extraction table will assist with this task.
- 3) Take advantage of the structure of the ICF during linking. The decision tree will assist with this task.

Discussion:

The 2002 and 2005 linking rules were useful to describe or represent assessment instruments using the ICF for the purposes of delineating content, establishing content validity for research planning and connecting goals between outcome measures and study outcomes. This update report focuses on teaching a method to extract content from assessments for the purpose of determining item equivalency

Since the earlier reports, the ICF has increasingly been used as a basis for measurement (15) and therefore requires comparative properties. This need for content equivalency was shown in a study that linked items from various generic questionnaires to the ICF. Items with interval scaling that mapped onto the same ICF category *b130 Energy& drive* were fit to a uni-dimensional Rasch model (15). Consequently, data from the various questionnaires could be combined.

We hypothesize that the method presented in this article will improve the fit between Rasch models and data collected using questions or items from separate instruments that have been deemed content equivalent. Additional testing of the method with different users and different use cases is necessary to test this hypothesis.

This version of the linking rules resolves certain open issues from the previous versions as well as issues identified from systematic review of the literature on ICF linking. The 2002 and 2005 rules were useful for describing the health domains found in QoL measures versus health-status measures. Since the differences between these measures were mainly in the approach or perspective from which questions are asked, as opposed to the health domains represented, use of rules did not result in a definitive conclusion about whether a measure truly represented QoL or health-status. Making this distinction is important in the field of functioning, disability and health where lack of consistency in terminology makes it difficult to relate results of one study to another, let alone to combine studies into meta-analyses (16).

The results of the systematic review showed that some users of the 2002 and 2005 ICF linking method were concerned with information loss when linking information to the ICF (9). This concern was also listed as a limitation in the 2005 linking rules. Response options as well as the relationships between health domains were discarded in the process of linking items or questions to ICF. Use of the data extraction table shows the complexity of items and the various components that are important for an understanding of a question or cue from an assessment.

In this paper, the modular way that content is extracted from the items demonstrates factors that must be taken into account when attempting content equivalency. Items analyzed in this manner are compatible with collecting information about items from assessment instruments in larger databases such as item banks. The PROMIS Cooperative Group uses their comprehensive item bank as a basis for developing a short-form instrument(17,18). However, item banks can also show the items that have already been created to assess various health domains and can be checked prior to the creation of new assessment instruments.

One on-going challenge of linking health and health-related information to the ICF is what to do about very general concepts such as physical health and QoL, or concepts that fit conceptually with the classification but do not have an existing ICF category. The 2005 linking rules provided some guidance to deal with this issue through the *not-defined* and *not covered* options. The new linking decision-tree and clarification

about the *other specified* and *unspecified* categories were introduced to support decision-making for these linking issues, in particular for users new to the ICF.

A continued limitation of the linking rules is the inability to connect health domains semantically once they are linked to the ICF. For example, in the item '*I have difficulty walking because of my leg pain'*, two separate concepts can be identified and linked to the ICF categories d450 (walking) and b28015 (pain in lower limb). Once linked to the ICF categories, the notion that walking was affected by pain is lost and hence the relationships between the health domains assessed by the item are also lost.

This report focuses on data preparation and data extraction from items in order to determine content equivalency between and among instruments. Many of the issues discussed here are additionally applicable to analysing other forms of qualitative health and health-related information. The targets of health interventions, information collected through interviews, or qualitative information found in policy reports all adhere to similar principles of data extraction such as asking the question: 'What is it about?' when preparing the data. Connecting that information to the ICF follows the same process found in the linking tree and through using *other specified* and *unspecified* categories.

Conclusion

The linking rules created in 2002 and updated in 2005 have been revised and are outlined in this report. Open issues from previous linking rules, the results of systematic review, six years of continued linking experience by the authors' and most importantly, the need for content equivalency as an outcome of the linking process all inspired the revisions contained in this update. Thus content equivalency is now a desired outcome of the linking process for assessment instruments.

The aim of this report is to illustrate the processes of data preparation and data extraction when attempting to link to the ICF for the purpose of content equivalency. Decision-making support for ICF linking in the form of the decision tree is presented for novice linkers and to support expert users with difficult linking problems. Empirical testing of this updated process is required in order to determine its usefulness, develop the perspectives options, and refine the process even further. Users of this method are encouraged to contact the developers with special use cases, difficult linking problems or proposed additions to the perspectives found in health assessments. These communications will help the authors, and the field, to refine our ideas even further.

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Table 1: Preparing and extracting data from various assessments for content equivalency purposes

Nr.	i Item as appeared in the	ii. Response options	iii. Quantifying the item*	iv. Perspective	v. What is it about?	vi. ICF-Category	vii. Annotation
Items from	self-report ques	tionnaires					
WHODAS-II #D4.3	How much difficulty did you have in getting along with people who are close to you?	None, Mild, Moderate, Severe, Extreme/ cannot do	Intensity (of problem)	Disability	Getting along with people who are close to you	d799 Interpersonal interactions and relationships, unspecified	People who are close to you
WHODAS-II #D4.5	How much difficulty did you have in sexual activities?	None, Mild, Moderate, Severe, Extreme/ cannot do	Intensity (of problem)	Disability	Sexual activities	d7702 Sexual relationships	
WHOQOL- 100 #21	How satisfied are you with your sex life?	Very dissatisfied, Dissatisfied, Neither satisfied nor dissatisfied, Satisfied, Very satisfied	Intensity (of satisfaction)	QoL	Sexual life	d7702 Sexual relationships	

WHODAS-II #H1	How do you rate your overall health in the past 30 days?	Very good, good, moderate, bad, very bad	Intensity (of problem)	Functioning / Disability	Overall health in the past 30 days	nd-gh not-defined general health	The past 30 days
WHODAS-II #D6.3	How much of a problem did you have living with dignity because of the attitudes and actions of others	None, Mild, Moderate, Severe, Extreme/ cannot do	Intensity (of problem)	Disability	 (1) living with dignity (2) attitudes and (3) actions of others 	 (1) pf: personal factors 2) e4: Attitudes (3) e3: Support & relationships 	Living with dignity
Cues from c	linical assessme	ents		<u> </u>			
Manual Muscle Test	Say: "I am going to push down and I want you to resist me. Keep your arm up as I push down." (while patient seated and glenofemoral	0 (no mm contraction can be seen or felt) to 5 (part moves through complete range of motion against gravity and maximal resistance)	Intensity (of strength)	Functioning / Disability	Shoulder (Middle deltoid) strength against gravity	b 7300 Muscle power functions of isolated muscles & muscle groups	Deltoid muscle(s) Against gravity

Item attribu	joint abducted to 90 degrees and elbow flexed to 90 degrees) tes from utility	measures					
Attribute #6 from the HUI-III	PAIN	 Free of pain and discomfort. Mild to moderate pain that prevents no activities. Moderate pain that prevents a few activities. Moderate to severe pain that prevents some activities. Severe pain that prevents most activities. 	Intensity (of pain / discomfort) Intensity (of activity limitation)	Disability	Pain & Discomfort	b280 Sensation of pain b298 Sensory functions and pain other specified	Discomfort

Appendix A: Working list of perspectives

Perspective name	Perspective definition
QoL	QoL perspectives seek to determine individuals' 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.
Functioning	Functioning perspectives refer to the interaction or the individual components of body functions, activities and participation
Disability	Disability perspectives probe impairments, activity limitations and or participation restrictions
Needs / Assistance	Needs perspectives assesses an individual's functioning or disability based on the needs that they draw from their social, physical and institutional environment
Impact	Impact perspective infers an individual's functioning or disability based on the effect that they have on their social, physical and institutional environment
Development / Norms	Developmental perspectives contrast an individual's functioning or disability with that of another reference individual or group
Health Barriers	Barrier perspectives reflect environmental factors that hinder the functioning of an individual
Health Facilitators	Facilitator perspectives reflect environmental factors that promote or allow for the functioning of an individual





*see section "linking to the ICF" ii. regarding the use of 8 and 9

Chapter 3

Illustrating Child-Specific Linking Issues using the Child Health Questionnaire

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This article is currently under review for the American Journal of Physical and Rehabilitation Medicine

NF provided the rationale for using the CHQ as the instrument to illustrate childspecific linking problems. NF performed the linking and identified the content analysis issues that are unique to children.

AC provided feedback and expertise about the linking problems that were encountered specific to children as well as all the scientific discussions presented in the article and edited the entire document.

JB suggested the layout and framework of the manuscript, assisted with structuring arguments related to child-specific linking and provided editorial feedback.
Abstract

The publication of the ICF-CY as a derived classification of the ICF has enabled child health and disability researchers to implement the classification into their work. There is little discussion available in the literature specifically about challenges associated with connecting the ICF-CY to child health instruments. The objective of this paper is to apply an updated approach to linking child-specific assessments using the Child Health Questionnaire as an example. We discovered the importance of knowledge about child-health assessment as a linking requisite, issues with linking information about child behavior, the importance of clarifying the vantage point from which one is linking (e.g., child, parent, or family), and that one should carefully consider the true purpose or targets of items before linking them to the ICF-CY, irrespective of the simple language used in the item. Finally, we propose the use of a new not-defined abbreviation to denote items that assess overall child development: *not-defined-development (nd-dv)*.

Introduction

Over the last three decades, many patient-oriented instruments for children have been developed and used^{1,2,3,4}. These instruments have been developed in different contexts based on a variety of conceptual understandings of health, disability or quality of life^{5,6,7}. Moreover, the instruments have been used with a multitude of clinical and research populations over time. It has therefore become difficult for clinicians and researchers to select the appropriate patient-oriented instrument for a specific purpose.

Connecting instruments to the International Classification of Functioning, Disability and Health (ICF)⁸ or the Children and Youth Version (ICF-CY)⁹ enables researchers and clinicians to understand and compare the content among and between measures so they can be matched to an intended purpose. Existing methods have been introduced and tested in the literature to connect the content of patient-oriented instruments to the ICF. These methods are called ICF linking rules^{10,11}. Following systematic¹³ review and 8 years of empirical use, further comments and refinements¹² have been made on the linking method specifically with regard to assessment instruments. In particular, this updated approach to linking involves determining health domains contained in items as well as health status versus quality of life (QOL) perspectives found in patient-reported questionnaires. It is important to know whether these refinements will also be applicable to linking child health instruments to the ICF-CY as opposed to adult-oriented health instruments to the ICF.

Prior to the introduction of the ICF-CY, child-specific instruments had been linked to the ICF^{14,15,16,17,18}. Since publication of the ICF-CY, there have also been studies that link child-specific instruments to this child and youth version^{19,20,21}. However, to our knowledge, there is no literature that is focused on methodological issues that are specific to linking child health questionnaires to the ICF-CY.

Given the recent refinements to the content analysis process attached to the linking rules, the objective of this paper is to apply the content analysis to an extensively used, questionnaire to determine whether there are child-specific issues that have to be taken into account. Those issues will be described for the purpose of making recommendations.

Application of linking rules

Instrument selection and rationale

The Child Health Questionnaire Child-Report 87 (CHQ CF-87)²² was the instrument chosen to illustrate the issues involved in linking to the ICF-CY. This choice was based on its frequent use in the literature for over a decade, its use across various health conditions in child health, its use across languages and cultures, its broad conceptual basis for health in its development and finally the variety of different domains it measures.

The CHQ CF87 was the first questionnaire among the CHQ series. This instrument was developed specifically for children and was based on the 1948 World Health Organization (WHO) definition of health as a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity²³. The manual of the CHQ states that "health was conceptualized as "having...dimensions of physical and psychosocial (emotional, behavioral, and social) well-being... that [could] affect a child's ability to perform important social roles including school work, usual activities at home, getting along with others, and developing friendships" (p. 31)²², which suggests that the conceptual basis of this instrument is consistent with the definition of health within the ICF-CY. Content for CHQ CF87 subscales, known as 'sections', was based on literature reviews, expert review, review of existing health instruments available for children and adults as well as focus groups and interviews with families. Initial psychometric validation work, occurred with children with ADHD, renal failure, cystic fibrosis and a school-based sample²⁴.

Data extraction and ICF-CY linking

We reviewed the conceptual basis and development of the CHQ from the manual²⁵ and selected literature reporting on the questionnaire^{26,27,28}. Using the 2005 linking rules¹¹ and additional information from the content equivalency paper¹², content was extracted from the CHQ CF-87. This content included: how the item was quantified (e.g., frequency, severity, type) from the response options, the perspective of the item (e.g., functioning/disability/health or QOL), the target domain of the item (the column labelled: What is it about?), the ICF-CY category and code based on the domain assessed in the item, any connections between the domains within the item (e.g., 'and' or 'because of'), and finally annotations that contain information not captured from the previous columns. Once the entire questionnaire was linked, we selected 1-3 items from

different CHQ sections that were illustrative of aspects of the content analysis that were unique to child health for display in Table 1.

Results

<Insert Table 1>>

Several issues and challenges emerged related to child-specific linking.

1.0 The aim is not apparent

During the linking process, when the purpose of the item was not clear, our knowledge of child health assessment was needed to interpret complex concepts from the item even if the questionnaire items were simply worded for children. This was evident for our first challenge: identifying the domain (What it is about?) when the aim of the item was not obvious. For example, without background knowledge about child health assessment, the aim of the item 5e "wanted to be alone" was not obvious. Previous knowledge about typical cues used to assess externalizing and internalizing behaviors in children was key to understanding that this question functioned as a screen to detect the presence or severity of a mental health condition. Since this item was also shown in the context of other questions probing internalizing and externalizing behaviors, we became more confident about the intent of the item domain within the subscale. Without knowledge of child mental health assessment and acknowledgement of the context from which the item was drawn, the item could have been interpreted as a question about temperament or personality.

2.0 Behavioural Problems

Our second challenge was similar to the first, although specifically related to questions about behavior problems. Questions about behavior were sometimes present in a literal sense (e.g., item 3.2) but they were also present as cues to screen for pathological problems or mental diagnoses. For example: the two items shown from "Section 5: Getting along/behavior" ask children about their externalizing and internalizing behaviors as indicators of deeper emotional or psychological problems. Also, few of the items in Section 5 made sense as components of child functioning on their own. The rationale for linking these items to a component of child functioning, disability or health in the ICF-CY therefore seemed weak.

As a solution, we decided to defer linking these items to ICD-10 codes (Table 1). This decision was supported by the description of the subscale in the manual of the CHQ, which stated that these items were based on the clinical assessment of "overt behavior as components of mental health [conditions]" (p. 36).

Finally, we used the dimensions of ICF-CY components as further criteria to decide if these items were appropriate to link. Thus if the item represented something that could be impaired (as with a body function), limited (as with an activity), restricted (as with a participation), presented barriers or facilitators (as with environmental factors) or described the unique features of the particular child (as with personal factors), then it was suitable for linking to the ICF-CY. Most items in Section 5 did not meet the previous criteria and therefore could not be linked to the ICF-CY.

3.0 Clarifying the vantage point of linking

Child health assessment is connected to family and caregivers. Consequently, there were items in the CHQ that asked about the child as well as the family and created confusion about whose health was being linked. The items in Section 9: "You and Your Family", asked both about the child's and the family's functioning. From the vantage point of the child, the activities that are performed with the family constitute his/her participation, and the family constitutes the environment of the child. We found that family functioning could only be linked from the vantage point of the child (so long as the child was the subject of the questionnaire). Thus if item 9.1a was interpreted as a family functioning item, it could not have been linked to the ICF-CY. The CHQ assesses child health as opposed to family health; this particular section assessed the child's level of health or disability based on the extent of impact on family activities. In ICF-CY terms, the item indirectly asks: 'How severe is the child's disability?' based on the extent to which the health or behavior of the child had an effect on the family environment. It was decided that the child's level of disability was being assessed based on the impact the child had on the family and the solution was to link from the child's point of view, placing the child's functioning as the aim of the item with the family as the environment or context of the child.

4.0 Quality of life questions for children

Issues about distinguishing quality of life from functioning, disability and health perspectives are discussed in the Cieza et al.'s 2010 linking refinements¹² and this approach was applied to the items. We found that the QOL perspective was worded and operationalized differently in child instruments as compared with adult instruments. Typical QOL cues from adult items such as 'how satisfied are you with...' or 'how

important is it for you to...' were not part of the CHQ. Items that began with 'how did you feel about...' seemed to be targeting the child's subjective perception of the ICF-CY domain. For example, item 7.1b asked "how good or bad did you feel about your school work?" We choose 'school work' as the health domain and QOL as the item perspective because the question seemed to be asking about the subjective experience of school work. Also in Section 8: "Your Health", item 8c tapped into children's expectations of their future health using the cue "I think..." and this created confusion about whether a QOL versus a disability perspective was present in the item.

For this challenge we decided, when in doubt about the perspective of an item, we returned to the WHO definition of QOL: the child's *perception* of their position in life as assessed by expectations, standards and concerns²⁹. These subjective elements must be mentioned in some form in the item in order to be assigned a QOL perspective. Thus one should consider whether an item incorporates the preferences, standards, expectations and concerns of the child (e.g. the child's perception of their functioning, disability or health) even if the language used in the item was presented in simpler terms than those usually used with adult items.

5.0 Questions about a reference group

Certain items cued children to compare their functioning with that of a **reference group**, which gave rise to questions about which perspective was represented in these items. For example, item 5.2: "compared to other children your age, in general would you say your behavior is..." evoked ideas about norm or reference-based approaches to measurement, which are not so frequently used in adult health questionnaires. In reviewing the introductory chapter of the ICF-CY, it became clear that cues about children's functioning relative to a reference group were tapping into the construct of development. In the ICF-CY disability may take the form of delays in development. Thus comparison with a reference group was a probe tapping into the developmental aspects of disability when linking to the ICF-CY and was therefore part of the disability perspective as applied to children.

Discussion

In this paper we identified child-specific issues resulting from applying the linking rules to a widely used child health instrument, the CHQ. Performing the linking exercise deepened our understanding of the content of the CHQ by identifying the domains and perspectives in the instrument. The diversity of conceptual content discovered in this

instrument might explain why different researchers use this instrument to assess different outcomes. For example, the CHQ has been mentioned in the literature as an instrument of QOL, HRQOL as well as a tool to assess physical and emotional wellbeing²⁵⁻²⁷. Linking the instrument to the ICF-CY provided a standard by which to evaluate CHQ content using an internationally accepted language of functioning and health. Using this language, the CHQ proved to be a questionnaire of physical and emotional health *and* QOL in some form or another, depending on the subscale or combination of subscales used. Thus, according to the analysis presented here, researchers should note that using the CHQ in its entirety does not assess solely any one of these outcomes but all of them to some degree.

There were other issues that we believe are salient to linking child assessments to the ICF-CY that were not brought forward by our analysis. One of these that needs more discussion is the issue of child development. In the 2005 linking rules, concepts that were considered conceptually part of the ICF but were too general to be assigned to a specific ICF component or category such as general mental and physical health were linked as *not-defined* and development seems to fit this scenario. For example, if there is an item such as "how is your child developing overall?", we would not be able to link it to body functions, body structures, activities or participation because development spans all of these components. Instead, we propose the use of the abbreviation *nd-dv* (*not-defined-development*) to denote such a case. This case is not the same as one in which items that compare children to a reference group to assess disability. Therefore the question "Is your child reading as well as most other children his or her age?", addresses the ICF-CY category, Reading d166 from a developmental disability perspective but should *not* be assigned the abbreviation nd-dv.

The issue of how to link information given by the child as opposed to information from proxy respondents did not arise but it is important to discuss. Even if a questionnaire or assessment queries a parent to answer on the child's behalf, one must link the actual domain as it applies to the child and not to the parent. Also, instruments intended to assess the child using proxies often ask about the child's disability based on the level of assistance the child requires. For example one might be tempted to link the item from the CP Child: "During the past 2 weeks, how difficult was changing diapers?"²⁹, to the domain 'Assisting others with self-care d6600', yet in the context of an infant health questionnaire, this item is about the child's positioning and toileting. Child assessments should thus always be linked to the ICF-CY from the child vantage

point, while separate caregiver assessments can be linked to the ICF from the vantage point of the caregiver.

Overall, there will always be linking challenges for which there is no precedent in the literature; because of this, knowledge of assessment instruments and the ICF-CY by those who perform the linking is vital to the quality of the process. As well the simple language contained in the CHQ questionnaire and other assessments intended for children can be deceptive because the items are short and seem to be clear, giving the appearance of simple linking, when in fact the linking might be very difficult. With child instruments, more so than with adult instruments, we suggest that there might be a layer of complex concepts beneath the surface of the item, and that linking in a mechanical way based on literal interpretations of items is often inaccurate. A table of the challenges that emerged from the process of linking this instrument and issues summarized in the discussion as well as solutions to these challenges are presented in table 2.

Conclusion

This paper illustrates that in linking items of an instrument to the categories of the ICF-CY there is often more to an item than meets the eye. Conversely, an instrument might probe functioning, disability and health with items that can be taken quite literally. A judgement is therefore required by those who perform the linking and this judgement will be improved by a general understanding of child health assessment and the ICF-CY. The results of this paper are intended to provide child-specific guidance to supplement the previous linking rules. These recommendations should be tested and applied to other child instruments to gather additional information about their validity and feasibility.

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Table 1: Data extraction table to determine content equivalency between items

Items with Instructions	Response Options	Perspective (functioning / disability, quality of life, impact or needs)	What is it about?	ICF Code & Category	Connectors	Annotation	
Section #1: YOUR GLUBA		Γ	Γ	I	1		
1.1 In general, would you say your health is:	Excellent Very good Fair Poor	Functioning/ disability	General health	Not-defined / general health nd-gh		Past 4 weeks	
	(Extent)						
Section #2: YOUR PHYSICAL ACTIVITIES							
The following questions of	ask about phys	ical activities yo	ou might do durin	g a day.			
2.1 During the past 4 weeks, has it been difficult for you to do the following activities due to your health problems?							
 a. do things that take a lot of energy, such as playing soccer, 	Yes very difficult Yes	Disability	 High energy physical activities 	Activity & participation d	because of	Past 4 weeks, energy, playing soccer, running,	

running or hiking?	somewhat difficult Yes a little difficult No not difficult (Difficulty)		- Health problems	Exercise tolerance b450 Not-covered: health condition nc- hc		hiking	
c. walk several blocks or climb several flights of stairs?	Yes very difficult Yes somewhat difficult Yes a little difficult No not difficult (Difficulty)	Disability	 Walking several blocks Climbs several flights of stairs Health problems 	Walking long distances d4501 Climbing d4551 Not-covered: health condition nc- hc	and because of	Past 4 weeks	
Section #3: YOUR EVERYDAY ACTIVITIES							
3.1. During the past 4 weeks, has it been difficult to do your school work or usual activities with friends because of problems like FEELING SAD OR	Yes very difficult Yes somewhat difficult Yes a little difficult No not difficult	Disability	 School work Usual activities with friends Feeling sad Feeling worried 	School education <i>d820</i> Informal relationships with friends <i>d7500</i> Managing	and because of and	Past 4 weeks	

WORRIED? 3.2. During the past 4 weeks, has it been difficult to do your school work or usual activities with friends because of problems with your BEHAVIOR?	(Difficulty) Yes very difficult Yes somewhat difficult Yes a little difficult No not difficult (Difficulty)	Disability	 Complete / finish schoolwork Accomplish activities with friends Behavior 	one's own behavior d250 School education d820 Informal relationships with friends d7500 Managing one's own behavior	and because of	Past 4 weeks
				d250		
Section #4: PAIN						
4.1. During the past 4 weeks, how much bodily pain or discomfort have you had?	Yes very difficult Yes somewhat difficult Yes a little difficult No not difficult (Difficulty)	Functioning	- Bodily pain - Discomfort	Sensation of pain b280 Sensory functions and pain, other specified b298	and	Past 4 weeks. No exact code for discomfort

4.2. During the past 4 weeks, how often have you had bodily pain or discomfort?	None to almost every day (Frequency)	Disability	- Bodily pain - Discomfort	Sensation of pain b280 Sensory functions and pain, other specified b298	and	Past 4 weeks. No exact code for discomfort
Section #5: GETTING ALO	NG					
Below is a list of items th	at describe chi	ldren's behavior	or problems the	y sometimes ha	ve.	
5.1. During the past 4 weeks how often did each of the following statements describe you?						
d. did not do what your teacher or parent asked you to do?	Very often Fairly often Sometimes Almost- never Never (Frequency)	None	Externalizing behavior as a symptom screen	code with ICD-10	and	Past 4 weeks
e. wanted to be alone?	Very often Fairly often Sometimes Almost-	None	Internalizing behavior as a symptom screen	code with ICD-10		Past 4 weeks

5.2. Compared to other children your age, in general would	never Never (Frequency) Excellent Very good Fair	Disability (development)	Behavior	Managing one's own behavior	
behavior is:	Poor			<i>azsu</i>	
	(Extent)				
Section #6: GENERAL WE	LL-BEING				
The following phrases are	e about childre	n's moods and fee	elings they may	have.	
6.1. During the past 4 weeks, how much of the time did you					
a. feel sad?	All of the time Most of the time Some of the time A little of the time None of the time (Frequency)	Disability	Feeling sad	Emotional functions b152	Past 4 weeks

Table 2: Summary table of child-specific ICF-CY content analysis

Challenge	Example Item	Solution
Linking items where the aim of the question is not apparent	During the past 4 weeks how often did you want to be alone?	Ensure that linkers are aware both of the instrument and the ICF-CY Consider the context of the item in the instrument and the development, applications and intent of the instrument when the purpose of the item is not clear
Linking items about child behavioral problems	During the past 4 weeks how often did you not do what your teacher or parent asked you to do? Would you say your behavior is excellent, very good, fair or poor?	 Decide whether the item is present as an example of internalizing/externalizing behavior for the purpose of screening or diagnosis OR Whether the item domain probes activity limitations or restrictions in the domain of behavior
Linking items about the family or items intended for caregiver proxy	During the past 2 weeks, how difficult was changing diapers?*	First decide if the item is about child or family health If the item is about child health, always link from the vantage point of the child
Deciphering the perspective of items written for children	During the past 4 weeks, how frequently have you had difficulty completing your schoolwork? During the past 4 weeks, how good or bad have you felt about	 Return to WHO definitions of functioning disability and health as stated in the ICF-CY or QOL as stated in the WHO-QOL BREF to see if they are asking children about: the frequency, severity, presence or absence, capacity, or performance of a functioning or contextual domain (funct/dis/health)

	your school work?	OR
		their subjective perception about their expectation, standards, concern and preference (QOL)
Which perspective compares the child to a reference group?	Compared to other children your own age	Comparing children to a reference group represents the disability perspective
Linking questions about child development	How is your child developing overall?	Use the category not defined: development (nd-dv)

Chapter 4

Health status and QOL instruments used in childhood cancer research: deciphering conceptual content using World Health Organization definitions

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NF devised the scientific rationale and interpretation of study results. NF performed the content analysis method with two other authors.

VS performed the content analysis method and provided input on the interpretation of the study results.

CB chose the appropriate statistical analysis and performed the statistical analysis as well as interpretation of the analysis.

AC provided scientific feedback on the rationale for the study and the interpretation of study results as well as editing. AC was the third party rater for settling disagreements.

AK conducted the literature review, scientific input on all portions of the study, performed the content analysis for negative items and provided editing feedback.

Introduction

After accidents, cancer is the most prevalent source of fatality in childhood [1]. Since the disease is now curable or treatable in 80-85% of children if diagnosed and treated appropriately [2], it has become increasingly important to capture the impact of cancer on children's lives. This impact can be assessed through various concepts including daily functioning, mental and physical health status and most importantly quality of life (QOL) [3, 4, 5]. QOL is a critical outcome to assess for many cancer therapies as well as clinical-decision making from the perspective of clinicians, children and their families. In health-care milieus, it was once believed to be sufficient to capture cancer symptoms to make inferences about the impact of cancer on a child's life [6]. Description of one's contextual and social environment, as well as daily functioning and emotions associated with the experience of cancer are now additionally important [6]. This biopsychosocial approach to assessment has the advantage of providing a more holistic understanding of a child's life, yet it also brings measurement challenges [7, 8].

One challenge associated with studying the impact of a health condition on the life of a child is related to the concepts or definitions used in measurement [9]. Instruments that espouse a health status approach according to one user might represent QOL to another [7,9]. Health status includes biopsychosocial information about health conditions, physical and emotional performance in daily life, and often social elements that facilitate or hinder health. This broad biopsychosocial definition of health is suitably guided by the International Classification of Functioning Disability and Health (ICF) or child version (ICF-CY) [10]. In the context of measurement, health status could include biological, psychological or social life domains quantified according to elements such as performance, capacity, frequency, severity, or presence.

In contrast to health status, QOL captures what is often called the subjective elements of life. The World Health Organization supports a theory of QOL which can be defined as: "the individual's perceptions in the context of their culture and value systems, in relation to their personal goals, standards and concerns" [11]. The implications of the WHO theory to measurement are that a QOL instrument must target a child's perceptions such as their personal goals, their expectations, their standards and concerns of themselves, their lives, and their future. Applying a WHO approach to QOL measurement thus requires that instruments target the child's perceptions, expectations, standards and concerns about their life/health domains irrespective of their actual health state. The WHO definition of QOL is interpreted to indicate that one

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must include the valuation of the person/child being assessed from *their point of view* in order to measure QOL properly. Consulting children during the content development stage of a QOL instrument can help address this issue.

The challenge for researchers and clinicians who wish to choose a questionnaire to measure the impact of cancer on a child's life is to decide whether they are interested in health-status or QOL. Using these terms interchangeably when they do not reflect the same concepts can lead to improper instrument use [7,9]. The use of instruments that do not measure what one intends to measure results in poor content validity [12, 13] which leads to confusion about how to interpret the results of research. For example, if the health status of a child does not improve following chemotherapy (eg: due to fatigue or discomfort) but the child values the impact the therapy has made on their life, the child's QOL has improved but NOT their health status. If one uses an instrument that emphasizes a health-status approach to measure QOL, they would conclude that the chemotherapy did not change QOL when in fact the child's health status is what did not change.

Although there is acknowledgement among researchers that the same instruments are often used for different applications, there is little consensus about how to address this issue of whether an instrument will actually evaluate health status or QOL. Both concepts measure health and related content including *biological*, *psychological* and *social* life. It is how these domains are measured; the *perspectives or approaches* with which the domains are measured that differ. Thus the distinction between health status and QOL is found in *how* something is measured (eg: sense of satisfaction versus actual performance) in addition to *what* is measured (eg: bio-psychosocial content).

The ICF-CY is a uniform terminology and classification system developed by the WHO that can classify biopsychosocial health content found in health status *and* QOL instruments [10]. The ICF or ICF-CY has been used to delineate the content of health and QOL instruments in over 57 studies to date using a standardized method of content analysis [14-17]. Linking information to the ICF or ICF-CY demonstrates a systematic process for showing what content is available in an instrument relative to the ICF and thus a WHO definition of biopsychosocial health (eg: body functions, body structures, activities and participation, environment and personal factors). Recently, this standardized method of linking questionnaires to the ICF or ICF-CY was updated to decipher health status from QOL approaches based on the perspective found in a

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questionnaire's items. Describing the biopsychosocial health content of health and QOL questionnaires using ICF-CY classification also remained an important part of the content analysis.

In addition to conceptual confusion about what instruments actually assess, the content of instruments for children should address the already compromised psychological state children might be experiencing when completing self-report questionnaires. Many of the most commonly used self-report health status and QOL instruments administered to children were developed for use in a clinical care context. An unanticipated consequence of a clinically oriented approach has led to the development of questionnaires that emphasize problems or negative aspects of life since the focus of clinical life is to improve upon the areas in which children experiences difficulty. Unfortunately, this problem-based approach to questioning children in self-report instruments can have implications regarding how they understand themselves and their condition and these issues should be examined further [18].

Although there are many reviews of QOL measures used in childhood chronic conditions [19-27] and to a lesser extent cancer [28, 29], the reviews on QOL in child instruments have focused on psychometric properties, general conceptual review or content development. Systematic appraisal of instrument content in the childhood cancer literature designed to address conceptual confusion between health and QOL is not in our knowledge available, while one review article discusses the issue of negative questioning for children [18].

This study has the following two objectives: 1) to decipher the perspectives as well as the health and related content for the most commonly used instruments assessing health status or QOL for children with cancer by using WHO definitions and ICF-CY classification, and 2) to analyze systematically the extent to which problem-based and negative questioning is present within those instruments. The first objective will be accomplished by analyzing the instruments' perspective based on WHO definitions of health, disability, functioning and QOL and also linking the health content for each item from the included measures to the ICF-CY. The second objective will be accomplished by completing an analysis of negative phrasing and content found in the included instruments.

Methods

Systematic Review

MEDLINE, CINAHL, EMBASE, PsycINFO, Cancerlit and Sociological Abstracts were searched from the inception of each database to 15th June, 2009. The search strategy, developed by a medical research librarian, involved searching for a subject heading or text word for the following terms: "quality of life", "health related quality of life", "quality adjusted life years", "health status", "functional status", "well-being", "patient reported outcome" along with specifically named generic and cancer-specific QOL questionnaires known to the research team (search strategy available upon request). These terms were combined with a subject heading or text word for cancer, and limited to children aged 0-18 years. No limitation was made on language. Included articles described the development, psychometric evaluation or use of a generic or cancerspecific health status or QOL questionnaire developed for use with children with cancer or childhood cancer survivors. Articles that included guestionnaires that focused exclusively on one specific health domain (e.g., fatigue, eating disorders, pain) were excluded. Articles that did not provide evidence of any development or validation process (i.e., ad hoc instruments) were excluded. Articles that described instruments developed for adult cancer patients were excluded unless the authors had validated the instrument in a sample of childhood cancer patients.

The titles and abstracts of all identified articles were examined independently by two reviewers. The full text of all potentially relevant papers was obtained and examined by two reviewers. Citations for included articles were examined to identify additional articles. For the purposes of this paper, the three most frequently used generic and the three most frequently used cancer-specific questionnaires were included in the analysis to delineate content issues for the most popular multi-dimensional patient reported instruments employed in the literature.

Overall, 125 articles employed 12 generic instruments and 14 cancer specific instruments reported in the reviewed literature. The most common measures were divided into a generic instrument category and a cancer-specific instrument category. The top three most frequently employed generic measures in the literature reviewed were the Child Health Questionnaire (CHQ) [30], the Peds QL [31,32], and the Health Utilities Index III (HUI-III)[33-34]. The most frequently utilized cancer-specific measures included the University of Minnesota Minneapolis-Manchester QOL Survey of Health

(MMMQOL) [35,36], the Peds QL Cancer[37] module and the Pediatric Oncology Quality of Life Scale (POQOLS) [38]. The frequency with which the measures were identified from the systematic literature search can be found in Table I. If any of the instruments published a child self-report in addition to proxy report, both versions were checked for inconsistencies in content on an item by item basis.

Overall procedure

Each of the six questionnaires that met the inclusion criteria were placed in an extraction matrix developed specifically for reviewing content and perspectives of health-status and QOL instruments. The matrix organized information about the perspective of the question or item, the health or health-related domain expressed in the question (i.e., the content), as well as the way the question or item was quantified or operationalized (e.g. extent of satisfaction, frequency, severity, etc.). The analysis was completed by NF and VS using an iterative (six step) process. In step one, analyzers NF, VS & AK met to review the purpose, background and typical applications of the six instruments so that the context in which instruments were developed could be reviewed. Secondly, the perspective was cooperatively extracted on an item by item basis by NF, VS & AC. Thirdly, the health or related information expressed in each item from each questionnaire was extracted (NF & VS). Details about how the perspectives and health and related content in steps two and three were extracted are described in the following sections.

In the fourth step NF and VS independently linked the information about the health or health-related domain to the ICF using the standardized linking rules [15,16]. The results from this independent linking stage were set aside for the reliability analysis. The fifth step was a summary of the dominant perspective (e.g., health-status vs. QOL) found in the measure as well as a consensus list of health or related content in the form of body functions, body structures, activities and participation, environmental or personal factors according to the ICF-CY. The sixth and final step was an analysis of the negative and positive information expressed in each instrument on an item by item basis by NF and AC.

Perspectives

Perspectives were extracted for each instrument item for the purpose of drawing conclusions about whether the instrument reflected QOL, Health (including disability/functioning as defined by the ICF-CY) or other approach (Table II). The

perspectives were assessed on an item by item basis for each instrument according to an updated draft of the linking method originally proposed by Cieza, et al. [15, 16]. This method used WHO definitions of QOL, disability, functioning, environmental facilitators and environmental barriers from the manuals of the WHOQOL-BREF and the ICF-CY to extract the *perspective* that each questionnaire item reflected. For example: an item was deemed to carry a QoL approach if an individual's perception of life or a life domain (e.g. as operationalized through 'importance', 'satisfaction', 'feelings about'), was the target of the item. The item: "I feel satisfied with my ability to manage my behaviour" therefore reflects a QOL approach whereas the item "I have difficulty managing my behaviour" reflects a disability (health) approach (Figure I).

Additional sub-perspectives of functioning, disability and health such as *impact*, *needs* or *norms* were empirically extracted from measurement approaches found in published instruments in the literature (in progress). Examples of assigning perspectives using example items from the MMMQOL, the CHQ and the POQOLS are shown below:

MMMQOL: I am happy with my life in general	\rightarrow QOL
MMMQOL: I cannot do as many activities because of my health	\rightarrow Disability
MMMQOL: I have a lot of energy for running or sports	\rightarrow Functioning
MMMQOL: My parents are usually patient with me	→ Facilitators / Barriers
POQOLS: My child has needed extra help with daily living skills	→ Needs / Assistance
CHQ: During the past 4 weeks, how often has your health or behaviour	\rightarrow Impact
interrupted various everyday family activities?	
CHQ: Compared to other children your age, in general would you say	→ Norms / Development

your behavior is excellent, very good, fair, poor

Items with no perspective such as: 'what is your sex?', were excluded from this perspective portion of the analysis even if they were included in the health domain content analysis.

Health and related domains: Translating content into ICF-CY categories

The content of the items was translated to the ICF-CY and grouped according to the components of the classification system using standardized linking rules developed and tested explicitly for analyzing the content of health instruments [30,31]. The ICF-CY classification scheme represents a variety of health and health-related domains that can be accessed on line at http://apps.who.int/classifications/icfbrowser/. Raters completed the item by item content analysis using a process inspired by a descriptive qualitative approach based on naturalistic observation. Sandelowski describes this process as 'staying close to the surface of the language' [39]. The raters cooperatively identified the health and health-related domains within each item prior to independently assigning it a specific alpha-numeric ICF-CY code. The first letter of the code represents the ICF-CY component of body functions (b) or structures (s), activities and participation (d), or environment (e). The following digits of the code were numeric. For example, item 9.1b from the Child Health Questionnaire: "During the past 4 weeks, how often has your health or behaviour interrupted various everyday family activities?" yielded the domains: health, behaviour and everyday family activities. An example of the linking for the domain 'behaviour' can be found in figure I. Once a health or related domain was linked to the ICF-CY by an individual rater (NF or VS), it was ready for the reliability analysis.

Concepts that were too general to be assigned to a code but still suited the concepts contained in the classification were assigned the code *nd* (*not defined*). For example, the concept 'general mental health' is part of the conceptual underpinnings of the classification but does not have a specific code. General mental health was thus assigned the code *nd-gmh*. Concepts that were not covered by codes in the classification or included as part of the conceptualization of the ICF-CY were assigned the code *nc* (*not covered*). The code *pf* was used to denote *personal factors*. All concepts that were labeled with ICF categories were grouped into the components as described above. The composition of health contents was determined for each instrument based on the proportion with which it contained these concepts.

Reliability analysis

Initial agreement between the two independent content analyzers (NF and VS) was computed using agreement statistics for categorical data using Kappa statistics and percentage agreement. Work completed on the best methods for calculating agreement in ICF linked data [40] indicates that the assumption of independence is violated for ICF

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linked data between raters on the same content. It also concludes that ICF data tend to show a skewed distribution, a bias which is most properly corrected using bootstrapped confidence intervals.

Percentage agreement is an appropriate statistical estimate of agreement between raters linking to the ICF while kappa is calculated for the purpose of creating a confidence interval with which to estimate error about the agreement co-efficient.

Negative content and phrasing

An additional dimension of the item-level content analysis, which is not part of the standardized method, was employed to identify the amount of negative information included in each questionnaire (NF & AK). This information cannot be captured by the ICF-CY because the domains from the classification are framed in a neutral way. Each item from each measure was analyzed individually and judged (yes/no) as having negative phrasing and/or negative content. If a question probed unpleasantness, disagreeableness, gloominess, pessimism, disfavour, detriment, hostility, disparity, or maliciousness as a result of phrasing (eg: "problems with, cannot do, difficulty with"), then it was labelled as containing negative phrasing.

A question was decidedly negative in terms of content if the main topic of the question was inherently unpleasant, disagreeable, gloomy, pessimistic, disfavourable, detrimental, hostile, disparaging, or malicious. Domains such as suicide, lying, stealing and problem behaviour were examples of content that were deemed negative. The proportion of negative to positive/neutral phrasing and content was tabulated for each measure. This information was collected to provide measure users with information about the possible impact completing the measure might have to children and their families.

Results

A summary of the characteristics of the included instruments is found in Table II.

Perspective

Overall, six instruments included in this analysis have perspectives weighted towards WHO definitions of functioning, disability and health according to the ICF-CY as opposed to QOL as defined by the WHOQOL initiative (Table III). Two questionnaires: the generic CHQ-PF87 and the cancer-specific MMMQOL Survey of Health contained 15% or greater proportion of questions that represented a QOL perspective. The questions from each of the remaining questionnaires contained less than a 15% QOL perspective.

Health and related content as represented by the ICF-CY

The content derived from all the generic instruments emphasized firstly the ICF-CY components *activities and participation* followed by *body functions* (Table IV). *Environment* components of health such as family or medical supports were negligibly represented in the CHQ and HUI-III relative to the total number of questions in the questionnaire.

The health domains emphasized by the cancer-specific measures were more varied (Table IV). The Peds-QL Cancer module emphasized environment components of health by virtue of the proportion of content about medical treatments and health professionals, followed by body functions content relating to side-effects of cancer and cancer treatments. The major ICF component emphasized in the MMMQOL Survey for children 8-12 concerned body functions such as b280 pain, b130 energy and drive and b152 emotional functions whereas the 13-20 version emphasized elements of activities and participation associated with socializing (e.g. d7200 Forming relationships). Both versions of the MMMQOL Survey contained a third or more questions pertaining to personal factors. The personal factors emphasized broad emotional traits such as loneliness or feelings of exclusion as opposed to specific emotional functions that can be impaired, limited or delayed from the *body functions* component of the ICF-CY. Finally, the POQOLS emphasized a variety of activities and participation domains such as recreational (d920 Recreation and leisure), sports activities (d9201 Sports) and socially based activities (d7 Interpersonal interactions and relationships) followed by body functions domains such as b152 Emotional functions, b130 Energy and drive and b280 Pain. Specific ICF-CY categories for each of the instruments are available upon request.

Reliability

Agreement statistics showing percentage agreement and kappa with bootstrapped confidence intervals for the ICF-CY linked data are shown in Table V. Interrater agreement fell between 0.6-0.8 using Cohen's kappa indicating moderate-good agreement.

Negative content and phrasing

Negative content varied greatly between measures from less than 1% in the POQOLS to 67% in the PedsQL Cancer Module (Table VI). Concepts such as pain, stealing, causing family tension, anger or teasing from other children are examples of negative content that appeared in the instruments.

The use of negative phrasing such as 'Do you have difficulty with?' ...or 'Problems with?.'..ranged from 0.33-100% across the instruments. Most of the questionnaires used combinations of negative neutral and positive phrasing techniques to cue respondents. Two extreme examples included the POQOL Scale, which used almost entirely neutral phrasing and the PedsQL, which consistently cued respondents to answer about health based on 'Problems with'.

Discussion:

A comprehensive and systematic content analysis of the most commonly employed generic and cancer-specific health status and QOL instruments were conducted for this review. Three criteria were described: the perspectives of the instruments (i.e. health status vs QOL), the health or related content (according to ICF-CY) and the negative phrasing and content of the questions in the instruments. The analysis showed that according to these criteria no one instrument demonstrated an ideal balance of content characteristics relative to the evaluated criteria and thus each one must be considered carefully relative to one's particular research or clinical evaluative purpose. A most notable finding is that the most popular instruments in childhood cancer favour a health status approach over a QOL approach in contrast to their mentioned use in the studies from which they were identified. Although most of the instruments were developed prior to the introduction or dissemination WHO concepts, there is a need to apply standard definitions to delineate what can actually be evaluated using such instruments.

The WHO defines QOL as a self-perceived construct including physical, emotional and social functioning of the person [11]. While many researchers and clinicians understand that QOL must contain these multi-dimensional domains of life, measurement activities in the field of childhood cancer have neglected to demonstrate how to distinguish health status from QOL. This confusion might be related to the overlap in content that is conceptually relevant to health while at the same time showing relevance to QOL. For example, internal psychological and physical states, the

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performance of life roles and the social environment of the individual can be important to both health status and QOL instruments. If one accepts the notion that it is the child's *perception* of his or her own life which differentiates health from QOL, then this perceptual element must be targeted in the questions. QOL perspectives using a WHO definition were assessed far less frequently relative to other health perspectives for all of the measures reviewed.

Imprecision of the concepts one wishes to measure has an impact upon content validity. Content validity requires representativeness of the concept one wishes to measure as well as overlap between measurement targets and measurement instruments [12]. For example, our findings reveal that the PedsQL Cancer module and the POQOLS instruments showed a relatively balanced distribution of questions across the ICF-CY health components of *body functions, activities and participation,* the *environment* and *personal factors.* In employing these instruments, one is more likely to demonstrate representativeness of health status in the ICF-CY conception. However, in combining this information with the perspective findings, we conclude that in using such instruments, one could assess the extent to which issues in these health domains are present or absent without learning much about a child's personal sense of these domains impact upon their life.

Qualitative methods are often recommended to explore a depth of understanding about children's personal perspectives and how health conditions can impact upon individual lives [5, 41]. However, quantitative instruments are tools that can assess the QOL of *many* children so that conclusions about the effectiveness of cancer therapies or treatments can be made. In theory, health status instruments will not be responsive to situations where children experience improvements in their outlook on life as a result of interventions while simultaneously experiencing few improvements in symptoms or disability as a result of cancer treatment. Although qualitative methods can help to explain this discrepancy between what disability instruments measure, and what children actually report during clinic visits or upon interview, they are not always available, appropriate or convenient for evaluating effectiveness on a larger scale. Thus the need for instruments that accurately probe a QOL versus a health status perspective using WHO definitions are needed [9].

Another issue pertaining to the content of childhood QoL measures is exposure to negative content and negative phrasing or questioning for the children and proxies to whom the questionnaires are administered. Including negative content from

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instruments can create psychological burden for respondents [18] and evokes an ethical dilemma for those who need to capture the negative as well as positive factors associated with childhood cancer. Including negative content is often necessary to target important concepts such as pain. Regardless, users of patient-reported instruments should be aware of the extent to which negative content is represented in a questionnaire so they can make balanced choices in assessing the experience of cancer [42, 43].

The issue of negative phrasing is complex because phrasing plays a critical role in questionnaire development, specifically item construction. For example, negative phrasing can be used for reverse scaling purposes to ensure that respondents are answering in an internally consistent way [43]. Measure developers and researchers must also consider that the questions posed in self-report measures shape the responses and the underlying cognitive thoughts one must call upon to answer such questions [43]. Asking about the extent to which a child experiences impairment, activity limitation and participation restrictions will therefore inform one's understanding of their disability while asking neutrally or positively about their performance or abilities will inform one's understanding of their functioning.

The method of content analysis used in this study was intended specifically for health and health-related information from patient-reported outcomes such as health and QOL questionnaires. The definitions used for classifying perspectives were based in the authors' understanding of WHO terminology from the WHOQOL manual and the ICF-CY standard classification document. WHO terms and classifications were used because they are rigorous, developed with multiple inputs and stakeholders and have the potential to evoke consensus among those concerned with health and QOL [10, 11]. This updated version of the Cieza et. al linking rules requires further empirical testing to determine its validity across a variety of patient outcomes including technical instruments and individualized measures, which were not included in this study. Since this method involves linking to the ICF-CY, which is based on mapping the health information of the individual, the method cannot be used to appraise instruments of family functioning. Finally, it is important to note that the analysis contained here is limited specifically to the content of instruments without discussing psychometric properties, which can be found in other reviews. Added quantitative dimensions of instruments such as the preference scores of the HUI III were not assessed for their contribution to health status versus QOL perspectives analysis.

The agreement between the initial rating of the ICF-CY categories assigned to health domains fell consistently in the 'moderate-good' range without reaching the excellent range of agreement. This range of agreement is commonly reported for studies that link health information to the ICF or ICF-CY [44, 45] and shows the importance of linking with more than one individual in an iterative way so that multiple points of view on the content of the instruments can be considered in the analysis. Although it is a goal of measurement that individuals understand and interpret the concepts from measures in a similar way, it is not the reality that questions from health status and QOL instruments are interpreted identically between individuals, let alone relative to the ICF-CY. Thus a 'good' or 'substantial' agreement represents a more realistic representation of how questions from QOL measures are viewed between individuals.

Conclusion

Measuring health status and QOL is important for understanding the impact of cancer in the lives of children. Inconsistencies in definitions or conceptual understanding of QOL have led to confusion about how to select or use instruments designed to measure this multidimensional concept. WHO definitions are consensus driven and can serve as the basis for content appraisal of different instruments. Our analysis of commonly used generic and cancer-specific instruments in childhood cancer research showed that the popular measures used to assess QOL reflect a variety of health and health-related domains but consistently use a health status approach as opposed to a QOL approach. The use of negative language and phrasing should also be considered by developers when revising instruments and by researchers who administer instruments. These findings should be consulted to determine content validity when selecting instruments for any particular research or clinical evaluation purpose.

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Table I: Definitions of WHO Perspectives and empirically-derived perspectives from theCieza et al updated linking rules

Perspective name	Perspective definition
QoL	QoL perspectives seek to determine individuals' 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.
Health (Functioning)	Functioning perspectives refer to the interaction or the individual components of body functions, activities and participation
Health (Disability)	Disability perspectives probe impairments, activity limitations and or participation restrictions
^A Needs/Assistance	Needs perspectives assesses an individual's functioning or disability based on the needs that they draw from their social, physical and institutional environment
^A Impact	Impact perspective infers an individual's functioning or disability based on the effect that they have on their social, physical and institutional environment
^A Development/Norm s	Developmental perspectives contrast an individual's functioning or disability with that of another reference individual or group
Environmental (Barriers)	Barrier perspectives reflect environmental factors that hinder the functioning of an individual
Environmental (Facilitators)	Facilitator perspectives reflect environmental factors that promote or allow for the functioning of an individual

^ADenotes the perspective is empirically-derived from the inductive analysis of multiple instruments (reported in the Cieza et al updated ICF linking rules) but not official WHO definitions

Figure 1: Different perspectives for the same health domain (ICF-CY category)



Table II: Characteristics of included measures

Measure Name	Measure Type	Child report available?	Population used for early measure development or testing	Domains / Factors / Subscales / Attributes studies)
Child Health Questionnaire (CHQ-87)	Generic	yes	Once developed based on literature review and theoretical work in child health, the CHQ-CF 87 was tested among 300, 10-15 year old children from a middle-school in northeastern USA; 54, 9-16 year old American children with ADHD; and 20 children aged 10-19 with end stage renal failure on hemodialysis between 1992-1996.	 Physical activities Role limitations (emotional) Role limitations (behavioral) Role limitations (physical) Bodily Pain Behaviour Mental health Self-esteem General health perception Change in health Family Activities Family Cohesion
Health Utilities Index III (HUI-III)	Generic	no	Developed from the HUI II (in which items came from HUI for infants & were modified with literature and 84 parent-child pairs) and first implemented in the 1990 Statistics Canada Ontario Health Survey & the 1991 Statistics Canada General Social Survey for	1. Vision282. Hearing3. Speech4. Ambulation5. Dexterity6. Emotion7. Cognition8. Pain

			the general Canadian population			
PedsQL 4.0	Generic	yes	961 children ages 5-18 and 1629 parents of children 5- 18 from pediatricians' offices, hospital specialty clinics: orthopedics, rheumatology & diabetes as well as in and outpatients from San Diego Hospital in USA	1. 2. 3. 4.	Physical functioning Emotional functioning Social functioning School functioning	37
University of Minnesota Minneapolis- Manchester QOL Survey of Health (MMMQOL)	Cancer- specific	yes	397 healthy, on or off treatment adolescent cancer patients Acute lymphocytic leukemia Hodgkin's & Non- Hodgkin's lymphoma from 9 institutions in USA	1. 2. 3. 4.	Family Dynamics & Outlook on Life Physical Symptoms Physical Functioning Psychological Functioning	6
Peds-QL 3.0 Cancer Module	Cancer- specific	yes	291 pediatric cancer patients with acute lymphocytic leukemia Hodgkin's & non- Hodgkin's lymphoma brain tumor and to their parents from Southern California USA	1. 2. 3. 4. 5.	Disease and treatment- related symptoms Physical functioning Psychologic functioning Social functioning Cognitive functioning	^A 20
Pediatric Oncology Quality of Life Scale POQOL	Cancer- specific	no	317 Parents of children with cancer outpatient oncology clinics and inpatient medical units in Florida & Atlanta USA.	1. 2. 3. 4.	Physical Function Role Restriction Emotional Distress Reaction to Current Medical Treatment	7

	Main diagnoses were	
	leukemia sarcoma or	
	Hodgkin's disease	

^AIncludes use of the earlier version of the PedsQL Cancer Module: PCQL

Table III: Proportion of perspectives (%) found within each measu

	Proportion (%) of Perspectives							
Measures	QOL	Health (disability & functioning)	Environmental (barriers & facilitators)	Norms or Development	Needs or Assistance (of child from the environment)	Impact (of child on environment)	Unable to Determine	Dominant Perspective(s)
CHQ-87	20.4	54.4	1.1	2.2	0	6.8	14.8	Health with QOL sub- component & other mixed approaches
HUI III	2.2	97.6	0	0	0	0	0	Health
Peds QL 4.0	4.3	78.2	8.6	8.6	0	0	0	Health with environmental & developmental sub- component
MMMQOL (8-12)	16.1	74.1	6.4	3.2	0	0	0	Health with QOL sub- component
MMMQOL (13-20)	17	76.5	4.3	2.1	0	0	0	Health with QOL sub- component
Peds QL	4	96	0	0	0	0	0	Health

Proportion (%) of Perspectives								
Cancer (toddler, child)								
Peds QL Cancer (older child, teen)	3.8	96.1	0	0	0	0	0	Health
POQOL	5	80	0	0	15	0	0	Health with a needs sub- component

Measures	Body functions	Activity & participation	Environment	Personal factors	*Not defined	**Not covered	Dominant ICF-CY Component(s)
CHQ-87	26	36	2	16	8	12	Activity & Participation
HUI III	33	58	0	0	0	1	Activity & Participation
Peds QL 4.0	32	50	10	4	0	4	Activity & Participation
Peds QL Cancer (toddler, child)	29	13	33	23	0	2	Environment
Peds QL Cancer (older child, teen)	27	18	31	22	0	2	Environment
MMMQOL (8-12)	39	12	8	29	4	8	Body Functions
MMMQOL (13-20)	19	30	4	30	3	15	Activity & Participation
POQOL	27	33	18	15	0	6	Activity & Participation

Table IV: Proportion of Health and Related Domains as Represented by ICF-CY Components (%)

*Not defined are codes that represent general concepts such as general physical health or general mental health that are part of the ICF-CY but are too broad to be assigned to a specific component

****Not covered** are codes that represent concepts that are not included in the ICF-CY such as diagnosis

	Percentage Agreement (%)	Kappa Coefficient	Bootstrapped Confidence Interval
Child Health Questionnaire	69.43	00.68	0.60-0.75
Health Utilities Index III	68.29	00.67	0.59-0.75
Peds QL	62.07	00.61	0.46-0.81
Generic Combined	68.28	00.67	0.62 - 0.73
Peds QL Cancer Module (all age versions combined)	74.07	00.71	0.58-0.83
University of Minnesota Minneapolis-Manchester QOL Survey of Health (all age versions combined)	84.80	00.83	0.75-0.90
Pediatric Quality of Life Scale	62.50	00.60	0.43-0.79
Cancer-specific Combined	78.67	0.77	0.71- 0.82

Table V: Initial agreement between raters of ICF categories in the first round of content analysis

	NI	N
	Negative content	Negative phrasing
Child Health Questionnaire-87	33.00	39.08
Health Utilities Index III	12.50	47.50
Peds QL 4.0	23.00	100.00
Peds QL Cancer Module (toddler, child)	66.66	100.00
Peds QL Cancer Module (older child, teen)	59.26	100.00
University of Minnesota Minneapolis- Manchester QOL Survey of Health (8-12)	32.25	31.01
University of Minnesota Minneapolis- Manchester QOL Survey of Health (13-20)	19.15	34.04
Pediatric Oncology Quality of Life Scale	00.33	00.00

Table VI: Percentage of questions with negative phrasing or content

Chapter 5

Health status instruments

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NF wrote this chapter according to the book format requested by the editor and conceived of the relevant sub-headings and framework to review relevant issues for child health status instruments.

VS assisted with the linking of these instruments.

AK provided scientific and editorial feedback on all sections of the chapter.

What is the Construct?

In 1948, the WHO defined health as "a state of complete physical, mental, and social well-being and not merely the absence of disease or infirmity" [1]. While this often quoted definition clarifies that health is much more than not being ill, on its own it fails to offer clarity or consensus about what aspects of a person's life *should* be included in the concept of health. Lack of consensus is problematic because a universal concept of health is needed to describe and to evaluate the needs of children all over the world. Ironically the original purpose of the 1948 definition was to provide such a definition.

Despite this confusion, the concept of health is viewed internationally as a resource for well-being and positive living [2, 3]. The productivity and potential of entire nations is determined at least in part by the health of their children [3]. Furthermore, the efforts of parents, communities and health providers are judged on the basis of improvements to child health. Health status instruments are multidimensional tools that can provide a snapshot of a child's health at any given point in time, but what definition of health do these tools reflect? The answer is that, however implicitly, instruments reflect the developers' ideas of health contextualized to the time, place and purpose for which the instrument was developed. Thus some historical background to the measurement of health provides a useful starting point to a consideration of health status instruments.

Since the advent of germ theory, health has been expressed using an absence-ofdisease approach useful in public health contexts. As early as the 1700s, public health proponents were interested in measuring the pattern of outbreaks and later the effects of clean water and community hygiene practices [2, 4]. At that time health indicators, such as the incidence and prevalence of diseases like cholera and dysentery, were useful because they showed the proportion of individuals with infirmity in the population. Since the development of antibiotics and vaccination, the incidence and prevalence of infectious disease has declined (at least in the developed world) [4]. Health indicators shifted their focus from prevalence and incidence to include symptoms for describing children and adults who were living with disabilities or morbidity as a result of extended illness. Symptom checklists were developed to measure pain or discomfort from the perspective of both the child and parent [5]. As many sources of mortality and morbidity of childhood have become treatable, questionnaires were developed to assess the experience of childhood chronic health conditions or disability and as indicators of health status.

More recently, a measurement approach has been developed based on child and parent report (otherwise known as patient-reported outcomes (PROs). PROs are any reports of the status of a patient's condition that come directly from the patient without interpretation by anyone else [6]. PROs measure concepts such as symptoms, satisfaction, health status and health-related quality of life. In order to capture PROs effectively, well-defined, reliable and valid instruments are needed. Such instruments are typically made up of multiple scales, which reflect the key aspects of a conceptual framework.

Health and the ICF/ICF-CY

Simultaneously with this shift towards measuring PROs over the last 20-30 years came a broadening of the definition of health. In the mid-1970s, Pless and Pinkerton described a non-categorical (i.e. non diagnosis-based) approach to conceptualizing health status [7], which shifted the emphasis of measurement away from diagnosis and onto various psychosocial indicators of health. Non-categorical thinking, combined with an increasing recognition of the importance of the child's daily functioning, has helped to prepare the clinical and research communities for a concept of health based on the International Classification of Functioning, Disability and Health (ICF) and more recently, the children and youth version (ICF-CY) [8,9].

The ICF-CY espoused a biopsychosocial approach to health, which has certain advantages to the measurement of health status. First, it created a definition of health based on the interaction of biopsychosocial factors (*body functions, body structures, activities and participation*), which occur in a context of the *environment* and *personal factors* [7,8]. The advantage of the ICF-CY definition of health is the inclusion of what constitutes health as opposed to a definition based on the exclusion of what health is not (i.e. the absence of disease). Second, the ICF-CY definition is accompanied by a classification that specifies details about the factors that compose health using a standard document. The advantage of the ICF-CY classification is that it serves as a taxonomy upon which health status instruments for children can be based (a practice that will also be applied to the measures we review in this chapter). Finally, despite similarities or differences in existing health status instruments, the ICF-CY taxonomy can be used to clarify the content of health status instruments so clinicians and researchers can select instruments that describe and compare the areas of importance to a given time, place and purpose within a biopsychosocial approach to health.

How is patient-reported health status unique relative to QOL?

As questionnaires are developed that include biopsychosocial outcomes of interest to children and their families, there is growing confusion about what distinguishes a health status instrument from other questionnaires that are called quality of life instruments (QOL) or health-related quality of life tools (HRQOL) [10]. Health status includes multidimensional information about health conditions and the ability to perform physically and emotionally in daily life. Ideally, in ICF-CY terms, health status includes the interaction of individual components of body functions, body structures, activities and participation, which occur in a context of environmental and personal factors. Given that these components span the entirety of the experience of life, it is difficult at first glance to see how health status is different from QOL.

Both health and QOL include factors pertaining to *biological, psychological* and *social* life. The distinction is in *how* these domains are measured. Chapter 34 by Colver on QOL outcomes specifies that it is the *subjective* element, which is common to all QOL outcomes and that makes QOL different from health status. Consistent with the WHO definition, the primary focus of QOL rests on the "individual's perception of his or her position in life" [11], with *perception* as the key word. Thus to measure QOL, the child's or parent's perceptions and values, such as satisfaction, importance or meta-appraisals about a biopsychosocial areas of life are the central/intended target of measurement (signal), while the actual presence, absence, severity, intensity or difficulty with the biopsychosocial area of life is secondary or unintended (noise) (see Figure I). Social life, for example, is a component that is relevant to the health of school-aged children and adolescents and hence addressed by most health status instruments in some way. The question: 'Does your child have difficulty getting along with other children?' represents a health status approach, whereas the question: 'Are you satisfied with your child's ability to get along with other children?' reflects a QOL approach.

Thus the concept of health status is distinct from QOL because it seeks to distinguish differences and commonalities in the experience of health for children based on health indicators that are common between children as opposed to individual or personal *perceptions* of those differences and commonalities. Although these distinctions can be made conceptually, deciding on whether an instrument reflects a health status or QOL approach requires some insight on the part of an instrument user.

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Most health status instruments were developed before these conceptual distinctions were made clear in the literature. This chapter describes considerations in the selection of instruments that are mainly described as health status instruments while recognizing that many contain mixed elements of health status and QOL. The chapter is also intended to provide instrument users with basic criteria with which they can appraise instruments of interest for their respective purposes assuming one is interested in measurement using a biopsychosocial definition of health such as is found in the ICF-CY.

We have selected for review in this chapter six popular instruments used to measure health status in children (see Figure II). Two of the six instruments reflect a health economics perspective. In order to appropriately capture PROs, well-defined, reliable and valid health status instruments are needed. In the next section we describe general factors to consider when choosing a health status instrument.

General factors to consider when measuring this domain

Content validity and content overlap

Content validity is an important measurement property that clinicians and researchers must consider when choosing an instrument [12]. Unfortunately, content validity is commonly overlooked and, once compromised, can lead to a false conclusion that vital outcomes were not affected by an intervention when in fact a change occurred. For example, an instrument that focuses on cognitive problems (e.g. attention, planning, mental flexibility) might not detect changes resulting from an educational intervention for a child with learning disability whereas an instrument focused on school performance detects the improvement based on the child's success in compensating for his/her cognitive issues. Instrument users are often satisfied to choose a so-called 'gold standard' instrument to assess health status; unaware that content validity is not a property of the instrument itself, but of the fit between an instrument and the purpose for which it is intended [12]. One can only say they expect to achieve content validity if the outcomes that are an issue for the children of interest are articulated by the chosen instrument in a comprehensive way. The basic issue is: What is the question for which any particular assessment tool is being selected?

Often clinicians and researchers are well aware of the aspects of health that they believe are important to the children and families they serve. From the perspective of body functions, these components could include emotional functions, pain, sleep, and specific aspects of cognition (e.g. memory and attention). Activities and participation

components can vary greatly depending on age and developmental level but typically include play, sports, learning and applying knowledge (e.g. school performance), mobility and self-care. Aspects of the environment that are important to child health, such as social support, the attitudes of others and the availability of educational, social and health services are often important. All of these health components are articulated in the ICF-CY classification. Thus the ICF-CY can serve as a standard tool by which clinicians and researchers can define their outcomes of interest, relate them to the classification system and, to ensure content coverage, seek instruments that ask children and families about those specific health outcomes.

Most of the instruments reviewed below were developed prior to the introduction of the ICF-CY. We performed a post-hoc analysis of the content of each instrument by linking each questionnaire item to the ICF-CY classification [8] in order to understand how the content maps onto a broad definition of health. Tables I through III provide information about the health domains that are covered by each instrument according to the ICF-CY components of body functions, activity and participation and environment. Table IV shows content that is consistent with personal factors which do not have specific ICF-CY categories; broad health concepts that are conceptually part of the ICF-CY but not specific enough to fit under an ICF-CY component 'not-defined' and concepts that are included in the instruments but not covered conceptually by the ICF-CY or using any specific ICF-CY category 'not-covered'. These tables are presented so that researchers and clinicians can determine for themselves whether content coverage is present for each of a number of commonly used instruments and the ICF-CY domains covered by each instrument.

Determining content coverage is a relatively simple step that instrument users can perform prior to selecting a tool. Publications are available that have systematically linked existing child self-report instruments to the ICF and the ICF-CY in addition to the instruments that will be reviewed in this chapter [13-16]. Users can refer to such reviews in addition to reviews of psychometric aspects of developed instruments [17-23] prior to deciding on a health status instrument.

Evaluation versus description

Evaluative instruments are tools that should be validated as being sensitive to changes that occur as a result of interventions or significant life events as well as development. If health status instruments were cameras, ideally they would have quick shutter speed so that they could capture even small change to an image between

consecutive photographs. When selecting evaluative instruments, one should consider which aspects of a child's health are expected to change following an intervention and determine whether there are questions representing those aspects of health in the instruments. In psychometric terms, this concept is described by Majnemer in Chapter 1 as responsiveness.

Guyatt and colleagues [24] described the issue of responsiveness in clinical research as well as methods for appraising responsiveness. The authors report a variety of statistical approaches (e.g. effect size, standard error of measures or SEM, Guyatt's responsiveness index, and receiver operating characteristic or ROC) that can be used as indicators of responsiveness. Conversely, they caution against over-reliance on statistical indicators to decide on responsiveness because the evaluative power of an instrument must also tap into content vital to represent the construct (in this case health status) for it validly to be measuring what it is intended to measure [24]. These conceptual and psychometric requirements for responsiveness are important because an instrument can show statistical change when it is not measuring the construct of interest, but some separate although related concept. For example, a questionnaire measuring physical energy (a body function) could show change for a sports intervention when participation is actually the construct one wants to measure.

A useful descriptive instrument should describe the main concerns of a given group of children. Such an instrument can be used to compare the issues that discriminate between groups of children (e.g. children with cerebral palsy show more health status problems related to mobility whereas children with epilepsy are concerned with memory issues). Well designed descriptive instruments are like cameras set to high resolution, able to capture a snapshot of an individual child or a group of children with accuracy and precision.

Contrary to evaluative instruments, descriptive tools need not focus on the elements of health that are expected to change. Instead, these instruments can probe health areas that are important for a specific group of children. This distinction is important in order to form a basis for comparison between clinical groups of children or to draw attention to the special needs of the children of interest. For example, children with a certain level of severity of cerebral palsy are not likely to show changes in their walking ability, but asking about mobility in these children and non-affected children would show how profoundly the issue of mobility affects one group versus the other. Descriptive instruments must have good discriminative validity in that they must be able

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to show quantitatively any differences that exist between groups of children that are expected to be different (e.g. children with cancer in active treatment would be expected to demonstrate more impairments in body functions than children in a remission phase).

In addition to appraising psychometric reports, the onus is on instrument users (i.e. clinicians and researchers) to consider the content of the instrument. Users must determine whether the items contained therein represent health domains that are expected to change following intervention (for evaluation) or items that describe their population's particular challenges or issues (for description). The ICF-CY content tables (Tables I-IV) presented in this chapter can assist with this task. However, additional issues that are unique to childhood will be addressed below.

Measurement issues unique to childhood:

Child versus parent-proxy

Parents and children typically report and emphasize different areas for targeting interventions and often view health differently from each other [25-27]. Whenever possible, children should be surveyed through the use of instruments that have child report questionnaires. Evidence shows that children can reliably report on their experiences beginning at school age [27]. Additionally, school-aged children are the only agents present for the entirety of their daily experience; therefore, in certain ways, they are the most knowledgeable about many aspects of their own daily health experiences. Using appropriately scaled and worded questionnaires, children with various chronic conditions can respond to self-report instruments. A crucial factor for selecting a health-status instrument, therefore, is whether a child self-report version is available with language and a developmentally tailored visual layout.

Negative language

In terms of the content of instruments, it is important to consider how the questions affect children's understanding of themselves [28-30]. Children might be experiencing vulnerable periods, such as an illness or a new diagnosis, when completing self-report questionnaires, and the questions found in an instrument can influence their understanding of themselves [30]. Many self-report health status instruments administered to children were developed for use in a clinical care context. An unanticipated consequence of a clinically oriented approach has often resulted in the

development of questionnaires that emphasize problems or negative aspects of life given that the focus of the clinical milieu is to improve upon the areas in which problems occur.

The content of childhood instruments can be considered in terms of negative content and/or negative phrasing or wording of questionnaire items and stems. The items in many questionnaires were developed using negative wording when the same item could easily have been developed using positive wording. For example the item: "Do you have difficulty remembering things you learn at school?" could be re-worded as: "Are you able to remember things you learn at school?". Including negative content in instruments can create psychological burden for respondents [30] yet also creates an ethical dilemma for those who need to capture the negative as well as positive factors associated with childhood. Including negative content is often necessary to target important concepts such as pain. Regardless, one should be aware of the extent to which negative content is represented in a questionnaire so as to make balanced choices in assessing child health.

Adult valuation of health applied to children

Applying a health utilities approach to health status measurement in children gives rise to special issues [31,32]. Health utilities questionnaires differ from other patient-reported health status instruments by placing an externally established value (a preference score) on the health status of individual children to obtain a single score value for that child's health. In this approach, one needs to consider the original population used to develop the preference scores (usually large numbers of community adults) and extrapolate what influences created the weighting or importance placed on any given health domain (attribute) over another. For example, imagine that the general population was surveyed and walking was determined to be the most important health attribute while social support was ranked low. Applying these preference scores to evaluate the health of children with cerebral palsy who have severe mobility restrictions is ethically questionable because the population of children with cerebral palsy might prefer or value very different health attributes than those chosen by the general population.

Development

Although there are studies that have used instruments developed for adults in research with adolescents and pre-teens, there is very little theoretical foundation or

psychometric validation for such practice. The use of instruments that were created for adults yet applied to developing groups (e.g. infants or adolescents) remains inappropriate. Health status domains that are universally applicable to all age groups are difficult to isolate and often do not exist. Social life for pre-school children focuses on rudimentary social skills with significant others such as siblings or caregivers, whereas the emphasis of social health for adolescents is tied to their peers. Children differ from adults in that their health is not only tied to their environment and context but is also dependent on others (e.g. reliance on parents and communities). Thus it can be argued that assessment of the child's social environment is a key to measuring their health. Overall, clues about developmental appropriateness should be found by examining the characteristics of the initial group of children with which the developers created and evaluated the content of the instrument, and look for evidence that the children and youth were involved in the development of the items of the measure.

Selecting health status instruments

Popular instruments for assessing health status for research and/or clinical purposes are diverse and plentiful. Given the breadth of instruments available, the emphasis of this chapter is to provide instrument users with the criteria to select the most appropriate instruments for their intended purposes. As one might expect, no one instrument likely represents all the desired characteristics a clinician or researchers might intend. Therefore we recommend balancing the health status instruments reviewed here with supplementary targets of measurement found in more specific ICF-CY components, as described in other chapters.

As outlined above, elements to consider when selecting a health status instrument include the following: content validity, including overlap of content with the ICF-CY framework; the purpose(s) and properties of the tools (evaluative versus descriptive); general performance in psychometric studies; and issues unique to childhood instruments, such as self-report and the impact of negativity in wording of items. Users should remain cognizant of these criteria when appraising the reviewed instruments.

The six questionnaires reviewed here differ from instruments that are focused on specific components of the ICF-CY, such as activity and participation because they span more than one ICF-CY component and represent a broadly defined concept of health. These six instruments are summarized in tabular format. It should be noted that the complete history of the psychometric properties of many of these instruments

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reviewed here is extensive and beyond the scope of this chapter. Instead, we limited our report of reliability and validity to the findings described by the original instrument developers. The content-based criteria proposed above have not been the focus of such reviews, yet are essential to the selection of any one of these instruments over another. Additionally, guidelines for interpreting the psychometric rigour of health-status instruments and utility instruments are available and can assist users with the task of sorting through such criteria.

Summary

Health status instruments are complex, due to their necessarily multidimensional nature and their dependence on the developer's contextual understanding or interpretation of health in a certain time and place. These instruments often have decades of implementation and use, therefore sorting through the literature and selecting the appropriate questionnaire for a specific purpose can be daunting. We have attempted to delineate the content of some very commonly used instruments using the ICF-CY, and provide additional criteria for appraising health status questionnaires that can be used with children. And we emphasize once again the importance of being clear about the clinical or research question(s) for which any instrument might be used.

Although one often selects instruments to test specific hypotheses about a group of children in a particular situation, every incidence of instrument use is also a test of the conceptual understanding of health supported by its use. Using instruments that reflect a biopsychosocial approach as represented by the ICF-CY taps into a broad understanding of health. Such an approach to measurement considers children's contextual factors (environment and personal) as well as their body functions and structures, activities and participation.

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Figure I: Differing emphasis of how health status and QOL are assessed for the health domain social interactions

	Health Status Approach Does your child have difficulty getting along with other children?	Quality of Life Approach Are you satisfied with your child's ability to get along with other children?
Signal	Actual performance, capacity, frequency, severity, extent of the child's social interactions	Perception, about expectations, standards or goals about the child's social interactions
Noise Unitended content	Perception, about expectations, standards or goals about the child's social interactions	Actual performance, capacity, frequency, severity, extent of the child's social interactions

Figure II: Instruments reviewed

Standard Approach	Health-Economics Approach
Child Health Questionnaire (CHQ)	Health-Utilities Index III (HUI-III)
Child Health Assessment Questionnaire (CHAQ)	EuroQol 5D-Youth (EQ-5D-Y)
Functional Status II Revised (FS-II R)	
Child Health Illness Profile (CHIP)	

Table 1: ICF body functions assessed according to frequency

First and second level categories are bolded

ICF Category	ICF Code	СНQ	СНІР					
			Core Section	Optional Section	FS II R	CHAQ	EQ5DY	HUIIII
Dispositions & Intra-	b125	1						
Personal Functions								
Responsivity	b1251				1			
Activity Level	b1252				1			
Temperament &	b126							
Personality Functions								
Agreeableness	b1261				1			
Psychic Stability	b1263		2		1			
Openness to	b1264				1			
Experience								
Energy & Drive	b130	2			1			
Functions								
Appetite	b1302							
Sleep Functions	b134	1			1			
Onset of Sleep	b1341		1					
Maintenance of Sleep	b1342				1			
Functions								
Attention Functions	b140	1						
Memory Functions	b144		1					1
Psychomotor Control	b1470	2						
Emotional Functions	b152	20	9	1	1		1	1

Appropriateness of Emotion	b1521			2			
Thought Functions	b160	3					
Expression of Spoken	b16710						
Language							
Experience of Self- &	b180	2					
Time Functions							
Experience of Self	b1800	1					
Seeing Functions	b210						1
Hearing Functions	b230						1
Pain	b280	2	1		1	1	1
Pain in Head & Neck	b28010	1	3				

Table 2: ICF Activities and Participation assessed according to frequencyFirst and second level categories are bolded

ICF Category	ICE	СНQ	СНІР					
	Code		Core Section	Optional Section	FS II R	CHAQ	EQ5DY	HUIIII
Activities &	d	1		1	1			
Participation								
Thinking	d163							1
Reading	d166		2					
Writing	d170					1		
Calculating	d172		1					
Solving Problems	d175		1					1
General Tasks & Demands	d2	1						
Carrying Out Daily Routine	d230		1					
Handling Crisis	d2402		1					
Managing One's Own Behaviour	d250	15			1			
Communication	d3							
Communicating by Receiving Spoken Messages	d310							
Speaking	d330		1					1

Producing Nonverbal Messages	d335		1				
Communication- Producing other specified & unspecified	d349			1			
Conversation	d350	1					
Discussion	d355	1	1				
Discussion with One Person	d3550		1				
Changing Basic Body Position	d410				5		
Lying Down	d4100	1					
Standing	d4104						
Bending	d4105	1					
Transferring Oneself	d420	1					
Lifting & Carrying Objects	d430						
Lifting	d4300	1					
Fine Hand Use	d440						1
Hand & Arm Use	d445						
Reaching	d4452		2		1		
Walking	d450	1			2	1	1
Walking Short	d4500	1	1				

Distances							
Walking Long Distances	d4501	2					
Climbing	d4551		1		1		
Running	d4552				1		
Moving Around in Different Locations	d460	3					
Using Private Motorized Transportation	d4701			2			
Driving Human Powered Transportation (eg: Bicycle)	d4750				1		
Self-Care	d5				1	1	
Washing Oneself	d510	1			1	1	
Washing Body Parts	d51000				1		
Washing Whole Body	d5101				1		
Drying Oneself	d5102						
Caring for Hair	d5202				1		
Caring for Fingernails	d5203				1		
Toileting	d530	1			1		
Dressing	d540	1			3	1	
Taking off Clothes	d5401				1		

Eating	d550	1	1	1		
Looking After One's Safety	d571			1		
Domestic Life	d6					
Acquisition of Goods & Services	d620			1		
Doing Housework	d640	1		1		
Interpersonal Interactions & Relationships	d7	4	1			
Complex Interpersonal Interactions	d720					
Relating with Person's in Authority	d7400	1	1			
Informal Social Relationships	d750		3			
Social Relationships with Friends	d7500	2	2			
Family Relationships	d760	5	1			
Family Relationships Unspecified	d7609		2			
Parent-child Relationships	d7600					
Major Life Areas	d8					
School Education	d820	8	3	1		
-------------------------------------------------------	-------	----	---	---	--	--
Maintaining Educational Program	d8201	3				
Work & Employment other specified & unspecified	d859					
Solitary Play	d8800		1			
Recreation & Leisure	d920	1	2			
Play	d9200		1			
Sports	d9201	1				
Socializing	d9205	11				

Table 3: ICF Environment content assessed according to frequencyFirst and second level categories are bolded

	ICF		C	СНІР				
ICF Category	Code	СНО	Core Section	Optional Section	FS II R	CHAQ	EQ5DY	HUIIII
Products &	e115					3		
Technology for								
Personal Use in Daily								
Living								
Products &	e120					4		
Technology for Indoor								
& Outdoor Mobility &								
Transportation								
Products &	e125					1		
Technology for								
Communication								
Assets	e165			1				
Immediate Family	e310			7				
Friends	e320		1					
Health Professionals	e355	1						
Attitudes	e4	1	1					
Media Services	e5600			1				
Health Services,	e580	1		7				
Systems & Policies								
Education & Training	e585	1						
Services, Systems & Policies								

Table 4: Life/Health/Personal Factors assessed by the health status questionnaires that are not classified in the ICF according to frequency

First and second level categories are bolded

			CI	HIP				
Life/Health/Personal Factor	Non-ICF Category	CHQ	Core	Optional	FS II R	CHAQ	EQ5DY	HUIIII
			Section	Section				
Functioning of Family	Not-covered	3						
Having Fun	Not-covered	1	1	2				
Illness	Not-covered							
Injury	Not-covered			9				
Health Condition	Not-covered	14	3	31		5		
Quality of Life	Not-covered	1						1
General health	Not-defined		1			1	1	
Mental health	Not-defined		1					
Physical Health	Not-defined	2						
Accomplishments (desired or actual)	Personal Factors							
Age	Personal Factors	1						
Cheating	Personal Factors	1	1					
Destroying Property	Personal Factors		1					
Language	Personal Factors			1				
Level of Education	Personal Factors	1						
Lying	Personal Factors	1	1					
Physical Abuse	Personal Factors			1				
Sex/gender	Personal Factors	1						
Solitude	Personal Factors	1						

Race	Personal Factors			1		
Running Away from Home	Personal Factors	1				
Stealing	Personal Factors	2	1			
Things you Want from Life	Personal Factors	1				
Threatening Others	Personal Factors		1			
Self-perception	Personal Factors	1				

	Child Health Questionnaire (CHQ)
Development	Developed in 1990s, CHQ's items are based on the WHO's 1948 definition of health, literature review and expert consensus.
	Early validation of the instrument occurred among 300, 10-15 year old children from a middle-school in north eastern USA; 54, 9-16 year old American children with ADHD; and 20 children aged 10-19 years of age with end stage renal failure on haemodialysis. Data were collected between 1992-1996.
References	[33,35,36,37,38,39]
ICF-CY Components & Categories	The CHQ is focused on body functions such as emotions and activities and participation such as socializing as well as some features relating to personal factors and the environment.
Negative Content	Negative content 33 %
Negative Phrasing	Negative phrasing 39%
Psychometric Highlights	Internal consistency (of subscales) reported as Cronbach's alpha > 0.7
	<i>Test-retest reliability</i> assessed by intra-class correlation coefficients, showed 8/14 subscales with test-retest reliability above 0.5
	<i>Discriminant validity</i> measured with F-statistics: differences between general school group, ADHD group and children with renal failure was significant for all subscales except 'Role/Social Behavioural'
	<i>Responsiveness</i> studies are available for Dutch children with acute asthma, ADHD and children with juvenile arthritis
Author's Note	This instrument is detailed and diverse; however it contains a mixture of health status and QoL items. Users who wish to obtain or partition information about children using health OR QoL would not be able to separate these two outcomes by using all scales of the CHQ.
Availability	Short and long versions of parent and child report forms are

Child Health Questionnaire (CHQ)
available in 98, 87, 50, 28 item versions. Child reports are for ages 10-19 years and parent/proxy reports are for ages 4-19 years. This instrument can be obtained through
http://www.healthact.com/chq.html for purchase.

	Child Health & Illness Profile (CHIP)
Development	Developed originally for adolescents to self-report on their health status. Focus groups with a sample of mothers and fathers, half of whom had children with chronic illness, reported on health issues to create the content of the questionnaire items.
	Early versions of the instrument were tested in paediatric outpatient settings in Baltimore with parents of diverse economic backgrounds. Field studies were conducted in four sites in the United States. The developers assert that use of the instrument is intended for research purposes and further validation is required to adapt the instrument to clinical settings.
References	[40,41,42]
ICF-CY Components & Categories	The core module of the CHIP focuses on activities and participation with an emphasis on interpersonal aspects such as playing and school performance. Body functions (e.g. emotional functions, experience of self and pain) are included Environment categories (e.g. attitudes of peers such as bullying, and social support from parents) are also assessed. The optional modules focus on additional features of the environment, such as health or social services, as well as a checklist of symptoms or diagnoses that can be represented with the International Classification of Diagnoses ICD10, not the ICF-CY.
Negative Content Negative Phrasing	Negative Content: 42%
	Negative Phrasing: 0%
Psychometric Highlights	Internal consistency using Cronbach's alpha =0.79 to 0.88 for the parent form and 0.70 to 0.82 for the child form
	<i>Test-retest reliability</i> was reported using intra-class correlation coefficients = 0.63-0.85 for parent form with the exception of the Restricted Activity sub-domain= 0.32; 0.63-0.76 for child report form
	<i>Discriminant validity</i> was reported for the adolescent version for the subscales only. A substantial variety in mean group differences were reported, less than half of which were significant and confidence intervals were reported about the means.

	Child Health & Illness Profile (CHIP)
	Responsiveness studies unavailable at this time.
Author's Note	This instrument has many items, which will reduce its usefulness in clinical settings. An advantage is the breadth of health components that span the body functions, activity and participation, environment and personal factors according to the ICF-CY, therefore indicating a true biopsychosocial approach to health status measurement.
Availability	Self-report is available for ages 6 to adolescent as well as parent- report forms.
	Contact: http://www.childhealthprofile.org/ for licensing information.

	Functional Status II (FSIIR)
Development	Developed conceptually based on Starfield's health framework. Adapted from an earlier version, the authors took 35 items from the Functional Status I and added some items based on literature review.
	The FSIIR was validated on a sample of over 700 American children that included children with significant chronic conditions, children with ongoing health conditions seen for regularly scheduled appointments and children seen for routine health care.
References	[43,44,45,46]
ICF-CY Components & Categories	The FSIIR focuses on body functions such as disposition, temperament, energy, sleep, cognition and emotion. General activities, communication and eating from activities and participation are touched upon. No environmental or personal factors are assessed.
Negative Content	Negative Content= 36%
Negative Phrasing	Negative Phrasing: 0%
Psychometric	Internal consistency was measured by Cronbach's alpha = 0.86-0.87
Highlights	<i>Discriminant validity</i> was reported as mean differences without significance testing or confidence intervals 86.8 SD=15.7 for the ill group and 96.1 SD=8.2 for the well group.
	<i>Test-retest reliability</i> not reported in instrument development studies.
	<i>Responsiveness</i> was shown in a study comparing chronically ill children with healthy children
Author's Note	This instrument is short although response errors are likely to occur if the questionnaire is not administered in interview format as the developers intended.
Availability	Parent-report forms for children 0-16 are administered in interview form. No child response form. The 14-item version is common to all age groups and longer age specific forms are available. To obtain the

Functional Status II (FSIIR)
instrument, contact:
Ruth E. K. Stein, M.D., Department of Pediatrics,
Albert Einstein College of Medicine/Montefiore Medical Center; Centennial 1, 111 East 210th Street; Bronx, NY 10467.

	Child Health Assessment Questionnaire (CHAQ)
Development	Developed based on an adaptation from the adult Stanford Health Assessment Questionnaire (HAQ) with at least one 'child specific' item added to each functional area in an attempt to adapt the measure for children with juvenile arthritis. The CHAQ is currently used by the Paediatric Rheumatology International Trials Organization (PRINTO) to collect international health data about children with arthritis.
References	[47,48,49,50]
ICF-CY Components & Categories	The CHAQ focuses on activities such as walking, bike riding, washing and dressing. There is no emphasis on participation such as interpersonal aspects of life and negligible information about the body functions of pain. Environment is assessed based on needs for assistive devices as opposed to social support or health and social services.
Negative Content	Negative Content= 3%
Negative Phrasing	Negative Phrasing=0%
Psychometric	From Validation Studies:
Highlights	Internal reliability was reported using Spearman correlation coefficient= 0.66 for children with juvenile arthritis
	From use by the Paediatric Rheumatology International Trials Organization (PRINTO): <i>Discriminant validity</i> reported as statistically significant mean scores differences. However there were small differences between the clinical groups and large standard deviations but no confidence intervals reported about the mean to establish whether it discriminates between clinical groups.
	Internal consistency was reported using Cronbach's alpha: >0.5
	<i>Test-retest reliability</i> was reported using intra class correlation coefficient= 0.6-0.9
	<i>Responsiveness</i> information is available among children with juvenile arthritis for participant countries of the PRINTO trials
Author's Note	This questionnaire first asks parents whether an activity is relevant to his/her child based on what is expected for age before assessing the child's level of ability for the activity. Conversely, norms for age

	Child Health Assessment Questionnaire (CHAQ)
	are not available. Therefore one should use caution applying this instrument to children with delays or chronic conditions.
	Authors not involved in CHAQ original development are attempting to validate its use beyond children with arthritis for children with cerebral palsy.
Availability	Parent- report form for all ages of children can be found in the public domain, no child report form available: <u>http://www.bspar.org.uk/downloads/registry_forms/CHAQForm.pdf</u>

	Health Utilities Index III
Development	The HUI III evolved from previous II & I versions designed to capture health outcomes of very low birth weight infants. The attributes assessed by HUI III were refined through literature and qualitative work with 84 parent-child pairs. The HUI III was first implemented in the 1990 Statistics Canada Ontario Health Survey & the 1991 Statistics Canada General Social Survey for the general Canadian population.
References	[51,52,53,54,55,56,57,58]
ICF-CY Components & Categories	The HUI III emphasizes body function attributes such as vision, hearing, cognition and emotion, and activities and participation items such as walking, hand function and communication. Environment domains assessed directly. There is one QoL probe.
Negative Content Negative Phrasing	Negative Content 12.50% Negative phrasing 47.50%
Psychometric Highlights	<i>Test-retest reliability</i> : using intra class correlation coefficient= 0.77 based on its use in the Canadian General Social Survey Health Questionnaire in 1991
	<i>Responsiveness</i> studies are available for children with asthma and Hodgkin's Lymphoma
Author's Note	As a health <i>utility</i> tool, the instrument can 1) classify the level of health based on norms from the population, but also 2) provides a final score where 0 indicates a valuation of death and 1 indicates perfect health for the purpose of health-economics or resource allocation decisions.
	When the HUI III classification is interfaced with the preference scoring function (the health preferences are based on adult preferences in Canada) to create a health utilities score value, application of that utility score (e.g., scores less than 1 = worse than death) to describe any one particular child is ethically questionable.

	Health Utilities Index III
Availability	Parent-report forms for children 6-18, no child report version. License and pricing information can be found at <u>http://healthutilities.com/</u>

	Euroqol Youth (EQ-5D-Y)
Development	The EQ-5D-Y was adapted from the adult EQ-5D for the purpose of providing children and adolescents the opportunity to report on their own health. However the content of the adult version was not created using a developmental approach to child health. The wording was adapted for child self-report using cognitive interviewing with children and adolescents.
	The original (adult) EQ-5D contains health components (states) that were described from literature review and formed by expert consensus in the early 1990s within the EuroQoL group.
References	[59,60,61,62]
ICF-CY Components & Categories	Body functions such as emotion and pain are covered. Walking, self-care, washing, dressing are contained in the instrument representing activities. No participation, environment or personal factors were represented by the ICF-CY.
Negative	Negative Content: 47%
Content Negative Phrasing	Negative Phrasing: 47%
Psychometric Highlights	<i>Test-Retest reliability</i> using percentage agreement = 69.8 to 99.7% and using Cohen's kappa= -0.003 to 0.549 for 8-18 year old children from Italy and Spain for a 7-10 day testing interval
	Discriminant validity was reported as percentage of responses selected for each health state (i.e., mobility, emotions and pain) between chronic and non-chronically affected children. The differences in the proportion of respondents of ill versus well children varied based on the country and were often non- significant for samples composed of at least 200 children. <i>Responsiveness</i> information not found for the youth version.
Author's Note	As with the HUI, this instrument is intended to produce a utilities score where 0 represents a state of health equivalent to death and 1 represents ideal health. Applying a valuation to any one individual child (as described for the HUI) based on the norms of

	Euroqol Youth (EQ-5D-Y)
	the majority European population, is an ethical concern.
	EuroQoL developers recommend supplementing the EQ 5D-Y with standard questionnaires to provide information about individuals. Thus, the EQ 5D-Y presents benefit only as a crude screen for body function and activities type health issues that are commonly valued by the adult population at large.
Availability	The form is intended for completion by individuals 8-18. Licensing information and agreements can be obtained through http://www.euroqol.org/home.html

Chapter 6

Generic child health and quality of life patient-reported outcome use in the literature: Analysis of instrument content versus instrument application

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This article will be submitted to Developmental Medicine Child Neurology.

NF conducted the systematic review (wrote the syntax, screened abstracts and extracted data) and devised the scientific rationale for this study. NF wrote the manuscript and supervised analyzed the perspectives found in the included instruments. She interpreted the findings with the input of other authors (75%).

OKDC assisted with data extraction of article characteristics and analyzed the perspectives for each the instruments included in the study using WHO definitions (6%).

EK analyzed the perspectives for each the instruments included in the study using WHO definitions and provided editing feedback (6%).

PLR provided scientific feedback on all post-review phases of the study. PLR made important contributions to the presentation of data and edited all drafts of the manuscript (3%).

PR provided feedback regarding the scientific rationale for the study and the purpose of the study (2%).

AC supervised the systematic review, provided feedback regarding various drafts of the syntax and provided the resources to conduct the systematic review and manage the extracted data found in this study. AC provided scientific input on the rationale and purpose of the study (8%).

Abstract

Background

More children than ever before are living with chronic, episodic or nonprogressive health conditions. The health services provided to children with chronic conditions need to assess the impact of care on day to day life. Health status and quality of life (QOL) instruments are useful for meeting this need. The conceptual basis for the instruments that are available are unclear and have an impact on instrument validity.

Method

This study will employ a review of the literature to find articles that employ health status and QOL instruments. Information regarding various aspects of instrument use will be extracted from the articles and analyzed using content analysis techniques that employ World Health Organization definitions of health status and QOL.

Results

Most of the instruments used to measure HRQOL or QOL do not meet WHO definitions of QOL. The studies that employ these instruments are typically descriptive in nature and apply the questionnaire to either the child or the parent to fill in the questionnaires but not both.

Discussion

Analysis of generic instruments showed that there is little agreement within the literature about what these instruments are intended to measure or how to make conceptual distinctions between health and QOL. Overlooking such issues will result in the propagation of study results that are incompatible and difficult to interpret.

Introduction

The effectiveness of health programs and interventions has traditionally been decided by clinicians, administrators, researchers and policy makers with little input from patients (1-4). With more children than ever before living with chronic, episodic or progressive health conditions (5,6), health services need to assess the impact of care on day to day life rather than restrict measurement, to morbidities that might or might not resolve from a biomedical perspective (7). In parallel, health services are becoming an increasingly precious commodity around the world and difficult decisions need to be made about the focus and allocation of health resources (8). It is important to recognize and value child and family assessments of the impact of services on child health in order to provide insights that can help with these difficult decisions.

Health status and quality of life (QOL) questionnaires are valuable tools for capturing child and parent experiences. When these questionnaires are administered directly to patients outside of a clinical testing situation and without clinicianinterpretation they can be called patient-reported outcomes (PROs) (9). PROs are essential patient appraisal tools, but they were often developed and implemented without a firm conceptual basis or a clear definition of what they intended to measure. Many PROs were developed prior to agreement in the literature about what constituted health versus QOL.

This lack of agreement about definitions affects the content of instruments. An instrument created for use in one particular context, without clear definitions, will contribute little to the advancement of knowledge about the effectiveness of interventions outside that study context. For example, one might choose to use the Peds QL 4.0 (10, 11) to evaluate the effectiveness of an intervention in one study while another study used the Child Health Questionnaire (CHQ) (12). In both cases, the instruments were applied in the hopes of measuring some aspect of health status or QOL, but the results obtained between studies were hardly comparable because they were likely measuring different although related things. Although the inconsistencies between instruments arose over decades, without proper content analysis, it will remain unclear what constructs they are measuring today.

Many clinicians and researchers believe that use of a generic PRO will overcome measurement discontinuity between situations so they choose to adopt the most popular instruments for their studies. Using this reasoning, the results of one's particular study are comparable to another's so long as the same generic instrument

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was employed. The changes observed between studies with the same instruments are comparable, however it remains unclear what exactly was being measured (e.g., health versus QOL) in the first place. The items, components and domains found in the PRO therefore need to be contrasted with a conceptual definition that clarifies what is being measured.

A practical illustration will provide further insight on why poor conceptual definition of a child self-report instrument is an important issue. Imagine the administration of a PRO that truly measures health status to evaluate whether a treatment improved a child's HRQOL. The child and parent report to their clinician that the health intervention made a difference in what they emphasize as healthy (e.g., my child cannot walk but he will have excellent mobility using a wheelchair) even though the actual health status (e.g., walking ability) did not change. In essence, the child's HRQOL changed (their subjective view on walking and mobility) but the health-status instrument was unable to show this because the PRO was not designed to target the expectations, standards and concerns of the child, but rather the presence, absence, frequency or severity of health or functional issues. The program who administered the instrument incorrectly concluded that the child's HRQOL did not change when in fact it was only the child's ability to walk that did not change. The intervention was falsely disregarded as ineffective.

Without a clear match between what one intends to measure and what the instrument is actually measuring, content validity is sacrificed (13). Content validity is cited as an important element to address on validity checklists (13), yet in the case of health and QOL PROs for children it has often been overlooked.

Reviews of health status or QOL instruments to date have attempted to discuss issues with conceptual definitions of measures but few have offered solutions to the problem (14,15). There is a great deal of data associated with the use of several popular instruments and it is unlikely that they will stop being used. It is, however, possible to complete thorough and systematic assessment of the perspectives and content contained in these instruments using standardized content analysis methods developed specifically for this purpose (16,17). These methods build on World Health Organization (WHO) definitions of QOL (18), and other WHO perspectives that define and classify components of and influences on health (19,20). WHO definitions and classifications are appropriate tools for establishing common ground between PROs because they are rigorously defined and internationally consensus driven, thus representing a firm base by which to appraise instruments from almost any context.

The content analyses employed in this study are based upon a method initially developed in 2002 by Cieza et al., (17), which included multiple iterations of validation in the subsequent two revisions (16). The latest version of the Cieza et al. content analysis method (the linking rules) can decipher the difference in perspectives between items in PROs (e.g., health versus QOL) (21). The method can also make the health and health-related content of these questionnaires explicit using the original standard classification of health called the International Classification of Functioning, Disability and Health (ICF) (19) or the child version (ICF-CY) (20).

The objectives of this review are to undertake a cross-sectional analysis of generic PRO use in the child health-based literature over a five year period in order to: 1) describe characteristics of studies associated with child health and QOL PRO use, 2) provide assessment of the perspectives found in the most popular PROs using the updated linking method and WHO definitions of health and QOL, and 3) discuss the extent to which there is correspondence between perspectives present in an instrument and the instrument's intended use. In essence, a content analysis based on WHO definitions will be used to highlight content validity issues in the literature. In part II of this study, we will perform an analysis of the health and health-related domains from each item of these instruments using the ICF-CY.

Objectives:

- 1. Describe basic characteristics associated with generic PRO use: e.g., what types of studies, ages of children and respondents are frequently reported?
- 2. Apply WHO definitions to determine what the most frequently used generic PROs in child health are actually measuring; i.e., what is their perspective (health status vs. HRQOL/QOL) using WHO definitions?
- 3. Describe what perspectives (health vs. HRQOL/QOL) instrument users intended to measure with these PROs
- 4. Discuss the match between what users intend to measure and what is actually measured using WHO perspectives, as well as the implications of these results.

Method:

Search strategy:

A literature review was conducted on Medline, PsychINFO and CINAHL for articles published between 2004 and 2008. The search was limited to three databases and these databases were chosen for their uniqueness of content relative to each other. Search terms for 'health' 'quality of life' 'disability', etc., were cross-referenced with search terms relating to questionnaires and instruments, which were also crossreferenced relative to the child and youth population. The syntax for the search was finally checked by a medical librarian specializing in systematic review and is available as an electronic supplement appendix A.

An overview of the search procedure is presented in Figure 1. Once the results of the search were obtained and duplicates deleted, 22 001 abstracts were screened by one reviewer with 60% random selection screened by a second reviewer. Next, 2145 full-text articles were screened by one of four calibrated reviewers and data extracted according to the criteria described in the following section. The inclusion criteria for the articles were any peer-reviewed study using a named and validated PRO on children to measure multi-dimensional health (functioning/disability), QOL (and related concepts such as well-being or HRQOL). We excluded articles that had less than 10 patients, did not focus on children or young adults (e.g., a clinical trial involving patients 16-80 years of age with no child-specific objectives), studies using only population surveys, ad hoc instruments, or unnamed instruments created for the study without prior validation, non-peer-reviewed studies, and review studies or editorials.

Data extraction

Data from each article were extracted using one of four reviewers, each of whom was 'calibrated' to the first author based on a sample of 10 articles at a time. A reviewer and the first author included or excluded 10 articles independently until their agreement reached 90%. In addition, all reviewers met regularly with the lead author to review difficult cases. A record was kept of the difficult cases and communicated and reviewed with all data extractors.

Article characteristics

Study characteristics were extracted in the following manner:

- 1. Study types were grouped according to the Cochrane group approach for observational studies (22).
- Ages were grouped as follows: 0-12 months, infants; 13-36 months, toddlers;
 >3-5 years pre-school; >5-12, school age; 13-19, adolescents; 20-25, young adults.
- 3. Language of data collection was recorded for each article. If data collection occurred in more than one country or in a country with more than one language, all countries and languages where PROs were extracted from children or their families were recorded

Characteristics pertaining to PRO instruments and their use were extracted in the following manner:

- 1. the name of the instrument and the instrument version if reported
- 2. the language in which the data collection occurred
- 3. whether the child only, proxy only or both child and proxy completed the questionnaire
- 4. the author's stated purpose in using the instrument, e.g. to measure health, HRQOL, QOL, well-being, physical or mental health etc.

Coding the stated purpose of instrument use

All coding was performed using WHO definitions of health, functioning and disability according to the ICF-CY or the WHOQOL task force definition of QOL (18) as well as the Cieza et al. linking rules (16,17). This method for interpreting instrument perspectives using WHO definitions has been reported in detail elsewhere (21). Prior to coding, the exact quotation from the text regarding the intent of questionnaire use was pasted into the database and the content analysis method was applied to code the outcome the instrument was used to measure. Statements about HRQOL or QOL, general mental or physical health, functioning or disability were mapped directly onto WHO/ICF-CY terms and were coded using the standard method mentioned. Examples of less straightforward statements were coded as follows: health satisfaction was coded as HRQOL, burden of [health condition] on daily life was coded as disability, adaptive behaviour and development were coded as functioning, and subjective well-being was coded as QOL.

Determining actual PRO perspectives (health vs. QOL)

The PROs that demonstrated 1% use or greater in this cross-section of the literature were obtained for this analysis. Generic instruments that appeared less than 1% of the time were not analyzed but the title and frequency of appearance of excluded questionnaires are available upon request. The included instruments were reviewed by 3 content assessors trained in the coding of PROs using the ICF or ICF-CY. These included three clinician-scientists (an occupational therapist NF, paediatrician OKC and neuropsychologist EK). The assessors reviewed the WHO definitions of health, functioning and disability (from the ICF-CY) and QOL (from WHOQOL task force) as well methods published for coding the perspectives of items using these. An illustration of the how the concepts were divided for the coding scheme can be found in Figure 2. Each assessor individually coded the perspectives found in the instruments on an item by item basis and met to combine the results, assess the initial level of agreement and develop consensus regarding the items on which there was disagreement or lack of clarity. The independently categorized coded items were set aside for a reliability analysis. A final consensus list of perspectives for each item is what is reported as the overall composition of perspectives in the PRO.

Initial reliability of the coding method

Agreement for the first round of independent coding of the PRO perspectives was analyzed on an item by item basis for each instrument using percentage agreement.

Results

709 studies yielded 1151 occasions of generic PRO instrument use applied to children. The characteristics associated with generic PRO use: the types of studies included in the paper, age groups represented by the instruments, and the respondents (proxy, child or both) are shown in Tables 1-3. The ten most used languages from which PROs collected data, in order of frequency, were English, Dutch, German, Spanish, French, Swedish, Italian, Chinese, Norwegian and Portuguese.

Perspectives found in the generic instruments coded by analysts using WHO definitions are found in Table 4 which can be contrasted with the reported use of the same instruments from the included studies in Table 5. A summary comparison of instrument use versus instrument content is found in Table 6. The agreement for the coding perspectives for each instrument is listed in Table 7.

Discussion

When standard WHO definitions of QOL and biopsychosocial health are applied to instruments, a conflict is revealed between their actual content and the purposes to which they are being applied. Of the 15 PROs analyzed, 12 were (most frequently) applied to measure HRQOL or QOL. Of those, only 4 were coded as having an HRQOL/QOL evaluation approach using WHO definitions. The strongest discrepancy between an instrument's content versus its application was found for the PedsQL (10,11). The PedsQL was found consistently to represent health status *not* an HRQOL or QOL perspective for all of its items. It should be mentioned that analysis of the Peds QL in our previous work showed that the items represented a broad biopsychosocial definition of health (21). Thus conceptually, the PedsQL spans a wide definition of health status that includes environmental factors, as well as factors connected to functioning and disability but the breadth of health content should not be confused with QOL.

To be clear, the assumption of this analysis is that the difference between a health status and HRQOL or QOL instrument is not so easily found in *what* is measured because both health status and HRQOL instruments can measure the same health domains. Health status and HRQOL instruments *should* address biological, psychological and social determinants of health. What distinguishes the health perspective from the HRQOL/QOL perspective is *how* domains of life and health are measured. For example, one could ask: "how difficult is it for your child to walk short distances?" or "how satisfied do you feel with your child's ability to walk short distances?" These items both ask about the same domain (walking): however, where the first targets performance (a dimension of health) the other targets satisfaction (a dimension of HRQOL).

Instruments that were named by developers as health, functioning or disability instruments (FDI, CHIP, FSIIR), were used appropriately relative to the others included in the analysis. The studies that sought to measure HRQOL/QOL displayed the most incongruity between what is actually measured versus how the instruments are used. Thus problems with content validity were the rule rather than the exception for researchers seeking to measure HRQOL/QOL.

Although it is unclear why this mismatch between content and application occurred, we suspect that the time and context in which these instruments were developed played a role in the problem. Instruments such as the CHQ and PedsQL were developed before conceptual differences between a biopsychosocial definition of health (as in the ICF-ICF-CY) and QOL (according to the WHO QOL task-force) were made clear in the health literature. Our results do not indicate that one should cease to use these instruments, but argue that it is important to be aware of the content, perspectives and composition of their items relative to an intended purpose as opposed to relying on their popularity or the content of the items alone.

The importance of having a firm conceptual understanding of the concept one wishes to measure before one selects an instrument is crucial to content validity (13). Guidelines about health-status measurement selection expressed in checklists such as COSMIN (23-26) place a strong focus on psychometric properties. Additionally, the FDA recommendations on the development and application of PROs caution that they must be developed in a context and therefore must be applied only for the populations for which they have been validated (9). These prescriptive recommendations might somehow detract from the importance of conceptual understanding of health and QOL by instrument users in place of a checklist approach. Irrespective of psychometric performance, conceptual understanding of what one intends to measure is essential to the process of obtaining validity with the use of health and QOL PROs.

The good news with PROs is that the items can often be taken at face value, meaning the content of an item and the thing the item is designed to measure, are usually much more straightforward than with clinical assessments. Thus if users have a firm conceptual understanding of the difference between health, HRQOL and QOL, they have critical skills that can help them to determine the face validity of these instruments or the content validity of the instruments for their purposes. Although these results do not intend to suggest that the importance of COSMIN and FDA recommendations be overlooked, we suggest a firm theoretical grounding of health and QOL as crucial to the process of measurement validity for these concepts.

Our descriptive analysis on the use of PROs showed these instruments were applied mostly to school-aged children and or adolescents, and to children or their parents but infrequently to both. There is much evidence to suggest that the correlations between child and proxy reports on health status and QOL are inaccurate or low (9,27-31). Proxies rarely actually report about their children from the vantage point of the child. Instead, parents and proxies typically report on health or QOL from their own vantage point. Knowing this, child and proxy reports need to be considered separately; neglecting to do so represents a missed opportunity to incorporate a more holistic picture of a child's life in the measurement process. We also found that the majority of the studies that used PROs for children were reported in cross-sectional research. The use of PROs in such research is descriptive as opposed to evaluative in nature. Descriptive instruments need to show the difference between groups of children (e.g. very ill from slightly ill) in order to be useful. It is possible that the PROs have demonstrated popularity despite issues brought forward by this analysis because researchers have not had enough experience attempting to apply the instruments in an evaluative context. When evaluating interventions, the responsiveness of a PRO depends on its ability to detect change (32). In theory, if an instrument does not have a strong and clear overlap with the domains that are expected to change, it will be unresponsive and ineffective for evaluative purposes. The literature available on whether most health interventions change health status or QOL is inconclusive at best and this could very likely be related to problems with content overlap.

Additionally, much of the validation work for the instruments mentioned in this study includes psychometric evidence that is useful for determining a PRO's use descriptively with much less evidence to show their ability to detect change. More effort needs to be applied to determine whether interventions are being accepted or rejected based on whether these instruments truly assess health status or HRQOL/QOL. It is therefore important that distinctions between health status and HRQOL/QOL are determined and applied at this descriptive phase of PRO use, before they are used for evaluative purposes.

Conclusion

We applied a systematic method for assessing the perspectives of PROs based on their content using WHO definitions of QOL and the ICF/ICF-CY. Analysis of generic instruments showed that there is little agreement in the literature about what they are intended to measure or how to make conceptual distinctions between health and QOL. Overlooking such issues will result in the propagation of study results that are incompatible and difficult to interpret. Providing evidence on the state of scientific knowledge about a health condition's impact on children's lives or the impact of interventions for the purposes of decision-making will also be interrupted.

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Table 1: Types of studies

	Percent
Cross-sectional	63.1
Cohort	12.2
Before & After/Uncontrolled Trial	6.9
Ecological	6.6
Controlled Trial (RCT, QR, NR)	5.5
Case-Control	2.7
Prevalence Survey	1.4
Case-Series	0.9
Surveillance Data	0.4
Routine Data Collection	0.3

Table 2: Age groups represented within 709 studies

Group	Percentage*
Infant	4.5
Toddler	16.1
Preschooler	24.4
School-aged	73.5
Adolescent	73.9
Young adult	11.8

*Percentage will not sum to 100 because many studies collected data from PROs from more than one age group

Table 3: Respondents

Respondent	Percentage
Child Only	41.5
Proxy Only	38.0
Both	20.1
Unknown	0.4

	Percentage use in the literature	Composition of items coded along the QOL/ HRQOL perspective	Composition of items coded along the health, functioning or disability perspective including health barriers and facilitators (Health Status)	Composition of items coded as having other or unknown perspective	
Peds QL 4.0	22.8	0.0	100.0	0.0	
Child Health Questionnaire (CHQ)	22.1	29.6	62.2	8.2	
DISABKIDs	1.3	76.9	23.1	0.0	
Functional Status Questionnaire (FSII)	1.7	0.0	100.0	0.0	
Health Utilities Index (HUI)	4.3	12.5	87.5	0.0	
KINDL	3.8	36.6	53.3	10.0	
KIDSCREEN	6.6	69.2	28.8	1.9	
SF_8_12_36	3.4	22.2	77.8	0.0	
Diener's Satisfaction with Life Scale	1.0	100.0	0.0	0.0	
TNO-AZL Series	5.6	36.6	63.4	0.0	
Child Health and Illness Profile (CHIP)	3.0	2.4	87.4	10.2	
Functional Disability Inventory (FDI)	2.9	0.00	100.00	0.00	

Table 4: Perspectives found in generic PROs according to WHO terms and definitions

Youth Quality of Life Instrument (YQOL)	1.4	76.6	17.1	7.3	
General Health Questionnaire	1.0	42.9	57.1	0.00	
Huebner (SLSS)/(MSLSS)	0.8 (0.5)	100 (65.0)	0.0 (30.0)	0.0 (5.0)	
Other*	18.2	n/a	n/a	n/a	

*Other generics include PROs that were used too infrequently i.e. <1.0% to be reported in this manuscript

	Stated Aim of Generic PRO Use (by %)							
	None	QOL	HRQOL	Functioning	Disability	General Health	Mental health	Physical health
Peds QL 4.0	5.0	27.9	60.7	1.9	0.8	2.7	0.7	0.4
Child Health Questionnaire (CHQ)	4.8	29.8	38.9	7.1	3.6	13.1	2.0	0.8
DISABKIDs	33.3	46.7	20.0	0.0	0.0	0.0	0.0	0.0
Functional Status Questionnaire (FSII)	10.0	0.0	5.0	20.0	30.0	35.0	0.0	0.0
Health Utilities Index (HUI)	2.0	8.2	59.2	2.0	0.0	28.6	0.0	0.0
KINDL	0.0	52.3	45.5	0.0	0.0	2.3	0.0	0.0
KIDSCREEN	0.0	34.2	64.5	0.0	0.0	1.3	0.0	0.0
SF_8_12_36	0.0	33.3	30.8	7.7	0.0	28.2	0.0	0.00
Deiner's Satisfaction with Life Scale	0.0	83.3	8.3	0.0	0.0	8.3	0.0	0.0
TNO-AZL Series	1.6	25.0	59.4	12.5	1.6	0.00%	0.00%	0.00%
Child Health and Illness Profile (CHIP)	5.9	20.6	26.5	0.0	0.0	47.1	0.0	0.0
Functional Disability Inventory (FDI)	0.0	0.0	0.0	27.3	69.7	3.1	0.0	0.00
Youth Quality of Life Instrument (YQOL)	0.0	100.0	0.0	0.0	0.0	0.0	0.0	0.0
General Health Questionnaire	0.0	8.3	8.3	16.7	0.0	0.0	66.7	0.0
Huebner's SLSS/MSLSS	0.0	100.0	0.0	0.0	0.0	0.0	0.0	0.0

Table 5: Reported use of generic PROs according to the included studies

Table 6: Comparison of instrument use versus instrument content

Instrument name	Instrument Use	Instrument Content		
	Most studies used this	Summary Measurement Perspectives from WHO		
	PRO to measure this	content analysis		
Peds QL 4.0	HRQOL/QOL	Health Status		
Child Health Questionnaire (CHQ)	HRQOL/QOL	Health Status (with QOL/ HRQOL & some unknown		
		features)		
DISABKIDs	HRQOL/QOL	HRQOL/QOL (with some health status features)		
Functional Status Questionnaire (FSII)	Health Status	Health Status		
Health Utilities Index (HUI)	HRQOL/QOL	Health Status (with one HRQOL/QOL attribute)		
KINDL	HRQOL/QOL	Health Status (HRQOL/QOL subcomponent)		
KIDSCREEN	HRQOL/QOL	HRQOL/QOL (with some health status features)		
SF_8_12_36	HRQOL/QOL	Health Status (with HRQOL/QOL subcomponent)		
Deiner's Satisfaction with Life Scale	HRQOL/QOL	HRQOL/QOL		
TNO-AZL Series	HRQOL/QOL	Health Status (HRQOL/QOL subcomponent)		
Child Health and Illness Profile (CHIP)	Health Status	Health Status (with an unknown subcomponent)		
Functional Disability Inventory (FDI)	Health Status	Health Status		
Youth Quality of Life Instrument (YQOL)	HRQOL/QOL	HRQOL/QOL (health facilitators/barriers		
		subcomponent)		
General Health Questionnaire	HRQOL/QOL	Health Status & QOL/HRQOL		
Huebner's SLSS/MSLSS	HRQOL/QOL	SLSS-HRQOL/QOL		
		MSLSS-HRQOL/QOL		
		(health subcomponent)		
Instrument name	% Agreement			
-----------------	-------------			
СНІР	75			
СНQ	88			
Diener's SWLS	100			
DISABKIDS	77			
FDI	100			
FSIIR	100			
GHQ-28	85			
HUI	100			
KIDSCREEN	80			
KINDL	46.7			
MSSWLS	76.7			
PEDsQL	91.3			
SF-36	89			
SSWLS	100			
TAPQOL	86			
YQOL	82.1			
total	81.1			

Table 7: Agreement statistics for coding perspectives of each instrument





Figure 2: Conceptual interpretation of WHO terms of health and QOL applied to instrument items



Electronic supplement: Appendix A Search strategies

Ovid MEDLINE(R) and Ovid OLDMEDLINE(R) 1950 to October Week 50 2008

#	Searches	Results
1	"quality of life".sh.	72418
2	disabled children.sh.	2598
3	health status/ or health status disparities/	41038
4	(health adj function*).ti,ab.	702
5	(school adj health*).ti,ab.	2705
6	exp Child Development/	35266
7	exp schools/ or exp schools, nursery/	60857
8	(health adj status).ti,ab.	23729
9	"Activities of Daily Living"/	37653
10	"activit* of daily living".ti,ab.	9928
11	Rehabilitation/	15473
12	rehabilitation.ti.	35049
13	adaptive behavio?r.ti,ab.	1359
14	(preschool or pre-school or pre school).ti,ab.	13434
15	(child adj development*).ti,ab.	2371
16	"disabilit*".ti,ab.	65525
17	quality of life.ti.	22195
18	"rehabilitation*".ti,ab.	69716
19	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18	^r 378730
20	health surveys/ or health status indicators/	42260
21	indices.ab. or indices.ti.	73368
22	index.ti. or index.ab.	272223
23	scale*.ti. or scale*.ab.	235369

24	pencil & paper.ti,ab.	8
25	"Instrument*".ti,ab.	118738
26	exp intervention studies/	3990
27	exp "Outcome and Process Assessment (Health Care)"/	422448
28	treatment outcome/	359374
29	epidemiologic studies/ or feasibility studies/ or intervention studies/ or pilot projects/ or sampling studies/	102257
30	clinical trial/	460026
31	trial.ti.	78690
32	intervention studies/	3990
33	"intervention*".ti.	45617
34	Questionnaires/	189500
35	(self report or self report).ti,ab.	17107
36	"survey*".ti,ab.	249872
37	"questionnaire*".ti,ab.	175770
38	(outcome adj5 measure).ti,ab.	22704
39	(outcome* adj assessment*).ti,ab.	2078
40	measure.ti.	14799
41	"interview*".ti,ab.	136553
42	disability evaluation.sh.	23774
43	(disabilit* adj evaluation*).ti,ab.	400
44	35 or 33 or 32 or 21 or 26 or 42 or 22 or 30 or 23 or 29 or 25 or 27 or 39 or 28 or 40 or 36 or 41 or 20 or 38 or 34 or 37 or 24 or 43 or 31	1950820
45	exp Child/	1305468
46	Adolescent/	1302716
47	"child*".ti,ab.	731404
48	"teenager*".ti,ab.	6985
49	"child*".ti,ab.	731404

50	"adolescent*".ti,ab.	97715
51	"infant*".ti,ab.	242628
52	exp infant/ or infant, newborn/	812682
53	(baby or babies).ti,ab.	38334
54	"toddler*".ti,ab.	3458
55	"preschooler*".ti,ab.	2170
56	50 or 53 or 51 or 48 or 47 or 52 or 46 or 49 or 45 or 55 or 54	2542982
57	56 and 19 and 44	48912
58	addresses.pt.	3209
59	bibliography.pt.	14224
60	biography.pt.	152563
61	"congresses".pt.	51671
62	("dictionary" or directory).pt.	7170
63	editorial.pt.	233379
64	government publications.pt.	185
65	"legal cases".pt.	8675
66	"letter".pt.	652579
67	"review".pt.	1437184
68	((systemat* or critical) adj (review* or search*)).ti.	13895
69	((systematic or critical) adj (literature or narrative or qualitative or quantitative or evidence or evidence based or Cochrane) adj (review* or search*)).ti.	383
70	evidence based review.ti.	348
71	(metaanaly* or meta analy*).ti.	11307
72	exp "Review"/	1438617
73	exp "Review Literature as Topic"/	4002
74	evidence based review.ti.	348
75	exp meta-analysis/	19982

76	(metaanaly* or meta analy*).ti.	11307
77	67 or 63 or 71 or 70 or 68 or 72 or 65 or 74 or 75 or 64 or 61 or 58 or 59 or 69 or 60 or 66 or 73 or 76 or 62	2551224
78	("2008" or "2005" or "2007" or "2004" or "2006").yr.	3008465
79	english.lg.	13875810
80	"pe?diatric*".ti,ab.	116792
81	56 or 80	2558326
82	81 and 19 and 44	49031
83	82 not 77	46242
84	83 and 78	18423
85	84 and 79	17001

PychINFO

#	Searches	Results
1	"quality of life".sh.	9759
2	quality of life.ab,ti.	13383
3	disabilities.sh.	2317
4	(health adj status).ab,ti.	5944
5	functional analysis.sh.	340
6	(function* adj abilit*).ab,ti.	841
7	adaptive behavior.sh.	1064
8	(health adj status).ab,ti.	5944
9	(function* adj status).ab,ti.	1613
10	"disabilit*".ab,ti.	40248
11	(disabilit* adj function*).ab,ti.	41
12	(function* adj health*).ab,ti.	348
13	adaptive behavio?r.ab,ti.	2353

14	(school* adj health*).ab,ti.	418
15	(school* adj function*).ab,ti.	293
16	("pre school*" or "pre-school" or "preschool").ab,ti.	18264
17	school. ti. {No Related Terms}	29251
18	exp "activities of daily living"/ or exp ability level/ or exp activity level/ or exp assisted living/ or exp daily activities/ or exp habilitation/ or exp hygiene/ or exp independent living programs/ or exp physical mobility/ or exp rehabilitation/ or exp self care skills/	51429
19	school.ab.	123079
20	"activities of daily living"/ or ability level/ or activity level/ or assisted living/ or daily activities/ or habilitation/ or hygiene/ or independent living programs/ or physical mobility/ or rehabilitation/ or self care skills/	22998
21	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20	237864
22	disability evaluation.sh.	232
23	evaluation.sh.	7750
24	exp Treatment Effectiveness Evaluation/	7385
25	exp treatment outcomes/	15894
26	self report.sh.	6603
27	intervention.sh.	4700
28	school based intervention.sh.	1536
29	family intervention.sh.	544
30	group intervention.sh.	175
31	crisis intervention.sh.	2128
32	evaluation.ab,tm,ti.	83170
33	("self report*" or "self-report*").ab,tm,ti.	36866
34	questionnaire.tm.	11329
35	report.tm.	845
36	"paper and pencil".ab,ti.	2124

37	"questionnaire*".ab,ti.	111805
38	(outcome adj measure).ab,ti.	1668
39	"treatment outcome".ab,ti.	4586
40	"therapy outcome".ab,tm,ti.	632
41	measurement.sh.	26165
42	(outcome* adj evaluation*).ab,ti.	756
43	(outcome* adj measure*).ab,ti.	7769
44	"measure*".ti.	33377
45	exp clinical trials/	997
46	exp School Based Intervention/	1536
47	exp Early Intervention/	4651
48	exp Family Intervention/	544
49	exp Group Intervention/ or exp Crisis Intervention Services/	1683
50	"intervention*".ti.	20834
51	"trial*".ti.	9202
52	(indices or index).ab,ti.	43242
53	scale*.ti. or scale*.ab.	158811
54	"Instrument*".ti,ab.	55598
55	(scale or index).tm.	32382
56	instrument.tm.	499
57	"Survey*".ab,tm,ti.	90941
58	"Intervention*".ab,ti.	96571
59	33 or 32 or 26 or 30 or 44 or 55 or 25 or 27 or 28 or 57 or 40 or 49 or 24 or 31 or 35 or 53 or 48 or 22 or 42 or 46 or 23 or 29 or 50 or 39 or 36 or 51 or 58 or 41 or 47 or 38 or 52 or 34 or 56 or 37 or 45 or 43 or 54	584393
60	neonatal birth 1 mo.ag.	6712
61	infancy 2 23 mo.ag.	27020

62	preschool age 2 5 yrs.ag.	61861
63	school age 6 12 yrs.ag.	139345
64	adolescence 13 17 yrs.ag.	189914
65	"toddler* ".ab,ti.	3024
66	("baby" or "babies").ab,ti.	5710
67	"teenager* ".ab,ti.	3211
68	"adolescen* ".ab,ti.	92430
69	"child*".ab,ti.	329875
70	infancy.ab,ag,ti.	8043
71	(preschooler* or Pre schooler or pre-schooler).ab,ti.	5752
72	"pe?diatric*".ab,ti.	8495
73	67 or 63 or 71 or 70 or 64 or 61 or 68 or 69 or 60 or 72 or 66 or 62 or 65	524195
74	59 and 21 and 73	51949
75	("2008" or "2005" or "2007" or "2004" or "2006" or "2009").yr.	183357
76	english.lg.	2064017
77	74 and 75 and 76	6569
78	((systemat* or critical) adj (review* or search*)).ti.	2414
79	((systematic or critical) adj (literature or narrative or qualitative or quantitative or evidence or evidence based or Cochrane) adj (review* or search*)).ti.	52
80	evidence based review.ti.	33
81	(metaanaly* or meta analy*).ti.	3654
82	exp "literature review"/	21722
83	(encyclopedia or edited book or authored book or dissertation abstract or book).pt.	517760
84	authored book.pt. or "literature review".md.	101779
85	book.pt.	249518

86	"literature review".md.	52699
87	meta analysis.md.	4868
88	"systematic review".md.	835
89	84 or 85 or 83 or 78 or 79 or 81 or 87 or 88 or 86 or 82	569260
90	(77 not (authored book or "literature review" or book or (encyclopedia or edited book or authored book or dissertation abstract or book) or ((systemat* or critical) adj (review* or search*)) or ((systematic or critical) adj (literature or narrative or qualitative or quantitative or evidence or evidence based or Cochrane) adj (review* or search*)) or (metaanaly* or meta analy*) or meta analysis or "systematic review" or "literature review" or "literature review")).ab,ag,ti.	6266
91	from 90 keep 2	1

CINAHL

#	Query	Limiters/Expanders	Last Run Via	Results
S83	S82	Limiters - Exclude MEDLINE records Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	3039
S82	S81	Limiters - Abstract Available Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	9913
S81	S80 and S79 and S78	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	11585
S80	LA english	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	1880150
S79	PY 2004 or PY 2005 or PY 2006 or PY 2007 or PY 2008	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	819406
S78	S63 not S77	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	23405
S77	S76 or S75 or S74 or S73 or S72 or S71 or S70 or S69 or S68 or S67 or S66 or	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search	284964

	S65 or S64		Database - CINAHL	
S76	literature review +	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	7978
S75	(MH "Systematic Review") or (MH "Literature Review+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	6611
S74	AB (metaanaly* or meta analy*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	4037
S73	TI (metaanaly* or meta analy*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	2867
S72	TI (review* or search*) or TI (systemat* or critical)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	58590
S71	PT systematic review	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	12844
S70	PT practice guidelines	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	4271

S69	PT letter	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	67127
S68	PT Directories	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	2919
S67	PT commentary	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	88650
S66	PT case study	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	98496
S65	PT Book review	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	2754
S64	PT Biography	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	4299
S63	S62 and S42 and S22	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	25823
S62	S61 or S60 or S59 or S58 or S57 or S56 or S55 or S54 or S53 or S52 or S51 or	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search	287578

	S50 or S49 or S48 or S47 or S46 or S45 or S44 or S43		Database - CINAHL	
S61	AB (baby or babies) or TI (baby or babies)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	9241
S60	AB adolescen* or TI adolescen*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	25849
S59	AB infant* or TI infant*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	24666
S58	AB child* or TI child*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	106837
S57	AB (pediatric* or peadiatric*) or TI (peadiatric* or pediatric*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	22535
S56	AB teenager* or TI teenager*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	1913
S55	AB (preschooler* or pre-schooler* or pre schooler*) or TI (preschooler* or pre-schooler* or pre schooler*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	785

S54	AB toddler* or TI toddler*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	1489
S53	(MH "Adolescence+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	114199
S52	(MH "Child, Disabled")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	4271
S51	(MH "Child, Preschool")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	57243
S50	AG preschool	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	57243
S49	AG adolescence	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	114199
S48	AG child	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	178006
S47	(MH "Child+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search	181328

			Database - CINAHL	
S46	MW Disabilities	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	4341
S45	CR child	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	701
S44	AG infant	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	81039
S43	(MM "Child, Disabled")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	3068
S42	S41 or S40 or S39 or S38 or S37 or S36 or S35 or S34 or S33 or S32 or S31 or S30 or S29 or S28 or S27 or S26 or S25 or S24 or S23	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	353027
S41	TI trial*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	21048
S40	AB Intervention or TI Intervention	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	48113

S39	MW Early Childhood Intervention	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	1574
S38	(MH "Clinical Trials+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	66966
S37	(MH "Self Report")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	18018
S36	IN self report* or self-report*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	783
S35	IN questionnaire*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	24825
S34	(MH "Research Instruments+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	212207
S33	(MH "Questionnaires+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	96483
S32	TI health indicator* or AB health indicator*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search	418

			Database - CINAHL	
S31	TI assessment*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	23133
S30	TI evaluation*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	21288
S29	AB (function* and assessment*) or TI (function* and assessment*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	11154
S28	(MW "Functional Assessment")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	10310
S27	(MH "Disability Evaluation+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	3935
S26	(MH "Patient Assessment+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	28680
S25	AB outcome* measur* or TI outcome* measur*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	19646

S24	AB outcome* assessment* or TI outcome* assessment*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	778
S23	MW outcome assessment	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	7906
S22	S21 or S20 or S19 or S18 or S17 or S16 or S15 or S14 or S13 or S12 or S11 or S10 or S9 or S8 or S7 or S6 or S5 or S4 or S3 or S2 or S1	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	254987
S21	TI school* and AB school*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	6674
S20	TI rehabilitation*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	15098
S19	(MH "Rehabilitation+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	89646
S18	AB (daily and function*) or TI (daily and function*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	5486
S17	AB ("ADL" or activit* of daily living*)	Search modes - Boolean/Phrase	Interface - EBSCOhost	4405

	or TI ("ADL" or activit* of daily living*)		Search Screen - Advanced Search Database - CINAHL	
S16	(MH "Activities of Daily Living+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	16119
S15	(MH "Assisted Living")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	978
S14	AB (school* and function*) or TI (school* and function*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	1965
S13	AB (Function* and disabilit*) or TI (function* and disabilit*)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	5881
S12	AB (Function* and health*) or TI (function* and health)	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	19102
S11	MW Function	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	5855
S10	(MW "Family Functioning")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	1148

-				
S9	AB School Health or TI School Health	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	1190
S8	(MW "School Health")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	10068
S7	AB health status or TI health status	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	6630
S6	MW "Child, Disabled"	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	4271
S5	(MH "Health Status") or (MH "Health+")	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	89250
S4	MW quality of life	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	21315
S3	MW health status	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	17729
S2	AB disabilit* or TI disabilit*	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search	27153

			Database - CINAHL	
S1	AB quality of life or TI quality of life	Search modes - Boolean/Phrase	Interface - EBSCOhost Search Screen - Advanced Search Database - CINAHL	21280

Final Discussion:

The work of this thesis is centred on the ICF. There is, however, another biopsychosocial approach to health that also has a classification system: the Disability Creation Process (DCP) model (1, 2). The DCP demonstrates the *process* of disablement while the ICF presents a snapshot in time of the *factors* that contribute to disability. In the DCP, disability is created when social participation is limited (1, 2). The DCP does have a classification system but it is currently available in only two languages.

The ICF conceptualizes disability as a negative consequence or a breakdown of functioning. In the ICF, one's body functions can be as relevant to health as participation (with no emphasis on one health domain over another) (3, 4), and this approach is useful for clinical and community contexts. The ICF has been tested and validated in many languages and countries. Being endorsed by the WHO is one of the most useful elements of the ICF/ICF-CY, granting international credibility to the analyses based on the framework and classification.

Conceptual implications of using WHO definitions.

Many have heralded the ICF/ICF-CY as a triumph of biopsychosocialism, a new way of thinking about health (5-13). The advantages of the ICF/ICF-CY have been articulated in this thesis, and by others, and do not need to be repeated here. The philosophical underpinnings of the ICF/ICF-CY are less often appreciated. The ICF/ICF-CY is based on a philosophy called realism (14), a subcomponent of which is universalism (15,16). Realism means that ideas about health and disability in the ICF/ICF-CY are applicable to all people. Universalism in the ICF means that all humans experience disability to greater or lesser degrees at some point in their lives depending on a variety of factors, only some of which are medical in nature (15,16). These philosophies conflict with popular post-modernist or relativist views that are widely espoused in the social sciences. Post-modern philosophies posit that the experience of health is too socially contextual and individually varied to be defined according to universalistic principles. The ICF approach contests a relativist view by proposing that health can be universally defined so long as biological, psychological, and social elements are taken into account using an interactionist approach.

The implication of the ICF/ICF-CY is that disability can be eradicated for *every* human, and health restored, so long as the interaction between people's body functions, activities and roles, and their context, is synergistic. Adopting the ICF/ICF-CY

implies that the allocation of resources and priorities for child health interventions will shift from medical services to include caregiver supports, educational opportunities and community supports. In the ICF conception, health services based primarily or solely in medical paradigms will be insufficient to promote child health.

A broad biopsychosocial view of health that is found in the ICF/ICF-CY does not necessarily mean that making children healthier will always make their lives better. Health is a resource for everyday life but it is only one element among many that individual children and families might value. Other elements of life that vary in importance from child to child or from family to family include human rights, material wealth, and spiritual needs(17). Depending on the environment, there are adolescents who place more value on their human rights than their physical safety and they are willing to risk health for future life opportunities. This phenomenon was observed by the recent youth movements emerging in Tunisia and Egypt that were directed at overthrowing political regimes deemed responsible for limiting life opportunities (18). Taking the value that individual children and families place on different health domains or life domains into account elevates the concept under discussion from health to HRQOL or QOL. The conceptual ideas presented in this thesis about health and QOL are often accepted as definitions but they are not as easily adopted in health service delivery or evaluation activities.

Methodological issues elicited by the WHO content analysis

I approached the use of the WHO content analysis with conceptual implications about health and QOL in the back of my mind while placing the potential for solving measurement problems at the forefront of my research activities. This thesis has demonstrated that overall, health and QOL instruments are not focused on body structure and function and they do incorporate activity and participation domains in their measures. It is the contextual factors (i.e., environmental and personal factors) surrounding health that are not often measured.

The most disconcerting result of the thesis is the misconception by child health researchers that they are measuring HRQOL/QOL when they are measuring health status. How did this gap in measurement use versus content occur, and what are the implications as to how we understand validity in health status and QOL measurement?

Traditionally, validity takes a three-pronged approach: 1) content validity requires a test to be representative of a construct and have items that overlap with that

construct, while 2) construct validity seeks to validate the construct being measured by determining the extent to which it behaves relative to other similar or contrasting constructs in an expected manner. Finally, 3) criterion validity determines how well the proxy or substitute instrument performs relative to the perfect instrument that captures the criterion (usually called the 'gold standard') (19). This approach to validity, along with other standards proposed in the literature (e.g. COSMIN standards) (20), has prompted health and QOL instrument developers to take a checklist approach to instrument validation. During this time a fundamental problem with comparability of instruments has arisen and confounded the literature on child health.

In contrast, unified approaches to validity have been proposed by methodologists such as Messick, Cronbach and Kane (21-23). The unified approach places the emphasis on processes of instrument validation relative to a variety of contexts and purposes. These scientists have a variety of views on how to unify validity but all share the notion that there are facets (content, construct, criterion, and others) that require greater or lesser emphasis depending on the measurement situation. Messick, for example, proposes that all facets of validity can be subsumed under construct validity, emphasizing the process of developing convergent and discriminant evidence in the test validation process (24,25).

Unfortunately, in the absence of a strong theoretical approach, the exercise of comparing a target construct to other constructs can become quite circular. I believe this is precisely what has occurred in the field of child health and QOL instrument development in the last fifteen years. The conceptual incongruity between the instruments was pushed aside because the correlation coefficients observed were adequate relative to other related but poorly theoretically defined instruments.

The hypothetico-deductive model of scientific theory proposed by Cronbach and Meehl (1955) overcomes the reliance on one facet of validity over another (21). In the hypothetico-deductive model the instruments are only one piece of the puzzle in the validation process. If the observations collected with the instruments were incompatible with the theory that was being tested, then either the theory was insufficient or the instruments were not appropriate, or both(21). In the body of child health research reviewed in Chapter 7, the theoretical rationale behind measuring health status or QOL was rarely articulated. Instead of testing a theory through hypotheses, researchers were often focused on answering one particular research question. The difficulty with doing health services research in this way is that there are insufficient time and resources to answer every important research question. Therefore every instance of health status and QOL measurement should be set up as a hypothesis to test a theory to some degree in order for health services research to be scientifically useful. By neglecting a theoretical approach, instrument users have missed identifying a fundamental problem standing in the way of their ability to interpret and compare the data collected with these instruments. The fundamental problem is that it is unclear what is being measured. I believe that this is an example of what social psychologist Kurt Lewin meant when he said: "There is nothing more practical than a good theory" (26).

The most appealing and personally heartening aspect of a unified validity approach is the discussion about consequential validity proposed by Messick (23). Consequential validity means that the evaluation, interpretation, use and most importantly social consequences of an instrument all affect validity. This facet of validity demands that both developers and users consider how the results of the instruments they employ affect social outcomes. Interpreting the results of this thesis through the lens of consequential validity, indicates that it is improper for a developer to create an instrument and place it in the public domain when there is still ambiguity about what theoretical approach to health or QOL is measured through use of that instrument. A discussion of the PedsQL will illustrate the points raised here about theoretical definitions and consequential validity.

In its initial stages the PedsQL was called a QOL instrument (27), and later an HRQOL instrument(28). There was a great deal of information provided on the populations used to develop the questions in the various versions of the PedsQL but little information on what constituted a QOL or HRQOL instrument or what theories these instruments were testing. Psychometric information about the PedsQL can be accumulated, but without theoretical guidance, the meaning of such scores cannot be interpreted.

Now imagine a situation where a service provider compares one intervention to another and decides to fund one on the basis that it resulted in greater changes in the PedsQL score than the other. The decision to base a choice of intervention on the grounds it resulted in a larger score change is unclear, atheoretical and therefore unethical.

There are other ethical issues brought forward by the methods used in this thesis, all of which fit under the umbrella of consequential validity. In Chapter 4 we

learned that the instruments pose largely negatively-worded questions that can affect a child's self-perception or a parent's view of their child with respect to many sensitive areas of life such as family functioning or the burden of the child on the family (29). These negative questions have social consequences but they also affect content validity. For example, negatively phrased items about activities and participation only probe the experience of disability, not functioning. In this situation of negative phrasing, the ethical aspects of measurement are connected to what exactly is being measured. Thus ethics and content are intertwined and both affect validity.

In Chapter 6 we learned that the health utility instruments (HUI & EuroQOL) are better defined theoretically than the other popular health status instruments (i.e., PedsQL and CHQ). However, no discussion by the developers of the HUI and the EuroQOL instruments about the ethics of assessing a child's health based on the preference scores of adults was found. Of even greater concern is the assessment of the health of a child who has a chronic condition, and who has likely prioritized health concerns in a different manner than people in the general population. For example, if a child with cerebral palsy cannot walk, and has strabismus, but experiences good mobility with the use of a wheelchair, they can still receive a HUI score to say their health is equivalent to death. Even more problematic, the improvements to their functioning made possible by their wheelchair will not be detected by the HUI. Applying a health utility approach to evaluate programs providing service to children with chronic issues is potentially unethical and a violation of consequential validity.

Content mapping using the standardized linking method

The content mapping method reviewed, revised and applied in this thesis has challenges, strengths and areas for growth that require as much thought as the implications of using the method. Applying the method almost always evokes questions about subjectivity versus objectivity as well as how to interpret reliability scores between raters in the first iteration of the linking process.

The process of linking to the ICF/ICF-CY has been criticized as being subjective because there is a human judgement involved which is not present in what physicians call *objective tests*. Examples of objective tests include blood pressure or laboratory blood investigations that do not involve the judgement of a person. Interestingly, in everyday medical culture, x-rays are considered objective tests despite the multiple (subjective) considerations a radiologist must take into account when interpreting a scan. Thus the *objective* test in medicine has come to mean an evaluation by a clinician, while the report of a patient is subjective regardless of the controls put in place to obtain a measurement. The implications are that clinicians are often unaware of the parameters that make a measurement 'objective' so the term should be approached with caution.

In measurement, however, the judgment of a person is only one potential source of error among many in assigning a categorical or numerical value to something observed. Error can be produced by a problem in the rater's judgment, from the instrument itself or even from the way an assessment is administered. In the linking method, appraising the process by which a rater arrives at an ICF category or a WHO perspective is systematically defined in order to reduce error. Rothstein cautions against confusion between what is a subjective concept (an experience that varies from person to person) versus a subjective measurement (a score or rating that was not systematically contained) (30). Thus the distinction between subjective experiences versus subjective measurements should be made when performing the linking process.

In the first steps of linking, a well trained rater must decide what an item is about. Doing this before one rater's ICF category is compared to another rater's category is essential for bringing forward many plausible candidates for interpreting an item. This comparison between the first impression of each linker is the basis for obtaining an agreement statistic. The thesis intimates that a kappa between 0.6-0.8 is sufficient to show that linkers are following the same system of obtaining ICF categories; the left-over non-agreement reflects true initial differences of opinion about an item (which is encouraged and desired). The final list of ICF/ICF-CY categories presented in a manuscript is exposed to multiple sources of input before it can be presented.

I believe that individuals, who work with the linking method (including myself) need to resist a temptation to overly bind the linking process with rules. As observed by Kane(22):

"standardization can be effective in controlling irrelevant variability, but it also restricts the range of observations included in the measurements relative to those that are potentially relevant to proposed interpretations and uses." (p. 20)

Thus, adding more "rules" to the linking process can increase the strength of the initial reliability between raters; the question remains whether this is a desirable practice. Increasing the reliability of linking can effectively serve to reduce the overall validity of the process.

The agreement obtained in the first phase of linking ICF/ICF-CY categories or a WHO perspective indicates variation between raters but also speaks to the clarity of the item. The more agreement that is expressed between well trained raters, the clearer the item in general. Thus well constructed items are more likely to show high agreement in ICF/ICF-CY linking. Those items that evoke consensus are the items that are better candidates for content equivalence. However, the observations that have been reported here are merely hypotheses that need to be empirically tested in future work.

Next steps following the thesis

Future work based on this thesis should develop along two lines: first, knowledge translation and second, further methodological work. The portion of this work that is valuable for knowledge translation is explicating the conceptual differentiation between health and QOL. Also, it would be ideal to help clinicians and community groups map their desired outcomes to existing instruments in order to describe the children they serve or to help them evaluate the interventions they perform.

The methods in this thesis are important for comparing health status or QOL data across studies, populations, services, and interventions. The linking method and the ICF-CY can be used as a platform for merging data sources across languages, clinical situations or settings. Certain items collected in one clinic using the PedsQL could be compared to the equivalent items collected in a different clinic with the CHQ. The advantage of this approach is that programs might not need to abandon their currently existing tools but they can supplement the domains of health and QOL that are important to their purposes with new instruments (following a validation process).

I hope that the work started here will give rise to a program of research where comparison of child health and QOL data from multiple sources will be possible. The expectation is that there will be a world with publically owned platforms for measurement that circumvent the need for privately owned data. This way programs and services will be able to show the value of their interventions regardless of the amount of resources they can devote to the measurement process, using a biopsychosocial view of health and a well-defined approach to health or QOL.

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