Paediatric health related quality of life : a European perspective : instrument development, validation, and use in clinical practice

Baars, R.M.

Citation

Baars, R. M. (2006, March 30). *Paediatric health related quality of life : a European perspective : instrument development, validation, and use in clinical practice*. Retrieved from https://hdl.handle.net/1887/18420

Paediatric Health Related Quality of Life: a European Perspective

Instrument development, validation, and use in clinical practice

Rolanda M. Baars

The DISABKIDS project described in this thesis was financially supported by:
European Commission (QLG-CT-2000-00716).

Cover photo: R.M. Baars (Vietnam)
Lay-out and printing: Febodruk BV, Enschede

ISBN 90-9020393-1

Paediatric Health Related Quality of Life: a European Perspective

Instrument development, validation, and use in clinical practice

Proefschrift

ter verkrijging van

de graad van Doctor aan de Universiteit Leiden,

op gezag van de Rector Magnificus Dr. D.D. Breimer,

hoogleraar in de faculteit der Wiskunde en

Natuurwetenschappen en die der Geneeskunde,

volgens besluit van het College voor Promoties

te verdedigen op donderdag 30 maart 2006

klokke 14:15 uur

door

Rolanda Maia Baars

geboren te Puttershoek
in 1970

Promotiecommissie

Promotor: Prof. Dr. J.M.Wit

Co-promotor: Dr. H.M. Koopman

Referent: Prof. Dr. R.J.B.J. Gemke (Vrije Universiteit Amsterdam)

Overige leden: Prof. Dr. S.P. Verloove-Vanhorick
Prof. Dr. A.A. Kaptein

“Que sera, sera”

For what the past has brought,
the present is,
and the future may bring.

Table of contents

1

General introduction and outline of the thesis

> The goal of all medical interventions should be to promote the patient's health, and thus, to increase their health related quality of life (H.I. Brunner, 2003).

General Introduction and outline of the thesis

The DISABKIDS project is a European collaboration of clinicians and investigators that received funding from the European Commission in 2001. Over the last four years the DISABKIDS project's aim was to cross-nationally develop a new European health related quality of life (HRQoL) instrument for children and adolescents with a chronic medical condition [1](Box 1). Some of the steps taken during the developmental process of the European DISABKIDS HRQoL instrument for children and adolescents with a chronic medical condition are described in this thesis. Data from the asthma population is a recurring theme in most chapters as the Dutch DISABKIDS centre operated as asthma consultant.

Four criteria must be met:

- If it occurs in children aged 0 to 18 years inclusively
- If its diagnosis is based on medical scientific knowledge and it can be diagnosed using reproducible and valid methods or instruments according to the professionals
- If it is not (yet) curable
- If it has been present longer than three months or if it will very probably last longer than three months, or if it has occurred three times or more during the past year

Box 1. Criteria used to define when a disease or condition is considered chronic in childhood [2].

This introduction will first supply the readers with an informative background on HRQoL research. The general HRQoL principles will be explained together with the impact of paediatric asthma on the life of a child or adolescent. The development of HRQoL instruments is also explained and the aim and developmental steps of the European DISABKIDS project are described.

Health related quality of life

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"[3]. This realisation has initiated the discussion on how to improve and measure health. At the same time the shift in mortality and morbidity rates of some chronic medical conditions (e.g. cystic fibrosis, cancer and metabolic disorders) have encouraged the discussion on how to improve quality of life (QoL) in patients. The first hit on QoL in PubMed* dates back to 1966 and although QoL has become a general concept in research and daily life since then, it is still an elusive concept.

---

* The National Library of Medicine's search service that provides access to an electronic database of over 15 million citations in biomedical literature dating back to the 1950's.

Definitions of QoL often include aspects as: "the perceptions of physical, psychological, social, cognitive, functional and behavioural dimensions of well-being and function as perceived by the person concerned" [1]. The World Health Organisation QoL (WHOQoL) group defines QoL as: "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns. It is a broad-ranging concept, incorporating in a complex way individuals' physical health, psychological state, level of independence, social relationships, personal beliefs and their relationships to salient features of the environment" [4]. When assessing the impact of health and illness on a person's life one hopes to measure the subjective perspective called health related quality of life (HRQoL) [5]. HRQoL can be defined as: "a psychological construct which describes the physical, mental, social, psychological and functional aspects of well-being and function from a patient perspective" [6]. HRQoL assesses the patient's functioning from a broader scope than clinical measures alone to help understand the effects of a medical condition on a patient's well-being. Not only the objective aspects related to illness and treatment are assessed, but also the more subjective concepts surrounding a condition, for example the patient's perception of their emotional and social situation [7,8].

Over the last decades there has been an increase in publications on HRQoL assessment in children and adolescents, healthy or with a chronic medical condition [9,10]. In general one can distinguish two main areas in which HRQoL assessment can be used: research and clinical practice. Most publications on paediatric HRQoL have psychometrically described the development and validation of questionnaires [11-17]. Numerous evaluative studies of children and adolescents with chronic conditions have been published [18-25]. There is also an increased interest in implementing HRQoL assessment in paediatric clinical trials, for instance when choosing between medications or comparing benefits or impact of a certain treatment regime [26-31].

The latest challenge is to implement HRQoL questionnaires in paediatric clinical practice for individual assessment [32-34]. There is an awareness of the importance of not only treating a child's physical condition, but also incorporating the psychological and social aspects, acknowledging that the child's total well-being is affected by a medical condition [35]. One reason for this awareness may be that objective measures of disease severity (e.g. pulmonary function) have shown modest correlations with how a patient feels and are thus seen as insufficient determinants of health status. The HRQoL of one patient can also differ extensively with another, even when their objective clinical parameters are similar [36-40].

There is evidence that HRQoL assessment can aid patient management in adults. Benefits include monitoring changes in patients, improving the clinician-patient relationship and communication, and potential screening for problems [41-48]. While HRQoL assessment is on the rise in adult clinical practice this is not being imitated in the paediatric setting. Although investigators have suggested to assess HRQoL in paediatric clinical practice, proven relevance of HRQoL implementation in paediatric clinical practice is unavailable and there are few indications that routine HRQoL assessment is being included in paediatric clinical practice [30,33,34,49-51]. Thus it is only assumed that, as in adults, individual HRQoL assessment in paediatric clinical practice can improve the clinician-patient relationship, facilitate communication, provide a complete impression of the child or adolescents health status, identify existing problem areas and initiate necessary intervention. Why HRQoL assessment is not widely included in paediatric clinical practice may depend on several factors. Some of these issues include the lack of valid and reliable questionnaires, minimal evidence of the benefit to patient care, the limited availability of disease specific measures, limited self-completion questionnaires for children and adolescents adapted to their age group, insufficient information on interpretation and use of questionnaires, and cultural barriers [9,30,52].

The clinician can add to these barriers through insufficient knowledge of HRQoL, constraints on financial and human resources, the belief that HRQoL assessment is unimportant, unawareness of available questionnaires or inexperience with questionnaires [32,42,47,53,54]. Encouraging is the confirmation that clinicians do consider their patient's HRQoL to be important and are interested in implementing HRQoL assessment in clinical practice [32,53-56]. This is reason enough to further develop and improve HRQoL assessment and eliminate any obstacles that stand in the way of implementation in clinical practice. Moreover, the use of HRQoL questionnaires is likely to improve if clinicians have access to valid and easy to use questionnaires with clinical value in areas they find important.

HRQoL and asthma

The most common chronic medical condition among children is asthma. Its prevalence varies greatly between countries, with a high prevalence in Australia, New Zealand and Ireland (20-28%) and a low prevalence (mostly under 5%) in countries in South-East Asia and Northern and Eastern Europe [57]. The majority of children develop their asthma before five years of age [58,59]. Asthma is a chronic inflammatory disease of the airways. A diagnosis of asthma is mainly based upon clinical observations [60]. In susceptible individuals inflammation and airway narrowing can cause recurrent episodes of wheezing, chest tightness, coughing (typically nocturnal or exercise related) and shortness of breath [60,61]. These episodes are usually reversible either spontaneously or with treatment [60] and can arise spontaneously or can be triggered by factors such as pollen, cigarette smoke, house dust mite, viral infections, exercise or weather changes [61-63]. In cases of acute asthma exacerbations symptoms can be more severe and in some cases even life threatening [58,60]. Asthma is linked to family atopy, and children with asthma are prone to allergies [61,62]. Asthma management consists of monitoring the course of the disease, avoiding or eliminating triggers (allergens, respiratory infections, irritants, chemicals, physical activity and emotional stress) that influence asthma symptoms or exacerbations, educating patients and pharmacological therapy [60,63]. Pharmacological treatment is focussed on suppressing inflammation and reversing bronchoconstriction. Initial treatment usually includes inhaled bronchodilators as reliever and, if this is insufficient, inhaled anti-inflammatory agents as protector in daily doses [61,63].

Asthma can impact a child or adolescent's life in several areas [64]. Children need to cope with taking their daily medication and some may be concerned about possible adverse effects. The symptoms they experience can lead to physical limitations, for example during sport or play. Nocturnal symptoms may disturb their sleep [60]. Children might miss school days or experience poor school performance [60,65]. Some may experience fear or panic due to the often sudden, life-threatening nature of the attacks [66,67]. Their social activities may be limited due to the necessity of avoiding potential trigger factors (e.g. cigarette smoke, house dust mite) or as a result of physical limitations [14]. The above factors can also add to the fear of being rejected by peers because of feeling "different" [68]. Children can be troubled because they cannot integrate fully with their peers, making them feel isolated and left out. Any of these experienced limitations in physical, social and emotional functioning can cause feelings of anger, depression, anxiety, embarrassment or frustration [69-72]. A study by Forrest et al. (1997) has also shown that teenagers with asthma experience more physical and emotional problems, lower perceived well-being, more activity restrictions and more negative behaviours that threaten social development than teenagers with no asthma [73]. A meta-analysis by McQuaid et al (2001) shows that children with asthma have more adjustment difficulties and more internalising and externalising problems related to the severity of their asthma than healthy children [74]. Asthma can disrupt the family routines and cause an increase in family stress [75] but can also be affected by family factors [76]. Parents may overprotect a child with asthma, creating the possibility of restricting them in their normal daily activities [65,77].

Overall, there are many factors that can cause extra stress and influence a child's or adolescent's mental and physical behaviour. These factors make it important for clinicians to become aware of the impact of asthma on a child or adolescent and their family. HRQoL questionnaires for children and adolescents are therefore likely to become increasingly important for the future of paediatric clinical practice and research.

Developing HRQoL instruments

An increasing number of paediatric generic and condition-specific HRQoL questionnaires have been developed since the first publications on paediatric HRQoL in the 1970's [5,9,78]. Some of the more well-known generic questionnaires include the Pediatric Quality of Life Inventory™ (PedsQL™4.0)[79], the Child Health Questionnaire (CHQ)[80] and the Child Health and Illness Profile (CHIP)[81]. Frequently used measures of HRQoL in children or adolescents with asthma are the Paediatric Asthma Quality of Life Questionnaire (PAQLQ)[36] and the Childhood Asthma Questionnaire (CAQ)[13,82].

In general the development of each new HRQoL questionnaire follows similar phases [83]. First, one should determine if there is a need for a new questionnaire [32]. Secondly a basic consensus within the research group should be reached on the content and structure of the questionnaire [84]. Choices include what one wants to measure, which group to test, their age range, their specific situation, and how long the questionnaire should be. Subsequently one should consider if a questionnaire is to be used for instance for individual assessment, group comparison or national screening surveys [85,86]. Nowadays there is a preference for cross-national questionnaires for use in multi-national clinical trials or to enable comparisons between different cultures or social groups [87]. Decisions need to be taken on whether to develop the instrument in one country and translate it for use in other countries (sequential), or develop it cross-nationally (simultaneously)[88].

The next phase is to generate questionnaire items through a top-down or bottom-up developmental process. Items can be collected in an expert consensus meeting (top-down development) and include clinical experience, literature or available questionnaires [11,15,18]. While the clinician's and even parent's opinion on HRQoL was regarded as sufficient, it is now known that their opinion can be different to that of the child or adolescent's [33,89-92]. For this reason the bottom-up (patient-derived) process is often applied in which the group of interest (e.g. children with asthma) provide the aspects they find important. Patient-derived methods can include interviews, focus group discussions or surveys [14,93,94]. The statistical data on selected items, collected in a pilot test, can then help to reduce the items and test the domain structure for the final instrument.

As mentioned before, there have always been restrictions to the use and availability of paediatric HRQoL questionnaires. So, although there are several HRQoL questionnaires for children and adolescents, new questionnaires are still being developed or improved, either for new chronic conditions or for use in new situations. Another reason for the ongoing development of new questionnaires is the growing need for (translated) HRQoL instruments in cross-national multi-centre studies. The aim of the DISABKIDS project was to take existing limitations into account and cross-nationally develop a new European instrument through a bottom-up process, consisting of a chronic generic and several condition-specific modules, for use in children and adolescents with a chronic medical condition and their parents.

DISABKIDS

The DISABKIDS project is a collaboration of investigators from seven European countries (Figure 1) that set out to develop a new European HRQoL instrument for children and adolescents with a chronic medical condition [1]. Included in the project were children and adolescents in three age groups (4-7, 8-12, 13-16) with one of the following chronic medical conditions: asthma, juvenile idiopathic arthritis (JIA), atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes or epilepsy. Each country had a consulting role in one of the chronic medical conditions. The project was funded through the Fifth Frame work Research Programme ‘Quality of life and management of living resources’ of the European Community and was oriented towards three tasks:

1. Developing modules for assessing HRQoL in children and adolescents with chronic medical conditions.
2. Psychometrically testing the instruments in different countries.
3. Assessing the value of the DISABKIDS instrument by implementing and evaluating it in paediatric clinical practice.

Figure 1. Participating countries in the DISABKIDS project

The DISABKIDS project is unique due to the simultaneous cross-national development, the patient-derived bottom-up procedure, the modular design, the inclusion of seven chronic medical conditions, the wide age range (4-16 years) and the availability of a self-assessment and a proxy version. The instrument will be available in paper-pencil and as computer version in several languages.

The DISABKIDS project runs parallel to the KIDSCREEN project, which is an epidemiological research project that aims to develop and test a generic HRQoL questionnaire in primarily healthy children and adolescents [84,95].

The DISABKIDS and KIDSCREEN projects have defined and developed a three level modular instrument (Figure 2) by combining the following modules:

1. The KIDSCREEN generic module
2. The DISABKIDS chronic generic module
3. The DISABKIDS condition-specific module

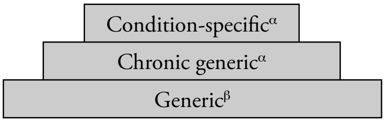


Figure 2. Modular design of the DISABKIDS$^{\alpha}$ and KIDSCREEN$^{\beta}$ instrument.

The generic module consists of items that are applicable to all children and adolescents, healthy or ill. This module is capable of measuring HRQoL across patient populations and can compare the outcome to a healthy population. The chronic generic module, as defined bij the DISABKIDS group, is applicable to any child or adolescent with a chronic medical condition (Box 1). Items relate to areas of life that are affected by chronic medical conditions. This module can be useful in situations where it is important to be able to measure HRQoL across different conditions and take into account common areas affected by chronic conditions. The condition-specific module assesses those aspects that are specific to patients with a certain chronic medical condition, often referred to as disease-specific. It can only compare between groups of patients with the same chronic condition but has the potential to identify smaller changes important to research or clinical practice [5,34,96,97]. This three modular design is unique to the DISABKIDS and KIDSCREEN projects and can supply the investigator or clinician with the opportunity to assess HRQoL at different levels.

International consensus was reached on the methodology of the questionnaire development. The procedure was derived from earlier experience of investigators in (international) instrument development [15,98-101] and consists of several work packages that reflect a stepwise instrument development procedure (Box 2).

WP 1: Literature review
WP 2: Focus groups
WP 3: Item development
WP 4: Translations
WP 5: Pilot study
WP 6: Analysis pilot study
WP 7: Field study
WP 8: Analysis field study
WP 9: Implementation and final results

Box 2. Work packages (WP) within the DISABKIDS project

DISABKIDS work packages

Literature Review (WP 1)

A literature search in Medline (1985-2000) was done for the identification of abstracts concerning HRQoL assessment in children and adolescents with a chronic medical condition. Available assessment instruments and known HRQoL dimensions were reviewed for their use in the project.

Focus Groups (WP 2)

Children and adolescents with asthma, juvenile idiopathic arthritis, atopic dermatitis, cerebral palsy, cystic fibrosis, diabetes mellitus and epilepsy participated in focus groups or interviews. Statements were collected from the literal transcripts for the item generation. A guideline manual insured that the groups were conducted in a similar method in all participating countries.

Item Development (WP 3)
The collected statements were used for the construction of the DISABKIDS instrument. The statements were divided into separate modules; a generic module, a chronic generic module and seven condition-specific modules. The statements underwent a reduction process to limit the amount of items for the pilot instrument.

Translations (WP 4)
Guidelines were established to harmonise the translations across countries. Two translators independently conducted a forward translation of the English pilot items into the target language. The translations were reviewed for conceptual equivalence and a single forward translation was decided upon. A backward translation was then performed, which was compared to the original item for the final translation. This process was performed in all the participating countries and compared across languages.

Pilot Study (WP 5)
Data were collected for the psychometric analysis. Each condition was tested in at least two countries, while asthma was tested in all countries. Children and adolescents participated in a cognitive interview to determine the comprehensiveness, clarity and acceptance of the questions.

Analysis pilot study (WP 6)
Data were collected in an international SPSS[†] data file. The analysis was carried out at an international level using classical multi-scaling as well as modern psychometric methods. The final domains were determined and item numbers reduced through quantitative psychometric analysis and the qualitative cognitive interview.

Field study (WP 7)
Data were collected to test the psychometric performance of the pilot instrument in populations of children and adolescents with selected chronic conditions. Asthma was again tested in all countries.

Analysis field study (WP 8)
The final scale structure of the DISABKIDS instrument was tested, including the reliability, validity, retest-reliability and the construct validity.

Implementation (WP 9)
The DISABKIDS instrument was implemented and tested in several settings. Analyses are still in progress. A paper and computer version was made available in several languages.

[†] Statistical Package for the Social Sciences: software package used for conducting statistical analyses.

Outline of this thesis

In this first chapter we set out to explain some general principles related to HRQoL, asthma, developing a questionnaire and the DISABKIDS project (**Chapter 1**). The second chapter describes a survey done under the members of the Dutch Paediatric Association. The objective was to learn more about the perspectives of the paediatricians on HRQoL assessment in clinical practice. Knowing what the opinion of a future user group is helps to implement a new HRQoL questionnaire, like the DISABKIDS instrument (**Chapter 2**). The third chapter describes the applied patient-derived methodology. Focus groups and interviews were used to identify the relevant aspects of HRQoL from children and adolescents with chronic medical conditions. Statements for the chronic generic and condition-specific modules were generated through this patient-derived method. Focus groups have proven to give a good indication of what the patient finds important and therefore emphasises the importance of the DISABKIDS bottom-up procedure (**Chapter 3**). The results of the asthma focus groups and interviews, conducted in four European countries, are described in chapter four. Recurring themes are discussed and aspects related to living with asthma are described in a quantitative manner (**Chapter 4**). Qualitative data was collected from the cross-national DISABKIDS focus groups and interviews. The sequential reduction steps necessary to cut down the vast amount of collected statements for use in the pilot study are set out in chapter five (**Chapter 5**). The development of the seven condition-specific modules is described in chapter six. A stepwise analysis was applied including the statistical results from the pilot and field study, the cognitive interview and expert opinions (**Chapter 6**). The validation study of the asthma population is presented in chapter seven. The relationship between HRQoL, disease severity and existing questionnaires is evaluated (**Chapter 7**). The discussion is laid out in chapter eight in which limitations of questionnaire development and future suggestions for research are presented. Concepts regarding the implementation of individual HRQoL assessment in clinical practice are discussed, including limitations, necessary requirements and clinical value (**Chapter 8**).

References

1. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. Int.J.Rehabil.Res. 2002;25(3):197-206.
2. Mokkink, L. B., van der Lee, J. H., Grootenhuis, M. A., Offringa, M., van Praag, B. M. S., and Heymans, H. S. A. Omvang en gevolgen van chronische aandoeningen bij kinderen. 2, 1-95. 14-12-2004. Emma Kinderziekenhuis AMC.
3. World Health Organization. WHO constitution, 1948.
4. The WHOQOL group. The World Health Organization quality of life assessment (WHOQOL): position paper from the World Health Organization. Soc.Sci.Med. 1995;41(10):1403-9.
5. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol Assess 2001;5(4):1-157.
6. Ravens-Sieberer U, Bullinger M. Assessing health-related quality of life in chronically ill children with the German KINDL: first psychometric and content analytical results. Qual.Life Res. 1998;7(5):399-407.
7. Spieth LE, Harris CV. Assessment of health-related quality of life in children and adolescents: an integrative review. J.Pediatr.Psychol. 1996;21(2):175-93.
8. Testa MA, Simonson DC. Assesment of quality-of-life outcomes. N.Engl.J.Med. 1996;334(13):835-40.
9. Andelman RB, Zima BT, Rosenblatt AB. Quality of life of children: toward conceptual clarity. In: Maruish ME, editor. The use of psychological testing for treatment planning and outcomes assessment. Mahwah, NJ: Lawrence Erlbaum; 1999. p. 1383-413.
10. Eiser C, Morse R. The measurement of quality of life in children: past and future perspectives. J.Dev.Behav.Pediatr. 2001;22(4):248-56.
11. Gee L, Abbott J, Conway SP, Etherington C, Webb AK. Development of a disease specific health related quality of life measure for adults and adolescents with cystic fibrosis. Thorax 2000;55(11):946-54.
12. Griffiths AM, Nicholas D, Smith C, Munk M, Stephens D, Durno C et al. Development of a quality-of-life index for pediatric inflammatory bowel disease: dealing with differences related to age and IBD type. J.Pediatr.Gastroenterol.Nutr. 1999;28(4):S46-S52.
13. Christie MJ, French D, Sowden A, West A. Development of child-centered disease-specific questionnaires for living with asthma. Psychosom.Med. 1993;55(6):541-8.
14. Creer TL, Wigal JK, Kotses H, Hatala JC, McConnaughy K, Winder JA. A life activities questionnaire for childhood asthma. J.Asthma 1993;30(6):467-73.
15. Bullinger M, Von Mackensen S, Fischer K, Khair K, Petersen C, Ravens-Sieberer U et al. Pilot testing of the 'Haemo-QoL' quality of life questionnaire for haemophiliac children in six European countries. Haemophilia. 2002;8 Suppl 2:47-54.

16. Ronen GM, Streiner DL, Rosenbaum P. Health-related quality of life in children with epilepsy: development and validation of self-report and parent proxy measures. Epilepsia 2003;44(4):598-612.
17. Juniper EF, Guyatt GH, Epstein RS, Ferrie PJ, Jaeschke R, Hiller TK. Evaluation of impairment of health related quality of life in asthma: development of a questionnaire for use in clinical trials. Thorax 1992;47(2):76-83.
18. Austin JK, Smith MS, Risinger MW, McNelis AM. Childhood epilepsy and asthma: comparison of quality of life. Epilepsia 1994;35(3):608-15.
19. Graue M, Wentzel-Larsen T, Hanestad BR, Batsvik B, Sovik O. Measuring self-reported, health-related, quality of life in adolescents with type 1 diabetes using both generic and disease-specific instruments. Acta Paediatr. 2003;92(10):1190-6.
20. de Jong W, Kaptein AA, van der Schans CP, Mannes GP, van Aalderen WM, Grevink RG et al. Quality of life in patients with cystic fibrosis. Pediatr.Pulmonol. 1997;23(2):95-100.
21. Rabbett H, Elbadri A, Thwaites R, Northover H, Dady I, Firth D et al. Quality of life in children with Crohn's disease. J.Pediatr.Gastroenterol.Nutr. 1996;23(5):528-33.
22. Vila G, Hayder R, Bertrand C, Falissard B, De Blic J, Mouren-Simeoni MC et al. Psychopathology and quality of life for adolescents with asthma and their parents. Psychosomatics 2003;44(4):319-28.
23. Schneider JW, Gurucharri LM, Gutierrez AL, Gaebler-Spira DJ. Health-related quality of life and functional outcome measures for children with cerebral palsy. Dev. Med.Child Neurol. 2001;43(9):601-8.
24. De Clercq B, De Fruyt F, Koot HM, Benoit Y. Quality of life in children surviving cancer: a personality and multi-informant perspective. J.Pediatr.Psychol. 2004;29(8):579-90.
25. Sawyer MG, Reynolds KE, Couper JJ, French DJ, Kennedy D, Martin J et al. Health-related quality of life of children and adolescents with chronic illness-a two year prospective study. Qual.Life Res. 2004;13(7):1309-19.
26. Everden P, Campbell M, Harnden C, McGoldrick H, Bodalia B, Manion V et al. Eformoterol Turbohaler compared with salmeterol by dry powder inhaler in asthmatic children not controlled on inhaled corticosteroids. Pediatr.Allergy Immunol. 2004;15(1):40-7.
27. Gupta M, Aneja S, Kohli K. Add-on melatonin improves quality of life in epileptic children on valproate monotherapy: a randomized, double-blind, placebo-controlled trial. Epilepsy Behav. 2004;5(3):316-21.
28. Zimmerman B, D'Urzo A, Berube D. Efficacy and safety of formoterol Turbuhaler when added to inhaled corticosteroid treatment in children with asthma. Pediatr. Pulmonol. 2004;37(2):122-7.
29. Weintrob N, Benzaquen H, Galatzer A, Shalitin S, Lazar L, Fayman G et al. Comparison of continuous subcutaneous insulin infusion and multiple daily injection regimens in children with type 1 diabetes: a randomized open crossover trial. Pediatrics 2003;112(3 Pt 1):559-64.

30. Clarke SA, Eiser C. The measurement of health-related quality of life (QOL) in paediatric clinical trials: a systematic review. Health Qual.Life Outcomes. 2004;2(1):66.
31. Bender BG. Measurement of quality of life in pediatric asthma clinical trials. Ann. Allergy Asthma Immunol. 1996;77(6):438-45.
32. Baars RM, van der Pal SM, Koopman HM, Wit JM. Clinicians' perspective on quality of life assessment in paediatric clinical practice. Acta Paediatr. 2004;93(10):1356-62.
33. Williams J, Williams K. Asthma-specific quality of life questionnaires in children: are they useful and feasible in routine clinical practice? Pediatr.Pulmonol. 2003;35(2):114-8.
34. Juniper EF. How important is quality of life in pediatric asthma? Pediatr.Pulmonol. Suppl 1997;15:17-21.
35. Harding L. Children's quality of life assessments: a review of generic and health related quality of life measures completed by children and adolescnets. Clin.Psychol. Psychother. 2001;8:79-96.
36. Juniper EF, Guyatt GH, Feeny DH, Ferrie PJ, Griffith LE, Townsend M. Measuring quality of life in children with asthma. Qual.Life Res. 1996;5(1):35-46.
37. Rutishauser C, Sawyer SM, Bond L, Coffey C, Bowes G. Development and validation of the Adolescent Asthma Quality of Life Questionnaire (AAQOL). Eur. Respir.J. 2001;17(1):52-8.
38. Carranza R, Jr., Edwards L, Lincourt W, Dorinsky P, ZuWallack RL. The relationship between health-related quality of life, lung function and daily symptoms in patients with persistent asthma. Respir.Med. 2004;98(12):1157-65.
39. Rowe BH, Oxman AD. Performance of an asthma quality of life questionnaire in an outpatient setting. Am.Rev.Respir.Dis. 1993;148(3):675-81.
40. Janse AJ. Quality of life of chronically ill children. Perception of patients, parents and physicians [dissertation]. University of Utrecht; Department of Pediatrics; 2005.
41. Detmar SB, Muller MJ, Schornagel JH, Wever LD, Aaronson NK. Health-related quality-of-life assessments and patient-physician communication: a randomized controlled trial. JAMA 2002;288(23):3027-34.
42. Greenhalgh J, Meadows K. The effectiveness of the use of patient-based measures of health in routine practice in improving the process and outcomes of patient care: a literature review. J.Eval.Clin.Pract. 1999;5(4):401-16.
43. Taenzer P, Bultz BD, Carlson LE, Speca M, DeGagne T, Olson K et al. Impact of computerized quality of life screening on physician behaviour and patient satisfaction in lung cancer outpatients. Psychooncology. 2000;9(3):203-13.
44. Espallargues M, Valderas JM, Alonso J. Provision of feedback on perceived health status to health care professionals: a systematic review of its impact. Med.Care 2000;38(2):175-86.
45. Jacobs JE, van de Lisdonk EH, Smeele I, van Weel C, Grol RP. Management of patients with asthma and COPD: monitoring quality of life and the relationship to subsequent GP interventions. Fam.Pract. 2001;18(6):574-80.

46. Higginson IJ, Carr AJ. Measuring quality of life: using quality of life measures in the clinical setting. BMJ 2001;322(7297):1297-300.
47. McHorney CA, Earl BD, Jr. A qualitative study of patients' and physicians' views about practice- based functional health assessment. Med.Care 2002;40(11):1113-25.
48. Velikova G, Booth L, Smith AB, Brown PM, Lynch P, Brown JM et al. Measuring quality of life in routine oncology practice improves communication and patient wellbeing: a randomized controlled trial. J.Clin.Oncol. 2004;22(4):714-24.
49. Juniper EF. Health-related quality of life in asthma. Curr.Opin.Pulm.Med. 1999;5(2):105-10.
50. Buller H. Assessment of quality of life in the younger child: the use of an animated computer program. J.Pediatr.Gastroenterol.Nutr. 1999;28(4):S53-S55.
51. Sherman AC, Simonton S, Latif U, Nieder ML, Adams RH, Mehta P. Psychosocial supportive care for children receiving stem cell transplantation: practice patterns across centers. Bone Marrow Transplant. 2004;34(2):169-74.
52. Eiser C, Morse R. A review of measures of quality of life for children with chronic illness. Arch.Dis.Child 2001;84(3):205-11.
53. Morris J, Perez D, McNoe B. The use of quality of life data in clinical practice. Qual. Life Res. 1998;7(1):85-91.
54. Taylor KM, Macdonald KG, Bezjak A, Ng P, DePetrillo AD. Physicians' perspective on quality of life: an exploratory study of oncologists. Qual.Life Res. 1996;5(1):5-14.
55. Tanaka T, Gotay CC. Physicians' and medical students' perspectives on patients' quality of life. Acad.Med. 1998;73(9):1003-5.
56. Walsh DL, Emrich LJ. Measuring cancer patients' quality of life. A look at physician attitudes. N.Y.State J.Med. 1988;88(7):354-7.
57. The International Study of Asthma and Allergies in Childhood (ISAAC) Steering Committee. Worldwide variations in the prevalence of asthma symptoms: the International Study of Asthma and Allergies in Childhood (ISAAC). Eur.Respir.J. 1998;12(2):315-35.
58. Silverstein MD, Reed CE, O'Connell EJ, Melton LJ, III, O'Fallon WM, Yunginger JW. Long-term survival of a cohort of community residents with asthma. N.Engl. J.Med. 1994;331(23):1537-41.
59. Croner S, Kjellman NI. Natural history of bronchial asthma in childhood. A prospective study from birth up to 12-14 years of age. Allergy 1992;47(2 Pt 2):150-7.
60. National Institutes of Health. Guidelines for the diagnosis and management of asthma. National Heart Lung and Blood Institute, National Asthma Education and Prevention Program. publication no. 97-4051. 1997. Bethesda MD, US Department of Health and Human Services, National Institutes of Health.
61. British guideline on the management of asthma. Thorax 2003;58 Suppl 1:i1-94.
62. International consensus report on diagnosis and treatment of asthma. National Heart, Lung, and Blood Institute, National Institutes of Health. Bethesda, Maryland 20892. Publication no. 92-3091, March 1992. Eur.Respir.J. 1992;5(5):601-41.

63. Ross MH, Mjaanes CM, Lemanske R. Asthma. In: Rudolph CD, Rudolph AM, editors. Rudolph's Pediatrics. 21st ed. New York: McGraw-Hill; 2003. p. 1950-63.
64. Sennhauser FH, Braun-Fahrlander C, Wildhaber JH. The burden of asthma in children: a European perspective. Paediatr.Respir.Rev. 2005;6(1):2-7.
65. Hilton S. Management of childhood asthma in general practice. In: Christie MJ, French D, editors. Assessment of quality of life in childhood asthma. Chur, Switzerland: Harwood academic publishers; 1994. p. 47-56.
66. Townsend M, Feeny DH, Guyatt GH, Furlong WJ, Seip AE, Dolovich J. Evaluation of the burden of illness for pediatric asthmatic patients and their parents. Ann.Allergy 1991;67(4):403-8.
67. Creer TL, Stein RE, Rappaport L, Lewis C. Behavioral consequences of illness: childhood asthma as a model. Pediatrics 1992;90(5 Pt 2):808-15.
68. French DJ, Christie MJ, West A. Quality of life in childhood asthma: development of the Childhood Asthma Questionnaires. In: Christie MJ, French DJ, editors. Assessment of quality of life in childhood asthma. Chur, Switzerland: Harwood Academic Publishers; 1994. p. 157-80.
69. Cohen R, Franco K, Motlow F, Reznik M, Ozuah PO. Perceptions and attitudes of adolescents with asthma. J.Asthma 2003;40(2):207-11.
70. Juniper EF. Quality of life in adults and children with asthma and rhinitis. Allergy 1997;52(10):971-7.
71. Okelo SO, Wu AW, Krishnan JA, Rand CS, Skinner EA, Diette GB. Emotional quality-of-life and outcomes in adolescents with asthma. J.Pediatr. 2004;145(4):523-9.
72. Nocon A. Social and emotional impact of childhood asthma. Arch.Dis.Child 1991;66(4):458-60.
73. Forrest CB, Starfield B, Riley AW, Kang M. The impact of asthma on the health status of adolescents. Pediatrics 1997;99(2):E1.
74. McQuaid EL, Kopel SJ, Nassau JH. Behavioral adjustment in children with asthma: a meta-analysis. J.Dev.Behav.Pediatr. 2001;22(6):430-9.
75. Schulz RM, Dye J, Jolicoeur L, Cafferty T, Watson J. Quality-of-life factors for parents of children with asthma. J.Asthma 1994;31(3):209-19.
76. Kaugars AS, Klinnert MD, Bender BG. Family influences on pediatric asthma. J.Pediatr.Psychol. 2004;29(7):475-91.
77. Gustafsson D, Olofsson N, Andersson F, Lindberg B, Schollin J. Effect of asthma in childhood on psycho-social problems in the family. J.Psychosom.Res. 2002;53(6):1071-5.
78. Garratt A, Schmidt L, Mackintosh A, Fitzpatrick R. Quality of life measurement: bibliographic study of patient assessed health outcome measures. BMJ 2002;324(7351):1417.
79. Varni JW, Burwinkle TM, Rapoff MA, Kamps JL, Olson N. The PedsQL in pediatric asthma: reliability and validity of the Pediatric Quality of Life Inventory generic core scales and asthma module. J.Behav.Med. 2004;27(3):297-318.

80. Landgraf JM. The Child Health Questionnaire (CHQ): a potential new tool to assess the outcome of psychosocial treatment and care. In: Maruish ME, editor. The use of psychological testing for treatment planning and outcomes assessment. Mahwah, NJ: Lawrence Erlbaum; 1999. p. 665-77.
81. Starfield B, Bergner M, Ensminger M, Riley A, Ryan S, Green B et al. Adolescent health status measurement: development of the Child Health and Illness Profile. Pediatrics 1993;91(2):430-5.
82. French DJ, Christie MJ, Sowden AJ. The reproducibility of the Childhood Asthma Questionnaires: measures of quality of life for children with asthma aged 4-16 years. Qual.Life Res. 1994;3(3):215-24.
83. Juniper EF, Guyatt GH, Jaeschke R. How to develop and validate a new health-related quality of life instrument. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trails. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 49-56.
84. Herdman M, Rajmil L, Ravens-Sieberer U, Bullinger M, Power M, Alonso J. Expert consensus in the development of a European health-related quality of life measure for children and adolescents: a Delphi study. Acta Paediatr. 2002;91(12):1385-90.
85. Wallander JL, Schmitt M, Koot HM. Quality of life measurement in children and adolescents: issues, instruments, and applications. J.Clin.Psychol. 2001;57(4):571-85.
86. Kirshner B, Guyatt G. A methodological framework for assessing health indices. J.Chronic.Dis. 1985;38(1):27-36.
87. Skevington SM. Advancing cross-cultural research on quality of life: observations drawn from the WHOQOL development. Qual.Life Res. 2002;11(2):135-44.
88. Bullinger M, Power MJ, Aaronson NK, Cella DF, Anderson RT. Creating and evaluating cross-cultural instruments. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trials. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 659-68.
89. Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. Qual.Life Res. 2001;10(4):347-57.
90. Janse AJ, Gemke RJ, Uiterwaal CS, van dT, I, Kimpen JL, Sinnema G. Quality of life: patients and doctors don't always agree: a meta-analysis. J.Clin.Epidemiol. 2004;57(7):653-61.
91. Theunissen NC, Vogels TG, Koopman HM, Verrips GH, Zwinderman KA, Verloove-Vanhorick SP et al. The proxy problem: child report versus parent report in health-related quality of life research. Qual.Life Res. 1998;7(5):387-97.
92. Sung L, Young NL, Greenberg ML, McLimont M, Samanta T, Wong J et al. Health-related quality of life (HRQL) scores reported from parents and their children with chronic illness differed depending on utility elicitation method. J.Clin.Epidemiol. 2004;57(11):1161-6.
93. Arunkumar G, Wyllie E, Kotagal P, Ong HT, Gilliam F. Parent- and patient-validated content for pediatric epilepsy quality-of- life assessment. Epilepsia 2000;41(11):1474-84.

94. Ronen GM, Rosenbaum P, Law M, Streiner DL. Health-related quality of life in childhood epilepsy: the results of children's participation in identifying the components. Dev.Med.Child Neurol. 1999;41(8):554-9.
95. Ravens-Sieberer U, Gosch A, Abel T, Auquier P, Bellach BM, Bruil J et al. Quality of life in children and adolescents: a European public health perspective. Soz.Praventiv med. 2001;46(5):294-302.
96. Fitzpatrick R, Davey C, Buxton MJ, Jones DR. Evaluating patient-based outcome measures for use in clinical trials. Health Technol.Assess. 1998;2(14):i-74.
97. Guyatt GH, King DR, Feeny DH, Stubbing D, Goldstein RS. Generic and specific measurement of health-related quality of life in a clinical trial of respiratory rehabilitation. J.Clin.Epidemiol. 1999;52(3):187-92.
98. Koopman HM, Baars RM, Segaar RW. The use of computer-aided health-related quality-of-life questionnaires for children with a chronic disease and their parents. 2002 May 17; Oxford: Hughes associates; 2003.
99. Simeoni MC, Auquier P, Antoniotti S, Sapin C, San Marco JL. Validation of a French health-related quality of life instrument for adolescents: the VSP-A. Qual.Life Res. 2000;9(4):393-403.
100. Power MJ. Development of a common instrument for quality of life. In: Nosikov A, Gudex C, editors. EUROHIS: developing common instruments for health surveys. Amsterdam: IOS Press; 2003.
101. Bullinger M, Alonso J, Apolone G, Leplege A, Sullivan M, Wood-Dauphinee S et al. Translating health status questionnaires and evaluating their quality: the IQO LA Project approach. International Quality of Life Assessment. J.Clin.Epidemiol. 1998;51(11):913-23.

2

Clinicians' perspective on quality of life (QoL) assessment in paediatric clinical practice

R.M. Baars, S.M. v.d. Pal, H.M. Koopman and J.M. Wit
Acta Paediatrica 2004;93:1356-1362

If respondents do not believe that QoL information is clinically relevant, it appears unlikely that it will be easily incorporated into routine clinical practice (K.M. Taylor, 1996).

Abstract

This study was undertaken to investigate paediatric clinicians' views on and use of quality of life (QoL) assessment in clinical practice. A survey was conducted among members of the Dutch Paediatric Association via e-mail. Over half of the 303 respondents (57%) believed that it was possible to use QoL questionnaires in clinical practice. The majority indicated that assessing QoL was beneficial and that it was especially necessary to assess QoL in children with a chronic disease (82%). Although only a minority (17%) currently used QoL questionnaires, most respondents would want to use QoL questionnaires in the future (76%). Obstacles that prevent the use of QoL questionnaires are the extra time needed for assessment, the unavailability of standardized questionnaires and insufficient knowledge about QoL. This survey shows that paediatric clinicians are positive towards QoL assessment, but that certain obstacles prevent the use of questionnaires. Thus, to facilitate future use, QoL questionnaires need to be developed from the perspective of the paediatrician. This means that they need to be developed for clinical use and brought to the attention of the paediatric health care community, demonstrating their validity to child health care.

Introduction

Questionnaires designed to measure quality of life (QoL) have been developed and tested since the 1970s [1]. They are increasingly being used as assessment and outcome measures in clinical research trials in adults [2]. Assessment of QoL has also improved in the paediatric field. There is an increase in the availability of generic and disease-specific questionnaires for use in children and adolescents [3-6]. However, QoL is seldom included as an outcome measure in paediatric clinical trials or in clinical practice [7-9].

Until recently most investigators have concentrated on the development and validation of QoL questionnaires for research. At present an increasing number of investigators have expressed their interest in using QoL questionnaires for individual assessment and see the implementation of individual QoL questionnaires into clinical practice as the current challenge in the field of QoL research [1,10-12]. In adult research QoL assessment has already proven to be helpful. Evidence has indicated that QoL assessment is beneficial as an aid to patient management. This includes improving the clinician-patient relationship and communication, better monitoring changes in patients, screening for potential problems, and if necessary, referring to other professionals [11,13-18]. Unfortunately, the paediatric field lacks studies that provide proof that QoL assessment has similar benefits for the child's health.

There is some information on the attitude of clinicians towards using QoL measurements for patient care. This research comes mostly from the adult oncology field [14,19-25]. Walsh's survey (1988) showed that although clinicians believe that QoL can be measured, only a few used a specific method or were aware of available instruments [25]. In the study by Taylor et al. (1996) the majority of respondents considered it important to collect QoL information from their patients but they tended to do this informally. Only 7% routinely assessed the QoL of their patients in a structured manner [24]. Identified obstacles for QoL assessment were: time and resource constraints (money and human resources), lack of evidence-based intervention studies on their benefit to patient care, a perceived lack of appropriate instruments, lack of knowledge, unavailable interpretation guidelines and a belief that QoL assessment is unnecessary [15,18,22,24]. No literature was found on paediatricians' views on QoL assessment in clinical practice, and there is no indication that paediatric health care professionals implement available QoL questionnaires on a regular basis. The objective of this study is to assess the use of QoL questionnaires and the perspective of paediatric clinicians towards QoL assessment in paediatric care.

Material and methods

The aim was to evaluate (a) the paediatric clinicians' perspectives on quality of life and QoL questionnaires in clinical practice, (b) their willingness to assess QoL and (c) the obstacles preventing the use of QoL questionnaires. As the familiarity with QoL questionnaires and their terminology was assumed to be minimal, we did not use a specific QoL concept but referred to QoL in general. We designed a questionnaire (see appendix) based on earlier studies [22-25]. Questions regarding the clinicians' gender, age, profession, years of working experience, sub-specialization and hospital affiliation were included. A pilot test was carried out among clinicians for comprehension, ease of use and completion time.

The registry of members of the Dutch Paediatric Association (n=1780) was used to identify the study group. Members mainly include house-officers, paediatric registrars, paediatricians and retired paediatricians. Between May and July 2002, the self-administered questionnaire was emailed to those members for whom an email address was available (n=1036). A reminder was sent a month after the first mailing. The electronically returned questionnaires were directly converted into a format of the Statistical Package for the Social Sciences (SPSS), the input of the mailed questionnaires was done by hand. Descriptive statistics were generated with the SPSS 10.0. The Pearson X^2 test was used to calculate the statistical difference within the population.

The heads of the paediatric departments of each of the eight university hospitals in the Netherlands were also contacted and asked to participate in a semi-structured interview addressing issues on QoL assessment and its future clinical use.

Results

Of the 1036 emailed members, 362 replied (35 %), either by email or mail. A total of 303 questionnaires were used for the analysis. The other 59 responses were excluded, mostly because the questionnaire was not attached to the email or an empty questionnaire form was returned. Some of these respondents indicated that they thought the questionnaire was not applicable to them or did not want to participate.

Demographics

The demographic characteristics of the study group are listed in Table 1. The studied population had a larger proportion of paediatricians and more respondents from university hospitals.

The respondents' perspectives on quality of life and QoL questionnaires

Sixty-nine percent of the respondents were familiar with the existence of QoL questionnaires for children. Most had heard of them through the literature (40%) and from conferences (32%). Aspects that were seen as most important for QoL were physical functioning, social contact, pain, self-respect and daily life activities. The majority of the respondents (72%) thought it was possible to assess QoL in a research setting, only 57% thought this was possible in a clinical setting. Sixty-one percent of responders indicated that they always assessed the patient's QoL informally during their consultation. Most clinicians did not use any formal method to assess QoL, only a few indicated ever using a paper (17%) or a computer-aided (6%) QoL questionnaire. If valid and reliable QoL questionnaires would be available to them in the future, 76% would find them useful.

Demographic	**Group**	**Studied population (n=303)**	**Dutch Paediatric Association (n=1477)**
Gender (*p* = 0.06)	Male	54%	48%
Age (y)	<30	10 %	
	30-39	34 %	
	40-49	28 %	
	>50	28 %	
Profession (*p* = 0.01)	House officer	3%	7%
	Paediatric registrar	16%	19%
	Paediatrician	73%	48%
	Retired	2%	11%
	Other	6 %	15%
Years of work experience	<5	30%	
	6-10	19 %	
	11-20	22 %	
	>20	29%	
Sub-speciality	Yes	49%	
Affiliation (*p* = 0.00)	University Hospital	57%	28%
	Community Hospital	35%	35%
	Other/combination	8%	37 %

Table 1. Demographics of the studied population (n = 303) compared to the remaining members of the Dutch Paediatric Association (n = 1477).

The majority (71%) indicated that a specialized nurse could be primarily responsible for the assessment of a patient's QoL. The paediatrician, the psychologist and the hospital play specialist were also seen as suitable assessors of QoL by 58%, 56% and 50% of the respondents, respectively. Ideal methods of assessing QoL (formally and informally) were found to be the doctor's consultation (64%), a paper QoL questionnaire (53%) and a computer-aided QoL questionnaire (50%).

Willingness to assess QoL

Although 76% of the respondents indicated that they would want to use QoL questionnaires in the future, only 60% expected to actually do this. QoL assessment was found relevant for use in paediatrics, clinical research and especially for children with a chronic disease. Eighty-two percent of the respondents think it is necessary to formally assess QoL in children with a chronic disease (Table 2).

Patient group	**Not necessary**	**In some cases**	**Necessary**
Children at outpatient clinic	16	79	4
Admitted children	12	79	9
Acutely ill children	63	36	1
Chronically ill children	1	17	82
Children with unrecognised complaints	8	57	35

Table 2. Opinions on the necessity to measure QoL in different patient groups (%).

Obstacles preventing the use of QoL questionnaires

Respondents were asked which main obstacles would prevent them from using QoL questionnaires in the future. The main obstacles were the extra time needed for assessment, the unavailability of standardized questionnaires and their insufficient knowledge about QoL (Table 3). When asked whether they think they now have the skill and knowledge to use QoL questionnaires, 70% answered negatively.

Perceived obstacles	**Percentage of respondents**
Extra time needed for assessment	59
Unavailability of standardized questionnaires	55
Insufficient knowledge about QoL	48
No assistance in administering questionnaires	41
Inexperience with questionnaires	40
Needed training in interpretation	40
Insufficient information on questionnaires	30
Needed training in administration	27
Resistance from child or parents	20
Own priorities are different	16
Availability of extra working space	15

Table 3. Perceived obstacles preventing paediatricians from using QoL questionnaires (%).

Subgroup analysis

The respondents who were familiar with the existence of QoL questionnaires (n=206) were significantly more positive about their use. They were more likely to see the possibility of their use in a clinical setting (p=0.00) and in research (p=0.023) and were also more positive about using QoL questionnaires in the future (p=0.049) than the respondents who were not familiar with QoL questionnaires. They also scored significantly higher on the feeling that they had the skill and knowledge to use QoL questionnaires (p=0.033). However, they saw the unavailability of standardized questionnaires as a larger obstacle (p=0.038). Respondents who worked at a university hospital (n=171) were significantly more familiar with the existence of QoL questionnaires than the group working in a community hospital (p=0.007). Respondents with less than 10 years of work experience (n=147) and registrars (n=58) were significantly less familiar with the existence of QoL questionnaires (p=0.03 and p=0.019, respectively). They also scored significantly higher on the obstacle: 'inexperience with questionnaires' (p=0.001 and p=0.008, respectively).

Interview with heads of the paediatric departments

Seven of the eight heads of the university paediatric departments participated in an individual semi-structured interview. They believed that most paediatricians were aware of the existence of QoL questionnaires, but suspected that most of them see QoL questionnaires primarily as a research instrument and that they perceive the use of QoL questionnaires to be subjective and unreliable for clinical use. They propose that paediatricians are only open to innovation when they can see the benefit and effectiveness of a new method. They stated, therefore, that it was important to further validate existing questionnaires and demonstrate their benefit in clinical practice. They indicated that implementation of QoL questionnaires in clinical practice would be prevented by the following problems: limited time and manpower, insufficient finances, lack of standardization, insufficient knowledge and unproven benefit. For these reasons, it was implied that paediatricians would probably not administer the questionnaires. They said that the professionals who could facilitate the assessment, analysis and interpretation of QoL questionnaires were registrars, specialized nurses and psychologists.

Discussion

The aim of the current study was to explore the clinicians' view on QoL assessment in paediatric clinical practice. Similar to earlier studies, we found that clinicians are positive towards the use of QoL questionnaires, but that assessing QoL formally is uncommon [23,24]. The respondents saw the assessment of QoL in children with a chronic condition as especially important, and they believed that assessment should mainly be a task for the specialized nurse. It was stressed both by the survey and semi-structured interviews with the heads of paediatric departments that greater acceptance of QoL measurement is reliant upon evidence-based research that shows that QoL questionnaires are valid, reliable and beneficial and that standardized questionnaires should be easily accessible and provide ample information on interpretation and use.

While the majority of the respondents indicated that they would be interested in using QoL questionnaires, only a few currently used them and a small majority expected that they would actually use them in the future. This can be explained by the number of obstacles that they expect to encounter, such as the unavailability of standardized questionnaires, limited time for assessment and insufficient knowledge about QoL. These obstacles were similar to those found by others outside the paediatric field [18,19,22,24]. To stimulate clinical implementation, it is thus important to do more research on validating questionnaires and provide evidence of their benefit for clinical practice. If, in the future, paediatricians have access to valid questionnaires in areas they indicate are important, such as the QoL assessment of children with a chronic disease, this is likely to substantially increase their use.

Limited time for the completion of the questionnaires in the clinical setting is a major problematic aspect. Administering QoL questionnaires and scoring them takes extra time. A possible solution would be to further develop computerized questionnaires that can be used in the clinical setting. These can easily be administered, supply automatic data analysis and give instant results with a possibility to compare them to earlier measurements or a norm population [16,26,27], thus eliminating one of the major obstacles to questionnaire use. Being familiar with QoL questionnaires was associated with a more positive attitude towards their use. Thus, steps need to be taken to inform clinicians and other professionals about available questionnaires and the possibilities for their use in clinical practice. This is especially important for clinicians who have just started their careers and clinicians working in community hospitals. This indicates the importance of QoL data being reported in journals familiar to paediatric clinicians. It was also indicated how important it is to introduce a more multidisciplinary approach towards QoL assessment, as respondents noted the specialized nurse as someone who would be an appropriate person to administer QoL questionnaires.

The major limitation of this study is that the findings are related to a sample, which represents the opinion of only 17 % of the Dutch Paediatric Association. However, the response rate in our study (35%) was higher than the percentage (26%) achieved in a recent study by McMahon et al (2003). They compared a fax, post and email survey of paediatricians and found that their response rate, after 2 mailings, was 26% for email, 41% for post and 47% for fax [28]. Our response rate might have been improved if we had sent reminders by postal mail; however, time and financial factors prevented this at that time. Another aspect influencing the response rate was that there was no possibility to check whether the email addresses were actually being used. We also found that some of the respondents were inexperienced in opening, completing and returning an attached file via email. Theoretically, members already interested in QoL issues might be more likely to return the questionnaire, which would lead to a more positive attitude towards QoL assessment. However, we propose that the representativeness of the sample might be reasonable. Our main argument is that the results from two sources, responses from the departmental heads and the survey group, correspond. The survey also consisted of a large number of respondents who were not familiar with QoL questionnaires (30%) or found it was not (yet) possible to use QoL questionnaires in clinical practice (37%), indicating that the group represented a wide range of experience with QoL measurement.

Conclusion

This study demonstrates the necessity of taking the clinicians' perspective into account in the development of QoL questionnaires for clinical practice. The paediatric clinicians in this survey were interested in QoL assessment and felt that this was especially necessary in the treatment of children with a chronic disease. However, they identified a number of obstacles for the use of QoL questionnaires. Thus, if QoL questionnaires are to become an important part of the patients' assessment, more consideration needs to be given to the obstacles, to their use in the clinical setting, and to promoting the questionnaires to the health care community.

Acknowledgements

We would like to thank Dr. R.A. Holl and Mr. P. Hoogensteijn for their advice, Dr. R. Brand, Mr. H.C.M. Franken and Dr. R. Wolterbeek for their help with the statistical work and Dr. J.E. Chaplin and Dr. C. Atherton for checking the final documents. We would also like to thank the heads of the paediatric departments and all participants who returned the questionnaires.

References

1. Wood-Dauphinee S. Assessing quality of life in clinical research: from where have we come and where are we going? J.Clin.Epidemiol. 1999;52(4):355-63.
2. Sanders C, Egger M, Donovan J, Tallon D, Frankel S. Reporting on quality of life in randomised controlled trials: bibliographic study. BMJ 1998;317(7167):1191-4.
3. Bender BG. Measurement of quality of life in pediatric asthma clinical trials. Ann. Allergy Asthma Immunol. 1996;77(6):438-45.
4. Connolly MA, Johnson JA. Measuring quality of life in paediatric patients. Pharmacoeconomics. 1999;16(6):605-25.
5. Eiser C. Children's quality of life measures. Arch.Dis.Child 1997;77(4):350-4.
6. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol.Assess. 2001;5(4):1-157.
7. Dolinar RM, Kumar V, Coutu-Wakulczyk G, Rowe BH. Pilot study of a home-based asthma health education program. Patient.Educ.Couns. 2000;40(1):93-102.
8. Grey M, Boland EA, Davidson M, Yu C, Sullivan-Bolyai S, Tamborlane WV. Short-term effects of coping skills training as adjunct to intensive therapy in adolescents. Diabetes Care 1998;21(6):902-8.
9. Kelly CS, Morrow AL, Shults J, Nakas N, Strope GL, Adelman RD. Outcomes evaluation of a comprehensive intervention program for asthmatic children enrolled in medicaid. Pediatrics 2000;105(5):1029-35.
10. Ganz PA. Quality of life and the patient with cancer. Individual and policy implications. Cancer 1994;74(4 Suppl):1445-52.
11. Guillemin F. Functional disability and quality-of-life assessment in clinical practice. Rheumatology.(Oxford) 2000;39 Suppl 1:17-23.
12. Wagner AK, Ehrenberg BL, Tran TA, Bungay KM, Cynn DJ, Rogers WH. Patient-based health status measurement in clinical practice: a study of its impact on epilepsy patients' care. Qual.Life Res. 1997;6(4):329-41.
13. Detmar SB, Aaronson NK. Quality of life assessment in daily clinical oncology practice: a feasibility study. Eur.J.Cancer 1998;34(8):1181-6.
14. Detmar SB, Aaronson NK, Wever LD, Muller M, Schornagel JH. How are you feeling? Who wants to know? Patients' and oncologists' preferences for discussing health-related quality-of-life issues. J.Clin.Oncol. 2000;18(18):3295-301.
15. Greenhalgh J, Meadows K. The effectiveness of the use of patient-based measures of health in routine practice in improving the process and outcomes of patient care: a literature review. J.Eval.Clin.Pract. 1999;5(4):401-16.
16. Higginson IJ, Carr AJ. Measuring quality of life: Using quality of life measures in the clinical setting. BMJ 2001;322(7297):1297-300.
17. Kazis LE, Callahan LF, Meenan RF, Pincus T. Health status reports in the care of patients with rheumatoid arthritis. J.Clin.Epidemiol. 1990;43(11):1243-53.
18. McHorney CA, Earl BD, Jr. A Qualitative Study of patients' and physicians' views about practice- based functional health assessment. Med.Care 2002;40(11):1113-25.

19. Bezjak A, Ng P, Taylor K, MacDonald K, DePetrillo AD. A preliminary survey of oncologists' perceptions of quality of life information. Psychooncology. 1997;6(2):107-13.
20. Bezjak A, Ng P, Skeel R, DePetrillo AD, Comis R, Taylor KM. Oncologists' use of quality of life information: results of a survey of Eastern Cooperative Oncology Group physicians. Qual.Life Res. 2001;10(1):1-13.
21. Gough IR, Dalgleish LI. What value is given to quality of life assessment by health professionals considering response to palliative chemotherapy for advanced cancer? Cancer 1991;68(1):220-5.
22. Morris J, Perez D, McNoe B. The use of quality of life data in clinical practice. Qual. Life Res. 1998;7(1):85-91.
23. Tanaka T, Gotay CC. Physicians' and medical students' perspectives on patients' quality of life. Acad.Med. 1998;73(9):1003-5.
24. Taylor KM, Macdonald KG, Bezjak A, Ng P, DePetrillo AD. Physicians' perspective on quality of life: an exploratory study of oncologists. Qual.Life Res. 1996;5(1):5-14.
25. Walsh DL, Emrich LJ. Measuring cancer patients' quality of life. A look at physician attitudes. N.Y.State J.Med. 1988;88(7):354-7.
26. Cramer JA. Quality of life assessment in clinical practice. Neurology 1999;53(5 Suppl 2):S49-S52.
27. Koopman HM, Baars RM, Segaar RW. The use of computer-aided health-related quality-of-life questionnaires for children with a chronic disease and their parents. 2002 May 17; Oxford: Hughes associates; 2003.
28. McMahon SR, Iwamoto M, Massoudi MS, Yusuf HR, Stevenson JM, David F et al. Comparison of e-mail, fax, and postal surveys of pediatricians. Pediatrics 2003;111(4 Pt 1):e299-e303.

Appendix

Questionnaire

Instructions

In this questionnaire you will find questions on the use of quality of life (QoL) measures within paediatrics. Each part of the questionnaire will be clarified with explanatory text. For the question you can tick that box that in your opinion is most appropriate. There are no right or wrong answers. What you think is of importance. Completing the questionnaire will take 10 minutes.

It is difficult to give an exact definition of quality of life (QoL) . Through the years several definitions have been presented. We have selected several subjects. Which of the following subjects are according to your opinion most important for QoL? (maximal 5 answers possible)

- ☐ Physical functioning
- ☐ Vitality
- ☐ Treatment load
- ☐ Pain
- ☐ Limitations
- ☐ Autonomy
- ☐ Home situation
- ☐ Daily life activities
- ☐ Future
- ☐ Religion
- ☐ Self respect
- ☐ Body image
- ☐ Mobility
- ☐ School functioning
- ☐ Satisfaction
- ☐ Emotions
- ☐ Cognition
- ☐ Social contacts
- ☐ Illness load
- ☐ Creativity
- ☐ Coping
- ☐ Other.......

We are curious about your opinion on measuring QoL with the help of questionnaires.
Do you think QoL can be....

	Yes	Not yet	No	No opinion
Defined?	☐	☐	☐	☐
Objectified?	☐	☐	☐	☐
Validly measured? (measure what it should measure)	☐	☐	☐	☐
Measured reliably? (continuously measure the same)	☐	☐	☐	☐
Used in the clinic?	☐	☐	☐	☐
Used in research?	☐	☐	☐	☐

In what way do you currently form an opinion on the child and parents QoL during (outpatient) consultation?

	Never	Sometimes	Always
Intuition	☐	☐	☐
Clinical experience	☐	☐	☐
In your consultation	☐	☐	☐
From another health care workers consultation	☐	☐	☐
A paper questionnaire	☐	☐	☐
A computerised questionnaire	☐	☐	☐
Other	....		

What, in your opinion, would be the ideal way to form an opinion of a child and parents QoL?

	Not	Sometimes	Useful
Intuition	☐	☐	☐
Clinical experience	☐	☐	☐
In your consultation	☐	☐	☐
From another health care workers consultation	☐	☐	☐
A paper questionnaire	☐	☐	☐
A computerised questionnaire	☐	☐	☐
Other	....		

Several questionnaires to measure QoL in children have been developed in the last years
Are you familiar with the existence of these questionnaires?

☐ No
☐ Yes, (more answers possible)

- ☐ While at university
- ☐ While specializing
- ☐ From literature
- ☐ During courses
- ☐ At a conference
- ☐ Other

In this next part we are curious about your opinion on the importance of quality of life questionnaires in the care for children and their parents. How relevant do you find the use of QoL questionnaires for:

	Not	Hardly	A little	Considerable	Very
Routine treatment	☐	☐	☐	☐	☐
Treatment of the child with a chronic disease	☐	☐	☐	☐	☐
Treatment of a child with unexplainable complaints	☐	☐	☐	☐	☐
Clinical research	☐	☐	☐	☐	☐
Improvement of the general health of a child	☐	☐	☐	☐	☐
Certain choices in your treatment	☐	☐	☐	☐	☐
Paediatrics	☐	☐	☐	☐	☐

Paediatricians in Holland:

	Never	Seldom	Sometimes	Often	Always
Think that questionnaires need to be included in the treatment	☐	☐	☐	☐	☐
Find it praiseworthy/commendable when I use QoL questionnaires	☐	☐	☐	☐	☐
Have influence on my choice to use QoL questionnaires	☐	☐	☐	☐	☐

In my opinion:

	No	Yes
It is completely my own choice whether I use QoL questionnaires	☐	☐
I have the skill and knowledge to use QoL questionnaires	☐	☐

Assuming that valid and reliable QoL questionnaires will be available in the future what would you in general think about using these QoL questionnaires your self?

	1	2	3	4	5	
Not useful	☐	☐	☐	☐	☐	Very useful
Very time-consuming	☐	☐	☐	☐	☐	Very time-saving
Very uninteresting	☐	☐	☐	☐	☐	Very interesting

Can you indicate what aspects would keep you from using QoL questionnaires in clinical practice? (several options)

☐ Your own priorities are different
☐ Insufficient knowledge about QoL
☐ Inexperience with questionnaires
☐ Resistance from child and parents
☐ Insufficient information on questionnaires
☐ Availability of standardized questionnaires
☐ Availability of extra working space for assessment
☐ Extra time for assessment of the questionnaire
☐ Training in administering the questionnaire
☐ Training in the interpretation of the questionnaires
☐ No assistance in administering the questionnaire
☐ Other.......

Do you think it is necessary to formally measure QoL through standardized questionnaires in …

	Not necessary	Necessary in some cases	Necessary
Children at the outpatient clinic	☐	☐	☐
Admitted children	☐	☐	☐
Acutely ill children	☐	☐	☐
Chronically ill children	☐	☐	☐
Children with unexplainable complaints	☐	☐	☐
Other	....		

What discipline should, according to you, be primarily be responsible for the administering QoL questionnaires from children and their parents. (more answers possible)

☐ Paediatrician
☐ General nurse
☐ Specialised nurse (e.g. diabetes nurse)
☐ General practitioner
☐ Social worker
☐ “ Well baby” health clinic
☐ Teacher
☐ Psychologist
☐ Hospital play specialist
☐ Other.......

Assuming that in the near future valid and reliable quality of life questionnaires (paper or computer) will be available in paediatrics.

Would you want to use QoL questionnaires in your treatment?

☐ Certainly not ☐ Probably not ☐ Maybe ☐ Probably will ☐ Surely will

Do you plan to use QoL questionnaires in your treatment?

☐ Certainly not ☐ Probably not ☐ Maybe ☐ Probably will ☐ Surely will

Do you expect to really use QoL questionnaires in your treatment?

☐ Certainly not ☐ Probably not ☐ Maybe ☐ Probably will ☐ Surely will

3

A child focus group methodology: experiences from the European DISABKIDS project

R.M. Baars, J.E. Chaplin, H.M. Koopman and the DISABKIDS group

> Without including children in the main stage of HRQoL research, we believe that children's fundamental beliefs, feelings and understanding of their disorders and their interface with society might not be revealed (G.M. Ronen, 2001).

Abstract

A cross-national patient-derived methodology was applied during the development of the European DISABKIDS health related quality of life (HRQoL) instrument to identify relevant HRQoL statements. Focus groups were used in the DISABKIDS project to include the child and adolescent's own concepts, language and culture, and to acknowledge the patient as the expert in their own lives. Participants included children and adolescents with a chronic medical condition, their parents and health care professionals. Focus groups were monitored by two moderators and facilitated by a series of semi-structured questions and probes. HRQoL statements were identified from the literal transcripts for the development of the European DISABKIDS instrument. The DISABKIDS manual served as a guide to assure that a similar method was used in all countries. The focus groups and interviews were conducted in eight institutions in seven European countries. Participants included 154 children and adolescents with a chronic medical condition, 142 family members and 26 health care professionals. Focus group progress was related to the developmental abilities of the child. A total amount of 3515 HRQoL statements were collected. These statements have subsequently been used for the development of the DISABKIDS instrument. This patient-derived procedure made it possible to reflect on aspects that are important to the patient group and phrase items in their own words. However, a number of issues need to be considered when adapting the focus group methodology for use in children and adolescents, which include the developmental abilities of the participants.

Introduction

In the past children and adolescents have often been treated as passive receivers of medical services and health care. Even paediatric health related quality of life (HRQoL) was often assessed by the parent instead of the child [1-3]. As proxy measures are not necessarily representative of how the child or adolescent feels the focus is currently directed towards the child and adolescent [4-7].

Including the young patient's opinion is now recognised as important, not only for the assessment of their HRQoL but also during the questionnaire's developmental process [8-12]. As HRQoL is inherently an attribute of the patient, the questionnaire should reflect the concerns and opinions that are important to the patient. Using only expert opinions for the development of a questionnaire is thought to lead to investigator bias and poor face and content validity [13]. Therefore the patients should be involved in the development of a questionnaire [13,14]. As a consequence patient-derived methods are being used as an aid in the development of HRQoL questionnaires. The focus group methodology is such a patient-derived method, which is regularly used in de development of paediatric HRQoL questionnaires [10,11,15].

Focus groups have been defined as "a research technique that collects data through group interaction on a topic determined by the researcher" [16]. It is a qualitative research method, which helps to understand beliefs, views, knowledge, ideas, attitudes and experiences in a certain group in relation to a certain topic. It gets its advantage from group interaction, which can deepen and clarify topics within a group [17-19]. The communication and interaction between the participants can generate useful data and allows the investigator to learn about the participants' perspectives. Focus groups were first used as a marketing research tool but are now more widely used for health care research and services. Within a focus group, one can address clinical questions, identify problems or learn how patients experience illness or health care services. Focus groups are also found to be useful in a preliminary phase of a study to identify important research issues or develop suitable items for a new questionnaire [8,14,18,19].

There is however little information available on the methodological adaptations that are necessary to run focus groups with children and adolescents [8,17]. One can assume that there is a need to consider issues related to the child and adolescent's developmental level. Such issues include the child and adolescent's verbal comprehension, their ability to report back on a specific time period, and the age appropriateness and changing importance of HRQoL issues [20]. When conducting focus groups or interviews with children and adolescents one especially needs to consider that their communication and social skills depend on their age and attained cognitive level [17,20,21]. Although their skills and developmental level may form a challenge in focus group research, the developmental variability of the child and adolescent is also an advantage. Focus groups can provide investigators with the child and adolescents own ideas, supplied in their language and including their view of health, which can help to adapt the questionnaire to their level of understanding.

This paper describes the cross-national patient-derived method that was applied in the European DISABKIDS project. Focus groups and interviews were conducted with children and adolescents with chronic medical conditions, their parents and health care professionals to capture their view on HRQoL. A description will be given of the methodology used in this large cross-national project. Positive and negative aspects will be discussed, which will help in the application of the focus group methodology with children and adolescents in the future.

Method

The DISABKIDS project

The DISABKIDS project is a European collaboration, which consists of eight partner institutions in seven countries (Austria, France, Germany, Greece, the Netherlands, Sweden and the United Kingdom). The aim was to cross-nationally develop a new European HRQoL instrument for children and adolescents with a chronic medical condition in several countries simultaneously [22]. Chronic medical conditions included in the project are asthma, juvenile idiopathic arthritis (JIA), atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes and epilepsy. The DISABKIDS project intends to provide for the growing need of multilingual cross-nationally validated paediatric HRQoL instruments for use in (international) clinical trials and clinical practice [23]. The final instrument has a modular build-up, with a chronic generic module that is applicable to all children with a chronic medical condition, and seven condition-specific modules.

The items for the DISABKIDS instrument were collected through a patient-derived (bottom-up) procedure based on focus groups and interviews with children and adolescents with a chronic medical condition. This child focused research method was applied in each of the DISABKIDS centres and provided the opportunity to capture the child's perspective on HRQoL. The identified HRQoL statements comprised the data pool from which the final instrument items were constructed. Focus group discussions and interviews were also conducted with parents and health care professionals in order to incorporate their views into the DISABKIDS instrument.

The DISABKIDS focus group methodology

To assure that a similar method was used in all countries a manual was developed which included the outline of the focus groups and the semi-structured questions. In view of time and resource constraint and if participants were unable to attend a planned focus group there was the option of participating in an individual interview. Each centre was responsible for training the moderators and recruiting participants for the focus groups and interviews.

Each of the eight DISABKIDS partners aimed to run focus groups for at least two to three chronic conditions. Separate focus groups were conducted for each chronic condition and were divided by age (4-7, 8-12 and 13-16). Each focus group did include a mixture of disease severity and gender. The general aim was to have between 4 and 8 participants in a focus group [17]. Parents and health care professionals participated in separate focus groups or interviews.

Two moderators ran the focus group discussions. Both moderators were familiar with group discussions as well as working with children. While the first moderator led the discussions the task of the second moderator included the technical aspects of tape-recording, observing the group and writing down content and non-verbal behaviour during the session.

At the start of each focus group the aim, duration and confidentiality was explained to the participants. Permission for the audio-taping was also asked. The moderators used the semi-structured focus group questions supplied in the manual (Box 1). The discussion was led in a non-directive fashion. The questions were chosen in order to lead the participants from a general discussion to more specific information about their quality of life. If necessary probe questions were used to stimulate conversation.

Semi-structured questions:
What do you like about your life?
What do you like best about your life?
What makes you happy?
What bothers you most in your life?
What kind of things keeps you healthy? (coping styles / activities)
If you could make a wish, what would you wish for in order to be happier or more satisfied?
Tell me about condition. (e.g. epilepsy / diabetes / asthma etc.)
Apart from yourself, do you know any other children with this condition?
How does their condition affect them at school / home?
How does your condition affect you at school / home?
What would you like people to say at school / home / hospital to help?
What would you like people to do at school / home / hospital to help?

More specific questions:
What do you think of your condition?
What do you think of your medication / treatment / hospital / the doctors.
What are the disadvantages of having your condition? Are there advantages?
Do you know what other kids think of you having this condition?
Are you different compared to children without this condition?

Box 1. Prepared semi-structured questions included in the focus group and interview manual.

The focus groups were organised into two parts (Table 1). Part one was intended to discuss various health and illness related topics. Part two allowed the children and adolescents to write down questions that would give a clinician a good impression of their HRQoL. They also listed which questions they found most important and possible question formats and response categories were discussed.

The focus groups and interviews were tape-recorded and transcribed verbatim to conserve the exact expressions used by participants. Statements related to HRQoL were identified from the literal transcripts to form a pool from which the final items were constructed.

Phase		**Time frame**	**Purpose**
Part 1	Introduction	5 min	Warm up
	Discussion	35 min	Discussing perceived HRQoL issues
	Break	10 min	
Part 2	Question construction	30 min	Writing individual questionnaire items
	Debriefing	10 min	Group evaluation and any missing topics

Table 1. Focus group structure.

Participants

The participants (children, adolescents and their parents) were chosen via the hospital register or patient associations and had a confirmed diagnosis of one of the seven chronic medical conditions included in the project protocol (asthma, JIA, atopic dermatitis, CP, CF, diabetes or epilepsy). All potential participants were contacted by letter with an explanation of the project and details of what they would be asked to do. Families willing to participate returned a form agreeing to the study and were contacted via telephone in order to co-ordinate a date suitable for all focus group participants to meet. All participants had inclusion criteria to comply to (Box 2). There was no requirement that the participants should be unknown to each other. The participants completed a standardised consent form prior to entering the focus groups and interviews. Where possible the location of the groups was outside the hospital premises. The focus groups were divided by chronic condition and stratified by age (4-7, 8-12 and 13-16 years). Each focus group consisted of a mixture of disease severity and gender. Parents were invited to participate in separate focus groups (grouped according to the chronic condition and age of their child). If participants were unable to attend a planned focus group they could take part in a personal interview. Health care professionals were contacted directly by the investigators and were often affiliated to the research centres. The European commission approved the study as well as each of the Medical Ethics Committees in each study centre.

Inclusion criteria:

- Consent to participate in the study
- Between 4 and 16 years of age
- Have one of the seven chronic conditions
- No other chronic medical condition
- Sufficient knowledge of language and able to express themselves

Box 2. Inclusion criteria for the child and adolescent.

Results

Focus groups and interviews were conducted in each of the DISABKIDS centres: the University of Vienna in Austria, the University Hospital of Marseille in France, the University Hospital of Hamburg in Germany, the Medical University of Lüebeck in Germany, Thessaloniki University Paediatric Clinic in Greece, Leiden University Medical Center in the Netherlands, the University Hospital of Lund in Sweden and the Royal Hospital for Sick Children in Edinburgh the United Kingdom. The University of Verona in Italy participated in the focus group research as an affiliated centre. Moderators were members of the research teams. Each centre included one to three chronic conditions (Table 2), which included separate focus groups for the different age ranges and parent groups.

Country	Condition A	Condition B	Condition C
Austria	Diabetes mellitus		
France	Epilepsy		
Germany (H*)	Atopic dermatitis		
Germany (L†)	Juvenile idiopathic arthritis	Cerebral Palsy	
Greece	Cystic Fibrosis	Asthma	
Italy	Asthma		
Netherlands	Asthma	Juvenile idiopathic arthritis	
Sweden	Diabetes Mellitus	Epilepsy	Asthma
United Kingdom	Cerebral Palsy	Epilepsy	

Table 2. Number of chronic conditions included in the focus groups and interviews per DISABKIDS centre.
*Hamburg / †Lüebeck

A total number of 322 participants contributed to the DISABKIDS focus groups and interviews. Of these 154 were children and adolescents, 142 were family members and 26 health care professionals. The data output consisted of the focus group and interview tape-recordings, notes from the moderators and cards or flipchart pages with suggested questions written by the children and adolescents. The group size ranged from three to six persons in each country and the time to complete the focus groups was between 60 and 90 minutes.

The Greek centre used videos as 'warming-up' activity at the start of the focus groups. This helped to relax the children. Others applied a general introduction round. The moderator's task was to stimulate new topics and guide the discussion, which was sometimes experienced as difficult, especially with the youngest age group. Young children were not always able to express their opinion or feelings in more than a few words. Answers were short and it was hard to stimulate group discussions. From the age of 6 there was a noticeable increase in their ability to express themselves. Interaction with group members was apparent from the age of 10 years. In some instances one speaker dominated the discussion while another child hardly spoke. The large age variation in the adolescent groups presented a few problems, for example the life experience of a 13 yr old boy can be very different to that of a 16 yr old girl. The amount of time children could focus on the topic increased by age and reached a maximum of 90 minutes in the adolescent groups. Parents, on the other hand, were reluctant to stop at the end of the sessions. In the parent and health care professional groups the discussions developed automatically. There was group interaction with participants discussing topics with each other and not just answering the moderator's questions. On the whole all the participants indicated that they welcomed this opportunity to talk to others in a similar situation, felt they where acknowledged as experts and would volunteer to participate in similar exercises in the future.

The tape-recordings were written or typed out literally, and included the notes and observations from the second moderator. This process was time consuming and took between 6 to 10 hours. Each country selected relevant HRQoL statements from their national focus group or interview transcripts. This resulted in a total amount of 3515 statements. Collected statements showed various similarities between countries. Medical aspects such as treatment and symptoms were discussed in all countries. Other recurring topics included experienced limitations, school, relationships with peers, and emotional reactions to having a chronic medical condition. The parents often discussed the health care system extensively. The collected statements, clustered by condition, were divided into three modules (generic, chronic generic and condition-specific). All the statements that were not directly related to health or a chronic medical condition (n=488) were merged to form a generic module. The statements that were applicable to any chronic medical condition (n=1647) were merged to form the chronic-generic module. The statements that were specific to each of the medical conditions (between 340 and 66 statements) formed the seven condition-specific modules (Table 3).

This division was the basis of the three-modular structure; the generic, chronic generic and condition-specific modules. From this point onwards the statements in each module underwent a reduction process for the development of the pilot instrument. These results are reported elsewhere [24].

Module		**Collected statements**
Generic		488
Chronic generic		1647
Condition-specific	Asthma	304
	JIA	340
	Atopic dermatitis	66
	CP	183
	CF	167
	Diabetes	141
	Epilepsy	179

Table 3. Number of collected statements per module.

Discussion

The focus groups and interviews enabled a bottom-up procedure for the development of the DISABKIDS instrument. Although focus groups and interviews have been used previously for the development of paediatric HRQoL questionnaires this has not been done simultaneously in several countries and for several chronic medical conditions. The applied patient-derived method has proven to be effective for data collection in the DISABKIDS project. All centres reported that there were many spontaneous responses, which suggests that participants were using their own way of expression and ideas.

There are some issues specifically related to running focus groups and interviews with children and adolescents that should be discussed. The methodology must be adopted to create the right kind of environment where the child can be open and motivated to participate. Being successful at child focus groups depends on the skills of the moderator and requires attention to several factors [16,25]. It cannot be emphasised enough that the moderators must be trained and aware of the cognitive and social capacities of children of different ages, and must be aware of their communication ability and attention span. The experienced age dependent change in ability to express themselves, interact with the group and stay focussed coincided with earlier described changes in childhood development [8,17,26,27]. As the quality of the supplied data is tied to the skills of the moderator we emphasise the importance of (international) training in this skill, especially for future cross-national studies.

Other problem areas concern group processes: the discussion may lack spontaneity, one participant can dominate the group and some participants may not join in the discussions or show disrespect. There are several guidelines to apply in such situations of which most were provided in the DISABKIDS manual (Box 3)[17]. We were also confronted with various developmental levels and different gender priorities in the adolescent groups [8,17,21]. For this reason we would choose to divide the adolescent group by gender and into two age groups in the future. Overall, one needs to keep in mind that focus groups are a time consuming method that needs careful planning and adaptation to the group of participants. It can also be difficult to recruit participants due to practical (previous appointments, summer time, travel distance) or personal reasons (shy, refusing to talk about medical condition).

There are limitations related to collecting data through focus groups and interviews. The generalisation of the collected data can be questioned as only a limited number of children, adolescents, parents and health care professionals participated (154, 142 and 26 respectively). Selection bias is also a limitation and creates the risk that the participants do not represent the general population of interest. It was also not possible to check that all relevant HRQoL aspects were discussed. Participants may have found it difficult to talk about certain (sensitive) issues, or topics were not thought of at the time. Other studies have continued conducting focus groups till no new data were presented [28,29]. However, this was not possible in the DISABKIDS project due to time constraints. Reassuring is that overall the number of participants was relatively large for focus group research and that a considerable amount of HRQoL statements were collected. The simultaneous setup in different countries supported the cross-national developmental process and demonstrated that various issues were relevant in all countries.

- Supply a good introduction and an opportunity to get to know the group
- Introduce some general rules on confidentiality and respect
- Let the children know that they are the experts and the investigator needs their input
- Consider the developmental age of the child and their ability to take part in the discussion
- Allow the children some control over the discussion
- Let them express themselves as freely as possible, listen attentively and don't interrupt
- Gather the information as objectively as possible
- Encourage descriptions of events by getting them to tell a story of their day or anecdotes
- Be relaxed during the interviews as this will relax the children as well
- Be sensitive to the mood of the group
- Do not insist on discussing painful themes
- Avoid making participants feel that they have failed if they can't answer the questions
- If you think the participants are telling lies then DO NOT press them for the truth
- Always end a focus group with a debriefing

Box 3. Aspects a moderator should apply during focus group work with children and adolescents.

Other issues concern the construction of the statement pool and what data to use from the focus groups and interviews. Qualitative patient-derived methods, like the focus group or interview, do not provide statistical data [29]. There is no statistical or standardised method to apply to the selection of HRQoL statements from the national focus group and interview transcripts. As the statement selection was a subjective process, based on general content analysis, it can be influenced by personal interpretations, interests and subjective factors [30]. Previous investigators have used the computer to facilitate this step, however as the qualitative data were in several different languages this was not applicable to the DISABKIDS project [31,32].

Conclusion

The applied child-focused patient-derived methodology aided in the collection of HRQoL statements for the development of the European DISABKIDS HRQoL instrument. The focus group and interviews were successful in embracing the child and adolescent's own point of view, concepts, culture and have acknowledged the child as the expert in their own lives. The described cross-national focus group and interview methodology made it possible to reflect on aspects that were important to the patient group, compare cross-national data and provided HRQoL statements for use in the DISABKIDS instrument. Being successful at focus group research does require attention to several factors (Box 4). In addition, sufficient consideration needs to be given to the organisational aspects related to focus group research, the developmental level of the participants and the method of processing qualitative data.

Preparations	Determine your goal and target population Agree on an available budget and timeline Get approval of the ethical committee Recruit at least two moderators Focus group manual with guidelines and questions Find suitable locations (avoid a hospital setting when possible) Send letters to potential participants
Inclusion criteria	Consent to participate in the study Fit the age requirements of the study Fit the group description (e.g. chronic medical condition) Sufficient language skills
Materials	Focus group manual Tape recorder and tapes with labels Pencils and paper or flipcharts Refreshments Token of appreciation
Moderator qualities	Familiar with group discussions Experience with children's cognitive and social capacities Aware of children's communication abilities and attention span Non-authoritarian and patient
Basic data	Participants' names Age and gender Type of school / grade Health Status Medication
Output	Literal transcription from tapes (takes 8-12 hours) General outline from tape (takes 4-8 hours) Data from notes (2-3 hours) Data from memory (1 hour)
Moderator debriefing	Did the moderator keep to the rules and suggested interview structure? Did the children understand the instructions? Did the discussion flow smoothly? Where there problems? What was the general atmosphere of the session? How did the session end?

Box 4. General guidelines for focus group research.

Acknowledgements

We would like to thank all the participants who contributed, by joining, organising, moderating and transcribing the focus groups. The DISABKIDS project was supported by the European Commission (QLG5-CT-2000-00716) within the Fifth Framework Program “Quality of Life and Management of Living Resources”.

References

1. Andelman RB, Zima BT, Rosenblatt AB. Quality of life of children: toward conceptual clarity. In: Maruish ME, editor. The use of psychological testing for treatment planning and outcomes assessment. Mahwah, NJ: Lawrence Erlbaum; 1999. p. 1383-413.
2. Bullinger M, Ravens-Sieberer U. Health related QOL assessment in children: a review of the literature. Revue Europeenne de Psychologie Appliquee 1995;45(4):245-54.
3. Ravens-Sieberer U, Bullinger M. Assessing health-related quality of life in chronically ill children with the German KINDL: first psychometric and content analytical results. Qual.Life Res. 1998;7(5):399-407.
4. Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. Qual.Life Res. 2001;10(4):347-57.
5. Guyatt GH, Juniper EF, Griffith LE, Feeny DH, Ferrie PJ. Children and adult perceptions of childhood asthma. Pediatrics 1997;99(2):165-8.
6. Theunissen NC, Vogels TG, Koopman HM, Verrips GH, Zwinderman KA, Verloove-Vanhorick SP et al. The proxy problem: child report versus parent report in health-related quality of life research. Qual.Life Res. 1998;7(5):387-97.
7. Sung L, Young NL, Greenberg ML, McLimont M, Samanta T, Wong J et al. Health-related quality of life (HRQL) scores reported from parents and their children with chronic illness differed depending on utility elicitation method. J.Clin.Epidemiol. 2004;57(11):1161-6.
8. Heary CM, Hennessy E. The use of focus group interviews in pediatric health care research. J.Pediatr.Psychol. 2002;27(1):47-57.
9. Somerville A, Knopfli B, Rutishauser C. Health-related quality of life in Swiss adolescents with asthma. Validation of the AAQOL-D and comparison with Australian adolescents. Swiss.Med.Wkly. 2004;134(7-8):91-6.
10. French DJ, Carroll A, Christie MJ. Health-related quality of life in Australian children with asthma: lessons for the cross-cultural use of quality of life instruments. Qual.Life Res. 1998;7(5):409-19.
11. Rutishauser C, Sawyer SM, Bond L, Coffey C, Bowes G. Development and validation of the Adolescent Asthma Quality of Life Questionnaire (AAQOL). Eur.Respir.J. 2001;17(1):52-8.
12. McEwan MJ, Espie CA, Metcalfe J. A systematic review of the contribution of qualitative research to the study of quality of life in children and adolescents with epilepsy. Seizure. 2004;13(1):3-14.
13. Gill TM, Feinstein AR. A critical appraisal of the quality of quality-of-life measurements. JAMA 1994;272(8):619-26.
14. Barbour RS. The use of focus groups to define patient needs. J.Pediatr.Gastroenterol. Nutr. 1999;28(4):S19-S22.

15. Ronen GM, Streiner DL, Rosenbaum P. Health-related quality of life in children with epilepsy: development and validation of self-report and parent proxy measures. Epilepsia 2003;44(4):598-612.
16. Morgan DL. Focus Groups. Annu.Rev.Sociol. 1996;22:129-52.
17. Kennedy C, Kools S, Krueger R. Methodological considerations in children's focus groups. Nurs.Res. 2001;50(3):184-7.
18. Krueger RA. Group dynamics and focus groups. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trails. Second Edition ed. Philadelphia: Lippin cott-Raven; 1996. p. 397-402.
19. Kitzinger J. Qualitative research. Introducing focus groups. BMJ 1995;311(7000):299-302.
20. Wallander JL, Schmitt M, Koot HM. Quality of life measurement in children and adolescents: issues, instruments, and applications. J.Clin.Psychol. 2001;57(4):571-85.
21. Horner SD. Using focus group methods with middle school children. Res.Nurs. Health 2000;23(6):510-7.
22. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. Int.J.Rehabil.Res. 2002;25(3):197-206.
23. Skevington SM. Advancing cross-cultural research on quality of life: observations drawn from the WHOQOL development. Qual.Life Res. 2002;11(2):135-44.
24. Petersen C, Schmidt S, Power M, Bullinger M, and the DISABKIDS group. Development and pilot-testing of a health-related quality of life chronic generic module for children and adolescents with chronic health conditions: A European perspective. Qual Life Res. 2005;14(4):1065-77.
25. Peterson-Sweeney K. The use of focus groups in pediatric and adolescent research. J.Pediatr.Health Care 2005;19(2):104-10.
26. Bibace R, Walsh ME. Development of children's concepts of illness. Pediatrics 1980;66(6):912-7.
27. Koopman HM, Baars RM, Chaplin J, Zwinderman KH. Illness through the eyes of the child: the development of children's understanding of the causes of illness. Patient. Educ.Couns. 2004;55(3):363-70.
28. McEwan MJ, Espie CA, Metcalfe J, Brodie MJ, Wilson MT. Quality of life and psychosocial development in adolescents with epilepsy: a qualitative investigation using focus group methods. Seizure. 2004;13(1):15-31.
29. Sim J. Collecting and analysing qualitative data: issues raised by the focus group. J.Adv.Nurs. 1998;28(2):345-52.
30. Weinberger M, Ferguson JA, Westmoreland G, Mamlin LA, Segar DS, Eckert GJ et al. Can raters consistently evaluate the content of focus groups? Soc.Sci.Med. 1998;46(7):929-33.
31. Aljunid S. Computer analysis of qualitative data: the use of ethnograph. Health Policy Plan. 1996;11(1):107-11.

32. Chapple A, Rogers A. Explicit guidelines for qualitative research: a step in the right direction, a defence of the 'soft' option, or a form of sociological imperialism? Fam. Pract. 1998;15(6):556-61.

4

Using cross-national focus groups to identify health related quality of life (HRQoL) aspects in children and adolescents with asthma and their parents: the European DISABKIDS approach

R.M.Baars, J.E.Chaplin, E. Hatziagorou, M. Niero, J.Tsanakas, J.M.Wit, H.M.Koopman and the DISABKIDS group.

Submitted

> The need to incorporate patients' values and preferences is what distinguishes quality of life from all other measures of health (T.M. Gill, 1994).

Abstract

Focus groups and interviews were conducted to support the bottom-up developmental process of the DISABKIDS health related quality of life (HRQoL) instrument for children and adolescents with a chronic medical condition and their parents. The HRQoL statements which were identified through focus groups and interviews were used to develop the European DISABKIDS HRQoL instrument. Participants included children and adolescents with a chronic medical condition, their parents and health care professionals across Europe. The asthma results are presented in more detail. The asthma focus groups and interviews were conducted in four European countries (Greece, Italy, the Netherlands and Sweden). A total of 43 children and adolescents with asthma, 33 family members and 7 health care professionals participated in the focus groups and interviews. Asthma symptoms and related medical aspects were discussed in all groups. Additional issues related to asthma included physical limitations in the youngest groups and social issues in the adolescent groups. Parents were worried about prevention, long-term effects, medication and school. The discussed topics were similar between countries. A total of 637 HRQoL statements were collected from the asthma focus groups and interviews for the development of the European DISABKIDS instrument. In addition to having to live with the medical implementations of asthma, children and adolescents are also effected in social and physical areas. The children and adolescents discussed the current limitations they experienced while parents concentrated on the long-term consequences. A greater understanding of the impact of asthma on the lives of children and adolescents was achieved. The focus group discussions were also a useful tool in generating statements from children and adolescents with a chronic condition like asthma for the development of the DISABKIDS HRQoL instrument.

Introduction

There are several aspects that need to be considered during the development of a paediatric health related quality of life (HRQoL) questionnaire. Ideally a HRQoL questionnaire should give an accurate representation of the aspects in life that are affected by an illness. Choosing which issues should be included in a questionnaire and how to generate these items are important decisions. There is no standard procedure for developing a HRQoL questionnaire for children or adolescents. Existing questionnaires have been constructed in several ways, they vary in their definition of HRQoL, they consist of different multidimensional constructs and differ in the content of the domains [1-7]. Selected items are often derived from one or more sources including relevant literature, existing questionnaires, health care professionals' opinions, investigators' opinions and the views of the child, adolescent or the parent [8-18].

To make sure that a paediatric HRQoL questionnaire accurately represents the child or adolescent's HRQoL, it is important that the items are appropriate to the representation of their illness. Numerous studies have shown that using the expert or parent opinions to construct a paediatric questionnaire can lead to bias and poor face and content validity as their opinions can differ from those of the child or adolescent [19-25]. As HRQoL is an attribute of the patient emphasis should be on the child and adolescent's perception of HRQoL. To accomplish this, the child and adolescent should be directly involved in the development of any paediatric HRQoL questionnaire by defining and identifying the issues that are important to them [26-28].

There are various ways of involving children and adolescents in the development of a new questionnaire. Their opinion can be collected through surveys, interviews or focus groups. A focus group is a qualitative research method, which allows the investigator to explore the opinions, attitudes, knowledge, concerns and experiences of a group [29-31]. Within HRQoL research focus groups can be used to explore the health care perception of a particular population, to study research questions or to support the development of a questionnaire [31,32]. Focus groups are also useful in generating questionnaire items as participants can determine the topics and identify HRQoL issues that are important to them [30,33]. This in contrast to having health care professionals, investigators or family members judge the importance for children and adolescents [26,34].

There are several publications on how a focus group can be used as an appropriate method to explore HRQoL issues in children and adolescents with a chronic medical condition [34-38]. The European DISABKIDS project included seven chronic medical conditions in the development of the paediatric HRQoL instrument. The findings reported in this paper are part of the larger DISABKIDS focus group study. We will only elaborate on the asthma results generated from the focus groups and interviews with children and adolescents with asthma, their parents and health care professionals. First, a qualitative description of the cross-national asthma focus groups and interviews is given. Secondly, the collected HRQoL statements will be described in a quantitative manner.

Method

The DISABKIDS project

The DISABKIDS project is a collaboration of eight research centres in seven European countries. The aim of the project was to develop a new HRQoL instrument for children and adolescents with a chronic medical condition and their parents [39]. Seven chronic conditions were included in the project: asthma, juvenile idiopathic arthritis (JIA), atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes and epilepsy. The instrument aims to consist of aspects that are important to the patient, be multidimensional, cross-nationally applicable, valid, reliable and sensitive [40]. The developmental process followed predefined steps (Box 1). The final HRQoL instrument consists of two modules: a chronic generic module that is applicable to all children or adolescents with a chronic medical condition and condition-specific modules for each of the seven included chronic conditions. Central to the DISABKIDS methodology was the 'bottom-up' construction or patient-derived method. Relevant HRQoL aspects were identified from the perspective of the child, the adolescent, their parents and health care professionals. Unique to this project is that focus groups were run cross-nationally. Subsequently the collected HRQoL statements are formulated into items and incorporated into the chronic generic or a condition-specific module [41].

1: Literature review
2: Focus groups
3: Item selection
4: Translations
5: Pilot testing
6: Analysis pilot study
7: Field study
8: Analysis field study
9: Implementation

Box 1. Developmental steps within the DISABKIDS project.

Participants

Children (aged 4-7 and 8-12) and adolescents (aged 13 -16) with a chronic medical condition and their parents were invited to participate in the focus groups. Families were identified through patient associations or their hospital clinicians. They received an invitation by mail and were phoned to ask whether they would participate. Patients with additional chronic conditions or who did not verbally master the national language were excluded. The focus groups were divided by chronic condition and stratified by age (4-7, 8-12 and 13-16 years). Each focus group consisted of a mixture of disease severity and gender. If participants were unable to attend a planned focus group they could take part in a personal interview. Parents were invited to participate in separate focus groups (grouped according to the chronic condition and age of their child). Health care professionals were contacted directly and where often affiliated to the research centres. The responsible local Ethics Committees approved the study and all participants signed a consent form.

Focus groups

To insure that a similar method was used in all participating countries a manual was written which included the outline of the focus group process and the question structure. A moderator led the focus groups and asked the questions, while an assistant observed, wrote minutes and operated a tape recorder. At the start of the focus groups the aim, duration and confidentiality was explained. Permission was also obtained for audio-taping. The moderator started with the semi-structured questions, given in the manual, and allowed the participants to direct the discussion. During the session the questions were directed towards more illness related topics. The moderator made sure to create a safe environment, encouraged involvement and probed for comments. As a closing exercise children and adolescents were asked to write their own questions of what would give a clinician a good impression of their HRQoL. Approximately 90 minutes was planned for each session. At the end, all participating children and adolescents received a gift. The focus groups and interviews were transcribed verbatim. This raw data was used for two purposes. Firstly, the transcripts were used to illustrate the discussed themes and the perceptions of the participants. Secondly, statements related to HRQoL were identified from the national transcripts by investigators in each country. These statements were entered into a database and used for the development of the DISABKIDS items [41].

Results

Population

Asthma focus groups and interviews were conducted in Greece, Italy, the Netherlands and Sweden from June to August 2001. A total of 9 child/adolescent focus groups and 8 parent groups were conducted, with an average of two of each in every country. In total, 43 children and adolescents with asthma (6, 23 and 14 in the age group 4-7, 8-12 and 13-16, respectively) and 33 family members participated in the focus groups and interviews. Generally the focus groups consisted of 3-6 participants. The Dutch centre also included two focus groups with experts (4 asthma nurses and 3 paediatricians), in order to collect their opinions and to enable comparison with the child or parent's view.

Qualitative focus group results

Child and adolescent

Children and adolescents indicated that it was bothersome to take medication on a daily basis. They often forgot to take the medication and some felt that they didn't need medication when they had no complaints. It was noted that those who discussed non-compliance to the medical regimen were also uncertain about how the medication worked or how it affected them (Box 2). Some found that they were insufficiently informed about side effects, worried about taking prednisolon or had an aversion to frequently changing their medication. The younger children complained about the taste of the medication. Most said that they did not mind taking medication in front of others but they tended to avoid this as much as possible. When discussing their relationship with the clinician some indicated that they would prefer the clinician to talk to them on a more personal level. Others were happy with the ways things were and felt understood. Especially the younger children disliked specific procedures such as getting injections, blowing peak flow or lung function tests. Hospital admittance generally had a negative impact on the child and adolescent.

The children and adolescents experienced several physical symptoms related to having asthma. They complained about their cough, feeling short of breath or feeling like they are breathing through a straw. Symptoms were influenced by cigarette smoke, dust, sports, weather seasons or emotions. They indicated that they got tired easily, had less energy than their peers and experienced limitations during physical activities. Sport is a recurring topic in the focus groups. The majority had experienced the need to stop during sport because they were out of breath or didn’t have enough energy to keep up with the rest of the group. This is troublesome because they feel left out of the group. Children and adolescents with allergies to pollen, dust or animals experienced even more limitations. These irritants are influential in classrooms, at a friends place and on school trips. Having to avoid certain places and activities has an impact on everyday life in terms of encountered limitations. The younger children often spoke about not being allowed to play with pets or missing their cuddly toys. The adolescents knew that they needed to avoid cigarette smoke but found it hard, as going to a friend's place or going out sometimes makes this unavoidable.

Non-compliance:
- The teacher said that medication is bad
- I shake because of the side effects …... so I don’t take my medication on the days I have a school-test
- I don't take medication ………to forget that I have asthma

Medical care:
- I am bothered by having to visit the hospital when I have an asthma attack
- I don't like going to the doctor to do injections for the allergy
- Lying in the hospital, that is most bothersome
- Family doctor never lets you finish talking

Limitations:
- Having asthma is not so nice, because you can't join in so many games
- I dislike the fact that I have to have a rest when I run
- Cannot go to parties at homes with pets

Social:
- The kids at school don't know that I have asthma
- Others go and tease you with it
- School parties are unpleasant because of the smoking
- They don't understand that you're short of breath
- I don't talk with my friends about my problem
- X understands that he is different from the other children and is worried about it

Box 2. Statement examples from the focus groups and interviews.

Children and adolescents don't want to feel any different from their peers. They find it important to belong to a group and to go out with friends. However, especially in the adolescent groups we found that due to the asthma they often felt different and left out of school activities. Experienced physical limitations made it harder to keep up with peers. Being ill, missing school or being behind in schoolwork sometimes prevented them from participating in social activities.

Some become angry when they are restricted in what they want to do. Others worry about getting teased because they cannot keep up with sports or need to take medication at school. For this reason some avoid mentioning that they have asthma. Most have come across not being understood by others, not only with peers but also with teachers or adults. School was not mentioned much by the younger group. Adolescents found it frustrating to miss school or school tests due to asthma complaints, hospital check-ups or admissions. This made it harder to keep up with the school's learning schedule. They indicated that it is important that the teachers understand the implications of having asthma.

Parents

The main issues discussed by the parents' concerned medication and emotional effects of asthma. Most parents indicated that there were frequent arguments because of their child not wanting or forgetting to take their medication. Parents worry about possible side effects from the asthma medication, especially about any long-term effects and whether their children can lead a healthy life in the future. Parents frequently discussed health care and the treatment they receive from clinicians. They want more information about health care facilities and medication. Having a doctor who listens and explains this is very important to them.

Parents know that their children are not able to participate in all activities. They acknowledged that their children get tired easily, cannot compete as well in some sports and avoid extreme activities. They also mentioned that their child misses out on certain activities as playing at a friend's house or having sleepovers. They believe that this makes it harder for them to make friends and may make them feel different from their peers. Parents also acknowledged that some children try to hide their asthma from friends. Some worry about their children being ashamed of having asthma. They find it hard to find out how their child really feels and how they cope with their asthma.

A few parents think that their children are more grown up, tougher, have more drive to prove themselves because of the asthma. Others parents worry about the amount of responsibility a child can have. They are constantly checking on their kids, what they can and can't do. Parents are concerned about their child’s vulnerability and try to protect them from exhaustion and irritants (i.e. limiting activities, keeping their home clean, getting rid of pets or cuddly toys). Their main aim is to prevent their child from getting asthma symptoms. The downside is that this sometimes restricts their child, for instance in developing social contacts. School performance was also a frequently discussed topic. Parents worry about the effect asthma had on the child's schoolwork due to getting tired easily, missing school or redoing tests. They try to inform the school on the impact of asthma and the importance of avoiding irritants. However, not all teachers understand the implications of asthma.

Country specific issues

In general similar topics were discussed in all four countries. There were some country specific aspects like riding a bike in the Netherlands, going to the beach in Greece and into the mountains in Italy. Greece was the only country where allergies, pets and difficulty to visit friends were not discussed in the focus groups.

Age differences

Children in the age group 4-7 did not spontaneously speak about their illness. They discussed basic things like what games they liked. Some discussed getting tired when running or playing. They often spoke about wanting pets. Children aged 8-12 talked about the more practical aspects as needing to use medication, not being able to do as well as other kids in sports or physical activities, about their symptoms bothering them and about not wanting to take medication. Some also discussed wanting a pet but not being allowed to have one. Next to the practical implication of using medication the teenaged group (aged 13-16) discussed more topics related to social activities. They discussed being less able to join in sports and physical activities or that some social activities were a problem (school parties, going out, disco, staying at a friends place, family gatherings). Most issues related to trying not to be any different from others and fitting into the peer group.

Health care professionals

The Dutch paediatricians and specialised asthma nurses that participated in the focus groups reported that it is difficult to understand the child's perspective. Current emphasis is on the presentation of symptoms and objective measurements of the disease. They try to specifically ask the child for their views as they realise that there might be a discrepancy between what parents tell them and what the children and adolescents experience. The problem as they see it is asking the right questions. They generally ask questions about sports and use of medication, but know that the answers may not reflect the actual problem the child is experiencing. Adolescents rarely talk about problems spontaneously. The clinicians and nurses acknowledge this as a problem as they know that they might miss valuable information about aspects in the adolescents' live, such as associated problems at school or in their social life. However, especially the clinicians feel limited by the time they have available when running a busy outpatient clinic.

Quantitative asthma focus group results

Item pool description

A total of 637 statements related to HRQoL were identified from the asthma focus group and interview transcripts and pooled in an asthma data bank. Three hundred and four statements were recognized as being specifically asthma related (condition-specific). These statements were used to develop the asthma specific module of the DISABKIDS instrument. Three hundred and thirty three statements could be appropriate to a child or adolescent with any chronic medical condition (chronic generic) and were not specific to having asthma. These chronic generic statements, collected from the asthma transcripts, were merged with the chronic generic statements collected from the focus groups and interviews of the other chronic medical conditions.

The statements collected from the asthma focus groups and interviews were given domain names by two investigators (RMB and JEC). Their interrater reliability was 0.52 (Cohen's kappa). When the given domain differed a third investigator (HMK) assigned the final domain name (Table 3). Most collected statements entered into the database were related to symptoms, treatment or medical care (35%). Statements related to limitations and restrictions mostly originated from the younger children and their parents. The children and adolescents generated most of the social statements. The adolescent's parents generated the majority of the statements related to school.

Domain	All 637 statements (%)	Percentile distribution of statements per group*			
		Child (8-12 yr)	Parent (8-12 yr)	Adolescent (13-16 yr)	Parent (13-16 yr)
Medical/hospital/doctor	20	20	19	18	18
Symptoms/complaints	15	21	12	15	11
Psychological / emotion	14	11	19	16	14
Limitations / restrictions	13	13	16	7	9
Social (friends/peers)	8	10	4	14	6
Physical / sport	7	8	5	6	7
School	7	4	7	8	13
Coping	7	5	11	6	11
Family / home	6	7	3	5	5
Health care	3	1	4	5	6

Table 3. Domain distribution of the collected statements, from children, adolescents and their parents in the asthma focus groups and interviews (%).

*The collected statements from children in the 4 -7 age group, their parents and health care professionals are not shown due to the small numbers.

Discussion

This study describes the qualitative patient-derived research method applied in the DISABKIDS project to explore the patient's view on the influence of asthma on their daily life. As stated earlier there are several advantages to using cross-national focus groups for item generation. Collected statements come directly from the target population and provide access to the child and adolescent's own language. In addition the probability of cultural bias will be reduced as a result of the cross-national setup where uniformity of the items between countries was sought after.

We found that focus groups with children and adolescents were more difficult compared to adult groups. Especially the youngest children (aged 4-7) were not really capable of expressing their opinions or feelings in more than a few words. Overall, children often preferred to wait until someone else had answered a question or the question was directed to them personally. This in contrast to the groups with parents and health care professionals were the conversations went almost automatically. Even for the adolescents, 90 minutes turned out to be the maximum time they could focus on the topic. Like others we found that near the end the motivation declined, the responses were less extensive and more irrelevant topics were raised [27]. In general the children and adolescents did feel acknowledged as being the experts and expressed that they would volunteer to participate again. The parents appreciated the chance to talk about their experiences and found it especially valuable to hear from others in a similar situation.

During the focus groups the children and adolescents admitted their non-compliance, they said that they forgot to take their medication, believed that it did not work, thought it was not necessary or wanted to avoid any side effects. Similar aspects related to non-compliance have been found in previous studies [42-45]. Social issues were found to be a dominant theme, especially for the adolescent, which was similar in other studies, even in other chronic conditions [34,35,46,47]. The impact of physical limitations and prevention measures on a child or adolescent's social life has also been illustrated before [45]. Adolescents indicated that they did not want to be seen or be treated as different and wished to be accepted by their peers. Younger children were more concerned with the actual physical limitations they experienced (running, riding a bike, sports). Parents were mostly troubled about medical aspects concerning insufficient confidence in medication, side effects, little knowledge of the medication or the treatment plan and fear of any long-term consequences. Similar outcomes have been discussed in other studies reporting that the children found the symptoms and limited physical activities most bothersome and that parents were worried about the medication, long-term effects of the illness and feeling helpless when a child has an asthma attack [11,46,48-50]. Not all topics were discussed in each country. For example allergy, pets and difficulty to visit friends were not discussed in Greece. This might be related to the fact that their living conditions are different from other countries and can be a reflection of selection bias as most of these children had exercise-induced asthma.

The focus groups with the health care professionals illustrated that parents and clinicians have different aims. While the parents and children often aim at limiting the use of medication as much possible, health care professionals accept a higher dose of medication to accomplish minimal physical limitations. This can cause a conflict in aims and influence non-compliance. Health care professionals indicated that they find it hard to recognise issues that are of importance to the patient, and that they mostly concentrated on the functional ability. The health care professionals do realise that if a child is not happy about using medication the chances increase that they will not use the medication properly. They also understand that they should explain their decisions more clearly to the child and parents to enhance compliance with medication. The risk of a child or adolescent becoming socially isolated or not being able to join in all activities was acknowledged. However, health care professionals suggest that this might also relate to a child's personality.

On the whole, asthma has a major impact on the child and adolescent's life. Keeping up with aspects important to the child or adolescent’s HRQoL is important to facilitate and improve clinical practice. Recognition and knowledge of any problems can enable clinicians and nurses to help and support these families. Another aspect illustrated by the focus groups discussions was that children and adolescents concentrate on the 'here and now'. Their focus is on the present limitations they experience and how this influences them in daily life. The parent's centre of attention is on the future (schooling, future jobs) and they aim for the lowest possible medication doses for fear of long-term consequences. Clinicians concentrate on the physical function, reducing the current symptoms, aiming for the best possible lung function and optimising treatment compliance.

Some methodological issues need to be discussed. One issue concerns selection bias. Participants may not be representative for the population of interest as the more confident individuals can be more willing to participate. Those who did not join may have a different view on their illness and the effect it has on their lives. Another point is that the total group of children, adolescents and parents was also relatively small. On the other hand the discussed topics were widespread and showed similarities between countries. As a result the collected statements were acknowledged as important to our research population. While some investigators have conducted focus groups or interviews till no new issues were presented, this was not possible in the DISABKIDS project due to time constraints [35,37,51].

Despite the fact that dynamic group interaction can stimulate additional information, interaction can also be inhibited. One person's opinion can prevail in the group by silencing less confident participants or by constantly changing topics [31,52]. Personal expectations of participants, group incompatibility or lack of respect may also cause problems, as can mixed gender groups [32,53-55]. In our focus groups the moderator needed to deal with limited interaction between participants, the more dominant child, the quiet child and, sometimes different priorities between the girls and boys.

Collected focus group data can only be interpreted or reported in a qualitative manner, with selected phrases and quotes as outcome. The strength of a certain viewpoint cannot be measured by counting the number of collected statements or by the intensity in which it is expressed [31]. Firstly, the amount of collected statements within a group can depend on the number of conducted focus groups and interviews. Secondly, the discussed issues can be biased as a result of the questions asked by the moderators or the interaction within the group [31]. Additionally, the selection of HRQoL statements from the transcripts depends on the investigator. The distribution of the collected statements (Table 3) therefore functions solely as illustration. Nevertheless, the fact that major topics were discussed in all four countries and that obtained results were similar to earlier studies helps to strengthen the validity of the findings.

There was no objective method for extracting the HRQoL statements from the literal transcripts. One DISABKIDS member identified the appropriate statements in each country. The quality of individual content analysis has been questioned, as the agreement between raters can be low and individual raters may not extract all information [56]. However, other investigators have suggested that individual judgement is valid and they find external or group ratings unnecessary as the context in which the data were conducted may be missed [57,58]. A computer program can also be used to code and analyse the transcripts but has its limitations and was not applicable in the DISABKIDS project due to the different languages of the transcripts [58,59].

Appointing domains to the statements also included a subjective element, with the first two raters disagreeing on quite a number of the statements. However, this does not indicate that one or the other is incorrect, as some statements can be applicable to several domains. The most frequently mentioned domains included the medical, symptoms, psychological and limitation domains, which is similar to the domains incorporated in most current paediatric questionnaires [60]. This means that these topics are either important or we recognised these HRQoL statements because we are familiar with these topics from earlier research [40].

The need to assess HRQoL in different countries and cultures is increasingly being discussed [61-63]. However, when a HRQoL questionnaire is developed in one country and is translated into another language this might cause cultural obstacles and true comparability may not be achieved [64-66]. Developing a HRQoL questionnaire cross-nationally can limit cultural and socio-economic influences. This brings us to the advantage of the cross-national method within the DISABKIDS project. The focus groups and interviews were an effective way to gather topics that were important and appropriate to children and adolescents in several countries. This benefited the international consensus that was achieved within the DISABKIDS group by selecting those items that reflected the universal concerns of our research population. Only a few HRQoL questionnaire have been developed in simultaneous collaboration with different countries [16,62,64,67]. One well-known project is the World Health Organization Quality of Life (WHOQOL) project, which works in collaboration with several countries around the world. However, this concerns adult research [68].

Conclusion

Overall, the focus groups and interviews gave insight into a range of important issues and viewpoints, identified recurring themes between groups and generated items for the new DISABKIDS instrument. It provided an impression of the HRQoL of children and adolescents with a chronic medical condition like asthma and ensured that aspects found important to the patients were included. However, one must keep in mind that a substantial amount of time and effort does need to be put into the organisation of focus groups and that there are special demands in moderating, transcribing and analysing the data. The information we have gathered is not only of importance to the DISABKIDS project but also for those who work with young patients in clinical practice or research.

Acknowledgements

We would like to thank S. Feith, N. Mourad, A. Engelberts, L. Bierlage, W. Maat, M. Mulder, T. Rodijk, P.L. Brand, F. Popma, K. Röben, N. Dors and all the participating families for joining, organising, moderating and transcribing the focus groups and interviews. The DISABKIDS project was supported by the European Commission (QLG5-CT-2000-00716) within the Fifth Framework Program "Quality of Life and Management of Living Resources".

References

1. Eiser C, Morse R. A review of measures of quality of life for children with chronic illness. Arch.Dis.Child 2001;84(3):205-11.
2. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol Assess 2001;5(4):1-157.
3. Juniper EF, Guyatt GH. Asthma and allergy. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trails. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 977-82.
4. Rutishauser C, Sawyer SM, Bowes G. Quality-of-life assessment in children and adolescents with asthma. Eur.Respir.J. 1998;12(2):486-94.
5. Harding L. Children's Quality of life assessments: a review of generic and health related quality of life measures completed by children and adolescnets. Clin. Psychol.Psychother. 2001;8:79-96.
6. Landgraf JM, Abetz N. Measuring health outcomes in pediatric populations: issues in psychometrics and application. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trails. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 793-802.
7. Rajmil L, Herdman M, Fernandez de Sanmamed MJ, Detmar S, Bruil J, Ravens-Sieberer U et al. Generic health-related quality of life instruments in children and adolescents: a qualitative analysis of content. J.Adolesc.Health 2004;34(1):37-45.
8. Fekkes M, Theunissen NC, Brugman E, Veen S, Verrips EG, Koopman HM et al. Development and psychometric evaluation of the TAPQOL: a health-related quality of life instrument for 1-5-year-old children. Qual.Life Res. 2000;9(8):961-72.
9. Creer TL, Wigal JK, Kotses H, Hatala JC, McConnaughy K, Winder JA. A life activities questionnaire for childhood asthma. J.Asthma 1993;30(6):467-73.
10. Ronen GM, Streiner DL, Rosenbaum P. Health-related quality of life in children with epilepsy: development and validation of self-report and parent proxy measures. Epilepsia 2003;44(4):598-612.
11. Townsend M, Feeny DH, Guyatt GH, Furlong WJ, Seip AE, Dolovich J. Evaluation of the burden of illness for pediatric asthmatic patients and their parents. Ann.Allergy 1991;67(4):403-8.
12. Usherwood TP, Scrimgeour A, Barber JH. Questionnaire to measure perceived symptoms and disability in asthma. Arch.Dis.Child 1990;65(7):779-81.
13. Eiser C, Havermans T, Craft A, Kernahan J. Development of a measure to assess the perceived illness experience after treatment for cancer. Arch.Dis.Child 1995;72(4):302-7.
14. Starfield B, Bergner M, Ensminger M, Riley A, Ryan S, Green B et al. Adolescent health status measurement: development of the Child Health and Illness Profile. Pediatrics 1993;91(2):430-5.

15. Griffiths AM, Nicholas D, Smith C, Munk M, Stephens D, Durno C et al. Development of a quality-of-life index for pediatric inflammatory bowel disease: dealing with differences related to age and IBD type. J.Pediatr.Gastroenterol.Nutr. 1999;28(4):S46-S52.
16. Bullinger M, Von Mackensen S, Fischer K, Khair K, Petersen C, Ravens-Sieberer U et al. Pilot testing of the 'Haemo-QoL' quality of life questionnaire for haemophiliac children in six European countries. Haemophilia. 2002;8 Suppl 2:47-54.
17. Juniper EF, Guyatt GH, Jaeschke R. How to develop and validate a new health-related quality of life instrument. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trails. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 49-56.
18. Groll D, Vanner SJ, Depew WT, DaCosta LR, Simon JB, Groll A et al. The IBS-36: a new quality of life measure for irritable bowel syndrome. Am.J.Gastroenterol. 2002;97(4):962-71.
19. Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. Qual.Life Res. 2001;10(4):347-57.
20. Theunissen NC, Vogels TG, Koopman HM, Verrips GH, Zwinderman KA, Verloove-Vanhorick SP et al. The proxy problem: child report versus parent report in health-related quality of life research. Qual.Life Res. 1998;7(5):387-97.
21. Guyatt GH, Juniper EF, Griffith LE, Feeny DH, Ferrie PJ. Children and adult perceptions of childhood asthma. Pediatrics 1997;99(2):165-8.
22. Rothwell PM, McDowell Z, Wong CK, Dorman PJ. Doctors and patients don't agree: cross sectional study of patients' and doctors' perceptions and assessments of disability in multiple sclerosis. BMJ 1997;314(7094):1580-3.
23. Janse AJ, Gemke RJ, Uiterwaal CS, van dT, I, Kimpen JL, Sinnema G. Quality of life: patients and doctors don't always agree: a meta-analysis. J.Clin.Epidemiol. 2004;57(7):653-61.
24. Williams J, Williams K. Asthma-specific quality of life questionnaires in children: are they useful and feasible in routine clinical practice? Pediatr.Pulmonol. 2003;35(2):114-8.
25. Sung L, Young NL, Greenberg ML, McLimont M, Samanta T, Wong J et al. Health-related quality of life (HRQL) scores reported from parents and their children with chronic illness differed depending on utility elicitation method. J.Clin.Epidemiol. 2004;57(11):1161-6.
26. Gill TM, Feinstein AR. A critical appraisal of the quality of quality-of-life measure ments. JAMA 1994;272(8):619-26.
27. Ronen GM, Rosenbaum P, Law M, Streiner DL. Health-related quality of life in childhood disorders: a modified focus group technique to involve children. Qual.Life Res. 2001;10(1):71-9.
28. McEwan MJ, Espie CA, Metcalfe J. A systematic review of the contribution of qualitative research to the study of quality of life in children and adolescents with epilepsy. Seizure. 2004;13(1):3-14.

29. Kitzinger J. Qualitative research. Introducing focus groups. BMJ 1995;311(7000):299-302.
30. Krueger RA. Group dynamics and focus groups. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trails. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 397-402.
31. Sim J. Collecting and analysing qualitative data: issues raised by the focus group. J.Adv.Nurs. 1998;28(2):345-52.
32. Heary CM, Hennessy E. The use of focus group interviews in pediatric health care research. J.Pediatr.Psychol. 2002;27(1):47-57.
33. Barbour RS. The use of focus groups to define patient needs. J.Pediatr.Gastroenterol. Nutr. 1999;28(4):S19-S22.
34. French DJ, Christie MJ, West A. Quality of life in childhood asthma: development of the Childhood Asthma Questionnaires. In: Christie MJ, French DJ, editors. Assessment of Quality of Life in Childhood Asthma. Chur, Switzerland: Harwood Academic Publishers; 1994. p. 157-80.
35. McEwan MJ, Espie CA, Metcalfe J, Brodie MJ, Wilson MT. Quality of life and psychosocial development in adolescents with epilepsy: a qualitative investigation using focus group methods. Seizure. 2004;13(1):15-31.
36. Ronen GM, Rosenbaum P, Law M, Streiner DL. Health-related quality of life in child hood epilepsy: the results of children's participation in identifying the components. Dev.Med.Child Neurol. 1999;41(8):554-9.
37. Rutishauser C, Sawyer SM, Bond L, Coffey C, Bowes G. Development and validation of the Adolescent Asthma Quality of Life Questionnaire (AAQOL). Eur.Respir.J. 2001;17(1):52-8.
38. Akobeng AK, Suresh-Babu MV, Firth D, Miller V, Mir P, Thomas AG. Quality of life in children with Crohn's disease: a pilot study. J.Pediatr.Gastroenterol.Nutr. 1999;28(4):S37-S39.
39. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. Int.J.Rehabil.Res. 2002;25(3):197-206.
40. Herdman M, Rajmil L, Ravens-Sieberer U, Bullinger M, Power M, Alonso J. Expert consensus in the development of a European health-related quality of life measure for children and adolescents: a Delphi study. Acta Paediatr. 2002;91(12):1385-90.
41. Petersen C, Schmidt S, Power M, Bullinger M, and the DISABKIDS group. Development and pilot-testing of a health-related quality of life chronic generic module for children and adolescents with chronic health conditions: A European perspective.
Qual Life Res. 2005;14(4):1065-77.
42. van Es SM, le Coq EM, Brouwer AI, Mesters I, Nagelkerke AF, Colland VT. Adherence-related behavior in adolescents with asthma: results from focus group interviews. J.Asthma 1998;35(8):637-46.

43. Chambers CV, Markson L, Diamond JJ, Lasch L, Berger M. Health beliefs and compliance with inhaled corticosteroids by asthmatic patients in primary care practices.
Respir.Med. 1999;93(2):88-94.
44. Buston KM, Wood SF. Non-compliance amongst adolescents with asthma: listening to what they tell us about self-management. Fam.Pract. 2000;17(2):134-8.
45. Penza-Clyve SM, Mansell C, McQuaid EL. Why don't children take their asthma medications? A qualitative analysis of children's perspectives on adherence. J.Asthma 2004;41(2):189-97.
46. Arunkumar G, Wyllie E, Kotagal P, Ong HT, Gilliam F. Parent- and patient-validated content for pediatric epilepsy quality-of-life assessment. Epilepsia 2000;41(11):1474-84.
47. Edwards TC, Huebner CE, Connell FA, Patrick DL. Adolescent quality of life, part I: conceptual and measurement model. J.Adolesc. 2002;25(3):275-86.
48. Kieckhefer GM. The meaning of health to 9-, 10-, and 11-year-old children with chronic asthma. J.Asthma 1988;25(6):325-33.
49. Bender BG. Overcoming barriers to nonadherence in asthma treatment. J.Allergy Clin.Immunol. 2002;109(6 Pt 2):554S-9S.
50. Kieckhefer GM, Ratcliffe M. What parents of children with asthma tell us. J.Pediatr. Health Care 2000;14(3):122-6.
51. McLafferty I. Focus group interviews as a data collecting strategy. J.Adv.Nurs. 2004;48(2):187-94.
52. Beyea SC, Nicoll LH. Learn more using focus groups. AORN J. 2000;71(4):897-900.
53. Kennedy C, Kools S, Krueger R. Methodological considerations in children's focus groups. Nurs.Res. 2001;50(3):184-7.
54. Horner SD. Using focus group methods with middle school children. Res.Nurs. Health 2000;23(6):510-7.
55. Peterson-Sweeney K. The use of focus groups in pediatric and adolescent research. J.Pediatr.Health Care 2005;19(2):104-10.
56. Weinberger M, Ferguson JA, Westmoreland G, Mamlin LA, Segar DS, Eckert GJ et al. Can raters consistently evaluate the content of focus groups? Soc.Sci.Med. 1998;46(7):929-33.
57. Webb C, Kevern J. Focus groups as a research method: a critique of some aspects of their use in nursing research. J.Adv.Nurs. 2001;33(6):798-805.
58. Chapple A, Rogers A. Explicit guidelines for qualitative research: a step in the right direction, a defence of the 'soft' option, or a form of sociological imperialism? Fam. Pract. 1998;15(6):556-61.
59. Aljunid S. Computer analysis of qualitative data: the use of ethnograph. Health Policy Plan. 1996;11(1):107-11.
60. Andelman RB, Zima BT, Rosenblatt AB. Quality of life of children: toward conceptual clarity. In: Maruish ME, editor. The use of psychological testing for treatment planning and outcomes assessment. Mahwah, NJ: Lawrence Erlbaum; 1999. p. 1383-413.

61. Guillemin F, Bombardier C, Beaton D. Cross-cultural adaptation of health-related quality of life measures: literature review and proposed guidelines. J.Clin.Epidemiol. 1993;46(12):1417-32.
62. Stahl E, Postma DS, Juniper EF, Svensson K, Mear I, Lofdahl CG. Health-related quality of life in asthma studies. Can we combine data from different countries? Pulm. Pharmacol.Ther. 2003;16(1):53-9.
63. French DJ, Carroll A, Christie MJ. Health-related quality of life in Australian children with asthma: lessons for the cross-cultural use of quality of life instruments. Qual.Life Res. 1998;7(5):409-19.
64. Schmidt S, Bullinger M. Current issues in cross-cultural quality of life instrument development. Arch.Phys.Med.Rehabil. 2003;84(4 Suppl 2):S29-S34.
65. Swaine-Verdier A, Doward LC, Hagell P, Thorsen H, McKenna SP. Adapting quality of life instruments. Value.Health 2004;7 Suppl 1:S27-S30.
66. Acquadro C, Jambon B, Ellis D, Marquis P. Language and translation issues. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trials. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 575-85.
67. Alonso J, Ferrer M, Gandek B, Ware JE, Jr., Aaronson NK, Mosconi P et al. Health-related quality of life associated with chronic conditions in eight countries: results from the International Quality of Life Assessment (IQOLA) Project. Qual.Life Res. 2004;13(2):283-98.
68. The WHOQOL group. The World Health Organization Quality of Life assessment (WHOQOL): position paper from the World Health Organization. Soc.Sci. Med. 1995;41(10):1403-9.

5

Item selection after focus group research: the European DISABKIDS approach

R.M. Baars, S. Schmidt, M. Bullinger, A.H. Zwinderman, J.M.Wit, H.M. Koopman and the DISABKIDS group.

Good answers come from good questions, not from esoteric analysis (H.M. Schoolman, 1968).

Abstract

A description is given of how items were selected for the DISABKIDS pilot instrument from a pool of health related quality of life (HRQoL) statements. To insure a bottom-up procedure the collected statements were generated from simultaneous cross-national focus groups and interviews with children and adolescents with a chronic medical condition, their parents and health care professionals. The DISABKIDS approach included a sequence of reduction methods. Steps included a redundancy scoring to delete the double, unclear and irrelevant statements. Secondly, experts rated the statements for importance. The statements were then rewritten into an appropriate questionnaire format. A final card sorting method was applied to select domains and dimensions. Last refinements were made to remove any further unethical, unclear or similar items. Three hundred and twenty-two participants contributed to the focus groups and interviews. A total of 3515 HRQoL statements were identified from the literal transcripts. These included generic quality of life statements (n=488) not related to any medical condition, chronic generic statements (n=1647) related to any chronic medical condition and condition-specific statements (n=66 to 340) that related to one of the seven included chronic medical conditions. After the applied redundancy steps a 100 chronic generic items and 26 to 44 condition-specific items were retained for testing in the DISABKIDS pilot study. Several sequential reduction steps were necessary to cut down the vast amount of collected statements from the DISABKIDS focus groups and interviews to develop our pilot instrument. These steps determine the final construct of a questionnaire. In our case it was found that the card sorting method was the most comprehensive and straightforward method to reduce the statements and divide the items into domain groups.

Introduction

A fundamental process in the development of health related quality of life (HRQoL) questionnaires is the selection of items. Qualitative patient-derived research methods, as the focus group and interview, have increasingly been used to collect items for new HRQoL questionnaires [1-6]. Organisational aspects related to running focus groups and the resulting qualitative data (i.e. discussed themes) have also been regularly described [7-13]. However, we found that existing literature gives no standard criteria or clear description on how to select questionnaire items from the collected qualitative data [14-16]. Processing raw qualitative data to form an item pool seems to be a variable and possibly subjective process. Steps for selecting the item pool should be objectively described, as failure to explain this process results in a difficulty to understand how selection choices were or can be made [17]. Applied development and selection methods can also influence the outcome of the final questionnaire [18]. A clear description of the development and processing of qualitative data is thus required to improve the quality of qualitative research [16,19,20].

Investigators have used patient-derived methods, as the focus group and interview, to generate items for new questionnaires but have not described or are unclear about the item selection process from their raw qualitative data [1,3,5,21-25]. If item reduction methods were described these concerned limited item pools or the selection of items after the pilot or field testing of a questionnaire. Described methods include clinicians or investigators selecting items, principal components analysis, criterion keying or asking patients to score items on importance, frequency or severity [1-3,23,26,27]. The clinimetric technique or "clinical impact" method is commonly used and provides an item rank order based on item importance, frequency or severity ratings provided by patients [28]. Scores of the individual patient or the means of the patient group are either multiplied or added to establish an item rank order (Box 1). The applied clinimetric technique does not seem to affect the general result of the item reduction [28]. The top ranking items are rated as the most important as they represent the highest experienced burden or impact.

*Multiplication or addition of the individual's importance score (I) on an item with the individual's severity (S) score of that item. The resulting product is an individual item importance-severity (M) or importance-plus-severity (P) score, which can be averaged over the group.
Formula: I x S =M   or   I + S = P

*The mean importance score of the group (I^m) can be multiplied with or added to the mean severity value for the entire group (S^m). The resulting overall group importance-severity score (M) or importance-plus-severity score (P) can be used to rank the items.
Formula: I^m x S^m = M   or I^m + S^m = P

*Multiplication or addition of the individual's importance score (I) on an item with the individual's frequency (F) score of that item. The resulting product is an individual item importance-frequency (M) score or importance-plus-frequency score (P), which can be averaged over the group.
Formula: I x F =M   or   I + F = P

*The mean importance score of the group (I^m) can be multiplied with the frequency ($F^{\%}$) of which an item occurs in a group. The resulting overall group importance-frequency score (P) can be used to rank the items.
Formula: I^m x $F^{\%}$ = P

I =importance, can be scored on a Likert scale (not important to very important)
S =severity of the complaint, can be scored on a visual analogue or ordinal scale
F = frequency, can be scored on a Likert scale (never, sometimes, often etc.)
$F^{\%}$ = proportion of patients that report the item as troublesome (max = 100%)
I^m = mean importance score given to an item by a group of patients who find this troublesome

Box 1. Examples of clinimetric techniques.

Above selection processes all assume that there is a limited list of items. The process of selecting items from a large statement pool generated from qualitative patient-derived data as in the DISABKIDS project has rarely been described [14,16]. The aim of this paper is to describe the stepwise item reduction process that was performed to develop the European DISABKIDS HRQoL instrument for children and adolescents with a chronic medical condition and their parents. Item selection, reduction steps and encountered problems are discussed.

Method

The DISABKIDS project

The DISABKIDS project is a collaboration of eight research institutions in seven European countries (Austria, France, Germany, Greece, the Netherlands, Sweden and the United Kingdom). The project's aim is to develop a European HRQoL instrument for children and adolescents with a chronic medical condition and their parents [29]. The project followed a cross-national approach, which combined a bottom-up and top-down strategy and where items were produced simultaneously in different countries. Only a few HRQoL questionnaires have been developed through such a strategy [30-32].

The final DISABKIDS instrument aims to include aspects that are of importance to the patients and is multidimensional, multilingual and cross-nationally applicable in several countries. Unique is that the DISABKIDS project developed a core chronic generic module, which is applicable to all children or adolescents with a chronic medical condition, supplemented by a condition-specific module, which is specific to a certain medical condition. These two modules make comparative clinical studies possible (i.e. comparison between illness samples) and also provide additional information on a specific disease [33]. Chronic conditions included in the project are asthma, juvenile idiopathic arthritis (JIA), atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes and epilepsy. Existing questionnaires and literature were reviewed to support the framework of the instrument. In order to focus on the child's perspective, statements for the DISABKIDS instrument were collected through a bottom-up procedure based on cross-national patient-derived methods, including focus groups and interviews. These procedures made it possible to reflect on aspects that are important to the patient group and adapt item phrasing to the child's level. The development of the DISABKIDS instrument followed consecutive work packages alternating inductive and deductive steps (Table 1) [31]. Patients and investigators were alternately seen as the appropriate experts during the instrument's construction phases. The DISABKIDS project is closely linked to the KIDSCREEN sister project. This project has developed a generic quality of life (QoL) questionnaire for children of the general population through similar methodology [34,35].

Work package		Scientific source	Process*
1	Literature review	Investigator	Top-down
2	Focus groups	Patient	Bottom-up
3	Item selection	Investigator	Top-down
4	Translations	Investigator	-
5	Pilot testing	Patient	Bottom-up
6	Analysis pilot study	Investigator	Top-down
7	Field study	Patient	Bottom-up
8	Analysis field study	Investigator	Top-down
9	Implementation plan	Investigator	-
10	Implementation study	Patient	Bottom-up
11	Final analysis	Investigator	Top-down

Table 1. Work packages (WP) within the DISABKIDS project.
*Inductive versus deductive processes are named as bottom-up and top-down approaches, respectively.

Collecting patient-derived data

Collected statements from the focus group and interview transcripts functioned as the main item source. Children and adolescents between 4 and 16 years old with one of the seven chronic conditions, their parents and health care professionals participated in the focus groups and interviews. Separate focus groups were organised for children and adolescents with each of the included chronic conditions and their parents. Each group was divided by age and consisted of a mixture of severity and gender. Health care professionals also gave their opinion on relevant HRQoL aspects through focus groups or interviews. Each of the eight DISABKIDS centres planned focus groups: Edinburgh (UK), Hamburg (DE), Leiden (NL), Lübeck (DE), Lund (SW), Marseille (FR), Thessalonica (GR) and Vienna (AU). Verona (IT) participated in the DISABKIDS focus group work package as an affiliated centre. Some participants were unable to attend the planned focus group and participated in individual interviews. To assure that a similar method was used in all centres a manual was developed which included the outline of the focus groups, the structured questions and general guidelines. The responsible Ethics Committees approved the study and all participants signed a consent form.

Item selection

The focus group discussions and interviews were literally transcribed. From these transcripts each country selected statements related to HRQoL. These statements were then translated into English and pooled into an ACCESS database grouped per condition. The ACCESS database was used to reproduce the taken steps, protect the input data with a password and give a clear overview. The statements per chronic medical condition were subdivided into three groups and a general domain name was designated to each statement by the investigator. One group consisted of generic statements, which could be applicable to all children, healthy or with a chronic medical condition. The generic statements from each condition were merged and transferred to the KIDSCREEN project. Merging those statements that were applicable to children with any chronic medical condition formed the second group; the DISABKIDS chronic generic module. The third division of statements formed the seven DISABKIDS condition-specific modules. The statements in each condition-specific module originated from the transcripts of a specific chronic medical conditions and reflected specific aspects related to that medical condition. These three groups were the basis of the modular build-up of the DISABKIDS and KIDSCREEN instrument.

The reduction of the statements was done per module and combined different methods in four steps (Figure 1). All steps have been documented and the final items can be retraced to the original source.

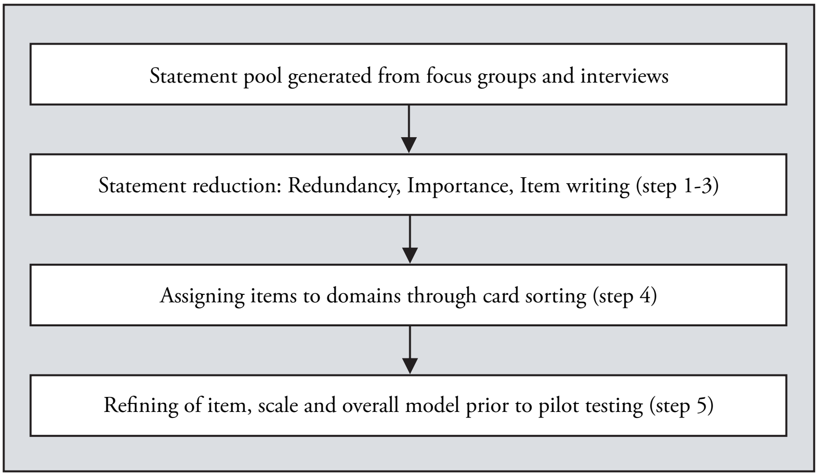


Figure 1. Item reduction progress.

Step 1: Redundancy scoring
The first step was aimed at limiting the excessive amount of statements. A DISABKIDS member from Scotland, Germany and the Netherlands each scored the collected statements derived from the focus groups and interviews on forms in a protected ACCESS database. The reduction of statements was based on criteria presented in the European Health Interview Survey (EUROHIS) study protocol [36]. Three independent raters marked the statements when they were redundant, semantically equivalent or unclear. Statements that did not meet the criteria of the project, like aspects related to health care needs, health care satisfaction or insurance problems, were also marked as failing the criteria. A statement was removed when two or more raters had marked it under one of the exclusion criteria.

Step 2: Importance scoring
The next step was to ask experts to rate the remaining statements in terms of importance. Each chronic generic statement was scored by all centres on importance to the

DISABKIDS HRQoL instrument, relevance to the child's age group (4-7, 8-12 and 13-16) and relevance for a parent questionnaire. Two to three experts from different DISABKIDS centres also scored the condition-specific statements on importance according to the same procedure. An importance scoring of 0-5 was given, with 5 being extremely important.

Step 3: Item writing

Each statement was rewritten into appropriate questionnaire items. Writing instructions were based on a general questionnaire format as presented in the World Health Organization Quality of Life (WHOQOL) study and the EUROHIS study protocol [32,36]. According to these guidelines, items should use simple language, avoid ambiguity in terms of either wording or phraseology, use short sentences, avoid double negatives, be convertible to a rating scale, ask about a single issue only, be applicable to individuals with a range of health status and should be stylistically comparable. At the same time the experts again checked statements for double, unclear or non-HRQoL statements. Each country wrote items for the condition-specific statements collected in their own centre. All chronic generic statements were rewritten and further reduced by the Dutch investigators.

Step 4: Card sorting

The next step consisted of a card sorting process in a DISABKIDS workshop to categorize the items into domains and dimensions [6,37]. This process was performed on the remaining item pool were each item was printed onto a card. The cards from each module (chronic generic and condition-specific) were sorted into three domains: the psychological, social and physical domain, each of which was perceived to be multidimensional. The process started with the chronic generic item pool, which needed to be reduced to roughly 100 items. Three to four DISABKIDS members worked with each of the domains. The cards in each domain were checked for correct placement and grouped according to similarity into several dimensions within this domain. This procedure was continued until all the cards in a domain had been placed in a dimension. If necessary, cards were replaced into another more appropriate domain for sorting. The same process was then applied to the item pool of the seven condition-specific modules with as aim to minimise the item pool to approximately 30 items. Formed expert groups identified the domains and dimensions. All sub-standard or double items were rejected and if the wording was unclear items were corrected.

Step 5: Refinement

In the final step the remaining chronic generic and condition-specific items were read out loud in a DISABKIDS workshop. All attending DISABKIDS members had the opportunity to comment on each statement. Some items were removed or rewritten in instances that they were unethical, had double meaning, and were still redundant or not applicable to all countries.

Results

A total of 154 children and adolescents, 142 family members and 26 health care professionals participated in either focus groups or interviews. A total of 3515 statements were identified from the DISABKIDS focus group and interview transcripts. The 488 generic statements, which were not related to having a disease were sent to the parallel KIDSCREEN project [35]. Of the remaining statements 1647 were applicable to any of the chronic medical conditions (chronic generic) while between 66 and 340 statements were specific to one of the seven chronic conditions (condition-specific). These two groups were the start of the two modules in the development of the DISABKIDS HRQoL instrument.

One DISABKIDS member in Scotland, Germany and the Netherlands scored all of the items in the first redundancy procedure (Step 1), each voted to remove 1404, 1614 and 2239 statements respectively, with between 50 and 68% being scored as semantically equivalent or redundant. Eventually 1802 of the 3027 items were marked under one of the rules by 2 or more raters (Box 2). This left us with 1225 statements, a reduction of 60%.

Fails criteria:
You have got to be well informed, either search the internet or read books.
She had had epilepsy for a long time but it was only noticed when she had a big seizure.
One feels so small at the hospital, you can't find a parking place, it's impersonal, stressed.
We didn't get any compensation for having a child with epilepsy and it costs money.

Semantically equivalent:
Do your teachers know that you have asthma?
The teachers didn't know that he had asthma.
We told them about asthma before the school started.

Box 2. Examples of removed statements in the first reduction phase.

After the redundancy scoring (Step 1) the general domain distribution, as given by each investigator after selecting statements from the literal transcripts, remained the same (Table 2). The health care needs domain was minimised, which was consistent with our aim, as this topic did not meet the criteria for our HRQoL instrument. These items were processed in a separate questionnaire development study [38].

Top 10 domains	Original statements	Remaining statements
Social	428 (14,1 %)	214 (17,5 %)
Coping	396 (13,1 %)	138 (11,3 %)
Health care needs	336 (11,1 %)	46 (3,8 %)
Psychological	262 (8,7 %)	143 (11,7 %)
Physical	253 (8,4 %)	112 (9,1 %)
Emotion	180 (5,9 %)	91 (7,4 %)
Medical	126 (4,2 %)	35 (2,9 %)
Treatment	123 (4,1 %)	52 (4,2 %)
School	95 (3,1 %)	39 (3,2 %)
Family	70 (2,3 %)	17 (1,4 %)

Table 2. General distribution of the top 10 original domains (as provided by each investigator after selection from the literal transcripts) for the combined 3027 chronic generic and condition-specific statements before reduction and the remaining 1225 statements after step 1 (%).

An importance score (Step 2) was given to each of the remaining statements by members of the DISABKIDS group. The interrater agreement between the expert importance ratings varied strongly and the interrater correlations were relatively low (overall ICC = 0.37, ranging from 0.05 to 0.69). Statements from the JIA and CF groups scored high on importance for a HRQoL questionnaire (3.44 (SD 0.59) and 3.48 (SD 0.48) resp.), while the asthma and epilepsy statements scored low on importance (2.77 (SD 0.74) and 2.76 (SD 0.78) resp.). After analysing the results the decision was made to omit this step. It was decided to go directly to the next phase (Step 3): a standard methodology of writing items on the basis of statements. Statements in the database were sent around for item writing, which was based on existing criteria [32,36]. The 1225 statements were divided into groups and rewritten by the DISABKIDS members into items appropriate for our instrument (Table 3). At the same time additional redundant or equivalent items and items not related to HRQoL were identified and removed. This further reduced the data file to 796 items.

Original statement	Rewritten statement
Leading a normal life and being together with others is important.	I can lead a normal life.
I'm glad that I've had arthritis for so long, so I don't know many things I can't do and therefore I didn't have to give anything up.	I accept that there are things I cannot do because of my condition.
It helps when others understand what you have, that they don't tease you.	Others understand what I have.
They want to do everything like the other children... but that is not possible…. afterwards she stays two days in bed, to recover.	I get exhausted easily.
I took her to psychosomatic counselling because she was so depressed (because of the illness) it was really bad.	I was depressed because of my condition.

Table 3. Examples of rewritten statements.

An iterative card sorting procedure (Step 4) was employed to assign items to (a) modules (chronic generic vs. condition-specific), (b) domains within modules and (c) dimensions within domains. At the same time remaining redundant or equivalent items were removed. This resulted in 148 items for the chronic generic module and between 25 to 54 items in each of the condition-specific modules. The rereading of the items in a DISABKIDS workshop again gave a slight reduction (Step 5). The final chronic generic pilot module consisted of 100 items and the condition-specific pilot modules consisted of between 26 and 44 items per condition (Table 4).

Module	Collected statements	After redundancy (step 1)	After item writing (step 3) *	After card sorting (step 4)	Refinement (step 5)
Generic					
Generic	488	-	-	-	-
Chronic generic					
Chronic generic	1647	583	307	148	100
Condition-specific					
Asthma	304	105	85	50	32
JIA	340	142	128	47	44
Atopic dermatitis	66	49	48	43	36
CP	183	121	34	25	26†
CF	167	86	73	54	38
Diabetes	141	62	61	31	28
Epilepsy	179	77	60	31	27
Total	3515	1225	796	429	331

Table 4. Results of the stepwise statement reduction process. Listed are the amounts of remaining statements after each reduction step.

*Step 2 was omitted in the statement reduction procedure.

†An item on independence was re-added.

Discussion

The current opinion in HRQoL research is that patient-derived approaches are required for valid questionnaire development in which a common methodology is the focus group approach [10,16,39,40]. The DISABKIDS project collected qualitative patient-derived data from seven chronic conditions. Unique is that the DISABKIDS project not only collected the items cross-nationally but that the selection steps were also carried out multi-centred, as experts from different countries participated in each step.

We have discussed a combination of steps to reduce the vast amount of collected statements from focus groups and interviews to develop the DISABKIDS pilot instrument. This stepwise process was done in an effort to make the item selection transparent and replicable. It is important to have a database showing all the taken steps so that one can refer to where each statement came from. It is conceivable that such an extensive process has not been presented before, as it is a complex system to describe in detail, often combining objective and intuitive judgements. The bottleneck was that investigators needed to make decisions about data that were supplied by children, adolescents and parents, causing the top-down procedure (investigators) to conflict with the bottom-up procedure (patients).

Before the pilot version items were selected, the DISABKIDS group had several moments of decision making. This started with which questions to ask in the focus groups, which HRQoL statements to select from the literal transcripts, how to translate these statements to one language, how to reduce the amount of statements, how to rewrite statements to items, which domains to utilize and whether all HRQoL aspects were selected. This process was complicated by the large amount of statements (3515 in total), the inclusion of seven chronic conditions and working in a multi-national group.

There are some issues in the DISABKIDS item selection methodology that need to be discussed. First, although the statements were generated from patient-derived methods the final item selection was based on the investigators (top-down) judgement (Table 1). As there was such an extensive amount of statements the DISABKIDS group decided to have the experts (instead of the patients) do the statement redundancy scoring (Step 1) and the importance scoring (Step 2). The data pool was so large that even the experts found it difficult to get an overview of all the statements. This is illustrated in the first reduction step where there was a distinct difference in the amount of statements removed by each of the experts and several redundant or equivalent items were missed. In an effort to bring back the children and adolescent's opinion, we asked them to approve the selected items and judge them on comprehension and applicability in a cognitive interview during the pilot test [41].

Other issues concern the construction of the statement pool. First, there was no opportunity to monitor how each country had selected the HRQoL statements from their national focus group and interview transcripts. The use of a computer was not applicable as the data were in several languages [42,43]. Thus personal interpretations, interests and subjective factors may have influenced the statement selection process [44]. In addition, there is a risk that when statements are taken out of their context they loose their original meaning [45]. Secondly, the investigator was responsible for the translation of the selected statement into English. As there was no official forward-backward translation the quality of the supplied English translation may have altered the meaning of the original statement and influenced the chance of it being used as a final item. Using expert or panel translators in this phase may have improved the quality of the statements and made the literal wording in the questionnaire more likely [46].

The DISABKIDS members provided the expert importance scoring, which was aimed at selecting those items that were fundamental to the DISABKIDS HRQoL instrument. Such an expert importance scoring has been used in other cross-cultural studies, such as the WHOQOL group [32]. Potential limitations resulted from this chosen method. Asking only the experts for the importance of a statement may have linked the importance scoring to their experienced severity of the condition. The JIA and CF statements where generally scored higher than the asthma and epilepsy statements. This indicates that the severity of the condition was probably scored and not the importance of each statement for the HRQoL instrument. Furthermore the given results may not reflect the highest frequency or importance as perceived by the patients with low and high disease severity. The expert scoring came with a risk that certain items (important to the patients) would be rejected in the selection process. Therefore the expert importance scoring (Step 2) was discarded by the DISABKIDS group. In future we would reconsider an importance scoring but per chronic medical condition and for separate degrees of severity, to avoid an item bias in the retained statements. Due to time and organisational constraints it was not possible to redo the importance rating and the step was omitted.

As most statements were literal phrases given by children and adolescents, the statements needed to be rewritten into items appropriate for the instrument. There was a danger revealed in the rewriting of the statements, as some items were no longer related to the original statement. For example: "My teacher thinks that I can't do anything" was changed to "Do you find schoolwork easy?" or "I can get colds much easier than others and have them longer" was rewritten into "Do you worry more than your friends about staying healthy?". Even though these changes lead to a conflict with our bottom-up approach the opinion prevailed that these new items were of importance, and thus remained in the item pool. In future an international item-writing group would be preferred to guard for uniformity and quality.

During the DISABKIDS item development process the card sorting was found to be a comprehensive and straightforward method to apply. As the item cards were grouped in piles, comparison was straightforward and it was easy to divide the items into domains and dimensions, identify redundant or equivalent statements or determine whether items had to be placed into another domain. Theoretically all previous reduction steps could have been performed in the card sorting method. A preferred strategy would have been to apply the earlier reduction steps in a card sorting method and to identify themes instead of statements to avoid a large data pool (Box 3).

• Statement selection	Have two or more experts select the statements from the transcripts
• Card sorting	Look for reoccurring themes per chronic condition and combine these in piles
• Redundancy	Remove excessive and semantically equivalent statements from the card sorting piles
• Modules	Compare the statements between the conditions and merge the overlapping statements to a chronic generic module
• Item writing	Rewrite the remaining statements into items

Box 3. Suggested steps in reducing qualitative data based on expert decisions.

Conclusion

We have described the DISABKIDS reduction process in detail and have shown that a few thousand HRQoL statements can make the item selection process of a new instrument quite complex. The current study shows that expert decisions were necessary to form a pilot instrument. By combining several reduction steps in this selection phase we tried to create the conditions for adequate face and content validity. Describing statement reduction methods has added value for future research as there is currently no basic guide of how to process qualitative patient-derived data. The DISABKIDS pilot and field study will provide further data on internal consistency, stability, validity and reliability [41].

Acknowledgements

To H.P.Sierat for creating an ACCESS database with all the statements. Thanks to J.Bruil, S. Detmar and U. Ravens-Sieberer from the KIDSCREEN group for brainstorming about all the possibilities. The DISABKIDS project was supported by the European Commission (QLG5-CT-2000-00716) within the Fifth Framework Program “Quality of Life and Management of Living Resources”.

References

1. Rutishauser C, Sawyer SM, Bond L, Coffey C, Bowes G. Development and validation of the Adolescent Asthma Quality of Life Questionnaire (AAQOL). Eur.Respir.J. 2001;17(1):52-8.
2. Ronen GM, Streiner DL, Rosenbaum P. Health-related quality of life in children with epilepsy: development and validation of self-report and parent proxy measures. Epilepsia 2003;44(4):598-612.
3. Griffiths AM, Nicholas D, Smith C, Munk M, Stephens D, Durno C et al. Development of a quality-of-life index for pediatric inflammatory bowel disease: dealing with differences related to age and IBD type. J.Pediatr.Gastroenterol.Nutr. 1999;28(4):S46-S52.
4. Akobeng AK, Suresh-Babu MV, Firth D, Miller V, Mir P, Thomas AG. Quality of life in children with Crohn's disease: a pilot study. J.Pediatr.Gastroenterol. Nutr.1999;28(4):S37-S39.
5. French DJ, Christie MJ, West A. Quality of life in childhood asthma: development of the Childhood Asthma Questionnaires. In: Christie MJ, French DJ, editors. Assessment of quality of life in childhood asthma. Chur, Switzerland: Harwood Academic Publishers; 1994. p. 157-80.
6. Starfield B, Bergner M, Ensminger M, Riley A, Ryan S, Green B et al. Adolescent health status measurement: development of the Child Health and Illness Profile. Pediatrics 1993;91(2):430-5.
7. McEwan MJ, Espie CA, Metcalfe J, Brodie MJ, Wilson MT. Quality of life and psychosocial development in adolescents with epilepsy: a qualitative investigation using focus group methods. Seizure. 2004;13(1):15-31.
8. Weinger K, O'Donnell KA, Ritholz MD. Adolescent views of diabetes-related parent conflict and support: a focus group analysis. J.Adolesc.Health 2001;29(5):330-6.
9. Kennedy C, Kools S, Krueger R. Methodological considerations in children's focus groups. Nurs.Res. 2001;50(3):184-7.
10. Ronen GM, Rosenbaum P, Law M, Streiner DL. Health-related quality of life in childhood disorders: a modified focus group technique to involve children. Qual.Life Res. 2001;10(1):71-9.
11. Bertram S, Kurland M, Lydick E, Locke GR, III, Yawn BP. The patient's perspective of irritable bowel syndrome. J.Fam.Pract. 2001;50(6):521-5.
12. Barlow JH, Shaw KL, Harrison K. Consulting the 'experts': children's and parents' perceptions of psycho-educational interventions in the context of juvenile chronic arthritis. Health Educ.Res. 1999;14(5):597-610.
13. van Es SM, le Coq EM, Brouwer AI, Mesters I, Nagelkerke AF, Colland VT. Adherence-related behavior in adolescents with asthma: results from focus group interviews. J.Asthma 1998;35(8):637-46.
14. McLafferty I. Focus group interviews as a data collecting strategy. J.Adv.Nurs. 2004;48(2):187-94.

15. McEwan MJ, Espie CA, Metcalfe J. A systematic review of the contribution of qualitative research to the study of quality of life in children and adolescents with epilepsy. Seizure. 2004;13(1):3-14.
16. Heary CM, Hennessy E. The use of focus group interviews in pediatric health care research. J.Pediatr.Psychol. 2002;27(1):47-57.
17. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol.Assess 2001;5(4):1-157.
18. Juniper EF, Guyatt GH, Streiner DL, King DR. Clinical impact versus factor analysis for quality of life questionnaire construction. J.Clin.Epidemiol. 1997;50(3):233-8.
19. Twohig PL, Putnam W. Group interviews in primary care research: advancing the state of the art or ritualized research? Fam.Pract. 2002;19(3):278-84.
20. Webb C, Kevern J. Focus groups as a research method: a critique of some aspects of their use in nursing research. J.Adv.Nurs. 2001;33(6):798-805.
21. Groll D, Vanner SJ, Depew WT, DaCosta LR, Simon JB, Groll A et al. The IBS-36: a new quality of life measure for irritable bowel syndrome. Am.J.Gastroenterol. 2002;97(4):962-71.
22. Younossi ZM, Guyatt G, Kiwi M, Boparai N, King D. Development of a disease specific questionnaire to measure health related quality of life in patients with chronic liver disease. Gut 1999;45(2):295-300.
23. Juniper EF, Guyatt GH, Epstein RS, Ferrie PJ, Jaeschke R, Hiller TK. Evaluation of impairment of health related quality of life in asthma: development of a questionnaire for use in clinical trials. Thorax 1992;47(2):76-83.
24. Eiser C, Havermans T, Craft A, Kernahan J. Development of a measure to assess the perceived illness experience after treatment for cancer. Arch.Dis.Child 1995;72(4):302-7.
25. Jhingran P, Osterhaus JT, Miller DW, Lee JT, Kirchdoerfer L. Development and validation of the Migraine-Specific Quality of Life Questionnaire. Headache 1998;38 (4):295-302.
26. Townsend M, Feeny DH, Guyatt GH, Furlong WJ, Seip AE, Dolovich J. Evaluation of the burden of illness for pediatric asthmatic patients and their parents. Ann.Allergy 1991;67(4):403-8.
27. O'Leary CJ, Jones PW. The influence of decisions made by developers on health status questionnaire content. Qual.Life Res. 1998;7(6):545-50.
28. Marx RG, Bombardier C, Hogg-Johnson S, Wright JG. How should importance and severity ratings be combined for item reduction in the development of health status instruments? J.Clin.Epidemiol. 1999;52(3):193-7.
29. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. Int.J.Rehabil.Res. 2002;25(3):197-206.
30. Schmidt S, Bullinger M. Current issues in cross-cultural quality of life instrument development. Arch.Phys.Med.Rehabil. 2003;84(4 Suppl 2):S29-S34.

31. Bullinger M, Von Mackensen S, Fischer K, Khair K, Petersen C, Ravens-Sieberer U et al. Pilot testing of the 'Haemo-QoL' quality of life questionnaire for haemophiliac children in six European countries. Haemophilia. 2002;8 Suppl 2:47-54.
32. The WHOQOL group. The World Health Organization Quality of Life assessment (WHOQOL): position paper from the World Health Organization. Soc.Sci. Med. 1995;41(10):1403-9.
33. Ware JE, Jr., Kemp JP, Buchner DA, Singer AE, Nolop KB, Goss TF. The responsiveness of disease-specific and generic health measures to changes in the severity of asthma among adults. Qual.Life Res. 1998;7(3):235-44.
34. Ravens-Sieberer U, Gosch A, Abel T, Auquier P, Bellach BM, Bruil J et al. Quality of life in children and adolescents: a European public health perspective. Soz.Praventiv med. 2001;46(5):294-302.
35. Herdman M, Rajmil L, Ravens-Sieberer U, Bullinger M, Power M, Alonso J. Expert consensus in the development of a European health-related quality of life measure for children and adolescents: a Delphi study. Acta Paediatr. 2002;91(12):1385-90.
36. EUROHIS: Developing common instruments for health surveys. Amsterdam: IOS Press; 2003.
37. Street H, Sheeran P, Orbell S. Exploring the relationship between different psychosocial determinants of depression: a multidimensional scaling analysis. J.Affect. Disord. 2001;64(1):53-67.
38. Petersen C, Schmidt S, Bullinger M, and the DISABKIDS group. Brief report: Development and pilot testing of a coping questionnaire for children and adolescents with chronic health conditions. J.Pediatr.Psychol. 2004;29(8):635-40.
39. Gill TM, Feinstein AR. A critical appraisal of the quality of quality-of-life measure ments. JAMA 1994;272(8):619-26.
40. Barbour RS. The use of focus groups to define patient needs. J.Pediatr.Gastroenterol. Nutr. 1999;28(4):S19-S22.
41. Petersen C, Schmidt S, Power M, Bullinger M, and the DISABKIDS group. Development and pilot-testing of a health-related quality of life chronic generic module for children and adolescents with chronic health conditions: A European perspective. Qual Life Res. 2005;14(4):1065-77.
42. Aljunid S. Computer analysis of qualitative data: the use of ethnograph. Health Policy Plan. 1996;11(1):107-11.
43. Chapple A, Rogers A. Explicit guidelines for qualitative research: a step in the right direction, a defence of the 'soft' option, or a form of sociological imperialism? Fam. Pract. 1998;15(6):556-61.
44. Weinberger M, Ferguson JA, Westmoreland G, Mamlin LA, Segar DS, Eckert GJ et al. Can raters consistently evaluate the content of focus groups? Soc.Sci.Med. 1998;46(7):929-33.
45. Carey M, Smith MW. Capturing the group effect in focus groups: a special concern in analysis. Qual.Health Res. 1994;4(1):123-7.
46. Swaine-Verdier A, Doward LC, Hagell P, Thorsen H, McKenna SP. Adapting quality of life instruments. Value.Health 2004;7 Suppl 1:S27-S30.

6

The European DISABKIDS project: development of seven condition-specific modules to measure health related quality of life (HRQoL) in children and adolescents

R.M.Baars, C. Atherton, H.M. Koopman, M. Bullinger, M. Power and the DISABKIDS group
Health and Quality of Life Outcomes 2005, 3:70.

> A prime question … is whether the academic psychometric principles, although perhaps elegant statistically, are satisfactory for the clinical goal of indicating what clinicians and patients perceive as quality of life (T.M.Gill 1994).

Abstract

The European DISABKIDS project aims to enhance the health related quality of life (HRQoL) of children and adolescents with chronic medical conditions and their families. A description is given of the development of the seven cross-nationally tested condition-specific modules of the European DISABKIDS HRQoL instrument in a population of children and adolescents. The condition-specific modules are intended for use in conjunction with the DISABKIDS chronic generic module. Focus groups were used to construct the pilot version of the DISABKIDS condition-specific HRQoL modules for asthma, juvenile idiopathic arthritis, atopic dermatitis, cerebral palsy, cystic fibrosis, diabetes and epilepsy. Analyses were conducted on pilot test data in order to construct field test versions of the modules. A series of factor analyses were run, first, to determine potential structures for each condition-specific module, and, secondly, to select a reduced number of items from the pilot test to be included in the field test. Post-field test analyses were conducted to retest the domain structure for the final DISABKIDS condition-specific modules. The DISABKIDS condition-specific modules were tested in a pilot study of 360 respondents, and subsequently in a field test of 1152 respondents in 7 European countries. The final condition-specific modules consist of an 'Impact' domain and an additional domain (e.g. Worry, Stigma, Treatment) with between 10 to 12 items in total. The Cronbach's alpha of the final domains was found to vary from 0.71 to 0.90. The condition-specific modules of the DISABKIDS instrument were developed through a step-by-step process including cognitive interview, clinical expertise, factor analysis, correlations and internal consistency. A cross-national pilot and field test were necessary to collect these data. In general, the internal consistency of the domains was satisfactory to high. In future, the DISABKIDS instrument may serve as a useful tool with which to assess HRQoL in children and adolescents with a chronic condition. The condition-specific modules can be used in conjunction with the DISABKIDS chronic generic module.

Introduction

The last few decades have seen an increase in the amount of constructed health related quality of life (HRQoL) questionnaires for use with children and adolescents [1,2]. Although a number of questionnaires have been used for evaluative studies the questionnaires are only occasionally used in paediatric clinical trials or clinical practice [3-5]. The expectation is that the implementation of HRQoL questionnaires will increase once a number of aspects of HRQoL research are improved.

One area of improvement concerns the need for valid cross-national questionnaires for use in international research [6-8]. Most questionnaires have been developed in one country and are then translated for use in other countries (sequential approach) [9]. This is thought to have its limitations as true compatibility is not necessarily reached [8,10]. A preferred design for the development of cross-national questionnaires is to construct a questionnaire in several countries through a simultaneous approach [8,9]. A questionnaire that was developed in simultaneous collaboration with different countries is the World Health Organization Quality of Life (WHOQOL) questionnaire, but it is only for use in adults [11].

Investigators have also suggested further improvement of HRQoL questionnaires by combining generic and condition-specific modules to offer sufficient detail in the assessment of HRQoL [12]. Generic questionnaires are generally used in HRQoL research and enable comparisons between groups of interest (i.e. different chronic medical conditions). Supplementing a generic module with a condition-specific module is suggested to provide additional information concerning a specific condition and has the potential to identify smaller changes important to research or clinical practice [12-14]. Examples of these are the 'How are you?' (HAY)-asthma [15,16] and the Paediatric Quality of Life Inventory (PedsQL™) [17,18], which both consist of a generic core scale with an additional asthma module.

However, thus far there were no HRQoL questionnaires that were developed in several countries simultaneously and consisted of a chronic generic and condition-specific module for use in children and adolescents with a variety of chronic medical conditions. The European DISABKIDS project aimed to provide in this need. The project was conducted simultaneously in collaboration with seven European countries and developed a series of modules to assess the HRQoL of children and adolescents who suffer from chronic medical conditions [19]. The unique combination consisted of the simultaneous cross-national development, the patient-derived bottom-up procedure, a two modular design and the inclusion of seven chronic conditions. This paper will illustrate the psychometric procedures that have been employed in the development of the condition-specific modules for the European DISABKIDS instrument. Results will be presented and limitations will be discussed. A pilot study was performed to test the basic domain structure and reduce the number of items. A larger field study was conducted to carry out the statistical analyses for the final version of the seven condition-specific DISABKIDS modules. The asthma-specific module will be described in more detail to illustrate the developmental process.

Method

The DISABKIDS group has developed a European HRQoL instrument for children and adolescents with a chronic medical condition and their parents [19]. The project is a collaboration of seven European countries (Austria, France, Germany, Greece, the Netherlands, Sweden and the United Kingdom) and included seven chronic medical conditions: asthma, juvenile idiopathic arthritis (JIA), atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes and epilepsy. The work was closely linked to the

KIDSCREEN project, which is concerned with the development of a generic quality of life (QoL) questionnaire for children of the general population through a similar methodology [20,21]. The instruments devised by these two projects form a three level modular structure (Figure 1).

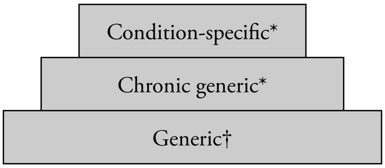


Figure 1: Modular design of the DISABKIDS* and KIDSCREEN† instrument.

The generic module is provided by the KIDSCREEN project and is a QoL questionnaire, suitable for all children, regardless of whether they enjoy complete health or suffer from a chronic medical condition. This generic module creates the possibility of comparing children with a chronic condition to healthy children. The DISABKIDS project has provided the other two modules. One is referred to as the chronic generic module, which is suitable for use with children and adolescents who suffer from any chronic medical condition. It can compare HRQoL across different conditions while taking into account specific areas affected by a chronic condition [22]. The third level consists of a condition-specific module, one for every chronic condition studied in the DISABKIDS project. Each one concerns aspects related to a specific chronic condition and can only compare between data from patients with the same chronic condition. In practice children and adolescents with a chronic medical condition can complete all three modules as each provides different information.

The DISABKIDS project has followed a stepwise methodology of questionnaire construction. Prior to the development of the instrument, an extensive literature review was conducted, and existing HRQoL questionnaires were reviewed in order to obtain an understanding of items in use. Central to the DISABKIDS project was the 'bottom-up' (patient-derived) nature of questionnaire construction, which was accomplished by involving children and adolescents with a chronic medical condition throughout the project. Focus groups and interviews were carried out in order to identify important HRQoL aspects from the perspective of children, adolescents and their parents. The participants were asked a series of semi-structured questions designed to facilitate discussion about their health and related quality of life issues. For example, "What kinds of things keep you healthy?" or "How does your condition affect you at school?". Participants were also asked to make suggestions as to what questions could be included in a HRQoL questionnaire suitable for others who suffer from the same condition as them. In this way the perspective of the child has been incorporated in order to ensure that the content of the questionnaire is directly relevant to the targeted age group [23].

HRQoL statements were selected from the collected qualitative data (focus group and interview transcripts) and merged into a data bank. Collected statements from each chronic condition group (asthma, epilepsy etc.) were then divided among the three modules of the instrument (figure 1). Statements that were considered relevant to all children and adolescents, either healthy or suffering from a chronic condition were entered in the generic module and passed on to the KIDSCREEN project. General statements concerning chronic medical conditions were entered into the chronic generic module. Every disease specific statement was placed in the appropriate condition-specific module. To minimise the number of items, a redundancy scoring, item writing and card sorting procedure was constructed [22]. The card sorting procedure was performed by the DISABKIDS investigators and assisted in the final item selection and provided a preliminary domain structure for each module for use in the pilot study. The selected items were translated to the appropriate languages following general guidelines [24].

The aim of the pilot test was to select a reduced number of items to be included in the field test and to determine a preliminary scale structure within each condition-specific module. At this stage it was considered important to integrate both statistical and subjective data during the item selection process. This included the percentage of 'not applicable' and 'never' responses, a cognitive interview and the clinical judgment of clinicians and investigators. The cognitive interview provided detailed feedback on the relevance, age appropriateness and comprehensibility of the condition-specific items [25-27]. Children and adolescents were asked to rate the difficulty of each item and to rephrase each item in their own words. This feedback was used in conjunction with statistical analyses in order to make informed decisions about the item reduction [22]. The aim of the field test was to re-analyse the final domain structure of each condition-specific module and to calculate the internal consistency of each domain with data from a larger cross-national sample. Items were also examined for distribution of responses, frequency of non-response, ceiling and floor effects.

Children and adolescents between 8 and 16 years of age and their parents were asked to participate in the DISABKIDS pilot and field study, completing the instrument either at the hospital or at home. Data from the children and adolescents were used for the statistical analyses. Condition-specific modules were generally tested in two or more countries; only asthma was tested in all seven countries. Analysis of the condition-specific modules was carried out centrally (in the UK) to ensure that the item selection was done in a consistent way across all seven conditions. The analyses were performed separately for each condition-specific module and were carried out using SPSS Version 11.

Results

Pilot study

The pilot study instrument included the pilot version of the chronic generic module (100 items) and the pilot version of the condition-specific modules (between 26 and 44 items) (Table 1). The applied answer categories were never, seldom, quite often, very often and always, which were scored on a scale from 1 to 5 and an additional 'not applicable' option. The pilot study was conducted between May and August 2002.

Condition-specific modules	**Number of items**	**Number of participants**	**Percentage of total sample (%)**
Asthma	32	132	37
Juvenile idiopathic arthritis	44	54	15
Atopic dermatitis	36	29	8
Cerebral palsy	26	21	6
Cystic fibrosis	38	28	8
Diabetes mellitus	28	59	16
Epilepsy	27	37	10

Table 1. Number of items and participants (n=360) in the pilot study for each condition-specific module.

The sample for the pilot study consisted of 360 participating families. An equal number of boys and girls (48% and 52%) were included, mean age 12.5 (SD 2.55). The asthma group was the largest group of the sample (n = 132). Questionnaire data were only included when more than 60% of the items were completed, resulting in a total of 342 cases for the analyses. This left a few missing values, which were replaced with their series mean to evade losing additional data.

Various sources of data were systematically considered in the selection of items for domains. Some of the data were qualitative in nature, for example the clinical opinion gained from the relevant consultants participating in the project, cognitive interview feedback from the children and adolescents, and the investigator's judgement of the quality of the item. These qualitative aspects were used in conjunction with quantitative results from statistical analyses of the pilot test data (missing values, floor and ceiling effects). Some items were removed solely on the basis of qualitative data when 3 or more qualitative factors were identified as problematic (for example: not understood in the cognitive interview, too many missing values and not sufficiently related to HRQoL).

The structure of the condition-specific modules, as derived from the card sort procedure, was used as a starting point for the identification of domains within the pilot test modules. Item-domain correlations and reliability (Cronbach's alpha) were calculated for these scales. The domain structures resulting from the card sorting method were not generally robust in the statistical analyses. Therefore, principal components analysis with varimax rotation was conducted in order to identify possible new domains. The sample size was quite low for some conditions, and therefore factor analyses were viewed with caution.

An iterative procedure was followed in order to examine possible domain structures. Item groupings, found in the principal components analysis as being similar to those of the original domain structures (from the card sort procedure), were identified. On the basis of a similarity between these two methods, 3-6 items were selected per domain. A scale was then computed and the reliability calculated. If the Cronbach's alpha (α) value was acceptable (above 0.6 to 0.7) and could not be improved by the removal of items, this was acknowledged as a domain [28]. The process was carried out for all feasible domains (typically two or three per condition). The resulting domains were then correlated with all the remaining condition-specific items. An item was added to a domain if it correlated with a domain, it loaded only on one domain, and it generally made sense to include the item in the domain [29]. The reliability of the domain, including the added items, was then re-calculated to ensure a good fit. In some instances items were removed on the basis of low corrected item-total correlations, which ideally should be above 0.4 [28].

If the constructed domains displayed an unsatisfactory (depending on group size and number of items) Cronbach's alpha value (i.e. α below a value of 0.7 to 0.6), the factor analysis was repeated, restricting it to two or three domains. This typically resulted in the grouping of similar items that could be formed into possible new domains (not necessarily those identified in the card sorting procedure). If a domain contained too many items and had a very high alpha value (α over 0.9), item-item correlations were carried out to identify and consequently exclude duplicate items.

When two or three domains had been identified with a total of around 15 items, a final check was run that consisted of the reliability of the domain, the item-domain correlation, and conceptual analysis that included whether or not the scale made sense. The internal consistency of the domains in each condition-specific module was between 0.75 and 0.89 (Table 2). Each domain was given a label that represented the semantic content. Consultants (with knowledge of a specific chronic condition) within the DISABKIDS project were given the opportunity of adding 1 or 2 items to a module on the basis of clinical importance; these items were not added to the domains, but were maintained as single items for separate analyses after the field study.

Condition	**Domain 1**	**n**	**α**	**Domain 2**	**n**	**α**	**Domain 3**	**n**	**α**
Asthma	Impact	8	.83	Worry	5	.86			
Juvenile idiopathic arthritis	Limitation	6	.82	Understanding	6	.75	Frustration	5	.77
Atopic dermatitis	Impact	7	.84	Skin	5	.77	Shame	4	.77
Cerebral palsy	Limitation	5	.84	Frustration	7	.81			
Cystic fibrosis	Impact	6	.77	Treatment	8	.87			
Diabetes mellitus	Impact	5	.84	Food	5	.76	Injections	5	.82
Epilepsy	Fear	8	.89	Social	6	.77			

Table 2. Domains, number of items (n) and the Cronbach's alpha (α) after the pilot analysis.

Example: the asthma pilot study analysis

After the card sorting methodology the asthma module originally consisted of 8 domains (Limitations, Symptoms, Worry, Allergy, Sleep, Medical, Interpersonal and Lack of energy) with a total of 32 items (Table 3). Analysis of the module as described above (including information from the cognitive interviews and clinical judgements) resulted in a 2 domain structure (13 items). The domains were labelled ‘Impact’ and ‘Worry’ due to their semantic content. The mean score on the 'Impact' domain was 3.63 (SD 0.82) and 4.15 (SD 0.89) on the 'Worry' domain. The DISABKIDS asthma consultants added two extra items, not selected through statistical analysis but based on clinical relevance.

Field study

The next step in the DISABKIDS project was the field study, which took place between April and July 2003. The sample for the field study consisted of 1152 participating families. The field study instrument included the chronic generic module (56 items)[22] and the seven condition-specific modules (between 14 and 19 items) (Table 4). An equal number of boys and girls (52% vs. 48%) were included, mean age 12.2 (SD 2.8). The asthma group was the largest in the sample (n = 405). Data from 1094 children and adolescents were used in the analysis, selected on the basis of more than 60% of the items in the module being completed.

Asthma condition-specific items	**Pilot study domains**	**Field study domains**	**Final domains**
Do you have problems sleeping at night because of your cough?	Sleep	Extra item	
Are you bothered by coughing during your sleep?	Sleep		
Are you scared at night because of your asthma?	Sleep	Worry	Worry
Does coughing give you attention from people?	Interpersonal		
Do other children understand that you are sometimes out of breath?	Interpersonal		
Do other kids make fun of your inhaler?	Medication		
Do you hate blowing into a peak flow meter?	Medication		
Do you need medicine to relieve your symptoms before going to bed?	Medication		
Do you worry that others do not know what to do if you have an asthma attack?	Worry	Worry	Worry
Are you worried that you might have an asthma attack?	Worry	Worry	Worry
Do you feel scared that you might have difficulty breathing?	Worry	Worry	Worry
Are you scared that you might have to go to the emergency ward?	Worry	Worry	Worry
Are you bothered that you have to stay indoors because of your allergies?	Worry		
Do you have to ask people not to smoke or to wear perfume?	Allergy		
Do you have to wear special clothes because of your asthma?	Allergy		
Are you bothered by hay fever?	Allergy		
Do you have to be careful about washing yourself due to the eczema?	Allergy		
Are you bothered by feeling sleepy?	Lack of energy		
Do you feel that you get easily exhausted?	Lack of energy	Impact	Impact
Do your parents prevent you from going out as much as your friends because of your asthma?	Limitations		
Does asthma bother you if you want to go out?	Limitations	Impact	Impact
Do you avoid going to people’s houses in case they are not clean enough?	Limitations		
Are you not able to take part in certain sports?	Limitations	Impact	Impact
Do you miss having a pet?	Limitations		
Do you miss cuddly toys?	Limitations		
Does your allergy stop you from doing what you want to do?	Limitations	Extra item	
Do you feel terrible when you are out of breath?	Symptoms	Impact	Impact
Do you feel short of breath when you do sports?	Symptoms	Impact	Impact
Are you bothered by the amount of time you spend wheezing?	Symptoms	Impact	Impact
Are you bothered by the amount of time you spend coughing?	Symptoms	Impact	
Have you been embarrassed about coughing in front of others?	Symptoms		
Do you cough when you do sports?	Symptoms	Impact	

Table 3. Item selection and domain appointment after the asthma pilot and field study.

Condition-specific modules	Number of items	Number of participants	Percentage of total sample (%)
Asthma	15	405	35
Juvenile idiopathic arthritis	19	150	13
Atopic dermatitis	19	65	5
Cerebral palsy	16	43	4
Cystic fibrosis	14	91	8
Diabetes mellitus	15	207	18
Epilepsy	16	191	17

Table 4. Number of items and participants (n=1152) in the field study for each condition-specific module.

At this stage the purpose of the analysis was to replicate the domains found in the pilot test analysis. Principal components analysis was carried out. Components that were found to be similar to the pilot test domains (like the asthma and CF module) were directly checked for reliability. A domain was kept if the alpha value was above 0.7 and could not be improved by the removal or inclusion of items.

All domains were correlated with each of the condition-specific items. An item was added to a domain if it correlated with the domain, it loaded clearly on one domain and it generally made sense to include the item in the domain. Items were removed if they loaded on more than one domain (above 0.4 for each domain) or on the basis of high item-item correlations (above 0.9) [29]. If necessary, items were also removed from a domain on the basis of low corrected item-total correlations and/or a substantial increase in alpha value if removed. The internal consistency of the domains was checked after each step. Each procedure was repeated until the optimal solution was found. In some cases domains were renamed or two domains were merged (for example for the diabetes, JIA, and atopic dermatitis modules). The internal consistency of the domains for each condition-specific module was between 0.71 and 0.90 (Table 5). It became clear that one domain of each condition related to the actual impact of the condition on a child or adolescent's life. These domains were relabelled 'Impact'. Over half of the extra items that were included on the basis of clinical relevance after the pilot study analysis were integrated in the final domains.

Condition	Domain 1	n	α	Domain 2	n	α
Asthma	Impact	6	.83	Worry	5	.84
Juvenile idiopathic arthritis	Impact	9	.87	Understanding	3	.73
Atopic dermatitis	Impact	8	.87	Stigma	4	.71
Cerebral palsy	Impact	10	.82	Communication	2*	.72
Cystic fibrosis	Impact	4	.80	Treatment	6	.85
Diabetes mellitus	Impact	6	.83	Treatment	4	.84
Epilepsy	Impact	5	.90	Social	5	.84

Table 5. Domains, number of items (n) and Cronbach's alpha (α) after the field study analysis.

*With only two items this is the inter-item correlation.

Example: the asthma field study analysis

The domain structure of the asthma pilot test analysis was successfully replicated resulting in a 2 domain structure of 'Impact' and 'Worry', which consist of 6 and 5 items respectively. Four items were removed on the basis of duplication and low item-domain correlations, including the two extra clinical items (Table 3). The cumulative proportion of the variance explained by the first two domains was 53% and the internal consistency (α) was 0.83 and 0.84 (Table 5). The mean score on the 'Impact' domain was 3.61 (SD 0.91) and 4.17 (SD 0.84) on the 'Worry' domain. The asthma-specific module was tested separately for all participating DISABKIDS countries. The reliability in each country was mostly above 0.8 (Table 6).

Asthma	**Impact α**	**p**	**Worry α**	**p**
Austria	.77	30	.80	30
France	.84	36	.77	34
Germany	.91	38	.88	41
Greece	.72	29	.61	38
Netherlands	.81	122	.84	127
Scotland	.86	48	.84	49
Sweden	.85	72	.86	73

Table 6. The Cronbach's alpha (α) and number of participants (p) for the final two asthma-specific domains calculated for each country.

Discussion

This study describes part of the developmental process of the seven DISABKIDS cross-national condition-specific modules (Box 1). The DISABKIDS instrument for children and adolescents is the first to be developed cross-nationally in collaboration with several European countries and to include a chronic generic and condition-specific module.

The DISABKIDS instrument has several advantages. First the construction of the chronic generic and condition-specific modules allows for a comprehensive assessment of HRQoL. The chronic generic module can be used in conjunction with any of the condition-specific modules. Combining these modules gives the clinician and investigator the unique opportunity to compare between countries and between different conditions.

The second advantage is the simultaneous cross-national patient-derived development of the DISABKIDS instrument. Children and adolescents from each DISABKIDS country were included in the developmental process of the instrument. HRQoL statements were collected from the cross-national focus groups and interviews. Investigators from the DISABKIDS centres were involved in the item selection process, assuring that all items where relevant in each country. This was again tested in the cognitive interview in the pilot study. This simultaneous setup in different countries supported the developmental process by taking into account cross-national consensus on important HRQoL issues.

In addition, the construction of the DISABKIDS instrument has been a reflective one, combining subjective and statistical procedures. Item selection and reduction was not carried out solely through the use of statistical methods, but also through the inclusion of qualitative factors, such as the views of children and adolescents (gained from cognitive interview) and clinical judgement. The domain structure that resulted from the pilot test was to a great extent successfully replicated after the field test. The reliability of each domain was satisfactory in each condition-specific module.

However, some limitations should be given consideration. The number of respondents in some condition groups in both the pilot and the field test was relatively small, CP (n=21 and 43) and atopic dermatitis groups (n=29 and 65) in particular (Table 1 and 4). It was therefore not possible to solely use statistical methods to develop these modules. It is important to carry out further data collection and to test the reliability and validity in larger patient groups for these conditions. It will also be necessary to carry out large cross-national studies in the future in order to use modern psychometric methods based on Item Response Theory (IRT), which will permit the testing of differential item functioning across cultures and inform the degree to which cross-national comparisons can be validly made. The use of such IRT-based tests was not possible at this stage of the development of the measure because IRT methods require very large sample sizes.

A second limitation is that the condition-specific modules were not tested in every country. Only asthma was tested in all the participating DISABKIDS countries. The Cronbach's alphas were adequate for each asthma domain in each country. The lower alphas in Greece might not only be due to lower numbers of tested participants but also to the fact that the researched population included mostly exercise-induced asthma, which might result in a different impact on their HRQoL. As the number of participants in the other chronic conditions was generally low the reliability per country will still need to be explored in more detail.

Exploring the data:
- Select data for each condition separately
- Remove children with more than 40% missing on condition-specific module
- Descriptives including mean, SD, missings, skewness, and kurtosis
- Check total domain alpha and each item's corrected item-total correlation
- Check item-item correlations for high or not correlating items

Checking existing domains:
- Compute original scales from card sorting
- Reliability of existing scales

Replicating domain structure:
- Factor analysis, if necessary restricted to four, three or two factors
- Comparing meaningfulness of solutions
- Computing domains, calculating reliability and the item-domain correlation
- Adding any correlated items to domains
- Item-domain correlations

Testing domains:
- Total reliability of the domains
- Checking item-domain correlations (ensure corrected item-total correlation is above 0.4)
- Domain-domain correlation
- Check deleted items against content of chronic generic items

Box 1. Summary of the analysis steps.

Future studies will be necessary to provide more details on the reliability and validity of the DISABKIDS modules, especially in larger groups and in different countries. Evidence also needs to be supplied on the value of the instrument in clinical practice. Further possibilities include testing the chronic generic module for applicability in other chronic medical conditions (e.g. haemophilia, heart disease or obesity).

The developmental steps within the DISABKIDS project have included a combination of qualitative and quantitative methods. The two methods were used in succession in order to complement each other, as has been the case throughout the DISABKIDS project. The qualitative data (cognitive interview and clinical judgement) collected in the pilot study was first used to disregard irrelevant items. This was followed by the psychometric calculations. In some cases the project members found removed items to be clinically relevant. These were therefore added as the two extra items in the field study.

Although the process of item reduction for each of the condition-specific modules was similar and included well know procedures [28,29], it remains difficult to describe the developmental process. As the value of each test depended on the size of the group and the number of items in the domain, and common sense judgements were also included, the taken steps may not always seem transparent. The number of countries included in the study meant that there were more national factors and individual opinions to include. Several processes within the DISABKIDS project (team meetings, group discussions) have influenced decisions. An example was the post-hoc decision to add extra items based on clinical relevance.

Conclusion

The condition-specific modules for the DISABKIDS instrument were developed through a step-by-step process including cognitive interview, clinical expertise, factor analysis, correlations and reliabilities. The seven condition-specific modules consist of an 'Impact' domain and an additional domain with a total of 10 to 12 items (See appendix). The DISABKIDS project has constructed a unique instrument, which was developed cross-nationally, included the patient's perspective and has a chronic generic module, which can be combined with one of the seven condition-specific modules. The expectation is that the instrument will be used in a wide variety of (international) studies of children and adolescents with common disorders of childhood.

Acknowledgements

The DISABKIDS project was supported by the European Commission (QLG5-CT-2000-00716) within the Fifth Framework Program “Quality of Life and Management of Living Resources”. The European Union has granted this project for the development of a modular questionnaire to assess health-related quality of life (HRQoL) in children and adolescents with chronic health conditions.

Appendix

The DISABKIDS condition-specific modules.

Asthma	
Impact	Do you feel that you get easily exhausted? Does asthma bother you if you want to go out? Are you unable to take part in certain sports? Do you feel short of breath when you do sports? Are you bothered by the amount of time you spend wheezing? Do you feel terrible when you are out of breath?
Worry	Are you worried that you might have an asthma attack? Do you worry that others do not know what to do if you have an attack? Do you feel scared that you might have difficulty breathing? Are you scared that you might have to go to the emergency ward? Are you scared at night because of your asthma?
Juvenile idiopathic arthritis	
Impact	Do you feel stiff in the mornings (like an old grandma/granddad)? Do you get exhausted easily? Does arthritis make you feel too exhausted to be with friends? Do you hate being in pain? Does it annoy you that the pain sometimes comes on so suddenly? Does pain stop you from doing what you want? Does it bother you that you can't do all sports/hobbies because of your arthritis? Do you hate being restricted in movement? Does it bother you that you have trouble writing/ drawing?
Understanding	Do others understand that your symptoms may change suddenly? Do your friends understand that you may feel poorly quite suddenly? Do teachers understand that you sometimes can't join in?
Atopic dermatitis	
Impact	Does the itching bother you? Does the appearance of your skin bother you? Does itching bother you during the night? Does your skin condition affect your concentration at school? Does looking at your skin scare you? Does your skin get worse when you are under stress? Does your skin condition affect your free-time (sports, playing)? Do you feel comfortable with the way your skin is?
Stigma	Do you try to hide your skin condition? Are you annoyed by others giving you strange looks? Do you dislike it when your friends see the cream being applied? Do you feel uncomfortable when others look at you?

Cerebral palsy	
Impact	Is it frustrating to be unable to keep up with other children? Do you wish that you could run around like everyone else? Do you wish that you could swim as well as other children? Does it bother you that getting dressed takes a long time? Do people think that you are not as clever as you are? Do you have trouble getting in and out of buildings? Are you able to do most things even though your legs don't move well? Does it upset you that you are unable to walk without help? Do you dislike being washed and dressed by other people? Does it upset you that you need help to use the toilet?
Communication	Can you communicate as well as you'd like? Does it upset you that you can't talk as well as other children?
Cystic fibrosis	
Impact	Do you get exhausted when you do sports? Do you feel tired during the day? Do you get out of breath? Do you need to rest more than others?
Treatment	Does it bother you that you must take your enzymes before every meal? Does it bother you that you have to eat a special diet to keep you healthy? Does it bother you that you have to spend a lot of time having treatment? Are you bothered because you have to do physiotherapy everyday? Have you felt that your treatment takes up too much of your free time? Do you feel bothered that you have to stop playing or doing things for treatment?
Diabetes mellitus	
Impact	Does diabetes stop you from doing the things you want to do? Does diabetes rule your day? Does it bother you that you have to be careful about what you eat? Is it difficult for you to stick to your diet? Do you worry about your blood sugar level? Does it bother you that others can always eat and drink as much as they like?
Treatment	Are you annoyed that you have to carry the testing equipment with you? Are you bothered that you have to plan everything? Do you mind taking insulin? Do you get fed up with measuring your blood sugar levels?
Epilepsy	
Impact	Are you afraid that you might hurt yourself during a seizure? Are you worried that you might have a seizure in public? Are you afraid of having a seizure? Do your seizures make you feel helpless? Are you scared that you could have a seizure at any time?
Social	Does it embarrass you when people take care of you when you have a seizure? Are you worried that people make fun of you when you have a seizure? Are you afraid that you can't remember what happens during a seizure? Are you ashamed of having seizures? Are you worried that other children will see you having a seizure?

References

1. Andelman RB, Zima BT, Rosenblatt AB. Quality of life of children: toward conceptual clarity. In: Maruish ME, editor. The use of psychological testing for treatment planning and outcomes assessment. Mahwah, NJ: Lawrence Erlbaum; 1999. p. 1383-413.
2. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol Assess 2001;5(4):1-157.
3. Clarke SA, Eiser C. The measurement of health-related quality of life (QOL) in paediatric clinical trials: a systematic review. Health Qual.Life Outcomes. 2004;2 (1):66.
4. Baars RM, van der Pal SM, Koopman HM, Wit JM. Clinicians' perspective on quality of life assessment in paediatric clinical practice. Acta Paediatr. 2004;93(10):1356-62.
5. Bender BG. Measurement of quality of life in pediatric asthma clinical trials. Ann. Allergy Asthma Immunol. 1996;77(6):438-45.
6. Quality of life and clinical trials. Lancet 1995;346(8966):1-2.
7. Acquadro C, Jambon B, Ellis D, Marquis P. Language and translation issues. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trials. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 575-85.
8. Skevington SM. Advancing cross-cultural research on quality of life: observations drawn from the WHOQOL development. Qual.Life Res. 2002;11(2):135-44.
9. Schmidt S, Bullinger M. Current issues in cross-cultural quality of life instrument development. Arch.Phys.Med.Rehabil. 2003;84(4 Suppl 2):S29-S34.
10. Swaine-Verdier A, Doward LC, Hagell P, Thorsen H, McKenna SP. Adapting quality of life instruments. Value.Health 2004;7 Suppl 1:S27-S30.
11. The WHOQOL group. The World Health Organization Quality of Life assessment (WHOQOL): position paper from the World Health Organization. Soc.Sci. Med. 1995;41(10):1403-9.
12. Fitzpatrick R, Davey C, Buxton MJ, Jones DR. Evaluating patient-based outcome measures for use in clinical trials. Health Technol.Assess. 1998;2(14):i-74.
13. Ware JE, Jr., Kemp JP, Buchner DA, Singer AE, Nolop KB, Goss TF. The responsiveness of disease-specific and generic health measures to changes in the severity of asthma among adults. Qual.Life Res. 1998;7(3):235-44.
14. Guyatt GH, King DR, Feeny DH, Stubbing D, Goldstein RS. Generic and specific measurement of health-related quality of life in a clinical trial of respiratory rehabilitation. J.Clin.Epidemiol. 1999;52(3):187-92.
15. Bruil J. Development of a quality of life instrument for children with a chronic illness [dissertation]. University of Leiden; 1999.
16. le Coq EM, Colland VT, Boeke AJ, Boeke P, Bezemer DP, van Eijk JT. Reproducibility, construct validity, and responsiveness of the "How Are You?" (HAY), a self-report quality of life questionnaire for children with asthma. J.Asthma 2000;37(1):43-58.

17. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. Med.Care 2001;39(8):800-12.
18. Varni JW, Burwinkle TM, Rapoff MA, Kamps JL, Olson N. The PedsQL in pediatric asthma: reliability and validity of the Pediatric Quality of Life Inventory generic core scales and asthma module. J.Behav.Med. 2004;27(3):297-318.
19. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. Int.J.Rehabil.Res. 2002;25(3):197-206.
20. Herdman M, Rajmil L, Ravens-Sieberer U, Bullinger M, Power M, Alonso J. Expert consensus in the development of a European health-related quality of life measure for children and adolescents: a Delphi study. Acta Paediatr. 2002;91(12):1385-90.
21. Ravens-Sieberer U, Gosch A, Abel T, Auquier P, Bellach BM, Bruil J et al. Quality of life in children and adolescents: a European public health perspective. Soz.Praventiv med. 2001;46(5):294-302.
22. Petersen C, Schmidt S, Power M, Bullinger M, and the DISABKIDS group. Development and pilot-testing of a health-related quality of life chronic generic module for children and adolescents with chronic health conditions: A European perspective. Qual Life Res. 2005;14(4):1065-77.
23. Gill TM, Feinstein AR. A critical appraisal of the quality of quality-of-life measurements. JAMA 1994;272(8):619-26.
24. Guillemin F, Bombardier C, Beaton D. Cross-cultural adaptation of health-related quality of life measures: literature review and proposed guidelines. J.Clin.Epidemiol. 1993;46(12):1417-32.
25. Grant EN, Turner-Roan K, Daugherty SR, Li T, Eckenfels E, Baier C et al. Development of a survey of asthma knowledge, attitudes, and perceptions: the Chicago Community Asthma Survey. Chicago Asthma Surveillance Initiative Project Team. Chest 1999;116(4 Suppl 1):178S-83S.
26. Bullinger M, Von Mackensen S, Fischer K, Khair K, Petersen C, Ravens-Sieberer U et al. Pilot testing of the 'Haemo-QoL' quality of life questionnaire for haemophiliac children in six European countries. Haemophilia. 2002;8 Suppl 2:47-54.
27. Barofsky I. Cognitive aspects of quality of life assessment. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trials. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 107-15.
28. Bullinger M, Power MJ, Aaronson NK, Cella DF, Anderson RT. Creating and evaluating cross-cultural instruments. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trials. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 659-68.
29. Hawthorne G, Richardson J, Osborne R. The Assessment of Quality of Life (AQoL) instrument: a psychometric measure of health-related quality of life. Qual.Life Res. 1999;8(3):209-24.

7

The European DISABKIDS health related quality of life (HRQoL) instrument for childeren and adolescents with a chronic medical condition: psychometric properties of the cross-national asthma sample

R.M. Baars, A.H. Zwinderman, J.E.Chaplin, J.M. Wit, M. Bullinger, H.M. Koopman and the DISABKIDS group
Submitted

A good quality of life can be said to be present when the hopes of an individual are matched and fulfilled by experience (K.C. Calman, 1984).

Abstract

This study was conducted to cross-nationally test the European DISABKIDS health related quality of life (HRQoL) instrument in a population of children and adolescents with asthma. The European DISABKIDS HRQoL instrument was developed through a step-by-step cross-national process. There is a core chronic generic module, with 37 items, covering 6 domains (Independence, Limitation, Emotion, Social inclusion, Social exclusion and Medication). In addition there are seven condition-specific modules, of which one is an asthma module that consists of 11 items and has 2 domains (Impact and Worry). Both DISABKIDS modules were tested in 7 countries within Europe on a total sample of 405 children and adolescents with asthma. The internal consistency for all the domains was between 0.66 and 0.85. Domain test-retest correlations were between 0.71 and 0.82, indicating good retest reproducibility. The correlation of the domains with the validation questionnaires was variable. The domain scores differentiated between asthma severity scores. The domain results differ systematically between countries but this has no significant effect on the validity of the instrument. The DISABKIDS HRQoL instrument is unique in being developed cross-nationally and in a modular structure. The psychometric properties of the DISABKIDS chronic generic and asthma-specific modules are sufficient for HRQoL assessment in children and adolescents with asthma.

Introduction

Asthma is the most common chronic medical condition among children, but the prevalence of asthma varies greatly, with up to a 20-fold difference between some countries. An average of 17 % of the children in Western Europe report wheezing and 13% have had asthma [1].

Children with asthma can experience limitations or impairments in various aspects of their life. Having asthma can lead to restrictions in activities [2-4], emotional problems [5,6], behavioural problems [6,7], adjustment difficulties [7], feelings of depression [5], a fear of being rejected by peers due to being "different" [8], lower perceived well-being [2], anxiety [3,9] and family stress [10,11]. In general asthma can be kept under control through pharmacological therapy and the avoidance of triggers that influence the asthma symptoms [12,13]. However, there are still indications that the health related quality of life (HRQoL) of children with asthma is decreased compared to their healthy peers [2,14]. It is therefore found crucial that, next to the medical treatment of a child with asthma, attention is paid to the child's HRQoL.

HRQoL information can help to assess the impact of a chronic medical condition on the daily life of a child and his or her family [15-17]. It can make clinicians aware of how the child perceives his or her illness. Consequently the increasing importance of HRQoL assessment in paediatric health care and research also makes it a new parameter in evaluating children with asthma.

In the last few decades there has been an increase in the development and testing of various paediatric HRQoL questionnaires [18,19]. Examples of some asthma-specific questionnaires are the Pediatric Asthma Quality of Life Questionnaire (PAQLQ) [20], the Childhood Asthma Questionnaire (CAQ) [21,22], the Life Activities Questionnaire for Childhood Asthma [23] and the Pediatric Quality of Life – Asthma module (PedsQL™ asthma module) [24]. Most of these questionnaires have been developed through a sequential approach, where the questionnaire was developed in one country and it has been translated into other languages [25]. Consequently, these translated questionnaires may need to be adapted due to different cultural or lifestyle aspects in certain countries, e.g. problems riding a bike may not be as relevant in Greece as they are in the Netherlands [26,27]. Developing a questionnaire in several countries through a simultaneous approach would diminish this problem and create the advantage of a cross-national questionnaire [28]. However, there have been only a few attempts to develop a HRQoL questionnaire in several countries simultaneously [25]. The WHOQOL questionnaire is a well-known example but this is a generic questionnaire and is only for use in adults [29].

With the exception of the PedsQL™, multi-language HRQoL paediatric questionnaires are either generic or condition-specific. Having both generic and condition-specific modules has the advantage of collecting information that can be compared with other illness groups and at the same time collecting specific data for a certain condition. Yet, until now, there was no questionnaire that combined a generic module, applicable to living with a chronic medical condition, with a condition-specific module. The DISABKIDS project's aim was to develop a cross-national paediatric HRQoL instrument simultaneously in several countries and at the same time developing a chronic generic and several condition-specific modules.

The aim of this paper is to evaluate the psychometric properties of the cross-nationally developed DISABKIDS HRQoL instrument in a population of children and adolescents with asthma in Europe. The results are a part of the DISABKIDS project conducted to develop and psychometrically test the DISABKIDS HRQoL instrument for several chronic medical conditions.

Material and Methods

The DISABKIDS project

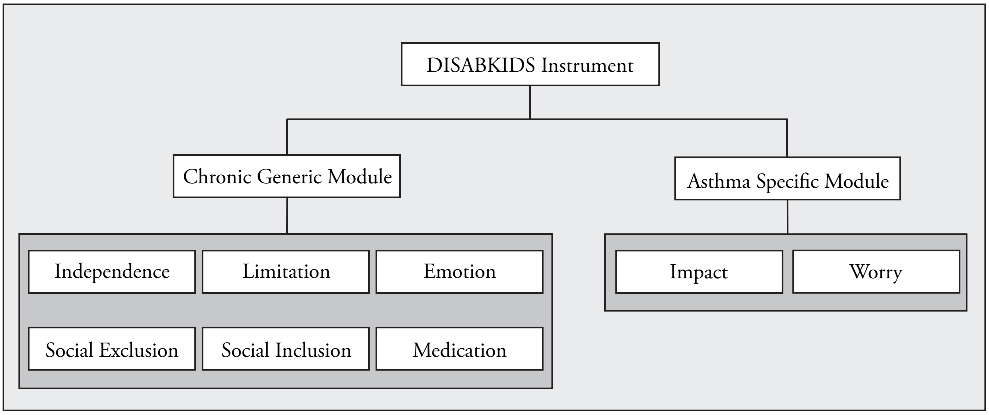
The European DISABKIDS project is a collaboration of eight research institutions in seven European countries (Austria, France, Germany, Greece, the Netherlands, Sweden and the United Kingdom) and aims at cross-nationally developing a European HRQoL instrument for children (aged 8-12) and adolescents (aged 13-16) with a chronic medical condition [30]. Chronic conditions included in the project were asthma, juvenile idiopathic arthritis (JIA), atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes and epilepsy. The final instrument should represent the child and adolescent's view by including aspects that are important to them, be multidimensional, cross-nationally applicable, valid and reliable. Central to the development is the bottom-up construction. This means that the children and adolescents with a chronic medical condition were involved in the development of the instrument by identifying the HRQoL aspects that they found important in their lives. However, what is new about the development of this paediatric HRQoL instrument is not so much the bottom-up construction, as the cross-national step-by-step process and the modular structure.

Instrument development

Focus groups and interviews were conducted with children, adolescents, their parents and health care professionals in all participating DISABKIDS countries to identify relevant HRQoL aspects from their perspective. Collected statements generated through these focus groups formed the basic item pool in which the common working language was English. Item selection was performed through redundancy scoring, item writing and card sorting [31,32]. Applied methods and results have been described elsewhere [33]. These items were then translated to the appropriate languages following established guidelines (forward-backwards translations) [26]. The DISABKIDS modules were tested in a pilot study (n=360). This included a cognitive interview in which the meaning of each national item was described by children and adolescents with the different chronic medical conditions and were internationally compared to assure similar meaning. The final DISABKIDS modules and domains were constructed through psychometric analyses and Rasch modelling using the field study data (n=1152) [33].

The final DISABKIDS instrument

The final European DISABKIDS instrument is a multi-module HRQoL questionnaire for children and adolescents with a chronic medical condition. The instrument, when used for one of the seven medical conditions, consists of two modules (Box 1). The first is a chronic generic module, which is applicable to all children and adolescents with a chronic condition regardless of the specific nature of their disease. This module consists of 37 items, which covers 6 domains (Independence, Limitation, Emotion, Social exclusion, Social inclusion and Medication). The second part consists of a series of condition-specific modules (e.g. asthma, JIA, atopic dermatitis, CP, CF, diabetes and epilepsy). All condition-specific modules consist of an 'Impact' domain and a complementary domain. The asthma-specific module consists of 11 items that form two domains (Impact and Worry).



Box 1. The DISABKIDS instrument: the chronic generic module with 6 domains and in this example the asthma-specific module with its two domains.

Children and adolescents are asked to think about a 4-week time frame and score each item on a 5-point Likert scale (1= never to 5 = always). The mean score of each domain forms a domain score. There is also a parent proxy version that consists of similar questions, but in the third-person tense. While the chronic generic module creates the opportunity to compare between different conditions, the condition-specific module should supply the clinician with more specific disease information [34-36]. Both modules can be used in conjunction with each other.

Validation measures

Integrated in the study were standard HRQoL instruments with a known relationship to HRQoL, including the Dutch DUX-25 (in the Netherlands and Sweden) and the German KINDL (in Austria, Germany and Greece). The DUX is a 25-item HRQoL questionnaire with four domains (Emotional, Home, Social and Physical) [37]. The KINDL is a HRQoL questionnaire with 24 items in 6 domains (Physical well-being, Emotional well-being, Self esteem, Family, Friends and Everyday function) and a 6-item disease module [38,39]. Sociodemographic and clinical items were also included, assessing age, gender, ethnicity, education, missed schooldays and asthma severity.

Asthma severity

Several classifications have been developed for asthma severity in recent years [12,40-43]. In daily practice asthma severity is frequently based on a combination of several parameters, including symptom frequency and severity, use of medication, physical limitations and pulmonary function tests [12,44]. Sometimes these parameters are combined with school or work absences, daily activities and use of health care facilities [5,45,46]. Severity was evaluated in several ways in the DISABKIDS project. Information was collected from the parents, the child and adolescent, and the clinician. There were single items, for children, adolescents and parents, assessing general health ('In general, how would you say your health is?') and disease severity ('How severe was your asthma during the last year?'). Parents were also asked to complete a symptom checklist for asthma severity based on a scale by Rosier (1994) [40]. Clinicians rated asthma severity through a single item ('How would you rate this child's asthma severity?') and a short questionnaire, in which the calculated score was based on questions concerning symptoms, medication and lung function [47].

Asthma field study population

The studied population consisted of children and adolescents with asthma and their parents. Participants were recruited through clinicians from paediatric clinics in all seven participating European countries. Children and adolescents were selected on the basis of: (a) their age being between 8-12 and 13-16 years, (b) diagnosed with asthma by a paediatrician, (c) ability to understand and read the questionnaire in the countries' national language, (d) absence of co-morbidity.

Field Procedure

Between April and July 2003, families were sent an information letter asking them to participate in the DISABKIDS study. The DISABKIDS instrument and additional questionnaires were administered to children and adolescents with asthma by an interviewer on the day of a doctor's appointment. If necessary the questionnaire was taken home to be completed. The parents completed the proxy version of the questionnaires at the same time, which also included the asthma severity rating. In addition questionnaires were posted to families who were not seen at the medical centres. Clinicians were also asked to complete a questionnaire, which included diagnosis, co-morbidity, development and disease severity. In each country the questionnaires were administered in the native language. All participants were asked to complete retest questionnaires at home 2 weeks later and to report if any major events had happened in the meantime and whether this was positive or negative. Where necessary a reminder phone call was made to stimulate the return of the retest. The European commission and each of the Medical Ethics Committees in the participating study centres approved the study. Informed consent was obtained from all participating families.

Statistical analysis

The Statistical Package for Social Sciences (SPSS 10.0; SPSS Inc., Chicago, IL) was used for the data analyses. Each country entered the anonymised data into a database to protect confidentiality and meet data protection requirements. Descriptive statistics were used to describe the range of responses to each question (variability) and the distribution of the domain scores (mean, SD). Further analyses focused on domain scores rather than individual item scores. Domain scores were obtained by adding item scores within domains and were only calculated if at least 70% of the items in the domain were completed. The scores were transformed to a linear scale from 0-100 to make comparisons between the domains possible in which higher scores indicate a better quality of life. The statistical level of significance was set at 0.01 in each analysis. The reliability, reproducibility, convergent validity and discriminant validity of the chronic generic and condition-specific domains were calculated.

The Cronbach's alpha (α) coefficient was used to measure the extent in which items within each domain correlate with each other to form a multi-item domain and how well the items within a domain fitted together as a single construct. An α coefficient of 0.70 or higher is considered acceptable for questionnaire validation, whereas an α of 0.90 or above is considered necessary for individual or clinical decision making [48,49]. The reproducibility was measured through a test-retest procedure. The DISABKIDS domains were correlated to existing HRQoL questionnaires (convergent validity) and parameters of asthma severity (discriminant validity) to assess the validity. The convergent validity was evaluated by calculating the Spearman's correlation coefficient between the DISABKIDS domains and the domains of the KINDL and the DUX-25 questionnaires. The discriminant validity was assessed with the spearman's correlation coefficient to explore the instrument's ability to distinguish levels of disease severity. The factors that were expected to influence the HRQoL were child and parent reported disease severity, last asthma attack, missed schooldays and clinician reported severity. The expectation was that children and adolescents with more severe asthma or missed school days would score lower on the domain scores and have a poorer HRQoL score.

Results

Respondents

Data were obtained from eight medical centres in the seven participating countries; Austria (AU), Germany (DE), Greece (GR), France (FR), the Netherlands (NL), Sweden (SW) and the United Kingdom (UK). The sample consisted of 405 children and adolescents who completed the questionnaires. Their age ranged between 8 and 17 years (mean age 11.4 years, SD=2.47): 66% were children (aged 8-12) and 34% adolescents (aged 13-17). The percentage of boys in the sample was 59%. Three hundred eighty two parents (85% mothers) participated in the study. Just over half of the questionnaires were completed in the clinic (51%), and the remainder were completed at home. Table 1 displays the demographic characteristics of the study sample.

Characteristics	**Total (n=405)**	**AU (n=30)**	**DE (n=42)**	**GR (n=38)**	**FR (n=37)**	**NL (n=133)**	**SW (n=75)**	**UK (n=50)**
%	100	7	10	9	9	33	19	12
Gender								
Male	59	57	55	66	57	54	68	60
Female	41	43	45	34	43	46	32	40
Age								
8-12	66	63	71	74	53	73	55	62
13-16	34	37	29	26	47	27	45	38
Ethnicity								
Born in own country	96	90	93	100	95	97	95	98
Born in other country	4	10	7	.	5	3	5	2
Education								
Primary school	51	42	34	68	34	70	29	50
Sp. primary school*	11	.	.	8	49	1	29	.
Secondary school	35	55	66	3	14	29	42	46
Sp. secondary school*	1	3	.	.	3	.	.	4
Other	2	.	.	21	.	.	.	.
General Health†								
Excellent	12	20	10	18	11	5	24	4
Very good	28	43	19	37	22	16	41	35
Good	40	20	52	32	46	49	29	40
Fair	19	17	19	10	22	29	5	19
Poor	1	.	.	3	.	1	1	2

Table 1. Demographic characteristics of the children and adolescents in the DISABKIDS asthma sample in percentages (n= 405).

* Sp. = special

† Assessed by the child and adolescent

Asthma severity

When asked how severe their asthma had been in the last year (single question), 27% of the children and adolescents rated not severe, 34% a little, 23% average, 12% said quite severe and 4% rated their asthma as bad (not shown in a table). Eleven percent of the children and adolescents reported having an asthma attack in the last week and 62% had missed one of more school days due to asthma in the last year. The parent and clinician asthma severity scores are presented in table 2. The parents' severity score, based on Rosier's (1994) [40] asthma symptom checklist, correlated 0.55 with the child's severity rating (single question) and 0.37 with the clinicians' questionnaire [47]. The correlation between both the clinicians' ratings (single item and short questionnaire) was 0.75, the correlations between the clinician and the child/adolescent or parent severity scores varied between 0.29 and 0.46.

Severity	Parent asthma symptom checklist [40] (n=382)	Clinician single severity question (n=246)	Clinician severity questionnaire [47] (n=255)
Low	43	21	20
Mild	28	42	45
Moderate	20	35	32
Severe	9	2	3

Table 2. Percentile distribution of the parent and clinician severity scores.

Descriptives

The percentage of missing items was low, <2.5% in the chronic generic module and <3.2% in the asthma-specific module. The mean domain scores, which were computed on a linear scale from 0-100, ranged from 65 to 89, in which a higher score represents a higher HRQoL. The percentile distributions show that the domains were slightly skewed, that the floor effects were minimal (% with a domain score of 0) but that there were substantial ceiling effects (% with a domain score of 100), especially for the chronic generic 'Social exclusion' domain. There were no significant differences between the domain scores for questionnaires that were completed in the clinic or at home. The Cronbach's alpha (α) coefficient was determined to assess the internal consistency reliability of the DISABKIDS domains. The α coefficient for the chronic generic domains ranged from 0.66 to 0.85. For the two asthma-specific domains the α was 0.83 and 0.84. The general descriptives of the DISABKIDS domains are shown in table 3.

Test-retest reliability

One hundred and forty-six children and adolescents completed both the test and retest questionnaires. The α coefficient for the retest ranged from 0.70 to 0.89 (n=146). The test-retest reliability was only calculated in the sub-sample that had completed the retest within 30 days (mean 16 days, SD 7) and included only those children and adolescents that had stated that no changes had taken place. A total of 59 children and adolescents fitted these conditions. The Pearson test-retest correlation of the DISABKIDS chronic generic and asthma-specific domains was between 0.71 and 0.83 (all $p < 0.01$). Most domain scores were slightly higher in the retest, but the differences were not significant ($p > 0.01$).

Domains	No of items	Original score (SD)	Transformed score (SD)	Skewness	Floor	Ceiling	α Coefficient (n=405)	Paired samples mean difference test-retest	Pearson correlation test-retest (n=59)
Chronic generic									
Independence	6	4.2 (.68)	79 (17)	-1.17	0.0%	9.4%	.77	.40	.75
Limitation	6	4.0 (.74)	74 (19)	-0.70	0.0%	7.4%	.75	-.89	.71
Emotion	7	4.3 (.75)	82 (19)	-1.19	0.0%	22.1%	.85	-.92	.81
Social exclusion	6	4.6 (.56)	89 (14)	-1.88	0.0%	35.7%	.73	-1.09	.71
Social inclusion	6	4.1 (.68)	78 (17)	-0.87	0.0%	10.1%	.66	-3.18	.78
Medication	6	4.1 (.85)	76 (21)	-0.82	0.3%	16.6%	.77	-1.12	.83
Asthma									
Impact	6	3.6 (.91)	65 (22)	-0.45	0.3%	5.9%	.83	-4.00	.78
Worry	5	4.2 (.84)	79 (21)	-1.15	0.0%	22.7%	.84	1.19	.73

Table 3. Domain descriptives for the DISABKIDS asthma sample aged 8-16 yrs (total n=405). The original (0-5) and transformed (0-100) domain scores, distribution, Cronbach's alpha (α) correlation coefficients (n=405), and the test-retest reliability (mean difference and Pearson correlation) for the children and adolescents where nothing changed and the retest was within 30 days (n=59)

Correlations

The relationship between the domains of the chronic generic and condition-specific module was computed by calculating the Spearman's correlation coefficients. The domain-domain correlations varied between 0.29 and 0.71 (Table 4). The highest correlation between the domains was seen between the asthma 'Impact' domain and the chronic generic 'Limitation' domain.

Domains	Ind.	Lim.	Emo.	Excl.	Incl.	Med.	Imp.
Chronic generic							
Independence	*						
Limitations	.58	*					
Emotion	.59	.68	*				
Social exclusion	.51	.60	.59	*			
Social inclusion	.50	.51	.47	.44	*		
Medication	.31	.41	.51	.34	.33	*	
Asthma							
Impact	.48	.71	.55	.54	.31	.41	*
Worry	.44	.56	.56	.45	.29	.39	.55

Table 4. Spearman's correlation coefficients of domains in the DISABKIDS modules for the asthma sample (n=405).

NB: all significant, p <0.01

The cross-sectional correlations between the chronic generic DISABKIDS domains and the domains of the Dutch DUX-25 ranged from 0.24 to 0.52. The correlations with the asthma-specific domains were between 0.15 and 0.46. Similar correlations were found with the German KINDL (Table 5).

Discriminant validity

Scores on the DISABKIDS domains were examined within the asthma severity subgroups based on parent, child/adolescent and clinician scores, missed school days and last asthma attack (Table 6). The Spearman's correlations for the parent and child/adolescent severity scores were between 0.23 and 0.50. The correlations with the clinician severity measures were between 0.09 and 0.18. The correlations were generally the highest for both the 'Limitation' and asthma 'Impact' domains.

Domains	DUX-25					KINDL							
	DUX Total scale	Emotion	Home	Social	Physical	KINDL Total scale	Physical well being	Emotional well being	Self esteem	Family	Friends	Everyday function	Disease module
Chronic generic													
Independence	.52	.46	.41	.42	.52	.48	.23*	.37	.41	.39	.44	.28	.46
Limitation	.47	.40	.34	.38	.49	.41	.38	.34	.31	.18*	.40	.20*	.50
Emotion	.35	.29	.24	.28	.36	.54	.37	.49	.48	.29	.44	.34	.61
Social exclusion	.40	.36	.30	.33	.36	.42	.27	.28	.33	.13*	.49	.23*	.44
Social inclusion	.46	.40	.36	.44	.39	.27	.18*	.27	.28	.17*	.38	-.02*	.29
Medication	.40	.34	.29	.27	.44	.25*	.15*	.40	.13*	.09*	.29	.18*	.21*
Asthma													
Impact	.38	.32	.29	.24	.46	.45	.52	.35	.38	.09*	.41	.24*	.54
Worry	.23	.25	.15*	.16*	.24	.37	.28	.24*	.35	.20*	.43	.30	.43

Table 5. Convergent validity: Spearman's correlation coefficients (r values) for the DISABKIDS domains compared to the DUX-25 total score and each domain score (n=207) and the KINDL total score, domain scores and disease module (n=110).

NB: all significant, p <0.01 with the exception of *

Domain	Parent severity	Child severity	Clinician severity	Missed school days	Last asthma attack
Chronic Generic					
Independence	.38	.34	.17	.30	.22
Limitation	.48	.47	.16	.42	.36
Emotion	.37	.39	.10*	.38	.25
Social exclusion	.29	.35	.12*	.31	.22
Social inclusion	.27	.28	.09*	.25	.22
Medication	.23	.23	.09*	.16	.22
Asthma					
Impact	.43	.50	.18	.40	.32
Worry	.32	.33	.12*	.43	.31

Table 6. Spearman's correlation coefficients were used to compare the DISABKIDS domains to the parent asthma symptom checklist [40], the child severity score (single question), the clinician severity questionnaire [47], the number of missed school days and the last asthma attack.
NB: all significant, p <0.01 with the exception of *

The relationship between the domain scores and the asthma severity score (based on the parents rating) is illustrated in figure 1. The DISABKIDS domain scores were significantly higher (better quality of life) in children and adolescents with low asthma severity than in those with severe asthma. Similar differences were observed for the child- and clinician severity scores (data not shown). There was also a relation between the HRQoL score and missed school days and last asthma attack (Table 6). Children and adolescents with more missed school days or with a recent asthma attack had significantly lower HRQoL scores.

Cross-national comparison

Univariate analysis of variance showed that the domain scores were not only dependent on the asthma severity but were also influenced by country. Relatively more severe asthmatic patients were included in the UK sample, whereas relatively fewer severe patients were included in the Swedish sample. Corrected for differences in asthma severity, the average domain scores remained significantly different between the countries (p<0.001). The linear association between asthma severity and the DISABKIDS domain scores however, did not differ significantly between participating countries (p>0.11). Thus, the relation between the domain scores and the asthma severity remains similar in all the countries.

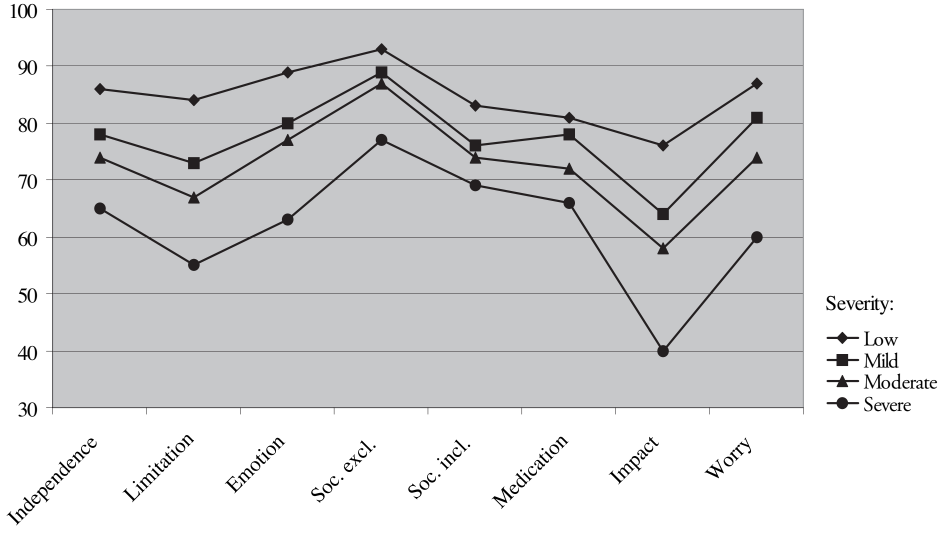


Figure 1. The DISABKIDS domains scores (0-100) of children and adolescents with asthma. Severity is based on the Rosier's (1994) [40] asthma symptom checklist completed by the parents.

Age and gender groups
To establish age and gender differences in the DISABKIDS instrument, independent sample t-tests were performed. In each analysis, a DISABKIDS domain was the dependent variable, while age group or gender were the independent variables. In general the HRQoL domain scores were similar in both gender and age groups. However, some differences were identified: girls scored significantly lower on the 'Limitation' and the asthma 'Impact' domain, adolescents (aged 13-16) had significantly lower scores on the asthma 'Impact' domain and children (aged 8-12) scored significantly lower on the 'Social inclusion' domain (data not shown).

Discussion

The DISABKIDS instrument was developed simultaneously in seven European countries and consists of a chronic generic and condition-specific module, which include HRQoL aspects that were identified through a patient-derived method. We have described the psychometric performance of the DISABKIDS chronic generic and asthma-specific module in a cross-national population of children and adolescents with asthma. The chronic generic module can provide information on the overall impact of a chronic medical condition on a child or adolescent's life and allows comparison across chronic conditions. The asthma-specific module can supplement this with information on specific asthma symptoms, which may be more closely related to the treatment regime [18,35].

The internal consistency of the domains was sufficient for the total asthma population, with the Cronbach's α ranging from 0.66 to 0.85 in the first test (Table 3) and between

0.70 and 0.89 in the retest. However, higher levels of reliability (Cronbach's alpha ≥ 0.9) are necessary for the DISABKIDS instrument to be psychometrically acceptable as an individual screening tool [48,49]. Further studies are being prepared to investigate the instrument's potential as an individual screening tool.

Within the population of children and adolescents that had completed the questionnaires within a month and had unchanged circumstances the measure generally reproduced similar results. The test-retest correlation was above 0.70 for all domains. This analysis supports the basic reliability of the instrument but needs to be taken cautiously. A selection bias might have taken place in the retest and only 59 questionnaires were completed within 30 days.

The inter-domain correlations suggest an overlap between the domain constructs. Correlations between some domains are to be anticipated (Limitation and Impact) while for some domains we expected a lower correlation (Social and Medication). This overlap can be explained when HRQoL aspects are closely intertwined in the lives of children and adolescents.

In the DISABKIDS project the face and content validity was achieved by the use of a bottom-up patient-derived construction. The children and adolescents further added to the item generation through their judgement of clarity and comprehension of items in the cognitive interviews [33,50-52]. The DISABKIDS instrument covers aspects of HRQoL as indicated by the patients as being important. However, coping and health care needs have not been included. New questionnaires including these aspects have been developed as separate entities [53].

Concurrent validity was evaluated by correlating the DISABKIDS domains with validated HRQoL questionnaires. The domains in the DISABKIDS chronic generic and asthma-specific modules displayed variable correlations with the DUX-25 and the KINDL domains. The scores indicate that the DISABKIDS domains correlate with some domains from the DUX-25 and the KINDL but that they also offer a different perspective through other domains (Medication, Worry). Due to the simultaneous testing of several chronic conditions the choice was made to include only generic questionnaires (KINDL and DUX-25) for the concurrent validity, thus missing the possibility to compare the asthma-specific module to existing asthma measures.

The construct validity was tested by examining the relationship between the domain scores to other measures at a single point in time. The DISABKIDS domains were sensitive to different ratings of asthma severity (parent and child/adolescent judgements of severity, missed schooldays and last asthma attack). The correlations between the severity scores and the DISABKIDS domains relating to physical aspects (Limitation and Impact) were the most apparent (Table 6). The correlations between severity and HRQoL were the highest for the parent and child ratings. The instrument is therefore sensitive to severity as judged by the parent and child or adolescent, which may be useful in clinical practice. In contrast, the correlations with the clinician's severity scores were distinctly lower. This again demonstrates that the child or adolescent's HRQoL is not directly related to clinician's disease severity rating or HRQoL judgement [54-57].

The cross-national focus and modular structure has been the specific approach of the DISABKIDS project. Cross-nationally developing a HRQoL questionnaire can limit the inclusion of national and socio-economic differences between countries in the measurement of health effects. A questionnaire applicable to countries across Europe can be of importance to cross-national research trials or individual HRQoL assessment [28]. We should however be aware of some disadvantages of this approach. The focus group and cognitive interview phase in the pilot test were used to collect information on face validity by asking children and adolescents what was important to them. However, the cross-national developmental process has caused some items (concerning pets, riding a bike to school, going to the beach or mountains) to be disregarded due to cross-national differences, as they were not found to be applicable in all countries.

We also need to consider some specific restrictions in this study. Firstly, there is a possible selection bias within the group that participated. Participants with a higher HRQoL and acceptance of their condition might be the ones to participate. The demonstrated ceiling affect may be related to this bias (Table 3). However, it is reassuring that the severity distribution was similar to the results reported by Rosier (1994) [40], who developed the symptom checklist, which suggests that we assessed a commonly found range of asthma patients. Secondly, the severity and domain scores differed between the countries ($p<0.001$). Fortunately, we could conclude that after correcting for the asthma severity, the linear association between asthma severity and the DISABKIDS domains remained the same and thus had no effect on the validity of the instrument. In follow-up research the psychometric properties should be assessed in sufficiently large groups for each country separately. Finally, there is no gold standard for HRQoL. We used several criterion variables as self scored severity of asthma and existing generic HRQoL questionnaires (Table 5 and 6). Future studies should provide more data on other criterion variables such as medication, lung function or asthma specific HRQoL and on responsiveness to clinical changes. The aim is to collect longitudinal data, setup intervention studies and test the applicability of the DISABKIDS instrument in clinical practice in ongoing studies.

Conclusion

Overall the DISABKIDS instrument displays a sufficient degree of reliability and validity. The domain scores correlate with measures of severity and existing HRQoL questionnaires. The DISABKIDS instrument is available as paper-pencil and computer version, is simple to administer and takes around 15 minutes to complete. The instrument has the advantage of a chronic generic and condition-specific module, is multilingual and has been tested cross-nationally. On the whole there is ample support for the use of the DISABKIDS instrument as a measure of HRQoL in a child or adolescent with asthma. In the future the instrument may prove to be relevant for clinical trials and individual assessment in clinical practice.

Acknowledgement

The DISABKIDS group would like to thank all the children, adolescents and parents for participating in the study. We would also like to thank all the clinicians and staff members for their participation and practical support in this extensive study. The DISABKIDS project was supported by the European Commission (QLG5-CT-2000-00716) within the Fifth Framework Program "Quality of Life and Management of Living Resources".

Appendix

Chronic generic module

1. Are you confident about your future?
2. Do you enjoy your life?
3. Are you able to do everything you want to do even though you have your condition?
4. Do you feel like everyone else even though you have your condition?
5. Are you free to lead the life you want even though you have your condition?
6. Are you able to do things without your parents?
7. Are you able to run and move as you like?
8. Do you feel tired because of your condition?
9. Is your life ruled by your condition?
10. Does it bother you that you have to explain to others what you can and can't do?
11. Is it difficult to sleep because of your condition?
12. Does your condition bother you when you play or do things?
13. Does your condition make you feel bad about yourself?
14. Are you unhappy because of your condition?
15. Do you worry about your condition?
16. Does your condition make you angry?
17. Do you have fears about the future because of your condition?
18. Does your condition get you down?
19. Does it bother you that your life has to be planned?
20. Do you feel lonely because of your condition?
21. Do your teachers behave differently towards you than towards others?
22. Do you have problems concentrating at school because of your condition?
23. Do you feel that others have something against you?
24. Do you think that others stare at you?
25. Do you feel different from other children/adolescents?
26. Do other kids/adolescents understand your condition?
27. Do you go out with your friends?
28. Are you able to play or do things with other children/adolescents (like sports)?
29. Do you think that you can do most things as well as other children/adolescents?
30. Do your friends enjoy being with you?
31. Do you find it easy to talk about your condition to other people?
32. Does having to get help with medication from others bother you?
33. Is it annoying for you to have to remember your medication?
34. Are you worried about your medication?
35. Does taking medication bother you?
36. Do you hate taking your medicine?
37. Does taking medication disrupt everyday life?

Answer category: Never – Seldom - Quite often - Very often – Always

Chronic generic domains
Independence: 1, 2, 3, 4, 5, 6
Limitation: 7, 8, 9, 10, 11, 12
Emotion: 13, 14, 15, 16, 17, 18, 19
Social exclusion: 20, 21, 22, 23, 24, 25
Social inclusion: 26, 27, 28, 29, 30, 31
Medication: 32, 33, 34, 35, 36, 37.

Asthma-specific module
1. Do you feel that you get easily exhausted?
2. Does asthma bother you if you want to go out?
3. Are you unable to take part in certain sports?
4. Do you feel short of breath when you do sports?
5. Are you bothered by the amount of time you spend wheezing?
6. Do you feel terrible when you are out of breath?
7. Are you worried that you might have an asthma attack?
8. Do you worry that others do not know what to do if you have an attack?
9. Do you feel scared that you might have difficulty breathing?
10. Are you scared that you might have to go to the emergency ward?
11. Are you scared at night because of your asthma?

Answer category: Never – Seldom - Quite often - Very often – Always

Asthma-specific domains
Impact: 1, 2, 3, 4, 5, 6
Worry: 7, 8, 9, 10, 11

References

1. The International Study of Asthma and Allergies in Childhood (ISAAC) Steering Committee. Worldwide variations in the prevalence of asthma symptoms: the International Study of Asthma and Allergies in Childhood (ISAAC). Eur.Respir.J. 1998;12(2):315-35.
2. Forrest CB, Starfield B, Riley AW, Kang M. The impact of asthma on the health status of adolescents. Pediatrics 1997;99(2):E1.
3. Townsend M, Feeny DH, Guyatt GH, Furlong WJ, Seip AE, Dolovich J. Evaluation of the burden of illness for pediatric asthmatic patients and their parents. Ann. Allergy 1991;67(4):403-8.
4. French DJ, Christie MJ, West A. Quality of life in childhood asthma: development of the Childhood Asthma Questionnaires. In: Christie MJ, French DJ, editors. Assessment of Quality of Life in Childhood Asthma. Chur, Switzerland: Harwood Academic Publishers; 1994. p. 157-80.
5. Okelo SO, Wu AW, Krishnan JA, Rand CS, Skinner EA, Diette GB. Emotional quality-of-life and outcomes in adolescents with asthma. J.Pediatr. 2004;145(4):523-9.
6. Klinnert MD, McQuaid EL, McCormick D, Adinoff AD, Bryant NE. A multimethod assessment of behavioral and emotional adjustment in children with asthma. J.Pediatr. Psychol. 2000;25(1):35-46.
7. McQuaid EL, Kopel SJ, Nassau JH. Behavioral adjustment in children with asthma: a meta-analysis. J.Dev.Behav.Pediatr. 2001;22(6):430-9.
8. French DJ, Carroll A, Christie MJ. Health-related quality of life in Australian children with asthma: lessons for the cross-cultural use of quality of life instruments. Qual.Life Res. 1998;7(5):409-19.
9. Cohen R, Franco K, Motlow F, Reznik M, Ozuah PO. Perceptions and attitudes of adolescents with asthma. J.Asthma 2003;40(2):207-11.
10. Gustafsson PA, Bjorksten B, Kjellman NI. Family dysfunction in asthma: a prospective study of illness development. J.Pediatr. 1994;125(3):493-8.
11. Schulz RM, Dye J, Jolicoeur L, Cafferty T, Watson J. Quality-of-life factors for parents of children with asthma. J.Asthma 1994;31(3):209-19.
12. National Institutes of Health. Guidelines for the diagnosis and management of asthma. National Heart Lung and Blood Institute, National Asthma Education and Prevention Program. publication no. 97-4051. 1997. Bethesda MD, US Department of Health and Human Services, National Institutes of Health.
13. Ross MH, Mjaanes CM, Lemanske R. Asthma. In: Rudolph CD, Rudolph AM, editors. Rudolph's Pediatrics. 21st ed. New York: McGraw-Hill; 2003. p. 1950-63.
14. Hallstrand TS, Curtis JR, Aitken ML, Sullivan SD. Quality of life in adolescents with mild asthma. Pediatr.Pulmonol. 2003;36(6):536-43.
15. Eiser C, Morse R. The measurement of quality of life in children: past and future perspectives. J.Dev.Behav.Pediatr. 2001;22(4):248-56.

16. Rutishauser C, Sawyer SM, Bowes G. Quality-of-life assessment in children and adolescents with asthma. Eur.Respir.J. 1998;12(2):486-94.
17. Juniper EF. How important is quality of life in pediatric asthma? Pediatr.Pulmonol. Suppl 1997;15:17-21.
18. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol Assess 2001;5(4):1-157.
19. Andelman RB, Zima BT, Rosenblatt AB. Quality of Life of Children: Toward Conceptual Clarity. In: Maruish ME, editor. The use of psychological testing for treatment planning and outcomes assessment. Mahwah, NJ: Lawrence Erlbaum; 1999. p. 1383-413.
20. Juniper EF, Guyatt GH, Feeny DH, Ferrie PJ, Griffith LE, Townsend M. Measuring quality of life in children with asthma. Qual.Life Res. 1996;5(1):35-46.
21. Christie MJ, French D, Sowden A, West A. Development of child-centered disease-specific questionnaires for living with asthma. Psychosom.Med. 1993;55(6):541-8.
22. French DJ, Christie MJ, Sowden AJ. The reproducibility of the Childhood Asthma Questionnaires: measures of quality of life for children with asthma aged 4-16 years. Qual.Life Res. 1994;3(3):215-24.
23. Creer TL, Wigal JK, Kotses H, Hatala JC, McConnaughy K, Winder JA. A life activities questionnaire for childhood asthma. J.Asthma 1993;30(6):467-73.
24. Varni JW, Burwinkle TM, Rapoff MA, Kamps JL, Olson N. The PedsQL in pediatric asthma: reliability and validity of the Pediatric Quality of Life Inventory generic core scales and asthma module. J.Behav.Med. 2004;27(3):297-318.
25. Schmidt S, Bullinger M. Current issues in cross-cultural quality of life instrument development. Arch.Phys.Med.Rehabil. 2003;84(4 Suppl 2):S29-S34.
26. Guillemin F, Bombardier C, Beaton D. Cross-cultural adaptation of health-related quality of life measures: literature review and proposed guidelines. J.Clin.Epidemiol. 1993;46(12):1417-32.
27. Wagner AK, Gandek B, Aaronson NK, Acquadro C, Alonso J, Apolone G et al. Cross-cultural comparisons of the content of SF-36 translations across 10 countries: results from the IQOLA Project. International Quality of Life Assessment. J.Clin. Epidemiol. 1998;51(11):925-32.
28. Skevington SM. Advancing cross-cultural research on quality of life: observations drawn from the WHOQOL development. Qual.Life Res. 2002;11(2):135-44.
29. The WHOQOL group. The World Health Organization Quality of Life assessment (WHOQOL): position paper from the World Health Organization. Soc.Sci.Med. 1995;41(10):1403-9.
30. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. Int.J.Rehabil.Res. 2002;25(3):197-206.
31. Starfield B, Bergner M, Ensminger M, Riley A, Ryan S, Green B et al. Adolescent health status measurement: development of the Child Health and Illness Profile. Pediatrics 1993;91(2):430-5.

32. Street H, Sheeran P, Orbell S. Exploring the relationship between different psychosocial determinants of depression: a multidimensional scaling analysis. J.Affect. Disord. 2001;64(1):53-67.
33. Petersen C, Schmidt S, Power M, Bullinger M, and the DISABKIDS group. Development and pilot-testing of a health-related quality of life chronic generic module for children and adolescents with chronic health conditions: a European perspective. Qual Life Res. 2005;14(4):1065-77.
34. Ware JE, Jr., Kemp JP, Buchner DA, Singer AE, Nolop KB, Goss TF. The responsiveness of disease-specific and generic health measures to changes in the severity of asthma among adults. Qual.Life Res. 1998;7(3):235-44.
35. Fitzpatrick R, Davey C, Buxton MJ, Jones DR. Evaluating patient-based outcome measures for use in clinical trials. Health Technol.Assess. 1998;2(14):i-74.
36. Guyatt GH, King DR, Feeny DH, Stubbing D, Goldstein RS. Generic and specific measurement of health-related quality of life in a clinical trial of respiratory rehabilitation. J.Clin.Epidemiol. 1999;52(3):187-92.
37. Koopman HM, Baars RM, Segaar RW. The use of computer-aided health-related quality-of-life questionnaires for children with a chronic disease and their parents. 2002 May 17; Oxford: Hughes associates; 2003.
38. Ravens-Sieberer U, Bullinger M. Assessing health-related quality of life in chronically ill children with the German KINDL: first psychometric and content analytical results. Qual.Life Res. 1998;7(5):399-407.
39. Rajmil L, Herdman M, Fernandez de Sanmamed MJ, Detmar S, Bruil J, Ravens-Sieberer U et al. Generic health-related quality of life instruments in children and adolescents: a qualitative analysis of content. J.Adolesc.Health 2004;34(1):37-45.
40. Rosier MJ, Bishop J, Nolan T, Robertson CF, Carlin JB, Phelan PD. Measurement of functional severity of asthma in children. Am.J.Respir.Crit Care Med. 1994;149 (6):1434-41.
41. Redier H, Daures JP, Michel C, Proudhon H, Vervloet D, Charpin D et al. Assessment of the severity of asthma by an expert system. Description and evaluation. Am.J.Respir.Crit Care Med. 1995;151(2 Pt 1):345-52.
42. Gautier V, Redier H, Pujol JL, Bousquet J, Proudhon H, Michel C et al. Comparison of an expert system with other clinical scores for the evaluation of severity of asthma. Eur.Respir.J. 1996;9(1):58-64.
43. Fritz G, Spirito A, Yeung A, Klein R, Freedman E. A pictorial visual analog scale for rating severity of childhood asthma episodes. J.Asthma 1994;31(6):473-8.
44. Williams SG, Schmidt DK, Redd SC, Storms W. Key clinical activities for quality asthma care. Recommendations of the National Asthma Education and Prevention Program. MMWR Recomm.Rep. 2003;52(RR-6):1-8.
45. Osborne ML, Vollmer WM, Pedula KL, Wilkins J, Buist AS, O'Hollaren M. Lack of correlation of symptoms with specialist-assessed long-term asthma severity. Chest 1999;115(1):85-91.

46. Sennhauser FH, Braun-Fahrlander C, Wildhaber JH. The burden of asthma in children: a European perspective. Paediatr.Respir.Rev. 2005;6(1):2-7.
47. Hargreave FE, Dolovich J, Newhouse MT. The assessment and treatment of asthma: a conference report. J.Allergy Clin.Immunol. 1990;85(6):1098-111.
48. Nunnally JC, Bernstein IH. Psychometric theory. 3rded. New York: McGraw-Hill; 1994.
49. Scientific Advisory Committee of the Medical Outcomes Trust. Assessing health status and quality-of-life instruments: attributes and review criteria. Qual.Life Res. 2002;11(3):193-205.
50. Grant EN, Turner-Roan K, Daugherty SR, Li T, Eckenfels E, Baier C et al. Development of a survey of asthma knowledge, attitudes, and perceptions: the Chicago Community Asthma Survey. Chicago Asthma Surveillance Initiative Project Team. Chest 1999;116(4 Suppl 1):178S-83S.
51. Bullinger M, Von Mackensen S, Fischer K, Khair K, Petersen C, Ravens-Sieberer U et al. Pilot testing of the 'Haemo-QoL' quality of life questionnaire for haemophiliac children in six European countries. Haemophilia. 2002;8 Suppl 2:47-54.
52. Barofsky I. Cognitive aspects of quality of life assessment. In: Spilker B, editor. Quality of life and pharmacoeconomics in clinical trials. Second Edition ed. Philadelphia: Lippincott-Raven; 1996. p. 107-15.
53. Petersen C, Schmidt S, Bullinger M, and the DISABKIDS group. Brief report: Development and pilot testing of a coping questionnaire for children and adolescents with chronic health conditions. J.Pediatr.Psychol. 2004;29(8):635-40.
54. Williams J, Williams K. Asthma-specific quality of life questionnaires in children: are they useful and feasible in routine clinical practice? Pediatr.Pulmonol. 2003;35(2):114-8.
55. Loonen HJ, Derkx BH, Griffiths AM. Pediatricians overestimate importance of physical symptoms upon children's health concerns. Med.Care 2002;40(10):996-1001.
56. Janse AJ, Gemke RJ, Uiterwaal CS, van dT, I, Kimpen JL, Sinnema G. Quality of life: patients and doctors don't always agree: a meta-analysis. J.Clin.Epidemiol. 2004;57(7):653-61.
57. Yawn BP, Fryer GE, Lanier D. Asthma severity: the patient's perspective. J.Asthma 2004;41(6):623-30.

8

Implementation of the DISABKIDS instrument: general discussion

> Thus, QoL, and its measurement, can seem nebulous or unscientific compared with traditional endpoints. However, the more elusive and subjective outcomes may, in the end, be more important (C. Eiser and R. Morse, 2001).

DISABKIDS's past

The aim of the DISABKIDS project was to develop, test and implement a new European health related quality of life (HRQoL) instrument for children and adolescents between the age of 4-16 with a chronic medical condition, and their parents [1]. The DISABKIDS instrument is the first paediatric measure that was developed cross-nationally in Europe, applied a patient-derived method and includes a chronic generic module and seven condition-specific modules.

The DISABKIDS project consisted of predefined work packages (WP) (Chapter 1). The literature review (WP 1) in Medline (1985-2000) identified 8233 abstracts concerning HRQoL assessment in children and adolescents with a chronic medical condition. Several HRQoL questionnaires were reviewed, and published HRQoL domains were considered for the DISABKIDS domain structure. A total of 154 children and adolescents participated in the focus groups or interviews, 142 family members and 26 health care professionals participated in either focus groups or interviews (WP 2). A total of 3515 statements were collected from the focus group transcripts. The item development steps led to a chronic generic module with 100 items and seven condition-specific modules with 26 to 44 items which made up the pilot study instrument (WP 3). The items were translated through a forward –backward –forward translation in each country (WP 4). A total of 360 children and adolescents with a chronic medical condition and 345 parents in seven European countries participated in the pilot study (WP 5). The analysis of the pilot study resulted in a field study instrument with 57 items in the chronic generic module and between 14 and 19 in each condition-specific module (WP 6). A total of 1152 families (including 405 children and adolescents with asthma), spread over 7 European countries, participated in the field study (WP 7). After the analysis of the field study data the final DISABKIDS instrument consisted of a 37-item chronic generic module with 6 domains and seven 10 to 12-item condition-specific modules consisting of 2 domains (WP 8). The implementation of the DISABKIDS instrument is still ongoing in several countries for the paper-pencil and computer versions (WP 9).

This thesis aimed to describe and discuss some of the steps taken within the European DISABKIDS project with a specific emphasis on the results obtained for asthma.

DISABKIDS at present

The two level modular DISABKIDS instrument is now available for children and adolescents between the age of 8 and 16 years with asthma, juvenile idiopathic arthritis (JIA), atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes and epilepsy

(Chapter 6). A short 6-item smiley module is available for the 4-7 year old children with any chronic medical condition. The instrument has been psychometrically tested in Austria, France, Germany, Greece, the Netherlands, Sweden and the United Kingdom, and is available in each of these languages in a paper-pencil and computer version. The instrument is simple to administer, the modules are relatively short and it takes about 15 minutes to complete.

Advantages
The DISABKIDS instrument has several advantages above other instruments (Box 1). The main advantages are the modular build-up, the multiple language versions and the cross-national validation.

- Two modules
- Several languages
- Applicable cross-nationally
- Several chronic conditions
- Short and easy to use
- Paper and computer version
- Wide age range
- Proxy version

Box 1. Advantages of the DISABKIDS instrument.

A unique combination is created when combining the generic module from the KIDSCREEN project with the DISABKIDS chronic generic and condition-specific modules (Chapter 1). The combination of the generic, chronic generic and condition-specific modules allows for a comprehensive assessment of HRQoL. The generic module assesses the HRQoL of any child or adolescent, with or without a chronic medical condition while the chronic generic module focuses on issues related to living with a chronic medical condition. The chronic generic module offers the possibility of comparing the HRQoL score between different chronic medical conditions. By supplementing the chronic generic module with a condition-specific module the clinician or investigator are given additional information concerning a specific condition. It is suggested that collected information from a condition-specific module relates more closely to the treatment regime and is more responsive to clinically significant changes [2-5].

Before actual HRQoL assessment can take place a questionnaire has to meet certain standards (Box 2). These criteria have been established to achieve a certain level of international conformity and facilitate the chance of incorporation in future studies or clinical use [6]. The DISABKIDS project aimed to meet the necessary requirements, yet some aspects such as responsiveness and interpretation still need to be further assessed.

- Sound theoretical basis and definition of HRQoL
- Multidimensional measurement
- Suitable for study question
- Domains are described and scored separately
- Adequate psychometrics (reliability and validity)
- Sensitive to changes over time (responsiveness)
- Norm group data available (for disease and age)
- Practical in use (administration and interpretability)
- Accepted by patients
- Appropriate to culture and lifestyle

Box 2. Recommended criteria for HRQoL measures [2,3,6-11].

Limitations

Although the design and aim of the DISABKIDS instrument sounds promising there are still several limitations and methodological issues that need to be addressed. A recurring problem is the recruitment and inclusion of participants. Selection bias is a possibility as the participants who were willing to contribute may not be representative for the population. Recruitment may have been influenced by the attitude or interest towards HRQoL, individual confidence, the willingness to do something for the paediatrician, better coping mechanism or a higher experienced HRQoL [12]. Non-responders might have more severe asthma, may lack the energy to participate or have a different view on illness and the effect it has on their lives. However, there are also several aspects that help to strengthen the validity of the findings. Central is the fact that the respondents were recruited from several European locations and the severity distribution of the asthma group was similar to other reports [13]. If a selection bias has occurred in the DISABKIDS project the observed HRQoL would probably be higher with a narrower severity distribution, compared to the population of interest. Thus, our results may underestimate the real variation in HRQoL among the different severity states and underrate the discriminative properties of the DISABKIDS instrument.

A further limitation was that the number of respondents in some chronic condition groups, cerebral palsy and atopic dermatitis in particular, was relatively small in both the pilot and the field studies. So even though the total number of participants over all conditions and countries was acceptable, the results of the separate analyses of some chronic conditions should be interpreted with caution. Although the cross-national focus has been an explicit approach of the DISABKIDS project, lack of time and resources stood in the way of testing each chronic condition in each DISABKIDS country. Only the asthma specific module was tested adequately in all seven countries (Chapter 6 and 7). However, a comparison of the asthma outcome between countries is still problematic, as some countries have only tested around 30 or 40 children and adolescents. All modules will need to be tested further in larger groups and across countries in future studies.

Another critical note relates to the DISABKIDS project as a European consortium. Within the European project, all countries worked individually on each work package. Although care was taken to stress uniformity in the group (such as supplying a manual for the focus groups, planning regular DISABKIDS meetings and describing every work package in detail) each DISABKIDS member had a considerable amount of autonomy. There was no opportunity to monitor how investigators in each country completed the work packages and no way of checking aspects such as the method of recruitment or data collection. Several factors could have played a role, including personal interpretations and interests, hospital facilities, time constraints and earlier research experience. For example, focus groups were used in the DISABKIDS project to take into account the child and adolescent's own ideas and language (Chapter 3 and 4). The literal transcripts were available in each national language and the investigator was responsible for the selection of statements and the translation into English. As there was no official translation the quality of the supplied English statement could not be guaranteed. The meaning of the original statement may have been altered, which could influence the chance of being selected as final instrument item. A possible solution would have been to use expert or panel translators, supply training sessions or perform these tasks with an international group to facilitate European conformity [14,15]. We emphasise the importance of training, as the quality of the collected data is very much tied to the skills of the investigator (Box 3).

The bottom-up (patient-derived) methodology that was applied in the DISABKIDS project was another reason for debate. The collection of the HRQoL statements from the focus groups and interviews were the basis of the DISABKIDS instrument and were applied to secure that the child and adolescent's opinion was incorporated (Chapter 3 and 4). This patient-derived method was followed by the (top-down) investigator's judgement for the selection of the final items (Chapter 5). This top-down procedure conflicted with the aim of developing the DISABKIDS instrument through a bottom-up procedure (child and adolescent input). With a patient-derived method one would prefer to have the children and adolescents select the important items but the extensive statement pool (3515 statements) was thought to be too large for them [16-19]. To compensate for the top-down procedure the child and adolescent's opinion was again included into the pilot test when they were asked to approve the selected items and judge them on comprehension and applicability in the cognitive interview [20-22].

- Recruitment of the participants
- Moderating the focus groups
- Identifying the appropriate statements from the focus group transcripts
- Translation of the HRQoL statements and items
- Rewriting statements to items
- Data input
- Statistical analyses

Box 3. Situations in which specific guidelines, expert translators, training sessions or international working groups would be advised.

On the other hand, because most aspects identified in the DISABKIDS asthma focus groups had been discussed in earlier publications (Chapter 4), one might question whether it is still necessary to include the patient's opinion when developing a questionnaire. Despite the extensive research of certain conditions (i.e. asthma) we still advocate to include the patients' opinions. Information on some chronic conditions (i.e. cerebral palsy or atopic dermatitis) is still limited and new results and different viewpoints may be yielded through patient-derived methods. Furthermore, the main advantage of the focus groups in the DISABKIDS project was the cross-national recurrence of issues and the combination of patient, parent and clinician's data.

A further drawback in our method is that we cannot assume that all important issues were included in the DISABKIDS instrument. Not all relevant topics may have been discussed in the focus groups or some children may have found it difficult to talk about certain topics. It is also possible that some topics were removed in the item selection phase (Chapter 5). The cross-national developmental process disregarded some items that may have been important in certain countries (for instance items concerning pets or riding a bike to school). A possibility would have been to run focus groups till no new issues were presented [12,23,24]. This was not possible in the DISABKIDS project as each work package was set in a certain time frame.

In short, there where a number of methodological issues and limitations during the development of the DISABKIDS instrument. Even so, the initial psychometric results and the first implementation experiences by clinicians and investigators are promising. Future research will help to explore the implementation possibilities.

DISABKIDS's future

The DISABKIDS instrument can play an important part in the future of paediatric HRQoL assessment. The two level modular build-up and the multiple language versions of the DISABKIDS instrument makes it utilizable in several circumstances, including population studies, clinical trials and individual assessment.

- **HRQoL evaluation:** includes mainly the description of a population group or a comparison between patient groups [25-31,31,32]. It can give the clinician a fair description of a group but is of little use for the care of the individual patient. The availability of the different DISABKIDS language versions makes the instrument suitable for group and cross-national comparisons.
- **HRQoL in clinical trials:** is mainly used to compare the outcome of different treatments within a group or to evaluate therapeutic effectiveness between groups [2,33,34]. While the use of HRQoL is increasingly being implemented in adult clinical trials the inclusion of HRQoL in paediatric clinical trials is still limited [35-38]. In the future the DISABKIDS instrument can be of use in (cross-)national clinical trials. Results are of use to the clinician and the general patient group but have no role in the individual evaluation and treatment of the patient.

- **HRQoL in clinical practice:** is aimed at the care of the individual. Objective measures of disease or clinical judgement in for instance asthma only weakly correlate with how a patient feels and functions (Chapter 7) [39-42]. HRQoL assessment can therefore provide a broader picture of health and provide insight into the impact of a chronic medical condition on the daily life of an individual child or adolescent. Knowledge of the HRQoL status can improve medical guidance to the children and their parents, identify those that need particular attention, screen for psychosocial problems or monitor the patient's progress [43]. The children and adolescent's HRQoL can be assessed with the help of a paper-pencil or computer-assisted instrument. The use of the DISABKIDS instrument in clinical practice will still need to be tested.

While the first psychometric results of the DISABKIDS instrument sound promising and the design comes with several advantages it is still essential to further test the current instrument in several situations to judge where improvements are necessary (Box 4). For instance additional testing of the modules is necessary in each country and for each module in sufficiently large groups. The chronic generic module can also be tested for applicability in other chronic medical conditions, for example haemophilia, heart disease or obesity. There is also need for more evidence that the instrument can function as an individual screening tool, which includes higher levels of reliability (Cronbach's alpha ≥ 0.9)[6,44].

- Psychometric properties in each country with sufficiently large groups
- Psychometric properties for each chronic condition in larger groups
- Comparisons to existing condition-specific HRQoL questionnaires
- Comparisons to clinical outcome and physiologic assessment of disease severity
- Sensitivity and the responsiveness to change in individual patients
- Longitudinal data to assess long-term changes in measured HRQoL
- Use in comparing interventions, treatment changes or different medications
- Relevance to clinical practice
- Appropriateness for cultural background of the patient

Box 4. Aspects that need to be studied further.

However, continuation of the DISABKIDS project is not uncomplicated and depends on external factors as time and resources. Since the European funding of the DISABKIDS project has ended there is a danger of discontinuation. Nevertheless, we still aim to interest investigators and clinicians in the continuation of testing and implementing the DISABKIDS instrument. The current advantages of the DISABKIDS instrument, especially the possibility of working with an international consortium, should give the instrument a fair chance. The available DISABKIDS manual should also assist in the proceedings to look for cross-national collaborations in the future to further validate and implement the DISABKIDS instrument.

HRQoL assessment in clinical practice; implementation philosophy

Since the management of chronic medical conditions revolves more around care than cure and HRQoL has been recognised as important to the care of children and adolescents, the number of paediatric HRQoL questionnaires has grown over the last decades [45,46]. There is however, still little evidence of their relevance in and influence on adult and paediatric clinical practice and the current need is to discuss why HRQoL assessment is not systematically implemented (Chapter 2)[37,47-51].

Experienced barriers

One problem is that there are several definitions of quality of life (QoL). There is no gold standard as to what it represents or how it can be measured as it includes subjective issues and the concept depends on the applied perspective (social, economical, psychological) [2,7,45,52,53]. A similar problem concerns HRQoL [54]. One can question whether we are able to reliably assess a subjective concept as HRQoL. A person's perception of health and expectations are related to the individual and can vary over time [55]. Further complicating is that when HRQoL is assessed in children and adolescents there are even more practical aspects to consider as cognitive development, changing perspectives, disease knowledge and age related activities, all of which can influence HRQoL outcome [56]. There is no straightforward way of solving these aspects.

Another issue that needs to be considered is that clinicians may feel that identified problems lay outside the traditional area of medical care and may not see it as their task to discuss HRQoL issues with their patients [33,47,49,50]. Clinicians were found to focus on symptoms and physical functioning but rarely on emotional or social problems (Chapter 4)[47,57-60]. A dilemma is that if psychosocial problems are revealed and the clinician feels incapable of interfering with these problems, they may be reluctant to adopt HRQoL measures. This is one of the reasons why the clinicians' perspective also needs to be taken into account during the development of a HRQoL questionnaire for clinical use (Chapter 2). The clinician may contribute by suggesting which aspects to measure so that the questionnaire relates to issues he feels he can intervene in. Strategies can also be discussed on how a questionnaire is best implemented and experienced barriers can be avoided. The possibility of giving HRQoL feedback to other health care professionals such as (specialised) nurses or psychologists also need to be considered [48,49].

Although some patients do not feel comfortable about discussing certain issues with their clinicians, the majority of patients want their clinicians to assess HRQoL aspects and feel this is useful to clinical practice [47,48,57-59,61,62]. Communication is seen as a crucial element in the quality of health care and can positively influence patient health outcome [63-66].

In summary, there are still ample problems that need to be solved before HRQoL is regularly assessed in clinical practice. Fortunately, there is evidence that clinicians are interested in HRQoL outcome, especially when it concerns a chronic medical condition

(Chapter 2 and 4)[48-50,67]. It is now essential to identify and reduce experienced barriers (Chapter 2) to encourage the implementation of HRQoL assessment in clinical practice on a regular basis (Box 5).

- HRQoL not seen as a priority in clinical practice
- Unfamiliar with HRQoL questionnaires
- Insufficient training in and knowledge of HRQoL
- Unavailability of appropriate questionnaires
- Unsatisfactory psychometric properties
- No proof of clinical relevance
- Insufficient feasibility (ease of collection and use)
- Costs of implementation
- Limited time and resources
- No intervention guidelines

Box 5. Main barriers for the use of HRQoL questionnaires in clinical practice [47-49,68].

Requirements necessary before clinical implementation

A fundamental concern is whether a questionnaire, like the DISABKIDS instrument, can and will be used for individual patient assessment. Current studies inform us more about experienced barriers and lack of clinical impact than about required essentials for successful and meaningful use of HRQoL assessment in daily clinical practice [69]. Thus, if HRQoL questionnaires are to be used routinely and become an important part of clinical practice (especially paediatric health care) the given obstacles (Box 5) need to be dealt with and HRQoL assessment needs to be promoted. Requirements to achieve acceptance of HRQoL assessment include: informing clinicians about available questionnaires, proving clinical relevance and providing guidelines for interpretation of HRQoL outcome scores (Box 6). If the necessary requirements are achieved the HRQoL questionnaire is more likely to be accepted by clinicians and to be included as outcome in the care for the patient.

Promotional needs:
- Information: increase familiarity with HRQoL and publish data in clinical journals
- Training: in implementation possibilities and interpretation of HRQoL outcome
- Health care professionals: stimulate a multidisciplinary approach in HRQoL assessment

Questionnaire factors:
- Content: includes items regarding important aspects for the patient
- Design: short, practical, computerised
- Psychometrics: reliable, valid, sensitive to change and availability of norm data
- Outcome: clinical relevance

Practical requirements:
- Implementation: easily available, quick to complete and administer
- Scoring: simple, provided promptly and in a useful format
- Interpretation: guidelines available for easy interpretation
- Intervention: strategies to translate outcome into specific interventions

Box 6. Requirements to promote HRQoL assessment in clinical practice [9,33,43,47-50,53,70-72].

Is there proof of clinical relevance?
The importance of HRQoL assessment in clinical practice is stressed as it is assumed to provide meaningful clinical information. Various suggestions are provided of how HRQoL assessment may be of benefit to individual patient care (Box 7) but clinical relevance is not always clear-cut [48,61,62,73,74]. Greenhalgh et al. (2005) have described the mechanisms between HRQoL intervention and expected outcome in a model, demonstrating its complexity [47]. The challenge is to decide what outcome to measure as several processes (communication, treatment response, recognition of problems) can be influenced before the final outcome of improved HRQoL or patient satisfaction is realized [47,61].

- Identifying and prioritising problems
- Assess treatment efficiency
- Monitoring disease progression
- Assisting in informed treatment changes
- Facilitating clinician-patient communication
- Improving patient satisfaction
- Allocating health care resources

Box 7. Suggested use of HRQoL measures in clinical practice [9,48,51,61,72,75].

A number of studies have reported on the impact of HRQoL feedback to clinicians. In general there is limited proof of influence on medical decisions (referring to others, treatment changes, clinical tests), patient satisfaction or HRQoL outcome [47,61-63,73,75]. Feedback of HRQoL assessment to clinicians does affect the extent in which HRQoL issues are discussed in a consultation, improves identification of psychological and social problems and increases the clinicians' awareness of the patient's HRQoL [47,61,63,73,75,76]. Only a few studies demonstrated that this increased recognition of HRQoL problems is subsequently associated with clinical intervention (follow-up appointments, counselling or referral) [48,59,76]. Clinicians may not consider HRQoL issues to be important enough to adapt their treatment or referrals to it. The facilitated communication, resulting from the HRQoL feedback, may be sufficient for clinicians [47-49,62]. Disappointingly there are currently no implementation studies available that describe individual HRQoL assessment in paediatric care.

What do we gain through HRQoL assessment?
There is a growing awareness that the clinician, parent and child or adolescent differ in their perception of HRQoL, disease severity and treatment expectations (Chapter 4 and 7) [77-81]. These differences, together with insufficient clinician-patient communication can lead to misunderstandings and dissatisfied patients [65]. If HRQoL assessment can improve the clinician-patient communication and patient health outcome, this may well be a sufficient reason to implement HRQoL measures [63,65]. Although this has not been proven in paediatric care, common sense tells us that improved communication can facilitate the recognition and acknowledgment of problems and can enable clinicians to improve the quality of care of the child and adolescent.

Implementation in asthma care

The asthma focus groups and interviews illustrated that there is a considerable impact on the life of a child and adolescent with asthma (Chapter 4). Physical limitations, which were often linked to social issues, were a dominant theme and non-compliance seemed to be linked to insufficient knowledge or denial. Clinicians found it hard to recognise these important issues in the life of a child or adolescent with asthma and felt that awareness of and familiarity with these problems might assist them in improving the care for their patients (Chapter 2 and 4). If children or adolescents feel misunderstood by their clinician, or for that matter their parents, this can negatively influence their clinician-patient relationship and may even affect their adherence to treatment.

The DISABKIDS instrument can evaluate a patient's HRQoL and help the clinician to focus on areas of particular importance to the child and adolescent. A future prospective could be to ask patients to complete the DISABKIDS instrument before consultation, preferably on a computer. The computerised instrument can be easily administered, save time and supply the clinician with immediate feedback of the patient's HRQoL status [9,73,82,83]. If the DISABKIDS computer version is implemented this can give an instant readout of the 0-100 score on each domain and compare this to a previous assessment or to the population norm data. The 37-item chronic generic module can provide general data on the impact of living with asthma. The 11-item asthma specific module can supply the clinician with asthma-related issues by concentrating on specific limitations and fears related to asthma. Any conspicuous scores can then be discussed with the patient. For instance if a low score on the medication domain is discussed with the child this could make clear that the child is rebelling against the medication because he or she doesn't feel it is doing any good. Clinical parameters or regular consultations may not have identified this problem. Problematic issues can be discussed, problems can be dealt with or explained and if necessary the child or adolescent can be referred to the appropriate health care professional (social worker, psychologist).

Conclusion

While HRQoL may seem ill defined and its assessment unscientific, to the patient this subjective outcome may be more important than biomedical endpoints [45]. Although the inadequate proof of clinical relevance may currently be the main reason for the limited use of HRQoL assessment in clinical practice, the expectation is that in the future a growing number of clinicians will incorporate routine HRQoL assessment [62,72]. In the mean time considerable work needs to be done to prove the benefit of HRQoL assessment in clinical practice and to overcome experienced barriers.

The European DISABKIDS project has come a long way in the development of a new cross-national HRQoL instrument for children and adolescents with a chronic medical condition. The DISABKIDS instrument can play an important role in future paediatric HRQoL assessment. The modular build up and cross-national development also offers advantages for assessment on a national and international level in HRQoL evaluation studies and clinical studies. However, further evaluation of the DISABKIDS instrument is needed to test its performance as individual measure in clinical practice and prove its relevance to clinicians. This refinement can only be achieved through future implementation, as understanding how current measures perform in practice facilitates improvements [84].

We may need to restrain our expectations of the impact of HRQoL assessment on clinical practice. As there is currently insufficient evidence that HRQoL assessment changes the treatment and referral plans of the clinician we might need to accept that an improved clinician-patient communication is sufficient reason to implement HRQoL questionnaires [47,63]. Improved communication can be an important component of the overall HRQoL assessment of a patient. Clinicians can benefit from the information presented to them and use it to facilitate communication and discuss problematic areas. Yet, one does need to keep in mind that HRQoL assessment will never address all issues that are important to the patient and that it can only supplement current clinical measures or communication and does not substitute them.

References

1. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. Int.J.Rehabil.Res.2002; 25:197-206.
2. Fitzpatrick R, Davey C, Buxton MJ, Jones DR. Evaluating patient-based outcome measures for use in clinical trials. Health.Technol.Assess.1998; 2:i-74.
3. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol.Assess.2001; 5:1-157.
4. Guyatt GH, King DR, Feeny DH, Stubbing D, Goldstein RS. Generic and specific measurement of health-related quality of life in a clinical trial of respiratory rehabilitation. J.Clin.Epidemiol.1999; 52:187-192.
5. McSweeny AJ, Creer TL. Health-related quality-of-life assessment in medical care. Dis.Mon.1995; 41:1-71.
6. Assessing health status and quality-of-life instruments: attributes and review criteria. Qual.Life.Res.2002; 11:193-205.
7. Spieth LE, Harris CV. Assessment of health-related quality of life in children and adolescents: an integrative review. J.Pediatr.Psychol.1996; 21:175-193.
8. Doward LC, Meads DM, Thorsen H. Requirements for quality of life instruments in clinical research. Value Health.2004; 7 Suppl 1:S13-S16.
9. Higginson IJ, Carr AJ. Measuring quality of life: using quality of life measures in the clinical setting. BMJ 2001; 322:1297-1300.
10. Cramer JA. Principles of health-related quality of life: assessment in clinical trials. Epilepsia.2002; 43:1084-1095.
11. Wallander JL, Schmitt M, Koot HM. Quality of life measurement in children and adolescents: issues, instruments, and applications. J.Clin.Psychol.2001; 57:571-585.
12. Sim J. Collecting and analysing qualitative data: issues raised by the focus group. J.Adv.Nurs.1998; 28:345-352.
13. Rosier MJ, Bishop J, Nolan T, Robertson CF, Carlin JB, Phelan PD. Measurement of functional severity of asthma in children. Am.J.Respir.Crit.Care Med.1994; 149:1434-1441.
14. Swaine-Verdier A, Doward LC, Hagell P, Thorsen H, McKenna SP. Adapting quality of life instruments. Value.Health.2004; 7 Suppl 1:S27-S30.
15. The World Health Organization Quality of Life assessment (WHOQOL): position paper from the World Health Organization. Soc.Sci.Med.1995; 41:1403-1409.
16. Townsend M, Feeny DH, Guyatt GH, Furlong WJ, Seip AE, Dolovich J. Evaluation of the burden of illness for pediatric asthmatic patients and their parents. Ann.Allergy 1991; 67:403-408.
17. Griffiths AM, Nicholas D, Smith C, Munk M, Stephens D, Durno C, Sherman PM. Development of a quality-of-life index for pediatric inflammatory bowel disease: dealing with differences related to age and IBD type. J.Pediatr.Gastroenterol.Nutr.1999; 28:S46-S52.

18. Richardson G, Griffiths AM, Miller V, Thomas AG. Quality of life in inflammatory bowel disease: a cross-cultural comparison of English and Canadian children. J.Pediatr. Gastroenterol.Nutr.2001; 32:573-578.
19. Rutishauser C, Sawyer SM, Bond L, Coffey C, Bowes G. Development and validation of the Adolescent Asthma Quality of Life Questionnaire (AAQOL). Eur.Respir.J. 2001; 17:52-58.
20. Petersen C, Schmidt S, Power M, Bullinger M, and the DISABKIDS group. Development and pilot-testing of a health-related quality of life chronic generic module for children and adolescents with chronic health conditions: a European per spective. Qual.Life.Res 2005. (In Press)
21. Quittner AL, Sweeny S, Watrous M, Munzenberger P, Bearss K, Gibson NA, Fisher LA, Henry B. Translation and linguistic validation of a disease-specific quality of life measure for cystic fibrosis. J.Pediatr.Psychol.2000; 25:403-414.
22. Barofsky I. Cognitive aspects of quality of life assessment. In Spilker B (ed). Quality of life and pharmacoeconomics in clinical trials. Philadelphia: Lippincott-Raven; 1996:107-115.
23. McEwan MJ, Espie CA, Metcalfe J, Brodie MJ, Wilson MT. Quality of life and psychosocial development in adolescents with epilepsy: a qualitative investigation using focus group methods. Seizure.2004; 13:15-31.
24. McLafferty I. Focus group interviews as a data collecting strategy. J.Adv.Nurs.2004; 48:187-194.
25. Brunner HI, Giannini EH. Health-related quality of life in children with rheumatic diseases. Curr.Opin.Rheumatol.2003; 15:602-612.
26. Kolsteren MM, Koopman HM, Schalekamp G, Mearin ML. Health-related quality of life in children with celiac disease. J.Pediatr.2001; 138:593-595.
27. Austin JK, Smith MS, Risinger MW, McNelis AM. Childhood epilepsy and asthma: comparison of quality of life. Epilepsia.1994; 35:608-615.
28. Hallstrand TS, Curtis JR, Aitken ML, Sullivan SD. Quality of life in adolescents with mild asthma. Pediatr.Pulmonol.2003; 36:536-543.
29. Austin JK, Huster GA, Dunn DW, Risinger MW. Adolescents with active or inactive epilepsy or asthma: a comparison of quality of life. Epilepsia.1996; 37:1228-1238.
30. Loonen HJ, Grootenhuis MA, Last BF, Koopman HM, Derkx HH. Quality of life in paediatric inflammatory bowel disease measured by a generic and a disease-specific questionnaire. Acta.Paediatr.2002; 91:348-354.
31. Sawyer MG, Reynolds KE, Couper JJ, French DJ, Kennedy D, Martin J, Staugas R, Ziaian T, Baghurst PA. Health-related quality of life of children and adolescents with chronic illness--a two year prospective study. Qual.Life.Res.2004; 13:1309-1319.
32. Okelo SO, Wu AW, Krishnan JA, Rand CS, Skinner EA, Diette GB. Emotional quality-of-life and outcomes in adolescents with asthma. J.Pediatr.2004; 145:523-529.
33. Wu AW, Cagney KA. The role of quality of life assessments in medical practice. In Spilker B (ed). Quality of life and pharmacoeconomics in clinical trails. Philadelphia: Lippincott-Raven; 1996:517-522.

34. Beenakker EA, Fock JM, Van Tol MJ, Maurits NM, Koopman HM, Brouwer OF, Van der Hoeven JH. Intermittent prednisone therapy in Duchenne muscular dystrophy: a randomized controlled trial. Arch.Neurol.2005; 62:128-132.
35. Quality of life and clinical trials. Lancet.1995; 346:1-2.
36. Sanders C, Egger M, Donovan J, Tallon D, Frankel S. Reporting on quality of life in randomised controlled trials: bibliographic study. BMJ 1998; 317:1191-1194.
37. Clarke SA, Eiser C. The measurement of health-related quality of life (QOL) in paediatric clinical trials: a systematic review. Health.Qual.Life Outcomes 2004; 2:66.
38. Bender BG. Measurement of quality of life in pediatric asthma clinical trials. Ann. Allergy Asthma Immunol.1996; 77:438-445.
39. Juniper EF, Guyatt GH, Feeny DH, Ferrie PJ, Griffith LE, Townsend M. Measuring quality of life in children with asthma. Qual.Life.Res.1996; 5:35-46.
40. van der MT, Postma DS, Schreurs AJ, Bosveld HE, Sears MR, Meyboom dJ. Discriminative aspects of two generic and two asthma-specific instruments: relation with symptoms, bronchodilator use and lung function in patients with mild asthma. Qual.Life Res.1997; 6:353-361.
41. Rowe BH, Oxman AD. Performance of an asthma quality of life questionnaire in an outpatient setting. Am.Rev.Respir.Dis.1993; 148:675-681.
42. Carranza R, Jr., Edwards L, Lincourt W, Dorinsky P, ZuWallack RL. The relationship between health-related quality of life, lung function and daily symptoms in patients with persistent asthma. Respir.Med.2004; 98:1157-1165.
43. Fitzpatrick R, Fletcher A, Gore S, Jones D, Spiegelhalter D, Cox D. Quality of life measures in health care. I: Applications and issues in assessment. BMJ 1992; 305:1074-1077.
44. Nunnally JC, Bernstein IH. Psychometric theory. New York: McGraw-Hill; 1994.
45. Eiser C, Morse R. The measurement of quality of life in children: past and future perspectives. J.Dev.Behav.Pediatr.2001; 22:248-256.
46. Andelman RB, Zima BT, Rosenblatt AB. Quality of life of children: toward conceptual clarity. In Maruish ME (ed). The use of psychological testing for treatment planning and outcomes assessment. Mahwah, NJ: Lawrence Erlbaum; 1999:1383-1413.
47. Greenhalgh J, Long AF, Flynn R. The use of patient reported outcome measures in routine clinical practice: lack of impact or lack of theory? Soc.Sci.Med.2005; 60:833-843.
48. Greenhalgh J, Meadows K. The effectiveness of the use of patient-based measures of health in routine practice in improving the process and outcomes of patient care: a literature review. J.Eval.Clin.Pract.1999; 5:401-416.
49. Baars RM, van der Pal SM, Koopman HM, Wit JM. Clinicians' perspective on quality of life assessment in paediatric clinical practice. Acta.Paediatr.2004; 93:1356-1362.
50. Taylor KM, Macdonald KG, Bezjak A, Ng P, DePetrillo AD. Physicians' perspective on quality of life: an exploratory study of oncologists. Qual.Life Res.1996; 5:5-14.

51. Gilbody SM, House AO, Sheldon T. Routine administration of Health Related Quality of Life (HRQoL) and needs assessment instruments to improve psychological outcome--a systematic review. Psychol.Med.2002; 32:1345-1356.
52. Anderson KL, Burckhardt CS. Conceptualization and measurement of quality of life as an outcome variable for health care intervention and research. J.Adv.Nurs.1999; 29:298-306.
53. Wood-Dauphinee S. Assessing quality of life in clinical research: from where have we come and where are we going? J.Clin.Epidemiol.1999; 52:355-363.
54. Doward LC, McKenna SP. Defining patient-reported outcomes. Value Health.2004; 7 Suppl 1:S4-S8.
55. Carr AJ, Gibson B, Robinson PG. Measuring quality of life: Is quality of life determined by expectations or experience? BMJ 2001; 322:1240-1243.
56. Harding L. Children's quality of life assessments: a review of generic and health related quality of life measures completed by children and adolescnets. Clin.Psychol. Psychother.2001; 8:79-96.
57. Schor EL, Lerner DJ, Malspeis S. Physicians' assessment of functional health status and well-being. The patient's perspective. Arch.Intern.Med.1995; 155:309-314.
58. Detmar SB, Aaronson NK, Wever LD, Muller M, Schornagel JH. How are you feeling? Who wants to know? Patients' and oncologists' preferences for discussing health-related quality-of-life issues. J.Clin.Oncol.2000; 18:3295-3301.
59. Jacobs JE, van de Lisdonk EH, Smeele I, van Weel C, Grol RP. Management of patients with asthma and COPD: monitoring quality of life and the relationship to subsequent GP interventions. Fam.Pract.2001; 18:574-580.
60. Rothwell PM, McDowell Z, Wong CK, Dorman PJ. Doctors and patients don't agree: cross sectional study of patients' and doctors' perceptions and assessments of disability in multiple sclerosis. BMJ 1997; 314:1580-1583.
61. Detmar SB, Muller MJ, Schornagel JH, Wever LD, Aaronson NK. Health-related quality-of-life assessments and patient-physician communication: a randomized controlled trial. JAMA 2002; 288:3027-3034.
62. Wagner AK, Ehrenberg BL, Tran TA, Bungay KM, Cynn DJ, Rogers WH. Patient-based health status measurement in clinical practice: a study of its impact on epilepsy patients' care. Qual.Life Res.1997; 6:329-341.
63. Velikova G, Booth L, Smith AB, Brown PM, Lynch P, Brown JM, Selby PJ. Measuring quality of life in routine oncology practice improves communication and patient wellbeing: a randomized controlled trial. J.Clin.Oncol.2004; 22:714-724.
64. Laine C, Davidoff F, Lewis CE, Nelson EC, Nelson E, Kessler RC, Delbanco TL. Important elements of outpatient care: a comparison of patients' and physicians' opinions. Ann.Intern.Med.1996; 125:640-645.
65. Stewart MA. Effective physician-patient communication and health outcomes: a review. CMAJ 1995; 152:1423-1433.
66. Detmar SB, Aaronson NK. Quality of life assessment in daily clinical oncology practice: a feasibility study. Eur.J.Cancer 1998; 34:1181-1186.

67. Walsh DL, Emrich LJ. Measuring cancer patients' quality of life. A look at physician attitudes. N.Y.State.J.Med.1988; 88:354-357.
68. Morris J, Perez D, McNoe B. The use of quality of life data in clinical practice. Qual. Life Res.1998; 7:85-91.
69. Thornicroft G, Slade M. Are routine outcome measures feasible in mental health? Qual.Health Care 2000; 9:84.
70. Guillemin F. Functional disability and quality-of-life assessment in clinical practice. Rheumatology (Oxford) 2000; 39 Suppl 1:17-23.
71. Bezjak A, Ng P, Skeel R, DePetrillo AD, Comis R, Taylor KM. Oncologists' use of quality of life information: results of a survey of Eastern Cooperative Oncology Group physicians. Qual.Life Res.2001; 10:1-13.
72. McHorney CA, Earl BD, Jr. A qualitative study of patients' and physicians' views about practice-based functional health assessment. Med.Care 2002; 40:1113-1125.
73. Taenzer P, Bultz BD, Carlson LE, Speca M, DeGagne T, Olson K, Doll R, Rosberger Z. Impact of computerized quality of life screening on physician behaviour and patient satisfaction in lung cancer outpatients. Psychooncology 2000; 9:203-213.
74. Kazis LE, Callahan LF, Meenan RF, Pincus T. Health status reports in the care of patients with rheumatoid arthritis. J.Clin.Epidemiol.1990; 43:1243-1253.
75. Espallargues M, Valderas JM, Alonso J. Provision of feedback on perceived health status to health care professionals: a systematic review of its impact. Med.Care 2000; 38:175-186.
76. Rubenstein LV, McCoy JM, Cope DW, Barrett PA, Hirsch SH, Messer KS, Young RT. Improving patient quality of life with feedback to physicians about functional status. J. Gen.Intern.Med.1995; 10:607-614.
77. Loonen HJ, Derkx BH, Griffiths AM. Pediatricians overestimate importance of physical symptoms upon children's health concerns. Med.Care.2002; 40:996-1001.
78. Janse AJ, Gemke RJ, Uiterwaal CS, van dT, I, Kimpen JL, Sinnema G. Quality of life: patients and doctors don't always agree: a meta-analysis. J.Clin.Epidemiol.2004; 57:653-661.
79. Theunissen NC, Vogels TG, Koopman HM, Verrips GH, Zwinderman KA, Verloove-Vanhorick SP, Wit JM. The proxy problem: child report versus parent report in health-related quality of life research. Qual.Life Res.1998; 7:387-397.
80. Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. Qual.Life Res.2001; 10:347-357.
81. Sung L, Young NL, Greenberg ML, McLimont M, Samanta T, Wong J, Rubenstein J, Ingber S, Doyle JJ, Feldman BM. Health-related quality of life (HRQL) scores reported from parents and their children with chronic illness differed depending on utility elicitation method. J.Clin.Epidemiol.2004; 57:1161-1166.
82. Velikova G, Wright EP, Smith AB, Cull A, Gould A, Forman D, Perren T, Stead M, Brown J, Selby PJ. Automated collection of quality-of-life data: a comparison of paper and computer touch-screen questionnaires. J.Clin.Oncol.1999; 17:998-1007.

83. Koopman HM, Baars RM, Segaar RW. The use of computer-aided health-related quality-of-life questionnaires for children with a chronic disease and their parents. The Ear Foundation. 33-44. 2003. Oxford, Hughes associates. Measuring the Immeasurable? Proceedings of a Conference on Quality of Life in Deaf Children. 17-5-2002.
84. Eiser C, Morse R. A review of measures of quality of life for children with chronic illness. Arch.Dis.Child.2001; 84:205-211.

9

Summary & Samenvatting

Summary

The World Health Organisation Quality of Life (WHOQoL) group defines quality of life (QoL) as: “an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns”. QoL has become an increasingly important concept in society as well as in research. Social and political questions are raised about the equal distribution of QoL in our own society and over the world. In cases where people are in poor health or have become ill, improving QoL has become a key issue. Reasons for the importance of assessing QoL include the awareness that objective measures of disease severity have shown poor correlations with how a patient feels. Of influence is also the change, caused by improved medical developments, in the impact of certain diseases from potentially lethal to chronic which has led to a treatment shift from cure to care. Hence the increased focus on health related quality of life (HRQol). In general QoL research is aimed at ensuring and improving physical and mental fitness and wellbeing. This has led to the development of several questionnaires to assess and describe QoL in various patient groups. In addition clinical research projects are increasingly including QoL as outcome measure when comparing treatment regimes. Existing QoL questionnaires are still being improved and new questionnaires are continuously being developed and tested for specific patient groups.

This thesis generally describes the developmental processes followed within the European DISADKIDS project, which was a collaboration of clinicians and investigators from seven European countries. Their aim was to develop a new European HRQoL instrument for children and adolescents with a chronic medical condition. The ***first chapter*** of this thesis begins with an explanation of the general principles related to HRQoL. It decribes the increased interest in the assessment of QoL but also shows that the use in clinical practice is still limited. Asthma is a main topic in the thesis which is why attention is also given to the mental and physical effects of asthma on children and adolescents. We decribe how questionnaires are generally developed through similar steps including defining the target group and the aim of the questionnaire, selecting items and testing the validity. The steps taken in the DISABKIDS project include focus groups in which patients indicate how their chronic condition influences them in daily life and what QoL aspects they find important. Participants were children and adolescents (and their parents) in three age groups (4-7, 8-12, 13-16) with the following chronic medical conditions: asthma, juvenile idiopathic arthritis, atopic dermatitis, cerebral palsy, cystic fibrosis, diabetes or epilepsy. The three level modular design, the inclusion of seven chronic medical conditions, the wide age range (4-16 years) and the availability of a self-assessment and a proxy version are aspects that make the DISABKIDS instrument unique.

As there is little information on the attitude of clinicians towards using QoL measurements for patient care the first objective was to learn more about the perspective of paediatric clinicians on implementing QoL assessment in clinical practice. ***Chapter two*** describes a survey done among the members of the Dutch Paediatric Association. More than half of the 303 respondents (57%) believed that it was possible to use QoL questionnaires in clinical practice. The majority indicated that assessing QoL was beneficial and that it was especially necessary to assess QoL in children with a chronic disease (82%), although only a minority (17%) currently used QoL questionnaires. Obstacles that prevent the use of QoL questionnaires are the extra time needed for assessment, the unavailability of standardized questionnaires and insufficient knowledge about QoL. These are aspects that need to be considered when developing a new instrument.

The patients were considered the main source of information for the basis of the instrument, especially for the development of the included items. Focus groups were applied to identify relevant HRQoL statements from the perspective of the patient for use as items in the DISABKIDS instrument. Participants included 154 children and adolescents with a chronic medical condition, 142 family members and 26 health care professionals. A total amount of 3515 HRQoL statements were identified. The focus group method needed to be adapted for children and adolescents. A number of issues need to be considered, including developmental abilities, language skills and attention span. There was also the need to create uniformity of the method between countries. The ***third chapter*** describes the applied focus group methodology.

The asthma focus groups and interviews were conducted in four European countries (Greece, Italy, the Netherlands and Sweden). A description of discussed topics and outcomes is given in ***chapter four***. A total of 43 children and adolescents with asthma, 33 family members and 7 health care professionals participated in the focus groups and interviews. The children and adolescents discussed the current limitations they experienced which included asthma symptoms, related medical aspects, physical limitations and social issues. Parents concentrated more on the consequences for the future, including prevention, long-term effects of medication and schooling possibilities. The focus group discussions were found to be a useful tool in generating statements from children and adolescents with a chronic condition for use in the development of a questionnaire.

The qualitative data, collected from the cross-national DISABKIDS focus groups and interviews, consisted of a total of 3515 HRQoL statements. These included generic QoL statements (n=488) not related to any medical condition, chronic generic statements (n=1647) related to any chronic medical condition and condition-specific statements (n=66 to 340) that related to one of the seven included chronic medical conditions. Sequential reduction steps were necessary to minimise the vast amount of collected statements for the development of the pilot instrument. These included redundancy scorings, importance scoring, item writing and card sorting. The steps are set out in ***chapter five***. After the applied redundancy steps 100 chronic generic items and 26 to 44 condition-specific items were retained for testing in the DISABKIDS pilot study.

It was found that the applied card sorting method was the most comprehensive and straightforward method to reduce the statements and divide the items into domain groups.

The development of the seven condition-specific modules is described in ***chapter six***. The DISABKIDS condition-specific modules were tested in a pilot study of 360 respondents, and subsequently in a field test of 1152 respondents in 7 European countries. The modules were developed through a step-by-step analysis process which included factor analysis, correlations and internal consistency. Results from the cognitive interviews and expert opinions were also considered. All seven final condition-specific modules consist of an 'Impact' domain and an additional domain (e.g. Worry, Stigma, Treatment) with between 10 to 12 items in total and a Cronbach's alpha varying from 0.71 to 0.90.

The questionnaire's validation results in the asthma population are presented in ***chapter seven***. The core chronic generic module (37 items, covering 6 domains) and the asthma specific module (11 items and 2 domains) were both tested in 7 countries within Europe on a total sample of 405 children and adolescents with asthma. The internal consistency for all the domains was between 0.66 and 0.85 with good retest reproducibility. The domain scores differentiated between asthma severity scores. The DISABKIDS HRQoL instrument is unique in being developed cross-nationally and in a modular structure. The psychometric properties of the DISABKIDS chronic generic and asthma-specific modules are sufficient for HRQoL assessment in children and adolescents with asthma.

The first results on the psychometric properties of the DISABKIDS chronic generic and asthma-specific modules are promising, however more work will still need to be done. The discussion in ***chapter eight*** presents remaining limitations, future research suggestions and possibilities regarding the implementation of individual HRQoL assessment in clinical practice. Overall the European DISABKIDS project has come a long way in the development of a new cross-national HRQoL instrument for children and adolescents with a chronic medical condition. The modular build up and cross-national development offer advantages for assessment on a national and international level in HRQoL evaluation studies and clinical studies. However, further evaluation of the DISABKIDS instrument is needed to test its performance as individual measure in clinical practice and prove its relevance to clinicians. In the future, the DISABKIDS instrument may serve as a useful tool to assess HRQoL in children and adolescents with a chronic condition to benefit the quality of their lives.

Samenvatting

De definitie van de Wereldgezondheidsorganisatie Kwaliteit van Leven (WHOQoL) groep definieert kwaliteit van leven (KvL) als: "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns". KvL is een steeds belangrijker concept geworden in zowel de maatschappij als in wetenschappelijk onderzoek. In de maatschappij en in de politiek is er aandacht voor het vraagstuk van de eerlijke verdeling van KvL in onze samenleving en de wereld. Daarnaast is het verbeteren van KvL een kernthema geworden voor mensen met een problematische gezondheid of een ziekte. Een van de redenen voor het belang van het bepalen van KvL zijn de aanwijzingen dat objectieve bepaling van de ernst van een ziekte slechts een zwak verband laat zien met het gevoel van welzijn van de patiënt. Van invloed is verder de verandering die is opgetreden als gevolg van medische ontwikkelingen in het beloop van bepaalde ziektebeelden van potentieel fataal naar chronisch, hetgeen heeft geleid tot een verschuiving van behandeling naar zorg. Vandaar de groeiende aandacht voor gezondheidsgerelateerde kwaliteit van leven ofwel 'health related quality of life' (HRQoL). Over het algemeen heeft onderzoek naar KvL als doel een bijdrage te leveren aan het waarborgen en verbeteren van fysieke en mentale gezondheid en welzijn. Dit heeft tot gevolg gehad dat er verscheidene vragenlijsten ontwikkeld zijn om de KvL van verschillende patiëntengroepen te bepalen en te beschrijven. Daarnaast wordt KvL steeds vaker opgenomen als afhankelijke variabele in klinische studies waarin verschillende behandelregimes worden vergeleken. Bestaande KvL vragenlijsten worden steeds verder verbeterd en nieuwe vragenlijsten worden voortdurend ontwikkeld en getest voor specifieke patiëntengroepen.

Dit proefschrift beschrijft het ontwikkelingsproces dat binnen het Europese DISABKIDS-project is gevolgd. Het project was een samenwerkingsverband van clinici en onderzoekers uit zeven Europese landen. Hun doel was om een nieuw Europees HRQoL instrument te ontwikkelen voor kinderen en adolescenten met een chronische ziekte. Het ***eerste hoofdstuk*** van dit proefschrift start met een schets van de algemene principes rondom HRQoL. Het beschrijft dat er een groeiende aandacht is voor het bepalen van KvL, maar ook dat het gebruik in de klinische praktijk nog beperkt is. Astma is een kernthema in dit proefschrift en daarom wordt in dit hoofdstuk ook aandacht besteed aan de psychische en lichamelijke effecten van astma op kinderen en adolescenten. We beschrijven tevens hoe vragenlijsten in het algemeen ontwikkeld worden in vergelijkbare stappen, die het definiëren van de doelgroep en het doel van de vragenlijst, itemselectie en validering omvatten. Het proces dat in het DISABKIDS-project gevolgd is omvat tevens het uitvoeren van focusgroepen, waarin patiënten konden aangeven op welke manier hun chronische ziekte hun dagelijks leven beïnvloedt en welke aspecten van KvL zij belangrijk vinden. Aan de focusgroepen hebben kinderen en adolescenten in drie leeftijdcategorieën (4-7, 8-12, 13-16) meegewerkt met de volgende chronische aandoeningen: astma, juveniele idiopathische artritis, atopische dermatitis, cerebrale parese, cystische fibrose, diabetes of epilepsie. De modulaire opzet in drie niveaus, het opnemen van zeven chronische aandoeningen, de brede leeftijdsgroep (4-16 jaar) en de beschikbaarheid van een kind- en ouderversie zijn aspecten die het DISABKIDS-instrument uniek maken.

Aangezien er weinig informatie beschikbaar is over de attitude van clinici ten opzichte van het gebruik van KvL bepaling ten behoeve van de patiëntenzorg, was het eerste doel meer te weten te komen over het perspectief van kinderartsen en arts-assistenten op de implementatie van KvL bepaling in de klinische praktijk. ***Hoofdstuk twee*** beschrijft een onderzoek uitgevoerd onder de leden van de Nederlandse Vereniging van Kinderartsen (NVK). Meer dan de helft van de 303 respondenten (57%) vonden dat het mogelijk is KvL vragenlijsten te gebruiken in de klinische praktijk. De meerderheid gaf aan dat de bepaling van KvL nuttig is en dat het in het bijzonder van belang is KvL te bepalen bij kinderen met een chronische ziekte (82%). Een minderheid (17%) gebruikte echter op dat moment al KvL vragenlijsten. De belemmeringen die het gebruik van KvL vragenlijsten in de weg staan zijn vooral: de extra tijd die nodig is voor de KvL bepaling, afwezigheid van gestandaardiseerde vragenlijsten en het ontbreken van voldoende kennis over KvL . Dit zijn aspecten die bij het ontwikkelen van een nieuw instrument overwogen moeten worden.

De patiënten werden gezien als de belangrijkste bron van informatie voor de basis van het instrumenten, vooral voor de ontwikkeling van de items. Om de relevante uitspraken over HRQol vanuit het perspectief van de patiënt te kunnen identificeren zijn er focusgroepen toegepast. Deze uitspraken dienden als basis voor de items in het DISABKIDS-instrument. Aan de focusgroepen hebben 154 kinderen en adolescenten met een chronische aandoening, 142 van hun familieleden en 26 zorgverleners meegedaan. In totaal zijn er 3515 uitspraken over HRQol geïdentificeerd. De focusgroep-methodiek moest voor het gebruik met kinderen en adolescenten aangepast worden. Een aantal kwesties dienen daarbij overwogen te worden, zoals cognitieve mogelijkheden, taalvaardigheid en concentratie. Daarnaast was het noodzakelijk om de methode over de verschillende deelnemende landen af te stemmen. Het ***derde hoofdstuk*** beschrijft de gebruikte methodologie van focusgroepen.

Astmafocusgroepen en interviews zijn in vier Europese landen uitgevoerd (Griekenland, Italië, Nederland en Zweden). Een overzicht van de besproken thema's en resultaten wordt gegeven in ***hoofdstuk vier***. In totaal namen 43 kinderen en adolescenten met astma, 33 familieleden en 7 zorgverleners deel aan de focusgroepen en interviews. De kinderen en adolescenten spraken vooral over de huidige beperkingen die zij ervoeren, zoals symptomen van astma, gerelateerde medische aspecten, fysieke beperkingen en sociale kwesties. Ouders richtten zich meer op de gevolgen van astma voor de toekomst van hun kinderen, zoals preventie, langetermijneffecten van de medicijnen en onderwijskansen. De focusgroepen bleken een bruikbaar instrument om uitspraken van kinderen en adolescenten met een chronische ziekte te genereren ten behoeve van de ontwikkeling van een vragenlijst.

De kwalitatieve data, verzameld via de DISABKIDS-focusgroepen en interviews in de verschillende landen, bestond uit 3515 HRQoL-uitspraken. Deze omvatten generieke KvL uitspraken (n=488) niet gerelateerd aan ziekte, chronisch generieke uitspraken (n=1647) gerelateerd aan chronische ziekten en ziektespecifieke uitspraken (n=66 tot 340) gerelateerd aan een van de zeven chronische ziekten die het project omvat. Een aantal opeenvolgende fasen van datareductie waren noodzakelijk om de grote hoeveelheid verzamelde uitspraken te minimaliseren voor gebruik in het pilot-instrument. De datareductie omvatte de identificatie van elkaar overlappende uitspraken, een score op belangrijkheid, het schrijven van items en een kaart-sorteermethode. De verschillende stappen zijn beschreven in ***hoofdstuk vijf***. Na de datareductie waren er 100 chronisch generieke items en 26 tot 44 ziektespecifieke items over om te testen in de DISABKIDS-pilotstudy. Uit dit proces kwam naar voren dat de gebruikte kaart-sorteermethode de meest volledige en minst gecompliceerde methode was voor het reduceren van de uitspraken en de verdeling van items in domeinen.

De ontwikkeling van de zeven ziektespecifieke modules staat beschreven in ***hoofdstuk zes***. De DISABKIDS ziektespecifieke modules zijn getest in een pilotstudy van 360 respondenten en vervolgens in een veldstudie van 1152 respondenten in de zeven Europese landen. De modules zijn ontwikkeld middels een stapsgewijs analyseproces, hetgeen factoranalyse, correlaties en interne consistentie omvatte. De resultaten van de interviews en de mening van experts zijn hierbij eveneens betrokken. De uiteindelijke zeven ziektespecifieke modules bevatten een ‘Impact’-domein and een additioneel domein (bijvoorbeeld ‘Worry’, ‘Stigma’, ‘Treatment’) met tussen de 10 en 12 items in totaal en een Cronbach’s alpha tussen de 0.71 en 0.90.

De resultaten van de validatie van het DISABKIDS-instrument in de astmapopulatie worden gepresenteerd in ***hoofdstuk zeven*** . De chronisch generieke module (37 items, verdeeld over 6 domeinen) en de astmaspecifieke module (11 items en 2 domeinen) zijn beide getest in zeven landen binnen Europa in een steekproef van 405 kinderen en adolescenten met astma. De interne consistentie voor alle domeinen was tussen de 0.66 en 0.85 met een goede test-hertestbetrouwbaarheid. De domeinscores differentieerden met de ernst van de astma. Het DISABKIDS-instrument voor het bepalen van de HRQoL is uniek door zijn crossnationale ontwikkeling en zijn modulaire structuur. De psychometrische eigenschappen van de DISABKIDS chronisch generieke en astmaspecifieke modules zijn voldoende voor het gebruik van het instrument voor de bepaling van HRQoL in kinderen en adolescenten met astma.

De eerste gegevens over de psychometrische eigenschappen van de DISABKIDS chronisch generieke en astmaspecifieke modules zijn veelbelovend. Er moet echter nog meer werk worden verricht. De discussie in ***hoofdstuk acht*** geeft een overzicht van de resterende beperkingen, suggesties voor toekomstig onderzoeken en mogelijkheden voor de implementatie van individuele HRQoL-vragenlijsten in de klinische praktijk.

Over het geheel genomen heeft het DISABKIDS-project veel bereikt in de ontwikkeling van een nieuw crossnationaal HRQoL-instrument voor kinderen en adolescenten met een chronische ziekte. De modulaire opbouw en de crossnationale ontwikkeling biedt voordelen voor het meten van HRQoL op een nationaal en internationaal niveau in beschrijvende en klinische studies. Het is echter van belang dat het DISABKIDS-instrument verder wordt onderzocht om te bepalen of het als individueel meetinstrument ook in de klinische praktijk te gebruiken is en zijn waarde kan bewijzen voor clinici. We verwachten dat het DISABKIDS-instrument in de toekomst een bruikbaar kan zijn voor het bepalen van de HRQoL van kinderen en adolescenten met een chronische ziekte en zo een bijdrage kan leveren aan de kwaliteit van hun leven.

Appendix

Appendix

The DISABKIDS chronic generic module

The DISABKIDS asthma specific module

Contact information

Chronic generic module

Questionnaire for children and adolescents

Hi,
We would like to ask you some questions about how you have been feeling during the past four weeks. These questions ask about some problems that children like you might have. We would like you to answer all the questions below.

Please
think back over the past four weeks when answering the questions and
choose the answer that fits you best and tick the appropriate box.

If you play with your friends 'very often' you would tick the box as shown in this example:

For example: __	never	seldom	quite often	very often	always
Do you play with your friends?	☐	☐	☐	☒	☐

There are no right or wrong answers. It's what you think that matters.

INDEPENDENCE

Please think back over the last 4 weeks…

		never	seldom	quite often	very often	always
1.	Are you confident about your future?	☐	☐	☐	☐	☐
2.	Do you enjoy your life?	☐	☐	☐	☐	☐
3.	Are you able to do everything you want to do even though you have a condition?	☐	☐	☐	☐	☐
4.	Do you feel like everyone else even though you have a condition?	☐	☐	☐	☐	☐
5.	Are you free to lead the life you want even though you have a condition?	☐	☐	☐	☐	☐
6.	Are you able to do things without your parents?	☐	☐	☐	☐	☐

PHYSICAL

Please think back over the last 4 weeks…

		never	seldom	quite often	very often	always
7.	Are you able to run and move as you like?	☐	☐	☐	☐	☐
8.	Do you feel tired because of your condition?	☐	☐	☐	☐	☐
9.	Is your life ruled by your condition?	☐	☐	☐	☐	☐
10.	Does it bother you that you have to explain to others what you can and can't do?	☐	☐	☐	☐	☐
11.	Is it difficult to sleep because of your condition?	☐	☐	☐	☐	☐
12.	Does your condition bother you when you play?	☐	☐	☐	☐	☐

EMOTION

Please think back over the last 4 weeks…

		never	seldom	quite often	very often	always
13.	Does your condition make you feel bad about yourself?	☐	☐	☐	☐	☐
14.	Are you unhappy because of your condition?	☐	☐	☐	☐	☐
15.	Do you worry about your condition?	☐	☐	☐	☐	☐
16.	Does your condition make you angry?	☐	☐	☐	☐	☐
17.	Do you have fears about the future because of your condition?	☐	☐	☐	☐	☐
18.	Does your condition get you down?	☐	☐	☐	☐	☐
19.	Does it bother you that your life has to be planned?	☐	☐	☐	☐	☐

SOCIAL EXCLUSION

Please think back over the last 4 weeks…

		never	seldom	quite often	very often	always
20.	Do you feel lonely because of your condition?	☐	☐	☐	☐	☐
21.	Do your teachers behave differently towards you than towards others?	☐	☐	☐	☐	☐
22.	Do you have problems concentrating at school because of your condition?	☐	☐	☐	☐	☐
23.	Do you feel that others have something against you?	☐	☐	☐	☐	☐
24.	Do you think that others stare at you?	☐	☐	☐	☐	☐
25.	Do you feel different from other children?	☐	☐	☐	☐	☐

SOCIAL INCLUSION

Please think back over the last 4 weeks…

		never	seldom	quite often	very often	always
26.	Do other kids understand your condition?	☐	☐	☐	☐	☐
27.	Do you go out with your friends?	☐	☐	☐	☐	☐
28.	Are you able to play or do things with other children/adolescents (e.g. sports)?	☐	☐	☐	☐	☐
29.	Do you think that you can do most things as well as other children?	☐	☐	☐	☐	☐
30.	Do your friends enjoy being with you?	☐	☐	☐	☐	☐
31.	Do you find it easy to talk about your condition to other people?	☐	☐	☐	☐	☐

MEDICAL TREATMENT

Do you take any medication or do you get treatment?
(e.g. pufs or sprays)

☐ yes, then please fill in the following questions

☐ no, you may go to the next page

Think about the past four weeks ….

		never	seldom	quite often	very often	always
32.	Does having to get help with medication from others bother you?	☐	☐	☐	☐	☐
33.	Is it annoying for you to have to remember your medication?	☐	☐	☐	☐	☐
34.	Are you worried about your medication?	☐	☐	☐	☐	☐
35.	Does taking medication bother you?	☐	☐	☐	☐	☐
36.	Do you hate taking your medicine?	☐	☐	☐	☐	☐
37.	Does taking medication disrupt everyday life?	☐	☐	☐	☐	☐

Asthma specific module

Now we would like to know some things about your asthma.

IMPACT

Please think back over the last 4 weeks…

		never	seldom	quite often	very often	always
1.	Do you feel that you get easily exhausted?	☐	☐	☐	☐	☐
2.	Does asthma bother you if you want to go out?	☐	☐	☐	☐	☐
3.	Are you unable to take part in certain sports?	☐	☐	☐	☐	☐
4.	Do you feel short of breath when you do sports?	☐	☐	☐	☐	☐
5.	Are you bothered by the amount of time you spend wheezing?	☐	☐	☐	☐	☐
6.	Do you feel terrible when you are out of breath?	☐	☐	☐	☐	☐

WORRY

Please think back over the last 4 weeks…

		never	seldom	quite often	very often	always
7.	Are you worried that you might have an asthma attack?	☐	☐	☐	☐	☐
8.	Do you worry that others do not know what to do if you have an attack?	☐	☐	☐	☐	☐
9.	Do you feel scared that you might have difficulty breathing?	☐	☐	☐	☐	☐
10.	Are you scared that you might have to go to the emergency ward?	☐	☐	☐	☐	☐
11.	Are you scared at night because of your asthma?	☐	☐	☐	☐	☐

Contact information

www.disabkids.de

Coordination centre
Monika Bullinger (bullinger@uke.uni-hamburg.de)
Silke Schmidt (sischmid@uke.uni-hamburg.de)
Corinna Petersen (copeters@uke.uni-hamburg.de)
Department of Medical Psychology
University Hospital of Hamburg-Eppendorf
Martinistr. 52 Pav. 69, 20246 Hamburg, Germany
Tel: +49-40-42803 6430

The Netherlands
Hendrik Koopman (H.M.Koopman@lumc.nl)
Rolanda Baars (R.M.Baars@lumc.nl)
Department of Paediatrics
Leiden University Medical Center, PO Box 9600
2300 RC Leiden, The Netherlands
Tel: +31-71-526 1666 / 2824

Germany
Ute Thyen (thyen@paedia.ukl.mu-luebeck.de)
Esther Müller-Godeffroy (mueller-g@paedia.ukl.mu-luebeck.de)
Department of Paediatrics
Medical University of Luebeck, Ratzeburger Allee 160
23538 Luebeck, Germany
Tel: +49-451-500 2615

United Kingdom
Mick Power (mjpower@staffmail.ed.ac.uk)
Peter Hoare (phoare@staffmail.ed.ac.uk)
Section of Clinical and Health Psychology
University of Edinburgh, Morningside Park
Edinburgh EH 10 5HF, United Kingdom
Tel: +44-131-537-6578/79

France
Marie Claude Siméoni (disabkids.phys-med-rehab@univie.ac.at)
Department of Public Health
University Hospital of Marseille, 27 BD. J. Moulin
13885 Marseille cedex 5, France
Tel: +33-49132-4474

Greece
John Tsanakas (tsanakas@spark.net.gr)
Athanasios Vidalis (vidalis@med.auth.gr)
Department of Psychiatry and Department of Paediatrics Respiratory Unit
Hippocratio Hospital, Konstantinoupoleos 49
Thessaloniki T.K. 54642, Greece
Tel: +30-31-838004

Sweden
John Eric Chaplin (john.chaplin@vgregion.se)
The Growth Centre (Växthuset),
Queen Silvia's Children's Hospital
SE-416 85 Gothenburg, Sweden
Tel: +46-31-343 5788

Austria
Michael Quittan (Michael.quittan@akh-wien.ac.at)
Othmar Schuhfried (Othmar.Schuhfried@univie.ac.at)
Department of Physical Medicine and Rehabilitation
University of Vienna, Währinger Gürtel 18-20
1090 Wien, Austria
Tel: +43-1-40400-5277

CV

Curriculum vitae

cv

Curriculum vitae

Rolanda Maia Baars was born on the 24th of September 1970 in Puttershoek, the Netherlands. Her child and teenage years were spent in different countries, including South Africa, Japan, Middle East, Malaysia and New Zealand. She passed her New Zealand high school certificate at Pompallier College, Whangarei in 1986 after which she returned to the Netherlands. There she continued her education within the Dutch school system and completed her VWO at the Andreas College in Drachten in 1990.

Her medical study started in 1990 at the University of Groningen. In 1994 she took up psychology at the same university to expand her area of interest and her knowledge of developmental psychology of the child and adolescent. Her final psychology research project was on ‘Quality of life and cystic fibrosis’. She received her medical degree on the 27th of May 1999 and her psychology degree a day later on the 28th of May 1999.

Her first clinical work experience was on the children's ward in ‘De Weezenlanden’ in Zwolle. In May 2001 she started her research work with the European DISABKIDS project at the University Medical Centre in Leiden. During the following four years she participated in this European consortium of which the main aim was to develop and test a European health related quality of life questionnaire for use with children and adolescents with a chronic condition. As DISABKIDS asthma consultant she concentrated most of her work around the collection and analyses of the asthma data.

Rolanda has now started her paediatric specialisation at the Juliana Children's Hospital in The Hague and the Leiden University Medical Centre.