Quality of Life Research (2005) 14: 1065–1077
DOI 10.1007/s11136-004-2575-z

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

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Accepted in revised form 26 August 2004

Abstract

Health-related quality of life (HRQOL) assessment in children and adolescents with chronic health conditions is increasingly considered as a relevant topic. The aim of the EU-funded DISABKIDS project is to develop, test, and implement European instruments for the assessment of HRQOL of children and adolescents with disabilities and their families. The current paper describes the development and pilot testing of a chronic generic HRQOL measure. Using literature searches, expert consulting and focus groups with children/adolescents and their families, items of the instruments were developed and translated into the respective languages. A pilot test with 360 children and adolescents was conducted. Children and adolescents (8–12, 13–16 years) with different chronic health conditions (asthma, epilepsy, diabetes, arthritis, atopic dermatitis, cerebral palsy, and cystic fibrosis) as well as their families were included. Data were analysed according to predefined psychometric and content criteria. Psychometric analyses resulted in a 56-item chronic generic HRQOL questionnaire with six domains ('Medication', 'Physical', 'Emotion', 'Independence', 'Social Inclusion', 'Social Exclusion') with acceptable internal consistency.

Key words: Children, Chronic condition, Health-related quality of life, Paediatric

Introduction

Advances in medical care have changed the focus of paediatric medicine from the treatment of infectious diseases to the management of chronic health conditions [1]. Because the prevalence of paediatric chronic health conditions is increasing, a significant proportion of children and adolescents is affected by chronic health conditions [2]. A child or adolescent with a chronic illness has to cope with psychological, social, and physical consequences related to having such conditions. The assessment of those consequences and their effect on the young peoples' health-related quality of life is a major task for medical research. With increasing criticism and growing acceptance of new health outcome parameters, such as health-

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relatedquality of life, the focus of health outcome measurement has shifted [3]. Health-related quality of life (HRQOL) is increasingly considered as an important health outcome parameter in medicine [4]. However, while theory and research on children with chronic health conditions and disabilities has grown in recent decades, adequate assessment methods for outcome measures still need to be provided. The EU-funded DISABKIDS project aims at the cross-national understanding of children's and adolescents' health-related quality of life by developing European instruments for HRQOL assessment from the perspective of children, adolescents, and their parents. More specifically, the DISABKIDS project aims at simultaneously developing a HRQOL chronic generic inventory as well as condition-specific questionnaire modules in seven European countries.

The conceptual background, overall objectives, and study outline of the DISABKIDS project have been described in detail by Bullinger et al. [5]. In sum, the DISABKIDS Group has defined HRQOL for their research work as a multidimensional construct with social, physical, emotional, and functional domains. Since childhood and adolescence show a rapid change in a variety of physical, emotional, and social aspects of the child's development, qualitative as well as quantitative changes in quality of life are expected. As a consequence, age differences in HRQOL domains have to be taken into account. One major objective of the study was to develop a reliable, valid, and sensitive measure to assess HRQOL across health conditions for different countries and for two age groups, 4–7 and 8–16 years. Approaches to develop such age appropriate versions can either be based on the assumption that there are qualitative transitions in HRQOL that require different domains in children and adolescents or that although some qualitative changes occur, it is important to provide an instrument to measure quantitative changes. The latter approach was taken, because it would enable the instruments to be employed in longitudinal studies.

The current paper focusses on the pilot testing and psychometric properties of the DISABKIDS chronic generic measure for children and adolescents aged 8–16 years. The initial developmental steps will be described briefly.

Development of the pilot form

Literature review

The first step involved an international literature review. The review aimed at identifying existing HRQOL questionnaires. Altogether 8233 articles were identified. All abstracts were evaluated by the collaborating centres according to predefined criteria. After a first round of evaluation, 19% of the studies were found to be relevant for the project. Of these, only a few studies (12%) had a research aim concerning HRQOL and described HRQOL instruments. In 34% of these studies, HRQOL was considered as a main aspect.

Focus groups

Focus groups were conducted with children and adolescents as well as their parents and caregivers. A focus group manual delivered a general outline of the procedure. The participating children and adolescents were stratified by age (4–7, 8–12, 13–16 years), severity of the disease as rated by a clinician (mild, moderate, severe), and by the type of disease (asthma, arthritis, epilepsy, cerebral palsy, diabetes mellitus, atopic dermatitis, or cystic fibrosis). A letter and a patient information sheet as well as a consent form were sent to the participants. Individual interviews instead of focus groups were conducted as a second option. A total of $n = 154$ children/adolescents, $n = 142$ parents, and $n = 26$ experts took part in focus groups or interviews (for the age group 4–7 years). The focus groups were designed as discussion groups. At the beginning, questions were asked about how the children and adolescents view their condition and how they cope with it. These questions were only meant to prompt the discussion.

Each centre filled out a focus group documentation sheet. The sheet contained three sections in which statements should be grouped: (a) generic, (b) chronic generic, and (c) condition-specific. The items were translated into English and were defined as generic, if they did not refer to any condition. They were defined as chronic generic if they pertained to being ill in an unspecific sense and as condition-specific if the covered aspects related specifically to a health condition (e.g.,

being afraid of seizures). In sum, 1647 chronic generic statements were derived from the focus group work.

Item generation

The objective was to generate items from the focus-statement-pool. A multistage process including rating items for redundancy, item writing, card sorting, and rereading was performed. The rating was conducted by an expert panel of researchers in three countries (The Netherlands, Germany, and UK) forming the item revision group. Statements were omitted if at least two countries indicated that it was repeated in the statement pool, semantically equivalent, or related to other constructs. After the reduction process, 307 items were included in a card sorting procedure. Therefore, one researcher per country was present in order to represent the different languages. The items were printed out and affixed on pieces of card. These cards were separated into three different dimensions of HRQOL (psychological, social, and physical) piles. Items that shared a common feature, e.g. they belonged to the physical domain, were put on the same pile. Once all the items had been sorted, a list of the categories (facets) within the dimensions according to the content of the items was created. Finally, 100 items were selected which represented 19 facets of HRQOL (Future, Perceived Impact, Self-Confidence, Emotion, Autonomy, Limitation, General Impact, Sleep, Overall Health Perception, Treatment, Medication, School, Acceptance, Stigma, Activities, Family Support, Differences, Contact, Family Functioning).

Translation

The HRQOL items were forward and backward translated. Statements generated by the focus groups and interviews were translated into English and re-translated after the item development process into the original language. First two independent translators translated the English pilot draft version into the target language (Dutch, French, German, Greek, or Swedish). The forward translators decided upon a reconciled forward translation. A bilingual speaker performed the backward translation into English. The backward translation was then compared with the pilot draft, generating the respective final forward translation. The international harmonisation took place during a project meeting with all DISABKIDS participants and served to ensure cross-national equivalence of items, i.e. that items were interpreted in the same way across countries. The equivalence of each item was examined (by comparing it with the English original and across the different languages). In addition, children and adolescents were involved in the assessment of the translation during the procedure of cognitive debriefing, where the clarity of the translated item was examined.

The HRQOL chronic generic pilot form

The 100 items of the pilot form represented 19 facets and five domains ('Psychological', 'Social', 'Physical', 'Overall Health Perception', and 'Medical') of HRQOL. The items were expressed as questions in the present tense. For the pilot version a six-point Likert response scale was utilised (1 = 'never', 2 = 'seldom', 3 = 'quite often', 4 = 'very often', 5 = 'always', 6 = 'not applicable'). The time frame referred to the past 4 weeks.

Pilot testing

The piloting and psychometric testing of the newly developed chronic generic module had several aims:

- to determine the item and scale characteristics,
- to analyse open questions with regard to relevance and difficulty of items,
- to explore the scale structure with exploratory, confirmatory factor and Rasch analyses, and finally,
- to select the best items for a field test version of the measure.

Methods

Design of the pilot test
The pilot study had a cross sectional design in each participating centre. An agreed-upon standardised pilot test manual was followed in the centres.

Sample
The preliminary version of the chronic generic HRQOL questionnaire was given to children and

adolescents treated in a participating centre in seven different countries (Austria, France, Germany, Greece, the Netherlands, Sweden, and United Kingdom). The inclusion criteria were

- a chronic health condition (asthma, arthritis, epilepsy, cerebral palsy, diabetes mellitus, atopic dermatitis, or cystic fibrosis) diagnosed according to international classification systems (ICD-10) by a physician,
- available consent form,
- age between 8 and 16 years,
- the ability to understand questions, articulate thoughts, and maintain a conversation.

With regard to the criterion of having a chronic health condition, the applied definition is in concordance with the definition as an illness that can last for an extended period, at least 3 months, often for life, and cannot be cured [6, 7]. Per participating centre and per condition it was planned to include as a minimum 12 families in the pilot test. In each centre at least two conditions had to be included in the pilot test. It was obligatory to test children with asthma in order to be able to compare one condition across all countries.

Procedure

Possible participants for the DISABKIDS study were contacted in advance with an information letter that included consent forms. Others were contacted during visits to specialist clinics. Researchers from each participating centre conducted the pilot testing. The pilot test procedure contained three parts. Part (A) involved filling out the questionnaire, part (B) a cognitive interview with children and adolescents about relevance, difficulty, and adequacy of items, and part (C) involved take home questionnaires. Part (B) was only conducted with a sub-set of questions with all participants- the sub-set being rotated so that all questions were covered in the cognitive debriefing.

Instruments

The children's questionnaire contained seven sociodemographic questions about gender, age, date of birth, number of siblings, years of schooling, class/ grade, and the type of school. In addition, the KINDL [8] and 10 items from the Child Health Questionnaire [9] were included. The next part of the questionnaire contained the newly developed 100 chronic generic HRQOL items. The condition-specific modules contained 38 items for cystic fibrosis, 36 items for atopic dermatitis, 28 items for diabetes, 32 items for asthma, 27 items for epilepsy, 44 items for arthritis, and 26 items for cerebral palsy. Information and data about the condition-specific modules will be presented in a subsequent publication.

The parents questionnaire started with 16 socio-economic status variables about the relationship to the child, age, date of birth, number of persons living in the household, type of school, profession, country, language, and current economic situation. Generic clinical variables about child age at onset of disease, diagnosis and treatment start, co-morbidity, development of the child, school absence, physical, social, emotional or behavioural problems were included. The clinical variables were followed by questions concerning the health status of the child/adolescent assessed by the FS-II-R. The FS-II-R [10] is a parental-report measure to assess behavioural manifestations of illness that interfere with a child's performance of age-appropriate activities. Subsequently, the DISABKIDS items were assessed as a parent proxy-report. Finally, for caregivers of the older age group the Children with Special Health Care Needs screener – CSHCN [11] was included which is a parent self-administered set of questions to identify children with special or chronic health care needs.

The medical documentation contained different sets of clinical variables with regard to the respective disease.

Data analyses

Data analyses for the pilot test were carried out using the SPSS (Windows), the Multitrait Analysis Programme for scale structure testing [12], EQS [13], and WINMIRA [14].

If out of range values or implausible values were entered into the database by any of the centres, they were recoded as missing values (0.4%). If two answers were coded, one answer was randomly picked (2.3%). The answer category 'not applicable' of the chronic generic items of the HRQOL module was treated as a missing value for scale calculations.

Classical multi scaling methods were applied at the item as well as at the scale level. Descriptive

item characteristics statistics including range, means, percentage of missing items, skew, and standard deviations of each item were calculated. χ^2, Fisher Exact and Mann–Whitney tests were used to explore differences between groups (age and gender). Item-scale correlations were calculated using the Pearson coefficient.

Descriptive scale characteristics statistics including range, means, and standard deviations of the scales were calculated. The item endorsement rates were analysed and items that demonstrated floor or ceiling effects identified. The reliability of the HRQOL facets was estimated using Cronbach's α coefficient (internal consistency). Scale intercorrelations were examined. The dimensionality of the chronic generic questionnaire was explored with exploratory (principal component analysis) and confirmatory factor analyses. In order to facilitate the interpretability of the exploratory factors each component matrix was rotated using the varimax procedure with the Kaiser normalisation method.

Answers for open-ended questions were reviewed for commonly occurring themes. Results concerning difficulty with understanding an item and clarity of the answer choices were examined.

Results were used to decide on retention, modification, or rejection of items using the following criteria:

Identification of item candidates for deletion

–Missing values: The percentage of missing items was treated as an estimation for the acceptance as well as for the feasibility of items. More than 5% of missing values were regarded as a first hint to delete an item.
–Item total correlation and changes in α: Each item in a hypothesised scale ought to correlate substantially with the construct measured. A low item-total-correlation (< 0.30) and a decrease of the α coefficient if the items was deleted were marked as an argument against keeping the item.
–Not applicable answers: If the percentage of non-applicable answers was high, this information was used as an indication for deletion. More than 5% of not applicable answer were regarded as a first hint to delete an item.
–Expert consensus: If the majority of a selected group of experts (including one representative per country) consented to omit or keep a certain item, this decision was accepted.

Examination of the scale structure

–Confirmatory Factor Analyses were used in a limited way together with the exploratory factor analyses in order to aid item selection rather than to test the overall scale structure.
Rasch analyses were employed in order to test whether the Rasch model applies to the dimensions identified by factor analyses and to identify over- and underdiscriminating items. The criterion for poorly fitting items was the item-Q index by Rost and von Davier [15]. Rasch analyses were not employed to test whether the items fitted the Rasch model criteria, but rather to give further evidence on the dimensions identified by factor as well as confirmatory factor analyses and to indicate which items were and which items were not consistent with a single underlying latent trait.
For the revised versions of the questionnaires, reliability (Cronbach's α), floor- and ceiling effects and scale fit values were reassessed.

Results

Description of the sample

The sample was composed of 360 children or adolescents and 345 parents (mostly mothers) or other caregivers. In accordance with the study plan, a large proportion of the sample (37%) had the diagnosis of asthma. Approximately equal numbers of the children were boys or girls. With regard to the parents' ratings, 83% of the children and adolescents had developed normally. The range of siblings was 0–4. 26% of the children and adolescents were tested in Germany (Table 1 gives the demographic and medical profile for the total sample).

Across the study partners the cognitive debriefing was performed for the items of the Psychological domain by 49 children and adolescents. 60 children and adolescents were cognitively debriefed for the Medical, Physical, and Overall Health Perception domain. 51 children and adolescents answered the questions with regard to the Social domain.

Table 1. Demographic and medical characteristics of the children/adolescents (n = 360)

Characteristic	N	%
Main diagnosis		
Arthritis	54	15.0
Asthma	132	36.7
Atopic dermatitis	29	8.1
Cystic fibrosis	28	7.8
Cerebral palsy	21	5.8
Diabetes mellitus	59	16.4
Epilepsy	37	10.3
Sex		
Female	171	47.5
Co-morbidity	93	28.7
Relatives with the same condition	99	30.5
Child mental development (parent rating)		
Normal	268	82.5
Slow	46	14.2
Retarded	11	3.4
	Range	*M* (SD)
Age	6–19	12.48 (2.55)
Child age at diagnosis	0–17	4.78 (3.98)
Years of schooling	1–13	6.80 (2.68)
Days absent from school/kindergarten (during the previous year)	0–150	12.06 (24.73)

Item total correlation and item characteristic

20 items were identified as candidates for deletion because of their poor item total correlation. Two items showed a poor item total correlation but were kept because of its clinical importance. Overall, the item distribution was skewed. The respondents predominantly scored at the ceiling.

With regard to the remaining items (without the 20 items with a low item-total-correlation), five items showed a high percentage of 'not applicable', responses and did not contribute to the scale consistency. Five items of the medical scale (items 57–68) had a high rate of 'not applicable', answers indicating the need to assess this scale in a different way (e.g., with a filter question). Eight items showed a high percentage of missing values.

Significant gender effects were found for ten items. Age effects were found for 30 items. The age effects were especially dominant in the social dimension. The gender effects were equally distributed across the dimensions. Table 2 gives an overview of the item characteristics.

Cognitive debriefing

Children and adolescents reported increased difficulties in understanding 32 items. The items that the children and adolescents rated as 'not relevant' were mostly items either with negative content (e.g., 'Are you the target of jokes?') or were about medication.

Scale characteristics

Minor ceiling effects were noted for the social domain. The internal consistency reliabilities (Cronbach's α coefficients) for the domains ranged from 0.45 to 0.89 (see Table 3).

The principal component analyses followed by varimax rotation revealed 27 factors with eigenvalues ranging from 21.73 to 1.02, accounting for 84.78% of the variance. Nine factors included more than three items loading ≥ 0.40. The first factor extracted referred to feelings and emotional states. The second factor extracted referred to social integration. The third factor referred to physical issues. The fourth factor was mainly about medication. The fifths factor referred to independence. The sixths factor was about the future. Altogether the six factors accounted for 44.48% of the variance.

Confirmatory factor analyses (CFA)

Separate CFAs were run with the 80 items (without 20 items with a low item-total-correlation and smiley items). The CFAs were run separately for the revised Psychological (or Emotional), Physical (primarily relating to Treatment), Medical, and Social domains in order to test the degree of fit for single factor structures for each of these domains and in order to assess item loadings on each of these domains. Values of the Comparative Fit Index (CFI) and the Non-Normed Fit Index (NNFI) range from 0 to 1 and are generally considered acceptable at levels above 0.90; the average of the standardized covariance residual matrix, the so-called RMSEA, are considered acceptable at values less than 0.05 [16]. The CFA for the revised 28-item

Table 2. Descriptive statistic for the chronic generic item pool (n = 360)

Item	*M* (1–5)	SD	Not appl. %	Missing %	Skewness	α	Corr.	Age dif	Gender dif	1	5
1. Do you have fears about the future because of your condition?*	4.11	1.00	5.8	2.8	0.92		0.44				
2. Are you confident about your future?	3.94	1.09	3.3	4.4	−0.95		0.33				
3. Do you wish your illness would go away?*	1.77	1.17	2.5	3.1	−1.34	⇧	0.15	++		•	
4. Do you feel that you will get better?	3.56	1.29	4.7	4.2	−0.62		0.22				
5. Do you feel lonely because of your condition?*	4.48	0.88	5.0	2.2	1.79		0.58				•
6. Do you enjoy your life?	4.45	0.79	1.4	1.9	−1.77		0.38				•
7. Do you feel under pressure because of your condition?*	4.11	1.08	3.1	3.6	1.05		0.62				
8. Does your condition get you down?*	4.02	1.15	3.6	3.3	1.08		0.64				
9. Does your condition restrict your life?*	3.88	1.23	2.5	4.2	0.93		0.56	++			
10. Do you forget your condition when you do certain things (e.g., when meeting friends)?	3.89	1.30	1.7	3.9	−0.98	⇧	0.19				
11. Do you have less free time because of your condition?*	4.07	1.22	5.0	3.1	1.14		0.40	+			•
12. Does it bother you that your life has to be planned?*	3.68	1.33	11.1	4.2	0.71		0.49				
13. Are you able to do everything you want to do even though you are ill?	3.80	1.21	3.9	3.6	−0.79		0.45				
14. Does your condition make you feel bad about yourself?*	4.19	1.05	3.6	3.3	1.29		0.21				•
15. Has your illness made you feel confident about yourself?	2.80	1.35	11.7	4.7	0.14		0.38				
16. Do you feel like everyone else even though you are ill?	4.12	1.20	3.3	3.6	−1.28		0.31				•
17. Has your condition made you more grown up than other children your age?	2.40	1.37	11.1	3.9	0.52		0.21				
18. Has your illness made you stand up for yourself?	2.96	1.40	9.2	3.9	0		0.38				
19. Are you shy because of your condition?*	3.80	0.61	5.6	3.6	1.98	⇧	0.11				•
20. Are you unhappy because our are ill?*	4.10	1.14	3.3	2.5	1.20		0.58		+		
21. Do you worry about your condition?*	3.82	1.15	2.8	2.8	0.87		0.60				
22. Do you have fun in spite of your condition?	4.42	1.00	1.1	2.8	−2.06	⇧	0.22				•
23. Does your condition make you angry?*	3.89	1.27	1.9	2.8	0.93		0.71				
24. Do you hate having your condition?*	2.93	1.49	3.3	2.5	0		0.56				
25. Do you think it is unfair that you are ill?*	3.54	1.48	5.3	2.8	0.56		0.56		+		
26. Do you feel nervous because of your condition?*	4.27	1.08	4.2	2.5	1.50		0.59				•
27. Do you feel embarrassed that you have an illness?*	4.35	0.97	4.4	1.7	1.40		0.63				•
28. Are you ashamed that you have an illness?*	4.65	0.86	4.7	2.8	2.92		0.48				•
29. Does your condition make you moody?*	4.00	1.07	6.4	3.9	0.85		0.58				
30. Do you hate having to depend on other people because.of your condition?*	3.81	1.23	13.6	3.3	0.79	⇧	0.06	+	+		
31. Are you free to lead the life you want even though you are ill?	3.82	1.31	3.3	3.1	−0.91		0.39				
32. Do you feel independent in managing your condition?	3.38	1.40	6.4	5.0	−0.41		0.21	++			
33. Are you able to do things without your parents?	4.10	1.11	0.3	3.9	−1.31		0.30	++			

Table 2. Continued

Item	*M* (1–5)	SD	Not appl. %	Missing %	Skewness	α	Corr.	Age dif	Gender dif	1	5
34. Are you able to run and move, as you like?	4.13	1.17	1.7	2.5	−1.27		0.52				
35. Are you limited in physical activities i.e. sports, biking, running?*	3.58	1.44	5.0	3.1	0.58		0.44				
36. Do you feel tired because of your condition?*	3.98	1.13	5.0	2.8	0.99		0.50	+			
37. Are you able to live with your condition the way it is?	4.25	1.03	2.8	3.9	−1.52		0.37				•
38. Is your life ruled by your condition?*	3.94	1.14	5.0	4.4	0.79		0.50	+			
39. Does it bother you that you have to explain to others what you can and can't do?*	3.49	1.42	6.4	3.9	0.46		0.51				
40. Do you have bad dreams or nightmares because of your condition?*	4.73	0.67	5.3	2.8	2.79		0.27	+			•
41. Is it difficult to sleep because of your condition?*	4.38	0.96	4.2	3.1	1.64		0.27				•
42. Is it okay for you to live with your condition?	3.76	1.41	2.5	4.2	−0.86	⇧	0.14				
43. Do you feel that everyone is healthy apart: from you?*	4.26	1.08	3.1	4.2	−1.50		0.19				•
44. Do you worry more than your friends about staying healthy?*	3.65	1.32	6.1	5.3	−0.66		0.25				
45. Is it a problem for you to go to doctor?*	4.10	1.18	1.9	3.6	1.30		0.23				•
46. Do you have enough time for yourself in spite of the treatment?	4.32	0.93	2.5	4.2	−1.55		0.23				•
47. Are you bothered by others watching you take your medicine?*	4.18	1.17	10.3	3.1	1.38		0.37				•
48. Are you bothered by the side effects of the medicine?*	4.04	1.22	17.5	4.4	1.23		0.39				
49. Has your schoolwork suffered because you have been on medication?*	4.42	1.05	12.2	3.6	1.88		0.27				•
50. Does having to get help with medication from others bother you?*	4.19	1.12	23.9	1.7	1.46		0.39				•
51. Are you worried that you will forget your medicine?*	3.24	1.77	9.4	3.1	0.20	⇧	0.11				
52. Is it annoying for you to have to remember your medication?*	3.44	1.45	10.6	3.6	0.46		0.48				
53. Are you worried about your medication?*	4.22	1.04	9.7	4.2	1.32		0.40				•
54. Do you accept that you need medication?	3.77	1.54	8.9	3.6	−0.87	⇧	0.11				•
55. Does taking medication bother you?*	3.60	1.49	8.1	5.3	0.63		0.54				
56. Do you hate taking your medicine?*	3.60	1.48	9.2	4.2	0.65		0.53				
57. Does taking medication disrupt everybody life?*	4.32	1.05	10.3	4.4	1.65		0.52				•
58. Do your teachers behave differently towards you than towards others?*	4.25	1.06	6.7	3.9	1.38		0.37				•
59. Are your teachers understanding your condition?	3.75	1.35	11.1	3.9	−0.79	⇧	0.15				
60. Do you have problems concentrating at school because of your illness?*	4.23	1.10	5.6	4.2	1.36		0.45				•
61. Do you have difficulties with keeping up with the course?*	4.30	1.09	4.7	3.6	1.52		0.55	+			•
62. Are your friends protective of you?	3.35	1.35	10.0	4.7	−0.31		0.52				
63. Are your friends supportive?	3.90	1.18	6.7	3.3	−0.84		0.56		+		
64. Do your friends accept you the way you are?	4.66	0.79	1.9	4.7	−2.70		0.32	+			•
65. Are others considerate to you?	3.86	1.20	6.7	5.6	−0.96		.41				
66. Do other kids understand your illness?	3.64	1.22	8.1	4.7	−0.63		0.40	+			

Table 2. Continued

Item	*M* (1–5)	SD	Not appl. %	Missing %	Skewness	α	Corr.	Age dif	Gender dif	1	5
67. Do you feel that others have something against you?	4.37	0.84	2.8	5.0	1.24		0.48				•
68. Do you think that others stare at you?*	4.42	0.89	3.9	4.4	1.64		0.48				•
69. Do you like it when people look at you?*	2.19	1.24	9.7	4.7	0.84	⇧	0.12	+			
70. Are you the target of jokes?*	4.26	1.08	4.7	5.0	1.56		0.38	+			•
71. Are you upset by other children teasing you?*	3.41	1.47	11.4	4.7	0.46		0.51				
72. Are you bothered by other people talking about you?*	3.52	1.38	6.4	5.3	0.61		0.47		+		
73. Do you feel excluded?*	4.40	0.95	3.9	4.4	1.77		0.61				•
74. Do you sleep over at a friend's house?	2.71	1.18	0.8	3.1	0.05		0.22	+			
75. Do you go out with your friends?	3.28	1.33	2.5	3.3	−0.31		0.32	+			
76. Are you able to play with other children?	4.49	0.85	3.1	3.6	−1.87		0.47				•
77. Do you take part in school sports despite having your condition?	4.40	1.10	1.9	3.6	−1.91		0.28				•
78. Does your condition bother you when your play?*	3.97	1.20	3.9	3.3	0.93		0.25	+			
79. Do your parents argue over things to do with your condition?*	4.59	0.81	8.6	5.0	2.38		0.30	+			•
80. Does your family bother you?*	4.39	0.88	4.4	3.9	1.47		0.17	+			•
81. Do your parents stop you from doing some things because of your condition?*	3.98	1.11	5.0	4.2	0.97		0.18				
82. Do others in your family have complaints about your condition?*	4.74	0.59	7.5	4.2	2.66		0.31				•
83. Do you get everything you want because of your illness?	4.02	1.19	7.5	5.0	1.14	⇧	0.03	+	+		
84. Do your parents support you in your treatment?	4.36	1.04	3.9	5.0	−1.70	⇧	0.10				•
85. Do you think that you can do most things as well as other children?	4.40	0.92	1.9	6.1	1.68		0.61				
86. Are you one of the group?	4.18	1.04	1.9	5.0	−1.31		0.41				
87. Do you feel different from other children?	4.22	1.12	3.9	6.1	−1.54	⇧	0.37				
88. Do you feel left out of things?*	4.14	1.05	3.1	6.1	1.19		0.53	+			
89. Do you worry that you will have problems finding a friend because of your condition?*	4.46	0.97	6.1	5.6	1.90		0.53	++			•
90. Do you get enough attention from other people?	3.69	1.16	3.3	5.3	−0.74		0.36	+			
91. Do your friends enjoy being with you?	4.41	0.77	2.5	5.0	−1.34		0.32				•
92. Is it difficult for you to make friends because of your condition?*	4.61	0.84	5.0	5.3	2.41	⇧	0.07	+			•
93. Dou you like being with other children with the same condition?	3.30	1.31	20.6	6.7	−0.19	⇧	0.15				
94. Do you find it easy to talk about your illness to other people?	3.30	1.42	4.7	5.3	−0.33		0.36				
95. Does your mother/father make too much of a fuss about you?*	3.91	1.17	5.0	6.4	0.91		0.30	+			
96. Does your condition affect the fami1y?*	4.04	1.16	8.6	4.7	1.05		0.19	++			
97. Do you think that you are a worry to your parents because of your condition?*	3.79	1.31	5.0	5.3	0.81		0.12	+			
98. Do your parents encourage you?	4.14	1.14	4.7	5.3	−1.34		0.14				•

Table 2. Continued

Item	*M* (1–5)	SD	Not appl. %	Missing %	Skewness	α	Corr.	Age dif	Gender dif	1	5
99. Are your brothers/ sisters nice to you when you are ill?	3.96	1.20	15.0	6.7	−1.03		0.20				
100. Do your parents talk to you about your condition?	3.17	1.23	4.2	5.3	−0.05	⇧	0.06	+			

⇧ = Alpha increases if item will be deleted of that facet.
+ = $p \leq 0.05$.
++ = $p \leq 0.001$.
• = ≥ 50% of the answers in answer category '1' or '5'.
* = Reversed item.

Table 3. Descriptive statistics and reliabilities: domain level

Domain	No. of items	*M*	Range	SD	Floor (%)	Ceiling (%)	α	Scale fit*
Medical	15	57.88	15–75	9.57	0.0	0.0	0.81	86.7
Overall Health	4	14.18	4–20	3.04	0.3	2.3	0.45	25.0
Physical	11	44.17	11–55	6.94	0.0	1.7	0.81	84.1
Psychological	38	146.35	38–190	20.09	0.0	0.0	0.91	88.8
Social	51	206.07	51–255	20.88	0.0	0.0	0.89	91.2

*Percentage of items that correlate higher with their own than with another scale.

Psychological Domain gave fit values for a one factor structure of RMSEA = 0.0314, CFI = 0.947, NNFI = 0.943, $\chi^2 = 1299.76$, df = 350, $p < 0.001$. The 7-item revised Physical Domain gave values of RMSEA = 0.0311, CFI = 0.970, NNFI = 0.955, $\chi^2 = 117.20$, df = 14, $p < 0.001$. The 11-item revised Medical Domain gave values of RMSEA = 0.0073, CFI = 0.982, NNFI = 0.978, $\chi^2 = 185.33$, df = 44, $p < 0.001$. The 34-item revised Social Domain gave values of RMSEA = 0.0392, CFI = 0.921, NNFI = 0.916, $\chi^2 = 2138.71$, df = 527, $p < 0.001$.

Rasch analyses

Based on the poorest fit indicated by high Q-indexes and high *z*-values in both the subscale as well as the domain analyses the Rasch analyses supported the elimination of four items from the Emotional domain and 2 items from the Social domain. These items showed Q-indices higher than 0.20 and *z*-scores higher than 3.0. In summary, therefore, the separate CFAs for each domain gave general support for each one being described by a single underlying latent trait because of the acceptable levels of the fit indices, though in combination with the exploratory factor analyses and the Rasch analyses a number of items were identified that made poor contributions to these latent traits.

Expert consensus

At a meeting with eight members of the DISABKIDS group from Germany, the Netherlands, Sweden, and the United Kingdom the candidates for deletion were discussed. The experts, a group of clinicians and statisticians, reviewed the remaining items and decided if any item should be kept or omitted because it may not provide any important clinical information. A further deletion of five items through expert consensus was agreed upon.

In sum, 44 items (Likert scale) were deleted because of a combination of the following reasons: a low item-total-correlation, a high percentage of

Table 4. Descriptive statistics and reliabilities

Facet	No. of items	*M*	Range	SD	Floor (%)	Ceiling (%)	α	Scale fit
Emotion	12	46.81	12–60	9.54	0.6	0.3	0.90	100.0
Independence	7	27.69	7–35	4.94	0	3.9	0.73	97.1
Physical	6	23.58	6–30	5.04	0	13.3	0.79	90.0
Social inclusion	9	36.21	9–45	5.42	0	3.0	0.71	97.8
Social exclusion	13	55.10	13–65	8.24	0	4.2	0.87	98.5
Medication	9	35.90	9–45	7.21	0.3	8.2	0.83	100.0

not applicable answer, a high percentage of missing values, bad fit in the Rasch analyses, and rejection by expert consensus.

Final version of the HRQOL chronic generic module

The number of items per facet ranged from 6 to 13. The reliability coefficients (internal consistency) of the final facets ranged from 0.71 to 0.90 and the scale fit values from 90% to 100% (see Table 4). Some ceiling effects were detected for the 'Physical' scale. The scale fit reached 100% for the 'Medication' and 'Emotion' domains. Item examples of the final version with 56 items are shown in Table 5.

Table 5. Item examples of the final chronic generic module (example for asthma)

Independence
Are you confident about your future?
Are you able to do things without your parents?
Physical
Are you able to run and move as you like?
Are you limited in physical activities, i.e. sports, biking, running?*
Emotion
Are you shy because of your asthma?*
Do you worry about your asthma?*
Social exclusion
Do you feel that others have something against you?*
Do you feel excluded because of your asthma?*
Social inclusion
Are your friends supportive?
Do you go out with your friends?
Medication
Are you bothered by the side effects of the medicine?*
Are you worried about your medication?*

*Reversed item.

Discussion

The current study has attempted to develop a pilot version of a chronic generic HRQOL questionnaire for children and adolescents with different chronic health conditions and in different European countries. The developmental steps have included focus group work, item development, translation, pilot test, and analyses. A standardised procedure has been carried out in seven European countries. Through the combination of various methods it has been possible to provide an international measures which needs to be further tested and validated in a field study. In conclusion, the stepwise approach to questionnaire development resulted in a 56-item chronic generic HRQOL measure. As the psychometric analysis of a newly developed measure can be viewed as an iterative process, specific values used as cut off points were not easy to define, especially since different methods were applied in parallel. The employed psychometric criteria proved to be a feasible approach to questionnaire development.

The perspective of children and adolescents was included especially at the beginning of questionnaire development. The statements of children and adolescents were the basis of questionnaire development. Focus group work provided a comprehensive starting point for the development and proved to be a useful procedure in prior studies [17]. It allowed us to use a bottom-up approach for developing the facets and the items of both questionnaires starting with the views of children and adolescents.

A crucial point for questionnaire development is that children may assign a different meaning to a wording an initially intended by the developers. This can lead to a misunderstanding of which the parties involved are unaware. In order to assess

potential misinterpretations, a cognitive debriefing was conducted during the pilot test. This technique is increasingly being used in questionnaire development [18]. Conducting cognitive interviews with children and adolescents is a special challenge because of their developmental ability and motivation to provide information. However, research showed that children and adolescents are able to handle the demands of a cognitive interview and provide important information. The results of the current study support this point of view. To gain better understanding of concepts of respondents, these techniques are a helpful method. Nevertheless, the amount of time necessary to carry this out and analyse it is a weakness of this approach.

Another important aspect for questionnaire development in children and adolescents are age-related differences. In the pilot test analyses, age-related differences were found for several items. For future developmental steps of the measure these differences will be further examined. For example, the DISABKIDS measure contains several questions pertaining to the future or relationship with the opposite gender which might be assessed only in adolescents.

Although the results of the current study are promising, there are several limitations which need to mentioned. The number of patients across health conditions is, with regard to the different types of health conditions, rather small. Therefore, psychometric data have to be carefully interpreted. In addition, the study meant to reflect a European approach, however, only seven countries were involved in the development and future studies are needed to test the applicability also in other countries.

The simultaneous approach of this study has been applied for questionnaire development only in the adult area so far. Although the simultaneous approach is a complex method for questionnaire development, it certainly improved the content of the measure. The derived dimensions are comparable to the dimensions of other HRQOL questionnaires, for example with the KINDL [8]. Nevertheless, the new measure emphases the impact of a certain chronic health condition and is not applicable for healthy children. The cross-cultural value and validity of the instrument will be clear after the international field testing. The clinical relevance of the measure, however, will be apparent in future applications in epidemiological surveys, descriptive cross sectional studies, longitudinal studies as well as clinical trials.

Acknowledgement

The DISABKIDS study is funded by the European Commission (QLG5-CT-2000-00716).

References

1. Eiser C, Morse R. A review of measures of quality of life for children with chronic illness. Arch Dis Child 2001; 84: 205–211.
2. Perrin JM, Shonkoff JP. Developmental disabilities and chronic illness: An overview. In: Behrman RE, Kliegman RM, Jenson HB (eds), Textbook of Pediatrics, Philadelphia: W. B. Saunders, 2000; 121–129.
3. Eiser C, Morse R. Quality-of-life measures in chronic diseases of childhood. Health Technol Assess 2001; 5: 1–157.
4. Bowling A. Measuring Disease: A Review of Disease-specific Quality of Life Measurement scales, Philadelphia; Open University Press, 1995.
5. Bullinger M, Schmidt S, Petersen C & the DISABKIDS Group. Assessing quality of life of children with chronic health conditions and disabilities: A European approach. Int J Rehabil Res 2002; 25: 197–206.
6. Eiser C. Psychological effects of chronic disease. J Child Psycho Psychiatry 1990; 31: 85–98.
7. Midence K. The effects of chronic illness on children and their families: An overview. Genet Soc Gen Psychol Monogr 1994; 120: 311–326.
8. 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: 399–407.
9. Landgraf JM, Maunsell E, Speechley KN, et al. Canadian-French, German and UK versions of the Child Health Questionnaire: Methodology and preliminary item scaling results. Qual Life Res 1998; 7: 433–445.
10. Stein REK, Jessop DJ. Functional Status II (R). A measure of child health status. Med Care 1990; 28: 1041–1055.
11. Bethell CD, Read D, Stein RE, Blumberg SJ, Wells N, Newacheck PW. Identifying children with special health care needs: Development and evaluation of a short screening instrument. Ambul Pediatr 2002; 2: 38–48.
12. Hays RD, Hayashi T, Carson S, Ware JE. Users Guide for the Multitrait Analysis Program (MAP). Rand Co-operation Report No. N-2786-RC, 1988
13. Bentler P Wu EJ. EQS for Windows. User's guide. Encino, CA: Multivariate Software, 2001.
14. Van Davier M. *Winmira*. A Method for Analysing Probabilistic Test Models. Kie1: IPN, 1996.

15. Rost J, Davier MV. A conditional item-fit index for Rasch models. Appl Psychol Measure 1994; 18: 171–182.
16. Byrne, BM. Structural Equation Modelling with AMOS. New York: Erlbaum, 2001.
17. Bullinger M, von Mackensen S, Fischer K, Khair K, Petersen C, Ravens-Sieberer U, Rocino A, Sagnier P, Tusell JM, van Den Berg M, Vicariot M. Pilot testing of the 'Haemo-QoL' quality of life questionnaire for haemophiliac children in six European countries. Haemophilia 2002; 8: 47–54.
18. The WHOQOL Group. The World Health Organization Quality of Life assessment (WHOQOL): Position paper from the World Health Organization. Soc Sci Med 1995; 41: 1403–1409.

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