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

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

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*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 <sup>1</sup>.

Children with asthma can experience limitations or impairments in various aspects of their life. Having asthma can lead to restrictions in activities <sup>2-4</sup>, emotional problems <sup>5,6</sup>, behavioural problems <sup>6,7</sup>, adjustment difficulties <sup>7</sup>, feelings of depression <sup>5</sup>, a fear of being rejected by peers due to being "different" <sup>8</sup>, lower perceived well-being <sup>2</sup>, anxiety <sup>3,9</sup> and family stress <sup>10,11</sup>. In general asthma can be kept under control through pharmacological therapy and the avoidance of triggers that influence the asthma symptoms <sup>12,13</sup>. However, there are still indications that the health related quality of life (HRQoL) of children with asthma is decreased compared to their healthy peers <sup>2,14</sup>. 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 <sup>15-17</sup>. 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<sup>18,19</sup>. Examples of some asthma-specific questionnaires are the Pediatric Asthma Quality of Life Questionnaire (PAQLQ)<sup>20</sup>, the Childhood Asthma Questionnaire (CAQ)<sup>21,22</sup>, the Life Activities Questionnaire for Childhood Asthma<sup>23</sup> and the Pediatric Quality of Life – Asthma module (PedsQL<sup>TM</sup> asthma module)<sup>24</sup>. 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<sup>25</sup>. 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<sup>26,27</sup>. Developing a questionnaire in several countries through a simultaneous approach would diminish this problem and create the advantage of a cross-national questionnaire<sup>28</sup>. However, there have been only a few attempts to develop a HRQoL questionnaire in several countries simultaneously<sup>25</sup>. The WHOQOL questionnaire is a well-known example but this is a generic questionnaire and is only for use in adults<sup>29</sup>.

With the exception of the PedsQL<sup>TM</sup>, 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<sup>30</sup>. Chronic conditions included in the project were asthma, juvenile idiopathic arthritis (JIA),

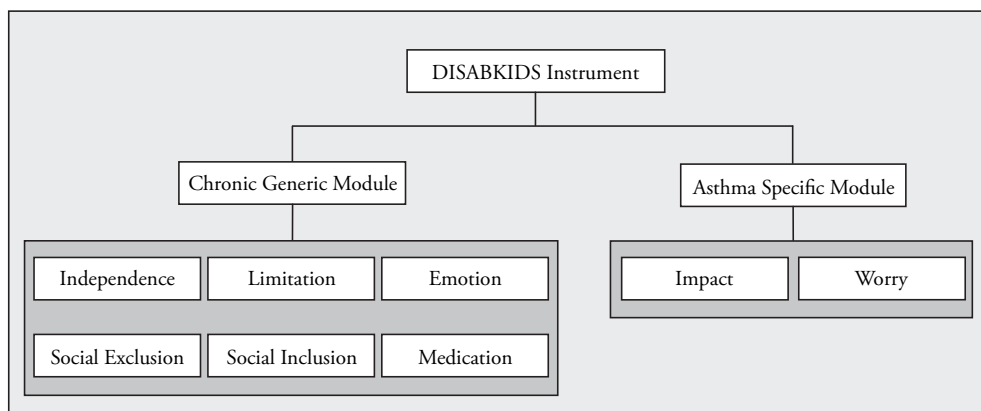
atopic dermatitis, cerebral palsy (CP), cystic fibrosis (CF), diabetes and epilepsy. The final instrument should represent the child and adolescent's view by including aspects that are important to them, be multidimensional, cross-nationally applicable, valid and reliable. Central to the development 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<sup>31,32</sup>. Applied methods and results have been described elsewhere<sup>33</sup>. These items were then translated to the appropriate languages following established guidelines (forward-backwards translations)<sup>26</sup>. 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)<sup>33</sup>.

### *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<sup>34-36</sup>. 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)<sup>37</sup>. 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<sup>38,39</sup>. 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<sup>12,40-43</sup>. 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<sup>12,44</sup>. Sometimes these parameters are combined with school or work absences, daily activities and use of health care facilities<sup>5,45,46</sup>. 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) <sup>40</sup>. 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 <sup>47</sup>.

### *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 ( $\alpha$ ) 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  $\alpha$  coefficient of 0.70 or higher is considered acceptable for questionnaire validation, whereas an  $\alpha$  of 0.90 or above is considered necessary for individual or clinical decision making<sup>48,49</sup>. 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
<b>Gender</b>								
Male	59	57	55	66	57	54	68	60
Female	41	43	45	34	43	46	32	40
<b>Age</b>								
8-12	66	63	71	74	53	73	55	62
13-16	34	37	29	26	47	27	45	38
<b>Ethnicity</b>								
Born in own country	96	90	93	100	95	97	95	98
Born in other country	4	10	7	.	5	3	5	2
<b>Education</b>								
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	.	.	.	.
<b>General Health†</b>								
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) <sup>40</sup> asthma symptom checklist, correlated 0.55 with the child's severity rating (single question) and 0.37 with the clinicians' questionnaire <sup>47</sup>. 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 <sup>40</sup> (n=382)	Clinician single severity question (n=246)	Clinician severity questionnaire <sup>47</sup> (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 ( $\alpha$ ) coefficient was determined to assess the internal consistency reliability of the DISABKIDS domains. The  $\alpha$  coefficient for the chronic generic domains ranged from 0.66 to 0.85. For the two asthma-specific domains the  $\alpha$  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  $\alpha$  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	$\alpha$ Coefficient (n=405)	Paired samples mean difference test-retest	Pearson correlation test-retest (n=59)
<b>Chronic generic</b>									
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
<b>Asthma</b>									
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 ( $\alpha$ ) 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.
<b>Chronic generic</b>							
Independence	*						
Limitations	.58	*					
Emotion	.59	.68	*				
Social exclusion	.51	.60	.59	*			
Social inclusion	.50	.51	.47	.44	*		
Medication	.31	.41	.51	.34	.33	*	
<b>Asthma</b>							
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
<b>Chronic Generic</b>					
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
<b>Asthma</b>					
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 <sup>40</sup>, the child severity score (single question), the clinician severity questionnaire <sup>47</sup>, 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.

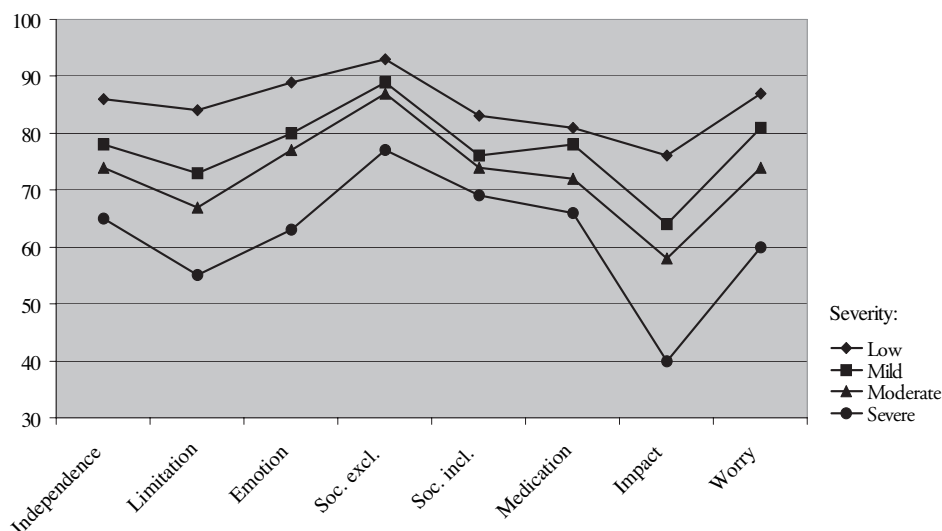


Figure 1. The DISABKIDS domains scores (0-100) of children and adolescents with asthma. Severity is based on the Rosier's (1994) <sup>40</sup> asthma symptom checklist completed by the parents.

### *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 <sup>18,35</sup>.

The internal consistency of the domains was sufficient for the total asthma population, with the Cronbach's  $\alpha$  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  $\alpha \geq 0.9$ ) are necessary for the DISABKIDS instrument to be psychometrically acceptable as an individual screening tool<sup>48,49</sup>. 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<sup>33,50-52</sup>. 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<sup>53</sup>.

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<sup>54-57</sup>.

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<sup>28</sup>. 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 effect 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)<sup>40</sup>, 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.

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## 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

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