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The Haemo-QoL Index: developing a short measure for health-related quality of life assessment in children and adolescents with haemophilia

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Summary. As quality of life (QoL) research is increasingly focusing on children and adolescents with haemophilia, the need for both age-appropriate and disease-specific assessment tools becomes apparent. Therefore, a set of questionnaires measuring QoL in children and adolescents with haemophilia was simultaneously developed in six languages within the European Haemo-QoL project (*Haemophilia*, **8**, 2002, 47; *Haemophilia*, **10**, 2004, 17). For implementation in larger studies and for use in daily clinical routine, a both short and psychometrically robust version of the questionnaire is needed. Using from the Haemo-QoL field study complete data sets of 306 children and adolescents (4–16 years) and their parents, a multivariate approach of item selection was applied to construct an eight-item instrument, the Haemo-QoL Index. The instrument is applicable to different age groups and represents the core content as well as the multidimensional structure of the original long versions. According to preliminary analyses, the index's psychometric performance concerning reliability and convergent validity is good. Further validation of the instrument's performance on a new and independent sample is needed.

Keywords: adolescents, children, Haemo-QoL, quality of life, questionnaire development, short form

Introduction

Quality of life (QoL) research in children and adolescents with haemophilia is an emerging area in haemophilia outcome assessment. Only over the past few years, instruments to measure QoL in haemophilia have been constructed [e.g. 1]. One of the first self-report tools is the Haemo-QoL instrument, which is a set of disease-specific and age-related questionnaires to measure quality of life in children and adolescents with haemophilia. The instrument was developed in the course of the Haemo-QoL project [2,3], a European cross-cultural study in six countries and is based on the World Health Organization's (WHOs) definition of health as patient-perceived wellbeing and function in terms of physical, emotional, mental, social and behavioural life domains [4,5]. In a series of steps ranging from item generation to pilot and field testing a number of different questionnaire sets were simultaneously created in six languages of the project partners (Dutch, English, French, German, Italian and Spanish): long versions for three age groups contain 21–77 items and cover 8–12 dimensions of QoL. Furthermore, two age-specific short form measures containing 16 and 35 items were developed. Each questionnaire version is available both for children's self-report as well as for parents' proxy report (see Fig. 1). All Haemo-QoL instruments, including the index, are available on the Haemo-QoL homepage [http://www.haemoqol.de].

The existing Haemo-QoL short forms are age specific and designed to be sensitive to different treatment forms. To be used as screening tools in daily clinical routine and to be implemented in larger studies, however, the need for developing a yet shorter and age generic instrument became obvious.

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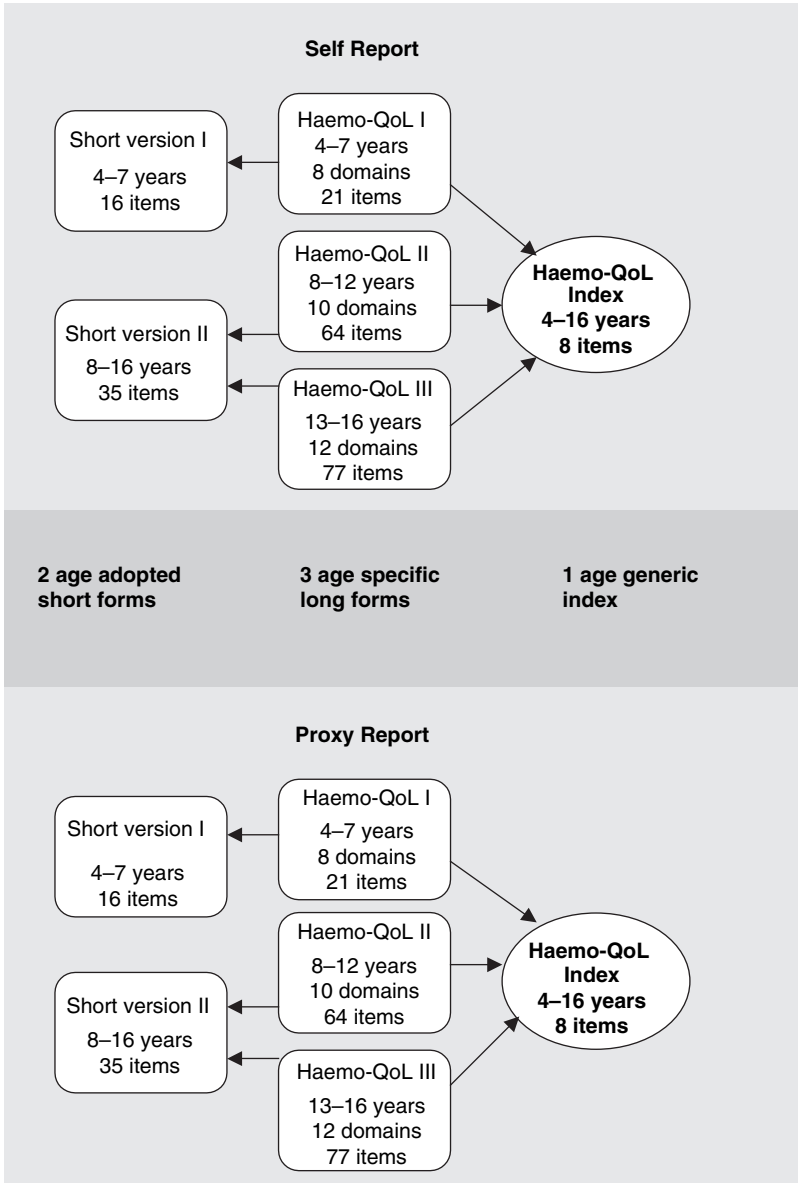

Fig. 1. Structure of the Haemo-QoL questionnaire set.

Depending on the purpose a short version intended for, several ways of construction are possible. For example, when developing a short form it can be considered important to preserve the (sub)scale structure of the original instrument. This aspect is usually important in QoL research, where the multidimensional nature of the concept is to be represented. Therefore, long form items which best represent their subscale or domain will be selected for the short form. Short form construction, however, can also neglect conceptual considerations and focus on optimizing the short version's psychometric performance. For example, selecting items on the basis of multiple regression analyses will lead to a short form that allows a good prediction of the long form's overall score, yet might not represent the original subscale contents [e.g. 6]. Such a short measure can serve as screening tool to differentiate between groups with a high or low overall QoL. Furthermore, construction can focus on the short form's homogeneity by selecting an item combination with high internal consistency [7].

The above approaches are examples for some widely used of many strategies in short form development [8]. By definition, a short form can never fully substitute for the long questionnaire in all aspects. Therefore, clarifying the major purpose the short form is intended for is a very important step within the development process [8].

As for the Haemo-QoL Index, the objectives were to: (i) develop an economical tool that represents the scale structure of the long version (index); (ii) merge

the age-specific instruments into an age generic measure that can be used in large clinical studies and for comparing data across ages.

Materials and methods

Sample

The Haemo-QoL Index was developed on the basis of the Haemo-QoL field study data [3]. A total of 339 children with haemophilia and their parents were recruited at 20 haemophilia treatment centres in six European countries (France, Germany, Italy, the Netherlands, Spain and UK). Boys were included in the study if they were aged 4 –16, understood the project language of the respective country and had either severe haemophilia A or B (factor activity level below 1% or between 1% and 2% but clinically severe). Subjects were excluded for cognitive impairment, human immunodeficiency virus infection or presence of inhibitors at the time of the study, which subsequently reduced the sample size to 320.

In the entire children's sample, the mean age was 10.00 years (SD = 3.7), 86% had haemophilia A and 87% had a factor activity level of <1%. Twenty-six per cent of the children had not experienced a joint bleed in the previous 12 months, but 11% reported having functional impairments. Sixty-seven per cent of the sample was on prophylactic treatment.

Seventy-seven per cent of the parents' sample (n = 309) were female, the mean age being 39 years (SD = 6.1); 91% were living with a partner, 28% had a college or professional degree; 38% were employed full time, 27% worked at home (see Table 1).

Defining the item pool for item selection

Calculations were based on the item pool of the Haemo-QoL questionnaire long versions: three age-specific versions are available both for children's self report as well as for parent's proxy report so that in total six different long versions can be distinguished (see Fig. 1). Haemo-QoL I for age 4–7 consists of 21 items, Haemo-QoL II for age 8–12 contains 64 items and Haemo-QoL III for age 13–16 contains 77 items. In the Haemo-QoL I, the items belong to eight dimensions considered relevant for the youngest children's QoL (physical health, feelings, view of

Table 1. Selected sociodemographic and clinical data of the Haemo-QoL field study sample (for further information see also [3]).

	Total	Age group I	Age group II	Age group III
Children				
Sample size	320	95	122	103
Mean age in years (SD)	10.00 (3.7)	5.56 (1.2)	10.00 (1.7)	14.09 (1.5)
Number of siblings				
None	19.0	26.7	12.3	19.8
One	50.5	44.4	57.0	48.5
More than one	30	28.9	30.7	31.7
Type of haemophilia				
A	85.5	84.5	83.1	89.3
B	11.6	14.4	12.7	7.8
Missing data	2.9	1.0	4.2	2.9
Level factor				
≤1	86.5	86.6	85.6	87.4
>1	12.9	12.4	14.4	11.7
Missing data	0.6	1.0	–	1.0
Treatment scheme				
Prophylactic	66.7	68.4	68.6	62.9
On-demand	31.8	30.5	30.5	34.3
Missing data	1.6	1.1	0.8	2.9
Parents				
Sample size	309	95	110	104
Mean age in years (SD)	39.30 (6.1)	36.27 (5.5)	39.23 (6.0)	42.54 (5.1)
Gender				
Female	77.2	81.9	72.6	77.5
Living with partner	90.9	92.3	90.6	88.9
Employment				
Full-time	38.4	35.1	38.4	39.6
Part-time	27.8	25.5	29.3	30.8

yourself, family, friends, others, kindergarten/school and treatment). In the Haemo-QoL II, two dimensions (dealing with haemophilia and perceived support) are added, complemented by another two dimensions (future and relationship) in Haemo-QoL III. Moreover, Haemo-QoL long forms not only differ in their number of items, but also in their number of answer categories. For the youngest age group, there are only three, frequency-related response options per item, while in all other versions five respective response options are provided.

Consequently, the first step towards a short form was defining a common item pool that all three long forms share: from the 21 items contained in Haemo-QoL I, 19 items are also part of the Haemo-QoL II and III. These 19 items belong to eight subscales, which finally formed the basis for item selection for the Haemo-QoL Index (see Table 2).

Item selection

To optimally represent the long versions' scale structure, it was decided that the index would consist of one item from each of the eight subscales that all three long versions share (i.e. the 19-item pool). Therefore, it was necessary to identify the best combination of eight of these 19 items. To maintain an acceptable consistency and homogeneity while including items originating from different subscales, an item selection strategy maximizing internal consistency was considered most useful for the Haemo-QoL Index.

As Haemo-QoL items (and factors) are intercorrelated, each item's reliability is dependent on the other items in the scale. Thus, in a selection of eight items, an item's reliability changes if one of the other items is taken out or replaced. To account for this variability, items were to be selected by a multivariate procedure calculating internal consistency (Cronbach's-alpha) for every possible combination of eight items within the 19-item pool (360 combinations). Of these 360 combinations, the best one, i.e. with the highest internal consistency was to be chosen. This procedure was to be repeated twice, once for the 19-item pool extracted from the children's instruments and once for the 19-item pool gained from the parents' versions. Assuming that the preferable item combination for children's self assessment would be different from the solution regarded best for parents' proxy assessment, it was decided to primarily focus on optimizing the children's tool for the following reasons: (i) QoL is by the WHOs definition considered a subjective construct for which self report is vital (see above); and (ii) psychometric properties of children's self-assessment tools are generally already weaker than those of adults' measures. Therefore, an optimal children's index was expected to yield also a psychometrically acceptable parents' version.

The index items are all assumed to be part of and contribute to children's overall QoL. To examine to what extent the items originating from different long-form subscales fulfil this assumption of a one-factor model, confirmatory factor analysis (CFA) was carried out using AMOS 4.01 [9] as software.

Table 2. Overview about scale structure and number of items in the Haemo-QoL long versions.

Dimension	Number of items per age group version			Number of items common across age groups
Age group	I	II	III	All
Age	4–7	8–12	13–16	
Physical health	4	7	7	4
Feeling	3	7	8	3
View	2	9	10	2
Family	4	5	8	2
Friends	1	4	4	1
Others	2	6	6	2
Sport and school	3	8	9	3
Treatment	2	7	8	2
Perceived support	–	4	4	–
Dealing	–	7	7	–
Future	–	–	4	–
Relationship	–	–	2	–
Total	21	64	77	19

Variables used for index validation

For validating the original Haemo-QoL questionnaires, information regarding sociodemographic, psychosocial and clinical variables was collected in the field study. In the index development process, clinical variables as assessed by physicians during the field study were used for determining discriminant validity (e.g. factor level activity, number of bleedings or joint bleedings, treatment modalities, etc.). Mann–Whitney *U*-tests comparing median scores between groups of children defined by these variables were calculated.

For convergent validity, three measures of the Haemo-QoL field study were taken into account: (i) the KINDL-R (Kinderlebensqualitätsfragebogen) [10], a generic and multidimensional measure of children's quality of life; (ii) the Child Health Questionnaire General Health Index (CHQ-GHI [11,12]), a global question about children's general health perception; and (iii) the 'Fragebogen zur Lebenszufriedenheit' (FLZ [13], adapted for children), a

questionnaire for self-assessment of satisfaction with important aspects of life. All three measures are inversely scored with respect to the Haemo-QoL, i.e. high values express high QoL in the KINDL-R, the CHQ and the FLZ. On the contrary, Haemo-QoL instruments are oriented towards impairment, so high QoL (i.e. little impairment) is indicated by low values. Thus, Pearson correlation coefficients, which were calculated to estimate the Haemo-QoLs convergent validity, were expected to be negative.

The intra-class correlation coefficient (ICC) was used to determine child–parent concordance (i.e. to which extent children and parents agree in their ratings) and test–retest reliability. Test–retest reliability was assessed after a 1- or 2-week interval in age groups II and III only and was calculated for subjects who did not report any major events between the two time points ($n = 167$ children and 148 parents).

To find out how well a person's total score in the Haemo-QoL long form is represented by the index, regression analyses were carried out.

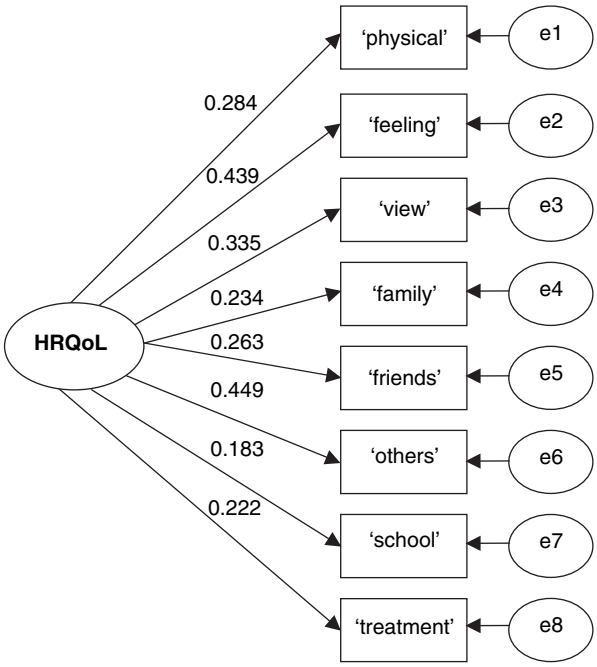

Fig. 2. Confirmatory factor analysis results of the theoretical model of the Haemo-QoL Index.

Results

Psychometric characteristics of the Haemo-QoL Index were calculated for complete cases only, i.e. for participants who responded to all eight index items ($n = 306$ children and 285 parents). As psychometric performance was estimated on the basis of the original Haemo-QoL field study sample which the index was developed on, these results have to be regarded as preliminary and require further validation.

Psychometric properties of the Haemo-QoL Index

Factor structure Fit measures as results of CFA showed that the items selected fulfilled the assumption of a one-factor model underlying the Haemo-QoL Index quite well (see Fig. 2). The Comparative Fit Index (CFI) was 0.916 and the root mean square error of approximation (RMSEA) was 0.072 (90% CI: 0.048–0.096), which indicates good model fit for the children's sample. Model fit for the parents' sample was slightly weaker but still acceptable (CFI = 0.887, RMSEA = 0.112, 90% CI: 0.089–0.136). Keeping the common factor constant, correlations of the remaining item regression residuals were low.

Descriptive statistics In the children's sample, the mean sum score for all eight index items was 15.13 (possible values ranging from 8 to 40, SD = 5.64, see Table 3). Transformed to a scale from 0 to 100, the mean total score was 22.27 (SD = 17.64). As low scores indicate high quality of life in the Haemo-QoL instruments, these results suggest a rather high overall QoL from children's self-report. The values are consistent with QoL ratings from the Haemo-QoL long forms where transformed mean scores vary from 20.18 to 23.67 across the three age groups (SD ranging from 9.30 to 12.81). The items' correlations with the index's total score (corrected for overlap) ranged from $r = 0.24$ (treatment item, i.e. item derived from the Haemo-QoL long-form subscale 'treatment') to $r = 0.55$ (feeling item). In the parents' sample, the mean sum score was 16.45 (SD = 5.72) and the transformed scale score was 26.39 (SD = 17.87). Parents thus rated their children's QOL slightly lower than did the children themselves. Corrected item–total correlation ranged from $r = 0.22$ (treatment item) to $r = 0.67$ (feeling item).

Reliability Cronbach's-alpha as a measure of internal consistency was $\alpha = 0.70$ in the children's sample and 0.78 in the parents' sample, suggesting acceptable homogeneity for the Haemo-QoL Index. The ICC indicating test–retest reliability was quite high in the children's sample (ICC = 0.76, $P < 0.01$), but rather low for parents' proxy ratings (ICC = 0.27, $P < 0.01$). Compared with the Haemo-QoL long versions with Cronbach's-alpha ranging from $\alpha = 0.85$ to $\alpha = 0.95$ and ICCs for test–retest reliability

Table 3. Item characteristics of the Haemo-QoL Index (self-report $n = 306$/proxy report $n = 285$).

Item wording	M	SD	Skewness	r_{IT}*	α_{-1}†
In the past 4 weeks					
I/my child was afraid of bleeds	1.50/1.62	0.95/1.04	2.01/1.75	0.40/0.50	0.68/0.75
I/my child was sad because of my/his haemophilia	1.52/1.73	1.01/0.95	2.04/1.21	0.55/0.67	0.65/0.73
I/my child envied healthy boys my/his age	1.85/1.93	1.32/1.12	1.32/0.90	0.52/0.54	0.64/0.75
My mother protected me/I protected my child too much	2.40/2.47	1.47/1.30	0.60/0.58	0.36/0.35	0.69/0.78
I/my child was unable to do as much with my/his friends because of my/his haemophilia	1.65/1.86	1.05/1.01	1.63/0.88	0.35/0.61	0.68/0.74
I/my child felt different from others because of my/his haemophilia	1.75/1.92	1.14/1.05	1.42/0.94	0.53/0.66	0.65/0.73
Because of haemophilia I/my child had to refrain from sports that I like/he likes	2.30/2.52	1.46/1.39	0.68/0.39	0.32/0.46	0.70/0.76
The injections annoyed me/my child	2.14/2.39	1.39/1.19	0.88/0.55	0.24/0.22	0.71/0.80
Total score (sum)	15.13/16.45	5.64/5.72	1.15/0.71	–	–
Total score (mean)	1.89/2.06	0.71/0.71	1.15/0.71	–	–
Total score (0–100)	22.27/26.39	17.64/17.87	1.15/0.71		

*r_{IT}, corrected item–total correlation.
†α_{-1}, Cronbach's-alpha if item deleted.

from ICC = 0.45 to ICC = 0.88, these index results were relatively high (see Table 4). Association between the Haemo-QoL Index and the long forms' total scores was determined by multiple regression analysis in the three different age groups. In predicting the respective long-form scores in the children's sample, the index achieved regression coefficients of $R = 0.94$ for the Haemo-QoL I, $R = 0.62$ for the Haemo-QoL II and $R = 0.85$ for the Haemo-QoL III. Adjusted R^2 indicating the explained common variance of the index with the respective long-form scores ranged from 0.34 to 0.88 in the children's sample. Consequently, a child's index score on average provides 34–88% of the information obtained by the respective long form's total score. For parents, the index achieved regression coefficients of $R = 0.96$ (Haemo-QoL I), $R = 0.88$ (Haemo-QoL II) and $R = 0.90$ (Haemo-QoL III) in predicting the long form total scores. Adjusted R^2 ranged from 0.76 to 0.92. for the different long forms. A parent's index score therefore on average predicts 76–92% of the respective long form's total score (see Table 4).

Validity Consistent with the Haemo-QoL long-form results, child–parent concordance in the QoL ratings was quite low for the index (ICC = 0.19, $P < 0.01$). Explanations for this result could not be identified: excluding the youngest age group from the analysis as well as calculating concordance rates on a single-item level (weighted kappas and ICCs) did not improve concordance results.

With respect to convergent validity, the index's correlations with instrument measuring related contents were moderate to high. In the children's sample, Pearson correlation coefficients with general QoL measures were $r = -0.22$ (KINDL-R total score, $P < 0.01$) and $r = -0.25$ (CHQ-GHI, $P < 0.01$). Correlations with (sub)scales measuring more similar contents such as the impact of the disease on QoL were notably higher: $r = -0.34$ for the FLZ ($P < 0.01$) and $r = -0.51$ for the KINDL-R Chronic Generic Scale ($P < 0.01$).

For parents, convergent validity was generally higher than in the children's sample: Pearson's r with the CHQ-GHI equalled $r = -0.37$ ($P < 0.01$), with the KINDL-R total score $r = -0.52$ ($P < 0.01$) and with the KINDL-R Chronic Generic Scale $r = -0.62$ ($P < 0.01$). In comparison with the Haemo-QoL long forms, the index's convergent validity values were mostly higher than the respective Haemo-QoL I values and not much lower than in Haemo-QoL II.

Results for the index's discriminant validity were less satisfactory. Calculating Mann–Whitney *U*-tests, hardly any significant differences in QoL between groups of children differing in their clinical presentation (factor level or number of joint bleeds) or in their treatment modalities (prophylactic vs. on-demand treatment, presence vs. absence of previous orthopaedic surgery, see Table 4) were found. However, the same is true for the original Haemo-QoL long versions.

Discussion

The multivariate development procedure outlined above resulted in an age generic, eight-item QoL index for children and adolescents with haemophilia.

Table 4. Psychometric characteristics of the Haemo-QoL Index in comparison with Haemo-QoL long forms† (self-report/proxy report)

	Haemo-QoL Index (8 items)	Haemo-QoL I (21 items)	Haemo-QoL II (64 items)	Haemo-QoL III (77 items)
Sample size (children/parents) (*n*)	306/285	95/95	122/110	103/104
Reliability				
Internal consistency (Cronbach's-alpha)	0.70/0.78	0.85/0.85	0.85/0.93	0.91/0.95
Child–parent concordance				
ICC	0.19**	0.42**	0.22*	0.13
P	<0.01	<0.01	0.02	*ns*
Test–retest sample size				
N	167/148	–	83/75	80/70
ICC	0.76**/0.27**	–	0.59**/0.45**	0.88**/0.64**
P	<0.01/<0.01		<0.01/<0.01	<0.01/<0.01
Association Index items- long form total scores (multiple linear regression)				
R		0.94/0.96	0.62/0.88	0.85/0.90
Adjusted R^2		0.88/0.92	0.34/0.76	0.69/0.78
P		<0.01/<0.01	<0.01/<0.01	<0.01/<0.01
Convergent validity (Pearson correlation coefficients)				
KINDL-R total score				
r	−0.22**/−0.52**	−0.25*/−0.25*	−0.39**/−0.72**	−0.59**/−0.72**
P	<0.01/<0.01	0.04/0.037	<0.01/<0.01	<0.01/<0.01
KINDL-R chronic-generic				
r	−0.51**/−0.62**	−0.51**/−0.58**	−0.48**/−0.76**	−0.61**/−0.71**
P	<0.01/<0.01	<0.01/<0.01	<0.01/<0.01	<0.01/<0.01
Health in general (CHQ-GHI)				
r	−0.25**/−0.37**	−0.11/−0.15	−0.21*/−0.45**	−0.52**/−0.41**
P	<0.01/<0.01	*ns*/*ns*	0.03/<0.01	<0.01/<0.01
Life satisfaction (FLZ)				
r	−0.34**/–	–	−0.44**/–	−0.57**/–
P	<0.01/–		<0.01/–	<0.01/–
Discriminant validity (Mann–Whitney U-tests)				
Factor activity level: <1% vs. ≥1%				
U	4415.5/3717.5	240.5/209.0	595.5/447.5*	316.0/393.0
P	*ns*/*ns*	*ns*/*ns*	*ns*/0.04	*ns*/*ns*
No. joint bleeds: <5 vs. ≥5				
U	4826.5/3932.5*	245.0/217.5	651.5/593.5	385.5**/453.5
P	*ns*/0.01	*ns*/*ns*	*ns*/*ns*	<0.01/*ns*
Treatment: prophylactic vs. on-demand				
U	9744.0/7896.0	591.0/512.0	1042.0/1009.5	924.5/705.0
P	*ns*/*ns*	*ns*/*ns*	*ns*/*ns*	*ns*/*ns*
Orthopaedic surgery: yes vs. no				
U	1412.0**/1765.5	49.0/47.5	127.5/161.5	169.0**/412.0
P	<0.01/*ns*	*ns*/*ns*	*ns*/*ns*	<0.01/*ns*

ns, not significant; CHQ-GHI, Child Health Questionnaire General Health Index; FLZ, Fragebogen zur Lebenszufriedenheit; *, result is significant at the 5% level; **, result is significant at the 1% level.
†For long form values see also [3].

Based on estimations from the Haemo-QoL field study data, the instrument reveals good psychometric characteristics.

Fit statistics using CFA proved satisfactory for the proposed one-factor model. Items loading highest on this factor, which is interpreted as 'health-related QoL', belong to subscales measuring feeling and wellbeing. These results are consistent with other findings within QoL research, where items measuring 'feeling' and 'wellbeing' also prove to be most important for explaining variance in QoL scores [e.g. Debensasson *et al.*, unpublished data]. Moreover, the index's item–total correlations reflect the importance of emotional contents for QoL ratings. However, recent studies indicate that the relative contribution of different facets to general quality of life also varies for different chronic conditions [14].

Internal consistency was good in comparison with the respective long-form values, suggesting that a homogenous index instrument could be developed despite the differences between the long-form questionnaires it originated from.

With respect to convergent validity, results were encouraging as the Haemo-QoL Index correlated higher with disease-related QoL scales as opposed to generic ones. Thus the index performed precisely as expected for a disease-specific QoL tool. This result not only underlines the validity of the Haemo-QoL Index but also supports the concept that generic instruments might not be sensitive enough for adequate QoL assessment in children with chronic diseases, so that disease-specific tools are necessary [4,15].

The index's discriminant validity however could not be demonstrated. This might be due to the lack of differences in clinical characteristics in the total sample and the general problem to validate a QoL measure against clinical criteria. The impact of different clinical conditions and treatment modalities on children's QoL and their potential to discriminate between groups of children in terms of QoL still remain to be explored [16]. For future research it would perhaps be more accurate to assess the clinical validity of QoL measures by considering the whole disease history of a patient including treatment instead of single clinical characteristics. Subjects could then be clustered according to their different disease histories and these clusters could be compared regarding QoL.

The rather low child–parent concordance of the Haemo-QoL measures could indicate that parents' impression of their children's QoL is neither directly reflecting nor consistent with the children's report. Empirical findings about child–parent concordance are diverse: Eiser and Berrenberg [17] and Eiser and Morse [18] suggest that parents of healthy children tend to overestimate their children's QoL, whereas parents of chronically ill children tend to underestimate their offspring's QoL in comparison with children's self report. In a European project aimed at enhancing the quality of life of children with chronic health conditions and their families (DISABKIDS), instruments measuring QoL in children with various chronic conditions (e.g. asthma, diabetes, etc.) were developed. Here, child–parent concordance rates were higher, varying around ICCs of 0.50–0.60 for different subscales [19].

The Haemo-QoL Index is an example for short-form development under very specific conditions that placed a number of restrictions on the development process: the original age-specific long forms not only differed in their number of items and dimensions, but also in their number of response options per item, which was intended to maximize age appropriateness of the measures. It could be argued that even the attempt of merging such diverse long versions into a common index tool should not have been made. On the other hand, considering the long forms' good reliability and convergent validity as well as the clinical need for an index questionnaire, developing it was judged acceptable and worthwhile even though some weaknesses had to be expected. Also, the strategy used to derive the index impacted on its properties and could be challenged. The *a priori* conceptual decision to include items from all long-form dimensions lead to an index differing from the one that would have emerged when choosing only items that best reflect the long versions' total scores. Given the multidimensional nature of health-related QoL, however, this strategy seemed both appropriate and psychometrically successful.

With its eight items, the index contains 38% of the 21 Haemo-QoL I items only, but is able to predict 88% of the Haemo-Qol I total score in the children's sample (adjusted R^2 of 0.88). In other words, when using the index instead of the Haemo-QoL I, one will on average lose about 12% of the information (for parents, 8%), but save about 62% of the time in terms of not filling out 62% of the items. Using the index instead of the Haemo-QoL II one will lose more information (66% for children, 24% for parents), but also save more time, as only 12.5% of the 64 items have to be filled out. Regarding the Haemo-QoL III, one will on average lose about 31% of the information when administering the index instead of the long form, but 90% of the time needed for filling out the questionnaire will be saved.

Contrasting time savings to the loss of information expected shows how important it is to identify which level of comprehensiveness is intended when implementing a QoL measure in a given study. An index might be a helpful and quick screener in clinical practice and an appropriate tool to describe overall disease-specific QoL in larger studies. Although in clinical trials examining treatment outcomes, longer and more sensitive QoL questionnaires such as the Haemo-QoL long forms are preferable.

The Haemo-Qol Index was both developed and preliminary validated on the basis of the Haemo-QoL field study data. It is therefore urgently needed to investigate its psychometric performance on at least one new and independent sample before drawing more definite conclusions about its quality. This could be achieved within the currently ongoing ESCHQoL project, a European study of clinical, health, economic and quality of life outcomes in haemophilia treatment, which also uses the Haemo-QoL instruments. The European ESCHQoL study assesses clinical outcomes of haemophilia care in

relation to quality of life and health economic aspects in adults as well as in children [http://www.eschqol.org].

In general, preliminary results suggest that the Haemo-QoL Index is a useful and economical instrument that reflects the multidimensional nature of HRQoL. It can be implemented in larger studies and – after further validation – in daily clinical routine when a quick screening of haemophiliac children's QoL is required.

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