

# Health related quality of life in disorders of defecation: the Defecation Disorder List

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**Background:** Constipation and encopresis frequently cause problems with respect to emotional wellbeing, and social and family life. Instruments to measure Health Related Quality of Life (HRQoL) in these disorders are not available.

**Methods:** A disease specific HRQoL instrument, the "Defecation Disorder List" (DDL) for children with constipation or functional non-retentive faecal soiling (FNRFS) was developed using accepted guidelines. For each phase of the process, different samples of patients were used. The final phase of development included 27 children. Reliability was assessed in two ways: internal consistency of domains with Cronbach's alpha, and test-retest reliability with intra-class correlation coefficients (ICC). To assess validity, comparable items and domains were correlated with Tacqol, a generic HRQoL instrument for children (TNO-AZL).

**Results:** In the final phase of the development, 27 children completed the instrument. It consisted of 37 items in four domains. The response rate was 96%. Reliability was good for all domains, with Cronbach's alpha values ranging from 0.61 to 0.76. Measures of test-retest stability were good for all four domains with ICCs ranging from 0.82 to 0.92. Validity based on comparison with the Tacqol instrument was moderate.

**Conclusion:** The DDL is promising as a measure of HRQoL in childhood defecation disorders.

Chronic constipation is a common problem in childhood, accounting for about 3% of consultations in paediatric practice.<sup>1</sup> In many children constipation is accompanied by encopresis, defined as voluntary or involuntary faecal soiling of underwear in children older than 4 years.<sup>2</sup> This condition often gives rise to behavioural, social, and emotional problems.<sup>3</sup> In the majority of cases no organic or psychiatric disorder is identified. Treatment is symptomatic and often prolonged. After five years of treatment 50% of children still suffer from constipation.<sup>4,5</sup>

Functional non-retentive faecal soiling (FNRFS) refers to encopresis as an isolated complaint, in the absence of constipation.<sup>6</sup> It is estimated that 20% of patients with encopresis suffer from FNRFS and the remaining 80% are constipated.<sup>7</sup>

Health related quality of life (HRQoL) encompasses the physical, social, and emotional effect of health disorders. It is now considered an important outcome measure in addition to other standard measures of disease activity. It provides complementary information that can guide the choice of treatment, especially when different regimes may appear equally effective.

Most available HRQoL questionnaires are generic rather than disease specific.<sup>8–11</sup> The major disadvantage with these is their tendency to lack sensitivity. Currently there are few paediatric disease specific questionnaires.

A disease specific questionnaire developed for Hirschsprung's disease in childhood<sup>12</sup> showed a relation between HRQoL and the severity of encopresis in those who had undergone surgery. Instruments to assess the impact of defecation disorders have been developed for adults with constipation ([www.qolid.org](http://www.qolid.org)) and for faecal incontinence (HRQoL instrument).<sup>13</sup> No HRQoL is available for children with constipation or FNRFS. The present study describes the development of such an instrument, the Defecation Disorder List (DDL).

## METHODS

The HRQoL questionnaire was developed using accepted guidelines.<sup>14</sup>

### Phase 1: Defining population and objectives for development

Patients with constipation or FNRFS (7–15 years old) referred to our tertiary care centre by school doctors, general physicians, and paediatricians were eligible. They were included in the study if:

- (1) They met at least two of four criteria for paediatric constipation:<sup>15</sup> fewer than three bowel movements per week (without laxatives); encopresis at least twice weekly; intermittent passage of very large stools (every 7–30 days); palpable abdominal or rectal mass
- (2) They fulfilled the criteria for FNRFS: encopresis at least twice weekly in the absence of constipation.

All had symptoms for at least six months.

### Phase 2: Item generation

Item generation was performed through group discussion among paediatric gastroenterologists and psychologists. A list of 102 items covering physical, social, emotional, and treatment issues was generated.

### Phase 3: Item reduction, phrasing, and formatting

The goals of item reduction are to eliminate redundant or inappropriate items, to include manageable numbers of questions, and to create a valid scale. Forty three patients (able to read and understand Dutch) were asked to rate the importance to their lives of each of the 102 items using a four

**Abbreviations:** FNRFS, functional non-retentive faecal soiling; HRQoL, health related quality of life; ICC, intra-class correlation coefficients; Tacqol, TNO-AZL Children's Quality of Life Questionnaire

point Likert scale. This ranged from least important (1 point) to most important (4 points). Initially they completed the questionnaire unsupervised, but if they did not understand items, they could receive help from a parent. Based on these scores, items were ranked, and the 33 most important for the total group, as well as for age and sex, were selected for inclusion. In addition, eight relevant general items were included from the Impact HRQoL instrument for children with inflammatory bowel disease.<sup>8</sup>

The questionnaire was also tested by one of the investigators (IJGK) using the Question Appraisal System (QAS-99) (Willis GB and Lessler JT, Research Triangle Institute, Rockville, USA) to identify problems of phrasing.

Five children completed the questionnaire and were then interviewed. This was to ensure that they understood the content and that all interpreted the question similarly. This led to some changes in phrasing. These children were also asked if they considered that important items were missing, but none were identified.

Questions were phrased in the first person. Answers were based on a five point Likert scale. The instrument now contained 41 items within four domains: constipation related, emotional functioning, social functioning, and treatment/interventions.

#### Phase 4: Pilot testing of the questionnaire

A further group of 26 children (18 boys) completed the questionnaire to ensure feasibility and to identify redundant items. These suffered from constipation with encopresis (n = 18), constipation alone (n = 3), and FNRFS (n = 5).

Spearman rank order correlation coefficients were calculated between all items to identify redundant items. This was considered if the coefficient was >0.6 and/or if items were of comparable content.

#### Phase 5: Modifications of pre-final instrument

Three of 41 items closely correlated and/or were of comparable content; they were excluded, leaving 38 items.

#### Phase 6: Reliability and validity

All children involved in the final testing phase kept a diary to record stool and encopresis frequency. They underwent a toilet training regime (three times daily) with a reward system. The constipated patients received laxative treatment.

The DDL and the generic Tacqol instrument (TNO-AZL)<sup>16</sup> were used in this phase. The latter examines health status in recent weeks and the emotional response to problems. Tacqol uses a four point Likert scale and contains seven domains, each with eight items: physical complaints, motor functioning, autonomy, cognitive functioning, social functioning, and positive and negative emotions.

**Table 1** Patient characteristics at time of completing the DDL

Characteristic	No.
No. of patients	27
Median years (range)	10.7 (7.3–14.3)
No. of boys	18
No. with constipation and encopresis	18
No. with constipation only	4
No. with FNRFS	5
Encopresis, episodes/week; median (range)	2 (0–8.5)
Duration of previous treatment in months; median (range)	33 (4.5–132)
Duration of previous symptoms in years; median (range)	6.4 (1.4–12.4)

**Table 2** Internal consistency (Cronbach's  $\alpha$ ) and ICCs

Domains	No. of items	Cronbach's $\alpha$	ICCs
Constipation related	3	0.65	0.92
Emotional functioning	13	0.76	0.89
Social functioning	11	0.63	0.82
Treatment/interventions	10	0.61	0.87
Total instrument	37		0.87

After consenting to participate, patients received the two questionnaires. If no response was received within two weeks, they were contacted to encourage participation. Two weeks later the DDL questionnaire was again sent to the participants. Again, if necessary the family were contacted.

This final phase included 28 subjects (19 boys), including children with constipation with encopresis (n = 19), constipation without encopresis (n = 4) and FNRFS (n = 5).

#### Statistical analysis

##### Reliability

First, "floor and ceiling effects" were examined. If a high proportion scored the highest (ceiling) or lowest possible (floor) score on an item, this item was considered to be of limited value in detecting change over time.<sup>17</sup> Reliability was assessed by examining "internal consistency" and "test-retest reliability". Internal consistency (or homogeneity), referring to the extent to which the items in a domain assess the same characteristic, was measured using Cronbach's alpha.<sup>18</sup> Homogeneity was considered satisfactory if the Cronbach's alpha value was >0.6 and excellent if >0.9. Reliability was assessed by repeat completion of the questionnaire after two weeks. It was assumed that symptoms would be unchanged over this period. For test-retest analysis, intra-class correlation coefficients (ICCs) were calculated for each item and each domain to assess the correlation between scores while correcting for systematic differences in scores.

##### Validity

"Validity" is defined as the extent to which the instrument measures what it is intended to measure. In developing the instrument, validity was tested in three ways. "Content validity" was tested during the early phases by ensuring that no issues of importance were omitted. "Construct validity" was tested by examining correlations with comparable items and domains in the Tacqol instrument. If this disease related instrument was measuring HRQoL, it should correlate with the generic instrument. Since disease specific instruments are more likely to detect small changes, such correlations would not normally be very high. A Spearman rank order coefficient between 0.4 and 0.6 was considered acceptable. "Discriminatory validity" was assessed based on the hypothesis that

**Table 3** Correlations of the DDL with overlapping items and domains in the Tacqol generic instrument

DDL item	Tacqol item	Spearman rank order
Stomach aches	Stomach item	0.74
Angry to have problem	Feeling angry	0.17
Being happy	Feeling happy	0.55
Being happy	Feeling pleased	0.55
DDL domain	Tacqol domain	Spearman rank order
Emotional functioning	Positive emotions	0.03
Emotional functioning	Negative emotions	0.19
Social functioning	Social functioning	0.14

**Table 4** Discriminant validity of the DDL in comparing low frequency ( $\leq 2$  times per week; group 1) and high frequency ( $> 2$  times per week; group 2) encopresis

Domains	Domain score Group 1	Domain score Group 2	p value	Mean difference (95% CI)
Constipation related	10.13	10.08	0.95	0.05 (-1.7 to 1.8)
Emotional functioning	25.73	26.75	0.69	1.02 (-6.2 to 4.2)
Social functioning	22.60	26.50	0.08	3.90 (-8.3 to 0.5)
Treatment/interventions	22.27	28.67	0.02	6.40 (-11.8 to -1.0)
Total instrument	87.00	97.92	0.09	10.92 (-23.8 to 2.0)

Independent Student's *t* test used to compare mean domain scores between groups.

HRQoL in those with very frequent encopresis would be lower than with lower frequency. Therefore, the mean domain scores in those with encopresis  $\leq 2$  times weekly, and those with  $> 2$  times weekly were compared, using the independent Student's *t* test.

## RESULTS

Table 1 shows patient characteristics. Overall 27 of the 28 subjects completed the first and second questionnaires, and each completed all items.

### Reliability: internal consistency and test-retest reliability (table 2)

Cronbach's  $\alpha$  values ranged from 0.61 to 0.76, indicating good consistency within the domains. ICCs ranged from 0.82 to 0.92, with an overall value for the questionnaire of 0.87. This indicates good to excellent reproducibility.

Four items (18, 31, 37, and 42) of the DDL suffered from floor effects, with 70% choosing the highest possible score (lowest in terms of HRQoL). Three of these items were in the social functioning domain. Only one item had to be removed from the final instrument because it did not contribute to homogeneity (Cronbach's alpha) in any of the four domains. Therefore, the final instrument consisted of 37 items.

### Validity (table 3)

The correlation between the DDL and the generic instrument with Spearman rank order coefficients was relatively low and ranged from 0.03 to 0.74. The domains related to "positive emotions" and "social functioning" showed an especially low correlation. With respect to discriminatory validity, only the treatment domain differed significantly between the groups (table 4). Subgroup analysis showed that in the male subgroup a statistically significant difference between those with a high and low encopresis frequency persisted in the treatment domain, but no differences were found in the other domains. In the female subgroup no domains were significantly different when comparing those with high and low encopresis frequency.

## DISCUSSION

This study describes the development of the first HRQoL instrument specifically for children with constipation and functional non-retentive faecal soiling. The questionnaire was developed using accepted standards for the development of such instruments. The final questionnaire consisted of 37 items in four domains. It appears to provide a valid and reliable instrument. The correlation with an established generic HRQoL instrument (Tacqol) was relatively low. The response rate to the questionnaire was excellent (96%). Patients filled in all items of the questionnaire and so it appears to be satisfactory in terms of patient acceptability.

Establishing the validity of an HRQoL instrument is difficult given the absence of gold standards.<sup>19</sup> Validity

of the DDL with respect to construct validity was moderate. Comparable items and domains correlated moderately well with the Tacqol instrument. This could be explained by subtle differences in wording. For example, an item from the Tacqol asks about "feeling angry" while an item from the DDL asks about only "feeling angry at having this disorder"; these items had a correlation coefficient of 0.17 (table 3).

Childhood constipation is common and about 30% of patients continue to have symptoms at 16 years of age;<sup>3</sup> it is therefore important to have available an HRQoL instrument. Generic quality of life instruments are considered unsatisfactory, and disease specific instruments are to be preferred.<sup>20-22</sup>

Many children with constipation suffer from encopresis. This often has a profoundly negative effect on the social and emotional aspects of life. Until now there have been few studies formally evaluating the impact of these conditions on quality of life. Two reports on children following surgical correction for anorectal malformations and Hirschsprung's disease have reported that faecal soiling has a major impact on the HRQoL.<sup>12, 23</sup>

The sensitivity of this instrument and its ability to detect changes over time require further evaluation using larger samples of patients. Currently the questionnaire is being translated into other languages, but these translations will require formal evaluation.

Apart from differences in wording, the low correlation between the Tacqol and DDL instruments implies that the instruments are not measuring the same thing. Either the Tacqol is unsatisfactory or our instrument is not valid. Thus if a patient with encopresis fills out an HRQoL questionnaire that does not address this specific symptom, the final score will be relatively lower than with a disease specific instrument.

Validity with respect to the discriminatory capabilities of the instrument was not as good as had been hoped. The confidence intervals of the domain scores comparing those with a low and high frequency of encopresis were wide (table 4). The mean differences between these groups were large for both the "social functioning" and "treatment/interventions" domains. The latter difference was significantly different ( $p = 0.02$ ). Comparing the total DDL scores of the two groups, the mean difference was 10.9 (97.9 minus 87.00).

Four items in the HRQoL showed significant "floor effects", so that they could not be expected to detect changes over time. Three of these were in the social domain, but they may nevertheless provide important information in other respects.

In summary, the DDL is a promising disease specific HRQoL instrument for children between 7 and 15 years of age with constipation or FNRFS. The ultimate purpose of this instrument is in the assessment of therapy and its impact on HRQoL. It now requires further study in a larger patient population.

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