
Development and Validation of the Fecal Incontinence and Constipation Quality of Life Measure in Children With Spina Bifida

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Purpose: Fecal incontinence and constipation in children with spina bifida are recognized to impact quality of life. Most disease specific quality of life instruments on fecal incontinence target adults and/or children without neuropathic bowel. We developed an instrument to evaluate bowel function and its impact on quality of life in children with spina bifida and their caregivers.

Materials and Methods: A 51-item questionnaire termed the FIC QOL (Fecal Incontinence and Constipation Quality of Life) survey was developed from expert opinion, patient interviews, and modification of previously published adult and pediatric studies for nonneuropathic bowel dysfunction. The items are divided into 7 quality of life factor groupings, including bowel program, dietary management, symptoms, travel and socialization, family relationships, caregiver emotional impact and financial impact. The questionnaire was given to caregivers of children with and without spina bifida. Discriminant validity was evaluated by comparing the spina bifida and control groups. Test-retest reliability was evaluated by having 41 patients complete 2 surveys within 4 to 6 weeks.

Results: Comparing questionnaires from 92 index patients and 52 controls showed a statistically significant difference for all 7 quality of life factor groupings. The FIC QOL instrument objectively demonstrated the negative impact of fecal incontinence and constipation on quality of life in these families. Comparing 82 questionnaires at 2 time points demonstrated the reliability of all FIC QOL questions.

Conclusions: The FIC QOL instrument provides a valid and reliable measure of the effect of fecal incontinence and constipation on the quality of life of caregivers and their children with spina bifida.

Key Words: abnormalities, spinal dysraphism, quality of life, questionnaires, fecal incontinence

Longitudinal studies following patients with SB from birth to adulthood show that these patients chronically experience neurogenic bowel, neurogenic bladder, hydrocephalus and issues related to mobility.¹ The impact of this chronic disease on the family of the patient is also significant.² Of patients with SB 34% of those 16 to 25 years old report bowel incontinence and 77% perceive that bowel incontinence has a negative impact on their lives.³ Krogh et al found that two-thirds of patients with myelomeningocele who were 6 years old or older and had fecal incontinence reported that incontinence significantly influenced social activities and QOL.⁴

We assessed the impact of bowel incontinence and constipation on QOL in patients with SB and their family members. To accomplish this goal we reviewed the literature for QOL instruments. Two general categories of instruments exist, that is generic and disease specific. Generic QOL instruments are useful for comparing outcomes across various populations. Disease specific instruments are more sensitive for detecting and quantifying small changes. We believed that a generic QOL instrument would fail to identify the specific effects of bowel incontinence in such a unique patient population.

We evaluated published QOL questionnaires for fecal incontinence and neurogenic bowel.⁵⁻¹⁰ Most of these instruments were validated in adults with nonneurogenic fecal incontinence or in children with normal sensation. There is only 1 SB specific QOL questionnaire in the literature.¹¹ This is a generic questionnaire that does not inquire about relationships in the family or about the specifics of bowel care. To our knowledge a QOL instrument or questionnaire to assess the impact of fecal incontinence and constipation on patients with SB and their families has not been described.

We describe the development, reliability and validity testing of a QOL measure specifically designed to assess the impact of fecal incontinence and constipation on the lives of children with SB and their families. This instrument would allow us and other investigators to objectively discriminate between populations of patients with SB based on the severity of symptoms and assess the impact of behavioral, medical or surgical therapy on QOL.

MATERIALS AND METHODS

Selection of Item Pool

This investigation was a cross-sectional questionnaire study. A list of questions/items that might plausibly be included in the final instrument was generated from cer-

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tain sources, including a pediatric urologist, a pediatric nurse practitioner, a physiatrist with a special interest in SB, information gained from patient/family interviews and a review of other pertinent QOL questionnaires. The goals of our proposed questionnaire were 2-fold, that is to discriminate between patients with SB in regard to bowel function and measure the magnitude of change in QOL after treatment. The focus was on features of daily life in which bowel incontinence and bowel care have a substantial impact. Items important to patients that were stable for at least short periods and performed by virtually all subjects were selected. Items that might be affected by change in bowel symptoms were also chosen. Questions that asked about pain or incontinence were specifically written in reference to pain from constipation or fecal incontinence, so that other sources of pain and urinary incontinence were excluded.

Item Scaling

Items concerned with identifying differences between individuals were scaled simply (2 options) to eliminate variability in interpretation. Other items had a more extensive scale to be sensitive enough to identify change after a treatment has been completed.

Item Reduction

Items were chosen from the total item pool for the first version of the questionnaire based on item frequency and importance, as assessed by patients and health providers. We performed field testing in 20 patients at a SB clinic. Content of the definitive instrument was based on item performance in this clinical setting. Items that effectively discriminated between patients with SB according to bowel function and were responsive to important differences in bowel care were chosen. Items that were unresponsive were not included. Ultimately a 51-item questionnaire termed the FIC QOL survey was developed, including 4 items describing demographic parameters (age, gender, diagnosis and race) and 47 describing fecal incontinence and QOL. The 47 items were separated into 7 QOL factor groupings based on subject, bowel program (16 items), dietary management (6), symptoms (4), travel and socialization (5), family relationships (4), caregiver support and emotional impact (8), and financial impact (4). Of these items 26 were considered functional, while 21 measured bother. The Appendix shows sample items from the final questionnaire.

Study Design

To accomplish validity and reliability testing the FIC QOL questionnaire was completed by 2 distinct populations from our institution, including 1 with SB and a control population that had urological issues without associated bowel dysfunction. Any patient with SB was eligible. Participants were sequentially recruited into the study during 4 years, starting in 2003 and ending in 2007. Questionnaires were distributed in the clinic or by mail.

For test-retest analysis a subgroup of the SB and control populations completed the questionnaire at 2 points separated by 4 to 6 weeks. Patients were randomly assigned to the test-retest group. The remainder of the control and SB groups completed the survey only once.

Test-retest reliability assesses the stability of a measurement within a period in which no change is expected to occur. A reliable instrument provides the same measurement at the 2 time points. The sign test and Wilcoxon signed rank test were used to evaluate test-retest reliability. The sign test was used on unordered and binary variables. The Wilcoxon signed rank test was used on continuous and unordered variables.

Discriminant validity was used to assess the ability of this questionnaire to distinguish between the SB and control populations. If our questionnaire is a condition specific QOL scale, the SB population should demonstrate lower QOL than the control population. For unordered variables we used the chi-square and Fisher exact tests. For continuous and ordered variables we used the Wilcoxon rank sum test. The Wilcoxon rank sum test is comparable to a 2 independent sample t test except the Wilcoxon rank sum test does not require normal assumption.

RESULTS

Of the 278 questionnaires that were distributed 144 were returned for an overall response rate of 52%. Of the 58 questionnaires given to the control population 52 were returned for a response rate of 89%. Of the 220 surveys distributed to the SB population 92 were returned for a response rate of 42%. Of the 59 surveys distributed to test-retest participants 41 were returned for a response rate of 69%.

The table lists the demographic characteristics of index patients and controls. Psychometric assessment of our questionnaire included test-retest reliability and discriminant validity. The test and retest administrations were completed an average of 29 days apart. None of the scales showed significant differences between the test and retest administration by the sign and Wilcoxon signed rank tests ($p = 0.25$ to 1.00). Validity testing with the chi square, Fisher exact and Wilcoxon rank sum tests showed that the index population had a significantly lower QOL score than controls for each questionnaire item tested ($p < 0.0001$ to 0.033).

	SB	Control
No. pts	92	52
Mean yrs age (range)	8.9 (4–16)	9.9 (2–23)
% Sex:		
M	50	64
F	50	36
% Race:		
Asian	2	4
Black	8	5
White	44	60
Hispanic	39	18
Other	7	5

DISCUSSION

Longitudinal studies in children with SB have shown that there is a significant impact on QOL of the patient and family members.^{1,12-14} Fecal incontinence is a socially difficult problem that affects at least a third of patients with SB.¹⁴ Advances in surgical treatment for neurogenic bowel, such as the antegrade continence enema, have allowed children to stop using diapers and be more like their peers.¹⁵ Studies have shown improved self-esteem following the antegrade continence enema procedure.¹⁶ These surgical interventions have an excellent success rate and studies have shown an improvement in QOL.¹⁷⁻¹⁹ These studies are limited by the fact that improvements in QOL were assessed using unvalidated instruments or clinician assessment. Assessing QOL in the pediatric SB population using a validated generic QOL instrument before and after reconstructive surgery did not show a difference.²⁰

To our knowledge a specific instrument to measure the impact of fecal incontinence on QOL in the SB population does not exist. The single published SB specific questionnaire focuses on general QOL and does not specifically measure QOL related to bowel issues.¹¹ We designed a questionnaire that is discriminative and evaluative, and specifically examines bowel issues that patients with SB and their families encounter. The FIC QOL survey passed the requisite psychometric testing to establish it as a useful tool.

Limitations of the FIC QOL questionnaire are that it may be difficult to tease out the effects of bowel care, and the morbidity of fecal incontinence and constipation on QOL, and separate that from the effects of urinary incontinence and other morbidities associated with SB. Also, the FIC QOL survey is meant to be completed by the caregiver, which may not be applicable as patients with SB enter adolescence. The response rate of the index population was significantly lower than that of the control population. This disparity was likely secondary to the fact that most control patients received and completed the questionnaire at the clinic. A larger portion of the SB population was mailed the questionnaire.

The FIC QOL instrument would be useful for determining what factors worsen or improve QOL in these families. A multifactorial analysis of demographics and QOL measures may manifest family factors or practices that affect QOL. As clinicians, we may learn what issues families actually perceive as a bother, rather than our projection on the situation. Likewise the success of medical and surgical continence remedies administered to improve QOL may be measured more objectively, rather than via the usual assessment of functional continence, which does not necessarily reflect QOL.

CONCLUSIONS

The FIC QOL questionnaire provides a valid and reliable measure of the effect of bowel care, fecal incontinence and constipation on QOL in children with SB and their caregivers. This instrument will allow clinicians and investigators to objectively discriminate between patients with SB based on the severity of symptoms and assess the impact of behavioral, medical or surgical therapy on QOL.

APPENDIX 1

Sample Items From FIC QOL Survey

Bowel Program: 4 sample questions

Does your child feel/sense the coming of a bowel movement (BM)?
 What percentage of time does your child produce a BM in the toilet?
 How many diaper changes per day does he/she require for BM (not urine)?
 How many bowel accidents (not in toilet) per week does child have?

Dietary Management: 3 sample questions

Do you notice any significant changes in bowel movements with diet?
 Is your child sensitive to foods in relation to bowel movements?
 Have you changed your child's diet to achieve better bowel care?

Symptoms: 4 sample questions

Does your child have abdominal pain from constipation?
 If so, how many episodes per month?
 Has your child ever been admitted to a hospital for constipation?
 If so, how many times?

Sample Likert Scale:

Never	Almost Never	Sometimes	Often	Almost Always
0	1	2	3	4

Travel and Socialization: 3 sample questions

How often does your child's bowel care prevent him/her from going out of the house?
 How often does your child's bowel care prevent you from going out of the house?
 My child is afraid to go out because of stool incontinence (Likert scale).

Family Relationships: 4 sample questions (all Likert scale)

My child's bowel problems affect his/her relationship with siblings.
 My child's bowel problems affect my relationship with my other children.
 My child's bowel incontinence affects his/her ability to socialize and meet friends.
 My child's bowel problems affect my relationship with my partner.

Caregiver Emotional Impact: 4 sample questions (all Likert scale)

My child's bowel care bothers me.
 My child's bladder/urine care bothers me.
 My child's bowel problems make me feel depressed.
 My child's bowel problems make me feel anxious.

Financial Impact: 2 sample questions (all Likert scale)

Are you employed?
 If no, does your child's bowel care prevent you from working?

APPENDIX 2

The FIC QOL survey is available for investigation and clinical use at http://www.ucdmc.ucdavis.edu/urology/specialties/pediatric_urology/.

Abbreviations and Acronyms

FIC QOL	=	Fecal Incontinence and Constipation QOL Measure
QOL	=	quality of life
SB	=	spina bifida

REFERENCES

- Oakeshott P and Hunt GM: Long-term outcome in open spina bifida. *Br J Gen Pract* 2003; **53**: 632.
- McCormick MC, Charney EB and Stemmler MM: Assessing the impact of a child with spina bifida on the family. *Dev Med Child Neurol* 1986; **28**: 53.
- Verhoef M, Lurvink M, Barf HA, Post MW, van Asbeck FW, Gooskens RH et al: High prevalence of incontinence among young adults with spina bifida: description, prediction and problem perception. *Spinal Cord* 2005; **43**: 331.
- Krogh K, Lie HR, Bilenberg N and Laurberg S: Bowel function in Danish children with myelomeningocele. *APMIS, suppl.*, 2003; 81.
- Rockwood TH: Incontinence severity and QOL scales for fecal incontinence. *Gastroenterology* 2004; **126**: S106.

6. Temple LK, Bacik J, Savatta SG, Gottesman L, Paty PB, Weiser MR et al: The development of a validated instrument to evaluate bowel function after sphincter-preserving surgery for rectal cancer. *Dis Colon Rectum* 2005; **48**: 1353.
7. Krogh K, Christensen P, Sabroe S and Laurberg S: Neurogenic bowel dysfunction score. *Spinal Cord* 2006; **44**: 625.
8. Pescatori M, Anastasio G, Bottini C and Mentasti A: New grading and scoring for anal incontinence. Evaluation of 335 patients. *Dis Colon Rectum* 1992; **35**: 482.
9. Eypasch E, Williams JI, Wood-Dauphinee S, Ure BM, Schmulling C, Neugebauer E et al: Gastrointestinal Quality of Life Index: development, validation and application of a new instrument. *Br J Surg* 1995; **82**: 216.
10. Otley A, Smith C, Nicholas D, Munk M, Avolio J, Sherman PM et al: The IMPACT questionnaire: a valid measure of health-related quality of life in pediatric inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* 2002; **35**: 557.
11. Parkin PC, Kirpalani HM, Rosenbaum PL, Fehlings DL, Van Nie A, Willan AR et al: Development of a health-related quality of life instrument for use in children with spina bifida. *Qual Life Res* 1997; **6**: 123.
12. Padua L, Rendeli C, Rabini A, Girardi E, Tonali P and Salvaggio E: Health-related quality of life and disability in young patients with spina bifida. *Arch Phys Med Rehabil* 2002; **83**: 1384.
13. Rendeli C, Ausili E, Tabacco F, Caliendo P, Aprile I, Tonali P et al: Assessment of health status in children with spina bifida. *Spinal Cord* 2005; **43**: 230.
14. Verhoef M, Barf HA, Post MW, van Asbeck FW, Gooskens RH and Prevo AJ: Functional independence among young adults with spina bifida, in relation to hydrocephalus and level of lesion. *Dev Med Child Neurol* 2006; **48**: 114.
15. Herndon CD, Rink RC, Cain MP, Lerner M, Kaefer M, Yerkes E et al: In situ Malone antegrade continence enema in 127 patients: a 6-year experience. *J Urol* 2004; **172**: 1689.
16. Aksnes G, Diseth TH, Helseth A, Edwin B, Stange M, Aafos G et al: Appendicostomy for antegrade enema: effects on somatic and psychosocial functioning in children with myelomeningocele. *Pediatrics* 2002; **109**: 484.
17. Bau MO, Younes S, Aupy A, Bernuy M, Rouffet MJ, Yepremian D et al: The Malone antegrade colonic enema isolated or associated with urological incontinence procedures: evaluation from patient point of view. *J Urol* 2001; **165**: 2399.
18. Ellsworth PI, Webb HW, Crump JM, Barraza MA, Stevens PS and Mesrobian HG: The Malone antegrade colonic enema enhances the quality of life in children undergoing urological incontinence procedures. *J Urol* 1996; **155**: 1416.
19. Yerkes EB, Cain MP, King S, Brei T, Kaefer M, Casale AJ et al: The Malone antegrade continence enema procedure: quality of life and family perspective. *J Urol* 2003; **169**: 320.
20. Parekh AD, Trusler LA, Pietsch JB, Byrne DW, DeMarco RT, Pope JC et al: Prospective, longitudinal evaluation of health related quality of life in the pediatric spina bifida population undergoing reconstructive urological surgery. *J Urol* 2006; **176**: 1878.

DISCUSSION

Dr. Bill Strand. Since predictability and bowel management sometimes can be achieved with a good program or with severe constipation, are you going to correlate your questionnaire results with some other measure of clinical control of the constipation to discern which ones are most happy with the program, whether it is those working hard or not?

Dr. Dana Nanigian. Some of the patients in the spina bifida group who completed the survey on the questionnaire had controlled constipation to the point that they were so constipated the incontinence was not bad. We gave the survey to all of our spina bifida patients, and so there is going to be a significant number of those who are happy with the bowel care and not incontinent. The entire group had spina bifida but not everyone was incontinent. We had no exclusion criteria.