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Validation of a bowel dysfunction instrument for adolescents with spina bifida

Katherine Hubert [a], Georgios Sideridis [b], Rebecca Sherlock [a], Jennifer Queally [c], Ilina Rosoklija [a], Greta Kringle [a], Kathryn Johnson [a], Samuel Nurko [d], Stuart Bauer [a]

[a]Department of Urology, Boston Children's Hospital, Harvard Medical School, Boston, MA, USA

[b]Clinical Research Program, Boston Children's Hospital, Harvard Medical School, Boston, MA, USA

[c]Department of Psychiatry, Boston Children's Hospital, Harvard Medical School, Boston, MA, USA

[d]Division of Gastroenterology, Hepatology & Nutrition, Boston Children's Hospital, Harvard Medical School, Boston, MA, USA

Correspondence to:
K. Hubert, 705 Riley Hospital Drive, Division of Pediatric Urology, Riley Outpatient Center, 4230, Indianapolis, IN 46202, USA

kchubert@iupui.edu
(K. Hubert)

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Summary

Introduction

Existing survey instruments for bowel dysfunction in the pediatric population are either parent-reported or focus on non-neurogenic bowel dysfunction.

Objective

The purpose of this study was to develop and validate an adolescent-reported survey to assess the severity of bowel dysfunction in spina bifida patients and examine its impact on quality of life (QOL).

Study design

We performed a cross-sectional study of patients in our Myelodysplasia Program, aged 11—17 years, with a history of constipation and/or fecal incontinence (FI) from November 2010 to June 2013. Control patients, aged 11—17 years, were recruited from the stone clinic. Exclusion criteria were lack of English fluency, insufficient reading skills, or an incontinent fecal diversion. A 29-item version of the Adolescent Fecal Incontinence and Constipation Symptom Index (A-FICSI) was developed with five domains (Figure). Test re-test reliability and correlation with the total global health-related QOL score from the Parkin survey were measured using the Pearson correlation coefficient. A factor analysis model with four-fold correlations was tested.

Results

Of the 65 study-eligible individuals approached, 25 (11 boys) completed the A-FICSI (median age 12.6 years, IQR 11.8—14.7 years) and 17 completed another survey on urinary incontinence (64.6% response rate). Twenty-one control patients with nephrolithiasis completed the A-FICSI. Nine of the 25 completed a second administration of the survey. The mean correlation between repeated administrations of the survey was $r = 0.43$. There was a significant negative correlation between severity of constipation ($r = -0.299$, $p < 0.05$) and severity of FI ($r = -0.316$, $p < 0.05$) with the total Parkin QOL score. The Comparative Fit Index (CFI) was 0.864. Most items loaded significantly on their respective factors. Between-factor correlations were all significant (>0.30) in the predicted direction. Unstandardized residuals were 8.7% (95% CI 6.4—10.9%). Item reduction was performed on the 29-item instrument based on results of the factor analysis. The finalized instrument contained 21 items.

Discussion

This is the first adolescent-reported bowel dysfunction instrument to undergo formal psychometric assessment in the spina bifida population. The instrument demonstrated adequate reliability and the five-factor structure fit the data well. This study highlights the negative impact of bowel dysfunction on the QOL of spina bifida patients. It is limited by the moderate sample size that is a common issue in relatively rare diseases.

Conclusion

The A-FICSI possesses desirable psychometric properties for the measurement of bowel dysfunction in the spina bifida population.

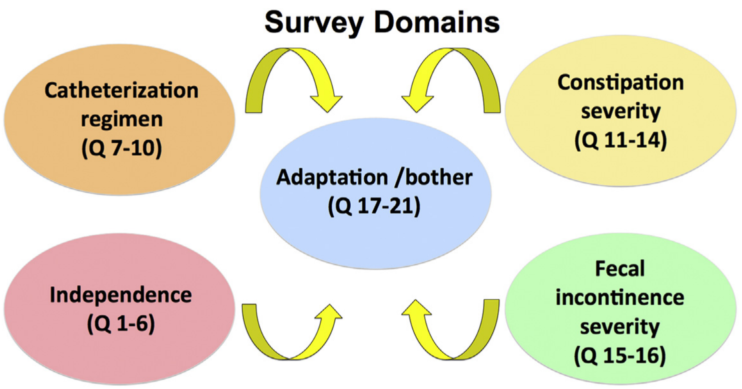

Figure Conceptual model of the Adolescent Fecal Incontinence and Constipation Symptom Index.

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Introduction

Children with spina bifida are reported to have a high frequency of neuropathic bowel problems (~78%) [1]. Problems related to the neuropathic bowel and its management include abdominal pain and distention, constipation, fecal incontinence (FI), prolonged duration of bowel management, autonomic dysreflexia, hemorrhoids, anal fissures, and rectal prolapsed [2]. Neuropathic bowel dysfunction also has a significant impact on social activities and/or quality of life (QOL) [3]. In an interview study of 134 adult spina bifida patients in Sweden, 51% reported daytime fecal leakage, although none had support to improve their bowel regimen [3].

Scientific evaluation of the impact of bowel dysfunction on QOL mandates the use of instruments that have undergone rigorous psychometric assessment. This may inform patient-centered decision-making among treatment alternatives and lead to improved QOL. To our knowledge, there are only two QOL questionnaires that are specific to the pediatric spina bifida population [4,5]. The survey developed by Parkin *et al.* is the "gold standard" measurement of multi-dimensional, health-related QOL for the pediatric spina bifida population [4]. One of the limitations of the Parkin questionnaire, however, is that does not inquire about relationships in the family or the specifics of bowel care [4]. The Fecal Incontinence and Constipation Quality of Life (FICQOL) Scale is a quality of life measure for children with spina bifida that is administered to the patient's caregiver [5]. Adult FI questionnaires and those developed for children with functional constipation and FI are not applicable to adolescents with spina bifida [6–8]. The purpose of this study was to develop an adolescent-reported survey, the Adolescent Fecal Incontinence and Constipation Symptom Index (A-FICSI), to assess the severity of bowel dysfunction in spina bifida patients and evaluate its reliability and factorial validity.

Materials and methods

Measures

The final A-FICSI is a 21-item, Likert-scaled instrument comprising five domains or factors: 1) independence, 2) catheterization regimen, 3) constipation severity, 4) FI severity, and 5) adaptation/bother (Appendix A, supplementary material). It was created using the Incontinence Symptom Index-Pediatric (ISI-P) as a template for its structure and question format [9]. In addition, a number of items regarding quantification of FI were borrowed from an adult self-report questionnaire that is used to measure FI and its risk factors in the community [10]. Input was also obtained from professional experts in the field (nursing, urology, gastroenterology, psychology). Initially, an expert interviewer performed focused key informant interviews with five patients and their respective caregivers to ensure the clarity and adequacy of the survey instrument. We also elicited suggestions for additional survey topics from each of the patient/caregiver dyads. We revised the instrument based on feedback from these families with the addition of two questions about dietary issues and sleepovers. The initial 29-item survey instrument contained questions specific to therapeutic interventions such as urinary catheterization, bowel regimen, and medication management (Appendix B, supplementary material). We included questions about medication management (relevant to a patient's bowel regimen) and urinary catheterization to provide an assessment of patient independence. The question about bladder emptying at school provides insight into patient independence outside of the home setting as patients are more likely to catheterize their bladder at school than perform an enema. We included questions related to both urinary incontinence and constipation/FI as these two conditions frequently co-exist in patients with spina bifida. Subjects also completed the Parkin questionnaire, a health-related QOL questionnaire for spina bifida patients [4]. This survey includes the following domains: social, emotional, intellectual, financial, medical, independence, environmental, physical functioning, recreational, and vocational.

Study design

We performed a cross-sectional cohort study of patients in our Spina Bifida Center from November 2010 to June 2013. Adolescents aged 11–17 with a history of FI and/or constipation and their caregivers were recruited during routine clinic visits. Control subjects, aged 11–17 without FI or constipation were recruited from the Pediatric Kidney Stone Clinic. Exclusion criteria were lack of English fluency (verbal or written), insufficient reading skills (IQ < 70 or <4th grade reading level), or an incontinent fecal diversion. The A-FICSI and Parkin questionnaires were self-administered to patients in a web-based format using handheld tablet devices. Patients were permitted to ask for assistance from their respective caregivers in completing the survey. An electronic mail with an embedded link to a web-based survey (A-FICSI only) was sent to a randomly selected subset of subjects 2 weeks after the initial survey administration in the clinic. Institutional Review Board approval was obtained. Psychometric assessment including reliability and factorial validity was performed. The responses of subjects who completed less than 50% of the survey were excluded from analysis. Missing data were first analyzed to detect their missing pattern and then were imputed using a discriminant analysis predictive model using the SOLAS software. Missing data ranged between 5% and 42%.

Data analysis

Validity of the A-FICSI

The validity of the A-FICSI was assessed using face, content, criterion-related, and construct validity. We assessed face validity during the key informant interview stage. We assessed content validity using an expert panel consisting of a pediatric urologist, a pediatric gastroenterologist, and a developmental pediatrician. This type of validity involves expert judgement of whether the instrument samples all of the important content or domains. We assessed criterion-