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Urinary incontinence in spina bifida: Initial instrument validation

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ABSTRACT

The purpose of this study was to perform a psychometric assessment of the Incontinence Symptom Index-Pediatric (ISI-P) in a cohort of adolescents with spina bifida (SB) and neuropathic urinary incontinence (UI) to test its validity and reliability. The ISI-P, an 11-item instrument with domains for symptom severity and impairment, was self-administered by subjects 11–17 years old with SB and UI. Controls were 11–17 years old, with nephrolithiasis and no history of UI. Formal psychometric assessment included an evaluation of internal consistency, test re-test reliability and factor analysis. Of 78 study-eligible subjects we attempted to contact, 33 (66.7% female) with a median age of 13.1 years completed the ISI-P (42.3% response rate). 21 control patients also completed the ISI-P. Cronbach's alpha was 0.936 and 0.792 for the severity and bother factors respectively. The delta Chi-square test for the two-factor (vs. one-factor) model was significantly [$\chi^2(89) = 107.823$, $p < 0.05$] in favor of the former model with descriptive fit indices being excellent (e.g., comparative fit index = 0.969). Furthermore, category information analysis showed that all categories were associated with different threshold values, namely that each category contributed unique information for the measurement of the latent trait. In conclusion, the ISI-P has desirable psychometric properties for the measurement of UI symptom severity and impairment in adolescents with SB.

1. Introduction

Daytime urinary incontinence (UI) is a common condition with an estimated prevalence in school-age children of 16.9% (Sureshkumar, Jones, Cumming, & Craig, 2009). There is evidence to suggest that the prevalence of UI significantly decreases with age (Kyrklund, Taskinen, Rintala, & Pakarinen, 2012). A large, cross-sectional survey of 10–14 year-old Belgian school children noted a prevalence of 9% (Bakker, van Sprundel, van der Auwera, van Gool, & Wyndaele, 2002). It is well established that UI is associated with diminished self-esteem, quality of life and comorbid clinical behavioral disorders (Butler, 1998; von Gontard et al., 1997; von Gontard, Mauer-Mucke, Pluck, Berner, & Lehmkuhl, 1999; von Gontard, Baeyens, Van Hoecke,

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Warzak, & Bachmann, 2011). Children with daytime UI have significantly increased rates of psychological problems including separation anxiety (11.4%), attention deficit (24.8%), oppositional behavior (10.9%) and conduct problems (11.8%) (Joinson, Heron, & von Gontard, 2006). UI is more common in children with developmental disabilities, such as spina bifida, than in typically developing children and has a significant negative impact on childhood quality of life (Frimberger, Cheng, & Kropp, 2012; von Gontard, 2013). In a parental survey of 433 children with spina bifida, 77% reported that UI was considered a moderate or severe stress factor for their children (Lie et al., 1991).

Scientific evaluation of the impact of UI on quality of life mandates the use of instruments that have undergone rigorous psychometric evaluation and meet all requirements of validity. Information on the psychosocial impact of UI is useful for quantifying the benefits or adverse effects of treatment. This can greatly inform decision-making among treatment alternatives and lead to improved quality of life. Our ability to quantify UI symptoms and bother in children with spina bifida, however, is limited by a lack of available patient-reported survey instruments that have undergone formal psychometric assessment (Vajda et al., 2009).

A number of instruments to assess UI in children without developmental disabilities have been developed (Afshar, Mirbagheri, Scott, & MacNeily, 2009; Akbal, Genc, Burgu, Ozden, & Tekgul, 2005; Bower, Wong, & Yeung, 2006b; De Gennaro et al., 2010; Farhat et al., 2000; Nelson et al., 2007). The Pediatric Incontinence Questionnaire (PinQ) is a cross-cultural, quality of life measurement that has undergone extensive psychometric assessment in children ages 6–16 with daytime UI and nocturnal enuresis (Bower, Sit, Bluyssen, Wong, & Yeung, 2006a; Bower et al., 2006b). The six domains of this scale include social relations with peers, self-esteem, family and home, body image, independence and mental health. Although the PinQ has been thoroughly investigated in children with UI, it has never been validated in patients with neuropathic bladder dysfunction. The dysfunctional voiding scoring system (DVSS) quantifies or grades the severity of abnormal voiding in children ages 3–10 years with daytime UI, abnormal voiding habits and/or recurrent urinary tract infections (Farhat et al., 2000). Although its validity was assessed, reliability was not. Furthermore, the study excluded patients with anatomic or neuropathic bladder dysfunction.

The Incontinence Symptom Index-Pediatric (ISI-P) is a pediatric UI instrument that is based on an adult version of the questionnaire, the incontinence symptom index (Wei, Hoag, Faerber, Dorr, & McGuire, 2003). The original instrument was altered to include a domain of insensate UI symptoms relevant to patients with neuropathic UI. Although it includes a neuropathic bladder domain, it was validated in a cohort of adolescents with primarily non-neuropathic etiologies of UI. Given that the psychometric properties of an instrument may not remain stable across populations, the ISI-P requires re-validation in a cohort of patients with neuropathic bladder dysfunction, such as those with spina bifida, prior to widespread use in this population. The purpose of the present study is to evaluate the psychometric properties of the ISI-P in cohort of spina bifida adolescents with neuropathic bladder dysfunction and UI in order to test its reliability and validity. The psychometric assessment consisted of internal consistency, test re-test reliability, confirmatory factor analysis and item response theory. The study tests the hypothesis that a two-factor model, consisting of UI severity and adaptation/bother factors, is the best fit for the data and therefore is the optimal structure for using the ISI-P to score health-related quality of life in adolescent patients with spina bifida.

2. Materials and methods

2.1. Study design

We performed a cross-sectional cohort study of patients in our Spina Bifida Center from November 2010 to June 2013. We recruited patients, ages 11–17, with a history of spina bifida and UI during routine clinic visits. We recruited control patients, ages 11–17, without UI from the Pediatric Kidney Stone Clinic. Patients with acute renal colic, obstructing stones or renal stents were not included as these patients are not seen in the stone clinic. Exclusion criteria were lack of English fluency (verbal or written), insufficient reading skills (IQ $<$ 70 or $<$5th grade reading level), absence of the legal guardian or an incontinent urinary diversion or presence of an indwelling Foley catheter. Patients self-administered the ISI-P in a web-based format using handheld tablet devices. Patients were permitted to ask for assistance from their respective caregivers in completing the survey. We performed psychometric assessment including reliability and factorial validity. The Institutional Review Board approved the study based on both ethical and methodological considerations.

2.2. Measures

The ISI-P is an 11-item instrument comprising two scales: UI symptom severity and impairment (Appendix A) (Nelson et al., 2007). It includes subdomains for stress UI, urge UI, insensate UI, nocturnal UI and pad use. A certified literacy expert provided input on the language of the instrument that was modified to a fifth-grade reading level. Formal validation in the pilot study demonstrated good internal consistency with a Cronbach's alpha of 0.84 for the complete instrument. Item-scale correlations were $>$0.60 for all except one item. Test–retest reliability was good with an $r = 0.97$ ($p < 0.001$). The instrument also had good discriminative validity with a total severity scale score of 9.3 in wet children and 0.7 in controls ($p < 0.0001$). Impairment scale scores differed by 2.2 points ($p < 0.001$). The mean scores were significantly different between subscales for all domains except for pad use.