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**The Toileting Habit Profile Questionnaire-Revised; Examining Discriminative and
Concurrent Validity**

Abstract

Sensory over-reactivity may be related to atypical defecation habits in children with constipation. The Toileting Habit Profile Questionnaire-Revised (THPQ-R) is designed to identify defecation-related sensory issues. This study examined discriminative and concurrent validity of the THPQ-R. Differentiating between children with and without constipation was used to establish discriminative validity. The relationship between scores on the THPQ-R and on the **defecation specific scale of the** Virginia Encopresis-Constipation Apperception Test (VECAT-**S**) was examined to establish concurrent validity. The difference in THPQ-R scores **between children with and without constipation** was statistically significant. There was a strong positive correlation between THPQ-R and VECAT scores. Results support using the THPQ-R to identify atypical defecation behaviors.

Background

Bowel management is an important activity of daily living and is key to an individual's independence, successful social participation and quality of life (American Occupational Therapy Association, 2014; Kovacic et al., 2015). Furthermore, acquiring continence of bowel is an important milestone of childhood. These two features of bowel management place it as a core component of occupational therapy (OT) practice.

Childhood defecation difficulties are complex and often result from an interaction between physiological, social and behavioral processes (Freeman, Riley, Duke, & Fu, 2014). In many cases, a specific organic cause cannot be identified, and the defecation disorder is considered functional (Tabbers et al., 2014). Functional defecation disorders (FDD) include functional constipation (FC) and functional non-retentive fecal incontinence (FNRFI) (The Rome Foundation, 2016). Fecal incontinence and challenging behaviors that interfere with age appropriate bowel management, are common to both children with FC (a result of fecal overflow) and those with FNRFI (Beaudry-Bellefeuille, Booth, & Lane, 2017). Children with long lasting symptoms of FDD are at risk for experiencing lower health-related quality of life (Kovacic et al., 2015). Moreover, childhood FDD are highly prevalent worldwide (0,7 – 29%; Mugie, Benninga, & Di Lorenzo, 2011) and are increasingly considered a public health problem (Rajindrajith, Devanarayana, Perera, Benninga, 2016). Finally, despite extensive reports of gastrointestinal and behavioral considerations in children with FDD (Pijpers, Bongers, Benninga, & Berger, 2010; Tabbers et al., 2014; Van Ginkel et al., 2003), gaps remain in our understanding of the factors involved in the emergence of FDD and the salient treatment elements to optimize outcomes (Freeman et al., 2014; Rajindrajith et al., 2016).

Reports support the hypothesis that issues in sensory reactivity (i.e. the process of modulating neuronal activity in response to sensory stimuli) may be related to atypical defecation habits in some children with FDD (Beaudry-Bellefeuille & Lane, 2017; Beaudry

Bellefeuille & Ramos Polo, 2011; Beaudry, Schaaf, & Ramos, 2013). For instance, using the Short Sensory Profile (McIntosh, Miller, Shyu, & Dunn, 1999), Beaudry-Bellefeuille and Lane (2017) reported significantly more sensory over-reactivity in children with FC than in typically developing children. Pollock (Pollock, Metz, & Barabash, 2014) studied the prevalence of sensory reactivity issues in children with dysfunctional elimination syndrome (DES), a diagnosis that includes a variety of urinary and bowel elimination difficulties (Neveus et al., 2006). The study revealed that 52.6% of participants with DES had sensory reactivity issues compared to 7.3% of the control group (n=55). Furthermore, preliminary reports of the effectiveness of OT interventions which consider the sensory issues of children with FDD are promising (Beaudry Bellefeuille & Ramos Polo, 2011; Beaudry et al., 2013). Clinical practice in this area, however, is limited by the lack of validated measures that can clearly identify defecation-related sensory issues which may be impacting participation in healthy age-appropriate toileting routines in children with FDD.

Currently, tools available for assessing and diagnosing defecation disorders consider behavioral characteristics, gastrointestinal symptoms or the impact of FDD on quality of life. For example, the Virginia Encopresis-Constipation Apperception Test (VECAT; Cox et al., 2003) focuses on the behavioral aspects of FDD. Other diagnostic tools such as abdominal X-Rays, transabdominal ultrasound (Tabbers et al., 2014) or Rome Foundation diagnostic criteria (The Rome Foundation, 2016) assess gastrointestinal symptoms. Finally, tools such as the Questionnaire on Pediatric Gastrointestinal Symptoms (Caplan, Walker, & Rasquin, 2005) assess the functional impairment associated with pediatric FDD. **Moreover, tools used to assess sensory reactivity consider reactions to sensory input in the context of a variety of activities of daily life but do not usually consider reactions to sensory input in the context of bowel management.** In order to further examine sensory reactivity in children with FDD there

is a need to validate assessment tools which focus on defecation-related sensory reactivity issues in this population.

The Toileting Habit Profile Questionnaire-Revised (THPQ-R) (Beaudry-Bellefeuille, Bundy, Lane, A., Ramos-Polo, Lane, S., 2018) is a screening questionnaire to help identify defecation behaviors and reactions related to sensory over-reactivity. This tool was originally developed through a collaboration between an occupational therapist and a gastroenterologist who observed that response to defecation related sensations appeared to influence acceptance of toilet training as well as the response to the urge to defecate in some children (Beaudry Bellefeuille & Ramos Polo, 2011; Beaudry et al., 2013). The Toileting Habit Profile Questionnaire (THPQ) emerged from available literature, clinical experience, and caregiver description of behaviors common in children with difficulties participating in toileting routines (Beaudry-Bellefeuille, Lane, & Ramos-Polo, 2016). More recently the construct validity of the THPQ was re-evaluated, resulting in a revised version of the tool (THPQ-R) designed to identify manifestations of sensory over-reactivity in children with FDD (Beaudry-Bellefeuille et al., 2018).

While preliminary studies regarding the THPQ and THPQ-R are promising, further examination of THPQ-R characteristics is needed before recommending its use in clinical practice. Current views on validity testing recommend a multidimensional perspective, drawing from diverse sources of evidence such as: 1) examination of the content of the measure, 2) the response patterns of data collected with the measure and, 3) the relationship of the measure under question to other instruments tapping into similar constructs, or known-group comparison studies to test hypotheses about expected differences in scores across groups (Goodwin & Leech, 2003). Previous examinations of the THPQ and the THPQ-R content and construct validity have drawn from evidence based on the content of the questionnaire using expert review and directed content analysis of recent literature for

verification of item relevancy (Beaudry-Bellefeuille et al., 2016; Beaudry-Bellefeuille et al., 2018). Additionally, we used Rasch analysis of response patterns of data collected with the THPQ-R as a means of examining construct validity (Beaudry-Bellefeuille et al., 2018). Consistent with the recommendation of Goodwin and Leech (2003), and with our aim to further the process of establishing validity of the THPQ-R, the current study examines discriminative and concurrent validity of the THPQ-R.

Methods

In addressing the aim of establishing discriminative validity, we determined if the THPQ-R could differentiate between children with and without FDD. The relationship between the responses given on the THPQ-R and those given on the defecation specific items of the VECAT (VECAT-S) were examined as a means of exploring concurrent validity.

Participants: Caregivers of children with and without FDD were recruited to the study. Participants whose children had FDD were eligible if they belonged to one of the following categories: 1) Parents of children aged 3 to 6 years old with FDD and no additional diagnosis; or 2) Parents of children with autism spectrum disorder (ASD) or/and attention deficit hyperactivity disorder (ADHD) aged 3 to 6 years old, identified by parent report of diagnosis, with FDD. We included children with ASD or ADHD due to the currently reported high prevalence of FDD in children with these diagnoses (Ibrahim et al., 2009; McKeown, Hisle-Gorman, Eide, Gorman, & Nylund, 2013). We chose the age range of 3–6 years because it is when symptoms such as feeling pain upon defecation or toilet refusal appear (Borowitz, Cox, & Sutphen, 1999), and ongoing toileting concerns become apparent (Schum et al., 2002; Wald et al., 2009). Parents of children with organic causes of defecation issues were excluded.

The control group consisted of parents of children in the same age range, without FDD and no additional diagnosis, or without FDD and a diagnosis of ASD or/and ADHD. FDD

(FC and FNRFI) was verified or ruled out using probe questions based on the Rome Foundation diagnostic criteria for FDD (The Rome Foundation, 2016).

Apart from parents of children with a diagnosis of ASD or ADHD, parents with children who had a curricular adaptation at school, who qualified for their school's special needs program, or who had been referred to early intervention or rehabilitation programs were excluded to insure that FC was not confounded by other diagnoses. Participants whose children had not yet initiated toilet training were also excluded given that both the THPQ-R and the VECAT-S are concerned with toileting. Initiation of toilet training was defined as asking the child to use the potty or toilet at least 3 times a day regardless of continence or use of a diaper.

Parents were recruited through parent support groups and through social media. Pediatric gastroenterologists and occupational therapists from both public and private clinics were also contacted for recruitment of parents of children diagnosed with FDD and/or children with ASD. Snowball recruitment was permitted. Recruitment was aimed at several English and Spanish speaking countries.

Data collection: Participants were sent a link to a series of online questionnaires. We collected responses to the questionnaires using a web-based survey tool (Qualtrics®; Qualtrics, 2017). Survey quality control was implemented to identify and exclude multiple entries and inconsistent reporting: (1) internet protocol address check; (2) e-mail invitation to the survey only after interested participants contacted the researcher; (3) exclusion of respondents who were not consistent on Rome Foundation questions or showed other evidence of indiscriminate responding. For example the question related to stool withholding is presented as part of the questions meant to identify FC and again with the questions designed to identify FNRFI. A parent is expected to give the same response in both sections.

Measures: *Toileting Habit Profile Questionnaire Revised (THPQ-R)*: The THPQ-R (Beaudry-Bellefeuille et al., 2018) is a revised version of the THPQ (Beaudry-Bellefeuille et al., 2016), a bilingual (English-Spanish) parent report questionnaire designed as a screening tool to help differentiate typical defecation behaviors and reactions from those that are associated with FDD and potentially related to sensory reactivity. There are 17 items (see Table 1) scored using a dichotomous scale (1=frequently or always; 2=never or rarely). For our analysis we used the 15 sensory over-reactivity items as recommended by the authors.

Probe questions based on Rome IV diagnostic criteria of FDD: The Rome Foundation is a non-profit organization dedicated to the creation of scientific evidence to assist in the diagnosis and treatment of functional gastrointestinal disorders (FGIDs). The Rome Foundation has developed diagnostic criteria concerning FGIDs. Probe questions based on the most recent diagnostic criteria, Rome IV (The Rome Foundation, 2016), were used to verify diagnosis of FDD. A child with two or more FC symptoms was included in the FC diagnostic group.

Virginia Encopresis-Constipation Apperception Test: The VECAT identifies toilet and bowel-specific behavior issues and can identify when these issues reflect more general behavior problems (Cox et al., 2003). The VECAT is a picture-based test which includes nine pairs of bowel-specific pictures and nine pairs of generic pictures addressing similar concerns. For example, the first pair of bowel-specific pictures refers to anticipation of pain versus relief upon expelling a stool, while the generic pair of pictures refers to anticipation of pain versus relief following the removal of a splinter. It can be administered to the parent or/and to the child. Respondents select the picture in each pair that best describes them or their child and then qualify if they or their child are little bit or a lot like the picture. In their study of validity of the VECAT with children with encopresis, investigators found that, when using the mothers' ratings, 87% of the children were correctly classified as patients or

controls (Cox et al., 2003). Additionally, they found the bowel-specific subscale (VECAT-S) to be sensitive to successful treatment intervention.

In order to adapt the VECAT for a Spanish speaking population a back-translation process was performed. The resulting Spanish version was presented to 11 parents of children with FDD to ensure comprehension and cultural adequacy. All agreed the items were adequate and did not indicate the need for any changes.

Data analysis: To establish discriminative validity, between-group comparison (control group and FDD group) of the THPQ-R scores was conducted using an independent-samples t-test. The difference was considered statistically significant at $p < .05$. Given that scores based on ordinal data were transformed into interval data, a parametric statistical test was chosen.

In our previous work with the THPQ-R, data from children with FDD subjected to Rasch analysis was found to effectively fit the Rasch model (Beaudry-Bellefeuille et al., 2018). Rasch analysis allows us to transform raw score data obtained from an ordinal scale (the THPQ-R) into equal interval data expressed in Log Odd Units (logits), the unit of Rasch measurement (Boone, Staver, & Yale, 2013). Item difficulty values obtained from this previous work were used to calibrate the analyses of the current data which was collected from children with and without FDD.

Using data from both the THPQ-R and the VECAT-S, a correlation coefficient was calculated to examine the concurrent validity of the THPQ-R. Results on the THPQ-R were not expected to distribute normally, based on pilot study data with the THPQ, therefore the Spearman correlation coefficient was calculated to analyze the relationships between the VECAT-S score and the THPQ-R score. To further examine the relationship between the THPQ-R and VECAT-S scores, linear regression analysis was performed.

Results

Participants

One hundred and ten caregivers participated in the study; 60 of whom had children with FDD. All children had FC; the recruitment process did not yield any participants with FNRFI. Participants' children details are presented in Table 2.

Discriminative validity of the THPQ-R

An independent-samples t-test was run to determine if there were differences in total THPQ-R score between children with and without FC. The minimum raw score on the THPQ-R is 15 and the maximum raw score is 30; based on Rasch transformation of ordinal scores into interval data, this is equivalent to -3.64 logits and 4.69 logits, respectively (see Table 3 for raw score Rasch measure equivalency). There were two significant outliers (unusually low scores) in the no FC group. We chose to keep the outliers in our analysis as clinical experience has shown that some children show atypical defecation behavior in the absence of FDD and removing the outliers would increase the mean for this group, widening the difference between groups. THPQ-R score was not normally distributed, as assessed by Shapiro-Wilk's test ($p < .05$). However, the independent-samples t-test can be considered robust to non-normality and we chose to carry out the test (Laerd Statistics, 2015). There was homogeneity of variance for THPQ-R scores for children with and without FC, as assessed by Levene's test for equality of variances ($p = .302$). As expected, mean THPQ-R score for children without FC (3.89 logits) was 1.65 (95% CI, 1.17 to 2.12) higher than mean THPQ-R score for children with FC (2.24 logits). The difference in mean THPQ-R score between children with FC and those without FC was statistically significant $t(108) = -6.809$, $p < .0005$.

Concurrent validity of the THPQ-R

An independent-samples t-test was run to determine if there were differences in mean VECAT-S scores in children with and without FC. The assumption of homogeneity of

variances was violated, as assessed by Levene's test for equality of variances ($p = .001$). As expected the mean VECAT-S score (3.38) in children with FC was 6.38 (95% CI, 4.40 to 8.35) lower than mean VECAT-S score (9.76) in children without FC. The difference in mean VECAT-S score between children with FC and those without FC was statistically significant, $t(99.035) = 6.41$, $p < .0005$. A Spearman's rank-order correlation was run to assess the relationship between THPQ-R total scores and VECAT-S scores in 110 children aged 3 to 6 years. Preliminary analysis showed the relationship to be monotonic, as assessed by visual inspection of a scatterplot (see Figure 1). As expected there was a strong positive correlation between THPQ-R scores and VECAT-S scores in children aged 3 to 6 years, $r_s = .644$, $p < .0005$, indicating the tools are measuring similar constructs.

As planned, simple linear regression was carried out to further examine and understand the relationship between VECAT-S scores and THPQ-R scores. Linearity was established by visual inspection of a scatterplot. There was independence of residuals, as assessed by a Durbin-Watson statistic of 1.727. There were no significant outliers with all cases having standardized residuals less than ± 3 . There was homoscedasticity, as assessed by visual inspection of a plot of standardized residuals versus standardized predicted values. Residuals were normally distributed as assessed by visual inspection of a normal probability plot. The linear regression model showed good fit for the data with average VECAT-S score accounting for 40.6% of the variation in THPQ-R score with adjusted $R^2 = 40.0\%$, a large size effect according to Cohen (1988). The analysis confirmed a linear relationship between VECAT-S scores and THPQ-R scores with a coefficient for VECAT-S score of .152 (95% CI, .117 to .187; $p < .0005$). Average VECAT-S score statistically significantly predicted THPQ-R score, $F(1, 108) = 73.77$, $p < .0005$.

Discussion

The construct of sensory over-reactivity is well established (Dunn, 2014; Lane, Reynolds, & Thacker, 2010; Parham & Ecker, 2007; Reynolds, Bendixen, Lawrence, & Lane, 2011; Su & Parham, 2014) and the use of caregiver questionnaires has become an accepted way to document its presence. However, available tools do not address sensory reactivity issues related to defecation, a crucial childhood occupation. The THPQ-R fills this gap (Beaudry-Bellefeuille et al., 2018). Here we document the discriminative validity of the THPQ-R. Our previous work had provided preliminary support for the discriminative validity of an earlier version of this questionnaire (Beaudry-Bellefeuille et al., 2016) in distinguishing between the defecation behaviors of typically developing children and those of children with retentive fecal incontinence who had not responded to conventional medical management (Beaudry-Bellefeuille & Lane, 2017). The present study adds support to the discriminative validity of the revised version of this tool and does so with children with FC, a much broader and more common diagnosis, thus increasing its relevance in research and clinical practice.

The VECAT-S served as basis for the examination of the concurrent validity of the THPQ-R. As expected we found that scores on both assessments were significantly lower for children with FC than for children without FC, supporting earlier validity studies with the VECAT (Cox et al., 2003). We add to the literature our new finding of a strong positive correlation between the THPQ-R and the VECAT-S, indicating that both tools have the capacity to tap into the construct of defecation specific behavior. However, the THPQ-R differs from the VECAT-S in that the defecation specific behaviors included in the THPQ-R have been carefully selected to help clinicians and researchers gain insight into potential sensory reactivity issues that may be impacting participation in healthy age appropriate toileting routines; the VECAT-S does not address sensory reactivity.

Evidence supporting the use of OT in improving participation in children with sensory issues and ASD is growing (Pfeiffer, Koenig, Kinnealy, Sheppard, & Henderson, 2011;

Omairi, 2018; Schaaf et al., 2014). There is also some evidence that addressing the sensory reactivity issues that appear to underlie the behaviors related to the development and maintenance of FDD may contribute to more successful treatment outcomes for children who experience these complex and often chronic conditions (Beaudry-Bellefeuille & Ramos-Polo, 2011; Beaudry et al., 2013; Handley-More, Richards, Macauley, & Tierra, 2009). Our current work supports the use of the THPQ-R in identification of atypical defecation behaviors; behaviors which previous work has identified as possible manifestations of sensory over-reactivity in children with FDD (Beaudry-Bellefeuille et al., 2018; Beaudry-Bellefeuille & Lane, 2017).

The main limitation to this study lies in the fact that a single FDD (FC) was examined when considering discriminative and concurrent validity. However, given that pediatric FC is highly prevalent and considered a global health issue, having tools validated with this diagnosis is of utmost importance.

Additionally, we must consider that only one diagnostic group (ASD) was included. Other diagnoses, in particular ADHD, are known to be associated with elevated prevalence of FDD (McKeown et al., 2013) and warrant investigation. While the current study was open to parents of children with ADHD, no parents of children with this diagnosis participated. The limited age range (3 to 6 yrs) is another factor to be considered as this may have contributed to the absence of parents of children with ADHD; this diagnosis tends to be given around age 6 or older (Atladottir et al., 2015). It is recognized that older children also experience FDD. However, we chose to focus on younger children given that the aim of the study was to validate the THPQ-R, a tool based on observation of behavior, and that behavioral manifestation of FDD appear to be more significant at younger ages (Borowitz et al., 1999). Finally, no tool that covers both sensory and bowel concerns was available, therefore

concurrent validity was conducted with the VECAT-S, which we considered the best available tool.

Future research is needed to strengthen the validity of the THPQ-R with a wider variety of defecation diagnoses and its proposed usefulness in identifying and tracking defecation behaviors related to sensory reactivity issues.

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