Original Article

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Examining Hyper-Reactivity to Defecation Related Sensations in Children with Functional Defecation Disorders

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Abstract

Background: Adequate sensory perception and reactivity to sensory stimuli associated with defecation is key to successful stool toileting. Preliminary reports suggests that some of the difficulties that many children with FDD experience with toileting could be related to sensory hyper-reactivity. Objective: This study investigated the relationship between sensory hyper-reactivity and functional defecation disorders (FDD).
Methods: Parents of three to six-year-old children with and without FDD completed two questionnaires; the Toileting Habit Profile Questionnaire-Revised (THPQ-R; tool that measures sensory hyper-reactivity to defecation related sensations) and the Short Sensory Profile (SSP). On both questionnaires, low scores indicate more concerns. Between group comparisons and the relationship between scores on the THPQ-R and on the sensory hyper-reactivity items of the SSP were examined.
Results: The sensory hyper-reactivity mean score of the SSP was lower for children with FDD. The difference was statistically significant ($P<0.0005$). There was a positive correlation between THPQ-R and SSP hyper-reactivity scores ($r274=0.485$, $P<0.0005$). Further, higher levels of sensory hyperreactivity (low SSP score) were associated with a higher frequency of the challenging defecation behaviors described in the THPQ-R (low THPQ-R score).
Conclusion: Health practitioners do not usually consider sensory hyper-reactivity as a possible factor contributing to the difficulties of the child with FDD. Our results indicate that routine screening for sensory hyper-reactivity may be an important practice element when working with children with FDD. In addition, the present study adds support to the validity of the THPQ-R in identifying behaviors potentially linked to sensory hyper-reactivity

Keywords: Functional constipation, Functional defecation disorders, Children, Sensation disorders

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Introduction

Childhood functional defecation disorders (FDD) are highly prevalent worldwide (1) and are increasingly considered a public health problem (2). Childhood FDD consist of functional constipation (FC) and functional non-retentive fecal incontinence (FNRFI) (3). Stool toileting refusal (STR), often associated with FDD, is not a diagnosis as such but is described in the literature as fear or refusal of defecation in the potty or toilet for a period lasting at least one month, without concomitant fear or refusal of micturition (4). Children with STR are at high risk of developing FDD, but may avoid developing FDD as long as their requests to be given a diaper to defecate are respected[4]. FDDs are observed in children with and without diagnostic concerns but are more prevalent in those with diagnoses such as autism spectrum disorder (ASD) (5) and attention deficit disorder (ADHD) (6). Unfortunately, despite extensive characterization of the gastrointestinal and behavioral features (3, 7, 8), our comprehension of the multiple factors involved in the emergence and maintenance of childhood FDD continues to be incomplete, and success rates for treatment remain limited (2, 7, 9, 10). As such, further inquiries regarding clinical factors associated with FDD are needed.

Normal defecation is a complex process that involves multiple sensorimotor and psycho-behavioral factors (11). Adequate sensory perception (i.e. the ability to recognize and interpret sensory stimuli) and reactivity (i.e., the modulation of neuronal activity in reaction to sensory stimuli) to the sensory stimuli associated with defecation is key to successful stool toileting. For example, perceiving the need to void the bowel and tolerance of the sensation of passing stool are necessary components of normal defecation (11). Although sensory perception is a longstanding consideration in the study of children with FDD (12-14), and despite the fact that sensory hyper-reactivity is increasingly considered in the study of other types of functional gastrointestinal disorders (15-17), specific links between sensory hyper-reactivity and FDD have not been thoroughly explored in the literature (18). Our recent work and the work of others suggests that some of the difficulties that many children with FDD experience with toileting could be related to issues in sensory hyper-reactivity (19-23). To further investigate this hypothesis, our research team has developed and validated a tool that measures sensory hyper-reactivity in relation to defecation-related sensations: the Toileting Habit Profile Questionnaire-Revised (THPQ-R) (24). The THPQ-R and its earlier version have been shown to adequately discriminate between children with and without FDD (19, 25); however, it´s efficacy in identifying bowel-related sensory hyper-reactivity concerns requires further investigation. This study aimed to: 1) investigate the relationship between sensory hyper-reactivity and FDD, and 2) further evaluate the construct validity of the THPQ-R in relation to its intended usefulness in identifying challenging defecation behaviors related to issues with sensory hyper-reactivity.

Methods

This investigation adopted a descriptive survey methodology in which parents of three to six-year-old children were invited to complete two different online questionnaires; the THPQ-R and the Short Sensory Profile (SSP). The ethics committee of the University of Newcastle (#H-2017-0079) approved the study.

Participants

Participants were caregivers of children with and without FDD or/and STR. Participants in the FDD/STR group were assigned to one of the following categories according to the characteristics of their children: 1) children aged 3 to 6 years old with FDD and/or STR and no additional diagnoses; or 2) children aged 3 to 6 years old with FDD and/or STR and ASD and/or ADHD identified by parental report of diagnosis. The age range of 3–6 years was chosen as it coincides with the time period when ongoing toileting concerns generally become apparent and when manifestations of pain upon defecation or refusal of defecation appear (26, 27). Children with a diagnosis of ASD or ADHD were included because of the reported higher prevalence of FDD in children with these diagnoses (5, 6). The comparison group was comprised of parents of children aged 3 to 6 years old without FDD and/or STR and no other diagnoses, or without FDD and/or STR and diagnosed with ASD and/or ADHD.

Probe questions based on the Rome Foundation diagnostic criteria for FDD were used to verify or rule out FDD (3). The diagnosis of interest was FC for children younger than 4 years and FC and FNRFI for children 4 years and older. A cover letter to the THPQ-R, which includes questions about initiation of toilet training and screens for STR, was part of the online survey (see appendix).

We excluded parents of children with organic causes of defecation disorders. We also excluded from both groups parents of children with neurological conditions, intellectual disability, or psychiatric disorders other than ASD and ADHD. Parents whose children were assigned to their school´s special needs program or had received early intervention services were excluded. Only those participants whose children had initiated toilet training were included.

Parent support groups of all types were contacted and social media were also used to recruit participants. Public and private pediatric gastroenterology and occupational therapy clinics were also contacted for recruitment of parents of children with FDD, STR, ASD and/or ADHD. Recruitment efforts were aimed at various English and Spanish speaking countries.

Data Collection

A web-based survey tool (Qualtrics®) (28) was used for data collection. We implemented several quality control strategies to identify and exclude multiple entries and minimize erratic reporting: (1) internet protocol address check; (2) access to the survey through e-mail invitation once interested participants contacted the researcher; (3) exclusion of respondents who were inconsistent on the probe questions of the Rome Foundation[3] diagnostic criteria or showed other evidence of indiscriminate responding.

Measures

Toileting Habit Profile Questionnaire Revised (THPQ-R): The THPQ-R (24) is a revised version of the THPQ (29). This tool is a parent report questionnaire designed to distinguish typical defecation behaviors and reactions from those that are potential manifestations of sensory reactivity issues associated with FDD and STR. The THPQ-R, available in English and Spanish, has 17 items scored using a dichotomous scale (1=frequently or always; 2=never or rarely), 15 items designed to document sensory hyper-reactivity, and 2 items designed to document sensory hypo-reactivity and/or poor perception (Table 1). Examination of the construct validity of the THPQ-R using Rasch and directed content analysis supports the use of the items designed to identify sensory hyper-reactivity (items 1 to 15) (24). The reliability index (similar to Cronbach's α) for the hyper-reactivity items is documented as 0.89 (24). Items 16 and 17 are recommended exclusively to gather clinical insight into possible links to sensory hypo-reactivity and/or poor perception (24). As such, only the sensory hyper-reactivity items (1 to 15) are part of the final score, with lower scores reflecting greater concerns in hyper-reactivity and defecation behavior. The THPQ-R has been shown to adequately discriminate between children with and without FC (25), with THPQ-R scores correlating highly with the defecation specific scale of the Virginia Encopresis-Constipation Apperception Test (30).

Short Sensory Profile (SSP): The SSP is a condensed version (38 items) of the original Sensory Profile (SP; 125 items) (31). Both the SP and the SSP are caregiver questionnaires that measure responses to sensory events in daily life in children aged between 3 and 10 years. The validity of these tools is well established. The data provided by these questionnaires allows therapists to examine how particular patterns in sensory reactivity may be linked to difficulties with participating in daily occupations (31). The SSP is scored on a five-point Likert scale ('Always' to 'Never'), with low scores indicating greater concerns regarding sensory reactivity. The SSP (32), translated to Spanish (S-SSP) and distributed by the publisher (Pearson), was revised for Spain (33) given that it was originally aimed at Spanish speakers living in the United States. This revised version was used in Spanish-speaking countries. The original English version of the SSP was used for English-speaking countries. In order

Table 1: Items of the Toileting Habit Profile Questionnaire-Revised

THPQ-R items		Sensory issue type
1.	My child hides to poop.	1
2.	My child asks for a diaper when he feels the need to poop.	1
3.	My child prefers to poop in his clothing although the potty or toilet is nearby.	1
4.	My child refuses to sit on the potty or the toilet to poop, but will accept to pee in the potty or toilet.	1
5.	My child refuses or seems uncomfortable sitting on the toilet or potty for both peeing and pooping, even at home.	1
6.	My child withholds poop or resists the urge to poop.	1
7.	My child follows an unusual ritual when pooping which involves actions or places not typically associated with pooping or with the age of the child.	1
8.	My child seems to feel pain when pooping, even if the poop is soft.	1
9.	My child refuses to poop outside of the home.	1
10.	My child shows exaggerated disgust at the smell of his poop.	1
11.	My child refuses to wipe or be wiped after pooping.	1
12.	My child shows fear or refusal related to certain features of the bathroom, such as fear of flushing the toilet.	1
13.	My child needs to pay attention to something else while pooping (a book, a game); this seems to help him/her tolerate the sensation of pooping.	1
14.	My child is sensitive to taste and/or food textures making it difficult to accept laxative medicine or high fibre foods.	1
15.	My child felt the urge to poop very early (younger than 12 months). My child would grunt in a certain way and I would sit him/her on the potty to poop.	1
16.	My child does not seem to feel the urge to poop.	2
17.	My child does not realize he/she has soiled (poop) his/her clothes or is not upset by soiling.	2

Sensory issue type: 1=Sensory hyper-reactivity; 2=Sensory hypo-reactivity and/or issues with perception; items in Spanish are available from the authors

to compare the results of this study with previous results from a pilot study (19), the first edition of the SSP was used. The newer version of the SSP, part of the Sensory Profile-2 family of assessments (34), was not available at the time of the previous study.

Analysis

Considering that the purpose of the study was to develop a better understanding of the relationship between defecation behaviors and sensory hyper-reactivity, our analysis was based on a subset of relevant items from the THPQ-R and the SSP. In relation to the THPQ-R, the analysis included the scores obtained on the first 15 items of the questionnaire as recommended by the authors (24). In the case of the SSP, analysis was done following the method described by Mazurek and colleagues (35) and used in a previous study (19). A sensory hyper-reactivity score was calculated using the items designed to detect sensory hyper-reactivity (items 1–14 and 34–38).

The Student-t test was used to compare the scores of both groups (comparison group and FDD/STR group) on the THPQ-R and SSP. Differences were considered statistically significant at $P<0.05$. To analyze the relationships between the SSP scores and the THPQ-R scores, a Pearson correlation coefficient was calculated. In order to use parametric statistical tests, THPQ-R and SSP raw scores, based on ordinal scaling, were transformed to linear measurements expressed in log odds probability units (logits) using Rasch computations (Table 2). For interpretation of SSP results, we used the published norms; typical scores on the hyper-reactivity items range from 77 to 95 (0.88 to 4.72 logits).

Results

The recruitment period yielded 299 participants whose children were aged between 3 and 6 years. After verification of inclusion and exclusion criteria, 276 participants were retained for the analysis (Table 3). The FDD/STR group (total n=136; ASD n=33, ADHD n=5) included children with FC (n=129), FNRFI (n=2) and STR (n=5). The comparison group (total n=140, ASD n=6, ADHD n=2) consisted of children without FDD or STR.

Sensory Hyper-Reactivity in FDD/STR

An independent-samples t-test was run to determine if there were differences in the sensory hyper-reactivity scores between children with FDD/STR and the comparison group. The sensory hyper-reactivity mean score was lower for children with FDD/STR (0.91±0.09) than for children without FDD/STR (1.34±0.08). The difference (0.44 (95% CI, 0.67 to 0.21)) was observed to be statistically significant: $t(274)=-3.759$; $P<0.0005$. The sensory hyper-reactivity mean score of the children without FDD/STR (1.34±0.08) fell clearly within the typical range (0.88 to 4.72). The mean score of children in the FDD/STR group (0.91±0.09), considering the

Table 2: THPQ-R and SSP Raw Scores Equivalence in Log Odds Probability Units

THPQ-R											
Raw	**Logits**	**S.E.**	**Raw**	**Logits**	**S.E.**	**Raw**	**Logits**	**S.E.**	**Raw**	**Logits**	**S.E.**
15	-4.41	1.87	19	-1.21	0.64	23	0.19	0.57	27	1.65	0.68
16	-3.08	1.08	20	-0.82	0.61	24	0.52	0.58	28	2.19	0.79
17	-2.23	0.81	21	-0.46	0.58	25	0.86	0.59	29	3.00	1.06
18	-1.66	0.70	22	-0.13	0.57	26	1.23	0.62	30	4.29	1.86
SSP											
Raw	**Logits**	**S.E.**	**Raw**	**Logits**	**S.E.**	**Raw**	**Logits**	**S.E.**	**Raw**	**Logits**	**S.E.**
19	-4.78	1.82	38	-0.82	0.23	57	0.01	0.20	76	0.83	0.23
20	-3.60	0.98	39	-0.77	0.23	58	0.05	0.20	77	0.88	0.23
21	-2.93	0.69	40	-0.72	0.22	59	0.09	0.20	78	0.94	0.24
22	-2.55	0.56	41	-0.67	0.22	60	0.13	0.20	79	1.00	0.24
23	-2.28	0.48	42	-0.62	0.22	61	0.17	0.20	80	1.06	0.25
24	-2.08	0.43	43	-0.57	0.22	62	0.21	0.20	81	1.12	0.26
25	-1.91	0.39	44	-0.53	0.21	63	0.25	0.20	82	1.19	0.27
26	-1.77	0.36	45	-0.48	0.21	64	0.29	0.20	83	1.26	0.28
27	-1.65	0.34	46	-0.44	0.21	65	0.33	0.20	84	1.34	0.29
28	-1.54	0.32	47	-0.40	0.21	66	0.37	0.20	85	1.43	0.30
29	-1.44	0.30	48	-0.35	0.21	67	0.42	0.21	86	1.52	0.31
30	-1.35	0.29	49	-0.31	0.20	68	0.46	0.21	87	1.63	0.33
31	-1.27	0.28	50	-0.27	0.20	69	0.50	0.21	88	1.75	0.36
32	-1.20	0.27	51	-0.23	0.20	70	0.54	0.21	89	1.88	0.39
33	-1.13	0.26	52	-0.19	0.20	71	0.59	0.21	90	2.05	0.42
34	-1.06	0.25	53	-0.15	0.20	72	0.64	0.22	91	2.25	0.48
35	-1.00	0.25	54	-0.11	0.20	73	0.68	0.22	92	2.51	0.55
36	-0.94	0.24	55	-0.07	0.20	74	0.73	0.22	93	2.89	0.68
37	-0.88	0.24	56	-0.03	0.20	75	0.78	0.22	94	3.54	0.98
									95	4.72	1.81

Raw: Raw Score; Logits: Log odds probability units; S.E.: Standard Error

Table 3: Distribution of children by age

Age (years)	3	4	5	6	Total
FDD/STR	45	43	23	25	136
NO FDD/STR	46	40	30	24	140
Total	91	83	53	49	276

FDD: functional defecation disorder; STR: stool toileting refusal

standard error of measurement (0.82-1.00), could potentially be outside the typical range and within the clinical range of sensory hyper-reactivity.

Relationship of THPQ-R and Sensory Hyper-Reactivity Scores

The THPQ-R mean score was lower for children with FDD/STR (1.83±1.59) than for children without FDD/STR (3.79±0.77). The difference (1.96; 95% CI: 2.26 to 1.66), was statistically significant: t(193.879)=-12.967; P<0.0005. When all children were considered together, there was a statistically significant, moderate positive correlation between THPQ-R and sensory hyper-reactivity scores: $r_{(274)}$=0.443; P<0.0005. This relationship was also found when children with FDD/STR were considered separately ($r_{(274)}$=0.485; P<0.0005). These results reveal that higher levels of sensory hyper-reactivity were associated with a higher frequency of the challenging defecation behaviors described in the hyper-reactivity items of the THPQ-R.

Discussion

The relationship between sensory reactivity concerns and FDD is a relatively recent area of study. On one hand, the construct of sensory reactivity (hyper-reactivity and hypo-reactivity) is well established within the Ayres Sensory Integration® framework (36-40), and issues in sensory reactivity are reported to be highly prevalent in children with ASD and ADHD (38, 41) as well as in children without developmental disorders[42]. Similarly, children with FDD have been extensively characterized by the medical and mental health fields (3, 8, 43). Furthermore, FDDs are reported to be highly prevalent in the general childhood population (1), and even more so among those with ASD or ADHD (5, 6). However, besides the publication of a few case studies (21, 44, 45) and program evaluations (20, 46), the interface between sensory reactivity issues and FDD has only been formally considered recently (19, 23, 35, 47).

A comprehensive understanding of the factors contributing to the emergence and maintenance of FDD is becoming increasingly important as the overall incidence rises[2] and success rates for treatment of children with defecation concerns remain limited (7, 9). This study adds to our understanding by identifying sensory hyper-reactivity in some children with FDD, and by showing that the two conditions are moderately correlated. Although the mean sensory hyper-reactivity score for children with FDD was within a sub-clinical range, there was a clear difference between the groups. Consequently, clinicians need to be sensitive to the possibility of sensory hyper-reactivity when working with FDD. Clearly identifying sensory hyper-reactivity related to FDD is of utmost importance to guide the assessment and intervention process with this population.

Assessment of sensory reactivity using caregiver questionnaires has become an accepted method of documenting issues in this area (34, 39); however, tools available up to this point have not addressed defecation. The THPQ-R fills this gap. The THPQ-R has been systematically developed and validated to measure a broad range of challenging defecation behaviors potentially linked to sensory hyper-reactivity in children aged 3 to 6 years. The THPQ-R is a caregiver questionnaire, which in the case of assessing the toileting behavior of young children, is the most ecologically valid way to obtain information (48). Our previous work had provided preliminary support for the hypothesis that the behaviors described in the sensory hyper-reactivity section of an earlier version of the THPQ were associated with sensory hyper-reactivity in children with fecal incontinence with concomitant functional constipation who had not responded to conventional medical management (19). The present study adds support to the validity of the revised version of the THPQ and does so with a bigger sample size of children with a variety of defecation issues, thus increasing its relevance in research and clinical practice.

Health practitioners do not usually consider sensory hyper-reactivity as a possible factor contributing to the difficulties of the child with FDD; however, the results presented here indicate that routine screening for sensory hyper-reactivity may be an important practice element. Tools such as the SSP may not be sufficiently sensitive to identify sensory hyper-reactivity issues related to FDD. The use of a refined tool, with items specifically designed to detect hyper-reactivity to defecation related sensations, is needed to complement tools that address general aspects of sensory reactivity. There is emerging evidence that interventions designed to address the sensory hyper-reactivity issues that appear to underlie the behaviors related to the onset and maintenance of FDD may contribute to more successful treatment outcomes (20-22). Moreover, there is growing evidence supporting the use of occupational therapy to enhance participation and performance in activities of daily living in children with sensory issues and ASD (49, 50). Our current work supports the use of the THPQ-R in the identification of sensory hyper-

reactivity-related FDD, which is an essential step in understanding some of the underlying issues that can potentially be addressed in intervention.

The main limitation to this study is the small age range (3 to 6 years); older children and teenagers also experience FDD. Nevertheless, our focus was on younger children as ongoing toileting concerns generally become apparent during this time and symptoms such as painful defecation or defecation refusal appear (26, 27). This age range was also chosen given that the THPQ-R has been validated with this age group. Additionally, the THPQ-R is based on observation of behavior, and behavioral manifestations of FDD seem to be more prominent in younger children (26). Furthermore, the diagnostic groups (ASD, ADHD) that were included in our study represented a small proportion of our sample that was unevenly distributed among our FDD/STR and comparison groups. We therefore could not analyze these cases separately. Thus, future studies with wider age ranges and larger samples of children with ASD and ADHD are needed to further validate the THPQ-R and better characterize children within the diagnostics groups known to be at risk for FDD.

Conflict of Interests: None declared.

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