



Contribution of Sensory Processing to Chronic Constipation in Preschool Children

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Objective To assess differences in sensory processing patterns between children with chronic constipation compared with a matched normative sample as well as to examine the extent to which specific sensory processing patterns and modality scores predicted atypical toileting behaviors.

Study design We used a cross-sectional comparative design to evaluate differences between children age 3 and 5 years old with constipation (n = 66) and those in a matched control sample (n = 66). We also examined the contribution of sensory processing patterns to atypical toileting behavior in the clinical sample.

Results Children with chronic constipation showed significantly higher sensory scores than a matched normative sample, specifically in oral processing ($P < .001$), visual processing ($P < .05$), sensory avoiding ($P < .001$), and sensory sensitivity ($P < .05$). Sensory registration, avoidance, and oral processing significantly predicted toileting behavior over-responsiveness, and attentional difficulties contribute to toileting under-responsiveness.

Conclusions Our findings revealed that children with chronic constipation have underlying sensory characteristics that contribute to toileting behavioral difficulties. By identifying sensory processing patterns of children with chronic constipation, we can optimize behavioral interventions to complement laxative therapy for this population. (*J Pediatr* 2019;210:141-5).

Chronic constipation is prevalent among young children, occurring in approximately 3%-29% of children.^{1,2} Chronic constipation, or fecal retention, is associated with infrequent and/or painful defecation and fecal incontinence; children with chronic constipation also experience fecal impaction, which may hinder the bowel training process. Treatment for chronic constipation typically includes a combination of interventions such as medical approaches (eg, laxative therapy, diet management), parent education and support, and behavioral interventions (eg, positive reinforcement, skill-building of techniques).³ Behavioral strategies hold great appeal given the potential to avoid invasive and challenging medical interventions, especially in very young children. Unfortunately, high-quality studies have been limited and do not provide definitive evidence to support the efficacy of behavioral interventions alone in the management of chronic constipation.⁴ A more comprehensive understanding of the etiology of chronic constipation in typically developing children is required to identify pathways for intervention that can prevent a chronic course and avoid recurrence of this common health problem.⁵

Emerging evidence suggests that sensory processing deficits (ie, detection and behavioral response to sensory information) may be an important but previously overlooked area of study that may shed light on potential risk factors in young children that contribute to chronic constipation. According to Dunn's Sensory Processing Framework,⁶ children show behaviors that fall within 4 sensory processing patterns: sensitivity (an increased perception of sensory stimuli); avoidance (an active withdrawal from sensory stimuli); seeking (an intense interest in sensory stimuli); and registration (a lack of or delayed response to sensory stimuli). Sensory processing differences occur across systems (eg, tactile, auditory) and reflect constellations of behaviors that reflect underlying neurologic thresholds as well as self-regulation strategies.⁶

In this study, we investigated sensory processing characteristics in children with chronic constipation and the contribution of sensory processing patterns to toileting behaviors. We addressed the following research questions: (1) To what extent do preschool-age children with chronic constipation differ from a control group on sensory processing patterns? and (2) To what extent does sensory processing predict toileting over- and under-responsiveness among children with chronic constipation?

Methods

Subjects were recruited from January 2015 to April 2017 from a hospital-based clinic using convenience sampling; all participants completed the study questionnaires during a clinic visit. This investigation was approved by the Human Subjects Review Committee at Ann and Robert H Lurie Children's Hospital of Chicago. Participants included children ages 3-5 years with chronic constipation (n = 66) and a matched sample of typically developing children based on age and sex (n = 66). In the clinical sample, children attending a pediatric gastrointestinal

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clinic for an initial visit with a chief complaint of chronic constipation were eligible for participation. We included children in the clinical sample based on Rome IV criteria,⁷ which included 2 or more of the following occurring at least once per week for a minimum of 1 month: 2 or fewer defecations in the toilet per week, at least 1 episode of fecal incontinence per week, history of retentive posturing or excessive volitional stool retention, history of painful or hard bowel movements, presence of a large fecal mass in the rectum, and a history of large diameter stools that can obstruct the toilet. We excluded children with autism spectrum disorder, severe developmental delay, and/or neurologic impairment.

Children in the control group were drawn from the standardization sample of the Sensory Profile-2. Using the fuzzy case control extension in SPSS (IBM Corp, Armonk, New York), subjects in the control group were matched to the clinical sample by age and sex. Children reported as receiving special education services were excluded from the control group, as were children with serious medical or psychiatric conditions and known sensory or motor impairments.

Measures

Parents provided basic demographic information and qualitative information about their child's toileting habits.

The Child Sensory Profile-2⁶ is a standardized, 86-item parent-report measure of children's sensory processing patterns in the context of everyday life. Parents indicate the extent to which each item describes their child's experience and/or functioning (almost always = 5 to almost never = 1). Summary scores are generated reflecting patterns in 3 domains: sensory modalities (auditory, visual, touch, movement, body position, oral), behavior (attention, conduct, social-emotional), and sensory processing pattern (registration, seeking, sensitivity, avoiding).

The Toileting Habit Profile Questionnaire⁸ is a 10-item, standardized tool used to evaluate sensory-based defecation difficulties. Parents rate the extent to which each item describes their child's behavior (almost always = 1 to never = 5). The tool includes 8 items that reflect over-responsiveness (ie, low sensory thresholds or sensitivity/avoiding) and 2 items that reflect under-responsiveness (ie, high sensory thresholds or registration). Lower scores on the Toileting Habit Profile are more indicative of behaviors hypothesized to be specific to children with fecal retention; that is, lower scores indicate greater difficulties.

Data Analyses

SPSS v 22.0 (SPSS Inc, Chicago, Illinois)⁹ was used to analyze data. The Sensory Profile-2 scoring was standardized in the general population and provides categorizations of scores based on the bell curve. Children are categorized as "much less than others/less than others," "similar to others," or "more than/much more than others"; such categorizations help practitioners understand individuals' scores as they relate to peers and to determine if children meet cut-off scores to show clear sensory differences. When children show "much less/less than others" scores, they scored at least

1 SD below the majority of peers and are showing decreased responses to sensory stimuli in that domain. When children show "more than/much more than others," scored at least 1 SD above the majority of peers and they are showing increased or exaggerated responses in that sensory domain. χ^2 tests were used to assess differences in the prevalence of sensory processing categories ("much less than others/less than others"; "similar to others"; "more than/much more than other") between groups (constipated vs controls). Understanding the categorical differences between children with constipation vs controls, as opposed to examining average scores per group, has clinical significance because practitioners consider only those that score outside of the typical range as having sensory processing difficulties. Multivariate regression was used to examine the extent to which sensory processing scores predicted toileting habit scores within the constipation group.

Results

The clinical population consisted of 66 children (% white = 75.8; % male = 54.5) and the control sample (n = 66) was matched on age and sex (% white = 63.1). Descriptive findings from the clinical sample included 45 of 64 (70%) having bowel movements at same time every day, 15 of 64 (23%) sitting on toilet at same time every day, 35 of 64 (55%) having access to toilet when needed (ie, do not have to share bathroom), 22 of 61 (36%) with fear of using toilet, 37 of 64 (58%) having successfully transitioned to underwear, 29 of 63 (46%) wiping without assistance, and 29 of 61 (48%) receiving rewards for using toilet. Bowel movements almost always occur in toilet in 21 of 62 (34%) subjects, in diapers in 17 of 59 (29%) subjects, in pull-ups in 13 of 62 (21%) subjects, and in underwear in 6 of 61 (10%) subjects. Urination almost always occurs in toilet in 38 of 61 (62%) subjects, in diapers in 11 of 59 (19%) subjects, in pull-ups in 11 of 60 (18%) subjects, and in underwear in 5 of 63 (8%) subjects.

χ^2 results (Figure) showed that children with chronic constipation significantly differed in oral processing ($\chi^2 = 20.137[2]$, $P < .001$), sensory avoiding ($\chi^2 = 6.405[2]$, $P < .05$), sensory sensitivity ($\chi^2 = 16.667[2]$, $P < .001$), and visual processing ($\chi^2 = 5.908[2]$, $P < .05$). Multivariate regression analysis (Table) showed that avoidance significantly predicted toileting habit over-responsiveness ($P < .05$). Registration also significantly predicted toileting habit over-responsiveness in the opposite direction ($P < .01$). No sensory scores predicted toileting habit under-responsiveness. With regard to sensory modality predicting toileting habit behaviors, the most parsimonious model showed that oral processing alone explained a significant proportion of variance in toileting over-responsiveness, $R^2 = .097$, $F(2,63) = 6.794$, $P < .05$. Attention explained a significant proportion of variance in toileting under-responsiveness, $R^2 = .128$, $F(2,63) = 4.495$, $P < .05$.

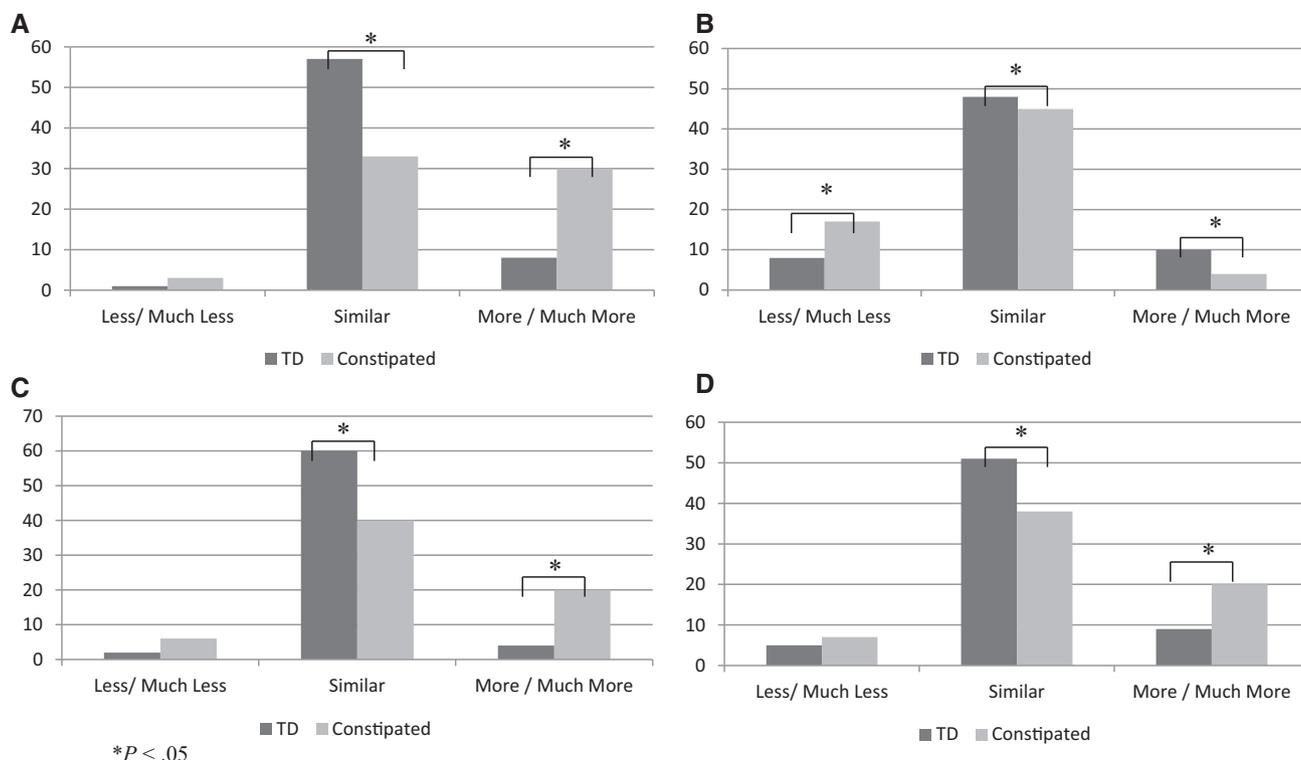


Figure. Differences in sensory categories by group. **A**, Oral processing; **B**, Visual processing; **C**, Sensory sensitivity; **D**, Sensory avoiding. TD, typical development; χ^2 analysis, *P < .05.

Discussion

Preschool age children with chronic constipation showed increased responses to sensory stimuli and increased avoidance behaviors as compared with a matched control group. Specifically, children in the clinical sample showed increased oral processing, sensory sensitivity, sensory avoiding, and visual processing vs controls. Prior investigations have related sensory sensitivity to toileting difficulties^{8,10,11}; however, we identified specific contributions of sensory systems and patterns as well as the role that attention plays in toileting behaviors.

Several indicators for adverse toileting behavior favored over-responsiveness more than under-responsiveness. Sensory registration, or under-responsiveness, emerged as an important marker in children with chronic constipation. It is possible that sensory under-responsiveness may serve as a protective factor and lessen the effect of sensory stimuli on toileting difficulties even among those with chronic constipation. Also, children who were rated as demonstrating high levels of sensory avoidance were also found to have toileting behavior over-responsiveness. Children with an avoidance pattern withdraw from sensory input that they perceive as being unpleasant. In the case of toileting, this may manifest as withholding of stool and overall bathroom avoidance. Taken together, these findings provide a rationale for how sensory patterns contribute to the development of chronic constipation: children with under-responsiveness may not notice that they must defecate whereas children with high sensitivity are avoidant of toileting and, therefore, experience higher levels of fecal retention.

When examining specific sensory modalities that may be predictive of chronic constipation, 2 factors emerged: oral processing and attention. Oral processing presented as the most significant factor in predicting toileting behavior over-responsiveness. Children with oral processing difficulties are often picky eaters and have a limited variety of food intake.^{12,13} On the surface, the association between oral processing and constipation may not seem intuitive; however, the association between food refusal and

Table. Relationship of toileting habit profile to sensory processing pattern

Toileting responsiveness	B	SE	T	Sig.
Toileting over-responsiveness				
Seeking	−0.80	.113	−.707	.483
Avoid	−.247	.097	−2.553	.014
Sensitivity	−.088	.130	−.673	.504
Registration	.355	.114	3.104	.003
Toileting under-responsiveness				
Seeking	−.001	.036	−.018	.986
Avoid	−.012	.029	−.423	.674
Sensitivity	−.006	.039	−.155	.877
Registration	−.013	.036	−.345	.731

B, least squares estimate; T, test statistic. Sig provides the P-value associated with the test statistic.

constipation/encopresis was first described over 50 years ago.¹⁴ More recently, research has suggested a bidirectional association between fussy eating and functional constipation was reported in preschool children.¹⁵ These findings point to a vicious cycle in which a child's gastrointestinal problems and problematic eating behavior mutually affect each other, and it may be that there are underlying factors that contribute to the co-occurrence of such behaviors.

In this investigation, we have demonstrated oral sensitivity as a prevalent "more intense" response in children with constipation and fecal retention. Children who are orally sensitive with regard to food texture and taste may have atypical sensory registration. Specifically, they may over register or under register the sensory properties of foods. Previous negative experiences with certain foods leads to avoidance of them, whereas foods that have pleasing sensory properties become overly represented in the diet (ie, crunchy foods, sweet foods). If registration difficulties play a role in understanding oral sensitivities, the same pattern may be affecting other areas of functioning, such as bowel functioning and toileting. Children with registration difficulties will apply similar compensatory strategies (ie, avoidance) when they experience discomfort with sensory input related to bowel movements. Thus, both challenges may develop as a result of sensory processing difficulties; however, over time, they lead to behavioral patterns that become fixed and rigid.

The second modality that emerged as a predictor for toileting behavior was attention. However, this finding was less robust and it was associated with toileting behavior under-responsiveness only (not over-responsiveness). Children who experienced more challenges with attention tended to have more features of toileting under-responsiveness (ie, not feeling the urge to defecate, not recognizing having had a bowel movement). Attention difficulties have been previously identified as markedly higher in children with "soiling difficulties."¹⁶ However, attention has been investigated through a psychological lens rather than a sensory processing lens. As a modality, attention skills are multifaceted and may differ depending on the sensory system (sight, hearing, touch, taste, smell). For example, children who are highly attentive to visual input may be less attentive to auditory input. In addition to the 5 basic senses, attention can also affect lesser known senses such as proprioception (body awareness), vestibular input (movement), and interoception (ability to register, understand, and feel sensations inside the body). This study suggests that attention differences may affect toileting behavior under-responsiveness, but the linkage needs to be better understood, especially with regard to variables that may have a more significant role in toileting (eg, proprioception, interoception).

We confirm that children with chronic constipation have underlying sensory characteristics that contribute to toileting difficulties. Specifically, young children with high sensory registration, high oral sensitivity, and increased sensory

avoidance behaviors may be at risk for toileting over-responsiveness. In addition, there is preliminary evidence to suggest a possible link between attention and toileting behavior under-responsiveness. As there are likely multiple pathways to the development of chronic constipation, a focus on constellations of variables is needed. By identifying sensory processing patterns of children with chronic constipation, we can better match behavioral interventions to areas of need, offering a valuable supplement to traditional medical approaches for treatment.

This study has several important limitations. The sample size was modest and data was not available for toileting behaviors in the control sample. In addition, the small number of items comprising the toileting behavior under-responsiveness score/domain may have contributed to limited findings for this behavioral pattern. Children recruited for this study represented a clinical sample; it is not possible to determine the generalizability to nonreferred preschool-age children. In addition, studies that can further explore the connection between sensory processing, differences in temperament and toileting difficulties are needed to establish different causal pathways. ■

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50 Years Ago in *THE JOURNAL OF PEDIATRICS*

Fatal Neonatal Hepatic Steatosis: A New Familial Disorder

Satran L, Sharp H, Schenken JR, Krivit W. *J Pediatr* 1969;75:39-46.

Three infants died from hemorrhagic complications likely owing to an underlying hepatic disorder. These infants were from the same family; however, no cause was identified for their presentation. All the infants died soon after birth, the longest surviving 12 weeks. Both parents remained healthy, and there was a healthy child born in between these siblings. All 3 deceased siblings had feeding difficulties and significant bleeding with prolonged prothrombin and partial thromboplastin times and hypofibrinogenemia. The first sibling had significant bleeding that was not responsive to vitamin K or transfusions, and did not have significant hepatosplenomegaly. The second sibling had a large cephalohematoma, enlarged liver to 5 cm, and renal failure with metabolic acidosis. The third sibling had vomiting and diarrhea as well as significant gastrointestinal bleeding.

On autopsy, the infants were found to have hepatic steatosis. The livers were enlarged and yellow in color. The hepatic cells had large vacuoles of fat on hematoxylin and eosin staining and Sudan black stain positivity. The kidneys also had prominent vacuoles that were consistent with deposition of fat. In 1 infant, the lungs showed signs of early bronchopneumonia, and in another the lungs showed congestion in the alveolar capillaries. One infant also had inflammatory exudate in the jejunum, but the gastrointestinal tract was otherwise normal. The remaining organs were normal.

For these infants, a broad differential was considered and extensive testing was done. High on the differential were inborn errors of glycogen or galactose metabolism, cystic fibrosis, tyrosinemia, Wolman disease, Reye syndrome, or a toxin. These diagnoses were excluded based on symptoms or diagnostic testing. A broader differential is possible today—an inborn error of metabolism or mitochondrial hepatopathy would be high on the possible causes for the presentation of these infants. Several mutations, such as *POLG*, *SLC25A20*, and *SCO1* are possibilities for potential mitochondrial hepatopathies.¹ These mutations can manifest as steatosis and lead to acute liver failure in the neonatal period. Additionally, Boison et al identified a disruption in the adenosine kinase gene as a potential cause for fatal neonatal hepatic steatosis.² However, further investigation is needed to better understand this process.

The diagnosis of these 3 infants, who developed neonatal liver disease and hepatic steatosis, was unclear 50 years ago and the same outcome may result today. However, with our increased understanding of hepatic metabolic disease and the wide availability of genetic testing panel and whole exome sequencing, it is likely that an answer would emerge!

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