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Toilet training in children with a functional defecation disorder and concomitant symptoms of autism spectrum disorder

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ABSTRACT

The purpose of this study was to determine the association between the presence of symptoms of Autism Spectrum Disorder (ASD) and the time of completion of toilet training in pediatric patients with a Functional Defecation Disorder (FDD). Consecutive children (4–12 yrs) presenting with FDD according to the ROME III criteria were screened for symptoms of ASD by two validated questionnaires; the Social Responsiveness Scale (SRS) and the Social Communication Questionnaire-Lifetime (SCQ-L). Children were defined as having symptoms of ASD when they scored at or above the cut-off value on one or two questionnaires (SRS $\geq$ 51; SCQ $\geq$ 15). This study included 96 age-matched controls from the general population and 242 pediatric patients with FDD of which 70 had symptoms of ASD. Significantly less children with FDD and ASD symptoms were toilet trained for stools and urine during daytime before the age of 4 yrs (41% and 58% respectively) than children with FDD only (56% and 72%), whereas almost all controls had completed toilet training daytime before this age (95% and 98%). Children with FDD and ASD symptoms completed toilet training both for stools and urine during daytime at a significantly later age than children with FDD only and controls.

1. Introduction

Functional Defecation Disorders (FDDs), such as functional constipation and functional non-retentive fecal incontinence, are common problems in childhood that may negatively affect quality of life. Environmental, genetic, behavioral factors, and child-parent interactions have all been subject of research and seem to play a role in the etiology or persistence of symptoms (Mugie, Benninga, & Di Lorenzo, 2011). However, the exact pathophysiology of FDD is incompletely understood. In a large proportion of children, symptoms of FDD arise in the period of toilet training (Blum, Taubman, & Nemeth, 2003; Schonwald, Sherritt, Stadtler, & Bridgemohan, 2004; Taubman, 1997; Taubman, Blum, & Nemeth, 2003). Furthermore, atypical toileting behaviors, for instance stool toileting refusal and hiding away while defecating, have been associated with late Completion of Toilet Training (CTT) and the presence of constipation (Taubman, 1997; Taubman et al., 2003).

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FDDs have frequently been associated with Autism Spectrum Disorders (ASDs) (e.g., Afzal, Murch, Thirrupathy, Berger, & Fagbemi, 2003; Chandler et al., 2013; Gorrindo et al., 2012; Ibrahim, Voigt, Katusic, Weaver, & Barbaresi, 2009; Pang and Croaker, 2011; Peeters, Noens, Philips, Kuppens, & Benninga, 2013; Peters et al., 2014; Wang, Tancredi, & Thomas, 2011). In the general population, a prevalence rate of ASD of 0.6% – 1% has been reported (Elsabbagh et al., 2012). ASDs are characterized by persistent impairments in social communication and social interaction and by restricted, repetitive patterns of behavior, interests or activities (DSM-5; American Psychiatric Association, 2013). Children with ASD are toilet trained later than controls, and after being toilet trained, regression in training skills often occurs when routines change (Dalrymple and Ruble, 1992; Sandhu, Steer, Golding, & Emond, 2009).

The association between the presence of concomitant symptoms of ASD and the time of CTT in children with a FDD is, however, unknown. It could be hypothesized that the presence of concomitant ASD symptoms further delays the time of CTT in children with FDD. Identifying children with FDD and concomitant symptoms of ASD is important as these children might benefit from an early adaptation of the regular treatment strategies, for instance by adding psycho-educational, psycho-therapeutic or psychiatric care to the standard therapy for FDD. Therefore, the aim of this study was to compare the time of CTT between children with FDD without ASD symptoms, children with FDD with concomitant symptoms of ASD and controls.

2. Methods

2.1. Participants

2.1.1. Children with FDD with and without symptoms of ASD

Between September 2009 and October 2011, consecutive pediatric patients, aged 4–12 years, referred to the outpatient clinic of a tertiary hospital in The Netherlands, with a diagnosis of Functional Constipation (FC) or Functional Non-Retentive Fecal Incontinence (FNRFI) according to the ROME III criteria (Table 1), were included after informed consent was obtained from the parents. This consecutive sampling technique involves that all individuals whose parents agreed to participate were selected, provided they met the mentioned pre-established criteria. Patients were excluded from participation if they suffered from Crohn's disease or ulcerative colitis, celiac disease, or when they had a history of large bowel surgery, congenital anorectal malformations, neurological disease (complete spinal cord transection, multiple sclerosis or spina bifida) or a genetic syndrome. Furthermore, patients with a known intellectual disability and/or an intelligence quotient below 70 were excluded.

2.1.2. Controls

General population controls, aged 4–12 years, were recruited from primary schools in Flanders, the Dutch-speaking part of Belgium. Children with FDD according to the ROME III criteria and/or ASD diagnosis and children with a history of a diagnosis of FDD made by a medical doctor or prior or current treatment with laxatives were excluded from participation.

2.2. Outcomes

The primary outcomes of this study were the time of CTT for stools, urine during daytime, and urine during nighttime in children with FDD without symptoms of ASD, children with FDD with concomitant symptoms of ASD and controls. A child was considered toilet trained for stools or urine during day- and nighttime when parents reported that the child wears underwear during the day and night and urinates and defecates in the toilet or potty with < 4 urine accidents per week and 2 or fewer episodes of fecal incontinence per month (Blum et al., 2003). Children, however, who had been completely toilet

Table 1
Diagnostic criteria for functional constipation and functional non-retentive fecal incontinence.

Functional constipation
Must include 2 or more of the following in a child with a developmental age of at least 4 years*:
1. Two or fewer defecations in the toilet per week
2. At least 1 episode of fecal incontinence per week
3. History of retentive posturing or excessive volitional stool retention
4. History of painful or hard bowel movements
5. History of large diameter stools that may obstruct the toilet
6. Presence of a large fecal mass in the rectum
Functional non-retentive fecal incontinence
Must include all of the following in a child with a developmental age at least 4 years**:
1. Defecation into places inappropriate to the social context at least once per month
2. No evidence of an inflammatory, anatomic, metabolic, or neoplastic process that explains the subject's symptoms
3. No evidence of fecal retention

* Criteria fulfilled at least once per week for at least 2 months before diagnosis.
** Criteria fulfilled for at least 2 months before diagnosis.