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Original Article

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Dyssynergic Defecation in Chronically Constipated Children in Korea

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ABSTRACT

Purpose: Dyssynergic defecation (DSD) is one of the important causes of chronic constipation in children. We aimed to analyze the clinical features, diagnostic test results, and treatments for DSD in children.
Methods: Children diagnosed with DSD using fluoroscopic defecography were enrolled in this study. Clinical data, including the results of colon transit time (CTT) test and biofeedback (BF) therapy, were collected from medical records retrospectively.
Results: Nineteen children were enrolled. The median age was 9 years (6–18 years), the median frequency of bowel movement was 1/7 days (1–10 days), the median duration of constipation was 7.0 years (2–18 years), the median age of onset of constipation was 2.5 years (1–11 years). In the CTT test, outlet obstruction type was noted in 10/18 (55.6%), slow transit type in 5/18 (27.8%), and normal transit in 1/18 (5.6%). The median CTT was 52 hours (40–142 hours). Initial medical therapy was performed with the polyethylene glycol 4000, and the response was good in 9/19 (47.4%), fair in 9/19 (47.4%), and poor in 1/19 (5.0%). BF was performed in 8/19, with good results in 6/8 (75.0%) children and failure in 2/8 (25.0%) children. After long-term medical therapy (11/19), 3/5 showed good response with medication alone, 6/8 showed good response with BF and medication combined.
Conclusion: DSD should be considered as a cause of chronic constipation in children, especially in those with abnormal CTT test results. BF combined with medical therapy is effective even with age-limited cooperation.

Keywords: Constipation; Dyssynergia; Child

INTRODUCTION

Although constipation is one of the most common digestive symptoms in children, algorithms for its accurate diagnosis and treatment are not well established in clinical practice.

Although most of pediatric functional constipation can be diagnosed clinically through Rome criteria and physical examination, not all pediatric constipation is functional, and significant number of patients need further tests for causes of constipation. Assessing bowel movement using radio-opaque markers is the primary basic test for intestinal motility function. Patients can be divided into normal and abnormal bowel movement groups, and abnormal groups

are then further classified into outlet obstruction type and slow transit type [1-4]. Although this test could provide directions for diagnosis and treatment, the way is not straight. And, in some patients who do not respond to conventional medical therapy or cannot taper the medicine, further studies are required to investigate the roles of outlet function, which includes the pelvic floor and puborectalis muscle, sigmoid colon, and anorectum, and slow transit, which includes the entire colon except the sigmoid colon [5-8].

Most recent studies on pediatric constipation have conducted aggressive tests, such as entire colon manometry, after a simple brief study without the evaluation of anorectal outlet function [8]. However, considering the frequency of the underlying diseases, feasibility of examination, and treatment in pediatric patients, it is reasonable to evaluate outlet function before aggressive end-stage studies, such as entire colon manometry. Some recent studies have reported the role of outlet function tests in the diagnosis and treatment of pediatric constipation [5-7].

In this study, the clinical characteristics of the patients, the results of diagnostic tests, including the colon transit time (CTT) test, and the response to treatments, including medications and biofeedback, were analyzed in children diagnosed with dyssynergic defecation (DSD) or pelvic floor dysfunction (PFD) by fluoroscopic defecography (FDG).

MATERIALS AND METHODS

Among children diagnosed with chronic constipation according to Rome III/IV criteria and showing abnormal CTT test results, from August 2005 to October 2020, 100 children underwent FDG. The CTT test was performed according to the Metcalf protocol [9] with 20 radio-opaque markers per capsule, and FDG was performed as described in a previous study [7]. Data on the clinical characteristics of the patients, results of the diagnostic tests, and response to medications and biofeedback (BF) were collected from medical records retrospectively. Classification of the response to therapy was as follows: 1) good response; same or more than two bowel-movement days per 3 days with soft to loose consistency (Bristol stool scale 5–7) without any symptoms; 2) fair response; same or more than one bowel movement day per 2 days without hard stools (Bristol stool scale 1–3) without any symptoms; 3) poor response; less than fair. This study was approved by the Institutional Review Board of the Konkuk University Medical Center (IRB No. KUMC 2021-08-034).

RESULTS

Nineteen children were diagnosed with DSD or PFD, including puborectalis relaxation failure and other pelvic outlets relax failure without a definite sphincter anomaly. The median age of the children was 9 years (range 6–18 years), the median age of onset of constipation was 2.5 years (range 1–11 years) and 12 of the children were males. The median frequency of bowel movement was one per 7 days (range 1–10 days), the median duration of constipation was 7.0 years (range 2–18 years), and fecal incontinence was noted in eight out of 19 (42.1%) children (**Table 1**). In the CTT test, outlet obstruction type was noted in 10 of 18 (55.6%) children, slow transit type was noted in five of 18 (27.8%) children, and normal transit was noted in one of 18 (5.6%) children. In two of 18 (11.2%), the result was not reliable. The median CTT was 52 hours (range 40–142 hours) (**Table 2**). Initial medical therapy was performed with

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Table 1. Clinical characteristics of the children with dyssynergic defecation

Parameters	Value (n=19)
Male:female	12:7
Age (yr)	9 (6–18)
Frequency of bowel movement per day	1/7 (1–10)
Duration of constipation (yr)	7.0 (2–18)
Age of onset of constipation (yr)	2.5 (1–11)
Fecal incontinence	8 (42.1)

Values are presented as number only, median (range), or number (%).

Table 2. Results of colon transit time test in children with dyssynergic defecation

Colon transit time test	Value (n=18)
Type of transit	
Normal	1 (5.6)
Outlet obstruction	10 (55.6)
Slow	5 (27.8)
Not reliable	2 (11.1)
Colon transit time (hr)	52 (40–142)

Values are presented as number (%) or median (range).

Table 3. Response to initial medication in children with dyssynergic defecation

Regimen	Patients (n=19)	Response		
		Good	Fair	Poor
PEG 4000	17	8	9	
PEG 4000+polycarbophil	1	1		
PEG 4000+mineral oil	1			1
Dosage of PEG 4000 (g/kg/d)		0.47 (0.25–0.88)		

Values are presented as number only or median (range).
PEG: polyethylene glycol.

Table 4. Results of long-term therapy, including biofeedback and medication in children with dyssynergic defecation

Therapeutic modality	Patients (n=19)	Response		
		Good	Fair	Poor
Biofeedback+PEG 4000 regimen	8			
Medication	8	2*	6	
Biofeedback	8	6		2* (fail)
PEG regimen only	3	1		2
Including biofeedback failure	5 (3+2*)	3		2
Follow-up loss	8			

*Two children who had failed in biofeedback therapy, however showed good response to long term medication.

polyethylene glycol (PEG) 4000 (Forlax®, Ipsen Pharma), and the response was good in nine of 19 (47.4%) children, fair in nine of 19 (47.4%) children, and poor in one of 19 (5.0%) children. The median initial PEG 4,000 dose was 0.47 g/kg/d (0.25–0.88 g/kg/d) (**Table 3**). BF was performed in eight of 19 children, with good results in six (75.0%) children and failure in two (25.0%) children who were aged 7–8 years. In good response cases to BF therapy, frequency of bowel movement, and stool consistency improved dramatically, and medicine was tapered rapidly. The response to long-term medical therapy which last more than 12 months was analyzed in 11 of 19 children; three of five children showed good response with medication alone (these included two children who failed biofeedback, and one child without biofeedback), two of five children showed poor response (two children with only medication), and six out of eight children showed good response with a combination of BF and medication. Follow-up loss was noted in eight of the 19 children (**Table 4**).

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DISCUSSION

The pelvic floor consists of a pelvic diaphragm and puborectalis muscles, which are striated muscles controlled by voluntary will. During defecation, the pelvic floor and both anal sphincters relax to facilitate fecal movement during the Valsalva maneuver. Most children instinctively learn how to properly control these muscles when they are young. However, some children have problems with the learning process, and some lose their control function after certain events (e.g., injury). The former is thought to be more common in children with DSD or PFD. Chronic constipation followed by difficulty in defecation occurs if any problems with the above processes occur [6,7,10,11].

Although DSD or PFD is not easy to diagnose in children and the prevalence of DSD or PFD in children is not known precisely [6,7], it is one of the most common causes of chronic constipation in adults [10,11]. Proper diagnosis is important, because it can be relieved with BF therapy, a relatively easy treatment [12-14]. In our study, most of the children had suffered for a long period before diagnosis, and we believe that DSD or PFD should be considered an important cause of chronic constipation in children.

Adult DSD has been classified into four types using the dyssynergy concept, which evaluates the functions of the puborectalis muscle, pelvic diaphragm, rectum, and anal sphincters; In type I, with an adequate propulsive force and paradoxical increase in anal sphincter pressure. In type II, without an adequate propulsive force and paradoxical anal contraction. In type III, with adequate propulsive force, but there is either absent or inadequate relaxation of the anal sphincter. In type IV, without an adequate propulsive force together with an absent or inadequate anal sphincters relaxation [10]. This classification has currently been simplified into three types: 1) high anal sphincter pressure at rest and during defecation, 2) inadequate propulsive force, and 3) a hybrid of both disturbances [15]. This concept is also thought to be applicable to children [6]. Therefore, we believe that the clinical characteristics and diagnostic and therapeutic pathway of DSD or PFD evaluated in our study has important implications for the practical diagnosis and treatment of chronic constipation in children.

DSD or PFD can be diagnosed using high-resolution manometry (HRM), which can be accompanied by balloon expulsion test or three-dimensional (3D) reconstruction [6] or defecography (fluoroscopic or magnetic resonance imaging) [5,7,16-19], and each test method has its advantages and disadvantages. For children, the cooperation of patients, and the safety and feasibility of testing methods, are more significant considerations than the accuracy and availability of test methods.

If the child is cooperative, HRM has the advantages of being less expensive, not related to radiation, identifying the recto-anal inhibitory reflex, and evaluating various subjective and objective anorectal functions, if combined with the balloon expulsion test or 3D reconstruction [6]. However, since the test is carried out while the child is lying down, it is in a non-physiological state, which is different from the actual defecation posture. In addition, there are considerable limitations due to the age-dependent ability of cooperation, at least until mid-primary school age. For the balloon expulsion test, the possibility of DSD or PFD is significantly low if the emission is successful, but age-associated limitations can be even more significant. Young adolescent girls are more reluctant to participate in this test because of a sense of shame. The test has limitations in the evaluation of structural problems such as intussusception, rectocele, and rectal prolapse.

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FDG is inexpensive and can evaluate the actual dynamic physiology of defecation in the physiological posture. It is a relatively easy test to perform at an early age of 4 years or more. In addition, almost all anorectal outlet functions (such as rectum, pelvic floor, puborectalis muscle, and anal sphincters) can be evaluated directly, providing more comprehensive and dynamic information. Structural problems, such as intussusception, rectocele, rectal prolapse, and descending perineum syndrome, can also be evaluated [5,7,16,17]. However, there is an issue regarding radiation exposure, especially for girls with ovaries in the abdominal cavity. To avoid this disadvantage, the tests should be performed within a few minutes.

Magnetic resonance defecography (MDG) has almost all the advantages of FDG, and it provides more information about surrounding structures, and there is no risk of radiation exposure [18,19]. However, it is expensive and less physiological because it is tested while the child is in the supine position. In addition, the availability of this test is limited.

Therefore, we believe that the choice of diagnostic test depends on the circumstances of patients and pediatricians. According to our experience, HMR with balloon expulsion tests can be considered preferentially if the patient is almost 10 years old and the child is able to cooperate with the examination. FDG is first choice at a younger age if testing is essential because the test is simple and can be performed with ease. We believe that the two tests are complementary.

A recent pediatric study using HRM reported that 70–80% of patients who underwent HRM without a screening process had DSD and that 40% of these had puborectalis relaxation failure [6]. In addition, at least 50% of adult patients had DSD in adult study [11]. However, we believe that the prevalence rate diagnosed with HRM or electromyography findings alone can be different from the actual clinical situation. The accurate prevalence of clinically significant DSD or PFDs in children should be based on age-appropriate tests that combine balloon expulsion tests with/without HRM, defecography, or CTT test.

According to our data, approximately 50% of patients with abnormal CTT test results showed various abnormalities on FDG, and PFD was observed in 12/51 (23.5%) patients [7]. In another study conducted during the same period, abnormal results of CTT testing were observed in 51.6% (98/190) of all patients [2]. Considering the children who did not undergo FDG despite abnormal CTT test results, it can be estimated that PFD could be found in approximately 12.0% (12/100) of the abnormal CTT test group. This corresponds to 2.7% of the 450 patients who underwent CTT tests during the same period. Considering the number of patients who visited the hospital during that period, it was estimated that 0.9% of patients had clinically significant PFD. The significant differences between our study and those of other studies could be derived from the diagnostic methods. In HRM studies, abnormalities are determined only by pressure values. By contrast, FDG can perform direct visual observation of the pelvic floor and anal sphincter movements. Therefore, we believe that FDG is more useful than HRM for diagnosing clinically significant DSD or PFD which requires BF therapy. However, for FDG/MDG tests based on CTT test, there is a risk of underestimation because mild abnormalities that are clinically unclear may be missed.

Many children diagnosed with DSD or PFD responded relatively well to initial medication based on PEG 4000. Approximately 25% of the children maintained a good response to low-doses and could finish the treatment. This has significant implications for frontline doctors. A good response to PEG does not imply that there is no considerable underlying problem.

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Therefore, as shown in our study, DSD or PFD must be considered when there is chronic constipation, a very low frequency of defecation, and failure to taper PEG medication, especially when the results of the preceding CTT test are abnormal. In our study, in children with DSD or PFD, the outlet obstruction type was observed in 55.6%, slow transit type in 27.8%, and normal transit type in 5.6% on the CTT test. This finding implies that the interpretation of the CTT test results is not straightforward.

If the results of the preceding CTT test were abnormal, we think that it is necessary to conduct a follow-up CTT test even if the medication had been tapered successfully. If abnormal results are observed again, further studies with HRM and/or DG should be performed considering the presence of DSD or PFD or other possibilities. Some patients in our study underwent this process to achieve an accurate diagnosis.

Although long-term outcomes of BF therapy in general pediatric constipation are poor [14,20,21], BF can be an excellent treatment for DSD or PFD [6,12,13]. However, as shown in our study, it is difficult for young children to understand and cooperate with the treatment due to age limitations. In such cases, we recommend that patients wait until they can cooperate with therapy while maintaining minimal effective medication. According to our experience, children more than 8-9 years old could undergo BF therapy successfully.

In this study, HRM with balloon expulsion was performed only in a few children to rule out Hirschsprung disease. If more children underwent simultaneous HRM, this could be a more valuable study.

In conclusion, it is a reasonable diagnostic flow to conduct radio-opaque marker tests as a basic test to classify subtypes and conduct further evaluation of anorectal outlet function for abnormal result groups, especially in the outlet obstruction type. The balloon expulsion test with HRM or FDG can be used to evaluate anorectal outlet functions. DSD or PFD could be an important cause of chronic constipation in children and can be treated successfully with BF combined with medical therapy, although age-dependent limitations still exist.

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