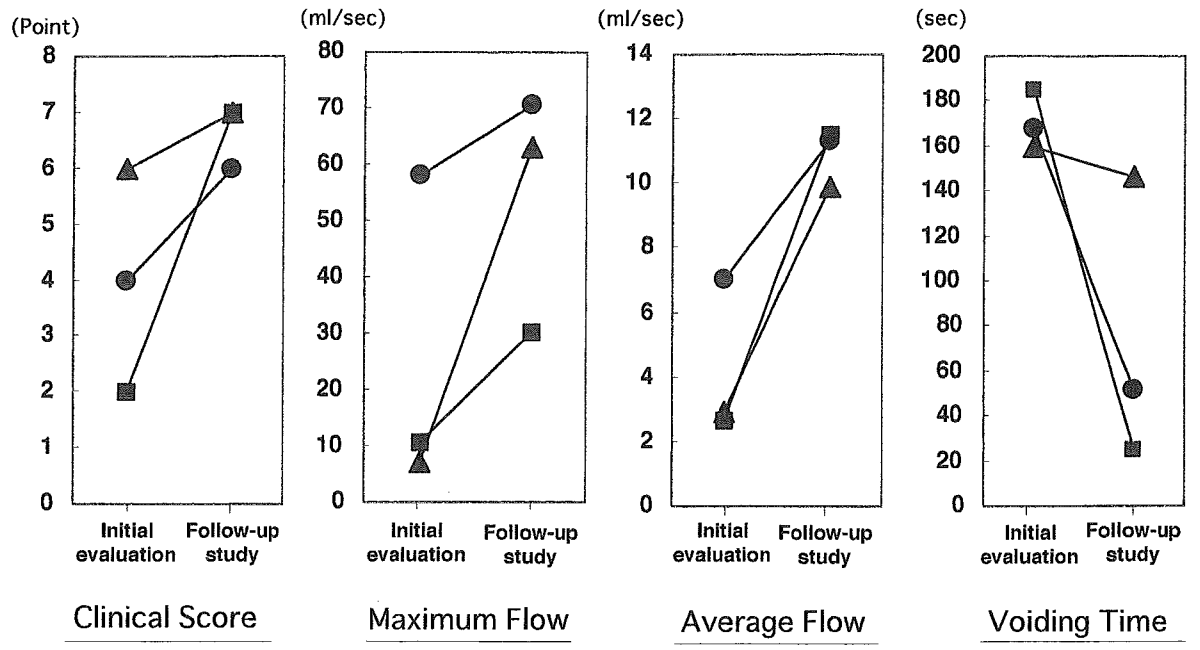

Fig 4. Results of follow-up studies for 3 patients with intermediate- or high-type anorectal malformations. The clinical scores and results of fecoflowmetric studies are shown for patients 4 (▲), 14 (■), and 16 (●). All the 3 patients exhibited improvement in clinical score and fecoflowmetric parameters in the follow-up study (compared with the initial evaluation). The patients' ages at the follow-up studies were 18, 12, and 7 years (patients 4, 14, and 16, respectively). The type of fecoflowmetric curves had changed from B to A in patients 14 and 16 at the time of follow-up.

study, 4 of 7 patients with RUF and 2 patients with AVF showed type I pressure fluctuations of the rectum and anal canal, which suggested the presence of functioning anatomic structures in these patients. In fact, these patients with type I pressure fluctuations had higher clinical scores than those of the patients with other types of pressure fluctuations. Patient 1 with anocutaneous fistula accompanying sacral bony defect and presacral meningomyelocele (Currarino Triad) had type II pressure fluctuation and low clinical score. Sacral neural defect is reported to be a major contributing factor for the disturbance of defecation in the patients with anorectal anomalies.[10,11] The unusually low clinical score in patient 1 as a patient with anocutaneous fistula may caused by the sacral bony and neural defects. In the patients with type II pressure fluctuations, pressure of anal canal dropped soon after saline infusion. Persistent relaxation of the anal canal during saline infusion may be responsible for incontinence seen in 4 of 6 patients with type II pressure fluctuations. Two patients of RUF (patient 15 and 16) who had type III pressure fluctuations suffered from severe constipation at the first evaluation, and required vigorous medical management such as daily use of glycerine enema, manual extraction of impacted stool, and colorectal irrigation. The lack of rectal contractions may indicate a disturbance in squeezing the contents out of the rectum. As was reported by Shafik,[12] an inactive rectum (rectal inertia) may cause severe chronic constipation. Sacral cord injury,[13] degenerative diseases, dysfunction of the intestinal neural network,[14] and chronic rectal distension by feces[15] have been reported to cause rectal inertia. Recently, Meier-Ruge reported the histologic neuromuscular changes of the rectal wall in high-type anorectal malformations.[16] Poor clinical bowel function of the 2 patients with type III pressure fluctuations is suggesting the importance of rectal contractile activity synchronized with the anal relaxations in defecation. The cause of rectal inertia of the 2 patients in our series was not known; however, the improvement of bowel functions of patient 16 at the follow-up study might suggest that the hypoactive rectum be reversible with patient 16. One important point observed in the follow-up study of the 3 patients was that they showed improvements not only in the clinical scores but also in the fecoflowmetric parameters. This means that these patients learned how to manage their social life with repaired anatomic components of the anorectum and also how to control them to achieve better bowel functions in the period between the initial evaluation and the second evaluation. In this study, fecoflowmetric parameters such as tolerable volume, the maximum flow rate, and average flow rate, as well as the configuration of fecoflowmetric curve itself were proved to be good clinical indicators of the bowel functions of the children after the repair of anorectal anomalies. In contrast to the controls, who had type I pressure fluctuations and type A fecoflowmetric

curves, only 3 of 7 patients of anorectal anomalies with type I pressure fluctuations exhibited type A fecoflowmetric curves. That is, rectal contractile activity synchronized with the relaxations of the anal canal does not directly connect to good findings in fecoflowmetry in patients of anorectal anomalies. Although patients with rectoanal coordination seen in type I pressure fluctuations had significantly higher clinical scores than those with other type of pressure fluctuations, other unevaluated factors such as motor activity of pelvic floor[17,18] and puborectalis muscle[19] may be playing roles for the bowel function of the patients after repair of anorectal malformations. Yagi et al[20] evaluated the ability to defecate with fecoflowmetric study and reported good correlations between fecoflowmetric parameters with Kelly's clinical score in children after repair of anorectal anomalies; however, they did not evaluated anorectal motilities. Anorectal motility plays an important role in defecation. We presented a simple and objective method consisting of saline enema test and fecoflowmetry for evaluating both the anorectal motilities and the ability to defecate. Therefore, not only the actual state of defecation but also the factors responsible for the malfunctioning anorectum are evaluated by this simple method. Without understanding the pathophysiology of the malfunctioning anorectum, no medical or surgical management could be done properly. The method of evaluating the anorectal functions presented in this report would be useful for pediatric surgeons in planning the management for anorectal malformations.

Fecoflowmetry, which is simple and noninvasive, gives us objective data for estimating the actual state of defecation along the clinical course of the patients after the repair of anorectal malformations. In the evaluation of bowel functions of patients with anorectal anomalies with altered or lacking anatomic structures of the anorectum, more attention should be paid to anorectal motility and fecodynamics as well as clinical scoring.

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資料4

ORIGINAL ARTICLE

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Fecoflowmetric evaluation of anorectal function and ability to defecate in children with idiopathic chronic constipation

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Abstract Idiopathic chronic constipation (ICC) is one of the most common clinical conditions in children. The pathophysiology is multifactorial and differs from case to case. To investigate the relationship between anorectal motility (ARM) and clinical course in children with ICC, anorectal function was evaluated using fecoflowmetry in nine children aged 2–14 years (mean 6.1). Three were boys and six were girls. Pressure fluctuations in the rectum and anal canal were simultaneously recorded during saline (250–500 ml) infusion into the rectum. The dynamics of defecation were evaluated using recordings of the saline evacuation curve from the rectum in each patient. Seven patients showed periodic contractions of the rectum accompanied (five) or unaccompanied (two) by relaxations of the anal canal during saline infusion. These patients achieved comfortable spontaneous defecation during follow-up periods ranging from 5 to 20 months. The other two exhibited no rectal contractions in spite of relaxations of the anal canal, and did not respond well to long-term medical management. In eight patients segmental fecoflowmetric curves showed a significantly lower flow rate and longer evacuation time than those of controls. Fecoflowmetry is a simple and non-invasive technique for evaluation of the ability to defecate. Disturbances of ARM may play an important role in patients with severe ICC. When evaluating anorectal function in children with chronic constipation, more attention should be paid to ARM and fecodynamics.

Keywords Fecoflowmetry · Anorectal function · Defecation · Constipation

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Introduction

Chronic constipation is a common complaint among patients in pediatric outpatient clinics. The majority of children with idiopathic chronic constipation CICC tend to improve on conservative treatment as they become older. On the other hand, some patients show no improvement in spite of vigorous medical treatment. In this study, we evaluated anorectal motility (ARM) and the ability to defecate using saline evacuation from the rectum in children with ICC. The importance of the evaluation of ARM and fecodynamics is emphasized.

Materials and methods

Children referred to Fujiwara Memorial Hospital because of ICC were examined. Patients with Hirschsprung's disease and metabolic, hormonal, or neurogenic disease were excluded. Nine patients (three boys, six girls) aged 2–14 years (mean 6.1) were included in this study. The duration of symptoms ranged from 4 months to 10 years (mean 3.6 years). The clinical details are summarized in Table 1. As controls, five children aged 6 to 13 years without problems in defecation were also evaluated.

After emptying the rectum by a glycerine enema, pressure fluctuations of the rectum and anal canal during saline infusion into the rectum were simultaneously recorded. The details of the procedures are described elsewhere [1]. In brief, while patients lie with the left side down, one pressure-monitor probe is positioned in the rectum and another in the anal canal. Transducers (DTS DX-360, Nihon Kohden, Tokyo) were connected via an amplifier (AD100F, Nihon Kohden) to a chart recorder (RTA-1100M, Nihon Kohden). Saline (300–500 ml) was infused at a rate of 30–50 ml/min.

After the pressure fluctuation curves were recorded, the patient was freed from the monitor and instructed to sit and evacuate the saline in the same manner as usual defecation. The saline evacuation curve was recorded with a uroflowmeter (UROFLO-MET

Table 1 Clinical characteristics of patients with constipation (*y* years, *m* months)

Patient no.	Age	Gender	Duration of symptoms	Symptoms at initial examination
1	2y10m	F	2y	Difficulty in evacuating hard stool, anal bleeding
2	2y11m	F	2y9m	Difficulty in evacuating hard stool, anal bleeding
3	3y0m	F	7m	Difficulty in evacuating hard stool, abdominal plain
4	3y10m	M	3y6m	Fecal impaction, anal bleeding
5	4y5m	M	4m	Fecal impaction with staining
6	4y8m	M	2y	Difficulty in evacuating hard stool, internal hemorrhoids
7	8y11m	F	4y	Fecal impaction with soiling, anal bleeding
8	10y	F	6y	Difficulty in evacuation hard stool, frequent abdominal pain and discomfort
9	14y0m	F	10y	Difficulty in evacuation hard stool, frequent abdominal pain and discomfort, no sense of accomplishment after defecation

Table 2 Findings of anorectal function tests

		Pressure fluctuations of rectum and anal canal		Fecoflowmetry
		Periodical rectal contractions	Synchronicity of rectal contractions and relaxations of anal canal	Shape of saline evacuation curve
Patients	1	Yes	Yes	segmental
	2	Yes	Yes	massive
	3	Yes	No	segmental
	4	Yes	Yes	not done
	5	Yes	No	segmental
	6	Yes	Yes	segmental
	7	Yes	Yes	not done
	8	No	No	segmental
	9	No	No	segmental
Controls	1	Yes	Yes	massive
	2	Yes	Yes	massive
	3	Yes	Yes	massive
	4	Yes	Yes	massive
	5	Yes	Yes	massive

SUF200, Sakura Tokyo). The shape of the evacuation curve, maximum flow rate (ml/s), flow time (s), mean flow rate (voided volume/flow time; ml/s), and total evacuation time (s) were evaluated. The flow time was defined as the sum total of the time with saline flow. The total evacuation time was the time from the beginning of evacuation to the end of the study; that is, the total of flow time and intermissions.

Results

The pressure fluctuations in the rectum and anal canal during saline infusion showed a periodic increase in rectal pressure synchronized with relaxations of the anal canal in five patients and all the controls (Table 2). One patient had irregular rectal pressure fluctuations unsynchronized with anal-canal pressure. One patient, who had incontinence as well as chronic constipation, showed relaxation of the anal canal soon after initiation of the saline infusion without recovery, thereafter accompanied by frequent leakage of saline from the rectum. In two patients there were no elevations in rectal pressure in spite of relaxations of the anal canal during saline infusion (Fig. 1).

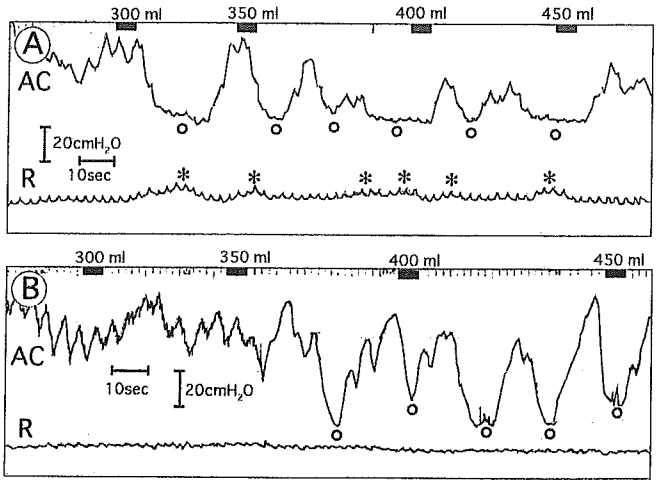

Fig. 1 Pressure fluctuations in rectum and anal canal in two patients: **A** Patient 2 shows periodic relaxations of anal canal (○) synchronized with increase in rectal pressure (*) after infusioin of 300 ml saline. **B** Patient 9 shows no contractions of rectum during saline infusion and only periodic relaxations of the anal canal (○). She had no urge to defecate even in presence of repeated anal-canal relaxations (*AC* anal canal, *R* rectum)

The saline evacuation curve was recorded in seven patients. Six showed a segmental fecoflowmetric curve, while one patient and all the controls showed a massive curve. The maximum flow rate of the patients ranged from 11.2 to 27.5 ml/s (mean 19.3 ml/s) and was significantly lower than that of the controls (58.1 ml/s). The average flow rate was significantly lower in the patients with ICC (5.1 ± 0.8 ml/s) than in controls (22.0 ± 4.3 ml/s). Both the total evacuation (127.9 ± 36.8 s) and flow times (43.1 ± 8.4 s) of the patients were significantly longer than those (16.5 ± 2.7 and 15.5 ± 1.4 s, respectively) of controls (Fig. 2). One patient exhibited a low-peaked segmental evacuation curve with a long evacuation time and had no sense of accomplishment for the evacuation in spite of her effort. Examples of fecoflowmetric curves obtained in this study are shown in Fig. 3.

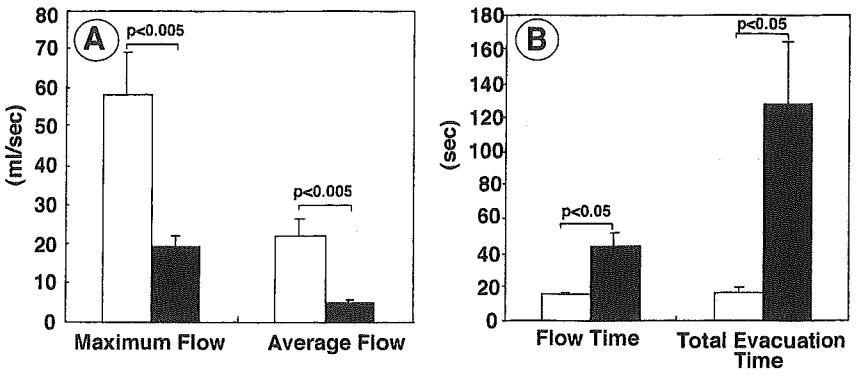

Fig. 2 Fecoflowmetric parameters of controls and patients with idiopathic chronic constipation. **A** Maximum and average flow rates of patients (*closed bars*) significantly lower ($P < 0.005$) than those of controls (*open bars*) **B** Flow time and total evacuation time of patients significantly longer ($P < 0.05$) than those of controls

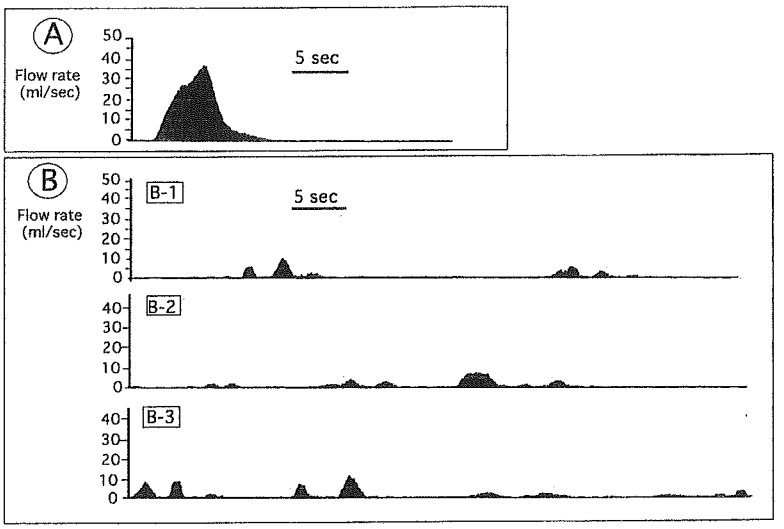

Fig. 3 Fecoflowmetric curves obtained from a control and a patient. **A** All children without defecation problems showed massive fecoflowmetric curves with high flow rate and short evacuation time. **B** Fecoflowmetric curve from patient 9: segmental and divided into small humps, reflecting poor ability and lack of urge to defecate (Tracings B-1, B-2, and B-3 continuously recorded; maximum flow rate and evacuation time 11.2 ml/s and 169.7 s, respectively)

The five patients who showed periodic rectal contractions synchronized with relaxations of the anal canal achieved comfortable spontaneous defecation without medication 6 to 20 months after the initial evaluation. Two further patients achieved daily bowel movements with laxatives. One patient still has abdominal pain and needs considerable effort and time to defecate. The frequency of her bowel movements is twice a week, and she often requires a glycerine enema to gain a sense of accomplishment. One patient still requires temporal use of glycerine enemas for complete evacuation of feces from the rectum. Discontinuation of medical support resulted in frequent abdominal pain and nausea (Table 3).

Discussion

Constipation represents a common problem in children, and is reported to account for 25% of visits to pediatric gastroenterology clinics [2]. The majority of pediatric patients with ICC recover with conservative treatment as they become older. On the other hand, there are children with severe, intractable constipation without an apparent causative disease. Both these patients and their parents experience anxiety over their intractable symptoms; however, the pathophysiologic grounds of ICC are poorly understood. Multiple factors such as dietary habits, autonomic nervous function, psychiatric problems, anal pain at defecation, and ARM may affect bowel habits.

Several abnormalities of anorectal function such as failure of rectoanal coordination [3], neuropathic rectal defects [4], abnormal motor activity of the pelvic floor [5, 6], decreased relaxation of the internal anal sphincter [7], paradoxical puborectalis contraction [8], and an abnormality of the colonic myenteric plexus [9] were reported in patients with ICC. These reports suggested the importance of ARM in the pathophysiology of ICC. It is not surprising that manometric parameters such as resting and squeeze pressures of the anal canal are not suitable clinical indicators for ICC, because ARM and the ability to defecate cannot be properly evaluated with these parameters [10, 11].

Although the presence of a rectoanal reflex is a key diagnostic finding for excluding Hirschsprung's disease (HD) [12], it may not, be a key factor regulating the clinical symptoms in children with ICC. In the present study, both squeezing and resting pressures were not significantly different between normal subjects and patients with ICC (data not shown). Fecoflowmetry [1, 13, 14] and scintigraphic defecography [15] have been

Table 3 Treatments and clinical courses of patients

Patient no.	Initial treatment	Follow-up period (months)	Present status of bowel movements		
			Stool frequency (times/week)	Maintenance treatment	Difficulty at defecation
1	Regular use of laxative	5	7 or more	None	None
2	Temporal use of laxative and glycerine enema	18	7 or more	None	None
3	Temporal use of laxative	20	7 or more	Laxative	None
4	Temporal use of laxative	36	7 or more	None	None
5	Manual extraction of impacted stool, Temporal use of glycerine enema	18	7 or more	Laxative	None
6	Temporal use of laxative	24	7 or more	None	None
7	Temporal use of laxative	6	7 or more	None	None
8	Regular use of laxative, temporal use of glycerine enema	12	6	Glycerine enema	Frequent abdominal pain
9	Regular use of laxative, temporal use of glycerine enema	29	2	Laxative and Glycerine enema	Difficulty in evacuting stool, No sense of accomplishment after defecation, Frequent abdominal pain

reported to be suitable methods for quantitative and dynamic assessment of the ability to defecate.

As we reported previously in adults [1], the type of anorectal pressure fluctuations during saline infusion into the rectum was in close relation with the clinical symptoms and the findings in a fecoflowmetric study. In our previous report, we classified the pressure fluctuations into five major types as follows: type I, the anal canal relaxes periodically and synchronously with the contractions of the rectum; type II, the anal canal relaxes soon after saline infusion, and anal canal pressure remains at the same level as rectal pressure; rype III, the anal canal relaxes periodically without rectal contractions; type IV, there is no relaxation of the anal canal during saline infusion; and type V, relaxations of the anal canal and rectal contractions occur irregularly and independently of each other. In the previous study, we found that type I and type II pressure fluctuations were dominant in normal and incontinent patients, respectively, which was consistent with the result reported by Read et al. [16]. Type III pressure fluctuation was found in patients with severe, long-standing constipation. Type IV was found in patients HD who responded poorly to surgical therapy.

The outcome of patients in this study, i.e., children with rectal contractions during saline enema achieving spontaneous daily bowel movements as they grew, may suggest that rectal contractions synchronous with anal-canal relaxations are important for normal defecation. As we anticipated, patients who failed to show rectal contractions (type III) at the first evaluation responded poorly to long-term medical management. The lack of rectal contractions may indicate a disturbance in detrusor activity of the rectum, and thus may predict their poor response to medical care. Poor improvement of clinical symptoms in patients with type III pressure fluctuations suggested the importance of rectal motility in the mechanism of defecation.

A decrease of rectal action potentials was reported in patients with chronic constipation with an inactive rectum (rectal inertia) [17]. An inactive rectum is caused by sacral cord injury [18], degenerative diseases, dysfunction of the intestinal neural network [2], and chronic rectal distension by feces [19]. The etiology of the hypoactive rectum in patients 8 and 9 may have been chronic rectal distension by feces, because they had no other apparent disease. Constipated patients with fecal impaction should be treated meticulously to prevent the rectum from becoming inactive.

The ability to defecate in children was well-evaluated in this study using fecoflowmetry. All the normal children exhibited a massive fecoflowmetric curve with short evacuation times and high flow rates. Six of seven patients with ICC had a segmental fecoflowmetric curve irrespective of the type of pressure fluctuations. The patients with ICC had significantly lower maximum and average flows and longer flow times and total evacuation times compared to normal children.

The fact that the patients with chronic constipation were well-distinguished from normal children by fecoflowmetric parameters suggests the usefulness of fecoflowmetry in the evaluation of the ability to defecate. Although it may be argued that the evacuation of saline from the rectum is not physiological and may not represent the actual state of fecodynamics, no significant differences were reported in configuration or flow parameters between water and paste fecoflowmetry [14]. Evaluation of the ability to defecate using saline evacuation from the rectum provided us with objective data for estimating the actual state of defecation in constipated children. Furthermore, it helped us show patients and their parents the results of management during the clinical course. For patients with an inactive rectum, a new therapeutic approach such as electrical pacing of the rectum [20] may be required to improve their clinical symptoms if they fail to respond to long-term vigorous medical management.

Fecoflowmetry is a simple and non-invasive technique for evaluation of the ability to defecate. When evaluating anorectal function in children with chronic constipation, more attention should be paid to ARM and fecodynamics.

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Evaluation of Anorectal Function in Patients With Tethered Cord Syndrome: Saline Enema Test and Fecoflowmetry

by

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Evaluation of anorectal function in patients with tethered cord syndrome: saline enema test and fecoflowmetry

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Object. Disturbance in anorectal function is a major factor restricting the activities of daily living in patients with spinal cord disorders. To detect changes in anorectal motilities due to a tethered spinal cord, anorectal functions were evaluated using a saline enema test and fecoflowmetry before and after patients underwent untethering surgery.

Methods. The bowel functions in five patients with a tethered cord syndrome (TCS) were evaluated by performing a saline enema test and fecoflowmetry. The contractile activity of the rectum, the volume of infused saline tolerated in the rectum, anal canal pressure, and the ability to evacuate rectal content were examined.

The characteristic findings in anorectal motility studies conducted in patients with TCS were a hyperactive rectum, diminished rectal saline-retention ability, and diminished maximal flow in saline evacuation. A hyperactive rectum was considered to be a major contributing factor to fecal incontinence. In one asymptomatic patient diminished anal squeezing pressure was exhibited and was incontinent to liquid preoperatively, but recovered after surgery. Two patients who underwent surgery for myeloschisis as infants complained of progressive fecal incontinence when they became adolescents. In one patient fecal incontinence improved but in another patient no improvement was observed after untethering surgery.

Conclusions. Fecodynamic studies allow the detection of neurogenic disturbances of the anorectum in symptomatic and also in asymptomatic patients with TCS. More attention should be paid to the anorectal functions of patients with TCS.

KEY WORDS • **tethered cord syndrome** • **anorectal function** • **saline enema test** • **fecoflowmetry** • **pediatric neurosurgery**

NEUROGENIC bowel dysfunction is one of the major life-limiting problems in patients with a spinal cord lesion.[7,17] Tethered cord syndrome affects mainly children and, in some cases, causes permanent neurological dysfunction of the bowel. Progressive sensorimotor changes in the legs and bladder/bowel dysfunctions as the patient matures are characteristic symptoms of TCS. The causes vary and include myelomeningocele, lipoma, epidermal cyst, and thickened tight terminal filum. A delay in the diagnosis and untethering surgery results in poor outcome such as persisting fecal and/or urinary incontinence and lower-extremity weakness. The proper time for untethering surgery depends on multiple factors such as patient age, concomitant disease, the surgery-related risk, and clinical symptoms. Disturbance of bowel functions is one of the major limiting factors for the daily life of patients; however, data derived from anorectal function tests in this condition are limited. To detect changes in anorectal motilities due to a TCS, anorectal functions were evaluated using saline enema test and fecoflowmetry before and after untethering surgery.

Abbreviations used in this paper: SD = standard deviation; TCS = tethered cord syndrome.

Clinical Material and Methods

Patient Population

Between 1996 and 2002, we evaluated anorectal functions in five patients with TCS before and after untethering surgery by using a saline enema test and fecoflowmetry. Patient age at the first evaluation ranged from 4 to 12 years (mean 9.2 ± 3.1 years [± SD]). Four were girls and one was a boy. Diagnosis was confirmed by magnetic resonance imaging. Clinical signs and symptoms were progressive sensorimotor changes in legs (three patients), neurogenic bladder (three patients), constipation (two patients), lumbago (two patients), and fecal incontinence (two patients). No apparent symptoms related to TCS were noted in one patient (Table 1). Seven children without bowel problems participated as controls. The age of controls ranged from 4 to 13 years (mean 9 ± 3.7 years).

Manometric and Fecoflowmetric Studies

Manometric and fecoflowmetric studies were performed as previously described.[10] Briefly, after the rectum was emptied by glycerine enema, patients lay with the left side down. One open-tipped probe was positioned in the rectum

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TABLE 1
Clinical features in five patients with TCS

Case No.	Age (yrs), Sex	Lesion(s) Responsible for Tethering	Bowel Movement Function	Neurogenic Bladder
1	11, F	sacrococcygeal lipoma, spina bifida occulta	chronic constipation	no
2	4, F	sacral lipoma	normal	no
3	12, M	repaired lumbosacral myeloschisis	incontinence & constipation	yes
4	10, F	spinal lipoma	normal	yes
5	9, F	repaired lumbosacral myeloschisis	incontinence & constipation	yes

and another in the anal canal. Each probe was perfused with water by a perfusion bag (Teruflex Medi-Quic ACS-222; Terumo Co., Tokyo, Japan) through a low-compliance infusion line coupled to a pressure transducer (DTS DX-360; Nihon Kohden Co., Tokyo, Japan). The transducer was connected via an amplifier (AD100F; Nihon Kohden Co.) to a chart recorder (RTA-1100M; Nihon Kohden Co.). The resting pressure of the anal canal was measured using the pull-through method. The patient was then instructed to squeeze maximally so that the greatest squeezing pressure could be recorded with the probe positioned at the site of the maximum anal canal resting pressure. Next, 500 ml of saline was infused into the rectum at a rate of 50 ml/minute, and the rectal and anal canal pressures were simultaneously recorded. The volume of saline required for inducing anal relaxations and rectal contractions was recorded. A relaxation of the anal canal seen just after initiating infusion was interpreted as an "initial drop" caused by abrupt saline infusion and was neglected in the analysis. The volume tolerated was also checked on the chart. The patient was then freed from the pressure-monitoring apparatus and instructed to sit on the commode to evacuate saline in the same manner as usual defecation. The commode was connected to a uroflometer (Uroflo-Met SUF200; Sakura Co., Tokyo, Japan, or Dantec Medical A/S, Menuet Compact, Denmark). The maximum flow rate and a saline-evacuation curve were recorded.

Statistical Analysis

All measured values are presented as the means ± SDs. When comparing three or more groups of data, the Fisher PLSD method was used as a post hoc test, and significance was set at a probability value less than 0.05 after analysis of variance.

Results

Clinical Course of the Cases

Case 1. This 11-year-old girl suffering from chronic constipation (one bowel movement every 7–10 days) for several years was referred to our clinic after a soft mass was found in the sacral region and lumbago persisting for several months. She was the fastest runner in her class until she was 8 years of age. At age 11 years, she was the slowest runner in her class. She and her family, however, were unaware of the pathognomonic condition until TCS, accompanied by sacrococcygeal lipoma and spina bifida occulta, was diagnosed. There was no sign of neurogenic bladder. An anorectal function test performed before untethering surgery showed poor contractions of the rectum and no urge to defecate during rectal saline infusion. The hypoactive rectum was thought to be one of the factors contributing to her chronic constipation. The fecoflowmetric curve was classified as "segmental," a classification usually seen in patients with chronic constipation. Reevaluation 3 months after treatment showed increased squeezing pressure compared with preoperative status (120 and 80 cm H_2O, respectively). The saline volume required to induce anal canal relaxation and tolerable volume were also increased after the treatment (100 and 500 ml, respectively) compared with those recorded before the treatment (50 and 250 ml, respectively). Moreover, rectal contractions synchronous with relaxations of the anal canal were shown during a saline enema test after treatment (Fig. 1). Postoperatively she experienced a strong urge to defecate when a 350-ml volume of saline was infused rectally. The maximum flow of saline from the rectum at evacuation increased nearly twofold over the preoperative flow (39.3 and 20.8 ml/second, respectively). The fecoflowmetric curve remained classified as segmental; nevertheless, she experienced a spontaneous bowel movement every day after treatment (Fig. 2).

Case 2. This 4-year-old girl with lipomyelomeningocele accompanying a TCS and sacral lipoma was referred to our clinic. She was asymptomatic. An anorectal function test demonstrated no abnormality except a prolonged relaxation of the anal canal after a rectoanal reflex elicited by balloon inflation in the rectum, which disappeared after untethering surgery. She remained asymptomatic during the 2-year follow-up period.

Case 3. This 12-year-old boy had suffered fecal and urinary incontinence and incomplete lower-leg palsy since birth. Lumbosacral myeloschisis, hydrocephalus, contracture of the hip, and pes calcaneus were present. He had undergone surgery for myeloschisis and hydrocephalus on the 2nd day after being born. Neurological examination demonstrated a disturbance below the L-5 level. A manometric study conducted at 1 month of age revealed a patulous external anal sphincter. When he reached puberty, he complained of progressing deformity, leg weakness, and lumbago. A diagnosis of secondary tethering of the spinal cord was made. He experienced complete bowel and bladder incontinence. No rectal contractions were observed during saline infusion. Untethering surgery was undertaken 6 months after he noticed worsening neurological symptoms. After surgery, his rectum became hyperactive. Anal canal relaxation was induced by a 30-ml infusion of saline. He experienced no clinical improvement in bowel function after surgery; however, the lumbago improved.

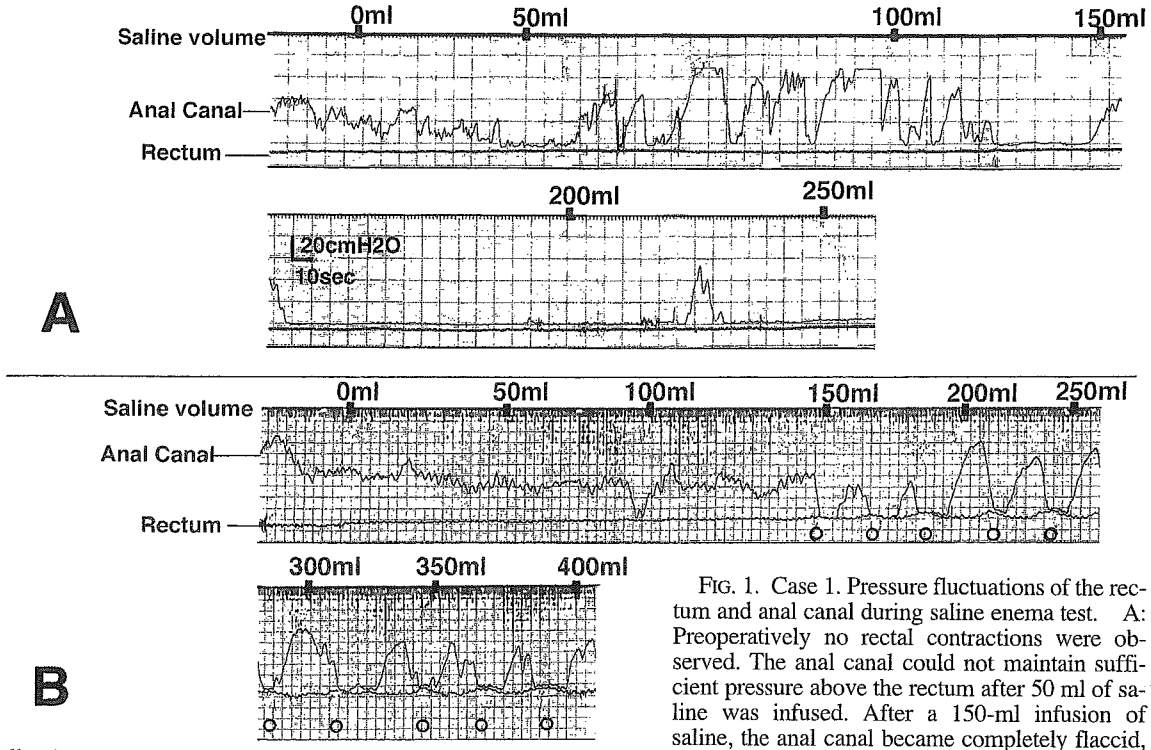

FIG. 1. Case 1. Pressure fluctuations of the rectum and anal canal during saline enema test. A: Preoperatively no rectal contractions were observed. The anal canal could not maintain sufficient pressure above the rectum after 50 ml of saline was infused. After a 150-ml infusion of saline, the anal canal became completely flaccid, allowing saline to flow out when 250 ml was infused. The urge to defecate was absent. B: A series of rectal contractions synchronous with the relaxations of the anal canal *(open circles)* appeared after surgery. The patient first felt the urge to defecate when 150 ml of saline was injected. She experienced a strong urge at 350 ml and was continent at 500 ml.

Case 4. This 10-year-old girl with spinal lipoma and neurogenic bladder was referred to our clinic to undergo anorectal function tests. At the age of 1 year TCS due to spinal lipoma was diagnosed. She was asymptomatic; however, at age 4 years her bladder was found to be neurogenic. At age 10 years, she began to complain of a dull pain in her right leg. Neurological examination revealed accentuated deep tendon reflex of the legs and the presence of patellar and ankle clonus. The neurogenic bladder was of the hypoactive type. She did not complain of difficulty with bowel function. Preoperative evaluation, however, revealed a low squeezing anal pressure (50 cm H_2O) and massive anal leakage of saline after a 150-ml infusion rectally. The neurosurgeons partially removed the sacral lipoma, untethered the spinal cord, and recapped the laminectomy. Squeezing pressure and the maximum volume tolerated improved (115 cm H_2O and 500 ml, respectively) by 1 month after untethering surgery. There was no apparent change in urodynamic status.

Case 5. This teenaged girl had undergone surgical repair of lumbosacral myeloschisis on the day of birth and a ventriculoperitoneal shunt was placed to treat accompanying hydrocephalus on the 6th day. When she was 4 months of age, a neurogenic bladder with Grade II vesicoureteral reflux was diagnosed. At age 9 years, anorectal function was evaluated after the patient complained of chronic constipation, which she had experienced since birth. Although her external anal sphincter was not active, the resting anal pressure was 100 cm H_2O at the first evaluation. A series of rectal contractions and relaxations of the anal canal were induced by a 120-ml infusion of saline rectally. She could tolerate up to 150 ml of saline rectally, and fecoflowmetry indicated a maximum flow rate of 47.6 ml/second. At 13 years of age, she was referred to our clinic for treatment of progressive leg weakness, lumbago, and bowel and bladder incontinence. The second anorectal evaluation revealed decreased anal canal pressure (35 cm H_2O) and a 10-ml threshold to induce rectal contractions (Fig. 3), resulting in low volume toleration (60 ml) and decreased maximum flow rate (21.2 ml/second) (Fig. 4). Deterioration of her anorectal functions was apparent. The neurosurgeons freed the spine of adhering dura from L-4 to the sacrum. Duraplasty and laminoplasty (L-5) were also performed after untethering. There were no remarkable improvements in urodynamic and fecodynamic functions 1 month after surgical treatment. It was almost 1 year postoperatively that she noted improved bowel habit and lower-extremity sensorimotor functions. At 2-year follow-up evaluation anal canal squeezing pressure, the threshold for rectal contractions, and tolerated saline volume had improved (80 cm H_2O, 250 ml and 300 ml, respectively [Fig. 3]).

Comparison of Anorectal Functions Between Patients and Controls

Data are shown as the means ± SD. Anal canal resting pressures before and after untethering surgery, respectively, were 51.3 ± 23.9 cm H_2O and 57 ± 11 cm H_2O, compared with that of 73.2 ± 23.7 cm H_2O in controls. There was no significant statistical difference. The mean anal canal squeezing pressures were 66.3 ± 29.3 cm H_2O

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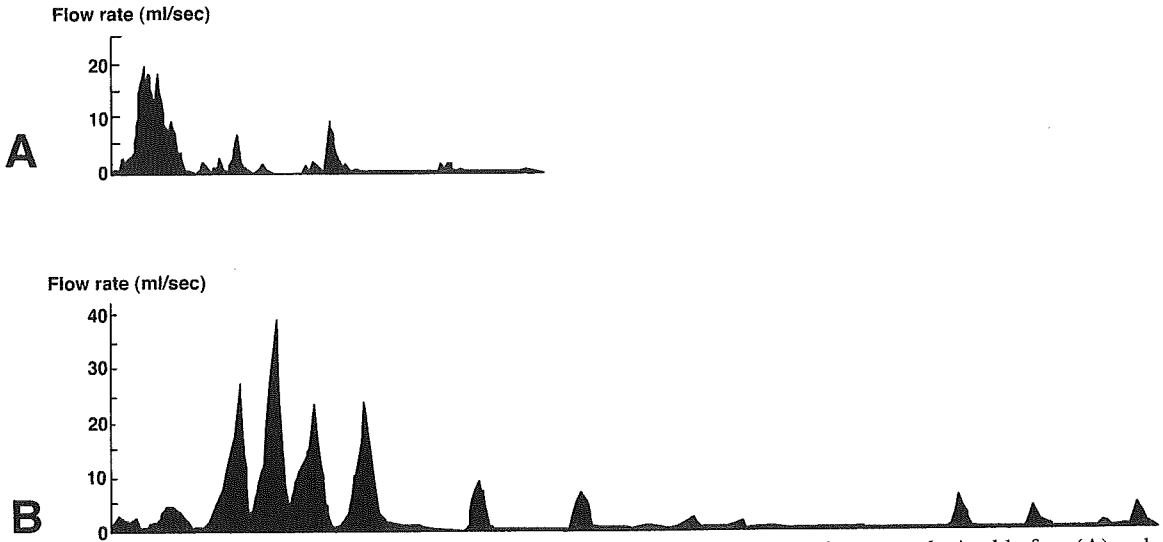

FIG. 2. Case 1. Fecoflowmetric curves before and after surgery. The fecoflowmetric curves obtained before (A) and after (B) untethering surgery were both classified as "segmental," which is usually seen in patients with chronic constipation. The maximum flow rate reached 39.3 ml/second, and 471 of 500 ml of injected saline was evacuated postoperatively.

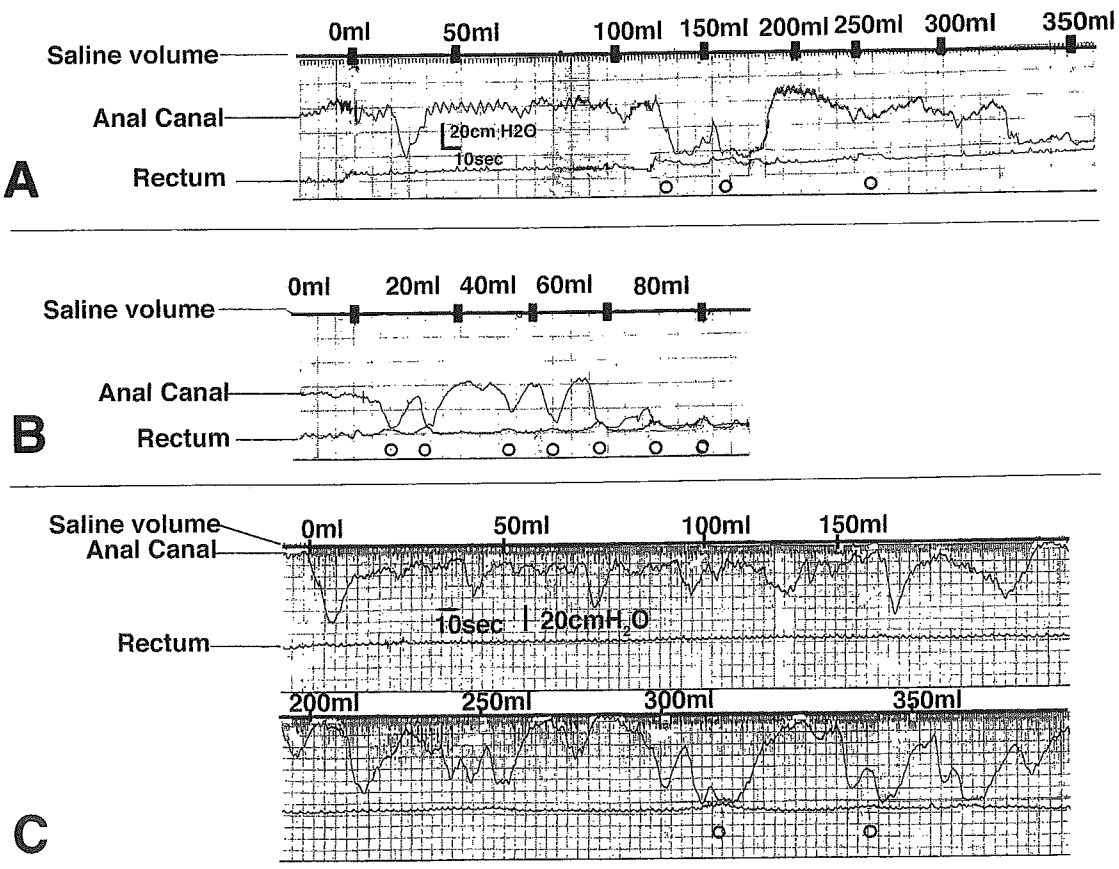

FIG. 3. Case 5. Pressure fluctuations of the rectum and anal canal during saline enema test. At 9 years of age the patient could tolerate a 150-ml saline injection. A: A series of rectal contractions *(open circles)* accompanying relaxations of the anal canal were induced after a 120-ml saline infusion. B: At 13 years of age, when she noticed progressive leg weakness and worsening of urinary and fecal incontinence, a hyperactive rectum and lowered tolerable volume (60 ml) were exhibited. C: Tracings recorded 2 years after untethering surgery demonstrated no sign of a hyperactive rectum. Full relaxation of the anal canal accompanied by rectal contraction was seen after a 300-ml infusion of saline.

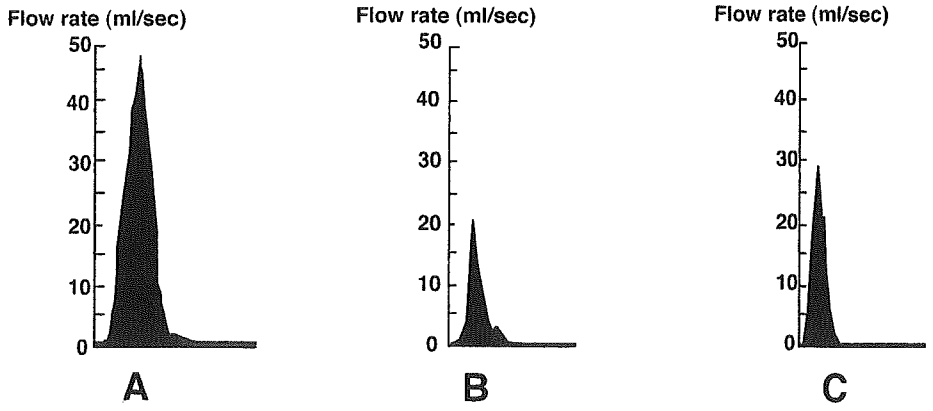

FIG. 4. Case 5. Fecoflowmetric curves before and after surgery. Fecoflowmetric curves recorded at ages 9 years (A), 13 years (B) and 15 years (C). Deterioration of control of bowel movement was obvious by age 13 years (B). After untethering surgery improvement was shown (C), even though it was not satisfactory.

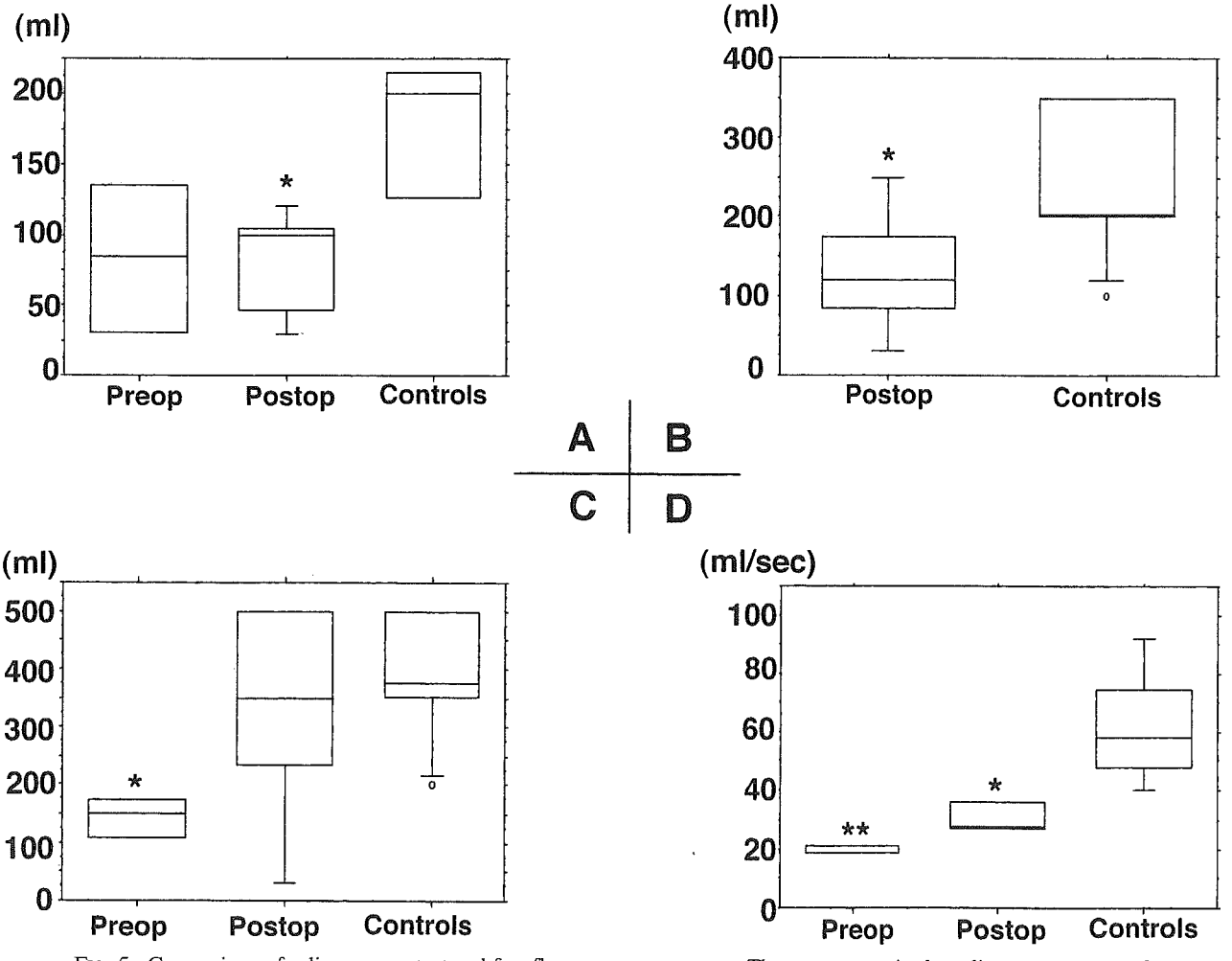

FIG. 5. Comparison of saline enema test and fecoflowmetry parameters. The parameters in the saline enema test and fecoflowmetry are shown in box plots. Horizontal bars represent the 10th, 25th, 50th, 75th, and 90th percentiles from bottom to top in that order. The saline volume required to induce anal canal relaxation was significantly lower in the patients who underwent surgery for TCS (A) than in controls, as was the volume required to induce rectal contractions (B). In the patients a significantly lower volume could be tolerated before untethering surgery (C) than in controls. The maximum flow rate was significantly lower in the patients than in controls (D). *$p < 0.05$, **$p < 0.01$.

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before surgery and 96.3 ± 25 cm H_2O after surgery compared with 110.5 ± 49.5 cm H_2O in controls. The mean postoperative volume of saline required to induce rectal contractions was 130 ± 80.3 ml, and this was significantly lower in controls (250 ± 100 ml). Because no rectal contractions were demonstrated in Cases 1 and 3 before untethering surgery, statistical analysis of rectal contraction threshold before treatment was not performed. The volume of infused saline tolerated was significantly smaller in the patients before surgery (140 ± 58.3 ml) than that in controls (383.3 ± 112.5 ml). The tolerated volume increased after the treatment (336 ± 193 ml). The ability to defecate was evaluated before and after surgery in all patients except the patient in Case 2. The maximum flow rate of saline evacuated rectally was significantly lower in the patients before surgery (19.7 ± 2.2 ml/second) than that in controls (53.8 ± 26.8 ml/second). The maximum flow rate increased after untethering surgery (31.1 ± 7 ml/second) (Fig. 5). The types of fecoflowmetric curves were "massive" in Cases 2 and 4 as well as in all controls, in whom no problem with bowel movements was present. In the patient in Case 5, who suffered constipation and incontinence, the massive fecoflowmetric curve was also exhibited during the series of evaluations. Because the maximum flow rate and the voided volume reflect, at least to some extent, the infused volume of saline tolerated by the rectum, the series of fecoflowmetric curves demonstrated in Case 5 were thought to indicate the progression or recovery of bowel functions. In the patient in Case 1, who suffered from chronic constipation, a segmental fecoflowmetric curve was demonstrated. The flat-type curve, usually observed in patients with severe incontinence, was seen in Case 3.

Discussion

Tethered cord syndrome is a unique pediatric neurosurgical condition. Lesions anchoring the spinal cord, such as myelomeningocele, intradural lipoma, and tight terminal filum create traction forces that stretch the spinal cord as the individual grows.[1] Symptoms induced by spinal cord tethering include neurogenic bladder dysfunction, anorectal dysfunction, lower-extremity sensorimotor changes, scoliosis, and back pain.[6,12,18] Early detection and treatment are reportedly major contributing factors to a better functional prognosis.[4,13] In addition to urological or orthopedic problems, bowel dysfunctions such as fecal incontinence and severe constipation are serious daily problems.[6,11,18] In contrast to the large number of reports on urological evaluations, only a limited number have been published on bowel functions in TCS.[5,6]

Combined with conventional manometric studies, saline enema tests and fecoflowmetry provides objective bowel function–related data reflecting the actual state of defecation.[8,9] Spinal lesion–induced bowel dysfunctions are classified into two major categories: upper motor neuron (hyperreflexive) bowel caused by lesions cranial to the conus medullaris (S2–4) and lower motor neuron (areflexive) bowel caused by lesions of the conus medullaris.[3,16,19] In our series, hyperreflexive bowel dysfunction was demonstrated before surgery in Cases 3 and 5. In the patient in Case 5 a higher volume-related threshold for inducing rectal contractions was exhibited after surgical treatment; consequently, continence improved. Preoperative and postoperative evaluations in the patient in Case 5 suggested that the hyperreflexive bowel was an important factor responsible for fecal incontinence. It has been reported that not fewer than 10% of patients with myelomeningocele developed TCS.[2,18] According to one report, the higher the level at which the myelomeningocele is present, the younger the age at which the patient develops TCS.[15] Any clinical presentation of TCS after the repair of myelomeningocele requires thorough investigation and early treatment to prevent the progression of neurological disturbance. In the patients in Cases 1 and 4, both of whom were unaware of the pathognomonic conditions of their bowel functions, insidious impairments of bowel functions were demonstrated on preoperative evaluation. Palmer, et al.,[14] reported that in 75% of children presenting with nonneurological symptoms of TCS subclinical changes in bladder function were revealed. Neurological centers controlling defecation are located close to the those controlling urination in the spinal cord at the sacral level, the pons, and the cerebral cortex. White, et al.,[19] have emphasized that the neurogenic mechanism is not necessarily upset to the same degree in both the bladder and colon. Furthermore, as reflected by findings in Case 1, the bowel dysfunction is not always accompanied by neurogenic bladder. Bowel function tests are essential to detect early neurological changes in patients in whom urological or sensorimotor manifestations are absent. Poor voluntary control of the external anal sphincter and a hyperactive rectum are probably major contributing factors to fecal incontinence in patients with TCS. Newly emerged or progressive anorectal dysfunctions, such as weakness of the external anal sphincter and hyperactive rectum, indicate the proper time for untethering surgery. It is not, however, always easy to determine the right time to undertake surgical untethering or to predict surgery-related improvements. A considerable delay in treatment may ultimately result in permanent poor bowel functions. Serial evaluation of bowel functions and meticulous observation of clinical symptoms are essential for deciding whether to perform untethering surgery.

Thus, evaluation of bowel functions involving a saline enema test and fecoflowmetry in patients with TCS proved to be beneficial in detecting insidious change. Furthermore, postoperative evaluation of anorectal functions provided objective data indicating the effect of the untethering surgery.

Conclusions

Evaluations of anorectal functions in patients with tethered spinal cord provide useful information for determining the proper time for untethering surgery. More attention should be paid to bowel dysfunctions in patients with TCS.

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― アンケートへのご協力のお願い ―

拝啓、

時下ますますご健勝のほどお喜び申し上げます。いつも一方ならぬお力添えにあずかり、誠にありがとうございます。

私は、秋田大学医学部統合医学講座臨床検査医学分野助教授の萱場広之と申します。

２０年来、秋田県を中心に地域医療に従事しております。平成１６年度より厚生労働省の長寿科学研究事業により、「高齢者の排便機能障害評価法と尊厳の回復に関する研究」という課題に取り組んでおります。地域医療をめぐる問題は我々医療従事者のみならず、この地で暮らす全ての人々にとって切実な問題であり、排便機能障害も例外ではありません。本研究の目的は、高齢者の排便機能障害の病態の分析・評価法を確立し、その病態に合った管理を通じて排便機能障害を有する高齢者の排便機能の向上やQOLの向上を目指すものです（別紙模式図）。研究の過程で派生するものとして、補助具や装具の工夫や社会基盤整備への提言も含まれるものと考えております。はなはだ困難な課題ではございますが、いくつかの補助具の考案と排便機能評価法の開発が軌道にのりはじめ、平成１７年度からは社会基盤や介護現場での実態調査に着手したところです。

介護現場の状況を知りたく、別紙のごときアンケート調査を計画いたしました。アンケートは無記名であり、また、ご協力頂いた皆様や施設の特定も行われぬよう、データのみの処理をいたします。

つきましては、本研究の主旨へのご理解を賜り、アンケート用紙にご記入の上、同封の返信用封筒にて３月１５日頃までにご返送頂ければまことに幸いに存じます。

日夜医療に携わる皆様のご健康を祈っております。今後ともご指導くださいますようお願い申しあげます。

敬具

平成１８年３月

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高齢者の排泄ケアに関するアンケート

＊四角い枠の中の解答欄の文字を丸で囲むか、必要事項のご記入をお願い致しします。

記入者（無記名）

性別 [男　女]

年齢 [10歳代　20歳代　30歳代　40歳代　50歳代　60歳代以上]

I, 勤務する施設の種類

a	訪問看護ステーション
b	介護老人保健施設
c	特別養護老人ホーム
d	在宅介護支援センター
e	デイセンター
f	ケアハウス

II, 勤務する施設でケア中の高齢者の人数

概数を記入してください [　　人]

III, ケアにあたる職員の人数

概数を記入してください [　　人]

IV, ケアにおける排便・排尿管理について

質問 IV-1　ケアを受けている方のうちで排泄の介護が必要な方の割合はおおよそどれくらいですか？

a	20%以下
b	20%～40%
c	40%～60%
d	60%～80%
e	80%以上

質問 IV-2　患者さんにとって排泄のコントロールが生活の質に及ぼす影響はどの程度と感じますか？

a	極めて大きい
b	大きい
c	小さい
d	極めて小さい
e	わからない

質問 IV-3　排泄のコントロールが患者さんの人間としての尊厳にどの程度重要と感じますか？

a	極めて重要
b	重要
c	あまり重要ではない
d	全く重要ではない
e	わからない

質問 IV-4　介護する側として排泄の介護はどの程度負担に感じていますか？

a	極めて大きい
b	大きい
c	小さくはないが大きな負担とも思わない
d	小さい
e	負担ではない

質問 IV-5　排便の介護のうちで最も負担に感ずるのはどれですか？（複数回答可）

a,	オムツ交換	e,	環境の清掃
b、	清拭,洗浄	f,	処置後の汚物処理
c、	摘便	g,	患者さんへの気遣い
d、	浣腸	h,	その他（　　）

質問 IV-6　介護する側から排便の介護上支障となるものは？　（複数回答可）

a, 人手不足	e, 介護への無理解など人的事項
b, 時間的制約	f, その他(　　)
c, 設備的制約	
d, 衛生用品の不足	

質問 IV-7　要介護者の具体的排泄の場所や時間について（複数選択可）

1）患者さんが尿意・便意を伝えられる、または周囲が察知できる場合

1)-1 排便介助を行う場所はどこですか？

a	ベッド（ふとん）上オムツ
b	ベッド（ふとん）上オマル
c	部屋の中のポータブルトイレ
d	普通トイレで介助

1)-2 排便介助の時期はどうしていますか？

a	患者さんが便意を訴えたとき
b	便意にかかわらず定時に行う
c	自分の手が空いたとき
d	自分では行わない

2）患者さんが尿意・便意を伝えられない場合

2)-1 排便介助を行う場所はどこですか？

a	ベッド（ふとん）上オムツ
b	ベッド（ふとん）上オマル
c	ポータブルトイレ
d	普通トイレで介助

質問 2)-2 排便介助の時期はどうしていますか？

a	患者さんが便意を訴えたとき
b	便意にかかわらず定時に行う
c	自分の手が空いたとき
d	自分では行わない

V,　排泄のケアに関して

どのような情報が必要でしょうか？（複数回答可）

a	介護用具や設備の情報
b	排泄の生理など医学的知識
c	排泄介護の実務的知識
d	排泄介護の研修会などの情報
e	他の施設での対応法や問題などの情報
f	その他（　　）

VI, 排泄ケアにおける器具や設備で、「こんなものがあればよいのに‥‥」と普段お考えものがあればどんなものでも結構ですので書いてください。

V, 在宅ケアにかかわっている方への質問

質問 V-1　普段の（訪問以外の時間帯）のケアの主な担い手は誰ですか？多い順に3つ選んでください。

a、ヘルパーさん
b、(ケアを受ける方の)実の娘
c、(ケアを受ける方の)実の息子
d、(ケアを受ける方の)孫
e、(ケアを受ける方の)夫
f、(ケアを受ける方の)妻
g、お嫁さん
h、おムコさん
i、ご近所の方
j, その他

1番目	(	)
2番目	(	)
3番目	(	)

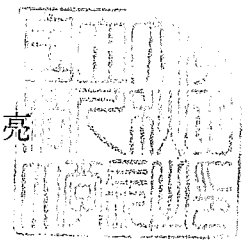
以上、ご協力ありがとうございました。

資料７

平成１７年８月１８日

萱場　広之　　殿

秋田大学長
三浦　亮



特許を受ける権利について（通知）

平成１７年８月５日付けで届出のありました発明について，下記のとおり決定しましたのでお知らせします。

記

発明の名称　　肛門機能障害者用自己洗腸療法補助具

決定事項　　承継する（本学に帰属する）

決定年月日　　平成１７年８月１８日