資料3

Evaluation of Anorectal Functions of Children With Anorectal Malformations Using Fecoflowmetry

By Hiroyuki Kayaba, Tatsuzo Hebiguchi, Hiroaki Yoshino, Masaru Mizuno, Mamiko Yamada, Junichi Chihara, and Tetsuo Kato

Akita, Japan

Background/Purpose: A simple and objective method evaluating the bowel functions of patients with anorectal anomalies is necessary. The authors applied fecoflowmetry and saline enama test for patients with anorectal anomalies to evaluate the fecodynamics and anorectal motilities in these children.

Methods: The bowel functions of 16 patients who underwent repair for anorectal malformations and 5 normal controls were evaluated by saline enema test and fecoflowmetry. The correlations between the clinical scores for the bowel functions and the parameters in fecodynamic studies were investigated.

Results: Seven of 16 patients exhibited periodical contractions of the rectum synchronized with relaxations of the anal canal during saline infusion as did the controls and had significantly higher clinical scores than the other patients. Two patients with severe chronic constipation lacked rectal contractions. Among the fecoflowmetric parameters, the maximum flow, average flow, and tolerable volume of saline infused into the rectum were significantly lower in the patients with low clinical scores than those of the controls. The maximal squeeze pressure and resting anal pressure were not significantly different between the patients and controls.

Conclusion: Fecodynamic studies, such as fecoflowmetry and saline enema test, help in obtaining clinical indicators for the bowel functions of patients with anorectal anomalies.

INDEX WORDS: Anorectal anomalies, bowel function, fecoflowmetry.

THE BOWEL FUNCTION after the repair of anorectal malformations generally is assessed by multivariate scoring method, because conventional anorectal manometry often fails to correlate with symptoms in the cases of altered anatomic structures in the anorectum. In these patients, not only anal pressure and rectoanal reflex, but also the ability to defecate should be evaluated. Incontinence and constipation are the major problems in patients after the repair of anorectal anomalies. Fecoflowmetry and saline enema tests provide us with objective data for continence, anorectal motilities, and the dynamics of defecation. The purpose of this study is to evaluate anorectal motility and the dynamics of defecation of the children treated for anorectal malformations.

MATERIALS AND METHODS

Patients

The bowel function of 16 patients (11 boys, 5 girls) who had undergone surgical repair for anorectal malformations and 5 normal controls (3 boys, 2 girls) was evaluated by fecodynamic study and anorectal manometry. The mean ages of the patients and normal controls at the time of evaluation were 8.8 ± 3.2 years and 11.0 ± 2.6 years, respectively. The types of anomalies of the patients were rectourethral fistula (RUF; 7 patients), rectocloacal fistula (RCF; 4 patients), rectobulbar fistula (RBF; 2 patients), anovestibular fistula (AVF; 2 patients), and anocutaneous fistula (ACF; 1 patient). Patient data are summarized in Table 1. Two patients with RUF and 1 patient with RBF were evaluated from 1 year to 5 years after the initial evaluation.

Saline Enema Test and Fecoflowmetric Study

Saline enema test and fecoflowmetric studies were performed in the manner previously described.[1] In brief, after emptying the rectum by glycerine enema, patients lay on the left side. One open-tipped probe was positioned in the rectum and another in the anal canal. Each probe was coupled to a pressure trasducer (DTS DX-360; Nihon Kohden, Tokyo, Japan). The transducer was connected via an amplifier (AD100F; Nihon Kohden) to a chart recorder (RTA-1100M; Nihon Kohden). The resting pressure of the anal canal was measured by pull-through method. Then, the patient was instructed to squeeze maximally to be recorded the maximal squeezing pressure by the probe positioned at the site of the maximal resting pressure of the anal canal. Next, 500 mL of saline was infused into the rectum at a rate of 50 mL/min, and the pressure of the rectum and anal canal were recorded simultaneously. Saline volume required for inducing anal relaxations and rectal contractions were recorded. The volume tolerated also was checked on the chart. Then the patient was 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 with a uro-flow meter (UROFLO-MET SUF200; Sakura Tokyo, Japan or Menuet Compact; Dantec Medical A/S, Denmark)

From the Central Clinical Laboratory and Department of Pediatric Surgery, Akita University Hospital, Akita, Japan.

This study was supported by Toyokichi Maeta memorial grant for gastroenterology.

Address reprint requests to Hiroyuki Kayaba MD, Central Clinical Laboratory, Akita University School of Medicine, 1-1-1, Hondo, Akita, Japan.

0022-3468/02/3704-0013$35.00/0

doi:10.1053/jpsu.2002.31622

Table 1. Clinical Charecteristics of Patients

Patient No.	Age (yr)	Gender	Type of Anomaly	Surgical Procedures
1	6	M	ACF*	CB
2	7	F	AVF	AT
3	13	F	AVF	AT
4	13	M	RBF	SPAP
5	13	M	RBF	SPAP
6	12	F	RCF	ASPAP
7	7	F	RCF	ASPAP
8	13	F	RCF	ASPAP
9	11	M	RCF†	ASPAP
10	5	M	RUF	SPAP
11	5	M	RUF	SPAP
12	9	M	RUF	SPAP
13	7	M	RUF	ASPAP
14	8	M	RUF	SPAP
15	5	M	RUF	SPAP
16	6	M	RUF	SPAP

NOTE. Patients 5, 14, and 16 were re-evaluated from 1 to 5 years after the initial evaluation.

Abbreviations: CB, Cut back procedure; AT, Anal transplantation; SPAP, Sacro-perineal anoplasty; ASPAP, Abdomino-sacro-perineal anoplasty.

*Anocutaneous fistula accompanied by Currarino triad.

†RCF accompanied by extrophy of the urinary bladder.

which consisted of an electric platform scale and a chart recorder to record a saline evacuation curve. A total amount of saline evacuated (milliliters), a maximum flow rate (milliliters per second), a flow time (seconds), a total evacuation time (flow time plus intermissions), an average flow (total amount of saline evacuated per flow time) were calculated. Clinical bowel function was converted into a score ranging from 0 to 8 using a questionnaire. The factors of clinical bowel function included in the questionnaire are summarized in Table 2.

Statistical Analysis

All measured values were presented as means ± SD. For analysis of significance of difference, Student's t test was used for differences between 2 groups. In comparisons of 3 or more groups, Fisher's PLSD (Protected Least Significant Difference) method was used as a post hoc test, regarding P less than .05 as significant after analysis of variance.

RESULTS

As we reported in a previous study on adult patients with problems in defecation,[1] the pressure fluctuations in the rectum and anal canal were classified into several types (Fig 1). Four of 7 patients with RUF and 2 patients with AVF showed periodical contractions of anal canal accompanied by rectal contractions, that is, type I pressure fluctuations. In 6 patients, the anal canal relaxed soon after the saline infusion, and the anal canal pressure remained at the same level as the rectal pressure, that is, type II pressure fluctuations. Incontinence was the major problem in 3 of 6 patients with type II pressure fluctuations. Two patients with rectourethral fistula who suffered from severe constipation lacked rectal contractions even in the presence of periodical relaxations of the anal canal (type III pressure fluctuations). In 2 patients, relaxations of the anal canal and increases in rectal pressure occurred irregularly and independently of each other (irregular type Table 3). The patients with type I pressure fluctuations had significantly higher clinical scores (5.3 ± 1.1) than that of the patients with other types of pressure fluctuations (3.4 ± 1.9). Fecoflowmetry curves were recorded in all the patients but patient 3 who refused to evacuate saline on the flowmeter. Fecoflowmetric curves were classified into 3 types as reported in a previous study on patients without anorectal malformation (Fig 2).[1] In type A, fecoflowmetric curve showed mountain peaks, which may indicate that evacuation was accomplished smoothly with high peak flow and a short evacuation time. In type B, the evacuation curve showed segmental peaks. Evacuation may have required some effort, and voiding time was longer than that of type A. In type C, fecoflowmetric curve was flat and had no apparent peak. The peak flow was less than 20 mL/sec. All of the 4 patients with middle or low anomaly were classified into type A. Only 4 of the 10 patients with high anomaly were classified into type A (Table 3). Patients with type A fecoflowmetric curve had significantly higher clinical scores (4.0 ± 1.0) for the bowel function than that (3.3 ± 1.8) of patients with other type of fecoflowmetric curves. Patients with clinical scores 5 or lower had significantly smaller tolerable volume (214.4 ± 175.6 mL) and the lower maximum flow rate (27.3 ± 27.1 mL/sec) compared with those of controls

Table 2. Clinical Bowel Function Scoring for Children With Anorectal Anomalies

Symptoms	Scores
Feels/reports the urge to defecate	
(a) Absent	0
(b) Not always	1
(c) Always	2
Constipation*	
(a) Unmanageable without bowel irrigation or manual extraction of rectal content	1
(b) Manageable with daily enemas or suppositorium	2
(c) Better than (b) and worse than (d)	3
(d) No constipation	4
Incontinence*	
(a) Daily	0
(b) More than twice a week	1
(c) Better than (b) and worse than (d)	2
(d) Only with diarrheal stool	3
(e) Never	4
Soiling	
(a) Daily	0
(b) Better than (a) and worse than (c)	1
(c) Never	2

*If one has score 4 and 2 in the categories of constipation and incontinence, respectively, the lower point (2) is added to the score. The perfect score is 8.

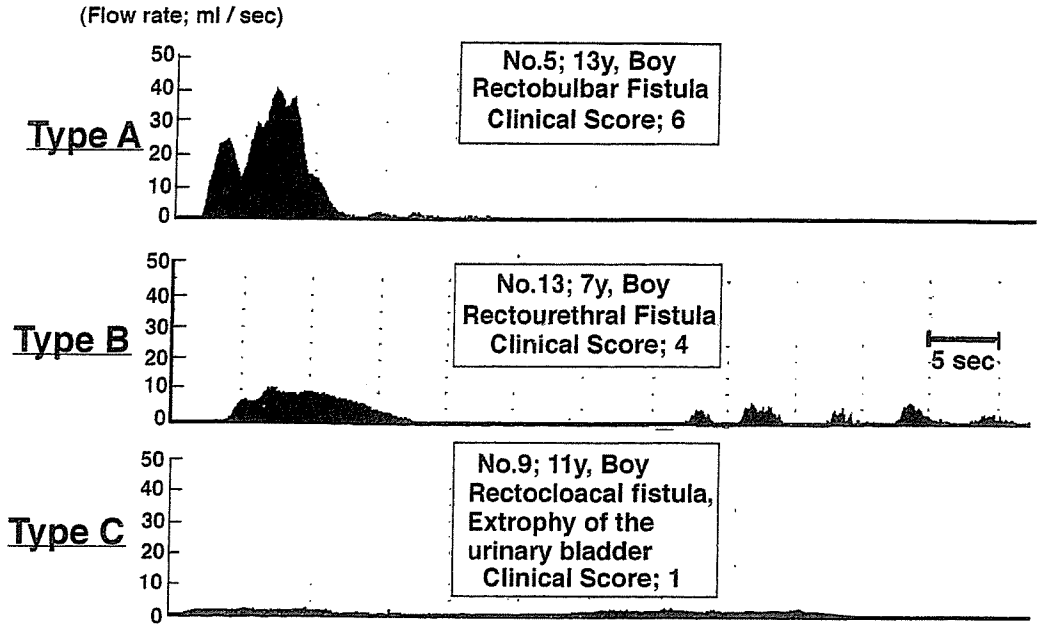

Fig 2. Classification of fecoflowmetric curve. The fecoflowmetric curves in patients 5, 14, and 9, representing type A, B, and C, respectively, are presented.

daily life. The evaluation of bowel functions of these children by conventional manometric studies such as resting and squeezing anal pressures often fail to reflect the actual bowel functions and should not simply be compared with the data of normal controls, because the children with anorectal malformations have altered anatomic structures of the anorectum.

Multivariate scoring methods have been utilized[2-4] for the evaluation of the ability to defecate in the children with anorectal malformations, because they are not invasive and need no special equipment. Fecoflowmetry,[1,5,6] defecography,[7] and scintigraphic defecography[8] have been reported to be suitable methods for quantitative and dynamic assessment of the ability to defecate. Defecography may be useful in the evaluation of morphologic abnormality as well as functional abnormality of the rectum, anus, and pelvic floor. Defecography requires, however, special equipment and has a restriction in applying to children because of its radiologic invasiveness. The bowel functions of children after repair of anorectal anomalies may be affected by the function of preserved or reconstructed anatomic structures including rectum, anus, the related muscles, and nerves. The periodical relaxations of the anal canal synchronized with contractions of the rectum are introduced in the majority of normal subjects.[1,9] In the current

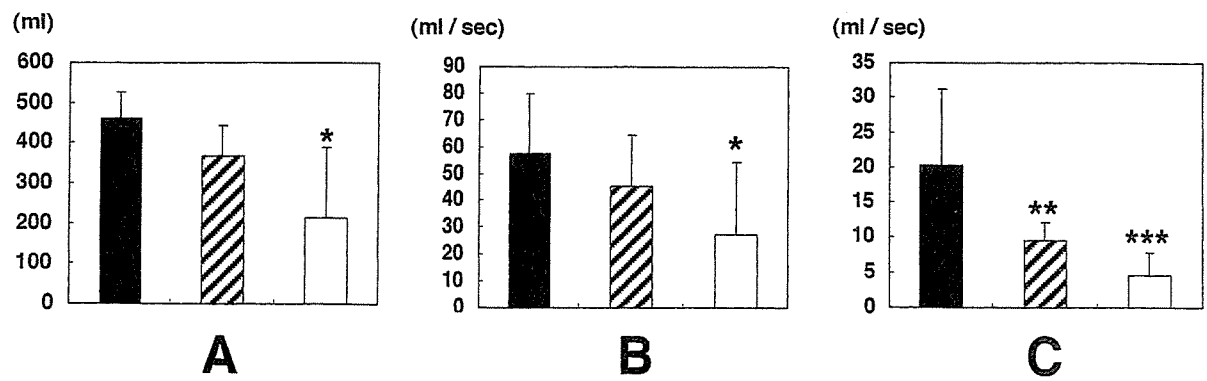

Fig 3. Results of fecoflowmetric study. (A) Comparison of the tolerable volume between controls (black bar) and patients with high (6-8) clinical score (hatched bar) or low (0-5) clinical score (white bar). The clinical score was 8 in all the controls. Results are expressed as mean values ± SD. The tolerable volume was significantly smaller in patients with low clinical score (versus that of controls). The mean tolerable volume of the patients with high clinical score was smaller than that of controls; however, there was no statistical difference. *$P < .05$. (B) Comparison of maximum flow. The maximum flow for patients with low clinical score (white bar) was significantly lower than that of controls (black bar). *$P < .05$. (C) Comparison of average flow. Both patients with low clinical score (white bar) and those with high clinical score (hatched bar) had significantly lower average flow than the controls (black bar). **$P < .01$; ***$P < .001$.

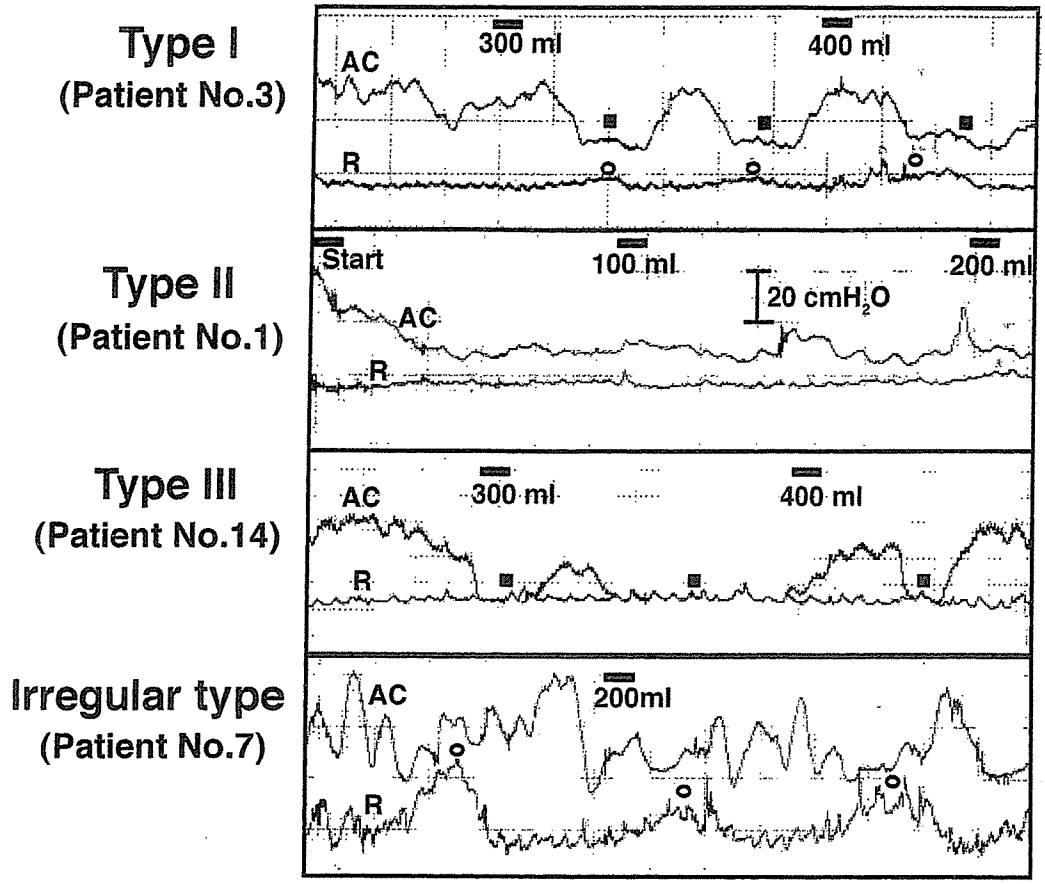

Fig 1. Classification of pressure fluctuations of the rectum and anal canal. The pressure fluctuations of the rectum and anal canal during saline infusion in the rectum in patients 3, 1, 14, and 7, representing type I, II, III, and irregular-type pressure fluctuations, respectively, were presented. In type I, the anal canal relaxes periodically (■), which is synchronous with rectal contractions (O). Type I is usually seen in normal subjects. In type II, which is popular in the patients with incontinence, the anal canal relaxes soon after the saline infusion without recovery thereafter. Rectal contractions are lacking in type III pressure fluctuations. Patients with type III pressure fluctuations tend to have longstanding severe constipation.

(462.5 ± 65.0 mL and 57.4 ± 22.6 mL/sec, respectively). The average flow rate of the patients with low (0 to 5) clinical scores (4.6 ± 3.2 mL/sec) was significantly lower than that of the patients with clinical scores 6 or higher (9.6 ± 2.6 mL/sec) and that of controls (20.4 ± 10.9 mL/sec, Fig 3). Resting anal pressure and maximal squeezing pressure failed to show statistical difference between the patients and controls. In 3 patients (patients 4, 14, and 16), follow-up studies were carried out after 1 to 5 years after initial evaluation. At the follow-up studies, all of them showed greater clinical scores, the maximum and average flow rate, and required shorter voiding time than those observed at the initial evaluations (Fig 4).

DISCUSSION

The children who underwent repair for anorectal malformations require long-term medical care and supportive treatment to achieve satisfactory bowel functions in

Table 3. Distribution of the Types of Pressure Fluctuation Curves and Fecoflowmetric Curves

Pressure Fluctuation Curve					Fecoflowmetric Curve		
Type I	Type II	Type III	Irregular	Type of Anomaly	Type A	Type B	Type C
	●●●		●	RCF	●●		●●
●●●●	●	●●		RUF	●●	●●●●●	
	●●			RBF	●●		
●●				AVF	●		
	●			ACF	●		
●●●●●				Controls	●●●●●		

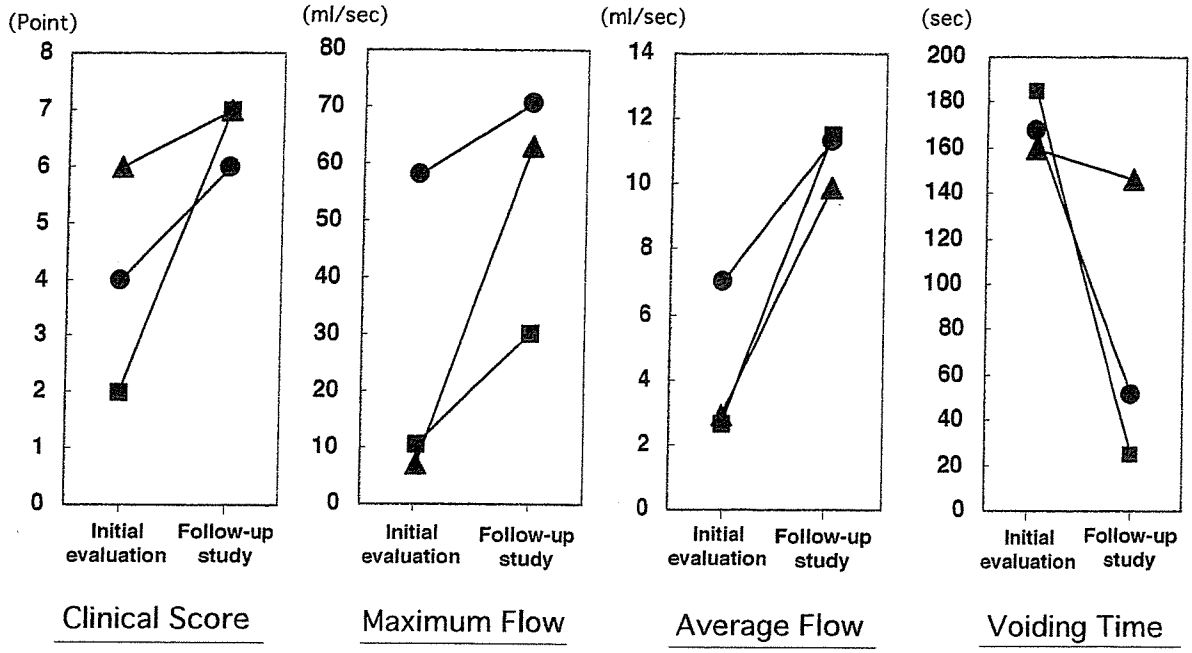

Fig 4. Results of follow-up studies for 3 patients with intermediate- or high-type anorectal malformations. The clinical scores and results of fecoflowmetric studies are shown for patients 4 (▲), 14 (■), and 16 (●). All the 3 patients exhibited improvement in clinical score and fecoflowmetric parameters 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.

REFERENCES

1. Kayaba H, Kodama K, Shirayama K et al: Evaluation of ability to defecate using saline evacuation from the rectum. Dis Colon Rectum 40:s96-s98, 1997 (suppl)
2. Rintala RJ, Lindahl H: Is normal bowel function possible after repair of intermediate and high anorectal malformations? J Pediatr Surg 30:491-494, 1995
3. Kelly JH: The clinical and radiological assessment of anal continence in childhood. Aust NZ J Surg 42:62-63, 1972
4. Katsumata K: A proposal for the clinical evaluation of the bowel functions after the repair of anorectal malformations. (in Japanese) J Jpn Soc Pedatr Surg 18:1458-1459, 1982
5. Shafik A, Khalid A: Fecoflowmetry in defecation disorders. Pract Gastroenterol 14:46-52, 1990
6. Shafik A, Abdel-Moneim K: Fecoflowmetry: A new parameter assessing rectal function in normal and constipated subjects. Dis Colon Rectum 36:35-42, 1993
7. Ekberg O, Nylander G, Fork FT: Defecography. Radiology 155: 45-48, 1985
8. Hutchinson R, Mostafa AB, Grant EA, et al: Scintigraphic defecography: Quantitative and dynamic assessment of anorectal function. Dis Colon Rectum 36:1132-1138, 1993
9. Read NW, Haynes DC, Bartolo DCC, et al: Use of anorectal manometry during rectal infusion of saline to investigate sphincter function in incontinent patients. Gastroenterology 85:105-113, 1983
10. Yuan Z, Bai Y, Zhang Z, et al: Neural electrophysiological studies on the external anal sphincter in children with anorectal malformation. J Pediatr Surg 35:1052-1057, 2000
11. Martins JL, Lederman HM, Pinu J: Clinical and radiological postoperative evaluation of posterior sagital anorectoplasty in patients with upper and intermediate anorectal malformations. Rev Paul Med 115:1566-1569, 1997
12. Shafik A: Electrorectography in chronic constipation. World J Surg 19:772-775, 1995
13. White JC, Verlot M, Ehrentheil O: Neurogenic disturbances of the colon and their investigation by the colonometrogram. Ann Surg 112:1042-1057, 1949
14. Loening-Baucke V: Constipation in children. Curr Opin Pediatr 6:556-561, 1994
15. Shafik A: Constipation. Pathogenesis and management. Drugs 45:528-540, 1993
16. Meier-Ruge WA, Holscneider AM: Histopathologic observations of anorectal abnormalities in anal atresia. Pediatr Surg Int 16:2-7, 2000
17. Shoulder P, Keighley MRB: Changes in colorectal function in severe idiopathic chronic constipation. Gastroenterology 90:414-420, 1985
18. Mertz H, Naliboff B, Mayer E: Physiology of refractory chronic constipation. Am J Gastroenterol 94:609-615, 1999
19. Wexner SD, Marchetti F, Salanga V, et al: Neurophysiologic assessment of the anal sphincters. Dis Colon Rectum 34:606-612, 1991
20. Yagi M, Iwafuchi M, Uchiyama M, et al: Postoperative fecoflowmetric analysis in patients with anorectal malformation. Surg Today 31:300-307, 2001

Pediatr Surg Int (2003) 19: 251–255
DOI 10.1007/s00383-002-0844-x

資料4

ORIGINAL ARTICLE

Hiroyuki Kayaba · Tatsuzo Hebiguchi · Hiroaki Yoshino
Masaru Mizuno · Norihiro Saitoh · Yoshimi Kobayashi
Tetsuya Adachi · Junichi Chihara · Tetsuo Kato

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

Accepted: 19 February 2002 / Published online: 24 April 2003

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

H. Kayaba · N. Saitoh · Y. Kobayashi · T. Adachi · J. Chihara
Central Clinical Laboratory,
Akita University School of Medicine, Akita, Japan

T. Hebiguchi · H. Yoshino · M. Mizuno · T. Kato
Department of Pediatric Surgery,
Akita University School of Medicine, Akita, Japan

H. Kayaba (✉)
Central Clinical Laboratory,
Akita University School of Medicine,
1-1-1, Hondo, Akita, Japan
E-mail: kayaba@hos.akita-u.ac.jp
Tel.: +81-18-884-6181
Fax: +81-18-884-6181

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

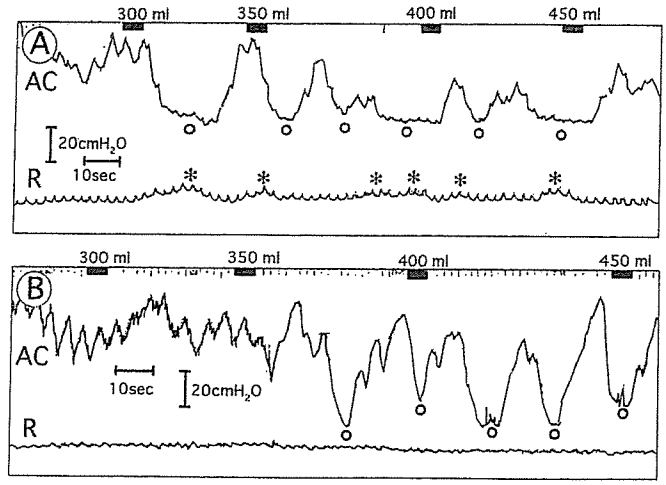

Fig. 1 Pressure fluctuations in rectum and anal canal in two patients: **A** Patient 2 shows periodic relaxations of anal canal (O) synchronized with increase in rectal pressure (*) after infusioin of 300 ml saline. **B** Patient 9 shows no contractions of rectum during saline infusion and only periodic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

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).

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.

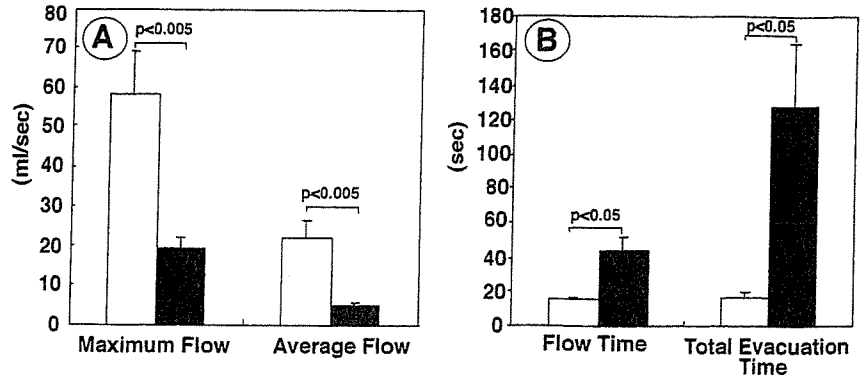

Fig. 2 Fecoflowmetric parameters of controls and patients with idiopathic chronic constipation. **A** Maximum and average flow rates of patients (*closed bars*) significantly lower ($P < 0.005$) than those of controls (*open bars*) **B** Flow time and total evacuation time of patients significantly longer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

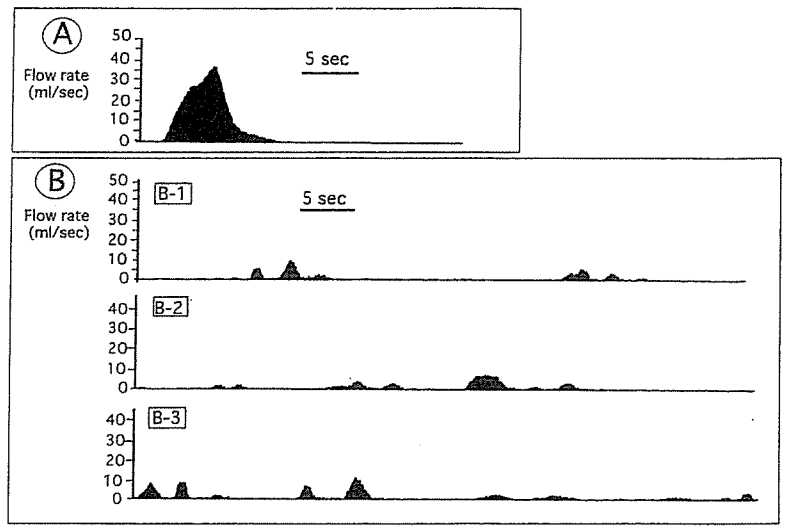

Fig. 3 Fecoflowmetric curves obtained from a control and a patient. **A** All children without defecation problems showed massive fecoflowmetric curves with high flow rate and short evacuation time. **B** Fecoflowmetric curve from patient 9: segmental and divided into small humps, reflecting poor ability 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.

Acknowledgements This study was supported by a Toyokichi Maeta memorial grant for gastroenterology.

References

1. Kayaba H, Kodama K, Shirayama K, Kodama M (1997) Evaluation of ability to defecate using saline evacuation from the rectum. Dis Colon Rectum 40[Suppl]: 96–98
2. Loening-Baucke V (1994) Constipation in children. Curr Opin Pediatr 6: 556–561
3. Rao SSC, Welcher KD, Leistikow JS (1998) Obstructive defecation: a failure of rectoanal coordination. Am J Gastroenterol 93: 1042–1050
4. Bassotti G, Chiarioni G, Vantini I, Betti C, Fusaro C, Pelli MA, Morelli A (1994) Anorectal manometric colonic propulsive impairment in patients with severe chronic idiopathic constipation. Dig Dis Sci 39: 1558–1564
5. Shoulder P, Keighley MRB (1985) Changes in colorectal function in severe idiopathic chronic constipation. Gastroenterology 90: 414–420
6. Mertz H, Nakibofff B, Mayer E (1999) Physiology of refractory chronic constipation. Am J Gastroenterol 94: 609–615
7. Loening-Baucke V (1984) Abnormal rectoanal function in children recovered from chronic constipation and encopresis. Gastroenterology 87: 1299–1304
8. Wexner SD, Marchetti F, Salanga V, Corredor C, Jagelman DG (1991) Neurophysiologic assessment of the anal sphincters. Dis Colon Rectum 34: 606–612
9. Krishnamurthy S, Shuffler MD, Rohrmann CA, Pope CE (1985) Severe idiopathic constipation is associated with a distinctive abnormality of the colonic myenteric plexus. Gastroenterology 88: 26–34
10. Corazziari E, Cucchiara S, Staiano A, Romaniello G, Tamburrini O, Torsoli A, Auricchio S (1985) Gastrointestinal transit time, frequency of defecation, and anorectal manometry in healthy and constipated children. J Pediatr 106: 379–382
11. Borowitz SM, Sutphen J, Ling W, Cox D (1996) Lack of correlation of anorectal manometry with symptoms of chronic childhood constipation and encopresis. Dis Colon Rectum 39: 400–405
12. Loening-Baucke V (1983) Anorectal manometry: experience with strain gauge pressure transducers for the diagnosis of Hirshsprung's disease. J Pediatr Surg 18: 595–600
13. Shafik A, Khalid A (1990) Fecoflowmetry in defecation disorders. Pract Gastroenterol 14: 46–52
14. Shafik A, Abdel-Moneim K (1993) Fecoflowmetry: a new parameter assessing rectal function in normal and constipated subjects. Dis Colon Rectum 36: 35–42
15. Hutchinson R, Mostafa AB, Grant EA, Smith NB, Deen KI, Harding LK, Kumar D (1993) Scintigraphic defecography: quantitative and dynamic assessment of anorectal function. Dis Colon Rectum 36: 1132–1138
16. Read NW, Haynes DC, Bartolo DCC, et al (1983) Use of anorectal manometry during rectal infusion of saline to investigate sphincter function in incontinent patients. Gastroenterology 85: 105–113
17. Shafik A (1995) Electrorectography in chronic constipation. World J Surg 19: 772–775
18. White JC, Verlot M, Ehrentheil O (1949) Neurogenic disturbances of the colon and their investigation by the colonometrogram. Ann Surg 112: 1042–1057
19. Shafik A (1993) Constipation: pathogenesis and management. Drugs 45: 528–540
20. Shafik A, El-Sibai O, Shafik AA (2000) Rectal pacing in patients with constipation due to rectal inertia: technique and results. Int J Colorectal Dis 15: 100–104

資料5

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

by

Hiroyuki Kayaba, M.D., Tatsuzo Hebiguchi, M.D., Yasunobu Itoh, M.D., Ph.D., Hiroaki Yoshino, M.D., Masaru Mizuno, M.D., Ph.D., Mayako Morii, M.D., Tetsuya Adachi, M.D., Junichi Chihara, M.D., Ph.D., and Tetsuo Kato, M.D., Ph.D.

Reprinted From: *The Journal of Neurosurgery*
Volume 98, April, 2003

J Neurosurg (Spine 3) 98:251–257, 2003

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

HIROYUKI KAYABA, M.D., TATSUZO HEBIGUCHI, M.D., YASUNOBU ITOH, M.D., PH.D., HIROAKI YOSHINO, M.D., MASARU MIZUNO, M.D., PH.D., MAYAKO MORII, M.D., TETSUYA ADACHI, M.D., JUNICHI CHIHARA, M.D., PH.D., AND TETSUO KATO, M.D., PH.D.

Central Clinical Laboratory, Department of Pediatric Surgery and Neurosurgery, Akita University Hospital, Akita, Japan

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

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.

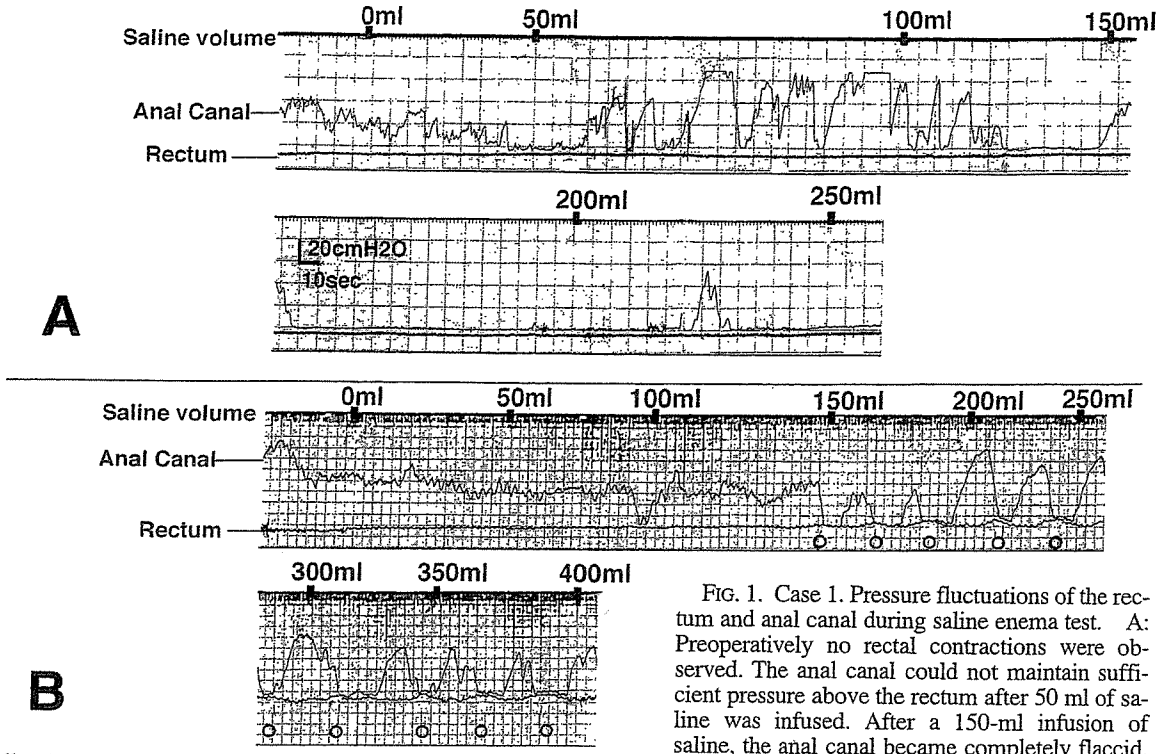

FIG. 1. Case 1. Pressure fluctuations of the rectum and anal canal during saline enema test. A: Preoperatively no rectal contractions were observed. The anal canal could not maintain sufficient pressure above the rectum after 50 ml of saline was infused. After a 150-ml infusion of saline, the anal canal 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

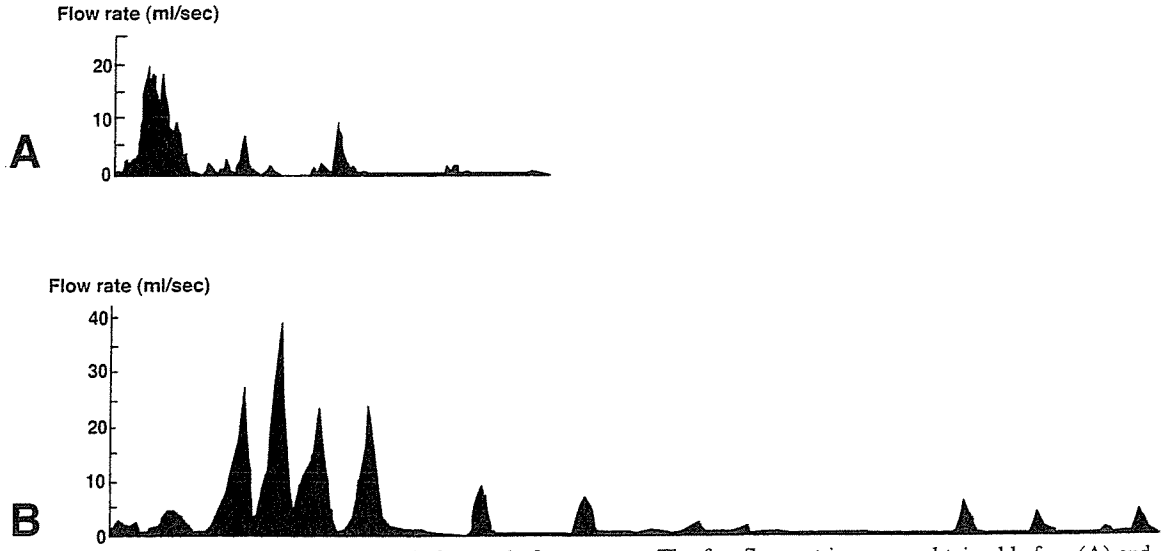

FIG. 2. Case 1. Fecoflowmetric curves before and after surgery. The fecoflowmetric curves obtained before (A) and after (B) untethering surgery were both classified as "segmental," which is usually seen in patients with chronic constipation. The maximum flow rate reached 39.3 ml/second, and 471 of 500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

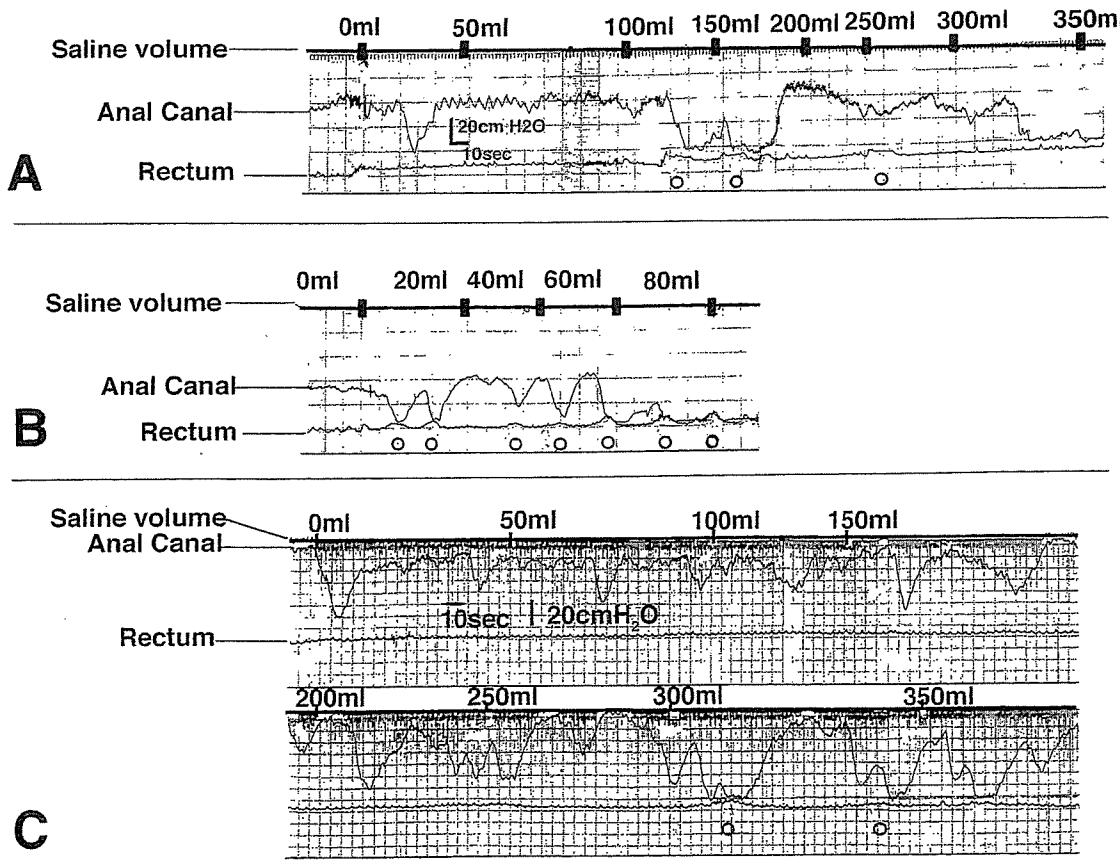

FIG. 3. Case 5. Pressure fluctuations of the rectum and anal canal during saline enema test. At 9 years of age the patient could tolerate a 150-ml saline injection. A: A series of rectal contractions *(open circles)* accompanying relaxations of the anal canal were induced after a 120-ml saline infusion. 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.

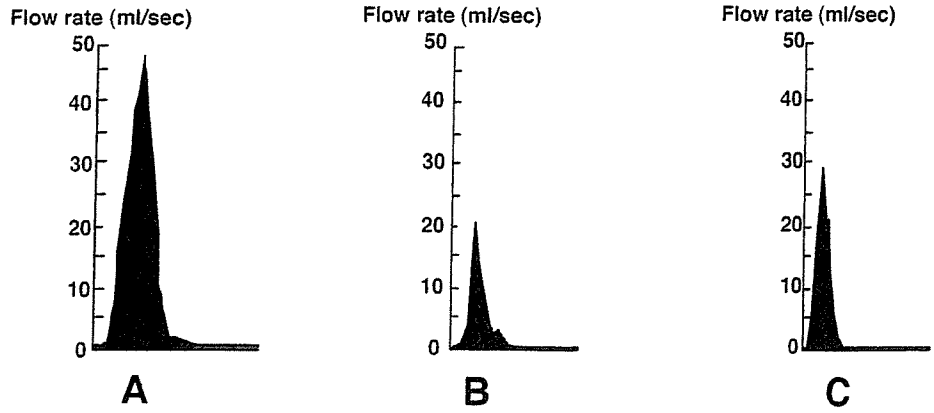

FIG. 4. Case 5. Fecoflowmetric curves before and after surgery. Fecoflowmetric curves recorded at ages 9 years (A), 13 years (B) and 15 years (C). Deterioration of control of bowel movement was obvious by age 13 years (B). After untethering surgery improvement was shown (C), even though it was not satisfactory.

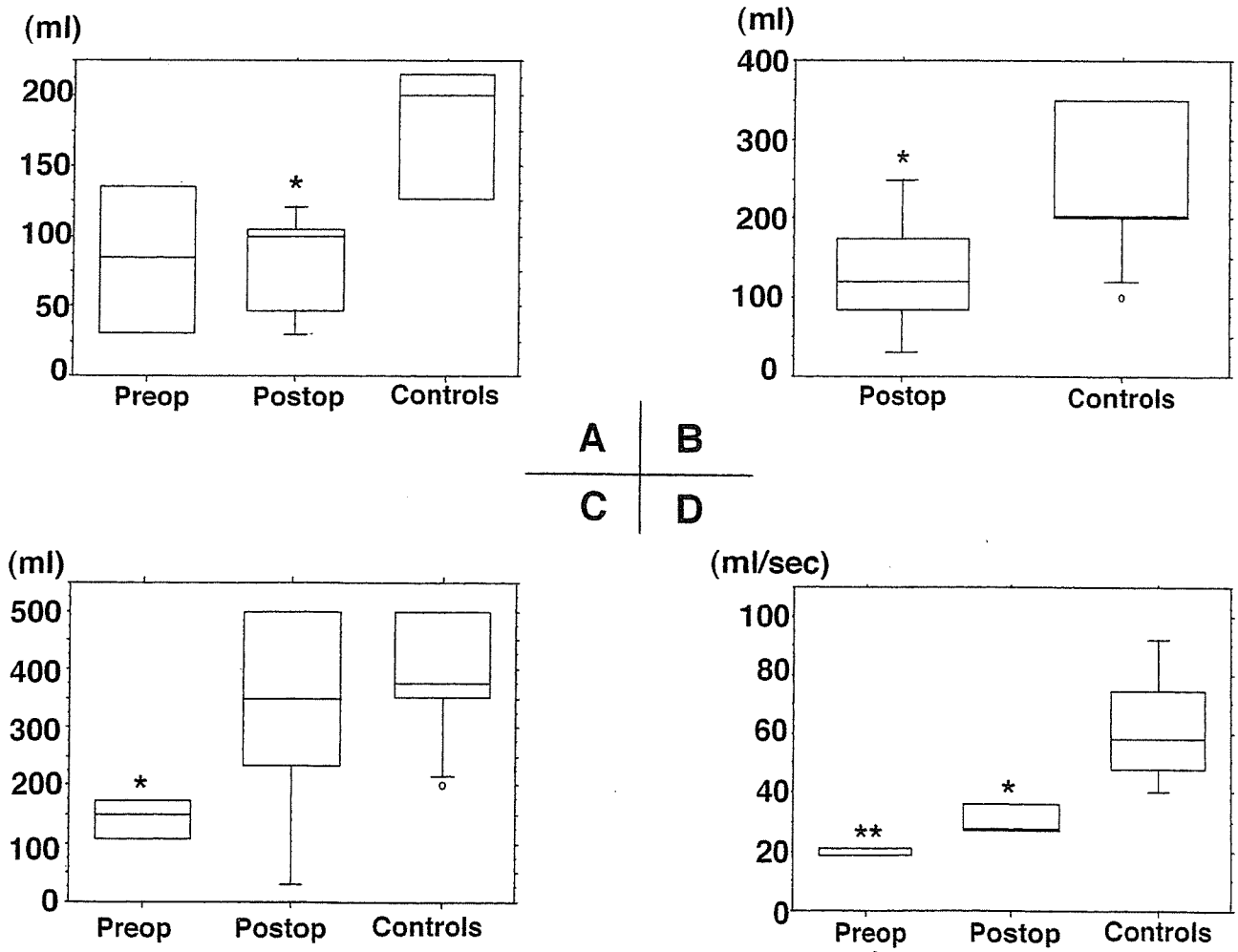

FIG. 5. Comparison of saline enema test and fecoflowmetry parameters. The parameters in the saline enema test and fecoflowmetry are shown in box plots. Horizontal bars represent the 10th, 25th, 50th, 75th, and 90th percentiles from bottom to top in that order. The saline volume required to induce anal 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). *$p < 0.05$, **$p < 0.01$.

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.

References

1. Anderson FM: Occult spinal dysraphism. Diagnosis and management. **J Pediatr 73:**163–177, 1968
2. Begeer JH, Meihuizen de Regt MJ, HogenEsch I, et al: Progressive neurological deficit in children with spina bifida aperta. **Z Kinderchir 41:**S13–S15, 1986

3. Bergman SB: Bowel management, in Nesathurai S (ed): **The Rehabilitation of People With Spinal Cord Injury.** Malden, MA: Blackwell Science, 2000, pp 53–58
4. Cornette L, Verpoorten C, Lagae L, et al: Tethered cord syndrome in occult spinal dysraphism: timing and outcome at surgical release. **Neurology 50:**1761–1765, 1998
5. Fukui J, Kakizaki T: Urodyamic evaluation of tethered cord syndrome including tight filum terminale: prolonged follow-up observation after intraspinal operation. **Urology 16:**539–552, 1980
6. Fukui J, Kodaira K, Watanabe S, et al: [Diagnosis, treatment and prognosis of congenital neurogenic bladder caused by traction of lower spinal cord segment with spina bifida occulta. (so called "tethered cord syndrome").] **Nippon Hinyokika Gakkai Zasshi 68:**569–589, 1977 (Jpn)
7. Glickman S, Kamm MA: Bowel dysfunction in spinal-cord-injury patients. **Lancet 347:**1641–1653, 1996
8. Kayaba H, Hebiguchi T, Yoshino H, et al: Evaluation of anorectal functions in children with anorectal malformations using fecoflowmetry. **J Pediatr Surg 37:**623–628, 2002
9. Kayaba H, Hebiguchi T, Yoshino H, et al: Fecoflowmetric evaluation of anorectal functions and ability to defecate in children with idiopathic chronic constipation. **Pediatr Surg Intern 18:** (in press), 2003
10. Kayaba H, Kodama K, Shirayama K, et al: Evaluation of ability to defecate using saline evacuation from the rectum. **Dis Colon Rectum 40:**S96–S98, 1997
11. Khoury AE, Hendrick EB, McLorie GA, et al: Occult spinal dysraphism: clinical and urodynamic outcome after division of the filum terminale. **J Urol 144:**426–429, 443–444, 1990
12. Kiechl S, Kronenberg MF, Marosi M, et al: Tethered cord syndrome as cause of spinal cord dysfunction. **Lancet 348:** 342–343, 1996
13. Kondo A, Kato K, Kanai S, et al: Bladder dysfunction secondary to tethered cord syndrome in adults: is it curable? **J Urol 135:**313–316, 1986
14. Palmer LS, Richards I, Kaplan WE: Subclinical changes in bladder function in children presenting with nonurological symptoms of the tethered cord syndrome. **J Urol 159:**231–234, 1998
15. Petersen MC: Tethered cord syndrome in myelodysplasia: correlation between level of lesion and height at time of presentation. **Dev Med Child Neurol 34:**604–610, 1992
16. Stiens SA, Bergman SB, Goetz LL: Neurogenic bowel dysfunction after spinal cord injury: Clinical evaluation and rehabilitative management. **Arch Phys Med Rehabil 78:**S86–102, 1997
17. Stone JM, Nino-Murcia M, Wolfe VA, et al: Chronic gastrointestinal problems in spinal cord injury patients: a prospective analysis. **Am J Gastroenterol 85:**1114–1119, 1990
18. Tamaki N, Shirataki K, Kojima N, et al: Tethered cord syndrome of delayed onset following repair of myelomeningocele. **J Neurosurg 69:**393–398, 1988
19. White JC, Verlot MG, Ehretheil O: Neurogenic disturbances of the colon and their investigation by the colonometrogram. **Ann Surg 112:**1042–1057, 1940

Manuscript received June 17, 2002.
Accepted in final form December 13, 2002.
This study was supported by a Toyokichi Maeta memorial grant for gastroenterology.
Address reprint requests to: Hiroyuki Kayaba, M.D., Central Clinical Laboratory, Akita University Hospital, 1-1-1, Hondo, Akita 010-8543, Japan. email: kayaba@hos.akita-u.ac.jp.

−55：2・2007−

原　著

高齢者の排便障害の病態分析

萱場広之*1　伊藤亘*2　山口一考*3
千葉貴人*4　竹田正秀*5　茆原順一*6

Physiopathological Evaluation of Defecatory Problems in Elderly People using Saline Enema Test and Fecoflowmetry

Hiroyuki KAYABA, MD*1, *Wataru ITO*, PhD*2, *Kazutoshi YAMAGUCHI*, PhD*3, *Takahito CHIBA*, PhD*4, *Masahide TAKEDA**5 and *Junichi CHIHARA*, MD*6

The population of the aged people is increasing rapidly in developed countries. Bowel care is recognized as an important factor to the wellbeing of the disabled elderly people. To evaluate the physiopathology of defecatory problems in the elderly, we applied saline enema test and fecoflowmetry; that is, pressure fluctuations of the rectum and anal canal were simultaneously recorded during saline infusion in the rectum, and then the saline evacuation curve was recorded in the elderly subjects. The patterns of the pressure fluctuations in the rectum and anal canal were analyzed in saline enema test. In fecoflowmetry, the shape of the evacuation curve and several parameters such as, flow rate and evacuation time were evaluated. The saline volume required to elicit rectal contractions and relaxations of the anal canal were significantly decreased in the patients with the lesions narrowing the spinal canal. The shape of evacuation curve represented the state of defecation in each subject. Subjects without defecatory problems had high flow rates and short evacuation time, while subjects with incontinence and/or constipation had low flow rates and long evacuation time. Furthermore, big and slow periodic pressure fluctuations of the anal canal, so called ultra slow waves, were seen in some patients with severe chronic constipation accompanying megacolon. These findings suggested that the physiopathology of defecatory problems in the elderly is variable and complicated, and that the appropriate treatment for these patients is achieved through appropriate evaluation.

[Rinsho Byori **55**：105〜111, 2007]

*1Department of Clinical and Laboratory Medicine, Akita University School of Medicine, Akita 010-8543

【**Key Words**】elderly(高齢者)，constipation(便秘)，incontinence(失禁)，fecoflowmetry(フェコフローメトリー)，anorectal function(直腸肛門機能)

高齢者人口比率の上昇が続く今日において老人介護は重要な社会問題であるが，中でも排泄介護の負担は大きい。高齢者の排尿障害に関しては多くの医学的アプローチがなされ，その治療にも生かされている一方で，排便障害，すなわち便秘や便失禁については，加齢に伴う生理的な変化として一括され，医学的検討対象として十分に認知されてこなかった。我々は，深刻でありながらも検討が遅れている排便

受付2006年12月21日・受理2007年1月30日

*1〜6秋田大学医学部統合医学講座臨床検査医学分野(〒010-8543 秋田市本道1-1-1)

本論文は第52回日本臨床検査医学会総会における座長推薦論文である。