Sacral Nerve Function in Child Patients After Ileal J-Pouch-Anal Anastomosis for Ulcerative Colitis

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To clarify the neurological function of the puborectalis muscle (PM) in child patients with soiling after ileal J-pouch-anal anastomosis (IPAA) for ulcerative colitis (UC), we examined the terminal motor latency in the sacral nerves that regulate the PM. Eight patients after IPAA for UC were studied (6 males and 2 females aged 11 to 13 years with a mean age of 12.8 years). All patients 6 months after IPAA showed soiling (group A) and these patients showed continence at 2 years after IPAA (group B). Group C serving as controls consisted of 16 subjects (10 males and 6 females aged 12 to 17 years with a mean age of 14.4 years). Left- and right-sided sacral nerve terminal motor latency (SNTML) tests were performed at 6 months and 2 years after IPAA in order to measure the latency of the response in the bilateral PM following magnetic stimulation of sacral nerve root segments 2 to 4 (S2–S4) of the spinal column overlying the cauda equina. The following results were obtained. (1) Right-sided SNTML: group A exhibited significant prolongation compared with groups B and C ($P < 0.0001$ and $P < 0.0001$, respectively). There was no significant difference between groups B and C ($P = 0.2329$). (2) Left-sided SNTML: group A exhibited significant prolongation compared with groups B and C ($P = 0.0002$ and $P < 0.0001$, respectively). There was no significant difference between groups B and C ($P = 0.2315$). Note that significant differences were not established between SNTML values measured on the right and left sides. Soiling in child patients 6 months after IPAA may be caused by damage to the bilateral sacral nerves during the operation. However, the damage to the sacral motor nerve improves within 2 years after IPAA.

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The functional results of total colectomy, mucosal proctectomy, and ileal J-pouch-anal anastomosis (IPAA) for ulcerative colitis (UC) have been acceptable and patient satisfaction has been very high because patients are free from stoma.^{1–3} In general, the functional outcomes after IPAA in children are better than those in adults.^{4–12} However, some child patients experience defecation function impairments after IPAA, including minor fecal incontinence (soiling), incontinence, increased bowel movement, urgency of defecation, evacuation difficulty, irregular bowel habitus, difficulty in distinguishing gas from feces, and diarrhea.^{8–10,12} The most common abnormality is soiling.^{8–10} Soiling in both children and adults is usually more severe in the early postoperative period of about 3 to 6 months after operation and improves with time, but may become permanent more than 1 year after IPAA.^{3–6,9} Generally, the frequency of soiling in child patients more than 1 year after IPAA is 0 to 12%,^{4–6,8,9,12} and in adults 30 to 40%.^{2,13,14} Over the past 20 years or so, the function of the anorectum in patients with soiling after IPAA has been studied in depth using anorectal manometry.^{15–17} According to the data of anorectal manometry in child patients after IPAA, the anal sphincter complex comprising the internal anal sphincter and external anal sphincter is important to prevent soiling.¹⁷ In contrast, the puborectalis muscle (PM) was considered the most important factor preventing soiling in other studies not using anorectal manometry, which cannot detect the function of the PM, although less is known about the neurological functions in patients with soiling after IPAA in regard to the sacral nerve (SN) that regulates the PM.^{18–20} To the best of our knowledge, there are no reports of electrophysiological studies of SN in child patients with soiling after IPAA for UC. We therefore studied bilateral SN function using sacral nerve terminal motor latency (SNTML) in child patients with soiling 6 months after IPAA and the same patients without soiling 2 years after IPAA for UC.

Patients and Methods

From 1999 to 2008, 12 children with UC (8 males and 4 females aged 11 to 13 years with a mean age of 12.2 years) visited our hospital. Four children refused to

participate in this study. A total of 8 patients after IPAA for UC were electrophysiologically studied (6 males and 2 females aged 11 to 13 years with a mean age of 12.8 years). All patients 6 months after IPAA showed soiling [minor incontinence: the seepage of mucus and/or stool during nighttime and/or daytime, every day, with a pad needed to protect against soiling (group A)]. All the patients showed continence 2 years after IPAA (group B). Patients with external inguinal hernia from our clinics with good control of bowel function acted as controls (group C). Group C consisted of 16 subjects (10 males and 6 females aged 12 to 17 years with a mean age of 14.4 years). A total of 8 patients after IPAA in this study had an uneventful postoperative course without severe complications and pouchitis 2 years after IPAA. Examinations were performed 6 months and 2 years after IPAA. All patients tolerated a general diet without medication at the time of the study. No patients had other diseases (without surgical histories, psychiatric diseases, metabolic diseases, endocrinological disorders, functional digestive tract impairments, and organic digestive tract diseases) before undergoing the IPAA.

The SNs innervating the PM can be excited transcutaneously by stimulating the spinal column overlying the cauda equina, thus allowing measurement of the motor latency to this muscle. The patient was placed in the left lateral position and a separate ground electrode was connected from the upper thigh to the preamplifier of the electromyography (EMG) apparatus (Isolator SS-104J; Nihon Koden, Tokyo, Japan; 7T18 Signal Processor; SAN-EI Instruments, Ltd, Tokyo, Japan). The stimulus was applied from a magnetic stimulator (Magstim-Model 200; The Magstim Company Limited, Carmarthenshire, UK) consisting of a flat coil with an outer diameter of 9 cm placed in the area overlying segments 2 to 4 (S2–S4). A large, brief pulse of current (peak value 4000 A after 110 s) was then passed from a high-voltage capacitor discharge system through the coil. The PM response was recorded with a glove-mounted electrode array. This consisted of two metal plates mounted 1 cm apart on the tip of the gloved index finger. The finger bearing this device was inserted into the anal canal so that the electrode array was in contact with the PM on the right and left sides. Magnetic stimulation at the S2 to S4 sacral

levels is proven to activate the SN root of the cauda equina. The latency of induced EMG activity (M wave: motor activity potential evoked from muscles directly regulated by the stimulated motor nerve) of the PM was recorded as a result of the SNTML test. In the experimental procedure, no bowel preparation was carried out; and after an overnight fast, subjects were given the opportunity to empty their colon before the experiment. No sedative was given. Measurements of SNTML were performed at 9 AM with the subjects in the left lateral decubitus position with the knees drawn up.

All data are expressed as mean $\pm$ SD. Statistical analysis was performed using a nonparametric Kruskal-Wallis test with Bonferroni correction (Stat-View 4.0, Macintosh; SAS Institute, Cary, NC). A *P* value of less than 0.05 was regarded as significant.

Informed consent was obtained from all subjects participating in the present study. Application of the magnetic stimulation method was also approved by the Ethical Committee of Nihon University School of Medicine.

Operative Technique

We took a 2-stage procedure for UC of the total colon and rectum. The anal mucosa was stripped from the level of the dentate line with a short cuff (the remaining rectum: 2 cm from the upper edge of the PM), and IPAA (length of the J-pouch: 10 cm) was carried out by hand-sewn anastomosis. In addition, we performed an ileostomy. Closure of the ileostomy was carried out 1 month after IPAA.

Results

Characteristics of the patients with UC

Four out of 12 children with UC (case numbers 9–12) refused to undergo the SNTML measurement. The operative indications were refractory symptoms (41.7%, 5/12); chronic symptoms (33.3%, 4/12); steroid side effects (16.7%, 2/12); and toxic dilatation (8.3%; 1/12). The surgical complications were wound infection (16.7%, 2/12); intestinal obstruction (16.7%, 2/12); and anastomotic stricture (16.7%, 2/12). These patients did not have severe complications in their daily life. Pouchitis was detected in only 2 out of 12 patients 2 years after IPAA. In 12 patients after IPAA, the frequencies of soiling were 83.3% (10/12) 6 months after IPAA and 16.7% (2/12) 2 years after IPAA (Table 1).

Comparison of the SNTML values on the right and left sides

On the right side, the SNTML values of patients in groups A, B, and C were 4.9 ± 0.4 , 3.5 ± 0.6 , and 3.3 ± 0.2 ms, respectively. Patients in group A exhibited significant prolongation compared with groups B and C ($P < 0.0001$ and $P < 0.0001$, respectively). There was no significant difference between groups B and C ($P = 0.2329$; Tables 2 and 3). On the left side, the SNTML values of patients in groups A, B, and C were 4.8 ± 0.5 , 3.6 ± 0.5 , and 3.4 ± 0.3 ms, respectively. Similar to the right side, patients in group A displayed significant prolongation compared with groups B and C ($P = 0.0002$ and $P < 0.0001$, respectively). There was no significant difference between groups B and C ($P = 0.2315$; Tables 2 and 3).

The conduction delay of the SNTML in group A was the longest, followed by that in group B and then that in group C on the right and left sides.

Discussion

Adult patients after IPAA often have impaired defecation conditions such as soiling, incontinence, high nocturnal bowel frequency, feeling of stool remaining, and urgency for more than 1 year after their operation. It is reported that child patients after IPAA show resolution of defecation impairments more than 1 year after IPAA. One of the main complications in child patients after IPAA is soiling and/or incontinence,²¹ just as for their adult counterparts. In this study, 10 out of 12 patients (83.3%) 6 months after IPAA showed soiling, but 10 out of 12 patients (83.3%) showed continence 2 years after IPAA. The reason why child patients in the early period after IPAA show soiling, but become continent more than 2 years after IPAA is still unknown.

Generally, disturbance of anal sphincter functions after IPAA has been accepted as a common cause in the pathogenesis of fecal soiling or incontinence.^{19,20} Among manometric studies, there are many reports that patients after IPAA who had soiling and/or incontinence showed dysfunction of the anal sphincter and neorectum (decreased sensation, compliance, volume, and motility). The cause of postoperative anal dysfunction is not completely understood, but it is thought to be due to overdistension of the internal and external anal sphincter during endoanal mucosectomy and partial destruction of the pelvic neural plexus.^{21–23} Continence is a

Table 1 Characteristics of child patients after IPAA for UC

Case	Sex	Age, y	Range of lesion	Operative indication	Surgical complication	Pouchitis	Soiling at 6 mo	Soiling at 2 y
1	Male	11.0	TCR	CS	—	—	+	—
2	Female	11.6	TCR	RS	Intestinal obstruction	—	+	—
3	Male	11.8	TCR	Steroid side effect	Wound infection	—	+	—
4	Male	12.0	TCR	CS	—	—	+	—
5	Male	12.5	TCR	Steroid side effect	—	—	+	—
6	Male	12.7	TCR	RS	—	—	+	—
7	Female	12.8	TCR	CS	—	—	+	—
8	Male	13.0	TCR	RS	Intestinal obstruction	—	+	—
9	Male	10.8	TCR	Toxic dilation	Anastomotic stricture	+	+	+
10	Female	12.1	TCR	RS	Wound infection	—	—	—
11	Female	12.8	TCR	CS	—	—	—	—
12	Male	13.0	TCR	RS	Anastomotic stricture	+	+	+

CS, chronic symptoms; RS, refractory symptoms; TCR, total colon and rectum.

complex function that depends on functional integrity of the anorectum. Anal sphincter functions, especially the PM, are primary factors in maintaining continence.^{19,20} Manometric studies have not clarified the function of the PM. There are no reports of the function of the PM in child patients after IPAA. Kroesen *et al* suggested that the main factor in sphincter impairment is surgical injury of the pelvic neural structures rather than disruption of the anal sphincters, including the internal anal sphincter, external anal sphincter, and PM, during dilatation.²⁴ We thus investigated the neural control of the PM in this study.

In electrophysiological studies, many researchers have considered that the regulatory pathway of the PM is derived from SNs from the spinal S2 to S4 roots.²¹ Less is known about the neurological functions in patients with soiling after IPAA with regard to the SNs that regulate the PM. To the best of our knowledge, there are no reports of electrophysiological studies of SNs in child patients after IPAA. In the magnetic stimulation approach, it is possible to measure the latency of the SNs extending from the spinal S2 to S4 nerve roots to the PM, and the

response is easily induced with high reproducibility, thus providing a reliably consistent index for evaluations.^{21,25} Diagnostic tests of SNTML measurements are valuable because they directly assess the underlying abnormality, which consists of damage to the innervation of the PM.²¹ In particular, the nerve stimulation technique using a magnetic stimulator may be useful in pinpointing the PM lesion causing soiling and incontinence.²¹ We thus studied the sacral nerve function using SNTML in patients with or without soiling after IPAA for UC in childhood.

In the present study, as measurements of the SNTML were conducted by an experienced investigator (RT), reproducible normal values with small discrepancies in values monitored at the right and left sides of the PM were established in the control subjects. Furthermore, in the SNTML investigations for patients after IPAA, a marked conduction delay in the SNTML was observed at the right and left sides of the PM of patients with soiling (6 months after IPAA) compared with that of the same patients who had become continent 2 years after IPAA. There were also no significant differences in the values of

Table 2 SNTML values in groups A and B

Case	Group A (6 mo after IPAA)		Group B (2 y after IPAA)	
	Right side, ms	Left side, ms	Right side, ms	Left side, ms
1	5.2	5.1	3.5	3.9
2	4.4	4.6	3.9	3.9
3	5.4	5.5	3.8	3.8
4	4.6	4.5	3.8	3.9
5	4.7	4.6	3.8	3.9
6	4.5	4.5	2.6	3.0
7	5.4	5.5	3.9	3.8
8	4.7	4.4	2.6	2.7
Mean $\pm$ SD	4.9 $\pm$ 0.4	4.8 $\pm$ 0.5	3.5 $\pm$ 0.6	3.6 $\pm$ 0.5

Table 3 Comparison of the SNTML values at the right and left sides

	Group A (n = 8)	Group B (n = 8)	Group C (n = 26)
Right side	4.9 ± 0.4 ms ^a	3.5 ± 0.6 ms ^b	3.3 ± 0.2 ms ^c
Left side	4.8 ± 0.5 ms ^d	3.6 ± 0.5 ms ^e	3.4 ± 0.3 ms ^f

a vs. b: $P < 0.0001$, a vs. c: $P < 0.0001$, b vs. C: $P = 0.2329$.

d vs. e: $P = 0.0002$, d vs. f: $P < 0.0001$, e vs. f: $P = 0.2315$.

SNTML: Sacral nerve terminal motor latency.

Group A: Patients with soiling 6 months after ileal J pouch-anal anastomosis.

SNTML at the right and left sides. It is well known that the PM is regulated by bilateral SN. Therefore, the authors performed bilateral SNTML measurements. The PM surrounds the rectum in a U-shape and may be regulated by the neural crisscrossing of the right and left sacral nerves. Therefore, the authors should carry out the SNTML measurement at a posterior site of the PM. The reason why the SNTML was delayed in patients with soiling after IPAA may be as follows: Child patients present practical difficulties for intrapelvic procedures and operative interventions through the anus because the pelvis is narrow. Therefore, the PM may have been excessively extended and injury might have been inflicted on both sides of the SN.

In summary, the conduction delay of the SNTML in child patients with soiling 6 months after IPAA was significantly longer than that of the same continent patients 2 years after IPAA or the control subjects. However, no significant differences in conduction delay between the continent patients 2 years after IPAA and control subjects were noted. These findings suggest that damage to the bilateral SNs, probably due to surgical stretching injury, may well be the cause of the soiling seen at 6 months after IPAA. However, the SNTML showed normal values 2 years after IPAA. These findings support the hypothesis that child patients with soiling after IPAA may recover the function of the bilateral SNs. The magnetic stimulation method may not directly stimulate the SNs itself and relies on a more general stimulation penetrating the surrounding tissue and activating the nerve. For more accurate measurement of the correct SNTML, the authors should use a concentric needle electrode to the PM. In this study, the child cases after IPAA were too few. The mechanism of anorectal function in humans is complicated and its control probably involves delicate functioning of various other factors.^{25,26} Studies from various aspects with larger cohorts are thus required in the future.

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