

Difference of Postoperative Stool Frequency in Hirschsprung Disease According to Anastomosis Level in a Single-Stage, Laparoscopy-Assisted Transanal Endorectal Pull-Through Procedure

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Abstract: Anorectal innervation that governs sensation, motor function, and rectal accommodation can be influenced by the type of surgical procedure used to treat children with Hirschsprung disease. At our institution, we began to perform single-stage, laparoscopy-assisted transanal endorectal pull-through (LATEP) with submucosal dissection and anastomosis of the ganglionated bowel at 2 different levels relative to the dentate line.

This retrospective study describes postoperative stool frequency changes in response to this procedure. Forty infants who underwent single-stage LATEP between September 2003 and April 2012 in a single center by the same surgeon were included in our analysis.

The patients were divided in 2 groups: Group A (n = 23) underwent submucosal dissection and anastomosis at 2 mm above the dentate line, and Group B (n = 17) underwent the same procedure with anastomosis 15 mm above the dentate line. Clinical characteristics, clinical findings on the first postoperative visit, and instances of coexisting anomalies did not differ between the 2 groups. Aganglionic segments were found in the rectosigmoid colon in 18 cases (78.2%) in Group A and in 15 cases (88.2%) in Group B. Although the stool frequency was no different at 1, 3, 6, and 12 months after the operation, Group B showed significantly fewer bowel movements than Group A after 2 years (3.77 in Group A vs 2.0 in Group B; $P = 0.035$) and after 3 years (3.92 vs 1.29; $P = 0.009$) in patients who had aganglionosis of the rectosigmoid colon. The mean follow-up period was 65.87 ± 28.08 months for Group A and 35.59 ± 18.68 for Group B.

The level of submucosal dissection and anastomosis in single-stage LATEP influenced the stool frequency in rectosigmoid aganglionosis.

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Abbreviations: HD = Hirschsprung disease, LATEP = laparoscopy-assisted transanal endorectal pull-through.

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INTRODUCTION

Hirschsprung disease (HD) is a congenital anomaly characterized by the absence of ganglion cells in the myenteric and submucosal plexuses. The anomaly is usually confined to the rectum and distal colon. However, 2% to 13% of all patients with HD have total colonic aganglionosis.^{1,2} The surgical goal in HD is to remove the aganglionic bowel and pull the normally innervated bowel through the anus to allow normal bowel movements. The most well-known operations are the Swenson, Duhamel, and Soave procedures. Each surgical procedure has its own advantages, making it difficult to judge which one is better, so the choice usually depends on the individual surgeon's preference. However, different procedures may have varying effects on anorectal innervation and sensation, motor function, and accommodation.³

Previous studies have reported that fecal incontinence occurs in 1% to 17% of patients after the Soave procedure.^{4,5} In 1986, Martin et al used the Soave procedure to treat patients with ulcerative colitis and reported that anal continence was related to the level of the anastomosis.⁶ Therefore, we designed this retrospective study to determine the clinical outcomes among pediatric patients who underwent single-stage, laparoscopy-assisted transanal endorectal pull-through (LATEP) in which the level of submucosal dissection and anastomosis was either 2 or 15 mm above the dentate line.

MATERIALS AND METHODS

Patient records from the Samsung Medical Center pediatric surgery database were retrospectively reviewed. Between September 2003 and April 2012, 40 infants underwent single-stage LATEP performed by the same surgeon and were selected for our study. Patients were diagnosed with HD based on the results of full-thickness rectal biopsy or rectal suction biopsy.

Birth weight, gestational age, age and body weight on the day of the operation, location of the aganglionic segment, coexisting anomalies, postoperative clinical course, and bowel movements at 1, 3, 6, 12, 24, and 36 months after the operation were analyzed. Every patient underwent the operation before 1 year of age and was not toilet-trained. Data of the patients' bowel movements were based on the protocolized bowel movement scale used in Samsung Medical Center (Table 1). This scale was developed to objectively quantify stooling frequency and patterns in patients younger than 3 years who have yet to achieve voluntary bowel movements. Only stool frequency was selected as the factor for comparison, whereas soiling was excluded. Patients who gained bowel continence during the study period were also assessed with continence evaluation scoring system.⁷ Constipation was defined as bowel frequency of <2 times per week. There was no patient who was lost during follow-up.

TABLE 1. Bowel Movement Check List Before Voluntary Bowel Movement

Details	Mean Frequency During the Week of Visit
Stooling: requiring change of diaper	Frequency/day
Soiling: staining on diaper	Frequency/week
Constipation: <2 times per week	Yes/No
Voluntary bowel movement	Yes/No

Operative Technique

Patients were divided into 2 groups according to the level of submucosal dissection and anastomosis. Before September 2008, we began submucosal dissection from 2 mm above the dentate and performed anastomosis at this level (Group A). Since September 2008, we changed operative policy to performing submucosal dissection and anastomosis from 15 mm above the dentate line (Group B). One day before surgery, all the patients underwent bowel preparation by rectal irrigation and anorectal stimulation. Patients were placed near the edge of the operating table and were covered with sterile drapes from nipple to toe. Intravenous broad-spectrum antibiotics were injected just before the incision was made. Three trocars were used, and a 5-mm transumbilical camera port and two 3-mm working ports were inserted bilaterally on the lateral two-thirds of the line from the umbilicus to the anterior superior iliac spine. The first

step during the intra-abdominal procedure was to locate the transitional zone. Full-thickness seromuscular biopsy was performed 2 cm above the transitional zone, and sutures were placed if there was perforation or bleeding at the biopsy site.

After the presence of ganglion cells was confirmed, rectal dissection was performed. During rectal dissection below the peritoneal reflection, only the posterior aspect of the rectum was dissected, sparing the lateral and anterior walls to preserve the nerve plexuses. After it was established that the part of the colon 5 cm above the biopsy-confirmed site was sufficiently redundant to reach the anal canal without tension, the transanal approach was begun. A self-retaining anal retractor (Lone Star Medical Products, Stafford, TX) was placed on the perineum for better exposure of the anus.

From this point, the procedures used for Groups A and B differed. In Group A, the hooks of the retractors were embedded at the dentate line (Figure 1). A circumferential incision was made 2 mm above the dentate line, and submucosal dissection was performed. When rectal intussusception was able to occur naturally because of the redundancy achieved after submucosal dissection, the muscle cuff was resected, and the colon was pulled through the anal canal and resected. The rectal muscular cuff was kept as short as possible, and the posterior muscular cuff was split at the level of the peritoneal reflection. The ganglionated end of the proximal colon was anastomosed 2 mm above the dentate line with the use of interrupted sutures (4-0 or 5-0 glyconate monofilament) (Monosyn, B. Braun, Melsungen, Germany).

Group B underwent the same procedure as Group A as described above except that the submucosal dissection and

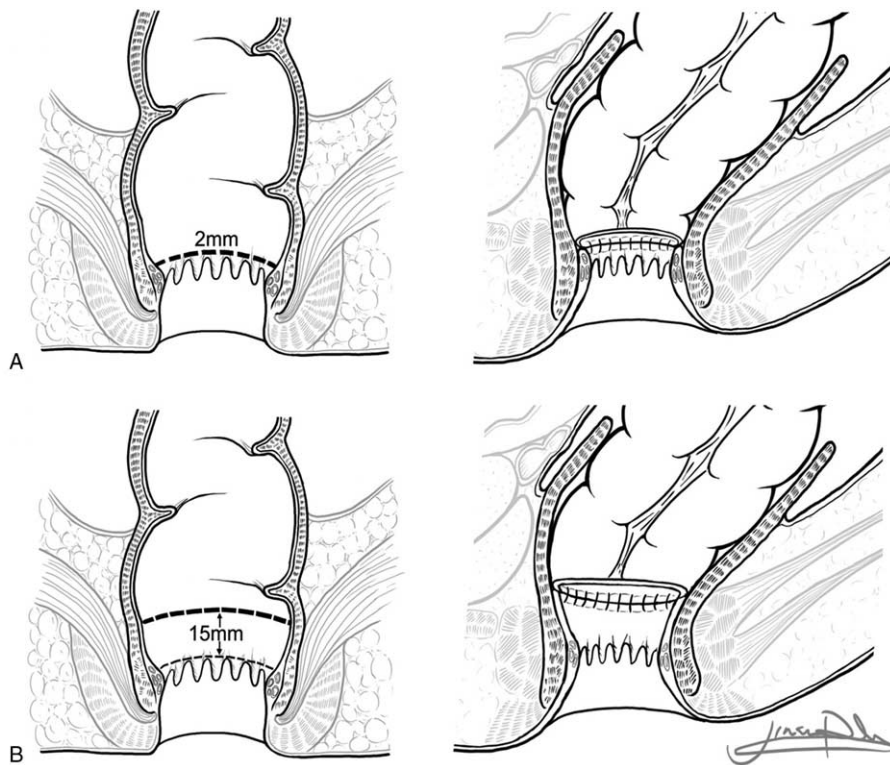


FIGURE 1. Differences in surgical technique. (A) In Group A, submucosal dissection and anastomosis of the ganglionated bowel was done at 2 mm above the dentate line. (B) In Group B, submucosal dissection and anastomosis of the ganglionated bowel were done at 15 mm above the dentate line.

anastomosis of the ganglionated proximal end were performed at 15 mm above the dentate line instead of 2 mm.

Statistical analyses were conducted using SPSS 21 (Statistical Package for the Social Sciences, SPSS Inc, Chicago, IL). Mean scores were compared using 2 independent sample *t* tests for continuous and normally distributed data, and Mann–Whitney *U* test for non-normally distributed data. The χ^2 test was used to analyze differences in sex distribution, site of aganglionosis, and combined anomalies. This study was approved by the institutional review board at Samsung Medical Center (IRB File No. 2015-08-116).

RESULTS

A total of 40 patients underwent single-stage LATEP, 32 (80%) of whom were male. The median gestational age was 39 + 3 weeks (range 37 + 2 – 42) and median birth weight was 3.27 kg (range 2.4–4.1). Of the 40 patients, 28 (70%) were treated within a month after birth; 8 patients (20%) were treated between 1 month and 6 months, and 4 patients (10%) underwent the operation between 6 months and 1 year after birth. The most common site of the aganglionic segment was the rectosigmoid colon, in 33 patients (82.5%) (Table 2).

There were 23 patients in Group A and 17 patients in Group B. No significant differences were found between the 2 groups in terms of male:female ratio, birth weight, gestational age, and age on the day of operation (24 vs 27 days; *P* = 0.730). Postoperative clinical course and coexisting anomalies were also similar between the 2 groups. Aganglionic segments were confirmed to be in the rectosigmoid colon in 18 patients (78.2%) in Group A and 15 patients (88.2%) in Group B. Aganglionic segments up to the descending or transverse colon were confirmed in 5 cases in Group A and 2 cases in Group B. Two patients in group A and 4 patients in group B were able to acquire voluntary bowel movement during the follow-up period (Table 3).

The number of postoperative stool frequency was compared between the 2 groups according to the aganglionic segment. There was no statistically significant difference in bowel movement frequency at 1, 3, 6, and 12 months after the operation in the patients with rectosigmoid colonic aganglionosis (Figure 2A). However, Group B showed significantly fewer bowel movements as compared with Group A at 2 years (3.77 in Group A vs 2.0 in Group B; *P* = 0.035) and at 3 years

(3.92 vs 1.29; *P* = 0.009) after the procedure. Although the patients in Group B who had transverse and descending colonic aganglionosis seemed to have fewer bowel movements, there was no statistically significant difference between the 2 groups (Figure 2B).

Enterocolitis occurred in 2 cases in each group. Mechanical ileus occurred in 1 patient in Group B but was resolved after conservative management. Mean follow-up was 65.87 ± 28.08 months in Group A and 35.59 ± 18.68 months in Group B.

DISCUSSION

Before adopting the Soave procedure, our primary choice of surgical procedure for HD was the Duhamel procedure. At that time, we believed it was important for the level of the anastomosis to be low, near the dentate line, to relieve sphincter pressure and thus prevent postoperative constipation. Antao et al⁸ had suggested that this low anastomosis approach in the Duhamel procedure was associated with lower sphincter pressure and prevented postoperative constipation. With this in mind, we were performing our LATEP procedure based on the same concept. However, the patients seemed to have more frequent bowel movements as compared with normal babies. Therefore, we hypothesized that a lower anastomosis in the LATEP procedure was the cause of more frequent bowel movements. In 1986, Martin et al reported that their patients with ulcerative colitis who underwent the Soave procedure with anastomosis at the level of the dentate line showed anal incontinence, whereas patients with anastomoses above the columns of Morgagni showed total continence.⁶ In 2005, Ishihara et al⁹ reported that in patients in whom transanal dissection was lower than the dentate line, continence scores were significantly worse than in patients in whom dissection was on the same level as the dentate line. Hollwarth et al¹⁰ explained these findings as being the result of damage to the delicate nerve endings on the dentate line that make up the reflex arch responsible for sensation and continence. For these reasons, we decided to revise our surgical procedure since September 2008. In 2 recently published reports on transanal pull-through procedure, comparisons were made between mucosal dissection procedures starting from the dentate line and anorectal line. Postoperative fecal continence scores were higher in the anorectal line group.^{11,12}

The number of cases of single-stage pull-through procedures in patients with HD is increasing, with this procedure being applied in up to 80% of cases and multiple-stage pull-through procedures being reserved for severely ill patients.^{13,14} The single-stage pull-through procedure has several advantages. Anesthesia needs to be administered only 1 time, there is no need for additional admissions, and the hospital stay is shorter.¹⁵ Our study included only those patients who underwent single-stage LATEP. Despite its retrospective design, our study revealed no differences between Groups A and B in terms of patient characteristics.

After 1995, when Georgeson et al¹⁶ introduced their first laparoscopy-assisted modified Soave procedure, many centers around the world adopted this new procedure in their practices. Three years later, De la Torre-Mondragon and Ortega-Salgado¹⁷ reported their use of an entirely transanal endorectal pull-through procedure. Although many centers adopted this procedure because of its simplicity and because it obviated laparoscopy, not enough randomized trials were conducted to compare the 2 procedures. In our view, precise localization of the transitional zone is necessary before the transanal endorectal dissection is begun, because in 8% of cases, the histological

TABLE 2. Characteristics of All Patients (n = 40)

Details	No. of Patients
Male sex, n (%)	32 (80%)
Birth weight,* kg	3.27 (2.4–4.1)
Gestational age,* wk	39 + 3 (37 + 2 – 42)
Time to operation	
At birth–1 mo	28 (70%)
1 mo–6 mo	8 (20%)
6 mo–12 mo	4 (10%)
Site of aganglionosis	
Rectosigmoid colon	33 (82.5%)
Descending colon	1 (2.5%)
Transverse colon	6 (15%)
Follow-up,* mo	50 (1–119)

*Median.

TABLE 3. Characteristics and clinical course by Group

Details	Group A	Group B	P
Patient numbers	23	17	
Male sex, n (%)	18 (78.2%)	14 (82.3%)	1.0
Birth weight,* kg	3.3 (2.7–4.1)	3.24 (2.4–4.0)	0.761
Gestational age,* wk	39+4 (37+2–42)	39+3 (38+1–40)	0.598
Age at first pull-through,* day	24 (9–306)	27 (7–191)	0.730
Weight at operation,* kg	3.46 (3–8.9)	3.8 (3.3–7.3)	0.056
Hospital days after operation*	6 (3–42)	6 (4–12)	0.951
Start of postoperative feeding,* day	2 (1–7)	2 (2–5)	0.704
Postoperative stool passing,* day	1 (1–4)	1 (1–4)	0.543
Site of aganglionosis			
Rectosigmoid colon	18 (78.2%)	15 (88.2%)	0.677
Descending colon	1	0	1.0
Transverse colon	4	2	1.0
Combined anomalies	4	4	0.702
Congenital heart disease	1	1	1.0
Down syndrome	0	1	0.425
Genitourinary anomaly	2	0	0.499
Others†	2	3	0.634
Voluntary bowel movement	2	4	0.373
Follow-up duration, mo	65.87 ± 28.08	35.59 ± 18.68	0.001

*Median.

†In Group A: 1 congenital dislocation of hip, 1 mental retardation of unknown origin. In Group B: 3 inguinal hernias.

transitional zone is seen to be proximal to the radiographically estimated rectosigmoid transitional zone on contrast studies.¹⁸ In such cases, the transanal approach can lead to failure owing to poor mesenteric dissection. However, the laparoscopic approach can be successful even in patients with long-segment aganglionosis.

If the rectal muscular cuff is longer than necessary, compression can disturb peristalsis of the pulled-through normal colon.¹⁹ A case of the muscular cuff rolling down during transanal pull-through and forming a constricting ring around the anastomosed normal bowel has been reported in the

literature.²⁰ In 2007, Nasr and Langer²¹ reported a lower incidence of postoperative rectal dilatation in the group that underwent shorter mucosectomy, with the rectal muscular cuff extending only 1 to 2 cm above the dentate line. In our practice, we also try to make the muscular cuff as short as possible during the transanal approach. According to another study, the mean length of the anal canal is 1.67 ± 0.34 cm in neonates and 3.03 ± 0.52 cm in older children.²² In our study, the median age of the patients on the day of operation was 26 days (range 7–306); therefore, we predict the rectal cuff to be 1 to 2.5 cm above the peritoneal reflection.

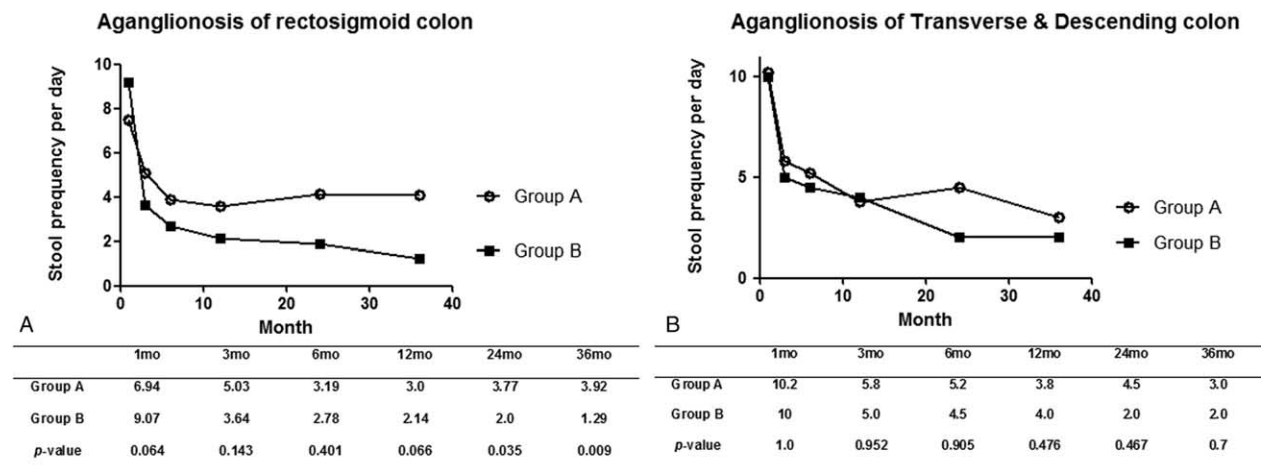


FIGURE 2. Bowel movement changes after the pull-through procedure. (A) Although no difference was found between Groups A and B until 12 months, Group B had fewer bowel movements starting 24 months after the operation. (B) No statistically significant differences were found between Groups A and B in patients with aganglionosis of transverse and descending colon.

It was difficult to clearly define stool frequency before the acquirement of voluntary bowel movement. Although a continence evaluation scoring system exists for patients older than 3 years,⁷ continence evaluation scale does not exist for patients younger than 3 years, who did not acquire voluntary bowel movement. In Samsung Medical Center, we applied our protocolized bowel movement scale for anorectal malformation patients since 2001. More effort will be needed to appropriately divide stooling and soiling in patients who lack voluntary bowel movement, despite the fact that this study defined soiling as staining of stool in a diaper or underwear. Moreover, a follow-up >3 years would precisely reflect the outcome of each surgical procedure in terms of stool frequency.

A major limitation of this study is that it was retrospective in design, with the possibility of imprecise data collection. Also there are only small number of patients in each group, limiting the significance of statistical analysis. Another limitation is the lack of objective data (eg, anorectal manometry or sensory test results) to back our hypothesis that the proximity of submucosal dissection and anastomosis to the dentate line may damage delicate nerve endings in this region and lead to the difference in bowel movement frequency.

CONCLUSIONS

The level of submucosal dissection and anastomosis in single-stage LATEP influenced postoperative stool frequency in the patients with rectosigmoid segment confined to HD. At 24 and 36 months after corrective surgery for aganglionosis of the rectosigmoid colon, the number of bowel movements was significantly smaller in patients with anastomoses created 15 mm above the dentate line than in patients with anastomoses created 2 mm above the dentate line. To improve the quality of future research on postoperative outcomes, prospective and randomized controlled studies should be performed and a clear distinction should be made between bowel movements and fecal soiling in whom voluntary bowel movement is acquired. In addition, more accumulation of the cases and longer-term follow-up is needed to evaluate which is the best anastomosis level of patients who have undergone surgical treatment for HD.

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