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Surgical approach for fecal incontinence with a patulous anus after transanal pull-through for Hirschsprung disease☆

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ABSTRACT

Background: We have performed transanal pull-through (TAPT) for Hirschsprung disease since 1998. Some of our patients after TAPT showed a patulous anus and suffered from severe true fecal incontinence. We performed anal canal plasty for these patients and evaluated its efficacy in restoring anorectal function.
Methods: Thirty-one patients who were ≥5 years old were previously operated on for Hirschsprung disease, and seven (22.5%) of these were indicated for this procedure. Anorectal function was evaluated using the Japanese Study Group of Anorectal Anomalies (JSGA) clinical assessment of defecation function score. For surgery, the patients were positioned in the prone jackknife posture. The posterior half of the anal canal was exposed and folded inward until the anal canal lumen was as narrow as the surgeon's index finger. External anal sphincter muscles were repaired, and the wound was closed vertically.
Results: The mean preoperative JSGA score was 1.42 ± 0.4. The mean JSGA scores at 2–6 months and 2 years after this procedure were 5 ± 2.1 and 5.8 ± 2.1, respectively. Postoperatively, the JSGA score significantly improved at both times ($p < 0.05$).
Conclusions: Anal canal plasty may be effective for true fecal incontinence and a patulous anus after TAPT. This surgical approach may be useful for these conditions.
Level of evidence: Type of study: Treatment study, Level IV.

One-stage transanal pull-through (TAPT) is one of the most widely accepted operative techniques for Hirschsprung disease (HD) [1]. We have also performed the transanal Soave procedure since 1998, and most patients have shown favorable anorectal function. However, some of our patients suffered from severe fecal incontinence for longer than 5 years after the operation. They had a patulous anus and true incontinence. Levitt and colleagues reported the same conditions [2–4]. Gosemann et al. [5] reported that the mean rate of fecal incontinence after TAPT (including laparoscopic-assisted TAPT) was 25% (94/860 cases), but there have been no previous reports on the rate of this true incontinence. Levitt et al. reported evaluation and a bowel management program for fecal incontinence after surgery for HD [2,3]. They also reported that 42.6% (29/68) of patients who were suffering from fecal incontinence after surgery for HD had true incontinence, and 20.6% (6/29) of patients did not improve with the bowel management program [2]. In our experience, patients presenting with true fecal incontinence with a patulous anus did not improve with bowel management. Levitt and colleagues presumed that these conditions were because of damage to the anal canal during the transanal procedure, and mentioned the effectiveness of reconstruction of the anal sphincter. Ohmi et al. reported that anal canal plasty was effective for idiopathic fecal incontinence in adult patients [6]. We modified this procedure for these patients with true incontinence after an operation for HD.

This study aimed to evaluate the efficacy of anal canal plasty for these patients in restoring anorectal function. To the best of our knowledge, this is the first report of surgical repair for children with fecal incontinence because of sphincter damage after the Soave procedure.

1. Materials and methods

1.1. Patients

Indications of anal canal plasty were as follows: (1) patients with a patulous anus (Fig. 1A); (2) radiologically, no fecal accumulation was present in the colon, and there was inability to retain contrast material in the rectum (Fig. 1B); and (3) severe fecal incontinence. Severe fecal incontinence was defined as patients having to change their underwear more than two to three times a day, in spite of bowel management with an enema or loperamide. At our institute, 31 patients who were 5 years old or older had previously been operated on for HD, and seven (22.5%) of these patients were indicated for this procedure. The patients'

Abbreviations: TAPT, transanal pull-through; HD, Hirschsprung disease; JSGA, Japanese Study Group of Anorectal Anomalies; 3D, three-dimensional.

☆ Conflicts of interest: None.

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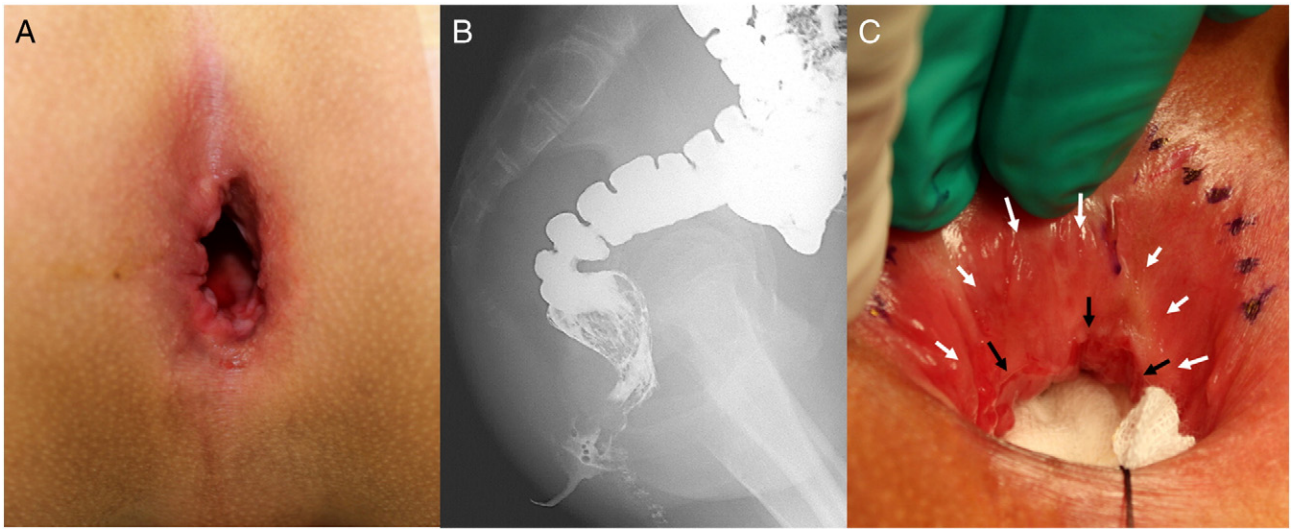

Fig. 1. Appearance of the anus and contrast enema before anal canal plasty. A, Patulous anus of patient 3. B, Contrast enema of patient 3 shows no fecal accumulation in the bowels and inability to close the anal canal. C, Dentate line and anastomosis line of patient 3. The dentate line is preserved, but the anastomosis line is close to the dentate line. White arrow, dentate line; black arrow, anastomosis line.

characteristics are shown in Table 1. The patients included six boys and one girl. Three patients had long segment aganglionosis that extended up to the transverse colon, two had rectosigmoid aganglionosis, and two had short segment aganglionosis. All patients had a one-stage procedure by irrigating and decompressing the colon and rectum with a transanally-inserted tube preoperatively. The mean (SD) ages at the initial operation and at this procedure were 41.14 ± 27.34 days old and 10.57 ± 2.44 years old, respectively.

At anal canal plasty, we assessed the dentate line and the anastomosis line under general anesthesia. In all of the patients, the dentate lines were preserved, but the anastomosis line was close to the dentate line, with a distance between lines of less than 5 mm (Fig. 1C).

1.2. Surgical procedure

For preoperative bowel preparation, laxative agents were administered for 3 days, and the patients had no oral intake with intravenous infusion on the previous day of the operation. We modified anal canal plasty for fecal incontinence of adult patients, as reported by Ohmi et al. [6]. A summary of the procedure is shown in Fig. 2A.

Under general anesthesia, the patient was positioned in the prone jackknife posture. A skin incision was made in a posterior half arc around the anus (Fig. 2B). The external sphincter muscle was confirmed by electrical stimulation. This muscle was stretched in most cases. We dissected between the muscle and the wall of the anal canal to expose the posterior wall of the anal canal. This dissection was extended to the cranial side for approximately 3 cm, and then the inferior part of the puborectal muscle was exposed. Further dissection between the posterior wall of the rectum and puborectal muscle was continued, and the posterior half of the anal canal was exposed for approximately 4 cm (Fig. 2C). The posterior wall of the anal canal was then folded inward until the anal canal lumen was as narrow as the surgeon's index finger. This wall was then held with interrupted stitches of 3–0 or 2–0 absorbable sutures by two to three layers (Fig. 2D). After folding the anal canal, some planes between the anal canal and the dissected muscles were created (Fig. 2E). These muscles were stitched in the posterior midline to close these planes and repair the muscle complex. The wound was closed vertically in a layer-to-layer fashion (Fig. 2F).

After surgery, the most frequent and serious complications were wound infection and dehiscence. To prevent these complications, prophylactic antibiotics were administered for 1 week, and the patients had no oral intake, except for clear liquid for 1 week. To protect the wound, the patients were asked not to defecate in the sitting position with abdominal pressure for 1 week. When the wound was intact, the patients then used a daily enema until they had voluntary bowel movement. After they had voluntary bowel movement, four patients who had hard stool or constipation were administrated laxatives or enemas if necessary. The patulous anus was narrowed to a normal size after this procedure (Fig. 3A).

1.3. Evaluation of anorectal function

Anorectal function was evaluated by a clinical score, anorectal manometry, and contrast enema.

1.3.1. Clinical score

We used the JSGA clinical score, which is composed of the following: minimum = 0, maximum = 8; presence of the urge to defecate = 0–2; severity of incontinence = 0–4 or severity of constipation = 0–4; and severity of soiling = 0–2. The total score is calculated as the sum

Table 1
The patient characteristics.

Patient	Sex	Age of this procedure (years)	Age of initial operation for HD (days)	Type of HD	Associate malformation
1	Male	6	19	long segment	
2	Male	9	20	Long segment	
3	Male	13	28	Short segment	
4	Male	12	24	Long segment	Mental retardation because of viral encephalitis
5	Male	12	82	Short segment	
6	Male	9	29	Short segment	
7	Female	13	86	Short segment	Mowat–Wilson syndrome

HD: Hirschsprung's disease.