



Long-term outcome of left- vs right-sided antegrade continence enema[☆]

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Abstract

Background/Purpose: This study compared long-term outcomes of left- vs right-sided antegrade continence enema (ACE) procedures.

Methods: We retrospectively analyzed medical records and performed a follow-up telephone survey of patients who underwent the ACE procedure at Severance Children's Hospital between 1999 and 2011.

Results: A total of 49 patients underwent 51 ACE procedures. The procedures were left-sided ACE in 25 patients (group 1) and right-sided in 26 patients (group 2), including 2 patients who underwent left-sided procedures a few years earlier. Fecal soiling was more common in group 1 than in group 2 (8/25 [32.0%] vs 1/26 [3.9%], respectively; $P = .0109$). Seven patients (28.0%) in group 1 had abdominal pain unrelieved by ACEs. Stomal reflux of fecal material was more frequent in group 1 than in group 2 (13/25 [52.0%] vs 0/26 [0%], respectively; $P < .0001$). Nonuse or infrequent use of ACE was more common in group 1 than in group 2 (11/25 [44.0%] vs 0/26 [0%], respectively; $P < .0001$). Patient satisfaction was higher in group 2 ($P = .0015$).

Conclusions: In our experience using the sigmoid colon for left-sided ACE, right-sided ACE was superior to left-sided ACE in achieving fecal continence, resolving intractable constipation, and providing patient satisfaction.

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For constipation or fecal incontinence in patients with anorectal malformations or meningomyeloceles, various conservative techniques have been used, such as digital evacuation, laxatives, retrograde enemas, and biofeedback. The Malone procedure for antegrade continence enemas

(ACEs), introduced in 1990, provides complete colonic emptying with antegrade washouts for patients who have fecal incontinence uncontrolled by conservative management [1]. ACEs by the Malone procedure or other modified procedures based on it have provided effective treatment of fecal incontinence or severe constipation where conservative methods have failed [2-6]. In 2002, left-sided ACE was introduced to shorten the duration of the enema because of the long and tedious process required for complete colonic emptying with ACE by the Malone procedure [7-10].

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In the last 2 decades, the results of ACE, either right-sided or left-sided, have been reported as satisfactory [4,6,9,11,12]. In only 3 reports, the outcomes and complications of the left-sided ACE were compared with those of the right-sided ACE. These reports, including 2 small studies and a review article, showed no significant differences between the procedures [13-15]. In the current study, we compared the surgical complications and the long-term outcomes of left-sided ACE to those of right-sided ACE in a relatively large cohort.

1. Methods

We retrospectively analyzed the medical records of patients who underwent the ACE procedure in Severance Children's Hospital between 1999 and 2011. Demographics, indication for ACE, surgical techniques, postoperative complications, and problems related to enemas were reviewed. We conducted a telephone survey to assess patient satisfaction and complaints.

The ACE procedure was suggested for patients who had persistent fecal incontinence despite conservative management. The selection process for those undergoing either left- or right-sided ACE was not dependent on preoperative studies but, rather, the year in which the procedure was performed. The right-sided ACE procedure was performed for the initial 3 years of the study. From 2002 to 2006, only the left-sided ACE procedure was performed. We subsequently returned to the right-sided procedure again from 2007 until present.

The left-sided ACE procedure was performed using the retubulized sigmoid colon flap or ileal flap as the conduit. This conduit was wrapped around a Foley catheter, implanted in the sigmoid colon at one end, and reinforced with several seromuscular sutures in the sigmoid colon to prevent reflux of fecal materials. The other end was exteriorized through a stab incision at either the umbilicus or the left lower quadrant of the abdomen. The other specifics of the left-sided ACE procedures were performed as described in previous reports [11,12]. The right-sided ACE procedure was performed using the appendix or cecal flap as previously described [1-3]. Specifically, the appendix or cecal flap was implanted in the cecal tunnel to prevent reflux of fecal materials. The end of the appendix or cecal flap was externalized through a stab incision at the umbilicus or the right lower quadrant of the abdomen. To create the stoma in both the left-sided and right-sided ACE procedures, an anastomosis was performed between the end of the conduit and the skin of stoma with several interrupted, absorbable sutures.

Postoperatively, an 8F or 10F Foley catheter was left for 1 to 2 weeks. Enemas were initiated 1 to 2 weeks after the ACE procedure with a small testing volume of 50 to 100 mL of normal saline. The patients were subsequently recommended to continue enemas with small amounts of normal saline or

tap water and to increase the volume of enema fluid every day or every other day until fecal continence could be maintained. Then, patients were instructed to optimize the volume of the washouts to clean the bowel as well as the frequency of ACEs needed to maintain fecal continence. After the initiation of ACE, the catheter was removed. If patients presented with abdominal pain during ACE, they were recommended to mix a small amount of dietary salt into the enema fluid. In the event patients experienced fecal soiling without fecal impaction during ACE, they were recommended to take antidiarrheal medication. Sodium phosphate enema solution was used when fecal soiling was not controlled by ACE with tap water with concurrent fecal impaction.

The telephone survey included patients who had maintained ACEs without undergoing subsequent repairs. Patients were asked to score their level of satisfaction using the following scale: 1 for poor; 2, unsatisfactory; 3, fair; 4, good; and 5, excellent. Also, they were asked about complications or episodes of fecal soiling, constipation, and abdominal pain in relation to ACEs.

Short-term complications were defined as those occurring within 1 month after the procedure and *long-term complications* as those occurring from 1 month after the procedure to the end of follow-up. Categorical parameters were analyzed with the χ^2 test or Fisher exact test, and continuous parameters were analyzed using the Wilcoxon rank sum test. Statistical analyses were performed with SAS version 9.2 software (SAS Institute Inc, Cary, NC). A *P* value less than .05 was considered indicative of statistical significance. The study was approved by the Ethics Committee of Severance Hospital with the approval number 4-2011-0749.

2. Results

A total of 49 patients underwent 51 ACE procedures during the study period. Left-sided ACE procedures were performed in 25 patients (group 1), and right-sided ACE procedures in 26 patients (group 2), including 2 patients who underwent left-sided ACE procedures a few years earlier. The clinical characteristics of the patients are presented in Table 1. The sex distribution and mean age did not differ significantly between the 2 groups. The mean age of all patients who underwent the ACE procedure was 8.7 ± 6.2 years (10.4 ± 8.1 years in group 1 vs 7.0 ± 2.9 years in group 2; *P* = .2749). The indications for the ACE procedure were (lipo)meningomyelocele, anorectal malformation, and Hinman syndrome. The most common indication for ACE was different between the 2 groups, being (lipo)meningomyelocele (84.0%) in group 1 and anorectal malformation (46.2%) in group 2 (*P* < .0001). The mean time between the ACE procedure and the last follow-up was also different between the 2 groups (64.5 ± 31.9 months in group 1 vs 33.4 ± 38.6 months in group 2; *P* = .0012).

Table 1 Clinical characteristics of groups 1 and 2

	Group 1, left-sided ACE (n = 25)	Group 2, right-sided ACE (n = 26)	<i>P</i>
Sex, male:female (%:%)	13:12 (52.0:48.0)	18:8 (69.2:30.8)	.2077
Age at ACE procedure (y)	10.4 ± 8.1	7.0 ± 2.9	.2749
Indication for ACE, n (%)	(L)MMC: 21 (84.0) ARM: 3 (12.0) HS: 1 (4.0)	(L)MMC: 10 (38.5) ARM: 12 (46.2) ARM, (L)MMC: 4 (15.4)	<.0001
Length of follow-up (mo)	64.5 ± 31.9	33.4 ± 38.6	.0012

(L)MMC, (lipo)meningomyelocele; ARM, anorectal malformation; HS, Hinman syndrome.

Short-term complications consisted of 3 cases of wound infections (11.5% [3/26] in group 2), 2 cases of intestinal obstructions (4.0% [1/25] in group 1 and 3.8% [1/26] in group 2), 1 case of disruption of the conduit (3.8% [1/26] in group 2), and 1 case of traumatic perforation at the anastomotic site (4.0% [1/25] in group 1) (Table 2). This perforation was caused by parastomal penetration during insertion of the catheter. Three patients (12.0%) in group 1 and 1 (3.8%) in group 2 underwent revision of their stomal stricture within 1 year after their ACE procedures. Redo ACE procedures were performed in 4 patients, including 2 patients who initially had left-sided ACE procedures revised to right-sided procedures, 3 and 5 years later. The incidence of surgical complications did not differ significantly between the groups (32.0% [8/25] in group 1 vs 26.9% [7/26] in group 2; $P = .4753$; Table 2).

Table 3 summarizes the long-term complications in both groups. These complications included uncontrollable fecal soiling, sustained abdominal pain or discomfort unrelieved by ACE, reflux of fecal materials from stoma, and stenosis of the stoma. All long-term complications occurred within 2 years after ACE procedures. Fecal soiling was not controlled by ACE in 8 patients (32.0%) in group 1 and 1 patient (3.9%) in group 2 ($P = .0109$). The outcome of fecal continence was not significantly affected by the preoperative indication for ACE ($P = .7091$). Sustained abdominal pain or discomfort uncontrolled by ACE occurred in 7 patients (28.0%) in group 1 but none (0.0%) in group 2

($P = .0042$). Patients in group 1 also complained of intractable constipation with abdominal pain in spite of ACE. Among the 7 patients who had sustained abdominal pain or discomfort in group 1, 6 were evaluated with simple x-ray or barium enema studies, which demonstrated their fecal impaction. In contrast, although a few patients in group 2 had abdominal pain, it was generally transient and tolerable, occurred during the infusion of enema fluid, and resolved after the washout. Reflux of fecal materials from the stoma occurred in 13 patients (52.0%) in group 1 but none (0.0%) in group 2 ($P < .0001$).

Nonuse and infrequent use (approximately less than once in 1-2 weeks) of ACE occurred in 11 (44.0%) of 25 patients in group 1 and 0 (0.0%) of 26 patients in group 2 ($P < .0001$). Stomal repair or conversion to right-sided ACE was performed in some nonusers because of malfunctioning of the ACE, as indicated by reflux of fecal material or failure to resolve fecal soiling or constipation. Stomal repair was performed in 3 patients in group 1, 9 to 15 months after the initial procedure. Another 2 patients in group 1 underwent conversion to right-sided ACE 3 or 4 years after the initial procedure. Among the nonusers, 3 patients in group 1 did not perform the ACE, nor did they have the conduit repaired because of the difficulty associated with another surgery. Another 3 infrequent users in group 1 used the ACE along with a transanal enema less than once every 1 or 2 weeks to control intolerable abdominal discomfort or abdominal distension from the fecaloma.

Table 2 Incidence of surgical complications in groups 1 and 2

	Group 1, left-sided ACE (n = 25)	Group 2, right-sided ACE (n = 26)
Short-term complications		
Wound infection, n (%)	0 (0.0)	3 (11.5)
Intestinal obstruction, n (%)	1 (4.0)	1 (3.8)
Perforation or disruption of conduit, n (%)	1 (4.0)	1 (3.8)
Long-term complications requiring surgical revision		
Stricture resolved by revision, n (%)	3 (12.0)	1 (3.8)
Redo of ACE, n (%)	3 (12.0) ^a	1 (3.8)
Total, n (%)	8 (32.0)	7 (26.9)*

^a Two patients who initially had a left-sided ACE that was revised to a right-sided ACE a few years later are included.

* $P = .4753$.

Table 3 Incidence of long-term complications in groups 1 and 2

	Group 1, left-sided ACE (n = 25)	Group 2, right-sided ACE (n = 26)	P
Fecal soiling, n (%)	8 (32.0)	1 (3.9)	.0109
Sustained abdominal pain/discomfort unrelieved by ACE, n (%)	7 (28.0)	0 (0.0)	.0042
Reflux from stoma, n (%)	13 (52.0)	0 (0.0)	<.0001
Nonuse/infrequent ^a use of ACE, n (%)	11 (44.0)	0 (0.0)	<.0001
Stenosis of stoma, n (%)	6 (24.0)	13 (50.0)	.0549

^a Less than once every 1 to 2 weeks.

Stomal stenosis was reported by 6 patients (24.0%) in group 1 and 13 patients (50.0%) in group 2 ($P = .0549$). This improved with frequent dilatation with Hegar dilator or Foley catheter in all but 3 patients in group 1 and 1 patient in group 2, who underwent revision to resolve the stenosis (Table 2). The incidence of stomal stenosis was higher in the patients whose conduit was constructed from the appendix rather than the ileal, cecal, or sigmoid colonic flap (68.8% [11/16] with an appendiceal conduit vs 21.4% [6/28] with an ileal, cecal, or sigmoid colonic flap as the conduit; $P = .0021$).

The duration of the ACE, the volume of fluid used per ACE, and the interval between 2 ACEs were compared between the groups (Table 4). The mean time required to perform enemas was about 22 minutes longer for right-sided than for left-sided ACE (34.4 ± 12.4 minutes in group 1 vs 56.8 ± 28.9 minutes in group 2; $P = .0260$). The volume of enema fluid required for ACE was greater in group 2 (1092.9 ± 659.2 mL in group 1 vs 2602.3 ± 2006.9 mL in group 2; $P = .0020$). The mean interval between 2 subsequent ACEs did not differ significantly between the 2 groups (3.5 ± 3.4 days in group 1 vs 2.4 ± 1.5 days in group 2; $P = .3896$). These results on the interval between ACEs included the 3 patients in group 1 mentioned above who used ACE infrequently (ie, less than once every 1–2 weeks).

The telephone survey excluded 3 patients in group 1 who underwent ACE repair. Of the remaining 46 patients, 35 could be reached by telephone and provided responses. The responders included 17 patients in group 1 and 20 patients in group 2, including the 2 patients who underwent conversion to right-sided ACE and responded to the both left-sided and right-sided ACE. The degree of satisfaction with ACE was higher in group 2; the mean satisfaction scores were 2.93 ± 1.49 in group 1 and 4.23 ± 0.93 in group 2 ($P = .0015$).

3. Discussion

Intractable constipation and fecal incontinence uncontrolled by conservative measures are indications for an ACE procedure. Initially, Malone et al [1] described the right-sided ACE in 1990. They used the appendix as a conduit to deliver washouts for complete colonic emptying and the prevention of fecal soiling. Modifications to this Malone procedure for ACE were subsequently introduced [2,3]. The original Malone procedure and its modification for right-sided ACE were reported as satisfactory in controlling fecal incontinence and intractable constipation [4–6].

However, some problems were associated with performing the Malone ACE. A long time and large amounts of fluid were required to clean the whole colon, especially from the dilated right and transverse colons. Several authors introduced the left-sided ACE for the following reasons: to reduce the prolonged time for the right-sided ACE; to avoid the proximity of the ACE stoma to a possible Mitrofanoff stoma, used for continent bladder catheterization; to facilitate gravity-assisted evacuation; and to avoid the reabsorption of the enema fluid in the right and transverse colon of a large-volume capacity [7–10].

Between 1999 and 2001, right-sided ACE procedures were performed at our institution. After the left-sided ACE procedure was introduced in 2002, it was performed until 2006 at our institution [11,12]. Beginning in 2007, right-sided ACE procedures are now performed again because frequent complications occurred among our patients who underwent left-sided ACE. Thus, in this study, we were able to compare right- and left-sided procedures in a relatively large number of patients.

We found that right-sided ACE was superior to left-sided ACE in maintaining fecal continence, which was achieved in 96% of the right-sided group but in only 68% of the left-

Table 4 Characteristics of performance of ACEs

	Group 1, left-sided ACE (n = 15 ^a)	Group 2, right-sided ACE (n = 23 ^a)	P
Duration (min)	34.4 ± 12.4	56.8 ± 28.9	.0260
Amount of water for ACE (mL/enema)	1092.9 ± 659.2	2602.3 ± 2006.9	.0020
Interval (d)	3.5 ± 3.4	2.4 ± 1.5	.3896

^a Numbers of patients for whom ACE data were available in their medical records.

sided group. In both groups, patients and their parents were advised to optimize the frequency of ACEs and the volume of the washouts to clean the bowel and maintain fecal continence. This required daily performance of ACE in some patients in each group. The frequency of ACE did not differ significantly between the groups. As expected, the mean volume of fluid used for ACE was larger and the mean time required was longer in the right-sided group. This makes sense given the larger volume of colon involved in the right-sided group. Despite the greater fluid volume and time required, the patients or their caregivers in the right-sided group reported higher satisfaction scores. Fecal continence and relief of abdominal symptoms from constipation were considered the main factors affecting satisfaction.

The incidence of sustained abdominal pain or discomfort was significantly higher in the left-sided group (28%) and was usually caused by intractable constipation unrelieved by ACE. This was probably caused by fecal impaction in the proximal colon or poor adherence to the ACE regimen. In the right-sided group, a few patients complained of transient abdominal pain or discomfort during ACE. However, none of their abdominal pain could be attributed to unresolved fecal impaction. Given that the 2 main purposes of ACE are to achieve fecal continence and resolve intractable constipation, right-sided ACE was superior to left-sided ACE in this study.

The percentage of patients who stopped performing ACE was 44% in the left-sided group. Their chief reasons for abandoning the procedure were perianal fecal soiling uncontrolled by ACE and additional fecal soiling from the stoma (reflux of fecal material). They also experienced constipation unresolved by ACE. Two patients, who could not maintain left-sided ACE and underwent conversion to right-sided ACE, were satisfied with the revisions.

The left-sided ACE was initially evaluated in several small studies in which the authors concluded that the outcomes were similar to those described in earlier reports on the right-sided ACE. Liloku et al [7] reported that 5 of 7 patients who underwent the left Monti-Malone procedure on the descending colon achieved fecal continence with a significant shortening of the duration of the enema. Churchill et al [9] reported on 18 cases of the left Monti-Malone procedure on the splenic or sigmoid colon. They found that 55% (10/18) of the patients had complete fecal continence; 28% (5/18) had partial continence with occasional accidents or soiling; 11% (2/18) failed to maintain fecal continence with improvement only in constipation; and 2% (1/18) underwent stomal closure after a month. Calado et al [16] reported on 9 cases of the Macedo-Malone ACE procedure in which 8 patients had complete continence and 1 patient had partial continence.

A few other studies directly compared the outcomes of left- and right-sided ACE [13-15]. Two of these studies of similar sample sizes found similar outcomes with both procedures. Meyer et al [13] studied 9 left-sided ACE cases and 22 right-sided ACE cases, and Kim et al [14] studied 7 left-sided and 23 right-sided cases. The following outcome

measures did not differ significantly between the groups: incidence of surgical complications, outcomes of ACEs, patient satisfaction, and the time, frequency, and fluid volume required for ACEs. In another study, Sinha et al [15] reviewed the literature and reported fecal continence in 93.5% (complete in 72.0%; partial in 21.5%) of left-sided ACE cases and 94.1% (complete in 80.6%; partial in 13.5%) of right-sided ACE cases.

Our results in the current study differed from those of these previous studies. With a comparatively larger population including 25 left-sided cases and 26 right-sided cases, we found the long-term outcome of right-sided ACE to be superior in terms of fecal continence and relief of constipation. In the previous study of our initial series of left-sided ACE cases, fecal continence was achieved in 73.7% (14/19), with a median follow-up period of 23 months [11,12], which is a similar finding to the current result of 68% (17/25). The direct comparison of the results in a relatively large cohort made a difference in the long-term outcome between left-sided and right-sided ACE. Several recent studies of long-term outcomes in relatively large cohorts of patients who underwent right-sided ACE reported that fecal continence was achieved in 93% to 96% of cases [17-19], which agrees with our results.

The other problems associated with performing ACE are reflux of fecal materials from the stoma and stomal stenosis. In the left-sided ACE group, to prevent reflux of fecal materials, we performed Witzel-type antirefluxing seromuscular reinforcement without any antireflux procedure-related complications. However, 52% in the left-sided ACE group had stomal reflux, compared with no patient in the right-sided ACE group. This could be explained by the assumption that the native tendency of the appendix is to act as a valve, preventing reflux of fecal materials in right-sided ACE procedures. On the other hand, stomal stenosis occurred in only 24% of the left-sided group but 50% of the right-sided group, and it was even about 70% in those who had an appendiceal conduit rather than ileal or colonic flap. Most of the stomal stenosis could be resolved with daily dilation of the stoma without revision.

We evaluated long-term outcome indicators including fecal continence, control of constipation, and complications between right-sided ACE with cecostomy and left-sided ACE with sigmoidostomy. In this study, right-sided ACE was superior to left-sided ACE in achieving fecal continence and managing constipation. Right-sided ACE led to no complications of stomal reflux but frequent stomal stenosis. Despite the common incidence of stomal stenosis and the long time required to perform the procedure, patient satisfaction was higher for right-sided ACE. By contrast, 44% of patients in the left-sided group gave up the use of ACE because of uncontrolled fecal incontinence and constipation.

The limitations of this study were the retrospective design, performance of ACE procedures by multiple surgeons, the different study periods at which ACE procedures were performed (early and middle periods in

group 1 vs initial and late periods in group 2), and the size of the cohort. Studies in even larger cohorts are needed. The mean period of follow-up was also significantly longer in the left-sided ACE group (65 months) than in the right-sided group (33 months). However, this did not contribute significantly to the outcome differences because most of the complaints were presented in the first 2 years after the ACE procedures in both groups. Furthermore, it was impossible to compare right-sided ACE procedures to left-sided ACE procedures using the splenic flexure or descending colon, as described in some cases of previous small studies. This was because the left-sided ACE procedure with the splenic flexure or descending colon was not performed in this study. In addition, the left-sided ACE procedure could be an important utility in patients who cannot undergo a right-sided ACE because of other surgeries. The splenic flexure or descending colon could be used for those patients with fewer problems than in our left-sided group among whom we used the sigmoid colon as the stoma site.

Performing ACEs is significantly more challenging for patients and caregivers than conservative methods of bowel management, such as digital evacuation and retrograde enemas. In particular, ACEs require longer time to perform and stricter adherence. In addition, abdominal discomfort may occur during ACEs, and pain or stomal bleeding may result from catheter insertion. Nonetheless, ACE is the optimal choice when conservative methods fail to provide fecal continence or to control intractable constipation. In this study comparing long-term outcomes between right-sided and left-sided ACE using the sigmoid colon, we found that right-sided ACE, although potentially more demanding in terms of time, provided greater patient satisfaction than did left-sided ACE.

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