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Cecostomy in Children with Defecation Disorders

HAYAT M. MOUSA, MD,* MAARTJE M. VAN DEN BERG, MD,* DONNA A. CANIANO, MD,† MARK HOGAN, MD,‡ CARLO DI LORENZO, MD,* and JOHN HAYES, PhD§

Administration of antegrade enemas through a cecostomy is a therapeutic option for children with severe defecation disorders. The purpose of this study is to report our 4-year experience with the cecostomy procedure in 31 children with functional constipation ($n = 9$), Hirschsprung's disease ($n = 2$), imperforate anus ($n = 5$), spinal abnormalities ($n = 8$), and imperforate anus in combination with tethered spinal cord ($n = 7$). Data regarding complications, antegrade enemas used, symptoms, and quality of life were retrospectively obtained. Placement of cecostomy tubes was successful in 30 of 31 patients. Soiling episodes decreased significantly in children with functional constipation ($P = 0.01$), imperforate anus ($P < 0.01$), and spinal abnormalities ($P = 0.04$). Quality of life improved in patients with functional constipation and imperforate anus. No difference in complications was found between percutaneous and surgical placement. Use of antegrade enemas via cecostomy improved symptoms and quality of life in children with a variety of defecation disorders.

KEY WORDS: antegrade enema; fecal incontinence; constipation; follow-up; imperforate anus.

Constipation and fecal incontinence affect several groups of patients, in particular, those with gastrointestinal neuromuscular disorders, such as anorectal and spinal anomalies (1). In these children and in a subgroup of children with functional constipation, intensive medical and/or psychosocial treatment may not always result in successful clinical outcome. In patients with imperforate anus the combination of impaired function of the anal sphincters, poor reservoir function, and colonic contractions propagating into the neorectum makes treatment of fecal incontinence particularly challenging (1).

The antegrade continence enema (ACE) has been reported as a therapeutic option for defecation disorders when maximal conventional therapy is not successful. This surgical procedure enables antegrade delivery of an enema to clean out the colon at regular intervals, avoiding accumulation of feces and reducing soiling episodes. Since the preliminary report in 1990 by Malone *et al.* (2), the ACE has been used increasingly in children with spinal abnormalities, Hirschsprung's disease, and functional constipation. Less information is available about the benefit of using a cecostomy in children with imperforate anus.

The original surgical procedure, regarding the use of the appendix as conduit for Malone antegrade continence enema (MACE), has undergone many technical modifications to improve outcome in the management of fecal incontinence (3). Recently, use of a percutaneously inserted cecostomy tube has been proposed for the management of defecation disorders (4). Compared to the MACE, significantly fewer complications were reported after cecostomy placement (5).

The purpose of this study is to report our 4-year experience with two different techniques of the cecostomy procedure and to compare the clinical outcome of cecostomy in

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From the Divisions of *Pediatric Gastroenterology, †Pediatric Surgery, and ‡Radiology, Columbus Children's Hospital, and §Division of Biostatistics, Ohio State University College of Medicine and Public Health, Columbus, Ohio, USA.

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Address for reprint requests: H. Mousa, MD, FAAP, Columbus Children's Hospital, 700 Children's Drive, ED 426, Columbus, Ohio 43205, USA; mousah@pediatrics.ohio-state.edu.

children with defection disorders secondary to functional constipation, imperforate anus, and spinal abnormalities.

METHODS

Patient Population. Between 1999 and 2003, 31 children (58% boys) received a cecostomy at Columbus Children's Hospital. The median age at the time of cecostomy placement was 8 (range, 3–18) years, with a median weight of 21 (range, 12–105) kg. The indications for creation of the cecostomy were constipation ($n = 7$), fecal soiling ($n = 9$), or a combination of both ($n = 15$). Underlying conditions included functional constipation ($n = 9$), Hirschsprung's disease ($n = 2$), imperforate anus ($n = 5$), imperforate anus combined with tethered spinal cord ($n = 7$) and spinal abnormalities ($n = 8$). Spinal anomalies included tethered cord and myelomeningocele.

Placement of Cecostomy. After a colonic cleanout, perioperative parenteral antibiotics (10 mg/kg metronidazole, 2.5 mg/kg gentamicin, 20 mg/kg ampicillin) were given. The placement of the cecostomy tube was performed percutaneously by an interventional radiologist in 16 patients. An open surgical approach was used in the other 15 patients. The following tubes were inserted; Chait (n =8; Cook Inc., Bloomington, IN) (6), MIC-Key ($n = 11$; Medical Innovations Corp., UT), and Corpak ($n = 2$; Viasys Healthcare Medsystems, Wheeling, IL). The decision to proceed with either the surgical or the radiologic placement of the cecostomy was based on parental preference and on the feasibility of the radiologic approach, with no loops of bowel overlying the cecum on a barium enema.

Percutaneous Approach. The percutaneous placement was performed as previously described (4). An abdominal ultrasound was used to identify the organs and vessels which are interposed between the expected puncture site and the cecum as well as the position of the inferior epigastric vessels. An enema tip was placed in the rectum, and air instilled to identify the cecum. Glucagon was administered intravenously for bowel paralysis. A 21-gauge needle was advanced into the cecum with fluoroscopic guidance. After advancing a sheath, suture anchors were deployed to secure the cecum. Standard techniques were then used to advance an 8-Fr drain into the cecum. The catheter was secured using the sutures from the anchors, which were tied through two holes made in a Molnar disk attached to the catheter.

Surgical Approach. The surgical approach was used in most patients with a right lower quadrant incision overlying the cecum. A small incision was placed in the anterior cecum after a purse string suture secured the full-thickness bowel wall. The catheter was inserted into the cecum from a separate stab wound, and the bowel was affixed to the anterior abdominal wall with sutures.

Intravenous antibiotics were continued for 48 hr after the surgical procedure. The patients were allowed to drink when bowel sounds returned, and the diet was advanced as tolerated. Oral metronidazole was continued for 7 days after discharge.

To secure patency, the tube was irrigated twice daily with 10 ml water until the antegrade irrigation was started 2 weeks after the procedure.

Clinical Outcome. Outcome measures were obtained by interviewing the parents to determine (1) complications, (2) type of antegrade enemas used, (3) effectiveness of treatment, and (4) quality of life. A standardized questionnaire was used to obtain data about the time taken to evacuation after antegrade washout, the type and volume of enema solution used, and the child's health status before and after the cecostomy placement. Prececostomy data were obtained retrospectively during the same interview. Bowel movement and fecal soiling frequency, number of medications, number of physician visits related to defecation problems, number of hospital admissions for disimpaction, number of missed school days per month, and quality of life were recorded. Frequency of bowel movements was scored as follows: 1 =< 5 bowel movements per week, 2 = 5/week to 3/day, and 3 => 3/day. Soiling frequency was scored as 1 = none, 2 = occasional, 3 = few episodes per week, 4 = few episodes per week to daily, and 5 = constantly. Quality of life was assessed by scoring limitations of activity (none, mild, moderate, and severe), global health score, and global emotional score (poor, fair, good, very good, and excellent).

Statistical Analysis. Outcome measures were analyzed in all the patients and in three subgroups: functional constipation ($n = 9$), imperforate anus ($n = 12$), and spinal abnormalities ($n = 8$). The patients with imperforate anus in combination with tethered cord were included in the group of imperforate anus. Baseline characteristics were analyzed in a descriptive way. The Mann–Whitney U and χ^2 tests were used to examine the relation between the type of cecostomy procedure and tube and the presence of complications. Wilcoxon nonparametric test was used to analyze the difference of the median of clinical factors before and after cecostomy placement. A P value of <0.05 was considered significant.

This study was approved by the Institutional Review Board of Columbus Children's Hospital.

RESULTS

Prececostomy Interventions. In all patients a maximal medical treatment, including a high dose of different oral laxatives, retrograde enemas, suppositories, and cleanouts, had failed. Diagnostic evaluation included anorectal manometry in 90%, magnetic resonance imaging (MRI) of the spine in 84%, rectal biopsy in 32%, contrast enema study in 94%, and colonic manometry in 84% of all patients. In patients with imperforate anus, anorectal manometry revealed a median anal resting pressure of 24 (range, 13–41) mm Hg.

Twelve of 13 children with a tethered spinal cord had undergone detethering surgery prior to cecostomy placement, without improvement of the defecation disorders. Tethered cord was identified as lipoma or low conus on MRI. Detethering surgery included laminotomy with the section involving filum terminale to release the spinal cord followed by reconstruction of lamina to avoid orthopedic deformity.

Antegrade Enema. The cecostomy was successfully placed in 30 of 31 patients. One patient needed surgical exploration immediately after percutaneous placement of the tube due to dilatation of the tract and pneumoperitoneum. A second cecostomy tube was then successfully placed surgically. The enemas were administered on a

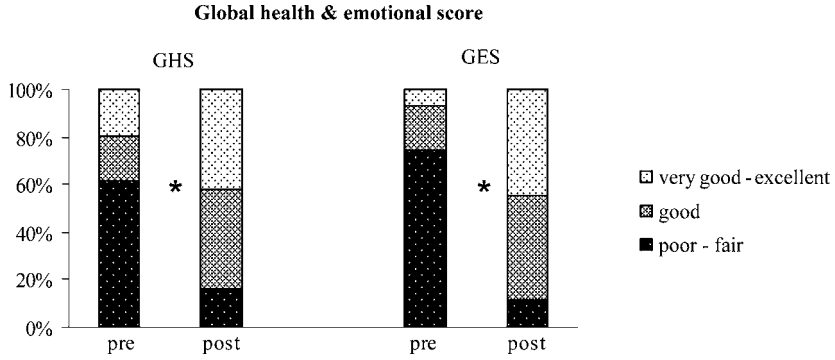

Fig 1. Global health score (GHS) and global emotional score (GES) pre- and postcecostomy; $N = 31$. $^*P < 0.01$.

daily basis in 25 of 31 children. Phosphate solutions (median, 0.5–1.0 ml/kg) followed by normal saline (median, 10–15 ml/kg) were used in most children (19/31). Other colonic irrigants were polyethylene glycol, mineral oil, saline, or bisacodyl diluted with saline. Complete evacuation of stools with antegrade enemas was achieved in 94% of the children within 60 min from the administration of the antegrade enemas.

Clinical Outcome in General Population. The median time of follow-up was 11 (range, 1 to 46) months after the cecostomy placement. Median episodes of fecal soiling decreased from constant soiling before cecostomy to no soiling postcecostomy ($P < 0.001$). The bowel movement frequency improved significantly, from a median of <5/week to 5/week–3/day at the time of follow-up ($P = 0.03$). No patient required admission for disimpaction. Health care use decreased from a median of 5 to 2 visits/year ($P < 0.001$), and number of medications from three to one per day ($P < 0.001$).

Quality of Life in the General Population. Global health score and global emotional score for all subjects are shown in Figure 1. Before cecostomy placement approximately 60% of the patients reported their global health as poor to fair. With the use of antegrade enema, this percentage decreased to 19% ($P < 0.01$). The median global emotional score improved from fair to good ($P < 0.01$). Limitations in activity due to soiling decreased significantly, from a median of moderate to none ($P < 0.01$). The percentage of children without activity limitations increased from 26% to 58% ($P < 0.05$) (Figure 2).

Clinical Outcome in Subgroups. We divided the patient population into three subgroups. Table 1 presents baseline characteristics and clinical outcome in children with functional constipation, imperforate anus, and spinal abnormalities before and during the use of antegrade enemas. In all groups, episodes of soiling, number of medications, and physician visits were significantly decreased. The number of patients with imperforate anus who reported constant soiling, decreased from 7 of 12 prececostomy to 0 of 12 postcecostomy ($P < 0.01$). In the group of functional constipation the frequency of bowel movements was significantly increased at the time of follow-up ($P < 0.01$). Children with imperforate anus had a normal frequency of bowel movements prior to cecostomy placement and no difference was found postcecostomy. In each

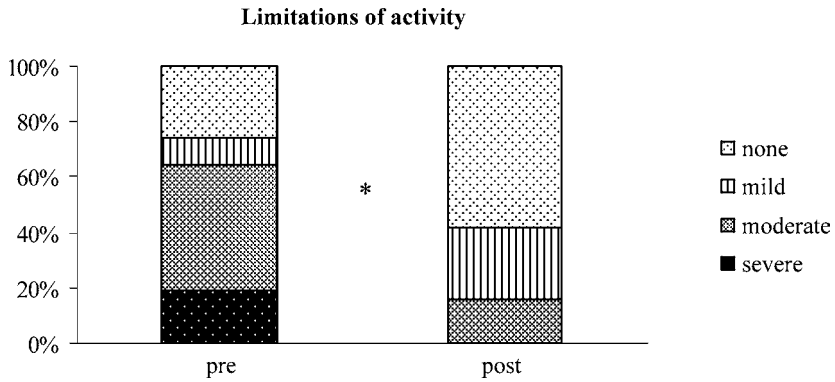

Fig 2. Limitations of activity pre- and postcecostomy placement; $N = 31$. $^*P < 0.01$.

TABLE 1. CLINICAL OUTCOME PRE- AND POSTCECOSTOMY

	Constipation (n = *9*)			*Imperforate anus* (n = *12*)			*Spinal abnormalities* (n = *8*)		
Onset of symptoms (number)	Since birth (5 of 9)			Since colostomy closure (12 of 12)			Since birth (4 of 8)		
Age at cecostomy placement (years)	12 (range, 3–16)			5 (range, 3–11)			9 (range, 4–18)		
Time of follow-up (months)	11 (range, 1–45)			11 (range, 1–27)			13 (range, 4–46)		
	Pre	*Post*	P *value*	*Pre*	*Post*	P *value*	*Pre*	*Post*	P *value*
Bowel movement frequency	<5/wk	5/wk–3/day	<0.01	5/wk–3/day	5/wk–3/day	n.s.	<5/wk	5/wk–3/day	n.s.
Soiling frequency	Constant	None	0.01	Constant	Occasional	<0.01	Occasional	None	0.04
Number of medications	4	1	0.01	2	0	<0.01	3	1	0.02
Number of physician visits per year	6	2	<0.01	4	2	0.05	4	1	0.04
Number of admissions for disimpaction	4	0	<0.01	1	0	0.04	1	0	n.s.
Number of missed school days per month	8–10	1–3	n.s.	8–10	1–3	n.s.	11–13	1–3	n.s.
Global health score	Poor	Good	0.01	Fair	Good	0.03	Fair	Good	n.s.
Global emotional score	Poor	Good	0.01	Fair	Good	0.01	Fair	Good	n.s.
Limitations of activity	Moderate	Mild	<0.01	Moderate	None	0.02	Mild	None	n.s.

Note. Baseline characteristics and clinical outcome pre- and postcecostomy in children with functional constipation, imperforate anus, and spinal abnormalities. All values are reported as median, and differences between medians are reported as *P* value (>0.05, not significant; n.s.).

group, children missed fewer school days but without a statistically significant difference.

Quality of Life in Subgroups. Global health score and global emotional score for different subgroups are reported in Table 1. In the groups of constipation and imperforate anus, global health and emotional score improved significantly with the use of antegrade enemas. Limitations of activity due to defecation disorders were also reduced significantly in these patient groups. In children with spinal abnormality, quality of life scores were not significantly improved postcecostomy.

Complications. Complications were reported in a total of 26 patients (84%). These complications included leakage around the tube (42%), dislodged tubes (26%), and granulation tissue at the site (68%). Sixteen children had two or more complications. No major complications such as perforation, stoma stenosis, or stoma prolapse occurred. No difference was found in occurrence or number of complications between percutaneous placement and surgical intervention. No difference regarding complications was found between MIC-Key and Chait tubes.

Approximately half of the patients reported pain during irrigation. With the use of phosphate solution, pain during irrigation occurred in 11 of 19 children. Three children used only normal saline; no pain during irrigation was reported in these children. Except for normal saline, no difference was found among the different enema solutions in the occurrence of abdominal pain.

DISCUSSION

This study confirms that the use of antegrade enemas through a cecostomy represents an effective means to treat fecal incontinence and/or constipation refractory to medical management. Symptoms of defecation disorders in children with different underlying etiologies improved significantly after a cecostomy was created. In addition, there was a benefit on the patients' physical activity, health-care use, and general well-being.

We subdivided our patient population into three groups: children with functional constipation, imperforate anus, and spinal abnormalities. All children with a combination of imperforate anus and tethered spinal cord underwent detethering surgery prior to cecostomy placement. These children were included in the imperforate anus group because we considered the altered anorectal anatomy due to anorectal abnormalities more important in the pathophysiology of fecal incontinence than the possible impaired rectal function secondary to spinal abnormalities. Defecation problems did not improve after detethering surgery in our children. This finding is of limited value since we might be dealing with a selective patient group, in which detethering surgery failed and therefore a cecostomy was indicated in these patients. Rosen *et al.* reported a high success rate of detethering surgery in constipated children with spinal abnormalities detected by spinal MRI (7).

Many children with a surgically corrected anorectal malformation continue to suffer from fecal incontinence (9), resulting in limited social interactions and impaired development of self-esteem (10). These patients may experience psychiatric and psychosocial dysfunction into puberty and adulthood (11). In order to prevent such consequences it is desirable to shorten the duration and to decrease the severity of their abnormal defecation. Lee at al. reported a favorable outcome using a cecostomy in infants with imperforate anus (12). In our study the soiling frequency decreased significantly in this category of patients. Also, the general health and emotional score improved dramatically, emphasizing that children with imperforated anus can be efficiently managed with regular antegrade colonic washouts through a cecostomy.

Antegrade enemas in the children with functional constipation resulted in a significant increase in defecation frequency, reduction of soiling frequency, and improvement of quality of life. A previous study on cecostomy placement in 12 patients with severe functional constipation revealed a similar outcome (8). Curry *et al.* reported a lower success rate (39%) after ACE placement in patients with functional constipation, compared to 73% for all other underlying conditions (13). The more favorable outcome in our series and the series of Youssef *et al.* (8) is most likely related to careful patient selection and prececostomy evaluation. Colonic manometry was used in all children with functional constipation to exclude previously undiagnosed intestinal neuromuscular disorders (14) and to determine the response to a colonic stimulant such as bisacodyl. A normal response to provocative testing in the proximal colon might predict a favorable response to the use of antegrade enemas. Further clinical research is required to substantiate the predictive value of colonic manometry on the outcome of cecostomy.

Although the study by Youssef *et al.* (8) evaluated a comparable patient group with functional constipation, an advantage of our study is that patients with imperforate anus and spinal abnormalities were included. Another difference from that study is the technique of placement, which was surgically in 11 and percutaneously in 1 patient. In our study we compared these different techniques in terms of outcome and complication rates.

A positive outcome of the use of antegrade enemas in patients with spinal abnormalities was previously reported, with a success rate of approximately 70% (13, 15). Our data in these patients showed a significant decrease

in fecal incontinence. Quality of life in this patient group was less obviously improved than in the patients with functional constipation and imperforate anus. This is probably due to more comorbidities and physical handicaps existing in children with spinal abnormalities.

In accordance with previous observations, the majority of patients achieved bowel emptying within 60 min after irrigation (5, 16, 17). Different irrigation solutions have been used as antegrade enemas. A phosphate solution followed by normal saline was effectively used in the majority of our patients without symptoms of phosphate toxicity. Data about the use and complications of phosphate solutions have been controversial in the literature. Although there are concerns about phosphate toxicity (13) and colonic ulcers (18), phosphate solutions have been reported to be safe and effective for colonic cleanouts in children .(19). We did not document electrolyte levels in our study patients. During irrigation abdominal pain occurred in 50% of our patients, confirming what has been reported previously (13). No patient had persistent abdominal pain; therefore suspicion of colonic ulcerations did not rise in any of our patients. The pain during irrigation was not related to the type of enema solution used, as it occurred in approximately 50% of the patients using each different solution. Only during the use of saline no abdominal pain was reported. Since the number of patients receiving each type of solution was small in our series, we cannot assess which irrigation regimen is optimal. It is reported that tap water enemas are safe for the use of antegrade enemas in children, without increased risk of water intoxication (20). One can speculate that saline might be less painful than irritant solutions such as phosphate and bisacodyl. Larger controlled studies are, however, required to evaluate the efficacy, safety, and cost effectiveness of different irrigation regimens.

Many modifications have been made to the original ACE procedure. These include variations on the technique of creating the cecostomy and appendicostomy, use of an antireflux procedure (16), open placement of a cecostomy tube (3), endoscopic placement (21), and percutaneous placement of a cecostomy tube (4). With the latter there seemed to be fewer complications requiring operative intervention than in patients with MACE (5). In one of our patients, the percutaneous cecostomy placement failed and a surgical placement was required. No other major complications occurred in our study, but minor complications were common. These problems were easily managed in most patients. No significant difference in the occurrence and numbers of complications between surgical approach and percutaneous placement was found in our study. The different cecostomy tubes used in this study had similar complication rates.

In summary, the cecostomy procedure for antegrade irrigation of the colon is safe and helpful in the management of intractable constipation and fecal incontinence in children with different underlying etiologies, including imperforate anus. Cecostomy tubes can be placed successfully by either surgical or percutaneous intervention, with similar complication rate.

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