Journal of Pediatric Surgery xxx (xxxx) xxx–xxx

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

A comparison of Malone appendicostomy and cecostomy for antegrade access as adjuncts to a bowel management program for patients with functional constipation or fecal incontinence☆,☆☆

Devin R. Halleran [a,*], Alejandra Vilanova-Sanchez [a], Rebecca M. Rentea [a], Mana H. Vriesman [a], Tassiana Maloof [a], Peter L. Lu [a], Amanda Onwuka [b], Laura Weaver [a], Karla KH Vaz [a], Desale Yacob [a], Carlo Di Lorenzo [a], Marc A. Levitt [a], Richard J. Wood [a]

[a] *Center for Colorectal and Pelvic Reconstruction, Nationwide Children's Hospital, Columbus, OH*
[b] *Center for Surgical Outcomes Research, Nationwide Children's Hospital, Columbus, OH*

ARTICLE INFO

Article history:
Received 21 September 2018
Accepted 1 October 2018
Available online xxxx

Key words:
Malone appendicostomy
Neoappendicostomy
Cecostomy
Antegrade enema
Bowel management
Fecal incontinence
Constipation

ABSTRACT

Background: Appendicostomy and cecostomy are two approaches for antegrade enema access for children with severe constipation or fecal incontinence as adjuncts to a mechanical bowel management program. Each technique is associated with a unique set of complications. The purpose of our study was to report the rates of various complications associated with antegrade enema access techniques to help guide which option a clinician offers to their patients.
Methods: We reviewed all patients in our Center who received an appendicostomy or cecostomy from 2014 to 2017 who were participants in our bowel management program.
Results: 204 patients underwent an antegrade access procedure (150 appendicostomies and 54 cecostomies). Skin-level leakage (3% vs. 22%) and wound infections (7% vs. 28%) occurred less frequently in patients with appendicostomy compared to cecostomy. Nineteen (13%) appendicostomies required revision for stenosis, 4 (3%) for mucosal prolapse, and 1 (1%) for leakage. The rates of stenosis (33 vs. 12%) and wound infection (13 vs. 6%) were higher in patients who received a neoappendicostomy compared to an in situ appendicostomy. Intervention was needed in 19 (35%) cecostomy patients, 15 (28%) for an inability to flush or a dislodged tube, and 5 for major complications including intraperitoneal spillage in 4 (7%) and 1 (2%) for a tube misplaced in the ileum, all occurring in patients with a percutaneously placed cecostomy. One appendicostomy (1%) patient required laparoscopic revision after the appendicostomy detached from the skin.
Conclusion: Patients had a lower rate of minor and major complications after appendicostomy compared to cecostomy. The unique complication profile of each technique should be considered for patients needing these procedures as an adjunct to their care for constipation or fecal incontinence.
Type of study: Retrospective comparative study.
Level of evidence: Level III.

Constipation and fecal soiling are common problems in the pediatric population and are prevalent in surgical patients following reconstruction for anorectal malformations (ARMs) and Hirschsprung disease (HD), as well as in patients with spina bifida and severe functional constipation [1–3]. The majority of patients can be successfully managed through dietary modifications or the use of stimulant laxatives, however there remains a subset of patient who fail to improve with these conservative measures and require a mechanical colonic wash out to achieve social continence.

The concept of utilizing either the appendix or the cecum to access the large intestine and give an antegrade flush to treat colonic disease was first described in 1906, when Dr. Holton Curl used colonic access for "irrigation of the colon with quinine solution" to treat amebic

☆ Author Contribution:Study conception and design: DRH, AVS, RMR, MHV, PLL, KKV, DY, CDL, MAL, RJWAcquisition of data: DRH, AVS, RMR, MHV, TM, LWAnalysis and interpretation of data: DRH, AVS, RMR, PLL, AO, KKV, DY, CDL, MAL, RJWDrafting of manuscript: DRH, AVS, RMR, MHV, TM, PLL, AOCritical revision of manuscript: DRH, AVS, RMR, MHV, TM, PLL, AO, LW, KKV, DY, CDL, MAL, RJW.
☆☆ How this paper will improve care: Our institutional experience with Malone appendicostomy and cecostomy demonstrates that the rate of both minor and major complications is higher after cecostomy, and the unique complication profiles of both should be considered for each individual patient.

* Corresponding author at: Center for Colorectal and Pelvic Reconstruction, Nationwide Children's Hospital, 700 Children's Drive, FOB 6B.2, Columbus, OH 43205. Tel.: +1 614 722 3881; fax: +1 614 722 3903.
E-mail address: devin.halleran@nationwidechildrens.org (D.R. Halleran).

https://doi.org/10.1016/j.jpedsurg.2018.10.008
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dysentery [4]. In the latter part of the 20th century, this concept was applied to the care of constipation and fecal incontinence with the description of the Malone antegrade continence enema [5] and the percutaneous cecostomy [6]. By providing an antegrade route for enema delivery, patients can self-administer enema flushes, improving their compliance and quality of life [7–10].

Appendicostomy and cecostomy are each associated with a unique complication profile, and no precise strategy exists to determine which procedure best suits each individual patient [11]. As such, the decision to perform either procedure is often based on patient preference and provider comfort. With this in mind, we aimed to review our institution's experience and report outcomes following appendicostomy and cecostomy for patients with medically unmanageable constipation and/or fecal incontinence.

1. Methods

We retrospectively reviewed all patients in our Center who received a Malone appendicostomy, neoappendicostomy, or cecostomy, either as an isolated procedure or in combination with additional intraabdominal surgery, from April 2014 to April 2017 as part of their treatment plan to manage severe constipation or fecal incontinence. Demographic information including age, gender, indication for antegrade enema access, surgical technique, perioperative complications, and need for any subsequent intervention was recorded. Patients were then divided into an appendicostomy or cecostomy group, depending on the procedure performed. The study was approved by the Institutional Review Board.

1.1. Technique

Our technique for performing in situ appendicostomy involves a 360° cecal plication around the base of the appendix taking care to protect the blood supply [Fig. 1] [12,13]. In patients needing urinary diversion, a split appendix technique was used [Fig. 2] [14]. If there was no appendix or the appendix was used for a Mitrofanoff, a neoappendicostomy was performed using a colonic (cecal) flap [Fig. 3] [12,15]. Appendicostomies were anastomosed to the skin either at the umbilicus [Fig. 4a] or in the right or left lower quadrants as dictated by the patient's anatomy and whether a Mitrofanoff was also needed (in such a case, the Mitrofanoff was placed in the umbilicus and the Malone in the right lower quadrant). These were performed as isolated procedures or in conjunction with other procedures depending on the reconstructive needs of the patients. Flushes were started once the child was advanced to a regular diet. For neoappendicostomies, half volume flushes were used for one month and then advanced to full volume. All patients were left with an indwelling 8 French tube for one month, at which point catheterization was taught, and then an ACE stopper was utilized for 6 months after appendicostomy. Cecostomy (with Chait tube) was performed using either an open, laparoscopic, or percutaneous approach and exteriorized through the abdominal wall in the right lower quadrant [Fig. 4b]. In cases of surgically-placed cecostomy tubes, the cecum was fixed to the anterior abdominal wall with 2–4 absorbable sutures. For percutaneous cecostomies, T-fasteners were used.

1.2. Statistical analysis

To assess the relationship between procedure type and key results, we performed Wilcoxon–Mann–Whitney and Kruskal–Wallis tests for continuous nonparametric outcomes, and for binary outcomes, we employed Chi-squared and Fisher's exact tests depending on the sample size. We assessed the distribution of age, gender, primary diagnoses, and surgical technique by procedure type. For the analysis of length of stay, we excluded combined procedures, which accounted for 112 of the 150 (75%) patients in the appendicostomy group and 18 of the 54 (33%) patients in the cecostomy group, and assessed average length of stay by procedure. We also assessed how key surgical outcomes and complications varied by Malone technique (in situ, cecal flap, split appendix), location of Malone (umbilicus vs. right or left lower quadrant), and antegrade access technique (surgical or percutaneous). Statistical analysis was performed using SAS 9.4 (SAS Institute Inc., Cary, NC).

2. Results

There were 204 patients who underwent an antegrade access procedure to administer their bowel regimen over the study period. 150 (74%) patients had an appendicostomy and 54 (26%) received a cecostomy. The mean duration of follow up was similar between the appendicostomy group (1.6 years, range 0.3–3.3) and the cecostomy group (1.7 years, range 0.3–3.2), and in all cases was at least 3 months. A summary of the patients' demographic information can be found in Table 1. Overall, patients who underwent appendicostomy were younger (7.6 years, range 2.4–31.0) than those who received a cecostomy (12.2 years, range 2.4–35.6) ($p = 0.0006$). Patients were more likely to have an appendicostomy as part of a combined intraabdominal procedure than patients who had a cecostomy (112/150, 75% vs. 18/54, 33%) ($p < 0.0001$). Patients with an anorectal malformation (ARM) (77/150, 51% vs. 3/54, 6%, $p < 0.0001$) and Hirschsprung disease (HD) (22/150, 15% vs. 3/54, 5.6%, $p = 0.11$) were more likely to have an appendicostomy, whereas patients with myelomeningocele (12/150, 8% vs. 25/54, 46%, $p < 0.0001$) or functional constipation (30/150, 20% vs. 19/54, 35%, $p = 0.04$) were more likely to receive a cecostomy. This pattern was mainly based on referral patterns within

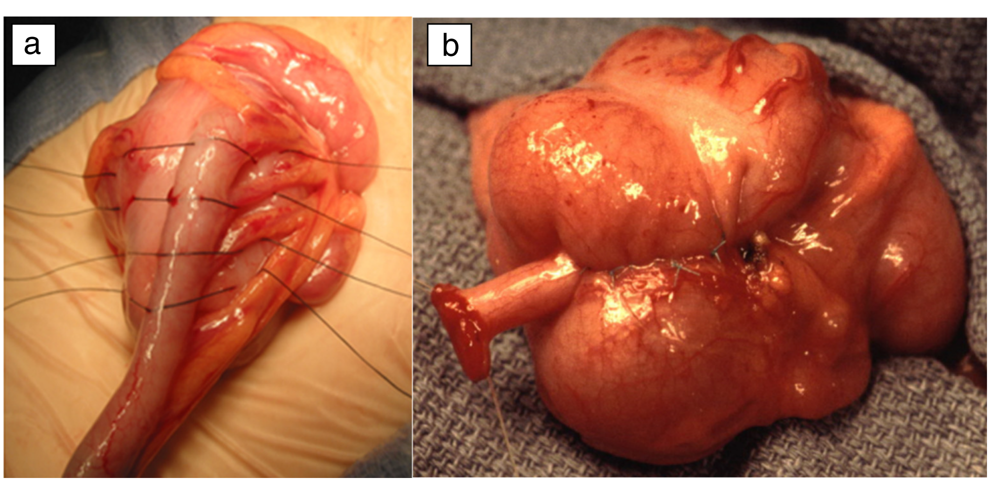

Fig. 1. 360° plication is planned around the base of the appendix (a) which avoids interrupting the blood supply. The completed plication demonstrating the valve mechanism (b).

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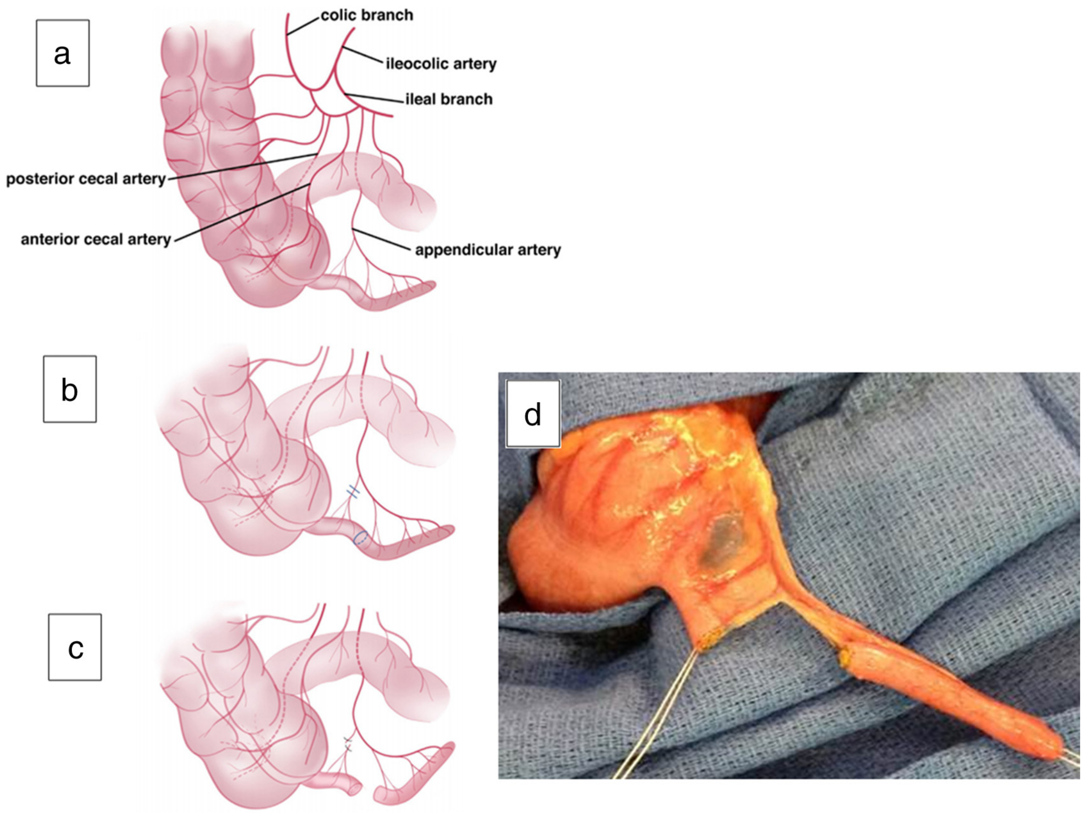

Fig. 2. Illustration of the split-appendix technique. The normal anatomy is illustrated in (a). The appendix is sharply divided (b) with the proximal end serving as the appendicostomy and the distal end used for the appendicovesicostomy. An intraoperative image of the appendix after division is shown in (d).

our institution. For appendicostomies it was primarily the surgical team conducting a bowel management program who identified those patients who would benefit from the procedure and the same team preferentially performed appendicostomies. For cecostomies it was the GI/motility team who referred the patient to interventional radiology. After the initial launch of our colorectal/motility collaborative program, the trend has been towards more patients being referred for appendicostomy.

Patients who underwent appendicostomy placement had a significantly lower length of stay compared to patients who received a cecostomy (3.0 days vs. 7.1 days, range 2–50, $p = 0.001$), which related to the higher incidence of complications in the cecostomy group. A comparison of complications seen in both groups is shown in Table 2. Patients were significantly less likely to experience skin leakage (4/150, 3% vs. 12/54, 22%, $p = 0.0001$) and wound infections (10/150, 7% vs. 15/54, 28%, $p < 0.0001$) after appendicostomy compared to cecostomy. One of the four patients with leakage after appendicostomy required surgical revision to tighten the plication; the remaining 3 resolved spontaneously. An intraabdominal intervention was required on postoperative day 24 owing to detachment of the appendix from the umbilicus in 1 patient (1%) after appendicostomy (which was managed using a laparoscopic approach) compared to 4 (7%) with cecal detachment after

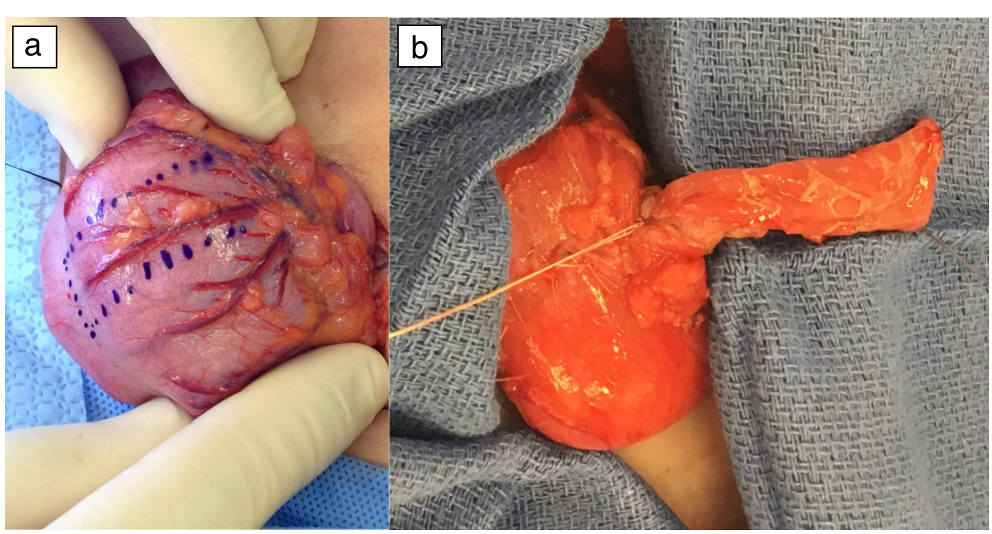

Fig. 3. Creation of a neoappendicostomy using a cecal flap. A U-shaped flap of cecum is incised over a single vessel (a) and tubularized over a feeding tube (b).

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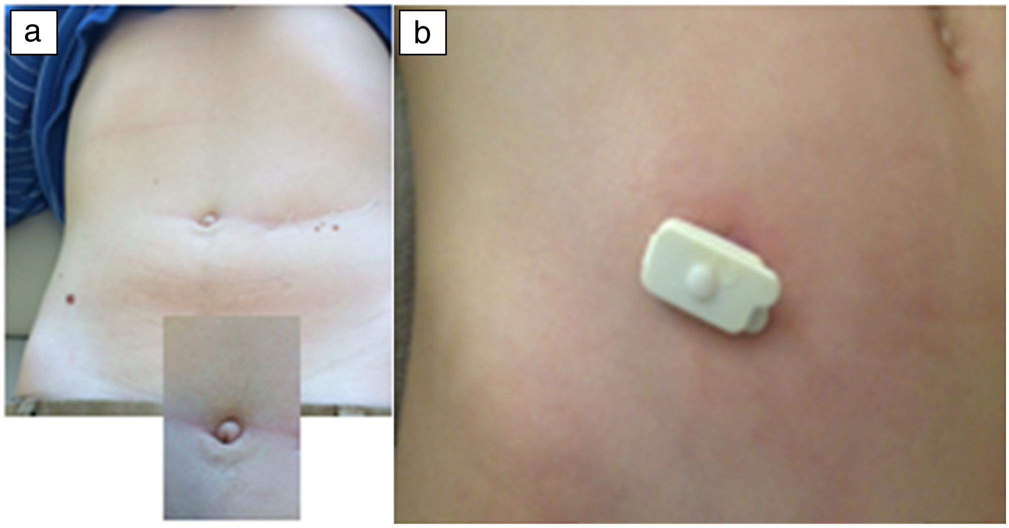

Fig. 4. External view of the (a) Malone appendicostomy at the umbilicus and (b) cecostomy (Chait) device in the right lower quadrant.

cecostomy ($p = 0.02$). All 4 of these patients required a laparotomy owing to fecal spillage. This complication was identified at a mean of 2 days (range 1–3) postoperatively. One patient (2%) required surgical revision after inadvertent placement of the cecostomy tube in the ileum compared to no patients (0%) with a malpositioned appendicostomy tube. Three patients (6%) with cecostomy underwent elective conversion to appendicostomy at the request of the patient or family, all for complaints of skin irritation. Overall, the age of patients who experienced a complication of any type was similar to those who did not experience a complication in both the appendicostomy (7.9 years vs. 7.6 years, $p = 0.88$) and appendicostomy (12.0 vs. 12.4, $p = 0.74$) group.

Malone orifice stenosis and mucosal prolapse requiring surgical intervention were seen in 19 (13%) and 4 (3%) patients, respectively, after appendicostomy. Five (31%) patients who developed stenosis had failed to perform the recommended daily catheterization or keep the Malone plug in place between catheterizations. Of patients who developed a stenosis, 10 (52%) experienced this complication within the first year after placement, while 6 (32%) and 3 (16%) patients had an intervention for stenosis within the 2nd and 3rd year, respectively.

Unplanned tube exchanges were required in 19 (35%) cecostomy patients for an inability to flush the cecostomy tube and in 15 (28%) patients after the tube fell out. After cecostomy, patients received an average of 3.8 (range 1–13) routine tube exchanges over the study period, and 34 (63%) patients underwent at least 1 unplanned procedure for a tube exchange.

Table 3 demonstrates the rate of complication by location of the appendicostomy. The umbilicus was the most common site for appendicostomy, used in 125 (83%) patients. The right lower quadrant, right upper quadrant, and left lower quadrant were used in 23 (15%), 1 (1%), and 1 (1%) patients, respectively. The rate of skin leakage, prolapse, stenosis, or wound infection did not vary by the location of the appendicostomy. Among patients who underwent appendicostomy, 121 (81%) underwent in situ appendicostomy using their native appendix, 13 (9%) underwent a split appendix technique, 15 (10%) had a neoappendicostomy using tubularized cecum, and 1 (1%) had a Monti channel using small bowel (12%) or split appendix (0/13, 0%) ($p = 0.03$). The rate of stenosis and wound infection was higher in patients who received a neoappendicostomy, while the rate of leakage and prolapse did not vary by the type of appendicostomy (in situ, split, or neoappendix) (Table 4).

Table 5 demonstrates the rate of complication by cecostomy technique. There were minor (skin leakage, inability to flush the tube, tube dislodgement, and wound infections) and major (intraabdominal leakage and misplaced tubes) complications associated with percutaneous

Table 1
Cohort demographics.

	Appendicostomy (n = 150)	Cecostomy (n = 54)	P
Age, years (mean, range)	7.6 (2.4–31.0)	12.2 (2.4–35.6)	0.0006
Male (%)	77 (51%)	21 (39%)	0.12
Mean duration of follow-up, years (range)	1.6 (0.3–3.3)	1.7 (0.3–3.2)	
Combined Procedure	112 (75%)	18 (33%)	<0.0001
Diagnosis			
Anorectal malformation	77 (51%)	3 (6%)	<0.0001
Functional constipation	30 (20%)	19 (35%)	0.04
Hirschsprung disease	22 (15%)	3 (6%)	0.11
Myelomeningocele	12 (8%)	25 (46%)	<0.0001
Tethered cord	3 (2%)	1 (2%)	0.95
Caudal regression	1 (1%)	0 (0%)	0.55
Cerebral palsy	2 (1%)	0 (0%)	0.43
Colonic dysmotility	1 (1%)	0 (0%)	0.55
Acute lymphoblastic leukemia	1 (1%)	0 (0%)	0.55
Prune belly syndrome	1 (1%)	2 (4%)	0.14
Sacrococcygeal teratoma	0 (0%)	1 (2%)	0.13

Table 2
Surgical outcomes by type of antegrade enema procedure.

	Appendicostomy (n = 150)	Cecostomy (n = 54)	P
Length of stay (SD)	3.0 (1.3)	7.1 (3.6)	0.001
[a]Minor Complications			
Skin leakage	4 (3%)	12 (22%)	0.0001
Prolapse	4 (3%)	N/A	
Stenosis	19 (13%)	N/A	
Unable to flush	N/A	19 (35%)	
Tube dislodgement	N/A	15 (28%)	
Wound infection	10 (7%)	15 (28%)	<0.0001
[b]Major Complications			
Detachment from abdominal wall	1 (1%)	4 (7%)	0.02
Misplaced tube	0 (0%)	1 (2%)	0.3
Mean number of tube exchanges	N/A	3.8 (1–13)	
Converted to appendicostomy	N/A	3 (6%)	

[a] Defined as one managed with antibiotics or an IR or superficial surgical procedure.
[b] Defined as one requiring an intraabdominal procedure (laparotomy or laparoscopy).

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Table 3
Surgical outcomes by location of appendicostomy.

	Umbilicus (n = 125)	Other (n = 25)	P
[a]Minor Complications			
Skin leakage	4 (3%)	0 (0%)	0.48
Prolapse	4 (3%)	0 (0%)	0.48
Stenosis	16 (13%)	3 (12%)	0.99
Wound infection	10 (8%)	0 (0%)	0.14
[b]Major Complications			
Detachment from abdominal wall	1 (1%)	0 (0%)	0.99

[a] Defined as one managed with antibiotics or an IR or superficial surgical procedure.
[b] Defined as one requiring an intraabdominal procedure (laparotomy or laparoscopy).

Table 5
Surgical complications by appendicostomy technique.

	Surgical (n = 18)	Percutaneous (n = 36)	P
[a]Minor Complications			
Skin leakage	4 (22%)	8 (22%)	0.99
Unable to flush	3 (17%)	16 (44%)	0.07
Tube fell out	3 (17%)	12 (33%)	0.33
Wound infection	4 (22%)	11 (31%)	0.75
[b]Major Complications			
Intraabdominal leakage	0 (0%)	4 (11%)	0.29
Misplaced tube	0 (0%)	1 (3%)	0.99

[a] Defined as one managed with antibiotics or an IR or superficial surgical procedure.
[b] Defined as one requiring an intraabdominal procedure (laparotomy or laparoscopy).

cecostomy tube placement compared to surgical placement, although this did not reach statistical significance.

3. Discussion

Appendicostomy and cecostomy permit antegrade access for the delivery of enema flushes in patients with constipation and/or fecal incontinence in whom these procedures were chosen as an adjunct to a bowel management program. Each procedure has a unique complication profile. Within our institution, we observed a statistically higher rate of both minor and major complications occurring in patients undergoing cecostomy compared to appendicostomy.

Cecostomy offers some practical benefits over appendicostomy. First, this procedure may be performed percutaneously using only sedation, a clear advantage over appendicostomy, which requires an abdominal surgery and general anesthesia. Additionally, it is technically easier to perform. However, the overall need for subsequent interventions, whether routine or unplanned, was higher in patients undergoing cecostomy, so these risks and benefits need to be discussed with the family. A laparoscopic as opposed to percutaneous technique appears to be a safer approach. Cecostomy requires periodic tube exchanges, both on a routine basis and in some cases of tube malfunction. The need for unplanned tube exchanges is another unique problem with cecostomies, and is most commonly the result of a dislodged or clogged tube. The risk of a clogged cecostomy is likely higher because the absence of a valve allows backflow into the tube where particles can solidify and make the tube difficult to flush. Furthermore, the risk of inadvertently misplacing the cecostomy tube in an adjacent organ is a concern in percutaneously-placed cecostomy tubes, which was seen in 1 (2%) patient in our series. This specific complication is avoided in patients who receive an appendicostomy or a surgically-placed cecostomy.

In contrast, appendicostomy offers several advantages over cecostomy. Suturing the appendix to the skin minimizes the risk of the ostomy separating from the anterior abdominal wall and thus the risk of intraabdominal spillage of cecal contents and peritonitis is lower. This complication did occur in one patient with spastic cerebral palsy after appendicostomy. The cecal plication valve mechanism likely prevented gross intraabdominal spillage and the appendicostomy was revised laparoscopically on an elective basis. Although the cecum can be fixed to the anterior abdominal wall if placed surgically, this is only possible in cecostomies placed percutaneously if T-fasteners are used. Despite this, four patients had their cecostomy detach from the anterior abdominal wall in the early postprocedure period and required urgent exploratory laparotomy. No patient who received a surgically-placed cecostomy experienced this complication.

Table 4
Surgical outcomes by appendicostomy technique.

	In situ (n = 121)	Split appendix (n = 13)	Cecal flap (n = 15)	Small bowel (n = 1)	P
[a]Minor Complications					
Skin leakage	3 (2%)	0 (0%)	1 (7%)	0 (0%)	0.57
Prolapse	3 (2%)	0 (0%)	1 (7%)	0 (0%)	0.57
Stenosis	14 (12%)	0 (0%)	5 (33%)	0 (0%)	0.03
Wound infection	7 (6%)	0 (0%)	2 (13%)	0 (0%)	0.03
Detachment from abdominal wall	1 (1%)	0 (0%)	0 (0%)	0 (0%)	0.99

[b]Defined as one requiring an intraabdominal procedure (laparotomy or laparoscopy).
[a] Defined as one managed with antibiotics or an IR or superficial surgical procedure.

Although revisions of an appendicostomy require general anesthesia, the rate of patients requiring reintervention was lower. In our study population, 25 patients (17%) required an intervention needing general anesthesia for some issue related to their Malone (see Table 2). By contrast, every patient with a cecostomy required at least one routine exchange and nearly two-thirds required an unplanned procedure. The most common reasons for appendicostomy revision were stenosis followed by prolapse and leakage. Each of the revisional procedures was performed on an outpatient basis.

There are certain complications that are shared between appendicostomy and cecostomy. Leakage is one such complication. In our cohort, the rate of leakage was significantly higher in patients after cecostomy than appendicostomy. This is expected as the cecostomy lacks a valve mechanism and leaves a plastic tube on the skin. Also the flush goes directly into and distends the cecum which may lead to leakage as opposed to a Malone which directs the flush into the midright colon. Plication of the appendix during creation of the appendicostomy to create a valve mechanism has been well described, and is our practice as well [13]. The rate of wound infection was also found to be significantly higher in our cohort of patients after cecostomy compared to appendicostomy. This complication was considered unmanageable for three patients who requested elective takedown of their cecostomy and creation of an appendicostomy.

The most common complication experienced by patients who underwent Malone was stenosis. The rate of reported stenosis after appendicostomy varies widely in the literature and may approach 20% [16–18]. Our stenosis rate of 13% is slightly lower than this. Our practice is to leave an ACE stopper in our patients for 6 months postoperatively with the hope that this will reduce stenoses. It is important to emphasize that the natural healing process tends to close the appendicostomy orifice at the skin level. Thus, regular cannulation is required to maintain patency of the lumen. Upon review of the patient charts for the purposes of this study, we noted that several of the patients who developed stenosis failed to perform daily catheterization or neglected to use the Malone plug. This may be the result of inadequate education or simple noncompliance. Proper education of patients' families of this potentially avoidable complication may reduce this problem. Cathing a second time during the day and using a 10 F catheter instead of an 8 F are interventions we are planning to employ in the future to try to reduce our stenosis rate.

We found that the rate of complication following appendicostomy did not vary by its location on the abdominal wall. Generally if no other conduit is required, our preference is to place the Malone in the

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umbilicus as this site allows for the best cosmesis. However, when a patient requires a simultaneous appendicovesicostomy (Mitrofanoff), we place the appendicostomy off the midline, typically in the right lower quadrant, and put the Mitrofanoff in the umbilicus, as the bladder is a midline structure.

The choice of conduit is determined by patient factors including anatomy, coexisting disease, and future reconstructive needs. For instance, patients with bladder dysfunction who may ultimately need urinary diversion can undergo a split appendix technique whereby the appendix is shared. We feel it is very important that Malone appendicostomies not be performed until the plan for urinary reconstruction is clear and advocate for a strong collaboration between urology and colorectal surgery [19,20]. A patient with a history of appendectomy or with an inadequately sized appendix may undergo neoappendicostomy using a cecal flap. Patients with a future need for a Malone, for example one with myelomeningocele, who have malrotation and undergo a Ladd's procedure, should have their appendix preserved.

Our results demonstrate a higher rate of skin level stenosis in patients undergoing neoappendicostomy. When performing a neoappendicostomy, we tubularize a flap of the cecum supplied by a single vessel [15]. Conversely an in situ or split appendicostomy has a mesentery and relatively rich blood supply compared to neoappendicostomy, and this may account for the difference.

We recognize several limitations of our study. First, as with all retrospective designs, we are dependent on the proper input of data into the patient chart. Similarly, the rationale behind a patient choosing one procedure over another cannot be determined from the charts, thus introducing possible selection bias. Also, as many of our patients are not local to our institution, it is possible that our records reflect an incomplete understanding of their postoperative course, although we try very hard to control for this with regularly scheduled remote follow-ups. Additionally, the underlying diagnoses of patients in either cohort varied considerably between the patients who received an appendicostomy or cecostomy and thus may contribute to the rate of complications seen in either group. This is likely the result of our previously mentioned institutional referral patterns. In addition, the relatively short follow up may underestimate the true rates of many of the reported complications, many of which are time-dependent, in children who require long-term antegrade enema access. Lastly, many of the complaints described are subjective in nature, and are dependent on a patient's and family's ability to tolerate a side effect, which is likely in part a function of how well the family was educated preoperatively.

Future studies comparing cecostomy and appendicostomy should address overall patient satisfaction and quality of life before and after each procedure which has not yet been covered in the literature. A thorough evaluation of the ongoing care requirements of each procedure will provide valuable information on the relative burden of therapy and overall costs of appendicostomy and cecostomy, which will be of value when advising patients prior to intervention.

4. Conclusions

Malone appendicostomy of all types in this cohort had a lower rate of complications and we believe is preferable to cecostomy for children who require access for antegrade enemas as part of a bowel management program. A thorough understanding of the risks and benefits of both procedures is required for patients and their families in order to make an informed decision prior to any intervention.

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Please cite this article as: Halleran DR, et al, A comparison of Malone appendicostomy and cecostomy for antegrade access as adjuncts to a bowel management program for patients wi..., J Pediatr Surg (2018), https://doi.org/10.1016/j.jpedsurg.2018.10.008