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Tube cecostomy versus appendicostomy for antegrade enemas in the management of fecal incontinence in children: A systematic review

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ABSTRACT

Background: Few studies have directly compared between cecostomy and appendicostomy for the management of fecal incontinence in pediatric population. This systematic review of the literature describes outcomes and complications following both procedures. We also reviewed studies reporting impact on quality of life and patient satisfaction.
Methods: MEDLINE, EMBASE, the Cochrane Central Register of Controlled Trials (CENTRAL), and Google Scholar were searched for chronic constipation pediatric patients who underwent cecostomy or appendicostomy. Two reviewers independently screened studies, extracted data, and assessed quality.
Results: An initial literature search retrieved 633 citations. After review of all abstracts, 40 studies were included in the final analysis, assessing a total of 2086 patients. The overall rate of complications was lower in the cecostomy group compared to the appendicostomy group (16.6% and 42.3%, respectively). Achievement of fecal continence and improvement in patient quality of life were found to be similar in both groups, however the need for revision of surgery was approximately 15% higher in the appendicostomy group.
Conclusion: Cecostomy has less post procedural complications, however rates of patient satisfaction and impact on quality of life were similar following both procedures.
Level of evidence: III

Contents

Fecal incontinence has a significant impact on quality of life in the pediatric population, resulting in increased burden of care, social isolation, and loss of independence. Initial management strategies may include dietary manipulation and the introduction of stool softeners and bulking agents, with escalating interventions including timed evacuation with stimulant laxatives or enemas. Enema administration can be problematic owing to patient discomfort, increased caregiver demand, and in older age groups, challenges with self-administration affecting independence. The use of antegrade enemas by accessing the cecum directly addresses many of the difficulties encountered during rectal enema administration.

The Malone antegrade continence enema (MACE) was first introduced in 1990 [1], and involves the creation of an intestinal conduit

Abbreviations: MACE, Malone antegrade continence enema; MINORS, Methodological Index for Non-Randomized Studies; PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

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for antegrade enema administration via the cecum. In most cases the appendix is used as the conduit, but in patients lacking a usable appendix the cecum can be tubularized to provide the same function. Initial reports on outcomes following the MACE procedure showed significant improvements in continence and quality of life, although reported rates of complications vary widely between studies. The MACE procedure can be done either laparoscopically or open.

A percutaneous approach to creation of a cecostomy conduit was reported in 1996 [2], offering a minimally invasive alternative with placement of a cecostomy tube or "button" device through the abdominal wall for antegrade enema administration. The tube can be placed by interventional radiologists using an image-guided technique, laparoscopically, or using an open technique. Many patients affected by chronic constipation and fecal incontinence have a history of previous abdominopelvic surgical interventions, leading to increased interest in alternatives to open procedures. Initial reports on outcomes following button cecostomy placement have been positive, with significant functional improvement which is defined as controllable fecal continence leading to clean underwear as well as enhanced quality of life.

Few studies have directly compared these procedures for the management of fecal incontinence, with little information available to guide clinical decision making with respect to the optimal approach for a given patient. We completed a systematic review of the literature describing outcomes following the MACE procedure and percutaneous cecostomy, with a focus on functional outcomes and complications. We also reviewed studies reporting impact on quality of life and patient satisfaction.

1. Methods

A comprehensive literature review was undertaken, with an initial electronic search of MEDLINE (1966 onwards), EMBASE (1980 onwards), the Cochrane Central Register of Controlled Trials (CENTRAL), and Google Scholar, using a series of keywords (antegrade continence enema, percutaneous cecostomy, laparoscopic cecostomy, button cecostomy, Malone antegrade continence enema). Gray literature was not searched as only evidence-based, peer-reviewed literature was included. To be eligible for inclusion, studies must have specified the underlying diagnosis, including those cases of constipation defined as idiopathic constipation, Hirschsprung's disease, spina bifida and anorectal malformations, or specified the outcomes or the complications of the procedures listed. We excluded case series reporting 3 or less patients.

In the cecostomy group, all percutaneous and laparoscopic procedures were included. In the appendicostomy group, all cecal access procedures were included with the classic appendicostomy in addition to tubularization procedures using the cecum, ileum, or transverse colon as a conduit. We included imbricated (whereby the cecum is overlapped with the base of the appendix [3]) and nonimbricated appendicostomy (in which the appendix is left without overlap of the cecum [4]). We also included appendicostomy and sigmoid colectomy performed simultaneously.

Methodological Index for Non-Randomized Studies (MINORS) was used to assess the quality of nonrandomized studies. The criterion is comprised of 12 items that evaluate the study's validity, methods, and completeness of reporting. A comparative study is assigned a score of 0–2 for each of the 12 items included, for a maximum score of 24. Higher scores are indicative of greater methodological quality. Two investigators assessed each study independently and compared their score afterwards to reach a consensus. If an agreement could not be reached, a third investigator was consulted [5].

The following outcome measures were extracted: patient or caregiver reported rates of continence or persistent significant soiling, subsequent conversion to a diverting ostomy for persistent constipation

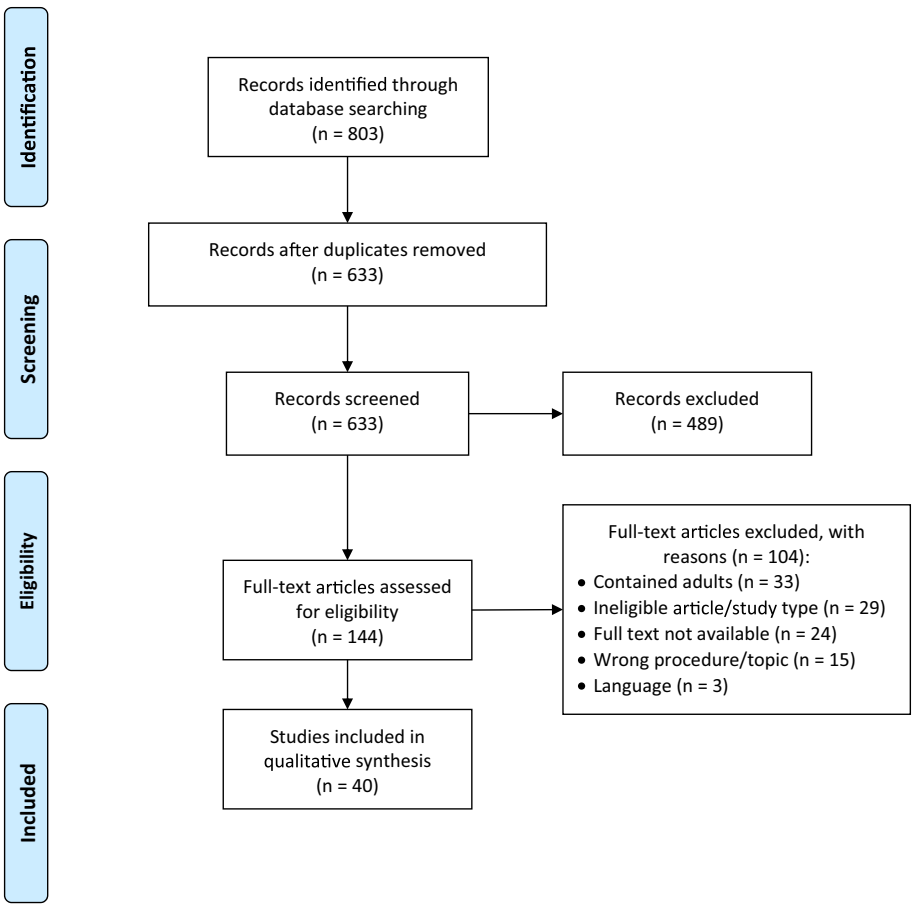

Fig. 1. PRISMA flow diagram for inclusion and exclusion of studies.

Table 1
Patient demographics.

	Appendicostomy	Cecostomy
Male	709 (55.8%)	242 (58.6%)
	From 24 studies ($n = 1270$)	From 7 studies ($n = 413$)
Mean age (range)	9.64 (0.5–18) years	9.39 (3–15) years
	From 31 studies ($n = 1534$)	From 7 studies ($n = 413$)
Mean FU	37.6 months	38.7 months
	From 25 studies ($n = 1266$)	From 7 studies ($n = 413$)

and incontinence, patient or caregiver reported improvements in quality of life, overall complication rates, and frequency of specific complications including wound infection, skin erosion or granulation tissue formation, leakage at the site of access, requirement for surgical revision, pain during irrigation, and procedure-specific complications including button dislodgement or mechanical dysfunction (button cecostomy) and stomal infection, stenosis, or other stoma complications including prolapse.

2. Results

An initial literature search retrieved 633 citations following deduplication. After review of all abstracts, 144 papers were retrieved for detailed evaluation of inclusion and exclusion criteria.104 papers were excluded after title and abstract screening, leaving 40 studies included in the final analysis (Fig. 1). Studies included 31 case reviews on patients undergoing appendicostomy procedures [6–36], 5 case reviews on patients undergoing cecostomy [37–41] [38–42], and 4 studies which included both approaches and provided demographics and outcomes clearly distinguishable between these two cohorts [42–45]· [3–6]. No randomized controlled trials were identified.

In the 9 studies on the cecostomy approach, a total of 432 patients were included; of the 35 studies on the appendicostomy procedure, a total of 1654 patients were included assessing a total of 2086 patients. Demographic data reported were variable but similar between the two categories (Table 1).

In both the cecostomy and appendicostomy cohorts, the most common mechanism of incontinence was neurogenic; with spina bifida representing the most often reported underlying diagnosis. Congenital anomalies, mostly involving various forms of anorectal malformation, were the second most common diagnostic category in the appendicostomy group and the third most common mechanism in the cecostomy group. Idiopathic constipation was the second most common mechanism in the cecostomy cohort and third in the appendicostomy group, followed by Hirschsprung's disease. In both cohorts, a small number of patients had other rare diagnoses including genetic and metabolic disorders (Table 2).

Clean underwear was achieved in 88.8% of patients following cecostomy and 87.5% of patients following appendicostomy procedures, while persistent soiling occurred postoperatively in 11.2% and 12.5% respectively. Revision of surgery owing to failure (either by complication or significant persistent soiling) occurred in 1.5% of patients following cecostomy and 16.5% of patients after appendicostomy. A small proportion of patients underwent creation of a diverting ostomy owing to failure of the cecostomy or appendicostomy procedures (0.5% and 3.0%, respectively).Overall, rates of patient satisfaction were high following both procedures, and a subset of studies quantified impact on quality of life, with high rates of improvement following both procedures (cecostomy: 92.4%; appendicostomy:89.8%). A relatively small number of patients had their surgery reversed, either surgically or nonsurgically, after complete resolution of symptoms (cecostomy: 4.2%; appendicostomy: 3.6%; Table 3).

Table 3
Patient outcomes.

Outcome	Appendicostomy $n = 1289$[a]	Cecostomy $n = 410$[b]
Continence	1128 (87.5%)	364 (88.8%)
Persistent soiling	161 (12.5%)	46 (11.2%)
Reversal owing to success	47 (3.6%)	17 (4.2%)
Revision owing to failure	213 (16.5%)	6 (1.5%)
Diverting ostomy	39 (3.0%)	2 (0.5%)
Improved QOL/total responders	194/216 (89.8%)	85/92 (92.4%)

[a] From 26 studies reporting on these outcomes.
[b] From 6 studies reporting on these outcomes.

The overall rate of complications was lower in the cecostomy group compared to the appendicostomy group (16.6% and 42.3%, respectively) and unique complications of each approach were apparent. Stenosis was the most common complication following appendicostomy while it was very rare following cecostomy (16.7% and 0.5%, respectively). Leakage was the second most common complication following appendicostomy, which was also quite rare following cecostomy (10.8% and 2.3%, respectively), while granulation tissue formation, difficult catheterization, stoma site infection, and pain during irrigation occurred at similar frequencies in both groups(Table 4). Two studies explicitly compared leakage rates for appendicostomy for those in which imbrication was performed or no imbrication and found a reduction in leakage for the imbricated group (imbricated: 2.9% and 0%; nonimbricated: 29.4% and 40%).

Studies reporting on follow up time reported a median of 2.4 years for the appendicostomy groups, and 1.9 years for the cecostomy group. Few studies explicitly reported the longer-term outcome of converting from one approach to the other. While no studies reported conversion of cecostomy to appendicostomy, three reported small conversion rates from appendicostomy to cecostomy (1.25%–6.25%).

3. Discussion

This study was designed to compare outcomes and complications following cecostomy and appendicostomy procedures for the treatment

Table 2
Mechanism of incontinence.

Mechanism	Appendicostomy $n = 1517$[a]	Cecostomy $n = 401$[b]
Neurogenic	561 (37.0%)	169 (42.1%)
Spina Bifida	401(26.4%)	168 (41.9%)
Myelomeningocele	127(8.4%)	1 (0.3%)
Other	33(2.2%)	0 (0%)
Anorectal malformation	521 (34.3%)	83 (20.7%)
Idiopathic constipation	309 (20.4%)	97 (24.2%)
Hirschsprung's disease	88 (5.8%)	9 (2.2%)
Other	38 (2.5%)	42 (10.5%)

[a] From 27 studies reporting on mechanism of incontinence.
[b] From 6 studies reporting on mechanism of incontinence.

Table 4
Postoperative stoma or device related complications.

Complication	Appendicostomy $n = 1515$[a]	Cecostomy $n = 432$[b]
Total complications	42.3%	16.6%
Stenosis	253 (16.7%)	2 (0.5%)
Prolapse	59 (3.9%)	1 (0.2%)
Dislodgement	11 (0.7%)	9 (2.1%)
Difficult catheterization (fistula/false passage)	26 (1.7%)	15 (3.5%)
Retraction	0	1 (0.2%)
Leakage	163 (10.8%)	10 (2.3%)
Infection	83 (5.5%)	13 (3.0%)
Granulation tissue	43 (2.8%)	14 (3.2%)
Pain during irrigation	49 (3.2%)	7(1.6%)

[a] From 31studies reporting on these outcomes.
[b] From 9 studies reporting on these outcomes.

of fecal incontinence in the pediatric population. Since it originated, modifications to the MACE procedure have been described, including the tubularization of intestinal conduits including the small bowel [7,8], cecum [22,30], and ascending colon [16,36] to permit antegrade continence enema creation in patients who have previously undergone appendectomy or in whom the appendix has already been used as a bladder conduit, as well as a minimally-invasive laparoscopic-assisted approach [11,20]. Similarly, a laparoscopic-assisted approach has been described for the percutaneous button cecostomy [37,40,41] [40–42], with no major differences in the achievement of continence or reported rates of complications in comparison to the original percutaneous procedure.

Given the similarities in the mechanism of enema delivery between the two approaches, it was expected that functional outcomes and rates of technical failure would be comparable. However, it was anticipated that complications would differ according to the mechanism of stoma creation. Indeed, overall rates of postprocedural complications were found to be lower in the cecostomy group while functional outcomes were similar in both groups. Establishing antegrade enema by creation of an appendicostomy or tubularized bowel segment increases the risk of stoma-related complications including stenosis and leakage, while granulation tissue formation, difficult catheterization, and stoma site infection were noted in both groups. Stenosis was reported more frequently in the appendicostomy group. This may be owing to the nature of the approach, whereby the stoma is left open, compared to the cecostomy approach in which the button remains in the stoma, thereby reducing or eliminating the risk of stenosis.

We cannot comment on the superiority of one technique over another with respect to cosmetic issues and psychological problems around intubation of a stoma, as these were not reported in most studies.

In this systematic review, achievement of fecal continence and improvement in patient quality of life were found to be similar following cecostomy and appendicostomy procedures; however, the need for revision of surgery was approximately 15% higher in the appendicostomy group compared to the cecostomy group. In addition, complications associated with cecostomy were minor and did not require operative intervention (e.g., granuloma, difficult catheterization, and site infection), while stoma stenosis and leakage following appendicostomy were found. Appendicostomy was more likely to require revisional surgery.

Follow up time in the identified studies was short. It is possible that other complications may emerge over a longer postoperative follow up period, such as further frequency of a need for conversion from one approach to another, as well as potential loss of long term continence. Ultimately, the decision must be individualized for each patient, after careful presentation of all risks and full discussion with the child and family.

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Conflict of interest

The authors do not have any conflicts of interest to declare.

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