



Malone appendicostomy versus cecostomy tube insertion for children with intractable constipation: A systematic review and meta-analysis

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ABSTRACT

Purpose: Children with intractable constipation are often treated with antegrade continence enemas. This requires the creation of a Malone appendicostomy in the operating room or insertion of a cecostomy tube using endoscopic, radiologic, or surgical techniques. The purpose of this study was to assess the evidence regarding these procedures. **Methods:** We conducted a search of Embase, Medline, CINAHL, and Web of Science up to October 2016. We included comparative studies of children treated with Malone appendicostomy or cecostomy tube insertion. Two reviewers screened abstracts, reviewed studies, and extracted data.

Results: We identified 166 children from three retrospective studies who underwent Malone appendicostomy ($n = 82$) or cecostomy tube insertion ($n = 84$). There were no differences in the number of patients who achieved continence (80% versus 70%, $p = 0.76$), but the need for additional surgery was higher in children treated with Malone appendicostomy (30% versus 12%, $p = 0.01$). Studies reported a variety of tube and stoma-related complications, but quality of life was not assessed using validated measures.

Conclusion: Malone appendicostomy and cecostomy tube insertion are comparable in terms of achieving continence. Children treated with Malone appendicostomy appear to be more likely to require additional surgery due to early or late complications.

Level of Evidence: Therapeutic, 1c.

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Constipation is common in the pediatric population and affects up to one in three children. The majority of cases are functional in origin, with only a minority resulting from pathologic causes such as spina bifida, anorectal malformation, cystic fibrosis, or hypothyroidism [1]. Functional constipation is defined by the Rome IV criteria and requires at least two of the following features: two or fewer defecations per week, stool retention, painful or hard bowel movements, large-diameter stools, presence of impacted stool in the rectum, and episodes of fecal incontinence following the acquisition of toileting skills [2].

Both functional and pathologic causes of constipation can have wide-ranging effects on quality of life for the child and family [3]. This association is even more significant in patients with overflow fecal incontinence [4]. Adequate bowel management is essential and early intervention may improve symptoms and psychosocial well-being [4].

The first step in the management of constipation in children is to rule out organic causes and address any factors that could be contributing to poor bowel function. This includes obtaining a thorough history from the parent or caregiver regarding bowel habits, weight gain, growth,

bladder function, and duration and timing of symptoms. Among patients who present acutely in distress, clinicians should inquire about fever, vomiting, diarrhea, rectal bleeding, abdominal distension, and history of delayed passage of meconium in the first 48 h of life. Factors that suggest a functional cause in an otherwise healthy child include stool withholding behavior, painful bowel movements, and onset of constipation associated with changes in diet or toilet training [2].

Most children with functional constipation respond to increasing fluid and fiber intake and a trial of laxatives. These include oral osmotic and stimulant agents in addition to rectal suppositories and enemas [2,3]. The Canadian Pediatric Society recommends referral to a pediatric gastroenterologist when children continue to have symptoms despite maximal medical management [3]. Evidence-based guidelines from the North American and European Societies for Pediatric Gastroenterology, Hepatology, and Nutrition suggest considering procedural interventions when patients experience intractable constipation refractory to medical management [5].

Procedural options include Malone appendicostomy, which is created using a surgical approach, and cecostomy tube insertion, which can be performed using endoscopic, radiologic, or surgical techniques. The purpose of these strategies is to create a conduit to the proximal colon to allow for the administration of antegrade continence enemas [5]. This prevents stool impaction and overflow fecal incontinence.

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The Malone appendicostomy was first described in 1990 by Dr. Patrick Malone [6] and may be performed using open or laparoscopic surgery. The appendix is used to create a conduit from the skin to the cecum. The advantage of this technique is that it eventually allows for intermittent catheterization and does not require an indwelling device. The most commonly reported complications include stoma stenosis, fecal leakage around the insertion site, and stoma site infections [7].

Cecostomy tube insertion was first described in 1996 and may be inserted with a variety of approaches, including endoscopic [8], radiologic [9], open surgery [10], or laparoscopic techniques [10,11]. Like the Malone, these options provide catheter access to the proximal colon and allow for the administration of antegrade continence enemas. Cecostomy tube insertion has the potential to be less invasive than Malone appendicostomy but requires an indwelling catheter, such as a Chait trapdoor [12]. These devices must be replaced on a routine basis and the tract may close if the tube becomes dislodged. The most common complications include tube blockage, fracture, or dislodgement, as well as pain with irrigations or leakage at the tube site [10].

Physician preference, accessibility, and local practice often determines which procedure is recommended. Previous research comparing these interventions consists of a limited number of retrospective studies and no randomized control trials. The purpose of this study was to review the evidence for Malone appendicostomy versus cecostomy tube insertion for children with constipation refractory to maximal medical management. We meta-analyzed data for outcomes such as complications, fecal continence, and quality of life whenever possible. Our hope is that this will better inform decision-making for clinicians, patients, and families, and provide direction for future research.

1. Methods

1.1. Study design

We conducted a systematic review and meta-analysis of children with functional and pathologic causes of intractable constipation refractory to maximal medical management. Our intervention and comparison groups were those treated with Malone appendicostomy versus

cecostomy tube insertion. Our primary outcomes were continence post-procedure and quality of life. Secondary outcomes included adverse events and complications. Our study design was registered with the International Prospective Register of Systematic Reviews (PROSPERO) on October 13, 2016 (number CRD42016048569) [13].

1.2. Search strategy

We conducted systematic searches of Medline, Embase, Web of Science, CINAHL, and Cochrane Central Register of Controlled Trials for studies of children with intractable constipation treated with Malone appendicostomy or cecostomy (see Appendix A for sample search strategy). A research librarian developed the search strategies for each database to select studies that included Malone appendicostomy or cecostomy. These concepts were further expanded to capture studies with variation in categorization, nomenclature, and syntax. The reference lists of included studies were also manually searched for any additional studies. We also performed searches of conference proceedings [14], theses and dissertations [15], and trial registries [16–19]. Citations up until October 2016 were included.

1.3. Study selection

Title and abstract screening was completed independently and in duplicate by two authors (CL, SS). Disagreements were resolved through review by a third author (MHL). Studies were included if participants were less than 18 years of age and compared outcomes for Malone appendicostomy and cecostomy tube insertion. We excluded studies that focused exclusively on adults, focused on one procedure only, or did not report outcomes related to continence.

1.4. Statistical analysis

The level of agreement for screening titles and abstracts was assessed with an unweighted kappa statistic. Outcome data were meta-analyzed using a random effects model and forest plots were created using Review Manager 5.3 [20]. Summary statistics were reported

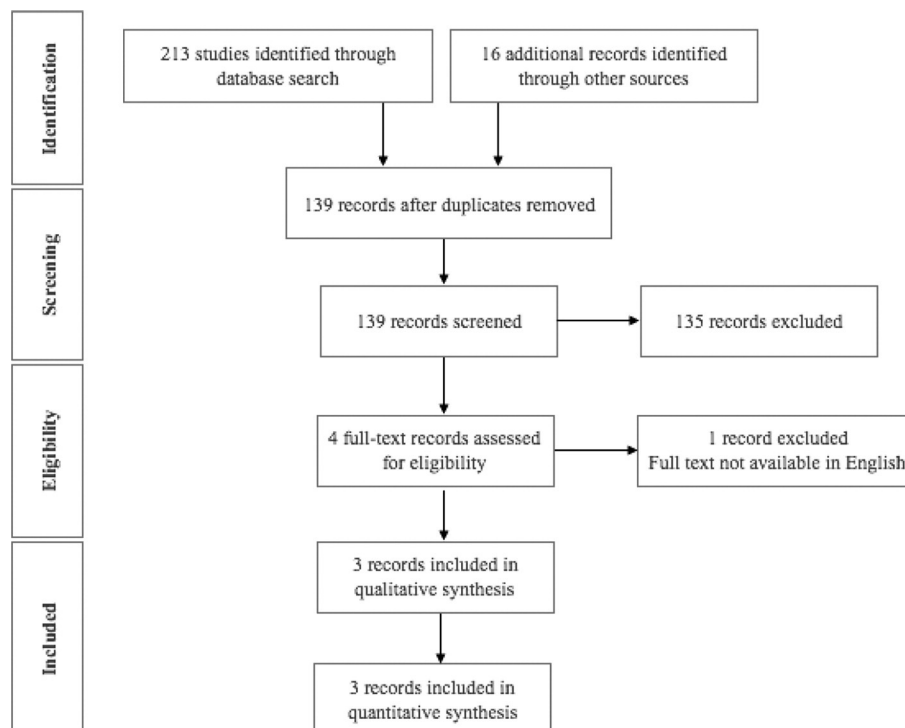


Fig. 1. Study selection using Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) format [25].

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