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What happens to children with idiopathic constipation who receive an antegrade continent enema? An actuarial analysis of 80 consecutive cases

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Abstract

Introduction: There is uncertainty about the prognosis for children with idiopathic constipation who opt for treatment by colonic lavage using an antegrade continent enema (ACE). The aim of this study was to perform an actuarial analysis of the outcomes of the ACE in children consecutively referred to our unit for this procedure, who suffered from idiopathic constipation and who had failed to respond to 3 years of medically supervised conservative management.

Methods: This study is a prospective analysis of the outcomes of 80 children with uncontrolled idiopathic constipation who underwent construction of an ACE by 1 surgeon.

Results: Twelve children were able to stop using their ACE because of resolution of their symptoms. The probability of a child who has idiopathic constipation being able to stop colonic lavage was 0.2, 6.2 years after construction of the ACE. In this group, the estimated mean time to have an ACE reversed was 8.8 years. Twelve children did not achieve satisfactory colonic lavage and either gave up (4) or deteriorated and had alternative treatment for their symptoms (8). The probability of ACE failure is 0.3 at 8.5 years after construction. Girls were significantly more likely to fail than boys, and colonic transit time was significantly longer among children who subsequently required alternative treatment for their symptoms.

Conclusions: Children with idiopathic constipation whose symptoms fail to resolve with medical management and who are treated with an ACE have 0.2 probability of cure, 0.3 probability of failure, and 0.5 probability of having to continue with colonic lavage after 6 years of colonic lavage.

The antegrade continent enema (ACE) was introduced by Malone et al [1] in 1990 as an aid to fecal continence in children with either a myelomeningocele or imperforate anus. The procedure has quickly become accepted in both pediatric and adult practice [2] as an effective and safe [3] option for patients with imperfect bowel control secondary to neuropathy [4] or as a consequence of congenital or acquired bowel pathology [5].

Although it might be expected that patients born with spina bifida or an imperforate anus would require lifelong assistance with bowel dysfunction, there is uncertainty about

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the prognosis for children with idiopathic constipation who opt for treatment by colonic lavage.

The aim of this study was to perform an actuarial analysis of the outcomes of the ACE procedure in children consecutively referred to our unit for this procedure, who have idiopathic constipation, and who did not respond to 3 years of medically supervised conservative management.

1. Patient and methods

All children who have idiopathic constipation undergoing ACE surgery by 1 surgeon were prospectively recorded in a customized database (Access, Microsoft Corporation). Children were followed up in a nurse-led continence clinic where details of their current status were recorded.

Antegrade continent enema was offered to children with idiopathic constipation where symptoms persisted despite medical management supervised by a pediatrician for at least 3 years. Previous medical management was heterogeneous but had always included prolonged treatment with laxatives, usually with periods of in-patient administration of surgical bowel-cleansing solutions, frequent manual disimpactions, and often involvement of a clinical psychology service.

In the first 32 cases, the diagnosis was confirmed by the use of marker studies using an established protocol [6]. However, because the marker studies did not alter the decision to undertake an ACE, and to avoid unnecessary radiation exposure, this practice was stopped. Hirschsprung disease was excluded by rectal biopsy in all cases.

The lavage regimen was supervised by specialized nurses and used a solution of saline prepared by parents at a volume of 20 mL/kg body weight. Details of studies on the physiological effects of saline lavage are published elsewhere and are not the subject of this study [3]. The complications of the ACE procedure have been reported in a previous communication and are not the subject of this study [7].

The outcomes of an ACE were categorized into 3 groups:

1. Ongoing lavage: The length of follow-up was calculated as time from the date of formation of the ACE to the current date.
2. Failure: either the parents had stopped using the ACE because colonic lavage had not been found to improve the child's bowel habit, or the child's colon had not proved to be lavageable and symptoms had deteriorated. The time to failure was calculated as the time from creation of the ACE to the clinic letter stating that the parents had ceased using the ACE or the date of commencement of alternative treatment. Children who could not be lavaged were defined as those having failed to have a bowel evacuation despite an appropriate volume of lavage fluid (defined elsewhere [3]). These children were assessed by performing continuous lavage through their appendicostomy over several days while in hospital. Typically, such children accommodate very large volumes of fluid in their colon, often in excess of 10 L, without bowel evacuation.
3. Cure: The appendicostomy was closed/reversed because the child achieved normal bowel habit. The criteria for ACE reversal were that for at least the previous 6 months, the child had stopped using the ACE, was stooling spontaneously at least every other day, was not requiring laxative therapy, and was not soiling. The ACE was reversed by dissecting the appendix to the cecal wall and ligating and removing it. The date of the operation to reverse the ACE was used as the censoring time.

A minimum of 6 months follow-up was judged to be appropriate because a decision regarding "cure" would take no less than 6 months to determine.

1.1. Statistical analysis

The probability of reversal or failure of ACE was calculated with Kaplan-Meier analysis, comparing groups using the log-rank test. Analysis of the effect of colonic transit time, sex, age at time of surgery, and duration of follow-up on the probability of an ACE being reversed or failing used binary logistic regression analysis, using a forward conditional model calculated using the SSPS statistical program, version 15. Comparison of colonic transit time used the Mann-Whitney test having confirmed the data were not normally distributed using a q-q plot. Significance was defined as $P < .05$.

2. Results

Eighty children (44 boys, 36 girls) underwent an ACE. They had a median age at surgery of 9.6 years (range, 3.4-18.7 years). Fifty-three had an appendicostomy established through a conventional grid-iron incision, whereas the most recent 27 children had a laparoscopic ACE. Follow-up ranged from 6 months to 10 years, with median follow-up of 6.2 years. No patient was discharged, and none was lost to follow-up.

Four children were identified where the appendicostomy was not being used. Although these children could be lavaged, the parent's had not found it to be of help in the child's bowel management and had ceased use.

In 8 children, deterioration of symptoms occurred despite ACE lavage, and this required alternative treatment of symptoms. These children could not be lavaged. Therefore, ACE lavage failed in 12. The Kaplan-Meier probability of an ACE failing was 0.3 at 8.5 years (Fig. 1), with an estimated mean time to failure of 8.6 years (95% confidence interval [CI], 79-9.2). None of the children who subsequently failed ACE therapy did so because of a complication of this surgery.