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Outcomes of Initial Subtotal Colectomy for Pediatric Inflammatory Bowel Disease



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ABSTRACT

Background: Subtotal colectomy with end ileostomy (STC-I) has been well established in the adult literature as an initial surgical treatment for refractory inflammatory bowel disease (IBD)-related colitis. However, in the pediatric population, the efficacy of this approach has been less well characterized, likely because of concerns regarding the advisability of leaving a diseased rectum *in situ*. Our aim was to examine the outcomes after STC-I for refractory IBD at our pediatric tertiary care center.

Methods: An institutional review board–approved retrospective review of patients aged 5–21 y who underwent operative treatment with initial STC-I for medically refractory IBD from January 2010 to August 2018. Only complications related to the STC-I were considered; complications subsequent to reconstruction are excluded from analysis. Early complications were defined as occurring within 60 d of STC-I. We performed descriptive statistics using the Fisher exact test and the Student t-test, as appropriate.

Results: Over the study period, 37 patients (aged 12.3 ± 4.2 y) underwent STC-I, with 73.0% performed laparoscopically. Patients were predominately male (51.4%) and Caucasian (48.6%). Thirty-one (83.8%) colectomies were performed for ulcerative colitis, two (5.4%) for Crohn disease, and four (10.8%) for indeterminate colitis. Nutritional status improved postcolectomy. Albumin levels of 3.3 ± 0.8 preoperatively increased to 4.3 ± 0.47 postoperatively ($P < 0.001$). Colonic bleeding was stopped by STC-I with increases in the hematocrit from 30.5 ± 6.8 preoperative to 38.9 ± 4.1 postoperatively ($P < 0.001$). Average time to discontinuation of IBD-related medications was 4 wk ($n = 27$). Forty-eight percent required outpatient rectal treatment for proctitis. Patients did well long term, with 67.5% reestablishing intestinal continuity at our institution. Average postoperative length of stay was shorter in the laparoscopic group compared with those undergoing open operations (5.1 ± 2.2 versus 6.9 ± 1.6 d, $P = 0.03$). Readmission rate at 30 d was 21.1%. Patients experiencing unplanned readmission or unplanned operations were similar between groups (30% versus 33.3%, $P = 0.85$ and 30% versus 18.5%, $P = 0.45$, respectively). Overall, 14 (37.8%) patients experienced a complication with many patients experiencing multiple complications. Early complications occurred in nine (24.3%) patients. Late complications also

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occurred in 24.3% of patients. There were four (10.8%) patients with five admissions for bowel obstruction, two of whom required operative intervention (5.4%).

Conclusions: Use of STC-I as an initial procedure in the treatment of refractory IBD-related colitis in children is a safe and reasonable surgical approach that allows weaning from immunosuppressing medications and stops colonic bleeding. Implementing a laparoscopic approach to subtotal colectomy provides further benefit by reducing postoperative length of stay.

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Background

Globally, the incidence of pediatric-onset inflammatory bowel disease (IBD) is increasing.¹ Despite advances in medical therapy, studies report between 14% and 44% of children with IBD will eventually require operative intervention.²⁻⁴ Indications for surgery in patients with IBD-related colitis include medically refractory disease and dysplasia, although dysplasia is uncommon in the pediatric population. Operative therapy for colonic IBD usually requires removal of the entirety of the colon and rectum and restoration of enteric continuity with an ileal reservoir connected to the anus. There are a variety of approaches to achieve this goal but the initial surgery is often either a total proctocolectomy with creation of an ileal pouch-anal anastomosis (IPAA) and a diverting ileostomy or an upfront subtotal colectomy with end ileostomy (STC-I) and a delayed completion proctectomy and creation of an IPAA, with or without a diverting ileostomy.⁵

STC-I (leaving a diseased but diverted rectal pouch in the pelvis) for medically refractory colitis has been well established in the adult literature as a viable initial surgical treatment for refractory IBD-related colitis; it is a safe initial operation that results in improved patient status before undergoing the IPAA procedure,⁶ with an associated decreased rate of anastomotic leakage.⁷ Elective IPAA creation additionally offers the added benefit of improved outcomes compared with emergency procedures.⁸

However, in the pediatric population, the efficacy and safety of an initial subtotal colectomy approach have been less well characterized, and some have argued that leaving a diseased rectum *in situ* at the initial operation is ill advised. Critics of this approach often argue that the retained rectum will continue to result in significant bleeding or that patients will still require sustained medical therapy. Pediatric-onset IBD is typically more aggressive and rapidly progressive compared with adult-onset IBD with what appears to be a stronger genetic component.⁴ In addition to the symptoms common to IBD of any age, pediatric-onset IBD can also result in growth failure, either from the disease itself or stemming from treatment of the disease. This failure to thrive can be the impetus to proceed to operative intervention.^{9,10} Given the differences in disease burden in children as compared with adults and taking into consideration the paucity of data examining the outcomes of a subtotal first approach to pediatric refractory IBD-related colitis, the aim of this study was to examine the outcomes after initial STC-I for refractory IBD at our pediatric tertiary care center.

Methods

After institutional review board approval, the records of pediatric and young adult patients, aged 5-21 y, who underwent operative treatment with an initial STC-I for medically refractory IBD from January 2010 to August 2018 at our tertiary care children's hospital were reviewed. A waiver of informed consent was obtained. The medical record was examined for details of the hospitalization and operation, including laparoscopic versus open approach and postoperative length of stay, and complications related to initial subtotal colectomy. Complications occurring after enteral reconstruction were not included in this analysis. Laboratory values and time to discontinuation of immunosuppressive medications were examined in both inpatient and outpatient records when available. Operations were classified as either semielective, indicating the patient was admitted for the purpose of the procedure, or emergent, indicating the patient was hospitalized before the decision to proceed with surgery during that admission. Early complications were defined as complications occurring within 60 d of the STC-I. Preoperative phlebotomy results describe bloodwork collected on an outpatient basis in preparation for the scheduled procedure or during the admission in which the operation took place for emergency procedures. Postoperative laboratory values were taken at least 2 mo postoperatively, typically in an outpatient setting and in the absence of acute illness. Descriptive statistics were calculated using the Fisher exact test and the Student t-test, as appropriate.

Results

A total of 37 patients were identified. The average patient age at the time of operation was 12.3 ± 4.2 y (range 5-21). Three (8.1%) patients were aged between 18 and 21 y. Patients were predominately male (51.4%) and Caucasian (48.6%) (Table 1). Time between IBD diagnosis and operation varied. Six (16.2%) patients underwent STC-I within 1 y of diagnosis, 17 (45.9%) between 1 and 3 y from diagnosis, seven (18.9%) between 3 and 5 y from diagnosis, and seven (18.9%) more than 5 y after initial diagnosis (Figure). Of the 37 STC-I, 73.0% were performed laparoscopically. All open procedures were performed in the first 2 y of the study period. Initial indications for STC-I include ulcerative colitis (UC) in 31 (83.8%), colonic Crohn disease in two (5.4%), and indeterminate colitis in four (10.8%) patients. Fourteen (37.8%) procedures were done on an emergency basis. Average preoperative length of stay for these patients was 10.1 ± 5.9 d (range 1-22 d).

Table 1 – Patient demographics.

Total, N	37
Age (y)	12.3 ± 4.2
Weight (kg)	45.2 ± 16.8
Body mass index	19.5 ± 3.7
Sex (male)	19 (51.4%)
Ethnicity	
Caucasian	18 (48.6%)
African American	5 (13.5%)
Asian	4 (10.8%)
Multiracial	4 (10.8%)
Not reported	6 (16.2%)
Diagnosis	
Ulcerative colitis	31 (83.8%)
Crohn colitis	2 (5.4%)
Indeterminate colitis	4 (10.8%)

Patients improved postoperatively (Table 2). As far as nutritional status, albumin levels of 3.3 ± 0.8 preoperatively increased to 4.3 ± 0.47 postoperatively ($P < 0.001$). In addition, bleeding per rectum improved significantly post-STC-I, with an increase in the hematocrit from 30.5 ± 6.8 (range 19.5-43) to 38.9 ± 4.1 (range 32-49), $P < 0.0001$. Before STC-I, 58% of patients were noted to have received blood transfusions on an either inpatient or outpatient basis. After recovery from STC-I, no patients required additional transfusions. Three patients with postoperative complications did receive transfusion in the immediate postoperative period, all within a week of operation.

Before STC-I, 86.5% of patients were maintained on steroid therapy, 51.4% of patients were on immunomodulators, and 75.7% of patients were on biologics. Among patients for whom complete data were available in the outpatient records ($n = 27$), average time to discontinuation of IBD-related medications was 4 wk, with only one patient with a known, preoperative diagnosis of colonic Crohn disease requiring continued medication in the form of biologic therapy (Table 2). A total of 48% of patients required enemas for treatment of proctitis; no patients required oral or intravenous steroid

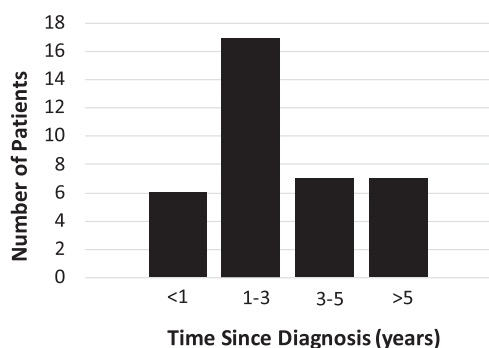


Fig – Time between diagnosis of IBD and operative intervention.

therapy. Twenty-five (67.5%) patients reestablished intestinal continuity at our institution.

In total, 14 (37.8%) patients experienced a complication with several patients experiencing multiple complications. All three patients in the age range 18-21 y experienced at least one complication. Of the 37 patients, six patients were readmitted eight times within 30 d postoperatively, for a 30-d readmission rate of 21.6%. Early complications occurred in nine (24.3%) patients. These included venous thromboembolism (5.4%), small bowel obstruction (5.4%), and intestinal perforation (2.7%). There were three patients with rectal stump dehiscence (8.1%), one resulting in mortality (The mortality occurred in a 10-y-old male with UC who was critically ill before the procedure. He had been hospitalized for more than a month, was cushingoid from chronic steroids, had several septic emboli, and required daily blood transfusion. He had developed a seizure disorder from posterior reversible encephalopathy.) after sepsis-induced cardiovascular dysfunction necessitating extracorporeal membrane oxygenation cannulation and one requiring emergency proctectomy. Late complications occurred in 24.3% of patients and included readmissions for dehydration (5.4%) or abdominal pain (2.7%). There were four (10.8%) patients with five admissions for bowel obstruction, two of whom required operative intervention (5.4%) (Table 3).

The number of patients experiencing unplanned readmission or unplanned operations at any time before enteral reconstruction was similar between open and laparoscopic groups. (30% versus 33.3%, $P = 0.85$ and 30% versus 18.5%, $P = 0.45$, respectively). When considering the total number of unplanned readmissions (six readmissions in three of 10 patients versus 13 readmissions in nine of 27 patients, $P = 0.5$) or unplanned operations (six procedures in three of the 10 patients versus seven procedures in five of the 27 patients, $P = 0.05$) there were decreased number of unplanned procedures in the laparoscopic group. Average postoperative length of stay was shorter in the laparoscopic group compared with those undergoing open operations (5.1 ± 2.2 versus 6.9 ± 1.6 d, $P = 0.03$), even excluding one patient in the open group with a postoperative stay of 43 d secondary to complications of extraintestinal Crohn disease and toxic megacolon.

Of note, during this timeframe, only one patient underwent a primary IPAA for colonic IBD at our institution. As such, we were not able to include a comparison between initial STC-I and primary IPAA in our data. The paucity of patients who underwent a primary IPAA likely reflects local surgical referral patterns. Interestingly, this patient also experienced multiple complications, including an abdominal collection requiring transrectal drainage and an eventual anostomy with dilation of the anastomosis.

Discussion

Pediatric IBD is a challenging disease; patients requiring operative therapy are typically on the severe end of the disease spectrum. They are often on toxic medications and are nutritionally and physically debilitated. Thus, postoperative complications are common. A 2016 study using a national French population-based database demonstrated that

Table 2 – Patient parameters post-STC-I.

Parameter	Preoperative	Postoperative	P value
Albumin	3.3 ± 0.8 (1.8-4.9)	4.3 ± 0.47 (3.7-5)	<0.001
Hematocrit	30.5 ± 6.8 (19.5-43)	38.9 ± 4.1 (32-49)	<0.001
% Patients on steroids	86.5%	0%*	<0.001
% Patients on immunomodulators	51.4%	0%*	<0.001
% Patients on biologics	75.7%	3.7%*	<0.001

* Some outpatient records not available, n = 27.

approximately 50% of children undergoing surgical treatment for IBD will experience at least one significant postoperative complication within 5 y of the procedure.¹¹ A 2018 study of nearly 800 pediatric patients with IBD examining the burden of surgery and postoperative complications again found that postoperative complications after IBD surgery were common, with 13% of patients with Crohn disease and 41% of patients with UC experiencing at least one complication.¹² Our data support these findings.

Given the frequency with which pediatric patients with IBD with colitis requiring colectomy will experience postoperative complications, it is a worthwhile endeavor to consider the life-long implications of these complications. Ultimately, one of the most feared complications of an ileal pouch is pouch failure and the subsequent need for a permanent life-long ostomy. The adult and pediatric literature reports pouch failure rates ranging from 1.5%-10%.¹³⁻¹⁹

The most common cause of pouch failure is pelvic sepsis because of intra-abdominal and peripouch abscesses secondary to anastomotic leak or, less commonly, disruption of the staple line at the tip of the J-pouch or within the pouch body.²⁰⁻²² Risk factors for pelvic sepsis include systemic inflammation, malnutrition, prolonged steroid usage, and hypoalbuminemia²³; all factors which are modifiable by an STC-I first approach.

A recent meta-analysis of over 2500 pediatric patients undergoing IPAA demonstrated a pouch failure rate of 16% in the setting of corticosteroid use compared with the overall pouch failure rate of 8% in their analysis.¹⁴ Although data are mixed, numerous studies have also demonstrated worse outcomes for patients undergoing IBD-related procedures when on

biologic therapy.²⁴⁻²⁶ A prospective, multicenter database review of 640 adult patients with IBD undergoing total proctocolectomy with IPAA in a one, two, three, or modified two-stage approach found a 15.0% anastomotic leak rate. When considering only patients undergoing total proctocolectomy with IPAA as part of the initial procedure, disease duration over 5 y and concurrent treatment with anti-tumor necrosis factor therapy and steroids were independent risk factors for anastomotic leakage on multivariate analysis. Analysis of patients undergoing a three-stage or modified two-stage approach (in which a subtotal colectomy is performed first) identified that when patients were in better condition and weaned off medication before IPAA the leakage rate decreased to 10%.²⁷

Despite the associated complications inherent to the treatment of pediatric patients with IBD, a subtotal colectomy first approach allows for a functionally improved patient before the construction of an ileoanal pouch, potentially decreasing the rate of pelvic sepsis and pouch failure rates associated with malnutrition and steroid or biologic treatment.

To our knowledge, this is one of the largest studies examining complications after the STC-I portion of a staged procedure for pediatric IBD. A study by Patton et al.²⁸ examining early and late complications for 31 pediatric patients undergoing colectomy for IBD included nine patients with initial STC-I and delayed IPAA and found complications in more than 50% of patients. A 2015 UK-based study of 19 subtotal colectomies for UC, of which 16 were open procedures, again found complications in nearly 50% of patients.²⁹ A 26 y experience looking at 30 initial STC-I for UC from 1983-2009 found complications in 33% of patients³⁰; however, advancements in

Table 3 – Early and late complication rates.

Complication	Early (<60 d postoperative)	Late (>60 d postoperative)
Total*	9 (24.3%)	9 (24.3%)
Readmissions	8 (21.6%)	8 (21.6%)
Venous thromboembolism	2 (5.4%)	N/A
Small bowel obstruction	2 (5.4%)	5 (13.5%)
Intestinal perforation	1 (2.7%)	N/A
Rectal stump dehiscence	3 (8.1%)	N/A
Mortality	1 (2.7%)	N/A

N/A = not applicable.

* Fourteen (37.8%) patients experienced a complication; some patients experienced multiple complications.

the medical treatment of IBD and the resultant changes in the patient population undergoing operative intervention in the past several decades may render these data outdated. Akin to these findings, overall, 37.8% of our patients experienced a complication.

Although a recent meta-analysis was not able to definitively recommend a laparoscopic approach for the surgical management of refractory pediatric IBD, overall data suggested that laparoscopic operations were associated with decreased postoperative pain and length of stay.³¹ Similarly, our data demonstrated reduced length of stay with a laparoscopic approach. We did not demonstrate a decrease in overall complications in patients undergoing a laparoscopic procedure, although it is not apparent if this is because of an underpowered study or a true equivalency between open and laparoscopic methods. Previous studies comparing the complication rates of open versus laparoscopic procedures for pediatric IBD demonstrate conflicting results; this is unsurprising as many studies are difficult to compare, often examining heterogeneous populations (a diagnosis of UC or Crohn disease) and different operations between studies.³¹ Our data did suggest a decrease in the number of unplanned reoperations needed with an initial laparoscopic approach.

We found improved nutritional status after STC-I. This portends significant implications for the subsequent restoration of enteral continuity, as hypoalbuminemia is an independent risk factor for the development of surgical site infection after gastrointestinal surgery³² and adequate preoperative nutrition has been shown to reduce rates of anastomotic leak, both in IBD³³ and in the general surgical population.^{34,35} In addition, as indicated by the significant increase in postoperative hematocrit, an STC-I was sufficient to stop or reduce colonic bleeding.

Our study is limited by its retrospective nature and relatively small sample size. However, a dearth of data exists regarding the outcomes of initial STC-I in the management of pediatric IBD, and this work expands on the previous literature. We have demonstrated that use of STC-I as an initial procedure in the treatment of refractory IBD-related colitis in children is a reasonable surgical approach with complication rates that are inherent to the patient population. In addition, we have demonstrated an improvement in postoperative nutrition, hematocrit, and reduction in immunosuppressive medications. Implementing a laparoscopic approach to subtotal colectomy provides further benefit by reducing postoperative length of stay. Future follow-up studies are needed to further examine the long-term outcomes of pediatric patients with an STC-I first approach in the management of refractory IBD.

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and data analysis and interpretation was done by N.-L.D., M.P.K., and C.L.K. Manuscript was drafted by N.L.D. and critical revision was performed by all authors.

Disclosure

The authors report no proprietary or commercial interest in any product mentioned or concept discussed in this article.

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