



# Transabdominal re-do pouch surgery in pediatric patients for failed ileal pouch anal anastomosis: a case matched study

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## Abstract

**Purpose** Data regarding safety and feasibility of re-do ileal pouch anal anastomosis (IPAA) for failed ileal pouch in children are limited. In this study, we compared the short- and long-term outcomes of re-do IPAA in pediatric and adult populations in a case-matched setting.

**Methods** Between March 2007 and June 2017, pediatric patients undergoing a transabdominal re-do IPAA by single surgeon were reviewed and case matched with adult counterparts. Short- and long-term outcomes including complications, functional outcomes, and quality of life of the two groups were compared.

**Results** 60 patients were included (pediatric,  $n = 30$ ; adult,  $n = 30$ ). Time between index IPAA and re-do IPAA was shorter in the pediatric group ( $30 \pm 26$  vs  $86 \pm 74$  months,  $p = 0.001$ ). In the pediatric population, the existing pouch was more commonly used to construct the re-do pouch ( $n = 19$  vs  $n = 12$ ,  $p = 0.07$ ). There was a trend towards the presence of less postoperative complications in pediatric group ( $n = 13$  vs  $n = 20$ ,  $p = 0.07$ ). There were no reoperations or mortality. Long-term pouch survival was comparable between two groups ( $p = 0.96$ ). Six re-do IPAA failed in the study period.

**Conclusion** Re-do IPAA is safe and feasible in pediatric population with failed IPAA and can be performed with similar short- and long-term outcomes compared to adults in experienced hands.

**Keywords** Ileal pouch anal anastomosis · Inflammatory bowel disease · Pediatric · Pouch failure · Redo pouch · Salvage surgery

## Introduction

Restorative proctocolectomy and ileal pouch anal anastomosis (RP/IPAA) is the surgical technique for treatment of mucosal ulcerative colitis (MUC), familial adenomatous polyposis (FAP), and selected cases of Crohn's disease (CD) [1]. It restores intestinal continuity with an acceptable functionality and good quality of life (QoL) [2]. Despite improved medical treatment strategies and increasing experience with the ileal pouch anal anastomosis (IPAA) procedure, up to 10% of the IPAA fail in long-term follow-up [3].

Re-do IPAA is the only option which may give such patients a chance to maintain their intestinal continuity. Due to the fact that revision or salvage of a failed IPAA is a complex and technically demanding procedure, few numbers of institutions and surgeons have gained unique experience on this type of surgery.

While it is known that outcomes of IPAA surgery may vary in adults and pediatric patients [4, 5], no data comparing the outcomes of re-do IPAA surgery in those patient populations have been reported. This is the first study evaluating the short- and long-term outcomes in pediatric versus adult patients undergoing transabdominal re-do IPAA for failed IPAA in a case-matched setting.

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## Methods

Between March 2007 and June 2017, patients undergoing a transabdominal re-do IPAA for a failed IPAA by single surgeon were evaluated. The institutional review

board approved the study protocol. Pediatric patients were case matched with adult patients based on final diagnosis and indication for re-do IPAA with a one-to-one fashion. Upper age limit of pediatrics were identified as 21 years old according to American Academy of Pediatrics' statement [6].

After case match was completed, patient characteristics (age, sex, and body mass index), primary pouch characteristics (pouch type, previous anastomosis type, and modality of the previous pouch surgery), time between index pouch and re-do pouch surgery, presence of diversion prior to re-do surgery, presence of new pouch creation, configuration of the re-do pouch, anastomosis type of the re-do pouch, pouch survival rates, patients' quality of life (QoL), functional outcomes, and short-term and long-term complications were compared between two groups.

### Perioperative work-up and follow-up

Patients undergo a complete clinical investigation prior to redo IPAA surgery. A detailed and accurate history of the presenting symptoms is taken. Operative records and pathology reports are reviewed. The following tests are being requested if needed: Exam under anesthesia, flexible pouchoscopy, gastrografin enema (GGE), magnetic resonance imaging (MRI), computed tomography (CT) or CT enterography, and anal physiology testing. Diverting ileostomy is created 6 months prior to re-do IPAA surgery minimum 20 cm proximally from tip of the existing pouch. Operative details were described in detail previously [7].

Patient-controlled analgesia is used for all the patients after surgery. Patients are allowed to ambulate and take oral liquids on the same day of surgery and use a spirometer. Patients who can tolerate liquids are offered solid food and are given oral analgesics. The urinary catheter is removed around postoperative day 1. Discharge criteria are similar in each group and included tolerance of meals without nausea or vomiting, established bowel or stoma function, adequate pain management with oral analgesics, and independent ambulation [8].

### Assessment of functional outcomes and quality of life

Patients' functional outcomes and quality of life (QoL) were evaluated during office visits. Frequency of bowel movements, daytime/nighttime seepage, pad use, and restrictions were assessed. Anal seepage, pad usage, and restrictions were simply recorded as being present or not (0 = no, 1 = yes). Quality-of-life assessment was performed using the Cleveland Global Quality of Life (CGQL) score, which includes three items: current QoL, current health, and current level of energy, each on a scale of 0–10 (0, worst; 10,

best). The scores are added and the final CGQL utility score is obtained by dividing the resulting number by 30 (0, worst; 1 best) [9].

### Definitions

Short-term follow-up time was defined as the first 30 days after re-do IPAA creation. Overall short-term morbidity rate was calculated by dividing the number of patients who had at least one postoperative complication by the total number patients undergoing surgery. Re-do pouch failure was defined as excision of the ileoanal pouch or permanent/definitive diversion with a proximal ileostomy [10]. Anastomotic leak was defined as a break in the integrity of the anastomosis, as documented by the combination of clinical, radiologic, and/or operative means [2]. Bowel obstruction was defined as the presence of at least three of the following five symptoms: nausea, abdominal pain, vomiting, abdominal distension, absence of flatus and/or stool within the last 24 h, findings indicating obstruction upon plain radiographic or contrast studies, or a diagnosis of intestinal obstruction as confirmed by surgery [2]. A clinically significant anastomotic stricture was defined as one requiring treatment. Pouch-related fistula was defined as an abnormal passage or sinus from the pouch to another surface or organ. Pouchitis was defined relying on symptoms including increased stool frequency, urgency, tenesmus, incontinence, nocturnal seepage, abdominal cramping, pelvic discomfort with high fever, dehydration, and malnutrition. Surgery was considered only in totally medical therapy refractory pouchitis cases [11].

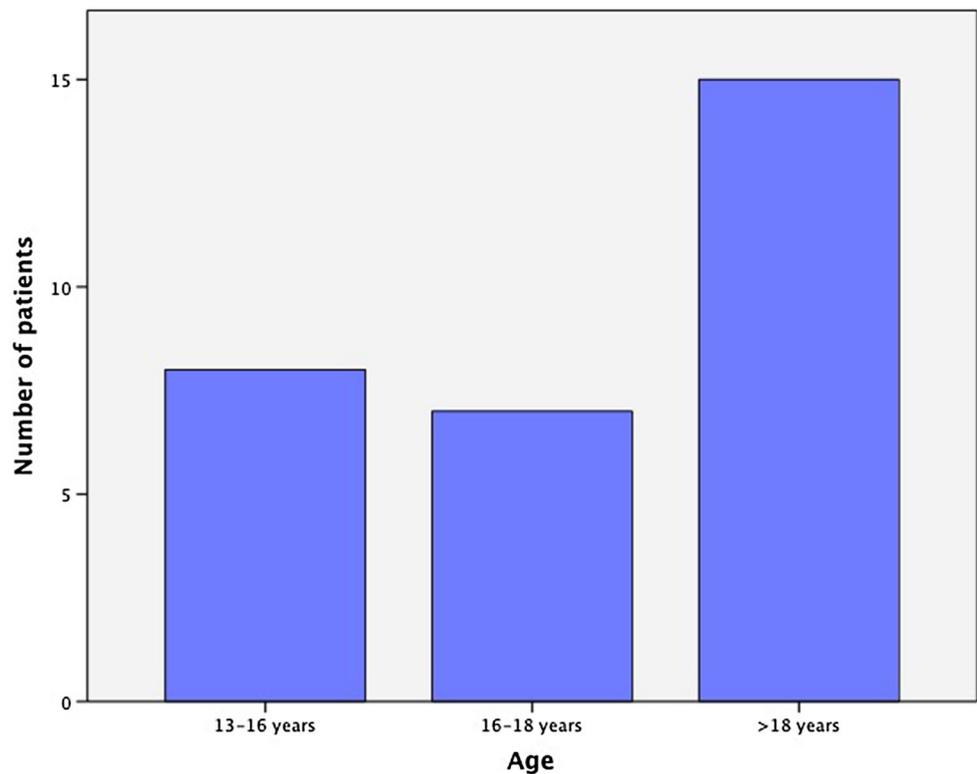
### Statistical analyses

Categorical variables and continuous variables were given as frequency (%) and mean  $\pm$  SD, respectively. Categorical variables were compared via Fisher's exact test or Chi-square test, and continuous variables were compared via independent *t* test or Mann–Whitney *U* test considering the normality. For pouch survival estimate comparison between two groups, Kaplan–Meier method with log-rank test was applied. Statistical significance was accepted when the 'p' value < 0.05.

### Results

Thirty pediatric patients (Fig. 1) were case matched with 30 adult counterparts. Primary diagnoses were ulcerative colitis ( $n = 56$ ), Crohn's disease ( $n = 2$ ), and familial adenomatous polyposis ( $n = 2$ ). Indications for surgery were leak/fistula ( $n = 40$ ), pouch dysfunction without preoperative definitive diagnosis ( $n = 10$ ), obstruction ( $n = 8$ ), and pouchitis ( $n = 2$ ) (Table 1). There were 14 and 20 females in pediatric and

**Fig. 1** Age distribution in pediatric group undergoing re-do IPAA



**Table 1** Re-do procedure indications that were used in case-matching process

	Pediatric (n=30)	Adult (n=30)
Indications, n (%)		
Leak/fistula	20	20
Pouch dysfunction without definitive preoperative diagnosis	5	5
Obstruction	4	4
Pouchitis	1	1

**Table 2** Patient characteristics between pediatric ( $\leq 21$ ) and adult ( $> 21$ ) age groups

	Pediatric (n=30)	Adult (n=30)	p
Age (years), mean $\pm$ SD	18 $\pm$ 2	38 $\pm$ 10	< 0.0001
Sex (female), n (%)	14 (47%)	20 (67%)	0.12
BMI (kg/m <sup>2</sup> ), mean $\pm$ SD	22 $\pm$ 4	23 $\pm$ 3	0.26
Diagnosis, n (%)			
MUC	28 (93%)	28 (93%)	
Crohn’s disease	1 (3%)	1 (3%)	
FAP	1 (3%)	1 (3%)	

SD standard deviation, BMI body mass index, MUC mucosal ulcerative colitis, FAP familial adenomatous polyposis

adult groups, respectively ( $p=0.12$ ) (Table 2). Primary pouch configuration, previous anastomosis type, and modality of previous surgery were comparable (Table 3). Time between index IPAA and re-do IPAA was shorter in pediatric group ( $30 \pm 26$  vs  $86 \pm 74$  months,  $p=0.001$ ). 24 and 26 patients were diverted prior to re-do IPAA in pediatric and adults, respectively ( $p=0.49$ ). All other patients who had no prior diversion had a diverting loop ileostomy during re-do IPAA surgery.

In 12 pediatric and 19 adult patients, primary pouch was reserved and used to construct the re-do pouch; there was a trend towards statistical significance ( $p=0.07$ ) (Table 4). There were three S pouches in pediatric group, while no S pouches were performed in adult group ( $p=0.24$ ). Short-term ( $\leq 30$  days) complications were comparable between the groups (Table 5). There were no reoperations. There was a trend towards the presence of less postoperative complications in pediatric group ( $n=13$ ,  $n=20$ ,  $p=0.07$ ). Re-do IPAA failed in six patients due to the following reasons: anastomotic leakage ( $n=4$ ), stricture ( $n=1$ ), and pouch prolapse ( $n=1$ ). 5-year survival estimation was 90% (CI 79.2–100) in both groups ( $p=0.96$ ). Survival curve is depicted (Fig. 2). Median follow-up for pediatric and adult patients was 47 (IQR 28.5–67) and 59 (IQR 34.75–76.5) months, respectively ( $p=0.12$ ). Similar long-term functional outcomes and QoL scores were observed in both study groups (Table 6).

**Table 3** Comparison of primary pouch characteristics in between two groups

	Pediatric ( <i>n</i> = 30)	Adult ( <i>n</i> = 30)	<i>p</i>
Previous pouch type, <i>n</i> (%)			0.42
J pouch	28 (93%)	25 (83%)	
S pouch	2 (7%)	5 (71%)	
Previous anastomosis, <i>n</i> (%)			
Stapled	83%	75%	
Handsewn	17%	25%	
Missing <sup>a</sup>	17	10	
Previous pouch surgery modality, <i>n</i> (%)			0.29
Minimally invasive	11 (42%)	8 (29%)	
Open	15 (58%)	20 (71%)	
Missing	4	2	
Time between index pouch and re-do pouch surgery (months), mean ± SD	30 ± 26	86 ± 74	0.001

*SD* standard deviation

<sup>a</sup>Missing data are the result of unavailability to reach postoperative reports

**Table 4** Comparison of the re-do operative characteristics in between two groups

	Pediatric ( <i>n</i> = 30)	Adult ( <i>n</i> = 30)	<i>p</i>
Diversion prior to re-do, <i>n</i> (%)	24 (80%)	26 (87%)	0.49
New pouch creation, <i>n</i> (%)	12 (39%)	19 (61%)	0.07
Re-do pouch type, <i>n</i> (%)			0.24
J pouch	27 (90%)	30	
S pouch	3 (10%)	0	
Re-do anastomosis, <i>n</i> (%)			0.74
Handsewn	25 (83%)	24 (80%)	
Stapled	5 (17%)	6 (20%)	
Postoperative morbidity, <i>n</i> (%)	13 (43%)	20 (67%)	0.07
Postoperative mortality, <i>n</i> (%)	0	0	

**Table 5** Short-term (≤ 30 days) complications

	Pediatric ( <i>n</i> = 30)	Adult ( <i>n</i> = 30)	<i>p</i>
Anastomotic leakage, <i>n</i>	7	8	0.77
SSI, <i>n</i>	3	4	0.69
Ileus, <i>n</i>	1	2	1
Stomal complications, <i>n</i>	0	2	0.49
Thrombophlebitis, <i>n</i>	1	0	1
Pulmonary, <i>n</i>	0	3	0.24
Urinary, <i>n</i>	1	4	0.35
Transfusion, <i>n</i>	1	1	1
Pouchitis, <i>n</i>	1	0	1

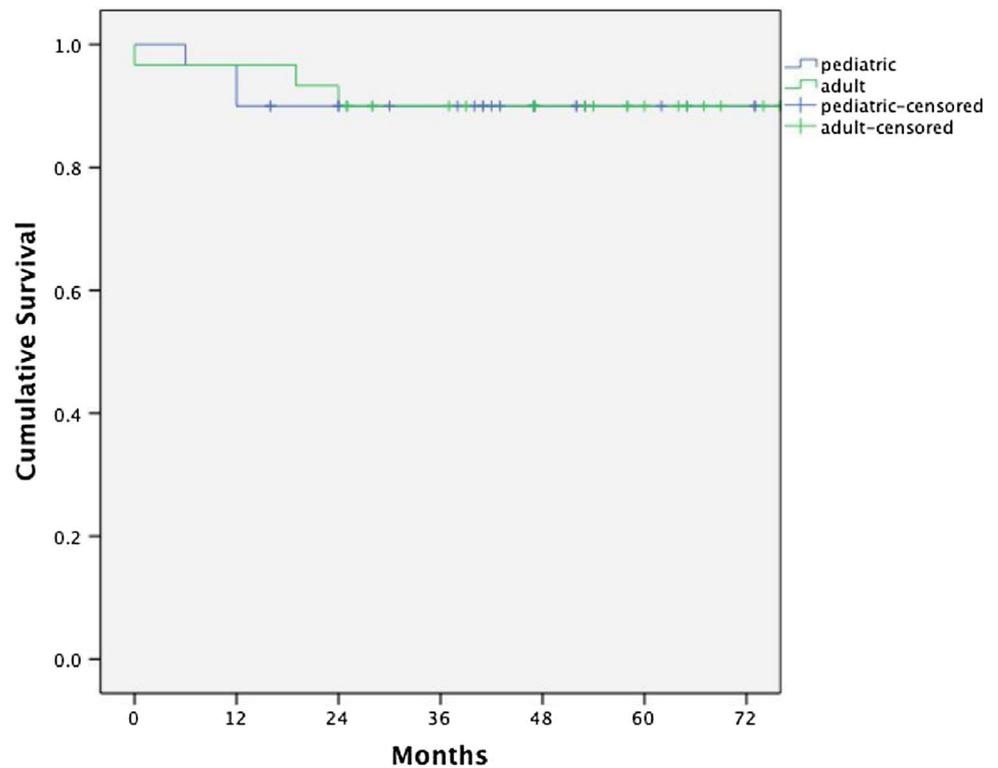
SSI superficial skin infection

## Discussion

Although there is an ongoing debate about timing of index IPAA for children, the aggressive course of pediatric inflammatory bowel disease might necessitate ileal pouch construction at early ages [12, 13]. However, the complexity of the procedure will ultimately conclude with some failures of the IPAA [11]. Re-do surgery is the only treatment of choice to maintain intestinal continuity and to avoid lifelong stoma in patients with failed IPAA. Our results reveal that transabdominal re-do surgery for failed IPAA is a safe and feasible option providing acceptable short- and long-term outcomes in both pediatric and adult patients.

Success of IPAA surgery in pediatric patients is highly related to operative experience and good patient selection due to risk of morbidity [5, 14]. Re-do IPAA surgery in children is expected to be more complicated and requires additional care due to fragile nature of those patients. As a technically demanding procedure, re-do IPAA can be associated with various short- and long-term complications. Anastomotic leak and surgical site infection were the most common postoperative short-term complications in our pediatric patients. Wu et al. [5] showed increased postoperative complications and stricture risk after primary IPAA surgery in pediatric population compared to adults in long term. Pediatric age was reported to be associated with stricture formation after IPAA construction in another study [4]. Hand-sewn anastomoses, proximal diversion prior to index IPAA surgery, postoperative complications including anastomotic leakage, and pelvic sepsis were shown to be associated with stricture formation [15–17]. Our study groups were comparable in terms of those risk factors. Clinically significant stricture was similar in both of our pediatric and

**Fig. 2** Kaplan–Meier survival plot for re-do IPAAs



**Table 6** Long-term complications and functional outcomes at the time of most recent follow-up

	Pediatric ( <i>n</i> =30)	Adult ( <i>n</i> =30)	<i>p</i>
Leak/fistula, <i>n</i>	0	3	0.24
Parastomal hernia, <i>n</i>	1	0	1
Pouch prolapse, <i>n</i>	1	0	1
Stricture, <i>n</i>	2	3	1
Functional results			
Bowel movements, mean ± SD			
Daytime <sup>a</sup>	6 ± 3	6 ± 3	0.86
Nighttime <sup>a</sup>	2 ± 1	2 ± 1	0.74
Seepage, <i>n</i>			
Daytime	16	15	0.8
Nighttime	19	17	0.6
Pad usage, <i>n</i>			
Daytime	13	13	1
Nighttime	20	18	0.59
Restrictions, <i>n</i>			
Dietary	11	10	0.79
Social	6	5	0.74
Work	6	6	1
Sexual	7	7	1
CGQL	0.6 ± 0.2	0.7 ± 0.2 <sup>a</sup>	0.45

CGQL Cleveland Global Quality of Life Score

<sup>a</sup>Decimals rounded to the nearest tenth

adult patients. Hand-sewn anastomosis is the fundamental step to achieve IPAA in re-do surgery, since stapled-IPAA is not doable in majority of the cases. We usually prefer proximal diversion with a loop ileostomy 3–6 months prior to the re-do IPAA surgery due to its benefits on controlling ongoing pelvic sepsis and massive dilation of the small bowel. Creation of hand-sewn anastomosis and proximal diversion with a loop ileostomy are needed for complex conditions related to IPAA failure. As a logical strategy, we strongly suggest three-stage approach for managing IPAA failure due to infectious and mechanical reasons.

Use of minimally invasive techniques for proctocolectomy was more frequent in our pediatric patients compared to their adult counterparts. Laparoscopic technique offers similar postoperative outcomes, pouch functions, and long-term QoL compared to open RP-IPAA in pediatric patients [11]. Although laparoscopic proctocolectomy seems to be superior in terms of postoperative recovery and preservation of fertility compared to open technique, some technical issues including twists in pouch mesentery and long rectal remnant cuff may be seen more commonly after laparoscopic restorative proctocolectomy [3, 18, 19]. Unless cardinal steps evaluating the position and the tension of the mesentery are checked properly, any type of minimally invasive restorative proctocolectomy should be abandoned. The cut edge of the small bowel mesentery should be straight and pass up and to the right toward the stump of the ileocolic artery during IPAA construction.

The staple line forming the ileal pouch should be straight and anteriorly located lining right down to the anastomosis. While there are no measurable data, anterior placement of pouch mesentery may provide extra length to create a tension-free anastomosis. Currently, safety and feasibility of both robotic and laparoscopic IPAA have been shown [20, 21]. Depending on the complexity of the IPAA failure, minimally invasive techniques can be used selectively for managing ileal pouch failures in experienced hands [22, 23]. In our series, all of the definitive steps of the re-do IPAA surgery were performed with the open technique due to their complicated nature. Preserving the bowel reserve with acceptable functional outcomes and maintaining the intestinal continuity should be the essential aim of the re-do IPAA operations which require extensive pelvic dissection and adhesiolysis.

In our study, there was a tendency to use the old pouch to construct a re-do pouch more commonly in pediatric population compared to the adults. Minimizing the loss of small bowel may facilitate further re-do pouch construction if needed in pediatric patients who have longer life expectancy and potential recurrent problem probability. The decision whether to use prior ileal pouch or construct a new one usually depends on viability of the index pouch and ability of the re-do pouch to reach anus without tension. Salvage or replacing of an ileal pouch provides similar long-term outcomes when a sound assessment performed at the time of redo IPAA surgery [24–26].

While the severe nature of the pediatric inflammatory bowel disease may be a preconditioning factor for eventual IPAA failure, postsurgical stress mechanisms are also different in children [27, 28]. Structurally, ongoing growth in pediatric pelvis in width may adversely affect IPAA functions and outcomes [29]. Index IPAA survival was shorter in pediatric patients of our study population. Pediatric patients who have ileal pouch failure desire to maintain their intestinal continuity and expect acceptable QoL due to their long-life expectancy. Our results showed transabdominal re-do IPAA surgery in pediatric patients provide comparable pouch survival, functional outcomes, and QoL with adult patients. An average number of bowel movements of 6 per day and 1 or 2 per night bowel movements with an acceptable daily life restriction directly reveal that re-do IPAA functions and QoL directly depend on patients' tolerance and expectations. Around one-third of patients had dietary and the other one-fifth of patients had social, work, or sexual restrictions. Those functional outcomes seem satisfactory for patients who desire to avoid a permanent stoma for any cost. Having hand-sewn anastomosis and a history of multiple pelvic operations with complications resulting in IPAA failure are the factors that result in worsening the functional outcomes in pediatric patients undergoing re-do IPAA surgery. In our previous study, we have shown that

stapled-IPAA provides better IPAA functions compared to hand-sewn IPAA [15].

While case-matching helped to create comparable study groups in terms of patients' characteristics, our study has several limitations due to non-randomized and retrospective design. The results of our study cannot be generalized and should be interpreted cautiously due to the fact that our senior surgeon has an expert in re-do IPAA field.

## Conclusions

The findings of our study suggest that re-do IPAA operation is safe and feasible in pediatric population with failed IPAA and can be performed with similar short- and long-term outcomes including gastrointestinal function and QoL compared to adults. This procedure can safely be offered to pediatric patients who have aversion to live with permanent ileostomy.

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## Compliance with ethical standards

**Conflict of interest** None of the authors have potential conflict of interest to disclose.

**Research involving human participants and/or animals** This article does not contain any studies with human participants or animals performed by any of the authors.

**Informed consent** A waiver of consent was taken from the legal guardians of the pediatric patients.

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