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Case Report

Pouch Salvage Surgery for Treatment of Colitis and Familial Adenomatous Polyposis: Report of Five Cases

Saeed Derakhshani,¹ Arash Mohammadi Tofigh,¹ Behzad Nemati Honar,¹ Gholamhossein

Hayatollah,^{1,*} and Maryam Derakhshani²

¹Imam Hossein Hospital, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran

²University of British Colombia, Okanagan, BC, Canada

^{*}*Corresponding author*: Gholamhossein Hayatollah, Imam Hossein Hospital, Shahid Beheshti University of Medical Sciences, Tehran, IR Iran. Tel: +98-9127240298, E-mail: dr.hayatollah@yahoo.com

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Abstract

Introduction: The restorative proctocolectomy (RPC) with ileal pouch-anal anastomosis (IPAA) is currently the preferred surgical method for most patients with ulcerative colitis and familial adenomatous polyposis and sometimes, functional bowel diseases. Infection around the pouch, remaining rectal stump, stricture at anastomosis site, pouch dysfunction and refractory pouchitis can lead to pouch failure. Pouch salvage surgery could prevent pouch failure in some cases.

Case Presentation: In this report, five patients were introduced, who underwent pouch salvage surgery after RPC/IPAA surgery failure. Two of the patients were male and three were female and the relevant age range was 16 to 41. Initially, RPC/IPAA surgery was performed on these five patients. Four of the patients underwent RPC/IPAA surgery as a result of ulcerative colitis and, one of the patients as a result of familial adenomatous polyposis. However, due to pouch failure from the RPC/IPAA surgery, pouch-salvage surgery was performed on each of these five patients. Two of the patients underwent pouch-salvage surgery due to infection and pouch fistula, and the other three underwent this surgery due to the remaining rectal stump, anastomosis stenosis and pouch dysfunction. The average time for when pouch-salvage surgery was performed was 3.5 years (three months to five years) after the initial operation and the patients were under follow-up care for two to seven years.

Conclusions: After performing pouch salvage operation, pouch function was acceptable in all patients and we could close ileostomies of all of them.

Keywords: Familial Adenomatous Polyposis, Ilioanal Pouch, Sepsis, Total Proctocolectomy, Ulcerative Colitis

1. Introduction

Today, restorative proctocolectomy with ileal-pouch anal anastomosis is a preferred surgical technique for most patients with ulcerative colitis and familial adenomatous polyposis (1, 2). Sometimes this method is also applied for patients with bowel dysfunction (3). Over the last two decades, this surgical technique and subsequent management has been substantially improved, and the functional results, complications and failure rates have been analyzed as well (4, 5). For those with five years of followup care, treatment failure has been reported as 17% (6, 7). The cumulative failure rate of pouch surgery was 15% within 10 to 15 years of follow-up. Pouch removal, permanent ileostomy or long-term bowel diversions have thus been inevitable. Treatment failure could occur at an earlystage, within the first postoperative year or later. Age is not a counter-indication for pouch surgery and does not increase the risk of pouch surgery failure (8). The most common complication (for 50% of cases) that results in surgical failure is sepsis (9). Among other factors, functional or

mechanical disorders of the pouch, inflammation of mucosal layer (inflamed pouch or remnant rectal stump), and neoplastic changes could be mentioned. In such conditions, pouch-salvage surgery with an attempt to solve the cause of failure could avoid permanent ileostomy or bowel diversion. Lifesaving abdominal surgical techniques have been successful for 20% - 80% of cases (depending on the follow-up period). Success rate has been 50 to 60% for perineal approaches such as pouch-vaginal fistula repair and 70% for abdominal surgery in cases of obstruction (9). In the current report, we present the results of five patients with colitis and familial adenomatous polyposis treated with pouch salvage surgery.

2. Case Presentation

2.1. Patient 1

This case was a 47-year-old male, who underwent a restorative proctocolectomy (RPC) with ileal pouch-anal anastomosis (IPAA) surgery as result of ulcerative colitis.

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Five months after the surgery, he underwent ileostomy creation due to the anastomotic stenosis and symptoms of ileus. Three years later, he was diagnosed by remnant long rectal stump and the anastomotic stenosis and symptoms of mucus discharge, and fecal incontinence. He then underwent pouch mobilization abdominal surgery, mucosectomy for the remnant rectal mucosa, pouch-anal anastomosis and diverting ileostomy. Three months later, ileostomy closure was performed. Fecal seepage was the only symptom, which still exists after three years of followup. However, it does not cause any disruption to the patient's life and work, and his quality of life has improved significantly.

2.2. Patient 2

This case was a 32-year-old male, who had RCP and IPAA for refractory treatment ulcerative colitis. Three months after the surgery, the patient referred to our clinic with symptoms of mucus discharge and obstructed defecation (70 times inadequate defecation per day). Pouchoscopy indicated anastomotic stricture and inflammation. He underwent pouch-salvage surgery (removal of the remnant rectum, mucosectomy, pouch re-anastomosis + ileostomy). Twenty days later, he developed mechanical small bowel obstruction due to the adhesive bands, which needed laparotomy and release of adhesions. Then, ileostomy was closed three months later, and within seven years of follow-up, no gastrointestinal symptoms remained.

2.3. Patient 3

The third patient was a 27-year-old female, who underwent RPC/IPAA due to familial adenomatous polyposis. Five days after the surgery, she developed peritonitis due to leakage from pouch-anal anastomosis. Ileostomy was then performed on this case. Three months later, ileostomy closure was performed. Three years later, for recurrent perineal abscess, that was related to pouch-perineal fistula, she underwent multiple fistula repair with the perineal approach; however, because of the lack of response, she underwent abdominal pouch mobilization, mucosectomy, pouch re-anastomosis to anal canal, and ileostomy. Ileostomy closure was performed three months later and then the fistula disappeared. No gastrointestinal symptom was observed during the four years of follow-up.

2.4. Patient 4

The fourth patient was a 40-year-old female, who had undergone RPC/IPAA due to the ulcerative colitis 24 years ago. After the surgery she developed sepsis complications around the pouch that ended to pouch-presacral fistula. Twenty-two years later, due to abscess formation and peritonitis, she underwent laparotomy and ileostomy and her symptoms were controlled. After the second surgery, the pouch-presacral fistula had remained. As a result of pouch failure, the patient underwent pouch resection surgery, creating a new pouch and ileostomy. The ileostomy closure was performed three months later and currently she does not have any gastrointestinal problems after seven years since her last surgery.

2.5. Patient 5

The last patient was a 35-year-old female, who underwent RPC/IPAA due to ulcerative colitis. After the operation, she developed intestinal obstruction due to severe stenosis at pouch anal anastomosis. For this reason, she underwent surgery to create ileostomy. Considering lower abdominal pain due to the mucus accumulation of the pouch, she underwent dilatation and transanal strictureplasty surgery and finally ileostomy was closed after three months. Currently, after two years, she has no stricture or gastrointestinal symptoms.

3. Discussion

Pouch salvage surgeries can avoid RPC/IPAA surgery failure. Pouch salvage operations are divided to two groups: Trans perineal and trans abdominal approaches (8). In the trans abdominal approach, the surgeon mobilizes the pouch, and depending on the pouch condition and small bowel mesentery length, the pouch will be removed or left in place. Then mucosectomy of rectal mucosa down to dentate line will be performed and at the end, hand sewn pouch anal anastomosis will complete the operation. Diverting ileostomy will be performed in most cases. Trans perineal approaches include fistulotomy for very low fistulas, advancement flap or muscle transposition for high fistulas and pouch-vaginal fistulas, or rectal mucosectomy plus pouch mobilization with pouch anal anastomosis for remained rectal mucosa or stricture at pouch anal anastomosis site. Four major complications resulting in pouch surgery failure were observed with sepsis being the most common factor. Sepsis could occur early, immediately after the surgery or with some delay with abscess (specifically, sacral abscess) or fistula (10).

Among other complications, pouch-vaginal fistula could be mentioned, which is more common in patients with ulcerative colitis. It appears in 2.6 to 16% of cases and the diagnosis depends on the follow-up period (11). Mechanical obstruction is another complication for pouch surgery that occurs at distal pouch and sometimes it is functional rather than mechanical. The initial treatment

for mechanical obstruction due to anastomosis stricture is a dilatation and in case of no improvement, re-do pouch surgery will be performed (12). Similar to non-mechanical cases, pouch inflammation is the most common long-term complication for patients with IPAA, which considerably affects life quality (13). The accumulated prevalence is 23% - 46% within the ten post-operative years. Cause of the inflammation is the abnormal mucosal immune response to microbial flora. The simulations are the increased defecation frequency and urgency, stool leakage, and crampy abdominal and pelvic pain. It is possible for extra intestinal symptoms such as joint pain to appear. The first-line treatment is antibiotic therapy and at the next stages or in case of a lack of response to the treatment, salicylate derivatives, corticosteroids and immunosuppressive could be used (14). "Afferent and efferent arm syndrome" could be mentioned as the unique complication. In this syndrome, the movable linear section of the ileum, which exists before the pouch, makes an acute angle with the pouch. For efferent arm syndrome, which is common in cases of spouch, the efferent arm makes an acute angle with the pouch and causes obstructive symptoms. The symptoms of the syndrome include obstructed defecation, and pelvic pain, which can be modified by operation (15). Irritable Pouch Syndrome (IPS) is one of the other complications of IPAA that strongly reduces patient's quality of life. Recent studies indicated that it was more prevalent among consumers of anti-depressants and anti-anxiety medications (16). Irritable Pouch Syndrome diagnosis is based on rejecting causes that could increase defecation times and change consistency of stool, and crampy abdominal pain together with endoscopy and normal pouch histology. Symptomatic treatments include low-fat, low carbohydrate, and caffeine and alcohol free diet. Sometimes, antibiotic therapy improves symptoms by eliminating bacterial overgrowth syndrome (17).

In three out of our five cases remaining rectal stump and rectal mucosa played an important role in pouch failure. It is crucial for surgeons, who are performing pouch surgery to remove the entire rectum and leave only the transitional zone mucosa above dentate line in place to prevent further bleeding, stool frequency and stricture at anastomosis site.

One of the most important causes of pouch failure is Crohn's disease. It can induce pouchitis, infection around the pouch and severe stricture in pouch or other parts of the small bowel. Unfortunately, 50% of these patients will have pouch failure and need to have permanent ileostomy.

In four of our cases patients had diverting ileostomy due to pouch failure and after pouch salvage surgery we could close their ileostomies, which improved their quality of life significantly. It is crucial for surgeons doing pouch surgery to remove the entire rectum and leave only the transitional zone mucosa above dentate line in place to prevent further bleeding, stool frequency stricture at anastomosis site and also development malignancy in the remnant of rectal mucosa.

3.1. Conclusion

Pouch salvage surgery is a technique to avoid RPC/IPAAS failure that may lead to a permanent or longterm ileostomy, which is not acceptable by patients and causes problems for them. As it was mentioned before, and as the results of this report indicate, the life-saving pouch salvage surgery creates a considerable improvement in the pouch function and the patient's quality of life.

Footnote

Authors' Contribution: Acquisition of data: Saeed Derakhshani, Arash Mohammadi Tofigh, Behzad Nemati Honar, Gholamhossein Hayatollah and Maryam Derakhshani; drafting of the manuscript: Saeed Derakhshani, Arash Mohammadi Tofigh, Behzad Nemati Honar, Gholamhossein Hayatollah and Maryam Derakhshani; critical revision of the manuscript for important intellectual content: Saeed Derakhshani, Arash Mohammadi Tofigh, Behzad Nemati Honar, Gholamhossein Hayatollah and Maryam Derakhshani; administrative, technical and material support: Saeed Derakhshani, Arash Mohammadi Tofigh, Behzad Nemati Honar, Gholamhossein Hayatollah and Maryam Derakhshani; study supervision: Saeed Derakhshani, Arash Mohammadi Tofigh, Behzad Nemati Honar, Gholamhossein Hayatollah and Maryam Derakhshani.

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