Contents lists available at ScienceDirect

# International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



# Case report

# A long-term follow-up after open ileal J pouch volvulus repair in a FAP patient – A case report

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### ARTICLE INFO

Keywords: Pouch volvulus Ileal pouch-anal anastomosis FAP J pouch

### ABSTRACT

Introduction: Most patients with familial adenomatous polyposis (FAP) undergo proctocolectomy with ileal pouch-anal anastomosis (IPAA). Pouch volvulus is one of the very rare complication of open IPAA surgery that can mimic small bowel obstruction secondary to adhesion. High index of suspicion and proper investigation is required.

Presentation of case: The patient is a 44-year-old female with FAP who is currently on the mend after a 2-stage total proctocolectomy with ileal pouch-anal anastomosis. Presented eight years after indexed surgery with pouch volvulus, successfully treated with bowel resection without pouchopexy.

Discussion: The surgery of choice for most patients with FAP is IPAA. Pouch volvulus is a rare complication of laparoscopic IPAA surgery and extremely rare after open surgery which can be treated surgically in various ways. But it can be catastrophic if not treated in timely manner. This case report presents a FAP patient with pouch volvulus after open IPAA treated with bowel resection and obliteration of space between the pouch mesentery and the sacral space without standard pouchopexy.

Conclusion: Volvulus is a rare side effect of an IPAA. Managing the initial procedure successfully is critical in order to avoid recurrence and consequences. The goal of this study is to identify symptoms and a successful management strategy for pouch volvulus.

# 1. Introduction

The surgery of choice for most patients with familial adenomatous polyposis (FAP) is proctocolectomy with ileal pouch-anal anastomosis (IPAA). Complications following IPAA occur in 19%-54% of the cases [1]. Pouchitis is one of the most common complications, affecting 18.8% of the patients [2]. On the other hand, pouch volvulus is a rare complication of IPAA surgery that can be treated with various surgical options. This complication has only been mentioned a few times in the literature, with one study reporting it in three out of 1700 patients (0.18%) [3]. Pouch complications have decreased significantly over the last four decades as surgeons have gained more experience [4]. This study presents a case report of a pouch volvulus after IPAA in our department and describes the symptoms of this rare condition and the successful management strategy used to preserve the pouch in a patient with IPAA volvulus. This case has been reported in line with the SCARE criteria [5].

In 1999, a 44-year-old woman with FAP with no past medical or surgical history, underwent open restorative proctocolectomy with a 15 cm doubled-stapled J pouch and a prophylactic loop ileostomy. Three months later, the ileostomy was successfully reversed. She had excellent function with regular follow up by examination and yearly endoscopy, until eight years after surgery, when she began experiencing constipation, discomfort, and nausea over 7 days, progressed acutely within one day to severe abdominal pain and vomiting. On physical examination, body mass index 18, she appeared frail, and apparently in discomfort. Her vital signs were: temperature:37.2 °C; blood pressure: 115/75 mmHg; heart rate: 110 beats per minute; respiratory rate: 20 respirations per minute. A distended abdomen, mild central tenderness with increased bowel sounds were discovered during the clinical examination. Blood tests were normal. An abdominal X-ray revealed a small bowel obstruction in the first diagnostic approach (Fig. 1). A contrast-enhanced CT scan of the abdomen revealed distended loops of

<sup>2.</sup> Presentation of case

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Fig. 1. Abdominal X-ray revealing small bowel obstruction.

the small intestine (Fig. 3). The ileo-anal pouch was also mildly distended (up to 6 cm) with mesenteric fat edema. A pouchogram revealed that there was some obstruction (Fig. 2). Then, the patient was then taken to the operating room by a colorectal surgeon, where she had an emergency exploratory laparotomy. A massive distention of the small bowel was discovered intraoperatively. We thoroughly examined the small bowel, but no adhesions up to the pouch were found.

Furthermore, the pouch was found to be distended, with a  $6\,\mathrm{cm}$  diameter, edematous, and congested and with a  $180^\circ$  anticlockwise



Fig. 2. Pouchogram showed dilated pouch and small bowel that may be due to torsion in the pouch.



Fig. 3. Segmental distention of the small intestine around the staples.

distortion behind the afferent loop. After the pouch was mobilized, resection of the bowel along with mesentery preservation and stapled anastomosis was performed. Moreover, the space between the pouch mesentery and the sacral space was obliterated to prevent future small bowel herniation. That was done with interrupted polyfilament suture being placed between the mesenteric side of the bowel and the sacral promontory in order to close the defect behind the afferent loop of mesentery where the twist found. The standard pouchopexy is to suture the pouch to the pelvic sidewall but here the suture were placed to close the defect behind the afferent loop of mesentery where the twist found, which doesn't make it standard pouchopexy. The reason behind that is to avoid the dissection of the pouch till the previous anastomosis which left intact. After surgery, the patient remained asymptomatic, tolerated a liquid diet well, and was discharged on the fifth postoperative day. She was admitted once one year after the procedure for a conservatively managed intestinal obstruction and then discharged home after 5 days. The patient has not had a recurrence of pouch volvulus after more than thirteen years.

### 3. Discussion

Total proctocolectomy with ileal pouch-anal anastomosis (IPAA) is the preferred surgical treatment for FAP. IPAA is a common procedure used to treat various conditions, including ulcerative colitis, Crohn's disease, indeterminate colitis, and FAP. Overall, a patient's quality of life is greatly improved and satisfactory after surgery, making IPAA the treatment of choice for these conditions [3,6]. Unfortunately, several complications are unique to IPAA patients. Some of the more common septic complications are anastomotic leak, abscess, and fistula, whereas the common nonseptic complications are stricture, pouchitis, and small bowel obstruction. Some of the less common complications are pouch prolapse and volvulus [3]. More than 40 years before the laparoscopic era, proctocolectomy using ileal pouch-anal anastomosis (IPAA) was introduced. Since then, minimally invasive surgery (MIS) technology has been applied to pelvic pouch surgery. One of the advantages of MIS is that it reduces complications related to adhesion. However, lack of adhesion can lead to a range of different complications. Specifically, when the ileal pouch is not adherent to the pelvis, volvulus can occur around the mesenteric axis. That's why the majority of volvulus cases were reported in the last five years [7].

Pouch volvulus is defined as the torsion of the ileal pouch around the mesentery axis or small intestine while maintaining the correct orientation of the ileal anal anastomosis. Twisting of the mesenteric axis due to misaligned anastomosis is not considered volvulus here.

After a thorough PubMed search, it appears that our case is the only reported case of a recurrent volvulus after open pouch surgery with a 20year follow-up period after treating the volvulus. In July 2014, the first case of recurrent volvulus was reported in the literature. Most IPAA volvulus cases have been reported in women with low BMIs and ulcerative colitis who underwent laparoscopic total proctocolectomies with IPAA. The higher prevalence in women is thought to be due to the anatomy of a wider pelvis, which allows for greater pouch mobility [7]. A low BMI and less adipose tissue in the pelvis allow for more space for pouch torsion. Most laparoscopically performed IPAAs result in fewer peritoneal adhesions. Some authors have hypothesized that the position of the mesentery of the pouch could facilitate pouch volvulus. In one study, 89 IPAAs were constructed with the mesentery positioned anteriorly, 35 posteriorly, and 27 on the right side. Three patients were diagnosed with an IPAA volvulus. All three patients were in the anterior group [8]. Finally, compared to open surgical procedures, laparoscopic procedures may increase the risk of volvulus due to the lack of adhesions formed [9]; however, in this case, an open surgery was performed. To explain the possible causes in this case, the following questions come to mind: does the way we construct the pouch matter? Moreover, are we creating a slight twist in the pouch when constructing the J-shaped pouch with the long arm of the J on the left side and ileostomy on the right side of the patient? These questions need to be answered.

To the best of our knowledge, this is the first case in the literature to report an open pouch volvulus and its successful treatment without pouchopexy with fourteen years follow-up after surgery.

#### 4. Conclusion

Pouch volvulus is a rare complication that can have serious consequences. Even without pouchopexy, attempting to obliterate the space between the pouch mesentery and the sacral hollow is critical during pouch volvulus repair as this is a potential site of small bowel herniation and, as a result, retorsion. For patients with unexplained abdominal pain, bloating, and other obstructive symptoms, surgeons should be highly skeptical.

# Sources of funding

No source of funding has been declared by the author.

## Ethical approval

Ethical approval was obtained from Research Advisory Council.

# Consent

Written informed consent was obtained from the patient for

publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

## Registration of research studies

Not applicable.

### Guarantor

Dr. Reem A. Alharbi.

### Provenance and peer review

Not commissioned, externally peer-reviewed.

## CRediT authorship contribution statement

The manuscript is written by Dr. Reem Alharbi.

## **Declaration of competing interest**

There are no conflicts of interest or disclosures.

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