

Systematic Review

A rare case of obstructive ileus due to ileal pouch torsion: a case report and systematic review

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ABSTRACT

Restorative proctocolectomy with ileal pouch-anal anastomosis (IPAA) has become the standard surgical treatment for ulcerative colitis patients. Among others, it is important for clinicians to be aware of a rare, though urgent situation, the ileal pouch torsion. A high degree of suspicion is essential as obstruction due to pouch torsion is not likely to resolve conservatively. Delay in diagnosis and treatment can lead to pouch jeopardy and life-threatening complications. The aim of this study was to present a systematic review of the literature combined with the presentation of a case report of our department regarding this rare entity. A systematic literature review according to QUORUM guidelines was conducted in July 2019 using MEDLINE, SCOPUS, and COCHRANE databases. We included studies reporting pouch torsion after IPAA in English or German language with no restriction regarding publication time. Outcomes after different treatment options, such as operative pouch detorsion, and/or pouchpexy, and/or pouch reconstruction, and/or end-ileostomy through laparoscopic or open procedures were evaluated. We identified 170 publications. After duplicates and irrelevant articles have been excluded, 25 publications remained for full-text review. Finally, 12 articles were included in this systematic review, concerning 14 cases. To the best of our knowledge, this is the first comprehensive systematic review on this topic to date.

Keywords: Ileoanal pouch torsion, IPAA complications, Ileal pouch volvulus, Pouch torsion

INTRODUCTION

Ulcerative colitis (UC) is a chronic diffuse nonspecific inflammatory disease that involves the colon. Surgical intervention is needed in 30% of the cases and meanwhile total proctocolectomy (TP) with ileal pouch-anal anastomosis (IPAA) has become the standard surgical treatment. Additionally, total proctocolectomy with IPAA is currently the surgical treatment of choice for familial adenomatous polyposis (FAP) without rectal involvement, indeterminate colitis, and some cases of Crohn's disease, as well.¹ Complications after IPAA occurs in 19% to 54% of the cases.² Small bowel obstruction (SBO) secondary to postoperative adhesions

is one of the most common complications occurring in 18%-23% of patients.^{3,4} On the other hand, there is an extremely rare entity related to the etiology of SBO, namely the ileal pouch torsion described only few times in the literature.

Case report

A 37- years old woman with a history of ulcerative colitis underwent a laparoscopic restorative proctocolectomy with a construction of a 15 cm doubled-stapled J-pouch and a prophylactic loop ileostomy. The ileostomy was reversed uneventfully three months later. She had excellent function until 5 years postoperatively, when she

presented with diffuse, colicky abdominal pain, nausea and retention of gas and feces. The clinical examination revealed a distended abdomen, with increased bowel sounds. Blood tests were normal, apart from leukocytosis (White Blood Cells: 14.880). The first diagnostic approach with abdominal X-ray revealed a small bowel ileus (Figure 1).

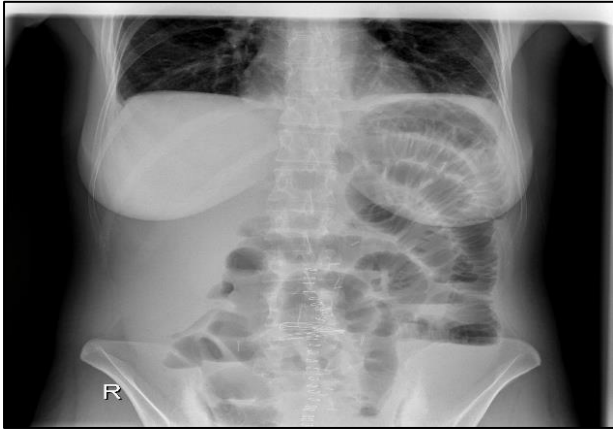


Figure 1: Abdominal X-ray revealing small bowel ileus.

The abdominal contrast-enhanced CT scan revealed distended loops of small intestine (Figure 2).



Figure 2: Segmental distention of the small intestine around the staples.

The ileo-anal pouch was also distended (up to 9 cm) with contemporary edema of the mesenteric fat (Figure 3).

A flexible pouchoscopy revealed complete obstruction without signs of ischemia. Subsequently, the patient was taken to the operating room, where she underwent an emergency exploratory laparotomy. Intraoperatively, a massive distention of small bowel and stomach was identified. We proceeded with a detailed examination of the small bowel, but no adhesions up to the pouch were

detected. In addition, the pouch was recognized distended, with a diameter of 10 cm, edematous, congested and with a 180° anti-clockwise distortion behind the afferent loop. After a careful adhesiolysis, the pouch was mobilized, repositioned and stabilized with 2/0 Vicryl sutures on both sides of the pelvis. Postoperatively, the patient remained asymptomatic, tolerated liquid diet well and was discharged on the 5th postoperative day.

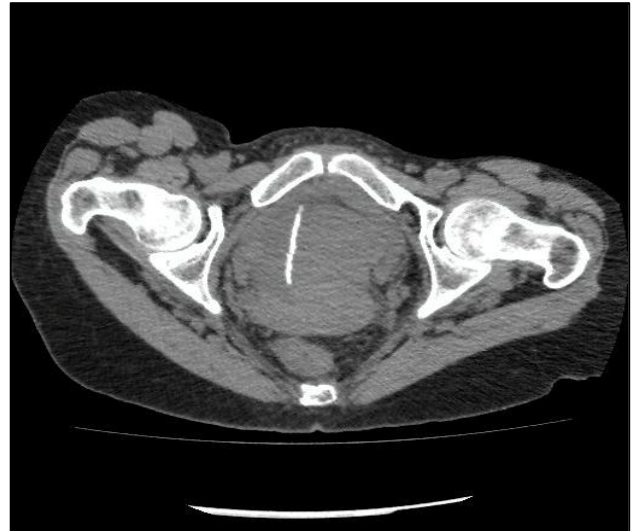


Figure 3: Distended ileo-anal pouch with contemporary edema of the mesenteric fat.

METHODS

Search strategy, study identification, and study selection process. This review is reported according to the Guidelines for the Quality of Reporting of Meta-analyses (QUORUM).⁵ A systematic review was conducted in the PubMed version of MEDLINE, SCOPUS and the COCHRANE database on 27/07/2019, of all the cases of ileal pouch torsion, by using different synonyms, Medical subject headings (MesH) and Emtree terms (for PubMed, Scopus and Cochrane Database respectively).

We included only studies in English or German language and published material with available data in print or on the web in full text. The time of publication was not considered as a restriction for our study. Details of the methods and materials of the literature review and the search strategy are mentioned briefly in Figure 4. Thus, the Cochrane Library was screened for all published trials indexed as pouch torsion. We performed intentionally a wider literature search with the aim not to overlook potentially relevant cases.

After duplicates were extracted the remaining record titles and abstracts were screened thoroughly. Full-text articles were obtained and evaluated in detail. The screening and selection process were performed by two investigators. Discrepancies were resolved by discussion.

The flow diagram summarizes the screening process (Figure 4).

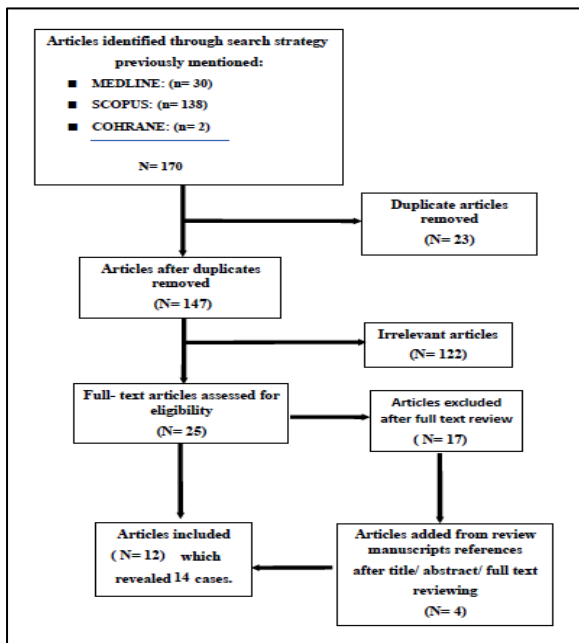


Figure 4: Flow diagram of screening process.

RESULTS

We identified 170 publications, of which 23 duplicates were removed, leaving 147 publications to be screened on the title and abstract. According the predefined criteria 122 articles were excluded, resulting in 25 publications for full text evaluation. Seventeen of them were also excluded as not relevant. The remaining 8 articles were included to the study. Moreover, 4 relevant articles were identified through the reference list of the reviews and were also included to this systematic review. Finally, 12 articles were included to this systematic review, revealing 14 cases. The characteristics of the included studies are presented in Table 1.

Surgical technique: recurrence

According to Table 1, six out of 14 cases were treated with a pouchpexy (2 laparoscopic vs. 4 open). One patient treated with an open pouchpexy with one row of interrupted non-absorbable sutures was readmitted to the hospital 2 months later with a recurrence of the volvulus. This time patient was treated with a new pouchpexy, but with 2 rows of continuous multifilament sutures. Ten months later the patient remained asymptomatic. Additionally, in the 2 cases of laparoscopic pouchpexy reported from Lee et al. and Brady et al. pouch was fixed to bilateral presacral tissue with interrupted sutures.^{6,7} Both patients had an uneventful recovery and remained asymptomatic during their follow up. Furthermore, in 3/14 cases there was a successful endoscopic detorsion of the pouch followed by insertion of a flatus tube, but patient's symptoms recurred during the next 24 hours.

Subsequently, a volvulus recurrence was diagnosed, resulting in an emergency surgical procedure.

Time to diagnosis: outcome

The preoperative diagnosis of this entity is not simple. Time needed to diagnosis and detorsion of the pouch is essential for pouch salvage. Of the 14 cases included to this systematic review, four cases required a pouch reconstruction due to ischemia of the pouch. However, time needed to diagnosis and definitive treatment is not mentioned from the authors. Furthermore, in one case where the patient was misdiagnosed and treated conservatively for over 82 hours resulting in deterioration, pouch was found to be gangrenous and perforated at its upper part. Due to reduced clinical status of the patient, in presence of metabolic acidosis, pyrexia and distension of the small bowel, the pouch was excised and an end-ileostomy was performed.⁸

DISCUSSION

Nowadays, ulcerative colitis has an annual incidence between 9 and 12 cases per 100,000 persons⁹, depending on the region studied and is similar between men and women.⁹ It can be presented with different symptoms such as diarrhea, abdominal pain, fever, weight loss and hematochezia. Thus, there are extraintestinal manifestations of the disease such as arthritis, uveitis, pyoderma gangrenosum, erythema nodosum, aphthous stomatitis, ankylosing spondylitis, primary sclerosing cholangitis and psoriasis.¹⁰

Despite the development and application of new medical agents as part of conservative treatment, there are three absolute indications for surgery: exsanguinating hemorrhage, frank perforation and documented or strongly suspected carcinoma.¹¹ Other relevant indications for surgery are severe colitis with or without toxic megacolon unresponsive to conventional maximal medical therapy and less severe but medically intractable symptoms or intolerable medication side effects.¹¹

Total proctocolectomy and ileal pouch-anal anastomosis is the gold standard surgical treatment for refractory ulcerative colitis. The percentage of patients who may experience surgical complications after IPAA ranges from 30% up to 60%.^{12,13} and the reported mortality related to pouch complications fluctuates between 3,5% and 17%.^{12,14} Small bowel obstruction (SBO) after ileal pouches occurs in 18% to 23% of cases³ with postoperative adhesions being the most common reason.^{4,12} Non- adhesive SBO of ileal pouches can occur due to postoperatively resulting anatomic changes, mostly located in pelvic region.¹⁵ The pouch may become stretched and redundant, flipping over itself causing obstruction. Furthermore, it is not clear yet, if certain pouch configurations are more prone to torsion than others.¹⁶

Table 1: Cases of ileal pouch torsion reported in the literature.

Authors	Type	Title	Journal/ DOI	No. of cases	Type of pouch	Type of torsion	Time after primary surgery (years)	No. of episodes	Time to operation (hours)	Operative procedure	Outcome
Arima et al ¹⁷	Case report	Volvulus of an ileal pouch-rectal anastomosis after subtotal colectomy for ulcerative colitis: report of a case	Surg Today (2014) 44:2382-2384 DOI 10.1007/s00595-013-0724-0	1	J- Pouch	360°	15	0	12	Open Pouchpexy	Uneventful
Brady et al ⁷	Video vignette	Laparoscopic Detorsion of an Ileal Pouch and Pouch Pexy	Dis Colon Rectum 2017;60:24 DOI: 10.1097/DCR.0000000000000698	1						Lap. Pouchpexy	Uneventful
Cárdenas et al ¹⁸	Letter to the editor	Recurrent volvular herniation of the ileal pouch: a case report and literature review	Int J Colorectal Dis (2016) 31:749-750 DOI 10.1007/s00384-015-2242-6	1	J- Pouch	Through mesenteric defect	9	2		Open Pouch Re-do	Uneventful
Jain et al ¹⁹	Letter to the editor	Volvulus of an Ileal J-pouch	Inflamm Bowel Dis (2010) Volume 16, Number, DOI 10.1002/ibd.20911	1	J- Pouch		~ 2	Recurrent		Open Pouch Redo	Uneventful
Lee et al ⁶	Case report	Laparoscopic rectopexy for recurrent volvulus of J pouch after total proctocolectomy and ileal pouch anal anastomosis	Issue Surgical Practice Surgical Practice (2015) Volume 19, Issue 3, DOI: 10.1111/1744-1633.12072	1	J- Pouch	Over Rectum	3	Recurrent	> 48	Lap. Pouchpexy	Uneventful
Mullen et al ²⁰	Case report	Ileal J-Pouch Volvulus Following Total Proctocolectomy for Ulcerative Colitis	J Gastrointest Surg (2016) 20:1072-1073 DOI 10.1007/s11605-015-3038-2	1	J- Pouch	360°	10	0	> 24	Open detorsion	Uneventful

Continued.

Authors	Type	Title	Journal/ DOI	No. of cases	Type of pouch	Type of torsion	Time after primary surgery (years)	No. of episodes	Time to operation (hours)	Operative procedure	Outcome
Myrelid et al ²¹	Case report	Recurrent Volvulus of an Ileal Pouch Requiring Repeat Pouchopexy: A Lesson Learnt	Hindawi Publishing Corporation Case Reports in Surgery, (2014) Volume 2014, doi.org/10.1155/2014/807640	1	J- Pouch	Along its longitudinal axis	11	Recurrent		Open Pouchpexy	Recurrency/ Reoperation 10 mo. later
Poggioli et al ²²	Original article	Redo Pouches: Salvaging of Failed Ileal Pouch-Anal Anastomose	Dis Colon Rectum 1993;36:492-496.	1		Along its longitudinal axis	2 weeks	0		Open Pouch Redo	Uneventful
Sagar et al ²³	Original article	Disconnection, pouch revision and reconnection of the ileal pouch- anal anastomosis	British Journal of Surgery, (1996) 83, 1401- 1405	3		Along its longitudinal axis				Open detorsion (2)/ Pouch Redo (1)	Uneventful
Tyagi et al ²⁴	Case report	Volvulus of ileal S-pouch: A rare complication of ileal pouch anal anastomoses	International Journal of Surgery Case Reports 5 (2014) 717-719 doi.org/10.1016/j.ijscr.2014.07.005	1	S- Pouch	Upon its axis	5	Recurrent	> 48	Open Pouchpexy + Loop ileostomy	Uneventful
Ullah et al ⁸	Case report	Long-Axis Rotational Volvulus in a W Ileoanal Pouch: An Unusual but Potentially Preventable Problem. Report of a Case	Dis Colon Rectum 2007; 50: 540-543 DOI: 10.1007/s10350-006-0825-6	1	W- Pouch	360°			>82	Open Pouch excision+ End-Ileostomy	
Warren et al ²⁵	Case report	Successful management of ileo-anal pouch volvulus	Colorectal Dis. 2011;13(1):106-7. doi: 10.1111/j.1463-1318.2010.02207.x	1		360°	10		> 24	Open Pouchpexy	Uneventful
Present study	Systematic Review	A rare case of obstructive ileus due to ileal pouch torsion: a case report and systematic review		1	J- Pouch	180°	5	0	3	Open Pouchpexy	Uneventful

However, pouch volume and asymmetric orientation around the longitudinal axis must be considered at the time of its construction, as these could be factors that determine predilection for rotational volvulus.⁸

The absence of adhesions to the pelvic wall, reported from most of the studies included in this systematic review, also explains the high mobility of the pouch, resulting in recurrent episodes of torsion treated non-operatively through endoscopic decompression. However, according to the results of our study, definitive surgical treatment is required, in order to protect the pouch from a possible strangulation in the future. Although the role of prophylactic pouchpexy has not been established yet, in our opinion it should be reconsidered for high volume pouches or for cases with an asymmetry to the longitudinal axis.

Finally, preoperative investigations and correlation with the anamnestic data of the patient are essential in pouch carrying patients, as a delayed diagnosis could be fatal for the viability of the pouch. For this reason, pouch torsion should be included in the differential diagnosis algorithm of SBO symptoms of these patients.

This study has certain limitations. As in all systematic reviews on observational studies, the epidemiological conclusions are limited, with a high heterogeneity among the studies, making outcomes not suitable for meta-analysis. Nonetheless, this is the first comprehensive systematic review on this topic to date.

CONCLUSION

Ileal pouch volvulus is an extremely rare complication after ileal pouch-anal anastomosis. Patients present with symptoms of small bowel obstruction (SBO), such as diffuse abdominal pain, absence of bowel movement, nausea, vomiting, diarrhea or hematochezia. A high degree of suspicion is essential as obstruction due to pouch torsion is not likely to resolve conservatively and operative intervention is required to prevent pouch jeopardy. For this reason, it is important to be aware of the existence of this rare entity, especially in patients presenting with ileus symptomatic and an ileal pouch-anal anastomosis in their anamnestic data.

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