J Gastrointest Surg (2012) 16:1581–1584
DOI 10.1007/s11605-012-1872-z

CASE REPORT

A Unique Variant of Afferent Limb Syndrome After Ileal Pouch–Anal Anastomosis: A Case Series and Review of the Literature

Hitoshi Ogawa · Sho Haneda · Kazuhiro Watanabe · Hideyuki Suzuki · Koh Miura · Shinobu Ohnuma · Hiroyuki Sasaki · Chikashi Shibata · Michiaki Unno

Received: 3 January 2012 / Accepted: 7 March 2012 / Published online: 23 March 2012

Abstract
Background Afferent limb syndrome is a relatively rare cause of small bowel obstruction after restorative total proctocolectomy with ileal pouch–anal anastomosis for patients with ulcerative colitis or familial adenomatous polyposis.
Discussion This report describes three patients who developed recurrent small bowel obstruction after ileal pouch–anal anastomosis. The bowel obstruction was caused by torsion of the ileum at the inlet of the ileal J-pouch, which was thought to be a variant of afferent limb syndrome. This variant of afferent limb syndrome is characterized by a flexible afferent limb of the pelvic pouch due to the lack adhesion of the ileum in the abdominal cavity. Preoperative diagnosis required multiple series of contrast small bowel enemas. Strictureplasty and ileopexy effectively resolved the recurrent bowel obstruction caused by this variant of afferent limb syndrome.

Introduction

Restorative proctocolectomy with ileal pouch–anal anastomosis (IPAA) is an established surgical procedure for patients with ulcerative colitis (UC) or familial adenomatous polyposis (FAP). Small bowel obstruction is a common complication after IPAA, and the incidence varies between 16 and 27 %.[1–6] This obstruction is most commonly due to pelvic adhesion or adhesions at the ileostomy closure site.[3, 4, 6] Afferent limb syndrome (ALS) is a relatively rare cause of intestinal obstruction after IPAA .[7]

This report presents three cases with small bowel obstruction after IPAA. These patients required surgery due to recurrent bowel obstruction. These three cases showed a similar mechanism of obstruction that may represent a unique variant of ALS.

H. Ogawa (✉) · S. Haneda · K. Watanabe · H. Suzuki · K. Miura · S. Ohnuma · H. Sasaki · C. Shibata · M. Unno
Department of Surgery,
Tohoku University Graduate School of Medicine,
1-1 Seiryo-machi, Aobaku,
Sendai, Japan
e-mail: hogawa@surg1.med.tohoku.ac.jp

H. Ogawa
Department of Surgery, Sendai Red Cross Hospital,
Sendai, Japan

Case Series

The clinical courses of the three patients are summarized in Table 1.

Case 1

A 31-year-old male underwent two-staged total proctocolectomy and IPAA with a J-shaped ileal pouch for FAP. Presented with abdominal pain and emesis 9 years later, he was diagnosed with a small bowel obstruction by X-ray analysis. He stayed in the hospital for 20 days and that was relieved by conservative treatment with long tube drainage. Transanal endoscopy revealed possible torsion of the small bowel just proximal to the pelvic pouch, but no strictures were evident and the fiberscope could readily pass through the torsion. The small bowel obstruction relapsed

Table 1 Summary of the patients with SBO due to the variant of ALS after IPAA

Patient (age, gender)	Primary disease	Duration between IPAA and onset of SBO	Hospitalization due to SBO	Surgical procedure for ALS	Recurrence of SBO after surgery
Case 1 (31, M)	FAP	9 years	3 times/6 months	SXPL and ileopexy	None for 6 months
Case 2 (40, F)	UC	13 months	3 times/2 months	SXPL and ileopexy	None for 10 months
Case 3 (46, M)	UC	3 months	8 times/3 years	SXPL and ileopexy	None for 4 months

IPAA ileal pouch–anal anastomosis, *SBO* small bowel obstruction, *ALS* afferent limb syndrome, *FAP* familial adenomatous polyposis, *UC* ulcerative colitis, *SXPL* strictureplasty

20 days after discharge from the hospital. He was again treated with long tube drainage and the symptoms ameliorated in a few days. The small bowel obstruction relapsed again 5 months after discharge. Contrast small bowel enemas demonstrated stricture of the small bowel proximal to the pelvic pouch, although contrast media passed through the stricture (Fig. 1). The symptoms disappeared by treatment with long tube drainage, and laparotomy was performed to identify the mechanism of the recurrent small bowel obstruction and resolve the obstruction. No adhesion of the ileum in the abdominal cavity was observed and the afferent limb of the pelvic pouch was flexible. The torsion of the small bowel was observed at the inlet of pelvic pouch, although no obvious stricture was found (Fig. 2). The bowel obstruction did not occur when the afferent small bowel passed straight to the pelvic pouch, instead developing when the afferent limb kinked by an axial clockwise turn due to the accumulation of the intraluminal contents, and flexion and torsion occurred at the inlet of the pelvic pouch, thus resulting in the bowel obstruction (Fig. 3). Heineke–Mikulicz type strictureplasty of the small bowel at the inlet of the pouch (Fig. 4) and ileopexy of the ileum oral to the pelvic pouch were performed. He was free of symptoms of obstruction for 6 months.

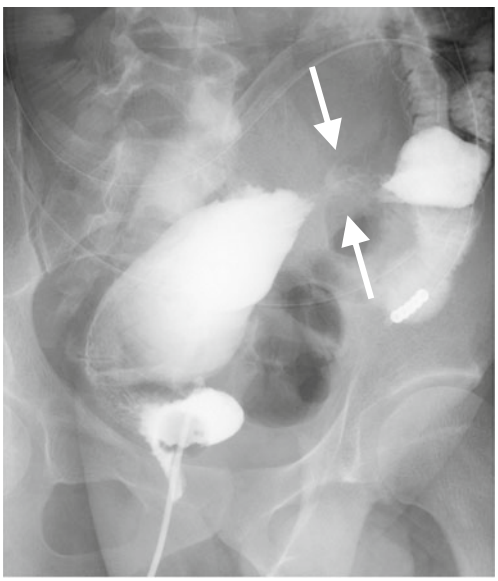

Fig. 1 Contrast small bowel enemas demonstrated stricture of the small bowel proximal to the pelvic pouch (*arrow*). (Case 1)

Case 2

A 40-year-old female underwent a two-staged total proctocolectomy and IPAA with a J-shaped ileal pouch for intractable ulcerative colitis. Thereafter, she complained of abdominal pain 13 months after surgery and was diagnosed with small bowel obstruction by CT, which demonstrated dilatation of the small bowel, although the mechanism of obstruction could not be identified. She stayed in the hospital for 10 days and recovered with long tube drainage. The small bowel obstruction relapsed 10 days after hospital discharge, and she was treated by long tube drainage again. Transanal endoscopy did not reveal any stricture; however, torsion of the small bowel proximal to the pelvic pouch was suspected as a cause of obstruction. Contrast small bowel enema demonstrated narrowing of the small bowel just proximal to the pelvic pouch. She discharged from the hospital because she did not complain of symptoms after dietary intake. The small bowel obstruction relapsed 5 days after discharge, and she underwent laparotomy. No adhesion of the ileum was seen in the abdominal cavity and the afferent limb of the pelvic pouch was flexible, as observed in the case 1. The obstruction appeared to be caused by

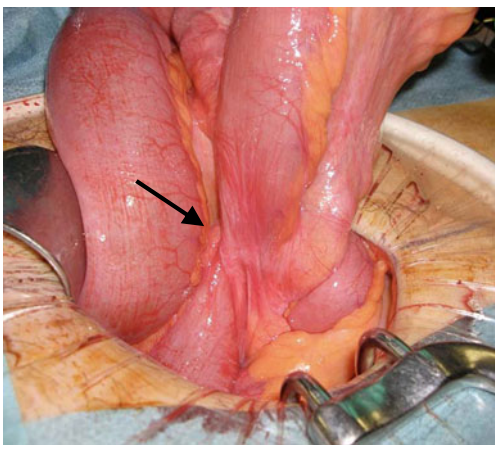

Fig. 2 Surgical finding of the small bowel in the case 1. Torsion was observed at the inlet of the pelvic pouch (*arrow*)

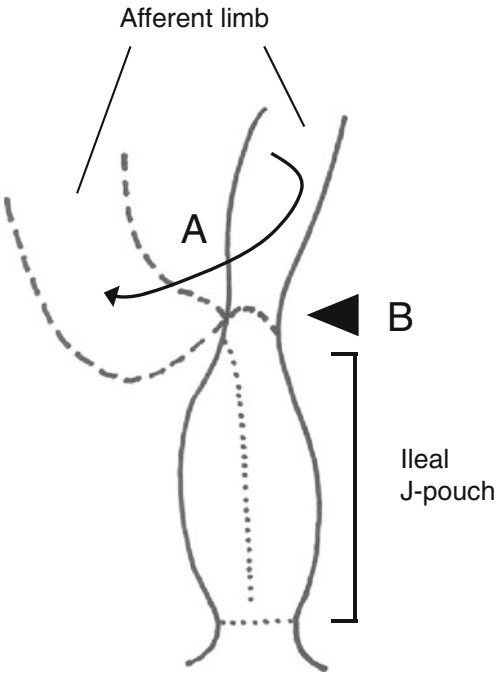

Fig. 3 A schematic drawing of the variant of ALS. When the afferent limb located in the pelvic floor (*A*), flexion and torsion occurred at the inlet of the pelvic pouch (*B*), thus resulting in the bowel obstruction

similar mechanisms as case 1; thus, an identical procedure (strictureplasty of the pouch inlet and fixation of the afferent limb) was performed. She is currently free of symptoms for 10 months after surgery.

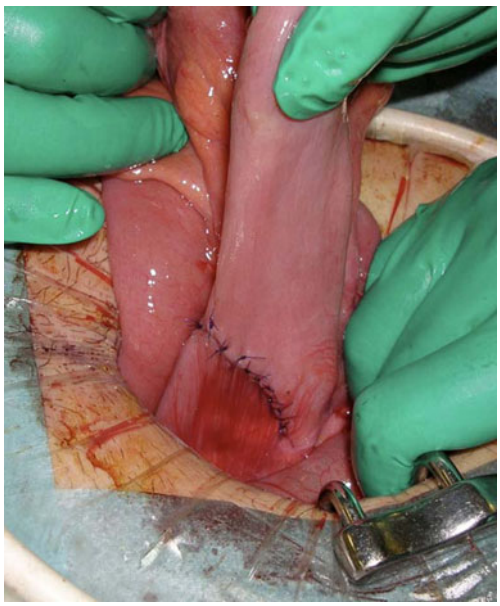

Fig. 4 Surgical procedure in case 1. Heineke–Mikulicz type strictureplasty of the small bowel was performed at the inlet of the pouch to prevent bowel obstruction

Case 3

A 46-year-old male underwent a two-staged total proctocolectomy and IPAA with a J-shaped pouch for intractable ulcerative colitis. He complained of abdominal pain and emesis 3 months after surgery and was diagnosed with a small bowel obstruction. Although the symptoms diminished after treatment with short tube drainage, he experienced recurrent bowel obstruction and required hospitalization eight times over the following 3 years. He underwent laparotomy due to the recurrent obstruction at his fifth hospitalization; however, the mechanism of obstruction was unclear and small bowel obstruction relapsed thereafter. Transanal endoscopy did not reveal any stricture or torsion within 80 cm of the anus, and repeated GI contrast study or CT examination could not identify the mechanism of the small bowel obstruction. The contrast small bowel enemas demonstrated narrowing of the small bowel just proximal to the pouch at his eighth episode of bowel obstruction, although contrast media passed through the stricture. This suggested that a similar mechanism of obstruction existed as that seen in the cases 1 and 2 had caused his recurrent bowel obstruction, and laparotomy was performed. There was no adhesion of the ileum in the abdominal cavity and the afferent limb of the pelvic pouch was flexible, as observed in the cases described above. The identical procedure was performed, and small bowel obstruction has not relapsed for 4 months.

Discussion

ALS after IPAA was defined as a distal small bowel obstruction caused by an acute angulation, prolapse, or intussusceptions of the afferent limb at the junction to the pouch, in the absence of intramural strictures.[7, 8] Read et al. reported six cases of ALS and described that afferent limb obstruction occurs because the limb of ileum just proximal to the pelvic pouch becomes trapped posteriorly between the pouch and sacrum.[9] Recently, Kirat et al. reported 18 patients with ALS after IPAA for UC, but the precise mechanism of obstruction was not detailed.[8] The mechanism of bowel obstruction shown in the present report was obviously different from that described by Read et al., and it did not meet the criteria for ALS described above. Therefore, the mechanism of bowel obstruction in the present cases could be categorized as an afferent limb variant.

The preoperative diagnosis of this variant of ALS is not simple. Narrowing or stricture of the small bowel just proximal to the pelvic pouch was revealed by contrast small bowel enemas in all three patients. However, these findings were not consistently obtained during the repeated examination, and the narrowing or stricture of the small bowel was not detected when the torsion of the afferent limb was

resolved due to the flexibility of the afferent limb. Therefore, a single contrast small bowel enema showing no narrowing or stricture could not exclude this variant of ALS, and multiple series of examinations are required for diagnosis. Kirat et al. described that angulation of pouch inlet and difficulty in intubating the afferent limb was detected at pouchoscopy in 15 out of 18 patients with ALS.[8] Torsion of the afferent limb was observed by endoscopy in two patients of the present report, but intubation was not difficult. Torsion was not observed in the remaining patient. No stricture was observed in all three patients. Therefore, although the finding of torsion by endoscopy might be helpful to diagnose this variant of ALS, endoscopic examinations would therefore be useful to exclude a stricture of bowel rather than to identify the mechanisms of obstruction.

Read et al. reported six patients with ALS and all patients underwent laparotomy for unresolved obstruction.[9] Shen et al. also described that patients with ALS typically required laparotomy to relieve the obstruction.[7] Likewise, the three patients described here eventually required laparotomy for the recurrent and frequent bowel obstruction, although each episode of obstruction was resolved by conservative therapy. Surgical procedures for ALS include enteroenterostomy, resection of angulated bowel, pexy of the pouch, ileostomy, and mobilization of the pouch with small bowel fixation .[8, 9] All of the patients in the present report underwent strictureplasty of the small bowel at the inlet of pouch where flexion and obstruction was thought to occur with kinking. Read et al. reported six patients with afferent loop syndrome and described that five of these six patients underwent side-to-side anastomosis of the afferent limb to the pouch (enteroenterostomy) to relieve the obstruction because of the risk of injury to either the pouch or its mesentery.[9] Enteroenterostomy therefore seemed to be a reasonable procedure in their cases because the afferent limb was adherent and trapped between the pouch and sacrum. It would be arduous and dangerous to manipulate the trapped loop of the ileum in those cases, although enteroenterostomy resulted in blind loop formation in their patients. In contrast to their cases, the afferent limb was flexible in the present cases, and it was easy to perform strictureplasty at the inlet of the J-pouch, which thus enabled us to relieve the obstruction without blind loop formation. In addition, the oral small bowel was fixed to the pelvic wall to prevent kinking at the pouch inlet. Either of the procedures may have been sufficient to resolve the bowel obstruction; however, both strictureplasty and bowel fixation were performed for assured prevention of bowel obstruction by relapse of this variant of ALS.

In conclusion, this variant of ALS should be considered when recurrent and frequent small bowel obstruction occurs after IPAA. Repeated contrast small bowel enema may be required to detect the narrowing or stricture. Endoscopic examinations are therefore useful to exclude intramural stricture, and the finding of torsion of the afferent limb may be helpful for making an accurate diagnosis. Strictureplasty of the pouch inlet and fixation of the afferent limb is therefore considered to be a safe and useful procedure to resolve this variant of ALS.

References

1. McMullen K, Hicks TC, Ray JE, Gathright JB, Timmcke AE. Complications associated with ileal pouch–anal anastomosis. World J Surg 1991;15:763-6; discussion 6-7.
2. Young CJ, Solomon MJ, Eyers AA, West RH, Martin HC, Glenn DC, Morgan BP, Roberts R. Evolution of the pelvic pouch procedure at one institution: the first 100 cases. Aust N Z J Surg 1999;69:438-42.
3. MacLean AR, Cohen Z, MacRae HM, O'Connor BI, Mukraj D, Kennedy ED, Parkes R, McLeod RS. Risk of small bowel obstruction after the ileal pouch–anal anastomosis. Ann Surg 2002;235:200-6.
4. Marcello PW, Roberts PL, Schoetz DJ, Jr., Coller JA, Murray JJ, Veidenheimer MC. Obstruction after ileal pouch–anal anastomosis: a preventable complication? Dis Colon Rectum 1993;36:1105-11.
5. Francois Y, Dozois RR, Kelly KA, Beart RW, Jr., Wolff BG, Pemberton JH, Ilstrup DM. Small intestinal obstruction complicating ileal pouch–anal anastomosis. Ann Surg 1989;209:46-50.
6. Aberg H, Pahlman L, Karlbom U. Small-bowel obstruction after restorative proctocolectomy in patients with ulcerative colitis. Int J Colorectal Dis 2007;22:637-42.
7. Shen B, Remzi FH, Lavery IC, Lashner BA, Fazio VW. A proposed classification of ileal pouch disorders and associated complications after restorative proctocolectomy. Clin Gastroenterol Hepatol 2008;6:145-58; quiz 24.
8. Kirat HT, Kiran RP, Remzi FH, Fazio VW, Shen B. Diagnosis and management of afferent limb syndrome in patients with ileal pouch–anal anastomosis. Inflamm Bowel Dis 2011;17:1287-90.
9. Read TE, Schoetz DJ, Jr., Marcello PW, Roberts PL, Coller JA, Murray JJ, Rusin LC. Afferent limb obstruction complicating ileal pouch–anal anastomosis. Dis Colon Rectum 1997;40:566-9.