Surg Today (2016) 46:1159–1165
DOI 10.1007/s00595-016-1307-7

ORIGINAL ARTICLE

Clinical features and management of afferent limb syndrome after ileal pouch-anal anastomosis for ulcerative colitis

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Received: 1 August 2015 / Accepted: 12 November 2015 / Published online: 22 January 2016

Abstract
Purpose Afferent limb syndrome (ALS) is a type of small bowel obstruction (SBO) caused by obstruction of the afferent intestinal limb after ileal pouch-anal anastomosis (IPAA). The aim of this study was to reveal the clinical features and management of ALS.
Methods Of 320 patients undergoing IPAA for ulcerative colitis, we analyzed data from patients presenting with SBO.
Results Six of 19 patients with SBO were diagnosed with ALS. All patients with ALS presented with recurrent intermittent obstructive symptoms before admission, whereas 15 % of patients without ALS presented with these symptoms ($P < 0.0005$). Among the six patients with ALS, four patients required surgery, although they underwent transanal decompression for each episode of obstruction. The proportion of surgery was higher in the ALS group ($P < 0.01$). Acute angulation of the afferent limb was recognized in four cases and followed by fixation of the afferent limb. No further surgery was required in any patient following the last fixation.
Conclusions SBO after IPAA may be caused by acute angulation of the afferent limb of the ileal J-pouch. Most patients with ALS may eventually require fixation of the afferent limb due to acute angulation of the afferent limb.

Keywords Afferent limb syndrome · Ileal pouch-anal anastomosis · Ulcerative colitis · Small bowel obstruction · Ileopexy

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Introduction

Restorative proctocolectomy and ileal pouch-anal anastomosis (IPAA) have become the surgical procedures of choice for the definitive management of ulcerative colitis (UC) and familial adenomatous polyposis [1–3]. However, small bowel obstruction (SBO) has remained a common postoperative complication after IPAA [4]. The risk of SBO may be higher than with other pelvic or abdominal surgeries because it combines wide abdominal and pelvic dissection and also may be performed as a staged procedure [5]. Pelvic adhesion or adhesions at the ileostomy closure site most commonly causes SBO [5–7]; however, Read et al. [8] first reported a subset of patients with SBO caused by acute angulation of the afferent limb, which had a different mechanism from that of adhesion or stricture. Kirat et al. [9] presented the outcomes of patients with afferent limb syndrome (ALS), which was defined by Shen et al. [10] as distal SBO caused by acute angulation, prolapse, or intussusception of the afferent limb at the junction to the pouch. To the best of our knowledge, there are no previous reports discussing the clinical features and management of ALS compared with SBO from causes other than ALS. Therefore, the aim of this study was to reveal the clinical features and management of ALS.

Materials and methods

Patients

This retrospective study was undertaken to evaluate the clinical features and management of SBO due to ALS by reviewing the surgical UC database of Mie University. We reviewed the records of 320 patients who underwent IPAA from January 2000 to December 2014 at Mie

University. Patients were not registered before ileostomy closure. The investigations were performed in accordance with the guidelines of the Declaration of Helsinki and were approved by the local institutional review board. Informed consent was obtained from all patients who agreed to have their personal data used for research purposes.

Surgery

The standard operation was mucosal restorative proctocolectomy with hand-sewn IPAA using an 18 cm, two-limbed, J-shaped ileal pouch and diverting ileostomy. An intact pouch without anastomotic leakage and an intact ileum without stricture between the stoma site and the junction to the pouch were confirmed just before ileostomy closure. Closure of the ileostomy was performed approximately 3 months after IPAA. In all patients, a sodium hyaluronate methylcellulose anti-adhesion barrier (Seprafilm®, Genzyme Corp., Cambridge, MA) was used before closing the incision.

Inclusion and exclusion criteria

Of 320 patients who underwent IPAA, patients without intestinal continuity due to pelvic sepsis, perianal sepsis, anorectal dysfunction after IPAA, or recurrence of UC-associated cancer were excluded. All patients presenting with SBO (including adhesive obstruction, ALS, and anastomotic stricture at the ileostomy closure site) were enrolled.

Definition of SBO

The diagnosis of SBO was defined as follows: the presence of abdominal symptoms (abdominal pain, bloating, nausea, vomiting, no gas or stools) and radiographic findings confirmed by an abdominal X-ray, radiographic enteroclysis, or radiographic contrast enema. Patients with obstructive symptoms during hospitalization for stoma closure were not defined as having SBO, i.e., paralytic ileus was distinguished from SBO. Patients with obstructive symptoms due to anal outlet obstruction were not defined as having SBO. Other types of abdominal pain were defined to be nonspecific. In this study, all patients with SBO were divided into an ALS and a non-ALS group.

Definition of ALS

ALS after IPAA was defined as distal SBO caused by acute angulation, prolapse, intussusception, or torsion of the afferent limb at the junction of the pouch in the absence of intraluminal mucosa-associated strictures that was confirmed endoscopically or radiologically [9–11]. Non-ALS was defined as SBO other than ALS.

Study variables

We evaluated clinical variables including sex, age at IPAA, type of anastomosis in IPAA, number of stages of operation, duration between intestinal continuity and first admission, hospitalization due to SBO during the follow-up period, recurrent intermittent obstructive symptoms, presenting symptoms, method of diagnosis, and treatment method. The follow-up period was defined as the duration from the first onset of SBO to the first operation for SBO or December 2014. Recurrent intermittent obstructive symptoms were defined as symptoms identified recurrently and intermittently before the first admission due to SBO. The definition of presenting symptoms was based on the symptoms at the first admission. The method of diagnosis was defined as the modality used for the diagnosis of ALS or non-ALS and the treatment method was defined as conservative or surgical intervention to treat SBO.

Statistical analysis

Statistical analysis of the comparisons was performed using the Chi square test or Fisher's exact test and the Mann–Whitney *U* test for quantitative and qualitative variables using the JMP software program (SAS Institute, Cary, NC). A P value <0.05 was considered to be statistically significant.

Results

Frequency of SBO in the ALS vs. non-ALS groups

The total number of patients before ileostomy closure or without intestinal continuity due to pelvic sepsis, perianal sepsis, anorectal dysfunction after IPAA, and recurrence of UC-associated cancer was 15. A total of 19 UC patients with SBO after IPAA were identified for review. The frequency of SBO was 6.2 % in 305 postoperative UC patients with intestinal continuity during the study period. In patients with SBO, the number of cases of adhesive SBO, ALS, and stricture of the ileostomy closure site was 12 (63.2 %), 6 (31.6 %), and 1 (5.2 %), respectively (Fig. 1).

Characteristics, diagnosis, and treatment methods in patients with ALS and non-ALS (Table 1)

The median number of hospitalizations for SBO during the follow-up period in the ALS group and non-ALS group

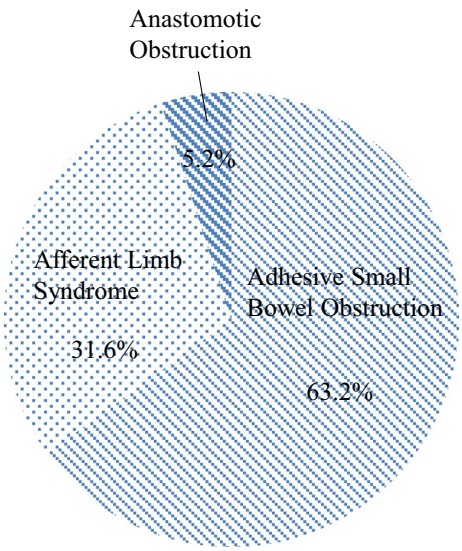

Fig. 1 SBO frequency and associated conditions. The frequency of SBO was 6.2 % in 305 postoperative UC patients with intestinal continuity during the study period. In patients with SBO, the number of cases of adhesive SBO, ALS, and stricture of the ileostomy closure site was 12 (63.2 %), 6 (31.6 %), and 1 (5.2 %), respectively

was 3 (range 1–17) and 1 (range 1–6), respectively, which was significantly different ($P < 0.05$).

All patients in the ALS group presented with recurrent intermittent obstructive symptoms before admission, whereas only 15 % of patients in the non-ALS group presented with these symptoms ($P < 0.0005$). Regarding the method of diagnosis, the proportions of radiographic contrast enema ($P < 0.005$), transanal endoscopy ($P < 0.001$), and abdominal computed tomography (CT) ($P < 0.05$) in the ALS group were significantly higher than that of the non-ALS group. Regarding the treatment method, the proportions of transanal decompression ($P < 0.001$) and surgery ($P < 0.05$) were significantly higher in the ALS group than that of the non-ALS group.

Diagnosis during operation, operative findings, surgical management, and outcomes in patients with ALS (Table 2)

Among six patients with ALS, two patients (Cases 2 and 5) did not require surgery, although they underwent transanal decompression for each episode of obstruction. Four patients underwent surgery and the median total number of operations for ALS was one (range 0–4). One patient (Case 1) was diagnosed incorrectly as having adhesive SBO during the first and second operation after IPAA, and adhesiolysis was performed. Because of a marked caliber change in the afferent limb of the pouch, Heineke-Mikulicz strictureplasty of the afferent limb was performed at the third operation. However, the patient developed recurrence of ALS after adhesiolysis and strictureplasty. At the fourth operation, acute angulation of the afferent limb was identified at 5 cm proximal to the junction of the pouch and dilatation of the oral side of the pouch was also observed. The proximal ileum of the angulated afferent limb was located on the left side of the pelvic cavity. No adhesion of the afferent limb in the abdominal cavity was observed and the afferent limb of the pouch was flexible. These findings were followed by fixation of the proximal ileum to the right side of the pelvic wall (ileopexy). The patient was free of any symptoms at the last follow-up, 42 months after surgery.

In Case 3, the patient was diagnosed as having ALS during the first operation for SBO. Acute angulation of the afferent limb was located at 7 cm proximal to the junction of the pouch and dilatation of the oral side of pouch was also observed. The proximal ileum of the angulated afferent limb was located on the left side of the pelvic cavity. No adhesion of the afferent limb in the abdominal cavity was observed and the afferent limb of the pouch was flexible. The patient underwent ileopexy due to the diagnosis of afferent limb angulation; however, she developed recurrence 1 week after the first operation. Strictureplasty was performed at the second operation because narrowing of the afferent limb of the pouch was regarded to be the cause of the recurrence; however, her symptoms recurred. At the third operation for ALS, a shortened fixed portion of the afferent limb was regarded to be the cause of acute angulation and additional ileopexy was performed. The suture site between the afferent limb and the right side of the pelvic wall was unfixed. The fixed portion of the afferent limb was extended to the level of the anterior superior iliac spine during the third operation (Fig. 2). She developed no further recurrence after the third operation.

The other patients with ALS (Cases 4 and 6) underwent ileopexy without additional procedures. Acute angulation of the afferent limb just proximal to the pouch and dilatation of the oral side of the pouch were recognized and followed by ileopexy. One patient (Case 4) had been free of symptoms of obstruction for 6 months as of the last visit to the outpatient clinic. The other patient (Case 6) was admitted once for obstructive symptoms, but had not required further surgery for 23 months as of the last visit to the outpatient clinic.

Discussion

According to a recent meta-analysis of 61 studies assessing SBO after open IPAA, the incidence of SBO was 11.4 % since the year 2000 [12]. The proportion rate of SBO ($N = 19$, 6.5 %) after IPAA for UC in our department was less than the SBO rates reported in this meta-analysis. Patients with SBO included six cases (31.6 %) with ALS

Table 1 Characteristics, diagnosis and treatment methods for patients with SBO

Variable	ALS ($N = 6$)	Non-ALS ($N = 13$)	P-value
Female	3 (50 %)	8 (61 %)	NS
Age at IPAA (years)	Median 40, range 27–61	Median 35.5, range 18–62	NS
Open surgery: laparoscopic surgery	6 (100 %): 0 (0 %)	12 (92 %): 1 (8 %)	NS
Hand-sewn IPAA vs. stapled IPAA	6 (100 %): 0 (0 %)	12 (92 %): 1 (8 %)	NS
Two-staged operation vs. three-staged operation	6 (100 %): 0 (0 %)	10 (76 %): 3 (24 %)	NS
Duration from restoration of gastrointestinal continuity to first admission (months)	Median 36.5, range 3–96	Median 25, range 0.1–122	NS
Hospitalizations because of SBO during the follow-up period	Median 3.5, range 1–17	Median 1, range 1–5	<0.05
Recurrent intermittent obstructive symptoms	6 (100 %)	2 (15 %)	<0.0005
Presenting symptoms[a]			
Abdominal pain	6 (100 %)	12 (92 %)	NS
Bloating	5(83 %)	6 (23 %)	NS
Nausea, vomiting	4 (66 %)	5 (38 %)	NS
No gas or stools	3 (50 %)	3 (23 %)	NS
Diagnostic method[a]			
Radiographic enteroclysis	2 (33 %)	11 (84 %)	NS
Radiographic contrast enema	6 (100 %)	3 (23 %)	<0.005
Transanal endoscopy	5 (83 %)	1 (7 %)	<0.001
Abdominal computed tomography	6 (100 %)	5 (38 %)	<0.05
Treatment method[a]			
Transanal decompression	5 (83 %)	0 (0 %)	<0.001
Nasointestinal decompression	2 (33 %)	5 (38 %)	NS
Endoscopic dilatation	1 (20 %)	0 (0 %)	NS
Surgery	4 (66 %)	2 (15 %)	<0.05

SBO small bowel obstruction, *ALS* afferent limb syndrome, *IPAA* ileal pouch-anal anastomosis

[a] Some patients had more than one presenting symptom, diagnosis, and treatment method

in our study. However, the meta-analysis did not describe the proportion of patients who developed ALS [12]. Read et al. [8] reported that 122 (22 %) patients had one or more episodes of obstruction among 567 patients with IPAA, and afferent limb obstruction was identified as the cause of obstruction in 6 of 122 (4.9 %).

In our study, ALS after IPAA was defined as distal SBO caused by acute angulation, prolapse, intussusception, or torsion of the afferent limb at the junction of the pouch, in the absence of intraluminal mucosa-associated strictures. Read et al. [8] described that afferent limb obstruction occurs because the limb of the ileum just proximal to the pelvic pouch becomes trapped posteriorly between the pouch and sacrum. Kirat et al. [9] reported 18 patients with ALS, which was defined as distal SBO caused by acute angulation, prolapse, or intussusception of the afferent limb. In their study, a difficulty in intubating the afferent limb at pouchoscopy due to angulation of the pouch inlet and kinking or narrowing of the pouch inlet on abdominal imaging could be identified in most patients. However, the precise mechanism of obstruction in each case (including obstruction caused by acute angulation of the afferent limb) was not described in detail. Ogawa et al. [11] stated that the mechanism of bowel obstruction in three cases could be categorized as an afferent limb variant, because the bowel obstruction was caused by torsion of the afferent limb, which was thought to be different from that described by Read et al. In our study, the cause of obstruction in four cases who underwent surgery was recognized as acute angulation of the afferent limb, which is similar to the afferent limb variant reported by Ogawa et al. [11].

To the best of our knowledge, no previous articles have described the formation of acute angulation of the afferent limb. Although the pelvic pouch is fixed in the pelvis, mobility of the ileum proximal to the pelvic pouch may be markedly increased in patients with ALS. Thus there may be a relationship between an increased mobility of the ileum proximal to the pelvic pouch and the formation of acute angulation of the afferent limb. It is speculated that the length of the pouch, mesenteric tension in the pelvis, position of the afferent limb at anastomosis or the adhesion of the stoma closure site in the pelvis is related to the development of ALS.

Table 2 Diagnosis during operation, operative findings, surgical management and outcomes in patients with ALS

Case	Gender	Age at IPAA (years)	Time between intestinal continuity and first admission (months)	Hospitalization because of SBO (/ months)	Diagnosis during primary operation	Age at first operation for SBO (years)	Operation	Total number of operations for ALS	Postoperative follow-up period (months)	Recurrence after ileopexy
1	Female	61	15	17/92	Adhesive SBO	67	Adhesiolysis × 2, SP, ileopexy	4	42	N
2	Male	61	3	4/75	–	–	–	0	–	–
3	Female	41	48	2/6	ALS	45	Ileopexy, ileopexy SP, ileopexy	3	6	Y
4	Female	40	25	6/60	ALS	44	Ileopexy	1	28	N
5	Male	28	96	1/16	–	–	–	0	–	–
6	Male	27	68	2/51	ALS	35	Ileopexy	1	23	Y

IPAA ileal pouch-anal anastomosis, *SBO* small bowel obstruction, *ALS* afferent limb syndrome

Read et al. [8] stated that afferent limb obstruction should be suspected in patients with recurrent obstruction after IPAA. Kirat et al. [9] reported that most patients presented with recurrent intermittent abdominal pain because of obstruction. Similarly, in our study, the proportion of patients with recurrent intermittent obstructive symptoms was significantly higher in the ALS group compared with the non-ALS group; however, there was no difference in the presenting symptoms between the two groups. Patients with ALS were hospitalized more frequently and required more examinations for the diagnosis compared with patients with non-ALS SBO.

In our cases, four of six patients eventually required laparotomy due to recurrent and frequent bowel obstruction, although each episode of obstruction was transiently relieved by transanal decompression.

Read et al. [8] described that five of six patients underwent side-to-side anastomosis of the afferent limb to the pouch (enteroenterostomy) to avoid injury to either the pouch or its mesentery. Therefore, enteroenterostomy appeared to be a reasonable procedure in these cases because the afferent limb was adherent posterior to the pouch and trapped between the pouch and sacrum, causing acute angulation of the afferent limb. Two patients required re-exploration and pexy of the pouch to the pelvic sidewall (pouchopexy) to relieve recurrent afferent limb obstruction.

Kirat et al. [9] reported that eight patients required surgery including resection of the angulated bowel, pexy of the pouch, pouch excision with end ileostomy, and mobilization of the pouch with small bowel fixation. Pexy of the pouch versus resection of the angulated bowel was elected according to the surgeon's preference or the severity of inflammation and adhesion. Six patients (except for two patients with pouch excision) did not develop recurrence of symptoms.

Ogawa et al. [11] reported three patients with torsion of the small bowel proximal to the pouch who underwent ileopexy to prevent kinking at the inlet to the pouch. These patients also underwent concurrent strictureplasty at the pouch inlet to relieve the obstruction without blind-loop formation. The authors described that both strictureplasty and bowel fixation were performed for the definitive prevention of bowel obstruction by relapse of this variant of ALS, although either of the procedures may have been sufficient to resolve the bowel obstruction [11].

In our study, ileopexy without strictureplasty was performed in two of four patients and neither required further surgery for recurrence. Because the obstruction in all four patients resulted from acute angulation of the afferent limb, ileopexy might have been a reasonable procedure for our cases. Strictureplasty of the afferent limb is not always required for all patients. At the third operation for ALS in Case 3, a shortened fixed portion of the afferent limb was

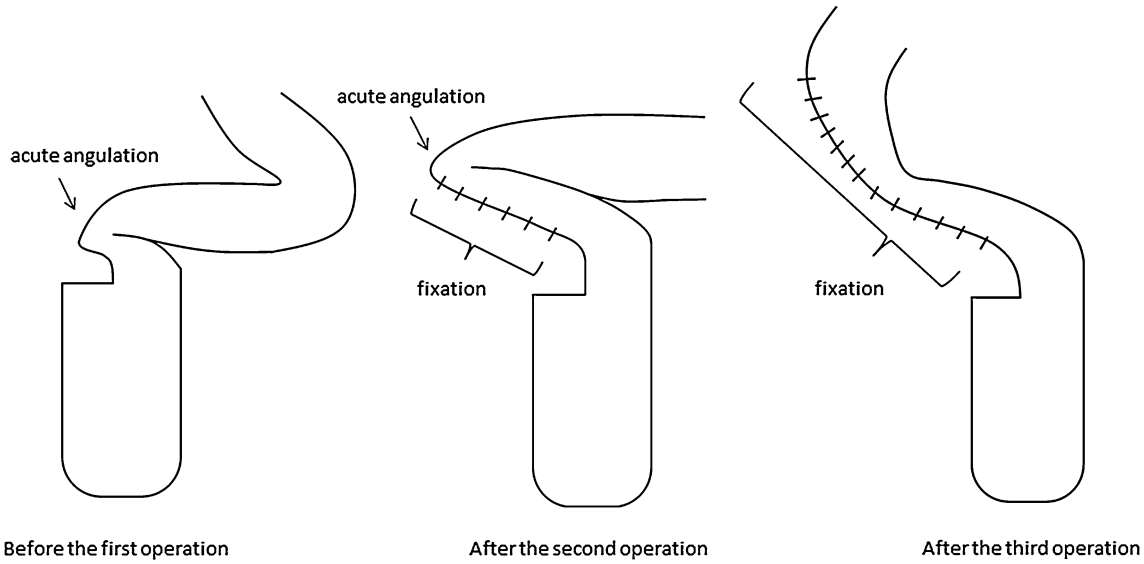

Fig. 2 Operative findings and procedure of the third operation in Case 3. The proximal ileum of the angulated afferent limb was located on the left side of the pelvic cavity during the first operation. The proximal ileum was fixed to the right side of the pelvic wall. During the third operation for ALS, a shortened fixed portion of the afferent limb was regarded to be the cause of acute angulation and additional ileopexy was performed. The fixed portion of the afferent limb was extended to the level of the anterior superior iliac spine

regarded to be the cause of acute angulation. A sufficient length of the fixed portion of the afferent limb to the pelvic wall may be needed to avoid recurrence.

Conclusions

SBO after IPAA in some patients might be caused by acute angulation of the afferent limb of the ileal J-pouch. When a patient presents with recurrent intermittent obstructive symptoms after IPAA, then we recommend considering ALS and conducting both endoscopy and abdominal imaging for the diagnosis. Although each episode of obstruction was transiently relieved by transanal decompression in our study, most patients with ALS due to acute angulation of the afferent limb may eventually require fixation of the afferent limb (ileopexy). Fixation of the afferent limb is considered to be a simple and effective procedure to resolve ALS caused by acute angulation of the afferent limb. The limitations associated with the present study include its retrospective nature and small sample size. Further prospective studies analyzing a large number of patients undergoing surgery for ALS may be needed to evaluate appropriate operative procedures and the long-term outcome.

Compliance with ethical standards

Conflict of interest All of the authors declare that they have no conflicts of interest in association with this study.

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