CASE REPORT

Cryptogenic repetitive severe colitis after ileostomy closure

Yuki Sakamoto¹ · Yasuo Sakamoto¹ · Yuji Miyamoto¹ · Ryuma Tokunaga¹ · Mayuko Ohuchi¹ · Yuki Kiyozumi¹ · Kenichi Nakamura¹ · Keisuke Kosumi¹ · Daisuke Izumi¹ · Kazuto Harada¹ · Junji Kurashige¹ · Yukiharu Hiyoshi¹ · Shiro Iwagami¹ · Yoshifumi Baba¹ · Naoya Yoshida¹ · Masayuki Watanabe¹ · Hideo Baba¹

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Abstract We describe a case of cryptogenic repetitive severe colitis after ileostomy closure. A 50-year-old man was referred to our hospital for treatment of advanced rectal cancer. The patient underwent laparoscopic intersphincteric resection combined with synchronous partial liver resection for colorectal liver metastasis detected intraoperatively and construction of a diverting ileostomy. Ileostomy closure was performed 3 months later. Five days following ileostomy closure, the patient complained of lower abdominal pain and developed multiple daily fever episodes. Colonoscopy revealed multiple ulcers in the sigmoid colon and rectum. Because symptoms did not improve despite fasting and antibiotic therapy, ileostomy reconstruction was performed. Re-closure was performed 6 months after the previous ileostomy closure; however, severe colitis recurred with similar symptoms, and construction of a colostomy at the transverse colon was then performed. Despite varied examinations, the cause of the colitis was not identified. To our knowledge, there are no previous reports of repetitive severe colitis after ileostomy closure.

Keywords Diverting ileostomy · Stoma closure · Colitis

Introduction

Diverting ileostomy is often performed to reduce the risk of anastomotic leakage in patients undergoing anterior resection for rectum [1, 2]. However, complications are reported after ileostomy closure, including wound infection and anastomotic leakage [3–6]. We report a case of cryptogenic repetitive severe colitis with severe symptoms such as a lower abdominal pain, high fever, and watery diarrhea, with the multiple deep ulcers after the every ileostomy closures. To the best of our knowledge, there are no previous reports of similar cases.

Case report

A 50-year-old man was referred to our hospital for treatment of advanced rectal cancer. The patient underwent laparoscopic intersphincteric resection with D3 lymph node dissection preserving the left colic artery combined with synchronous partial liver resection for colorectal liver metastasis detected intraoperatively and construction of a diverting ileostomy. Administering of 12 cycles of mFOLFOX6 was planned as adjuvant chemotherapy after surgery. In fact, ileostomy closure was performed after 4 cycles of mFOLFOX6, according to the patient's request.

Five days after ileostomy closure, the patient complained of lower abdominal pain and watery diarrhea that occurred more than four times per day, with multiple daily fever episodes. Colonoscopy revealed multiple, deep, and sub-circumferential ulcers in the sigmoid colon and rectum (Fig. 1). Laboratory examination showed elevated inflammatory markers (white blood cell count 22500/μL, neutrophil 91.5 %, lymphocyte 4.3 %, eosinophil 0.2 %, basophil 0.1 %, monocyte 3.9 %, C-reactive protein 23 mg/dL). Bacterial

✉ Hideo Baba
hdobaba@kumamoto-u.ac.jp

¹ Department of Gastroenterological Surgery, Graduate School of Medical Sciences, Kumamoto University, 1-1-1 Honjo, Chuo-ku, Kumamoto 860-8556, Japan

Fig. 1 Colonoscopy revealed multiple ulcers at colon

culture showed no evidence of infectious colitis caused by *Clostridium difficile* or other bacteria. Histopathologic examination showed only non-specific inflammatory change without evidence of inflammatory bowel disease (IBD) such as noncaseating granuloma in ulcerative colitis or crypt abscess in Crohn's disease. Because his symptoms did not improve despite immediate fasting and antibiotic therapy, reconstruction of the ileostomy was performed, with the severe colitis improving markedly following the reconstruction. Colonoscopy following 2 months of adjuvant mFOL-FOX6 revealed that the multiple colonic ulcers had resolved (Fig. 2a) and computed tomographic angiography showed sufficient blood supply from the left colic artery (Fig. 2b). Re-closure of the ileostomy was performed 6 months after the previous ileostomy closure; however, severe colitis recurred with similar symptoms (Fig. 3). We constructed a permanent colostomy at the transverse colon because colitis was localized to the sigmoid colon and rectum. Symptoms improved after construction of the colostomy and to date, the colostomy remains in place.

Fig. 2 Preoperative examination of re-closure.
a Mucosa of colon recovered.
b CT angiography showed the flow of the left colic artery

The patient gave written informed consent to publish the details of his case.

Discussion

A diverting stoma is constructed to prevent anastomotic leakage that can lead to panperitonitis and sepsis [1]. Huser et al. stated that a defunctioning stoma reduced the rate of clinically relevant anastomotic leakages, especially for low rectal cancers [2]. Diverting ileostomies are usually closed a few months after construction; however, high morbidity after ileostomy closure has been reported. Chow et al. reported that bowel obstruction and surgical-site infection were two of the most frequent complications and that the overall morbidity rate was 17.3 % by meta-analysis [4].

Pseudomembranous colitis after ileostomy closure has been often reported with an incidence rate of 1.6–4.3 % [3, 6–9]. Pseudomembranous colitis is diagnosed by detecting cytotoxin and confirming the presence of pseudomembranes by colonoscopy [10]. In our case, we suspected pseudomembranous colitis and repeated the cytotoxin assay four times; however, *C. difficile* toxin was never detected. Additionally, colonoscopy revealed multiple ulcers, not pseudomembranes. Based on these findings, we suspected ischemic colitis and repeated the colonoscopy, which revealed hemorrhagic nodules, bleeding, longitudinal ulcers, pseudopolyps, and pseudomembranes, indicating ischemic colitis [11]. However, we found only multiple dotted ulcers, not the characteristic findings of ischemic colitis. Furthermore, we preserved the left colic artery and blood supply to the left colon was confirmed by computed tomographic angiography before the second ileostomy closure. We then considered the possibility of IBD such as ulcerative colitis and Crohn's disease. However, there was no past history about IBD in this patient, and histopathologic findings showed only non-specific inflammatory changes without findings of IBD. Although we suspected colitis caused by cytomegalovirus, a

Fig. 3 Severe colitis relapsed

cytomegalovirus antibody was never detected. Eosinophilic colitis was negative because the eosinophil rate (0.2 %) in the white blood cell was within normal limit. Antibiotic-induced colitis was also negative because antibiotics was only administered during operation and the symptoms was recognized 15 days after surgery. We also considered the possibility of mucosal damage by anticancer agents used during adjuvant chemotherapy. We performed a second ileostomy closure 3 months after the final chemotherapy; however, colitis relapsed with similar symptoms. Our final diagnosis was cryptogenic repetitive severe colitis.

Colonic mucosal injury caused by intestinal secretions may have been a cause of the recurrent colitis in this case. Patients often complain of frequent watery diarrhea related to passing intestinal secretions or dietary residue after ileostomy closure with the diarrhea caused by atrophic changes in the intestinal mucosa [12]. Diarrhea gradually disappears with improvement of the mucosal atrophy in most cases. However, our patient's condition worsened despite fasting and antibiotic therapy. His condition improved soon after re-construction of the ileostomy, which led us to speculate that intestinal secretions, and not diet, stimulated and damaged the mucosa directly. Intestinal intramural vascular insufficiency is another possible cause, although blood flow from the left colic artery was confirmed by computed tomographic angiography. The fact that colitis was localized to the left colon supports this possibility and both causes may have contributed to the repetitive severe colitis in our case. We discussed three treatment options: (1) the construction of transverse colostomy, (2) the re-construction of ileostomy, and (3) the continuation of conservative therapy. Finally, we constructed a colostomy at the transverse colon because the patient hoped the construction of a colostomy and we

considered that improving bowel function would be more difficult in the future. To date, the colostomy remains in this patient with no recurrent symptoms of colitis.

To our knowledge, there are no previous reports of repetitive severe colitis after ileostomy closure. We consider it necessary to inform patients regarding adverse events, as the present case demonstrated, and the possibility of the need for a permanent stoma.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical standards This case report has been approved by the appropriate ethics committee and has, therefore, been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

References

1. Bax TW, McNevin MS (2007) The value of diverting loop ileostomy on the high-risk colon and rectal anastomosis. *Am J Surg* 193:585–587
2. Huser N, Michalski CW, Erkan M et al (2008) Systematic review and meta-analysis of the role of defunctioning stoma in low rectal cancer surgery. *Ann Surg* 248:52–60
3. Mansfield SD, Jensen C, Phair AS et al (2008) Complications of loop ileostomy closure: a retrospective cohort analysis of 123 patients. *World J Surg* 32:2101–2106
4. Chow A, Tilney HS, Paraskeva P et al (2009) The morbidity surrounding reversal of defunctioning ileostomies: a systematic review of 48 studies including 6,107 cases. *Int J Colorectal Dis* 24:711–723
5. Akiyoshi T, Fujimoto Y, Konishi T et al (2010) Complications of loop ileostomy closure in patients with rectal tumor. *World J Surg* 34:1937–1942
6. Rubio-Perez I, Leon M, Pastor D et al (2014) Increased postoperative complications after protective ileostomy closure delay: an institutional study. *World J Gastrointest Surg* 6:169–174
7. Randall JK, Young BC, Patel G et al (2011) Is *Clostridium difficile* infection a particular problem after reversal of ileostomy? *Colorectal Dis* 13:308–311
8. Abe I, Kawamura YJ, Sasaki J et al (2012) Acute fulminant pseudomembranous colitis which developed after ileostomy closure and required emergent total colectomy: a case report. *J Med Case Rep* 6:130
9. Wilson MZ, Hollenbeak CS, Stewart DB (2013) Impact of *Clostridium difficile* colitis following closure of a diverting loop ileostomy: results of a matched cohort study. *Colorectal Dis* 15:974–981
10. Fekety R (1997) Guidelines for the diagnosis and management of *Clostridium difficile*-associated diarrhea and colitis. American College of Gastroenterology, Practice Parameters Committee. *Am J Gastroenterol* 92:739–750
11. Theodoropoulou A, Koutroubakis IE (2008) Ischemic colitis: clinical practice in diagnosis and treatment. *World J Gastroenterol* 14:7302–7308
12. Williams L, Armstrong MJ, Finan P et al (2007) The effect of faecal diversion on human ileum. *Gut* 56:796–801