

Case Report

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Ileal Perforation in a Patient with Ulcerative Colitis after Proctocolectomy

Mikiya Takao¹, Kazuhito Sasaki^{1,2*}, Keisuke Hata², Tatsuo Iijima³, Fuyo Yoshimi¹, Toshiaki Watanabe²

¹Department of Surgery, Ibaraki Prefectural Central Hospital and Cancer Center, 6528 Koibuchi, Kasama, Ibaraki 309-1793, Japan

²Department of Surgical Oncology, Faculty of Medicine, The University of Tokyo, 7-3-1, Hongo, Bunkyo-ku, Tokyo 113-8655, Japan

³Department of Pathology, Ibaraki Prefectural Central Hospital and Cancer Center, 6528 Koibuchi, Kasama, Ibaraki 309-1793, Japan

*Corresponding author: Kazuhito Sasaki, Department of Surgical Oncology, Faculty of Medicine, The University of Tokyo, 7-3-1, Hongo, Bunkyo-ku, Tokyo 113-8655, Japan, E-mail: sasakik-ky@umin.ac.jp

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Abstract

Ulcerative colitis (UC) is a form of inflammatory bowel disease that typically involves the colorectum; it has been reported that a certain proportion of patients with UC also develop ileitis, leading to ileal perforation in very extreme cases. We report a 66-year-old male with UC who presented with ileal perforation eight days after proctocolectomy. Although this situation is very rare, differential diagnoses for small bowel perforation after UC surgery could include backwash ileitis, cytomegalovirus (CMV) infection, Crohn's disease, diffuse enteritis, ischemic enteritis, Behçet's disease, medication adverse effect, and iatrogenic injury. Of these, backwash ileitis or diffuse enteritis is the most probable diagnosis in our case. Granulomas and transmural lymphoid aggregates with associated mucosal ulceration were absent. In addition, no signs or symptoms suggestive of Crohn's disease were seen postoperatively. Thus, the original diagnosis was likely fulminant UC. Infectious enteritis (including CMV), ischemic enteritis, and Behçet's diseases were clinically ruled out. Stool cultures and CMV antigen testing were negative. Moreover, histopathology revealed no evidence of CMV infection. Only a few cases of ileal perforation after UC surgery have been reported thus far. Surgeons should evaluate for perforation of the small bowel intraoperatively. Resection of the affected ileum is still a matter of debate. Although the inflammation is usually reversible and preservation of the distal ileum is vital for the creation of an ileal pouch and the avoidance of high output, the rare possibility of ileal perforation should be kept in mind in extreme cases of fulminant UC.

Keywords: Backwash ileitis; Ileal perforation; Postoperative course; Ulcerative colitis

Introduction

Ulcerative colitis (UC) is characterized by chronic intestinal inflammation, which is usually continuous from the rectum and is typically confined to the colorectum. As opposed to Crohn's disease (CD), small bowel inflammation is relatively rare in UC. Moreover, small bowel perforation after colectomy is extremely rare in UC patients. Here we report a 66-year-old male with UC who developed ileal perforation on postoperative day (POD) eight after proctocolectomy. To the best of our knowledge, ileal perforations after colectomy for UC are extremely rare, and only a few cases have been reported in the English literature [1,2].

Case Report

A 66-year-old male presented with bloody diarrhea. His medical history included cerebral infarct and hypertension. He took aspirin and cilostazol orally to prevent recurrent cerebral infarct. His family history was unremarkable. Colonoscopy revealed mucosal granularity, edema, and erythema diffusely and continuously from the rectum to the cecum (Figure 1A). The terminal ileum was normal (Figure 1B). Biopsy showed inflammatory cell infiltration in the lamina propria, cryptitis, and crypt abscesses, and he was diagnosed with pancolitis-type UC.

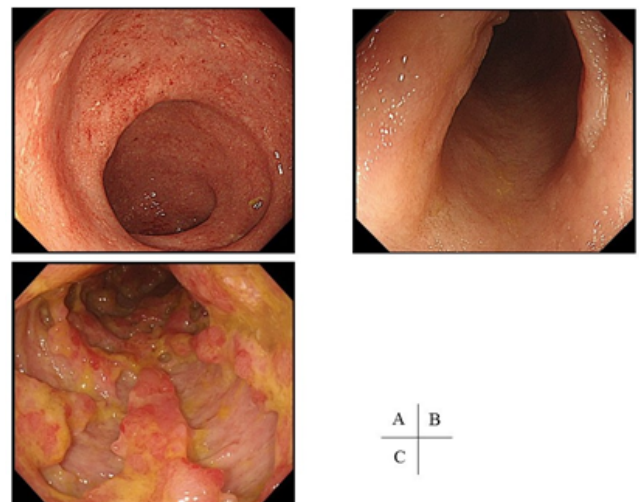


Figure 1

Figure 1: (A) Colonoscopy revealed mucosal granularities, edema, and erythema diffusely and continuously from the rectum to the cecum. (B) The terminal ileum was normal. (C) Follow-up colonoscopy was performed up to the transverse colon the day prior to proctocolectomy, revealing fissuring ulcerations and mucosal inflammation.

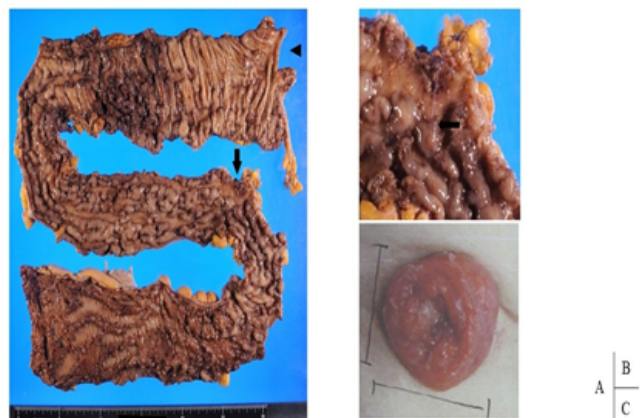


Figure 2: (A) The proctocolectomy specimen showed a small perforation of the sigmoid-descending colon junction (black arrow) and widespread deep ulcerations, but inflammation of the cecum and the ileocecal valve (black arrowhead) appeared relatively mild macroscopically. (B) A magnified view of the perforation of the sigmoid-descending colon junction (black arrow). (C) The mucosa of the ileostomy on the day following proctocolectomy was grossly normal.

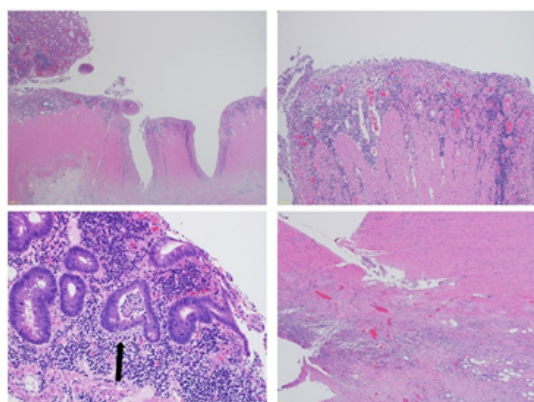


Figure 3

Figure 3: Histopathology of the proctocolectomy specimens (hematoxylin and eosin stain). (A) Widespread deep ulceration ($\times 20$), (B) inflammatory cells infiltrating the muscularis proper ($\times 100$), (C) crypt abscesses (black arrow, $\times 200$), and (D) transmural inflammation near the perforation site ($\times 40$) were seen.

He was unresponsive to intravenous prednisolone infusion therapy (Figure 1C), and because his general condition deteriorated, proctocolectomy and ileostomy were performed. Laparotomy showed a large amount of ascites and a small perforation of the sigmoid-descending colon junction. Intraoperatively, edema and erythema involved the entire colorectum and the ileum 20 to 70 cm proximal to the ileocecal valve. We preserved the affected ileum for future pouch surgery and resected only the proximal ileum beginning at the ileocecal valve (Figure 2A and 2B). The mucosa at the ileostomy was

grossly normal (Figure 2C). Histopathology of the surgical specimens showed widespread deep ulceration (Figure 3A and 3B), crypt abscesses (Figure 3C), and transmural inflammation most notable near the perforation site (Figure 3B and 3D), but no evidence of granulomatous lesions; these findings were consistent with fulminant UC. Though the inflammation of the cecum and the ileocecal valve appeared relatively mild macroscopically (Figure 2A), it was microscopically severe with erosions and ulceration.

From POD four, he required blood transfusion because of bloody discharge from the ileostomy. On POD eight, he suddenly developed abdominal pain, and his systolic blood pressure decreased to 70 mmHg. Computed tomography revealed a large amount of ascites and free air, indicating gastrointestinal perforation. Laparotomy showed ileal perforation approximately 30 cm proximal to the ileostomy. We resected the entire affected ileum, approximately 70 cm in length, and reconstructed the ileostomy (Figure 4).

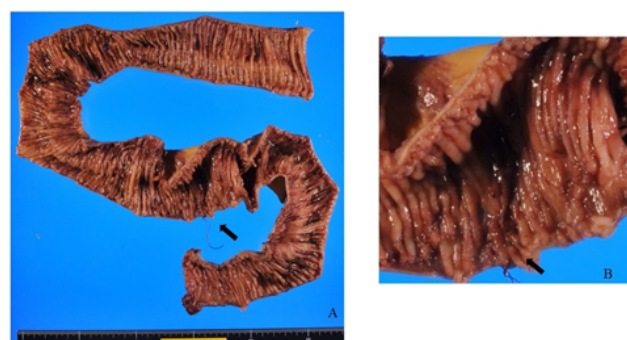


Figure 4

Figure 4: (A) The ileectomy specimen showed multiple ulcerations with a perforation (black arrow) approximately 30 cm proximal to the previous ileostomy. (B) A magnified view of the perforation of the ileum (black arrow).

Histopathology of the ileal resection specimens showed multiple ulcerations with perforation; the remaining mucosa contained scattered crypt abscesses (Figure 5A). The inflammatory cell infiltrate was primarily limited to the submucosal layer (Figure 5B), but showed focal transmural involvement, particularly near the perforation site (Figure 5C). No granulomatous lesions or gland metaplasia were identified. The patient required intensive postoperative care, but was discharged ambulatory with a good clinical course on POD 34 following the second surgery.

Discussion

To the best of our knowledge, ileal perforations after UC surgery are extremely rare, and only one case with cytomegalovirus (CMV) reactivation is reported in the English literature [1,2]. The differential diagnosis for small bowel perforation after UC surgery includes baskwash ileitis (BWI), CMV infection [1], CD [3], diffuse enteritis [4], ischemic enteritis, Behçet's disease, medication adverse effect, and iatrogenic injury. Of these, BWI or diffuse enteritis is the most likely diagnosis in our case. The most specific markers of CD, granulomas and transmural lymphoid aggregates with associated mucosal ulceration [3], were absent. In addition, no symptoms or signs suggestive of CD were seen postoperatively. Thus, the original

diagnosis was likely fulminant UC. Other causes such as infectious enteritis (including CMV), ischemic enteritis, and Behçet's disease were clinically ruled out. Stool cultures and CMV antigen testing were negative. Moreover, histopathology revealed no evidence of CMV infection. Medication-induced enteritis due to aspirin could not be completely ruled out, but was unlikely, as he had taken aspirin for 16 years.

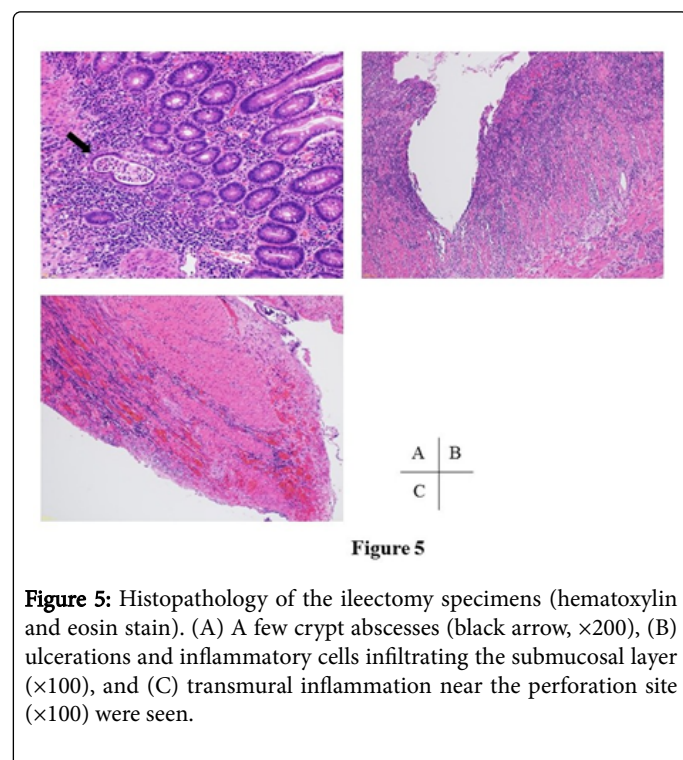


Figure 5

Figure 5: Histopathology of the ileectomy specimens (hematoxylin and eosin stain). (A) A few crypt abscesses (black arrow, ×200), (B) ulcerations and inflammatory cells infiltrating the submucosal layer (×100), and (C) transmural inflammation near the perforation site (×100) were seen.

BWI was originally thought to be the result of reflux of colonic contents into the terminal ileum through an incompetent ileocecal valve, causing continuous ileal involvement. However, no evidence supports this theory, and its precise etiology, criteria, and surgical treatment strategy remain controversial [5-7]. Recent reports indicate that ileitis is found in about 17-22% of resected specimens in UC, and is usually superficial, mild, and confined to the short-segment terminal ileum, consistent with the backwash theory [5-7]. However, some authors are skeptical about this theory, because a minority of ileitis cases are not consistent with BWI, showing deep, diffuse, or discontinuous involvement [5,6] Okita et al. [8] reported a case of perforated backwash ileitis. They resected the perforation site, 15 cm from the ileocecal valve, but were able to preserve the majority of the

ileum. Inflammation of the cecum, ileocecal valve, and ileostomy was macroscopically much milder than that of the ileum near the perforation site, which was atypical for BWI.

Corporaal et al. [4] reviewed 42 cases of enteritis in patients with well-established UC and reported a case of multiple jejunal perforations shortly after colectomy. Eighty-one percent of patients presented with enteritis postoperatively, and responded well to steroids or calcineurin inhibitors, suggesting that a postoperative change in inflammatory mediators or inhibitors may be an underlying cause. In our case, we assume that preexisted atypical ileitis was exacerbated postoperatively. Considering that perforation occurs after colectomy in most cases [2], the postoperative exacerbation might be related to the underlying etiology of diffuse enteritis after UC surgery, such as a sudden change in inflammatory mediators [4]. Furthermore, steroids were being tapered in our patient, which might have played a role in the development of ileitis.

Surgeons should evaluate for possible perforation of the small bowel intraoperatively. Resection of the affected ileum is still a matter of debate. The inflammation is usually reversible and preservation of the distal ileum is vital for the creation of an ileal pouch and the avoidance of high output [8]. Despite its rarity, ileal perforation should be kept in mind in extreme cases of fulminant UC.

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