International Journal of Surgery Case Reports 84 (2021) 106020

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr

Case report

Whole jejunoileal diverticulosis with recurrent inflammation and perforation: A case report

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ARTICLE INFO

Keywords:
Diverticulitis
Perforation
Abscess
Jejunoileum
Small intestine
Hemodialysis

ABSTRACT

Introduction: Jejunoileal diverticulitis is uncommon and poorly understood. We report a case of whole jejunoileal diverticulosis with recurrent inflammation and perforation.

Case presentation: A 72-year-old man with hemodialysis presented with fever and abdominal pain. The patient had a medical history of twice having jejunoileal diverticulitis. Serum testing indicated a white blood cell count of 15,670/μL and a C-reactive protein level of 10.31 mg/dL. Contrast-enhanced computed tomography showed jejunoileal diverticulosis with the concomitant mesenteric fat opacity and a 60-mm × 45-mm mass lesion containing extraluminal air bubbles. Jejunoileal partial resection was performed. Multiple diverticulosis was recognized over the entire jejunoileum, and the pouches existed along entry points of the bowel vascular supply through the mesentery. Intestinal resection was limited to the intestinal loop associated with complicated diverticulitis with abscess. Macroscopic examination revealed multiple jejunoileal diverticulosis. In the reddened mucosa, the diverticulitis and mesenteric perforation were recognized. Microscopic examination showed protrusion of mucosal and submucosal layers through a defect in the muscular layer with gangrenous inflammation. These findings supported a diagnosis of jejunoileal diverticulitis with perforation and abscess. The patient had no postoperative complications and no recurrence within 6 months.

Discussion: Treatment for jejunoileal diverticulitis should be individualized for each patient according to their degree of inflammation, recurrence, and the patient's background.

Conclusion: Extensive diverticulosis over the entire jejunoileum is very rare. In this case, the section of the inflamed diverticulosis can be distinguished and resected to avoid a short-bowel syndrome, which should lead to an uneventful postoperative course.

1. Introduction

Diverticulosis can occur in any portion of the gastrointestinal tract from the upper esophagus to the colon. In the small intestine, jejunoileal diverticulosis is rarer than Meckel and duodenal diverticulosis [1], with an incidence of 2.0%–2.3% on small bowel follow through [2,3] and 0.26% at autopsy [4]. Only 10% of the patients have complications, such as bleeding and inflammation [5]. Therefore, jejunoileal diverticulitis is uncommon in clinical practice and poorly understood. Here we report a case of a patient with whole jejunoileal diverticulosis with recurrent inflammation and perforation and review the relevant literature. This work has been reported in line with the SCARE 2020 criteria [6].

2. Case presentation

A 72-year-old man presented with fever and abdominal pain. The patient underwent hemodialysis and had a medical history of twice having jejunoileal diverticulitis with perforation 7 months and 2 weeks before this hospitalization (Fig. 1a, b). The patient had been treated with tazobactam/piperacillin antibiotics therapy because the perforation had been localized and was accompanied only by mesenteric air.

On this admission, physical examination revealed periumbilical tenderness. Serum testing indicated an elevated white blood cell count of 15,670/μL, and a C-reactive protein level of 10.31 mg/dL. Contrast-enhanced computed tomography showed multiple jejunoileal diverticulosis (Fig. 1c) with concomitant mesenteric fat opacity and a 60-mm × 45-mm mass lesion containing extraluminal air bubbles (Fig. 1d).

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https://doi.org/10.1016/j.ijscr.2021.106020
Received 10 April 2021; Received in revised form 22 May 2021; Accepted 22 May 2021
Available online 27 May 2021

Ascending and sigmoid colonic diverticulosis were also observed. Therefore, recurrent and exacerbating jejunoileal diverticulitis with perforation and abscess was diagnosed preoperatively.

Jejunoileal partial resection was performed. Multiple diverticulosis was recognized over the entire jejunoileum, and the pouches were present along entry points of the bowel vascular supply through the mesentery (Fig. 2a). The abscess complicating the diverticulitis involved the adjacent non-inflammatory small intestine (Fig. 2b) and contained pus identified as *Escherichia coli* and Lancefield group F-Streptococcus after culturing (Fig. 2c). The intestinal resection was limited to the intestinal loop associated with complicated diverticulitis with abscess. The resected intestinal length was 100 cm, and the residual small intestinal length was 320 cm. Macroscopic examination revealed multiple jejunoileal diverticulosis (Fig. 3a, b). In the reddened mucosa, diverticulitis, and mesenteric perforation were recognized (Fig. 3b, c). Microscopic examination revealed protrusion of the mucosal and submucosal layers through a defect in the muscular layer (Fig. 4a). Additionally, inflammatory cells, including macrophages, had infiltrated, and inflammatory granulation tissue under the serosa was observed (Fig. 4b). These findings were consistent with a diagnosis of jejunoileal diverticulitis with perforation and abscess. Negative pressure wound therapy was performed without surgical skin suture for wound closure and there were no postoperative complications. The patient was discharged 39 days after the surgery and recurrence of jejunoileal diverticulitis was not observed within 6 months after the surgery.

3. Discussion

Extensive diverticulosis in the entire jejunoileum is very rare. Our patient's clinical course differed from that of uncomplicated jejunoileal diverticulitis, and surgical intervention was performed considering the patient's severe and recurrent inflammation and his background of hemodialysis. Even though the multiple diverticulosis was present over the entire jejunoileum, the section of diverticulitis was localized, so only a part of the intestine was resected to avoid short-bowel syndrome. A favorable outcome was achieved.

The cause of diverticulosis is explained by the theory of locus minoris resistentiae [7]. The presence of a weakened area in the bowel wall, which is the entry point of the blood vessels through the muscular coat, and increased intraluminal pressures induce herniation of the mucosa and submucosa through the muscular wall. In fact, jejunoileal diverticulosis is pathologically an acquired pseudodiverticulosis and usually seen in the mesenteric border of the small intestine in people $\geq$60 years old [5,8]. It is unclear why diverticulosis occurs in the jejunoileum and not the colon, but nearly 60% of patients with small-bowel diverticulosis have coexisting colonic diverticulosis [1–4,9]. Our case also showed that diverticulosis was present in the entire jejunoileum in addition to the colon, and it was assumed to have been caused by tissue vulnerability.

Treatment for jejunoileal diverticulitis should be individualized for each patient according to their degree of inflammation, recurrence, and the patient's background. First, inflammation of diverticulitis can range from uncomplicated to severe conditions associated with gangrene and a localized or generalized perforation. Jejunoileal diverticulitis tends to perforate and develop abscesses [10]. Most of the perforations are localized in the root of the mesentery because the diverticula are usually walled off by adjacent small-bowel mesenteric leaves [11]. The first-line treatment against jejunoileal diverticulitis with localized perforations is controversial. Rangan [12] reports that nonoperative management,

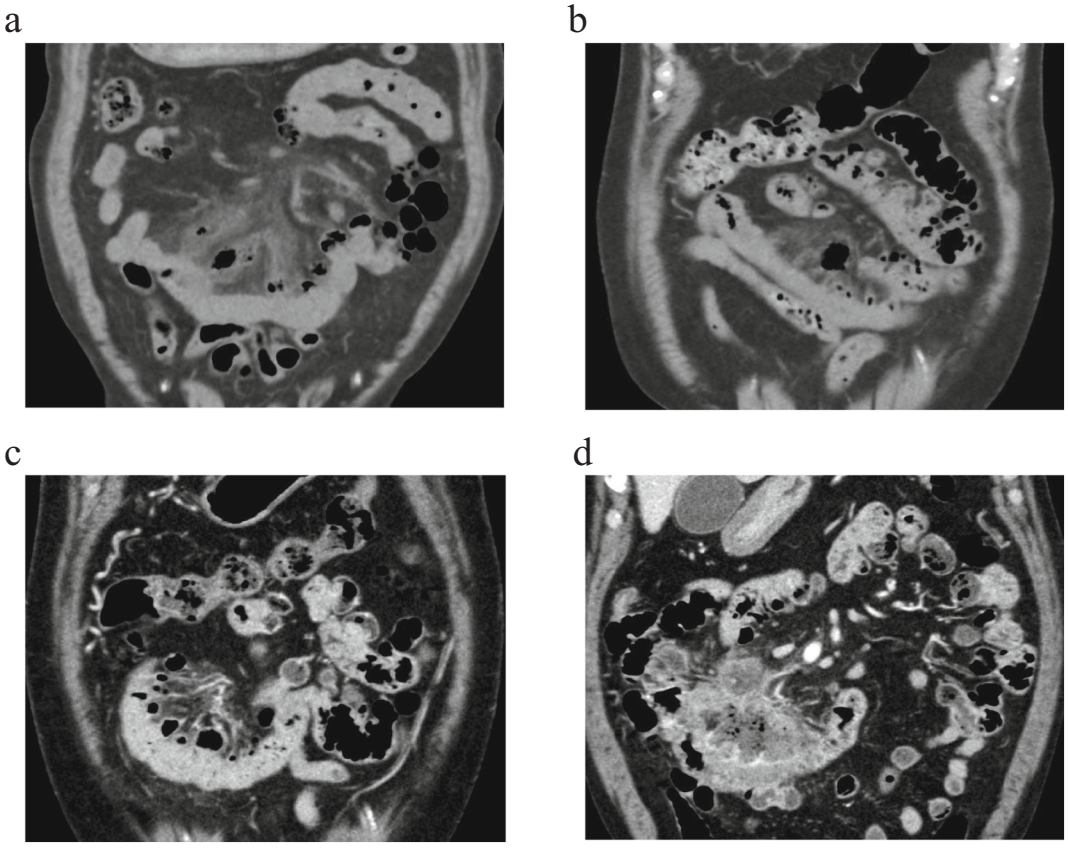

Fig. 1. Preoperative computed tomography images.
(a) Jejunoileal diverticulitis with perforation 7 months before this hospitalization. (b) Jejunoileal diverticulitis with perforation 11 days before this hospitalization. (c) Multiple jejunoileal diverticulosis along the blood vessels. (d) Jejunoileal diverticulitis accompanied by mesenteric fat opacity and a 60-mm × 45-mm mass lesion containing extraluminal air bubbles.

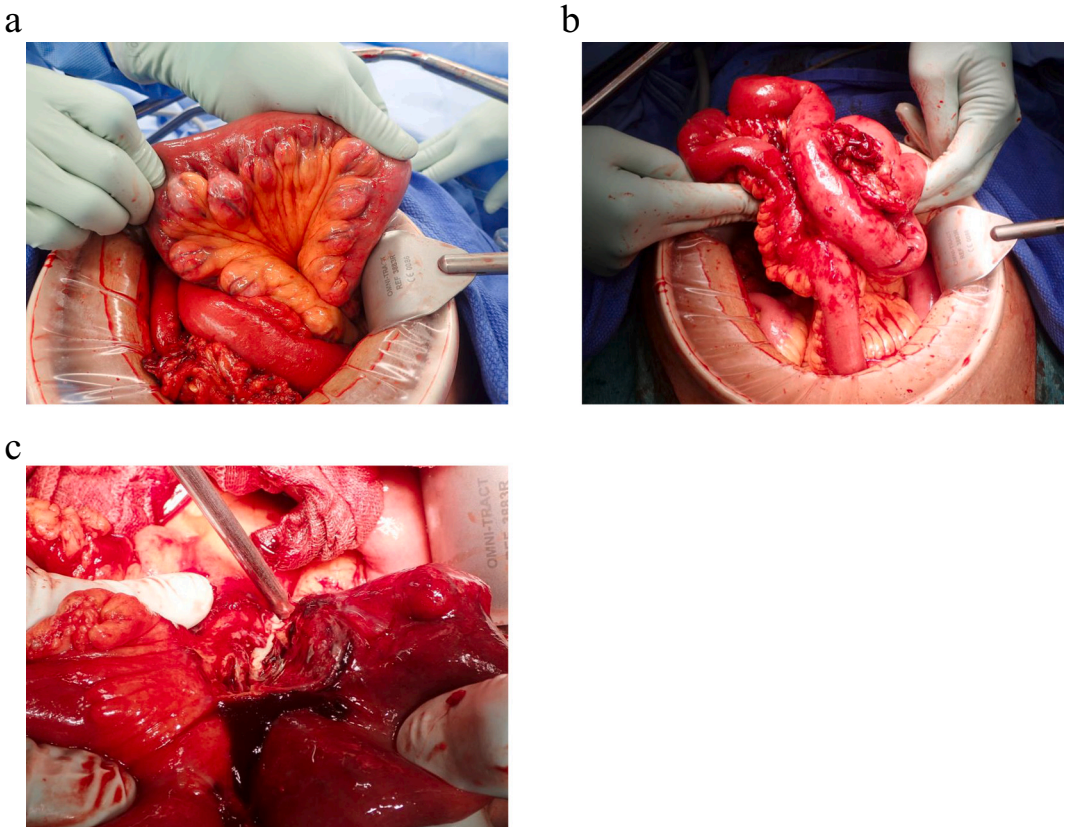

Fig. 2. Intraoperative findings.
(a) Extensive jejunoileal diverticulosis formed along entry of the blood vessels in the mesentery. (b) The abscess complicating the diverticulitis involved non-inflammatory intestine. (c) Pus contained in the abscess.

including antibiotics and percutaneous drainage, can be considered. However, some conservative treatment reportedly has failed for exacerbation, and surgery was finally performed [13,14]. In our case, surgery was performed because of recurrent and exacerbating inflammation although the diverticulitis with abscess was localized.

Second, regarding the recurrence rate of jejunoileal diverticulitis, there is no practice data available. Our patient had been previously treated with antibiotics twice but resulted in prolonged inflammation due to recurrence and exacerbation of jejunoileal diverticulitis with perforation. Consequently, an abscess formed and involved the adjacent non-inflammatory small intestine, so resection of a longer intestinal length was needed. Our case suggested the possibility that surgical intervention at the earlier onset stage of diverticulitis with perforation avoided combined resection of the adjacent non-inflammatory small intestine.

Finally, the patient's background is very important when selecting treatment. Hemodialysis is a lifesaving treatment for patients with end-stage renal disease, but patients with hemodialysis have immune-system dysfunction. The immunodeficiency is considered to be involved in monocytes. Both the uremia itself and hemodialysis membrane could induce an increased monocyte apoptosis [15]. Other studies have reported that monocytes in patients with hemodialysis are hyporesponsive to lipopolysaccharide for their interleukin-1β, tumor necrosis factor-α, and interleukin-6 production [16]. In our case, *Escherichia coli*, the bacterial outer membrane of which is mainly composed of lipopolysaccharide, was detected in the abscess. The compromised condition in hemodialysis might have been a factor in the recurrent and exacerbating diverticulitis. For these reasons, surgical intervention in our case was appropriate. When considering the surgery for the inflamed jejunoileum, we thought that less invasive surgery should be performed and that the optimal range of intestinal resection should only include the severe inflamed small intestine to avoid short-bowel syndrome, similar to Crohn's disease [17,18]. Our patient had multiple diverticulosis over the entire jejunoileum, but partial resection of only the inflamed intestine was performed. The postoperative course was uneventful.

4. Conclusion

We treated a patient who had whole jejunoileal diverticulosis with recurrent inflammation and perforation. In the case, the section involving only the inflamed diverticulosis could be distinguished and was resected to avoid short-bowel syndrome. It led to the uneventful postoperative course.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

The case report is exempt from ethical approval in our institution.

Funding

This research did not receive any specific grant from funding

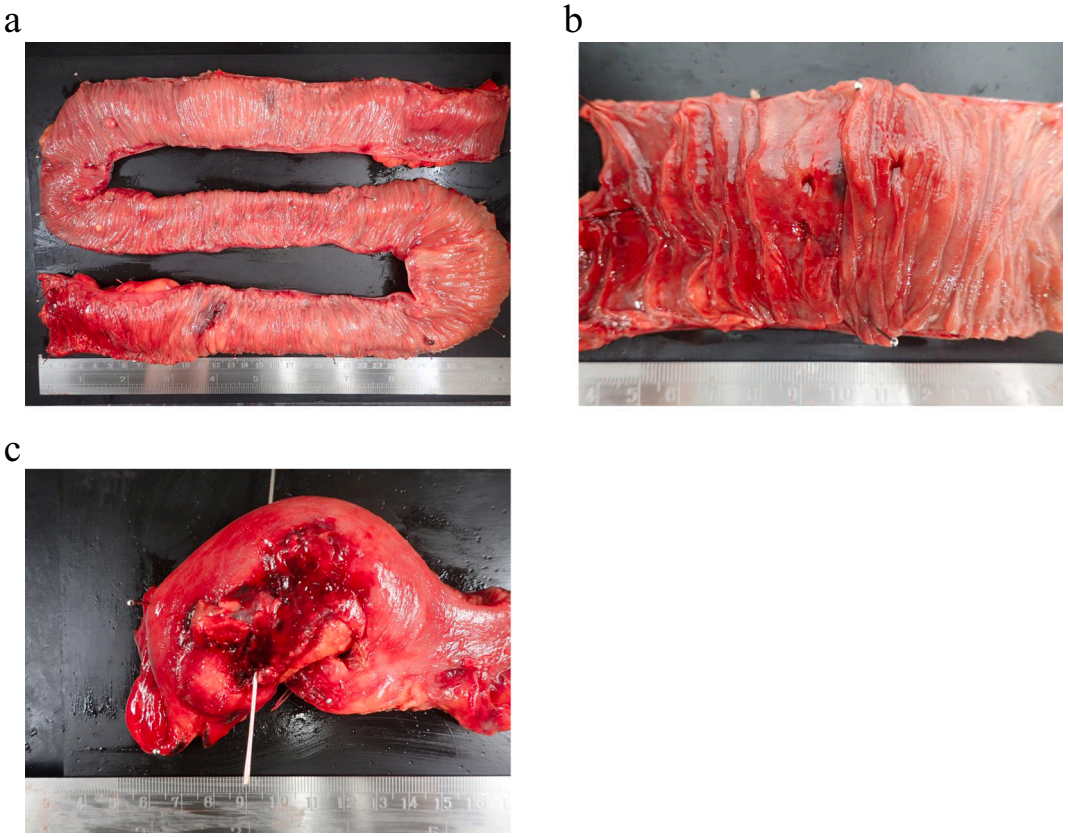

Fig. 3. Macroscopic examination of the resected specimen.
(a) Resected jejunoileum. (b) Diverticulitis with the mucosal reddening. (c) Perforated diverticulum in the resected segment of the jejunoileum.

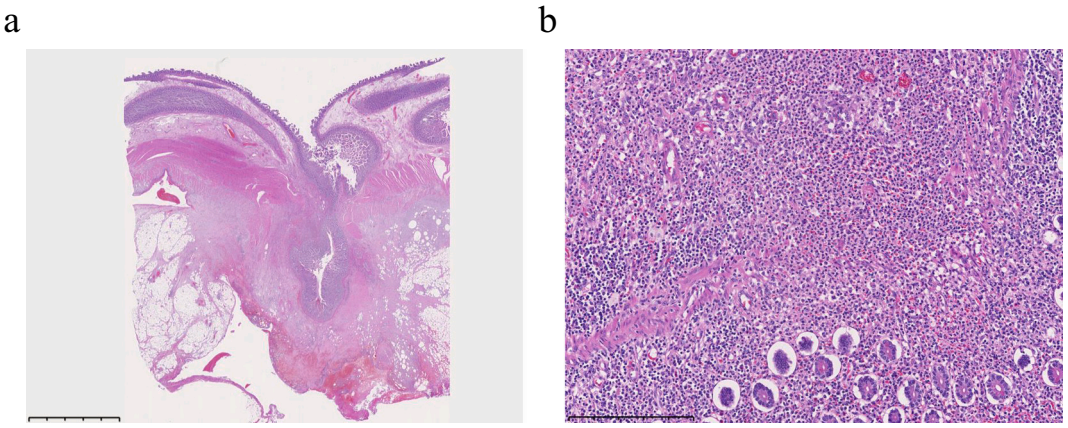

Fig. 4. Histopathological examination.
(a) Protrusion of mucosal and submucosal layers through a defect of the muscular layer. Scale division: 1 mm. (b) Inflammatory cell infiltration and inflammatory granulation tissue under the serosa. Scale division: 50 μm.

agencies in the public, commercial, or not-for-profit sectors.

Guarantor

Yoshifumi Watanabe.

Research registration number

Not applicable.

CRediT authorship contribution statement

YW drafted the manuscript and provided the original pictures. MM, MH, and RS participated in treating the patient and revised the manuscript. All authors read and approved the final manuscript.

Declaration of competing interest

None.

Acknowledgements

None.

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