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Ileo-ileal fistula with severe malnutrition caused by strangulated ileus surgery while preserving ischemic ileum: A case report

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ABSTRACT

INTRODUCTION: Entero-enteric fistulas are rare complications that occur in patients with inflammatory bowel disease and other intestinal diseases. In this report, we present an ileo-ileal fistula accompanied by severe malnutrition caused by strangulated ileus surgery while preserving the ischemic ileum in a very elderly patient.

CASE PRESENTATION: A 90-year-old woman underwent emergency surgery without bowel resection for strangulated ileus in another hospital. Minor abdominal pain and slight fever persisted after surgery. She lost weight, losing approximately 10 kg within half a year. She gradually became difficult to move due to dyspnea upon exertion and generalized edema and visited at our hospital. Pleural effusions, ascites and severe malnutrition were observed. An elastic hard mass with mild tenderness was palpated in her abdomen. Computed tomography showed a loop-like ileum and ileo-ileal fistula with adjacent fat stranding. We performed a partial small bowel resection. The resected specimen demonstrated an ileo-ileal fistula and circumferential ulceration in the loop-like adhesion. After the operation, the nutrition status was resolved immediately without any medications.

DISCUSSION: In cases of strangulated ileus, there are no deterministic criteria for evaluating intestinal blood flow. This is the first report of ileo-ileal fistula onset after surgery for strangulated ileus without intestinal resection. Furthermore, this fistula caused severe malnutrition duo to chronic inflammation, ulcer formation, and the blind-loop syndrome.

CONCLUSIONS: When preserving the intestinal tract in the operation of strangulated ileus, the occurrence of entero-enteric fistulas should be considered. Since malnutrition in the elderly is a serious problem, it should be treated promptly.

1. Background

In cases of strangulated ileus, intestinal preservation should be attempted to the greatest extent possible; however, there are no deterministic criteria for evaluating intestinal blood flow [1–3]. Therefore, judgment may be difficult in some cases [1–3]. Entero-enteric fistula is a rare complication that occurs in patients with inflammatory bowel disease and other intestinal diseases [4]. However, there has been no reported onset in cases where surgery was performed for strangulated ileus without intestinal resection. Herein, we present an ileo-ileal fistula accompanied by severe malnutrition caused by strangulated ileus surgery while preserving the ischemic ileum in a very elderly patient. This work has been reported in line with the SCARE criteria [5].

2. Case presentation

A 90-year-old woman underwent emergency surgery for a strangulated ileus caused by an adhesion band in another hospital in July 2016. The patient had no previous disease other than a surgical history of uterine fibroids and hypertension. Approximately 50–100 cm from ileocecal valve, a loop-like adhesion of the ileum approximately 15 cm in length was observed. The bowel resection was not performed, because after cutting the band, intestinal blood flow improved upon observing the serosal surface. However, the details of these surgeries were unknown because there was no surgical picture. Sub-ileus had repeatedly occurred after surgery, but it was improved by conservative treatment. Several months after surgery, the symptoms of intestinal obstruction gradually dis-

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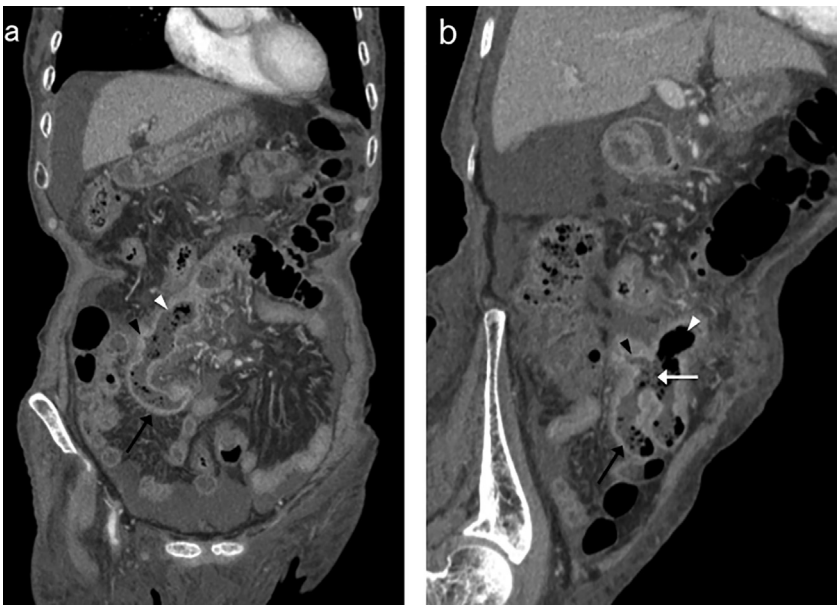

Fig. 1. CT revealed a loop-like ileum and ileo-ileal fistula with adjacent fat stranding. Black arrow: a loop-like ileum, white arrow: ileo-ileal fistula, white arrow-head: oral side ileum, black arrow-head: anal side ileum.

appeared, but minor abdominal pain and slight fever persisted. The patient lost weight, losing approximately 10 kg within half a year after surgery. After mid-January 2017, dyspnea upon exertion and generalized edema appeared. She gradually became difficult to move and visited the department of General internal medicine in our hospital in February.

Pleural effusions, ascites and whole body edema were observed, and a blood test revealed severe hypoproteinemia: albumin, 1.11 g/dL; prealbumin, 7.09 mg/dL. In addition, hyper-inflammatory response, hypocholesterolemia, anemia, and low Triiodothyronine syndrome were observed (Table 1). Due to these findings, a chronic, severe, debilitating malnutrition caused by chronic inflammation was considered. The abdominal wall was soft, and bowel sounds were regular. An elastic hard mass with mild tenderness was palpated in the right lower quadrant. The patient had no history of diarrhea or any abdominal trauma.

Computed tomography (CT) was performed to investigate the abdominal mass. CT showed a loop-like ileum and ileo-ileal fistula with adjacent fat stranding (Fig. 1). We considered this site responsible for inflammation.

There was no evidence of nephrotic syndrome or protein-losing gastroenteropathy. The patient and her family refused both a small bowel enema and an endoscopy. Although oral feeding was possible, combined oral feeding and total parenteral nutritional support did not improve her nutritional status. Furthermore, mild abdominal pain and slight fever continued. We thought that surgical treatment was necessary, and the patient and her family agreed to surgery after a clear preoperative conversation. After receiving albumin preparation and a blood transfusion, the patient received an exploratory laparotomy two weeks after hospitalization.

Approximately 50 cm from ileocecal valve, a loop-like adhesion of the ileum approximately 15 cm in length was observed. These intestines were markedly reddened and indurated (Fig. 2). The omentum surrounded the loop-like adhesion of the ileum and became mass-like. The proximal bowel was slightly distended. We determined this site to be responsible for this disease condition and performed a partial small bowel resection. The postoperative course was uneventful. After a month of rehabilitation, she was discharged on her foot. Nutritional statuses including hypoproteinemia, anemia and hypocholesterolemia were completely improved after the operation without any medication (Fig. 3). Consequently, the pleural effusion, ascites and edema disappeared.

The resected specimen demonstrated an approximately 2 cm ileo-ileal fistula in the loop-like adhesion, as well as circumferential ulceration continuous with the fistula (Fig. 4). A pathological examination found ulceration and inflammatory granulation tissue around the fistula. In addition, acute and chronic inflammation and marked fibrosis were noted. These findings were consistent with the ischemic change caused by strangulated ileus. There was no evidence of inflammatory bowel disease or malignant transformation (Fig. 5).

3. Discussion

Intestinal fistulas represent a serious complication after abdominal surgery [6]. Entero-cutaneous fistulas are the most frequent of all intestinal fistulas and are the easiest to diagnose [6,7]. However, entero-enteric fistulae are a rather rare complication. They are typically diagnosed in a delayed manner due to a lack of specific and obvious symptoms. Most reports of entero-enteric fistulae were caused by Crohn's disease [4,8,9]. In several other cases, including duodenal ulcer, intestinal tuberculosis, Henoch-Schönlein purpura and necrotizing enteritis in children, iatrogenic and magnetic foreign body ingestion have been reported [4,10–13]. However, to the best of our knowledge, this is the first report of ileo-ileal fistula onset after surgery for strangulated ileus without intestinal resection was performed. In this study, it was hypothesized that the fistula formation was caused by ulceration and inflammatory adhesion due to an ischemic change of the ileum. This ischemic change was likely caused by a strangulated ileus, and the pathological examination was consistent with these findings. In surgical cases of strangulated ileus, intestinal blood flow and the possible need for bowel resection must be evaluated after the incarceration has been reduced [2]. However, there were no clear criteria for judging whether to preserve or remove the ischemic intestinal tract [1]. Therefore, evaluating bowel ischemia is difficult in some patients. Bowel resection is necessary when signs of irreversible ischemia are seen in the strangulated intestine. However, intestinal resection has risks of anastomotic leakage and stenosis. There are