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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 due 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.

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## 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 mal-

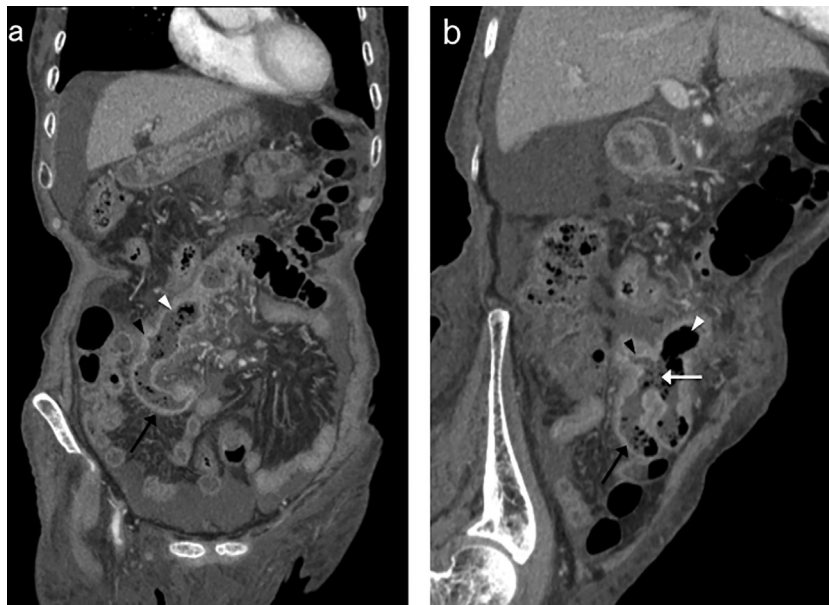
nutrition caused by strangulated ileus surgery while preserving the ischemic ileum in a very elderly patient. This work has been reported in line with the SCARE criteria [5].

## 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 post-operative course was uneventful. After a month of rehabilitation, she was discharged on her foot. Nutritional statuses including hypoproteinemia, anemia and hypocholesterolemia were com-

pletely 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

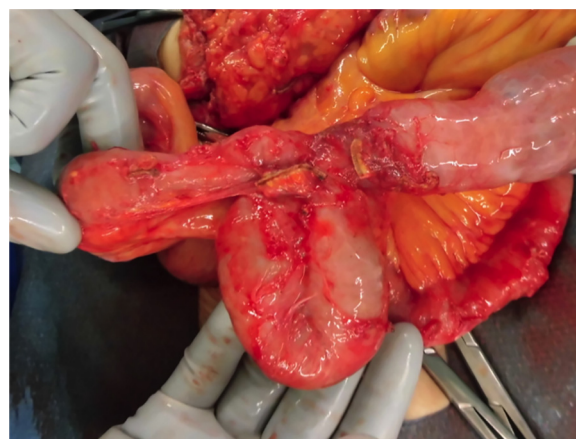
**Table 1**  
Laboratory Findings on Visiting Our Hospital.

		normal range
Peripheral blood		
WBC	6000/ $\mu$ L	(3300–8600)
RBC	$274 \times 10^4$ / $\mu$ L	(386–492)
Hb	7.4 g/dL	(11.6–14.8)
Ht	23.8%	(35.1–44.4)
MCV	86.9 fL	(83.6–98.2)
MCH	27.2 pg	(27.5–33.2)
MCHC	31.3 g/dL	(31.7–35.3)
PLT	$40.1 \times 10^4$ / $\mu$ L	(15.8–34.8)
Blood chemistry		
TP	3.76 g/dL	(6.60–8.10)
Alb	1.11 g/dL	(4.10–5.10)
Pre-Alb	7.09 mg/dL	(20.00–40.00)
T-Bil	0.17 mg/dL	(0.40–1.50)
AST	16 IU/L	(13–30)
ALT	11 IU/L	(7–23)
LDH	187 IU/L	(124–222)
ALP	173 IU/L	(106–322)
$\gamma$ -GTP	10 IU/L	(9–32)
AMY	43 IU/L	(44–132)
Fe	24 $\mu$ g/dL	(40–188)
UIBC	70 $\mu$ g/dL	(137–325)
TIBC	94 $\mu$ g/dL	(260–420)
Ferritin	33.3 ng/mL	(12.0–60.0)
BUN	19.5 mg/dL	(8.0–20.0)
CRE	1.16 mg/dL	(0.46–0.79)
UA	3.93 mg/dL	(2.60–6.99)
TC	107 mg/dL	(142–219)
TG	60 mg/dL	(30–149)
HDL-C	29.4 mg/dL	(40.0–103.0)
LDL-C	68 mg/dL	(65–139)
Glucose	102 mg/dL	(73–109)
CRP	4.65 mg/dL	(<0.30)
Na	134.7 mEq/L	(139.0–145.0)
K	4.37 mEq/L	(3.60–4.80)
Cl	106.4 mEq/L	(101.0–108.0)
Ca	6.91 mg/dL	(8.80–10.10)
TSH	2.28 $\mu$ IU/mL	(0.50–5.00)
FT3	1.15 ng/dL	(2.30–4.00)
FT4	1.14 pg/mL	(0.90–1.70)
Vit B1	23 ng/mL	(24–66)
Vit B12	1280 pg/mL	(180–914)
Folic acid	2.8 ng/mL	(>4.0)
Coagulating system		
PT	100.4%	(80.0–120.0)
APTT	36.1 s	(24.0–39.0)

several reports on intestinal blood flow evaluation methods, such as intraoperative indocyanine green fluorescence imaging, CT perfusion imaging, laser Doppler velocimetry, and tissue reflectance spectrophotometry [1–3,14,15]. To confirm these effects, further clinical studies are required.

In this study, the short-circuit due to ileo-ileal fistula was a short distance, and the main causes of severe malnutrition were thought to be chronic inflammation due to fistula, ulcer formation, and the blind-loop syndrome. The clinical symptoms of entero-enteric fistulas differ depending on which intestinal tract form the fistula. Diarrhea, weight loss, abdominal pain and other symptoms have been reported, but they are not specific [4]. Furthermore, elderly people may not have confirmed subjective symptoms. In addition, physicians occasionally hesitate to re-operate and invasively examine for elderly patients. We thought that these factors led to a delay in diagnosis in this case. With the increasingly aging population, the number of elderly patients also continues to increase worldwide. The appropriate examinations and treatments should not be avoided simply based on age.

It is widely accepted that chronic inflammation causes malnutrition [16–18]. Malnutrition causes various systemic dysfunctions such as decreased immunity [16,17]. Even with mild etiological rea-



**Fig. 2.** Intraoperative findings: Approximately 50 cm from the terminal ileum, a loop-like adhesion of the ileum approximately 15 cm in length was observed. These intestines were markedly reddened and indurated.

sons, the elderly easily deteriorate the general condition due to their frailties [7,19,20]. Especially in the very elderly, malnutrition may be fatal. Therefore, it is necessary to promptly investigate the cause and treat it.

There are a few limitations in this report. First, there is no detailed information on initial surgery. Therefore, it is difficult to conclude that strangulation ileus was the cause of the intestinal fistula. Second, since patient consent was not obtained, gastro-intestinal endoscopic examination was not performed. Therefore, inflammatory bowel disease has not been completely ruled out. However, since the general condition improved immediately by resection of the intestinal fistula, it is unlikely that other pathological conditions are related.

In addition, intraoperative findings showed no obvious abnormal findings in other intestinal tracts. Taken together with the patient's medical history, it is likely that strangulated ileus caused the ileo-ileal fistula.

**4. Conclusions**

We presented, to the best of our knowledge, the first case of an ileo-ileal fistula with severe malnutrition that occurred after strangulated ileus surgery in a very elderly patient. When preserving the intestinal tract in the operation of strangulated ileus, this complication should be considered. Since malnutrition in the elderly is a serious problem, it should be treated promptly.

**Conflicts of interest**

The authors have no conflicts of interest.

**Funding**

Satoshi Nishiwada and other participating authors have no involvement as above.

**Ethical approval**

This case report is not research study, therefore approval was not given.

The ethical approval has been exempted by our institution.

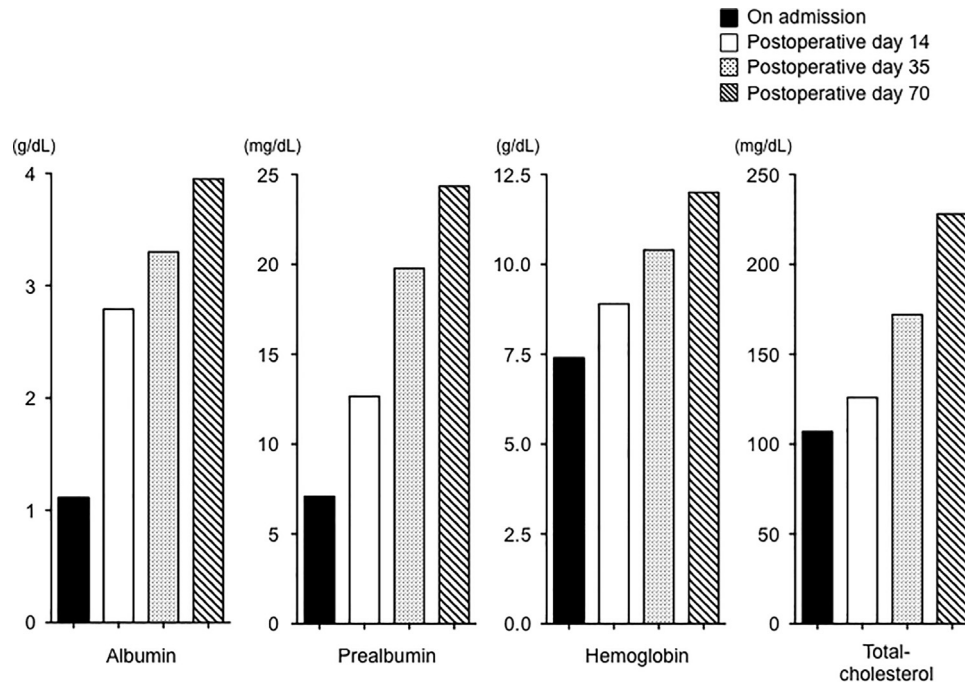


Fig. 3. Comparison of nutritional data before and after surgery: After operation, hypoproteinemia, anemia and hypocholesterolemia were resolved without any medication.

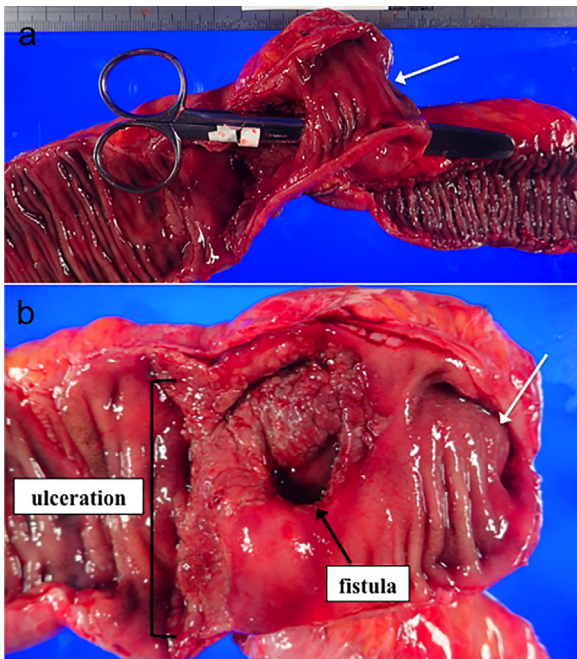


Fig. 4. The resected specimen demonstrated an approximately 2 cm ileo-ileal fistula in the loop-like adhesion (white arrow). Circumferential ulceration was observed around the fistula.

**Consent**

Written informed consent was obtained from the patient for publication of this case report.

**Authors contribution**

SN collected the patient’s clinical data, analyzed the data and was a major contributor in writing the manuscript; SN designed and drafted the manuscript; TT was involved with acquisition of

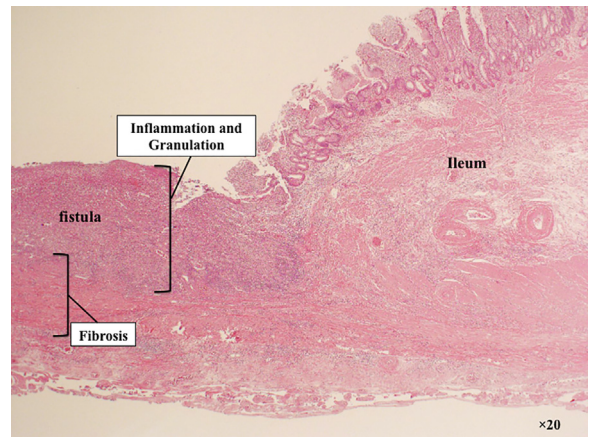


Fig. 5. A pathological examination found ulceration and inflammatory granulation tissue around the fistula. In addition, acute and chronic inflammation and marked fibrosis were noted. There was no evidence of inflammatory bowel disease or malignancy.

data as well as analysis and interpretation of data and drafting the manuscript; YK collected the clinical data; DN collected the radiological data; NS was involved with acquisition of data as well as analysis and interpretation of data and drafting the manuscript; NF and HI collected data; AY was involved with acquisition of data and revising it critically for important intellectual content. All authors read and approved the final manuscript.

**Guarantor**

Satoshi Nishiwada and Atsushi Yoshimura accept full responsibility for the work.

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