CASE REPORT

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Ileo-ileal knot: a rare case of acute strangulated intestinal obstruction

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ABSTRACT

Strangulated intestinal obstruction is one of the most common types of acute abdomen and requires urgent surgical treatment. Herein, we report a very rare case of strangulated intestinal obstruction caused by an ileo-ileal knot. An 80-year-old woman was admitted to our hospital with suspicion of strangulation ileus and underwent emergency laparotomy after investigation by exploratory single-port laparoscopy. During surgery, a small bowel gangrene caused by an ileo-ileal knot was found. The gangrenous segment was resected, and primary anastomosis was performed. Post-operative recovery was uneventful except for a minor wound infection. Our extensive search of the literature found only 7 case reports of ileo-ileal knot including ours. An ileo-ileal knot should be considered in the differential diagnosis of acute intestinal obstruction, because this rare phenomenon requires urgent surgical treatment; and some complications should be considered during or after surgery.

Key Words: ileo-ileal knot, ileo-ileal knotting, intestinal obstruction, gangrene, acute abdomen

INTRODUCTION

Strangulated intestinal obstruction is a relatively common type of acute abdomen and requires urgent surgical treatment. The causes of strangulated intestinal obstruction are many including primary volvulus, hernias, adhesions, bands, and intussusceptions. Intestinal knot formation is the obstruction of an intestinal segment displaying the closed loop phenomenon due to a knot of the mesentery and is an unusual cause of strangulated intestinal obstruction.[1)] Several types of intestinal knot formation have been reported, i.e., appendico-ileal, ileo-caecal, ceco-sigmoid, ileo-sigmoid, and ileo-ileal.[2, 3)] Among them, the ileo-ileal knot is a very rare entity.[2)] Here, we present a rare case of an acute strangulated intestinal obstruction caused by an ileo-ileal knot in an elderly female who required emergency surgical treatment to save her life.

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CASE PRESENTATION

An 80-year-old woman was transferred to our hospital from another hospital due to suspicion of a strangulation intestinal obstruction. She had a history of advanced rectal carcinoma, and chemotherapy had been performed after loop colostomy of her transverse colon. Also, adnexectomy had been performed previously to allow torsion of the ovarian tube. Severe abdominal pain suddenly occurred during infusion for chemotherapy at the previous hospital about 6 hours before admission to our hospital. In the physical examination, she seemed to be severely ill; however, her vital signs were stable. Her abdomen was distended with rebound tenderness, especially in the lower quadrant. Laboratory investigation yielded the following results: white blood cell count, 8220/μL (normal, 3300–8190/μL); neutrophils, 93.2% (normal, 37.4–68.5%); C-reactive protein, 0.99 mg/dL (normal, <0.25mg/dL); procalcitonin, 0.09 ng/ml (normal, <0.05 ng/ml); base excess, –4.3 mmol/L (normal, 0±2 mmol/L); and lactic acid, 39.6 mg/dL (normal, 4.5–18.0 mg/dL). An abdominal X-ray taken at the previous hospital showed multiple distended small bowel loops with air-fluid levels (Fig. 1A). Also, contrast-enhanced computed tomography (CT) performed at that hospital showed that a part of small intestine was poorly enhanced, showing a whirl sign (Fig.1B and 1C). CT performed at our hospital indicated increment of ascites. Based on these findings, an emergency exploratory operation was required.

Starting an operation by using single-port laparoscopy is recommended in our department

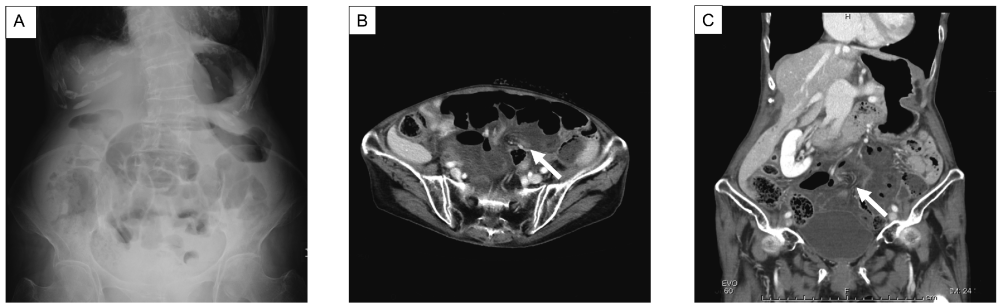

Fig. 1(A) Abdominal radiograph showing multiple distended small bowel loops with air-fluid levels. (**B, C**) Axial and coronal contrast-enhanced CT images (B: axial, C: coronal). CT images showed that a part of small intestine was poorly enhanced with torsion (arrows).

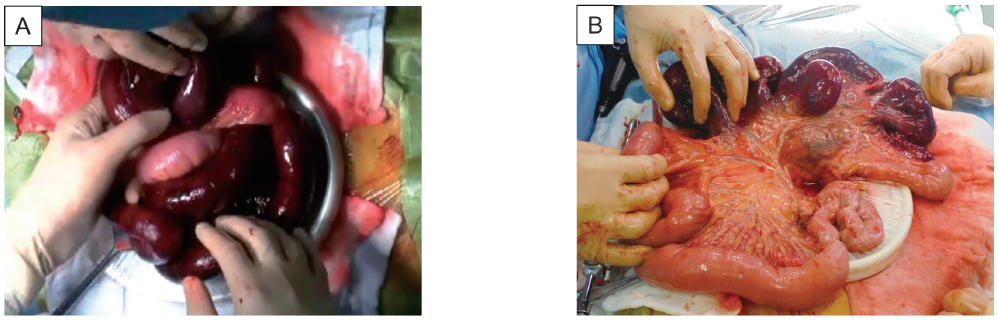

Fig. 2 Intraoperative image highlights. **(A)** Loop of the small intestine constituted a knot on the distal ileum segment showing gangrene. **(B)** The ileal knot could be untied after decompression by enterotomy.

for final confirmation of a diagnosis.[4-6] Laparoscopy showed a gangrenous small intestine and a massive amount of dark hemorrhagic fluid. Then, we converted to open surgery by making an adequate skin incision. At laparotomy, the proximal loop of ileum was knotted on the distal ileum. The gangrenous segment was about 2.5 m, extending up to 10 cm from the ileo-cecal junction (Fig. 2A). En bloc resection or untying of the gangrenous segment was very difficult. Therefore, the bowel contents were discharged by enterotomy of the gangrenous segment, which was then untwisted (Fig. 2B). Resection of the whole gangrenous segment and primary anastomosis of jejuno-ileal segment were performed. Post operatively, she was managed in the intensive care unit with intra-venous fluid, antibiotics (flomoxef sodium), and analgesia. Except for a minor wound infection, the patient made an uneventful recovery and was discharged on postoperative day 12.

DISCUSSION

Intestinal knot formation was first described by Riverius in the 16th century and later by Rokitansky in 1836. As mentioned earlier in the Introduction section, several types of intestinal knot formation are known. Among them, the ileo-sigmoid knot, termed ISK, is the most common type of intestinal knot syndrome.[7, 8] On the other hand, the ileo-ileal knot is a very rare entity. In fact, an ileo-ileal knot was found in only 1 case out of 92 cases of intestinal knot.[2] Also, our extensive search of the literature found only 7 reports (including ours) of ileo-ileal knot since 1988, as shown in Table 1.[1, 3, 9-12] Common clinical characteristics of ileo-ileal knot were not found by our literature search. On the other hand, causations of ISK are divided into anatomical and dietary habit factors. It has been reported that freely mobile small intestines and redundant sigmoid colon with a long and narrow mesentery are anatomical factors of ISK.[8, 10, 11] Also, several reports indicated that the ingestion of a single daily meal is a dietary habit factor of ISK.[8, 10, 11] Namely, when a semi-liquid bulky meal progresses into the proximal jejunum, the heavier segment of the proximal jejunum falls into the lower quadrant due to increased mobility of the intestine. The empty loops of the ileum and distal jejunum twist and rotate around the base of the narrow sigmoid colon.[8, 10, 13] In our present case, the small intestine was longer as compared with the mesentery. Also, a freely movable space of small intestine was ensured, because the patient was extremely thin, with little fat tissue in her abdominal cavity. In addition, previous adnexectomy may have contributed to making a free space for moving of her small intestine. Also, her abdominal pain occurred after lunch. Furthermore, infusion for chemotherapy may have triggered peristalsis of the intestines instead of bulky meal progression.

Preoperative diagnosis of intestinal knotting is extremely difficult. A single report showed that CT findings help in the preoperative diagnosis of ISK.[14] In most cases, intestinal knotting is diagnosed intraoperatively, as in the present case. Hence, early intervention is needed; and the most effective diagnosis and treatment for this unusual condition is surgery. At surgery, when all segments of the intestine are viable, trying to untie the knot is recommended. However, prolonged attempts to do so are unwise, because the gangrene of the bowel progresses rapidly; and this procedure has a high risk of causing perforation. When an irreversible ischemic segment is found, controlled decompression by enterotomy before untying a gangrenous knot is recommended.[11] In our present case, the ileo-ileal knot was untied smoothly by controlled enterotomy decompression. Of course, appropriate fluid resuscitation and antibiotic therapy are required.[3, 8] Depending on the length of the remaining small bowel, short bowel syndrome should be considered after surgery.[3]

Ileo-ileal intestinal knotting is a very rare entity. However, it should be often considered in the differential diagnosis of causation of a strangulated intestinal obstruction, because of the high rate of morbidity and mortality. A high index of suspicion and immediate surgical intervention

Table 1 The details of seven cases of ileo-ileal knot including ours.

Author	Year	Age	Sex	Chief complaint	Past surgical history	Knot point	Duration until operation	Operation	Outcome
Pendse	1988	11 M	N.D.	Vomiting, Constipation, Abdominal distension	–	20cm from ileo-cecal junction	48 h	En block resection of gangrene segment and anastomosis	Alive
Uday	2012	68	M	Abdominal pain, distention, Vomiting	–	15cm from ileo-cecal junction	48 h	Resection of gangrene segment after untying of knot and anastomosis	POD8, discharge
Andromanakos	2014	26	M	Abdominal pain	–	Ileum-ileum, Sigmoid-ileum	6 h	Resection of gangrene segment afetr untying of knot by enterotomy decompression and anastomosis	POD15, discharge
Kumar	2015	75	F	Abdominal pain, distention, Vomiting, Constipation	Left inguinal hernia Vaginal hysterec-tomy	Ileum-ileum	72 h	Enterotomy decompression after untying knot	Alive
Abebe	2015	55	F	Abdominal pain	–	8cm from ileo-cecal junction	48 h	En block resection of gangrene segment and anastomosis	POD14, discharge
Gopivallabh	2016	54	M	Swelling in the right groin Abdominal distention, Vomiting, Constipation	Appendicectomy	One foot proximal to the ileocecal junction	48 h	En block resection of gangrene segment and anastomosis	POD6, discharge
Our case	2016	80	F	Abdominal pain	Colostomy, Adnexectomy	10 cm from ileo-cecal junction	6 h	Resection of gangrene segment after untying of knot by enterormy decompression and anastomosis	POD12, discharge

N.D., not described.

are the most useful tools for this critical condition.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

CONFLICT OF INTEREST

The authors declare that they have no conflict interests.

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