

Case Report

A rare case of ileo-ileal knotting with strangulated intestinal obstruction

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Abstract

The ileo-ileal knot is an extremely rare surgical emergency that results in gangrene of the affected bowel segment. Only a few cases have been reported in the literature. Most of the time, it is an intraoperative diagnosis. Intestinal knot formation, specifically the ileo-ileal knot, is a comparatively uncommon aetiology of strangulated intestinal obstruction. Here we report a case of strangulated intestinal obstruction in a 45-year-old male patient secondary to ileo-ileal knotting. Emergency laparotomy, en-bloc ileal resection of 80 cm, and end-to-end ileo-ileal anastomosis were performed. On the 12th postoperative day, the patient was discharged after being treated for a superficial surgical site infection. The purpose of this case report is to remind physicians that ileo-ileal knotting should be considered in the differential diagnosis of a patient with intestinal obstruction, and that if the knot is strangulated, en-bloc resection is the primary intraoperative measure to consider in the management of these patients.

Key words: ileo-ileal knotting, strangulated intestinal obstruction, intestinal knotting, strangulated small bowel obstruction

Introduction

Riverius and Rokitansky authored the earliest reports of intestinal knotting in the 16th and 19th centuries, respectively (1–7). Various configurations of intestinal knots have been reported, including ileo-sigmoid, ileo-ileal, appendico-ileal, and ileal-Meckel's. Almost all cases of intestinal knots occur between the ileum and sigmoid colon. The most common form of intestinal knotting is ileo-sigmoid knotting (3,6,8–10). Intestinal knot formation is

characterized by tying together of two intestinal segments. The ileo-ileal knot is an infrequent entity in which a proximal portion of the ileum volvulizes or twists around the distal portion of the ileum or its mesentery, causing intestinal obstruction (1,3,6,8,10). This process eventually results in ischemia and may result in gangrene. The most common causes of intestinal obstruction are adhesions, which account for 60% of cases, followed by intestinal neoplasms,

which constitute 20% of cases, and hernias, which account for about 10% of cases. Intussusception and volvulus, each accounting for less than 5%, and intraabdominal abscesses, gallstones, and foreign bodies, all constituting less than 2%, are less common causes of intestinal obstruction (6,10). Because of the rarity of this diagnosis, data are lacking on the age and sex predilection of ileo-ileal knotting and the exact cause. The diagnosis of intestinal knotting is even rarer in western countries (4–7). The purpose of this case report is to remind physicians that ileo-ileal knotting should be considered in the differential diagnosis of a patient with intestinal obstruction, and that if the knot is strangulated, en-bloc resection is the primary intraoperative measure to consider in the management of these patients.

Case Description

A 45-year-old male from a rural health center was referred to our hospital and diagnosed with strangulated intestinal obstruction. He presented with severe, colicky abdominal pain of two days duration associated with repeated episodes of bilious emesis, abdominal distension, fever, and failure to pass feces and flatus. He had no history of previous surgery. He had an unremarkable past medical history.

Physical Examination

He appeared acutely ill with hypotension (BP = 90/50 mmHg), tachycardia (pulse rate = 120 bpm), and fever (Temperature=38.1 °C).

He had a dry tongue and buccal mucosa.

On abdominal examination, the abdomen was grossly distended with hypoactive bowel sounds, diffuse tenderness, and rebound tenderness. The percussion note was hypertympanic.

Investigation

On laboratory workup, the patient was found to have leukocytosis with neutrophil predominance

and anemia (WBC = 28,000/uL; neutrophils = 98%; Hct = 33%, platelet count = 220,000/ uL).

He had a plain abdominal x-ray taken two days prior from the referring health center, which is of poor quality. The x-ray demonstrated multiple dilated small bowel loops with air-fluid levels (Figure 1).



Figure 1: Erect plain abdominal x-ray

Treatment

Based on the above findings, he was deemed volume-depleted and resuscitated as fast as possible with 5 L of normal saline, producing adequate urine. A nasogastric tube was inserted. Antibiotic therapy (Ceftriaxone 1 g IV BID and Metronidazole 500mg IV TID) was initiated. The patient was then transferred to the operating room for an emergent intervention.

Intraoperative findings: Approximately 1 liter of dark hemorrhagic fluid was immediately encountered within the peritoneal cavity. An ileo-ileal knot was identified. The involved segment of the bowel was frankly gangrenous (Figure 2). A schematic picture of the gangrenous ileo-ileal knot is illustrated in Figure 3.

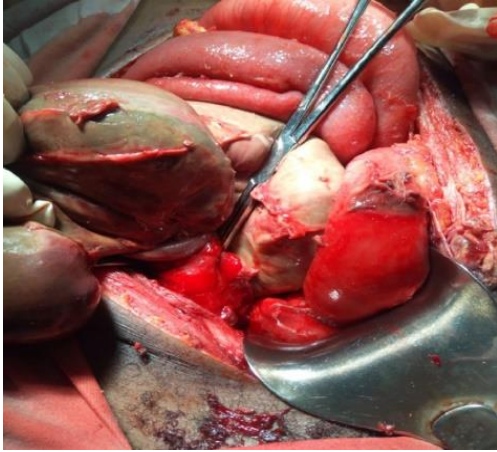


Figure 2: The intraoperative picture showing the gangrenous ileo-ileal knot

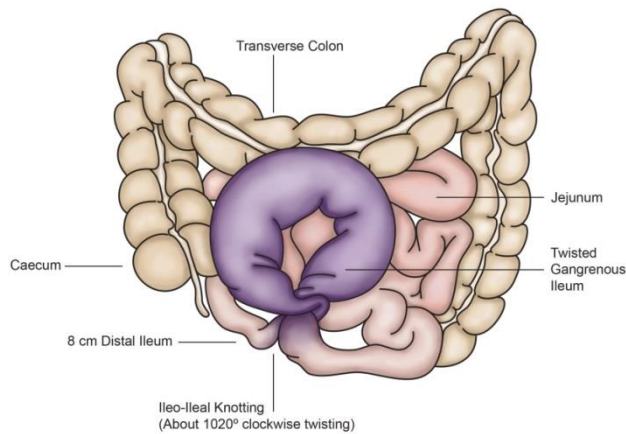


Figure 3: A schematic picture of the gangrenous ileo-ileal knot

The base of the knot was approximately 8 cm proximal to the ileocecal junction. The gangrenous ileal segment was approximately 80 cm in length.

Procedure done: Hemorrhagic fluid was suctioned, an en-bloc resection of the 80 cm of gangrenous ileum was performed, and an end-to-end ileo-ileal anastomosis was done. The patient was extubated safely and transferred to the post-anesthesia care unit. A superficial surgical site infection complicated the patient's postoperative course, which was treated successfully with local wound care over the following days. The patient

was discharged in stable condition on postoperative day 12. The patient was seen in follow-up after two weeks and was doing well.

Discussion

The ileo-ileal knot is an extremely rare surgical emergency that may result in gangrene of the affected bowel segment (3,4). In the literature, very few cases have been reported; only 12 cases have been reported since 1990, including the Japanese literature. There have only been six case reports in Ethiopia, two of which are in the pediatric age group (8–13). In general small bowel volvulus and abdominal wall hernias are the commonest causes of small bowel obstruction in the developing countries, including Ethiopia. Ileo-ileal knotting is common in regions where small bowel and sigmoid volvulus are prevalent (10,11,13). For unknown reasons, the condition is rare in western countries but common in India, Africa, Eastern Europe, and Asia (6,10,11,13,14). A diagnosis is often made intraoperatively as a preoperative diagnosis is challenging, especially in settings where radiographic modalities are limited (1–3,5,6). Even intraoperatively, the diagnosis can be missed, as many surgeons are not aware of the gross appearance of this rare condition.

The presentation of a patient with ileo-ileal knotting in terms of signs and symptoms is indistinguishable from other types of intestinal obstruction; like most cases of small bowel obstruction, it may result in rapid deterioration. The average duration of symptoms before the patient presents to the hospital usually lasts two days (6,10). The clinical course of our patient reflects this scenario. If erect plain abdominal films are taken, we see features of intestinal obstruction. Though the quality of the image was poor, this was the case with our patient. Whenever needed, one can do an abdominopelvic CT scan to pick up other associated pathologies like intussusception (6). Treatment should be initiated expeditiously with

aggressive IV fluid resuscitation, nasogastric tube insertion, and broad-spectrum IV antibiotics. Once the patient is adequately resuscitated, an emergency laparotomy should be prioritized (3). In most cases, the knots are found in the terminal ileum.(6) This was the case with our patient. If the bowel appears viable intraoperatively, detorsion or untying the knot may be sufficient, and the bowel should be reassessed for signs of ischemia or injury. When the knot is found to be gangrenous, an en-bloc resection of the gangrenous segment and an ileal anastomosis are recommended. In cases of gangrenous ileo-ileal knotting, controlled enterotomy for decompression can be done, followed by en-bloc resection (4,6,12,13). No attempt should be made to untie a gangrenous segment as it risks bowel perforation and peritoneal contamination. If the distal segment is less than 10 cm from the ileocecal valve, then an ileocolic anastomosis should be performed, which is an end-to-side or ileo-ascending anastomosis (1,2,4,6,11). In our case, we did an en-bloc resection of the gangrenous segment. Despite the remaining length of the ileum being less than 10 cm, we wanted to preserve the ileocecal valve, so we did an end-to-end ileo-ileal anastomosis. If a patient is hemodynamically unstable, an ileostomy is another possibility.

Postoperatively, the patient should be assessed for hydration status, anemia, and signs of an anastomotic leak if an anastomosis is performed (3,11). Depending on the length of the remaining small bowel, the clinician should consider and monitor for short bowel syndrome. If a large segment of small bowel is resected or the ileocecal valve is bypassed or resected, there is a high chance of developing a short bowel syndrome. Whenever possible, it is good to preserve the ileocecal valve. Once a short bowel syndrome develops, dietary modification should be the first measure to be taken (1,11). During the patient's two-week postoperative follow-up, there were no signs of short bowel syndrome.

Overall, ileo-ileal knotting is a potentially lethal diagnosis that merits a high index of suspicion, with mortality rates approaching 50% in the reported literature. According to some studies, mortality can range from 8% to 25% (3,9,11). Ileo-ileal knotting resulting in intestinal obstruction has been recorded infrequently in the medical literature. To date, six cases of ileo-ileal knotting have been reported in Ethiopia; ours will be the seventh. This will greatly contribute to raising awareness of this rare case.

Conclusion

Ileo-ileal knotting is a rare cause of small bowel obstruction. In a patient with intestinal obstruction, it should be considered in the differential diagnoses. Prompt diagnosis and intervention are critical because bowel strangulation can occur quickly. In the management of patients with strangulated ileo-ileal knotting, en-bloc resection is a primary intraoperative measure that should be considered.

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Consent for publication

The patient consented to the publication of this case report.

Data Availability statement

Not applicable.

Conflict of interest

The authors declare that they have no conflicts of interest.

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Authors' Contribution

ABA is the P.I. who participated in the entire process, from the report's inception to its final draft. Moreover, he participated in the operation of the case. AYK reviewed the initial draft of the case report, took intraoperative photographs, and participated in the operation.

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