

Case Report

A Rare Case of Gangrenous Ileosigmoid Knotting Explored as a Case of Ruptured Hemorrhagic Ovarian Cyst in a Young Woman

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Abstract

Introduction: The wrapping of the ileum around the sigmoid colon and its mesentery, or *vice versa*, is known as Ileosigmoid Knotting (ISK). Although the frequency of ISK is unknown, adult males are more likely to experience it, and it typically manifests in regions with a high sigmoid volvulus incidence. There is debate concerning the cause of ISK. Abdominal distention and discomfort are the primary indicators, whereas vomiting, pain, and distention are the main symptoms.

Case presentation: a 25 year of female patient presented to our hospital with 4 days history of abdominal distention and pain with associated failure to pass feces and flatus. She was explored by gynecology side with the impression of ruptured hemorrhagic cyst of the ovary and surgical side took over after intraoperative consultation with the diagnosis of gangrenous ISK.

Discussion: Theoretically, ISK has been linked to anatomic anomalies such as hypermobile small bowel, enlarged small intestine mesentery, and redundant sigmoid colon with a narrow mesenteric pedicle. Meckel diverticulitis with band, ileocecal intussusceptions, floating cecum, and trans-mesenteric herniation are risk factors for ISK in addition to these morphological abnormalities. Due to the nonspecific clinical pictures and lack of gold standard diagnostic preoperative imaging high index of clinical suspicion and urgent surgery are corner stones in the management of such patients.

Conclusion: High index of suspicion as well as early diagnosis and intervention are important for better outcome.

Keywords: Ileosigmoid knotting; Acute Abdomen; Women of reproductive age; Ovarian cyst

Introduction

The wrapping of the ileum around the sigmoid colon and its mesentery, or *vice versa*, is known as Ileosigmoid Knotting (ISK) [1]. Although the frequency of ISK is unknown, adult males are more likely to experience it, and it typically manifests in regions with a high sigmoid volvulus incidence. There is debate concerning the cause of ISK. Abdominal distention and discomfort are the primary indicators, whereas vomiting, pain, and distention are the main symptoms [2-5]. To diagnose ISK, there are no particular blood tests available. Multiple minor intestinal air-fluid levels and a dilated sigmoid colon are visible on plain abdominal X-ray images. An abdominal CT scan shows twisted and dilated small intestine segments along with a whirling sigmoid mesentery and a twisted and dilated sigmoid colon even though it is challenging to accurately diagnose ISK prior to surgery [1,5].

Case Presentation

A 25-year-old female patient presented to our gynecologic

emergency room with a vague abdominal pain of 4 days duration. She has associated 1 episode of vomiting of ingested matter and associated failure to pass feces and flatus. She has a regular menses that comes every 28 days, and her last menses was 2 weeks back. The other part of the history was unremarkable.

On physical examination she was acutely sick looking and in pain. Her vital signs were pulse rate 112-120 beats per minute, blood pressure of 100 mmhg/60 mmhg. She was not febrile, and her oxygen saturation and respiratory rates were in the normal range. On abdominal examination she has a slightly distended abdomen that moves with respiration and tenderness all over the abdomen more on the pelvis area. Digital rectal examination was not done but on per vaginal examination done by the gynecology side she had cervical motion tenderness.

The patient brought an ultrasound examination result that was done in a private hospital which suggests "left adnexal complex predominantly cystic lesion with free thick hypoechoic intraperitoneal fluid collection suggestive of ovarian hemorrhagic cyst with rupture". The repeated ultrasound in our hospital suggests a consistent report. On Complete Blood Count (CBC): she had leukocytosis of 16,000 with neutrophilia of 85% and the other CBC parameters were in the normal range.

She was taken to the Operating Room (OR) by the gynecology side with the diagnosis of acute abdomen secondary to ruptured left ovarian hemorrhagic cyst. The abdomen was approached through pfannenstiel incision and intraoperative finding was around 300 ml of hemorrhagic fluid in the general peritoneum with gangrenous small and large bowel loops (Figure 1). Surgical side was consulted intraoperatively and on further exploration the patient had a gangrenous

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ileosigmoid knotting with the wrapping of the distal ileum around the sigmoid 360-degree counterclockwise and a gangrenous ileum and sigmoid colon (Figures 2 and 3). For this we extended the incision vertically (inverted T) and did an en block resection of the volvulated segment and reconstituted bowel continuity with end-to-end ileo-ileal anastomosis, with a 5 cm of distal ileal remnant close to the ileocecal valve.

We exteriorized the distal descending colon as end colostomy as it was distended and edematous. We lavage the abdomen thoroughly and closed the abdominal wall in layer. Post operatively the patient had an uneventful recovery and she was discharged on 5th post-operative day with appointment for reversal of the stoma. She was seen in an outpatient clinic twice and she is having a smooth recovery and waiting for reversal of the colostomy.



Figure 1: A non-ruptured simple left ovarian cyst.



Figure 2: Gangrenous small and large bowel loops found upon laparotomy.

Discussion

Parker reported the first instance of Ileosigmoid Knotting (ISK) in 1845. More than 280 instances have been documented globally since that time. The sigmoid colon and its mesentery are encircled by an ileum loop in ISK, an uncommon cause of intestinal blockage. This condition can lead to gangrene of the ascending colon, caecum,

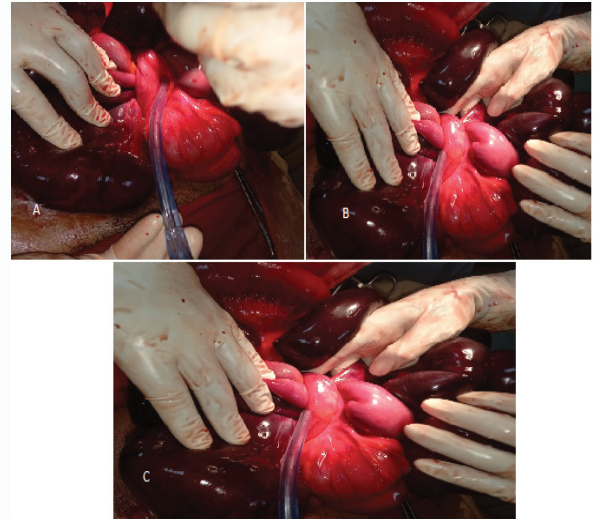


Figure 3: A, B and C) A gangrenous ileosigmoid knotting with the wrapping of the distal ileum around the sigmoid 360-degree counterclockwise.

and even the sigmoid colon. Although the exact prevalence of ISK is unknown, some Asian, Middle Eastern, and African ethnicities seem to be more affected than others. Eighty percent of ISK patients are men, and the average age at presentation is forty years old (with age variations from four to ninety-nine years old). The precise etiology, however, remains unknown [3,5].

Theoretically, ISK has been linked to anatomic anomalies such as hypermobile small bowel, enlarged small intestine mesentery, and redundant sigmoid colon with a narrow mesenteric pedicle. Meckel diverticulitis with band, ileocecal intussusceptions, floating cecum, and trans-mesenteric herniation are risk factors for ISK in addition to these morphological abnormalities. A review of the literature showed that eating a large meal while having an empty small bowel can cause ISK, which explains why Muslims are more likely to get ISK during Ramadan. After eating a large meal, moving down the meal increases small bowel peristalsis, which causes the jejunum's heavier sections to fall to the left lower quadrant. The jejunum's and ileum's empty sections then wrap around the sigmoid's narrower base. This patient had an enlarged small bowel mesentery and a redundant sigmoid with a constricted mesenteric pedicle intraoperatively [1,2,4,6].

For ISK, there are two classification systems. Four forms of ISK have been found in the first classification: type I involves the ileum twisting around the sigmoid colon, type II involves the sigmoid colon twisting around the ileum, and type III involves the ileo-cecal segment twisting around the sigmoid colon. It is impossible to identify the segment that is twisted in type IV, often known as uncertain type. The patient we saw had ISK type II, as it appears the ileum have twisted around the sigmoid [5,7].

In the second classification of ISK, patients are classified in 6 classes as follows: in class I, there are no other risk factors for mortality (such as associated disease, advanced age, etc.); in class II, risk factors of mortality mentioned above are present; but the patient is not in shock and bowel gangrene is absent; in class III, the patient is in shock; in class IV, gangrene of the ileum or sigmoid colon is present; in class V, patient has shock as well as gangrene of the ileum or sigmoid colon and in class VI the gangrene of both ileum and sigmoid colon is present. Since our patient had both large bowel (Sigmoid colon) and

small bowel gangrene, based on this classification, she is categorized in class VI [7].

Despite the severe nature of the disease pre-operative diagnosis is challenging due to the non-specific presentation. This problem becomes more prominent in cases of a reproductive age woman with long list of differential diagnosis for an acute abdomen. Even with advanced imaging modalities available today only a small proportion of patients are suspected to have ISK preoperatively due to lack of a specific imaging features. Hence, a high index of clinical suspicion is of paramount importance in preoperative consideration of this entity [3,5,8].

Prolonged attempts at untying the knot are unwise especially in the presence of a gangrenous bowel as the risk of septic shock and sudden deterioration from liberation of toxin into the blood stream is high. Another challenge in the management of such cases is, after resection usually there will be a short segment of terminal ileum left close to the Ileocecal Valve (ICV) and previous literatures recommend against end-to-end anastomosis rather to close the stump and do an end to side ileocectomy. But with significant amount of small bowel resected in most of such cases, like ours, bypassing the ICV might predispose the patient for short bowel syndrome, and numerous recent reports confirm the safety of end-to-end ileo-ileal anastomosis closer to the ICV, and we were successful at our anastomosis [3,5,6,8].

As to the colon Primary anastomosis has been recommended in 3rd world situations where patient compliance with follow-up is poor. If the duration of illness has been short, and the colon is not distended proximal to the obstruction, a primary anastomosis is feasible. But in our case due to the edematous distended colon, we prefer to do end colostomy and plan future reversal.

Conclusion

In summary, the ileosigmoid knot, also known as compound volvulus, is an uncommon ailment that quickly results in gangrene of the sigmoid colon and a significant section of the ileum. Early laparotomy is required due to the clinical picture of severe abdominal discomfort that is out of proportion to the physical findings. To prevent irreversible endotoxemia, the knot should be cut and removed if the small bowel is gangrenous. A primary colonic anastomosis can be performed successfully if the patient has a brief medical history and the residual colon is clean, well-vascularized, and not significantly. The small bowel ends almost always can safely be primary anastomosed and its crucial not to bypass the ICV as much as possible to decrease the risk of SBS.

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