



Case report

Ileo-sigmoid knotting- an unusual cause of intestinal obstruction: A case report

Ehsanollah Rahimi-Movaghar^a, Tahmineh Tahouri^{b,*}^a Department of Surgery, Farhikhtegan Hospital, Faculty of Medicine, Tehran Medical Science, Islamic Azad University, Tehran, Iran^b Shahid Modarres Educational Hospital, Shahid Beheshti University of Medical Science, Faculty of Medicine, Tehran, Iran

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ABSTRACT

Introduction and importance: Ileosigmoid knotting is an unusual cause of intestinal obstruction in which the ileum wraps around the base of the sigmoid colon and its mesentery, which leads to a closed loop intestinal obstruction. **Presentation of case:** A 59-year-old male patient was referred to our center with acute abdominal pain, obstipation and a few episodes of vomiting. On physical examination, he had hypotension and tachycardia as well as distension, diffused tenderness, guarding and rebound tenderness of the abdomen. Bowel sounds were absent. Abdominal Plain X-ray showed dilatation of the large bowel associated with the distended small bowel. After resuscitation with aggressive intravenous fluid therapy, the patient underwent an emergency laparotomy. Exploration revealed gangrene of the intestinal loops, including the jejunum and ileum, which was secondary to a 360° clockwise twisting of the ileal loops around the sigmoid colon. Gangrene of the sigmoid colon along with torsion of the superior mesenteric arteries was also obvious. Resection of the gangrenous loops of the small bowel as well as a sigmoidopexy was carried out and a primary anastomosis of the small intestine and colon was performed. He did well post-operatively.

Clinical discussion: It is crucial to distinguish Ileosigmoid knotting from sigmoid volvulus because it can rapidly, even within hours, progress to bowel gangrene and peritonitis; moreover, in this situation the endoscopic reduction is contraindicated.

Conclusion: Ileosigmoid knotting is an unusual cause of bowel obstruction which has relatively high mortality rates and should be considered in patients with acute abdominal pain and presentations of bowel obstruction.

1. Introduction

Ileosigmoid knotting is an unusual cause of intestinal obstruction in which the ileum wraps around the base of the sigmoid colon and its mesentery. This situation, also known as compound volvulus, leads to a closed loop intestinal obstruction. It may briskly progress to gangrene in both the ileum and the sigmoid colon within hours; thereupon, peritonitis and resultant sepsis with high mortality can occur [1,2].

Theoretically, anatomical factors such as redundant sigmoid colon with a narrow mesenteric pedicle and a long small bowel mesentery and mobile and free small bowel can be responsible for the development of the Ileosigmoid knotting; however, the main etiology is not well-known [3].

Ileosigmoid knotting is commonly seen in the areas of the world in which the incidence of sigmoid volvulus is high, so it is relatively

unusual in the west and most of the reported cases are from Asian and African nations. Most patients with Ileosigmoid knotting are males in their third to fifth decades [4,5].

Diagnosis of Ileosigmoid knotting can be difficult. CT scan is the diagnostic modality of choice in which multiple air-fluid levels in the small intestine as well as dilated sigmoid colon and “whirl sign” can be demonstrated. Anyway, once the diagnosis is made, adequate resuscitation and surgical intervention are necessary. The surgical approach is mainly focuses on untwining of the knot, resection of the gangrenous segments of the sigmoid colon and the small intestine and re-establishment of small bowel continuity. Besides, Stoma can be required in the elderly as well as in patients with comorbidities, unstable hemodynamics and peritonitis [6,7].

It is crucial to distinguish Ileosigmoid knotting from sigmoid volvulus because it can rapidly, even within hours, progress to bowel

* Corresponding author at: Shahid Modarres Educational Hospital, Shahid Beheshti University of Medical Science, Faculty of Medicine, Yadegar Imam Highway, Saadat-Abad Intersection, 1998734383 Tehran, Iran.

E-mail address: tahmineh.tahouri@hotmail.com (T. Tahouri).

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gangrene and peritonitis; moreover, in this situation the endoscopic reduction is contraindicated [8].

2. Case presentation

The case described was a 59-year-old male patient, previously healthy, brought to the emergency department of Firuzabadi General Hospital, an academic hospital in Tehran, Iran, with diffuse abdominal pain as well as obstipation of 1 day duration. He also had a few episodes of vomiting. The patient was otherwise healthy and did not take any medication.

He had no past psychological history and family history was also unremarkable. On physical examination, the patient was conscious and oriented. His vitals were a pulse rate of 120 beats per minute regular, BP of 80/60 mmHg, temperature of 37.7°C and a respiratory rate of 21 breaths per minute. Abdominal examination revealed abdominal distension, diffused tenderness, guarding and rebound tenderness. Bowel sounds were absent. Laboratory investigations demonstrated a white blood cell count of 12,400/mm³ and arterial blood gas analysis showed features of metabolic acidosis with pH of 7.15 and HCO₃ level of 12 meq/L.

Abdominal Plain X-ray showed dilatation of the large bowel associated with the distended small bowel. There was neither air-fluid level nor free air under the right hemidiaphragm (Fig. 1). Since the patient had features of peritonitis and hypovolemic shock, computed tomography scan could not be performed. Resuscitation with aggressive intravenous fluid therapy was immediately started and broad-spectrum antibiotics were also prescribed and afterward the patient underwent an emergency laparotomy by an experienced general surgeon. Exploration revealed gangrene of the intestinal loops, including the jejunum and ileum which was secondary to a 360° clockwise twisting of the ileal loops around the sigmoid colon (Fig. 2). The sigmoid colon was viable and torsion of the superior mesenteric arteries was also obvious. Resection of the gangrenous loops of the small bowel (about 200 cm in size) as well as a sigmoidopexy was carried out and a primary anastomosis of the small intestine and colon was performed. Postoperatively, except for an ileus, the patient made an uneventful recovery and he was discharged after 9 days in a good general condition. In 3 years follow-up, the patient is in his usual state of health and free of symptoms.

3. Discussion

The first case of Ileosigmoid knotting (ISK) was described by Parker in 1845. Since then, over 280 cases have been reported worldwide [8]. ISK is a rare cause of intestinal obstruction in which a loop of ileum, wraps around the sigmoid colon and its mesentery. In this situation, gangrene of the ileum, sigmoid colon, and even caecum and ascending

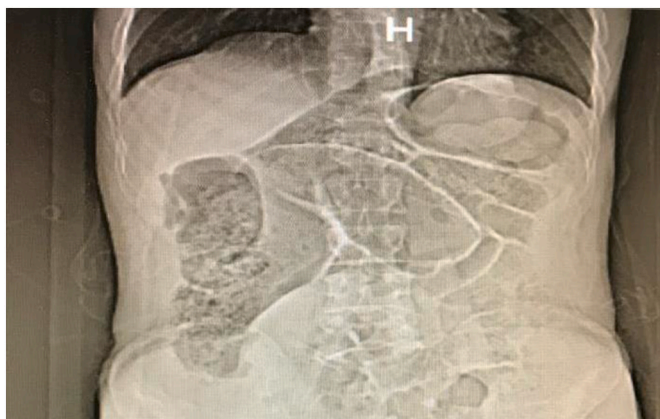


Fig. 1. Abdominal Plain X-ray showed dilatation of the large bowel associated with distended small bowel.



Fig. 2. Intraoperative findings. Intestinal loops including jejunum and ileum which was secondary to a 360° clockwise twisting of the ileal loops around the sigmoid colon. Gangrene of the sigmoid colon along with torsion of the superior mesenteric arteries was also obvious.

colon can occur [3]. The incidence of ISK is not known [1] but it is more common in certain Asian, Middle East and African nationalities. About 80% of ISK patients are male and the mean age of presentation is about 40 years (ranges between 4 and 90 years old) [9].

However, the exact etiology is unknown. Anatomical abnormalities such as redundant sigmoid colon with a narrow mesenteric pedicle, elongated small intestine mesentery, hypermobile small bowel, have been theoretically responsible for the occurrence of ISK [1]. Other than these anatomical factors, Meckel diverticulitis with band, ileocecal intussusceptions, floating cecum and trans-mesenteric herniation are risk factors for ISK [2,6]. Our patient was a Muslim male who presented with ISK during Ramadan. Reviewing the literature revealed that ingestion of a heavy meal in presence of an empty small bowel may lead to ISK, so that there is a higher incidence of ISK in Muslims in Ramadan. Moving down a heavy meal increases the small bowel peristalsis which leads to falling of the heavier parts of jejunum to the left lower quadrant and the empty parts of ileum and jejunum wrap around the narrowed based sigmoid [6]. Moreover, Intra-operatively he had a redundant sigmoid with narrowed mesenteric pedicle as well as an elongated small bowel mesentery.

There are two classifications for ISK. In the first classification, 4 types for ISK have been identified: in type I, the ileum twists around the sigmoid colon; in type II the sigmoid colon twists around the ileum and in type III, the ileo-cecal segment twist around the sigmoid colon. In type IV (or indeterminate type), it is not possible to determine which segment is twisted. Our patient had type II of ISK. 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 shock and small bowel gangrene, based on this classification, he had been categorized in class V [10].

Due to confusing clinical and radiographic presentations of ISK, in most cases, making a precise diagnosis is not easy. The Rate of pre-operative accurate diagnosis of this unfamiliar entity ranges between 0 and 25 % in literature [3]. Clinical signs of small intestine obstruction such as vomiting are present while the plain X-ray of the abdomen often shows distension of the colon, which is often mistaken for simple sigmoid volvulus [9]; accordingly, a diagnostic triad of ISK has been defined which includes the following: clinical presentations of small intestine obstruction, radiographic presentations of colon obstruction and inability to insert a sigmoidoscope [3].

Various surgical techniques have been reported in the literature for the management of ISK. Indeed, choosing a surgical approach in ISK patients mainly depends on hemodynamic stability of the patient as well as the pathologic and anatomic intraoperative findings. So, in patients with unstable hemodynamics (either pre-operatively or intra-operatively), damage control laparotomy is the best strategy and a second looked laparotomy should be done within 24–48 h later. In contrast, in patients with stable hemodynamics, a definitive surgical procedure which depends on intraoperative anatomical and pathological findings can be done at the initial laparoscopy. In most (about 75 %) ISK cases, the small bowel is gangrenous and in about 53 % to 60 % of patients, both the sigmoid colon and small bowel are gangrenous, whereas in 20–25 % of patients both the small and large bowels are found viable in surgery. Our patient had an unstable hemodynamic pre-operatively and a viable sigmoid colon with a gangrenous sigmoid colon intra-operatively. In the current case, successful resection of the gangrenous loops of the small bowel as well as a sigmoidopexy was carried out and a primary anastomosis of the small intestine and colon was performed. Although the preferred definitive procedure in ISK patients with a viable sigmoid colon is sigmoid resection [10], this procedure has not been done for the patient due to his relatively hemodynamics. Anyway, in 3-years follow-up, the patient remains asymptomatic.

Since most patients with ISK present with frank obstruction, the mortality rate of ISK is relatively high and estimated about 48 % [1,2]. Keys for appropriate management are Prompt intravenous fluid resuscitation, correction of the acid-base and electrolyte imbalance, empiric antibiotic therapy (with aminoglycosides and cephalosporins) and immediate surgical exploration [1,2].

In overall, the prognosis of ISK is relatively poor with a mortality rate of up to 48 %. Toxic shock is the most frequent cause of death. Other common complications are including wound dehiscence or infection, stoma problems or leak of anastomosis. Fortunately, the recurrence of ISK is very rare and only one case has been reported to date in literature [11].

4. Conclusion

Ileosegmoid knotting is an unusual cause of bowel obstruction which has relatively high mortality rates and should be considered in patients with acute abdominal pain and presentations of bowel obstruction as a differential diagnosis.

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Ethical approval

Ethic committee of Islamic Azad University Tehran Medical Science approved this study. This case report is exempt from ethical approval in our institution.

Consent

The patient gave written consent about her willingness to participate in the study after she was completely informed about study. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

Not applicable.

Guarantor

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CRediT authorship contribution statement

Ehsanollah Rahimi Movaghar, M. D.: Conception and Design of the study, writing the paper, Critical review, Supervision.

Tahmineh Tahouri, M.D.: Data collection, writing the paper.

Declaration of competing interest

The authors declare that they have no conflict of interests.

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