CASE REPORT

The Ileosigmoid Knot: A Case Report

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Summary
Ileosigmoid knotting (ISK) is an uncommon cause of acute intestinal obstruction where the ileum wraps around the base of the sigmoid colon. Seen with higher frequency in Africa, the condition rapidly progresses to gangrene of the ileum and sigmoid colon. Preoperative diagnosis is not easy partly due to the unfamiliarity of this entity and the confusing and contradictory features of the disease. Awareness of the condition is essential for prompt diagnosis and optimal management.

Key Words: Ileosigmoid knotting, Intestinal obstruction

Introduction
Ileosigmoid knotting (ISK), also known as a compound volvulus or double volvulus, is a rare but life threatening cause of closed loop obstruction (1). It is reportedly more common in many parts of Africa. In this condition the ileum wraps around the base of the sigmoid colon and forms a knot on itself resulting in vascular compromise to one or both loops (2). Mortality rates of gangrenous ISK vary from 20% - 100% (3). Despite the seriousness of this condition, preoperative diagnosis is difficult because the clinical signs are difficult to define and the radiological signs are inconstant (4). We describe a case of ileosigmoid knotting in a 40 year old male and review relevant literature.

Case Report
A 40-year-old male psychotic patient was admitted with severe acute onset lower abdominal pain for one day. The pain was worsening and was associated with progressive abdominal distension, constipation and vomiting. On examination he was in distress, blood pressure of 114/94 mm Hg, pulse rate of 83/min, and temperature of 39.5° C. Abdomen was distended, tense and had generalized tenderness, bowels sounds were absent. Rectal examination was negative for stool, blood or mass. Laboratory data revealed a leucocytosis of 17000/mm³, hemoglobin level of 11 gm/dl. Plain erect abdominal radiograph showed dilated loops of bowel with gas and air fluid levels (Figure 1). The patient was started on intravenous fluids and antibiotics and optimized for explorative laparatomy. At laparatomy, serous fluid was encountered and a loop of ileum was found encircling a dilated sigmoid colon (Figure 2). There was gangrene of terminal ileum (involving approximately 20 cm of the terminal ileum) 15cm from the ileocecal junction. The sigmoid colon was viable and the knot was undone by traction of the sigmoid loop. Following resection of gangrenous ileum, a primary anastomosis was carried out. Postoperatively the patient developed some episodes of vomiting on the first post operative day, was managed conservatively and eventually recovered and was discharged after 8 days.

Figure 1: Erect abdominal radiograph showing dilated loops of bowel filled with gas in the left upper, left mid and right upper regions of the abdomen. An air fluid level is seen at the left mid abdomen.
Figure 2: Intra-operative photograph showing a distended loop of sigmoid colon and a gangrenous ileal loop wrapped around its base.

Discussion
Ileosigmoid knotting, the name popularized by Shepherd (2), was first described by Parker in 1845 (5). It is comparatively common in Asia, Africa and the Middle East (3, 6) in areas where sigmoid volvulus is common. (7). ISK is common in adult males (80.2%) with a mean age of 40 years (range 4-90 years) (1). The factors thought to be responsible for ISK are a long small bowel mesentery and freely mobile small bowel; a long sigmoid colon on a narrow pedicle; and finally the ingestion of a high bulk diet in the presence of an empty small bowel (2, 7, 8). These features predispose to a freely movable active ileum, which can wind itself around the pedicle of an elongated passive sigmoid. Other secondary causative factors include late pregnancy, trans-mesenteric herniation, Meckel’s diverticulitis with a band, ileocecal intussusceptions, and floating cecum. (1)

ISK has been classified into four types (I, II, III, Undetermined) by Alver et al (9). Type I is the most common and occurs when the ileum (active component) revolves around the sigmoid colon. In Type II the sigmoid colon (active component) revolves around the ileum, Type III the ileocaecal portion revolves around the sigmoid colon and Undetermined is when it is difficult to decipher which is the active or the passive component. The case we described was Type I.

The predominant symptoms and signs of ISK include abdominal pain and tenderness (100%), abdominal distension (94% to 100%), nausea and vomiting (87% to 100%), rebound tenderness (69%) and shock (0% to 60%) (1). There are no specific laboratory findings of ileosigmoid knotting. The laboratory reports may show a drop in hemoglobin, leucocytosis (as in the present case), raised blood urea nitrogen and electrolyte imbalance suggestive of peritonitis (1).

Preoperative diagnosis is difficult and often infrequently made (0-28%) (1). This has been attributed to the unfamiliarity of this disease and the confusing and self contradictory features of the disease. Clinical features suggest small bowel obstruction, while radiographic findings are that of colonic distension, which is uncommon in small bowel obstruction. Plain abdominal radiographs may show the characteristic double closed-loop obstruction, with the sigmoid colon in the right upper quadrant and the small bowel loops in the left but this is only an occasional finding (9). More often, the picture is that of either simple sigmoid volvulus or small bowel obstruction. In the present case the radiographic findings were suggestive of small intestinal obstruction. Lee et al (11) described the characteristic computed tomography (CT) findings of ISK to include: the whirl sign, which is created by the twisted intestine and mesentery, which is more marked on contiguous slices than the sigmoid volvulus and the medial deviation of the cecum and descending colon with a beak appearance on its medial border. CT scan was not available in the present case. Its availability could be useful in making the diagnosis preoperatively. In the present case the diagnosis was made at laparatomy.

Prompt relief of obstruction by an emergency laparatomy is a life saving procedure. The anatomical and pathological changes of the involved loops of bowel dictate the surgical procedure (1, 3). In 73.5-79.4% of the cases, gangrenous bowel is encountered (1).

When both loops of intestine are viable, untying alone has been considered to be inadequate because of recurrence of the volvulus (8). However, some case studies have reported that the recurrence of volvulus is uncommon (2,7). In our case the sigmoid colon was not resected. Our decision was based on the findings of Shepherd (2), who reported no recurrences in patients in whom the sigmoid colon was not resected. Untwisting the knot is not recommended when both loops of bowel are gangrenous because it is difficult, time consuming and can cause accidental damage to the bowel leading to perforation, fecal peritonitis and eventually septic shock (2,7,10). En bloc resection of the knot and double primary anastomosis of both the ileal and the sigmoid ends is preferred (2,7,8,10).

Primary anastomosis is preferable, however, if the terminal ileum is gangrenous to within 10 cm to the ileocaecal valve, an end to end anastomosis should not be attempted (2, 9); the distal stump must be
closed and an end to side ileocaecostomy anastomosis performed or an end to-side enterotransverse colostomy leaving the caecum in situ (2,10). If the ischaemic process extends to the caecum and the ascending colon, a right hemicolectomy must be performed (1).

Recent data suggest that primary colonic anastomosis may be undertaken. A tension free anastomosis requires adequate mobilization of the rectum and descending colon. The Hartmann’s procedure is considered suitable, when the viability of the distal loop is doubtful or when the viable distal loop is very short to be exteriorized without exerting undue tension (2,10). The risks of primary anastomosis with unprepared bowel must be balanced against the need for a second operation with the two-stage procedure (3).

ISK has a grave prognosis with a mean mortality rate of 6.8-8% in non gangrenous and 20-100% in gangrenous cases. The average mortality rate has reduced from 0-73.6% before 1990 to 0-47% after 1990 (3). The morbidity rate is also high. The most common cause of death is shock. The presence of advanced age, associated medical problems, shock, bowel gangrene or perforation increases the mortality rate (9).

**Conclusion**

Ileosigmoid noting is a rare cause of acute intestinal obstruction with poor prognosis. Surgeons should be aware of this condition and select an appropriate surgical option based on individual requirements to optimize the survival of these patients.

**References**