

CASE REPORT

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Ileosigmoid knotting: A rare cause of acute intestinal obstruction and peritonitis

Addis Y Kidane

ABSTRACT

Introduction: Ileosigmoid knotting (ISK) is one of the rarer causes of bowel obstruction compared to others. This is also called compound volvulus or double loop obstruction. The presentation symptoms are similar to those of other large bowel or distal intestinal obstructions. But the pathogeny's or disease's progression is rapid. So the patient develops ischemia or strangulation of the involved segment of the bowel earlier than the sigmoid or cecal volvulus. One of the challenges of the disease is that, especially in low-income countries like ours, there is no access to contrast computed tomography (CT), which is more sensitive diagnostic imaging.

Case Report: A 58-year-old male patient presented to our hospital with abdominal pain for two days. He was referred from the primary hospital. The pain was initially central and later diffused to other parts of the abdomen. Associated with this, he was vomiting of ingested matter for more than two episodes. On physical examinations, he was in acute pain, and his vital signs showed a pulse rate of tachycardia at 127 BPM. On abdominal examination, he had distention and diffused tenderness with guarding. So we diagnosed him with peritonitis secondary to gangrenous sigmoid volvulus obstruction (SVO). So I chose the imaging that is available in our hospital, a plain abdominal X-ray. So the patient was resuscitated with normal saline, an antibiotic was initiated, and he was taken to the operation after giving consent. The ileum and sigmoid colon were gangrenous and en bloc resection was done, and the jejunum and remaining 7 cm of ileum from the ileocecal valve were

anastomosed. The patient was taken to or for a relook, and the anastomosis was disrupted, then refreshed, and both the proximal and distal segments were taken out as stomas. The output was significant and later developed a controlled enterocutaneous fistula, which was closed after three weeks.

Conclusion: The prognosis is poor for ISK compared to other common types of volvulus and worse prognosis challenging management in conditions with both active and passive bowel ischemia which happened to our patients. Diagnosis is also challenging before the surgery. The abdominal X-ray and ultrasound are available and commonly the surgeon commonly will not send for this unless the patient is stable. Blind rectal tube deflation the common intervention in our setup with no access for sigmoidoscopy is not indicated for this non-surgical initial management rather with a highly suspicious diagnosis early resuscitation with early surgical treatment.

Keywords: Compound volvulus, Ileosigmoid knotting, Intestinal obstruction, Peritonitis

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INTRODUCTION

Very uncommon conditions include ileosigmoid knotting (ISK), the ileum turning off or vice versa, and the sigmoid colon surrounding the other segment. Intestinal blockage may result from ISK, sometimes referred to as compound volvulus or double volvulus, a rare surgical emergency. It can spread quickly to the ileum and sigmoid colon, becoming gangrenous. Parker was the first to document this phenomenon in print in 1845 [1].

It is crucial to distinguish between ISK and simple sigmoid volvulus (SV) because ISK is contraindicated for endoscopic decompression [2]. A severe condition's morbidity will increase if a significant portion of the colon is removed. There is also a chance of short bowel syndrome (SoBS), which is characterized by people having fewer than 200 cm of residual small bowel. A broadly applicable functional definition posits that inadequate intestinal absorptive capacity leads to clinical signs of diarrhea, dehydration, and malnutrition [3].

A 58-year-old man was treated in our emergency department for a gangrenous ileosigmoid knot. Sharing this example will help us better understand the illness, which will enable an earlier diagnosis and the avoidance of death.

CASE REPORT

This 58-year-old male patient initially complained of acute, continuous, colicky stomach pain that persisted for two days without any aggravating or alleviating causes or radiation. Along with this, he also experienced flatus, growing abdominal distension, inability to pass feces, two to three episodes of vomiting the ingested material, and a subjective, short-lived, high-grade intermittent temperature. He had been experiencing constipation and sporadic cramping in his abdomen for three months, which prevented him from doing his farming duties. He attended a traditional healer on multiple occasions for this compliance, receiving traditional medication and stomach massages. However, he did not see any progress. After being admitted to Sheno Primary Hospital in Oromia, Ethiopia, the patient received a referral, an intravenous (IV) line was set up, a catheter was placed, and an unidentified drug was provided.

Additionally, the patient disclosed no past history of surgery or traumatic trauma. Upon physical examination, the patient showed signs of consciousness, irritability, and disorientation. Vital signs included a 127 bpm pulse, 125/75 mmHg blood pressure, 37.8 °C temperature, and 94% saturation. A chest check revealed nothing unusual. There were symptoms of peritonitis and a tight, enlarged abdomen in the patient. The results of laboratory tests showed that the platelet count was 312×10^9 , the neutrophil count was 93%, and the white blood count was 42×10^9 (Table 1). Since there was an obvious bowel blockage in the patient, an erect abdominal X-ray was not performed. Broad-spectrum antibiotics and vigorous intravenous fluid (two bags of normal saline) were used to revive the patient (ceftriaxone 1 mg IV and metronidazole 500 mg IV). Once the patient's condition was relatively stabilized (blood pressure of 130/85 mmHg and pulse of 120 bpm), we moved them to the operating room. After performing a damage control laparotomy (exploratory laparotomy), it was discovered that the distal ileum was knotted around the sigmoid loop base (see Figures 1–3). The inflated proximal ileum, twisted ileum, and closed sigmoid loop

were all gangrenous. As the ileum was difficult to untie, sigmoid resections were done in block (Figure 4A and B). After the ileum was untwisted, it was filled with warm, saline-soaked abdominal packs and given a 20-minute recovery period. The ileum, both distal and proximal, has somewhat recovered. Thus, a block of the gangrene-affected ileum was removed, leaving 7 cm of terminal ileum. After a second operation on the second post-operative day, the patient's general intestinal edema subsided, and there was a frank ischemia of the residual distal ileum, along with a recall of pus mixed with gastrointestinal (GI) contents from the ruptured anastomosis site. When the patient returned to the operating room (OR) after a full 72 hours, the results revealed a disruption in the ileocecal anastomosis along with an increase in notable memory loss. Laboratory tests revised on the first post-operative day (see Table 2). A functional jejunostomy and ileostomy was performed approximately 6 cm from the teres ligament and 6 cm from the ileocecal valve, utilizing a double-barrel (proximal end jejunum) stoma to extrude the necrotic ileum from the distal ileum. The patient's hemodynamic instability and severe intra-abdominal sepsis prevented re-anastomosis from being done. The lower abdomen was sealed. On the sixth post-operative day, the laparotomy wound was completely closed. The post-operative period for the patient was uneventful. On the six post-operative day, a regular oral diet was started and the patient was moved to the surgical ward. Three weeks after the initial bowel reconstruction,

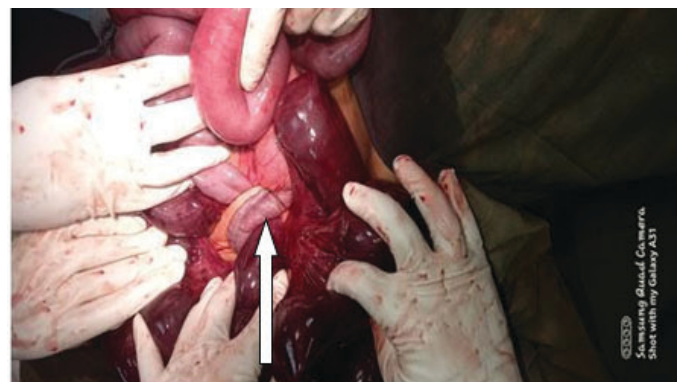


Figure 1: Intraoperative pictures showed the ischemic distal ileum and knotting part of the sigmoid colon.



Figure 2: Intraoperative picture taken using mobile camera, showing the small bowel which is gangrene.

an entero-cutaneous fistula developed, and the patient was released under conservative care. On the 15th day following the re-continuation surgery, the patient was released from the hospital after updating the laboratory (see Table 3).

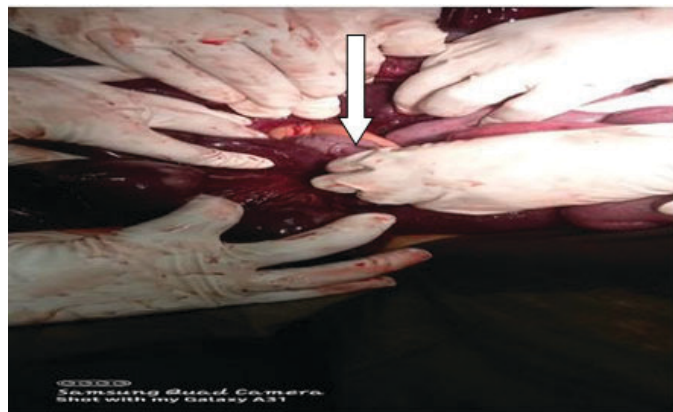


Figure 3: This intraoperative picture, the arrow is to show the part of large bowel (the most proximal part of sigmoid colon) which knotted the ileum.

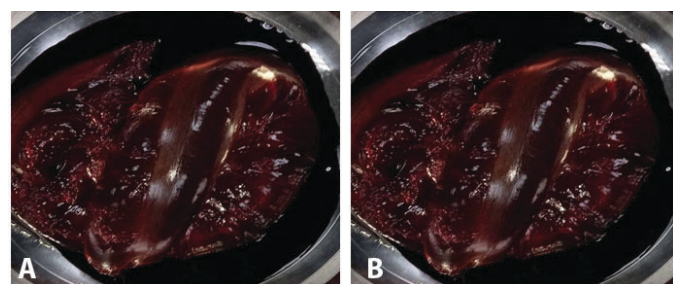


Figure 4: (A) This picture showed the distended and ischemic sigmoid colon, which is resected in block. (B) The resected part of sigmoid colon.

Table 1: Laboratory reports on the preoperative period

Lab result	Change
• WBC: 28.39	• PT: 10.4 sec
• Neutrophil: 89.3	• aPTT: 26 sec
• Lymphocyte: 3.7	• INR: 0.95
• Hgb: 10.6	• Urea: 91
• Hct: 27.3	• BUN: 43
• PLT: 156	• S.cr: 1.5
• Na: 136	• RBS: 107
• K: 6	• PT: 10.4 sec
• Cl: 110	• aPTT: 26 sec
• Ca: 1.27	• INR: 0.95
• Urea: 91	
• BUN: 43	

Abbreviations: WBC: white blood cell, PT: prothrombin time; aPTT: activated partial thromboplastin time; RBS: red blood sugar, Hct: hematocrit, Hgb: hemoglobin, INR: international normalized ratio, BUN: blood urea nitrogen, PLT: platelet.

Table 2: Laboratory results on the first and second post-operative day laboratory results

Serum chemistry 07/08/2023	Results
• Urea	58
• BUN	27
• Creatinine	1.39
• Na: 145	145
• K: 2.7	2.7
• Cl: 108	108
• Ca: 0.76	0.76
Laboratory report On second post-operative day 08/08/2023	
Results	Results
• WBC	• 7.27
• Neutrophil	• 82.2
• Lymphocyte	• 14.4
• Hgb	• 10.5
• Hct	• 31
• PLT	• 167
• Na	• 141
• K	• 4
• Cl	• 113
• PT: 9.2 sec	
• aPTT: 18.7	
• INR: 0.85	
• Urea: 37	
• BUN: 17	
• S.cr: 0.97	
• AST-SGPT: 99	
• ALT-SGPT: 75	
• TB: 0.36	

Abbreviations: WBC: white blood cell, Hgb: hemoglobin, Hct: hematocrit, PLT: platelet, BUN: blood urea nitrogen, PT: prothrombin time, INR: international normalized ratio, AST: aspartate aminotransferase, ALT: alanine transaminase, SGPT: serum glutamic pyruvic transaminase, TB: total bilirubin.

Table 3: Laboratory result prior to the discharge of the patient

WBC: 10.24	ALT-SGPT: 18
• Neturophil: 80.52	• S.cr: 1.36
• Lymphocyte: 7.15	• TB: 0.36
Hgb: 10.64	Total protein: 3.9
• Hct: 34	• Albumin: 2
• PLT: 409	

Abbreviations: WBC: white blood cell, Hgb: hemoglobin, Hct: hematocrit, PLT: platelet, ALT: alanine transaminase, SGPT: serum glutamic pyruvic transaminase, TB: total bilirubin.

Table 4: Taken from Schwartz's principle of surgery, 10th ed.

Risk factors for development of short bowel syndrome after massive small bowel resection
Small bowel length <200 cm
Absence of ileocecal valve
Absence of colon.
Diseased remaining bowel (e.g., Crohn's disease)
Ileal resection
Small bowel length <200 cm

This showed the cause of short bowel syndrome which is taken from Schwartz's principle of surgery, 10th ed.

DISCUSSION

Compound volvulus, sometimes referred to as double volvulus or ISK, is an uncommon surgical emergency that causes intestinal blockage. It's critical to distinguish ileosigmoid knot from simple SV since endoscopic decompression in the former can be dangerous or even lethal. A puncture or damage could result during the ileosigmoid knot's endoscopic decompression. Ileosigmoid knots can generally have a 48% fatality rate [2]. Ileosigmoid knotting is a rare case of acute abdomen or intestinal obstruction that later leads to bowel necrosis.

The underlying ethology of acute abdomen considerably influences the course of treatment and prognosis; yet, in most cases, this is a surgical emergency with accompanying morbidity and death [4].

One frequent acute surgical emergency is small bowel blockage (SBO). A wide range of conditions are included in the differential diagnosis of SBO, such as intestinal knotting, primary volvulus, hernias, adhesions, bands, and intussusceptions. The literature reports on a number of intestinal knotting varieties, including ISK. There have also been reports of appendico-ileal, ileocecal, ceco-sigmoid, and ileo-ileal knotting as other causes of intestinal knotting. The difficulty in making an early and preoperative diagnosis is the primary issue associated with intestinal knotting [5].

Parker was the first to report ISK in 1845, and Burkitt was the first to report a case in Africa in 1952. Shepherd introduced the term "Ileosigmoid knotting" in 1967 to describe a particular ailment. With a peak incidence in the third and fifth decades of life, it is more common among men. It is frequently found in regions where SV rates are higher. Because of its rarity, vague radiographic features, and resemblance to SV, misdiagnosis is common. Usually, the diagnosis is made during surgery. The cause is not well understood.

Ileosigmoid knot ethology is influenced by three variables: Three conditions have been identified: (I) a long small bowel mesentery with a free-moving small bowel; (ii) a long sigmoid with a narrow pedicle; and (iii) a large diet combined with an empty small bowel. According to certain research, a large diet may move up to the proximal jejunum, where it may improve small bowel movement and land in the left lower quadrant of the abdomen [6–8]. Small bowel loops have the ability to rotate around the sigmoid tight pedicle. With more peristalsis, this rotation can be advanced to a closed loop of the intestine. In addition to the physical reasons previously described, there are secondary causes. These include but are not limited to late pregnancy, transmesenteric herniation, Meckel diverticulitis with band, and ileocecal intussusceptions. Ileosigmoid knots are said to be uncommon among white populations and to be more common in some Middle Eastern, African, and Asian countries [9].

Depending on the active bowel segment, volvulus direction of the sigmoid colon (clockwise or counter

clockwise), and prevalence (sporadic or endemic), ileosigmoid knots are classified in different ways (ileum or sigmoid colon). Nevertheless, none of these classifications provide any details regarding the prognosis and available treatments for these illnesses. With regard to bowel viability, bowel anastomosis risk, age (mean life expectancy in Turkey is 75 years), and the American Society of Anaesthesiologists' physical status classification, Atamanalp presented newly updated classification systems with treatment algorithms and prognosis-estimating data for SV in 2020 and for ISK in 2021 [1].

A new classification system based on the patient's age, coexisting conditions, and gangrenous bowel existence has been proposed by Atamanalp. It falls into one of four categories: Types I–IV. The ileum, the active component, forms a knot around the passive sigmoid colon in type I, the most prevalent kind. The inverse of type I, type II involves the sigmoid colon—the active component—knotting itself around the ileum. The ileocecal segment and the sigmoid colon form a knot in type III. Type IV is unknown, and it is not possible to identify the factor that knots the other [9]. A new classification system was described by Atamanalp et al. in 2009 based on pre-operative and operative criteria that also correlate with mortality. Class-one patients are those with no risk factors. Class-two patients are those with no shock or bowel gangrene but who have the predisposing risk factors mentioned above. Class-three patients present with shock. Class-four patients have gangrene of the ileum or sigmoid colon. Class-five patients are those with both shock and ileum or sigmoid colon gangrene; those with both ileum and sigmoid colon gangrene are class six [9].

As ileosigmoid knots don't always present, diagnosing them is challenging. When ileosigmoid knot is misinterpreted radiographically as simple SV, attempts to do a sigmoidoscopy endoscopic decompression may be made. A puncture or damage could result during the ileosigmoid knot's endoscopic decompression. Ileosigmoid knots generally have a 48% fatality rate. While the overall diagnostic accuracy rate was 20.8%, abdominal X-ray radiography, computed tomography (CT), and magnetic resonance imaging (MRI) were found to be diagnostic in 8.2%, 96.2%, and 100.0% of cases, respectively. Of the patients, 85.6% had gangrenous bowels. In 14.0% of instances, ileum resection was performed, in 7.6% sigmoid colon resection, and in 67.1% of cases, both segments were resected. There was a 22.7% death rate and a 22.7% morbidity rate [6].

Of the 923 instances that were considered serious, the majority came from nations in Asia and Africa. With a male to female ratio of 79.9% to 20.1%, the average age was 43.9 years. The primary duration of symptoms was 48.1 hours, and the most frequent clinical characteristics were distention (88.3%), stomach pain/tenderness (99.1%), and constipation/obstipation (constipation) (58.8%). Of the patients, 85.6% had gangrenous bowels.

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Acid-base imbalance correction and vigorous intravenous fluid resuscitation are the first steps in the management of ileosigmoid knots. Stabilizing hemodynamic should be the first step toward surgical intervention. For 5–7 days prior to and following surgery, antibiotic medication should be administered. Examples of antibiotics to consider are aminoglycosides, metronidazole, and cephalosporin. In the literature, a number of surgical techniques for treating ileosigmoid knots are listed. The surgical approach is determined by the pathological and anatomical discoveries made throughout the surgery. If both the ileum and the sigmoid are gangrenous, the most frequently carried out operation is ilea and sigmoid resection with primary enteroenteric anastomosis ileum and Hartmann's procedure/colostomy for the descending colon. Because of the small terminal ileum in our case, which was less than 10 cm from the ileocecal valve, primary anastomosis was not carried out [7].

A rare complication of ISK is short bowel syndrome. In most cases, the removal of less than 50% of the small intestine is well tolerated. However, when more than 50–80% of the small intestine is removed, clinically substantial malabsorption happens [8].

However, residual bowel length is not the only factor predictive of achieving independence from total parenteral nutrition (TPN) (enteral autonomy). Other determinants of the severity of malabsorption include the presence or absence of an intact colon [10], as indicated earlier (see Table 4).

Following a large-scale small bowel resection, the residual intestine experiences compensatory adaptation during the first one to two years. The period of adaptation is linked to decreased frequency and volume of bowel movements, increased ability to absorb nutrients through the enteral route, and decreased need for total parenteral nutrition. Upon completion of this process, some patients are effectively weaned off of TPN [11].

Patients with SoBS who are unable to become independent off TPN may benefit from efforts to improve adaptation that are suggested by an understanding of the mechanisms mediating intestinal adaptation. Currently, there is still much to learn about the phenomenon of intestinal adaptation in patients [12].

In adults, the most common etiologist of SoBS (Table 4) are acute mesenteric ischemia, malignancy, and Crohn's disease. A significant portion of the small bowel is removed during a single procedure in 75% of cases. In 25% of instances, the cumulative impact of several small intestinal resection surgeries is the cause. Patients with Crohn's disease who experience small bowel syndrome typically follow the latter pattern, but those who experience acute mesenteric ischemia and intestinal infarction typically follow the former pattern.

Data on the chronic home total TPN administration were last collected in 1992. About 40,000 people were chronically getting TPN at home at that time.

Among these patients, SoBS was the most common reason for TPN; however, this estimate does not include patients with SoBS who are not getting TPN at home. It is unsuccessful as well [3, 13–16].

CONCLUSION

Ileosigmoid knotting is a rare cause of intestinal obstruction and bowel ischemia. To prevent morbidity, it is crucial to distinguish between simple SV and ileosigmoid knot. Early diagnosis could be made easier using computer tomography. Bowel resection with or without primary anastomosis is a common surgical procedure. Its morbidity and mortality rates are higher than those of other volvulus species. According to several research, the death rate from ISK ranges from 2% to 55% (mean 36%). Several prognostic indicators are discussed, including the patient's overall health, the existence of shock, the presence of gangrene, and the length of the sickness. Gangrenous patients have a significant death rate. There is also a significant rate of morbidity. Shock is the most frequent cause of death. There is a higher rate of morbidity but also a good survival rate after second opinion or stoma creation surgery. According to a review of the literature, there has been a consistent decrease in mortality over the past 20 years as a result of increased awareness of the illness, the availability of broad-spectrum antibiotics, and intensive care unit treatment. Because of the ischemia caused by delayed intervention, it may be the cause of SoBS. Our patients produce short bowel, which is about 6 cm from the ileocecal valve and the ilium was viable, after considerable portions of the small bowel and resection of part of the small bowel are removed.

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Author Contributions

Addis Y Kidane – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Author declares no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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