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Ileoileal knotting as a rare cause of acute small bowel obstruction: a case report

Noeud iléo-iléal comme cause rare d'occlusion aiguë de l'intestin grêle : à propos d'un cas

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Cas clinique

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ABSTRACT

Ileo-ileal knotting is the twisting of ileum around itself and is a rare cause of obstruction. The high morbidity and mortality are caused by the disease's quick course. A 23-year-old male presented with sudden onset abdominal pain and vomiting. Preoperative investigations suggested acute small bowel obstruction. Immediate surgical intervention revealed an ileoileal knotting with gangrenous bowels. Patient benefitted from intestinal resection and stoma fashioning. Outcome was favourable. A high index of suspicion and immediate surgical intervention is needed in the management of this entity because it can quickly progress to strangulation.

RESUME

Le nœud iléo-iléal est la torsion de l'intestin grêle sur lui-même et constitue une cause rare d'occlusion. La morbidité et la mortalité élevées de cette pathologie sont dues,à l'évolution rapide de la maladie. Il s'agissait d'un patient de 23 ans reçu en urgence pour des douleurs abdominales et des vomissements. Les bilans préopératoires avaient suggéré une occlusion aiguë de l'intestin grêle. L'intervention chirurgicale a révélé un nœud iléo-iléal avec nécrose intestinale. Le patient avait eu une résection intestinale et une stomie. Les suites opératoires étaient favorables. Un degré de suspicion élevé et une intervention chirurgicale immédiate sont nécessaires dans la prise en charge de cette pathologie car elle peut rapidement évoluer vers la strangulation.

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Introduction

Small Bowel Obstruction (SBO) is a common surgical emergency that occurs when the gut becomes mechanically blocked. It can be partial or complete and can be non-strangulated or strangulated. Bowel obstruction is a surgical emergency that requires significant healthcare resources and financial costs for its management. It accounts for 15% of surgical admissions for abdominal pain, with an annual incidence of 12/100000 cases in Africa (1,2). Early identification and treatment are critical to reducing the morbidity and mortality of SBO. The mortality rate for strangulated gut is about 100% if treatment is not received; however, if surgery is performed within 24 to 48 hours, the mortality rate is less than 10% (3). The age of the patient, presence of comorbidities, and treatment delay are factors that affect morbidity (3). Nonetheless, intestinal obstruction still results in a 5-8% overall mortality rate (3). Adhesions (65%), hernias (15%), and neoplasms (5%) are the main causes of SBO in developing countries (4). Other conditions such as Crohn's disease, volvulus, tuberculosis, parasite infections, and intestinal knotting have also been reported (4).

The occlusion of an intestinal segment with a closed loop phenomenon as a result of mesentery knotting is known as intestinal knotting. Rokitansky et al. documented intestinal knot development in 1836, although Riverius did so in the 16th century (5). Although there are a number of theories, the precise reason of this knotting has not yet been identified. The commonest cause of intestinal knotting reported is ileo-sigmoid knotting, others are appendico-ileal, ileocaecal, and ceco-sigmoid (7). Few examples of ileo-ileal knotting have been documented in the literature, making it the rarest type of intestinal knotting (6). The difficulty in making early and preoperative diagnoses is the primary issue with intestinal knotting that has been observed. We describe an adult male patient who had an abrupt strangulated intestinal blockage due to an ileo-ileal knot, which is rare and presented atypically.

Case presentation

A 23-year-old male, with non-remarkable past medical history, presented to the emergency department with a one-day history of severe sudden onset of constant, non-radiating periumbilical abdominal pains associated with multiple episodes of vomiting. There was fever and loss of appetite but otherwise he had no failure to pass faeces or flatus. On examination he was acutely sick looking, however his vital signs were stable. His abdomen was mildly distended, there was generalized tenderness and guarding marked at the umbilicus bowel sounds were not audible. A digital rectal examination showed that the rectum was empty. Laboratory investigations showed leucocytosis with neutrophil predominance, no other anomaly found. Plain abdominal Xray showed dilated bowels but no obvious air fluid levels.

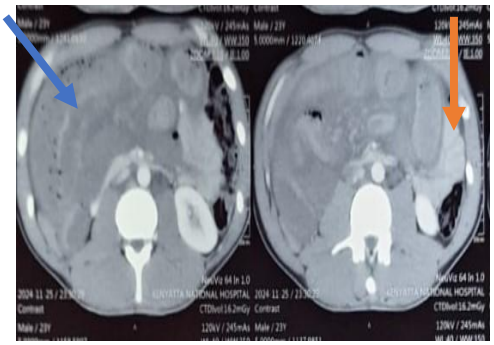

Figure 1: *preoperative axial view of abdominal CT scan image of patient. Blue arrow shows multiple pneumatosis intestinalis and orange arrow shows one of the multiple dilated bowel loops.*

Computed Tomography scan done (**Figure 1**) revealed dilated proximal small bowel loops with multiple air fluid levels, circumferential wall thickening and pneumatosis intestinalis and a transition point seen in the small bowel at the right lower abdomen with collapsed distal ileum. No obvious mass seen. There was crowding of the mesentery and twisting of the vessel. Features highly suggestive of SBO querying volvulus or internal hernia. Following resuscitation, a laparotomy was performed. Intraoperatively, 1.5l of haemorrhagic fluid was encountered, there was an ileoileal knotting, with twisting at the terminal ileum, strangulating about 200 cm of ileum (**Figures 2-3**).

Detorsion of the mesentery was attempted but not successful. A bloc resection of the gangrenous bowel and a right hemicolectomy were done. The remaining small bowel was about 250cm. postoperative period was unremarkable, close clinical and biochemical monitoring was ensured. Feeding was instituted gradually and was well tolerated. Progress in the ward was favourable and was then discharged on postoperative day 7. He was due for stoma reversal after 6 weeks.

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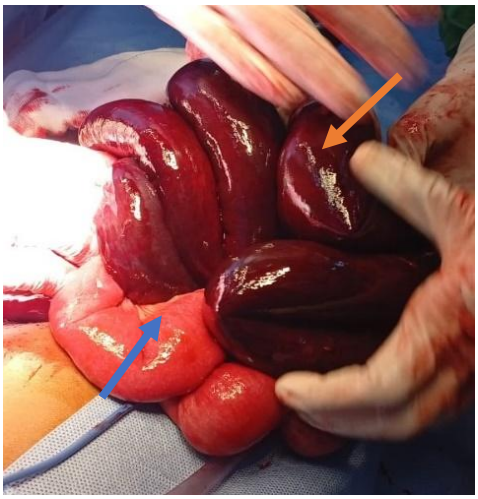
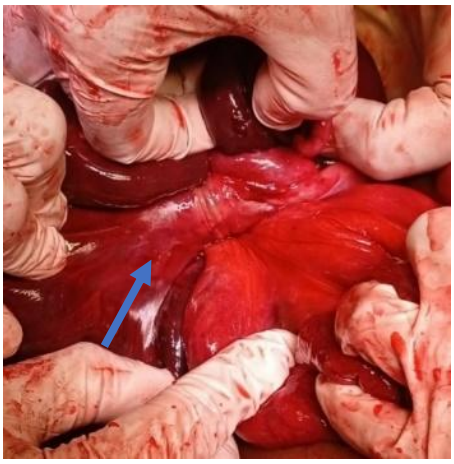


Figure 2: *intraoperative image showing viable bowel (blue arrow) and dilated gangrenous small bowel (orange arrow)*



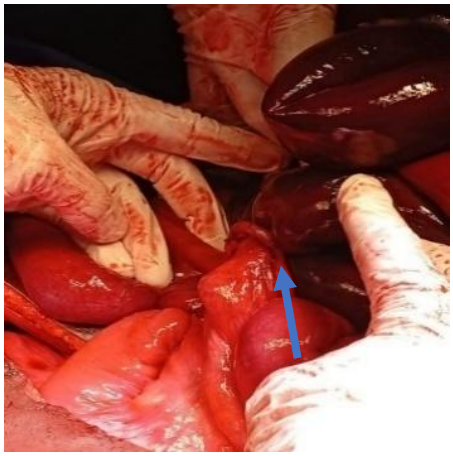


Figure 3: *intraoperative image showing exact point of knotting at ileal mesentery (blue arrow)*

Discussion

Intestinal knotting is a phenomenon whereby one dynamic coil of intestine twists and loops around another coil of intestine that is relatively static, giving rise to a knot. According to published research, the ileoileal knot is the rarest intestinal knot, with just a small number of occurrences reported worldwide (8). Its geographic range includes Africa, India, Asia, and East Europe, although it is extremely uncommon in the West. This might have something to do with their heavy, high-fiber diet. The majority of recorded occurrences in Africa occured in East Africa (7,9,10).

It is unclear exactly what causes ileoileal knotting. However, a number of processes have been proposed to account for these causes. Anatomical variations such as freely mobile small intestines from long and narrow mesentery with risk increasing if the bowels are loaded. Other mechanisms like having a single bulky meal especially during fasting, pregnancy as it causes displacement of bowel loops, intussusception especially with the presence of a lead point along with sudden vigorous peristalsis have also been described (8). None of which was noticed in our patient.

Despite being slightly more common in men, ileoileal knotting can occur in people of any age, with some studies reporting occurrences as young as 11 months and as old as 80 years (8). Although individuals with ileoileal knots may exhibit the traditional clinical tetrad of SBO abdominal pain, nausea and emesis, abdominal distention, and constipation-to-obstipation, these symptoms are not always present. According to studies, 93% of patients' primary complaint was always abdominal pain (8,11). He presented within a day of the onset of symptoms, which is consistent with previous research which stipulate that patient typically presented within 48 hours (8). This could be explained by the fact that once a knot forms, it initiates a vicious cycle of intestinal blockage, followed by persistent peristalsis and vascular compromise, all of which quickly leads to gangrene (8,10) prompting the early consultation. Due to its rarity, intestinal knotting is very challenging to diagnose preoperatively. In contrast to conditions like intussusception, which includes red currant jelly stools and pathognomic imaging signs, intestinal knotting only exhibits symptoms suggestive of acute abdominal obstruction and necessitates a high index of suspicion. A double closed-loop blockage, the "whirlpool sign," which indicates the twisting of the intestine and mesentery typical of volvulus, and pneumatosis, which suggests intestinal ischaemia caused by strangulation, are some of the radiological findings that have been reported to be highly suggestive (6). Our case's radiological findings reported a few of these characteristics. Because endoscopic reduction is contraindicated, it is critical to distinguish intestinal knots from other disorders like volvulus or intussusception (6,8,12). In any case diagnosis is only conclusively made intraoperatively just as in the present case.

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The anatomical and pathological changes explain the operative procedure of choice. Gangrenous bowel is encountered in 73 - 80% of cases (12). It is advisable to undo the knot when viable bowel is met. Given how rare the chance of recurrence is, this can be tried safely. Attempting to remove the knot in cases where gangrene is discovered may not work and result in a bowel perforation with spillage of gut contents and may also cause reperfusion injury. Untying the knot in this case is therefore not advised; instead, a bloc resection of the affected segment with either end-to-end anastomosis or stoma fashioning should be carried out, depending on the patient's clinical condition (9). In our case, we attempted to unravel the knot in order to define our anatomy and do adequate resection without taking out viable segments in the mix but failed. The lengthy surgical procedure and the hemodynamic instability made us fashion a stoma rather than primary anastomosis as there was increased risk of anastomotic leaks. A right hemicolectomy is the standard treatment in intestinal knotting when the lesion occurs less than 10-15cm of the ileocecal valve. Anaemia, electrolyte abnormalities, nutritional condition, and hydration status should all be evaluated in the initial postoperative period. Anastomotic leak symptoms should also be monitored in cases where anastomosis was performed. Depending on how much of the small bowel is left and whether the ileocecal valve is removed, long-term follow-up should look for signs of short bowel syndrome (5). The favourable prognosis for our patient may be explained by the early hospital presentation, appropriate preoperative care, and prompt surgical intervention. Due mostly to the disease's quick progression, the mortality rate in the majority of evaluations for ileoileal knotting cases is approximately 50% (7).

Conclusion

Ileoileal knotting is a very rare cause of acute intestinal obstruction with only a handful of cases reported worldwide. It is extremely difficult to diagnose preoperatively however proper patient history and imaging help a lot. It can only be confirmed intraoperatively. Clinicians should have a high index of suspicion and early surgical intervention has paramount importance in the management to limit the risk of gangrene of this unusual but deadly condition.

Authors contribution

BFA conceived and designed the manuscript. BFA and SER wrote the manuscript. FS, FP and OD review the manuscript. FP accepted the final version.

Conflict of interest: The authors declare that the research was conducted in the absence of any commercial or financial relationship that could be construed as a potential conflict of interest.

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