Small bowel knots

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ABSTRACT

INTRODUCTION Intertwining of bowel loops to form a knot is very rare cause of intestinal obstruction. Among intestinal knots, ileoileal knotting is the most rare, with only a handful of cases reported in literature. We present a rare case of ileoileal knotting and review of small bowel knots. The aim of this review was to summarise the existing evidence on small bowel knots and to postulate the possible mechanisms for knotting.

METHODS A systematic search was conducted for literature published up to December 2019 using MEDLINE, PubMed and Google Scholar databases, together with the references of the full-text articles retrieved. Papers with case reports of small bowel knots were considered to be eligible for inclusion in the review.

FINDINGS A total of 14 case reports were evaluated. There was no clear predilection for age or sex. Mostly cases were from Asia and Africa with no cases from the West. The presenting complaints were abdominal pain (93%), vomiting (64%), abdominal distention (57 %) and obstipation (43%). The bowel was gangrenous in 78% of cases. All underwent exploration, with the majority requiring resection and anastomosis of the involved segment.

CONCLUSION Ileoileal knotting is a very rare cause of intestinal obstruction. Possible mechanisms include loaded bowel with longer mesentery, vigorous peristalsis, single bulky meal, pregnancy and intussusception. The condition is extremely difficult to diagnose preoperatively and it is usually diagnosed intraoperatively. The standard of treatment is resection of gangrenous part and anastomosis.

KEYWORDS

Ileal knot – Ileoileal knot – Small bowel knot – Intussusception – Intestinal obstruction

Accepted 23 March 2020

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Introduction

Acute intestinal obstruction is a surgical emergency and accounts for approximately 15% of all emergency department visits for acute abdominal pain.¹ Complications of intestinal obstruction include perforation peritonitis and bowel ischaemia. The most common causes of intestinal obstruction include adhesions (60%), neoplasms (20%) and herniation (10%).² Adhesions resulting from previous abdominal surgery are the most important cause of small bowel obstruction, accounting for approximately 60% of cases. Less common causes of intestinal obstruction are intestinal intussusception (less than 5%), volvulus (less than 5%), intra-abdominal abscesses, gallstones and foreign bodies (less than 2%).^{3–5}

Twisting of bowel loops with an intervening knot is a very rare cause of acute intestinal obstruction and gangrene of the bowel. Among intestinal knotting, the most common is ileosigmoid knotting. Ileoileal knotting is a very rare entity, with only a handful of cases reported across the globe in the indexed literature.⁶ We present a review of small bowel knots in the indexed literature, together with our experience of ileoileal knotting with long-segment intussusception in a 55-year-old patient. The

objective of this review was to summarise the existing evidence on small bowel knots reported in indexed literature, with special emphasis on the mechanism, diagnostic difficulty and need for urgent treatment of the condition.

Methods

Computer literature searches of PubMed, MEDLINE, the Cochrane Library and Google Scholar were undertaken. Search terms used were ‘ileoileal knot’, ‘ileal knot’ and ‘small bowel knot’ and the time period was 1988–2019. The keywords were kept to a minimum to identify potentially relevant papers that could be easily searched while simultaneously reducing the potential loss of eligible articles. All appropriate articles and cases concerning bowel knots were studied and only those reporting ileoileal knots were considered eligible for inclusion in this review. No language restrictions were employed. Two of the authors (YB and LB) independently performed a meticulous search of the literature, excluded duplicate records and compiled the relevant data.

The search on PubMed yielded 3 results, while MEDLINE and the Cochrane Library did not produce any results.

Further search was carried through Google Scholar, which yielded 67 results, of which only 9 were cases of ileoileal knotting. A total of 13 cases were found across all platforms (Fig 1). In addition, we reviewed the reference lists of the original articles to understand presentation and pathophysiology. Data recorded included age and sex of the patient, chief complaints, location of the knot, presence of gangrene of the bowel or not, surgical procedure performed and patient outcome.

Findings

Case presentation

A 55-year-old woman presented to surgery emergency with pain abdomen and non-passage of faeces and flatus for two days. The pain was acute onset, started from the right lower abdomen and rapidly progressed to become severe and involved the whole of the abdomen. It was followed by abdominal distension and bilious vomiting (three episodes, about 800ml total volume). There was a history of loose stools for four days, with no history of fever or any other chronic disease or any previous surgery. Her medical history was unremarkable.

On examination, the patient was dehydrated, cold to touch, with a pulse rate of 122 beats/minute, blood pressure

of 86/68mmHg and respiratory rate of 16 breaths/minute, maintaining a saturation of 100% on room air. The abdomen was grossly distended with tenderness present all over. On rectal examination, the rectum was found to be distended with no faecal or bloodstaining of a gloved finger. The patient was resuscitated with intravenous fluids, following which she improved to a pulse rate of 102 beats/minute, blood pressure of 100/72mmHg, respiratory rate 17 breaths/minute and urine output of 200ml over four hours. Initial arterial blood gas analysis showed mild metabolic acidosis (pH 7.27; PaO₂ 92mmHg; PaCO₂ 34mmHg; HCO₃ 20 mEq/l) due to hypovolemic shock, which was corrected with fluid resuscitation.

After stabilisation of the patient, an x-ray of the abdomen was done, which showed dilated small bowel loops and multiple air-fluid levels. Her haemoglobin was 9.2g/dl, Total leucocyte counts were 14000/mm³, blood urea was 24mg/dl and serum creatinine 0.7mg/dl. Serum electrolytes and coagulation profile were within normal range (Na 138 mEq/l, K 4.0 mEq/l, INR 1.2). Computed tomography (CT) of the abdomen showed ileocolic intussusception involving a long segment of bowel with dilated intussuscepted ileum, a tubo-ovoid lesion in the intussusceptum component, probably appendix, with a thickened wall with a possible underlying tumour and consequent acute small bowel obstruction.

Figure 1 Flowchart of selection of articles included in review

The patient underwent emergency laparotomy and intraoperative findings were suggestive of jumbled terminal bowel loops with long segment intussusception, with an ileoileal knot in the preceding segment that was causing obstruction and hence resultant dilatation of proximal loops, together with strangulation of blood supply to terminal ileum and caecum. Examination revealed a loop of ileum intussuscepting into the ileal loop and the whole loop within loop intussuscepting into the caecum and ascending colon, traversing the ileocaecal junction and reaching just short of the hepatic flexure. An ileoileal knot was present in the bowel loop just proximal to intussusception. The knot was not amenable to release

and the involved segment of ileum was found to be gangrenous. The appendix was found to be normal. There was a firm 3 × 2cm polypoidal intraluminal mass in the terminal ileum, perhaps acting as the lead point for intussusception. The involved part of the bowel was resected and ileoascending anastomosis was performed.

Examination of the specimen showed gangrenous terminal ileum and caecum, with ileal segment intussuscepting into the large bowel and forming a knot just proximal to intussusception. There was 3 × 2cm polyp in the terminal ileum, which on histopathological examination was found to be a tubulovillous adenoma; the appendix was normal. The patient had an uneventful recovery and was discharged on day 6. The patient was healthy and symptom free to five months of follow-up in the outpatient department.

Results

There is no predilection for age for such cases. Knotting has been reported in an 11-month-old and an 80-year-old. Eight cases (57%) were found in male patients and six cases (43%) in female patients, showing no clear predilection for any sex. The geographical distribution extends to Africa, India, Asia and East Europe; for obscure reasons, the entity is very rare in the West. This may be related to a diet that is bulky and high in fibre. The presenting complaints were abdominal pain (93%), vomiting (64%), abdominal distention (57%), obstipation (43%), and the average duration of complaints was two days before presenting to the hospital.

Diagnosis was established as an acute intestinal obstruction on clinical and radiological findings, and the actual cause was only found intraoperatively. All cases underwent laparotomy. Most of the knots were found in the terminal ileum. Three cases (22%) were managed by de-rotation of the knot and the remainder (78%) required resection of gangrenous segment. In 10 cases (71%), anastomosis of the bowel loop was performed, while in one case an ileostomy was made. A mortality rate of 50% has been noted in the case of ileosigmoid knotting, although there were no deaths in the review of small bowel knots.

Discussion

Intestinal knot formation was first described by Riverius in the 16th century as 'intertwining of two loops of intestine' and later by Rokitsky in 1836 as 'compound volvulus'.^{7,8} Several types of intestinal knot formation are known. The ileosigmoid knot is the most common type of intestinal knot syndrome.⁹ Ileosigmoid knotting is defined as the wrapping of the ileum around the sigmoid colon and its mesentery or vice versa.¹⁰

The ileoileal knot is a very rare entity. In ileoileal knotting, one dynamic coil of ileum twists and loops around another coil of ileum that is relatively static, giving rise to a knot. In fact, an ileoileal knot was found in only 1 of 92 cases of intestinal knot.^{11,12} Also, our extensive search of the literature found only 14 reports (including ours) of ileoileal knot since 1971 (Table 1).

Figure 2 Computed tomography showing long-segment intussusception (target sign)

Figure 3 Ileoileal knot proximal to gangrenous segment

As there have been very few reported cases of ileoileal knotting, the exact cause is not properly understood. Factors such as freely mobile small intestine with a redundant sigmoid with a long and narrow mesentery of small bowel have been implicated in the causation of the ileosigmoid knot.¹⁵ Other studies have reported a relationship between dietary habits and ileosigmoid knots that the ingestion of a single daily meal is associated with a higher incidence of knotting.¹⁴ Most cases of ileal sigmoid knot reported during Ramadan fasting are due to a narrow mesentery and a high-volume intake after fasting.¹⁵ When a semi-liquid large quantity, bulky meal progresses into the proximal jejunum, it increases the mobility of the intestine. The heavier segments of the proximal jejunum then fall into the left lower quadrant. The empty loops of the ileum and jejunum twist and rotate around the base of the sigmoid colon which is loaded. The resultant peristalsis causes formation of an ileosigmoid knot with an obstruction involving two closed loops: one in the small intestine and the other in the sigmoid colon.^{16,17}

No such mechanism has explained an ileoileal knot, although it has been attributed to a mobile small intestine longer than the mesentery. We propose the following mechanisms involved in causing ileoileal knotting:

> loaded bowel with longer mesentery;

- > sudden or vigorous peristalsis;
- > single bulky meal as during fasting;
- > pregnancy as it causes displacement of bowel loops;
- > intussusception along with sudden vigorous peristalsis.

In our case, we hypothesise that the polyp acted as a lead point for long-segment intussusception, which led to sudden peristalsis of the bowel and twisting and turning of bowel loops. A history of loose stools for one week further explains the hypermotility of the bowel, eventually leading to a knot formation in the ileal loops. Once the knot is formed, it starts a vicious cycle of intestinal occlusion and ischaemia due to continuous peristalsis and vascular impairment, all of which leads to gangrene of the bowel. Gangrene of the bowel was found in 11 cases (78%) in our review. When all segments are viable untying the knot may be considered as an option as recurrence is uncommon. However, in most such cases resection of the affected segment is needed as there is usually an ischaemic gut at the time of presentation owing to delay in presentation and diagnosis.

The diagnosis of intestinal knotting preoperatively is extremely difficult. Coexisting pathology such as intussusception may be diagnosed based on clinical history such as passage of 'redcurrant jelly' stools, examination of the abdomen (any lump) and ultrasonographic and CT findings.¹⁸ It is always diagnosed intraoperatively. The usual presentation will be

Figure 4 Loop within loop intussusception with a proximal small bowel knot

Figure 5 Ileoileal knot

Figure 6 Ileal polyp acting as lead point for intussusception

Table 1 Reported cases of ileoileal knots in the indexed literature

Author (date)	Age (years)	Sex	Chief complaints	Location of knot	Surgery	Gangrene of the knot or segment	Patient outcome
Cowley (1971) ²⁵	25	F	abdominal pain, vomiting	50cm from ileocaecal junction	Resection of gangrene segment and anastomosis	Yes	Discharged
Pendse <i>et al</i> (1988) ²⁶	11 months	M	vomiting, constipation, distension	20cm from ileocaecal junction	Resection of gangrene segment and anastomosis	Yes	Discharged
Uday <i>et al</i> (2012) ²⁷	68	M	abdominal pain, distention, vomiting	15cm from ileocaecal junction	Resection of gangrene segment after untying of knot and anastomosis	Yes	Discharged
Andromanakos <i>et al</i> (2014) ²⁸	26	M	abdominal pain	ileum-ileum, sigmoid-ileum	Resection of gangrenous segment after untying of knot by enterotomy decompression and anastomosis	Yes	Discharged
Kumar <i>et al</i> (2015) ²⁹	75	F	abdominal pain, distention, vomiting	ileum-ileum	Enterotomy decompression after untying knot	No	Discharged
Abebe <i>et al</i> (2015) ³⁰	55	F	abdominal pain	8cm from ileocaecal junction	Resection of gangrenous segment and anastomosis	Yes	Discharged
Gopivallabh <i>et al</i> (2016) ³¹	54	M	swelling in the right groin abdominal distention, pain vomiting, constipation	1ft proximal to the ileocecal junction	Resection of gangrenous segment and anastomosis	Yes	Discharged
Taniguchi <i>et al</i> (2016) ³²	80	F	abdominal pain	10cm from ileocaecal junction	Resection of gangrenous segment after untying knot by enterotomy decompression and anastomosis	Yes	Discharged
Kalaichelvan <i>et al</i> (2016) ³³	65	M	diffuse abdominal pain, vomiting, constipation, distension	Terminal ileum also associated with volvulus of terminal ileum	Release of ileoileal knot and volvulus de-rotation	No	Discharged
Rajesh <i>et al</i> (2018) ³⁴	23	M	abdominal pain, vomiting, obstipation	20cm from ileocaecal junction	Derotation of knot	No	Discharged
Krishna <i>et al</i> (2018) ³⁵	22	M	abdominal pain, vomiting, abdominal distention, obstipation	1ft proximal to ileocaecal junction with volvulus of distal ileum	Resection of involved part of bowel with double barrel ileostomy	Yes	Discharged
Ahuja and Bhuta (2018) ³⁶	52	F	abdominal pain, abdominal distension	mid-ileal loops	Laparoscopic Resection of gangrenous bowel with extracorporeal anastomosis	Yes	Discharged
Nallagounder (2019) ³⁷	28	M	abdominal pain, vomiting, fever	5cm from ileocaecal junction	Resection of gangrenous bowel with end to end anastomosis with proximal protective loop ileostomy	Yes	Discharged
Beg, Bains <i>et al</i> (this study)	55	F	abdominal pain, distention, vomiting, constipation	25cm from ileocaecal junction	Resection of gangrene segment and ileoascending anastomosis	Yes	Discharged

that of an acute abdomen with sudden-onset abdominal distension. A plain x-ray of the abdomen in erect posture will demonstrate the features of intestinal obstruction. CT of the abdomen and pelvis may be done where necessary. An additional advantage of CT is that any coexisting pathology may be found. Characteristic signs of intussusception such as the target sign or loop within loop sign can be seen.¹⁹

Characteristic radiological features including a double closed-loop obstruction, are often misdiagnosed preoperatively. Abdominal CT also exhibits characteristic findings that may reveal a volvulus, including a characteristic 'whirlpool sign' showing the twisted intestine and mesocolon, both created by the twisted intestine and mesentery and medial deviation of the descending colon with a beak-like appearance on its medial border.²⁰ CT can also reveal signs of bowel ischaemia caused by strangulation, such as pneumatosis.

When irreversible ischaemia is present, controlled enterotomy decompression may be done prior to resection of the congested segments to release the tension within the bowel lumen. The mortality rate noted in the case of ileosigmoid knotting is 50%.²¹ In our review, there were no deaths. Manipulation of the knot with the intention of untying is not recommended in case gangrene has set in or the bowel shows signs of vascular compromise, and there is a high risk of perforation. Once the necrotic ileum is removed, a primary end-to-end anastomosis of the small bowel should be done if the distal ileum is not affected. On the other hand, if the remaining segment is closer than 10cm to the ileocaecal valve, end-to-side ileocolic anastomosis or ileoascending anastomosis is preferred.²² Only three cases were managed by de-rotation of the knot; the remainder required resection of gangrenous segment and anastomosis.

Despite reports that describe the radiographical appearance and clinical features of the ileoileal knot, the diagnosis of an ileoileal knot is difficult because of its rarity and atypical radiographical findings. Consequently, this rare abdominal condition is usually diagnosed during exploratory laparotomy. Even during surgery, ileoileal knotting can be misdiagnosed because the macroscopic features of this unusual condition are not familiar to surgeons.^{23,24}

Conclusion

Ileoileal knotting is a very rare cause of acute intestinal obstruction with only a handful of cases reported worldwide. Possible mechanisms include loaded bowel with a longer mesentery, sudden or vigorous peristalsis, single bulky meal (as during fasting), pregnancy and intussusception. It is extremely difficult to diagnose preoperatively, thus it is diagnosed intraoperatively and the standard of treatment is resection of gangrenous part and anastomosis.

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