

Appendicular knot causing closed-loop obstruction, volvulus and strangulation of ileum in a 9-year-old: a case report

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Appendicular knot/tie syndrome is a rare cause of mechanical intestinal obstruction. It is rarer still in the paediatric age group. We present the case of a 9-year-old boy admitted with clinical features and radiological evidence of acute intestinal obstruction, which at laparotomy revealed an inflamed/gangrenous knotted appendix, ileal volvulus and strangulation. This is a 'three in one' case of acute abdomen associated with appendicoileal knotting. In the presence of viable ileum, a retrograde appendectomy was performed. Postoperative period and follow-up were uneventful. Appendicular knot/band syndrome should be considered a possible differential in acute intestinal obstruction. Computed tomographic scan as an investigative modality should be selective in the presence of acute intestinal obstruction

Introduction

An inflamed appendix as a direct cause of acute small bowel obstruction is rare. Even rarer is an appendicular knot or a tie around the small intestine as a cause of mechanical intestinal obstruction. Preoperative diagnosis is usually difficult, and diagnosis is usually made laparotomy. Epidemiology of the reported cases suggest this condition to be commoner in patients in their third decade and above. Only three cases have been reported in patients less than 15 years of age worldwide. One case has been reported in Africa, but of an adult.

We report a 'three in one' case of acute abdomen in a 9-year-old in sub-Saharan Africa, presenting with features of acute mechanical intestinal obstruction with intraoperative findings of ileal volvulus and strangulation secondary to appendicoileal knotting by an inflamed/gangrenous appendix.

Case report

DA, a 9-year-old boy, presented at the Emergency Paediatric Unit of our facility, referred on account of colicky abdominal pain, bilious vomiting, abdominal distension and partial constipation of 4-day duration. There was no previous history of abdominal surgery.

On examination, he was found to be in acute pain; he was dehydrated and his body temperature was 37.2°C. Abdomen was centrally distended with visible intestinal patterning and peristalsis. Bowel sounds were exaggerated, and the digital rectal examination revealed tenderness in the rectovesical pouch. Chest was clinically clear, pulse rate was 110/min and the blood pressure was 120/90 mmHg.

Plain abdominal radiography revealed dilated loops of small bowel and multiple fluid levels on supine and erect

with or without peritonitis, with emphasis on adequate resuscitation and surgery. *Ann Paediatr Surg* 12:111–114 © 2016 Annals of Pediatric Surgery.

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views. Haemoglobin concentration was 13 g/dl, sodium 138 mmol/l, potassium 4.9 mmol/l, chloride 109 mmol/l and bicarbonate 17 mmol/l. An assessment of the acute intestinal obstruction with peritonitis was made.

Fig. 1



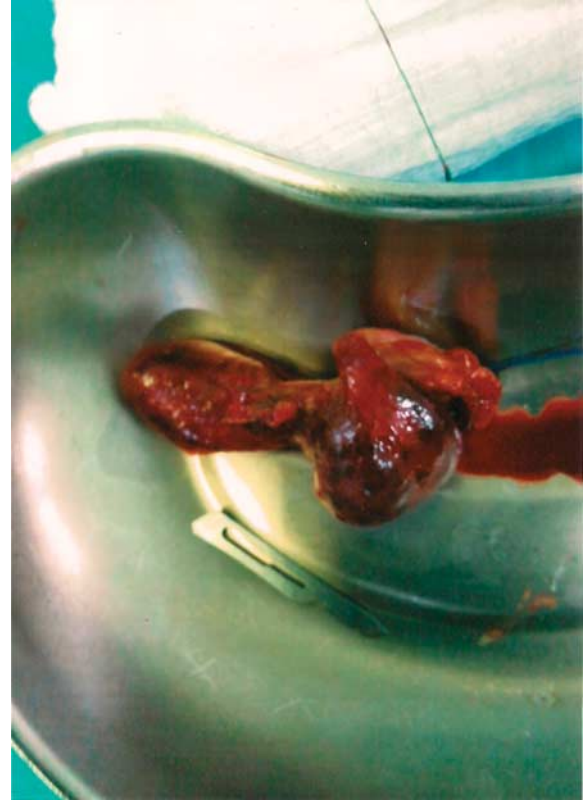
Ileal loop with inflamed appendix knotted tightly at its base.

Fig. 2



Appendix with gangrenous tip knotted around the base of a cyanosed loop of twisted ileum.

Fig. 3



Inflamed appendix with gangrenous distal end.

Fig. 4



Pink ileal loop after untwisting it and appendectomy.

The patient was vigorously resuscitated and underwent laparotomy 24 h after admission. Access was gained via a lower midline incision; intraoperative findings revealed a volvulus of 22 cm of ileum, 10 cm from the ileocaecal junction with cyanotic appearance. A 10-cm-long vermiform appendix was found knotted at the base of the ileal loop (Figs 1 and 2).

The Volvulus was detorted and retrograde appendectomy was performed to undo the knot and release the ileal loop. The appendix was grossly inflamed with 3 cm of gangrenous distal end (Fig. 3). The ileal segment was demonstrated to be viable (Fig. 4) and the peritoneal cavity lavaged. Histology of the appendix showed features in keeping with acute appendicitis.

Postoperative events were uneventful except for a mild, superficial surgical site infection, which was managed by wound dressings. The patient started oral intake 48 h after the operation, and was discharged on the seventh day after the operation. Follow-up was uneventful.

Discussion

An inflamed appendix as a direct cause of small bowel obstruction is rare; this is despite the first case being reported by Hotchkiss [1] in 1901. A review of the literature suggests that the pathology of small bowel obstruction by an inflamed appendix could be the following:

- (1) Adynamic: Ileus secondary to periappendicular inflammation and local sepsis.
- (2) Mechanical without strangulation: compression, kinking or traction of bowel trapped in an appendicular mass.
- (3) Mechanical with strangulation, the strangulating agent being
 - (i) tip of the inflamed appendix adhering to the posterior peritoneum across the ileum causing luminal compression [2].
 - (ii) tip of the inflamed appendix adhering directly to the bowel wall with associated compression and kinking of the loop [3].
 - (iii) tip of the inflamed appendix adhering to large bowel or posterior peritoneum forming a sling through which bowel herniates [4–7].
 - (iv) inflamed appendix encircling a loop or segment of ileum as a knot [8–16].
- (4) Mesenteric ischaemia with resultant gangrene of terminal ileum. This is the rarest form with only one reported case [3].

Mechanical obstruction and strangulation by an encircling inflamed appendix as a knot or inflamed appendix adhering to caecum, colon, small bowel or posterior peritoneum (i.e. 3c and 3d above) has been collectively termed the appendicular band syndrome or the appendicular tie syndrome [12,15,16]. This syndrome is characterized by a closed-loop obstruction, and the possible complications are intestinal obstruction, volvulus, small bowel and appendiceal strangulation [16]. There are 18 reported cases in the literature [4–7,14–16], but only two cases by Bose and colleagues [9,11] were complicated by ileal volvulus. In instances where the appendix knots at the base of an ileal loop causing obstruction and strangulation, the term appendicoileal knotting has been used descriptively [12,16].

Only five (27.78%) cases have been described in the paediatric age group [3,7,10,15], none of which was from the African continent. This probably conforms to the described bimodal age incidence rate of acute appendicitis [11].

The appendix by virtue of its mobility exhibits many variations in its normal positions relative to the ileum and caecum [17]. The initial phase of appendicular inflammation is characterized by an increased ability to adhere to contiguous structures. Progressively increasing oedema and fibrosis in an inflamed appendix wrapped around a loop of bowel may lead to increasing mechanical obstruction and strangulation of both structures. These dynamics in the presence of a relatively long appendix are considered the aetiopathogenesis of appendicular knot/band syndrome. In addition to these, narrowing of the base of mesentery of the involved loop of bowel predisposes it to volvulus as was the case in this report.

The clinical presentation of appendicular band syndrome is either of the following two:

- (1) predominant picture of acute intestinal obstruction, which is the more common presentation reported.

This obscures significantly the features of appendicular disease. This was the case in this report.

- (2) predominant features of appendicitis with some evidence of intestinal obstruction [4].

Although the clinically correct diagnosis of acute intestinal obstruction was always made, the diagnosis of appendicular band syndrome in all reported cases was made at laparotomy. It has been suggested that preoperative diagnosis may be improved by computed tomographic (CT) scans. However, in the light of the fact that this condition invariably requires urgent surgical exploration, complications of CT scan (radiation, possible contrast reaction and high cost) and its unavailability in some developing countries, selective scanning as suggested by Narjis *et al.* [5] and Assenza *et al.* [11] is advised. Prompt and accurate diagnosis, adequate resuscitation and treatment is desirable as morbidity and mortality rise significantly with delay. In Fevang's retrospective study of 877 patients undergoing surgery for small bowel obstruction, mortality rate rose from 3% in the event of viable loop strangulation to 16% in the event of nonviable loops. Likewise, the complication rate rose from 16 to 36%, respectively [18]. On the strength of the clinical diagnosis of acute mechanical intestinal obstruction, we recommend urgent resuscitation and exploration to forestall devitalisation of bowel and gangrene, which significantly increase morbidity and mortality.

An incision offering adequate access during exploratory laparotomy is essential, as the cause of mechanical obstruction remains unknown preoperatively. In most studies, the vertical midline incision was used, as it provides adequate access for the range of surgical procedures employed in the treatment, ranging from appendectomies to right hemicolectomies [13]. This was the incision used in this case.

Conclusion

Appendicular band/knot syndrome, though relatively rare, has become a recognized entity in which the inflamed appendix is a direct cause of mechanical small bowel intestinal obstruction. It should be considered a differential diagnosis in patients with features of mechanical small bowel obstruction. Diagnosis is however usually made at laparotomy, and valuable resources need not be expended on CT scan when features of obstruction with or without peritonitis are present.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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