CLINCAL CASE

APPENDICULAR BAND SYNDROME: A RARE CAUSE OF INTESTINAL OBSTRUCTION IN NEONATE WITH REVIEW OF LITERATURE

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Abstract

Acute appendicitis is a quite common condition in children, but it is very less common in neonates and infants. Acute appendicitis has several ways of presentations but its presentation as mechanical obstruction is very rare. We report a case of a 16 days old male patients who presented with acute intestinal obstruction due to appendicular band syndrome, and had been treated successfully in our institute.

Keywords*: appendicular band syndrome, appendicitis, intestinal obstruction*

Introduction

Acute appendicitis is problem which is quite common in children and adults, but extremely rare condition in neonates and infants accounting to 0.04% [1,2]. In neonates, it may be associated with an underlying pathology like necrotizing enterocolitis, meconium plug syndrome or Hirschsprung's disease. Acute appendicitis can lead to both the adynamic and mechanical obstruction of bowel. Again, presentation as mechanical obstruction is very rare [3]. We would like to share our experience of appendicular band syndrome presenting as acute intestinal obstruction.

Case presentation

A 16 days old male child was admitted with complaints of abdominal distension and bilious vomiting for 1 day, not passing stool for 2 days. The baby was born by Lower segment caesarean section (LSCS), at a gestation of 33 weeks, with a birth weight of 1.6 kg. The baby had passed meconium within 24 hours after birth and bowel movements were regular following that, and he was on breast feeds. There was no history of fever.

On examination, the baby was lethargic, dehydrated and was ill looking. There was tachycardia and tachypnoea, the heart rate was 170/min, and the respiratory rate was 66/min. The abdomen was distended with visible bowel loops, tenderness was present and there was visible intestinal peristalsis. A provisional diagnosis of acute intestinal obstruction due to bowel stenosis or adhesive band was made.

Routine investigations revealed a high total leucocyte count at 51,140/mm^3 with neutrophil count of 76%. The blood urea and serum creatinine were elevated, values were 113 mg/dl and 0.9mg/dl respectively. The venous blood gas revealed a pH of 7.25 and lactate level of 6.4 mmol/L. The baby was resuscitated with intravenous broad-spectrum antibiotics and fluid. Nasogastric aspiration showed a bile stained aspirate.

The supine plain X-ray of the abdomen showed evidence of numerous distended small bowel loops with no pelvic gas (Figure 1). There was no sign of free air in the abdomen. A contrast enema revealed a normal sized colon with non-opacified dilated small bowel loops in the background.

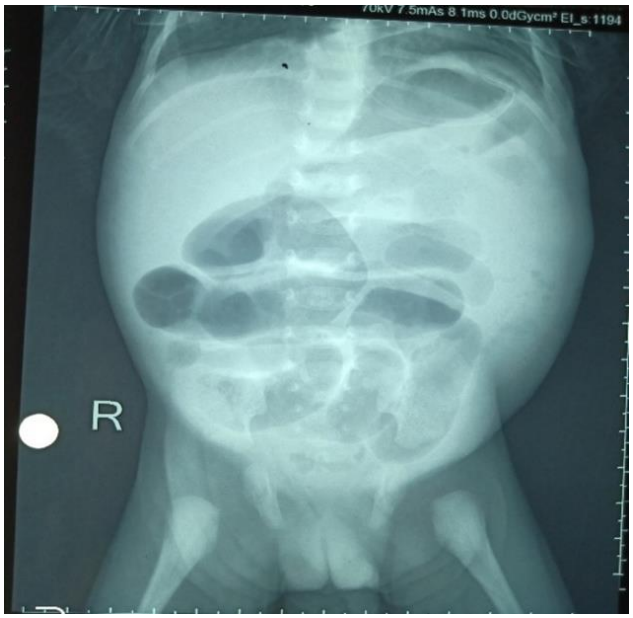

Figure 1 – X-Ray abdomen showing distended small bowel loops with no pelvic gas

An emergency laparotomy was done with a view to relieving the small bowel obstruction. We found that the distal small bowel was being compressed and constricted by a thick fleshy tight band adherent to the antimesenteric portion of the distal ileum (Figure 2), 10 cm proximal to the ileocecal valve. The band, we found, was arising from the tip of a thickened appendix. The constricted portion of the ileum was necrotic and almost perforated. Because the baby was very ill, temporary loop ileostomy with appendicectomy was done, after releasing the tight appendicular band. The baby recovered well from the operation and was discharged on the 15th postoperative day, after gaining adequate weight. After 3 months, it was planned for intra-operative rectal biopsy to rule out underlying pathology of Hirschsprung disease, and for closure of loop ileostomy. Distal loopogram was normal, with patent distal bowel. Intra-operative frozen rectal biopsy report came out to be normal. Ileostomy was closed in same sitting. Patient is in follow up and is doing well.

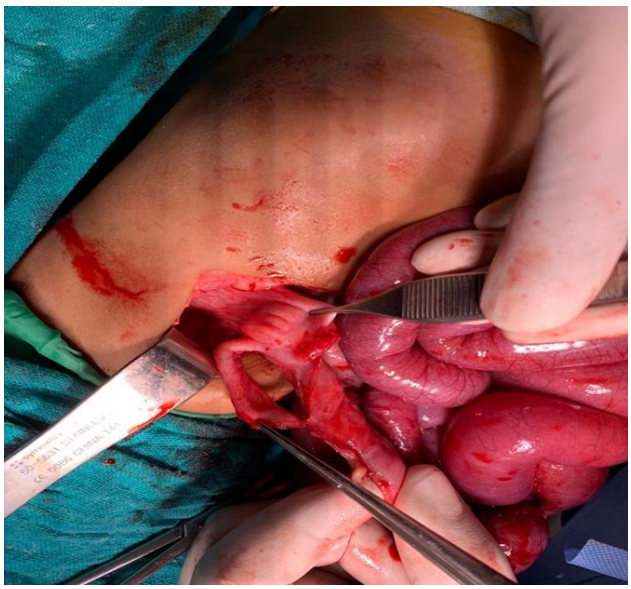

Figure 2 – Appendix tip adhered to antimesenteric border of ileum entangling bowel inside it

Discussions

Appendicular knot syndrome, also known as appendicular band syndrome or appendicular tie syndrome is an extremely uncommon condition where a segment of bowel is entrapped by appendix causing constriction ring which can cause obstruction or strangulation and finally can lead to gangrene of entrapped segment [4]. It was first described by Hotchkiss in 1901 [5]. It is extremely rare in neonates and infants [6]. Appendicitis itself is very rare in newborn, though, when it does occur in this age group, the risk of appendicular perforation is very high, about 85% [7].

In our case, it is probably the inflammation or perforation of the inflamed appendix which resulted in a band forming between the tip of the appendix and the mesentery of the terminal ileum.

Bhandari et al in 2009, had classified intestinal obstruction due to appendicitis into four types: mechanical, adynamic, strangulation and mesenteric ischemia [3]. Assenza et al have described an adult patient with operative findings similar to ours, and have suggested that mechanical bowel obstruction caused by appendicitis, can be due to the tip of the appendix becoming adherent to the mesentery or the serosa of the bowel leading to direct compression of the intestine [8].

Till now, we could find out only one case published in neonates, in literature. Though

many cases are published in children and adults. Sarkis P. had described in 21day old, intestinal obstruction due to appendicular band syndrome. He also performed appendicectomy [9].

Appendicitis in neonates is very rare and its occurrence frequently denotes an underlying significant etiology like necrotizing enterocolitis, meconium plug syndrome or Hirschsprung's disease. Our patient's history and findings did not indicate necrotizing enterocolitis. We plan to do a rectal biopsy for this patient before closure of the ileostomy to rule out Hirschsprung's disease.

Initial resuscitation and appendicectomy is standard treatment, though may need stoma formation, if there is gangrenous bowel or patient is very ill with impending stricture or perforation, as was in our case.

Conclusion

Though appendicular band syndrome is extremely rare in neonates and infants, it should be considered, in cases of acute intestinal obstruction. During the preoperative period, it is very difficult to suspect this etiology of mechanical bowel obstruction in this age group. Any delay in operating on these patients can have an adverse influence on their prognosis, and prompt intervention is needed.

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