CASE REPORT

Appendiceal Knotting: A Rare Complication Causing Intestinal Obstruction in a Child
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ABSTRACT
Appendicitis is a common cause of acute abdomen in children. Its common complications are; perforation, gangrene and appendix mass. The most dreaded complication is portal pyemia which could be fatal. Appendiceal knotting is a rare complication. No patient with this complication has been reported in literature. We therefore present the management of this rare complication seen as a cause of intestinal obstruction in a child at our centre.

KEY WORDS: Appendicitis Complication, Knotting.

INTRODUCTION
Appendicitis is the most common cause of surgical abdomen in children. Currently it accounts for approximately one third of childhood hospitalisation for abdominal pain. It is also considered as a cause of acute non specific abdominal pain in children. The life time risk of appendicitis is estimated to be 8.67% for boys and 6.7% for girls. Appendicitis is most common in older children and adolescents with a peak incidence between 12 and 18 years. This condition is less common in those who take high fibre diet.

A genetic predisposition appears operational in some cases particularly in children in whom appendicitis develops before the age of 6 years. Luminal obstruction causing appendicitis may result from lymphoid hyperplasia, inspissated faecal matter and sometimes parasites. The diagnosis of appendicitis in a child deviates from the classic description of the disease. This is because the presentation and manifestation in a child is completely different from an adult. It is also pertinent to note that complications ensue quite early in children when compared to adults.

Appendiceal knotting is a rare complication of this disease. No patient has been managed in our environment or reported in literature with this condition. There is also no report of this condition as a cause of intestinal obstruction. We therefore report the management challenges of a patient who presented with appendiceal knotting causing intestinal obstruction at Usmanu Danfodiyo University Teaching Hospital, Sokoto, Nigeria.

CASE HISTORY
HM was a 10 year old boy who presented to our hospital with a 6-day history of inability to pass stool, associated with central abdominal pain, progressive abdominal distension and bilious vomiting which became prominent 5 days after the onset of illness. He had absolute constipation. There was no history of febrile illness prior to presentation. No past history of similar abdominal pain. No history of trauma to the abdomen in the past or previous abdominal surgery.

At presentation, the child was acutely ill looking, in obvious painful distress. He was however not pale, anicteric and acyanosed. He was dehydrated with a temperature of 37.0 C. He weighed 18 kg (90% of his expected body weight). Pulse rate was 100 beats per minute, regular, good volume. Blood pressure was 130/90 mm Hg. He had normal heart sounds. He was dyspnoic with a respiratory rate of 40 per minute. He had vesicular breath sounds, no added sounds.

The abdomen was grossly distended. However it moved with respiration, with visible peristalsis from left to right. The abdomen was tense but non tender. No palpable organomegaly. No masses were felt. The bowel sounds were exaggerated. Inguinal hernial orifices were intact. Rectal examination revealed an empty rectum, no mass, no tenderness, gloved finger stained with normal rectal secretions, non mucoid and non bloody.
A provisional diagnosis of intestinal obstruction possibly from malrotation with midgut volvulus was made with differentials of colonic obstruction and intestinal duplication entertained.

He was resuscitated. The packed cell volume was 37% and urea and electrolytes were within normal limits after resuscitation. There was delay in obtaining plain abdominal X-ray and abdominal ultrasound before intervention. He had intervention without these investigations to avoid further deterioration.

At laparatomy, via a long lower midline incision, the intra-operative findings were; an inflamed 10 centimetre appendix with the tip knotted at the inferior surface of the terminal ileum and an inflammatory mass surrounding the tip. There was herniation of part of the terminal ileum through the knotted appendix causing a closed loop obstruction as well as simple obstruction proximal to the closed loop portion. The proximal portion of the ileum, the jejunum, duodenum and stomach were dilated. The distal ileum and the entire colon were collapsed. There were no adhesions and no intestinal duplication seen. There were no features of malrotation. There was an ischaemic portion on the terminal ileum at the level of the neck of the closed loop obstruction which recovered after relief of the obstruction, figure 1 and 2.

The obstruction was released by detaching the tip of the knotted appendix from the terminal ileum. Appendectomy was done but the stump was not buried, figure 3.

The post operative recovery was satisfactory and the patient was discharged home in good condition on the 7th day post operatively after removal of stitches. Histology revealed schistosomal appendicitis and schistosomal appendiceal lymphadenitis.

**DISCUSSION**

Diagnosis of appendicitis could be quite challenging in children. The classic description of this disease in adults often deviates in children, thereby making diagnosis difficult. More so in children the various differential diagnosis may be difficult to differentiate by clinical and laboratory means. For example, neonatal appendicitis could be indistinguishable from focal necrotising...
enterocolitis confined to the appendix. 

Intestinal obstruction in children may also be difficult to diagnose. Intestinal obstruction from internal herniation is particularly difficult to diagnose pre-operatively. Most often the diagnosis is made intra-operatively as was seen in our patient. The common areas for internal herniation are the hiatus orifice, foramen of Winslow, para-duodenal recesses, the pre-ileal recess and the obturator foramen. There may also be internal herniation if there are defects on the mesentery.

Appendiceal knotting causing intestinal obstruction is a rare and strange cause of intestinal obstruction as seen in our patient. It therefore suffices to say that appendicitis when it occurs could be complicated to any dimension. The natural history of appendicitis is quite varied. There are instances where appendicitis has resolved spontaneously. Perforation is a very common complication in children especially those younger than 6 years and even more in those less than 1 year. The other complications are gangrene and appendix mass which could either be an appendix phlegmon or an appendix abscess. Portal pyemia is a rare and dreaded complication which may result in high mortality. Due to these complications, it is advocated that appendicitis should be treated urgently in children. Appendicitis when it occurs may sometimes cause intestinal obstruction in children. This obstruction occurs due to ileus caused by local inflammation around the terminal ileum and caecum. Obstruction may also occur when there is generalised peritonitis causing ileus. This obstruction is adynamic and different from the mechanical obstruction seen in our patient.

Diagnosis of appendicitis was not entertained in our patient because the child presented with features of mechanical obstruction. There were no features suggestive of appendicitis before the features of intestinal obstruction. This was however not surprising since the presentation of appendicitis in a child could be quite deviant. There was a delay in obtaining appropriate radiological and ultrasonographic investigations which might have assisted in making a diagnosis or eliminating differential diagnosis. This was a major management challenge. Computerised tomographic scan which could have been useful here was not readily available. Sometimes when available, affordability becomes a problem. Delay was a major issue. Due to these limitations, laparotomy was therefore employed in the patient as a delay might have resulted in deterioration of the clinical state of the patient or might have resulted in morbidity or mortality of the patient.

Appendiceal knotting is a very rare complication of appendicitis. This condition has not been reported in literature. The other conditions that might have simulated this condition could have been malrotation with mid gut volvulus, ileo-sigmoid knotting, strangulating intestinal adhesions or bands, intestinal duplication or intussusception.

The treatment for appendicitis is appendectomy. Our patient with appendiceal knotting had relief of obstruction by detaching the tip of the inflamed appendix from the terminal ileum with appendectomy performed without burying the stump. This was necessary in this patient as there was associated surrounding inflammation involving the terminal ileum and caecum.

Appendicitis which is a common condition in children. It may continue to be complicated variably and dimensions may continue to be added as seen in our patient with appendiceal knotting causing intestinal obstruction from internal herniation into the recess created by the knotted appendix. It is therefore important to continue to report these new dimensions when they occur in order to inform paediatric surgeons with a view to improving morbidity and mortality.

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