Duplication cyst of the bowel causing ileal volvulus. A case report

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Abstract

A 10 month old male infant presents to us with features of acute intestinal obstruction and a palpable abdominal mass of five days duration. A prior ultrasound report had made a diagnosis of intussusception. Subtle but important details in history and examination cast doubts on the ultrasound diagnosis. Exploratory laparotomy revealed ileal volvulus secondary to duplication cyst. This case is reported because of its rarity and to show the strength of sound clinical evaluation even in the face of technological advancements.

***Key Words**: Duplication cyst, alimentary tract, ileal volvulus*

Date Accepted for Publication: 19th April 2010
NigerJMed 2010: 230 -232

Introduction

Duplications of the alimentary tract are rare congenital malformations and may occur in any part of the tract. The worldwide incidence is in the range of 1:4500 livebirths. Various theories have been put forward to explain the occurrence of these duplications. The most accepted theory is that which relates the duplications to the persistence of portions of the neuroenteric canal during embryogenesis. In this regard, alimentary tract duplications are considered part of the split notochord syndrome. These duplications constitute an important clinical entity due to the wide variation of presentations which frequently make a preoperative diagnosis difficult,. Additionally, they can result in very serious complications like intestinal obstruction, ulceration, bleeding and perforation. Volvulus secondary to intestinal duplication is rare. We report this case of ileal volvulus secondary to duplication cyst of the ileum because of its rarity and to highlight the essence of attention to fine details in evaluation of patients and in making diagnosis.

Case Report

A ten month old male infant was brought to our children emergency ward with five days history of abdominal pain, vomiting, and non passage of stool. Vomiting was bilious and followed soon after meals. Abdominal pain came on in paroxysms associated with patient wriggling and straining. There was no associated fever or passage of blood in stool.

Patient was well nourished, not pale, afebrile, anicteric, acyanotic, but in obvious painful distress. The abdomen was full, with obvious intermittent peristaltic waves with bowel markings. There were no umbilical or groin swellings. A mobile intra-abdominal mass was palpable, spanning the periumbilical and left iliac regions. Digital rectal examination showed an empty rectum; bimanual palpation could not feel mass in the rectum. Exam finger was stained with scanty faeces but no blood.

An abdominal ultrasound report had identified the abdominal mass as intussusceptions. Plain abdominal xray showed dilated bowel loops and air fluid levels.

With a working diagnosis of acute intestinal obstruction, ? cause, to rule out intussusception, we optimized the patient and embarked on an exploratory laparotomy.

At surgery, a 12x10 cm cystic swelling was intimately applied to the mesenteric border of the ileum, about 30 cm from the ileocaecal junction. With this cyst as a pivot, there was a 360 degrees twisting of the bowel proximal and distal to cyst (Fig 1).

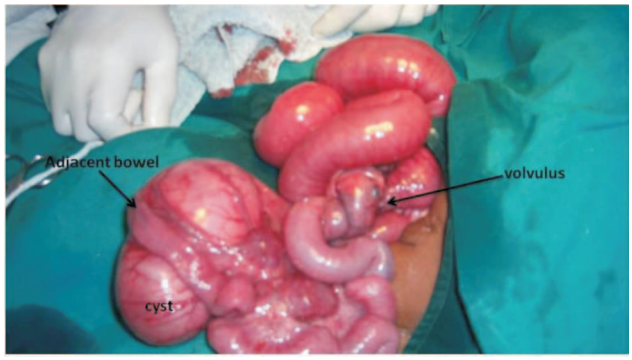

Figure 1 : Shows duplication cyst and the bowel twisted around its mesentary

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The volvulus was detorted and there was no evidence of ischaemic injury or gangrene. The cyst was excised incooperating the bowel adjacent to it. This was then followed by an ileoileal anastomosis. Post operative recovery was uneventful.

Histopathological analysis showed that the cyst shared a common muscular wall with the adjacent bowel and so confirmed the diagnosis of duplication cyst (Fig 2).

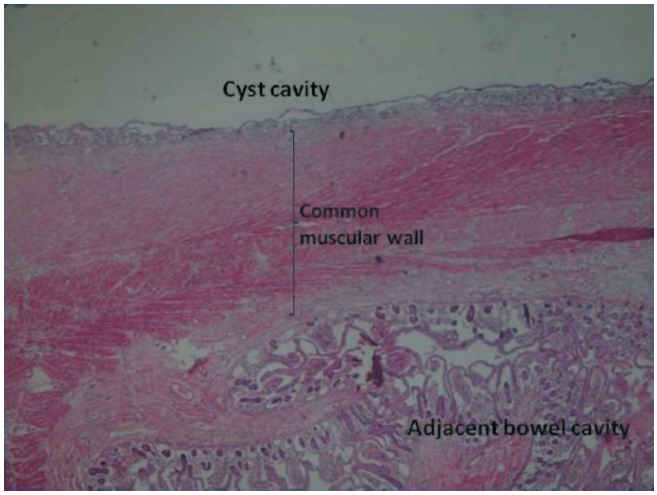

Figure 2: Histology picture of cyst and adjacent bowel showing a common muscular wall

Discussion

Intestinal duplication cyst(IDC) is a rare cause of intestinal obstruction in children. The rarity of this entity and our low index of suspicion made a preoperative diagnosis difficult. This is often the scenario with cases of intestinal duplications[4]. IDC causing obstruction may do so by kinking the bowel, bulging into bowel lumen or twisting of the bowel(volvulus). Volvulus is a rare complication of IDC. Even in regions where small bowel volvulus has been recognized as a common cause of mechanical intestinal obstruction, IDC is hardly encountered as the cause of volvulus A PUBMED search yielded two reports of volvulus occurring in association with IDC but there were no clear cause effect relationship.,. Our intraoperative findings (Fig. 1) suggest that the IDC provided the moments for the volvulus.

Even if we had made a preoperative diagnosis of volvulus, we would have considered intestinal malrotation rather than IDC since the former is a more likely cause of volvulus in that age group. In this patient, the diagnosis of intussusception was supported by the age of ten months which is around the peak occurrence of primary intussusceptions. In addition , the presence of a palpable abdominal mass and intestinal obstruction, and an ultrasound report diagnosing intussuception could have swayed our judgment towards intussusception. Given the clinical state of the patient, a non operative reduction (hydrostatic reduction) would have been the appropriate line of treatment.

However, a bit of attention to some details saved us from this otherwise inappropriate and possibly harmful line of treatment for this patient. Firstly, a patient presenting five days after onset of symptoms of intussusception would be expected to be more ill than we saw. Secondly the absence of the red currant jelly stool and blood on digital rectal examination were strong pointers that the diagnosis of intussusception was unlikely. The fact that the mass was not felt by the finger in the rectum despite being felt in the left iliac fossa cast doubts as to whether the mass was actually within the bowel. We decided to explore being convinced that patient did not have intussusception but we did not consider duplication cyst as a diagnosis due to our low index of suspicion. However, having discovered it at surgery, it became necessary to resect the normal bowel along with the cyst to achieve a satisfactory outcome. This points to the essence of surgeons being familiar with the various management options for the treatment of IDC as advocated by some researchers. Most authors recommend that internal drainage of IDC should be avoided as much as possible since 30-50% of them may have heterotopic acid secreting mucosa which can lead to ulceration and bleeding subsequently[2].

This report highlights how unreliable or misleading ultrasound reports and indeed other investigative tools could be in our environment. With thorough evaluation of patients and attention to details, clinical judgement may be considered superior in the face of investigative reports contradicting compelling clinical features.

It is interesting that despite a 5 day history, the volvulus did not cause bowel gangrene. This is unlike volvulus due to intestinal malrotation which tends to be more extensive and more dramatic. Volvulus due to duplication cyst is a far less severe problem and only involves the portion of the bowel around the cyst.

Conclusion

Volvulus and intestinal obstruction in children can result from duplication cyst of the bowel. In infants it could easily be mistaken for intussusception. However, with high index of suspicion, a correct preoperative diagnosis can be made and appropriate and timely treatment instituted.

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