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Case Report

Radiology Section

Ileocolic Intussusception in an Infant with Ileal Duplication Cyst as a Lead Point- A Case Report

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ABSTRACT

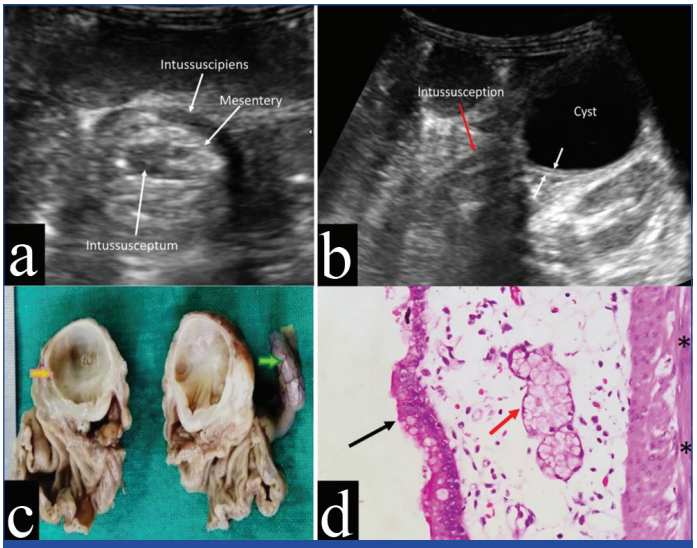
Gastrointestinal duplication cysts are uncommon congenital malformations, with small intestine being the most common site, followed by colon and stomach. It can have variable presentations such as intestinal obstruction, bleeding, palpable mass, or rarely volvulus. Here, the authors report a case of intussusception in a two-year-old child, presented with complaints of bilious vomiting and abdominal distension for three days with X-ray features suggestive of bowel obstruction. Ultrasonography revealed intussusception with ileal duplication cyst as the lead point, which was confirmed on exploratory laparotomy. In a paediatric patient, enteric duplication cyst should be included in the differential diagnosis of a cystic lesion as the lead point in intussusception.

Keywords: Gastrointestinal duplication cyst, Intestinal obstruction, Jejunal loops

CASE REPORT

A two-year-old child presented to the Emergency Department with complaints of bilious vomiting and abdominal distension since three days. The child was dehydrated and sick with a distended abdomen and step ladder pattern of peristalsis on examination. He had a pulse rate of 100 beats/min and a temperature of 37.5°C. Patient's laboratory parameters were within the normal limits except for a high White Blood Cell (WBC) count of 14700/mm^3. Plain radiograph of the abdomen showed multiple air-fluid levels involving the jejunal loops. Emergency ultrasound of abdomen revealed bowel loop, telescoping into bowel loop has given a target appearance located in the right iliac fossa, suggestive of intussusception [Table/Fig-1a]. Bowel loops proximal to intussusception were dilated. On further ultrasound scanning, a cystic lesion measuring 3.6×4.2 cm was noted at the leading end of the intussusception. The cyst showed a double-layered wall, suggestive of gut signature [Table/Fig-1b]. Based on gut signature in a cyst that acts as a lead point of intussusception, diagnosis of ileal duplication cyst causing intussusception and intestinal obstruction was made on ultrasound.

At laparotomy, ileocolic intussusception was seen with a duplication cyst as lead point [Table/Fig-1c]. Segmental resection of the ileum, caecum, and right-sided colon, including the duplication cyst was done, and gut continuity was restored by an ileocolic anastomosis. Postoperative stay in hospital was uneventful. Histopathological examination confirmed the lesion to be a duplication cyst [Table/Fig-1d]. The child was completely asymptomatic and having growth and development appropriate for his age at the eight-month follow-up visit.



[Table/Fig-1]: (a) USG using curvilinear 5 MHz transducer shows bowel within bowel appearance (target sign) in the right iliac fossa, suggestive of intussusception; (b) An anechoic cystic lesion is seen as the lead point of the intussusception with double-layered cyst wall (white arrows), indicating gut signature sign; (c) Gross image of the cut surface of the intestinal duplication cyst (thick yellow arrow) showing the unilocular cyst on the serosal aspect of the ileum (thick green arrow); (d) Histopathological section (Haematoxylin & Eosin, 400x) from the cyst wall showing columnar epithelium (black arrow) with focal gastric glands (red arrow), lymphocytes, and thin-walled capillaries in subepithelium along with a definite organised muscle coat (black asterisks).

DISCUSSION

Duplication of the gastrointestinal tract is a rare congenital malformation which was initially termed as Duplication of Alimentary Tract (DAT) by Fiorani C et al., [1]. Enteric duplication cysts are uncommon cystic structures present along the mesenteric border of the intestine. It can be seen in any part of the gut, however ileum is the most common site, accounting for 45% of cases. Other common sites include colon and stomach [2,3]. Two different types have been described: cystic (more common) and tubular. Duplication cyst shares a common muscular wall and blood supply with its adjacent bowel wall. In children, these duplications are considered to be benign lesions, but malignancy has been reported in adults [2,3].

The presentation of enteric duplications varies depending on the size, shape, location, and type of mucosa [1]. Although many cases of duplication cysts are detected on the antenatal scan, they may be asymptomatic and discovered accidentally at the surgery [4]. They may be minimally symptomatic and present with vague abdominal pain, constipation, or failure to thrive. They present as an emergency with acute intestinal obstruction due to sudden distension or intussusception [2]. Rectal bleeding is another emergency presentation when the ectopic gastric tissue in the epithelial lining bleeds. Duplication cysts may present at any age, from the foetus to the geriatric patient, but the majority (80%) of the patients present before two years of age [5]. Khan RA et al., reported seven cases of duplication cyst causing intestinal obstruction in neonates, and ultrasonography was a very useful modality in diagnosing these cases [6]. Paradiso FV et al., also reported a case of terminal ileal duplication cyst that presented with intussusception [7].

The radiological diagnosis of enteric duplications may be difficult, particularly in an emergency when associated with intussusception, as in the present case. Plain abdominal X-ray is non specific and usually shows the features of intestinal obstruction. On ultrasound, intussusception shows the characteristic bowel within bowel appearance [8]. However, ultrasound should not be stopped only at the diagnosis of intussusception but also to look further for the cause of the same. Intussusception in infant is often idiopathic and is assumed to be due to hypertrophied lymphoid tissue in the ileum. Nevertheless, it could be due to a mass lesion acting as a lead point. A lead point may be benign tumours, meckel's diverticulum, haemangioma, foreign body, ectopic pancreatic tissue, lipoma, as well as malignant tumours such as lymphoma [2,3,8]. Rarely, a duplication cyst can be the lead point, as in this case. It demonstrates an inner hyperechoic rim of the mucosal-submucosal tissues and an outer hypoechoic muscular layer [9].

Computed Tomography (CT) scan can help to localise the cyst and its relations with the contiguous organs. An enhancing rim of the tissue surrounding a fluid-filled cyst is diagnostic; it may also help to identify synchronous lesions if present [8]. However, further investigations were not done considering our patient's clinical condition.

Surgical treatment is advocated for all symptomatic duplication cysts [1,10]. Asymptomatic cysts discovered accidentally should also be resected to prevent future complications. Complete excision of the cyst and the adjoining intestine should be carried out because of the intimate attachment of the common wall and because isolated resection of the cyst would compromise blood flow to the adjacent intestinal segment. An alternative approach to its management may be considered that involves cyst marsupialisation, partial cystectomy, and mucosal stripping, particularly in extensive tubular duplication cysts, where excision may necessitate near-total colectomy in a child [2].

Histologically, most duplication cysts are lined by the mucosa native to the lesion, but ectopic tissue is present in 25-30% of the specimens [1,11]. In the present case, the cyst was lined by the intestinal type of mucosa. The most common type of ectopic tissue is gastric, followed by exocrine and endocrine pancreatic tissues. Peptic ulceration causing perforation or haemorrhage may occur in the duplications with ectopic gastric mucosa [1,8].

CONCLUSION(S)

Intussusception with duplication cyst as a lead point is a relatively uncommon entity usually present in the early period of life. It needs prompt diagnosis and early surgery to prevent further complications. Cyst wall showing gut signature on ultrasound examination is an important clue to its diagnosis. Enteric duplication cyst should be included in the differential diagnosis of cystic leading lesions for intussusception.

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