Recurrent Abdomen Pain and Vomiting in a Child: A Congenital Condition Rarely Reported - Ileal Duplication Cyst

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ABSTRACT

Gastrointestinal duplications are rare but interesting clinical entities. The presentation depends on their anatomical location, size and other characteristics. The most common variety is the small bowel cystic duplication. We report a case of a 13 year old male who presented with recurrent abdominal pain and vomiting. USG abdomen suggested a possibility of Meckel's diverticulum. Laparotomy revealed a duplicated segment of ileum of about 10cm. Duplicated segment was resected and end to end anastomosis was done. This report highlights ileal duplication though rare, but a possible cause of recurrent abdomen pain in children. The key in the management of a case of Alimentary duplication cyst is the preoperative suspicion and intraoperative examination of the entire bowel.

Keywords: Ileal duplication cyst, recurrent abdomen pain, variable presentation

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INTRODUCTION

Gastrointestinal duplications are rare clinical entities. They have a varied presentation, with most of them showing up in the paediatric population. Clinical features may vary from asymptomatic abdominal masses to bowel obstruction or perforation. This review traces the embryological origin and describes the anatomical types of duplications. An outline of the principles of management is described.

CASE REPORT

A 13 year old male presented with recurrent non bilious vomiting and abdominal pain since 15 days. Abdomen pain was more in right iliac region and dull aching. There was no history of fever or melena. The child had many such episodes in last 5 years. On examination, abdomen was soft, right iliac fossa tenderness was present and bowel sounds were present. X Ray abdomen erect appeared normal. USG abdomen revealed thick walled small bowel with diverticulum, suggesting a

possibility of Meckel's diverticulum (Figure 1). Computed Tomography of abdomen and radio isotope Technetium scan(Tc-99m) was advised but patient could not afford the same.

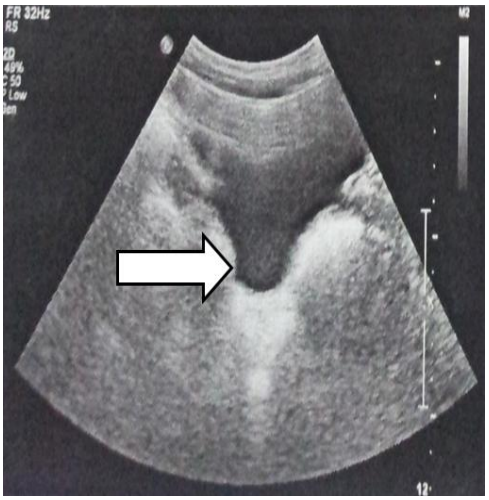

Figure 1: Ultrasound of abdomen showing a thick walled small bowel with diverticulum (arrow); suggesting a possibility of Meckel's diverticulum

Hence child was taken for laparotomy, which revealed a duplicated segment of ileum of about 10cm (Figure 2, 3). Duplicated segment was resected and end to end anastomosis was done. HPE was consistent with a duplicated segment of small bowel with ileal mucosa. The post operative period was uneventful and the child was discharged on 4th day.

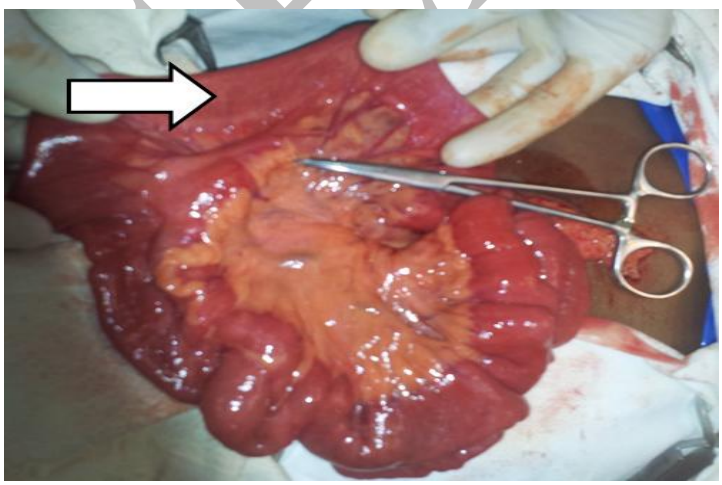

Figure 2 : Intra-operative photograph, showing the normal ileum (arrow) and duplicated segment of ileum (artery forceps)

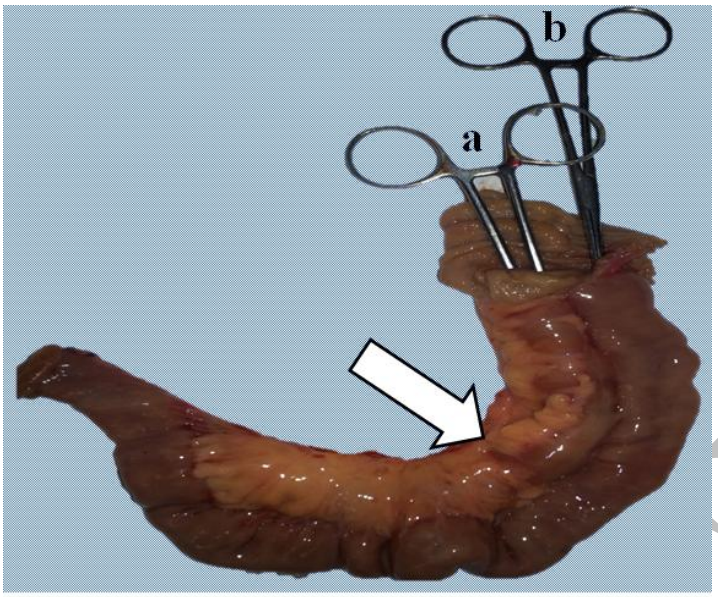

Figure 3: Postoperative photograph of the specimen, showing resected duplicated segment with the normal ileum (artery forceps-b) and duplicated segment of ileum (artery forceps-a) with one end terminating blindly (arrow)

DISCUSSION

Alimentary tract duplication was firstly reported by Fitz [1] and subsequently defined by Ladd et al [2] as a spherical- or tubular-shaped anomaly that was attached or adherent to and shared the identical phenotypic characteristics with the normal alimentary tract. Although, Alimentary tract duplication is a rare congenital malformation (1/10000 live births), this anomaly can occur anywhere along the gastrointestinal tract, with the ileum being the most frequently affected segment [3,4], either on the mesenteric margin or the contralateral side [5,6]. Daudet et al [7] reviewed 764 cases, the majority of which occurred at infancy but rarely at adulthood, with a male dominance.

Ileal duplication normally exhibits highly variable and nonspecific clinical manifestations in adults. The most frequent complaints are mainly of symptoms suggestive of gastrointestinal bleeding and intestinal obstruction [8,4,9,10] and abdominal pain. Palpable abdominal mass is also reported by approximately 50% of patients [9]. Furthermore, gastrointestinal bleeding and refractory abdominal pain may be underlain by heterotopic gastric mucosa lining the duplication cyst. Therefore,

antacids can be effective in relieving abdominal pain as was in this patient.

Multiple diagnostic tools are reported to be useful in the investigation of Duplicated alimentary tract cyst, including contrast-enhanced gastrointestinal ultrasonography and radiography, abdominal CT scan, and gastrointestinal endoscopy [10,12]. Moreover, Tc-99m pertechnetate scintigraphy is recommended as the first-line option of choice for the work-up of Duplicated alimentary tract cyst [13]. The positive result depends mainly on the abnormal enrichment of radionuclides accumulated by the heterotopic gastric mucosa. Ileal duplication cyst mainly needs to be differentiated from Meckel's Diverticulum. In pathogenesis, Meckel's diverticulum is a true congenital diverticulum derived from the remnant of the omphalomesenteric duct during the development of the terminal ileum, while Duplicated alimentary tract cyst can occur anywhere along the gastrointestinal tract but most frequently in the ileum. Meckel's Diverticulum is normally located on the contralateral side of the mesenteric margin, while ileal duplication cyst occurs either on the mesenteric margin or the contralateral side. Meckel's Diverticulum is often complicated with ectopic gastric mucosa; therefore, abdominal pain responsive to antacids is more frequently present in Meckels Diverticulum patients than in ileal duplication cyst patients. Furthermore, Meckel's Diverticulum is known to often cause a series of complications, such as diverticulitis, gastrointestinal bleeding or perforation, and intestinal obstruction, whereas these complications are relatively less common in ileal duplication cyst. However, it is almost impossible to distinguish Duplicated alimentary tract cyst from Meckel's Diverticulum prior to operation if the ileum is involved. A previous study conducted in Japan reported that only 11.2% of ileal duplication cases could be correctly diagnosed before operation, 18.2% misdiagnosed as ileal intussusception, 15.1% as ileal mass, 14.4% as ileus, and 26.7% as abdominal pain of unknown cause [14]. Therefore, it easily leads to the misdiagnosis of ileal Duplication cyst as Meckel's Diverticulum. As Meckel's Diverticulum is the most frequent gastrointestinal malformation, a suspected diagnosis of Meckels Diverticulum was made and indicated for exploratory laparotomy in our case. Appendicitis also needs to be excluded as the primary complaint was

right lower quadrant pain in this patient, especially if complicating infection occurs in the duplication cyst. The possibility of ileal duplication should be excluded for a patient diagnosed with suspicious Meckels Diverticulum or appendicitis but exhibiting gastrointestinal bleeding and/or intestinal obstruction, recurrent abdomen pain leading to frequent hospitalisation. Use of laparoscopy may be helpful in identifying any suspicious ileal diseases when a diagnosis of Meckels Diverticulum or appendicitis is doubtful.

Symptomatic treatment, such as acid-suppressing medications, may be effective in some cases, if the symptoms are primarily associated with ectopic gastric mucosa. Like the possibility of adenocarcinoma in Meckels Diverticulum with complicating ectopic gastric mucosa, malignant transformation of ileal duplication cyst with complicating gastric mucosa heterotopia is also a major concern in adult patients as epithelial instability is seen in long-standing duplication cysts. A historic review published by Johnson and his colleagues [8] reported that three out of 13 (23.1%) adult ileal Duplication cyst patients had ileal cancer, including adenocarcinoma in two patients and squamous cell carcinoma in one patient. Thus, radical resection of the duplication cyst along with the affected native intestinal segment remains the mainstay modality of definitive treatment [8].

Conclusion

In conclusion, the key in the management of a case of Alimentary duplication cyst is the preoperative suspicion and intraoperative examination of the entire bowel. Our report describes an ileal duplication cyst, a rare congenital gastrointestinal malformation uncommonly seen in adults. Surgical resection is the most effective treatment modality. The importance of this rare condition is that it exhibits no specific manifestations and are rarely suspected preoperatively so as to be looked into in the intraoperative period.

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