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CASE REPORT

Gastric Duplication Cyst Associated with Esophageal Atresia and Anorectal Malformations: A Rare Association

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ABSTRACT

Esophageal atresia (EA) and tracheoesophageal fistula (TEF) and anorectal malformations (ARM) in a newborn usually present with frothing of saliva and massive abdominal distension rendering it difficult to diagnose associated intrabdominal pathologies. We report a large gastric duplication (GD) cyst in a 2-day-old neonate with EA and TEF. GD cyst was detected in the early post-operative period as the abdominal distension persisted after repair of EA and TEF. The baby underwent excision of GD cyst with good outcome thereafter.

Key words: Anorectal malformations; Esophageal atresia; Gastric duplication cyst; Neonate

INTRODUCTION

Gastric duplications (GDs) are rare accounting for about 4% of all enteric duplications [1]. It should be kept as a differential diagnosis in an infant with non-bilious vomiting and can mimic hypertrophic pyloric stenosis. GDs are usually cystic, not communicating with the stomach, and located most commonly in the greater curvature [2]. GD cyst in a neonate usually presents with non-bilious vomiting, abdominal mass or distension, and failure to thrive. However, its association with esophageal atresia (EA) along with anorectal malformation (ARM) is a very rare entity and may present as a diagnostic dilemma. Herein, we report a case of GD cyst which was missed initially owing to massive abdominal distension in a case of EA and tracheoesophageal fistula (TEF) associated with ARM.

CASE REPORT

A 2-day-old preterm, low birth weight male child, presented with complaints of excessive frothing of saliva from mouth along with respiratory difficulty. The child had not passed stools since birth. There was gross abdominal distension. There was no history of cyanosis or jaundice. The baby was diagnosed to have EA and TEF as the red rubber catheter could not be passed into the esophagus beyond 10 cm from mouth which was further confirmed on X-ray findings (Figure 1). On perineal examination, there was absent anal opening along with meconium pearls suggestive of low ARM. Antenatal ultrasound was reported to be normal with adequate liquor. The child underwent right posterolateral thoracotomy through extrapleural approach, and TEF ligation and end-to-end esophageal anastomosis along with anoplasty were done. There was no evidence of any other anomaly in the chest. Postoperatively, the child recovered well but continued to have abdominal distension despite passage of stools. On abdominal examination, on the 3rd post-operative day, a cystic mass was incidentally detected. The abdominal mass was around 8 cm × 6 cm in size, transversely mobile, and also moves with respiration. All borders of the mass were well made out, and it was mainly occupying the upper abdomen. However, the child tolerated breastfeeds well, and there were no complains of vomiting.

Plain radiograph of chest with abdomen (Figure 1) done previously to confirm EA was revisited, which revealed an upper abdominal mass shadow displacing the surrounding bowel. Ultrasound abdomen was suggestive of a mesenteric cyst or a choledochal cyst. As the baby was asymptomatic, he was discharged on full breast feeds and daily anal dilatation. MRCP done after the discharge revealed a 7.2x6.2x5.1cm unlocu-

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lar cyst that was reported as mesenteric/ enteric duplication cyst (Figure 2).

After 10 days, the baby presented again in emergency with complaints of non-bilious vomiting along abdominal lump for which exploratory laparotomy was planned after initial stabilization. On exploration, a cystic mass of size 8 cm × 6 cm arising from the greater curvature of the stomach, consistent with a cystic duplication (Figure 3) was excised without gastric wall resection (mucosal sparing). A gastric lining in which all of the layers of the gastrointestinal tract had a typical appearance was identified on histology, confirming GD cyst (Figure 3). Heterotopic pancreatic tissue was also seen in the cyst wall lining. The baby was discharged on the 5th post-operative day after the second surgery on full breastfeeds. The baby is doing fine on follow-up.

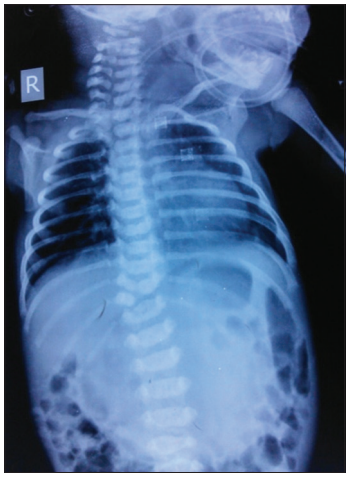

Figure 1: X-ray chest and abdomen showing red rubber catheter in blind upper pouch of esophagus along with mass per abdomen displacing the surrounding bowel loops

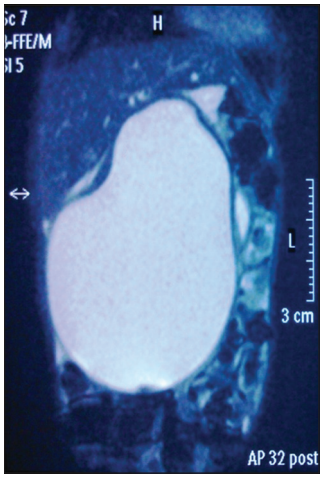

Figure 2: Magnetic resonance cholangiopancreatography (coronal view) suggestive of mesenteric cyst or less likely enteric cyst

DISCUSSION

GDs are usually cystic, non-communicating, and commonly located at the greater curvature of the stomach [1]. The pathological diagnostic criteria of GDs include lining with the gastrointestinal mucosa, attachment to the gastrointestinal tract, the presence of a smooth muscle coat, communicating with gastric lumen or not, and sharing blood supply with the stomach [1]. GDs present with vague abdominal pain, vomiting, and occasionally a palpable abdominal mass. They may remain asymptomatic or may present with complications such as hematemesis, melena, or perforation [2].

The etiology of enteric duplications remains hypothetical and many theories have been proposed, but it is mainly based on the split notochord theory which proposes that the abnormal separation of the notochord for the endoderm causes enteric duplications [3]. Spataru *et al.* found 17 cases of foregut duplication cyst associated with EA on literature search [4]. However, to the best of authors' knowledge, GD cyst along with EA and TEF, and ARM is a very rare association and has not been reported so far in the literature.

Ultrasonography and contrast-enhanced computed tomography (CECT) abdomen are done to investigate the nature and origin of the mass. Plain X-ray abdomen, done in our case, also suggested a mass

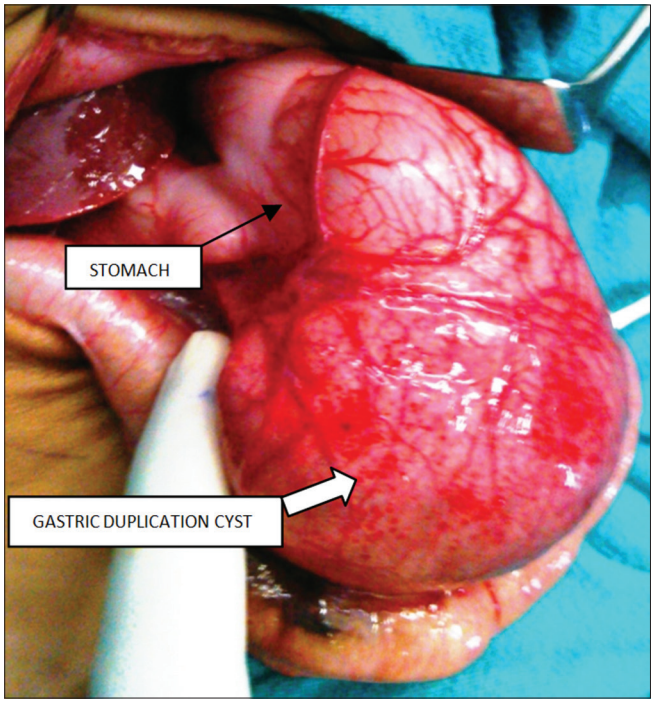

Figure 3: Intraoperative picture showing a large gastric duplication cyst (white arrow) arising from the greater curvature of the stomach (black arrow)

abdomen displacing the surrounding bowel loops although we missed that finding initially. Instead of CECT abdomen, an MRCP was done in our case as ultrasound was suggestive of a possibility of choledochal cyst.

Non-communicating GD cyst can be excised completely while, in a rare communicating variety, a segmental gastric resection is usually required. Further, in a large GD cyst, as in our case, after decompression, it can be excised while sparing the mucosa, thus avoiding segmental gastric resection. The common wall with the stomach is left intact, and ablation of the residual mucosa or mucosal stripping is done to prevent complications associated with the retained mucosa [3].

An antenatally diagnosed GD cyst should be carefully evaluated in postnatal period. As most of them are asymptomatic on presentation, surgical management of these duplication cysts may be done after the treatment of the associated emergent conditions as done in our case. Had an asymptomatic duplication cyst been diagnosed initially, even then we would have gone for only surgical management of EA/TEF and ARM initially.

In conclusion, concurrence of GD cyst with EA and ARMs is a very rare association. The diagnosis should be suspected in an infant where abdominal distension and non-bilious vomiting persist along with a palpable abdominal mass in the post-operative period.

Author's contribution

All authors contributed equally in concept, design, literature review, drafting the manuscript, and approval of the final manuscript.

Consent statement

Authors declared that they have taken informed written consent, for publication of this report along with clinical photographs/material, from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient however it cannot be guaranteed.

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