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Case Report

Gastric Duplication Cyst in Children: A Case Report and Literature Review

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Abstract

Objective: Gastric duplication cyst is rare. The symptoms and signs are nonspecific. Thus, preoperative diagnosis often becomes difficult. It should be differentiated from other possible cases of gastric outlet obstruction. Inspite of this, the prognosis has improved due to improvised operative techniques. We report a rare case of gastric duplication cyst which was successfully treated by mucosal tripping of the cyst wall.

Case Report: A 1-year 5-months boy presented with features of gastric outlet obstruction. A thorough preoperative investigation failed to reach the diagnosis. On exploration, a gastric duplication cyst was detected. It shared a common wall with the stomach and thus, complete excision was not possible. The major part of the cyst was excised. Rest of the cyst wall was treated by mucosal stripping to avoid major resection of the stomach. At 2-year follow up, there was no evidence of recurrences of the symptoms.

Conclusion: Gastric duplication cyst should be kept as a differential diagnosis while dealing with any case of gastric outlet obstruction in children. And when the cyst is not amenable for complete resection, mucosal stripping can be done safely to achieve the fruitful outcome.

Background

Gastric Duplication Cyst (GDC) constitutes (2%-7%) of all gastrointestinal duplications. Clinically, it is indistinguishable from pancreatic pseudocyst and cystic neoplasm of the pancreas [1]. The usual location of GDC is along the greater curvature of the stomach. Sometimes, the GDC share a common wall with the stomach. Complete resection of the cyst is the choice of surgery. Sometimes, it shared a common wall with the stomach and not amenable for complete resection. In these cases, mucosal striping is considered as an alternate method of treatment.

Case Report

A 1 year 5 months boy presented with non-bilious, non-projectile vomiting after feeding for two months. He had no history of abdominal trauma, jaundice or abdominal pain. The patient was dull, pale and having a poor cry. On examination, a nontender, cystic lump (8cm X 5cm) was detected in the epigastrium and left hypochondrium. Blood Hemogram and serum amylase/ lipase were normal. The Ultrasonography (USG) revealed an ill-defined partly cystic space occupying gut related lesion (12cm X 6.6cm) near the splenic flexure. Contrast Enhanced Computed Tomography (CECT) showed a large cystic lesion between the stomach and pancreas that raises the possibility of gastric duplication cyst or pseudocyst of pancreas (Figure 1). On exploration, there was no evidence of inflammation in the pancreas or peripancreatic fluid collection. A large cystic lesion was detected which was adherent to the posterior wall of the stomach and transverse Mesocolon. The cyst had no communication with the lumen of the stomach but it shared a common wall. Thus, complete resection of the cyst was not possible. The major part of cyst was excised. For the residual cyst wall which was sharing a common wall with the stomach, mucosal striping was done (Figure 2). Post-operative recovery was uneventful. Oral feeding was started on 2nd Post-Operative Day (POD) and the patient was discharged on 5th POD. Biopsy of the cyst wall showed gastric mucosa and smooth muscles, suggestive of gastric duplication cyst. At 2 years follow up, there was no recurrence and the patient was doing well.

Discussion

Foregut duplication is relatively rare congenital anomaly. But it may occur at any level from oral cavity to rectum. According to the embryonic origin, it may be esophageal, bronchogenic, and neuroenteric. About 50-70% of foregut duplication cysts are enterogenous and 7-15% is bronchogenic [2]. The duplication cyst is named on the basis of their site rather than the lining mucosa it possesses. GDC comprises less than 7% of all foregut duplication. Usually, GDC occurs along the greater curvature of the stomach. But, it may be found in the lesser curvature (5.5%), in the upper part of the stomach at the level of the cardia, in the anterior or posterior wall of the fundus and near the gastro esophageal junction [3].

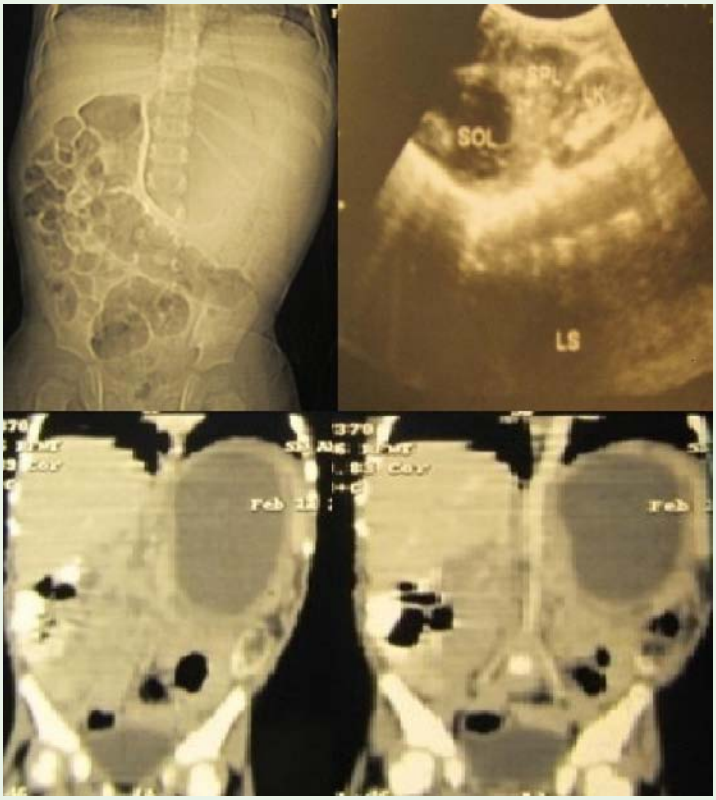

Figure 1: X-ray abdomen shows displacement of gut loops from left upper abdomen, USG and CECT of abdomen show a cystic lesion between stomach and pancreas with mass effects.

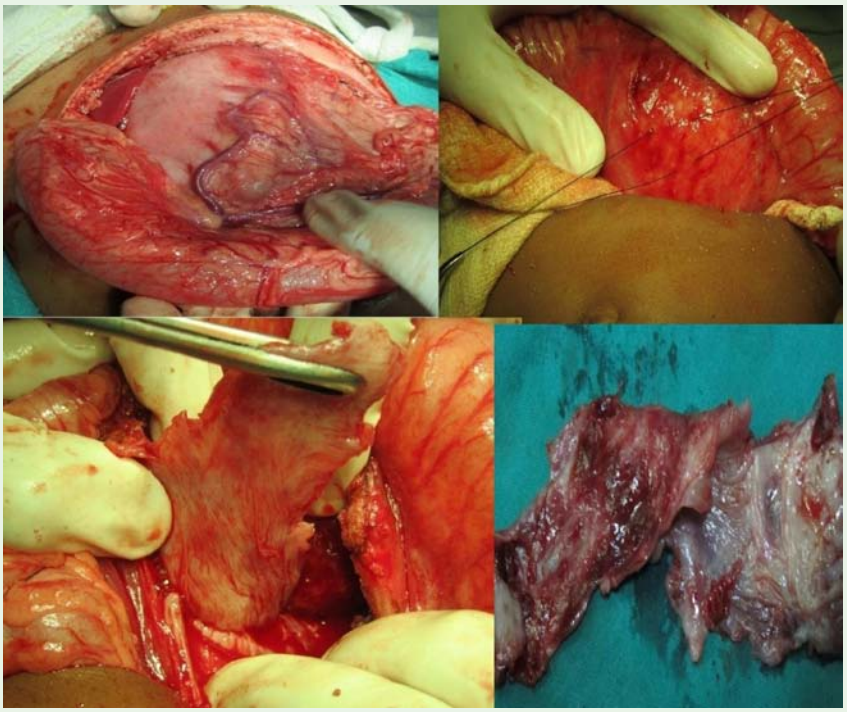

Figure 2: Intra-operative picture shows a cystic lesion adherent to stomach, the cyst was opened in between two stay sutures, the procedure of mucosal stripping and the specimen of stripped mucosa.

Several theories have been attempted to explain the exact pathogenesis. Bremer proposed the theory of aberrant luminal recanalization and fusion of longitudinal folds. According to McLetchieet al, a large GDC or GDC lying outside the stomach wall are caused by faulty endoderm and notochord separation. Other hypothesis is abortive twinning, persistent embryological diverticuli, and hypoxic or traumatic events. In about 50% cases, it may be associated with various other congenital anomalies like; esophageal diverticulum, alimentary tract duplications, genitourinary abnormalities, vertebral and spinal cord abnormalities (16-26%) [4]. GDCs are of two types; cystic (80%) or non-communicating and tubular (20%) or communicating. The presentations of GDC merely depend on the site of occurrence, size and type of the cyst and presence of ectopic mucosal lining. Usually, 67% cases of GDC become symptomatic within the first year of life. In younger infants, the symptoms are related to the mass effect leading to gastric outlet obstruction. Sometimes, it does simulate the other diseases, like pseudocyst of pancreas or cystic neoplasm of pancreas. The ectopic pancreatic tissue may be present in 10% cases which may lead to pancreatitis. Sometimes, there is a communication with the pancreatic ductal system that also gives rise to abdominal pain and/or pancreatitis [5]. A case of gastric duplication cyst simulating leiomyoma has been reported in literature. The GDC may be complicated by infection, gastrointestinal bleeding, perforation, ulceration, fistula formation, obstruction and compression due to mass effect, which may be the presenting symptoms, especially in adults.

The natural history of GDC is not clear. A series of investigations are engaged to clear out the diagnostic dilemma. A contrast x-ray may suggest an intramural filling defect indenting the gastric contour. The CECT demonstrates a thick-walled cystic lesion with enhancement of the inner lining with or without calcification which also may be found in mucinous cystic tumors of the pancreas. Importantly, the MRI does not provide any additional information over the CECT. The Endoscopic Ultrasound (EUS) is the investigation of choice for GDC as it delineates the cyst with an echogenic internal mucosal layer and a hypo echoic intermediate muscular layer which is specific for GDC [6]. The image-guided aspiration has little value in the diagnosis because; the cytological features of GDC are indistinguishable from the mucinous pancreatic neoplasms. Moreover, the nature of the cyst-fluid may be changed because of intra-cystic bleeding, chronic inflammation or infection.

Surgical intervention must be initiated soon after the provisional diagnosis of GDC. The fatal complications like; intracystichemorrhage, torsion, perforation and obstruction are common in long standing cases. A definite risk of malignant transformation has been described. Hence, complete resection of the cyst has been considered as a 'Gold standard' procedure. For communicating type, marsupialization and drainage procedure in the form of cystojejunostomy have been suggested [7]. A risk of ulceration is there inmarsupialization procedure. Whereas, anastomotic stenosis and blind loop syndrome are the common in cystojejunostomy.

Non-communicating cyst is treated by complete excision without violation of the gastric lumen whenever possible. In difficult cases, where the cyst is practically sharing a common wall with the stomach, the "Wrenn" method of mucosa stripping can be performed safely even in tubular variety [8]. Although, multiple incisions are necessary for this method. But, the procedure definitely reduces the risk of future malignancy. Recently, a few cases of laparoscopic resection and laparoscopy assisted resection of the GDC have been also reported. In accordance to the literature review, the laparoscopy is gaining more demand for the treatment of foregut duplication cyst.

Conclusion

The clinical behavior of GDC is unpredictable. Moreover, it is often difficult to choose the proper surgical procedure, especially when major part of the stomach is involved. Mucosal tripping is an effective operative technique which can be done safely in cases of GDC which are not amenable for surgical resection.

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