Gastric duplication cyst: A rare congenital anomaly

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ABSTRACT

Gastrointestinal (GI) duplication is defined as a spherical structure, with a muscular coat lined by a mucous membrane in the alimentary tract is a rare congenital anomaly. In this report, we present a case of a 14-year-old male child with complaints of epigastric pain for 1 month. Computed tomography scan of the abdomen was suggestive of two cystic lesions in the stomach. Exploratory laparotomy was performed with excision of cysts. Histopathology of both cysts was suggestive of gastric duplication cyst.

Keywords: Abdomen, Congenital anomalies, Gastric duplication cyst, Gastrointestinal duplication

Gastrointestinal (GI) duplications are rare congenital malformations that may vary greatly in presentation, size, location, and symptoms. A GI duplication is defined as a spherical structure, with a muscular coat lined by a mucous membrane [1]. They are a rare phenomenon and account for only 4% of all GI duplications [2]. About one-third of patients with gastric duplication cysts (GDCs) also have other congenital anomalies such as the annular or heterotopic pancreas, or vertebral anomalies such as spina bifida [3]. They can be found anywhere in the stomach, with the majority being located on the greater curvature.

GDCs are rarely diagnosed in the adult population and occur more commonly in young children, who may present with symptoms of abdominal pain, gastric outlet obstruction, or a palpable abdominal mass. Accurate diagnosis of these cysts before resection is difficult. Differential diagnoses are varied, including GI stromal tumors, neuroendocrine tumors, pancreatic heterotopia, pancreatic pseudocysts, and neurogenic tumors. Malignant transformation of a GDC is very rare [4,5].

CASE REPORT

A 14-year-old male child presented to the department with a chief complaint of recurrent episodes of pain in the epigastric region for 1 month. There was no associated history of vomiting, hematemesis, melena, and weight loss.

The patient was afebrile and vitally stable. The abdominal examination was non-tender and did not reveal any palpable mass. He did not have any congenital anomalies or any other significant history.

The serum amylase was 20 units/L and serum lipase was 94 units/L. The rest of the hematological workup was also within normal limits.

The ultrasonography was suggestive of a well-defined complex cystic lesion of size 7.3 cm \times 6.4 cm in the splenorenal pouch. The lesion showed multiple thin septations and debris within, likely to be a pseudocyst of the pancreas.

Contrast-enhanced computed tomography (CT) of the abdomen demonstrated a cystic lesion of size $6 \text{ cm} \times 8.2 \text{ cm} \times 6.8 \text{ cm}$, presented at the superior aspect of the fundus of the stomach, and seen along the posterosuperior aspect of gastroesophageal junction (Fig. 1). The cyst at the fundus of the stomach was not communicating with the gastric lumen. There was another small cystic lesion of size $1.6 \text{ cm} \times 2.8 \text{ cm} \times 2.9 \text{ cm}$ along the anterior wall at the antral region, which was also not communicating with the gastric lumen (Fig. 2). A differential diagnosis of pseudocyst of the pancreas or GDC was made.

The patient underwent an exploratory laparotomy. A roof-top incision was given and abdomen was opened. On aspiration of the larger cyst (Fig. 3) presents at the fundal region, mucinous fluid was demonstrated. This cyst was excised leaving a seromuscular sleeve attached to the greater curvature of the stomach, of which mucosal stripping (Fig. 4) was done. The sleeve was later approximated and sutured.

On histopathological examination of the cystic mass on the posterosuperior wall showed a single layer of mucin-secreting cells. At places, gland formation was seen. Underlying tissue composed of smooth muscle and loose fibrovascular tissue.

A wedge resection of the stomach was done along with the cyst present on the anterior aspect of the stomach (antrum) (Fig. 5) and the histology of this cyst composed of a varied type of epithelium varying from gastric type to ciliated columnar epithelium. Subepithelial tissue showed fibroconnective tissue. The histological features of both cysts were suggestive of a GDC of an anterior and posterior wall of the stomach.

The patient was started on an oral diet on post-operative day 3 and discharged on the 5th post-operative day. He has been on regular follow-up and has gained around 9 kg in 6 months postoperatively.



Figure 1: Computed tomography scan showing a cyst in the antral region



Figure 2: Computed tomography scan showing cyst in the fundal region

DISCUSSION

GI duplication is a relatively rare anomaly that may occur at any level from the oral cavity to rectum with ileum being the most common site. The duplication cyst is entirely separated from the adjacent bowel but shares a common wall; they are mostly non-communicating [6]. Its pathogenesis is said to be abnormal recanalization after the solid epithelial stage of embryonic bowel development [7]. GDCs are usually located along the greater curvature. GDCs comprise 4% of all GI duplications [6].

There have been several studies conducted on this topic including case reports by Yamasaki A *et al.* from Cincinnati, USA, in 2016 [7] and Kim *et al.* from Seoul, Korea, in 2015 [8]. The first case report of a GDC was published in 1911 by Wendel [8]. The essential criteria for the diagnosis of a GDC are that the wall of the cyst is contiguous with the stomach wall, it is lined by the epithelium of gastric or any other type of gut mucosa and the cyst is surrounded by smooth muscle, which is continuous with the muscle of the stomach [9].

Various other congenital anomalies such as alimentary tract duplications, esophageal diverticulum, or spinal cord



Figure 4: Mucosal stripping of the excised cyst

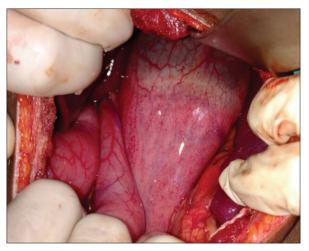


Figure 3: Gastric duplication cyst in the posterior-superior region of stomach (fundal region)



Figure 5: Gastric duplication cyst on the anterior wall of stomach (antral region)

abnormalities are encountered in up to 50% patients of gastric duplication. >80% of gastric duplications are cystic and do not communicate with the lumen of the stomach. The remainders are tubular with some communication. The mucosal lining of duplication may be histologically similar to the segment of the gut to which it is topographically related. However, some duplication may include lining from other segments of the alimentary or respiratory tract [6].

Gastric duplications typically become symptomatic during childhood. Among them, 67% are diagnosed within the 1st year of life, and <25% are discovered after the age of 12 [9]. The duplication cysts of the stomach are usually diagnosed intraoperatively in adults [10]. Our patient presented with the complaint at the age of 14.

The clinical presentation of GDC can be highly variable and non-specific ranging from vague abdominal pain to nausea, vomiting, epigastric fullness, weight loss, anemia, dysphagia, dyspepsia with abdominal tenderness, and epigastric mass on physical examination [2]. The cysts can compress the adjacent organs such as the pancreas, kidney, spleen, and adrenal gland [11]. The cysts may also be manifested by complications such as infection, GI bleeding, perforation, ulceration, fistula formation, obstruction, compression, or carcinoma arising in the cysts. Up to 10% of gastric duplications may contain ectopic pancreatic tissue which may lead to pancreatitis and mimic a pancreatic pseudocyst [12].

Duplication cysts have the potential for neoplastic transformation. Of 11 reported cases of malignancy arising within the duplication cysts, eight were adenocarcinomas. Five of the carcinomas originated from gastric duplications. Adenomyoma arising from a gastric duplication has also been reported [6].

Although it is difficult to diagnose GDC preoperatively, recent imaging modalities have provided some informative findings. CT scan and endoscopic ultrasound (EUS) are the best ways to identify GDC. EUS is useful in distinguishing between the intramural and extramural lesions of the stomach. The role of EUS-guided fine-needle aspiration in GDC is uncertain because (a) the cytological features of GDC may closely resemble those of mucinous pancreatic neoplasms and (b) GDCs with elevated levels of carcinoembryonic antigen and CA19-9 have been reported, mimicking mucinous pancreatic neoplasms [4,13].

Multiple treatment modalities have been reported in the literature including enucleation, formation of cystgastrostomy, minimally invasive laparoscopic excision, and even endoscopic removal. Complete removal is the treatment choice to avoid the risk of possible complications such as obstruction, torsion, perforation, hemorrhage, and malignancy [2]. A GDC is classically

treated by complete excision of the cyst and resection of the shared wall between the stomach and the duplication cyst [12].

CONCLUSION

A GDC is a rare congenital anomaly. The diagnosis of GI tract duplications may be done by imaging studies; more often, the correct diagnosis is not established before surgery. Due to the risk of malignant transformation and other complications, GDC should be treated surgically by complete resection.

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