



Gastric duplication cyst with hemoptysis: an unusual presentation

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Abstract

Gastrointestinal duplications are rare developmental anomalies that may occur at any level from the oral cavity to the rectum, with the ileum being the most common site. Gastric duplications are rare gastrointestinal duplications. Various theories have been proposed for the development of duplication cysts; however, there is no single theory that explains all types of duplications. Complete removal is the treatment of choice to avoid the risk of possible complications and malignant transformation. Most cases present within the first year of life. The clinical presentation of gastric duplications can be highly variable; however, they usually present with abdominal pain and a lump. Clinical presentation with hemoptysis is rare. Only one such case has been reported previously. We describe a three-year-old male with gastric duplication who presented with hemoptysis.

Keywords: Duplication, gastric, hemoptysis

Introduction

Gastrointestinal duplications are rare developmental anomalies. Gastric duplications are even rarer. The usual presentation is abdominal pain and a lump. Presentation with hemoptysis or chest symptoms is extremely rare. Careful preoperative imaging can preclude thoracotomy with lobectomy. We report a rare case of a gastric duplication cyst presenting with hemoptysis.

Case

A three-year-old male child with sickle cell trait presented with vague abdominal pain, recurrent cough, and multiple episodes of hemoptysis over a one-year period. His vitals were stable. Air entry was decreased in the left lower zone. His abdominal examination was unremarkable.

The patient was anemic. The coagulation profile was normal. Multiple chest X-rays were suggestive of persistent consolidation in the lower lobe of the left lung (Figure 1). Computed tomography (CT) of the chest and abdomen revealed left lower lobe consolidation suggestive of sequestration (Figure 2). A 2.4x1.5x2.2-cm cystic structure was visualized in the lesser sac extending up to the left hemidiaphragm, raising suspicion of a cyst arising from the pancreas. Magnetic resonance cholangiopancreatography (MRCP) revealed a normal pancreas with a cyst in the lesser sac (Figure 3). Suspecting a cystic lesion in the left upper abdomen, the patient was explored. At laparotomy, an elongated cystic structure was seen in close proximity to the upper part of the greater curvature sharing a common wall with the stomach (Figure 4, 5). There was no communication between the stomach and the cavity of the cyst. The cyst extended upwards up to the left hemidiaphragm but was blind ended. The cyst was excised completely.

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Figure 1. Pre-operative chest X-ray showing consolidation in the left lower lobe



Figure 3. Magnetic resonance cholangiopancreatography showing a normal pancreas with a cyst in the lesser sac

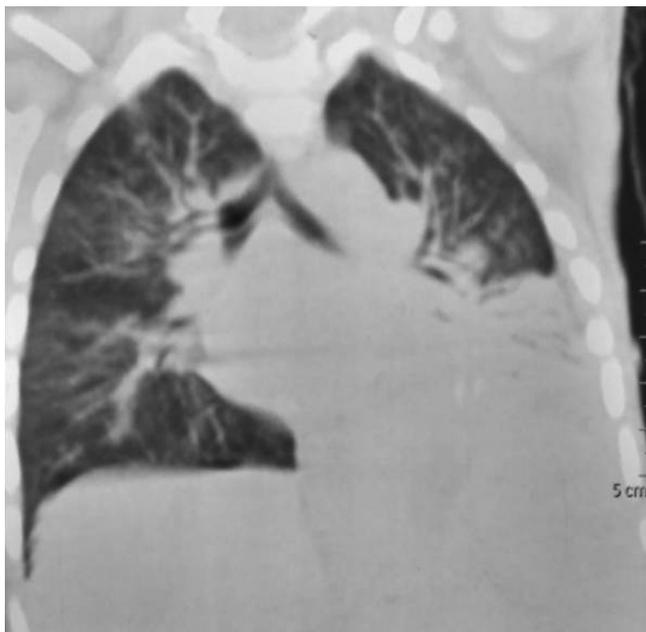


Figure 2. Computed tomography of the chest and abdomen showing left lower lobe consolidation suggestive of sequestration

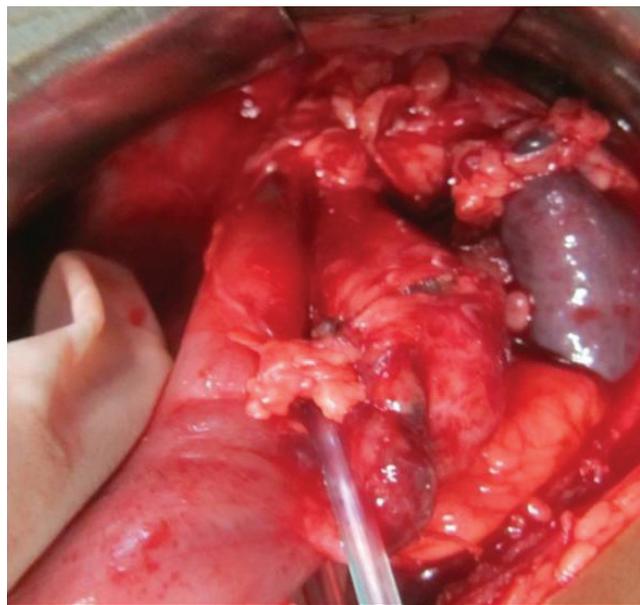


Figure 4. Intra-operative image showing an elongated cystic structure in close proximity to the upper part of the greater curvature sharing a common wall with the stomach

Histopathology was suggestive of gastric duplication cyst with lymphocytic infiltrate in the mucosa.

The patient had an uneventful post operative recovery. He remains asymptomatic and the postoperative X-ray shows resolution of the consolidation (Figure 6). Written informed consent was obtained from the patient's family.

Discussion

Gastrointestinal duplications are a rare developmental anomaly that may occur at any level from the oral cavity to the rectum, with the ileum being the most common site. Duplication cysts of the stomach are even rarer, comprising 4% of all gastrointestinal du-

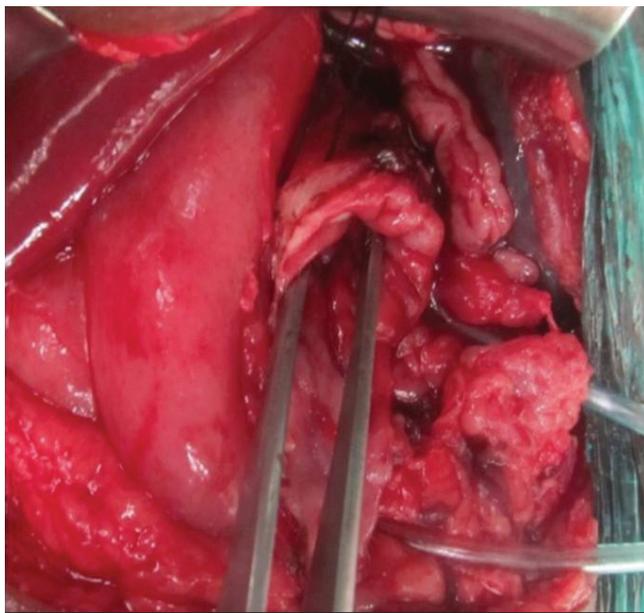


Figure 5. Intra-operative image showing the lumen of the cyst, but no communication with the stomach

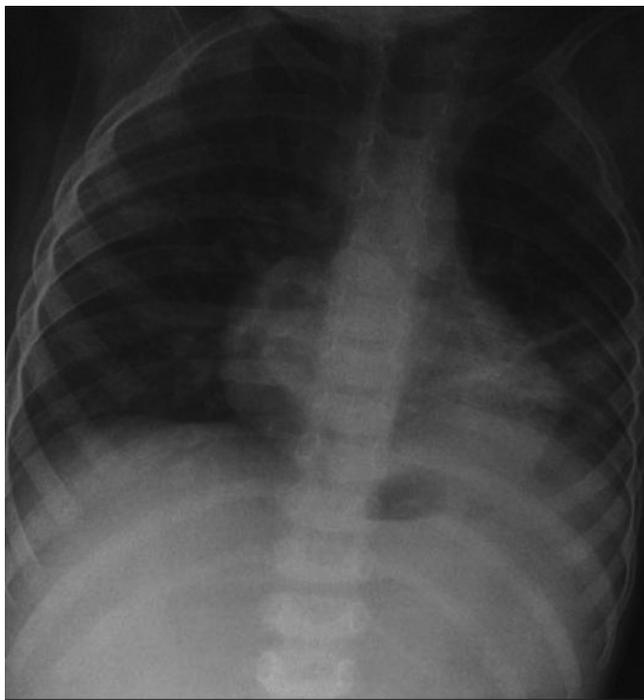


Figure 6. Post-operative chest X-ray showing resolution of the consolidation

plications (1). The usual location for gastric duplication cysts is along the greater curvature (2). The essential criteria for the diagnosis of a gastric duplication cyst are - (a) the wall of the cyst is contiguous with the stomach wall; (b) the cyst is surrounded by smooth muscle, and (c) the cyst wall is lined by epithelium of gastrointestinal mucosa (1). More than 80% of gastric duplications are cystic and do not communicate with

the lumen of the stomach. The rest are tubular with gastric communication (1).

Various theories have been proposed for the development of duplication cysts. However, there is no single theory that explains all types of duplications. Sixty-seven percent of gastric duplications are diagnosed within the first year of life, and less than 25% are discovered after the age of 12 years (1).

The clinical presentation of gastric duplication cysts can be highly variable ranging from vague abdominal pain, nausea, vomiting, epigastric fullness, weight loss, anemia, dysphagia, dyspepsia with abdominal tenderness, and epigastric mass on physical examination. The cysts can potentially compress the adjacent organs such as pancreas, kidney, spleen, and adrenal gland because most cases occur along the greater curvature of the stomach. The cysts may also be manifested by complications such as infection, gastrointestinal bleeding, perforation, ulceration, fistula formation, obstruction, compression, or carcinoma arising in the cysts (1). Less than 10% of gastric duplications may contain ectopic pancreatic tissue (3). There are many clinical and radiologic similarities between gastric duplication cysts and pancreatic pseudocysts, but normal levels of pancreatic enzymes and no history of pancreatitis may suggest a duplication cyst (4).

The primary presentation of a gastric duplication cyst with hemoptysis is extremely rare. Only one such case was reported previously by Menon et al. (5), a two-year-old child who presented with hemoptysis. The proposed possible etiology was an associated lung lesion such as a sequestration, associated thoracic duct duplication cyst ulcerating into lung tissue or direct communication through a fistulous tract. Bronchopulmonary foregut malformations commonly involve the left lower lobe and can present with hemoptysis. However they are not known to be associated with gastric duplications. The regression of the lung consolidation after excision of the cyst alone could be due to ulceration leading to lung erosion through a diaphragmatic communication.

It is difficult to outline the natural history of gastric duplications with certainty because of their rarity. As with the native gastric mucosa, the cyst lining may undergo erosions, ulceration, and regenerative changes. Duplication cysts have the potential for neoplastic transformation (1). Although it is difficult to diagnose these cysts preoperatively, recent imaging modalities can provide

some informative findings. Classically, radiographic studies show an intramural filling defect indenting the gastric contour (1). Contrast-enhanced CT scans can demonstrate them as thick-walled cystic lesions with enhancement of the inner lining. Complete removal is the treatment of choice to avoid the risk of possible complications and malignant transformation (1).

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