Journal of Pediatric Surgery 53 (2018) 189–191

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Pediatric Surgical Images

Accessory pancreatic lobe in association with a gastric duplication cyst

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ARTICLE INFO

Article history:
Received 19 April 2017
Received in revised form 25 August 2017
Accepted 11 September 2017

Key words:
Accessory pancreatic lobe
Pancreatitis
Gastric duplication cyst
Enteric cyst

ABSTRACT

Gastric duplication cysts are an extremely rare anomaly with few reported cases in association with accessory pancreatic tissue. Diagnosis can be challenging given a presentation of recurrent pancreatitis and resemblance to pancreatic pseudocysts. We report the case of a 6-year old boy with multiple episodes of pancreatitis who was discovered to have an accessory pancreatic lobe connected to a gastric duplication cyst, successfully treated with surgical excision.

Enteric duplication cysts are uncommon, present in 1 out of every 4500 live births [1]. Duplication cysts of the alimentary tract can occur anywhere from the oropharynx to rectum and are named for the organ with which they are associated. Gastric duplication cysts represent 3.8% of all gastrointestinal duplications and the majority manifest within the first year of life [2]. Depending on the anatomical location and size, these lesions can range from asymptomatic to causing abdominal pain, nausea and vomiting with resemblance to pancreatic pseudocysts. We report the case of a 6-year old boy with recurrent pancreatitis who was discovered to have a gastric duplication cyst on the posterior stomach wall connected to an accessory pancreatic lobe, successfully treated with surgical excision.

1. Presentation

A 6-year old boy, with a past medical history of *Helicobacter pylori* gastritis three years prior, presented to an outside hospital with a three month history of abdominal pain, associated with meals, that was increasing in intensity. He was diagnosed with acute appendicitis and underwent an uneventful laparoscopic appendectomy. However, pathologic analysis of the appendix revealed no acute inflammatory process. On postoperative day two, the patient experienced nausea and intense epigastric pain with radiation to the back worsened by eating. The serum lipase of the patient was elevated at 1412 IU/L consistent with pancreatitis and the patient was treated with intravenous fluids and a low-fat diet and discharged several days later. One month following this episode, the patient once again suffered from intense epigastric pain with elevated pancreatic enzymes and underwent a CT scan that revealed a 1 cm pseudoaneurysm with surrounding hematoma anterior to the pancreatic head and adjacent to the gastroduodenal artery. The patient underwent coiling of a gastroduodenal artery branch involving the pseudoaneurysm. The serum lipase and amylase normalized and the patient was discharged on postoperative day three. Yet, six weeks after the coiling procedure, the boy once again suffered from intense abdominal pain after consuming fatty foods. At this time, a work-up for cystic fibrosis and other genetic studies were performed which were all found to be negative. He was treated conservatively, and over the course of the following two months had continued bouts of epigastric pain and documented pancreatic enzyme elevations.

After his sixth episode of pancreatitis, the patient was transferred to our institution with an initial serum lipase of 2985 IU/L and serum amylase of 241 IU/L. Physical examination was unremarkable, but the patient's parents endorsed a 30 lb weight loss over the past year. An MRI of the abdomen was significant for accessory pancreatic parenchyma measuring 1.3 × 2.7 × 1.8 cm originating from the midpancreatic body with ventral extension (Fig. 1). Additionally, a 1.9 × 2.2 × 2.6 cm well-marginated cystic lesion near the pancreatic head and 2nd part of the duodenum was present with a differential diagnosis of an enteric duplication cyst versus a pancreatic pseudocyst (Fig. 2). An ultrasound was performed for further evaluation of this cystic lesion, which failed to demonstrate gut signature (Fig. 2). Additionally on the MRI, a

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https://doi.org/10.1016/j.jpedsurg.2017.09.007
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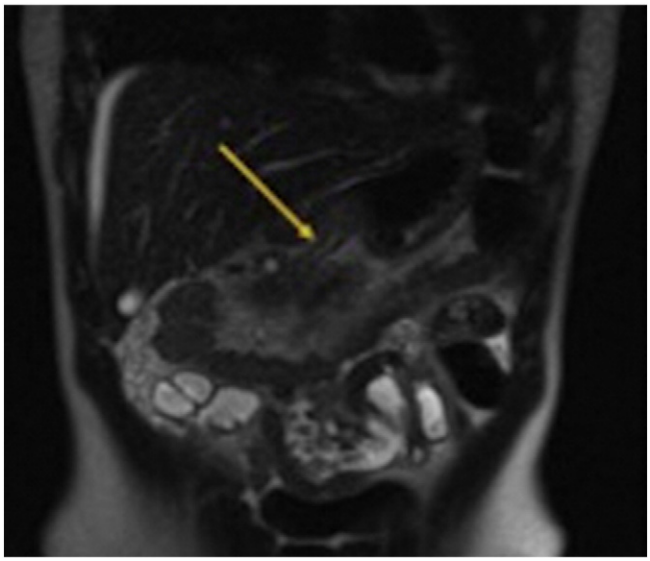

Fig. 1. Coronal T2-weighted single-shot turbo spin-echo image shows pancreatic parenchyma (arrow) more ventral than the expected location of normal pancreas. Not shown, the accessory pancreatic lobe and normal pancreas are continuous with each other on contiguous slices.

persistent gastroduodenal artery pseudoaneurysm was present. Given the multiple lesions present, a plan was formulated to first ameliorate the pseudoaneurysm and once stable, surgically remove the accessory pancreatic tissue and cystic lesion. The patient underwent coiling of a 1.6 cm pseudoaneurysm of the gastroduodenal artery. The patient tolerated the procedure well and was discharged on postoperative day eight.

Two weeks following discharge, the patient was taken to the operating room for a planned laparoscopic, possible open, resection of the accessory pancreatic tissue and cystic structure. Following exposure of the greater omentum, a 2×3 cm cystic mass was discovered attached to the posterior wall of the stomach as well as to the pancreas (Fig. 3). Owing to the limited exposure and adjacent adhesions from the previous pseudoaneurysm and bouts of pancreatitis, the case was converted to an open approach. The cyst wall was contiguous with the posterior stomach wall, over an approximate 1 cm in diameter area but the lumens did not communicate. The involved stomach wall was then repaired with interrupted Lembert sutures. Next, the accessory pancreas, which was in continuity with the cyst, was transected from its connection to the body of the pancreas with a vascular GIA stapling device (Fig. 4).

The patient tolerated the procedure and was discharged after a five day hospitalization with no signs of recurrent pancreatitis upon follow up. Pathologic examination of the surgical specimen confirmed the diagnosis of a gastric duplication cyst and accessory pancreatic lobe.

2. Discussion

Duplication cysts are a rare anomaly, most commonly arising from the ileum and least commonly from the stomach. The most frequent location of a gastric duplication within the stomach is the greater curvature (66%) and second most common is the posterior wall (14%) [3]. Gastric duplication cysts may communicate with the gastric lumen as well. The majority are diagnosed within the first year of life and less than one quarter are diagnosed after the age of 12 [4]. Presentation is highly variable, depending on the size and location of the lesion and can range from asymptomatic to causing nonspecific symptoms of abdominal pain, nausea, vomiting, weight loss, dysphagia or present as an epigastric mass on examination [4]. Additionally, in 50% of cases, other anomalies may be evident such as gastrointestinal tract duplications, esophageal diverticulum or spinal cord abnormalities [5]. Diagnostic criteria for gastric duplications include: (1) a cyst wall that is contiguous with the stomach wall, (2) the cyst is surrounded by smooth muscle which is continuous with stomach muscle, and (3) the cyst wall is lined by either gastric epithelium or any other type of gut mucosa or respiratory tract epithelium [6].

An accessory pancreatic lobe involves pancreatic tissue arising from the pancreatic gland with an aberrant duct. The anomaly has been associated with gastric duplication cysts as the irregular duct may communicate with both the main pancreatic duct and the duplication cyst. Traverso et al. reported that patients with gastric duplication cysts contiguous with the stomach tend to have recurrent episodes of pancreatitis [7]. This is believed to be owing to obstruction of the accessory lobe pancreatic duct by mucus secretion, biliary sludge or ulcer bleeding. This likely led to this patient's symptomatology as well as the pseudoaneurysm of the gastroduodenal artery which was prone to injury by the vessel's proximity to the chronically inflamed accessory pancreatic lobe. A recent literature review revealed that since the first reported case by Bradbeer in 1959, only 18 cases of an accessory pancreatic lobe in association with a gastric duplication cyst have been reported, with a strong female predilection [8]. Although not present in this case, up to 10% of gastric duplications contain ectopic pancreatic tissue leading to pancreatitis with resemblance to a pancreatic pseudocyst [5,9]. Multiple theories have been proposed, but the embryologic cause for the development of a gastric duplication cyst and an accessory pancreatic lobe is unknown.

Multiple imaging modalities may assist with the diagnosis of a gastric duplication cyst. On magnetic resonance imaging, duplications are typically well-circumscribed, homogeneous, and hypointense lesions on T1-weighted images and hyperintense on T2-weighted images [10]. This is variable as demonstrated in our case in which the lesion was T1 hyperintense. T1- and T2-hyperintensity suggests a combination of fluid with protein and/or hemorrhage, findings which can be seen with a pseudocyst and a duplication cyst. On sonography, the combination of an echogenic inner mucosa with a hypoechoic outer muscular layer is suggestive of gut signature and therefore a gastric duplication

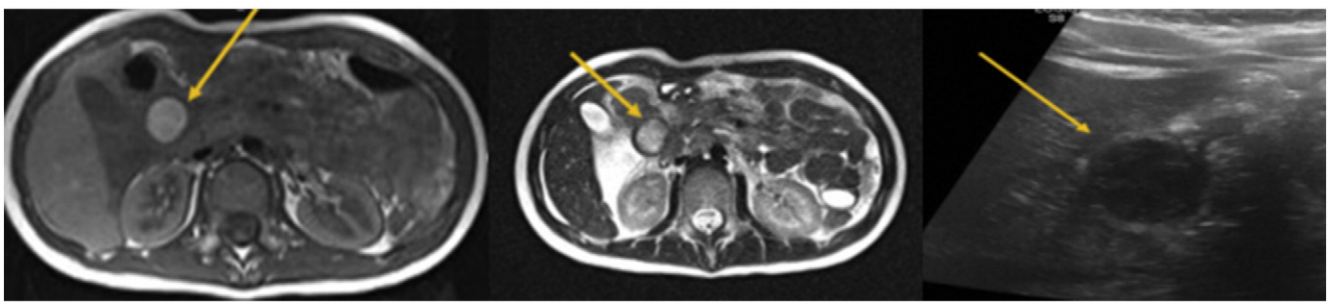

Fig. 2. T1- and T2-weighted turbo spin-echo MR images (left and middle) of a cystic lesion near the descending duodenum. Ultrasound image (right) shows a hypoechoic cystic lesion with low-level internal echoes and a thickened wall without typical gut signature.

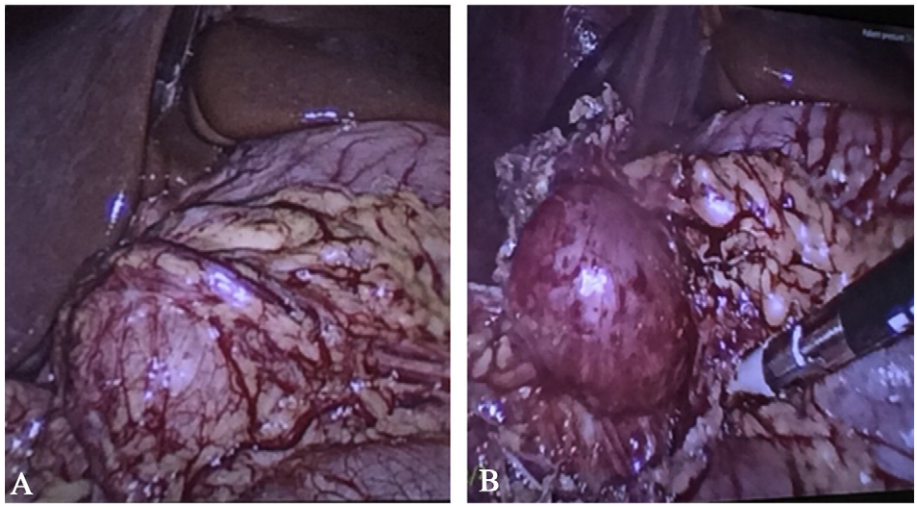

Fig. 3. A. Laparoscopic view of the gastric duplication cyst upon initial visualization. B. Laparoscopic view of the duplication cyst after dissection of the greater omentum.

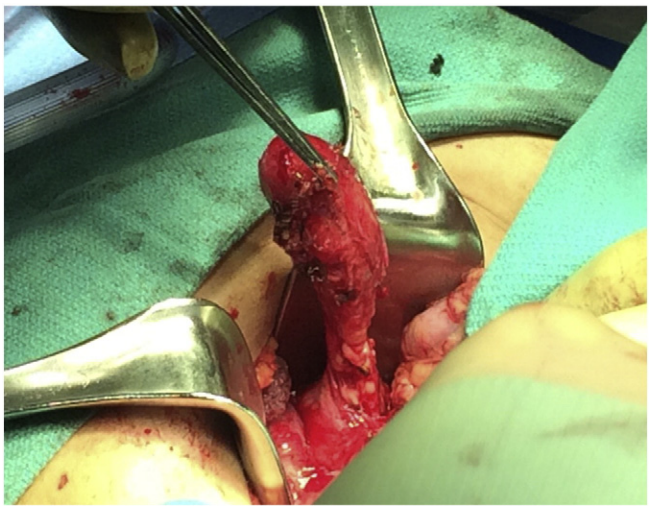

Fig. 4. Gastric duplication cyst, detached from the stomach, with the accessory pancreatic lobe in continuity with the middle body of the pancreas before transection.

cyst [11]. This patient's lesion was thick and single-layered on ultrasound, which would suggest a pseudocyst; however, gut signature can be obscured by inflammation as was the case with this cyst. The recognition of accessory pancreatic tissue raised the level of suspicion for a duplication cyst. Of note, the cyst and the accessory pancreatic tissue were not visualized on the previous computed tomography performed at the outside hospital. This was performed earlier in the patient's disease course and at that time, perhaps the cyst was draining into the stomach and was not obstructed and therefore not visualized. The accessory pancreatic tissue may also have been obscured by the inflammation from the pancreatitis as well as the pseudoaneurysm.

Once a correct diagnosis is made, management involves local excision of the gastric duplication with removal of the accessory pancreatic lobe [12]. This treatment is based on limited evidence in the literature of this rare association.

3. Conclusion

Gastric duplication cysts are an unusual phenomenon and association with an accessory pancreatic lobe is even rarer. These anomalies have been associated with multiple episodes of pancreatitis and may cause diagnostic difficulty owing to their resemblance to pancreatic pseudocysts. When involving the stomach, surgical excision is the treatment modality of choice for both the accessory pancreatic tissue as well as the gastric duplication cyst.

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