CLINICAL VIGNETTE

Late Complication of a Rare Type of Gastric Duplication Cyst

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A retired 76-year-old male radiologist with a history of hypertension presented to an emergency department for syncope associated with melena. The patient has been asymptomatic prior to presentation. He was found to have a hemoglobin which dropped from 11.3 g/dL down to 8.4 g/dL and was transfused 2 units of packed red blood cells. Upper endoscopy showed the presence of blood and a single gastric ulcer with extrinsic compression from a large mass in the fundus. Computed tomography (CT) of the abdomen showed a large 12 cm cyst at the gastric fundus with small high-density material within the gastric lumen adjacent to the large cystic mass (Figure 1). He underwent percutaneous drainage of the cyst performed by interventional radiology, and 800 ml of brown-tan colored cloudy fluid was aspirated. Cytology of the fluid showed abundant necrotic cellular material, a few histiocytes and acute inflammatory debris. The patient remained hemodynamically stable and was discharged when he did not have any recurrent signs of active bleeding. Repeat CT of the abdomen one month later showed the recurrent stomach cyst.

The patient underwent an elective laparoscopic excision of the cyst without any complications. Pathology showed a benign 10.5cm foregut cyst which was unilocular and attached to the serosal aspect of the stomach. Sections of the cyst wall showed benign epithelial lining with pseudostratified columnar cells with cilia, attenuated cuboidal cells and squamous epithelium (squamous metaplasia) at different areas. Goblet cells were not identified. There are large areas of denudation of the cyst lining where granulation tissue with dense mixed inflammatory cells and hemosiderin deposition and associated fibrosis. The gastric mucosa shows features suggestive of proton pump inhibitor effect. No H.pylori organisms are identified. No intestinal metaplasia, dysplasia or malignancy is seen.

Gastrointestinal duplication cysts are rare congenital abnormalities characterized by an epithelial lining and attachment to some part of the gastrointestinal tract. They are surrounded by at least one layer of smooth muscle. They can affect any part of the gastrointestinal tract but primarily involve the ileum, esophagus, colon and stomach.[1] While the ileum is the most common, the stomach is the least common, with gastric duplication cysts representing only approximately 4% of all the duplication cysts.[2,3] Gastric duplication cysts are twice as common in female than male patients.[1]

Gastrointestinal duplication cysts are most commonly diagnosed in infancy and childhood and rarely in adulthood. Adults are generally asymptomatic but may present with non-specific symptoms, such as abdominal pain, vomiting, gastrointestinal bleeding, and obstructive symptoms. There are rare reports of malignant transformations to adenocarcinoma.[4] Although there are no clear data regarding the incidence or the rate of bleeding in gastric duplication cysts, it is thought that the bleeding occurs more often with the cystic type due to ulceration from the increased intracystic pressure or chemical erosion.[5]

Diagnosis was traditionally made by barium studies. Ultrasound is particularly helpful as a diagnostic tool in infants. Cross-sectional imaging including CT and magnetic resonance imaging (MRI) are useful to characterize the lesion but do not accurately provide the diagnosis. Increasingly endoscopic ultrasound is playing a major role in the diagnosis because it can show the inner echogenic mucosal as well as the outer hypoechoic muscle layers typical of gastrointestinal duplication cysts.[6] Differential diagnoses include any cystic abdominal masses stemming from pancreas, biliary ducts, liver, or spleen as well as gastrointestinal stromal tumors.[7] On pathology, the cyst wall should be lined by epithelium of the gastric mucosa or any other type of gastrointestinal or respiratory mucosa.[7] In our case, sections of the cyst wall showed benign epithelial lining with pseudostratified columnar cells with cilia, attenuated cuboidal cells and squamous epithelium (squamous metaplasia) at different areas. Gastric duplication cyst lined by pseudostratified columnar ciliated epithelium apparently is extremely rare, with fewer than 20 cases reported as of 2017.[8]

There is no consensus on the management of asymptomatic gastric duplication. In the setting of complications, non-communicating gastric duplication cyst is treated by complete surgical resection. Communicating cyst usually requires no treatment if not complicated.[7] Although these lesions are considered benign, it has been proposed that those occurring in individuals over 50 year of age should be completely removed to eliminate the potential risk for malignant transformation.[9]

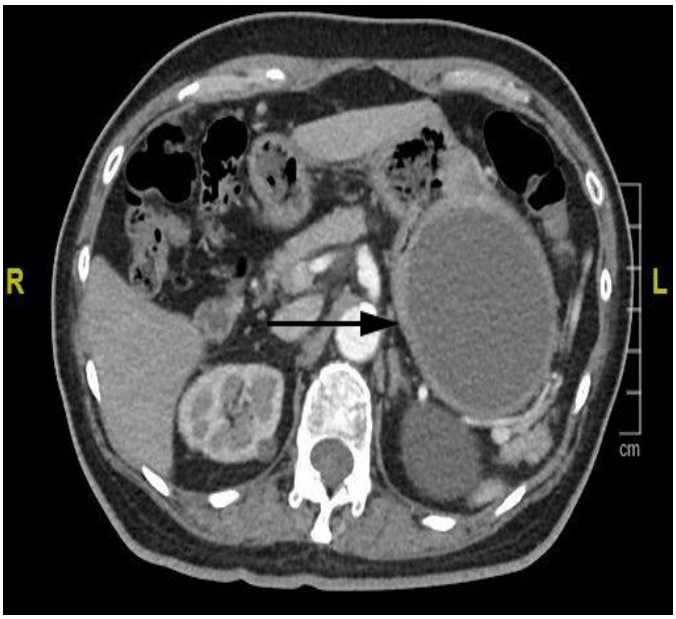

Figure 1. Abdominal computed tomography (CT) scan demonstrating a large cystic lesion adjacent to the gastric fundus (arrow).

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