

Complicated Gastric Duplication Cyst in an Adult Patient: Uncommon presentation of an uncommon disease

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ABSTRACT

Gastric duplication cyst is a very rare congenital anomaly accounting up to 4% of all gastrointestinal tract duplications. It is a quite rare anomaly in adults, the majority of the cases are diagnosed in the neonatal period. Gastric duplication cysts are usually asymptomatic or they do present with non-specific symptoms. They are usually discovered incidentally during endoscopy or laparotomy or very rarely after getting complicated. We describe herein, along with literature review, a case of an adult patient who presented with abdominal pain and bloody vomiting and turned out to have a gastric duplication cyst complicated by internal bleeding.

CASE REPORT

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A 28-year-old female patient presented to the emergency department complaining of three-days history of exacerbating epigastric pain, irradiating to the back and not relieved by over the counter medications. The pain was associated with nausea and episodic vomiting containing streaks of blood. One month prior to presentation, the patient started complaining of early satiety and epigastric discomfort with reported weight loss estimated around 7.5 Kg over the last month.

She denied any history of fever, fatigue or changes in bowel habits. No urinary or respiratory symptoms as well.

The physical examination was unremarkable except for mild discomfort at the epigastric area upon deep palpation. Basic laboratory investigations showed normal hematocrit

level and normal white count. The pancreatic and liver panels were within normal ranges.

Initial workup in the emergency room was done with an ultrasound showing a bi-lobulated cystic mass in the right upper quadrant measuring approximately 15 x 6 cm having smooth internal surface with dependent echogenic component. There was no significant vascularization on Doppler. The lesion lies between the liver, the pancreatic head and the stomach with a close contact with the gastric wall (Figure 1).

Further investigations with a contrast injected CT scan showed a bi-lobulated cystic mass with thickened enhancing wall, smooth inner border and dense content (Figure 2). There was associated fatty infiltration of the surrounding mesenteric fat with minimal layer of perihepatic ascites. The scan delineated better the lesion showing its total separation from

the pancreas and its close abutment with the lower aspect of the greater curvature of the stomach as better demonstrated on the MRI (Figure 3).

Endoscopic ultrasound was then performed showing a multilayered lesion containing echogenic content with some of its layers continuous with those of the gastric wall; features are highly suggestive of gastric duplication cyst (Figure 4). The content of the cyst was aspirated yielding serosanguinous fluid and explaining the streaks of blood described by the patient. FNA needle biopsy, performed at the same time during the endoscopic ultrasound, showed numerous polynuclear neutrophils, macrophages and numerous red blood cells. No malignant cell was detected. There were no endoscopic signs of active bleeding. Upper gastrointestinal gastrografin swallow was also performed to assess the presence of communication between the stomach and the cystic lesion. It shows early contrast filling of the cystic lesion as the contrast arrives to the stomach and before reaching the duodenum (Figure 5).

The patient was hemodynamically stable with significant improvement of her symptoms on medical treatment. She was started on antibiotics because of the inflammatory pattern of the lesion on the CT scan (enhancement and fatty infiltration) and the dominance of the polymorphonuclear neutrophils on the FNA. The patient was discharged home after resolution of her symptoms, to be followed clinically and radiologically. Elective surgical excision of the lesion was planned after the resolution of the inflammation or earlier in case of recurrence of the symptoms.

One year after, the patient presented again with recurrent similar symptoms including abdominal pain, intermittent vomiting and streaks of blood. Decision was then to intervene surgically. Laparoscopic excision of the cyst was performed successfully with no postoperative complications. The lesion showed an identical wall as the gastric wall. It was found adhering to the greater curvature of the stomach at the inferior antral and pyloric border with no clear separation. Pathological analysis of the lesion confirmed the diagnosis of gastric duplication cyst showing the multilayered aspect of the wall similar and continuous with the mucosa and submucosa layers of the gastric wall at the surgical margins (Figure 6). The patient was seen in outpatient clinics as routine follow up 3 months after the procedure. She denied having any post-operative symptoms.

The patient did well after the surgery. She denied any recurrent symptoms on her outpatient follow up visit 3 months after discharge.

DISCUSSION

Gastrointestinal tract duplication is a rare congenital anomaly that can affect any part of the gastrointestinal tract from the esophagus until the rectum with the ileum, esophagus, colon and the stomach are the most commonly affected sites respectively [1].

The gastric duplication cysts (GDC) represent 4% of these duplication cysts [2], they are classified either as cystic or tubular depending on their communication and the contiguity with the stomach. The cystic gastric duplication cysts represent more than 80% of the cases and they are usually separate of the stomach and not communicating with its lumen. The remaining 20% represent the tubular GDC, they are contiguous with the stomach and show some communication with its lumen [3, 4].

Etiology & Demographics:

The majority of gastric duplication cysts are diagnosed in younger age groups with more than two-thirds of the cases diagnosed in infants. Gastric duplication cysts occur twice as often in female patients [1].

Many theories have been reported concerning the pathogenesis and formation of gastric duplication cysts. Some have suggested that duplications share the same pathogenesis of the diverticula formation as outpouching of the gastrointestinal tract [5]. Others hypothesized that supernumerary foregut buds are the origin of the duplications during embryological life [6]. However, the most plausible theory for the origin of duplications is an abnormal vacuolization or recanalization of the intestinal tract after the solid epithelial stage during the embryonic development [7].

Clinical & Imaging findings:

The majority of the GDC are usually symptomatic within the first year of life presenting either with a palpable mass or by upper intestinal obstruction. In adults, they are usually asymptomatic and detected incidentally during cross-sectional imaging or endoscopy done for other indications. However, they may present with non-specific symptoms that vary according to the location, the size, and the type of fluid secreted by their mucosa. Symptoms are usually attributed to infection, hematemesis secondary to ulceration, obstructive symptoms secondary to compression of the adjacent structures or rarely to malignant transformation [8, 9].

Various other congenital anomalies such as other gastrointestinal duplications, esophageal diverticulum, or spinal cord abnormalities may be present [2].

There are no clear data about the incidence or the rate of the bleeding in GDCs. However, it is believed that the bleeding occurs more often with the cystic type due to ulceration secondary to increased intracystic pressure or chemical erosion [10, 11].

Strict radiological and morphological criteria have been established for a definitive diagnosis of duplication cysts with CT scan and endoscopic ultrasound considered the best ways to make the diagnosis [12, 13].

On conventional radiographic examination such as barium studies, the cyst appears as an intramural filling defect or an outpouching of the greater gastric curvature [8].

Ultrasound is a noninvasive imaging modality used as first intention in infants or in the setting of upper gastrointestinal symptoms in adults. It shows the lesion in the majority of the cases as hypoechoic cystic lesion in the upper abdomen seen usually adjacent to the stomach, pancreas, the liver and biliary tracts.

Cross-sectional imaging studies including the CT and MRI are important to determine the cystic nature of the lesion as well as its extent and relation with the adjacent structures. Gastric duplication cysts may appear round or tubular on cross-sectional imaging with thin and slightly enhancing wall. Although cross-sectional imaging is important to characterize the lesion but it does not accurately provide the diagnosis [10].

Endoscopic ultrasound plays a major role in the diagnosis by showing the inner echogenic mucosal and the outer hypochoic muscle layers that are typical of a gastrointestinal tract duplication [14]. The role of FNA in GDCs remains controversial. Some authors argued that a cytological and histological examination of the cyst via EUS-FNA was necessary to rule out malignancy [14].

On pathology, the cyst wall should be lined by epithelium of gastric mucosa or any other type of gastrointestinal or respiratory mucosa. For instance, the mucosal lining of the cyst may include those of the thorax, tongue, liver, pancreas and stomach suggesting that the undifferentiated epithelium of foregut might undergo transition to differentiated specialized epithelium during embryonic period [15].

Our present case fulfilled all these criteria of communicating gastric duplication cyst.

Treatment & Prognosis:

There is no accepted consensus of management of gastric duplication cysts especially if they are asymptomatic.

Complete surgical removal is considered the treatment of choice of GDC to avoid the potential complications such as obstruction, torsion, perforation, hemorrhage, and because the risk of malignant transformation that have been described in some cases [16, 17].

Non-communicating GDC is classically treated by complete resection, performing excision of the shared wall between the stomach and the duplication whereas communicating GDC usually requires no intervention if not complicated [18]. Laparoscopic resection is being performed successfully [19].

Differential Diagnoses:

The differential diagnosis includes any cystic abdominal masses, especially arising from the adjacent organs (pancreas, liver, biliary ducts and spleen). History, clinical presentation and radiological exams are usually helpful to orient toward the right diagnosis.

Pancreatic cystic lesions: GDC can easily mimic a pancreatic pseudocyst which is usually associated with clinical history and imaging findings of recurrent or chronic pancreatitis. Other differential diagnosis includes malignant pancreatic cystic tumor-like pancreatic mucinous cystadenoma especially in patients in whom the duplication is contiguous with the stomach. The diagnosis of pancreatic mucinous cystadenoma is based on the clinical presentation, the CT scan, EUS, and the high concentrations of CEA [18]. For instance, malignant pancreatic mass, usually seen in elderly patients, arises from the pancreas and shows solid and/or cystic lesion with heterogeneous enhancement on cross-sectional imaging [15].

Biliary and hepatic cysts: include mainly hepatic cysts of the left hepatic lobe and biliary cysts of the extrahepatic biliary ducts. Ultrasound and cross-sectional imaging help usually making the diagnosis showing hypochoic or hypodense lesion arising from the liver (left lobe) or extrahepatic biliary tracts. They are commonly seen in adults/elderly patients and they are usually asymptomatic.

Splenic cystic lesions: Any splenic cystic lesions can present like a gastric duplication cyst. Differential considerations include epithelial inclusion cyst of the spleen, post-traumatic pseudocyst of the spleen and parasitic cyst of the spleen. Cross-sectional imaging is usually sufficient to make the diagnosis.

Gastrointestinal stromal tumors: they are also among the differential diagnoses. They usually present in early adulthood. They arise from the gastric wall and may show cystic or mixed cystic and solid enhancing component [15].

Other differentials considerations include mesenteric and omental cysts [16].

TEACHING POINT

Gastric duplication cyst (GDC) is a congenital abnormality diagnosed in the neonatal period and very rarely in adults. CT scan and endoscopic ultrasound are the best ways to identify GDC showing a cystic lesion that is contiguous to or continuous with the gastrointestinal tract.

REFERENCES

1. Bower RJ, Silber WK, Kiesewetter WB. Alimentary tract duplication in children. *Ann Surg* 1978;188:669-74. PMID: 718292
2. Blinder G, Hiller N, Adler SN. A double stomach in an adult. *Am J Gastroenterol* 1999;94:1100-2. PMID: 10201493
3. Kim DH, Kim JS, Nam ES, Shin HS. Foregut duplication cyst of the stomach. *Pathol Int* 2000;50:142-5. PMID: 10792773
4. Murakami S, Isozaki H, Shou T, Sakai K, Toyota H: Foregut duplication cyst of the stomach with pseudostratified columnar ciliated epithelium. *Pathol Int.* 2008, 58: 187-190. PMID: 18251783
5. Veeneklaas GMH. Pathogenesis of intrathoracic gastrogenic cysts. *Am J Dis Child* 1952;83:500-7. PMID: 14902104
6. Theodosopoulos T, Marinis A, Karapanos K, Vassilikostas G, Dafnios N, Samanides L, Carvounis E. Foregut duplication cysts of the stomach with respiratory epithelium. *World J Gastroenterol* 2007; 13: 1279-1281. PMID: 17451215
7. Gross RE, Holcomb GW Jr, Farber S. Duplications of the alimentary tract. *Pediatrics* 1952; 9: 448-468. PMID: 14920118
8. Macpherson RI. Gastrointestinal tract duplications: clinical, pathologic, etiologic, and radiologic considerations. *RadioGraphics* 1993;13:1063-1080. PMID: 8210590
9. Kim SK, Lim HK, Lee SJ, Park CK. Completely isolated enteric duplication cyst: case report. *Abdom Imaging* 2003;28:12-4. PMID: 12483377
10. Gupta S, Sleeman D, Alsumait B, et al. Duplication cyst of the antrum: a case report. *Can J Surg* 1998;41:248-50. PMID: 9627554
11. Wiczorek RL, Seidman I, Ranson JH, et al. Congenital duplication of the stomach: case report and review of the English literature. *Am J Gastroenterol* 1984;79:597-602. PMID: 6465107
12. Holcomb GW, Gheissari A, O'Neill JA, Shorter NA, Bishop HC. Surgical management of alimentary tract duplications. *Annals of Surgery.* 1989;209:167-174. PMID: 2916861
13. Rowling JT. Some observations on gastric cysts. *Br J Surg* 1959;46:441-5. PMID: 13651616
14. Seijo Ríos S, Lariño Noia J, Abdulkader Nallib I, et al. Adult gastric duplication cyst: Diagnosis by endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA). *Rev Esp Enferm Dig* 2008;100:586-90. PMID: 19025312
15. Kuraoka K, Nakayama H, Kagawa T, et al. Adenocarcinoma arising from a gastric duplication cyst with invasion to the stomach: a case report with literature review. *Journal of Clinical Pathology* 2004;57:428-431. PMID: 15047751
16. Barr LL, Hayden CK Jr, Stansberry SD, et al. Enteric duplication cysts in children: Are their ultrasonographic wall characteristics diagnostic? *Pediatr Radiol* 1990;20:326-8. PMID: 2190153
17. Mardi K, Kaushal V, Gupta S: Foregut duplication cysts of stomach masquerading as leiomyoma. *Indian J Pathol Microbiol.* 2010, 53: 160-161. PMID: 20090253
18. D'Journo XB, Moutardier V, Turrini O, et al. Gastric duplication in an adult mimicking mucinous cystadenoma of the pancreas. *Journal of Clinical Pathology.* 2004;57:1215-1218. PMID: 15509688
19. Tayar C, Brunetti F, Tantawi B, et al. Laparoscopic treatment of an adult gastric duplication cyst. *Ann Chir* 2003;128:105-8. PMID: 12657549

FIGURES

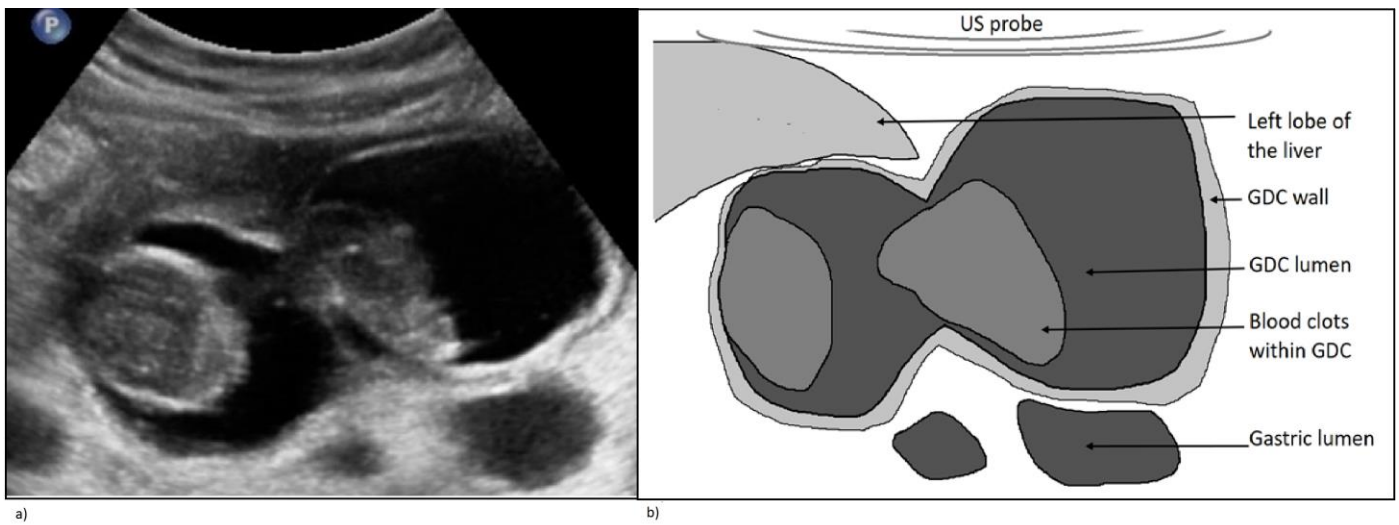


Figure 1: 28-year-old female patient with gastric duplication cyst.

Findings: Bi-lobulated cystic lesion consistent with gastric duplication cyst (GDC) showing smooth internal surface and inner echogenic component representing the blood clots seen on the CT scan.

Technique: Trans-abdominal ultrasound using a curved transducer (3.5 MHz) at the epigastric region (a) with its schematic drawing (b).

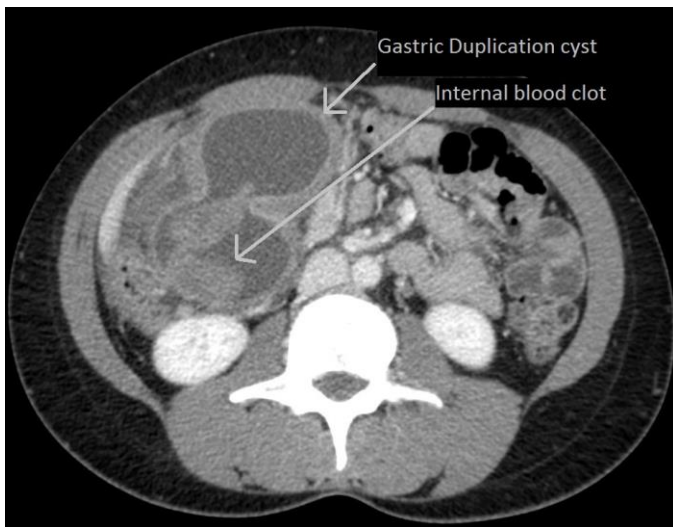


Figure 2: 28-year-old female patient with gastric duplication cyst.

Findings: Bi-lobulated cystic lesion in the right upper quadrant with thickened enhancing wall, smooth inner border and dense content. Findings are consistent with gastric duplication cyst with internal blood clots

Technique: Axial contrast-enhanced CT scan (Multislice, 64-detector) of the abdomen acquired in the portal phase (120 kV, 106 mAs), contrast: 70 ml, Isovue-370 @.



Figure 3: 28-year-old female patient with gastric duplication cyst.

Findings: The gastric duplication cyst (GDC) (partially included in this coronal cut) shows very bright T2 signal intensity. It is seen in close contact with the inferior aspect of the gastric greater curvature (marked by *), the liver and the gallbladder (GB).

Technique: Coronal T2-weighted image of the abdominal cavity (3T magnet, 915//80.00, TA 90, 512x512, ST: 4 mm).

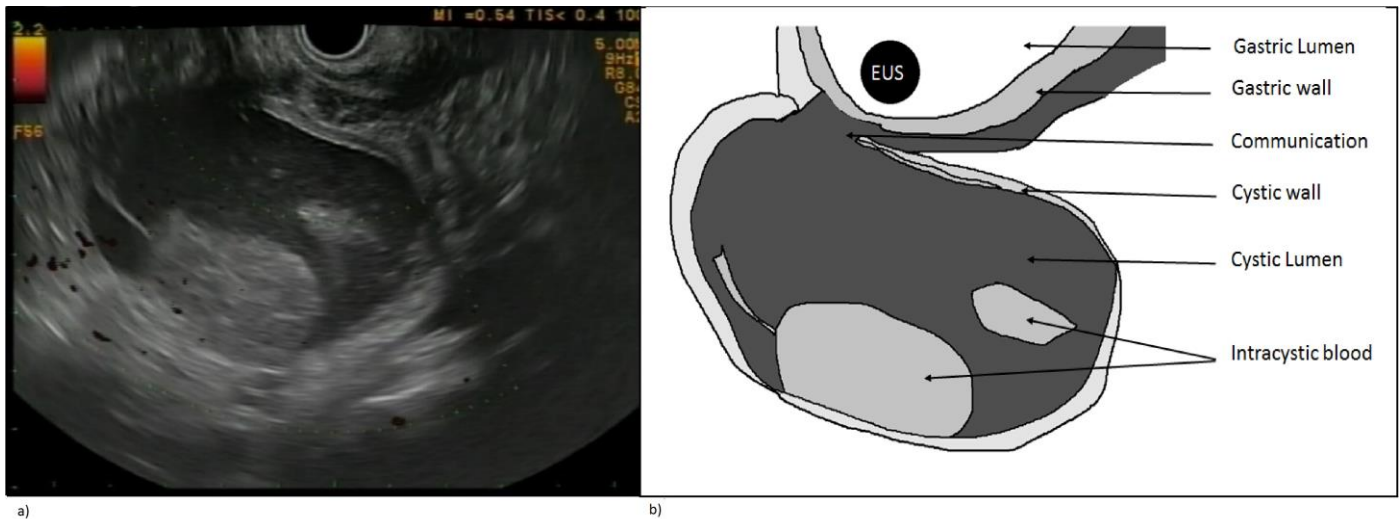


Figure 4: 28-year-old female patient with gastric duplication cyst.

Findings: Endoscopic ultrasound (EUS) showing the multilayered aspect of the gastric duplication cyst wall with the smooth internal surface of the cystic wall and inner echogenic component (intra-cystic blood). The lesion is communicated with the gastric lumen as demonstrated on the drawing.

Technique: Endoscopic ultrasound (9 Hz) (a) with its schematic drawing (b).

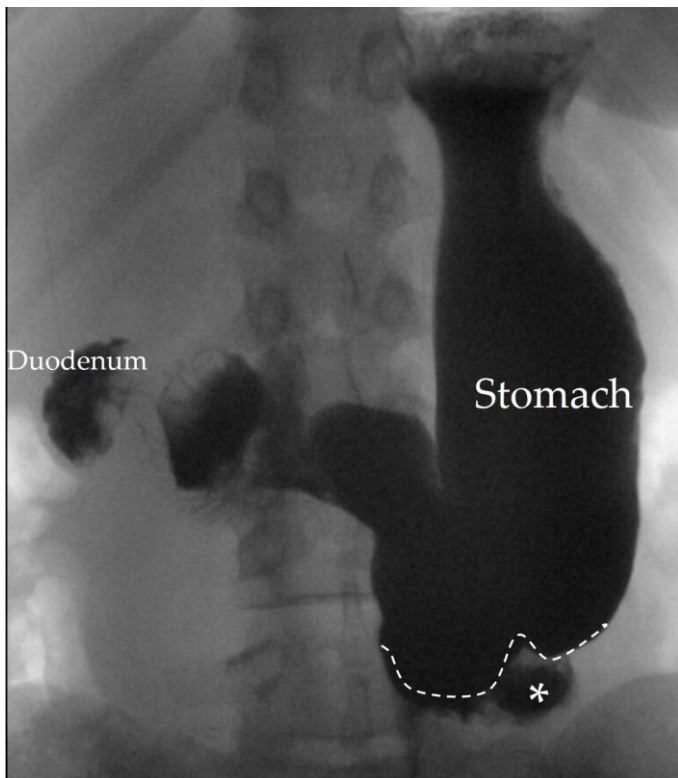


Figure 5: 28-year-old female patient with gastric duplication cyst.

Findings: Demonstration of the early contrast filling of the gastric duplication cyst (marked by *) see as an outpouching from the greater gastric wall as the contrast fills the stomach and the proximal duodenum.

The greater curvature of the stomach is marked by white dotted line.

Technique: Anteroposterior radiograph of an upper gastrointestinal Gastrografin swallow.

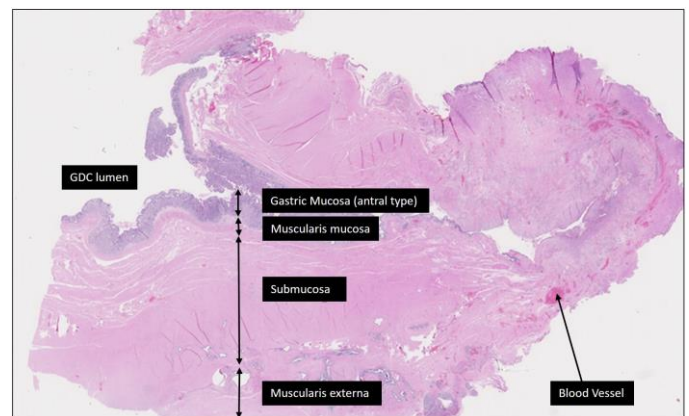


Figure 6: 28-year-old female patient with gastric duplication cyst.

Findings: Demonstration of the gastric duplication cyst lumen and the gastric mucosa type (Antrum) with inflammatory thickening of the wall.

Technique: Microscopic pathological analysis of the surgical specimen after resection of the gastric duplication cyst (Hematoxylin and Eosi Staining, 7.5x).

Etiology:	Congenital
Incidence	4% of all duplication cysts
Gender ratio	F:M = 2:1
Age predilection	<ul style="list-style-type: none"> • Neonatal period • Very rarely in adults
Risk factors	None described
Treatment	<ul style="list-style-type: none"> • Surgical resection if non-communicating • Follow up if communicating (except if symptomatic)
Prognosis	Favorable if treated
Findings on imaging	Cystic lesion contiguous to the stomach wall

Table 1: Summary table of gastric duplication cyst.

	General	Ultrasound	CT	MRI	Endoscopic Ultrasound
Gastric Duplication Cyst	<ul style="list-style-type: none"> • Neonatal period • Rarely in early adulthood 	<ul style="list-style-type: none"> • Hypoechoic lesion adjacent to the stomach 	<ul style="list-style-type: none"> • Non-enhancing cystic lesion (if not complicated) 	<ul style="list-style-type: none"> • Non-enhancing cystic lesion (if not complicated) 	<ul style="list-style-type: none"> • Cystic lesion with inner echogenic mucosa and outer hypoechoic muscle layer • May be or not communicating with the stomach
Gastrointestinal stromal tumor	<ul style="list-style-type: none"> • Early adulthood 	<ul style="list-style-type: none"> • It may show a solid and/or cystic lesion adjacent/within the stomach 	<ul style="list-style-type: none"> • Enhancing solid lesion (sometimes mixed solid and cystic) arising from the gastric wall 	<ul style="list-style-type: none"> • Enhancing solid lesion (sometimes mixed solid and cystic) arising from the gastric wall 	<ul style="list-style-type: none"> • Solid and/or cystic lesion adjacent/within the stomach.
Pancreatic Cystic mass	<ul style="list-style-type: none"> • Late adulthood/elderly 	<ul style="list-style-type: none"> • Cystic or mixed solid and cystic mass arising from the pancreas 	<ul style="list-style-type: none"> • Cystic or mixed solid and cystic mass arising from the pancreas 	<ul style="list-style-type: none"> • Solid and/or cystic mass arising from the pancreas 	<ul style="list-style-type: none"> • Cystic or mixed solid and cystic mass arising from the pancreas
Pancreatic Pseudocyst	<ul style="list-style-type: none"> • History of chronic pancreatitis 	<ul style="list-style-type: none"> • Hypoechoic lesion arising from the pancreas 	<ul style="list-style-type: none"> • Cystic lesion (Usually not enhancing) arising from the pancreas 	<ul style="list-style-type: none"> • Cystic lesion (usually not enhancing) arising from the pancreas 	<ul style="list-style-type: none"> • Hypoechoic lesion arising from the pancreas.
Biliary/Hepatic /Cystic lesions	<ul style="list-style-type: none"> • Adulthood/elderly • Usually asymptomatic 	<ul style="list-style-type: none"> • Hypoechoic lesion arising within the liver or the biliary ducts 	<ul style="list-style-type: none"> • Hypodense lesion arising within the liver or the biliary ducts 	<ul style="list-style-type: none"> • Cystic lesion arising from the liver or the biliary ducts 	<ul style="list-style-type: none"> • Hypoechoic lesion arising within the liver or the biliary ducts
Splenic Lesions	<ul style="list-style-type: none"> • Inclusion or simple cyst • Post traumatic pseudocyst • Parasitic cyst 	<ul style="list-style-type: none"> • Hypoechoic lesion arising within the spleen 	<ul style="list-style-type: none"> • Hypodense lesion arising the spleen 	<ul style="list-style-type: none"> • Cystic lesion arising from the spleen 	<ul style="list-style-type: none"> • Not applicable

Table 2: Differential diagnosis table for gastric duplication cyst.

ABBREVIATIONS

CEA = Carcinoembryonic Antigen
CT = Computed Tomography
EUS = Endoscopic Ultrasound
FNA = Fine Needle Aspiration
GDC = Gastric Duplication Cyst
MRI = Magnetic Resonance Imaging
US = Ultrasound

KEYWORDS

Gastric duplication cyst; Duplication cyst; Upper gastrointestinal bleeding; Hematemesis; endoscopic ultrasound

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