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Adult Gastric Bronchogenic Cyst With Elevated Tumor Marker in Containing Fluid: A Case Report and Literature Review

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Introduction: Duplication cysts are rare malformations that exist in the alimentary tract, and the mucosa of gastric bronchogenic cyst lined by pseudostratified columnar ciliated epithelium (PCCE) is even more rare. We reviewed related literatures to depict this unique abnormality.

Case presentation: Herein we report an abdominal mass that was found incidentally in a 52-year-old female. The biochemical test of contents revealed a high concentration of tumor markers unusually. A laparoscopic surgery was undertaken to remove the lesion. The cyst was found to originate from the stomach but did not show any anatomic communication with the stomach lumen. Pathologic examination confirmed the mucosa was lined by PCCE.

Conclusion: The symptoms and radiologic presentations of GDC are nonspecific. It should be distinguished with other abdominal cystic masses. Surgery is advised in respective of malignancy.

Gastrointestinal duplication cysts are uncommon malformations in the gastrointestinal tract with an uncertain location anywhere from the esophagus to the colon. Gastric duplication cyst (GDC) accounts for about 2% to 8% of all gastrointestinal duplication cysts.[1] More than half of the patients were accurately diagnosed in early childhood, as a result of unspecific symptoms associated with GDC in children.[2–4] Conversely, most adult patients are asymptomatic and cysts are often found incidentally.[5] The requisite criteria for diagnosing of GDC is: (a) the wall of the cyst is contiguous with the stomach wall; (b) the cyst is surrounded by smooth muscle, which is continuous with the

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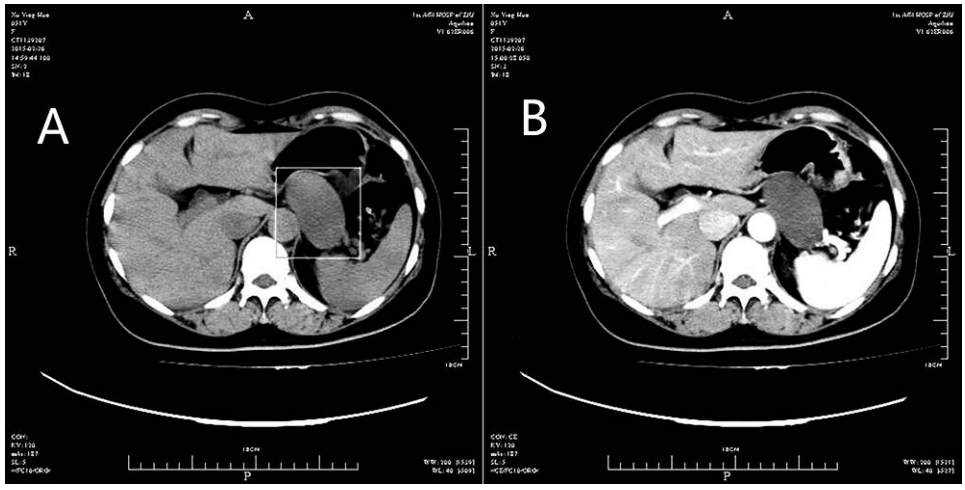

Fig. 1 CT of this patient showed the cyst clearly. (a) Non-contrast enhanced CT revealed a thick-walled cyst (red box) posterior to the gastric wall. (b) The cyst did not show contrast enhancement in arterial phase of contrast enhanced CT.

muscle of the stomach; and (c) the cyst wall is lined by epithelium of gastric or any other type of gut mucosa.[6,7]

In this paper, we presented a case of GDC lined by pseudostratified columnar ciliated epithelium (PCCE), with extremely high concentrations of CEA, CA19-9, and CA-125 in the content fluid. All available relevant literature was collected and analyzed comprehensively.

Case Report

A 52-year-old female admitted into our hospital for further consultation after an abdominal mass was detected.

No symptoms were presented. Her weight did not drop sharply. She admitted a history of pulmonary tuberculosis for 3 years. Apart from this, further history did not reveal any other chronic diseases. Examination was unspecific. Liver function test and tumor marker test were within normal ranges.

The ultrasound test indicated a hypoechoic lesion located in the less curvature of the stomach. A computed tomography (CT) scan of the abdomen and pelvis showed an oval thick-walled collection posterior to the gastric wall and anterior to the pancreatic body without any contrast enhancement (Fig. 1). The cyst size was about 63 × 37 mm. An ulcer could be seen in the gastric mucosa, but no cyst or communication between the cyst and stomach lumen was found under gastroscopy endoscopic ultrasonography (EUS; Fig. 2). EUS was introduced, and the results showed part of the cyst wall maybe connected to the pancreas. Drainage of the cyst was done with a needle under EUS; a coffee-colored mucoid fluid of about 11 mL was drained. The biochemical examination of the fluid showed ascending amylase (3815U/L) and notable elevated levels of CEA (13,550 ng/mL), CA19-9 (40742U/mL), CA-125 (>10,000U/mL), CA15-3 (262.5U/mL), ferritin (1928.5ng/mL) separately, while AFP was undetectable. A diagnosis of pancreatic mucinous cystadenoma was established.

A laparoscopic laparotomy was performed and found the cyst was surprisingly isolated from the pancreas and diaphragm legs, but originated from the posterior wall of the less curvature of the stomach (Fig. 3). The cystic lesion was resected with a stapling cutter from the stomach wall. Gross check on-table revealed normal gastric mucosa of the medial surface of the specimen (Fig. 4).

The pathologic examination confirmed that the cyst was embedded within the gastric muscular layer and did not communicate with the gastric lumen. Microscopically, the cyst wall was lined with PCCE. The smooth muscle layers underneath the mucosa were consecutive and intact (Fig. 5). Ultimately, the diagnosis was determined as gastric bronchogenic cyst.

The patient was uneventful recovered and was discharged on postoperative day 5. A 1-year follow-up showed the patient was well and free of tumor recurrence.

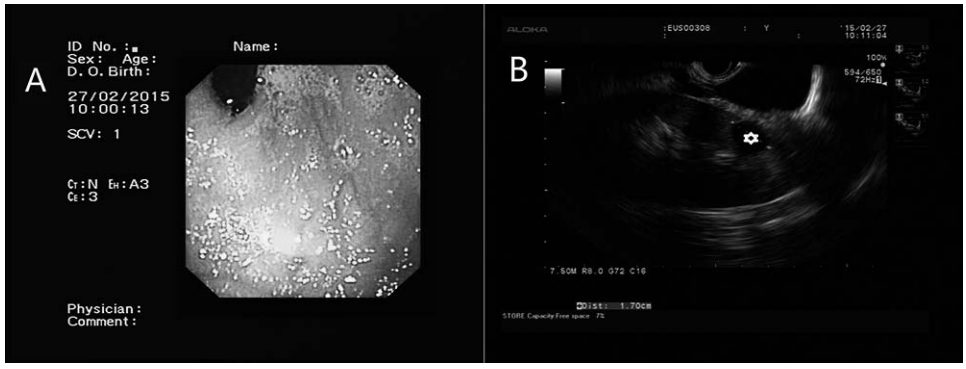

Fig. 2 EUS showed nonatrophic gastritis without erosion. (a) The cyst did not communicate with gastric cavity. (b) The size of cyst's cross section (red asterisk) was 6.6 × 3.0 cm.

Discussion

Duplication cyst is a broad definition, irrespective of its location, morphology, or pathology, by way of simplifying the nomenclature.[8] This means alimentary duplication cysts are abnormalities of different size with the possibility of arising from anywhere along the gastrointestinal tract. The symptoms can be variable, including vomiting, anemia, nausea, distension, dysphagia, and dyspepsia.[5] Most of them are discovered incidentally during routine physical examination.

Aberrant tissues such as pancreas and respiratory epithelium were confirmed with pathologic examination in some duplication cysts.[9–11] PCCE is typical tissue which exists in respiratory mucosa. To elucidate the outline of this special malformation, we reviewed all reported cases of GDCs lined by PCCE, presented in Table 1.[11–31] Patients were confirmed with a cystic mass in adult ages, without difference in prevalence in males and females. Most patients presented as asymptomatic. No patient was found in a condition which the cyst communicated with the gastric lumen. Accurate diagnoses were quite few, yet replaced with gastrointestinal stromal tumor (GIST), neurogenic tumor, adenocarcinoma, or other abdominal diseases.

Adenocarcinoma arising from GDCs lined by PCCE was not always found in reports, but there are still debates about GDC malignant transformations.[32–36] The author related it to the lasting action of chronic inflammation in duplication cysts. Fuku-

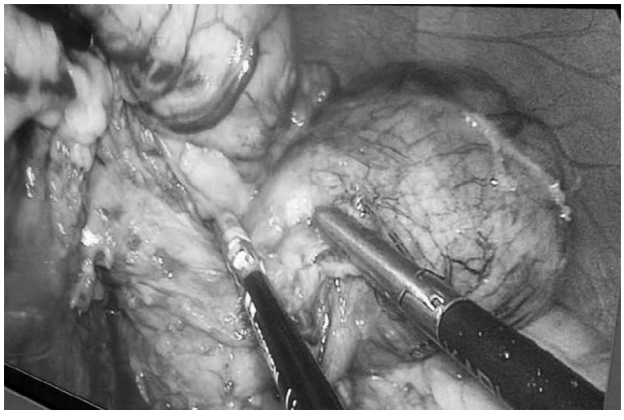

Fig. 3 Findings from the perspective of laparoscope.

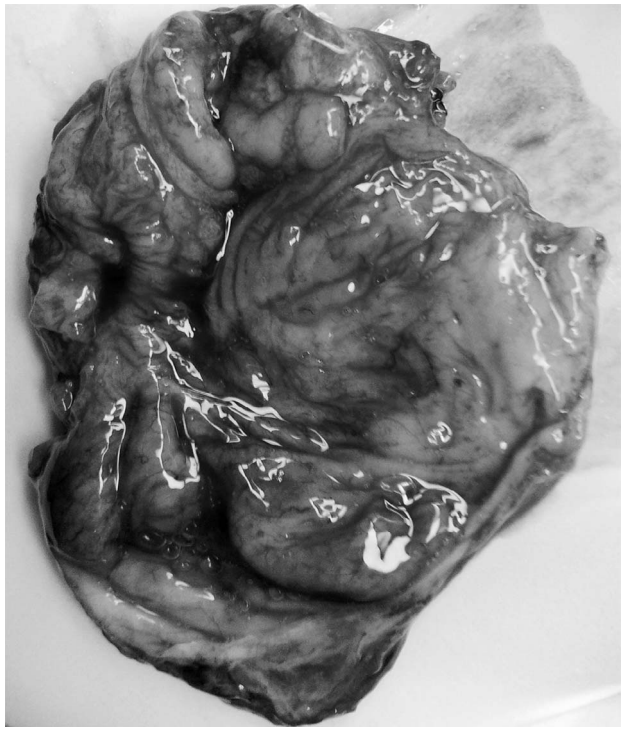

Fig. 4 The inner wall of the cyst looks demonstrated normal gastric mucosa on the whole.

Table 1 Reported gastric duplication cysts lined by PCCE

References	Sex	Age	Location	Size (cm)	Communicate with the stomach or not	Symptom	First diagnosis
Falleti *et al*, 2013	M	55	Gastroesophageal junction	NR	NR	Asymptomatic	GIST
Belli *et al*, 2013	F	45	Gastric cardia	4.8 × 2.1	NR	Abdominal pain	GIST
Kurokawa *et al*, 2012	M	71	Gastric cardia	NR	N	Throat discomfort	NR
Wu Yulian *et al*, 2011	M	76	NGEJ, LC	4.0 × 4.0	N	Asymptomatic	GIST
Mardi *et al*, 2010	M	42	LC	4.5 × 5.2	NR	Left lumbar pain	Gastric leiomyoma
Jiang *et al*, 2010	F	25	Gastric fundus	3.0 × 2.5	N	Epigastric pain	GIST
Shibahara *et al*, 2009	M	43	LC	NR	NR	Epigastric pain	Gastric adenocarcinoma
Sato *et al*, 2008	F	60	LC	3	N	Asymptomatic	Cystic neoplasm
Murakami *et al*, 2008	F	72	LC	2.0 × 1.5	N	Asymptomatic	GIST
Wakabayashi *et al*, 2007	M	37	NGEJ, LC	4.0 × 4.0	N	Epigastric pain	NR
Hall *et al*, 2007	M	40	NGEJ, LC	6.0 × 5.0	N	Epigastric discomfort	GIST or enlarged lymph nodes
Theodosopoulos *et al*, 2007	F	46	Posterior of fundus	8.0 × 5.5	NR	Vomiting	NR
Lee *et al*, 2006	F	38	LC	3	N	Asymptomatic	NR
Cunningham *et al*, 2006	F	63	Posterior of fundus	10.0 × 7.6	N	Abdominal pain	GDC
Melo *et al*, 2005	F	39	Fundus	4.0 × 2.5	NR	Asymptomatic	GIST
Rubio *et al*, 2005	M	26	NR	NR	NR	Epigastric pain	NR
Song *et al*, 2005	F	62	NGEJ, LC	3.5 × 2.5	NR	Asymptomatic	Neurogenic tumor or lymph node
Hedayati *et al*, 2003	F	59	Posterior of stomach	7.0 × 5.0	N	Asymptomatic	NR
Kim *et al*, 2000	M	35	NGEJ, LC	7.0 × 6.0	N	Epigastric pain	GDC or liver cyst
Takahara *et al*, 1996	M	25	Posterior of fundus	6.5 × 5.0	NR	Asymptomatic	GIST
Gensler *et al*, 1966	F	46	NGEJ, LC	6.0 × 8.0	N	Asymptomatic	Leiomyoma or lipoma of the stomach
Present	F	52	LC	7.0 × 6.5	N	Asymptomatic	Pancreatic mucinous cystadenoma

F, female; LC, lesser curvature; M, male; NGEJ, near gastroesophageal junction; N, negative; NR, not reported.

moto[33] insists that it is a red alert for those who are identified with GDCs and older than 50, because of continuous exposure to carcinogenic substances. In the case we presented, the fluid drained from the cyst contained extremely high levels of CEA, CA19-9, and CA-125, which questioned the meaning of this phenomenon. Only 2 other articles have pointed out similar findings, but neither of both were the same pathologic characteristics.[37,38] Tumor markers are referred as cancer biomarkers. They are produced by tumors or by the bodies in response to cancer. Different elevated levels of CEA, CA19-9 and CA-125 often arose in patients who suffer from pancreas cancer, colon cancer, etc.[39] Combined with previous studies, we have reasons to believe that the elevated tumor marker indicated a potential risk of malignancy, regardless of its leading role or possibly just a byproduct in this process.

Interestingly, expression of TTF-1, MUC1, MUC5AC, and surfactant were confirmed in different cases.[40] TTF-1 and surfactant participate in lung embryogenesis and maturation. With the ciliated epithelium, the authors assumed a branch arising from the respiratory diverticulum gives rise to the development of gastric bronchogenic cyst. The expression of TTF-1 begins at weeks 11-12 gestation in fetus, hence it is possible that the expression of TTF-1 is negative if a duplication cyst developed at a very early stage during gestation.[29]

Duplications presented in spherical or tubular forms mostly. The former is much more common, accounting for about 82% of all reported cases and separate with the gastrointestinal lumen. Tubular cysts are usually seen in the bowel, communicating with the lumen.[41] Atypical dumbbell-shaped cells have been found in some cases.[42] Gastric duplication cysts are frequently located in the greater curvature of stomach, adjacent to the pancreas, spleen, or other abdominal organs.

Different imaging examinations display duplication cysts well. Contrast-enhanced CT revealed a thick-walled cystic lesion with enhancement of the inner lining.[43,44] Calcified plaque emerges occasionally. Solid foci may suggest tumor formation in the

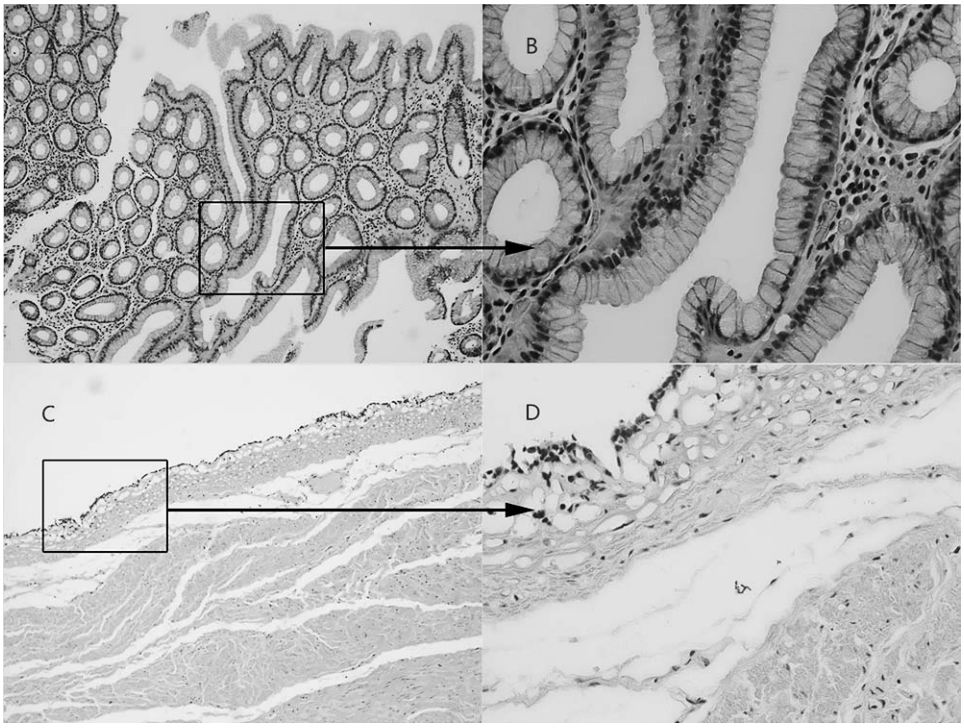

Fig. 5 Histopathologic examination revealed the specific characteristic of the mucosa. (a, b) Hematoxylin-eosin stained histologic section showed cyst wall lined by PCCE. (c, d) The smooth muscle layers underneath the mucosa were consecutive and intact. Original magnifications were ×100 high field (a, c) and ×400 high field (b, d) separately.

cyst. Septa in the cyst may suggest infection. CT and magnetic resonance imaging (MRI) remain superior in diagnosing GDC because of its ability to show cyst margins clearly. However, the advantage of MRI over CT is its ability to make a clear distinction between the thin walls separating the cysts and other organs on T2-weighted images.[45] This also provides more information about mucous or serous contents in the cysts. The fluid content of cysts are diverse due insults such as bleeding, infection, or inflammation.

EUS also provides more details about GDC compared to ordinary ultrasound. Meanwhile, it can aid in the pathologic examination and even treatment. Eom *et al* [46] used this technique to operate a complete dissection of GDC. GDCs should be considered when ultrasound examination reveals an echogenic internal mucosal layer and a hypoechoic intermediate muscular layer, defined as "double wall" or "muscular rim."[47] Nevertheless, these instruments are not sufficient to identify duplication cysts accurately, due to the nonspecific features of these cysts. GDC can be misdiagnosed as gastrointestinal stromal tumor, pancreatic cyst, splenic cyst, etc. It is notable that neoplasms from GDCs should be distinguished from those associated with cystic neoplasms or tumors with cystic degeneration, such as a GIST, neuroendocrine tumor, neurogenic tumor, or heterotopic pancreas.[9,48,49]

Some authors recommended conservative therapy in asymptomatic patients.[47] However, given the unknown risk of malignant transformation, surgery is by far considered as the primary suggested treatment. Laparoscopic surgery is quite safe as it ensures an en bloc excision and minimize invasive injury for patients. If a complete excision is not achievable, a partial gastrectomy can be considered as an alternative method.

Acknowledgments

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