

Case Report

Gastric duplication cyst lined by pseudostratified columnar ciliated epithelium masquerading as a pancreatic mucinous cystic neoplasm: a case report and literature review

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Abstract: Gastric duplication cyst (GDC) with a pseudostratified columnar ciliated epithelial (PCCE) is a congenital rare cystic neoplasm, which is often difficult to distinguish from other entities by imaging techniques, and as a consequence it may be wrongly overtreated. We herein report a case of a 52-year-old female incidentally found to have an abdominal mass by ultrasonography and computed tomography. Additionally, endoscopic ultrasonography and fluid analysis were consistent with a pancreatic mucinous cystic neoplasm with a markedly elevated fluid amylase, carcinoembryonic antigen, and carbohydrate antigen 19-9. Then, laparoscopic resection of the cyst originating from the stomach and wedge gastrectomy were performed. Final pathology revealed a GDC with PCCE. In addition, we also performed a literature review of 31 reports of GDC with PCCE. Although rare, GDC lined by PCCE should be included in the differential diagnosis of pancreatic cystic neoplasms or a gastric wall mass.

Keywords: Gastric duplication cyst, laparoscopic resection, pancreatic mucinous cyst, pseudostratified columnar ciliated epithelium

Introduction

Gastric duplication cyst (GDC) is an uncommon congenital malformation and is usually lined by gastrointestinal mucosa. Most cases present during childhood with nonspecific symptoms, such as nausea, vomiting, weight loss, or an epigastric mass. In adults, most cases are discovered incidentally on radiological examination or gastric endoscopy [1, 2]. Accurate diagnosis of GDCs before resection is difficult. Differential diagnoses are varied, including gastrointestinal stromal tumors (GISTs), neuroendocrine tumors, pancreatic heterotopia, pancreatic pseudocysts, pancreatic mucinous cysts, and neurogenic tumors. GDC is typically treated surgically because of the symptoms and risk of malignant change. Additionally, GDC lined by pseudostratified columnar ciliated epithelium (PCCE) is extremely rare, and only 31 cases have been reported in the English literature by April 2017 [1-31]. Herein, we describe a

52-year-old female with a GDC lined by PCCE, preoperatively misdiagnosed as a pancreatic mucinous cystic neoplasm. We make a comprehensive review of the literature.

Case presentation

A 52-year-old woman was referred to the Second Affiliated Hospital of Zhejiang University School of Medicine for evaluation of a large abdominal mass that had been found incidentally on ultrasonography during an annual health examination. Her medication history and physical examination were unremarkable. All laboratory test results, including liver function and tumor markers, were normal. An abdominal computed tomography (CT) demonstrated a homogeneous, low-density mass, measuring 6.3 cm×3 cm, between the stomach and body of the pancreas (**Figure 1A**), without enhancement in the arterial phase (**Figure 1B**) and portal phase (**Figure 1C**). Endoscopic ultrasonogra-

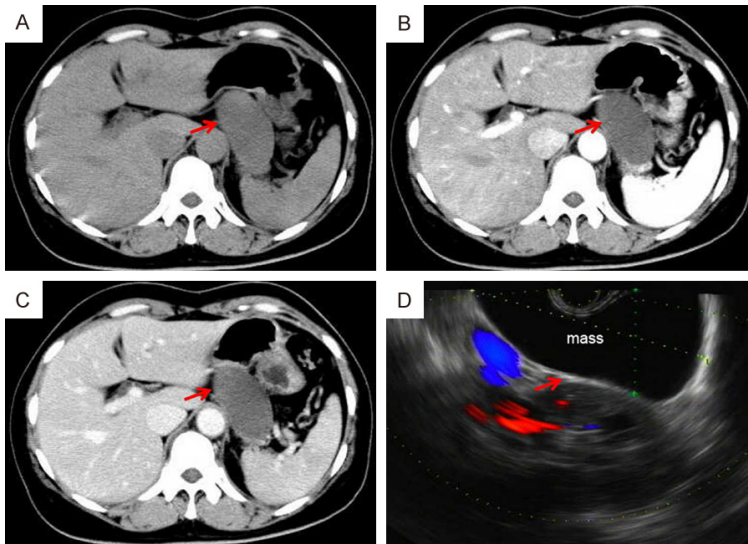


Figure 1. Abdominal computed tomography scan confirmed a homogeneous, low-density mass, measuring 6.3 cm×3 cm (arrows), between the stomach and body of the pancreas (A), without enhancement in the arterial phase (B) and portal phase (C). Endoscopic ultrasonography showed an anechoic cystic lesion near the body of the pancreas not connected with the pancreatic duct or the stomach wall (D).

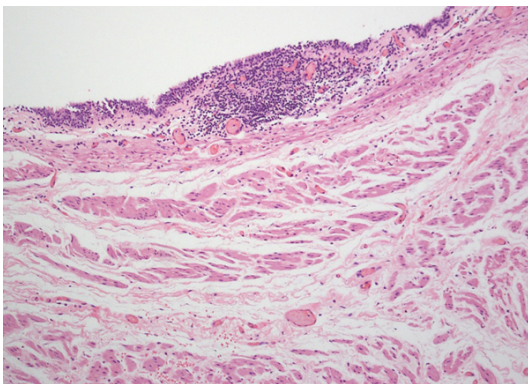


Figure 2. The pathologic examination demonstrated that the cyst wall was lined by pseudostratified columnar ciliated epithelium, and surrounded by benign smooth muscle (hematoxylin and eosin, ×50).

phy guided fine needle aspiration (EUS-FNA) visualized an anechoic cystic lesion near the body of the pancreas not connected with the pancreatic duct or the stomach wall (**Figure 1D**). FNA of the cyst demonstrated no evidence of malignancy, but showed a markedly elevated amylase of 3815 U/L, carcinoembryonic antigen (CEA) level of 13550 ng/ml and carbohydrate antigen (CA) 19-9 level of 40742 U/ml, suggesting the diagnosis of a pancreatic mucinous cystic neoplasm. Given the lesion's malignant potential, high cystic fluid CEA and CA 19-9, and amylase suspicious for neoplastic

process, the patient was therefore referred for surgical resection.

At the time of the laparoscopic exploration, the lesion was noted to be tightly adherent to both the stomach and pancreas. The pancreas was mobilized free of the cyst and it was evident that the lesion originated from the stomach. Then, laparoscopic resection of the cyst and wedge gastrectomy were performed. The pathologic examination demonstrated that the cyst was embedded within the gastric muscular layer, and did not communicate with the gastric lumen. Microscopically, the cyst wall was lined by PCCE, morphologically consistent with respiratory epithelium, and surrounded

by benign smooth muscle (**Figure 2**). The patient has been well since hospital discharge, and the postoperative radiologic work-up and upper endoscopy showed no evidence of recurrence during the 2-year follow-up period.

Discussion

Gastrointestinal duplication is a relatively rare anomaly that may occur at any level from the oral cavity to the rectum with the ileum being the most common site. GDC, accounting for 2-8% of all gastrointestinal duplications, is relatively rare [2]. Most cases will occur in females compared with males (2:1), with the majority of cases being diagnosed in the pediatric population within the first 3 months of life and rarely after 12 years of age [18, 23]. Histologically, they have a gastrointestinal mucosal membrane (usually gastric) with the submucosal smooth muscle layer [5-9]. PCCE is usually found in an esophageal duplication cyst. However, it is extremely rare in the GDC, with only a few reports describing their clinical characteristics.

Since Gensler et al. reported the first case of GDC lined by PCCE in 1966 [1], 30 articles and 31 patients have been reported in the English literature up to April 2017 (**Table 1**) [2-31]. Including the present case, 16 cases were men

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Table 1. Reported literature of gastric duplication cyst lined by pseudostratified columnar ciliated epithelium in the English language

Case	Authors	Sex/Age	Complaint	Location	Size (cm)	CT	MRI	EUS	Preoperative diagnosis	Surgery
1	Gensler et al. [1]	F/46	No	NGEJ, GC	6×8	NA	NA	NA	leiomyoma, lipoma of the stomach	cystectomy
2	Takahara et al. [2]	M/25	No	PW of fundus	6.5×5×5	cystic mass with homogeneous density	fluid level separating an upper layer of low signal intensity and lower layer of high signal intensity inside the cyst	well-defined mass located adjacent to the fourth layer of the gastric wall	benign intramural gastric cyst	partial gastrectomy
3	Kim et al. [3]	M/35	epigastric pain	NGEJ, LC	7×6×5	cyst lesion between stomach and the left lobe of the liver	NA	NA	GDC, simple cyst of liver	cystectomy
4	Hedayati et al. [4]	F/59	No	PW of stomach, LC	7×5	homogeneous mass thought to be arising from the left adrenal gland	NA	NA	adrenocortical carcinoma	cystectomy
5	Song et al. [5]	F/62	No	NGEJ, LC	3.5×2.5×1.5	homogeneous, solid, low-density nodule in the hepatogastric ligament near the LC	NA	NA	perigastric neurogenic tumor, reactive hyperplasia of lymph node	gastric wedge resection
6	Rubio et al. [6]	M/26	epigastric pain	NA	NA	NA	NA	NA	GDC (by biopsy)	No, omeprazol medication
7	Melo et al. [7]	F/39	No	fundus	4×2.5×1	mass in the fundus of the stomach	NA	NA	GIST	gastric wedge resection
8	Cunningham et al. [8]	F/63	fever, abdominal pain	PW of fundus	10×7.6	cystic lesion at the tail of the pancreas	NA	NA	pancreatic mucinous neoplasm	partial gastrectomy
9	Lee et al. [9]	F/38	No	cardia, LC	7×5	solid mass originating from the gastric wall	NA	an echo-poor mass lesion arising within the submucosal layer	GIST, developmental or complicated cyst	cystectomy through endoscopic mucosal resection
10	Theodosopoulos et al. [10]	F/46	vomiting	(1) PW of fundus (2) Gastrosplenic ligament	(1) 8×5.5 (2) 3	well circumscribed cystic lesion attached to the posterior wall of the gastric fundus	NA	NA	NA	excision of both cysts and splenectomy
11	Hall et al. [11]	M/40	epigastric discomfort	NGEJ, LC	6×5	slightly hypodense and homogeneous soft tissue mass	NA	subepithelial hypoechoic lesion	congenital foregut cyst (by EUS-FNA)	No, antibiotics
12	Wakabayashi et al. [12]	M/37	epigastric pain	NGEJ, LC	4×4	cystic tumor adjacent to the LC of the stomach and lateral segment of the liver	NA	the cystic tumor to be attached to the gastric wall	duplication cyst of the stomach	gastric wedge resection
13	Murakami et al. [13]	F/72	No	middle body, LC	2.0×1.5	well-circumscribed subserosal cystic mass	NA	NA	benign cyst of the stomach, GIST	distal gastrectomy with systematic lymph node dissection
14	Sato et al. [14]	F/60	No	cardia, LC	3	low-density, rounded lesion	NA	well-defined cystic lesion without intracystic septa	bronchogenic type of foregut cyst	No, follow-up
15	Shibahara et al. [15]	M/43	epigastric pain	cardia, LC	NA	cystic lesion between the liver and the stomach	cystic lesion between the liver and the stomach	NA	gastric cancer	total gastrectomy with D2 lymph node dissection

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16	Jiang et al. [16]	F/25	epigastric pain	fundus	3.0×2.5×2.0	NA	NA	NA	GIST	gastric wedge resection
17	Mardi et al. [17]	M/42	left lumbar pain	AGJ, LC	4.5×5.2	well defined soft tissue density with mild contrast enhancement	NA	NA	gastric leiomyoma	complete excision of the mass lesion
18	Jiang et al. [18]	M/76	No	NGEJ, LC	4×4	homogeneous, low-density mass, without contrast enhancement	NA	hypoechoic lesion, arising within the tunica muscularis	GIST	surgical excision
19	Khoury et al. [19]	M/29	abdominal pain	fundus, GC	8.5×5.5×4.8	mass at the GC of the stomach	NA	cystic mass in the submucosa of the fundus	GDC	partial gastrectomy
20		F/26	epigastric pain	middle body, LC	5×2.2×2	cystic mass near the gastroesophageal junction along the LC of the stomach	NA	NA	congenital gastrointestinal duplication cyst	partial gastrectomy
21	Hosomura et al. [20]	F/44	No	PW of the stomach	7.5×6.5×6.5	well-circumscribed homogenous low density mass between the PW of the upper third of the stomach and the tail of the pancreas	homogenous low-intensity mass on T1-weighted imaging and a homogenous high-intensity mass on T2-weighted imaging	NA	benign cyst of the pancreas, GIST	gastric wedge resection
22	Napolitano et al. [21]	M/56	No	NGEJ, AW	5×3×3	NA	cystic mass with complex content, located anteriorly to the gastroesophageal junction	hypoechoic mass with slightly heterogeneous internal echoes and regular margins	GDC (by EUS-FNA)	partial gastrectomy
23	Falletti et al. [22]	M/55	No	NGEJ	5×3×3	NA	cystic mass with complex content located forward to the gastroesophageal junction	hypoechoic mass with a slightly heterogeneous internal echo and regular margins	GIST	surgical excision
24	Belli et al. [23]	F/45	abdominal pain	adjacent to the gastric cardia	8×7	well-defined, dense, cystic or semi-solid mass	NA	NA	GIST	NA
25	Leepalao et al. [24]	F/29	left upper quadrant pain	(1) PW (2) anterolateral upper abdomen (3) GC	(1) 9.2×6.6 (2) 1.8×1.7 (3) 3.0×2.8	three benign appearing well demarcated thin-walled simple cystic masses	NA	NA	enteric duplication cysts	wedge resection
26	Geng et al. [25]	M/52	epigastric discomfort	NGEJ, LC	3.0×4.2	well circumscribed, homogeneous, non-enhancing, low density, submucosal cystic mass	NA	NA	GIST	proximal gastrectomy with lymph node dissection
27	Kim et al. [26]	F/43	No	NGEJ, LC	2.5	ovoid submucosal cystic tumor	NA	NA	cystic hygroma, duplication cyst	laparoscopic gastric wedge resection
28	Laurent et al. [27]	M/23	No	NGEJ, PW	4.5×4×4	NA	increased, round shaped mass, with no infiltration of adjacent organ and no gadolinium enhancement	hypo-echogenic mass with slightly heterogeneous internal echoes and regular margins	GIST, gastric leiomyoma, simple cyst of the stomach	gastric wedge resection

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29	Sultan et al. [28]	F/43	abdominal pain	adjacent to the pancreatic tail	7.4×3.6	well-defined sausage-shaped retroperitoneal cystic lesion	no communication between the lesion and the stomach or pancreatic duct	anechoic cystic lesion near the tail of the pancreas not connected with the pancreatic duct or the stomach wall	potential malignant tumor	laparoscopic-assisted distal pancreatectomy, splenectomy, and partial gastrectomy
30	Sun et al. [29]	M/67	epigastric pain	fundus	5	well-defined, low density, cystic lesion with cyst wall enhancement and punctate calcification	NA	lesion located in the muscularis propria of stomach	GIST	cystectomy
31	Tjendra et al. [30]	M/65	progressive dysphagia	NGEJ	4.3×4.2	low density soft tissue mass	NA	hypoechoic round mass with well-defined borders in the subhepatic peritoneal space along the gastrohepatic region	GIST, GDC	transhiatal esophagectomy and partial gastrectomy
32	Namdarglu et al. [31]	M/25	epigastric pain	PW, GC	4×5	well-marginated cystic lesion	cystic lesion, with heterogeneous signal intensity on T2-weighted images	NA	GIST	gastric wedge resection
33	Our case	F/52	No	between the stomach and body of the pancreas	6.3×3	homogeneous, low-density mass	NA	anechoic cystic lesion near the body of the pancreas not connected with the pancreatic duct or the stomach wall	pancreatic mucinous cystic neoplasm	gastric wedge resection

CT: computed tomography; MRI: magnetic resonance imaging; EUS: endoscopic ultrasonography; M: male; F: female; AGJ: anterior of gastrointestinal junction; NGEJ: near gastroesophageal junction; LC: lesser curvature; GC: greater curvature; PW: posterior wall; AW: anterior wall; GDC: Gastric duplication cyst; GIST: gastrointestinal stromal tumor; NA: not available.

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and 17 cases were women. The mean age of the patients was 45.1 years (range 23 to 76 years), and 21 cases (63.6%) were younger than 50 years. Additionally, 15 cases were incidentally detected, while the others had symptoms that included epigastric pain and discomfort, vomiting, abdominal pain, progressive dysphagia and fever. Most of the tumors were located in the lesser curvature of stomach (14 cases) or near the gastroesophageal junction (12 cases). The average tumor size was 5.6 cm (range 2 to 10 cm). Malignant change of GDC lined by PCCE is rare. Shibahara et al. reported a case of bronchogenic cyst of the stomach located at the opposite side wall to the gastric adenocarcinoma, and they thought that chronic inflammation from the bronchogenic cyst extending to the gastric mucosa might be the cause of adenocarcinoma [15].

Despite advances in imaging, it frequently fails to identify the cystic nature of the gastric duplication due to the thick proteinaceous cyst fluid. It has been reported that the rate of CT misdiagnosis ranges from 43 to 70% [2]. Magnetic resonance imaging does not seem to significantly improve diagnostic accuracy [32]. Moreover, EUS may help to differentiate between the solid and cystic component and the relation between the cyst wall and adjacent gastrointestinal structures. EUS-FNA has been proposed as a valuable tool for the diagnosis and characterization of the cyst, and can be therapeutic [2, 21, 32, 33]. However, the fluid analysis of GDCs may resemble other neoplastic processes, such as pancreatic cystic neoplasm given the high amylase, CEA, or CA19-9, especially when located outside the alimentary tract. In our case, we misdiagnosed the mass as a pancreatic mucinous cyst preoperatively, which was similar to Cunningham [8]. Therefore, most cases of GDC are diagnosed during surgical resection or by pathologic examination of surgical specimens. Of the 33 cases, only 3 cases avoided surgical resection, because of the correct preoperative diagnosis of GDC based on EUS-FNA or biopsy [6, 11, 14].

The recommended treatment for symptomatic patients with GDC is surgical resection. Complete cystectomy is the best surgical option and alternatively segmental gastrectomy can be performed. The management of asymptomatic patients with GDC is controversial. Eloubeidi et al. [33] suggested watchful waiting after con-

firmed the benign nature of these cysts by EUS-FNA, while other experts recommend prophylactic surgical resection because of the potential for malignant transformation.

Conclusion

GDC lined by PCCE should be included in the differential diagnosis of a pancreatic cystic neoplasm or gastric wall mass, especially if the lesion is located between the stomach and body/tail of the pancreas.

Disclosure of conflict of interest

None.

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