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Case Report

Gastric Bronchogenic Cyst Histologically Diagnosed After Laparoscopic Excision: Report of a Case

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Abdominal computed tomography of a 71-year-old man revealed a 3-cm mass in gastric cardia. Although the mass was widely attached to the gastric wall, no clear contrast enhancement was observed. Abdominal magnetic resonance imaging revealed the mass to have homogenous high intensity on T2W1 images and isointensity on T1W1 images. On diffusion-weighted imaging, no high intensity was observed. However, the mass had a smooth surface and was widely attached to the gastric wall, consistent with computed tomography findings. A gastric submucosal tumor was suspected. Laparoscopic tumor resection was performed. Histopathologic diagnosis of the mass was a bronchogenic cyst derived from the respiratory primordium originating in the foregut of the primitive intestine. Such cysts are mostly found in the mediastinum or thoracic cavity; their occurrence on the gastric wall is extremely rare. Despite this, we think that bronchogenic cysts should be considered in the differential diagnosis of abdominal unilocular cystic diseases.

Key words: Bronchogenic cyst – Gastric wall – Abdominal cyst

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Bronchogenic cysts originate from the primitive foregut and are believed to be formed from abnormally displaced lung buds.

In normal embryonic development, the lung bud protrudes from the abdominal wall of the foregut in the fourth week. It extends distally, separates from the foregut, and subsequently forms the trachea and bronchus.

Bronchogenic cysts are thought to develop in the period before the formation of the diaphragm, in which the abnormally divided lung bud is displaced in the abdomen without being connected to the lung and bronchus.[1] The lung bud formed from abnormal germination and separation of the primitive trachea is not continuous with the bronchus, and it adheres to the esophagus, which is formed from the posterior part of the foregut. The esophagus then grows caudally and brings about formation of the adhered lung bud structure.[2]

Because they lack continuity with the tracheobronchial system, they are considered to be congenital cysts.[3] Bronchogenic cysts are often found in the thorax and mediastinum. Their occurrence in the abdominal cavity is extremely rare.[2] Because preoperative imaging diagnosis is difficult, they are usually diagnosed after excision. We report our experience with a patient who had a bronchogenic cyst involving the gastric wall, which was preoperatively diagnosed as a gastric submucosal tumor and for which the patient underwent laparoscopic excision. The imaging findings in this case were not typical of bronchogenic cysts.

Table 1 Laboratory findings on admission

	Value
Hematology	
WBC	4500/μL
Hb	14.1 g/dL
Plt	19.4×10^4/μL
Biochemistry	
AST	23 U/L
ALT	21 U/L
BUN	8.6 mg/dL
Cre	0.9 mg/dL
Tumor marker	
CEA	3.1 ng/mL
NSE	8.4 U/mL
SCC	1.0 ng/mL

AST, aspartate aminotransferase; ALT, alanine aminotransferase; BUN, blood urea nitrogen; CEA, carginoembryonic antigen; Cre, creatinine; Hb, hemoglobin; NSE, neuron-specific enlase; Plt, platelet; SCC, squamous cell carcinoma antigen; WBC, white blood cell.

Case Report

Our patient provided his consent for the publication of this case report.

He was a 71-year-old man whose chief complaint was throat discomfort. At the age of 65 years he had been diagnosed with prostate cancer, for which he underwent hormone therapy and radiation therapy. At age 67 years, he was diagnosed with variant angina pectoris and hypertension. He did not have a very remarkable family history. He did not have a history of smoking. For the last 2 years, the patient had been experiencing throat discomfort. He visited

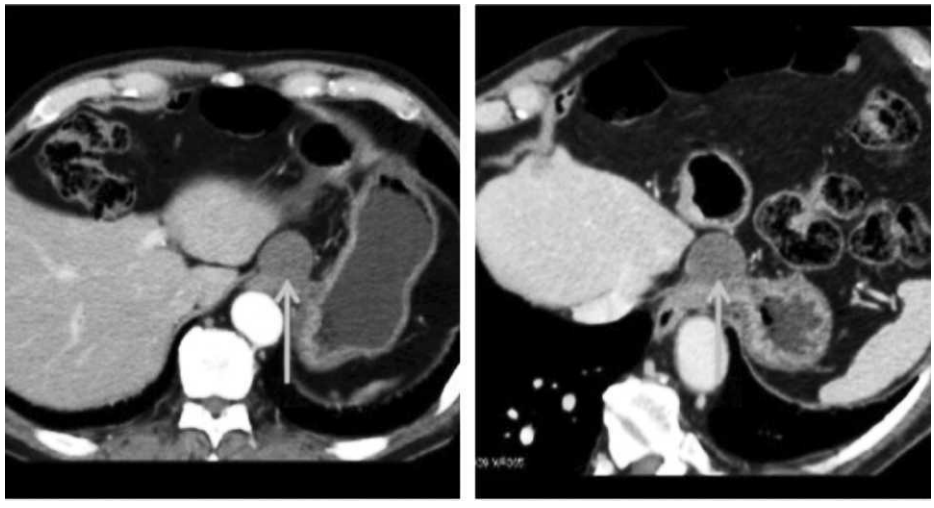

Fig. 1 Findings from contrast-enhanced CT. A tumor lesion of 32 mm in diameter is found anterior to the cardia with soft tissue density, homogenous internal region, and a clear margin.

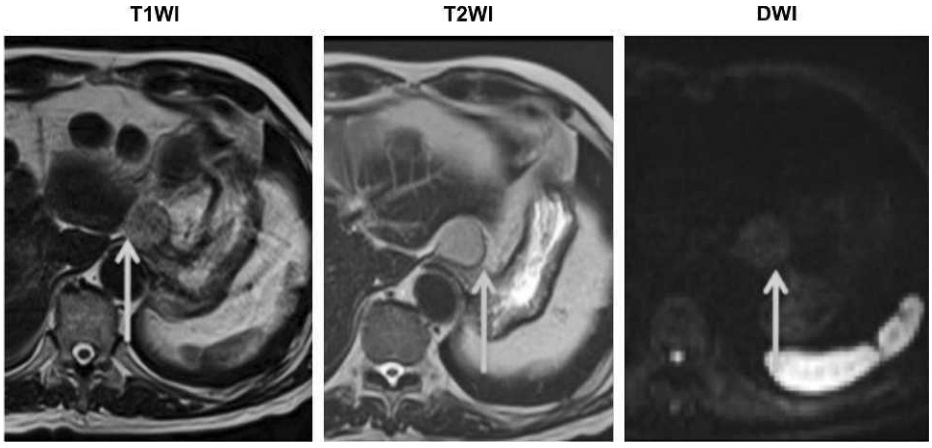

Fig. 2 Findings from abdominal MRI with contrast enhancement. The tumor in the gastric cardia shows homogenous high intensity on T2W1 images and isointensity on T1W1 images.

the nearest clinic, where computed tomography (CT) imaging revealed a 3-cm mass in the anterior wall of the gastric cardia. He was referred to our hospital for further investigation in October 2009, and surgery for the gastric submucosal tumor was scheduled.

A physical examination on admission identified superficial lymph nodes of the neck that were impalpable. They were detected in CT scan. The abdomen was flat and soft with no tenderness. Laboratory tests did not show an increase in the levels of inflammatory response and tumor markers (Table 1).

Abdominal CT showed a tumorlike lesion of 32 mm in diameter in the anterior gastric cardia, with soft tissue density, homogenous internal region, and a clear margin (Fig. 1). Abdominal magnetic resonance imaging (MRI) showed a mass of the gastric cardia with homogenous high intensity on T2W1 images, and isointensity on T1W1 images. The tumor was not found to have high intensity on diffusion-weighted images, but showed a smooth surface and was found to be strongly adherent to the gastric cardia wall (Fig. 2).

Endoscopic examination of the upper digestive tract could not confirm the findings of the abnormal mass on the lesser curvature of the gastric cardia, as seen on the CT and MRI images (Fig. 3).

Based on the above findings that CT shows homogenous internal region without cystlike structure, the differential diagnosis was gastrointestinal stromal tumor, lymphangioma, or double intestinal tract. Laparoscopic partial gastrectomy was carried out.

The tumor was in close proximity to the gastric cardia, and it protruded into the lesser omentum cavity. The lesser omentum was opened, and the structure surrounding the tumor was removed. A bright yellow and viscous cyst fluid was discharged from the cyst wall. The mass was excised as far as 5 mm from the esophageal wall along the direction of the lesser curvature. An automatic sewing machine

Fig. 3 Endoscopic findings of the upper digestive tract. Endoscopic examination could not confirm the presence of the lesion (observed in the CT images) in the lesser curvature.

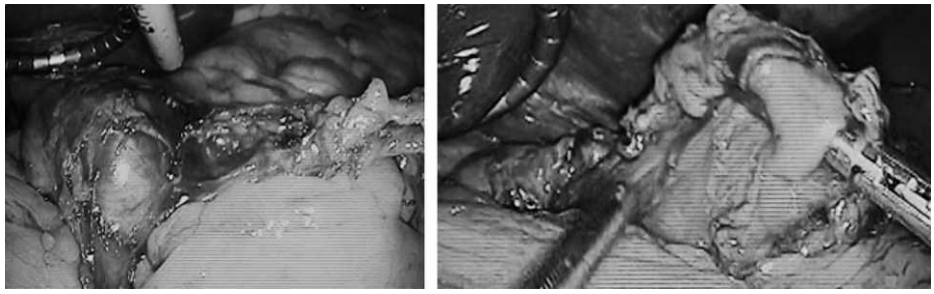

Fig. 4 Intraoperative findings. The tumor is located in proximity to the gastric cardia, and it protrudes into the lesser omentum cavity. The cystic fluid is yellow, bright, and viscous.

(Echelon 60; Ethicon Endo-Surgery, Cincinnati, Ohio) was used for suturing (Fig. 4).

Microscopic examination showed that the mass was a cystic tissue lined with typical bronchial epithelium. A bronchogenic cyst was considered. A pseudostratified ciliated epithelium was found in the thin-wall (0.5–1 mm thick) cyst lumen. Further, unlike the esophageal cyst, squamous epithelium and Auerbach nerve plexus were not found, and a dual muscular layer was also not present (Fig. 5).

On postoperative day 3, the patient had fever and cough, and he was confirmed positive for type A influenza. Only a liquid diet was allowed on postoperative day 4. From postoperative day 5, the patient started having proper meals. The patient was discharged without any complications. No recurrence has been observed after 1 year after surgery.

Discussion

Bronchogenic cysts are usually located in the retroperitoneal space; their occurrence in the abdominal cavity is extremely rare. So far, only 6 bronchogenic cysts involving the gastric wall examined using CT have been reported in the English literature.[4–9] They were searched on PubMed using the terms "gastric" and "bronchogenic cyst" (Table 2).

There are no specific symptoms for this cyst, and in this case the patient did not have any related pain.

Bronchogenic cysts are typically observed on CT images as a mass with a smooth surface and clear margin. Homogenous density is seen in the internal region of the mass. In addition, the MRI findings are typical of cystic lesions, such as low intensity on T1 and high intensity on T2 images. However, it is usually difficult to establish a definitive diagnosis based on imaging studies.

In the 6 cases of cysts involving the gastric wall found in the literature, which employed CT imaging, they were all found with low-density masses. MRI was conducted in 1 case, where the cyst showed low intensity on the T1 image and high intensity on the T2 image.

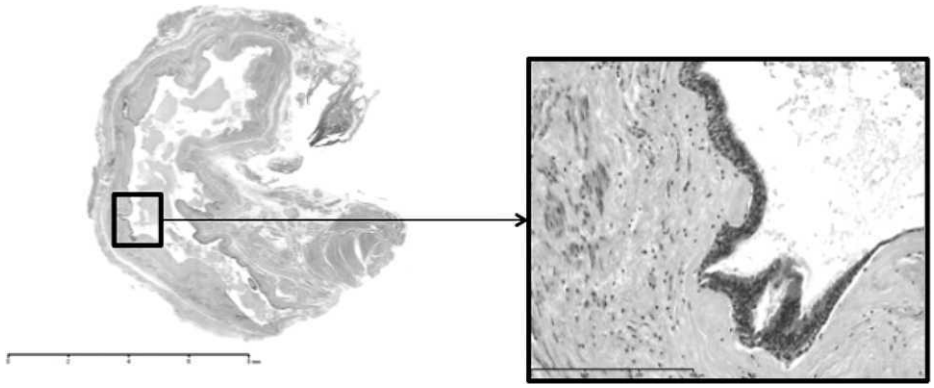

Fig. 5 Histopathologic findings. Hematoxylin-eosin staining (left: loupe image-micrograph; right: original magnification ×200). Pseudostratified ciliated epithelium, which is similar to bronchial epithelium, is found in the 0.5- to 1-mm–thick lumen of the thin-wall cyst. Squamous epithelium, Auerbach nerve plexus, and dual muscular layers were not observed.

Table 2 List of the published English literature on bronchogenic cysts involving the gastric wall diagnosed using CT images

	Year	Author's name	Age, y	Sex	Size, mm	Chief complaint	Location	CT	MRI	Single/ multiple	Preoperative diagnosis	Treatment
1	2003	Hedayati *et al*[4]	59	F	70 × 50	No symptom	Posterior gastric cardia	Low density	N/A	Single	Adrenal incidentaloma	Laparotomy
2	2005	Song *et al*[5]	62	F	35 × 25	No symptom	Lesser curvature–gastric cardia	Low density	N/A	Single	Gastric neurogenic tumor or reactive lymph nodes	Laparotomy
3	2006	Lee *et al*[6]	38	F	70 × 50	No symptom	Lesser curvature–gastric cardia	Low density	N/A	Single	Gastric submucosal tumors	EMR
4	2008	Sato *et al*[7]	60	F	30	No symptom	Lesser curvature–upper gastric	Low density	N/A	Single	N/A	Observation
5	2010	Tan *et al*[8]	30	F	51 × 46	No symptom	Posterior gastric wall	Low density	N/A	Single	N/A	Laparoscopic surgery
6	2011	Ubukata *et al*[9]	81	F	26	No symptom	Gastric lesser curvature	Low density	T1 low intensity T2 high intensity	Single	Congenital cysts	Laparotomy
7	2012	Present case	71	M	32	Throat discomfort	Anterior wall of lesser curvature–gastric cardia	Low density	T1 isointensity T2 high intensity	Single	Gastric submucosal tumors	Laparoscopic surgery

EMR, endoscopic mucosal resection; N/A, not applicable.

In this report, the lesion did not have the typical appearance of a cyst on CT and MRI. Further, because it had adhered to the gastric wall and CT showed homogenous internal region without cyst-like structure due to internal viscosity, it was preoperatively diagnosed as a gastric submucosal tumor. Bronchogenic cyst was diagnosed based on histopathologic examination of the excised specimen. Bronchogenic cysts are extremely difficult to diagnose from physical and radiologic examination. Even in the previous reports on 6 bronchogenic cysts, none of them were successfully diagnosed from CT images only.

Histopathologic diagnosis of bronchogenic cysts is based on the presence of normal bronchial tissue in the cysts, such as ciliated columnar epithelium, mucous gland, smooth muscle fiber, fibrous tissue, elastic tissue, and cartilage. However, very few cysts show all of these components.[10] Incomplete defective form of bronchogenic cysts—those without cartilage and glandular tissue—are often hard to differentiate from esophageal cysts, gastroduodenal cysts, and ectopic bronchial pulmonary sequestration. Cartilage and glandular tissue were also not found in this case. The mass was diagnosed as a bronchogenic cyst from the presence of pseudostratified, ciliated, typical bronchial epithelium in the 0.5- to 1-mm specimen of the thin-wall cyst lumen, and from the absence of squamous epithelium, Auerbach nerve plexus, and dual muscular layer, unlike in esophageal cysts.

The risk of recurrence is high in cases with only cyst reefing, and complete surgical resection is necessary.[11–13] Patients who underwent complete resection were reported to have no recurrence.[14]

Even though the incidence of bronchogenic cysts in the abdomen is extremely low, bronchogenic cysts should be considered in the differential diagnosis of abdominal unilocular cystic diseases, which are difficult to diagnose preoperatively.

References

1. Sander TW. *Langman Human Developmental Biology.* 8th ed. Tokyo, Japan: Medical Science International; 2001:241–249
2. Braffman B, Keller R, Gendal ES, Finkel SI. Subdiaphragmatic bronchogenic cyst with gastric communication. *Gastrointest Radiol* 1988;**13**(4):309–311
3. Maier HC. Bronchogenic cyst of the mediastinum. *Ann Surg* 1948;**127**(3):476–502
4. Hedayati N, Cai DX, McHenry CR. Subdiaphragmatic bronchogenic cyst masquerading as an "adrenal incidentaloma". *J Gastrointest Surg* 2003;**7**(6):802–804

5. Song SY, Noh JH, Lee SJ, Son HJ. Bronchogenic cyst of the stomach masquerading as benign stromal tumor. *Pathol Int* 2005;**55**(2):87–91
6. Lee SH, Park DH, Park JH, Kim HS, Park SH, Kim SJ *et al*. Endoscopic mucosal resection of a gastric bronchogenic cyst that was mimicking a solid tumor. *Endoscopy* 2006;**38**(suppl 2): E12–E13
7. Sato M, Irisawa A, Bhutani MS, Schnadig V, Takagi T, Shibukawa G *et al*. Gastric bronchogenic cyst diagnosed by endosonographically guided fine needle aspiration biopsy. *J Clin Ultrasound* 2008;**36**(4):237–239
8. Tan KK, Nandini CL, Ho CK. A case of gastric bronchogenic cyst in Singapore with multiple intrigues. *ANZ J Surg* 2010; **80**(4):286–287
9. Ubukata H, Satani T, Motohashi G, Konishi S, Goto Y, Watanabe Y *et al*. Intra-abdominal bronchogenic cyst with gastric attachment: report of a case. *Surg Today* 2011;**41**(8): 1095–1100
10. Bagwell CE, Schiffman RJ. Subcutaneous bronchogenic cysts. *J Pediatr Surg* 1998;**23**(11):993–995
11. Otsuka N, Ookuma T, Hongo H, Sada A, Sugihara S, Miyauchi Y *et al*. A case diagnosed with adenocarcinoma in mediastinal bronchial cysts [in Japanese]. *Clin Cancer* 1985;**31**(15):1941
12. Murphy JJ, Blair GK, Fraser GC, Ashmore PG, LeBlanc JG, Sett SS. Rhabdomyosarcoma arising within congenital pulmonary cysts, report of three cases. *J Pediatr Surg* 1992;**27**(10):1364–1367
13. Matsuo S, Uchiyama Y, Kimino K, Yamaoka N, Akamine S, Tsuji K. Analysis of cases underwent bronchogenic cystectomy [in Japanese]. *Oita Med Mag* 1990;**19**:52–57
14. Nagatani N, Kashiwabara T, Ooki A, Kawakami F, Okuno G, Yamamoto M *et al*. A case of intraperitoneal bronchial cyst with high amylase fluid diagnosed with ultrasonography [in Japanese]. *J Pathol* 1996;**93**(8):594–598