

## Diagnostic Challenge to Distinguish Gastric Duplication Cyst from Pancreatic Cystic Lesions in Adult

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### Abstract

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We report herein a 63-year-old female with gastric duplication cyst (GDC), of which resected specimen was histologically shown to be composed of gastric foveolar epithelium and thin bundles of smooth muscle. Computed tomography revealed a thin-walled cystic lesion surrounded by the pancreatic tail, spleen, left kidney, and the stomach. Magnetic resonance imaging demonstrated a thin layer between the cyst and either the spleen or kidney, successfully excluding the possibility that the cyst originated from these organs. Endoscopic ultrasonography failed to show a smooth muscle bundle in the cyst wall, which is a diagnostic finding for GDC. Even retrospectively, these preoperative findings could not distinguish GDC from pancreas-originating cystic lesions. Despite the recent advances in diagnostic imaging modalities, preoperative diagnosis of GDC in adults remains difficult due in part to its rarity and the absence of characteristic findings.

**Key words:** gastric duplication, cyst, endoscopic ultrasonography

(DOI: [10.2169/internalmedicine.46.0009](https://doi.org/10.2169/internalmedicine.46.0009))

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### Introduction

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Gastric duplication cyst (GDC) is a congenital anomaly that is rarely seen in adults (1). Approximately a half of the patients with GDC were found within the first year after birth with various symptoms such as vomiting, abdominal pain, and/or weight loss, and more than 70% of reported cases have been found at less than 12 years of age (1). Ladd and Gross (2) and Rowling (3) reported the essential features of GDC as follows: 1) the cyst wall is contiguous with the stomach; 2) it is surrounded by a smooth muscle coat, that fuses with the muscle layer of the stomach; and 3) it is lined with alimentary epithelium.

We encountered a Japanese female with GDC that was preoperatively diagnosed as a pancreatic cyst. A variety of imaging modalities failed to indicate GDC before the operation. We present herein the clinical course of the case and discuss the difficulties and problems in diagnosing GDCs.

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### Case Report

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A 63-year-old female was admitted without any symptom

to our hospital for the examination and treatment of a developing abdominal cyst. At the age of 56, she began suffering from right renal tuberculosis. The abdominal computed tomography (CT) revealed a low-density mass with a maximum diameter of 34 mm in the left upper abdomen that was suspected to be a pancreatic cyst. The cyst size had not increased on periodic CT for years, but the latest CT demonstrated an enlargement of the cyst (measuring 45 mm in diameter) over the previous year (Fig. 1).

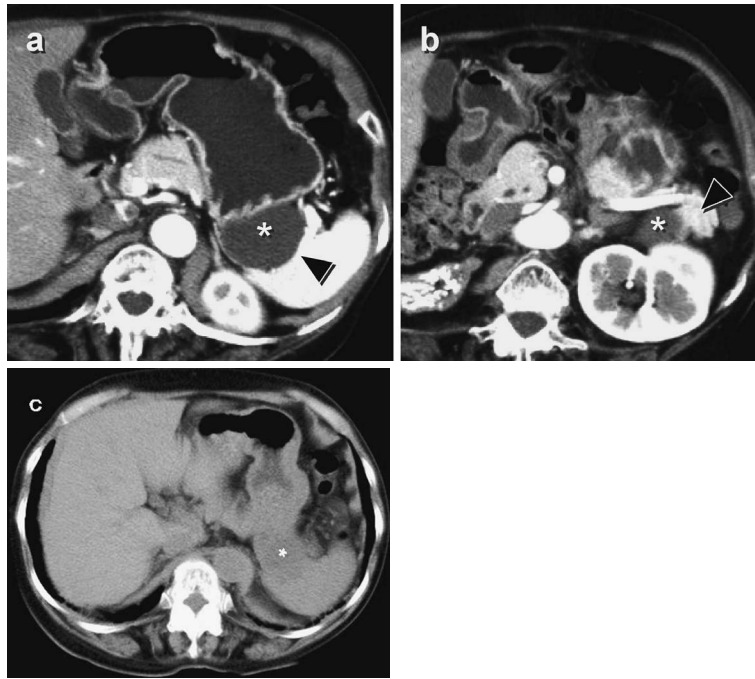
On admission, physical examination revealed no palpable mass on her abdomen and no vertebral anomalies. Blood tests showed normal levels of hepatobiliary and pancreatic enzymes, renal function, and tumor markers, including carcinoembryonic antigen and CA19-9. Enhanced CT demonstrated a cystic lesion surrounded by the greater curvature of stomach, spleen, right kidney, and pancreatic tail (Fig. 1). The cyst did not come into direct contact with the left adrenal gland. The cyst wall was thin without any detectable nodule or septa. Magnetic resonance imaging (MRI) demonstrated the cyst content to be low intensity on T1-weighted images and relatively high intensity on T2-weighted images (Fig. 2). It is noteworthy that MRI demonstrated a thin layer between the cyst and either the spleen or kidney, and thus

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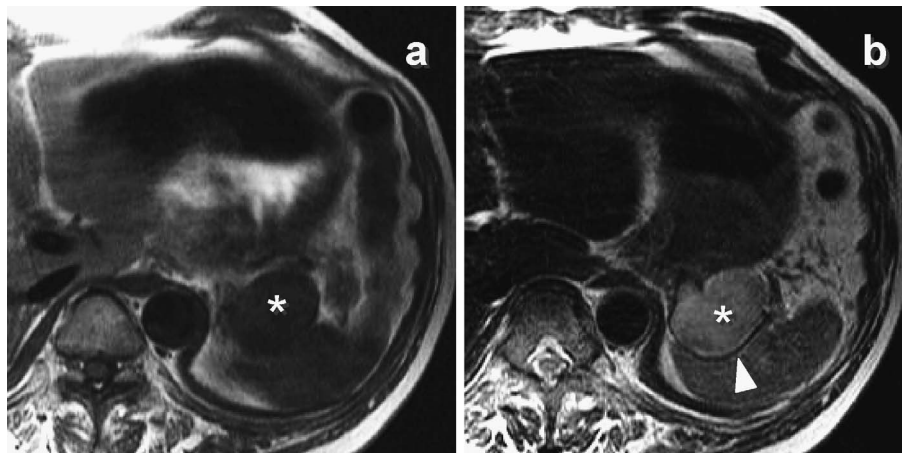
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Received for publication January 9, 2007; Accepted for publication April 17, 2007

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**Figure 1.** Enhanced CT demonstrates a cystic lesion (indicated by asterisks) surrounded by the greater curvature of the stomach, spleen, right kidney, and pancreatic tail. The cyst does not contact with the left adrenal gland. The cyst seems to be contiguous with the spleen (a, arrowhead) and pancreas (b, arrowhead). The plain CT taken one year before the operation shows the same cyst with the maximum diameter of 3.4 cm (c, indicated by an asterisk).

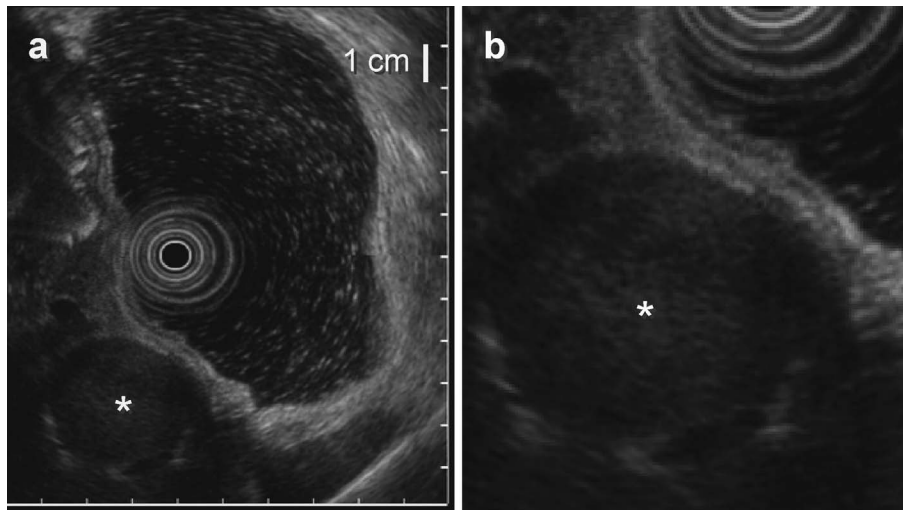


**Figure 2.** MRI demonstrated the cyst content (indicated by asterisks) to be low intensity on T1-weighted images (a) and relatively high on T2-weighted images (b). It is of note that a thin layer between the cyst and spleen is seen on T2-weighted images (arrowhead).

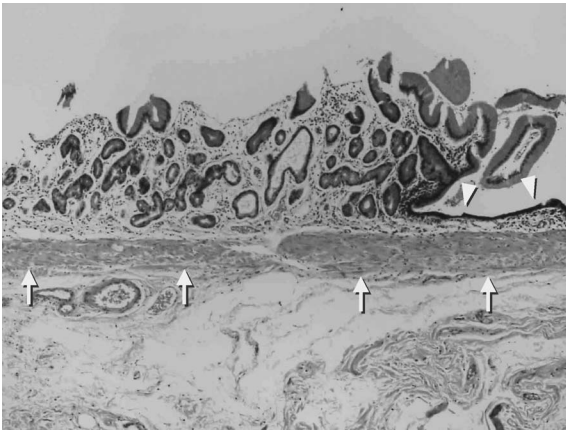
ruled out the possibility that the cyst was derived from these organs (Fig. 2-a; arrowhead). Endoscopic retrograde pancreatography revealed no particular findings such as communication between the cyst and the pancreatic duct system. Endoscopic ultrasonography (EUS) showed that the cyst was walled with a thin echogenic layer and filled with a sludge-like material (Fig. 3), which was growing outward from the pancreas tail. The proper muscle layer of the gastric wall adjacent to the cyst was well preserved, but there was no detectable hypoechoic interlayer in the cyst wall. These findings indicated that the cyst was a mucinous cystic tumor of the pancreas rather than GDC.

Considering the malignant potential of the cystic lesion based on its increasing size, a distal pancreatectomy was planned. On laparotomy, the cyst was unexpectedly found to be contiguous to the greater curvature of stomach and easily separated from the pancreas, indicating the gastric origin of the cystic lesion. The cyst was subsequently totally resected with partial gastrectomy. The postoperative course was uneventful, and the patient was discharged on the 10th postoperative day.

The gross observation of the resected specimen showed a cyst 45 mm in maximum diameter with a clear margin. There was no communication between the cyst lumen and



**Figure 3.** EUS demonstrates a cystic lesion (indicated by asterisks) posterior to the stomach (a) and shows that the cyst is encapsulated by a thin echogenic layer and fulfilled with sludge-like material (b). The structure of the gastric wall is well-preserved. There is no solid component within the cyst.



**Figure 4.** Photomicrograph of the cyst wall. The inner wall of the cyst is lined by ciliated pseudostratified epithelium (arrowheads) and gastric foveolar epithelium with metaplastic change. The cyst wall contains subjacent thin bundle of smooth muscle (arrows).

the stomach. The pathological examination revealed that the inner wall of the cyst was lined by ciliated pseudostratified epithelium and, in part, gastric foveolar epithelium with metaplastic change (Fig. 4). The cyst wall contained subjacent thin bundles of smooth muscle that appeared contiguous in part with the proper muscle layer of the stomach. No malignant change was observed. From these findings, the cyst was diagnosed as GDC.

## Discussion

GDCs are rarely seen in adults, because the majority of cases are diagnosed in childhood (1, 3). In adults, GDCs are incidentally discovered with ultrasonography, CT scan, or gastric endoscopy (4, 5). Because most cases occur along the greater curvature of the stomach (1), the cysts are found

to compress the adjacent organs such as the pancreas, kidney, spleen, and adrenal gland. Accordingly, the differential diagnosis would include cysts derived from these organs. Misdiagnosis of GDCs has been reported as pancreatic pseudocyst, pancreatic mucinous cystadenoma, and splenic cyst (4-7).

Although it is difficult to obtain a preoperative diagnosis of GDC, recent imaging modalities have provided some informative findings. Contrast-enhanced CT typically demonstrates GDC as a thick-walled cystic lesion with enhancement of the inner lining (8, 9). Calcification is occasionally observed on CT (6, 10). These findings are of diagnostic significance for GDCs. However, since mucinous cystic tumors of the pancreas also show similar radiological features, cases with GDCs adjoining the pancreas are indistinguishable from pancreatic mucinous cystic tumors based on these CT findings. Moreover, because the wall is sometimes thin as seen in the present case, enhancement of the inner cyst wall is not always demonstrated.

In the present case, CT did not provide informative findings regarding the cyst origin. However, MRI revealed a thin layer lying between the cyst and either the spleen or kidney on T2-weighted images and successfully eliminated the possibility of a cyst from these organs. Generally, MRI can provide additional information about the cyst content compared to CT findings. However, the nature of the fluid in the GDC was reported to differ in each case according to bleeding, chronic inflammation, or infection (8). Therefore, MRI seems to be of less significance than expected in diagnosing GDCs.

EUS is useful in distinguishing between the intramural and extramural lesions of the stomach. When EUS demonstrated an echogenic internal mucosal layer and a hypoechoic intermediate muscular layer of the cyst, GDCs was reported to be highly likely (8). However, the multi-layered cystic wall is not always depicted by EUS, and diagnosis

may be difficult in such cases. Also in this case, the smooth muscle bundle of the cyst wall was so thin that EUS could not demonstrate it as a low echoic layer.

In previously reported cases, aspiration of the cyst content was attempted and sometimes showed high concentrations of CA19-9 and/or carcinoembryonic antigen in its content, making the diagnosis of GDC rather complex (6, 11). According to reports of cases for which endoscopic fine needle aspiration biopsy was carried out, diagnosis is relatively easier when the pseudostratified ciliated epithelium was obtained (12). However, the sensitivity, specificity, and diagnostic accuracy of endoscopic fine needle aspiration biopsy

for GDCs are not known. Further, in several cases, carcinoma was reported to be derived from the epithelial cells lining the cyst wall (7). Together with the risk of infection and bleeding, the potential risk of disseminating the malignant cells along the needle tract and into the peritoneum should always be considered (13).

In conclusion, we encountered a patient with GDC that was proven histologically in the resected specimen. Even though a panel of imaging modalities is available, it is still difficult to obtain a preoperative diagnosis of GDC. In cases with a cystic lesion adjoining the gastric wall, we should consider GDC as a candidate for differential diagnosis.

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