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Successful laparoscopic partial gastrectomy and spleen-preserving distal pancreatectomy for gastric duplication cyst connecting with the pancreatic tail

Hiromichi Kawaida^{a,*}, Ayako Kimura^a, Mitsuaki Watanabe^a, Hidenori Akaike^a, Naohiro Hosomura^a, Yoshihiko Kawaguchi^a, Hidetake Amemiya^a, Makoto Sudo^a, Hiroshi Kono^a, Masanori Matsuda^a, Hideki Fujii^a, Daisuke Ichikawa^a, Mitsuharu Fukasawa^b, Ei Takahashi^b, Katsuhiko Sano^c, Tomohiro Inoue^d

^a First Department of Surgery, Faculty of Medicine, University of Yamanashi, Japan

^b First Department of Internal Medicine, Faculty of Medicine, University of Yamanashi, Japan

^c Department of Radiology, Faculty of Medicine, University of Yamanashi, Japan

^d Department of Human Pathology, Faculty of Medicine, University of Yamanashi, Japan

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ABSTRACT

INTRODUCTION: Gastrointestinal duplication cyst is a congenital rare disease that may occur in any region from mouth to anus. Among them, gastric duplication cysts are very rare.

CASE REPORT: Here we report A 23-year-old Japanese man who visited our hospital to evaluate an abdominal tumor. Abdominal computed tomography showed a well-circumscribed homogenous low-density mass measuring 6.2 × 6.0 cm between the pancreatic tail and the upper posterior wall on the gastric greater curvature, and the mass seemed to originate from the pancreatic tail. We found intraoperatively that the mass adhered to the stomach and pancreatic tail strongly, so we performed laparoscopic partial gastrectomy and spleen-preserving distal pancreatectomy. Pathological findings showed that the lining epithelium of the cystic mass consisted of the gastric foveolar epithelium with fundic glands. Furthermore, the pancreatic tissue of the pancreatic tail and the muscular layer of the cystic mass were intermingled.

DISCUSSION: GDCs are usually diagnosed at a younger age and in adults, they are very rare. Therefore, surgical resection is considered to be the best treatment due to the difficulty of diagnosis, and also that it mimics a pancreatic cystic tumor, and malignant transformation. Complete resection of the cyst is the ideal technique and laparoscopic surgery should be selected whenever possible.

CONCLUSION: We experienced a case of GDC continuous to both stomach and pancreatic tail. Laparoscopic surgery is safety and useful even if GDC is continuous with both the stomach and the pancreas.

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1. Introduction

Gastrointestinal duplication cyst is a congenital rare disease that may occur in any region from mouth to anus. Among them, it most commonly occurs in the ileum and ileocecum and gastric

duplication cysts(GDC) are very rare [1,2]. Due to their position and mass effect, GDCs are usually diagnosed in a younger age. The most common location of GDCs is along the greater curvature [3] and continuation to both stomach and pancreas is extremely rare. We experienced an adult case of a Gastric duplication cyst(GDC) continuous to both stomach and pancreas, on which laparoscopic surgery was performed.

2. Case report

A 23-year-old Japanese man visited our hospital to evaluate an abdominal tumor detected incidentally by MRI. He is a radiologist. As MRI's machine was newly purchased, he underwent MRI examination as a test. The patient had no past medical history and the abdomen was flat and no definite mass was palpable. Laboratory studies showed within normal range, and carbohydrate antigen

* Corresponding author at: First Department of Surgery, Faculty of Medicine, University of Yamanashi, 1110 Shimokato, Chuo-shi, Yamanashi, 409–3898, Japan.

E-mail addresses: kawaidah@yamanashi.ac.jp (H. Kawaida), ainoue@yamanashi.ac.jp (A. Kimura), mitsuakiw@yamanashi.ac.jp (M. Watanabe), hakaike@yamanashi.ac.jp (H. Akaike), naohiroh@yamanashi.ac.jp (N. Hosomura), ykawa@yamanashi.ac.jp (Y. Kawaguchi), hamemiya@yamanashi.ac.jp (H. Amemiya), msudoh@yamanashi.ac.jp (M. Sudo), hkouno@yamanashi.ac.jp (H. Kono), masam@yamanashi.ac.jp (M. Matsuda), hfuji@yamanashi.ac.jp (H. Fujii), dichikawa@yamanashi.ac.jp (D. Ichikawa), fmitsu@yamanashi.ac.jp (M. Fukasawa), etakahashi@yamanashi.ac.jp (E. Takahashi), snkthr@yahoo.co.jp (K. Sano), tomohiroi@yamanashi.ac.jp (T. Inoue).



Fig. 1. (a,b) CT shows a well-circumscribed homogenous low-density mass measuring 6.2×6.0 cm (dotted arrow) between the upper posterior wall on the gastric greater curvature (thick arrow) and the pancreatic tail (thin arrow). The mass seems to originate from the pancreatic tail. The pancreatic tail stretches to the caudal side.

19-9 or carcinoma embryonic antigen was unremarkable. Abdominal ultrasonography (AUS) showed a low echoic lesion measuring 5.6×4.5 cm adjacent to the stomach. Abdominal computed tomography (CT) showed a well-circumscribed homogenous low-density mass measuring 6.2×6.0 cm between the pancreatic tail and the upper posterior wall on the gastric greater curvature, indicating that the mass originated from the pancreatic tail (Fig. 1a,b). The wall of the mass was not enhanced, and the pancreatic tail stretched to the caudal side (Fig. 1b). Magnetic resonance imaging (MRI) showed a homogenous low intensity mass on T1-weighted imaging and a homogenous high intensity mass on T2-weighted imaging. Magnetic resonance cholangio-pancreatography showed no communication between the tumor and main pancreatic duct. Endoscopic ultrasonography (EUS) demonstrated a cystic mass adjacent to the stomach wall showing a heterogenous low-echoic

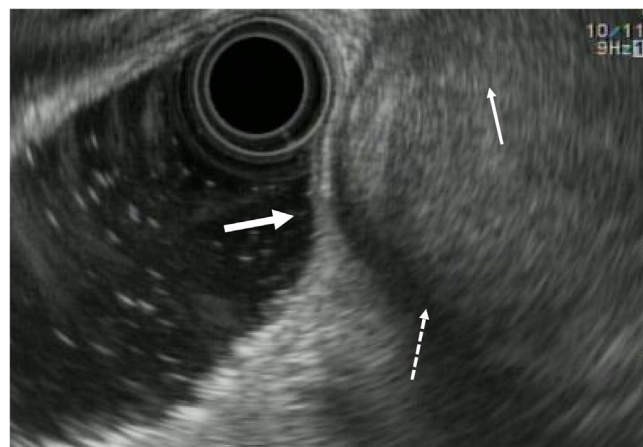


Fig. 2. EUS shows a cystic mass adjacent to the stomach wall (thick arrow) and a heterogenous low-echoic lesion (dotted arrow) is circumscribed by a high-echoic thin wall.

lesion circumscribed by a high-echoic thin wall (Fig. 2). From these findings, the tumor was suspected to be a gastrointestinal duplication cyst arising from the pancreatic tail or gastric wall; however, mucinous cystic neoplasm could not be ruled out.

To achieve complete resection, we decided to perform laparoscopic extirpation. We created a pneumoperitoneum through the umbilicus by open technique and a 10-mm trocar under direct vision and a total of five trocars were used. Intraoperatively, a soft 6.0 cm \times 6.0 cm mass adhered to the upper posterior wall on the gastric greater curvature and shared a smooth lining with the surface of the stomach (Fig. 3a). Furthermore, the mass was tightly adhered to the pancreas (Fig. 3b). Since it was difficult to separate the tumor from both the stomach and the pancreas, we performed laparoscopic partial gastrectomy and spleen-preserving distal pancreatectomy. Both connecting parts between the cyst and the stomach wall, and between the pancreatic tail were transected with using Endo GIA™ Reloads with Tri-Staple™. The specimen was placed in a bag (Endo Catch Gold, Covidien, Mansfield, MA, USA) and removed via an umbilicus incision.

The postoperative course was uneventful and the patient was discharged on the fifth postoperative day.

Pathological findings

Macroscopically, the specimen was a unilocular cyst, $6.0 \times 5.6 \times 4.0$ cm in size (Fig. 4) and the cyst did not communicate with the gastric lumen or pancreatic duct. The lining epithelium consisted of gastric foveolar epithelium with fundic glands (Fig. 5a). Furthermore, pancreatic parenchyma of the pancreatic tail and muscular layer of the cystic mass were intermingled (Fig. 5b). The cyst had no communication with the pancreatic duct.

3. Discussion

Gastrointestinal duplication cysts may occur in any region from mouth to anus and are present in 1 out of 4 500–10 000 live births [4]. Among them, they most commonly occur in the ileum and ileocecum. GDCs are very rare and account for 3.8–17% of gastrointestinal duplications [1]. Due to their position and mass effect, GDCs are usually diagnosed in a younger age and 80% of patients are under the age of 12. The most common location of GDCs is along the greater curvature [3] and continuation to both the stomach and the pancreas is extremely rare.

Despite the development of laparoscopic surgery with advances in equipment and technology, our search in the English-language literature revealed only 10 cases of laparoscopic surgery for GDC (Table 1. [5–13]). Furthermore, to the best of our knowledge, this

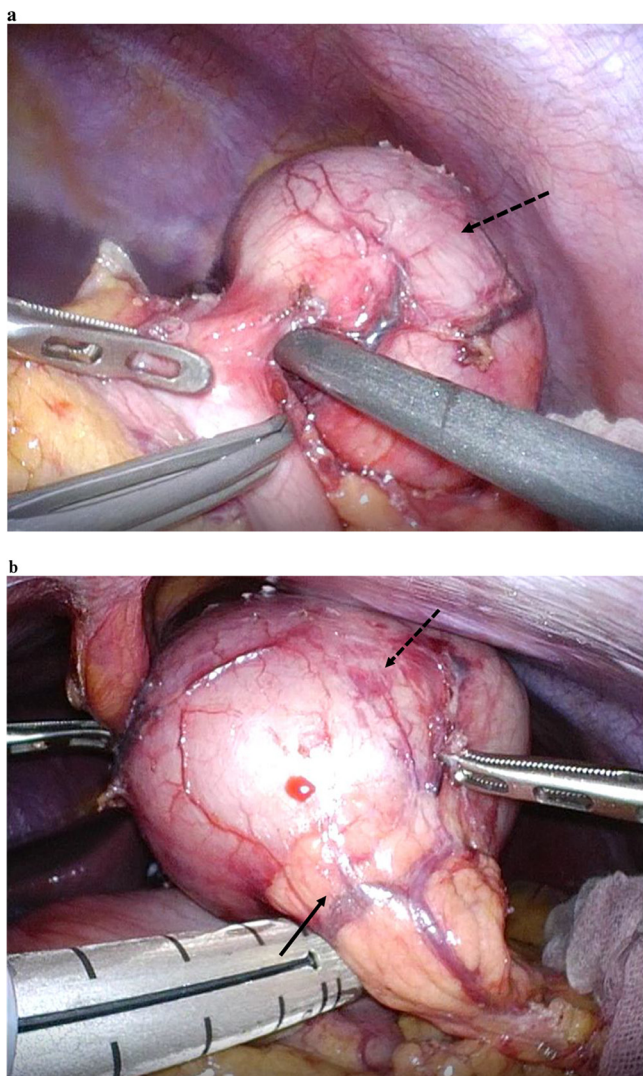


Fig. 3. (a) A soft 6.0 cm × 6.0 cm mass (dotted arrow) adheres to the upper posterior wall on the gastric greater curvature (thick arrow) and it shares a smooth layer with stomach. (b) The mass (dotted arrow) is tightly adhered to the pancreas (thin arrow).



Fig. 4. The specimen was a unilocular cyst and measured 6.0 × 5.6 × 4.0 cm in size. Thick arrow shows the cutting line of the stomach and thin arrow shows the pancreatic tail.

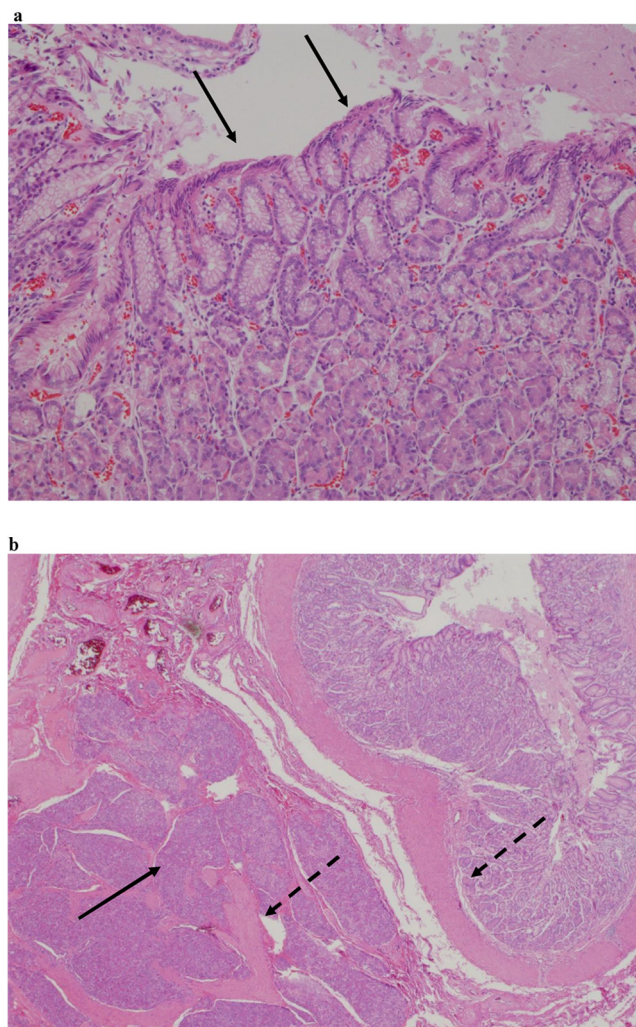


Fig. 5. (a) The lining epithelium consisted of the gastric foveolar epithelium with fundic gland (thin arrow) (HE × 200). (b) Pancreatic tissue of the pancreatic tail (thin arrow) and muscular layer of the cystic mass (dotted arrow) are intermingled (HE × 200).

is the first report of a laparoscopic partial gastrectomy and spleen-preserving distal pancreatectomy for GDC in the English literature.

GDCs have no specific symptoms or signs. Depending on their location, symptoms may be vague abdominal complaints, recurrent abdominal pain, nausea, vomiting, epigastric fullness, weight loss, anemia, dysphagia, dyspepsia, inflammation, pancreatitis. However, in adults, GDCs are usually asymptomatic [14].

Because of their position and mass effect, GDCs are usually diagnosed at a younger age with approximately 67% of cases confirmed in the first year of life. In adults, they are very rare [1,2] and the diagnosis may be difficult despite numerous imaging modalities such as abdominal ultrasonography(AUS), endoscopic ultrasonography(EUS), contrast-enhanced CT, MRI. However, technetium-99 m imaging will identify duplications containing gastric mucosa [14]. AUS has been reported to be effective, that the “double wall sign” represents the hyperechoic inner layer representing the submucosa surrounded by a hypoechoic outer layer representing the smooth muscle is suggestive of GDCs [1]. EUS is useful for demonstrating the relation between the cyst and gastric wall [5]. In our case, we also could demonstrate the relation between the cyst and gastric wall, but could not visualize the pancreatic tail. Contrast-enhanced CT may demonstrate GDC as a thick-walled cystic lesion with enhancement of the inner lining [15], but sometimes it shows a nonspecific

Table 1
Clinicopathological data for reported cases of laparoscopic surgery for a GDC.

No	Sex	Age(yr)	Size(cm)	Location	Symptoms	Operation	Pathological findings	References
1	F	14	unknown	The posterior wall of the cardiac stomach	Epigastric tenderness	Cystectomy	Squamous epithelium and glandular epithelium	Sasaki T et al. [28]
2	M	45	6.0	The lesser curvature of the gastroesophageal junction	Epigastric discomfort	Cystectomy	A smooth muscle wall with unclassified mucosal lining	Machado MA et al. [29]
3	F	2 mo	2.2	The greater curvature of the stomach in the gastroesophageal junction	Unknown	Enucleation	Mucinous columnar gastric epithelium	Ford WD et al. [30]
4	M	37	5.0 × 5.0	The lesser curvature of the stomach in the gastroesophageal junction	Upper abdominal pain	Partial gastrectomy	Pseudostratified ciliated epithelium	Wakabayashi H et al. [31]
5	M	42	5.2 × 4.5	The lesser curvature of the stomach	Pain in the left lumbar region	unknown	Pseudostratified ciliated columnar epithelium	Mardi K et al. [32]
6	F	3	4.5 × 3.5 × 3.0	The tail of the pancreas	Left lateral abdominal pain	Enucleation	Gastric foveolar epithelium, fundic gland, and pyloric gland	Kohno M et al. [18]
7	M	56	5.0 × 3.0 × 3.0	The anterior of the gastroesophageal junction	None	Partial gastrectomy	Gastric foveolar epithelium with cardiac gland and pseudostratified ciliated columnar epithelium	Napolitano V et al. [27]
8	M	2 mo	8.0 × 3.0 × 3.0	The greater curvature of the stomach	Vomiting	Partial gastrectomy	Gastric epithelium	Takazawa S et al. [33]
9	M	23	4.5 × 4.0.4.0	The posterior wall of the fundus	None	Partial gastrectomy	Pseudostratified ciliated columnar epithelium and gastric epithelium and cardiac glands	Laurent S et al. [34]
10	F	28	8.0 × 5.0 × 5.0	The greater curvature of the stomach	Epigastric tenderness	Partial gastrectomy	Gastric epithelium	Thomopoulos T et al. [35]
Present	M	23	6.0 × 5.6 × 4.0	The greater curvature of the stomach and pancreatic tail	None	Partial gastrectomy and distal pancreatectomy	Gastric foveolar epithelium with fundic gland	Present case

GDC: gastric duplication cyst; M: male; F: female; yr: year; mo: month.

cystic structure [15]. Magnetic resonance imaging does not seem to improve diagnostic accuracy significantly [5]. To obtain further information, there are reports of endoscopic needle aspiration being performed [5]. However, this procedure may cause complications, such as hemorrhage, peritoneal dissemination if the tumor is malignant.

These malformations are believed to be congenital. Gastric duplication cysts result from abnormal foregut development. The etiology of alimentary duplications is unknown, although the two most widely held hypothesis to explain gastrointestinal duplications are Bremer's defect in recanalization and McLetchie's neuroenteric band theory [16,17]. Kohno, et al. classified GDCs into four categories based on whether it has communication with the pancreatic duct and contiguity with the stomach: those without any communication with the pancreatic duct, noncontiguous with the stomach (category 1); those without communication with the pancreatic duct, contiguous with the stomach (category 2); those with communication with the pancreatic duct, noncontiguous with the stomach (category 3); and those with communication with the pancreatic duct, contiguous with the stomach (category 4) [18]. Cases of GDC contiguous with the stomach and heterotopic pancreas have been reported previously, but this is the first case contiguous with the stomach and pancreatic tail.

Surgical resection is considered to be the best treatment due to the difficulty of diagnosis, and also that it mimics a pancreatic cystic tumor [18], and malignant transformation [19]. Complete resection of the cyst is the ideal technique achieved by both open and laparoscopic approaches [7]. However, With the progress of

devices, safety of distal pancreatectomy is increasing. Laparoscopic surgery is less invasive and is becoming standard surgery, and should be selected whenever possible.

4. Conclusion

We experienced a case of GDC continuous to both stomach and pancreatic tail. Laparoscopic surgery is safety and useful even if GDC is continuous with both the stomach and the pancreas.

Conflict of interest

Hirromichi Kawaida and the other co-authors have no conflict of interests to declare.

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Ethical approval

Given that this is a case report with no identifiable information included in the manuscript, ethical approval was not obtained. Our institution has been exempted ethical approval on case reports.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request [20].

Author contributions

Hiromichi Kawaida, Hideki Fujii, and Daisuke Ichikawa designed the research and wrote the paper, and Masanori Matsuda, Ayako Kimura, Mitsuaki Watanabe, Hidenori Akaike, Naohiro Hosomura, Yoshihiko Kawaguchi, Hidetake Amemiya, Makoto Sudo, Hiroshi Kono, Mitsuharu Fukasawa, Ei Takahashi, Katsuhiko Sano, and Tomohiro Inoue contributed to collect and analyze the data.

Registration of research studies

I already registered this case report.
Research registry No 3297

Guarantor

Hiromichi Kawaida
First Department of Surgery, Faculty of Medicine, University of Yamanashi
1110 Shimokato, Chuou-shi, Yamanashi, 409-3898 Japan

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