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Asymptomatic adenocarcinoma arising from a gastric duplication cyst: A case report

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ABSTRACT

INTRODUCTION: Duplication of the alimentary tract is a relatively uncommon congenital anomaly and most cases occur in childhood. Malignancy arising from a gastric duplication cyst is extremely rare. We herein report a very rare case of malignant transformation of a gastric duplication cyst.

PRESENTATION OF CASE: A 47-year-old asymptomatic Japanese woman was referred to our hospital with a large abdominal mass adhered to the stomach. Since there was a possibility of malignant transformation, complete resection of the cyst and segmental gastrectomy without regional lymphadenectomy were performed.

DISCUSSION: To our knowledge, this is the 2nd report of asymptomatic adenocarcinoma arising from a gastric duplication cyst in the English-language literature. Unfortunately, the patient developed peritoneal metastasis and ascites seven months after the surgery and died.

CONCLUSION: From our long-term follow-up experience of this gastric duplication cyst, we recommend making accurate diagnosis as soon as possible with biopsy using endoscopic ultrasonography. When the disease is diagnosed as malignant, we recommend gastrectomy with lymphadenectomy. Even if the disease is diagnosed as benign, we recommend close observation with imaging modalities.

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1. Introduction

Duplication of the alimentary tract is a relatively uncommon congenital anomaly that can occur anywhere from the oral cavity to the anus. Gastric duplications are quite rare (incidence of 4%–8%), and most cases occur in childhood [1]. Malignancy arising from a gastric duplication cyst is extremely rare; only 10 cases have been reported in the English-language literature to date (Table 1: Ref No. [2–7]). Some reports suggest their poor prognosis [6,8]. We herein describe the clinical and pathological features of this disease through a case report.

2. Case report

A 42-year-old Japanese woman was referred to our hospital for evaluation of a large abdominal mass that had been found incidentally on ultrasonography during an annual health examina-

tion. Her medical history was unremarkable. Physical examination revealed an elastic-soft mass palpable in her left upper abdominal quadrant, but she was asymptomatic. All laboratory test results were normal except for a CA19-9 concentration of 72.4 U/ml (normal, <37 U/ml). Ultrasonography revealed a 10 cm homogeneous cyst adjacent to the greater curvature of the stomach, pancreas, spleen, and left adrenal gland. Computed tomography also revealed the cyst, which was well-defined and had a homogeneous density (Fig. 1a). We followed up this large cyst every 6 months with computed tomography because the patient was asymptomatic (Fig. 1b). At 46 years of age, she developed a cerebellar infarction and began taking acetylsalicylic acid every morning. At 47 years of age, she was still asymptomatic, but computed tomography revealed mural nodules in the cyst (Fig. 1c) and an increased CA19-9 concentration of 167.1 U/ml. Mural nodules are a well-known sign of malignancy in association with some types of pancreatic tumors. We therefore recommended that the patient undergo advanced examinations and surgical treatment; however, she refused. Six months later, she agreed with our recommendation and underwent further examinations.

Computed tomography and ultrasonography showed an increased number of mural nodules containing calcification, but the cystic lesion had slightly decreased in size (Fig. 1d). No lym-

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Table 1
Clinicopathological data in cases of carcinoma arising from gastric duplication cysts.

Reference	Age/Sex	Symptoms	Histology	Size (cm)	Extension	Follow-up (month)
Mayo et al. [2]	64/F	Weakness, anorexia	ADC	6	Wall of the stomach and cyst	NED/12
Coit and Mies [3]	50/M	Vomiting	Epithelial malignancy	17	NA	NA
	72/F	Abdominal pain	ADC	3.2	Wall of the stomach and cyst	NED/72
Kuraoka et al. [4]	40/M	Fever, back pain	ADC	7	Wall of the stomach and cyst	Liver metastasis/7
	71/F	Abdominal pain, anorexia	ADC	8	Wall of the cyst	NED/1
	56/M	Vomiting, weight loss	ADC	10	Wall of the cyst	NED/28
Home et al. [5]	40/M	Abdominal pain	NEC	12	Wall of the cyst	Multiple metastases/14
Barussaud et al. [6]	67/F	Abdominal mass, weight loss	Mixed ADC and SCC	NA	Wall of the stomach and cyst	DFD/6
Zheng et al. [7]	25/M	Asymptomatic	ADC	8	Wall of the stomach and cyst	NED/13
Present case	47/F	Asymptomatic	ADC	10	Wall of the stomach and cyst	Peritoneal metastasis/7

Age is presented in years.

F: female; M: male; NA, not available; ADC: adenocarcinoma; NED: no evidence of disease; NEC: neuroendocrine carcinoma; SCC: squamous cell carcinoma.

phadenopathy or ascites was noted, and her CA19-9 concentration had decreased to 58.6U/ml. Magnetic resonance imaging (MRI) showed multiple mural nodules and septa separating pockets of serous and hemorrhagic fluids. Our preoperative differential diagnoses were bronchogenic cyst, gastric duplication cyst, and pancreatic pseudocyst.

Laparotomy revealed a large cystic mass strongly adhered to the greater curvature of the stomach but to neither the pancreas nor left adrenal gland. We performed complete resection of the cyst and partial wedge resection of the stomach. We did not perform regional lymphadenectomy.

Gross pathology showed a 10.0 × 9.5 × 8.0 cm unilocular cystic mass adherent to the greater curvature of the stomach (Fig. 2a). The cyst contained abundant dark mucilaginous fluid and necrotic tissue. There was no direct communication between the cyst lumen and the gastric lumen. The cyst was granular and had a relatively uniform thickness except for six mural nodules that projected into the cystic lumen (Fig. 2b).

Microscopic examination showed that the cyst was located in the muscularis propria of the stomach and that the mural nodules were composed of moderately differentiated adenocarcinoma (Fig. 3a and b). The adenocarcinoma cells invaded the whole thickness of the cyst wall and serosa of the stomach and extended laterally on the surface of the cyst (Fig. 3c, red arrows). However, a few non-neoplastic gastric glands were found in a small part of the cyst wall (Fig. 3c, blue arrows). Immunohistochemical studies revealed that MUC expression in the epithelium was consistent with that of the gastric glands: positive for MUC5AC and MUC6 and negative for MUC2 (Fig. 3d). These findings indicated that the cyst was a gastric duplication cyst, and that the tumor was an adenocarcinoma arising from the cyst.

Seven months later, the patient developed ascites due to peritoneal metastasis with gross elevation of her serum CEA and CA19-9 concentrations. After recurrence, she underwent oral chemotherapy (TS-1). Two months after she began the chemotherapy she died of cancerous peritonitis.

3. Discussion

Alimentary tract duplications are rare congenital anomalies that most frequently occur in the ileum (35%). They occur in the stomach with an incidence of only 4%–8%. Most duplications are diagnosed during the first year of the life; they are rarely diagnosed in adults [9,10]. Several theories have been proposed for the pathogenesis of alimentary tract duplications, but most researchers believe that abnormal recanalization after the solid epithelial stage of embryonic bowel development is the most likely mechanism [1]. The following morphological criteria are used for diagnosis of these rare anomalies: First, generally speaking, they are spheric cysts

or tubular structures located in or immediately adjacent to part of gastrointestinal tract. Second, they are surrounded by at least one coat of smooth muscle, fusing with the muscularis propria of the alimentary canal (usually all three muscle layers are present without a plane of cleavage between the cyst and the alimentary canal). Finally, they are lined by typical gastrointestinal mucosa [9]. The present case met the first and second above-mentioned morphological criteria for gastric duplication cysts.

Gastric duplications are most frequently located at the greater curvature of the stomach, as in the present case, followed by the posterior wall, lesser curvature, anterior wall, and pylorus [7,11]. These lesions may or may not communicate with the gastric lumen. In our case, the cyst did not communicate with the gastric lumen.

The associated symptoms are nonspecific and include nausea, vomiting, epigastric pain, and weight loss [12]. However, our patient had been clinically asymptomatic until surgery. She is the second reported patient who was asymptomatic until surgery [7].

Differentiation of a duplication cyst from other cystic lesions is often difficult. Such lesions include a pseudo cyst of the pancreas [13], bronchogenic cyst, epithelial inclusion cyst of the spleen, and GIST [5]. In fact, our preoperative differential diagnoses were bronchogenic cyst and duplication cyst.

This is the 11th report of adenocarcinoma arising from a gastric duplication cyst in the English-language literature (Table 1). To our knowledge, this is the second asymptomatic patient to be reported. And our case added some kind of findings about the tumorigenesis of this carcinoma in gastric duplication cyst.

Despite undergoing complete resection, the patient developed metastasis in 7 months. We speculate that there were already micro-metastases that could not be detected macroscopically at the time of laparotomy because the cancer cells invaded to the gastric serosa pathologically. On the other hand, the tumor size may have no relationship with the malignant change of the duplication cyst, because the size of these malignant tumor varied from 3.2 cm to 17 cm as shown in Table 1. Taken together, we strongly recommend having an accurate diagnosis with biopsy using endoscopic ultrasonography as soon as possible once mural nodules can be seen in the cyst wall. When the disease is diagnosed as malignant with biopsy, subtotal or total gastrectomy with regional lymphadenectomy should be performed because of their poor prognosis as shown in Zheng et al. [7] and as in the present case [6,8,13]. Alternatively, very close observation should be performed even when the disease is diagnosed as benign.

4. Conclusion

A case of adenocarcinoma arising within a gastric duplication cyst has been presented. To our knowledge, this is the second report of an asymptomatic patient with adenocarcinoma in a gastric dupli-

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