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A rare case of enlarged gastric heterotopic pancreas with retention cysts: A case report and literature review

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ABSTRACT

INTRODUCTION: Gastric heterotopic pancreas (HP) is usually asymptomatic and benign; however, it may become evident when it is complicated by pathological changes such as inflammation, bleeding, and malignant transformation.
PRESENTATION OF CASE: A 43-year old man was diagnosed with gastric HP 18 years prior suffered a haemorrhage from the enlarged gastric HP with multiple cystic lesions. Although endoscopic ultrasonography–guided fine needle aspiration showed no malignancy, he underwent a partial gastrectomy for diagnosis and treatment. Postoperative histological findings revealed ectopic pancreatic tissue with retained cysts that consisted of dilated pancreatic ducts without malignancy.
DISCUSSION: This is a first report of enlarged gastric HP due to the expansion of retained cysts. Gastric HP is rarely enlarged by pathological changes including inflammation, retention cysts, or malignant neoplasms.
CONCLUSION: Symptomatic enlarged gastric HP should be respected and further examined histologically to ensure diagnostic accuracy.

1. Introduction

Heterotopic pancreas (HP) is defined as pancreatic tissue occurring in the upper gastrointestinal tract that lacks anatomical or vascular communication with the normal pancreas. HP is a relatively common congenital anomaly that is reported in 1–2% of autopsy cases [1]. Although the majority of HP cases are asymptomatic and benign, symptoms or bleeding may be caused by malignant transformation or inflammation [2].

Here we report a case of an enlarged and bleeding gastric HP caused by retention cysts within HP tissue. We also conduct a literature review of studies on enlarging gastric HP. This work is reported according to SCARE criteria [3].

2. Presentation of case

A 43-year-old healthy man was admitted to our hospital with recurrent episodes of melena; he had no relevant medical history. Physical examination findings were normal except for anaemia, as his haemoglobin level was 8.1 g/dL. Other findings including tumor markers such as carcinoembryonic antigen and cancer antigen 19-9 were also within the normal range. Eighteen years prior, a submucosal tumor measuring 25 mm at the posterior wall of the gastric angle was revealed by esophagogastroduodenoscopy (EGD), and he was diagnosed with gastric HP by fine needle aspiration (FNA). Five years prior, the tumour was documented to be the same size as it was originally (Fig. 1a). In our physical examination, EGD revealed that the submucosal tumour had erythematous mucosa and erosion measuring 40 mm in diameter on the posterior wall of the gastric angle (Fig. 1b). Contrast-enhanced computed tomography revealed multiple cystic lesions with wall thickening on the lesser curvature of the stomach (Fig. 2a). Endoscopic ultrasonography (EUS) showed multiple cystic formation within the third layer of the gastric wall (Fig. 2b). EUS-FNA revealed non-atypical crypt epithelium and stroma without malignancy. Magnetic resonance cholangiopancreatography confirmed hypointense multiple cystic lesions with thin septations on T2-weighted images. No mural nodule or solid component was detected in any of the cysts (Fig. 2c). Although these findings did not indicate that the tumour was malignant HP, the clinical findings that the gastric HP was larger than it had been five years prior and had caused haemorrhaging suggested the coexistence of a malignant tumour or inflammation.

Abbreviations: HP, heterotopic pancreas; EGD, esophagogastroduodenoscopy; FNA, fine needle aspiration; EUS, endoscopic ultrasonography.

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Table 1
Review of enlarged gastric heterotopic pancreas.

Author	Age	Sex	Symptom	Follow-up period (year)	Surgical procedure	Heinlich type of HP	Component in HP
Phillip [9]	80	Male	Dyspepsia	13	Antrectomy	NR	IPMN
Fukumori [10]	76	Male	Weight loss	2	Partial gastrectomy	2	Pancreatic cancer
Okamoto [11]	75	Female	Pain	2	NR	1	Invasive ductal carcinoma
Straatman [12]	61	Female	Dysphagia	3	Partial gastrectomy	1	Pancreatitis
Present case	43	Male	Melena	5	Partial gastrectomy	1	Retention cysts

Abbreviations: HP, heterotopic pancreas; IPMN, intraductal papillary mucinous neoplasm; NR, not record.

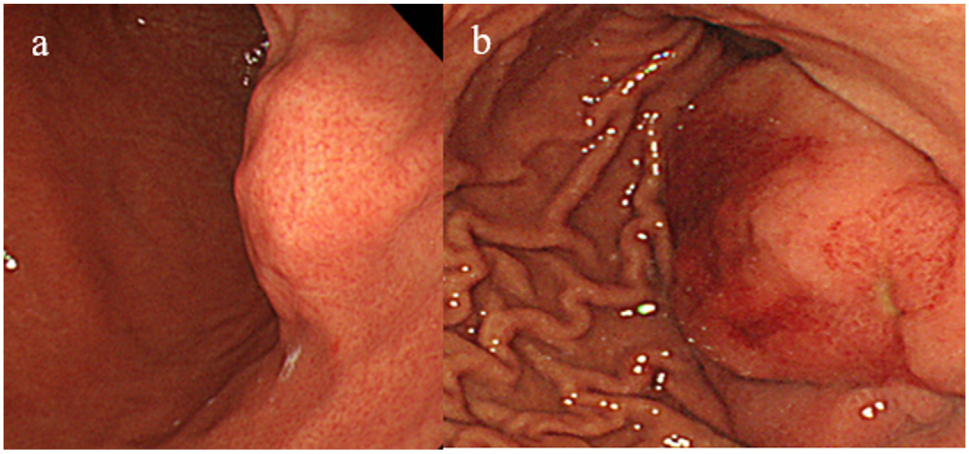

Fig. 1. a) Endoscopic findings at 5 years prior. A subepithelial mass with smooth surface measuring 4.0 × 3.0 cm is visible on the posterior wall of the distal antrum. b) A subepithelial mass measuring 4.0 × 3.0 cm at the same site. Erythematous discoloration with erosion is visible on the surface of the mass.

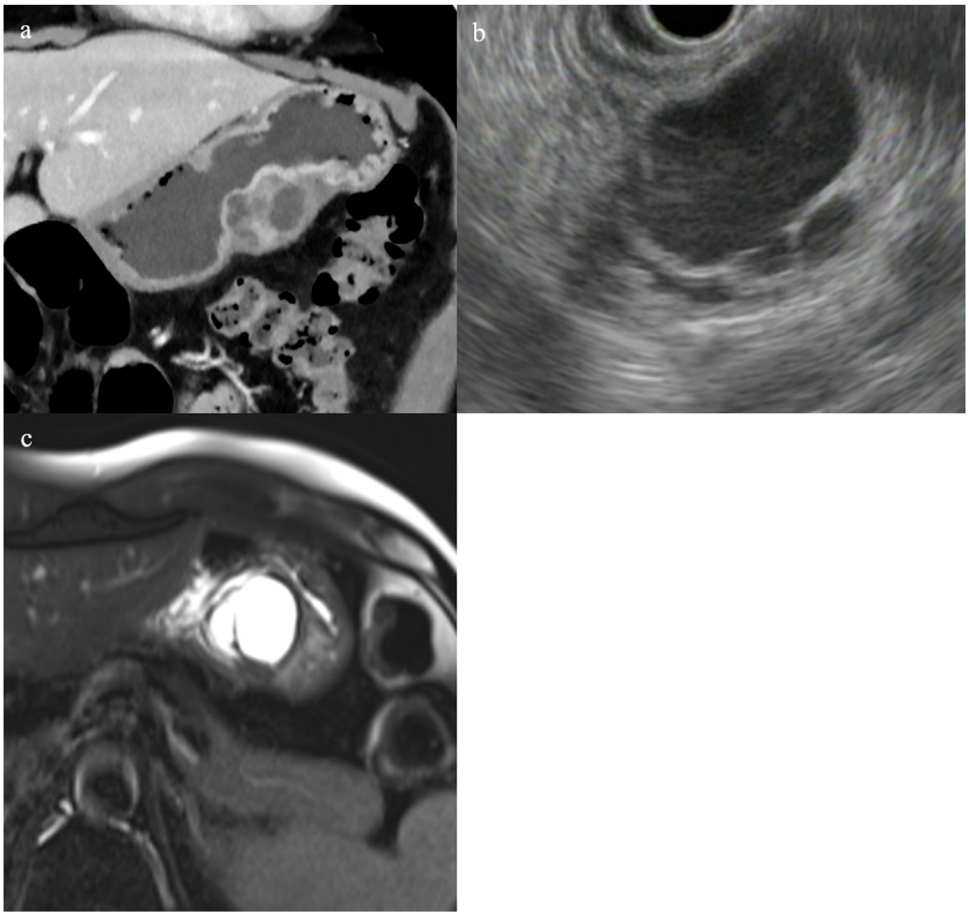

Fig. 2. a) Contrast-enhanced computed tomography scan of the portal phase showing a round mass with multiple cystic lesions on the lesser curvature of the stomach. b) Endoscopic ultrasound image showing multiple cystic formation located in the third layer of the gastric wall. c) Magnetic resonance T2-weighted image showed a hypointense mass with hyperintense multiple cystic lesions.

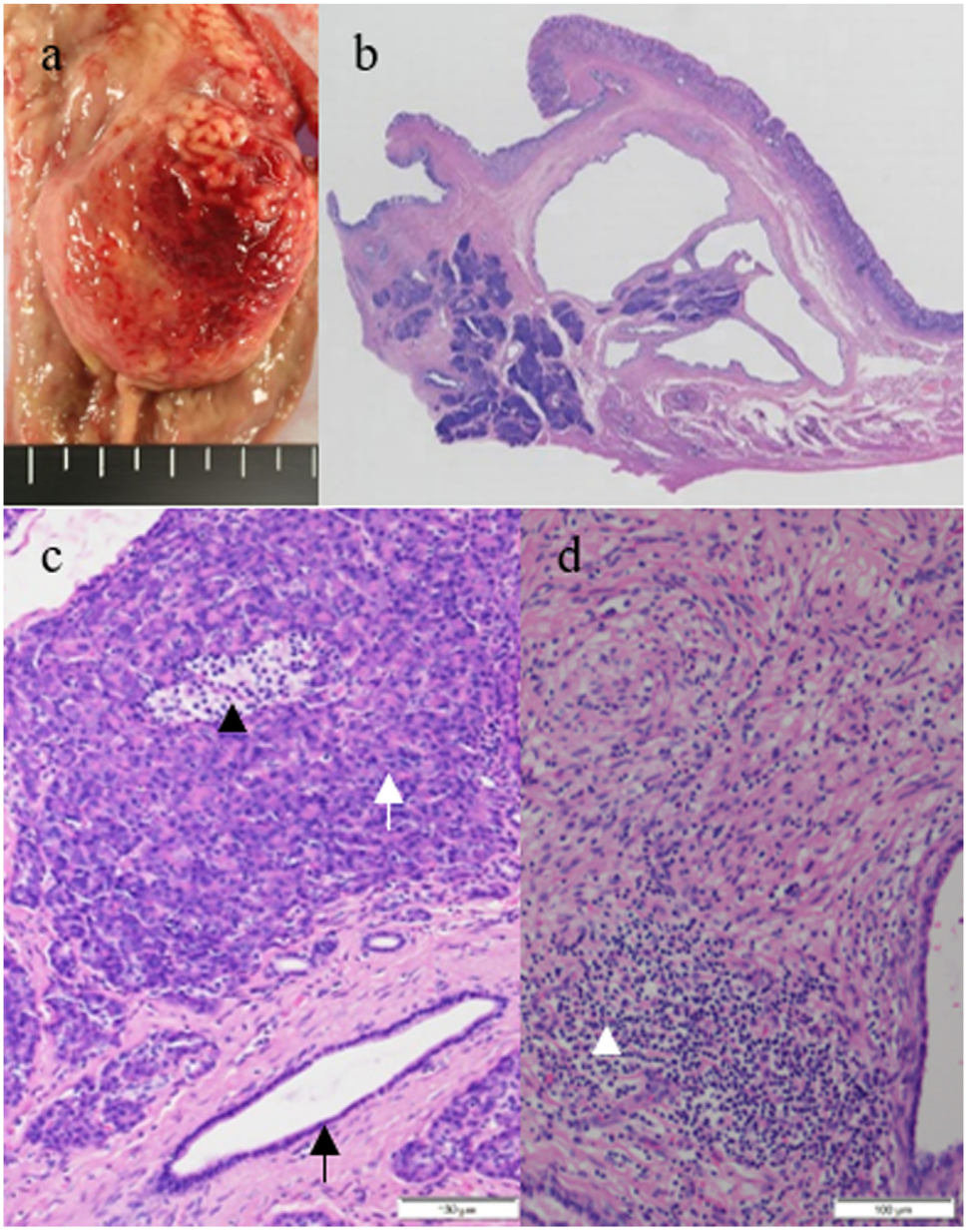

Fig. 3. Pathological findings. a) Tumour specimen. b) Hematoxylin and eosin staining revealing a heterotopic pancreas with multiple dilated cysts lined by columnar epithelium (original magnification, ×10). c) Pancreatic lobules with acini (white arrow), ducts (black arrow), and islets of Langerhans cells (black triangle) (original magnification, ×100). d) Periductal chronic inflammation with some dense fibrous tissue and lymphocyte invasion indicative of chronic inflammation (white triangle) but no acute inflammation such as pancreatitis (original magnification, ×100).

Hence, the patient underwent laparotomy for diagnosis and treatment. Surgical findings showed that the tumour was palpable, elastic, and soft in the gastric wall without serous invasion. We performed a partial gastrectomy. The intraoperative frozen section analysis revealed no malignancy; thus, a lymph node dissection was unnecessary. Macroscopically, the resected specimen showed a tumour (40 × 30 mm) covered with smooth gastric epithelial mucosa (Fig. 3a).

The postoperative histological examination of the specimen revealed ectopic pancreatic tissue with a partially cystic component. The mass was located at the submucosa and part of the muscularis propria of the stomach. Ectopic pancreatic tissue consisted of acinar and ductal structures with scattered islets of Langerhans cells (Heinlich type 1). The cystic components were retention cysts that consisted of dilated pancreatic ducts without malignancy (Fig. 3c). We could not confirm communication between the retention cysts and gastric lumen. Periductal chronic inflammation with some dense fibrous tissue and lymphocyte invasion indicated chronic inflammation but no acute inflammation like pancreatitis (Fig. 3d). The postoperative period was uneventful and he was discharged on postoperative day eight.

3. Discussion

HP is defined as pancreatic tissue that lacks anatomical or vascular communication with the normal body of the pancreas [1]. Histologically, HP is classified as Type 1, composed of acini, duct and islets; Type 2, composed of ducts only; Type 3, consisting of acini only (exocrine pancreas); and Type 4, composed of islets only (endocrine pancreas) [4].

HP most commonly occurs in the upper gastrointestinal tract, with occasional cases reported in the ileum, mediastinum, bile

ducts, gall bladder, fallopian tubes, splenic hilum, omentum, and lungs [4]. Gastric HP is discovered in the antrum in 85–95% cases on the posterior or anterior wall, being more common along the greater curvature [5].

Endoscopically, HP is identified as a submucosal tumour; the differential diagnosis is necessary from other gastric submucosal tumours such as gastrointestinal stromal tumours, schwannomas, and malignant lymphomas [6]. Endoscopic biopsy may be difficult since gastric HP is typically covered with non-malignant epithelial mucosa [7]. Although tissue sampling of the submucosal tumour using EUS-FNA could aid in establishing the accurate preoperative diagnosis, it cannot completely rule out malignancy.

Gastric HP is generally asymptomatic and can be followed up without surgical excision. However, its function is like a normal pancreas; hence, practically any pancreatic pathology can occur within gastric HP, including enlargement due to pancreatitis or neoplastic transformation. Symptoms due to enlargement may include ulceration, bleeding, obstruction, and intussusception. Gastric HP patients with these symptoms and suspected malignancy should undergo surgery for symptom relief and to ensure diagnostic accuracy [8].

Here we reviewed a case of enlarged gastric HP. We performed a systemic search of the PubMed database for the English-language literature up to June 2020 using the search term 'heterotopic pancreas' and 'ectopic pancreas'. Our review of the literature identified only five cases including ours published to date (Table 1) [9–12]. The mean patient age was 75 (43–80) years; three patients were men (60%). All cases had certain symptoms and were treated with resection. Histological diagnoses revealed malignant neoplasms in three cases (60%), pancreatitis in one case, and a benign tumour in one case (our case). Four cases (80%) consisted of type 1 or 2 HP, which include the pancreatic ducts.

Our patient had been diagnosed with asymptomatic gastric HP by EUS-FNA 18 years prior and only now experienced lesion enlargement, multiple cystic changes, and haemorrhaging from the gastric mucosa. Although EUS-FNA showed no malignancy, surgery was recommended to alleviate symptoms and exclude the presence of a malignant lesion. Since intraoperative frozen section findings revealed no malignancy, we were able to forego lymphadenectomy. The final histological findings revealed that enlargement of the gastric HP was caused by expansion of retention cysts.

Retention cysts are cystically dilated pancreatic ducts induced by duct obstruction [13]. Such obstructions are caused by fibrous- or duct-obstructing tumours [14–17]. Gananadha et al. reported the first case of retention cysts in HP at the distal oesophagus. Although the enlargement was caused by pancreatitis, the authors concluded the importance and difficulty of the excluding malignancy [18]. They also emphasized that frozen section was useful for excluding malignancy and avoiding major surgery.

The present case is the first report of an enlarged retention cysts in gastric HP. Causes other than malignant tumours or acute inflammation may obstruct communication between the ectopic tissue and the gastric lumen, leading to an enlarged retention cysts. Especially, type 1 or 2 gastric HP, which include pancreatic ducts, might be susceptible to the development of enlarged retention cysts.

4. Conclusions

Here we reported a rare case of enlarged gastric HP caused by retention cysts. Symptomatic enlarged gastric HP should be respected and further examined histologically to ensure diagnostic accuracy.

Declaration of Competing Interest

None of the authors have any commercial or financial involvement in connection with this study that represents or appears to represent any conflicts of interest.

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Ethical approval

This research was conducted in accordance with the regulations of the Hiroshima City Hiroshima citizens hospital ethics committee.

Consent

Written informed consent has been obtained from the patient for the publication of this case report and any accompanying images.

Author contribution

KM and MI participated in treatment of the patient, collected case details, literature search and draft the manuscript. TM performed the pathological analysis. The other co-authors participated in treatment planning of the patient. MI participated in treatment planning of the patient and helped to draft the manuscript. MO contributed to study concept, and review of the final manuscript. All authors read and approved the final manuscript.

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