Premalignant Lesion of Heterotopic Pancreas Combined with Gastritis Cystica Profunda in Gastric Fundus

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INTRODUCTION

Heterotopic pancreas, also known as ectopic pancreas, is found mainly in the stomach, duodenum, or jejunum. Pancreatic intraepithelial neoplasia (PanIN) is the non-invasive precursor of pancreatic cancer and gastritis cystica profunda (GCP) is considered a precursor of gastric cancer. As with most putative cancer precursor lesions, the diagnosis and treatment of these lesions has been controversial. A patient with no history of gastric surgery visited our institution for a regular evaluation. Endoscopy showed a 2 x 2 cm sized, protruding mass lesion with overlying normal mucosa on the fundus of stomach. Endoscopic ultrasound (EUS) and computed tomography (CT) led to the possible diagnosis of a gastrointestinal stromal tumor with cystic change. Laparoscopic gastric wedge resection was performed with intra-operative endoscopic guidance. Microscopic examination identified the mass as pancreatic tissue. Furthermore, it demonstrated PanIN, grade 3 (PanIN-3) mixed pancreatobiliary and intestinal type, arising in the heterotopic pancreas and associated with GCP. This report describes a rare case of a PanIN lesion combined with GCP as precursors of precancerous lesions in heterotopic pancreas and stomach.

Key words: heterotopic pancreas — gastritis cystica profunda — PanIN — stomach.

CASE REPORT

A 64-year-old asymptomatic male with no history of gastric surgery visited our institution for a regular evaluation. He had a history of hypertension with medication. The physical examination was unremarkable. When the esogastroduodenoscope was introduced into the stomach, a 2 x 2 cm sized, protruding mass lesion with overlying normal mucosa was seen on the fundus of the stomach (Fig. 1). Endoscopic ultrasound (EUS) revealed a multi-septated, not well-demarcated mass, measuring 2.3 x 2.0 cm (Fig. 2). In addition, the lesion was shown to have a heterogeneous echogenicity, mixed hypoechoic solid component with anechoic component, arising from muscularis propria interface. The laboratory study was not significant, with normal CEA, CA19-9 and pancreatic amylase level. Computed
tomography (CT) scan of the abdomen also demonstrated a submucosal multi-septated cystic mass at the gastric fundus (Fig. 3). No perigastric or periesophageal lymph nodes were identified. The pancreas showed no abnormal findings. A preoperative evaluation led to the possible diagnosis of a GIST with cystic change. A laparoscopic gastric wedge resection was performed with intraoperative endoscopic guidance due to the short distance to the gastroesophageal junction of the mass. Microscopic examination identified the mass as pancreatic tissue. Furthermore, it demonstrated pancreatic intraepithelial neoplasia, grade 3 (Pan IN-3), mixed pancreatobiliary and intestinal type, arising in the heterotopic pancreas including acinar glands and pancreatic ducts and associated with GCP (Fig. 4). The patient's postoperative recovery was uneventful.

**DISCUSSION**

Heterotopic pancreas is a relatively uncommon lesion that may be found incidentally during surgery or endoscopy. These lesions can be found virtually anywhere along the gastrointestinal tract, but typically involved sites are the stomach, duodenum, or jejunum. In the stomach, it is most commonly located in the distal stomach along the greater curvature of the gastric antrum, in contrast with our case, located at the fundus [7]. Heterotopic pancreas is usually asymptomatic and benign. It can often be mistaken for GIST or leiomyoma because it is difficult to diagnose definitively even though invasive endoscopic biopsy and its radiologic characteristics are similar to US and CT scanning [8]. Additionally, the heterotopic pancreas functions like a normal pancreas, so the same pathology that affects the pancreas can affect the ectopic pancreatic tissue, such as pancreatitis, cysts, pseudocysts, and malignant transformation to ductal adenocarcinoma. Therefore, complete surgical resection should be performed when heterotopic pancreas is found incidentally during surgery or when it is symptomatic, in order to prevent complications [9].

PanINs are defined as neoplastic epithelial proliferations in the smaller caliber pancreatic ducts and are divided into PanIN-1 to PanIN-2 to PanIN-3 based upon the degree of architectural and nuclear atypia present [10]. PanIN has been known to be the precursor to invasive carcinoma, histologically and genetically progressing to invasive ductal adenocarcinoma in the pancreas [11-13]. Fukumori et al reported a case of ectopic pancreatic cancer developing from the ectopic pancreas in the stomach [14]. Though treatment policy for PanIN has not been established, complete surgical resection should be performed, since it is considered a rare but surgically curable, localized disease with a good prognosis following radical resection [15].
Gastritis cystica profunda is a rare gastric lesion characterized by the presence of gastric glands in the submucosa of the stomach with normal overlying mucosa and is often mistaken for other more common gastric pathologies [16, 17]. An unspecified mucosal insult such as a history of gastric surgery is widely accepted as the cause for GCP genesis. Some authors suggest that GCP may develop secondary to chronic inflammation, foreign body reaction or ischemic injury, in order, interruption of the muscularis mucosa, migration of epithelial cells into the submucosal layer, and cystic dilatation [3]. However, in our case, there is no history of gastric surgery and no definitive evidence of inflammation in the heterotopic pancreas. Although significant debate has ensued over the malignant potential of these lesions, more recent reports describe dysplastic changes within the submucosal glands of GCP, suggesting an adenocarcinoma precursor lesion [3, 4]. Deery et al reported a case of gastric adenocarcinoma developing in association with GCP in patients without previous gastric surgery [18]. Though a defined treatment strategy for GCP has not been well described due to the rarity of the lesions, those should be managed with definite surgical oncology principles.

CONCLUSION

To our knowledge, PanIN-3 lesion combined with GCP as the precursor of invasive cancer in the fundus of an unoperated stomach has not yet been reported in the literature. Because heterotopic pancreas as well as GCP is a rare gastric submucosal tumor that is mistaken for other more common gastric pathologies such as GIST and the preoperative diagnosis is difficult, it is important to consider GCP or heterotopic pancreas in the differential diagnosis of patients presenting with suspicious submucosal gastric lesions.

Conflicts of interest. No conflict to declare.

REFERENCES


