Heterotopic Pancreas in the Stomach which Caused Obstructive Stenosis in the Duodenum

Shinya Kobayashi¹, Yasutaka Okayama², Kazuki Hayashi¹, Hitoshi Sano¹, Shigehiro Shiraki¹, Kazuo Goto², Hirotaka Ohara¹ and Takashi Joh¹

Abstract

The patient, a 43-year-old Japanese man suffering from duodenal ulcer and reflux esophagitis, was admitted to our hospital because of submucosal tumor in the antrum and obstructive stenosis of duodenum. Several imaging tests could not rule out the possibility of malignant disease. Therefore, the patient was surgically treated. Pathohistological examination of resected tissue demonstrated Heinrich type I heterotopic pancreas in the gastric lesion and submucosal abscess in the duodenal lesion with stenosis. In this case, it was considered that the heterotopic pancreas caused chronic inflammation to form the gastric tumor, and submucosal abscess leading to the severe duodenal stenosis.

Key words: Heinrich type I, submucosal abscess, chronic inflammation, duodenal stenosis, gastric tumor

(DOI: 10.2169/internalmedicine.45.1814)

Introduction

Most cases of heterotopic pancreas in the stomach are endoscopically detected by chance because patients with this disorder are typically asymptomatic. Treatment is not generally required. Therefore, heterotopic pancreas in the stomach is not a great problem in the clinical setting.

Here, we report a very rare case of heterotopic pancreas in the stomach, which caused an obstructive duodenal stenosis. Surgical treatment was performed and histological examinations demonstrated that heterotopic pancreas located in the gastric antrum caused chronic inflammation around the heterotopic pancreatic tissue to form a submucosal tumor in the antrum, and resulted in the formation of a submucosal abscess in the duodenum. This duodenal abscess was considered to have caused duodenal stenosis.

Case Report

The patient was a 43-year-old Japanese man. Three years previously, he complained of epigastralgia and heartburn, and received ambulatory treatment based on the diagnosis of duodenal ulcer and reflux esophagitis. The patient was admitted to our hospital, because obstructive stenosis in the duodenum was found during follow-up study for duodenal ulcer by gastrointestinal radiologic examination and gastro-duodenoenoscopic study. He had no remarkable symptom. Physical examinations on admission did not demonstrate any abnormalities, nor did laboratory data on admission. Anemia was not found on peripheral blood examination, and inflammatory reaction was not observed on biochemical blood analysis. Serum levels of tumor markers (CEA, AFP, CA19-9, DUPAN II, and SPAN-1), pancreatic enzymes (AMY, trypsin, lipase, elastase 1, and PSTI), and gastrin were all normal. Upper gastrointestinal radiologic examination demonstrated dilation failure in the gastric antrum and the duodenal bulb, as well as stenosis over the entire circumference of the post-bulbal part of the duodenum (Fig. 1a, b). Although endoscopic study demonstrated an elevated lesion at the posterior wall of the antrum, there was erosion in the surface of the lesion. These findings indicated that this lesion was formed by a submucosal tumor or extrinsic pressure. The duodenal bulb was severely narrowed. Redness and erosions were observed in the mucosa of duodenal bulb, but endoscope could not be inserted into the anal site.
Figure 1.  a) Upper gastrointestinal radiologic examination demonstrated dilation failure in the gastric antrum and the duodenal bulb. b) Stenosis over the entire circumference of the post bulbal part of the duodenum.

Figure 2.  a) Upper gastrointestinal endoscopic study demonstrated extrinsic pressure between the greater curvature of the antrum and the posterior wall, there was erosion in the center of submucosal tumor. b) The duodenal bulb was narrowed, and redness and erosions were observed in the duodenal mucosa.

(Fig. 2a, b). Biopsies obtained from the duodenal bulb showed no neoplastic changes. Computed tomography (CT) demonstrated a tumor measuring 4×5 cm between the gastric antral and the duodenal wall, and a part of the tumor was densely stained with a contrast medium. No abnormality was detected in the liver, the pancreas, or the biliary tract (Fig. 3a, b). Abdominal ultrasonography (US) did not demonstrate a clear tumor lesion detected by CT. These findings suggested that duodenal stenosis was caused by a submucosal tumor of the antrum and/or the duodenum. In addition to the severity of stenosis, the possibility of malignant disease could not be ruled out. Therefore, the patient was surgically treated. The tumor was palpated between the gastric antrum and the duodenal bulb intraoperatively. Rapid intraoperative pathology did not demonstrate any malignant findings. Therefore, subtotal gastrectomy and partial duode-
notomy were performed. When resected tissue specimens were macroscopically examined, a round tumor measuring 5×4 cm was observed at the region 4 cm from the duodenal stamp on the gastric serosa. There was erosion in the center of submucosal tumor (Fig. 4a, b). In addition, an ulcer and erosions were observed in the duodenal mucosa, and the wall was hypertrophied. When fixed specimens were examined, there was no capsule and the inside of the tumor was whitish and yellowish (Fig. 5a, b). Pathohistological findings demonstrated that the lesion was present between the proper muscular layer and the serosa of the stomach, while, in the lesion, pancreatic tissue with acinar cells, duct cells, and islets of Langerhans was observed (Fig. 6a, b). Therefore, the patient was diagnosed as having Heinrich type I heterotopic
Figure 5. a) b) Cut surface shows the whitish and yellowish colored tumor with fibrosis.

Pancreatic tissue is heterotopically observed in various abdominal organs, and termed, heterotopic pancreas, aberrant pancreas, or accessory pancreas. Pearson et al reviewed 589 cases of heterotopic pancreas, and reported the frequencies of this disorder in the abdominal region; 30% in the duodenum, 25% in the stomach, 15% in the jejunum, 3% in the ileum, and 6% in Meckel diverticulum (1). Thus, almost half of the cases of heterotopic pancreas developed in the stomach and the duodenum. Moreover, they noted that heterotopic pancreas was also observed in the gallbladder, the common bile duct, the liver, and the spleen. The frequency of heterotopic pancreas found in the resected stomach was also reported to range between 0.25 and 0.8% (2). It was also reported that heterotopic pancreas in the stomach predominantly develops in male subjects between 30 and 50 years of age (the male-to-female ratio is 2:3:1). Most cases (90%) of heterotopic pancreas in the stomach are single, and are observed in the antrum. Moreover, heterotopic pancreas in the stomach macroscopically exhibit a morphology of submucosal tumor (3). Heinrich classification (4) is frequently used to classify this disorder, and it was reported that Heinrich type II, which consists of acinar and duct cells excluding islets of Langerhans, was most frequently observed. Although it was reported that the symptoms of heterotopic pancreas in the stomach include epigastralgia, epigastric discomfort, nausea, and eructation, most cases follow asymptomatic courses (5, 6). Heterotopic pancreas in pediatric patients is generally detected as a small nodule measuring 1-2 mm in diameter. However, such a small nodule is reported to increase in size along with the growth of pediatric patients (7, 8). When such nodules increase in size at the pyloric zone of the stomach, the most common site of heterotopic pancreas, it may cause pyloric stenosis (9, 10). Moreover, although the frequency is lower than that in normal pancreatic tissue, heterotopic pancreas was reported to induce secondary changes, such as acute or chronic pancreatitis, bleeding, or cyst formation (11-15). A report has been published describing elevated serum amylase levels in patients with heterotopic pancreatitis in the stomach. Although the levels of pancreatic enzymes were all normal in the present patient, it was considered that repeated inflammation caused by heterotopic pancreas in the stomach resulted in tumor formation at the serosal side of the antrum. Submucosal abscess formation was observed in the duodenal region with stenosis. Although the real reason for abscess formation was unknown, it is conceivable that inflammation and tumor formation around the gastric heterotopic pancreas first, and then the submucosal abscess was induced in the duodenum, which caused such an obstructive stenosis.

Discussion

Pancreatic tissue is heterotopically observed in various abdominal organs, and termed, heterotopic pancreas, aberrant pancreas, or accessory pancreas. Pearson et al reviewed 589 cases of heterotopic pancreas, and reported the frequencies of this disorder in the abdominal region; 30% in the duodenum, 25% in the stomach, 15% in the jejunum, 3% in the ileum, and 6% in Meckel diverticulum (1). Thus, almost half of the cases of heterotopic pancreas developed in the stomach and the duodenum. Moreover, they noted that heterotopic pancreas was also observed in the gallbladder, the common bile duct, the liver, and the spleen. The frequency of heterotopic pancreas found in the resected stomach was also reported to range between 0.25 and 0.8% (2). It was also reported that heterotopic pancreas in the stomach predominantly develops in male subjects between 30 and 50 years of age (the male-to-female ratio is 2:3:1). Most cases (90%) of heterotopic pancreas in the stomach are single, and are observed in the antrum. Moreover, heterotopic pancreas in the stomach macroscopically exhibit a morphology of submucosal tumor (3). Heinrich classification (4) is frequently used to classify this disorder, and it was reported that Heinrich type II, which consists of acinar and duct cells excluding islets of Langerhans, was most frequently observed. Although it was reported that the symptoms of heterotopic pancreas in the stomach include epigastralgia, epigastric discomfort, nausea, and eructation, most cases follow asymptomatic courses (5, 6). Heterotopic pancreas in pediatric patients is generally detected as a small nodule measuring 1-2 mm in diameter. However, such a small nodule is reported to increase in size along with the growth of pediatric patients (7, 8). When such nodules increase in size at the pyloric zone of the stomach, the most common site of heterotopic pancreas, it may cause pyloric stenosis (9, 10). Moreover, although the frequency is lower than that in normal pancreatic tissue, heterotopic pancreas was reported to induce secondary changes, such as acute or chronic pancreatitis, bleeding, or cyst formation (11-15). A report has been published describing elevated serum amylase levels in patients with heterotopic pancreatitis in the stomach. Although the levels of pancreatic enzymes were all normal in the present patient, it was considered that repeated inflammation caused by heterotopic pancreas in the stomach resulted in tumor formation at the serosal side of the antrum. Submucosal abscess formation was observed in the duodenal region with stenosis. Although the real reason for abscess formation was unknown, it is conceivable that inflammation and tumor formation around the gastric heterotopic pancreas may mechanically irritate the duodenal wall and cause bacterial infection to form a submucosal abscess. In this case, this abscess formation was considered to have caused duodenal stenosis. To date, there have been no reports describing severe duodenal stenosis induced by heterotopic pancreas in the stomach. Therefore, the present case was considered to have been a very rare case of such a disorder. We would like to emphasize that when duodenal stenosis with gastric submucosal tumor has developed within a relatively short time, not only malignant diseases, but also inflammatory changes associated with gastric heterotopic pancreas should be considered.
Figure 6.  a, b) Pancreatic tissue with acinar cells, duct cells, and islets of Langerhans was observed. (HE stain ×100) b) Arrow shows islet of Langerhans. c) Histologic examination of resected specimen demonstrated marked neutrophil proliferation between the muscular layer of mucosae and the proper muscular layer of the duodenum. Submucosal abscess was formed in the duodenum. (HE stain ×40)

References