Single Case

Early Gastric Cancer Just above a Heterotopic Pancreas

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Abstract
We report the first case of early gastric cancer just above a heterotopic pancreas for which the differential diagnosis was carcinoma arising from heterotopic pancreas. Routine upper gastrointestinal endoscopy in an 83-year-old man with sigmoid colon cancer revealed a gastric cancer in the lesser curvature of the antrum. Endoscopic ultrasonography (EUS) for evaluating the depth of tumor invasion revealed a hypoechoic mass in the submucosal layer. The depth of tumor invasion was diagnosed as muscularis propria. Distal gastrectomy and sigmoidectomy were performed. Histologically, the resected specimen of the stomach unexpectedly revealed a heterotopic pancreas just below the gastric cancer. They were not linked, and the heterotopic pancreas had no dysplasia. The gastric cancer had slightly invaded the submucosa. The hypoechoic mass on EUS was not the invasive tumor but the heterotopic pancreas. The preoperative staging of the gastric cancer on EUS was confounded by the presence of the heterotopic pancreas just below the gastric cancer.
Introduction

Heterotopic (aberrant or ectopic) pancreas is defined as pancreatic tissue that lacks anatomical or vascular connections with the original pancreas. Heterotopic pancreas is relatively uncommon, and is divided into three types according to von Heinrich’s classification [1]. Type I is composed of a complete structure including ducts, acini, and islets. Type II is composed of ducts and acini. Type III is composed of ducts only. Most cases of heterotopic pancreas are asymptomatic and found incidentally during endoscopy, surgery, or autopsy. Complications associated with heterotopic pancreas include ulceration, bleeding, luminal obstruction, inflammation, and malignant transformation. There are some reports of adenocarcinoma arising from heterotopic pancreas [2, 3]. On the other hand, carcinoma concomitant with heterotopic pancreas can also occur [4]. In the case of carcinoma above or close to a heterotopic pancreas, it is necessary to make a histological determination of whether it arises from the heterotopic pancreas or coexists with the heterotopic pancreas. Three criteria for the diagnosis of carcinoma arising from heterotopic pancreas are proposed [5]: (1) the tumor must be found within or close to the heterotopic pancreatic tissue; (2) a direct transition between the pancreatic structures and the carcinoma must be observed (i.e. duct cell dysplasia or carcinoma in situ), and (3) the nonneoplastic pancreatic tissue must comprise at least fully developed acini and ductal structures.

No report of gastric cancer above or close to a heterotopic pancreas has been reported. We report the first case of early gastric cancer just above a heterotopic pancreas.

Case Report

An 83-year-old man was admitted to our hospital, complaining of lower abdominal pain and abdominal distension of 2 weeks’ duration. He had undergone an operation for descending colon cancer 13 years previously. On examination, he was afebrile, his blood pressure was 151/88 mm Hg, and his pulse was 99 bpm. His abdomen was soft, distended, and tender in the left lower quadrant. Muscle defense and rebound tenderness were not present. Laboratory tests were as follows: white blood cell count, 6,000/μl; hemoglobin, 12.5 g/dl; platelets, 301,000/μl; albumin, 3.9 g/dl; total bilirubin, 0.5 mg/dl; aspartate aminotransferase, 24 IU/l; alanine aminotransferase, 15 IU/l; amylase, 115 IU/l; creatinine, 1.01 mg/dl (which was similar to his previous level); carcinoembryonic antigen, 8.8 ng/ml, and carbohydrate antigen 19-9, 6.7 U/ml. CT and colonoscopy revealed an obstructing sigmoid colon cancer. Thus, self-expandable metallic stent placement as a bridge to surgery was performed. After achieving luminal patency and the disappearance of his symptoms, upper gastrointestinal endoscopy was performed routinely, which revealed an irregular ulcer measuring 3 cm in diameter in the lesser curvature of the antrum (fig. 1a). A biopsy specimen obtained from the lesion revealed poorly differentiated adenocarcinoma. Endoscopic ultrasonography (EUS) for evaluating the depth of tumor invasion revealed a hypoechoic mass in the submucosal layer, and the border between the mass and proper muscular layer was unclear (fig. 1b). Thus, the depth of tumor invasion was diagnosed as muscularis propria. Enhanced abdominal CT revealed no metastasis in the lymph nodes or distant organs. Under the preoperative diagnosis of advanced gastric cancer and sigmoid colon cancer, distal gastrectomy and sigmoidectomy were performed.

The resected specimen revealed superficial, depressed-type (type 0-Iic), poorly differentiated adenocarcinoma in the lesser curvature of the antrum, measuring 25 × 35 mm. The
tumor was located mainly in the mucosa, and it had slightly invaded the submucosa. The cut surface of the resected specimen revealed a yellowish mass measuring 12 mm in diameter just below the gastric cancer, located mainly in the submucosal and muscular layers (fig. 2). Histologically, this yellowish mass was heterotopic pancreatic tissue consisting of acini, ducts, and islets (von Heinrich type I) (fig. 3). The heterotopic pancreas had no dysplasia. It was located just below the gastric cancer, but the two were not linked (fig. 2, fig. 3). Therefore, we concluded that the gastric cancer did not arise from the heterotopic pancreas but coexisted with it.

**Discussion**

This is a case of gastric cancer concomitant with a heterotopic pancreas. The differential diagnosis was carcinoma arising from heterotopic pancreas. Three criteria for the diagnosis of carcinoma arising from heterotopic pancreas are proposed [5]: (1) the tumor must be found within or close to the heterotopic pancreatic tissue; (2) a direct transition between the pancreatic structures and the carcinoma must be observed (i.e. duct cell dysplasia or carcinoma in situ), and (3) the nonneoplastic pancreatic tissue must comprise at least fully developed acini and ductal structures. Our case did not fulfill criteria 1 and 2. The gastric cancer was distant from the heterotopic pancreas, and the heterotopic pancreas was free of dysplasia. Therefore, we concluded that the gastric cancer was concomitant with the heterotopic pancreas.

To our knowledge, this is the first report of gastric cancer just above or close to a heterotopic pancreas. Malignant transformation in heterotopic pancreas is rare. Matsuki et al. [3] reviewed the literature from 1963 to 2003 and found only 12 cases of adenocarcinoma arising from a heterotopic pancreas in the stomach. A case of gastric cancer concomitant with heterotopic pancreas such as our case might be included in case series of gastric cancer arising from a heterotopic pancreas, because it is not always easy to make a histological determination of whether a carcinoma arises from a heterotopic pancreas. In our case, if the tumor had progressed and invaded the heterotopic pancreas, a misdiagnosis of gastric cancer arising from heterotopic pancreas might have been made.

It is difficult to obtain a definite diagnosis of heterotopic pancreas preoperatively [6, 7]. Tanaka et al. [6] reported that most of heterotopic pancreas lesions were found unexpectedly at the time of operation, and only 2 cases in 15 heterotopic pancreas lesions were diagnosed as submucosal tumor preoperatively. We were not able to diagnose the heterotopic pancreas preoperatively, because there were no findings of submucosal tumor by upper gastrointestinal endoscopy. On the other hand, EUS imaging was able to detect the mass of heterotopic pancreas, which was preoperatively misdiagnosed as tumor invasion of the submucosal and muscular layers. Histology of the resected specimen revealed that the mass was heterotopic pancreas and the depth of tumor invasion was the submucosa. The presence of heterotopic pancreas just below the gastric cancer was confounding the preoperative evaluation of the depth of tumor invasion on EUS. Matsushita et al. [8] reported that characteristic EUS features of heterotopic pancreas are an indistinct margin, a heterogeneous appearance, lesions accompanied by an anechoic area, fourth-layer thickening, and a location within either the third and fourth layers or only in the third layer. In our case, some of these characteristics were observed on EUS. There are three case reports of gastric cancer just above the gastric lipoma [9–11]. With regard to preoperative diagnoses, one was a confident diagnosis of gastric lipoma, the second submucosal tumor, and the third not obtained. When
we find a mass below a gastric cancer on EUS that appears to be cancer invasion, we may also consider a diagnosis of concomitant submucosal tumor including heterotopic pancreas and lipoma.

In conclusion, to our knowledge, this is the first report of gastric cancer just above a heterotopic pancreas for which the differential diagnosis is carcinoma arising from heterotopic pancreas. In the case of gastric cancer above or close to a heterotopic pancreas, preoperative evaluation of the depth of tumor invasion might be difficult.

Acknowledgments

None.

Statement of Ethics

All procedures have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. Informed consent was obtained from the present patient for being included in the study.

Disclosure Statement

The authors declare no conflict of interest.

References

Fig. 1. a Endoscopic finding of a gastric lesion. An irregular ulcer measuring 3 cm in diameter was observed in the lesser curvature of the antrum. b EUS image of the gastric lesion. A hypoechoic mass was observed in the submucosal layer, which appeared to be tumor invasion (arrows).
Fig. 2. a Cut surface of the resected specimen. A yellowish, smooth-bordered mass was observed mainly in the submucosal and muscular layers (arrows). b Gross findings from the resected specimen after cutting. The gastric cancer is visible along the blue lines. The heterotopic pancreas is to be seen along the white lines. The heterotopic pancreas was just below the gastric cancer, measuring 12 mm in diameter.
Fig. 3. Histological finding of heterotopic pancreas and gastric cancer. The heterotopic pancreas was observed in the submucosal layer, and it had no dysplasia (arrows). The adenocarcinoma was observed in the mucosa (arrowheads). They were not linked. HE. ×20.