Endoscopic treatment of pancreatic fluid collection in gastric heterotopic pancreas. A case report

Ivana Mikoviny Kajzrlikova[1], Jan Kuchar[1], Petr Vitek[1,2], Barbora Horavova[3], Hana Vankova[4], Martin Chrostek[5]

Background. Heterotopic pancreas is defined as pancreatic tissue located outside the pancreatic parenchyma that lacks an anatomic or vascular connection to the normal pancreas. Symptomatic gastric heterotopic pancreas is a rare condition that can manifest as acute or chronic pancreatitis. Asymptomatic heterotopic pancreas does not require treatment, while symptomatic lesions should be resected. The modality of final resection of heterotopic pancreas depends on its size and the depth of gastric wall involvement.

Methods and Results. A 36-year-old woman was admitted for recurrent epigastralgia. Abdominal computed tomography (CT) scan revealed that an abscess had formed in the gastric antrum. After multidisciplinary discussion we decided for conservative treatment with intravenous antibiotics and further detailed endoscopic diagnostic. Esophagogastroduodenoscopy revealed a submucosal mass with a central fistula and intermittent pus secretion in the prepyloric region of the gastric antrum, which was subsequently drained with a double pigtail stent under endoscopic ultrasound (EUS) and fluoroscopy. The possibility of pancreatic fluid collection in the case of heterotopic pancreas was suggested during the EUS examination, and histology subsequently confirmed heterotopic pancreatic tissue. The patient was in good condition and without any abdominal pain. According to a control CT scan after 10 weeks, the fluid collection was completely resolved. Due to the possible recurrence of pancreatitis, resection of heterotopic pancreas was proposed to the patient. Since the lesion involved the muscularis propria of the gastric wall, surgical resection of the mass was indicated.

Conclusion. Fluid collections after acute pancreatitis in heterotopic pancreas in the gastric antrum can be successfully managed by endoscopy.

Key words: heterotopic pancreas, pancreatitis, drainage, endoscopy, surgery

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[1]Department of Internal Medicine, Beskydy Gastrocentre, Hospital Frydek-Mistek, El. Krasnohorske 321, 73801 Frydek-Mistek, Czech Republic
[2]Department of Internal Studies, Faculty of Medicine, University of Ostrava, Czech Republic
[3]Department of Pathology, Hospital Frydek-Mistek, El. Krasnohorske 321, 73801 Frydek-Mistek, Czech Republic
[4]Department of Radiodiagnostic, Hospital Frydek-Mistek, El. Krasnohorske 321, 73801 Frydek-Mistek, Czech Republic
[5]Department of Surgery, Hospital Frydek-Mistek, El. Krasnohorske 321, 73801 Frydek-Mistek, Czech Republic
Corresponding author: Ivana Mikoviny Kajzrlikova, e-mail: kajzrlikova@nemfm.cz

BACKGROUND

Heterotopic pancreas, also known as ectopic pancreas, aberrant pancreas, and pancreatic rest, is defined as pancreatic tissue located outside the pancreatic parenchyma that lacks an anatomic or vascular connection with the normal pancreas.

The vast majority of lesions are detected incidentally. Heterotopic pancreas is rarely symptomatic, but it can manifest as abdominal pain, gastrointestinal bleeding, obstruction, and acute or chronic pancreatitis, and rare cases of malignant transformation have also been described[1-4]. Heterotopic pancreatic tissue is usually located in the submucosa and/or the muscularis or subserosa. Lesions are most frequently found in the stomach, duodenum, or proximal jejunum but have also been reported within Meckel's diverticulum, the navel, spleen, Fallopian tubes, the gallbladder, bile ducts, the minor and major papillae, the mediastinum, and brain[5-8].

Asymptomatic heterotopic pancreas does not require treatment. Symptomatic lesions, however, should be resected. Endoscopic resection can be performed if the muscularis propria is not involved, and in cases with deeper involvement, surgical resection is preferred to endoscopic resection[6].

Here, we report a case of successful endosonography-navigated endoscopic drainage of pancreatic fluid collection in heterotopic pancreas in the gastric antrum.

CASE PRESENTATION

A 36-year-old woman was admitted for recurrent epigastralgia and nausea. She had no vomiting or diarrhea. Patient's symptoms started three days before admission. The patient had an unremarkable medical history except two cesarean sections.

The patient's temperature was 36.6 °C, heart rate was 106 bpm, blood pressure was 175/100 mmHg, and oxygen

saturation in room air was 97%. The clinical examination revealed pain in the epigastrium with no peritoneal signs. Blood analysis showed leukocytosis (16.3×109/L) as well as normal hematocrit and platelet count. The serum C-reactive protein (CRP) level was increased at 142.3 mg/L. Liver enzymes, pancreatic enzymes, and tumor markers were normal. Abdominal CT revealed that an abscess had formed in the gastric antrum with regional lymphadenopathy, and a diagnosis of subepithelial gastric tumor (i.e. gastrointestinal stromal tumor) was also considered (Fig. 1). The chest X-ray was normal.

After multidisciplinary discussion we decided for conservative treatment with intravenous antibiotics and further detailed endoscopic diagnostic. Esophagogastroduodenoscopy revealed a submucosal mass with central fistula and intermittent pus secretion in the prepyloric region of the gastric antrum (Fig. 2). Linear endosonography (EUS)-navigated drainage was planned for the following day. By EUS, a 32 mm hypoechoic collection with heterogenous tissue fragments (about 50% of the volume) was observed. The lesion was found to originate from the second and third layers of the gastric wall (muscularis mucosae and submucosa).

The collection was punctured with a 19-G needle, and sanguinolent pus fluid was aspirated and sent for bacterial culture, cytology, and histology. Then, drainage of the collection under combined endosonographic and fluoroscopic control was performed using a 10F cystotome, dilatation balloon, and 10F 5 cm long double pigtail plastic stent (Fig. 3, 4). The possibility of pancreatic fluid

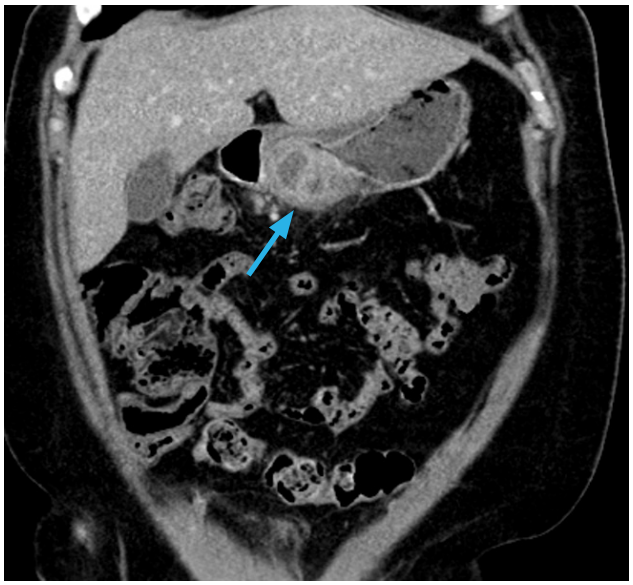

Fig. 1. Abdominal CT scan of abscess formation in gastric antrum.

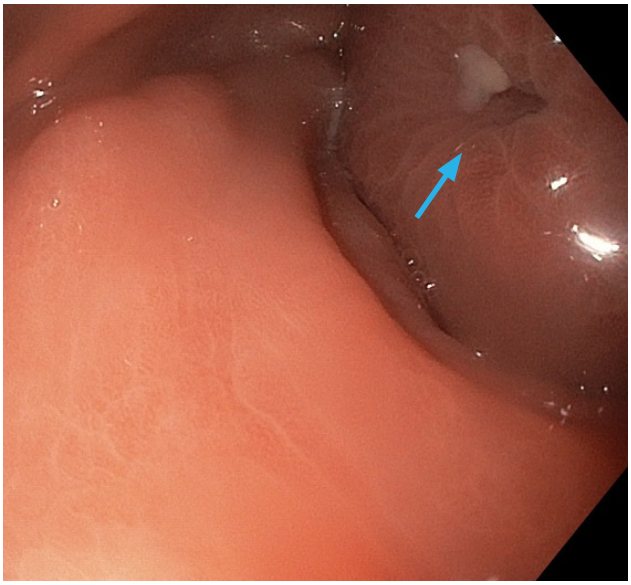

Fig. 2. Gastroscopic appearance of submucosal mass in gastric antrum.

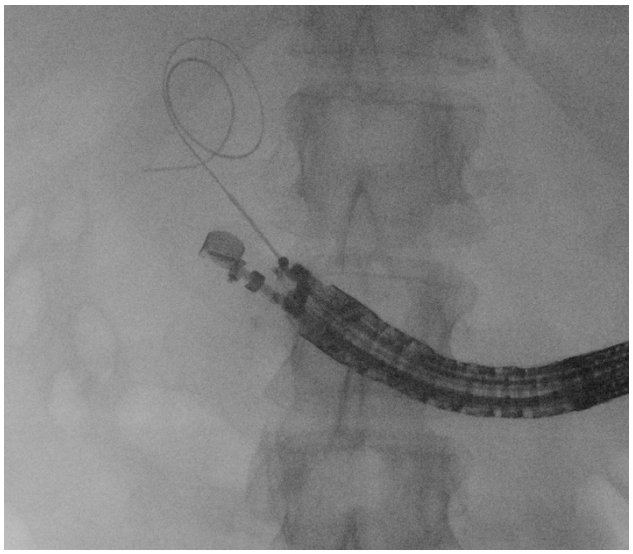

Fig. 3. Fluoroscopic picture of guidewire inside the lesion.

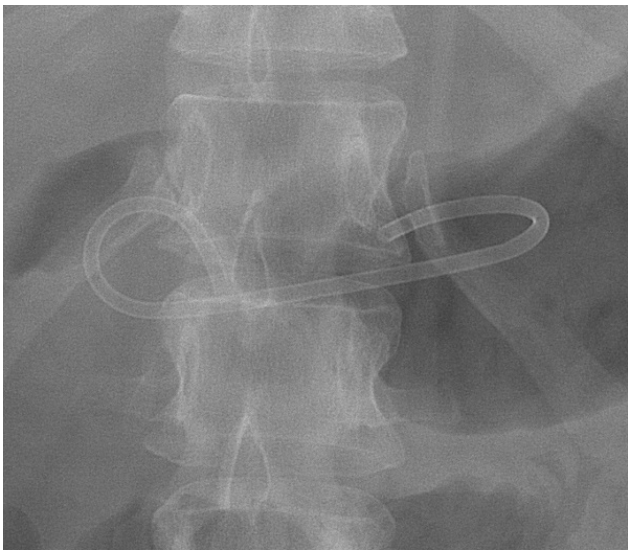

Fig. 4. Fluoroscopic picture of double pigtail stent in situ.

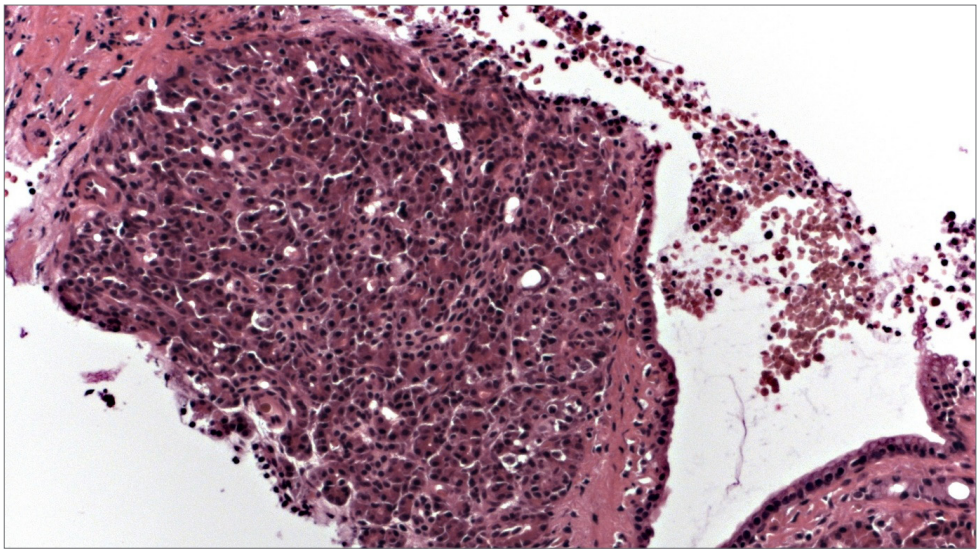

Fig. 5. Histologic picture of heterotopic pancreatic tissue.

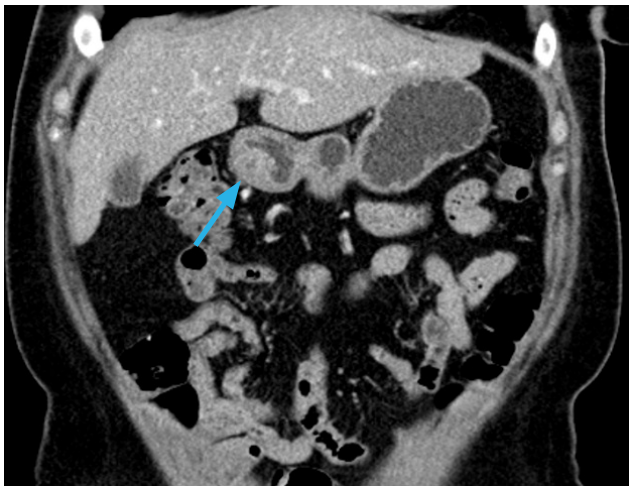

Fig. 6. Abdominal CT scan 10 weeks after the drainage.

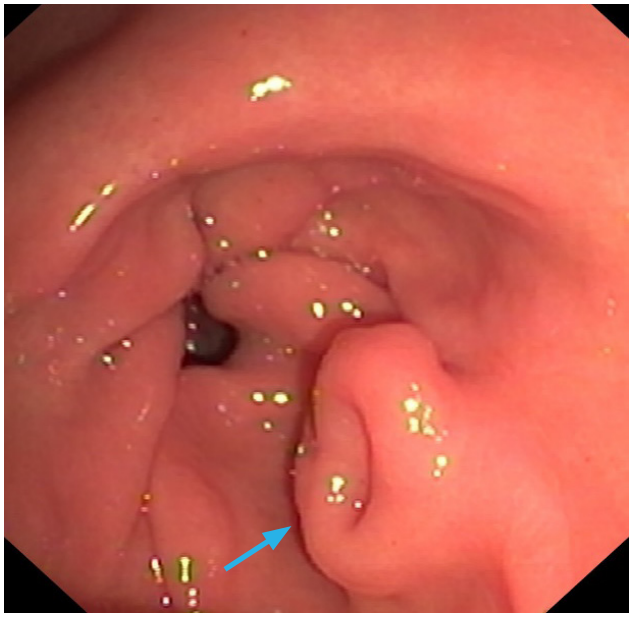

Fig. 7. Endoscopic appearance of heterotopic pancreas 3 months after the drainage.

collection in heterotopic pancreas was suggested during the EUS examination.

Histologic analysis subsequently confirmed heterotopic pancreatic tissue with ducts and an exocrine component, together with blood cells, pus and, fragments of the gastric wall (Fig. 5).

The patient's condition improved, as evidenced by a decreased CRP level and leukocyte count, and the patient was discharged. The patient was in good condition without any abdominal pain. Abdominal ultrasonography after 7 weeks showed regression of fluid collection from 32 mm to 12 mm with the double pigtail stent *in situ*, and her blood test was normal. Ten weeks after drainage, she underwent a control CT scan, which revealed full regression of the fluid collection in heterotopic gastric pancreas and spontaneous migration of the plastic stent (Fig. 6). Control EUS revealed a 28 mm lesion with a salt and pepper structure and anechoic ducts in the gastric antrum (Fig. 7, 8). The fluid collection was completely healed, and the lesion was shown to involve four gastric layers with intact serosa (Fig. 8).

Due to possible recurrence of pancreatitis, resection of the heterotopic pancreas was proposed to the patient. Endoscopic resection was determined not to be safe because of involvement of the muscularis propria of the gastric wall, and thus, surgical resection was indicated after multidisciplinary discussion. The patient underwent surgical resection of the mass with pyloric preservation at another hospital and is currently under surveillance. Final histopathological examination of resected specimen confirmed the diagnosis of heterotopic pancreas.

DISCUSSION

Heterotopic pancreas is a variant of foregut embryologic dystopia, which was first reported by Jean Schultz in 1727 when it was observed in an ileal diverticulum, but the first histological confirmation was described by Klob in 1859 (ref.[9,10]). This condition is defined as pancreatic tis-

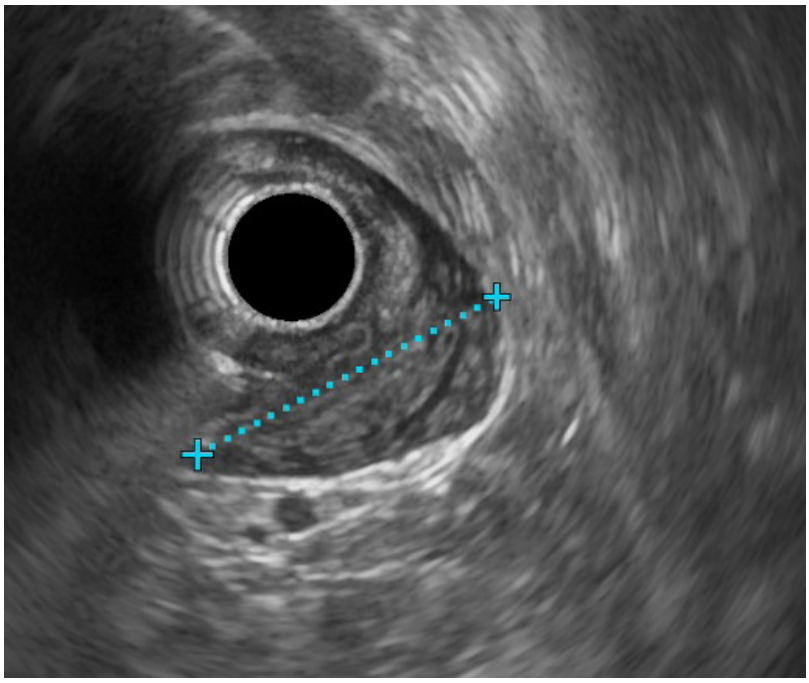

Fig. 8. EUS appearance of heterotopic pancreas 3 months after the drainage.

sue located outside the pancreatic parenchyma that lacks an anatomic or vascular connection with normal pancreas. Four morphopathological types of ectopic pancreas have been described: type 1 is represented by specific pancreatic tissue, type 2 contains only pancreatic ducts, type 3 includes only acinar tissue (exocrine pancreas), and type 4 contains only islets (endocrine pancreas) (ref.[11]). More complex pancreatic tissue may be more susceptible to acute pancreatitis. Our patient had histologic finding of pancreatic tissue with ducts and an exocrine component.

The incidence of heterotopic pancreas in autopsies ranges from 0.5%–13.7%. This condition is more common in males and in individuals between 30 and 50 years of age, and most lesions are detected incidentally[12]. Lesions are most frequently found in the stomach (25%–38%), duodenum (17%–21%), and proximal jejunum (15%–21%) but have also been reported within Meckel's diverticulum, the navel, spleen, Fallopian tubes, gallbladder, bile ducts, the minor and major papillae, the mediastinum, and brain[5-8]. Overall, 90% of gastric lesions are located in the gastric antrum, as in our patient.

In heterotopic pancreas, pancreatic tissue is more complex, whereas pancreatitis is a greater risk. Our patient experienced abdominal pain with elevated leukocyte count and CRP level but normal pancreatic enzyme levels. The diagnosis of abscess within the gastric wall was suggested and the EUS-drainage was planned. During EUS examination under fluoroscopic control, the drainage was successfully performed and fluid collection in heterotopic pancreas was suspected because of the appearance of the mass on endoscopy and EUS. Histologic examination confirmed the diagnosis.

Considering the appearance of the collection on EUS and the time interval from the beginning of abdominal pain, the collection can be classified as acute necrotic collection according to guidelines[13]. The standard approach to pancreatic fluid collections is to wait, if necessary, a minimum of four weeks after the collection is well demarcated followed by drainage.

In heterotopic pancreas in the gastric antrum, the fluid collection is located within the gastric wall, and thus, the drainage can be performed early, and the anatomic conditions are different.

EUS is the most accurate modality that can differentiate subepithelial lesions. Typical findings in heterotopic pancreas are hypoechoic or intermediate echogenic heterogeneous lesions with indistinct margins. Lesions most commonly arise from the third or fourth layer, or from a combination of two layers of the gastrointestinal tract[6]. Uncommonly, EUS reveals anechoic duct-like structures, as in our case. An advantage of EUS is an ability to perform guided biopsies for histologic evaluation.

Asymptomatic heterotopic pancreas does not require treatment, but symptomatic lesions should be resected. Endoscopic resection can be performed if the muscularis propria is not involved and if it can be performed by a standard snare, band ligation-assisted, or cap-assisted polypectomy technique[6]. Successful endoscopic resections of heterotopic pancreas using endoscopic submucosal dissection are described in the literature[14,15]. Full-thickness resection overcomes deep wall involvement but is limited by the horizontal extent of the mass. In our case, the lesion involved four gastric layers with intact serosa and surgical resection was indicated. Our patient underwent partial gastric resection of gastric antrum with the mass with pyloric preservation.

CONCLUSION

Symptomatic gastric heterotopic pancreas is a rare condition that can manifest as acute or chronic pancreatitis. Fluid collections after acute pancreatitis can be successfully managed by endoscopy. Symptomatic heterotopic pancreas should be resected, the modality of final resection (endoscopic or surgical) depends on its size and the depth of gastric wall involvement.

ABBREVIATIONS

CRP, C-reactive protein; CT, Computed tomography; EUS, Endoscopic ultrasound.

Author contributions: IMK: cared for the patient, performed endoscopic treatment, and wrote and corrected the manuscript; JK, PV: cared for the patient, performed endoscopic treatment and reviewed and corrected the manuscript; BH: interpreted the pathological findings; HV: performed radiologic examinations; MC: provided surgery consultations; all authors issued final approval for the version to be submitted.

Conflict of interest statement: The authors state that there are no conflicts of interest regarding the publication of this article.

Ethical Approval /Patient consent: Ethical approval was not necessary for this work due to its design (case report). Written informed consent for publication of a case report was obtained from the patient.

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