

CASE REPORT

An Unusual Case of Gastric Heterotopic Pancreas

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ABSTRACT

Context Submucosal lesions of the gastrointestinal tract represent a diagnostic challenge for the physician. Endoscopic ultrasonography may provide useful information before deciding on therapeutic strategy.

Case report We report on a case of a young female presenting with a large gastric submucosal mass, 32 mm in size. Endoscopic ultrasonography identified a non-homogeneous lesion, with three cystic spaces suggesting a degenerated gastrointestinal stromal tumor. An exploratory laparoscopy was performed. Surprisingly, the final diagnosis was gastric heterotopic pancreas.

Conclusion Heterotopic pancreas should always be kept in mind when facing extramucosal gastric masses, especially in young people. A perioperative biopsy is recommended to prevent unnecessary extensive surgery.

INTRODUCTION

Submucosal lesions of the gastrointestinal tract usually appear as intraluminal protrusions with normal overlying mucosa. Most frequently, these lesions are found incidentally during routine endoscopy. Surface biopsies are usually normal. The differential diagnosis includes a number of

benign and malignant gastric wall tumors, intramural vessels, and extrinsic compression from extramural structures [1].

Heterotopic pancreas is a congenital disorder which consists of the presence of normal pancreatic tissue located outside the pancreatic frame [2]. It often remains asymptomatic throughout life, but may sometimes cause symptoms including symptomatic gastrointestinal bleeding, gastric ulceration, gastric outlet obstruction, pancreatitis and even malignant degeneration.

CASE REPORT

A 25-year-old woman without a significant past medical history developed progressive dyspeptic symptoms over a few weeks without concomitant weight loss. On physical examination, the patient was in good health; bowel sounds were normal and there was no tenderness in the epigastric area; the liver and the spleen were not palpable. Blood count and liver function tests were normal. An upper gastrointestinal endoscopy was performed and revealed an intraluminal protrusion with normal overlying mucosa in the antrum. A surface biopsy revealed normal mucosa. Abdominal CT scan showed a polycystic gastric mass of 32 mm in diameter, without any lymphadenopathies. Endoscopic ultrasonography (EUS) identified an iso- to hypoechoic lesion with well-defined margins, located in the gastric wall and originating from within the submucosa and the muscular propria layer (fourth layer). The lesion was

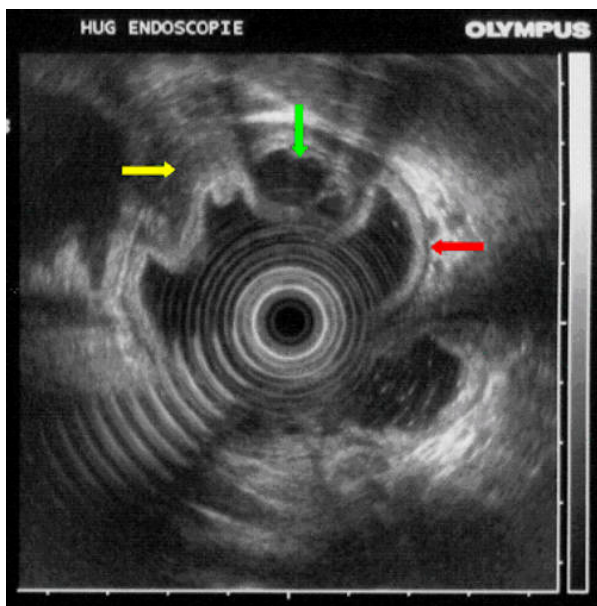


Figure 1. Endoscopic ultrasonography of the lesion. Large gastric mass originating from within the submucosa and muscle layer (yellow arrow) with cystic spaces (green arrow). Red arrow: normal gastric wall.

non-homogeneous, with three cystic spaces, the largest one being 12 mm in size (Figure 1). There were no lymphadenopathies. From this information, we postulated that the patient had a potentially malignant tumor such as a malignant gastrointestinal stromal tumor (GIST). EUS-guided fine needle aspiration was not performed because we thought that the lesion had already degenerated. She was then referred to surgery. An exploratory laparoscopy revealed a 30 mm cystic tumor located in the posterior wall of

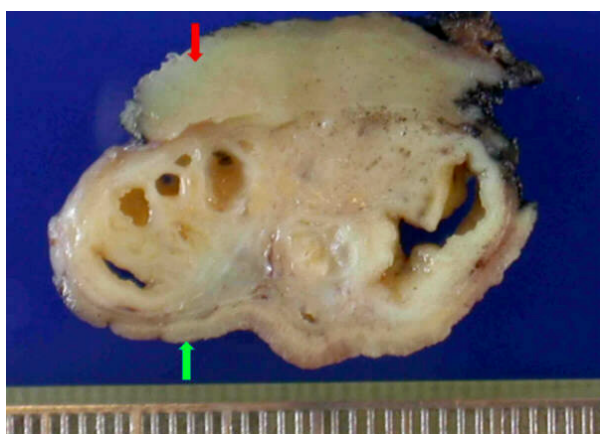


Figure 2. Gross examination: Well-delimited mass of 30 mm in size with several cysts located predominantly in the submucosa (green arrow: mucosa; red arrow: muscle layer).

the gastric antrum, with a distal limit 10 mm from the pylorus. A laparoscopic resection was performed, with only a 5 mm resection margin distally, due to the proximity of the pylorus. Frozen section analysis identified a heterotopic pancreas. Based on this diagnosis, no further resection was performed. The stomach was then closed by continuous suture. The postoperative course was uneventful and she was discharged four days after surgery. The final histological diagnosis was gastric heterotopic pancreas without any evidence of malignancy (Figure 2 and 3). The lesion was composed of exocrine acinar tissue, ducts and endocrine cells. The cysts corresponded to a dilated duct with erosions and inflammation located essentially in the submucosa and in the muscle layer.

DISCUSSION

The most common locations of heterotopic pancreas are the stomach, and the small intestine. Heterotopic pancreas has also been reported to occur in unusual sites such as the esophagus, the lungs, the gallbladder and even the spleen [3, 4, 5]. It has been suggested that heterotopic pancreas results from the separation of pancreatic tissue during the embryonic rotation of the dorsal and ventral buds. It usually remains asymptomatic throughout life and is found incidentally during an upper GI endoscopy or

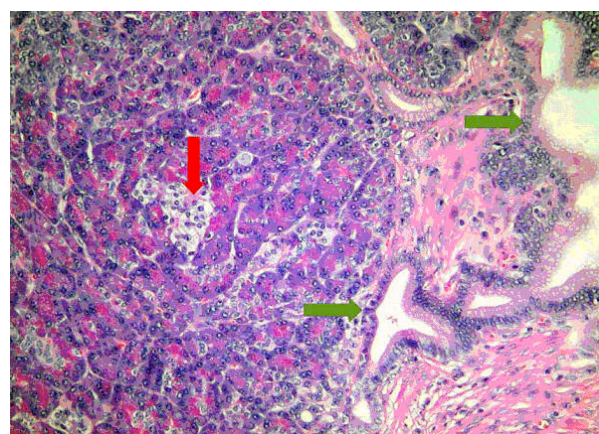


Figure 3. High power field magnification (x200) illustrating acinar and ductal components (green arrows) surrounding an endocrine aggregate (red arrow).

other radiological imaging modalities in patients between 40 and 70 years of age [5]. The lesion can sometimes induce abdominal symptoms. Among these, epigastric pain is common and seems to be due either to intussusception of the overlying gastric mucosa or to pancreatitis [6]. Pancreatic abscess and the malignant transformation of heterotopic pancreas have been reported [7]. Gastric lesions are located in the antrum in 85-95% of cases and originate from the submucosa in two-thirds of cases, the remaining being localized in the *muscularis mucosae* or the subserosal layer [8].

EUS has provided a major breakthrough for characterizing submucosal lesions [1]. It can show whether the lesion arises from the digestive wall or from an extrinsic structure compressing the gastrointestinal tract. It can determine the layer of origin of intramural lesions, an important clue for arriving at a diagnosis. Stromal cell tumors, for example, can be seen as developing from the *muscularis propria* [9], a feature found in the present case, while lipomas typically arise from the submucosa. Specific EUS characteristics can be helpful in differentiating between benign and malignant tumors. One study demonstrated that some EUS criteria can predict malignancy: a tumor diameter greater than 4 cm, irregular extraluminal border, heterogeneity, echogenic foci, and cystic spaces greater than 4 mm [10]. Some of these features were found in the present case.

At EUS, heterotopic pancreas is usually hypoechoic and heterogeneous with indistinct margins. It most commonly arises from the third or fourth layer, or a combination of the two layers of the GI tract. Anechoic areas within the lesion correlate with ductal structures [11].

GISTs are more frequently seen in older people. At EUS, GISTs are typically hypoechoic, homogeneous lesions with well-defined margins, although they can occasionally have irregular margins and

ulcerations. Most GISTs originate from within the *muscularis propria*. Small lesions may originate from the *muscularis mucosa* [10].

Although EUS-guided fine needle aspiration has been shown to be a valuable tool for staging such lesions [12, 13, 14], the multidisciplinary panel of physicians in charge of the patient decided not to perform it because it was thought that the lesion had already degenerated. Performing EUS-guided fine needle aspiration in this particular patient might have helped avoid unnecessary surgery. If a heterotopic pancreas is found during routine endoscopy, the treatment should be dictated by the symptoms. If malignancy is suspected or if the lesion is responsible for a gastric outlet obstruction, a surgical resection is indicated. Surgical resection is preferred to endoscopic mucosal resection when the *muscularis propria* is involved.

In conclusion, heterotopic pancreas should always be kept in mind when facing extramucosal gastric masses, especially in young people. A perioperative biopsy is recommended in order to prevent unnecessary extensive surgery.

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Keywords Choristoma; Diagnosis, Differential; Endosonography; Pancreas

Abbreviations GIST: gastrointestinal stromal tumor

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