Endoscopic removal of gastric ectopic pancreas: An initial experience with endoscopic submucosal dissection

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INTRODUCTION

Ectopic pancreas, also called heterotopic or aberrant pancreas, is defined as pancreatic tissue lying outside its normal location and lacking anatomic or vascular connections with the pancreas. It has been found in 0.6% to 13% of autopsies and has also been noted in approximately one of every 500 surgical operations involving the upper abdomen[1]. Ectopic pancreas is mostly found in the upper gastrointestinal tract adjacent to the pancreas; in 90% of patients with ectopic pancreas, it was found in the stomach, duodenum, or proximal part of the jejunum[2]. Histologic diagnosis of ectopic pancreas is usually difficult when tissue specimens are obtained using a standard endoscopic biopsy forceps[3].
Results

The eight patients included one man and seven women and ranged in age from 18 to 57 years (mean, 36 years). Four patients presented with dyspepsia or epigastric pain. The subepithelial lesions were incidentally diagnosed in the other four patients without preceding symptoms. Five lesions were located at the antrum and three lesions were located at the lower body. None of the lesions showed endoscopic findings such as umbilication or central dimpling. Conventional biopsies were performed on five lesions, but none were diagnosed as ectopic pancreas as based on pathology.

According to EUS, the lesions were mainly located in the second (deep mucosal) or third (submucosal) layer and ranged from 6 to 12 mm (mean 8 mm) in size (Table 1). All lesions were hypoechoic; five lesions were homogeneous and three lesions were heterogeneous. The border was distinct in five lesions (5/8, 62.5%) and indistinct in three lesions (3/8, 37.5%). An undulated margin was observed in six lesions (6/8, 75%) and anechoic cystic or tubular structures appeared in three lesions (3/8, 37.5%).

For accurate diagnosis of the subepithelial lesions, EMR was performed. To decrease the risk of perforation or bleeding, we first planned to remove the lesions by EMR. In all cases, we injected saline solution including a small amount of epinephrine and indigo carmine beneath the lesions. In four cases, the lesions were properly elevated and then were resected by the injection-and-cut technique. However, in the other four cases, the saline spread into surrounding normal tissue and the lesions became flattened, which made it impossible to remove the lesions via the injection-and-cut technique. Therefore, we decided to perform ESD on these four lesions and removed them successfully without any complications. There were no recurrences during the median follow-up period of 30 mo (range 24 to 40 mo).

Discussion

Most patients with ectopic pancreas are asymptomatic, but symptoms may rarely occur due to the irritating effect of hormones and enzymes secreted by the ectopic pancreas. Rare complications resulting from ectopic pancreas have been reported, including gastric outlet obstruction, obstructive jaundice, intestinal obstruction, and intussusceptions. Asymptomatic patients with ectopic pancreas can generally be monitored with treatment reserved for patients who are symptomatic, have enlarging lesions or require diagnostic certainty.

Ectopic pancreas is most often detected as an incidental finding during routine upper endoscopy. The typical endoscopic finding is a firm round or oval subepithelial lesion with a central depression, which corresponds to the opening of a duct. The gross appearance of central dimpling or umbilication implies a presumptive diagnosis of ectopic pancreas during preoperative endoscopy. The characteristic EUS features of ectopic pancreas, including indistinct margins, heterogeneous echogenicity (mainly hypoechoic accompanied by scattered small hyperechoic areas), presence of an anechoic area and fourth-layer thickening, and location within the second, third, and/or fourth layers are very useful for establishing a preoperative diagnosis of...
ectopic pancreas⁴,¹². Heterogeneous hypoechoic or mixed echogenicity, resembling that of the normal pancreatic parenchyma, corresponds to the presence of acinous tissue with scattered adipose tissue within the lesion⁴. Anechoic areas indicate duct dilatation, and fourth-layer thickening is considered a consequence of the hypertrophy of the muscularis propria⁹. Although these endoscopic and EUS findings are suggestive of ectopic pancreas, the accuracy for the diagnosis of subepithelial tumors is limited⁶,¹³. In fact, none

Table 1  Summary of clinicopathologic and endoscopic ultrasonography features in eight patients with ectopic pancreas

| Case | Sex | Age (yr) | Symptoms | Location | EUS features | Treatment | Follow-up period (mo) |
|------|-----|---------|----------|----------|--------------|-----------|----------------------|
| 1    | F   | 18      | Dyspepsia| Antrum   | 2, 3         | Hypoechoic | Homogenous           | Distinct  | Absent | EMR   | 28 |
| 2    | F   | 44      | None     | Antrum   | 2, 3         | Hypoechoic | Homogenous           | Distinct  | Absent | EMR   | 40 |
| 3    | F   | 37      | None     | Antrum   | 3            | Hypoechoic | Heterogeneous        | Indistinct| Absent | EMR   | 35 |
| 4    | M   | 45      | None     | Antrum   | 3            | Hypoechoic | Homogenous           | Distinct  | Present| ESD   | 21 |
| 5    | F   | 43      | None     | Lower body| 3           | Hypoechoic | Homogenous           | Distinct  | Present| ESD   | 27 |
| 6    | F   | 22      | Epigastric pain | Antrum | 3            | Hypoechoic | Heterogeneous        | Indistinct| Present| ESD   | 38 |
| 7    | F   | 57      | Epigastric pain | Antrum | 2, 3       | Hypoechoic | Heterogeneous        | Indistinct| Present| ESD   | 26 |
| 8    | F   | 24      | Dyspepsia | Lower body| 2, 3       | Hypoechoic | Homogenous           | Distinct  | Absent | ESD   | 24 |

EUS: Endoscopic ultrasonography; EMR: Endoscopic mucosal resection; ESD: Endoscopic submucosal dissection.
of our cases showed typical endoscopic findings such as central dimpling or umbilication and four of them did not exhibit a characteristic anechoic duct structure by EUS. Therefore, three lesions with anechoic duct structure were diagnosed as ectopic pancreas but the other five lesions, without anechoic duct structure, were suspicious as ectopic pancreas or were diagnosed as other diseases such as inflammatory fibrinoid polyp.

Histological diagnosis of ectopic pancreas is usually difficult when tissue specimens are obtained using conventional endoscopic biopsy forceps. For precise histological diagnosis, endoscopic techniques for obtaining deeper specimens are necessary, such as EUS-guided biopsy or combined strip biopsy and bite-on-bite biopsy. Endoscopic removal of gastric ectopic pancreas is also useful for accurate diagnosis and treatment. The diagnosis of ectopic pancreas was not made based on the pathological appearance of specimens taken with standard endoscopic biopsy forceps in any of our cases.

EUS provides the most useful information regarding tumor location within the gastric wall, helps to distinguish subepithelial lesions, and assists in establishing indications for endoscopic removal. Endoscopic removal of submucosal lesions, especially ESD, is considered dangerous because of the risk of perforation or bleeding. There have only been a few reports describing EMR methods for gastric ectopic pancreas, such as strip biopsy, cap-assisted EMR, or ligation-assisted EMR. In the present study, we first planned to remove the lesions by EMR and we therefore injected saline beneath the lesions. However, in four cases, we were forced to switch to ESD and removed the lesions without any complications. The current series, to our knowledge, is the first to describe the use of ESD for removal of gastric ectopic pancreas. Therefore, in cases for which conventional EMR is difficult or impossible, ESD may be used as an alternative method for successful removal of ectopic pancreas.

**COMMENTS**

**Background**

Ectopic pancreas is mostly found in the upper gastrointestinal tract adjacent.
to the pancreas; in 90% of patients with ectopic pancreas, it was found in the stomach, duodenum, or proximal part of the jejunum. Histologic diagnosis of ectopic pancreas is usually difficult when tissue specimens are obtained using a standard endoscopic biopsy forceps. Recently, endoscopic ultrasonography (EUS) was reported to be helpful for diagnosing ectopic pancreas. However, the accuracy of EUS for the diagnosis of subepithelial tumors is limited.

Research frontiers
Options for treatment for gastric ectopic pancreas include observation, surgery, or endoscopic resection. There have only been a few reports describing endoscopic mucosal resection (EMR) for gastric ectopic pancreas, such as strip biopsy, cap-assisted EMR, or ligation-assisted EMR. In the present study, the authors first planned to remove the lesions by EMR and we therefore injected saline beneath the lesions. However, in some cases, they were forced to switch to endoscopic submucosal dissection (ESD) and removed the lesions without any complications. The current series is the first to describe the use of ESD for removal of gastric ectopic pancreas.

Innovations and breakthroughs
To decrease the risk of perforation or bleeding, EMR is usually used to remove submucosal lesions. Usually saline solution including a small amount of epinephrine and indigo carmine is injected beneath the lesions. Then, the lesions are properly elevated and are resected by the injection-and-cut technique. However, in some cases, the saline spreads into surrounding normal tissue and the lesions become flattened, which makes it impossible to remove the lesions via the injection-and-cut technique. In these cases, ESD may be used as an alternative method for successful removal of subepithelial lesions.

Applications
When conventional EMR is difficult or impossible, ESD may be used as an alternative method for successful removal of subepithelial lesions such as ectopic pancreas.

Terminology
Ectopic pancreas, also called heterotopic or aberrant pancreas, is defined as pancreatic tissue lying outside its normal location and lacking anatomic or vascular connections with the pancreas. It has been found in 0.6% to 13% of autopsies and has also been noted in approximately one of every 500 surgical operations involving the upper abdomen.

Peer review
The paper is well written and easy to read. It is also well supported by excellent endoscopic images and pathology slides.

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