ACG CASE REPORTS JOURNAL

CASE REPORT | ENDOSCOPY

# Submucosal Tunneling Endoscopic Resection for the Management of Heterotopic Pancreas With Cystic Degeneration

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## ABSTRACT

Heterotopic pancreas is pancreatic tissue present outside of the normal location of the pancreas. In the presence of cystic degeneration, heterotopic pancreas is clinically significant because of the symptoms it causes and its physical resemblance to cancerous growth. A diagnosis of heterotopic pancreas is achieved with the aid of various endoscopic techniques for tissue removal. Submucosal tunneling endoscopic resection has proven successful for the resection of gastric subepithelial masses. We present a 53-year-old woman undergoing submucosal tunneling endoscopic resection for the resection of a subepithelial gastric cyst caused by heterotopic pancreas with cystic degeneration.

## INTRODUCTION

Heterotopic pancreas is pancreatic tissue that is found outside of the normal location of the pancreas and lacks a physical connection to the organ.[1] The tissue may become problematic if it causes symptoms, undergoes malignant transformation, or resembles malignancy on imaging. Although the lesion is usually asymptomatic, symptoms may occur if the heterotopic pancreas is of a large size or has high involvement of the mucosa.[2] These symptoms may include gastrointestinal bleeding, ulceration, abdominal pain, heterotopic pancreatitis, pseudocyst formation, and obstruction.[1,3] The management of heterotopic pancreas is not only important for the control of symptoms. Malignant transformation has been noted to occur to a higher degree in heterotopic pancreas in the stomach, making malignancy a pressing concern if the ectopic tissue is found in this location.[4] Even if there is no malignant transformation, the manner in which the tissue can resemble malignancy may pose problems in the clinical assessment of patients.

## CASE REPORT

A 53-year-old woman with gastroesophageal reflux disease and peptic ulcer disease underwent an esophagogastroduodenoscopy for chronic dyspepsia. Her history was unremarkable for excessive alcohol intake or chronic pancreatitis. A subepithelial lesion (SEL) in the gastric body was incidentally found. Further exploration by endoscopic ultrasound revealed a 13-mm round lesion involving the submucosa in the proximal antrum. The lesion was well-circumscribed, multiloculated, and anechoic-hypoechoic, suggestive of a cystic lesion (Figure 1). Bite-on-bite biopsies were negative for pathology, and there were no indications of enlarged perigastric lymph nodes. The lesion raised suspicion for a pancreatic cyst with cystic degeneration, a granular cell tumor, a carcinoid tumor, and other neoplasms. Given the presenting symptoms, unclear etiology, and preference to avoid long-term surveillance, endoscopic resection was performed.

After discussing risks and benefits, the patient consented to submucosal tunneling endoscopic resection (STER). The lesion was marked with the tip of an endoknife; a premixture of hetastarch and methylene blue was injected into the submucosa, and the tumor

*ACG Case Rep J* 2020;7:e00419. doi:10.14309/crj.0000000000000419. Published online: July 9, 2020

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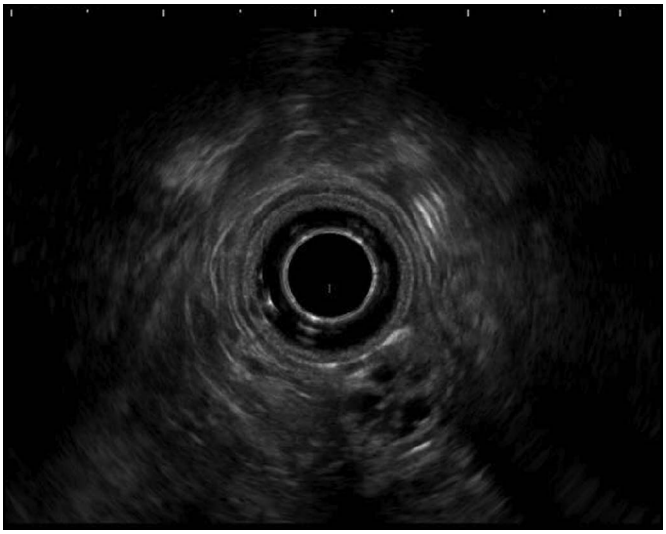

**Figure 1.** Endoscopic ultrasound finding of a multiloculated, anechoic-hypoechoic lesion in the proximal antrum.

was removed by submucosal dissection (Figure 2). This created a submucosal pocket with intact muscularis propria that was closed with endoscopic clips, resulting in an en bloc resection without complications (Figure 3).

Pathology revealed an encased tan brown, mottled, and cystic nodule (1.8 × 1.1 × 0.6 cm) consistent with pancreatic heterotopia with cystic degeneration. The cystic structures were lined by benign-appearing columnar cells with mucin production consistent with the pancreatic ducts. The solid areas of the sample revealed pancreatic acinar cells (Figure 4). The final diagnosis was pancreatic heterotopia with cystic degeneration.

After the procedure, the patient's omeprazole dose was increased from 20 to 40 mg/d for 30 days. She was instructed to follow a liquid diet for 24 hours and then a soft food diet for 48 hours. Because pathology showed a benign lesion, an esophagogastroduodenoscopy follow-up was not scheduled. The patient has since reported feeling well with improvement in her dyspepsia.

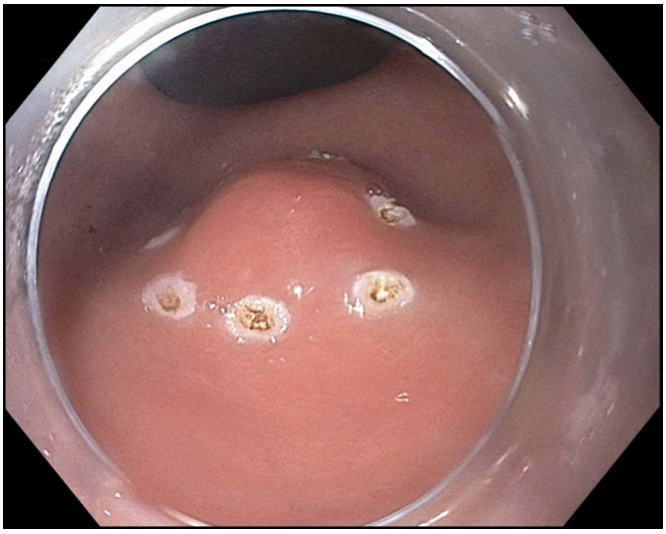

**Figure 2.** Marking of the subepithelial lesion with an endoknife.

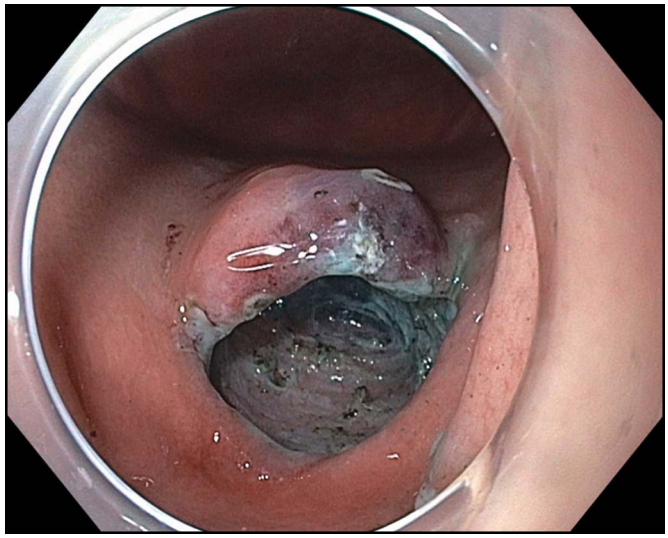

**Figure 3.** Submucosal pocket with intact mucosa at the top and preserving the muscularis propria underneath.

## DISCUSSION

In the stomach, heterotopic pancreas may resemble gastrointestinal stromal tumors, the most prevalent submucosal gastric lesions.[5,6] This similarity lies not only in the shared physical appearance but also the fact that both masses are found in the submucosal layer.[6] This difficulty in correctly distinguishing between pancreatic heterotopia and a tumor has been noted to persist even with the use of advanced imaging techniques.[7] Cystic degeneration in pancreatic heterotopia is rare. Although the etiology is unknown, it has been hypothesized that the causes of cyst formation in ectopic pancreas are similar to those

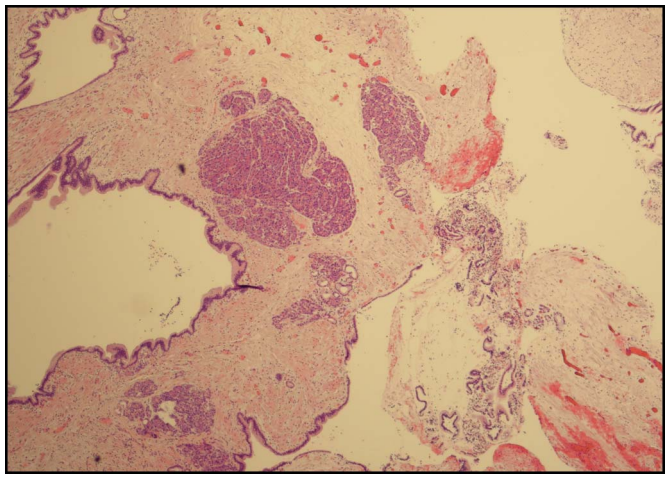

**Figure 4.** Cystic structures with pancreatic acinar cells.

in the pancreas. For example, factors that lead to pancreatitis, such as heterotopic pancreatic duct obstruction or high alcohol intake, have been thought to play a role.[8] Cystic dystrophy in heterotopic pancreas may be problematic because it may cause epigastric pain and emesis. A more pressing concern, however, is that pancreatic heterotopia with cystic degeneration may resemble a tumor with cystic degeneration, making proper characterization of the lesion necessary.[9] We present a case of a gastric SEL, later determined to be heterotopic pancreas with cystic degeneration, excised by STER.

The diagnosis of heterotopic pancreas may be accomplished after biopsy, but it is often the case that a diagnosis is made after resection because of the challenges faced in sampling tissue for a diagnosis. One manner of diagnosing heterotopic pancreas involves the use of endoscopic ultrasound-guided fine needle aspiration (EUS-FNA).[10] EUS-FNA, however, may be challenging to perform in this setting, and accuracy rates for achieving a proper diagnosis of subepithelial gastric lesions have varied greatly. Furthermore, EUS-FNA has a low diagnostic yield for small SELs, such as the one in our case.[11] Owing to these factors, EUS-FNA was not the diagnostic approach of choice in this patient. Resection was deemed necessary to both properly characterize the lesion and to remove a potential contributor to the patient's dyspepsia.

Endoscopic resection techniques such as endoscopic mucosal resection (EMR), endoscopic submucosal dissection (ESD), STER, and endoscopic full-thickness resection (EFTR) are options for the excision of gastric SELs that are difficult to diagnose. EMR has been described as an alternative to both jumbo forceps biopsy and surgery with few complications.[12] Submucosal tunneling procedures, such as ESD, however, have higher en bloc resection rates than EMR and may be good substitutes when complete resection proves challenging by EMR.[13] Any of the submucosal tunneling procedures (ESD, STER, or EFTR) would have been appropriate for the management of the mass in our patient. However, STER's reported effectiveness in the complete resection of small SELs, relatively short healing time, coupled with the fact that complications from the procedure are generally well-managed with conservative treatment, made the technique suitable for our patient. The advantage of STER over ESD is the preservation of the mucosa, which could improve healing time. EFTR, although an option, is primarily indicated for lesions involving the muscularis propria, which was not the case in our patient.[11] We demonstrated a patient with an unknown SEL that was difficult to diagnose without resection. The mass was successfully resected by STER, which allowed for a diagnosis of heterotopic pancreas with cystic degeneration.

## DISCLOSURES

Author contributions: E. Almazan wrote the manuscript, reviewed the literature, revised the manuscript for intellectual content, approved the final manuscript, and is the article guarantor. T. Runge revised the manuscript for intellectual content and wrote and approved the final manuscript. QK Li wrote and revised the manuscript for intellectual content. S. Ngamruengphong revised the manuscript for intellectual content and approved the final manuscript.

Financial disclosure: S. Ngamruengphong is a consultant for Boston Scientific.

Informed consent could not be obtained from the family of the deceased. All identifying information has been removed from this case report to protect patient privacy.

Received February 24, 2020; Accepted May 8, 2020

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