

CASE REPORT

불명료한 출혈로 발현된 공장의 이소성 췌장

최우형, 장형진, 승지환, 고봉석, 강상범

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A Case of a Jejunal Ectopic Pancreas Presenting as Obscure Gastrointestinal Bleeding

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A jejunal ectopic pancreas, where pancreatic tissue is found outside of the usual anatomical location, is a rare submucosal tumor that may cause obscure gastrointestinal (GI) bleeding. After initial negative endoscopic evaluation of the obscure GI bleeding, including colonoscopy and/or upper endoscopy, it is reasonable to proceed with further evaluation of the small bowel. Diagnostic options for the evaluation of the small bowel may include capsule endoscopy, push enteroscopy, or barium contrast small bowel studies. Here, we report a case of obscure GI bleeding caused by a jejunal ectopic pancreas, diagnosed through capsule endoscopy and barium contrast small bowel studies, which was treated successfully with single incision access laparoscopy. (*Korean J Gastroenterol* 2013;62:165-168)

Key Words: Pancreas; Gastrointestinal hemorrhage; Capsule endoscopy

INTRODUCTION

An ectopic pancreas is pancreatic tissue found outside of the usual anatomical location and it may cause inflammation, bleeding, obstruction, and lead to malignant transformation.¹ Capsule endoscopy allows for the evaluation of obscure gastrointestinal (GI) bleeding.

Here, we report a case of a jejunal ectopic pancreas that led to melena. In the present case, a jejunal ectopic pancreas was identified through capsule endoscopy and barium contrast small bowel studies and was treated with single incision access laparoscopy.

CASE REPORT

A 68-year-old woman with diabetes and essential hypertension was admitted to our hospital for evaluation of melena without abdominal pain. Physical examination on admission revealed signs and symptoms of anemia, including dizziness, headache, and pale conjunctiva. She denied any other change in bowel habit or a history of hemorrhoids. The patient had been taking sulfonyleurea (glimepiride 2 mg per day) for her diabetes and angiotensin receptor blocker (losartan 50 mg per day) for her hypertension. On presentation, her hemoglobin was 4.7 g/dL (normal, 12-15 g/dL), hematocrit 14.9% (normal, 36-46%), total bilirubin 0.3 mg/dL (normal, 0.2-1.2 mg/dL), and lactate dehydrogenase 232 IU/L (normal, 218-472 IU/L). The reticulocyte production index was 1.16.

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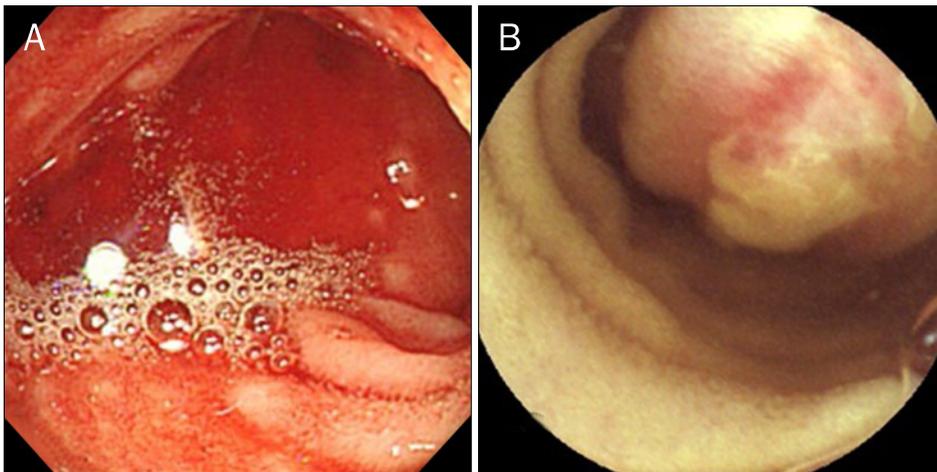


Fig. 1. (A) Fresh blood gushed out from above the terminal ileum. (B) Capsule endoscopy revealed a submucosal lump with erosion located in the jejunum.

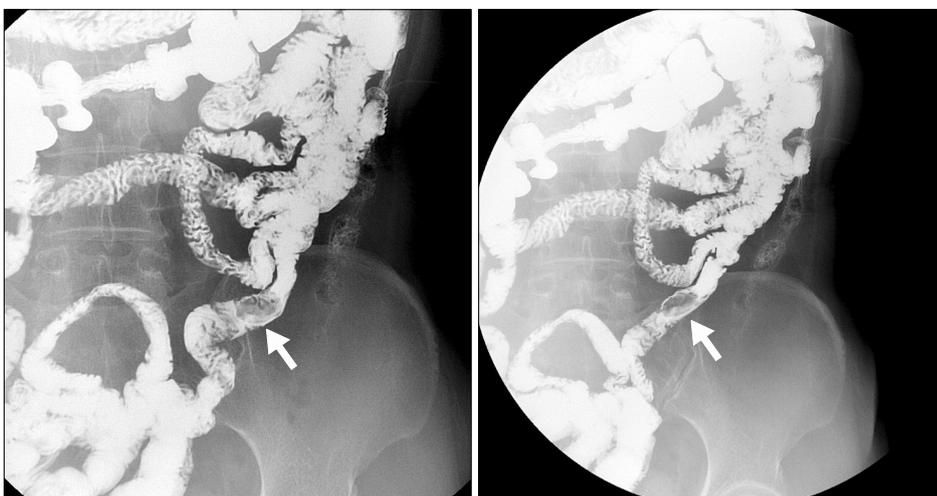


Fig. 2. The small bowel series demonstrated a 2-cm smooth, oval defect (arrow) without irregularity of the overlying mucosa in the distal jejunum.

Esophagogastroduodenoscopy and abdominal CT scanning did not reveal any significant abnormalities, such as a mass or obstruction, and colonoscopy revealed small bowel bleeding. In the terminal ileum, up to 20 cm from the ileocecal valve, fresh blood gushed out from above (Fig. 1A). Capsule endoscopy, performed to investigate the source of the obscure GI bleeding, revealed a submucosal lump with erosion located in the jejunum (Fig. 1B). A small bowel series was performed to verify the presence of the mass and it demonstrated a 2 cm-sized, smooth, oval filling defect without irregularity of the overlying mucosa in the distal jejunum (Fig. 2). The submucosal lesion in the jejunum was presumed to be the source of the obscure GI bleeding and the cause of the patient's melena. Surgical wedge resection of the submucosal lesion was carried out using a single incision laparoscopic approach. A 1.5×2.0 cm submucosal mass with an ul-

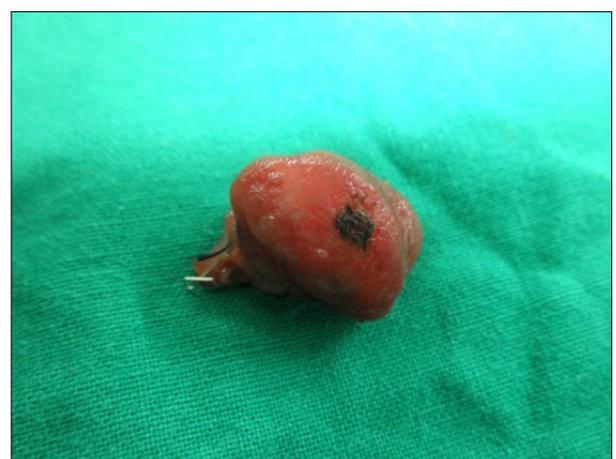


Fig. 3. The gross specimen revealed a 1.5×2.0 cm submucosal mass. The erosion with the black sanguineous crust was considered to be the cause of the obscure gastrointestinal bleeding.

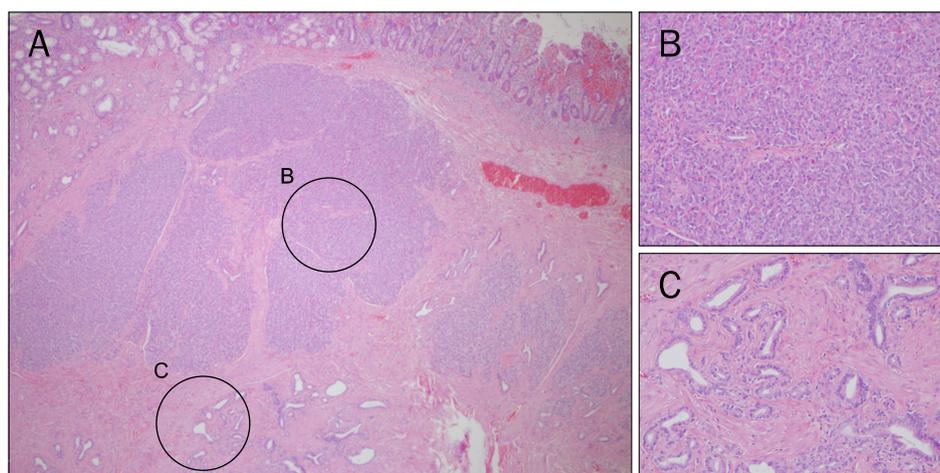


Fig. 4. A portion of the jejunum characterized as longer and with more irregular villi, no submucosal glands, and no aggregated lymphatic nodules. (A) An ectopic pancreas is located in the submucosa of the jejunum (H&E, $\times 40$). This lesion demonstrates pancreatic acini (B, $\times 200$) and secretory ducts (C, $\times 200$) in the submucosa of the jejunum (H&E).

cerated tip was noted in the gross specimen (Fig. 3). Microscopic examination revealed the presence of pancreatic ducts and secretory acinar cells without islet cells within the submucosa of the jejunum (Fig. 4). The patient recovered from surgery and had no GI symptoms during the follow-up.

DISCUSSION

An ectopic pancreas, also called a heterotopic or aberrant pancreas, was first described by Jean Schultz in 1729 and refers to pancreatic tissue that lacks anatomical and vascular connections with the pancreas.² It occurs most commonly in the stomach, duodenum, and jejunum and has been reported in other locations, including the ileum, Meckel's diverticulum, colon, gall bladder, umbilicus, fallopian tube, mediastinum, spleen, and liver.³ The diameter of the ectopic pancreatic tissue is generally 1-33 cm and it was 1.5 cm in the current case.⁴

Gaspar Fuentes and colleagues⁵ modified von Heinrich's classification of ectopic pancreas and divided this entity into four types. Type I, a total ectopic pancreas, is composed of all pancreatic cell types, type II contains pancreatic ducts only, type III is composed of acinar cells only, and type IV is composed of islet cells only.⁶ The lesion in the present case was most consistent with a type I lesion, although no islet cells were identified.

An ectopic pancreas is usually discovered incidentally during radiographic or endoscopic examination of the gut, surgery for other abdominal conditions, or upon autopsy.⁴ A patient with an ectopic pancreas can be normal or present with symptoms such as bleeding, vomiting, or abdominal pain due

to pancreatitis, intestinal obstruction, or intussusception.³ Rarely, an ectopic pancreas can be associated with other pancreatic diseases, including islet cell tumors, pancreatic carcinomas, and pancreatic cysts.¹ In the present case, the patient presented with obscure GI bleeding, and an ectopic pancreas was diagnosed during evaluation of this complaint. The prevalence of GI bleeding due to ectopic pancreas is not well established as the rarity of its morbidity. Dolan et al.⁷ reported that GI bleeding has been reported in 3 out of 73 symptomatic cases among 212 ectopic pancreas patients.

Obscure GI bleeding has been defined as bleeding of unknown origin that persists or recurs after an initial negative endoscopic evaluation, including colonoscopy and/or upper endoscopy.⁸ The majority of lesions causing obscure GI bleeding are eventually found in the small intestine. Accordingly, if conventional evaluation of GI bleeding has failed to reveal a source, it is reasonable to proceed with further evaluation of the small bowel.⁹ Diagnostic options include capsule endoscopy, push enteroscopy, or barium contrast small bowel studies.⁹ However, push enteroscopy only evaluates the proximal jejunum, and barium contrast small bowel studies are not particularly sensitive for small lesions. Additionally, push enteroscopy has potential complications, such as bowel perforation and acute pancreatitis, as well as the rare occurrence of bleeding and infection.^{10,11} In this setting, capsule endoscopy has a high sensitivity and specificity for detecting a bleeding source in patients with obscure GI bleeding.¹² For the previously mentioned reasons, capsule endoscopy is well-suited as the first choice for evaluating obscure GI bleeding. In the current patient, a source of obscure GI bleeding was detected by capsule endoscopy and confirmed by ba-

rium contrast small bowel studies.

Although endoscopic ultrasound and double balloon enteroscopy are helpful for diagnosis, an ectopic pancreas is usually buried in the submucosa, which makes it difficult to distinguish from other submucosal tumors.⁴ Endoscopic diagnosis is rarely conclusive on endoscopic biopsies because the lesion is usually located too deeply in the submucosa to easily biopsy.³ Surgical resection and histologic evaluation should be undertaken to distinguish an ectopic pancreas from malignant tumors.¹³

In regard to surgical procedures to address an ectopic pancreas, single incision laparoscopic surgery constitutes the next step in the development of minimally invasive surgery.¹⁴ In the current patient, access to the peritoneal cavity was achieved through a single port in the umbilicus.

In summary, a jejunal ectopic pancreas was revealed to be the source of obscure GI bleeding through capsule endoscopy and barium contrast small bowel studies. The number of reported cases of an ectopic pancreas of the GI tract will likely increase due to the increased use of both capsule endoscopy and double balloon enteroscopy.¹⁵

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