

Gastroduodenal intussusception of a gastrointestinal stromal tumor: a rare cause of acute pancreatitis

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Patients with symptomatic gastrointestinal stromal tumor (GIST) typically present with gastrointestinal bleeding and abdominal pain. This report presents an unusual case of fundic GIST complicated by gastroduodenal intussusception, manifesting as acute pancreatitis. The patient presented with epigastric pain and pancreatic enzyme elevation; thus, he was diagnosed with acute pancreatitis. Computed tomography showed evidence of pancreatitis and a 4×4.7 cm well-defined hyperdense lesion in the 2nd part of the duodenum, compressing the pancreatic head and pancreatic duct. Esophagogastroduodenoscopy revealed invagination of the gastric folds into the duodenum, causing pyloric canal blockage consistent with gastroduodenal intussusception. Spontaneous reduction of the lesion during endoscopy revealed a 4 cm pedunculated subepithelial mass with central ulceration originating from the gastric fundus. Endoscopic ultrasound demonstrated a heterogeneous hypoechoic lesion originating from the 4th layer of the gastric wall. Laparoscopic-endoscopic intragastric wedge resection of the fundic lesion was subsequently performed, and surgical histology confirmed GIST.

Keywords: Acute pancreatitis; Gastroduodenal intussusception; Gastrointestinal stromal tumor

INTRODUCTION

Gastrointestinal stromal tumor (GIST) accounts for approximately 1% of all gastrointestinal tract neoplasms.¹ GIST usually presents as an incidental gastric subepithelial lesion during upper endoscopy or abdominal radiologic studies. In symptomatic cases, abdominal pain and gastrointestinal bleeding are common manifestations. Gastrointestinal obstruction is a less frequent presentation, occurring in only 10%–30% of patients,²

while gastroduodenal intussusception and acute pancreatitis are extremely unusual presentations of gastric GIST. This report presents the case of a patient diagnosed with gastric GIST complicated by gastroduodenal intussusception, resulting in acute pancreatitis.

CASE REPORT

A 55-year-old male patient presented with worsening epigastric pain and vomiting for 10 days. He reported intermittent abdominal pain for 10 months prior to presentation. Abdominal examination revealed tenderness at the epigastrium. His serum amylase level was 320 U/L, compatible with acute pancreatitis. Abdominal computed tomography (CT) revealed a 4×4.7 cm well-defined hyperdense mass in the 2nd part of the duodenum, compressing the pancreatic head, edematous pancreatic parenchyma, peripancreatic fat stranding, and pancreatic duct dilatation (Fig. 1). Esophagogastroduodenoscopy was signifi-

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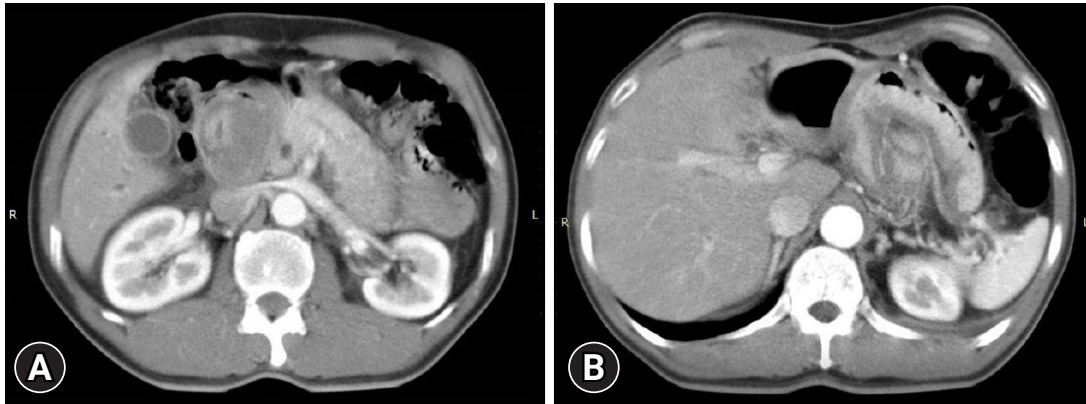


Fig. 1. (A) Computed tomography scan of the abdomen revealed a 4×4.7 cm well-defined hyperdense tumor in the 2nd part of the duodenum, edematous change of the pancreatic parenchyma, and peripancreatic fat stranding. (B) The mass was originated from the gastric fundus.

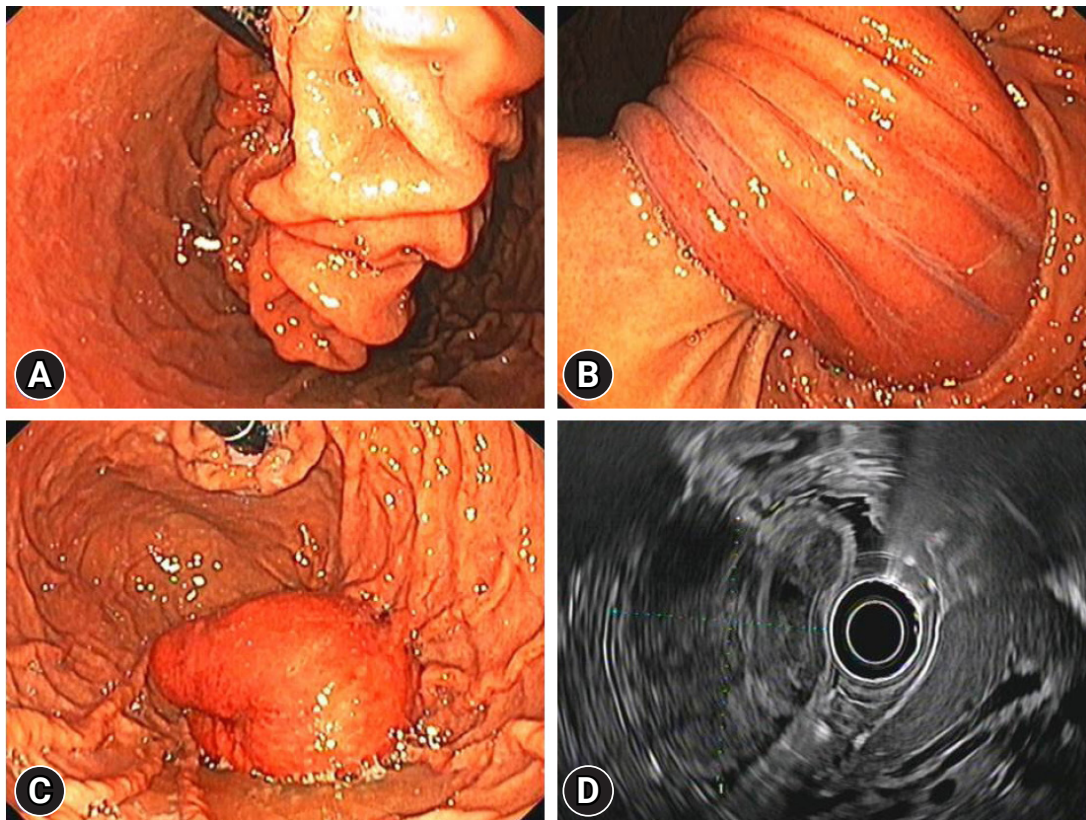


Fig. 2. Endoscopic images of the lesion. (A) Twisting of the gastric folds in the gastric cardia and fundus. (B) Torsion of the gastric folds running across the incisura into the pyloric canal. (C) A 4-cm sized, pedunculated subepithelial lesion arising from the fundus was visualized after spontaneous reduction of the intussuscepted gastric folds. (D) Endoscopic ultrasound of the subepithelial lesion demonstrating a heterogeneous hypoechoic mass originating from the 4th layer of the gastric wall.

cant for abnormal torsion of gastric folds that originated from the gastric fundus, coursed along the lesser curvature, and invaginated into the pyloric canal, thereby blocking the passage

of the scope into the duodenal bulb (Fig. 2A, B). These findings were consistent with those of gastroduodenal intussusception. The lesion spontaneously reduced back into the stomach during

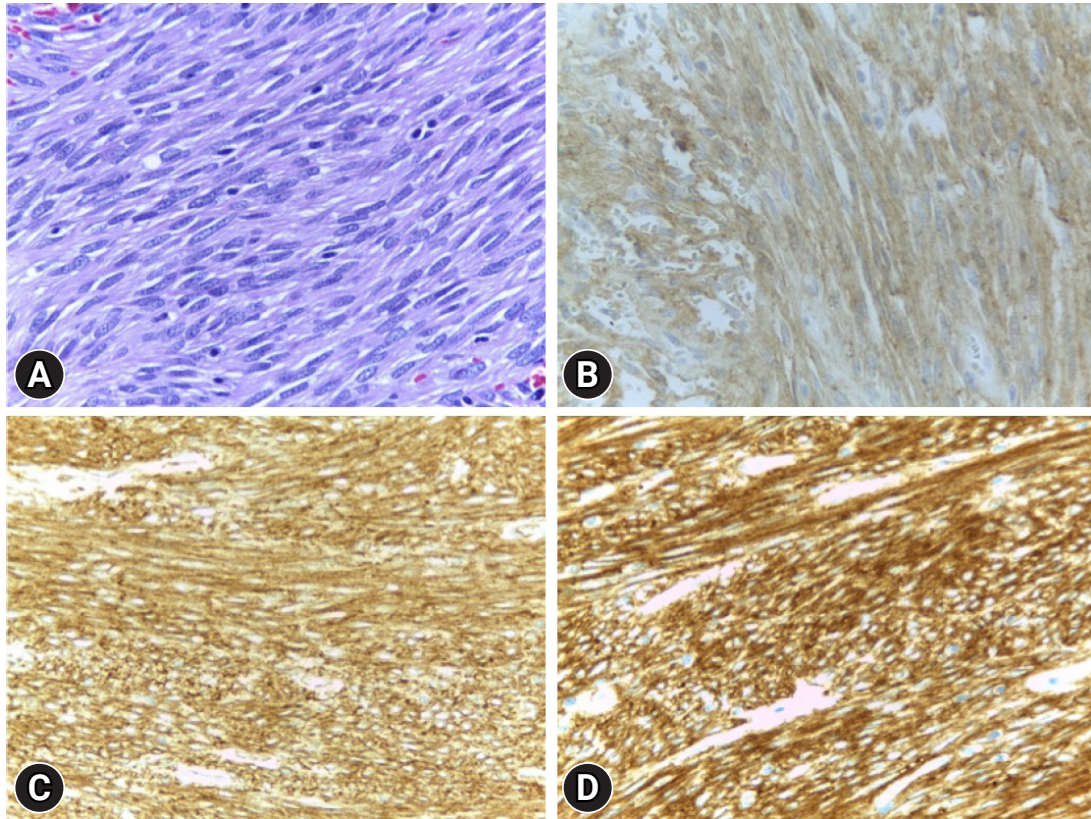


Fig. 3. (A) Pathologic examination showed spindle-shape cells on hematoxylin & eosin stain. Immunohistochemical staining of the specimen was positive for (B) CD117, (C) CD34, and (D) DOG-1, respectively. All images were taken in high-power-field magnification ($\times 400$).

the procedure, allowing visualization of an approximately 4-cm sized, pedunculated subepithelial mass with inflamed mucosa and central ulceration (Fig. 2C). Endoscopic examination of the duodenal bulb, the 2nd part of the duodenum, and the ampulla were unremarkable (Supplementary Video 1). Endoscopic ultrasound was notable for a heterogeneous hypoechoic mass originating from the 4th layer of the gastric wall (Fig. 2D), highly suggestive of GIST. Given the lesion size and location, we elected to proceed with combined laparoendoscopic intragastric wedge resection. The procedure was successfully performed without any complications. The histology revealed a 5.5 \times 4.3 \times 4 cm-sized, intramural spindle cell tumor with a mitotic rate of $<5/50$ high power field (G1). Immunohistochemical analysis showed that the tumor cells were positive for CD34, CD117, and DOG-1, but not for S100 and 1A4, confirming the diagnosis of GIST (Fig. 3). Therefore, the final diagnosis was gastric fundal GIST complicated by gastroduodenal intussusception, triggering acute pancreatitis. The patient remained asymptomatic without evidence of recurrence after 2 years of follow-up.

DISCUSSION

Gastroduodenal intussusception, first described by Hobbs and Cohen, and also known as ball valve syndrome, is an uncommon condition in which a part of the stomach invaginates into the pylorus and duodenum.³ This condition accounts for less than 10% of all intussusceptions in adults.⁴ The possible etiologies of this condition include adenomatous polyps, lipomas, hamartomas, leiomyomas, leiomyosarcomas, adenocarcinomas, and GISTs. Among the gastrointestinal tract mesenchymal tumors, GIST is the most common, with the stomach being the most predominant location, accounting for 60% of cases.⁵ The presentations of GIST generally include abdominal pain, gastrointestinal bleeding, and, to a lesser extent, bowel obstruction, although cases can also be asymptomatic.⁵ In contrast, gastroduodenal intussusception caused by GIST is incredibly infrequent; only 17 case reports of GIST complicated by gastroduodenal intussusception have been identified thus far. Most reported patients present with acute or intermittent

Table 1. Case reports of gastroduodenal intussusception causing acute pancreatitis

Study	Age (yr)	Sex	Etiology	Location	Size (cm)	Treatment
White (1991) ⁸	67	Male	Leiomyoma	Body	3.5	Gastrostomy excision
Herman (1992) ⁹	44	Female	Adenomatous polyp	Body	6	Gastrostomy excision
Senadhi (2011) ¹⁰	58	Female	GIST	Antrum	6	Partial anterior gastric resection
Jones (2012) ¹¹	65	Male	GIST	Fundus	8.2	Laparoscopic wedge resection
Sun (2012) ¹²	82	Female	GIST	Fundus	8	Surgical resection
Chahla (2014) ¹³	76	Male	Hyperplastic polyp	Antrum	12	Partial endoscopic resection
Prakash (2017) ¹⁴	65	Male	GIST	Antrum	-	Subtotal gastrectomy
Yildiz (2016) ¹⁵	85	Female	GIST	Fundus	6	Subtotal gastrectomy
Salazar-Alarcón (2017) ¹⁶	68	Male	Adenocarcinoma	Antrum	7	Subtotal gastrectomy
Jetha (2018) ¹⁷	86	Female	Adenomatous polyp	Fundus	8.8	Laparoscopic resection
Bejjani (2018) ¹⁸	84	Female	Adenocarcinoma	Antrum	7.5	Partial lateral gastrectomy

GIST, gastrointestinal stromal tumor.

episodes of epigastric pain and vomiting that are compatible with clinical gastric outlet obstruction for a day to months leading up to diagnosis. The most common tumor location is the antrum and fundus, with sizes ranging from 2.5 cm to 8 cm in maximum dimension. All patients underwent surgical resection.⁶

Our patient developed acute pancreatitis, which is an exceedingly peculiar complication of gastroduodenal intussusception. This condition was believed to be triggered by pancreatic duct distortion or compression secondary to the duodenal wall traction or direct obstruction of the pancreatic duct orifice by the intussusceptum.⁷ Extensive literature search to date only yielded reports of 11 cases of such exceptional circumstances, and only five cases of acute pancreatitis secondary to gastroduodenal intussusception of gastric GIST (Table 1).⁸⁻¹⁸ The mean age of these five patients was 71 years, with a slight female predilection. Three patients had pedunculated GISTs dangling from the fundus, causing intussusception, while the remaining two patients had GISTs located in the antrum. Tumor size ranged from 6 cm to 8.8 cm. All patients underwent successful surgical resection.

Endoscopically, GIST appears as a smooth bulge of subepithelial lesions covered by normal overlying mucosa, with or without central ulceration, and with a firm consistency when probed with biopsy forceps.¹⁹ Endoscopic ultrasound usually demonstrates hypoechoic homogeneous lesions arising from either the 2nd or 4th layer, although heterogeneous lesions with anechoic spaces and calcifications may also be found.¹⁹ Contrast-enhanced CT scans can help evaluate lesion extension, evidence of malignant metastasis, and complications such as intussusception and obstruction. As in our case, endoscopy and CT helped in

the diagnosis.

Reduction of the intussusception can be performed endoscopically; however, this technique may not always be feasible in cases with totally obliterated lumens. In such a scenario, open surgical reduction may be required unless spontaneous reduction occurs, as demonstrated in our case. Resection by either laparoscopic or open surgery remains the only definitive treatment for GIST to date. Endoscopic submucosal dissection, submucosal tunnel resection, and full-thickness resection are viable options for 2 to 4 cm GIST.¹⁹

In conclusion, gastric GIST complicated by gastroduodenal intussusception is uncommon. Here, we present a case of acute pancreatitis secondary to gastroduodenal intussusception from a pedunculated gastric GIST at the fundus. Our patient subsequently underwent successful combined laparoscopic and endoscopic intragastric wedge resection. This diagnosis should be considered in patients presenting with acute pancreatitis and gastric outlet obstruction.

Supplementary Material

Supplementary Video 1. Upper endoscopy demonstrated gastroduodenal intussusception with spontaneous reduction of the pedunculated subepithelial mass originating from the gastric fundus (<https://doi.org/10.5946/ce.2021.073.v001>).

Supplementary materials related to this article can be found online at <https://doi.org/10.5946/ce.2021.073>.

Conflicts of Interest

The authors have no potential conflicts of interest.

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Author Contributions

Conceptualization: PN, SN, NP; Data curation: PN, SN, TP, NP; Formal analysis: PN, SN, NP; Investigation: PN, TP, NP; Project administration: NP; Supervision: NP; Validation: SN, NP; Visualization: PN, SN, TP, NP; Writing-original draft: PN, SN; Writing-review & editing: SN, TP, NP.

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