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CASE REPORT

Gastroduodenal intussusception caused by gastric gastrointestinal stromal tumor: A case report and review of the literature

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Abstract

BACKGROUND

Gastric gastrointestinal stromal tumor (GIST) is the most common etiology of gastroduodenal intussusception. Although gastroduodenal intussusception caused by gastric GIST is mostly treated by surgical resection, the first case of gastroduodenal intussusception caused by gastric GIST was treated by endoscopic submucosal dissection (ESD) in Japan in 2017.

CASE SUMMARY

An 84-year-old woman presented with symptoms of postprandial fullness with nausea and occasional vomiting for a month. Initially, she visited a local clinic for help, where abdominal sonography revealed a space-occupying lesion around the liver, so she was referred to our hospital for further confirmation. Abdominal sonography was repeated, which revealed a mass with an alternating concentric echogenic lesion. Esophagogastroduodenoscopy (EGD) was performed under the initial impression of gastric cancer with central necrosis and showed a tortuous distortion of gastric folds down from the lesser curvature side to the duodenal bulb with stenosis of the gastric outlet. EGD was barely passed through to the 2nd portion of the duodenum and a friable ulcerated mass was found. Several differential diagnoses were suspected, including gastroduodenal intussusception, gastric cancer invasion to the duodenum, or pancreatic cancer with adherence to the gastric antrum and duodenum. Abdominal computed tomography for further evaluation was arranged and showed gastroduodenal intussusception with a long stalk polypoid mass 5.9 cm in the duodenal bulb. Under the impression of gastroduodenal intussusception, ESD was performed at the base of the

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gastroduodenal intussusception; unfortunately, a gastric perforation was found after complete resection was accomplished, so gastrorrhaphy was performed for the perforation and retrieval of the huge polypoid lesion. The gastric tumor was pathologically proved to be a GIST. After the operation, there was no digestive disturbance and the patient was discharged uneventfully on the 10th day following the operation.

CONCLUSION
We present the second case of gastroduodenal intussusception caused by GIST treated by ESD. It is also the first case report of gastroduodenal intussusception by GIST in Taiwan, and endoscopic reduction or resection is an alternative treatment for elderly patients who are not candidates for surgery.

Key Words: Gastric gastrointestinal stromal tumor; Endoscopic submucosal dissection; Gastro-duodenal intussusception; Elderly; Esophagogastroduodenoscopy; Gastrointestinal obstruction; Case report

Core Tip: This is the first case report of gastroduodenal intussusception caused by gastrointestinal stromal tumor in Taiwan and endoscopic reduction or resection is an alternative treatment for elderly patients who are not candidates for surgery.

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INTRODUCTION

Gastric outlet obstruction (GOO) is a clinical syndrome characterized by epigastric abdominal pain and postprandial vomiting due to mechanical obstruction. Benign disease such as peptic ulcer disease was responsible for 90% of cases until the late 1970s[1]. With the decline in the incidence of peptic ulcer disease, it is estimated that 50-80% of all cases of GOO are attributable to malignancies. Distal gastric cancer remains a relatively common cause of malignant GOO, accounting for up to 35% of GOO cases[2]. Gastro-duodenal intussusception is a rare cause of GOO in adults, and it is typically caused by a pathological leading point, malignant in over one half of cases[3]. Herein, we report an 84-year-old woman with gastroduodenal intussusception caused by a gastric gastrointestinal stromal tumor (GIST).

CASE PRESENTATION

Chief complaints

An 84-year-old woman presented with symptoms of postprandial fullness with nausea and occasional vomiting for a month.

History of present illness

The patient suffered from persistent hematemesis and tarry stool complicated with orthostatic hypotension over the past 2-3 years. She complained about abdominal distress, abdominal fullness, nausea, and vomiting in recent one month. She first visited a local clinic, where abdominal sonography showed a liver tumor.

History of past illness

The patient had a history of hypertension, chronic kidney disease, and hepatitis B virus infection.

Personal and family history

The patient denied any personal history of alcohol, betel nuts, and cigarette consumption. She also denied travel, contact, and cluster history in recent 6 mo. As a housewife, she did not have any occupational history. Regarding her family history, she had one elder brother and four younger sisters. All of them did not have any malignancy history.

Physical examination

On the physical examination, the patient's consciousness was alert (E4V5M6); her conjunctiva was not pale; she had anicteric sclera; her chest had symmetric movement with respiration; Her breath sound was bilaterally clear; and she had regular heart beat, flat abdomen, normoactive bowel sound, no muscle guarding, no tenderness, no rebound pain, and no pitting edema.

Laboratory examinations

The results of laboratory examinations are shown in Table 1.

Imaging examinations

Abdominal sonography was repeated, which revealed a mass with an alternating concentric echogenic lesion (Figure 1). EGD was performed under the initial impression of gastric cancer with central necrosis and showed a tortuous distortion of gastric folds down from the lesser curvature side to the duodenal bulb with stenosis of the gastric outlet (Figure 2A). EGD was barely passed through to the 2nd portion of the duodenum and a friable ulcerated mass was found (Figure 2B). Several differential diagnoses were suspected, including gastroduodenal intussusception, gastric cancer invasion to the duodenum, or pancreatic cancer with adherence to the gastric antrum and duodenum. Abdominal computed tomography for further evaluation was arranged and showed gastroduodenal intussusception with a long stalk polypoid mass (5.9 cm) in the duodenal bulb (Figure 3).

Further diagnostic work-up

Under the impression of gastroduodenal intussusception, endoscopic submucosal dissection (ESD) was performed at the base of the gastroduodenal intussusception; unfortunately, a gastric perforation was found after complete resection was accomplished, so gastrorrhaphy was performed for the perforation and retrieval of the huge polypoid lesion (Figure 4).

FINAL DIAGNOSIS

The gastric tumor was pathologically diagnosed as a GIST (Figure 5).

TREATMENT

Endoscopic resection and laparotomy were performed for gastric tumor removal and gastrorrhaphy.

OUTCOME AND FOLLOW-UP

The patient had a complete remission.

DISCUSSION

Regarding gastrointestinal obstruction in adults, symptoms are variable depending on the locations of obstruction, which range from small bowel obstruction followed by large intestine and gastric outlet complications[4]. It is mostly caused by reasons such as adhesion, malignancy, and volvulus. In adults, intussusception accounts merely for 1% of mechanical gastrointestinal obstructions, representing a very rare cause[5]. The symptoms of intussusception are nausea, vomiting, gastrointestinal bleeding, change

Table 1 Laboratory examinations

	Result	Reference range
WBC	9.37	4.4-11.3 × 10^9 L
Hb	9.1	12.3-15.3 g/dL
Plt	302	(160-370) × 1000/uL
CRP	2.04	mg/L
Crea	1.53	mg/dL
BUN	25.9	mg/dL
Na	139	mmol/L
K	3.9	mmol/L
GOT	16	IU/L
GPT	11	IU/L
INR	0.97	
PTT	24.3	sec
CEA	3.58	

WBC: White blood cells; Hb: Hemoglobin; Plt: Platelets; CRP: C reactive protein; Crea: Creatinine; BUN: Blood urea nitrogen; GOT: Glutamic oxalacetic transaminase; GPT: Glutamic pyruvic transaminase; CEA: Carcinoma embryonic antigen.

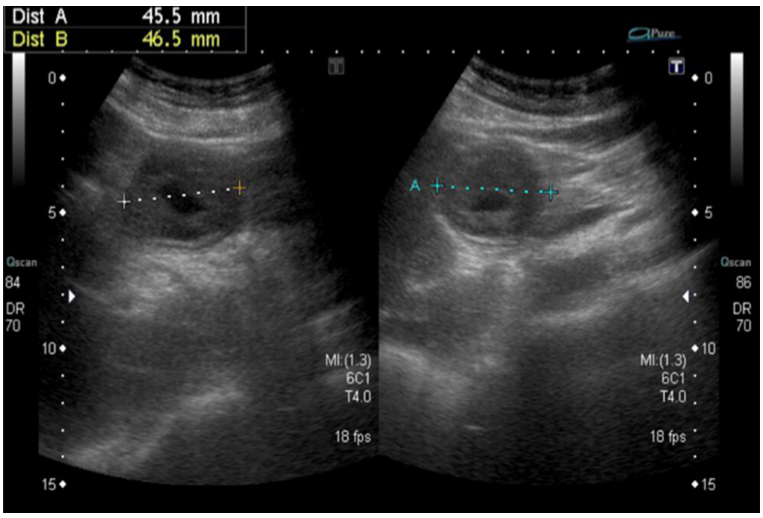

Figure 1 Abdominal ultrasound revealed the doughnut sign, measuring 4.5 cm × 4.6 cm.

in bowel habits, constipation, or abdominal pain[6]. Ischemic change and peritonitis seldom occur but represent major critical complications of intussusception. In adults, intussusception is usually the result of lesions, including scar-like tissue in the intestine (adhesions) and prior surgery such as gastrointestinal bypass surgery for weight control, polyp, or tumor. The presenting case suffering from partial gastric outlet obstruction by gastroduodenal intussusception was managed by ESD and gastrorrhaphy proved that is was caused by a GIST.

GIST accounts for around 0.2% of all gastrointestinal tumors and occurs anywhere along the gastrointestinal tract, but most commonly in the stomach (40%-60%) and jejunum/ileum (25%-30%)[7]. GIST is typically asymptomatic or has nonspecific symptoms (*i.e.*, early satiety and bloating), unless they ulcerate, bleed, or grow large enough to cause pain or obstruction. Conceivably, gastroduodenal intussusception caused by GIST most commonly presents with nonspecific symptoms of acute or intermittent abdominal pain with vomiting lasting from days to several months[8]. By reviewing the relevant literature, we found 41 cases of gastroduodenal intussusception within the past 20 years (Table 2)[9-44]. Gastric GIST is the most common etiology and

Table 2 Review of case reports on gastroduodenal intussusception

Ref.	Year	Age	Sex	Diagnosis	Pathology report	Management	Size
Nakagawara *et al*[9]	2000	50	F	EGD	Gastric heterotopia	Endoscopic polypectomy	30 mm × 36 mm
Sankaranunni *et al*[10]	2001	48	M	CT	Gastric lipoma	Laparotomy	NA
Harrison *et al*[11]	2001	76	M	EGD	Leiomyoma	Laparotomy	50 mm × 42 mm
Mouës *et al*[12]	2002			EGD and CT	Gastric lipoma	Laparotomy	50 mm × 100 mm
Crowther *et al*[13]	2002	59	F	CT	GIST	Partial gastrectomy	60 mm
Vinces *et al*[14]	2005	72	M	Laparoscopy	Gastric lipoma	Exploratory laparotomy	NA
Vinces *et al*[14]	2006				Gastric lipoma		NA
Juglard *et al*[15]	2006				Ménétrier's disease		NA
Adjepong *et al*[16]	2006	84	M	CT	GIST	Laparoscopic Billroth II partial gastrectomy	40 mm × 30 mm
Samamé *et al*[17]	2007				GIST		NA
Shum *et al*[18]	2007	34	F	CT	GIST	Partial gastrectomy	50 mm × 50 mm
Shum *et al*[18]	2008	67	M	Ultrasound and EGD	Gastric carcinoma	Surgical resection	45 mm × 40 mm
Alamili *al*[19]	2008			CT	Duodenal lipoma	Surgical resection	NA
Siam *et al*[20]	2008	29	M	EGD	GIST	Partial Gastrectomy	60 mm × 60 mm
Su *et al*[21]	2009	24	M	EGD	Gastric carcinoma (PJS)	Surgical resection	30 mm
Hillenbrand *et al*[22]	2009	42	F	CT	Post banded gastroplasty	Surgical reduction	
Chan *et al*[23]	2009	34	F	CT	GIST	Laparoscopic wedge resection	65 mm × 44 mm
Eom *al*[24]	2011	73	F	CT and EGD	Gastric carcinoma	Subtotal gastrectomy	78 mm × 75 mm
Euanorasetr *et al*[25]	2011				Gastric carcinoma	Subtotal gastrectomy	NA
Gyedu *et al*[26]	2011	59	F	CT and US	GIST	Partial gastrectomy	70 mm × 60 mm
Seok *et al*[27]	2012	51	M	CT and EGD	GIST	Gastric partial resection	55 mm × 42 mm
Seok *et al*[27]	2012	62	F	EGD and CT	GIST	Billroth II partial gastrectomy	52 mm × 35 mm
Wilson *et al*[28]	2012	78	F	CT	GIST	Laparoscopic wedge resection	44 mm × 33 mm
Chen *et al*[29]	2013	63	F	CT and EGD	Gastric hamartomatous polyp	Endoscopic mucosal resection	NA
Rittenhouse *et al*[30]	2013	52	F	CT	GIST	Laparoscopic wedge resection	50 mm × 50 mm
Chahla *et al*[31]	2014	76	M	CT	Gastric hyperplastic polyp	Endoscopic resection	< 30 mm
Khanna *et al*[32]	2014	33	M	CT and EGD	Brunner's gland hamartoma	Duodenostomy and polypectomy	35 mm × 70 mm
Kadowaki *et al*[33]	2014	77	F	Laparotomy	Gastric collision tumor	Gastrotomy followed by duodenotomy	120 mm
Yang *et al*[34]	2015	63	M	CT	Gastric schwannoma	Conventional laparotomy	55 mm × 48 mm
M S *et al*[35]	2015	74	M	CT	GIST	Partial gastrectomy	NA
Indiran *et al*[36]	2015				GIST		NA

Yildiz *et al*[37]	2016	85	F	CT	GIST	Subtotal gastrectomy	60 mm × 50 mm
Komatsubara *et al*[38]	2016	90	F	EGD	GIST	Wedge resection	50 mm × 45 mm
Yamauchi *et al*[9]	2017	95	F	CT	GIST	Endoscopic submucosaldissection	42 mm × 39 mm
Jameel *et al*[39]	2017	65	F	EGD and CT	GIST	Laparoscopic resection	60 mm × 60 mm
Casimiro Pérez *et al*[40]	2018	55	M	EGD and CT	Gastric submucosal lipoma	Laparoscopic transgastric excision	63 mm × 55 mm
Zhou *et al*[41]	2018	69	M	EGD and CT	GIST	Laparoscopic resection	45 mm × 40 mm
Ssentongo *et al*[42]	2018	85	F	CT	GIST	Wedge resection	25 mm × 25 mm
De *et al*[43]	2018	42	F	EGD	GIST	Surgical resection	80 mm × 70 mm
Đokić *et al*[8]	2019	62	M	CT and US	GIST	Laparotomy resection	75 mm × 55 mm
Suda *et al*[44]	2019	81	F	EGD and CT	Gastric carcinoma	Laparoscopic gastrectomy	55 mm
Our case	2020	84	M	US and EGD and CT	GIST	Endoscopic submucosaldissection and surgical repair	59 mm

EGD: Esophagogastroduodenoscopy; F: Female; CT: Computed tomography; GIST: Gastrointestinal stromal tumor; M: Male; NA: Not available.

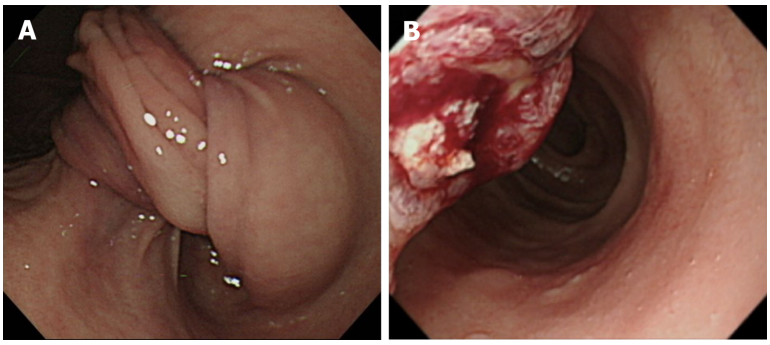

Figure 2 Esophagogastroduodenoscopy. A: Gastro-duodenal intussusception; B: Ulcerated polypoid lesion.

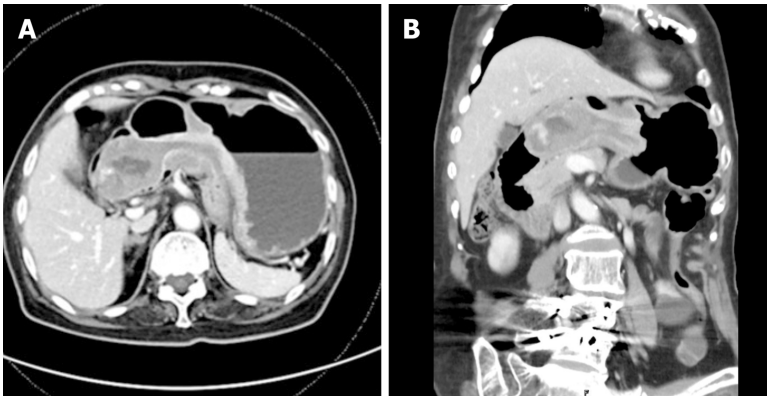

Figure 3 Abdominal computed tomography revealed intussusception with a long stalk polypoid mass 5.9 cm in the duodenal bulb. A: Axial view; B: Coronal view.

accounts for more than half of these cases, with the mean size of the GIST being 54.8 mm and the average age being 64.25 years (range, 29-95 years). Management of gastroduodenal intussusception included surgical intervention and endoscopic

Figure 4 Cardia submucosal tumor measuring 5.6 cm × 4.5 cm × 3.5 cm.

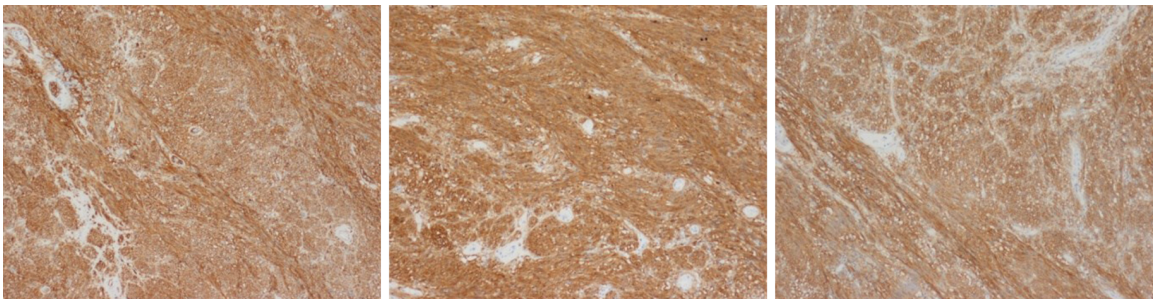

Figure 5 Microscopic examination.

reduction in the past, and for the present case, endoscopic reduction of the invagination was tried but failed due to its large size (5.9 cm). Although gastroduodenal intussusception caused by gastric GIST is mostly treated by surgical resection, the first case of gastroduodenal intussusception caused by gastric GIST was treated by ESD in Japan in 2017[45], so ESD was also tried for this case with the result of complete resection although complicated with perforation. Finally, gastrorrhaphy repair and retrieval of the huge polypoid lesion were accomplished. Here we present the second case of gastroduodenal intussusception caused by GIST treated by ESD. It is also the first case report of gastroduodenal intussusception caused by GIST in Taiwan, and endoscopic reduction or resection is an alternative treatment for elderly patients who are not candidates for surgery.

CONCLUSION

We present the second case of gastroduodenal intussusception caused by GIST treated by endoscopic submucosal dissection. It is also the first case report of gastroduodenal intussusception caused by GIST in Taiwan, and endoscopic reduction or resection is an alternative treatment for elderly patients who are not candidates for surgery.

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