

# Gastroduodenal Intussusception with a Gastric Gastrointestinal Stromal Tumor Treated by Endoscopic Submucosal Dissection

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## Abstract

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A 95-year-old Japanese woman presented to our hospital with intermittent vomiting and several episodes of melena. Abdominal computed tomography revealed intussusception of the gastric tumor into the duodenum. After endoscopic reduction, endoscopic ultrasonography identified a hypoechogenic lesion limited to the submucosal layer. Endoscopic resection was performed as a localized treatment for the prevention of recurrent gastroduodenal intussusception. To our knowledge, there have been no other reports describing a gastric gastrointestinal stromal tumor presenting with gastroduodenal intussusception and treated using an endoscopic submucosal dissection technique.

**Key words:** gastrointestinal stromal tumor, gastroduodenal intussusception, ball valve syndrome, endoscopic submucosal dissection

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## Introduction

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Gastroduodenal intussusception is a rare occurrence that accounts for less than 10% of all intussusception cases in adults. As the majority of such cases are secondarily caused by a tumorous lesion that serves as a lead point, intussusception in adults often recurs even after successful non-operative reduction and thus requires surgical resection. We herein describe a case of a gastric gastrointestinal stromal tumor (GIST) presenting with gastroduodenal intussusception repositioned via endoscopy and resected using an endoscopic submucosal dissection (ESD) technique.

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## Case Report

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A 95-year-old woman with dementia and a gastric submucosal tumor presented to the emergency room with intermit-

tent vomiting and several episodes of melena. Although the tumor had been identified 10 years prior, considering the patient's underlying diseases, no further examination was performed at the time.

Anemia of the conjunctiva and epigastric tenderness were observed on a physical examination. Laboratory findings revealed microcytic hypochromic anemia [hemoglobin, 8.1 g/dL (normal range: 11.3-15.2 g/dL); hematocrit, 23.8% (normal range: 33.4-44.9%)] and decreased levels of albumin [2.8 g/dL (normal range: 3.8-5.2 g/dL)]. Abdominal computed tomography (CT) revealed intussusception of the gastric submucosal tumor into the duodenum with no distant metastasis (Fig. 1). An emergency endoscopic examination revealed invagination of the gastric body into the duodenum (Fig. 2). After disinvasion by endoscopic manipulation, a submucosal tumor was identified in the lower gastric body, approximately 40 mm in diameter. Bleeding from the ulceration of the tumor was also observed (Fig. 3). All visible

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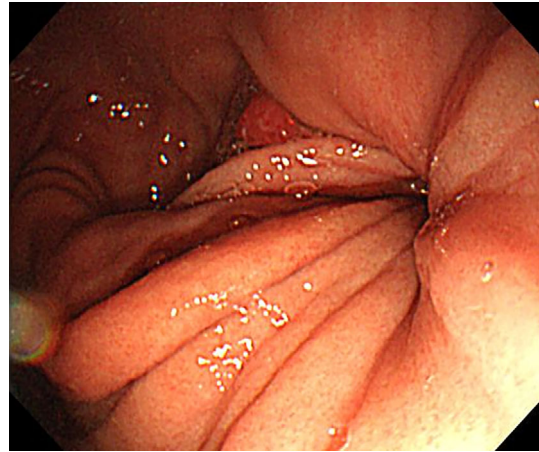
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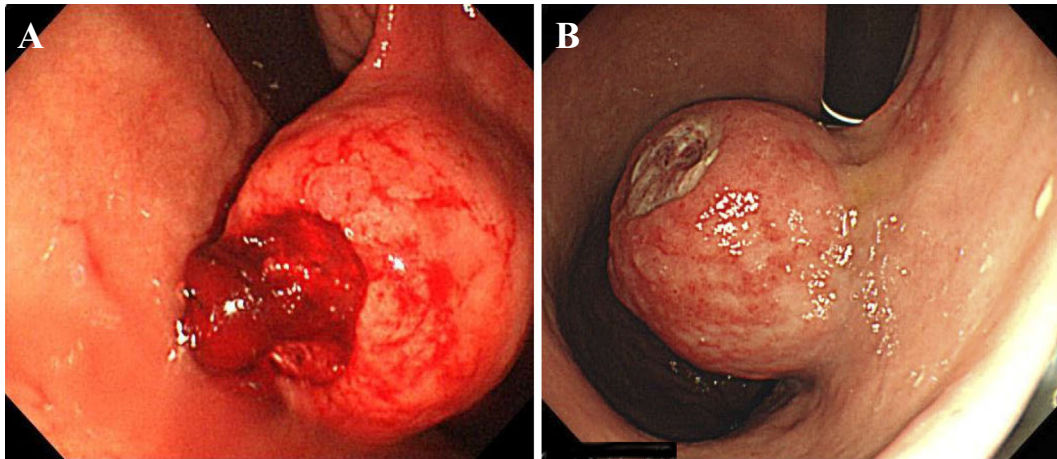
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**Figure 1.** Computed tomography scan at the level of the intussusception. The gastric tumor invaginated into the duodenum with extravasation of contrast medium.



**Figure 2.** Endoscopic image showing invagination of the gastric body into the duodenum. The distal stomach appeared folded.



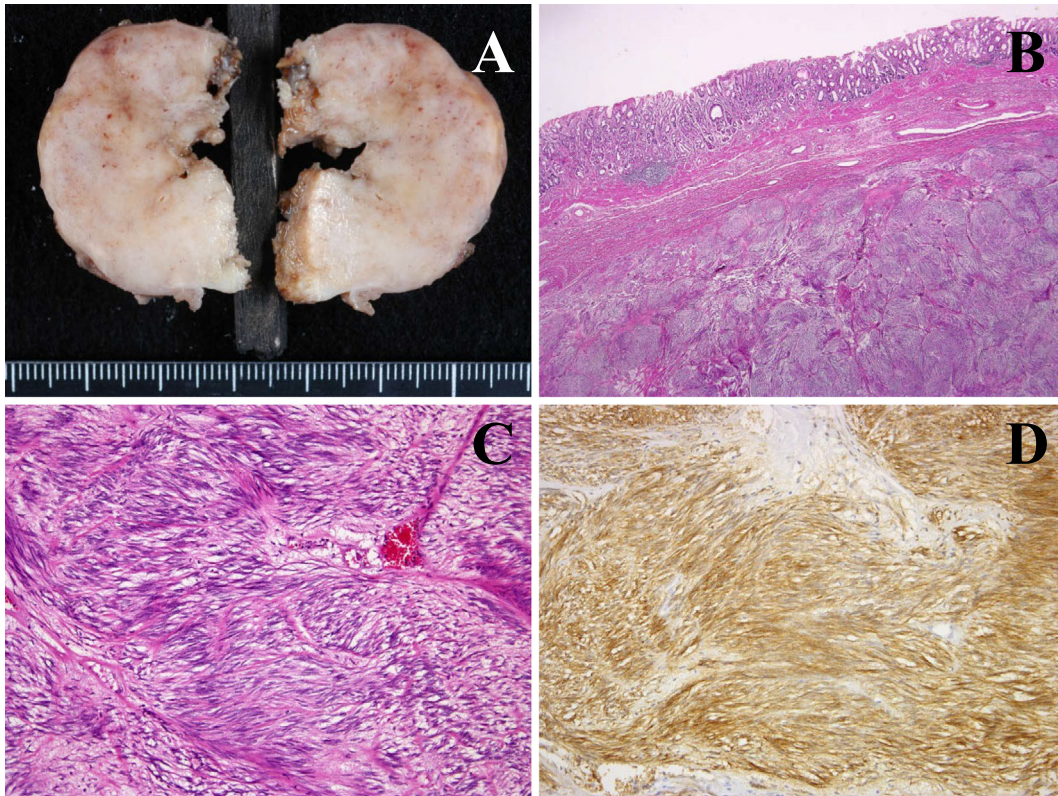
**Figure 3.** A: Endoscopic image showing a submucosal tumor with an ulcerated surface and active bleeding on the posterior wall of the gastric body. B: Retrograde distant view after endoscopic hemostasis.



**Figure 4.** Endoscopic ultrasound scan showing a hypoechoic lesion with hyperechoic foci limited to the third (submucosal) layer.

blood vessels were coagulated via argon plasma coagulation. Endoscopic ultrasonography (EUS) revealed a hypoechoic lesion with hyperechoic foci limited to the third (submucosal) layer (Fig. 4). The tumor size had increased slightly over the 10-year period, and the esophageal hiatal hernia had worsened. In addition to the EUS findings, narrowing of the shape in the base and easy mobility of the tumor suggested that the tumor was derived from the submucosal layer as opposed to the muscularis propria.

Several days after exvagination, the tumor again invaginated into the duodenum, resulting in bleeding. Open laparotomy and resection of the tumor were proposed; however, the patient and her family rejected surgery under general anesthesia. Endoscopic resection was therefore performed for local treatment. ESD was performed using a dual knife (Olympus, Tokyo, Japan). The tumor was completely removed en bloc within 160 minutes. The tumor was cut into segments after ESD in order to pass it through the esophagogastric junction and retrieve it endoscopically. Al-



**Figure 5.** Pathological analysis of the gastric mass confirmed a gastric gastrointestinal stromal tumor via positive immunohistochemical staining for c-kit and CD34. **A:** Macroscopic findings of the segmented tumor. **B:** Sheet formatted spindle cell spread in submucosal layer. **C:** The tumor showed the typical features of a spindle cell tumor with long, oval nuclei. **D:** C-kit protein immunostaining demonstrated strong cytoplasmic immunoreactivity.

though no major perforations were observed during ESD, chest radiography performed the following day showed free air in the abdomen. A minor perforation was suspected and conservatively treated with antibiotics and fasting.

Microscopically, the tumor consisted of sheets of spindle cells with long, oval nuclei. Immunohistochemically, the tumor cells were stained positive for both c-kit protein (CD 117) and CD34 and negative for  $\alpha$ -smooth muscle actin and S-100. The staining for Ki-67, a proliferation marker, was only positive in a few cells (MIB-1 labeling index, 0.06%). The tumor was histologically diagnosed as a low-grade GIST (Fig. 5). No tumor recurrence was detected, and the patient died of old age 55 months after endoscopic treatment.

## Discussion

Gastroduodenal intussusception, also known as gastroduodenal invagination or ball valve syndrome, is an extremely rare event (1). It typically occurs when a gastric tumor herniates into the duodenum, causing gastric obstruction. Although gastroduodenal intussusception accounts for less than 10% of all intussusception cases in adults (2), the exact prevalence remains uncertain. Zenda et al. previously speculated that greater mobility of the tumor, a higher tumor weight resulting in drooping of the tumor, increased gastric

peristalsis, and loosening of the pyloric orifice are all predisposing factors for gastroduodenal intussusception (3). In the present case, the gastric tumor size increased only slightly over a 10-year observation period, although the esophageal hiatal hernia was exacerbated. Therefore, although the exact mechanism is unknown, loosening of the surrounding tissue of the stomach may have been a major contributor towards gastroduodenal intussusception.

Gastroduodenal intussusception caused by gastric GIST is an exceedingly rare cause of gastric obstruction. To our knowledge, there have been 12 previously reported cases of gastroduodenal intussusception caused by gastric GIST (4-15). The characteristics of these 12 cases in addition to the case described here are presented in Table. These cases involved 9 women and 4 men, ranging from 29 to 95 years of age. The tumor sizes ranged from approximately 4 to 7 cm in diameter and were found to have originated in various locations, from the antrum to the fundus. Symptoms appeared from several weeks to several months before presentation at a hospital and were non-specific, including abdominal discomfort, nausea, vomiting, abdominal pain, and loss of appetite. The initial clinical presentations varied and depended on the degree of obstruction. Partial obstruction may cause chronic intermittent symptoms, while complete obstruction may cause abrupt symptoms. Indeed, Shum et al. reported a case of gastroduodenal intussusception that,

**Table. Review of Case Reports on Gastroduodenal Intussusception with GIST.**

No.	Reference	Age	Sex	Location	Size (cm)	Presentation	Treatment
1	15	59	F	Anterior wall of antrum	6	Intermittent epigastric pain and vomiting for 3 weeks	Partial gastrectomy
2	14	84	M	Antrum	4×3×3	Intermittent abdominal pain, nausea, weight loss, and vomiting for 6 weeks; with dark stools	Bilroth's II partial gastrectomy
3	13	34	F	Fundus	5×5, 3×2, 2×1	Intermittent epigastric pain	Partial gastrectomy
4	12	29	M	Antrum	6×6	Intermittent epigastric pain, nausea, and vomiting for 5 months; with dark stools	Bilroth's I partial gastrectomy
5	11	34	F	Posterior wall of fundus	6.5×4.4×3.8	Epigastric pain	Laparoscopic wedge resection
6	10	59	F	Anterior wall	7×6×5	Intermittent vomiting for 5 months	Partial gastrectomy
7	9	62	F	Posterior wall of distal body	5.2×3.5×3.2	Worsening epigastric pain and dark stools for 3 days	Bilroth's II partial gastrectomy
8	8	78	F	Antrum	4.5×3.3×3.4	Persistent epigastric discomfort and vomiting for 1 week	Laparoscopic wedge resection
9	7	52	F	Fundus	5×5	Worsening epigastric pain and vomiting for 1 day	Laparoscopic wedge resection
10	6	No data	M	Fundus	No data	Intermittent pain and vomiting for 1 year	No data
11	5	74	M	Posterior wall	No data	Intermittent vomiting for 3 weeks	Partial gastrectomy
12	4	85	F	Fundus	6×5	Abdominal discomfort, nausea, and weight loss for 6 months; acute, severe abdominal pain and vomiting	Subtotal gastrectomy
13	Present case	95	F	Posterior wall of distal body	4.2×3.9	Vomiting and loss of appetite for 1 week and dark stools	Endoscopic submucosal dissection

incidentally, was found on routine follow-up CT scan for GIST with no symptoms of gastric outlet obstruction.

Gastroduodenal intussusception caused by gastric GIST is essentially treated via surgical resection of the tumor. In the case presented here, although reduction of intussusception was endoscopically performed, the tumor invaginated again within several days. We performed endoscopic resection as opposed to surgical resection to remove the tumor, due to the patient's age (95 years). Several authors have described the usefulness of ESD for small and asymptomatic gastric submucosal tumors (16, 17). It was recently reported that endoscopic resection of subepithelial tumors of the stomach has a high success rate with few adverse events (18). In the present case, EUS suggested that the tumor was located within the submucosal layer without involvement of the muscular layer. Therefore, ESD was performed as an alternative to surgery. The tumor was resected en bloc despite its relatively large size. However, because the tumor was divided into segments in the stomach in order to retrieve it perorally, it was difficult to pathologically identify the origin of the GIST and whether it was derived from the muscularis mucosae or proper muscular layer. Furthermore, a minor perforation was suspected in the present case. Several stud-

ies have reported that perforation during ESD for gastric GIST occurs in 5% to 15% of cases (19), and no cases of seeding or plantation have been reported. Moreover, recent investigations have demonstrated that perforation during ESD for early gastric carcinoma does not lead to dissemination (20). Similarly, it is assumed that GISTs are at low risk for plantation.

The standard treatment for gastric GISTs is complete surgical resection (21) through conventional open surgery. Less invasive approaches, such as laparoscopic-endoscopic cooperative surgery (LECS) (22), non-exposed endoscopic wall-inversion surgery (NEWS) (23), and a combination of laparoscopic and endoscopic approaches to neoplasia with non-exposure technique (CLEAN-NET) (24), have been developed in recent years. However, these techniques and devices are only available at select institutions. In addition, these treatments are invasive for elderly patients, as general anesthesia and artificial ventilation are required. In contrast to treatments involving laparotomy or laparoscopy, endoscopic resection using the ESD technique is minimally invasive. Therefore, endoscopic resection is considered an option for the treatment of gastroduodenal intussusception caused by gastric GIST originating in the muscularis mucosae, par-

ticularly in patients of advanced age and/or with severe underlying disorders in whom general anesthesia and artificial ventilation are risky procedures. If surgical treatment is inadequate, conservative management should be considered before endoscopic treatment. The operative stress, risks of anesthesia, and patient's performance status should be included as evaluation criteria, as well as the age and underlying disease. The method of management must be discussed from a comprehensive perspective.

In summary, we present a case of a gastric GIST with gastrointestinal hemorrhaging and gastroduodenal intussusception that was treated with ESD. We believe that endoscopic resection is an acceptable alternate treatment for elderly patients who are not candidates for surgery.

**The authors state that they have no Conflict of Interest (COI).**

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