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
gastroduodenal intussusception; unfortunately, a gastric perforation was found after complete resection was accomplished, so gastorrhapsy 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

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 × 10⁹/L |
| **Hb**         | 9.1    | 12.3-15.3 g/dL   |
| **Plt**        | 302    | (160-370) × 10⁶/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.

Gastroduodenal intussusception caused by gastric GIST is the most common etiology and in bowel habits, constipation, or abdominal pain. 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 gastorrhaphy 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. 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             |
| Vincennes et al.[14] | 2005 | 72  | M   | Laparoscopy      | Gastric lipoma          | Exploratory laparotomy                      | NA                |
| Vincennes et al.[14] | 2006 |     |     |                  | Gastric lipoma          | NA                                          | NA                |
| Juglard et al.[15] | 2006 |     |     |                  | Ménétrier’s disease     | NA                                          | 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                                          | 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 et 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                         | NA                |
| Chan et al.[23] | 2009 | 34  | F   | CT               | GIST                    | Laparoscopic wedge resection                | 65 mm × 44 mm     |
| Eom et al.[24] | 2011 | 73  | F   | CT and EGD       | Gastric carcinoma       | Subtotal gastrectomy                        | 78 mm × 75 mm     |
| Eianorasetr 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     |
| MS et al.[35] | 2015 | 74  | M   | CT               | GIST                    | Partial gastrectomy                        | NA                |
| Indiran et al.[36] | 2015 |     |     |                  | GIST                    | NA                                          | NA                |
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
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, gastorrhaphy 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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