

# Gastroduodenal intussusception caused by gastric gastrointestinal stromal tumor in adults: a case report and literature review

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## Abstract

Intussusception mostly occurs in childhood and is rare in adults. Although intussusception can occur in any part of the gastrointestinal tract, gastroduodenal intussusception caused by a gastric tumor is relatively uncommon in clinical practice. A PubMed search identified 24 published cases of gastroduodenal intussusception caused by gastric gastrointestinal stromal tumor (GIST); however, it is possible that we missed other cases not included in PubMed. Here we report a case of gastroduodenal intussusception caused by gastric GIST in an 85-year-old man. He came to the hospital because of recurrent black stools. Plain computed tomography (CT) scan indicated a mass in the gastric antrum, with slight enhancement in the arterial phase on enhanced CT scan. He was diagnosed with GIST. In addition, images indicated that the mass overlapped into the duodenum, and gastroduodenal intussusception was thus considered. Gastroscopy showed a huge mass in the gastric body. According to the gastroscopy and CT results, gastroduodenal intussusception caused by a gastric tumor was considered. The patient underwent complete

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surgical removal, which revealed a mass originating from the gastric antrum and overlapping into the duodenum. The postoperative pathological diagnosis was intermediate-risk gastric GIST. The patient was followed up for 4 months without tumor recurrence.

### **Keywords**

Gastroduodenal intussusception, gastric tumor, gastrointestinal stromal tumor, adult, surgery, duodenum

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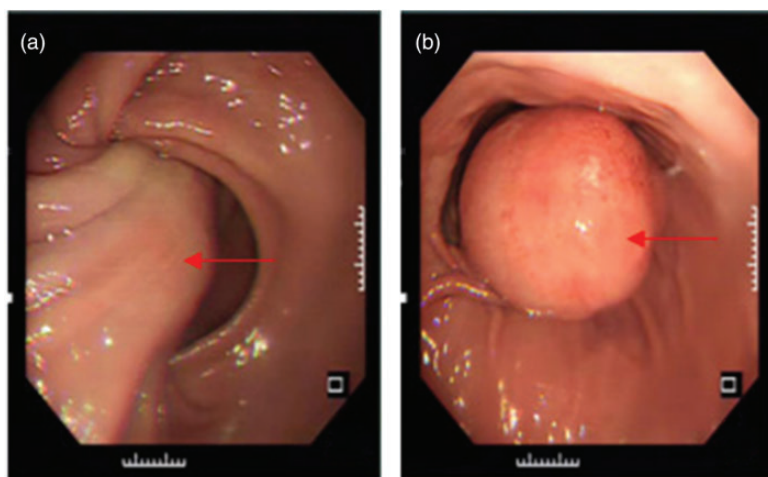
### **Introduction**

Gastroduodenal intussusception (also called gastroduodenal invagination or ball valve syndrome) is an extremely rare event and is the most infrequent form of intussusception, accounting for less than 10% of cases in adults.<sup>1,2</sup> Case reports of gastroduodenal intussusception caused by gastric tumors, such as gastric gastrointestinal stromal tumor (GIST),<sup>3–5</sup> gastric carcinoma,<sup>6</sup> gastric lipoma,<sup>7,8</sup> gastric schwannoma,<sup>9</sup> and gastric leiomyoma,<sup>10</sup> are uncommon. Hsieh et al.<sup>11</sup> reviewed the relevant literature regarding gastroduodenal intussusception within the past 20 years and found that gastric GIST was the most common etiology, accounting for more than half of these cases in adults. Here, we report a case of gastroduodenal intussusception caused by gastric GIST in an 85-year-old man. We review the clinical manifestations, diagnosis and differential diagnosis, treatment, and prognosis of gastroduodenal intussusception due to gastric GIST in the current case and in the literature.

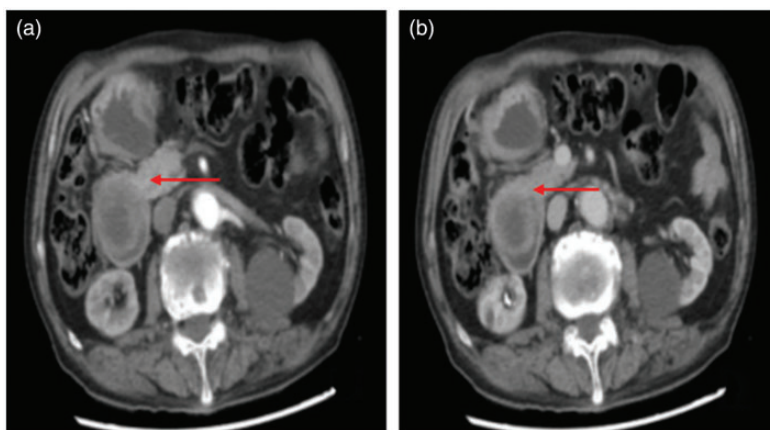
### **Case presentation**

An 85-year-old Chinese man came to the First People's Hospital of Taicang City on 23 July 2021 with a 1.5-year history of recurrent black stools. He had previously

been hospitalized in the Gastroenterology Department on 4 October 2019 because of his black stools and epigastric discomfort. Gastroscopy examination showed uplifting of the mucosa of the gastric body. The patient was diagnosed with gastroduodenal intussusception and underwent endoscopic reduction (Figure 1a, b). Pathological examination showed moderate chronic superficial gastritis of the gastric body. An abdominal plain computed tomography (CT) scan on 13 October 2019 indicated a 4.8-cm × 2.9-cm mass in the gastric antrum. We then received a consultation request and recommended that the patient should be transferred to our department for surgical treatment; however, the patient rejected this suggestion. He then attended Shaxi People's Hospital of Taicang City on 16 June 2021 due to further black stools. Gastroscopy revealed a GIST of the gastric body and superficial gastritis. The patient again presented with epigastric distention and pain and attended the surgical Outpatient Department of our hospital (First People's Hospital of Taicang City) on 18 July 2021. Review of a plain abdominal CT scan at that time showed that the mass in the gastric antrum had increased. The patient had a history of gout, hypertension, and inguinal hernia surgery over several years. Abdominal examination revealed no obvious positive signs. The laboratory results were normal, except for hemoglobin



**Figure 1.** Gastroscopic examination revealed uplifted mucosa (arrows) of the gastric body and overlap into the duodenum (a) and Endoscopic reduction was performed (arrows) (b).

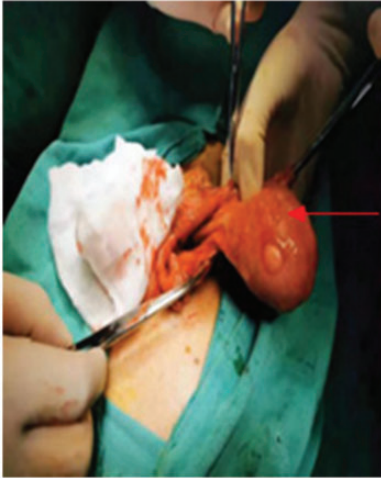


**Figure 2.** Enhanced computed tomography scan showed a mass (arrows) with slight enhancement in the arterial (a) and venous (b) phases on 27 July 2021.

77 g/L (normal range, 110–150 g/L). An enhanced CT scan on 27 July 2021 (Figure 2a, b) showed slight enhancement in the arterial and venous phases, and a diagnosis of GIST was considered. A review of the gastroscopy results showed a huge 5-cm  $\times$  3.5-cm mass in the gastric body. The patient underwent laparoscopic exploration and local resection of the tumor. During surgery, a 6-cm  $\times$  5-cm

mass was found originating from the gastric antrum (Figure 3) and overlapping into the duodenum. Gastroduodenal intussusception was diagnosed intraoperatively and the mass was completely resected after intussusception with surgical reduction. Postoperative histopathological examination revealed that the cells were epithelioid, the tumor was 5.5 cm in diameter, and mitotic figures showed two mitoses per

50 high-power fields. Immunohistochemical (IHC) staining showed CKP (–), CD117 (weak +), CD34 (+), DOG-1 (+), and SDHB (+) reactions (Figure 4a–f). The pathological diagnosis was intermediate-risk epithelioid GIST of the gastric



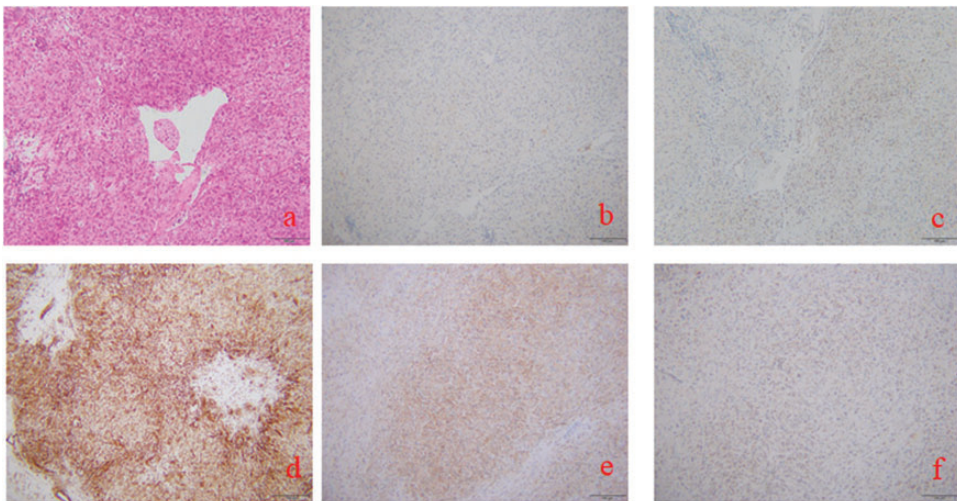
**Figure 3.** A mass measuring 6 cm × 5 cm (arrows) overlapped into the duodenum and was found to originate from the gastric antrum.

antrum. His final diagnosis was confirmed as gastroduodenal intussusception caused by epithelioid gastric GIST. The patient recovered well and was discharged 8 days after surgery. No recurrence or metastasis was observed after 4 months of follow-up.

This study was approved by the Ethics Committee of the First People's Hospital of Taicang City, China. All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

## Discussion

Gastroduodenal intussusception is a very rare complication of gastric tumors, caused by prolapse of the tumor and subsequent invagination of a portion or the full



**Figure 4.** Pathological examination. (a) Hematoxylin–eosin staining showed that the cells were epithelioid ( $\times 100$ ). Representative immunohistochemistry for (b) CKP, (c) CD117, (d) CD34, (e) DOG-1, and (f) SDHB ( $\times 100$ ).

thickness of the gastric wall into the duodenum.<sup>12,13</sup> We identified 43 reported cases of gastroduodenal intussusception caused by gastric tumors in adults in the literature (PubMed), including 24 cases of gastric GIST.<sup>3–5,11,14–33</sup> The clinical characteristics of these previous cases and the current case are presented in Table 1. There were 9 men and 15 women, ranging from 23 to 95 years of age. The tumors ranged from approximately 2.5 cm to 8.0 cm in diameter, and originated from different parts of the stomach, with most in the gastric antrum and gastric fundus. The symptoms of gastroduodenal intussusception caused by gastric GIST included upper abdominal discomfort, abdominal pain, nausea, vomiting, anorexia, appetite loss, postprandial fullness, hematemesis, black stools, dizziness, and palpitations. Most patients had varying degrees of obstruction, from partial obstruction causing chronic intermittent symptoms to complete obstruction, causing abrupt symptoms. Three cases of gastroduodenal intussusception caused by GIST induced acute pancreatitis.<sup>25,32,33</sup> In addition, an epigastric mass may be palpated by physical examination in individual patients. The current patient presented with epigastric pain and black stools, which were consistent with the above clinical symptoms of gastroduodenal intussusception. However, unlike other cases, this patient was hospitalized twice for gastrointestinal bleeding.

The preoperative diagnosis of gastroduodenal intussusception caused by gastric tumors can be difficult and may require different modalities of investigation, including CT and endoscopy.<sup>4</sup> Gastric tumors may be detected by upper gastrointestinal endoscopy as an endophytic mass protruding into the lumen, or even the full thickness of the gastric wall extending into the duodenum, and most patients can undergo endoscopic reduction. Unlike endoscopy, CT can provide details of the intussusception and the

adjacent organs and is critical for the preoperative assessment of patients.<sup>34</sup> The current patient was admitted to our hospital twice. At the first visit, gastroscopy showed a portion of the gastric wall protruding into the duodenum and mucosal uplift of the gastric body was observed after endoscopic reduction. Meanwhile, CT scan showed a mass in the gastric antrum and slight enhancement in the arterial phase during contrast-enhanced scanning. At the second visit, gastroscopy showed that the mass in the gastric body had increased, and CT images showed that the mass overlapped into the duodenum.

Although the radiologic and gastroscopic findings in the current patient were consistent with previous reports of gastroduodenal intussusception, the final diagnosis of GIST-related gastroduodenal intussusception could not be confirmed until after surgery and postoperative pathological examination.<sup>35</sup> In addition, although CT, endoscopy, and surgery can diagnose gastroduodenal intussusception, they cannot diagnose the specific type of gastric tumor. In this case, surgery revealed that the mass originated from the gastric antrum and overlapped into the duodenum, and the postoperative pathological diagnosis was gastric GIST. A diagnosis of GIST can only be confirmed by histological and IHC methods. Histologically, strong expression of c-Kit protein (CD117, a type III tyrosine kinase receptor encoded by the c-KIT proto-oncogene) suggests that GISTs originate from interstitial cells of Cajal or their precursors. Immunologically, GISTs are defined by positive immunostaining for CD117 (overexpressed in 95%) and CD34 (positive in 60%–70%),<sup>36</sup> and identification of CD117 expression by IHC staining is thus enough to make a clinical diagnosis of GIST. However, approximately 40% of tumors with *PDGFRA* mutations are weakly



**Table 1.** Twenty-five reported cases (including this case) of gastroduodenal intussusception caused by gastric gastrointestinal tumor:

Case no.	Ref.	Age/sex	Year	Symptoms	Location	Tumor size (cm)	Diagnostic method	Treatment
1	Crowther et al. <sup>3</sup>	2002 59/F	2002	Indigestion, epigastric pain, vomiting	Anterior wall of antrum	6.0	Upper gastrointestinal endoscopy and CT	Surgical resection
2	Adjepong et al. <sup>4</sup>	2006 84/M	2006	Abdominal pain, nausea and vomiting	Gastric antrum	4.0	Upper gastrointestinal endoscopy and CT	Laparoscopic Billroth II distal gastrectomy
3	Samamé et al. <sup>5</sup>	2007 69/–	2007	Epigastralgia, hematemesis and vomiting	Gastric antrum	–	CT	Exploratory laparotomy
4	Hsieh et al. <sup>11</sup>	2021 84/F	2021	Postprandial fullness, nausea and vomiting	Gastric	5.6	CT	Endoscopic resection
5	Yamauchi et al. <sup>14</sup>	2017 95/F	2017	Vomiting and melena	Lower gastric body	4.0	CT and endoscopy	Endoscopic submucosal dissection
6	Shum et al. <sup>15</sup>	2007 34/F	2007	Epigastric pain	Gastric fundus	5.0	CT and upper endoscopy	Partial gastrectomy
7	Siam et al. <sup>16</sup>	2008 29/M	2008	Abdominal pain, nausea, vomiting and anemia	Gastric antrum	6.0	Capsule endoscopy and enteroscopy	Billroth I partial gastrectomy
8	Chan et al. <sup>17</sup>	2009 34/F	2009	Gastrointestinal bleeding and epigastric pain	Posterior wall of gastric fundus	6.5	Upper endoscopy and CT	Laparoscopic wedge resection
9	Gyedu et al. <sup>18</sup>	2011 59/F	2011	Vomiting	Anterior wall of the stomach	7.0	Abdominal ultrasound	Wedge resection
10	Seok et al. <sup>19</sup>	2012 51/M	2012	Nausea, vomiting, melena and severe anemia	Gastric	5.5	Upper endoscopy and CT	Surgical resection
11	Wilson et al. <sup>20</sup>	2012 78/F	2012	Upper abdominal discomfort, vomiting and anorexia	Distal body and antrum of the stomach	4.5	Gastroscopy and CT	Laparoscopic wedge resection of the stomach
12	Basir et al. <sup>21</sup>	2012 62/F	2012	Epigastric pain, anorexia, and black stools	Posterior wall of the distal body of the stomach	5.2	Upper gastrointestinal endoscopy and CT	Billroth II partial gastrectomy
13	Rittenhouse et al. <sup>22</sup>	2013 52/F	2013	Epigastric abdominal pain, and vomiting	Fundus of the stomach	5.0	CT	Laparoscopic exploration and resection of the tumor
14	Babannavar et al. <sup>23</sup>	2015 74/M	2015	Vomiting	Posterior wall of the stomach	–	CT and upper gastrointestinal endoscopy	Laparotomy and resection of the tumor
15	Indiran et al. <sup>24</sup>	2015 –/M	2015	Intermittent pain and vomiting	Gastric	–	Ultrasound, CT, barium meal and endoscopy	–
16	Yildiz et al. <sup>25</sup>	2016 85/F	2016	Epigastric discomfort, nausea, and weight loss	Upper gastric body	6.0	CT	Subtotal gastrectomy and Roux en Y anastomosis

(continued)

Table 1. Continued.

Case no.	Ref.	Year	Age/ sex	Symptoms	Location	Tumor size (cm)	Diagnostic method	Treatment
17	Komatsubara et al. <sup>26</sup>	2016	90/F	Appetite loss and vomiting	Gastric fundus	5.0	Esophagogastroduodenoscopy and CT	Wedge resection
18	Jameel et al. <sup>27</sup>	2017	65/F	Upper abdominal pain and vomiting	Posterior wall of stomach	6.0	Endoscopy and CT	Laparoscopic resection
19	Zhou et al. <sup>28</sup>	2018	69/M	Abdominal pain, nausea, and vomiting	The posterior wall of the gastric antrum	4.5	Endoscopy, upper gastroenterography, and CT	Laparoscopic exploration and wedge resection
20	Ssentongo et al. <sup>29</sup>	2018	85/F	Epigastric pain, dyspepsia, dizziness, and palpitations	Gastric fundus	2.5	CT	Laparotomy and wedge resection
21	De et al. <sup>30</sup>	2018	42/F	Upper abdominal pain and vomiting	Anterior wall	8.0	Endoscopy and CT	Laparotomy and surgical resection
22	Đokić et al. <sup>31</sup>	2019	62/M	Epigastric pain and black stools	Lesser curvature of the gastric body	7.5	CT	Explorative laparotomy and circular radical resection
23	Michael et al. <sup>32</sup>	2021	23/F	Epigastric pain, intermittent vomiting, and loss of appetite	Gastric antrum	7	Esophagogastroduodenoscopy and abdominal CT	Wedge resection
24	Numphrphut et al. <sup>33</sup>	2021	55/M	Epigastric pain and vomiting	Gastric fundus	5.5	CT and esophagogastroduodenoscopy	Laparo-endoscopic intragastric wedge resection
25	Current case	2021	85/M	Black stools and epigastric pain	Gastric antrum	5.5	CT and endoscopy	Laparoscopic exploration and local resection of the tumor.

M, male; F, female; CT, computed tomography scan.

positive or negative for CD117.<sup>37,38</sup> The positive-expression rate of DOG1 in GIST is up to 94.8%, with a high sensitivity of 89% and a higher positivity rate in epithelioid GIST than CD117.<sup>37,39,40</sup> In the current case, IHC revealed CD117 (weak +), CD34 (+), and DOG-1 (+) expression, and routine hematoxylin–eosin staining revealed that the cells were epithelioid, thus confirming the final diagnosis of epithelioid GIST. Based on the above evidence, the patient was eventually diagnosed with gastroduodenal intussusception caused by epithelioid gastric GIST.

The treatment of gastroduodenal intussusception caused by gastric GIST mainly includes surgical resection and endoscopic treatment. Twenty-two previous cases<sup>3–5,15–33</sup> were treated by surgical resection and only two cases<sup>11,14</sup> were treated by endoscopic submucosal dissection (ESD). Notably, 12 cases underwent laparoscopic surgery, but no studies have reported the use of a robotic approach. Surgery is thus the main treatment option for most cases of gastroduodenal intussusception caused by gastric GIST, with different surgical methods depending on the size and location of the tumor. Endoscopic treatment includes endoscopic reduction and endoscopic resection. Faulx et al.<sup>41</sup> suggested that endoscopic full-thickness resection, submucosal tunnel endoscopic resection, and full-thickness ESD were viable options for GISTs of 2 to 4 cm. Among the previous 24 case reports, only one patients underwent combined laparoscopic and endoscopic intragastric wedge resection given the lesion size and location.<sup>33</sup> In the current case, the patient initially underwent endoscopic reduction but refused further surgical resection. However, he was admitted to the hospital for further surgery 1.5 years later because of the occurrence of more severe symptoms. In summary, laparoscopic resection is the main treatment for this disease. If necessary, intraoperative

gastroscopy can be used to locate the lesion to ensure the smooth completion of laparoscopic surgery. Endoscopic reduction may be an alternative option for patients who are unsuitable for resection, such as elderly patients who are not candidates for surgery or general anesthesia.<sup>11,14</sup>

## Conclusion

Gastroduodenal intussusception caused by gastric GIST usually manifests with upper abdominal discomfort, abdominal pain, and nausea, with acute pancreatitis in a few cases. Endoscopy and abdominal CT examination have important clinical value for the diagnosis of gastroduodenal intussusception caused by gastric GIST, and tumor reduction can be carried out endoscopically. The final diagnosis depends on postoperative pathological examinations, and regular postoperative follow-up is mandatory.

## Availability of data and materials

All data and materials are presented in this manuscript and the datasets are available from the corresponding author on reasonable request.

## Declaration of conflicting interest


The authors declare that they have no competing interests in regard to this research paper.

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