



Case Report

Unusual gastroduodenal intussusception secondary to a gastrointestinal stromal tumor of the gastric fundus

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ARTICLE INFO

Article history:

Received 11 October 2016

Accepted 12 October 2016

Available online 17 October 2016

Keywords:

Gastroduodenal intussusception

Gastrointestinal stromal tumor

Gastric fundus

Surgery

ABSTRACT

Introduction: Intussusception is uncommon in the proximal gastrointestinal tract. Although there are a few case reports of gastroduodenal intussusception, it is extremely rare that a tumor located in the gastric fundus causes gastroduodenal intussusception. We report a patient with gastroduodenal intussusception secondary to a gastrointestinal stromal tumor (GIST) in the gastric fundus.

Presentation of case: A 90-year-old woman was admitted with appetite loss and recurrent vomiting. Esophagogastroduodenoscopy showed cord-like mucosa of the gastric fundus pulled into the duodenal bulb. Abdominal computed tomography scan showed a well demarcated round tumor in the duodenum. The patient underwent a laparotomy, since endoscopic reduction of the tumor was not successful. The tumor was reduced by gently reducing the mass. A small incision was made in the anterior wall of the stomach, and the tumor was resected. The pathological findings of the tumor showed GIST. The post-operative course was uneventful.

Discussion: There were only eight patients in the world literature, including the present case, reported with gastroduodenal intussusception secondary to a GIST in the gastric fundus. All patients presented with ball valve syndrome, known as a cause of acute abdomen. Three tumors were reducible preoperatively. Five of the eight tumors were resected with an intra-gastric open surgical approach.

Conclusion: We report a patient with gastroduodenal intussusception caused by a rare GIST in the gastric fundus, which needed resection.

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1. Introduction

Gastroduodenal intussusception is rare and reported to be the least common type of intussusception in the gastrointestinal tract [1]. Gastroduodenal intussusception most commonly occurs due to an anatomical abnormality or at the site of a gastrojejunostomy in the English literature [2–5]. The ball valve syndrome, first described in 1946 by Hobbs and Cohen [6], was recognized as a rare but important cause of an acute abdomen caused by prolapse of gastric tumors through the pylorus into the duodenal bulb. There are few reports of tumors in the gastric fundus, located far from the pylorus ring, causing ball valve syndrome. We report a patient with gastroduodenal intussusception secondary to a gastrointestinal stromal tumor (GIST) in the gastric fundus, which presented with ball valve syndrome.

2. Presentation of case

A 90-year-old woman was referred with appetite loss and vomiting. Her medical history was not significant. Physical examination was negative for masses, hernias, tenderness or peritoneal signs. Laboratory data showed hypokalemia (serum potassium 2.6 mEq/L), but otherwise there were no remarkable findings. Esophagogastroduodenoscopy showed cord like mucosa extending from the gastric fundus into the duodenum (Fig. 1). Although a small diameter endoscope could pass through the pylorus, endoscopic reduction of the tumor which was the lead point of the gastroduodenal intussusception was not successful. Computed tomography scan of the abdomen showed a well demarcated round tumor with a central dimple in the duodenum (Fig. 2). Based on the gross appearance and radiological findings, the tumor was most likely a gastrointestinal stromal tumor (GIST). Laparotomy was then performed. After confirming the presence of the mass in the duodenum, reduction of the tumor was done by gently reducing the mass back along the duodenum. This required a great deal of force, which may explain why endoscopic reduction was unsuccessful. A small incision was made in the

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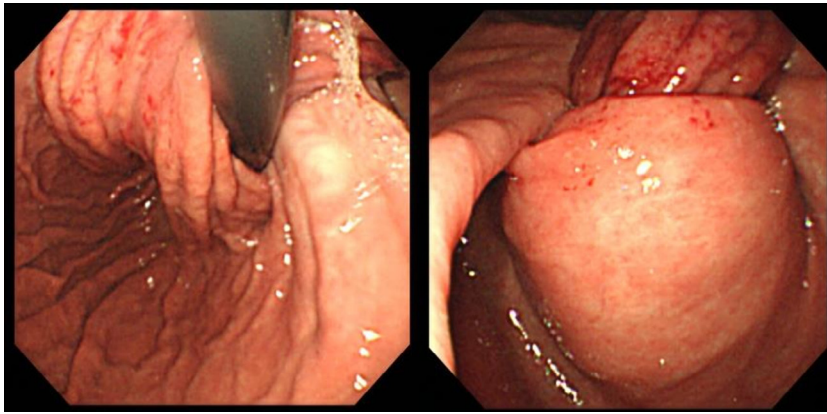


Fig. 1. Esophagogastroduodenoscopy shows cord-like mucosa from the gastric fundus pulled into the duodenal bulb.

anterior wall of the stomach, and the tumor was resected with a stapler from the mucosal side (Fig. 3). The postoperative course was uneventful and the patient was able to resume oral intake. Histologically, the resected tumor was composed of spindle-

shape cells. The resected tumor was 50 × 45 × 40 mm in size, and the pathological diagnosis was a GIST, which was positive for c-kit, CD34, and had a 5–10% MIB-1 index. By contrast, α -smooth muscle actin, S100, and cytokeratin AE1/AE3 were negative (Fig. 4).

3. Discussion

In gastroduodenal intussusception, a pedunculated benign gastric tumor is usually known to become the lead point [7]. Polyps constitute 40% and intramural smooth muscle tumors 40% [1]. In Japan, 143 cases of gastroduodenal intussusception caused by gastric tumors were reviewed [8]. About 67% of the tumors were epithelial tumors, such as an early gastric cancer and benign polyp. Intramural tumors, which were less common than epithelial tumors, comprised 25% of tumors. The tumors included 97 in the gastric antrum (68%), 36 in the gastric body (25%), and 10 in the gastric fundus (7%). While gastroduodenal intussusception is a rare clinical entity, tumors in the gastric fundus are least commonly associated with intussusception.

A review of 16 patients with gastroduodenal intussusception caused by tumors in the gastric fundus reported that all tumors were located in the submucosa, and the average maximum diameter was 5.4 cm [9]. These patients were often older females with a small physique. All patients presented with ball valve syndrome. Other features in common included larger tumors than those in the gastric antrum which cause gastroduodenal intussusception, the tumors were mobile since submucosal tumors usually do not infiltrate surrounding tissue, and the supporting tissue of the gastric wall was fragile because of aging. These features meet the requirements to develop ball valve syndrome. The authors suggest it is more often seen in women because women survive longer than men.

There were eight patients, including the present patient, reported with ball valve syndrome secondary to a GIST in the gastric fundus (Table 1) [9–15]. Seven patients were Japanese. The average age was 77 years (44–93), and 7/8 are women. The average maximum diameter of the tumors is 57 mm (32–87). Endoscopic reduction succeeded preoperatively in three tumors. Three tumors were resected laparoscopically, and the remaining five were resected at open surgery.

GIST is a non-epithelial, mesenchymal tumor first described in 1983, and the majority are gastric in origin [16,17]. The presentation of a GIST is usually nonspecific and depends on the size and location of the tumor. Small GISTs, 2 cm or less, are usually incidentally found during workup for other unrelated conditions because they are often asymptomatic. A gastric GIST causing duodenal obstruction is not common [18]. As described above, tumors located in the gastric fundus need to be submucosal and large to cause gas-



Fig. 2. Computed tomography scan showed a well demarcated tumor in the duodenum (arrow). The duodenum was pulled by the tumor and shifted to the left side.

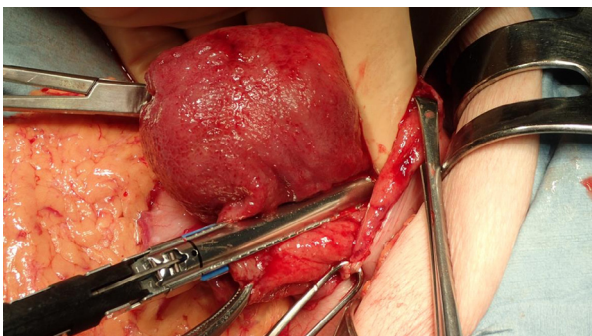


Fig. 3. The tumor was resected with a linear stapler, from the mucosal side through the gastrostomy.

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