Internal Medicine
The Japanese Society of Internal Medicine
doi: 10.2169/internalmedicine.1916-18
Intern Med 58: 1267-1271, 2019
http://internmed.jp

【 CASE REPORT 】

The Development of Gastric Outlet Obstruction due to a Lumen-occupying Protruding Duodenal Ulcer Mimicking a Submucosal Tumor

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Abstract:
Even in the era of *Helicobacter pylori* eradication and proton pump inhibitors, peptic ulcer remains an important disease. Stricture due to a duodenal ulcer in the healing stage is a well-known etiology of benign gastric outlet obstruction. However, a duodenal ulcer-induced submucosal tumor-like change with gastric outlet obstruction is a very rare manifestation. We herein present a rare case of a patient with deteriorating symptoms of gastric outlet obstruction caused by an unusual manifestation of a lumen-occupying protruding duodenal ulcer mimicking a submucosal tumor.

Key words: gastric outlet obstruction, peptic ulcer, duodenal ulcer, submucosal tumor

(Intern Med 58: 1267-1271, 2019)
(DOI: 10.2169/internalmedicine.1916-18)

Introduction

Gastric outlet obstruction (GOO) has both benign and malignant etiologies. Since the emergence of eradication therapy for *Helicobacter pylori* and proton pump inhibitors for the treatment of peptic ulcer, malignant disease has become the leading etiology of GOO (1). Nevertheless, GOO caused by numerous benign etiologies remains important, and peptic ulcer is the leading cause of benign GOO (2). The diagnostic criteria for duodenal ulcer-related GOO are gastric retention and severe stenosis of the duodenal bulb on esophagogastroduodenoscopy (EGD), because an endoscope cannot be advanced into the duodenal bulb due to severe stricture (3). Although peptic ulcers typically appear as mucosal defects with various degrees of excavation and/or mucosal scarring, there are few reports of gastric ulcers with features that mimic a submucosal tumor (SMT) (4-6). We herein report a case of GOO that developed due to a lumen-occupying protruding duodenal ulcer mimicking a submucosal tumor.

Case Report

A 70-year-old man was referred to our department with a one-year history of a deteriorating abdominal fullness after meal ingestion with no significant medical history of peptic ulcer or malignancy. The patient had no history of using non-steroidal anti-inflammatory agents or other ulcer inducing drugs. The patient smoked approximately 40 cigarettes per day. The laboratory findings on admission were as follows: white blood cell count, $5.7\times10^3/\mu L$; hemoglobin 13.1 g/dL; C-reactive protein, 0.1 mg/dL; albumin, 4.22 g/dL; serum carcinoembryonic antigen, 1.7 ng/mL (upper limit of normal, ULN: 5.0 ng/mL), and serum carbohydrate antigen 19-9, 2.0 U/mL (ULN: 37.0 U/mL). The serum *Helicobacter pylori (H. pylori)* antibody titer was 3.0 U/mL (ULN: 10.0 U/mL). Contrast enhanced computed tomography demonstrated an extended stomach with food residue and an enhanced lesion that protruded into the duodenal bulb (Fig. 1). EGD revealed an expansive space-occupying lesion in the duodenal bulb (Fig. 2). The lesion included a reddish mu-

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Received: August 3, 2018; Accepted: September 11, 2018; Advance Publication by J-STAGE: January 10, 2019
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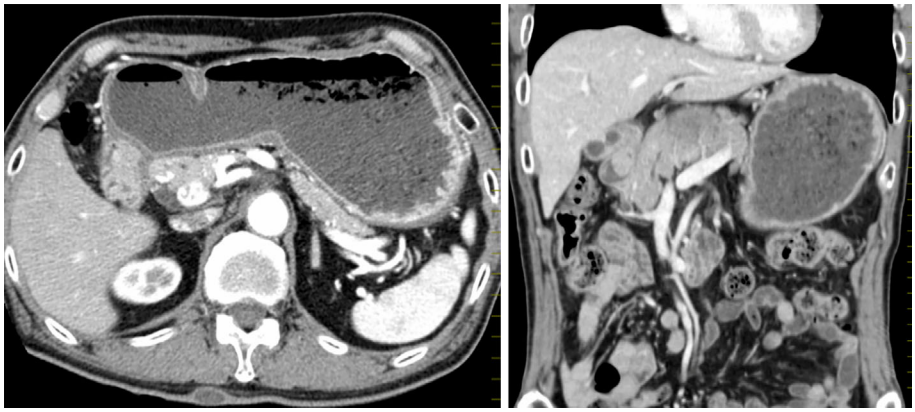

Figure 1. **Contrast enhanced computed tomography (CE-CT). An image of a CE-CT demonstrating an enhanced lesion protruding into the lumen of a duodenal bulb. The stomach is extended with food residue.**

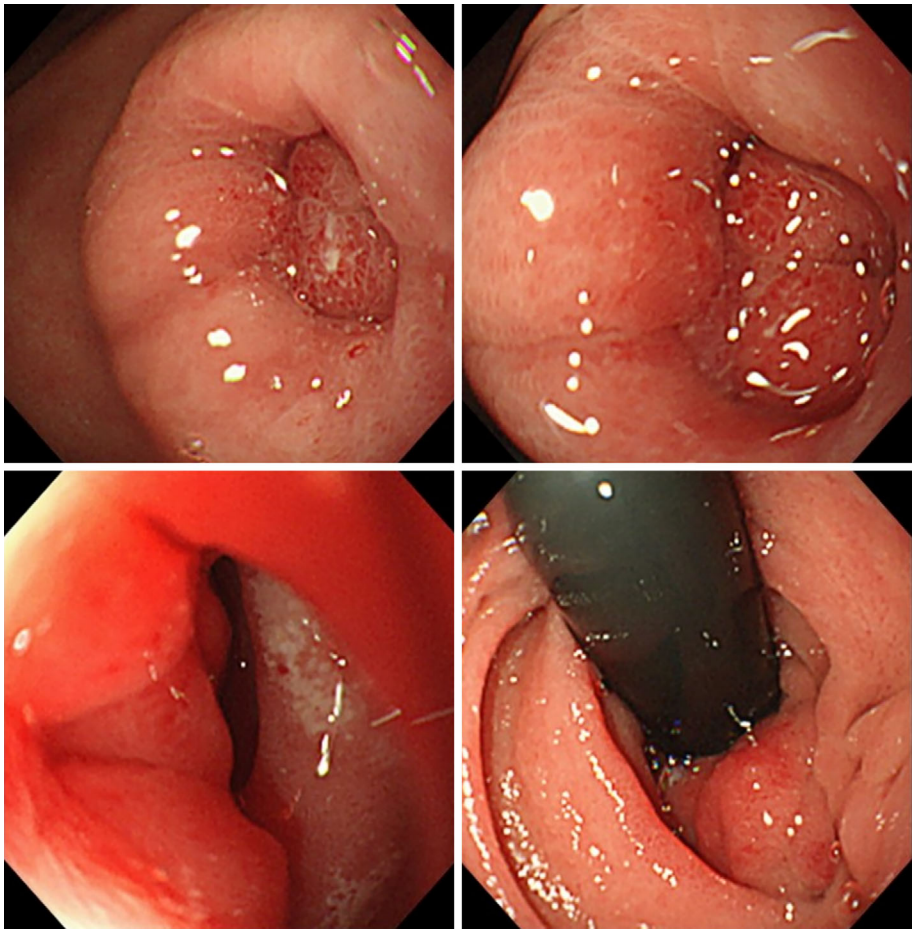

Figure 2. **Endoscopic views of the duodenal lesion. Esophagogastroduodenoscopy (EGD) demonstrating a space-occupying lesion at the duodenal bulb. A pyloric ring was expanded and obstructed with this lesion. EGD in the duodenal J-turn position demonstrating an opposite hemisphere of the lesion mimicking a submucosal tumor. The lesion was capped with a reddish normal mucosa with a central depression.**

cosa with indentation. Double-contrast fluoroscopy under EGD using a J-turn position in the duodenal bulb revealed a filling defect that was acting like a ball valve. A forceps biopsy from the duodenal mucosa showed no evidence of malignancy and a non-specific pathological diagnosis. Positron emission tomography (PET) demonstrated an intensive abnormal uptake [maximum standardized uptake value (SUV) 6.1] at the duodenal bulb (Fig. 3), which was suggestive of malignancy. Next, we conducted an endoscopic ultrasonography-guided fine-needle biopsy (EUS-FNB) us-

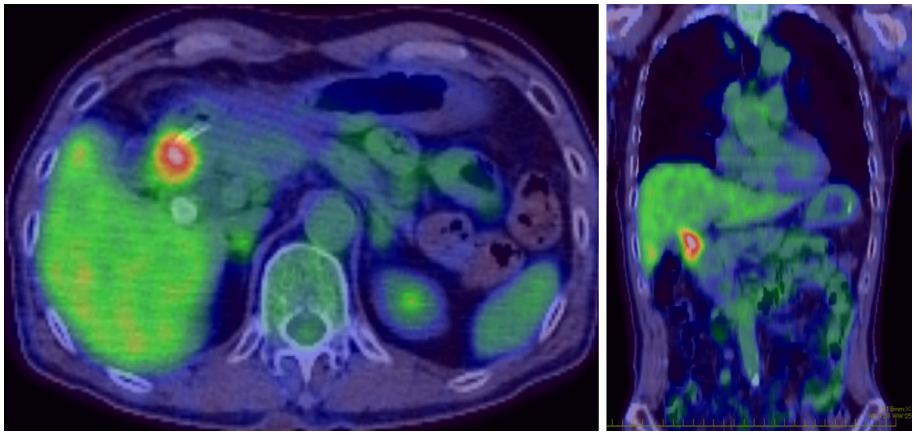

Figure 3. **Positron emission tomography (PET). PET demonstrating an intensive abnormal uptake at the duodenal bulb. A maximum standardized uptake value of 6.1 is suggestive of malignancy.**

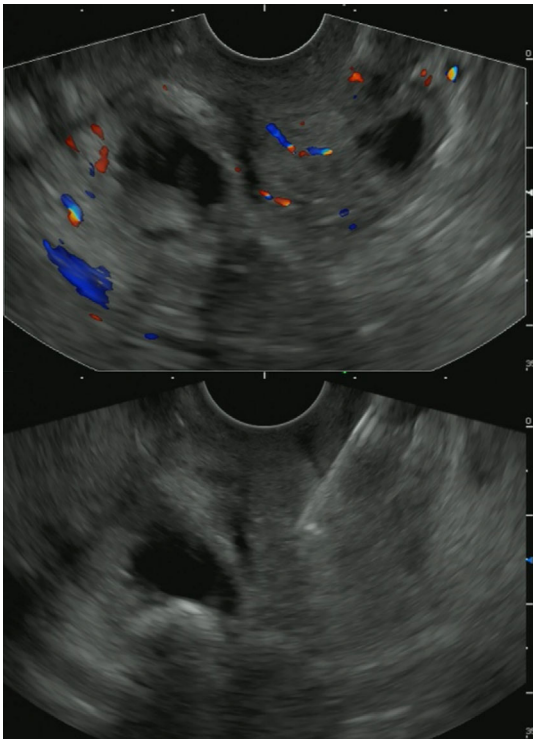

Figure 4. **Endoscopic ultrasonography (EUS) and EUS-guided fine needle biopsy (EUS-FNB) images. (Top) An EUS image demonstrating a slight echogeneous mass of 11 mm in diameter with a vasculature signal. The lesion was contiguous to the duodenal submucosal layer. The muscular layer at the base of the mass was thickened. (Bottom) An EUS image obtained during EUS-FNB with a 22-gauge Franseen needle.**

ing a 22-gauge Franseen needle with suction under rapid on-site evaluation. The EUS image revealed a homogeneous echoic mass of 11 mm in diameter with a vasculature signal that was contiguous to the duodenal submucosal layer. The muscular layer at the base of the mass was thickened (Fig. 4); however, the specimen procured during EUS-FNB only showed the intestinal mucosa.

The patient was scheduled to undergo surgery with curative intent for the following reasons: 1) the GOO status was deteriorating, 2) the maximum SUV in the PET was 6.1, and 3) there possibility of a false-negative cytopathological EUS-FNB result was a concern. Laparotomy revealed severe adhesion between the gallbladder and the hepatoduodenal ligament. An intraoperative frozen section analysis revealed no malignant tissue. Thus, in addition to a distal gastrectomy, cholecystectomy, distal bile duct resection, and Roux-en-Y reconstruction with hepaticojejunostomy was performed. One of the resected specimens showed a duodenal ulcer associated with marked tissue granulation and severe chronic active inflammatory infiltration in the submucosal and muscular layers. There was no evidence of malignancy in the other resected organs (Fig. 5). The patient's postoperative course was uneventful and there was no evidence of ulcer recurrence.

Discussion

We described a rare case of deteriorating GOO caused by a benign duodenal ulcer mimicking a submucosal tumor. Peptic ulcers are the leading cause of benign GOO (2). Severe stenosis of the duodenal bulb revealed by EGD is one of the diagnostic criteria for duodenal ulcer-related GOO (3). In the present case, the deteriorating GOO was caused by a lumen-occupying protruding duodenal ulcer that was behaving like a ball valve. We proceeded with a differential diagnosis according to the characteristics observed in the EUS image (7). The duodenal mass demonstrated echogenicity and was contiguous to the duodenal submucosal layer (third layer). The muscular layer (fourth layer) at the base of the mass was thickened; thus, the primary differential diagnosis was ectopic pancreas (7, 8). The first EGD biopsy specimens did not provide a specific pathological diagnosis. We therefore performed EUS-FNB to obtain submucosal tissue for a cytopathological diagnosis. In a previous article, EUS-guided tissue acquisition provided a diagnostic accuracy of 92.7% for endoscopic biopsy-negative gastrointestinal tumors (9). However, the sensitivity was 87.3% and

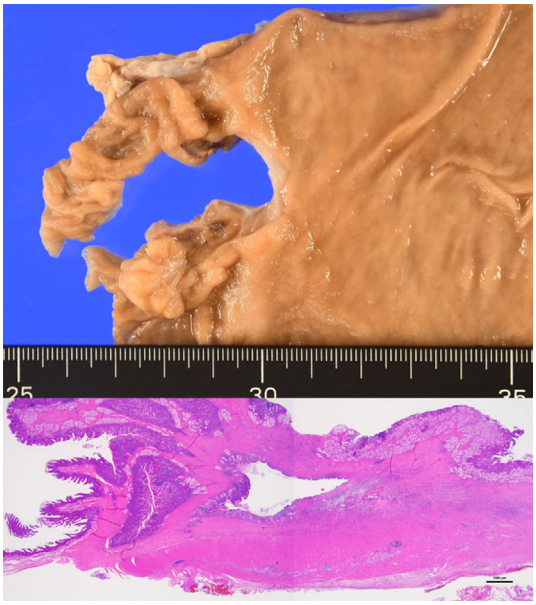

Figure 5. The macroscopic and microscopic findings of the resected stomach and duodenal bulb. (Top) A surgical specimen demonstrating the protruded lesion mimicking a submucosal tumor with an ulcer at the duodenal bulb. A defect site was submitted for intraoperative frozen section analysis. (Bottom) The pathological specimen showing the ulcer with severe chronic inflammatory infiltration predominantly in the bulky submucosal layer. The mucosal defect was consistent with an ulcer. There was no evidence of malignancy (Hematoxylin and Eosin staining, scale bar: 1,000 μm).

the negative predictive value was 85.2%, which suggested the possibility of false-negative results (9). Regarding the PET-CT finding of an intense abnormal uptake in the duodenal bulb, this lesion was suggestive of the malignant transformation of an ectopic pancreas (8). This suspected malignant transformation originated from the knowledge that a cut-off SUV (max) of 3.5 had high sensitivity (92.6%) and specificity (76.9%) in the diagnosis of malignancy in patients with solitary pancreatic lesions (10). The postoperative pathological conclusion in the present case was GOO mimicking SMT caused by a benign duodenal ulcer with severe chronic active inflammatory infiltration predominantly in the submucosal layer. Although a bite-on-bite jumbo biopsy might provide additional information for making a clinical decision, the result was definitive in only 65.1% and 40.0% of specimens from third and fourth layer lesions, respectively-even with a jumbo biopsy, Furthermore, the risk of bleeding was significant, with an incidence rate of 34.9% (11).

Benign gastric ulcers occasionally develop SMT-like features when marked submucosal fibrosis during the healing process leads to the elevation and protrusion of the ulcer bed (4-6). However, the present case is unique because the GOO developed from a lumen-occupying protrusion of a benign duodenal ulcer with SMT-like features. This is the first report of duodenal ulcer-induced SMT and GOO. Unlike the submucosal protrusion of a gastric ulcer (4-6), the submucosal protrusion of a duodenal ulcer easily occupies the duodenal lumen resulting in GOO (as described above). In rare cases, the manifestation results in intussusception (12).

With the exception of heavy cigarette consumption, the present patient had no risk factors for duodenal ulcer. In recent articles, a smoking habit, the intake of aspirin, and *H. pylori* infection were identified as independent risk factors for duodenal ulcer formation (odds Ratio: 2.84, 4.28, and 3.56, respectively) (13). Furthermore, cigarette smoking aggravates ulcer formation and delays ulcer healing in the gastrointestinal mucosa (14). We deduced that severe protracted inflammation in the submucosal layer due to smoking might eventually lead to the development of submucosal protrusion that could mimic SMT in the duodenal bulb. We also considered that severe adhesion around the duodenal bulb might develop as a result of micro-perforation of the ulcer. Severe inflammation could explain the intensive abnormal uptake at the duodenal bulb on PET.

In conclusion, severe stricture caused by a duodenal ulcer is a well-known cause of GOO. However, we should keep in mind that duodenal ulcers rarely have unusual manifestations of GOO caused by a lumen-occupying protrusion that mimics a submucosal tumor.

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

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