A giant Brunner gland hamartoma successfully treated by endoscopic excision followed by transanal retrieval

A case report

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Abstract

Rationale: Brunner gland hamartoma (BGH) is a rare tumor of the duodenum. Although BGH is a benign tumor, larger lesion with gastrointestinal symptoms requires tumor removal. We report a giant BGH, successfully treated by endoscopic excision followed by transanal retrieval.

Patient concerns: A 38-year-old woman complained of severe anemia, tarry stool, and vomiting.

Diagnoses: Esophagogastroduodenoscopy (EGD) showed a pedunculated giant submucosal mass at the duodenal bulb.

Interventions: We attempted to remove it because the lesion seemed to be responsible for patient’s anemia and vomiting. The lesion had clear but bulky stalk. We carefully cut the stalk using needle-knife and IT knife\textsuperscript{2}. We tried to retrieve specimen, but the mass could not pass through the pyloric ring because of its size. Then we tried to obtain the specimen from anus. Polyethylene glycol solution was administered to accelerate rapid excretion.

Outcomes: The mass was successfully removed and was histologically confirmed as a giant BGH, measuring 55mm in size.

Lessons: Reports about endoscopic resection of giant BGH are rare. Moreover, our case is the first report of transanal retrieval of resected specimen using polyethylene glycol solution. Endoscopic resection of BGH is less-invasive but can be more challenging if the mass is large. Our case provides useful option for endoscopic treatment of giant BGH.

Abbreviations: BGH = Brunner gland hamartoma, CT = computed tomography, EGD = esophagogastroduodenoscopy, EMR = endoscopic mucosal resection, ESD = endoscopic submucosal dissection, HSP = hot snare polypectomy.

Keywords: anemia, Brunner gland hamartoma, endoscopic resection, transanal retrieval

1. Introduction

Brunner gland hamartoma (BGH) is a rare tumor of the duodenum arising from the Brunner glands.\textsuperscript{[1]} Although BGH is a benign tumor, patients who have larger lesion with gastrointestinal symptoms such as bleeding, nausea, vomiting, and abdominal pain,\textsuperscript{[2–4]} require tumor removal. Larger BGHs have been generally treated by surgical intervention,\textsuperscript{[5,6]} while some lesions have been treated by endoscopic resection despite its larger
Achievement of en bloc specimen would be important for histological confirmation of BGH, however, in larger lesions, transoral retrieval of the resected specimen may be difficult because of its size. Here, we report a BGH measuring about 5.5 cm in proximal duodenum, successfully treated by endoscopic excision followed by transanal retrieval.

2. Case report

A 38-year-old woman presented with severe anemia, tarry stool, and vomiting. She had no remarkable medical history. Before being referred to our hospital, she visited to a clinic and was diagnosed as severe anemia with 6.3 g/dL hemoglobin by hematological test. She was also informed of a protruding lesion at the duodenal bulb by esophagogastroduodenoscopy (EGD).

On physical examination, pallor was noted, while her abdomen was soft and free from pain and tenderness. Slight fever and tachycardia (temperature 37.2°C, pulse 117 beats/min) were noted but other vital signs (pressure of 142/99 mmHg, oximetry saturation 100%) were normal. Since oral iron supplement has been offered to the patient for 3 weeks before she was admitted to our hospital, hematological test showed improvement of anemia with 9.7 g/dL hemoglobin. Other laboratory results were within normal limits, including tumor marker levels. Helicobacter pylori test was negative.

EGD showed a pedunculated giant submucosal mass at the anterior wall of the duodenal bulb (Fig. 1A). From the body to the top of the lesion, congested hyperplastic change and erosive appearance were seen on the mucosal surface (Fig. 1B). Using magnifying endoscopy with narrow band imaging (NBI), dilated villous and pit structures were shown at the erosive area but there was no evidence of endoscopic appearances associated with neoplastic lesions such as irregular surface and capillary patterns (Fig. 1C). At the base of the lesion, the width of the stalk was > 10 mm (Fig. 1A). Computed tomography (CT) analysis and barium x-ray examination also showed a mass near the proximal duodenum measuring about 55 mm in size (Fig. 2A and B). Neither nodal metastasis nor extramural extension was indicated. Although a conclusive diagnosis had not been made for the lesion, we could not rule out the possibility of malignancy. Because the lesion seemed to be responsible for patient’s anemia and vomiting, we thought that removing the mass was necessary.

Figure 1. EGD findings. (A) A pedunculated giant submucosal mass seen the anterior wall of the duodenal bulb. At the base of the lesion, a bulky stalk is seen. (B) From the body to the top of the lesion, the mucosal surface revealed congested hyperplastic changes. Erosive appearance was also seen. (C) Magnifying endoscopy with NBI showed dilated villous and pit structures at the erosive area but there was no evidence of endoscopic appearances associated with neoplastic lesions such as irregular surface and capillary patterns. EGD = esophagogastroduodenoscopy, NBI = narrow band imaging.

Figure 2. CT analysis (A) and barium x-ray examination (B). A mass measuring about 55 mm in size was shown near the proximal duodenum. (White arrows). CT = computed tomography.
Since it was a pedunculated lesion, we attempted to remove it by endoscopic resection. The lesion had clear but bulky stalk. We carefully cut the stalk using needle-knife (Zeon Medical Co., Tokyo, Japan) and IT knife2 (Olympus medical systems Co., Tokyo, Japan). After removing the lesion, hemoclips were immediately placed to prevent bleeding (Fig. 3A). We tried to retrieve the resected specimen from the duodenum to the stomach, but the mass could not pass the pyloric ring because of its size. We finally gave up retrieving it transorally. Instead, we tried to obtain the specimen from the anus. Although the patient had vomiting when she visited a clinic, there was no evidence of bowel obstruction and the entire lesion was within the duodenum (Fig. 1A). Polyethylene glycol solution (Nifec) was offered to accelerate rapid excretion. The mass was successfully obtained after 10 hours without any complications. The macroscopic examination showed lobulated mass measuring 5.5 × 4.5 × 2 cm.

![Figure 3. Endoscopic resection. (A) After cutting the stalk of the lesion using needle-knife and IT knife2, hemoclips were immediately placed to prevent bleeding. (B) EGD findings after 2 months showing no evidence of recurrence or tumor remnants. EGD = esophagogastroduodenoscopy.](image)

![Figure 4. Pathological evaluation of resected specimen. (A) Macroscopic examination showed lobulated mass measuring 5.5 × 4.5 × 2 cm. (B) Histologic section demonstrated mucus cells of the proliferating Brunner glands with basal nuclei without atypia. These proliferating Brunner glands formed lobules of various sizes. The mass was histologically confirmed as a giant BGH. BGH = Brunner gland hamartoma.](image)
endoscopic resection is indicated for relatively smaller BGHs that can be retrieved orally. If the resected specimen could be retrieved from the anal, physician can positively consider endoscopic resection for giant BGH. Our case provides useful information for endoscopists who perform endoscopic resection of giant BGH, but the safety of this procedure needs further evaluated. It should be noted that the possibility of bowel obstruction cannot be excluded if the mass is larger.

In conclusion, we reported a patient with a giant BGH who was successfully treated by endoscopic excision followed by transanal retrieval. Although BGH is a benign tumor, resection should be considered because large tumors may cause symptoms such as bleeding and pain. Endoscopic resection would be preferable to conventional surgery because of less-invasiveness but would be more challenging if the mass is large. We believe that our case provides salient finding for many endoscopists who plan endoscopic treatment of giant BGH.

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