

## Case Report

## Massive Upper Gastrointestinal Bleeding Secondary to a Large Pedunculated Brunner Gland Hamartoma in the Duodenum

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### INTRODUCTION

Brunner glands are deep mucosal and submucosal alkaline secreting glands mostly located in the proximal duodenum with their quantity and size diminishing in the distal regions.<sup>1</sup> These alkalotic secretions neutralize the acidic contents of the stomach, therefore maintaining the duodenal mucosal epithelium and alkalotic milieu necessary for absorption.<sup>2</sup>

Brunneroma, or Brunner gland hamartoma (BGH), is a rare benign tumor with prevalence of 0.008%, comprising 10% of all benign tumors in the duodenum.<sup>3</sup> Most cases occur in patients aged 50 to 70 years.<sup>3</sup> Visualization of BGH is done by esophagogastroduodenoscopy (EGD), although endoscopic ultrasonography has been increasingly utilized for the evaluation of the origin, extent, and vascularity of the lesion.<sup>3</sup> Computed tomography (CT) scans may serve as an initial tool to detect large BGHs. It also can be done to confirm the absence of extraluminal extension.<sup>2</sup> Confirmation of diagnosis is established with histopathology, which shows Brunner glands, ducts, smooth muscle, fibrous tissue, and lymphocytes with immunohistochemistry positive for Mucin 6 (MUC6).<sup>4</sup> We present a rare case of massive upper gastrointestinal (GI) bleed caused by a large pedunculated BGH in a 70-year-old patient.

### CASE REPORT

A 70-year-old female with previous history of iron deficiency anemia presented to the emergency department for fatigue and lightheadedness accompanied by new onset black tarry stools of two days' duration. The patient had no previous history of non-steroidal anti-inflammatory drug (NSAID) or aspirin use. Upon presentation, she was hemodynamically stable. The abdominal examination revealed no significant findings, with the absence of tenderness or distension. Initial labs revealed a hemoglobin level of 5.4 g/dl, significantly lower than her baseline of 9 g/dl. Labs also indicated iron deficiency anemia. Intravenous (IV) fluids and IV pantoprazole were started. Two units of packed red blood cells were transfused. Although the patient's hemoglobin improved to 7.9 g/dl following the transfusion, it dropped back to 6.8 g/dl, while still having persistent black stools. EGD showed a polypoid mass with irregular mucosa and ulceration in the second portion of the

duodenum away from the major papilla, so biopsies were taken (Figure 1). A subsequent CT of the abdomen and pelvis showed no extraluminal masses. With ongoing hemoglobin decline, dropping to 5.1 g/dl, CT angiography of the abdomen and pelvis revealed bleeding from the gastroduodenal artery, which was managed by embolization. Initially, pathology indicated inflamed and ulcerated benign mucosa with reactive epithelial changes. *Helicobacter pylori* testing was negative on biopsy. The patient underwent another EGD for complete endoscopic mucosal resection (EMR) and an endoscopic ultrasound (EUS). EUS demonstrated a hypoechoic pedunculated mass measuring up to 21 mm confined to the mucosal layers. The lesion was noted to be well away from the ampulla. Epinephrine was injected to decrease the bleeding risk, followed by successful placement of a Polyloop™ ligature (Figure 2). Hot snare mucosal resection was successful at the level of the polyp and the BGH was completely retrieved (Figure 3). A hemostatic clip was placed to prevent rebleeding afterwards. Pathology results after complete resection revealed prominent lamina propria capillaries, gastric mucin cell metaplasia, Brunner's gland hyperplasia, and cystic dilatation, indicative of BGH (Figure 4). After resection, the patient's tarry stools resolved, and her hemoglobin stabilized at around 9 g/dl.



Figure 1. Brunner Gland Hamartoma (BGH) in the second portion of the duodenum away from the major papilla.

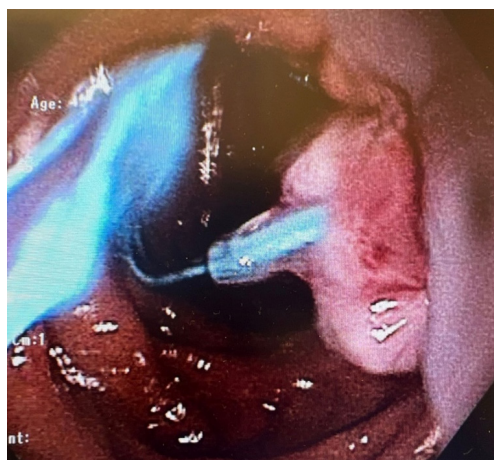


Figure 2. Polyloop™ ligature placed on Brunner Gland Hamartoma (BGH).

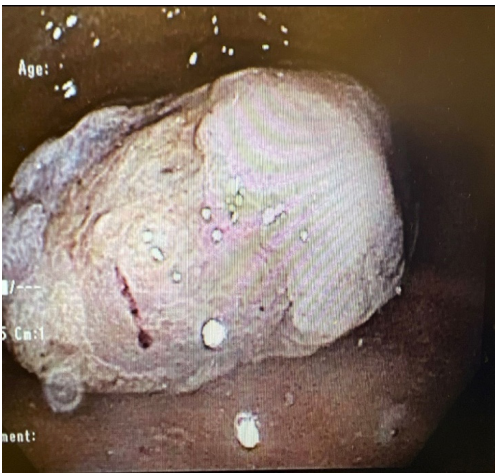


Figure 3. Brunner Gland Hamartoma (BGH) post endoscopic mucosal resection (EMR).

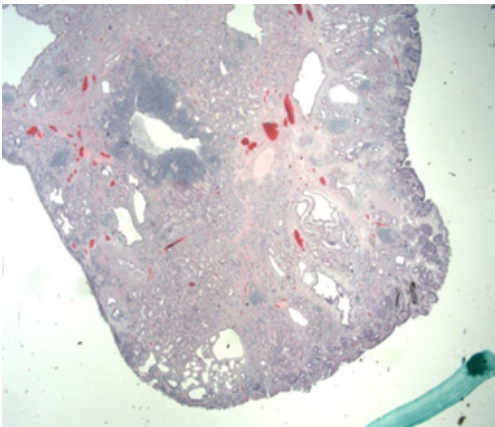


Figure 4. Brunner Gland Hamartoma (BGH) under 1.25x magnification hematoxylin and eosin (H&E) staining.

## DISCUSSION

BGHs are rare benign duodenal tumors ranging from 1 to 3 cm in size.<sup>5</sup> Up to 70% of these tumors have been found to occur near the duodenal ampulla.<sup>6</sup> BGHs generally are solitary and become pedunculated and polypoidal as they grow.<sup>7</sup> While they often are asymptomatic and detected incidentally, they can occasionally present with symptoms such as intestinal obstruction or upper GI bleeding.<sup>6</sup> Symptomatic patients with upper GI bleeding often present with anemia from chronic blood loss. They also report melena four times more commonly than hematemesis.<sup>8</sup> In rare cases where a tumor extends into the pancreas, it can obstruct the ampulla of Vater or pancreatic duct, potentially leading to complications such as inflammation, pancreatitis, or obstructive jaundice.<sup>8</sup>

In this case, we found a large pedunculated BGH measuring 21 mm in the second portion of the duodenum away from the major papilla, which is an atypical location. The patient presented with lightheadedness, weakness, fatigue, and black tarry stools, indicative of anemia and melena secondary to BGH bleeding.

To diagnose BGH, EGD and CT scanning are done initially to visualize the lesion causing the bleed.<sup>8</sup> CT scans often may show a polypoid filling defect, indicating extraluminal extension of the tumor.<sup>9</sup> In this case, we first visualized the tumor on EGD, and a subsequent CT scan showed no filling defects or extension into the duodenal wall.

For a definitive diagnosis of BGH, histological examination of the tumor is needed.<sup>10</sup> Endoscopic biopsies usually are superficial and not

sufficient to reach the BGH in the submucosa, which often leads to falsely negative results initially.<sup>2</sup> Hence, the final diagnosis is made based upon histological features after tumor resection.<sup>10</sup> Microscopically, BGHs show polypoidal growth of Brunner glands, with fibromuscular and adipose tissue components, lymphoid aggregates, and cystic dilatation of Brunner glands.<sup>1</sup>

Treatment usually involves endoscopic polypectomy for pedunculated tumors >2 cm in size.<sup>3</sup> It is generally safe, minimally invasive, and cost-effective.<sup>5</sup> However, surgical resection sometimes is necessary if endoscopic measures are not possible or have failed.<sup>8</sup> In the literature, there are no reported cases of BGH recurrence after complete resection, and the long-term outcomes are highly favorable.<sup>5</sup>

This case underscores the importance of complete resection for accurate diagnosis, as initial biopsies may not always reveal characteristic features due to its submucosal location.

## CONCLUSIONS

BGHs typically are benign lesions diagnosed incidentally during EGD or imaging. However, in some cases they can present with significant complications, such as GI hemorrhage or obstruction. Given the diagnostic challenges associated with biopsy sampling, complete resection of the tumor is imperative for achieving a definitive diagnosis. Both diagnosis and treatment of symptomatic BGH involve complete resection of the tumor with endoscopic or surgical approaches.

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