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CASE REPORT | SMALL BOWEL

Recurrence of Brunner Gland Adenocarcinoma After Duodenectomy and Gastrojejunostomy

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ABSTRACT

Malignant neoplasms arising from the Brunner gland are exceedingly rare. A 62-year-old man with a history of surgical resection of Brunner gland adenocarcinoma presented with upper extremity cellulitis. Hospital course was complicated by atrial fibrillation and hematochezia. Bidirectional endoscopy was negative; however, small bowel enteroscopy revealed recurrence of Brunner gland adenocarcinoma 6 years after surgical resection. To our knowledge, this is the first reported case of recurrent Brunner gland adenocarcinoma after curative resection.

INTRODUCTION

Small intestine cancers are rare, comprising only 2% of gastrointestinal cancers and 0.42% of total cancers.[1–4] Malignant neoplasms arising from the Brunner gland are exceedingly rare, with only a handful of cases reported in the literature. We describe a case of recurrence of Brunner gland adenocarcinoma (BGA) 6 years after surgical resection with a Billroth II gastrojejunostomy.

CASE REPORT

A 62-year-old man with a history of gastroparesis presented with worsening anemia of several months. Esophagogastroduodenoscopy revealed a 20 mm, villous-appearing mass in the first portion of the duodenum; biopsy revealed high-grade dysplasia. Positron emission tomography and computed tomography were without uptake outside of the duodenum. Endoscopic ultrasound showed the lesion extending from the mucosa and invading the muscularis propria; fine needle aspiration of surrounding lymph nodes did not show invasion. The patient was referred to surgical oncology where a surgical resection and gastrectomy with gastrojejunostomy were considered because the tumor seemed to involve all layers and patient's long-standing history of gastroparesis. During the operation, the endoscope located the mass on the lateral wall of the second and third portions of the duodenum. A partial duodenectomy was performed to excise the mass; gastrectomy and gastrojejunostomy were performed between the jejunum and the posterior wall of the stomach, creating a Billroth II anatomy. Immunohistochemistry (IHC) performed at the Massachusetts General Hospital showed a strong expression of MUC6 with coexpression of MUC5AC. Final diagnosis of BGA was made. The patient was discharged without postoperative complications but was lost to follow-up for surveillance.

Six years later, the patient was admitted for cellulitis of right upper extremity. During hospitalization, he developed atrial fibrillation and was started on anticoagulation; few days later, he was noted to have hematochezia and significant anemia requiring multiple transfusions. Bidirectional endoscopy did not show evidence of bleeding or other abnormalities. Small bowel enteroscopy revealed a 25 mm polypoid appearing mass in the second portion of the duodenum upstream from the ampulla (Figure 1). Histological analysis revealed the mass to be an adenoma with high-grade dysplasia (Brunner gland type), indicating recurrence of the previous malignancy (Figure 2). After an uneventful clinical course, the patient was discharged with a plan for surgical oncology and interventional gastroenterology follow-up.

Two weeks after discharge, the patient presented with melena. Repeat small bowel enteroscopy revealed a bleeding ulcer at the gastrojejunostomy anastomosis. The ulcer was treated with epinephrine injection and hemostatic clips, but the patient continued to have hematochezia and eventually developed hemodynamic instability requiring vasopressor support. Unfortunately, the patient developed respiratory failure requiring mechanical ventilation and ultimately died in the intensive care unit.

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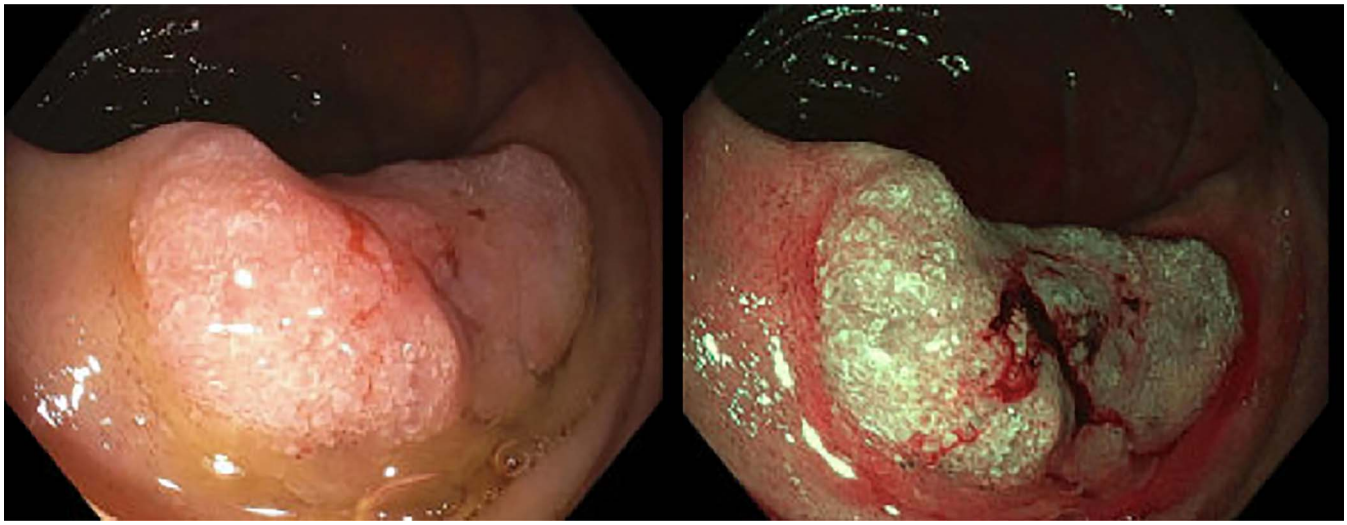

Figure 1. Duodenal polypoid mass upstream from ampulla.

DISCUSSION

Brunner glands are deep or submucosal acinar glands of the duodenum that secrete a viscous alkaline mucus to perform digestive and metabolic functions. They may extend distally from the pylorus to the first and second portions of the duodenum. Rarely, these glands undergo hypertrophy or hyperplasia which is termed a Brunner gland adenoma. It is controversial whether asymptomatic Brunner gland adenomas need to be removed; generally, these adenomas do not undergo malignant transformations.[5] Many experts argue they can be watched unless become symptomatic. The adenocarcinoma type of Brunner gland is extraordinarily rare; around 25 cases have been reported in the literature from 1994 to 2017.[6] Clinical presentations are nonspecific and may include obstructive symptoms (abdominal pain, nausea, vomiting), weight loss, and gastrointestinal bleeding. Diagnosis is established through endoscopic evaluation or intraoperatively during intraabdominal surgeries; radiography is not sensitive.[7] Current opinion suggests that a high gastric acid state (chronic hyperchlorhydria) leads to glandular hyperplasia, which may further develop into a thickened adenoma or hamartoma.[8] No exclusive IHC marker exists for the Brunner glands; therefore, published cases have reported diverse IHC markers such as MUC6 (pyloric/Brunner gland-type mucin) and MUC5AC (gastric foveolar-type mucin).[6,9,10] Koizumi et al conducted a literature review of 20 reported cases of BGA: The mean age was 67.4 years (range 39–85), more men were affected than women (15:5), and the mean tumor diameter was 25.6 mm (range 7–70 mm).[10]

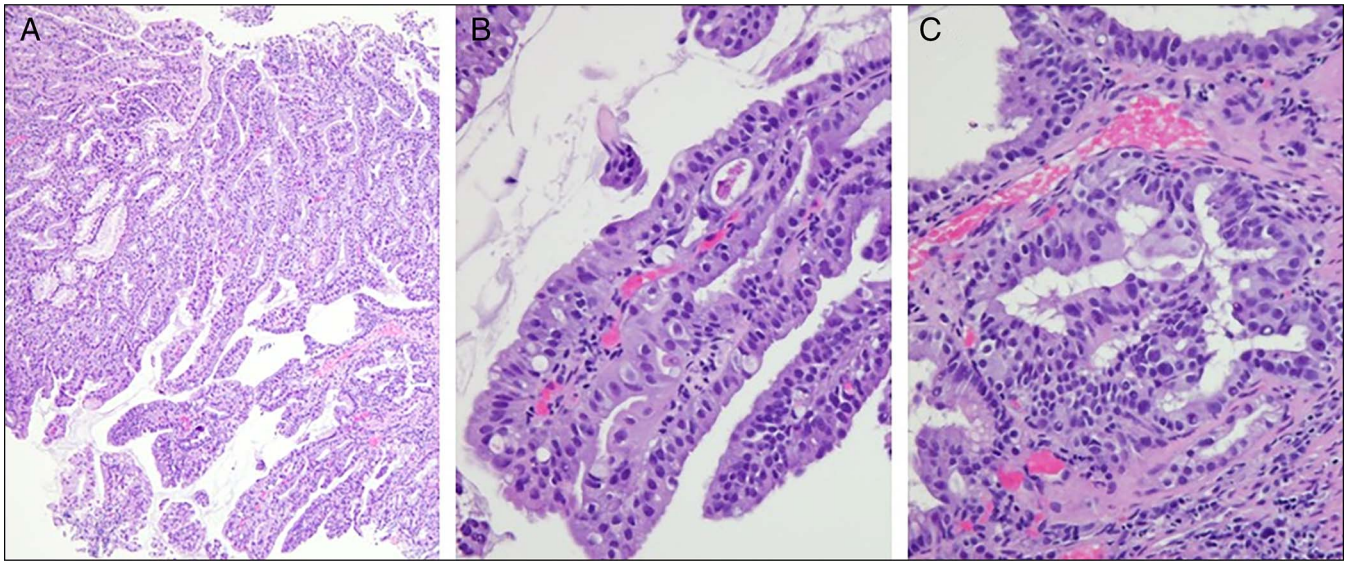

Figure 2. Histological sections (hematoxylin and eosin): (A) duodenal tumor composed of crowded atypical glands lined by columnar epithelium with pale eosinophilic cytoplasm (×40), (B) high-grade dysplasia characterized by enlarged pleomorphic nuclei (×200), and (C) complex glandular architecture (×200).

Treatment for duodenal malignancies is changing: In the past, surgical resection (pancreaticoduodenectomy, segmental resection) was the primary approach.[11] However, a systematic review showed that endoscopic removal of duodenal lesions (snare resection, endoscopic submucosal dissection, endo loop technique) was associated with safe outcomes.[12] Several cases of small or pedunculated Brunner gland lesions successfully treated with endoscopic polypectomy have been reported.[13,14] Ultimately, the treatment modality depends on the tumor location, staging, and patient characteristics. Gold et al reported that lymph node involvement was a significant prognostic factor in the decision-making process regarding surgical or endoscopic therapy.[5] The presence of a vessel within the stalk increases the risk of post-resection bleeding, and pretreating with epinephrine and/or mechanical hemostasis can yield safe outcomes.[12] Surgical excision may be desired for large tumors or cases that failed snaring. Koizumi et al reported that resected BGAs (both surgical and endoscopic) had relatively good survival over a mean observation period of 21.6 months (range 3–45 months), although follow-ups after 45 months were not ascertained.[10]

Presently, there are no established recommendations regarding surveillance because a clear malignant potential is not yet determined. Sakurai et al reviewed 722 cases of Brunner gland hyperplasia, and 15 (2.1%) had dysplastic changes, suggesting the possibility of carcinogenesis.[16] Thus, patients with risk factors (symptomatic, positive IHC markers) may benefit from endoscopic ultrasound/fine needle aspiration and cytopathologic examination. DNA mismatch repair and microsatellite instability test were not performed in our patient. Although there are some reports on its utility in preventing small bowel adenocarcinomas, association with malignancies other than colorectal cancer has not been fully investigated.[17] Of the few reported cases of BGA, Moon et al reported conducting a microsatellite instability test; however, none of the markers showed mismatch repair failure.[5]

This case is exceptional because of late recurrence of BGA after curative resection. In addition, it illustrates the point that an altered anatomy of Billroth II operation can complicate endoscopic detection of culprit lesions in patients with gastrointestinal bleeding. Clinicians encountering this rare malignancy may need to surveil for a longer period after surgical resection and remain vigilant for signs of recurrence.

DISCLOSURES

Author contributions: All authors have contributed to the conception, acquisition, interpretation, writing, and revision of this paper. P. Hong is the article guarantor.

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