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Single Case

En Bloc Endoscopic Resection of Large Pedunculated Brunner's Gland Hamartoma: A Case Report

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Keywords

Brunner hamartoma · Gastrointestinal hemorrhage · En bloc endoscopic resection

Abstract

Brunner's gland hamartoma (BGH) is an uncommon benign tumor arising from alkaline-based mucin-secreting glands of the duodenum. These lesions are typically discovered incidentally during upper gastrointestinal endoscopy or radiological diagnostics, even though they can eventually lead (in the case of increasing size) to obstructive or hemorrhagic symptoms. We report the case of a 67-year-old Caucasian man with two episodes of melena and epigastric pain during the last month. Esophagogastroduodenoscopy revealed a large pedunculated and eroded polyp inside the antrum with a 2-cm-long stalk arising from the anterior wall of the duodenal bulb. Endoscopic ultrasonography showed a submucosal lesion with homogeneous hyperechoic parietal thickening and some central gaps. The muscularis was undamaged. No lymphadenopathy was observed. We performed an en bloc endoscopic resection of the polyp. The size of the whole piece was approximately 6 × 3 × 2 cm, pseudocapsulated and tough in consistency. In the case presented, the en bloc endoscopic removal was successful despite the size of the tumor.

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Published by S. Karger AG, Basel

Introduction

Brunner's gland hamartoma (BGH), also known as Brunneroma or Brunner's gland adenoma, is an uncommon benign tumor arising from alkaline-based mucin-secreting glands of the duodenum [1]. These lesions account for 10% of benign tumors of the duodenum [2]. BGHs are predominantly found in the duodenal bulb (57%) with decreasing frequency in the second (27%) and third parts (5%) of the duodenum. This distribution corresponds to the anatomical distribution of Brunner's glands [3]. They secrete an alkaline fluid of mucin as well as pepsinogen and urogastrone, which protect the epithelium from and inhibits secretion of gastric chyme, respectively [4–6]. BGHs are most prevalent in the fifth and sixth decades and represent genders and race equally [5, 6]. These lesions are typically discovered incidentally and during upper gastrointestinal endoscopy or radiological diagnostics, even though they can eventually lead (in the case of increasing size) to obstructive or hemorrhagic symptoms such as abdominal pain, nausea, dyspepsia, vomiting, intussusception, upper gastrointestinal bleeding, and recurrent pancreatitis [7].

Case Report

A 67-year-old Caucasian man came to our hospital for a second opinion because a surgical resection was proposed to him at the time of first esophagogastroduodenoscopy (EGD) performed in another hospital. One month before, he had two episodes of melena accompanied by epigastric pain for which he went to the nearest emergency room. There he had an EGD performed that showed a big polyp of the duodenum to be surgically removed. When the patient came to us, he was in good health, he was only under chronic antihypertensive therapy, and transfusions were not necessary for the previous episode of melena. He denied nonsteroidal anti-inflammatory drugs abuse. The new EGD revealed a large pedunculated polyp with regular mucosa but with eroded apex, with a 2-cm-long stalk arising from the anterior wall of the duodenal bulb (Fig. 1) and spontaneously migrating between antrum and second duodenum. Endoscopic ultrasonography (EUS) performed with high-frequency mini-probe showed a submucosal lesion with homogeneous, hyperechoic parietal thickening, and some central gaps. The muscularis propria was undamaged. No lymphadenopathy was observed (Fig. 2). We proposed to the patient an endoscopic resection of the polyp to prevent other episodes of bleeding or obstruction, and he agreed. To perform the procedure in deep sedation and to promptly treat any complications, the patient was admitted to our gastroenterology ward. He underwent ECG and chest X-ray, which resulted normal. The anesthetic examination did not place any contraindications to anesthesia, the blood counts were in the normal range. The anesthesia was done with Diprivan and ketamine iv. The EGD confirmed that the polyp was attached to the anterior wall of the bulb immediately downstream of the pyloric ring. Resection was made en bloc by positioning a diathermic loop at the base of the polyp from the stomach. The size of the polyp, which was located inside the duodenal bulb, made the procedure challenging at the time of the scheduled EGD; therefore, we decided to pull it up into the stomach and cut the stalk from this position (Fig. 3). A plastic cap was used to obtain a stable position of the scope inside the pylorus. There was no bleeding from the resected section, and there were no visible blood vessels (Fig. 4). No complication occurred, the polyp was recovered through a Basket Polyp Retriever and sent for histological analysis (Fig. 5). When the patient

was brought back in the ward, he was conscious and did not complain of pain. His blood pressure was 120/80 mm Hg, saturation 98%, and heart rate 54. He fasted for 24 h and underwent infusion therapy with proton pump inhibitors at maximum dosage and hydration.

The size of the whole piece was approximately 6 × 3 × 2 cm, pseudocapsulated and tough in consistency. The gross endoscopic resection specimen showed a well-defined yellow-white solid mass lined by a normal duodenal mucosa measuring 6 × 3 × 2 cm (Fig. 6). The histological examination showed nodular physiognomy of Brunner glands with ductular structures and epithelial coating and without significant atypia and scattered stromal elements in the submucosa (Fig. 7). This morphological picture is compatible with BGH (Fig. 8).

Discussion

BGH was described for the first time by Johann Conrad Brunner (1653–1727) in 1688. He was a Swiss anatomist who gave a precise anatomic description of the duodenal submucosal glands and coined the term “pancreas secundarium.” These glands are branching acinotubular glands that arise in the duodenum submucosa. In 1934, Feyter proposed a classification of the abnormal proliferation of Brunner's glands into three different subsets: type 1, diffuse nodular hyperplasia; type 2, circumscribed nodular hyperplasia; and type 3, glandular adenoma [8]. According to this classification, the diagnosis of BGH is based on histological features, such as lack of surfacing capsule, network combination of acini, smooth muscle, adipose tissue, Paneth cells and mucosal glands, and mostly a lack of cellular atypia [9]. The pathogenesis of BGH is still unknown, but there are some hypotheses. Three of the most quoted are as follows: hyperchlorhydria-like trigger of disease, correlation to pancreatic diseases, and inflammation [10–12]. Most patients are asymptomatic, but some may present with nonspecific symptoms, such as abdominal bloating, abdominal pain, or nausea, and other patients may present with hemorrhage or obstruction. EUS is specifically useful in evaluating extension, morphological characteristics and the vascularity of BGH [13]. The CT scan is a second-line study that permits the physician to analyze the extent of BGH and its relationship with adjacent structures [14]. The differential diagnosis of duodenal growth includes leiomyomas, adenomas, lipomas, adenocarcinomas, carcinoid tumors, lymphomas, leiomyosarcoma, pancreatic or ampullary tumors, and the lesions of Peutz-Jeghers syndrome [15, 16].

Because upper endoscopic biopsies usually fail, an accurate study using EUS and, if necessary, CT scan is mandatory to study the extension, morphological characteristics, and vascularity of the lesion. In particular, the position in the duodenum, dimensions, and invasion of muscularis propria are the three fundamental elements when deciding whether to do an endoscopic or surgical resection. Endoscopic polypectomy is the suggested treatment for duodenal lesions occupying the first part of the duodenum, whereas more aggressive treatment has been reported in the case of D2 or D3 BGH localization. Endoscopic resection is indicated for the removal of pedunculated polyps. Regarding acceptable size, some things have changed compared to the past – now a safe endoscopic resection is possible also for lesions measuring more than 3 cm. This is possible thanks to advanced endoscopic techniques and to a careful study of the morphological characteristics of the lesion. In our case, the polyp was pedunculated, did not invade the muscularis propria, and satellite lymphadenopathies were absent, which suggested a benign nature of the lesion. Endoscopic resection in selected cases prevents demolition surgery. Treatment choice depends on tumor size, symptoms, and malignancy. Endoscopic treatment is preferred for pedunculated lesions, whereas surgical resection is man-

datory in the presence of malignancy or if endoscopic methods fail. Asymptomatic lesions usually have low neoplastic potential, and therefore, they only require a follow-up. In the case presented, en bloc endoscopic removal was successful despite the size of the tumor.

Statement of Ethics

The study was reviewed and approved by the Comitato Etico of Sacro Cuore Don Calabria Hospital of Negrar Institutional Review Board.

Disclosure Statement

The study participant provided informed consent prior to study enrollment.

Author Contributions

P.B. and E.C. designed and performed the work. E.C., G.Q., and R.M. contributed to the drafting. A.P. wrote the paper.

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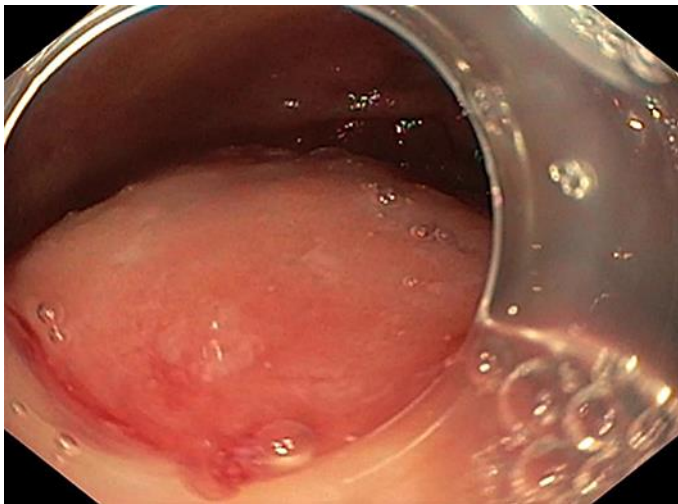


Fig. 1. Endoscopic view of a large duodenal polyp inside the bulb.

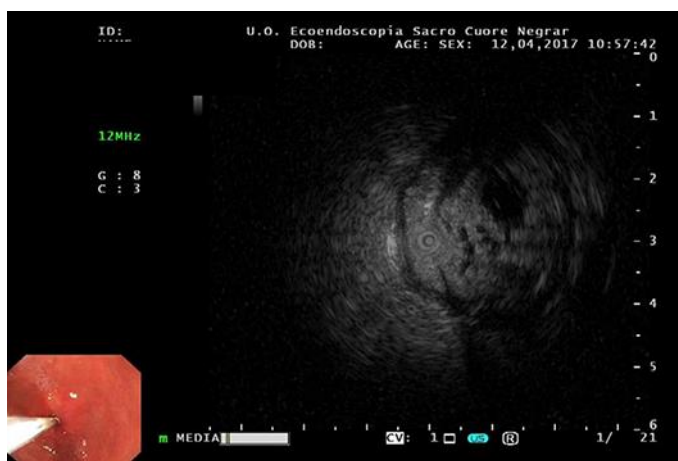


Fig. 2. Endosonographic view (Olympus 12-MHz EUS miniprobes under water). The polyp seems to originate from the submucosa without affecting the muscularis layer.

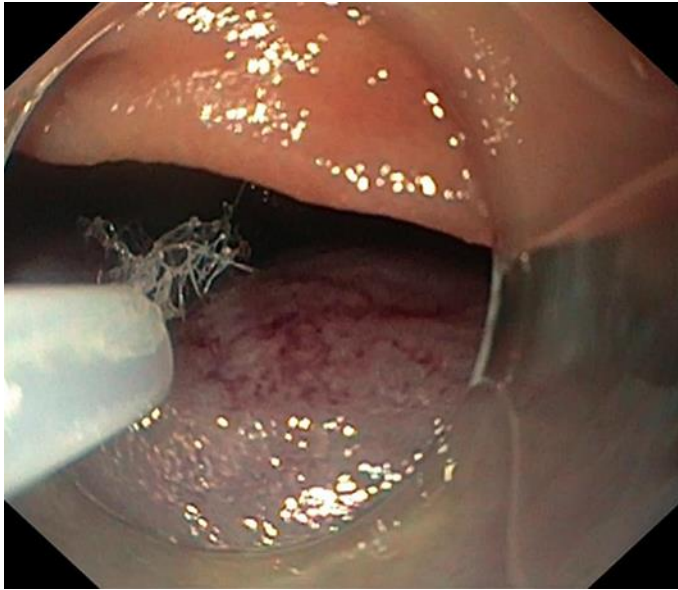


Fig. 3. The appearance of the newly resected polyp.

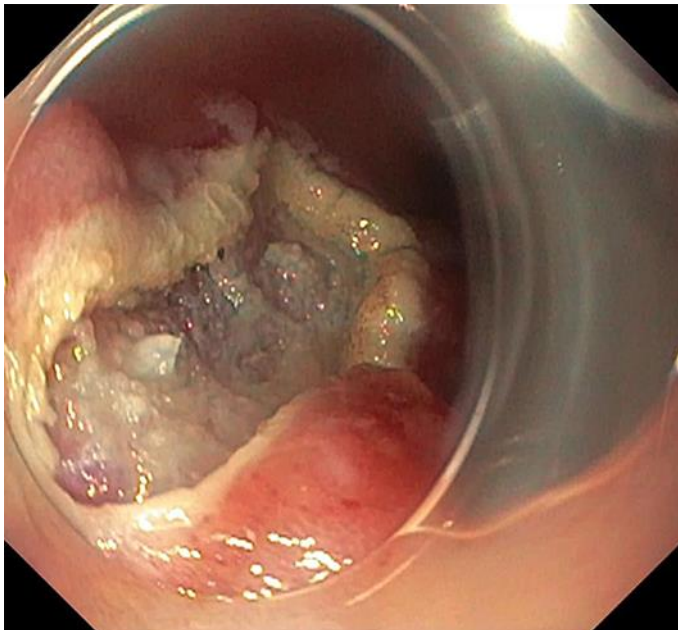


Fig. 4. Base of the polyp resection in the duodenal bulb.

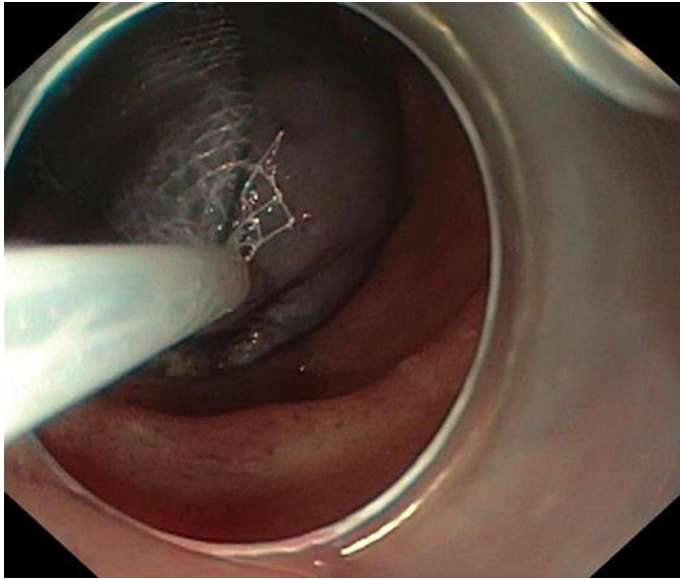


Fig. 5. The polyp is recovered by means of the Memory Basket Polyp Retriever.

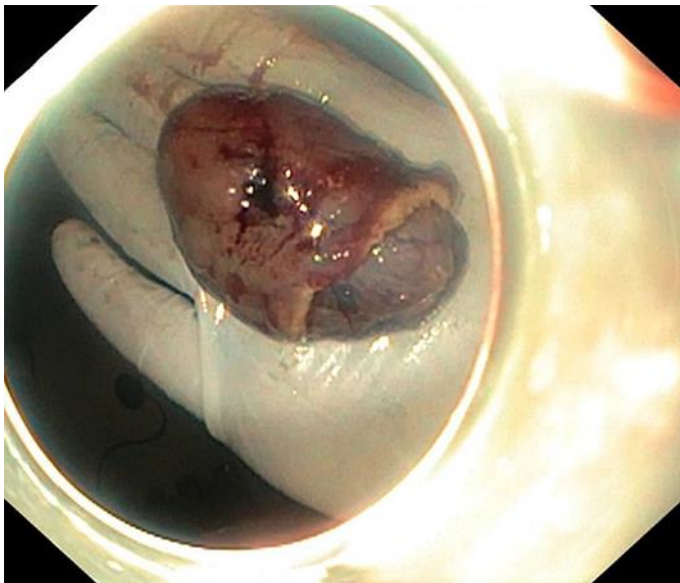


Fig. 6. The resected specimen in the operator's hand. The dimensions were 6 × 3 × 2 cm.

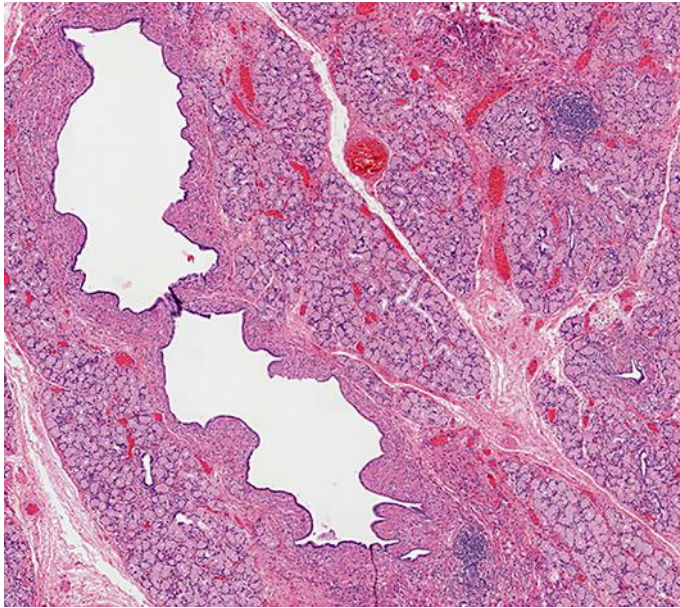


Fig. 7. Dilated ducts with some structural irregularity and cuboidal cells with scant cytoplasm are present in this sclerotic glandular focus, which retained a lobular architecture. Note the bland cytology with absent mitotic activity. HE. $\times 10$.

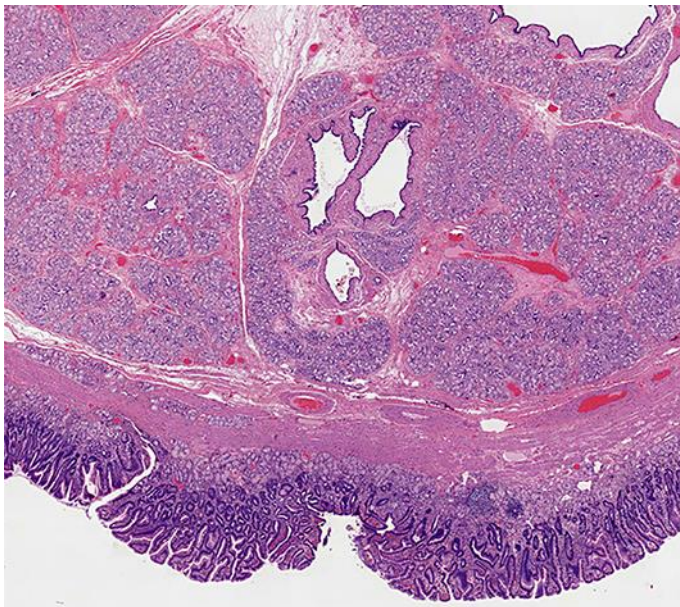


Fig. 8. Histological examination shows a submucosa proliferation of Brunner's glands in a lobulated pattern with dilated ducts and fibrous septa. HE. $\times 4$.