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A Large Brunner's Gland Hamartoma Causing Gastrointestinal Bleeding and Obstruction

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Key Words

Brunner's gland hamartoma · Duodenal neoplasms · Gastrointestinal hemorrhage

Abstract

Context: Clinical symptoms of Brunner's gland (BG) hamartoma are rare. Most lesions are small and asymptomatic. Symptomatic BG hamartoma may mimic a malignancy of the duodenal-pancreatic area. Though standard mucosal biopsies are inferiorly diagnostic due to normal duodenal mucosa surrounding, a biopsy was indicated in this case. **Case Report:** We report the case of a 37-year-old man with a large BG hamartoma mimicking a malignancy. Preoperatively the biopsy led to an inconclusive diagnosis. Due to its large size and its presence of symptoms, a surgical excision was performed. **Conclusion:** BG hamartoma may be a rare indication for surgery.

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Introduction

Benign small bowel neoplasms are uncommonly encountered. Brunner's gland (BG) hamartomas comprise a small portion of these benign tumors. BG hamartoma, also known as BG adenoma or Brunneroma, is a rare proliferative lesion from the BG of the

duodenum, first described by Salvioli in 1872. These glands are located in deep mucosa and submucosa of the duodenum, functionally protecting duodenal epithelium from acid.

They have a rapid response to presence of acid in the first part of the duodenum and respond by a large secretion of alkali mucus. BG hamartomas are commonly small and asymptomatic, and in most cases they are discovered incidentally. Clinical symptoms are caused by obstruction, leading to postprandial pain or bleeding, which is often occult [1]. The location of the lesion is often at the bulb of the duodenum and the incidence decreases with increase of the distance from the pyloric ring [2]. When symptoms occur, it can be effectively treated with endoscopic removal; however, in some cases large lesions should be surgically removed.

We present an unusual presentation of a large BG hamartoma with gastrointestinal bleeding and obstruction.

Case Report

A 38-year-old black male was presented with upper abdominal discomfort postprandial, reduced appetite, fatigue, dizziness and weight loss. He had no complaints of nausea, vomiting or a history of gastrointestinal bleeding. Family history was negative for gastrointestinal diseases. Physical examination revealed no abnormalities. Laboratory evaluation showed a microcytic hypochromic anemia.

Upper tract endoscopy showed a large mass with normal intact mucosa in the third part of the duodenum extending to the proximal jejunum, which could not be passed. A biopsy specimen was non-diagnostic.

Contrast computed tomography scan showed a polypoid mass in the proximal jejunum with filling defect and a feature suggestive of a mass outside the intestines, obstructing the duodenal bulb, suggesting a lymphoma or other tumor (fig. 1). There were no signs of metastatic disease. However, barium radiographic study revealed no impression from outside.

Fig. 1. Radiological examination of the upper gastrointestinal tract reveals circumferential small bowel wall thickening. This can be a feature of neoplasm, intussusception or inflammation.

Fig. 2. Surgical finding of a large pedunculated polypoid tumor with a benign appearance in the duodenum.

Fig. 3. Organoid appearance of a hyperplastic gland consistent with a BG hamartoma.

Duodenotomy disclosed a pedunculated polyp within the posterior wall of the pars horizontalis of the duodenum. On gross examination of the resection specimen, a mass with lobulated, polypoid appearance and hemorrhagic cover, without ulceration, measuring 10 cm in diameter was observed (fig. 2).

Histological assessment revealed a pedunculated convolute with nodular hypoplastic BGs consistent with a BG hamartoma. No signs of malignancy were found (fig. 3).

Discussion

BG hamartomas represent 5–10% of small bowel tumors and occur most commonly in the fifth and sixth decade of life, with no gender or race predominance [3]. BGs are branched acinotubular glands, secreting mucus. They are mainly found in the submucosa of the duodenum and incidentally in the pylorus and jejunum [1].

Levine et al. [2] reported that 70% of BG hamartomas are located in the first portion of the duodenum. They are either pedunculated (89%) or sessile (11%), located in submucosa and mucosa, decreasing progressively in size and number more distally. In our patient, the mass was located in the third part of the duodenum, which is more likely to hemorrhage [2, 3]. In the group of patients with tumor-related hemorrhage, manifested as hematemesis or melena, blood loss is often chronic with evidence of ulceration. Patients with tumor-related obstructive complaints tend to have hamartomas >2 cm.

Many mechanisms have been postulated to explain the etiology and pathogenesis of BG tumors. Local irritation, parasympathetic activity, chronic pancreatitis, *Helicobacter pylori* infection and Billroth II reconstruction are suggested [4]. The real stimulus is unknown.

Considerable variation in classification and terminology consists for BG proliferation: circumscribed nodular hyperplasia, diffuse nodular hyperplasia and localized adenomatous hy-

perplasia. Pathology of the categories is suggested to be identical with similar histologic features [5]. BG hamartoma is the most appropriate name. It is composed of a disorganized admixture of lobules of BGs and variable combination of ducts, adipose tissue, smooth muscle, sclerotic glands, lymphoid tissue and lack of encapsulation circumscribed under muscularis mucosa [4].

Accurate diagnosis can be difficult. Differential diagnosis usually includes leiomyomas, adenomas, lipomas, Peutz-Jegher's syndrome, adenocarcinomas, carcinoids, lymphomas, leiomyosarcomas, pancreatic or ampullary neoplasm [6].

Enteroclysis is offered to detect small intestinal lesions. It is believed to give accurate visualization of the intestinal lumen and mucosal surface [7]. Computed tomography is believed to be insensitive, but helpful for distinguishing relation of the lesion and its adjacent structures [8]. Mumtaz et al. [4] suggested that endoscopic ultrasound is the best imaging modality for diagnosis. The lesion is seen as a cystic or heterogeneous solid mass. Endoscopy can reveal the mucosal origin and the location of the hamartoma. Histological examination of local tissue biopsy specimens may not be representative due to the normal surrounding and submucosal position. Despite several investigation modalities, endoscopic or surgical exploration is often the only tool to achieve definitive histological diagnosis.

Either endoscopic polypectomy or confined surgical resection should be performed in symptomatic patients. Lesions >2 cm in diameter are best treated by surgical resection [9]. When feasible, the surgical procedure should be a transduodenal polypectomy or segmental duodenal resection for hamartomas located in the first, third and fourth portion of the duodenum. Lesions in the second portion may require duodenal bypassing, in the form of pancreaticoduodenotomy.

Very few cases of BG hamartomas have been reported in association with epithelial dysplasia, adenocarcinoma, and carcinoid tumors [10].

Our patient was treated because of the symptomatic nature of the large polyp. Its location hampered endoscopic resection.

Conclusion

This case illustrates a patient in the fourth decade of life with abdominal pain and anemia due to occult gastrointestinal bleeding, caused by a BG hamartoma of unusual size. However, symptomatic BG tumors are rare; they should be considered in the differential diagnosis of a duodenal tumor since they account for a significant part of small bowel neoplasms. BG hamartomas are benign in nature. Up until now, malignant recurrence after resection of a BG hamartoma has not been reported. Consequently, therapy is only necessary in symptomatic lesions preferably by minimal invasive therapy. In large tumors such as in the present case, surgical resection is necessary. Endosonographic follow-up of patients treated for BG hamartoma is not warranted.

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Giant Brunner's Hamartomas of the Duodenum and Obstructive Jaundice

An Overview of the Literature and Suspicion of Malignancy in a Case

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Key Words

Adenocarcinoma of papilla of Vater · Brunner's gland adenoma · Brunner's hamartoma · Giant Brunner's hamartoma · Obstructive jaundice · Whipple's procedure

Abstract

Background/Aims: 150 cases of Brunner's gland hamartoma (BGH) have been reported in the literature. BGHs are benign and are thought not to cause bile obstruction. **Methods:** In this case report, a 60-year-old male is presented with unexplained obstructive jaundice who was also known for over 17 years with diffuse adenomatous hyperplasia of Brunner's glands in the duodenum. Despite the benign preoperative diagnosis, the choice of treatment was Whipple's procedure due to suspicion of a coexisting malignancy. **Results:** Pathological analysis of the resection specimen revealed multiple BGHs and an adenocarcinoma of the papilla of Vater (PoV). Molecular pathology using loss of heterogeneity analysis was used to confirm that both were different entities. **Conclusion:** It is likely that previous reports of malignant degeneration of BGHs may actually have been cases involving the coexistence of a PoV adenocarcinoma. Physicians need to be alert when a patient presents with BGH accompanied with obstructive jaundice for simultaneously occurring PoV adenocarcinoma.

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Introduction

Approximately 150 cases of Brunner's gland adenoma have been reported in the literature since the first report in 1876 by Cruveilhier [1]. These tumors are discovered incidentally as an abnormal cause of duodenal obstruction or due to bleeding, but not as a cause of obstructive jaundice [2, 3].

Case Report

A 60-year-old male known with extreme diffuse adenomatous hyperplasia of Brunner's glands for over 17 years was referred to us with a reduced appetite, weight loss, and jaundice. Our patient's Brunner's gland adenomas were first diagnosed and reported in the literature in 1989 when he presented with meleana and anemia [1].

Physical examination revealed a thin man with jaundice with a palpable mass in the right upper abdomen. Liver enzymes were elevated and total bilirubin was 82 $\mu\text{mol/l}$.