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 $\mu\text{mol/l}$.

1

2

3

4

Fig. 1. Operation picture showing enormous enlargement of duodenum with a diameter of 25 cm (arrows).
Fig. 2. Operative resection specimen with large duodenal hamartomas and PoV tumor (with forceps through papilla).
Fig. 3. Detailed picture of resection specimen with arrow at PoV carcinoma and multiple hamartomas at proximal side (left side).
Fig. 4. Microscopic image of hamartoma. Lendrum stain; obj. 2.5×.

Intra and extra bile duct widening as well as a hydropic gallbladder filled with sludge was found during an echo of the abdomen. The ductus pancreaticus was normal. A CT scan showed an extreme widened duodenum with a thickening of the wall around the papilla of Vater (PoV). Multiple intraluminal swellings were found in the antrum, bulbus duodeni, pars descendens duodeni and pars horizontalis duodeni. The pancreas, especially the pancreas head region and the ductus pancreaticus, was normal. No abnormalities were found in the jejunum and ileum. Additionally, no evidence for malignancy was discovered.

Biopsies taken from the duodenum confirmed Brunner's gland hamartoma (BGH) without evidence of malignancy. Because it was known from the literature that BGHs are normally

benign and do not cause bile obstruction, the obstructive jaundice remained unexplained. Therefore, despite a preoperative benign diagnosis, the clinical presence of jaundice and CT scan showing duodenal wall thickening around the PoV made us decide to perform a radical/oncological resection of the duodenum and pancreas head via Whipple's procedure.

A pancreaticoduodenectomy, choledochojejunostomy, gastropancreaticostomy and gastrojejunostomy with a feeding jejunostomy was performed. The suspicion of a PoV carcinoma was confirmed during the procedure and radically removed (fig. 1–3). Macroscopic pathological analysis revealed large polyps throughout most of the proximal duodenum ranging from 0.5 to 10 cm with most being around 5 cm. All polyps were mucosal and loose

Fig. 5. Microscopic image of adenocarcinoma PoV. HE stain; obj. 2.5X.

Fig. 6. Microscopic image of adenocarcinoma PoV. Lendrum stain; obj. 5X.

from the intestinal wall. A 7.5 × 5 cm frayed and granular mass was found within the polyp located at the area of the PoV. At the height of the pancreas situated against the duodenum, noduli were seen. The largest nodule had a diameter of 1.5 cm and when dissected had a fatty appearance. During lamination it was found that the ductus of Wirsung led to the tumor mass.

Microscopic pathological analysis of the PoV tumor revealed a tubular structure with outspoken cytonuclear atypia. The adenocarcinoma did grow into, but not through the muscularis propria. All resection planes were free of tumor. Analysis of hamartomas showed Brunner's gland hypertrophy, no cytonuclear atypia, and increased lobular fibrous septa. Here and there, zones were found with large amounts of fat cells mimicking lipomas. Epithelium of hamartomas was normal. All lymph nodes were without tumor metastases (fig. 4–6).

In summary, multiple BGHs were found plus an adenocarcinoma of the PoV within the duodenum. Additional molecular pathology using loss of heterogeneity analysis (LOH) confirmed that BGHs and the adenocarcinomas of the PoV were two different entities.

Postoperative recovery was complicated by a retroperitoneal bleeding whereby a relaparotomy was required. Further recovery was uneventful and patient was discharged in good condition after 4 weeks. At follow-up 6 months later, the patient was found to be in perfect condition without symptoms of gastrointestinal bile duct obstruction.

Discussion

Benign tumors of the duodenum are rare with an incidence rate of 0.008% in patients at autopsy. Brunner's gland adenomas comprise 10.6% of these tumors [4]. Brunner's gland adenoma is specifically known as a benign tumor with a good prognosis, however a small number of 7 malignant cases have been reported in the literature [5, 6]. The molecular LOH pathological analysis

used in our case confirms that BGHs are benign and should not be confused during pathological analysis with a simultaneously occurring PoV adenocarcinoma.

Brunner's glands are mostly located proximally in the duodenum, deep in the mucosa and submucosa. They are branched acinotubular structures, which secrete mucus that empties into the crypts of Lieberkuhn. In addition, Brunner's glands secrete urogastrone and pepsinogen when stimulated by acid chyme from the stomach [7].

Obstructive symptoms can also be the chief complaint leading to discovery [8]. Two unique cases have been presented in the literature in which intestinal obstruction was caused by intussusceptions of the duodenal wall by hamartoma migration [3]. Additionally, 3 cases of bile duct obstruction have been described in combination with BGHs [2]. In these rare cases no clear explanation is given however.

Available literature regarding giant duodenal hamartomas demonstrates an inherent difficulty to correctly preoperatively diagnose the condition as well as an incurrent risk of surgical overtreatment. These are known factors clinicians have to face when dealing with BGHs and this case nicely illustrates these dilemmas.

Generally, BGHs are benign and have a good prognosis [6, 8]. Therefore, treatment should normally be conservative and directed towards relieving symptoms experienced by the patient. It is preferable to radically excise symptomatic tumors via an endoscope. Alternatively, if endoscopic treatment fails and digestive obstruction indicates surgical treatment, the pancreas-preserving duodenal replacement forms an attractive alternative [9]. In this case, the pancreaticoduodenectomy according to Whipple was chosen due to a suspected malignancy. Pathological follow-up confirmed that surgical treatment was the correct decision. As this case demonstrates, physicians need to be alert when a patient presents with BGH accompanied with obstructive jaundice for simultaneously occurring PoV adenocarcinoma.

Previous case reports have suggested malignant degeneration of BGHs. However, because none of these case reports used LOH analysis to discriminate between a coexisting malignancy of the PoV, it is more likely that these cases actually involved simultaneous malignant degeneration.

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