CASE REPORT

Brunner's gland hamartoma causing gastric outlet obstruction: case report

Hamartoma de glándulas de Brunner causando obstrucción piloroduodenal: reporte de caso

César Vivian Lopes[1], Sophia Andreola Borba[1], Felipe Luzzatto[3], Carolina Scherer Dornelles[1], Carlos Eduardo Oliveira dos Santos[1,2], Júlio Carlos Pereira Lima[1]

[1] Department of Gastroenterology and Digestive Endoscopy, Santa Casa Hospital, Porto Alegre, Brazil.
[2] Department of Endoscopy and Gastroenterology, Santa Casa Hospital, Bage, Brazil.
[3] Department of Pathology, Santa Casa Hospital, Porto Alegre, Brazil.

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Conflict of interest
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ABSTRACT
Proliferative lesions of Brunner's glands > 5 mm in size are called hamartomas. Brunner's gland hamartomas are located mainly in the first and second portions of the duodenum and are mostly asymptomatic. However, large hamartomas can lead to serious complications, such as gastric outlet obstruction. Herein, we describe the case of a patient with a large duodenal Brunner's gland hamartoma admitted with gastric outlet obstruction, which was resected successfully endoscopically. A 59-year-old female was admitted with a history of recurrent episodes of postprandial vomiting and upper abdominal pain. Esophagogastroduodenoscopy revealed a pedunculated polyp on the duodenal bulb, causing intermittent pyloric obstruction. Endoscopic biopsies were inconclusive. Linear endosonography showed a solid lesion arising from the submucosal layer, and samples obtained by EUS-guided tissue acquisition confirmed a Brunner's gland hamartoma. The lesion was endoscopically resected with a hot snare, and the specimen was transorally retrieved. After polyp removal, the patient's symptoms were completely resolved. A patient with recurrent episodes of postprandial vomiting and upper abdominal pain, in the presence of a subepithelial duodenal lesion, either pedunculated or sessile, should have ruled out a Brunner's gland hamartoma. EUS-guided tissue acquisition can confirm the diagnosis, and endoscopic resection is feasible for most cases.
Keywords: *Brunner's Glands; Hamartoma; Gastric Outlet Obstruction; Duodenum; Endoscopy, Gastrointestinal (fuente: DeCS Bireme).*

RESUMEN
Las lesiones proliferativas de las glándulas de Brunner > 5 mm de tamaño se denominan hamartomas. Los hamartomas de las glándulas de Brunner se localizan principalmente en la primera y segunda porciones del duodeno, y en su mayoría son asintomáticos. Sin embargo, los hamartomas de gran tamaño pueden conducir a complicaciones graves, como la obstrucción de la salida gástrica. En este documento, describimos el caso de una paciente con un gran hamartoma duodenal de glándulas de Brunner ingresada con obstrucción piloroduodenal, que fue resecado exitosamente por vía endoscópica. Una mujer de 59 años fue ingresada con antecedente de episodios recurrentes de vómitos posprandiales y dolor abdominal superior. La esofagogastroduodenoscopia reveló un pólipo pediculado en el bulbo duodenal, causando obstrucción pilórica intermitente. Las biopsias endoscópicas fueron inconclusas. La ecoendoscopia lineal mostró una lesión sólida originada en la capa submucosa, y las muestras obtenidas mediante adquisición tisular guiada por USE confirmaron un hamartoma de glándulas de Brunner. La lesión fue resecada endoscópicamente con un asa caliente, y la pieza fue recuperada por vía transoral. Después de la resección del pólipo, los síntomas de la paciente se resolvieron por completo. En un paciente con episodios recurrentes de vómitos posprandiales y dolor abdominal superior, en presencia de una lesión subepitelial duodenal, ya sea pediculada o sésil, debe descartarse un hamartoma de glándulas de Brunner. La adquisición tisular guiada por USE puede confirmar el diagnóstico, y la resección endoscópica es factible en la mayoría de los casos.
Palabras clave: *Glándulas de Brünner; Hamartoma; Obstrucción de la Salida Gástrica; Duodeno; Endoscopía Gastrointestinal (source: MeSH NLM).*

Correspondence:
Carlos Eduardo Oliveira dos Santos
Department of Gastroenterology and Digestive Endoscopy, Santa Casa Hospital, Porto Alegre, Brazil.
E-mail: ddendo@uol.com.br

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INTRODUCTION

Brunner's glands are exocrine mucin-secreting glands normally located in the deep mucosa and submucosa, mainly in the first and second portions of the duodenum [1]. A Brunner's gland proliferative lesion > 5 mm in size is called hamartoma [2]. Brunner's gland hamartoma (BGH) is an uncommon benign lesion, accounting for 5-10% of all benign duodenal lesions and less than 1% of all gastrointestinal tumors [3,4]. These lesions are mostly asymptomatic, but large hamartomas can present nonspecific symptoms and, rarely, lead to serious complications, such as gastric outlet obstruction [4].

Herein, we describe the case of a large duodenal Brunner's gland hamartoma, which was admitted with gastric outlet obstruction, confirmed by Endoscopic Ultrasound (EUS)-guided tissue acquisition, and successfully resected endoscopically.

CASE REPORT

A 59-year-old female presented at our hospital with a history of recurrent episodes of postprandial vomiting, upper abdominal pain, and epigastric dullness. The general physical examination was unremarkable. Her vital signs were normal, and the blood parameters were within normal range.

Esophagogastroduodenoscopy revealed a 4-cm multilobulated pedunculated polyp with a smooth surface on the posterior wall of the duodenal bulb. The polyp was freely mobile, moving back and forth through the pylorus with peristalsis, and causing intermittent pyloric obstruction (Figure 1 A-C). Endoscopic biopsies were non-diagnostic.

Linear endosonography showed a homogeneous hypoechoic solid lesion, with no cystic spaces, arising from the submucosal layer, with regular borders, measuring 3.8 x 1.8 cm, with a thick and long pedicle, and presenting a feeding artery in the base (Figure 1 D-E). EUS-guided tissue acquisition using a 22-gauge Franseen needle with a single needle pass and multiple to-and-fro movements was performed. Tissue cores obtained were processed as cell blocks for histologic evaluation. Hematoxylin-eosin-stained sections demonstrated a benign neoplasm composed of duodenal submucosal Brunner's glands lined by epithelial cells with bands of fibrous tissue, suggestive of a Brunner's gland hamartoma.

Resection of the lesion was performed with a hot snare, transecting the stalk using coagulation current for approximately 30 seconds. As the stalk could be easily snared, mechanical hemostasis or submucosal epinephrine injection were not used prior to the resection, despite the presence of vessels in the base of the lesion. No bleeding was observed after the procedure, and the specimen could be retrieved *en bloc* transorally (Figure 1F).

Histopathology of both EUS-guided tissue acquisition samples and resection specimen demonstrated hyperplasia of non-dysplastic Brunner's glands with bands of fibrous tissue, suggestive of Brunner's gland hamartoma (Figure 2). The lesion was confined exclusively to the submucosal layer, with no extension into the muscularis propria.

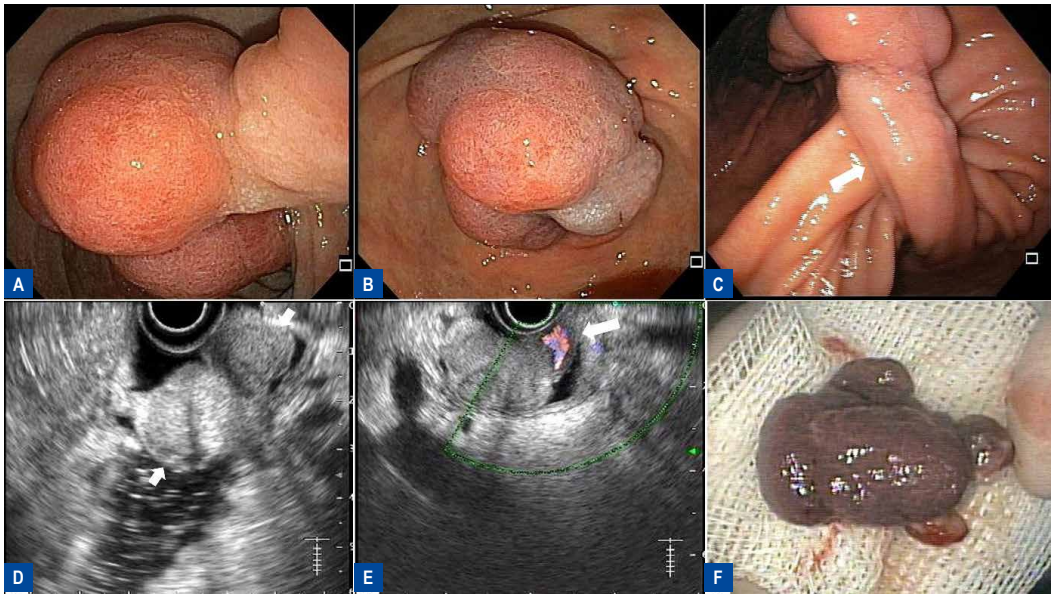

Figure 1. (A) Esophagogastroduodenoscopy showed a multilobulated, pedunculated, large polyp in the posterior wall of the duodenal bulb. **(B)** The duodenal polyp retrogradely prolapsed through the pylorus to the antrum, with congested mucosal surface, and causing a near-complete gastric outlet obstruction. **(C)** A bulky and long stalk prolapsed through the pylorus (arrow). **(D)** Linear endosonography revealed a homogeneous hypoechogenic lesion compromising the submucosal layer (arrows). **(E)** A feeding artery was detected in the base of the lesion (arrow). **(F)** Endoscopically resected specimen of 3.7 x 3.0 x 1.5 cm in size.

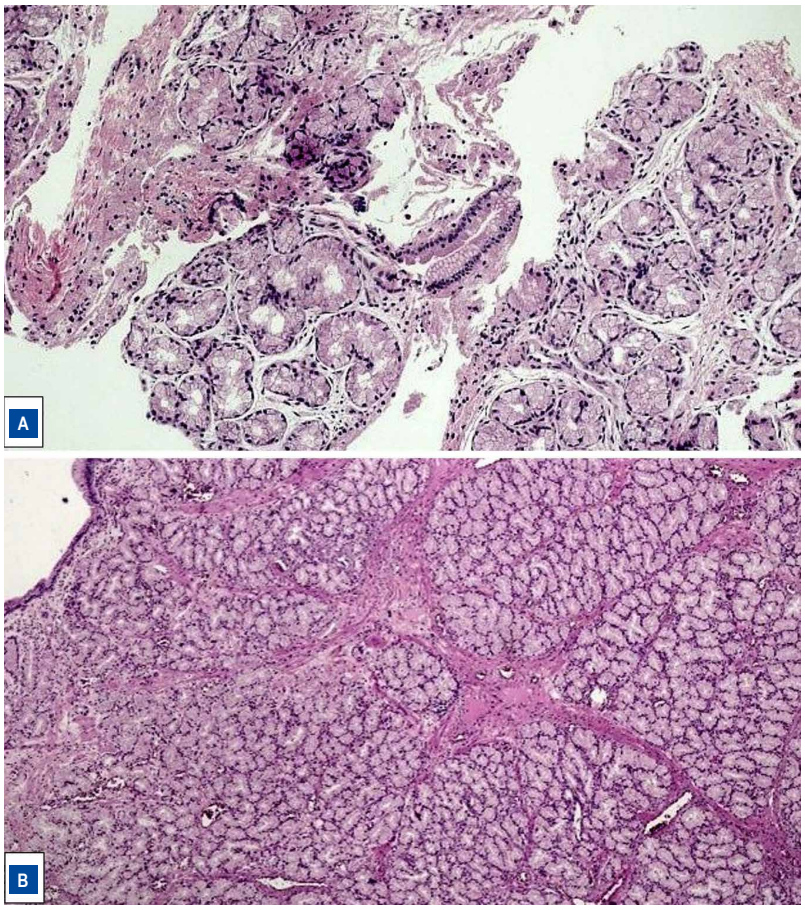

Figure 2. (A) Histopathology of EUS-FNB tissue samples revealed non-dysplastic duodenal submucosal Brunner's glands lined by cuboidal to columnar epithelial cells (H&E; x200), suggestive of Brunner's gland hamartoma. **(B)** Resected specimen confirmed a well-circumscribed lesion composed of massive hyperplastic Brunner's glands in a fibrous stroma, with no cytological atypia (H&E; x40).
EUS-FNB – Endoscopic ultrasound guided fine needle biopsy.

The patient was discharged the same day of the endoscopic procedure with no adverse events. After polyp removal, the patient's symptoms resolved completely. The follow-up endoscopy after 2 months demonstrated no residual tumor.

DISCUSSION

Brunner's glands are exocrine mucin-secreting acinar glands, located in the deep mucosa and especially submucosa. The duodenal bulb (70%) is the most common location of these glands [2], and their presence decreases caudally, being rarely found in the jejunum [1]. The alkaline mucous secreted from these glands protects the duodenal epithelium from the gastric acid and guarantees an alkaline environment for the best activity of the intestinal enzymes [4,5].

A Brunner's gland proliferative lesion, solitary and larger than 5 mm is called hamartoma [2]. A BGH is an uncommon benign subepithelial tumor of the duodenum [1], representing 5-10% of all benign duodenal tumors and less than 1% of the primary tumors of the small intestine [3]. The hamartomas usually occur between the fifth and sixth decades of life, without any gender or race preference. Lesions are usually identified incidentally as pedunculated or sessile lesions during upper digestive endoscopy or imaging studies performed for diverse reasons [1,5]. The pathogenesis of this uncontrolled hamartomatous growth remains unclear, though some risk factors such as *H. pylori* infection and chronic damage to the duodenal mucosa have been postulated [4].

Most of BGHs are asymptomatic and usually < 2 cm. However, large sized hamartomas (≥ 2 cm) may present nonspecific clinical manifestations, such as chronic abdominal pain, nausea, vomiting, iron-deficiency anemia, and more serious acute complications, such as digestive bleeding and gastric outlet or duodenal obstruction [1,4,6]. In a meta-analysis by Joseph *et al.* [6] analysing 93 patients

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with BGH, the most common presenting symptom was abdominal pain, which occurred in 56.6% of patients. Melena was found in 53% of patients.

Brunner's gland hamartoma are more usually a solitary pedunculated polyp, and the average lesion size is 4.2 cm (range: 0.5-12 cm) for symptomatic patients [6,7], almost the same size observed in our case. However, endoscopic appearance alone is not characteristic for BGHs, and conventional endoscopic biopsies are often non-diagnostic because these lesions are deeply located in the mucosa and submucosa. Therefore, BGH should be included in the differential diagnosis of other subepithelial duodenal lesions, such as gastrointestinal stromal tumors, neuroendocrine tumors, lymphomas, pancreatic rests, lipomas, and duplication cysts [2,4]. At this point, the information obtained by endoscopic ultrasound may be useful to narrow the number of differential diagnoses. More often, BGH is a hypo or hyperechoic lesion depending on the amount of fibromuscular tissues in the stroma, with heterogeneous echogenicity and indistinct borders, containing small or large cysts, and arising from deep mucosa or submucosa layer [8,9]. In addition, the presence of blood vessels within the stalk of pedunculated lesions may be easily evaluated [9], as observed in our case.

Although the information provided by imaging studies is sufficient for planning the therapeutic approach in most cases, deep tissue samples for histopathology are essential to confirm the diagnosis of BGHs. In addition to imaging data, endoscopic ultrasound can also guide tissue acquisition for histopathologic evaluation [10]. In our case, the BGH presented as homogeneous hypoechoic submucosal lesion with well-circumscribed borders and no cysts. In spite of a ultrasonographic appearance not characteristic, endoscopic ultrasound-guided tissue acquisition was paramount to confirm the diagnosis of a BGH prior to resection.

Histologically, the hamartomas are frequently composed of hyperplastic Brunner's glands with some cystic dilatation, fibromuscular septae, and a variable mixture of adipose tissue, lymphoid aggregates, and pancreatic heterotopic elements [1,2]. Malignant transformation of Brunner gland's hamartomas is exceptionally rare. In a study of 722 Brunner gland's hyperplasia specimens, Sakurai *et al.* [11] observed dysplasia in 2.1%, and invasive adenocarcinoma in 0.3% of cases. Both cases of adenocarcinoma were found in flat elevated lesions with central depression. For polypoid lesions, there was no case of cancer, but only 2 cases of low-grade dysplasia and a single case of high-grade dysplasia.

The vast majority of BGHs are managed conservatively because the lesions are small, asymptomatic, and their neoplastic potential is very low. However, symptomatic and large lesions should be removed, which can be done successfully endoscopically or surgically [3,6,12]. The best treatment for these lesions has not been established because the experience is restricted to a few number of published case reports and small case series [4,13]. Endoscopic management has been the preferred therapeutic approach over the surgery, especially for accessible and pedunculated lesions, because it is an effective and less-invasive procedure with fewer adverse events [12-14]. For lesions with thick stalks, pretreatment of the stalk with endoloop or clip for bleeding prevention may be recommended.

In regard to the effectiveness of endoscopic treatment for BGH, the literature is scarce. Concerning the most common neoplastic lesions of duodenum, in a meta-analysis with 485 nonampullary duodenal adenomas from 14 studies [15], endoscopic mucosal resection (EMR) was successfully accomplished in 93% of the cases. However, most lesions were sessile (92%) and located in the second portion of the duodenum (80%), with a mean size smaller than that of symptomatic BGHs, ranging from 13mm to 35mm. The intraprocedural and delayed bleeding rates were, respectivelly, 16% and 5%. Perforation occurred in 1% of the cases. Our case was a pedunculated lesion, similar to solely 8% of the cases of this series. Nonetheless, despite good results of EMR or polipectomy for adenomatous lesions, there is still no consensus about the best endoscopic therapeutic approach and periodic surveillance after endoscopic resection for BGHs.

Surgery for BGH should be reserved for emergency complications, large sessile lesions, pedunculated lesions with a thick stalk, when a malignancy cannot be ruled out, or in case of endoscopic interventions failure [13]. Most common surgical interventions include duodenotomy with resection, or more invasive procedures, such as duodenectomy, or even pancreaticoduodenectomy. Outcomes are excellent with no reported recurrences after complete surgical resection.

In our reported case, a Brunner's gland hamartoma causing gastric outlet obstruction, once confirmed by EUS-guided tissue acquisition, could be successfully resected by endoscopic polypectomy.

Brunner's gland hamartoma should be considered in patients presenting with recurrent postprandial vomiting and a duodenal subepithelial lesion. EUS-guided tissue acquisition may facilitate the diagnosis, and endoscopic resection is feasible for most cases.

Ethical considerations

Informed consent was obtained from the patient for the publication of her information and images.

Declaration of use of generative AI and AI-Assisted technologies in writing

The authors declare that no generative artificial intelligence (AI) or AI-assisted technologies were used in the writing, analysis, or preparation of this manuscript. All content was conceived, drafted, and critically reviewed by the authors, ensuring the originality, accuracy, and integrity of the work.

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