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Case Report

Diagnosis and Surgical Management of Brunner's Gland Hamartoma of the Duodenum: Case Report

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Abstract

We present a clinical case of a 27-year-old male patient who presented with symptoms of intestinal obstruction of approximately one month's duration. He sought emergency care with abdominal pain described as burning, moderately intense, located in the epigastrium, intermittent, aggravated by food intake, and associated with abdominal distension and oral intolerance. Radiological studies were performed, and an upper endoscopy revealed an exophytic mass with inflammatory characteristics obstructing the passage to the second portion of the duodenum. Additionally, a contrast-enhanced computed tomography of the abdomen was conducted, which showed an exophytic mass at the same level and a polypoid lesion in the jejunum.

Due to the clinical and imaging characteristics, the patient was scheduled for surgery, during which the tumor was resected. His postoperative course was uneventful, and he was discharged home after 12 days. In this article, we present our diagnostic and therapeutic approach to a Brunner's Gland Hamartoma in the duodenum.

Keywords: Hamartoma; Brunner's gland; Management; Diagnosis; Uncommon; Surgical resection; Diagnosis approach

Introduction

A duodenal Brunner's Gland Hamartoma (BGH) is a highly uncommon pathology, accounting for approximately 10% of benign duodenal neoplasms, typically presenting in a pedunculated form. Most of these lesions are found in the duodenal bulb, accounting for up to 57%, and their frequency decreases as we move towards the more distal portions of the duodenum [1]. Typically, they are asymptomatic and discovered incidentally during an endoscopic examination. However, in some cases, when they grow to a considerable size, referred to as giant hamartomas, measuring more than 5 cm, they can lead to symptoms, primarily abdominal pain, nausea, vomiting, hematochezia or melena, obstruction, and in rare instances, even intussusception.

Up to 57% of these lesions are found in the duodenal bulb, and their frequency decreases as we move towards the more distal portions of the duodenum [1]. They are often asymptomatic and discovered incidentally during an endoscopic examination. Nevertheless, when they grow to a considerable size, referred to as giant hamartomas, measuring more than 5 cm they can cause abdominal pain, nausea, vomiting, hematochezia, obstruction and, in rare instances, intussusception. When symptomatic, treatment options include resection using endoscopic or surgical approaches. In the following section, we will discuss a clinical case in which we can highlight the clinical characteristics mentioned earlier. In meticulous detail, we will describe our approach to reach a diagnosis and subsequently outline the treatment plan, along with the histopathological findings. This is a case of a Brunner's gland hamartoma, which was surgically removed due to obstructive symptoms and apparent signs of intussusception on abdominal tomography. Subsequently, the patient exhibited a favorable postoperative course without complications.

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Case Presentation

A 27-year-old male patient with no significant medical history presented with symptoms that had been ongoing for one month prior to admission. He described insidious, burning, moderately intense abdominal pain located in the epigastrium. The pain was intermittent and worsened after meals, with no alleviating factors, and was accompanied by abdominal distension. The patient denied experiencing fever, asthenia, adynamia, or any other associated symptoms. The after mentioned symptomatology persisted and progressed, with an increase in the intensity and frequency of the pain, along with the onset of nausea that progressed to vomiting of gastric contents on multiple occasions. Consequently, the patient sought evaluation and management.

Upon physical examination, vital signs within normal range, the patient was neurologically intact, with no abnormalities noted in the cardiovascular or pulmonary systems. Abdominal examination revealed a distended abdomen, reduced peristalsis, soft, depressible, and tender to palpation in the epigastrium, with no signs of peritoneal irritation. Laboratory tests were conducted, and among the results, relevant findings included iron-deficiency anemia, which has been associated with this type of pathology, with the following parameters: Hemoglobin at 12.5 g/dL, Mean Corpuscular Volume (MCV) at 78.7 fL, Mean Corpuscular Hemoglobin (MCH) at 25.4 pg, white blood cell count at 10,520 cells/mm^3, with absolute neutrophil count at 7,280 cells/mm^3, and a platelet count of 529,000/mm^3. An amylase test was performed to explore differential diagnoses, yielding normal results

at 57 U/L. Initially, an upper abdominal ultrasound was conducted, which revealed only moderate hepatic steatosis, with no evidence of other abnormalities. Hence, an upper gastrointestinal endoscopy was performed, which reported an extrinsic inflammatory process of undetermined origin, causing an obstruction in the passage to the second portion of the duodenum, along with an apparent polyp at the location of the Vater's ampulla (Figure 1). As part of the diagnostic approach, a contrast-enhanced abdominal CT scan was requested, revealing thread-like passage at the level of the second and third portions of the duodenum, attributed to mucosal thickening. Additionally, a polypoid lesion extending into the jejunum was identified, characterized by heterogeneity with negative densities, with intussusception at this level considered likely (Figure 2).

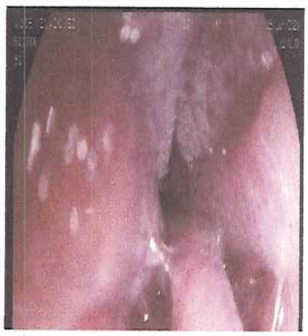

Figure 1: An extrinsic inflammatory process of undetermined origin obstructing the passage to the second portion of the duodenum, with an apparent polyp at the location of the Vater's ampulla.

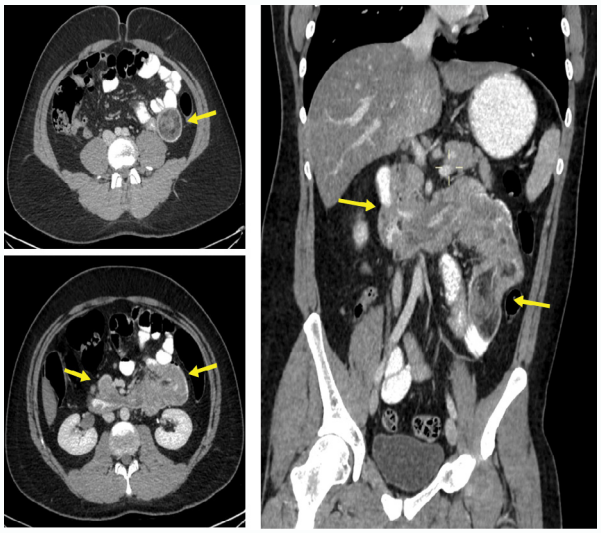

Figure 2: Contrast-enhanced abdominal CT scan revealed a thread-like passage of contrast material at the level of the second and third portions of the duodenum due to mucosal thickening. An apparent polypoid lesion extending into the jejunum was observed, displaying heterogeneity with negative densities, suggesting a possible intussusception.

In light of the previously described findings and the persistence of symptoms, the decision was made to schedule the patient for surgery. A diagnostic laparoscopy was initially performed, revealing a significantly dilated small intestine indicative of a probable chronic obstructive process. However, no masses or intussusception were evident. Therefore, the procedure was converted to an exploratory laparotomy. During the laparotomy, a tumor was located in the proximal jejunum, approximately 30 cm from the ligament of Treitz. An enterotomy was performed at this site, revealing a tumor originating from the mucosa and submucosa. Additionally, a lymph node measuring 4 cm × 3 cm was found, dissected, and sent for intraoperative biopsy, which indicated a probable low-grade lymphoma (Figure 3).

Considering a high suspicion of a duodenal origin for the tumor, the retroperitoneum was approached. Further inspection of the duodenum revealed retraction of its second portion. An incision at this site to create a duodenotomy evidenced a tumor measuring approximately 10 cm × 5 cm dependent of the duodenum, with areas of caseous necrosis (Figure 4).

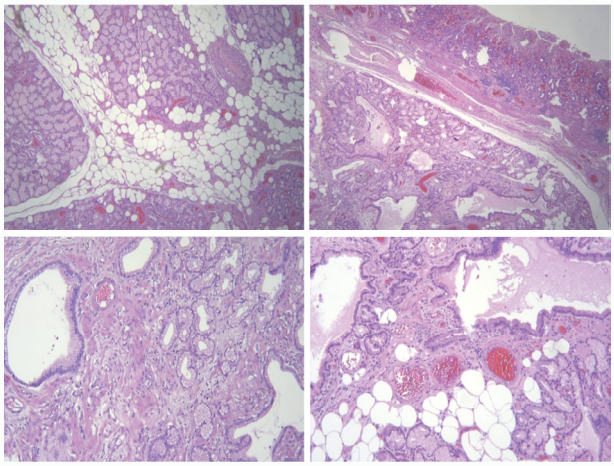

Figure 3: "The lesion consists of a proliferation of mucous glands lined by clear cells with vacuolated cytoplasm and basal nuclei. No atypia is noted. These glands are arranged in a lobular pattern with some glandular dilation. Large ducts are observed, along with lobules of mature adipose tissue. The lesion is well-demarcated with viable resection margins. No evidence of dysplasia or malignancy is observed".

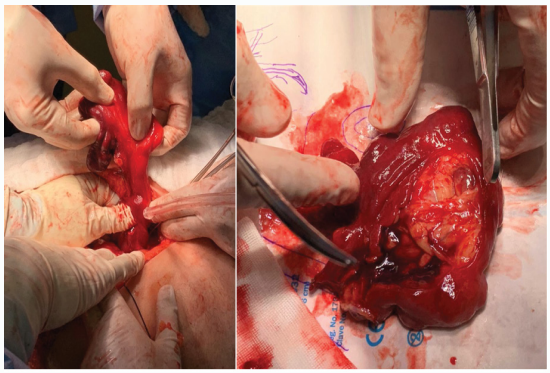

Figure 4: Tumor originating from the duodenum, with areas of caseous tissue and necrosis, measuring approximately 10 cm × 5 cm.

The tumor was resected, and subsequent closure of the enterotomies was performed in two layers using Vicryl 3-0, employing a continuous Connell-Mayo pattern followed by Lembert pattern with Silk 3-0. Fibrin adhesive (Tisseel) was applied over the anastomoses. Two Blake-type drains were placed, one on the right side directed toward the duodenal enterotomy, and the other on the left side directed toward the jejunal enterotomy. The laparotomy incision was closed with Stratafix 2-0 sutures, and subcutaneous tissue was

closed with Vicryl 3-0. Finally, the skin was closed with staples. An intraoperative endoscopy was performed, confirming the enterotomy site with well-approximated edges, no signs of active bleeding, and a pneumatic test was conducted, revealing no evidence of leakage.

The patient remained stable with a favorable progression. He remained on fasting for 5 days, after which a diet was initiated, gradually progressing from clear liquids to a normal diet. The patient was discharged after a 12-day hospital stay without complications. In the end, a curative resection of the lesion was achieved, as mentioned in the literature. Treatment for this type of pathology, when symptomatic, is definitive, with no recurrence or progression to malignancy reported. During follow-up, the patient has remained stable, free from recurrent symptoms, and has shown excellent progress. The definitive pathology report confirmed a Brunner's gland hamartoma (measuring 7.1 cm × 3.1 cm × 1.6 cm) and identified a lymph node with lymphoid follicular hyperplasia.

Discussion

Regarding anatomy, the duodenum is the initial segment of the small intestine and is considered retroperitoneal, apart from the duodenal bulb. It has an approximate length of 25 cm and assumes a 'C' shape, extending from the pyloric sphincter to the ligament of Treitz. The duodenum is divided into four segments: the duodenal bulb, the descending portion, the horizontal portion, and finally, the ascending portion. These lesions typically present as a pedunculated polyp or mass with normal mucosa on its surface. Their distribution along the small intestine decreases with progression, starting at 70% in the proximal portion and decreasing in frequency throughout the rest of the intestine. They are found in 57% of cases in the duodenal bulb, 27% in the second portion, and 5% in the third portion [2].

Brunner's glands were first described in 1688 by the Swiss anatomist Conrad Brunner [3]. They are primarily located in the submucosa of the duodenum, decreasing in number towards the ampulla of Vater. These glands secrete alkaline fluid, which help protect the duodenal epithelium from erosion by gastric acid [4].

Among the benign lesions of Brunner's glands, hyperplastic nodules or polyps and hamartomas are commonly encountered [5]. Some define lesions smaller than 2 cm as hyperplastic nodules or polyps, whereas lesions larger than 2 cm are termed hamartomas [6]. They represent 5 to 10% of benign tumors that can be found in the duodenum, with an incidence of less than 0.01%. A hamartoma is characterized by an abnormal proliferation of Brunner's glands, along with a mixture of adipose tissue, fibrous septa, smooth muscle, and other components of normal tissue [7-9]. The exact etiology of these lesions remains unclear; however, chronic pancreatitis and Helicobacter pylori infection are believed to play a role in their presentation [4,10]. In a study of 19,000 individuals, 5 out of 7 who were diagnosed with Brunner's gland hamartoma also had *H. pylori* infection [11].

The nomenclature can appear confusing when exploring the literature. From a histopathological perspective, some consider a lesion smaller than 5 mm in size, whether solitary or multiple, to be hyperplasia, while lesions larger than this size is labeled as hamartomas. This differentiation is important as size is a parameter considered when determining the type of management, as larger solitary lesions are more likely to cause symptoms [12]. These types of lesions are frequently encountered as incidental findings during upper gastrointestinal endoscopy. In the case of hamartomas, which are uncommon benign tumors of the duodenum, endoscopy typically reveals a submucosal mass that may have a peduncle [12]. They are generally asymptomatic but can present with symptoms such as bleeding, intestinal obstruction, or, insidiously, with symptoms like dyspepsia or anemia [12-14].

Gastrointestinal symptoms that may occur include abdominal pain as an initial symptom, abdominal distension, diarrhea, and hematochezia [14]. In a study of 27 cases, 37% presented with bleeding, 37% with intestinal obstruction, and the rest were incidental findings [9]. When conducting imaging studies such as CT scans, hyperplasia of Brunner's glands is commonly found as solitary or multiple lesions smaller than 5 mm. Hamartomas, on the other hand, tend to be larger, typically averaging around 2 cm in size, with internal cysts in one-third of cases. Most often, they appear as isoattenuating images compared to the pancreas on non-contrast CT scans in up to 83% of cases and as hypoattenuating images following the administration of iodinated contrast in half of the cases. A peripheral enhancing rim around the lesion can sometimes be observed during the arterial phase in two-thirds of the studies [15].

In some studies, both ultrasound and CT scans have been reported to have diagnostic rates of up to 100% [14]. When performing magnetic resonance imaging, hamartomas may display internal cystic-appearing images that are hyperintense on T2-weighted sequences [16]. Treatment for asymptomatic cases is typically conservative, while symptomatic cases are managed through resection, either endoscopically for small lesions or surgically for larger ones, especially those >5 cm, when symptomatic. Surgical intervention is commonly recommended for cases causing bleeding, obstruction, and in rare instances, intussusception [1,17]. Recurrence has been described very rarely in the literature, and it is not considered a malignant condition, thus providing an excellent long-term prognosis.

Methods

This case report has been reported in line with the SCARE Criteria [18].

Conclusion

In this case, we can conclude that, despite being a rare cause, we should consider such lesions when addressing this type of symptomatology or during the approach of anemia of unclear origin. This condition may produce symptoms or warrant monitoring if incidentally discovered and asymptomatic to prevent complications that may hinder appropriate treatment. It is essential to be aware of this pathology to avoid misdiagnosis since it is not commonly considered as one of the primary differential diagnoses when evaluating a patient with such characteristics. Furthermore, there have been no reported cases of malignancy following treatment, and recurrence is exceedingly rare.

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