Case report

Brunner's Gland Hamartoma of the Duodenum as a Cause of Intestinal Obstruction in a Young Patient : A Surgical Approach

Abstract :

The Brunner's gland hamartoma is a benign tumor of the duodenum, but it has malignant potential with a very low risk of progressing to adenocarcinoma. It is rare, accounting for less than 1% of primary tumors of the small intestine. Furthermore, its clinical manifestations are nonspecific, its etiology is poorly understood, and the treatment strategy needs to be tailored accordingly. We report a case of a patient with a Brunner's gland hamartoma of the duodenum who presented with intestinal obstruction and was treated with surgical resection of the polyp via duodenotomy.

Keywords: Brunner's gland; Duodenum; Hamartoma; Hyperplasia

1. Introduction :

« The Brunner's glands, described by the anatomist Brunner in 1688, are submucosal mucous glands that secrete mucin » [1]. « Hyperplasia and hamartoma of Brunner's glands are benign proliferative lesions of the duodenum. Hyperplasia of Brunner's glands is typically a lesion less than 0.5 cm, characterized by neutral mucin-secreting glands occupying at least 50.0% of the duodenal mucosa in a biopsy specimen » [2, 3]. In contrast, hamartoma of Brunner's glands, also known as Brunner gland adenoma, is generally a lesion larger than 0.5 cm [4, 5], involving proliferative glands within the duodenal submucosa, mixed with dilated cystic glands and smooth muscle proliferation [6]. The fundamental difference lies in the mixture of other benign components (smooth muscle fibers) with the glands to qualify as an "hamartoma".

« The majority of Brunner gland hamartomas are isolated pedunculated polyps, with a minority being sessile. Their diameter ranges from 1.0 to 2.0 cm, rarely exceeding 5.0 cm, and up to 12.0 cm in some cases » [7, 8]. « Most lesions are located in the proximal duodenum, with occurrence decreasing with distance from the pyloric ring: 57.0% in the duodenal bulb, 27.0% in the descending part, and 7.0% in the horizontal part » [9].

Brunner gland hamartoma is an extremely rare tumor, with an estimated incidence of 0.008% in a single series of 215,000 autopsies [1].

In this article, we report a clinical case of a patient with a Brunner gland hamartoma of the duodenum who presented with intestinal obstruction and was treated with surgical resection of the polyp via duodenotomy.

2. Case presentation :

He is a 22-year-old young man with no notable medical history. He was admitted with high intestinal obstruction without signs of gastrointestinal bleeding.

Clinical examination and laboratory tests were normal. An abdominal computed tomography (CT) scan showed a tissue lesion in the bulboduodenal region measuring 57mm x 34mm x 22mm, which enhanced moderately after injection of iodinated contrast.

The patient was taken to the operating room after preparation. During exploratory surgery, a duodenal polyp was identified (Figure 1). The patient underwent surgical resection of the polyp via duodenotomy (Figure 2 and 3). Postoperative recovery was uneventful.

Histopathological examination of the surgical specimen revealed a hamartoma of the Brunner's glands, measuring 4.5 cm.

During a 6-month follow-up, the patient remained asymptomatic. A follow-up esophagogastroduodenoscopy showed no residual lesions.

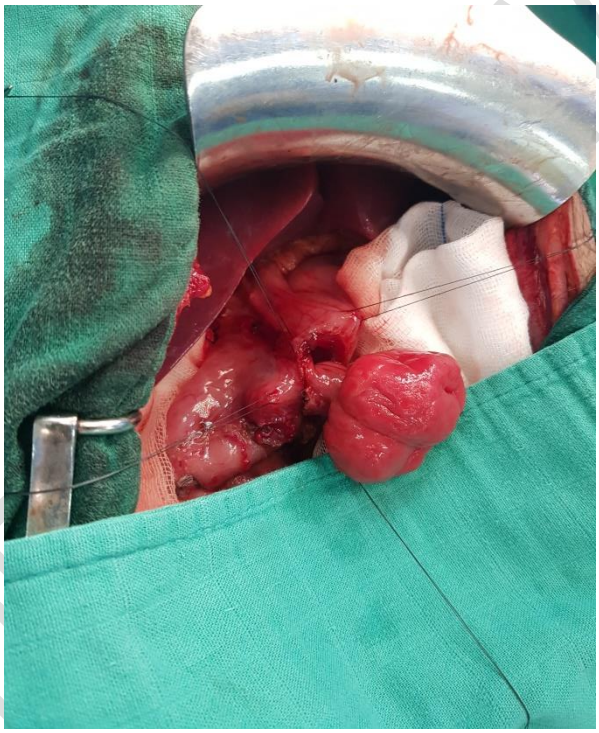

Fig.1 : Operative view of duodenal polyp.

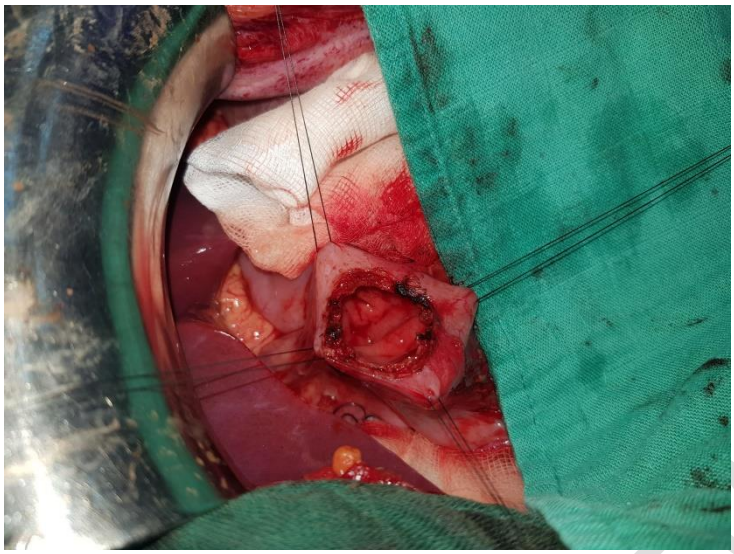

Fig.2 : View after resection of duodenal polyp.

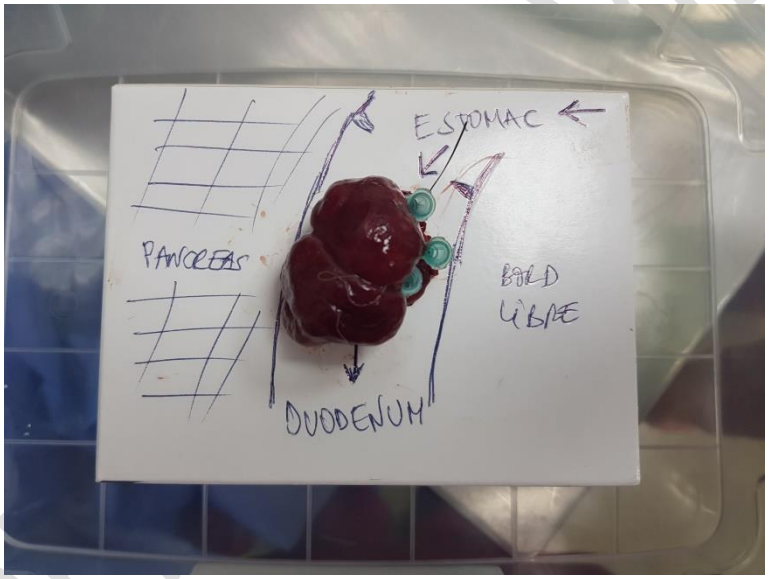

Fig.3 : Surgical specimen

3. Discussion :

Brunner's gland hamartoma tends to be predominant in the fifth or sixth decade of life with an equal distribution between sexes [7]. The clinical presentation varies; however, the majority of cases are asymptomatic or manifest with nonspecific and vague symptoms such as abdominal pain, nausea, or bloating.

In symptomatic patients, the most common clinical manifestations are gastrointestinal bleeding (37%) and obstructive symptoms (37%) [7]. Gastrointestinal bleeding most often presents as chronic blood loss leading to iron deficiency and anemia [5]. Less frequently, when erosions or ulcerations of the tumor occur, patients may present with melena or hematemesis. These observations are typically described in Brunner's gland hamartomas located beyond the first portion of the duodenum, likely

because these lesions are subject to more stress and vascular damage due to gastrointestinal motility [5]. In our case, the patient was admitted for intestinal obstruction.

The etiology and pathogenesis of Brunner's gland hamartoma remain unclear. Due to the "anti-acid" function of Brunner's glands, increased acid secretion has been postulated to stimulate these structures to undergo hyperplasia [3]. Franzin et al. [6] reported an association between Brunner's gland hamartoma and hyperchlorhydria in patients with chronic gastric erosions and duodenal ulcers, but Spellberg et al. [8] did not observe regression of the lesion with acid secretion inhibitors [8]. A second hypothesis suggests an inflammatory origin due to dense infiltration of inflammatory cells [10]. Since lymphocytes are usually present in the normal submucosa of the intestinal tract, the presence of inflammatory foci in Brunner's gland hamartoma is not sufficient to support the inflammatory hypothesis. Finally, it has been suggested that H. pylori infection could play a role in the pathogenesis of Brunner's gland hamartoma. In a recent study of 19,100 subjects, H. pylori infection was found in five out of seven cases of Brunner's gland hamartoma (71%) [11]. In our patient, no H. pylori infection was found. The extremely rare occurrence of Brunner's gland hamartoma and the high prevalence of H. pylori infection in the general population do not allow us to establish a clear pathogenic link.

The duodenal bulb is the most frequent location of Brunner's gland hamartoma (57%) [12]. In most cases, these lesions develop into a polypoid mass, usually pedunculated (88%), measuring between 1 and 2 cm [13].

« Diagnosis is not always straightforward. Radiological findings are often non specific [14]. Indeed, duodenal filling defects can mimic several other lesions such as leiomyoma, lipoma, lymphoma, aberrant pancreatic tissue, or carcinoid tumors » [15]. « CT scan is only useful to confirm the absence of extraluminal extension of Brunner's gland hamartoma » [15].

« Diagnosis can be confirmed by histological examination of the excised mass. Traditional endoscopy with pinch biopsies is generally negative because pinch biopsies cannot reach tumor tissue located entirely in the submucosal layer » [16].

« Endoscopic or surgical removal of Brunner's gland hamartoma has been suggested to prevent the development of complications (bleeding, severe anemia, intestinal obstruction, or intussusception). Endoscopic polypectomy is the preferred approach, being more cost-effective and less invasive than abdominal surgery » [17, 18]. However, success depends on the location and size of the hamartoma as well as the presence of a pedicle. Several cases of successful endoscopic resections have been reported [14, 19-20]. In our case, the patient had a 57 mm duodenal process complicated by intestinal obstruction, so surgical treatment was necessary. Duodenotomy resection of the duodenal polyp was performed without complication, and there was no recurrence of the lesion during the 6-month follow-up.

4. Conclusion :

The Brunner's gland hamartoma is a rare benign tumor of the duodenum with nonspecific clinical manifestations. Its mechanisms remain unclear. Malignant transformation should be cautiously evaluated through histology combined with immunohistochemistry, especially if the lesion size increases and its morphology changes. An observation strategy is employed in the majority of

Brunner's gland hamartoma cases. If necessary or in cases of complications, endoscopic and/or surgical resection is required.

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Details of the AI usage are given below:

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