Kassab et al., J Clin Case Rep 2016, 6:5
DOI: 10.4172/2165-7920.1000802

Journal of Clinical Case Reports

Case Report Open Access

Giant Duodenal Hamartoma with Gastro Duodenal Intussusception Treated with Whipple Operation

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Abstract

Duodenal Brünner gland hamartomas are usually asymptomatic and benign. They are relatively rare with around 300 cases described in the literature. In general the patients present mild symptoms, sometimes with silent bleeding leading the patient to anemia. In other cases they may present an acute episode of bleeding or obstruction1. In cases of bigger tumors, occupying almost the total duodenal lumen, the endoscopic treatment is difficult.

Keywords: Duodenal Brünner gland; Silent bleeding; Anemia; Whipple operation

Introduction

Duodenal Brünner gland hamartomas are usually asymptomatic and benign. They are relatively rare with around 300 cases described in the literature. In general the patients present mild symptoms, sometimes with silent bleeding leading the patient to anemia. In other cases they may present an acute episode of bleeding or obstruction [1]. In cases of bigger tumors, occupying almost the total duodenal lumen, the endoscopic treatment is difficult. The CT scan may show a duodenal mass. The obstruction can be purely due to mechanical factors or relates to duodenal intussusception. The local endoscopic resection is, in general, the best option. Rarely the Whipple operation (pancreaticoduodenectomy) may be necessary.

Case Report

A 36 year-old woman presented symptoms of persistent vomiting and weight loss during the last year. During the physical examination it was possible to palpate a huge mass in the epigastrium. She had been seen in several medical institutions and submitted to several endoscopies that showed a big gastric polyp with hyperplastic aspect and another big duodenal mass. It was impossible for the endoscope to pass through the duodenal tumor (Figures 1 and 2). The cholangio resonance revealed a big solid mass with a diameter of 14 cm invading the duodenal wall, extending to the inferior portions of the duodenal arch, promoting duodenal and gastric dilation (Figure 3). The endoscopic resection was impossible so the surgical option was chosen with two possibilities: laparotomy with trans duodenal resection opening the duodenum, or Whipple procedure. During the surgery we noted that the mass had big size (Figure 4), and that there were big lymph nodes around the tumor, near the mesenteric vein. It was then performed the pancreaticoduodenectomy associated with distal gastrectomy including the area where the polyp was located. The reconstruction was made by an end-to-end pancreaticojejunostomy and a hepaticojejunsotomy with a Roux-en-Y and finally a gastrojejunostomy. The specimen opening revealed a huge vegetating intraluminal duodenal mass and a big gastric polyp in the antrum (Figures 5 and 6).

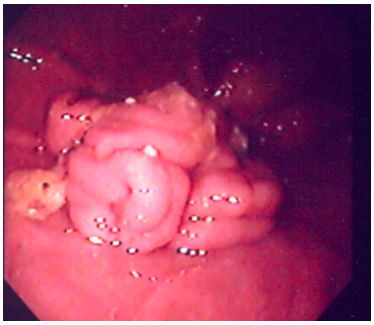

Figure 1: Endoscopy revealing a big gastric polyp.

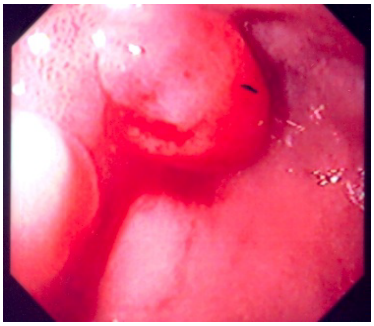

Figure 2: Endoscopy: polypoid tumor obstructing the duodenum.

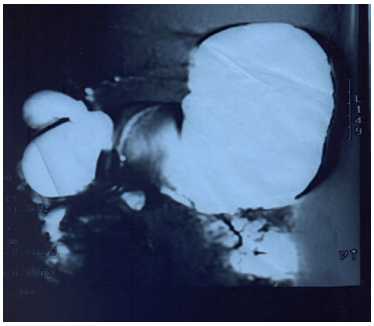

Figure 3: Magnetic resonance image.

The post-operative period was uneventful and the patient was discharged in the 11th day. The pathologic analysis showed a giant duodenal Brunner gland Hamartoma and hyperplastic gastric polyp.

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Received April 22, 2016; **Accepted** May 26, 2016; **Published** May 31, 2016

Citation: Kassab P, Ilias EJ, Castro OAP, Nara F, Thuler FR, et al. (2016) Giant Duodenal Hamartoma with Gastro Duodenal Intussusception Treated with Whipple Operation. J Clin Case Rep 6: 802. doi:10.4172/2165-7920.1000802

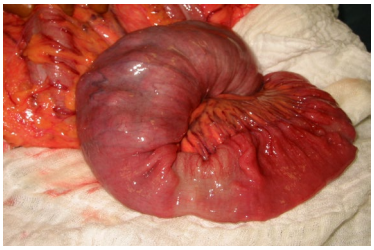

Figure 4: Intra operatory view of the tumor inside the duodenum.

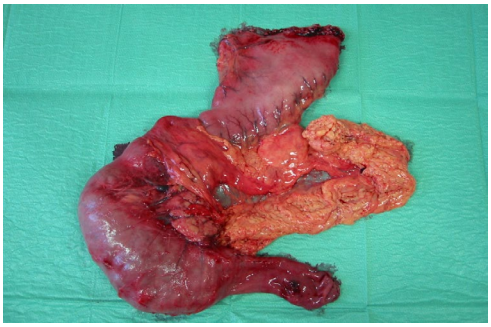

Figure 5: Specimen: stomach, pancreas head and duodenum.

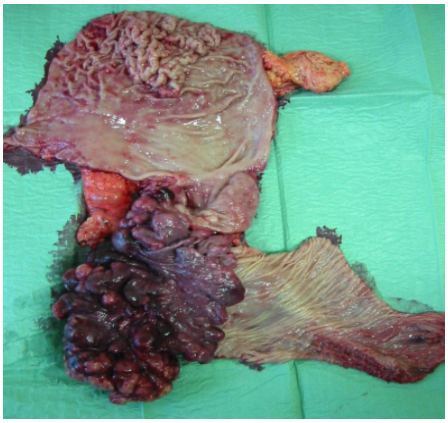

Figure 6: Specimen opened.

The margins and the lymph nodes were free. The patient is followed after 12 years and does not present any sequelae.

Discussion

Brunner glands are alkaline secreting located in the submucosal duodenal layer. They are mainly located in the first portion and decrease progressively in direction of the third duodenal portion. The duodenal Brünner gland hamartomas (BGH) follow that prevalence. In a study with 27 patients, approximately 70% were located in the first duodenal portion, 26% in the second and only 4% in the third portion. It is rare to find them in the pyloric area or in the jejunum. Their size varies between 0.5 and 12 cm, nevertheless tumors bigger than 2 cm are not common [2-5]. These lesions may be referred as BGH, because of their large size (>2cm) and the combination of mesenchymal and epithelial elements [6]. The growing of the BGH is typically polypoid and pedunculated, similar incidence among men and women with a peak in the fifth and sixth decades of life. Because most patients are asymptomatic, it is normally an occasional finding during upper GI endoscopy. In the symptomatic patients the clinical signs may include hemorrhage, obstruction as in the case we describe which presented obstruction and [2,7] chronic unperceived bleeding might cause anemia due to iron deficiency [8]. Brunner's gland hamartoma represent 5-10% of small bowel tumors [9] and among the differential diagnosis, we should include: leiomyoma, adenomas or adenocarcinomas, lymphoma, neuroendocrine tumors, heterotopic pancreas or even gastrointestinal stromal tumors (GIST). The histology shows the presence of normal tissue amidst ducts, Brunner glands, fatty and lymphoid tissue. Five out of seven cases of BGH have H. pylori infection [10] although its relation to H. pylori infection is not clear [11]. The malignant transformation of BGH is rare; nevertheless, cases of multiple foci of dysplasia have been described [12]. Those findings suggest that one should be cautious when treating this type of tumor1. The treatment options include endoscopic mucosal (EMR) or submucosal resection (ESD) to small pedunculated tumors [7,11], and surgical resection for bigger lesions [13]. The fact that the majority of BGH are benign with asymptomatic evolution, make the endoscopy eventually the best choice for the first treatment. Sometimes the surgery can be necessary when the endoscopic resection fails, in the suspicion of malignancy or even in huge tumors [1]. Because of the size, the presence and obstructive symptoms and impossibility of EMR or ESD, the patient in this report underwent pancreaticoduodenectomy associated with distal gastrectomy through laparotomy. The gastrectomy was necessary to remove the big antral polyp. Although there were enlarged lymph nodes, the malignancy was not confirmed. The Whipple procedure for BGH is rare with few cases reported in the literature and there is no report of association with synchronous gastric hyperplastic giant polyp. In all described cases as in the present, there was concern about possible malignancy, but it was not confirmed in any of the descriptions [1,14,15].

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