

## Recurrent giant Brunner's gland adenoma

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**SUMMARY: Recurrent giant Brunner's gland adenoma.**

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*Benign duodenal tumours are extremely rare, with an incidence of 0.008% among general population; those originating from Brunner's Gland represent 11% of this neoplasms. Most cases remain asymptomatic and are often diagnosed during routine endoscopic pro-*

*cedures, however their clinical presentation may be variable making resection treatment of choice in order to prevent complications. Recurrence has not been reported previously on literature.*

*59-year-old male presented to the emergency department nine months following endoscopic resection of a 2x1.5cm Brunner's gland adenoma complaining of bloating, weight loss and gastro intestinal bleeding, diagnostic approach revealed a 10x4cm mass occupying the duodenum. Tumour size did not allow for endoscopic resection and surgical removal was performed with excellent outcome and no further recurrence at 30 months.*

KEY WORDS: Endoscopic surgery - Duodenal tumours - Gastrointestinal neoplasia - Brunner's gland adenoma.

## Introduction

Benign duodenal tumours are extremely rare, with an incidence of 0.008% among general population. Broadly speaking, they are diagnosed during the fifth or sixth decades of life and have equal gender distribution (1, 2). Beneath all such neoplasms, those that originate from Brunner's gland, which is a sub mucosal alkaline mucin-secreting structure located solely in the duodenum, account barely for 11% of cases (3).

Brunner's gland adenoma's (BGA) pathogenesis is still not entirely understood. Studies have shown an association with hyperchlorhydria and peptic ulcer disease, yet they characteristically show no response to acid suppression which suggest there might be other factors involved in their development (4, 5).

Clinical presentation is quite variable, most are small asymptomatic lesions that remain undiagnosed until identified throughout routine endoscopic procedures. When symptomatic, BGA are usually larger and present with mild gastrointestinal bleeding, intermittent vomit, oral intolerance and weight loss. Also in exceptional circumstances substantial lesions may cause severe melena/hematemesis in the context of ulceration as well as acute pancreatitis and jaundice when the duodenal papilla becomes obstructed (6).

Imaging findings are non-specific in general terms, contrasted studies may show duodenal filling defects with limited sensitivity for small lesions and computed tomography is mostly useful ruling out malignancy assessing for extra luminal and lymph node invasion.

Differential diagnosis must include gastrointestinal diseases including leiomyoma, intestinal lipoma, lymphoma, aberrant pancreatic tissue and carcinoids.

Upper endoscopy is considered the gold standard for diagnosis in this context, as it allows for direct visualization, biopsy and therapeutic resection in

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most cases. BGA appears as polypoid or nodular intraluminal masses classically revealing a slit-shaped hole covered with columnar epithelium under magnified vision (7).

## Case report

A 59-year-old male with limited access to health services and no other relevant medical, familiar or social history underwent upper endoscopy after complaining of 2 months with dyspepsia, he had already received proton pump inhibitors (Omeprazole 40 mg daily) for 30 days with no relief. During the procedure a 2x1.5 cm polypoid lesion was identified on the duodenum and then resected completely allowing for patient discharge upon completion. Histopathologic analysis reported Brunner's gland

adenoma as diagnosis. The patient lost follow-up and presented 9 months later in the emergency department complaining of 3 months with progressive abdominal bloating, oral intolerance, diarrhoea, weight loss (7 kg) and two weeks of intermittent melena. Physical exam revealed pain during deep epigastric palpation, but was otherwise entirely normal.

Laboratory analysis presented normocytic normochromic anaemia with haemoglobin of 7.4 g/dl (normal range 13.5-17 g/dl) and no other abnormalities.

Upper endoscopy disclosed an ulcer in the pyloric canal as well as an anterior sub mucosal lesion in the duodenal bulb occluding more than 70% of the lumen, hindering the advancement of the endoscope (Figure 1). The extent of the lesion could not be outlined, making a safe endoscopic resection technically unfeasible, still biopsies were taken dur-

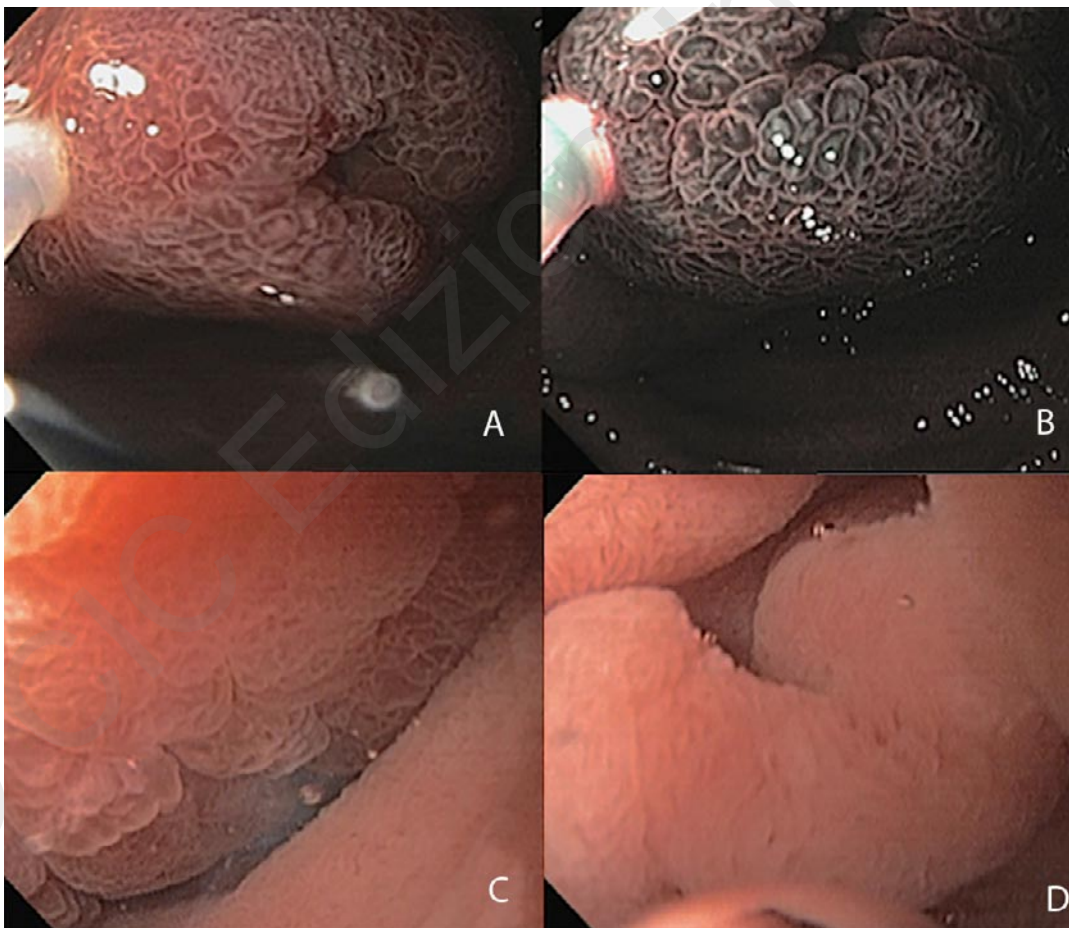


Figure 1 - (A, B) Endoscopic images showing a protruding tumour occluding more than 70% of intestinal lumen. The mucosa has a smooth surface, hyperplastic changes and a slit-shaped hole. (C) Duodenal lumen narrowing on lateral view. (D) Close-up to the slit-shaped hole.

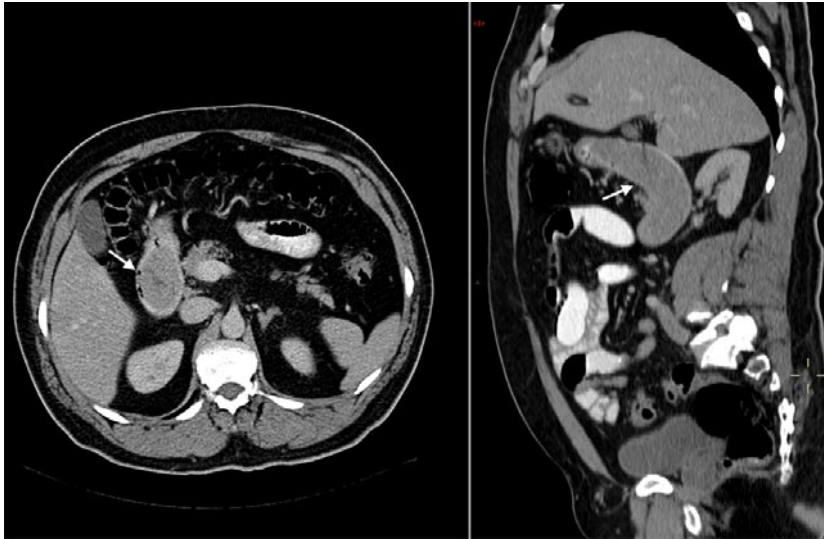


Figure 2 - Abdominal CT scan (axial left, sagittal right) showing tumour extending from duodenal bulb to the third portion (arrow). There is no invasion of the duodenal wall or surrounding abnormal lymph nodes.

ing the procedure. Computed tomography depicted an intraluminal tumour extending from the first to third portion of the duodenum and helped ruling out extraluminal invasion and locally advanced disease (Figure 2). Histopathology once again reported Brunner's gland adenoma as diagnosis.

Our patient underwent surgical resection through laparotomy during which a Kocher manoeuvre was initially performed for exposure of the pre pyloric vein followed by a longitudinal incision from the pylorus to the first portion of the duodenum. Digital exploration of the intestinal lumen revealed a 10x4x3 cm polypoid tumour which was excised completely succeeded by transverse closure of

the incision and the abdominal wall.

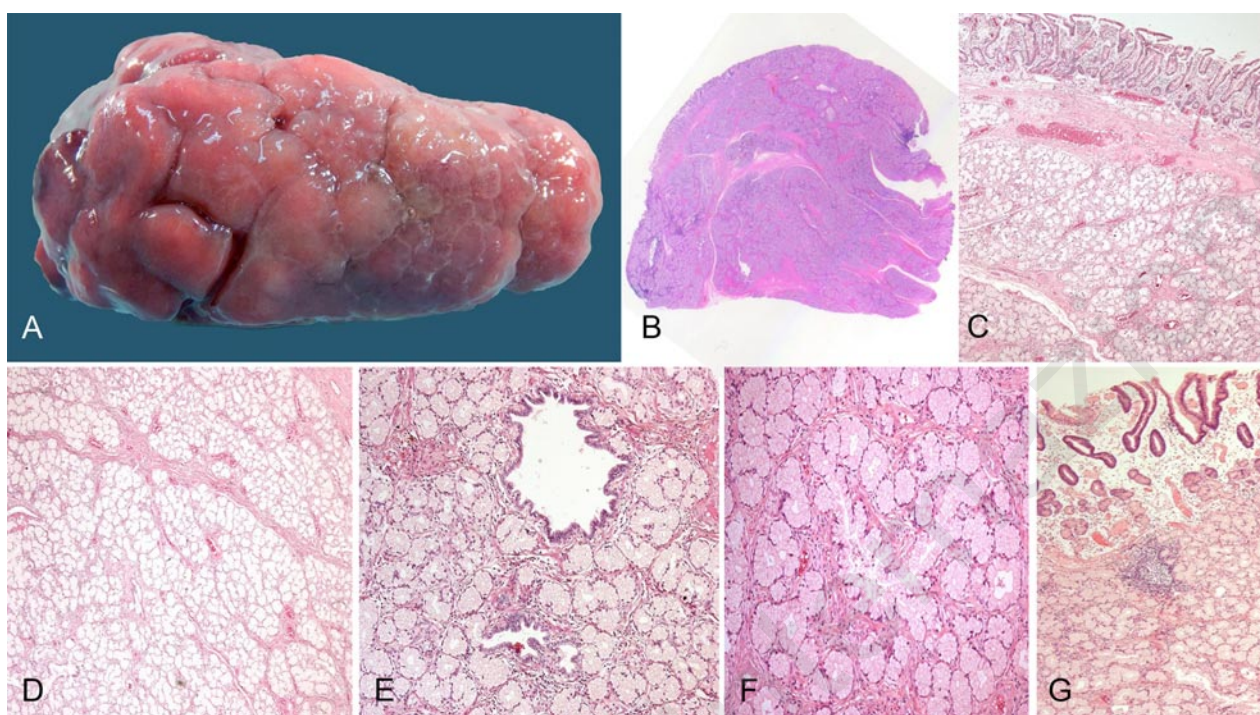
Postoperative course was uneventful, the patient was discharged home after 7 days in excellent conditions. Histological analysis confirmed the diagnosis and presented no signs of dysplasia or malignancy (Figures 3, 4). The patient received ambulatory follow-up and at 30 months there has been no further recurrence.

## Discussion

There are no specific guidelines for the management of BGA, however it is currently agreed that re-



Figure 3 - The excised polypoid lesion (10x4x3 cm).



**Figure 4 - Brunner's gland adenoma (H&E stain).** A) gross appearance of the giant polypoid lesion, 10x4 cm in size; B-C) microscopic image showing nodular proliferation of Brunner's glands in the duodenal submucosa; D) Glands with branched tubule alveolar growth with variable proliferation of stroma and smooth muscle; E- F) glands covered by epithelium without atypia, nuclei arranged towards the periphery with abundant clear cytoplasm and formation of lights in the centre, accompanied by dilated ducts; G) mild chronic inflammatory infiltrate in the periphery.

regardless the approach it should always be resected to avoid complications as bleeding or obstruction. Also there is current data supporting BGA has the potential of exceptionally evolving towards malignancy, Brookes et al. reported dysplastic changes are present in 2.1% of cases and invasive carcinoma in 0.3% (8) which must be taken into consideration when selecting a treatment plan. Considering no studies have been made regarding safety of a conservative/expectant conduct, caution must be taken when deferring treatment for any reason.

In any case, there have been no previous reports of recurrence after complete resection and in our patient neither the initial nor the second specimen showed any signs of malignant transformation on microscopic analysis. We consider this unusual presentation could help raise awareness on how important is to follow this individuals on a regular basis, especially in the context of those with poor access to health services who may be prone to under estimate their condition.

In general terms, endoscopic resection is the pre-

ferred method of treatment as most BGA are small and accessible. For lesions larger than 5 cm a surgical approach is usually required and may be performed by either laparoscopy or laparotomy (9). Endoscopic and hybrid (laparoscopic assisted) techniques for substantial tumours have been attempted in experienced centres, but results have not been evaluated on a large scale and should be performed with caution selecting patients on an individual basis (10).

#### *Acknowledgements*

We would like to thank the Pathology Department of Instituto Nacional de Ciencias Medicas y Nutrición "Salvador Zubirán" for providing insight that helped this project.

#### *Financial Disclosure:*

The Authors have no financial interest to declare in relation to the content of this article. The Article Processing Charge was paid for by the Authors.

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