

BRUNNER'S GLAND ADENOMA PRESENTING WITH GASTROINTESTINAL BLEEDING: A DISCUSSION OF WHAT WE KNOW SO FAR

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SUMMARY

Brunner's gland adenoma (BGA) is an exceedingly rare benign neoplasm of the small bowel, accounting for about 5-10% of the total benign duodenal lesion and typically found in the 5th or 6th decade of life (3,14), though there are also cases where the adenoma was discovered in children (3). It can symptomatically mimic dangerous duodenal lesion such as adenocarcinoma and normally diagnosed histopathologically (8)

Brunner's gland is typically found at the highest concentration in proximal duodenum. It secretes highly thick viscous alkaline fluid that protects the intestinal mucosa from any insult from the gastric secretions. It also produces hormone that inhibit gastric acid secretion called enterogastrone.

Brunner's gland adenoma is usually asymptomatic and occasionally manifest as duodenal obstruction and upper gastrointestinal hemorrhage (3,5). Since most patient of Brunner's gland adenoma is asymptomatic, it is usually found incidentally during endoscopies of upper gastrointestinal tract (8,12) different from our clinical case. We are reporting a 48 year old lady who presented to our clinic with persistent vomiting after meal and hematemesis secondary to Brunner's Gland Adenoma which required a polypectomy by endoscopy under sedation as a definitive treatment for her symptoms.

Keywords: Brunner's Gland, Adenoma, Case Report, Anemia, Peptic Duodenitis

1.0 CASE REPORT

A 48-year-old lady present to the surgical clinic of our institution with persistent vomiting after meal, usually occurred 30 minutes after meal for the past 2 months. The vomiting is non projectile and contained only food content. Further history taking revealed that she had 2 episodes of hematemesis 1 week prior. Otherwise, there are no other symptoms such as abdominal pain, melaena and anemic symptoms. She had a long-standing history of hypertension and currently a housewife. She denied history of smoking, drinking alcohol and taking additional medication.

Upon physical examination, there was a mild tenderness in the epigastric region but otherwise the abdomen was soft. PR examination shows no melanic stool. Other systemic reviews were normal.

Full blood count revealed a hemoglobin level to be 9.8 g/dl indicating that the patient was anemic. Otherwise, other blood investigation parameters were normal. An OGDS was performed on the patient. We discovered a large polyp at duodenum D1 with central ulcer and sign of recent bleeding from the ulcer. However, no active bleeding was noted. During the exploration, a biopsy was taken. The HPE was reported as chronic active duodenitis.

We discussed the options of treatment with patient and the patient consented for an endoscopic polypectomy under sedation. During the OGDS, an Endoloop was applied to the base of the polyp before performing the polypectomy. The polyps were sent for a histopathological examination. The histopathological results reported as an extensive Brunner's gland hyperplasia in the submucosal tissue, with no evidence of dysplasia or malignancy with a size of roughly 3cm confirming the diagnosis of Brunner's Gland Adenoma. Patient didn't experience any immediate complications and was discharged the next day of the procedure. She was asymptomatic during the follow up. She was not experiencing any hematemesis after the polypectomy.

2.0 DISCUSSION

Brunner's gland adenoma (BGA) also known as Brunner's gland hamartoma is a rare benign tumor of the duodenum (10) which was first described by Cruveilhier as the first case of benign duodenal gland adenoma in 1835. It usually presents as a single pedunculated polyp commonly located at the first part of duodenum (13) like our patient, rarely larger than 5 cm with an average size of 2 cm. They represent 5-10% of benign duodenal tumors (14) and estimated to have less than 0.01% of incident occurring (5,10,11, 14). Currently there are less than 200 papers published on BGA (12). It was also coined the term Brunner's gland hamartoma as they can also show an admixture of normal tissue including the Brunner's gland, smooth muscle and adipose tissue histologically thus elucidating the very definition of Hamartoma: which is an overgrowth of mature normal cells in an organ, composed of identical cellular elements (10). Brunner's Gland Adenoma normally manifest itself in middle age patients usually in the fifth or sixth decade of life with no gender or race prominence (1,4,7), thus correspond directly to

our patient who is a 48 years old female. There is also a report of a rare case of BGA in children (3) which confirmed that BGA is not only present in adults.

Friedrich Feyrter, an Austrian Pathologist was frequently cited by other authors (5,9) when classifying BGA. In his work (15), He describes BGA into three types: type 1, diffuse nodular hyperplasia; type 2, circumscribed nodular hyperplasia; and type 3, glandular adenoma. However, since the classification of BGA is still vehemently debated, some authors opted to classify the lesion according to size (3,5): Lesion less than 1 cm is classified as “Hyperplasia”; Lesion more than 1 cm is classified as “Adenoma”. With our patient’s lesion having the size of 3 cm, its more than appropriate to classify it as Adenoma.

It was hypothesized that hyperacidity of Upper Gastrointestinal System causes the hyperplasia of the Brunner’s glands (5,7-9,12). Akaki et al (16) particularly tried to associate Brunner’s Gland Hyperplasia with duodenal mucosal damage because they discovered that mucosal surface with ulceration damage contain Brunner’s Gland proliferation beneath it. Incidentally, Frenkel et al (4) discovered that their BGA specimen contain focal ectopic gastric mucosa as well as extensive gastric metaplasia due to chronic peptic duodenitis. However, this postulation seemed unlikely as a study concerning BGA described that 20% of their patient had gastric hypoacidity. Conversely, only 45% of the patient had increased gastric acid secretion (5,9). So far this theory partly explains BGA in our patient with chronic active duodenitis in her HPE. Nonetheless, the patient does not have any past history of complain regarding Gastritis / Duodenitis. It was also suggested that presence of helicobacter pylori may play a role in invoking the proliferation of Brunner gland (8-9,11-12). In a study done consisting of 19100 patients, it was discovered that 5 out of 7 patients diagnosed with Brunner’s Gland adenoma have concurrent Helicobacter Pylori infection (17). However, histopathological examination of our patient does not reveal the presence of Helicobacter pylori. Additionally, it is peculiar that BGA prevalence is low despite H. Pylori infection being so common in our population. Thus, there must be an additional cause that might be contributing to the pathogenesis of Brunner’s Gland Proliferation. Unfortunately, a clear correlation between BGA and these theories cannot be established as there are lack of strong evidence.



Fig. 1: Endoscopic view of the D1 duodenal mass during biopsy

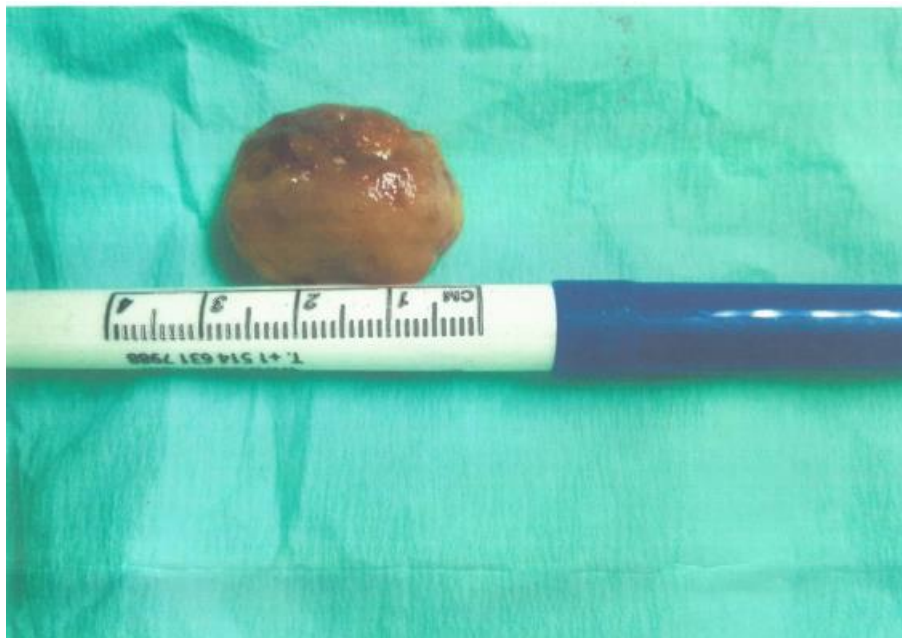


Fig. 2: Endoscopically resected specimen. The size is roughly 3 cm

Most patient that have BGA are asymptomatic. Therefore, the adenoma is often discovered incidentally during endoscopy or imaging (12). The most common clinical presentation of symptomatic patients would be Gastrointestinal Bleeding or Gastric Outlet Obstruction (6,8,11). Clinical manifestation would depend on the size, location and type of the adenoma (3). The patient will present with epigastric pain, nausea and vomiting and weight loss if the mass grows too large causing obstruction. Meanwhile, the patient will present hematemesis (as in our patient), fatigue and anemia if there's erosion or ulceration of the adenoma (5). Therefore, the symptoms present a diagnostic challenge for surgeons as the symptoms are relatively common in the medical practice. It was also found that symptomatic patients who presented with obstructive and bleeding symptoms have larger and similar size lesion (mean 2.1 cm and 2.8 cm, respectively) while asymptomatic patients have smaller lesion (mean 1.6 cm) (8).

Since physical examination does not provide any significant findings, diagnostic investigation would be necessary. This include abdominal CT Scan and Endoscopy (12). CT Scan allow us to localize the location of the lesion. It is useful when used in a large lesion to determine extra luminal involvement (11). Otherwise in small lesion it has poor sensitivity (1). With intravenous contrast administration enhancement, we can identify whether the lesion is homogenous or heterogenous lesions with solid or cystic components (9). Finally, we can do an endoscopy to visualize the lesion. It is also useful as it facilitates the process of taking a biopsy (5). It is imperative to take a deeper biopsy to obtain a conclusive histopathological specimen (1,5,8-9). Unfortunately, in our case, we're unaware of this fact and BGA was not diagnosed until after surgical resection.

There are two approaches in treating symptomatic BGA, either by open / laparoscopic surgery or endoscopic resection (3,6-7,9-10). While some supports the notion of conservative approach in treating the lesion due to its benign nature and often small size, there's a fear that it might undergo malignant transformation. So, some doctors opted for interventional approach instead (4,10). Most authors advocated Endoscopic Resection of the lesion instead of a more conventional open surgery (3,6-7,9-10), which is in line with our choice of treatment which is endoscopic polypectomy. Many advanced modalities of endoscopic resection had been introduced such as Snare Resection, Endoloop technique and Endoscopic Submucosal Resection to improve the safety of surgery (7,12). However, endoscopic resection is only preferred if: 1) Tumor is pedunculated and in the size that can be resected and pass through the pylorus to the oral cavity, 2) Lesion that are <1cm or between 1cm to 2cm and limited to the mucosa (7). Conversely, the conditions that are unfavorable for Endoscopic Resection include: 1) The tumor is too large (>2cm), 2) Difficult to be accessed by endoscopy due to its location from the pylorus and anatomical location (2) or have a submucosal extension. In this condition, surgical excision should be opted instead (3-4,7,9-10). However, despite the tumor in our patient being slightly less than 3 cm, we still managed excise it endoscopically. Fortunately, the long-term outcome of Brunner's Gland Adenoma is good as they are no report of recurrence post resection (5,8). Most patient will remain on long term proton pump inhibitors post resection (3).

3.0 CONCLUSION

Brunner's gland adenoma is a rare condition and the exact cause and mechanism that contributed to Brunner's Gland proliferation is still not understood fully. It's normally asymptomatic but when manifested physically, such as hematemesis and anemia like in our patient, the symptoms will mimic common conditions that might cause a diagnostic challenge for the treating doctors. Fortunately, the adenoma can be treated by both endoscopic resection and the outcome of the treatment is very good and uneventful.

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