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Case Report
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# Giant Brunner's Gland Hamartoma of the Proximal Jejunum: A Rare Cause of Obscure Gastrointestinal Bleed

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## Abstract

Brunner's gland hamartoma is a rare benign proliferative lesion that develops most commonly in the posterior wall of the proximal duodenum and ectopic locations such in the proximal jejunum are extremely rare. We report a case of a giant Brunner's gland hamartoma in the proximal jejunum presenting as obscure gastrointestinal bleed and discuss the diagnostic challenges that are faced.

## Introduction

Brunner's gland hamartoma, are rare, benign, proliferative lesions arising from the submucosal Brunner's glands which are usually found in the proximal duodenum. Majority of cases are discovered incidentally during upper endoscopy, and are usually asymptomatic and small. Occasionally, they may be larger and can present with common gastrointestinal symptoms like anemia, overt bleeding, obstruction, or chronic abdominal pain [1].

## Case Presentation

Our patient is a 65 year old lady who has had multiple hospital admissions since 2010 for symptomatic iron deficiency and persistent vague abdominal discomfort. Since then, she has undergone endoscopic evaluation with gastroscopy and colonoscopy several times, retrograde balloon assisted small bowel enteroscopy, video capsule endoscopy, tagged RBC scan and multiple non-contrasted CT scans of the abdomen and pelvis (as she is allergic to CT contrast), all of which have been negative in identifying a source of gastrointestinal bleed.

She again presented this year to the emergency department with symptomatic anemia and a hemoglobin level of 4.8 g/dL. Video capsule endoscopy was performed. Ten minutes into the study, an ulcer with small amounts of fresh blood was identified in the proximal jejunum (Figure 1). Push enteroscopy was then performed, demonstrating an elongated polypoidal mass in the proximal jejunum (Figure 2). Biopsies were taken and the small bowel wall opposite the lesion was tattooed and clipped to mark its location (Figure 3). Histology from the endoscopic biopsies was reported as normal small bowel mucosa.

Subsequently, the patient underwent an exploratory laparotomy. The site of the polyp was easily identified at the site of the pre-operative endoscopic tattoo. After running the rest of the bowel to ensure that there were no other masses, an enterotomy was made close to the tattoo and an elongated jejunal polyp with mucosal ulceration was found within the lumen, its long stalk originating approximately 20 cm distal to the duodenojejunal flexure. The short segment of jejunum was resected with subsequent end-to-end handsewn anastomosis. The patient recovered well post-operatively with no further drop in hemoglobin level.

The resected specimen showed a pedunculated polyp measuring 3.5 cm × 1.7 cm × 1.1 cm, with a stalk measuring 12 cm in length and 1.8 cm in width (Figure 4A). Microscopic examination of the polyp and its stalk showed submucosal lobules of Brunner's glands (Figure 4B) with occasional dilated ducts (Figure 4C). These were separated by a fibromuscular stroma with bundles of smooth muscle (Figure 4D) as well as some irregularly dilated blood vessels. As such, the final diagnosis was that of a Brunner gland hamartoma.

## Discussion

Brunner's glands are branched acinotubular alkaline secreting submucosal which function to neutralize the acidic chyme and optimize intestinal absorption. They are mainly located in the

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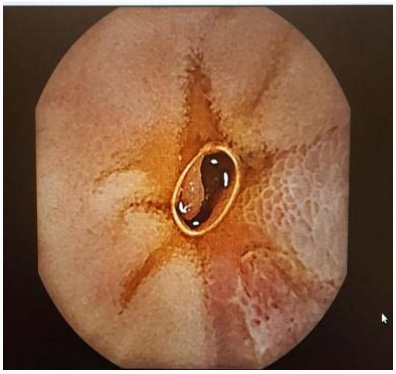

**Figure 1:** Video capsule endoscopy–ulceration in the proximal jejunum with a small amount of fresh blood seen approximately 10 minutes into the study.

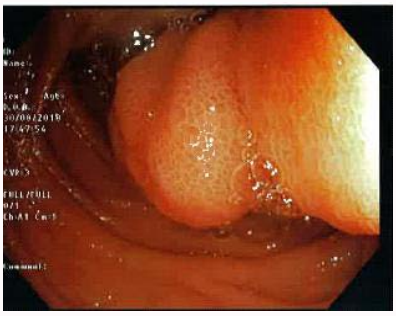

**Figure 2:** Push enteroscopy–elongated polypoidal lesion in the proximal jejunum.

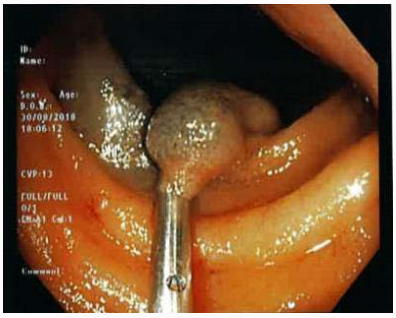

**Figure 3:** Small bowel mucosa opposite the jejunal polyp marked with tattoo and clip to facilitate intra-operative identification of the lesion.

proximal duodenum between the pyloric ring and the ampulla of Vater. Ectopic locations are uncommon and include the pylorus and jejunum. Brunner's gland hamartoma are rare benign tumors arising from these glands. Histologically, they consist of increased numbers of normal appearing Brunner glands accompanied by smooth muscle and adipose tissue [2].

The exact pathogenesis of Brunner's gland hamartoma remains uncertain. It has been postulated that hyperacidity stimulates the proliferation of Brunner's glands. Another proposed etiology is that the glands undergo hyperplastic reactive proliferation to inflammation and associations with helicobacter pylori infection, peptic ulcer disease, chronic pancreatitis and chronic renal failure have been described.

Cruveilhier described the first case of a Brunner's gland lesion causing intussusception in 1835. Since then, there have been less than 200 cases reported in the literature, majority of which describe

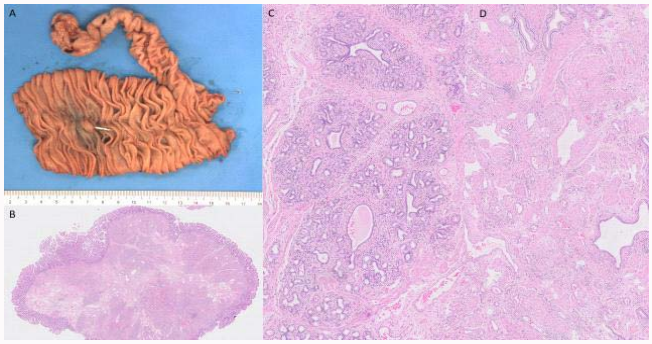


**Figure 4A:** Macroscopic photo of specimen. Pedunculated polyp present at top of photo. **4B:** Hematoxylin and eosin-stained photomicrograph showing a low power view of one section of polyp showing it to be covered by small bowel epithelium with lobulated Brunner glands seen in the submucosa. **4C:** Hematoxylin and eosin-stained photomicrograph showing a medium power view of the lobules of Brunner glands with dilated ducts. **4D:** Hematoxylin and eosin-stained photomicrograph showing a medium power view of the fibromuscular stroma from the center of the polyp.

duodenal Brunner's gland hamartoma. We have found only one previous case report in the literature describing a Brunner's gland hamartoma of the jejunum causing iron deficiency anemia and mimicking intussusceptions on radiologic studies [3].

Since Brunner's gland hamartoma are most commonly found in the proximal duodenum, gastroduodenoscopy is usually helpful for diagnosis. However, in rare instances like in our patient where the lesion is located more distally, conventional gastroduodenoscopy is inadequate in identifying the lesion and other modalities of evaluating obscure gastrointestinal bleed need to be considered. Useful diagnostic tools that can aid in identifying Brunner's gland hamartoma include small bowel contrast studies, CT enterography, video capsule endoscopy, as well as push enteroscopy and balloon-assisted enteroscopy.

Our patient has severe allergy to CT contrast which made diagnosing the lesion more challenging. Furthermore, the lesion was not demonstrated on the initial Video Capsule Endoscopy (VCE), which has a false negative rate of 11% for small bowel lesions. There is greater yield of VCE and a higher likelihood of positive findings in patients with a hemoglobin level of less than 10 g/dL, longer duration of bleeding (>6 months), more than 1 episode of bleeding, overt (rather than occult) bleeding (60% *vs.* 46%), and when VCE is used within 2 weeks of the bleeding episode (91% *vs.* 34%) [4]. Push enteroscopy is useful in accessing the proximal jejunum up to 100 cm distal to the ligament of Treitz, which allows the endoscopist to biopsy and better evaluated the lesion. As Brunner's glands are mainly located in the submucosal layers, biopsies of Brunner's gland lesions often show negative findings with slight changes of inflammation.

As there is insufficient knowledge about the natural history of Brunner's gland hamartomas, there is no consensus on its management and resection is reserved patients who are symptomatic. Recently, there have been reports of successful endoscopic removal of Brunner's gland hamartomas using snare resection and endoloop technique, and it appears to be a less invasive and effective approach compared to surgical resection [5]. Limitations include the size of the polyp and the thickness of the stalk. Large lesions which cannot be safely excised endoscopically should be referred for surgery. In our patient, in the view of the size of the polyp with its long stalk, wide base and its location in the proximal jejunum, surgical resection was

the preferred option. Small bowel resection is preferred over local excision of the polyp in such cases as the origin of the polyp stalk can be resected to ensure clear margins.

In conclusion, we report a very rare occurrence of Brunner's gland hamartoma in the proximal jejunum presenting as a diagnostically challenging case of obscure gastrointestinal bleed.

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