
Case report

A large Brunner's gland hamartoma presenting with severe anaemia

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Introduction

Johann Conrad Brunner (1653-1727) who was a Swiss anatomist discovered the duodenal submucosal structures known today as Brunner's glands (1). Brunner's glands are branched acinotubular glands in the submucosa of the duodenum. They are located mainly in the duodenal bulb, proximal duodenum and progressively decrease in number and size in the distal duodenum. The term "Brunner's gland adenoma" or "Brunneroma," has been applied to lesions that are histologically similar to Brunner's gland hamartoma (2). Cruveilhier, in 1835, and Savioli in 1876, described the first cases of "adenoma" of the Brunner's gland. Several features of these lesions favour their designation as hamartomas, including the lack of encapsulation, the mixture of acini, smooth muscles, adipose tissue, Paneth cells, and mucosal glands and the lack of any cellular atypia (3).

Grossly, Brunner's gland hamartomas have a smooth surface and tend to be well-circumscribed, solitary polyps that can be sessile

or pedunculated. They are pink or tan on cut surface with a lobular appearance due to fibrous septae. Histologically, the solitary mass contains a mixture of acini. The cells of Brunner's glands have eosinophilic to clear cytoplasm, and they typically contain basally oriented nuclei, ducts, smooth muscle, adipose tissue, and lymphoid tissue. They may occasionally contain heterotopic pancreatic acini and ducts. The presence of both ductal and glandular components is further evidence of a hamartomatous origin (2).

Diagnosis is usually confirmed by the use of imaging studies such as computerised tomography, endoscopic ultrasound scan, and upper gastrointestinal endoscopy. Brunner's gland hamartoma is a benign lesion, which can be treated by limited surgical resection or polypectomy. The long term outcome after Brunner's gland hamartoma resection is favourable without any reported recurrences of lesions (3,4,5). A few cases of Brunner gland hamartoma have been reported in association with epithelial dysplasia, duodenal adenocarcinoma, and carcinoid tumors (4).

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Case Report

A 37 year old male presented to the general surgical ward with fatigue, weakness and several episodes of malaena for 7 months duration and two episodes of coffee ground vomiting for one day. The patient had been treated for gastritis and anaemia with antacids and haematinics during the last 7 month period. The family history revealed that his paternal aunt had similar symptoms and signs. She had also undergone upper gastrointestinal polypectomy and was found to be in good health after surgery. Clinically, the patient was severely pale with a tachycardia (100 beats/min) and a blood pressure of 120/80mmHg. On examination, other systems were normal.

On admission, his haemoglobin was 4.2g/dl and the platelet count was 423x10³/mm³. The blood picture showed a hypochromic microcytic anaemia. Neither an ultrasound scan nor a CT/

MRI scan was done preoperatively. Upper gastrointestinal endoscopy revealed a polyp in the first part of the duodenum. The patient was treated with transfusion of 5 pints of blood, tranexemic acid and antacids prior to surgery. A polypectomy was performed and the polyp was sent for histology. The patient was discharged on haematinics. His haemoglobin value was 11.8g/dl two months after surgery and the patient was found to be currently in good health.

Pathological findings

A formalin-fixed resected specimen of a pedunculated, firm polyp measuring 4.5x2x1.5cm was received. The peduncle was 2cm in length and 0.6cm in diameter. There was a focus of ulceration (1x0.7cm) close to the peduncle (Fig. 01). The cut section was firm and white and showed streaks of haemorrhage (Fig. 02).



Fig. 1. A pedunculated polyp with a focus of ulceration close to the peduncle



Fig. 2. Cut section showing haemorrhagic streaks

Histological sections revealed a polypoid lesion composed of lobules of proliferated benign acini of Brunner's glands separated by fibromuscular septae. The septae showed foci of lymphoid aggregates, mature adipocytes (Fig. 3 and 4), congested blood vessels and a diffuse infiltrate of lymphocytes, plasma cells and eosinophils. The surface epithelium showed an ulcer (Fig. 7). The rest of the epithelium showed multiple foci of healed ulcers with regeneration (Fig. 8). Appearances were those of an ulcerated Brunner's gland hamartoma.

Discussion

Duodenal tumours, both malignant and benign, are rare. Brunner's gland hamartomas/adenomas account for up to 1% of small bowel tumours (6). The incidence of Brunner's gland hamartoma in Sri Lanka is not known because of its rarity. In 2012, The Ceylon Journal of Medical Science, published a case of a 35 year old female presenting with anaemia due to a Brunner's gland hamartoma in the junction between 1st and 2nd part of the duodenum (7).

Most symptomatic patients present in the fifth or sixth decade and there is an equal sex distribution (6). Brunner's gland polyps are mostly located in the proximal duodenum and are usually pedunculated, and a size greater than 2 cm is exceedingly rare (8). This patient was somewhat younger at presentation with the lesion arising from the 1st part of the duodenum. The size of the lesion was greater than usual. Brunner's gland adenomas/hamartomas are detected incidentally and are usually

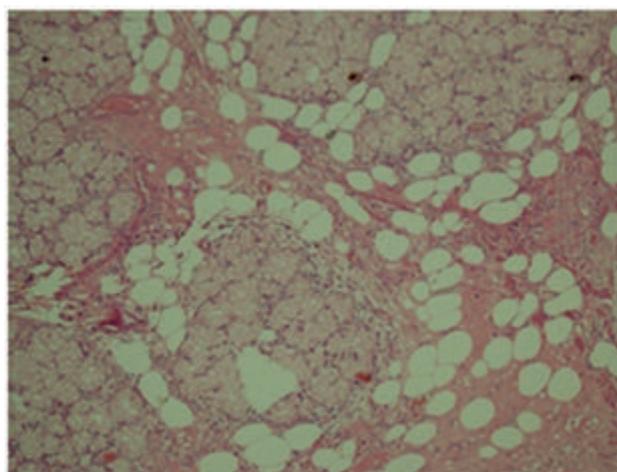


Fig.3. Proliferated benign Brunner's gland separated by fibromuscular septae with foci of mature adipocytes(H &E x 100)

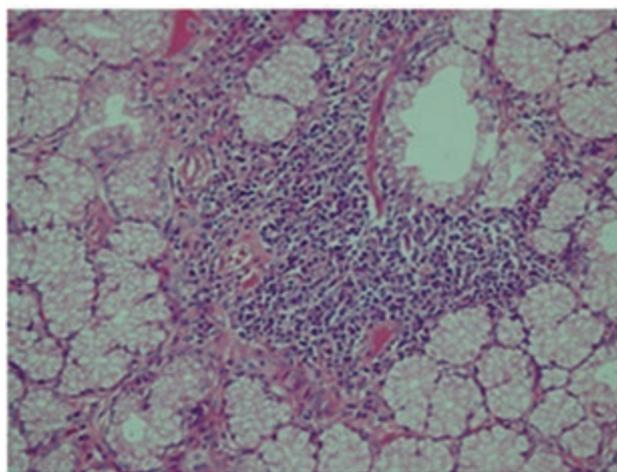


Fig.4. A lymphoid aggregate within the lesion (H &E x 200)

asymptomatic or have nonspecific complaints such as nausea, bloating, or vague abdominal pain. The symptomatic patients can present with a variety of clinical manifestations.

The most common presentations in symptomatic patients are gastrointestinal bleeding (37%) and obstructive symptoms (37%). Occasionally, they present with intussusception

(3,9). Involvement of the Ampulla of Vater has been associated with uncommon manifestations, including obstructive jaundice, biliary fistula, and recurrent pancreatitis. On rare occasions, patients can present with gastric outlet obstruction (3). This patient had upper gastrointestinal bleeding denoting a common mode of presentation and with a macroscopic focus of ulceration. Even though gastrointestinal bleeding was a common presentation of Brunner's gland hamartoma, it is a rare cause of anaemia due to chronic gastrointestinal bleeding.

The pathogenesis of Brunner's gland hamartoma remains unclear. Because of the anti-acid function of Brunner's glands, it has been postulated that hyperchlorhydria could stimulate these structures to undergo hyperplasia. In one study, 45% of patients with Brunner's gland hyperplasia had increased gastric output; however, 20% of these patients were reported to have gastric hypoacidity. It has been suggested that *Helicobacter pylori* infection may play a role in the pathogenesis of Brunner gland hamartomas. In a study involving 19,100 subjects, Kovacevic and coworkers found that 5 of 7 patients diagnosed with these hamartomas had concurrent *H. pylori* infection. A pathogenic link, however is difficult to establish, given the rarity of these lesions and the high prevalence of *H. pylori* in the general population.

Another theory is that these hamartomas may form as a hyperplastic reaction to inflammation, given the inflammatory cell infiltrate occasionally witnessed histologically. Supporting evidence for this assertion is insufficient given that the presence of

lymphocytes is not unusual in the normal submucosa throughout the gastrointestinal tract (3). Goldman, a well-known reviewer of the Brunner's gland "hyperplastic" polyps, pointed out that the histologic architecture of these lesions consists of a combination of ductal and acinar structures with fibromuscular and adipose elements. Accordingly, he was the first to stress that these features strongly suggest a hamartomatous origin for Brunner's gland polyps. The distribution of Brunner's gland hamartomas include the duodenal bulb (57%), the second part (27%) and third part (5%) of the duodenum, the pyloric canal (5%), jejunum (2%), and proximal ileum (2%)

The differential diagnosis usually includes leiomyoma, adenoma of the mucosal glands, carcinoid tumour, lipoma, prolapsed pyloric mucosa or antral polyp (8). The histopathological picture of this patient's polyp was that of a typical Brunner's gland hamartoma showing a focus of acute surface ulceration and features of regenerative atypia due to healed ulceration. An entire gastrointestinal examination is mandatory since hamartomas are commonly associated with a generalised polyposis syndrome. Even though these tumors are dysembryoplastic lesions and exhibit a benign course without malignant predisposition, the literature reports a proven case of cancer developing from this origin (8). The patient was asymptomatic after 2 months of follow up with a gradual increase in haemoglobin level. An unrecognised familial inheritance should be considered in this patient due to the presence of a positive family history in this patient.

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