Brunner's gland hamartoma of the duodenum

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ABSTRACT

Brunner's gland hamartoma (BGH) is a rare benign tumour of the duodenum. We present a case of duodenal obstruction caused by a BGH, which required surgical resection after a failed attempt at endoscopic removal.

KEYWORDS

Duodenal polyp – Brunner's gland hamartoma

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Brunner's gland hamartoma (BGH) is a rare benign tumour formed from a proliferation of Brunner's glands, typically forming a polypoidal growth in the proximal duodenum. It may be an incidental finding during an investigation but can also present acutely with haemorrhage or obstruction, requiring endoscopic or surgical resection.

Case History

A 66-year-old otherwise fit and healthy woman was admitted through our emergency services with an episode of sudden onset, severe epigastric pain of one day's duration. She also complained of recurrent nausea and vomiting of similar duration. The pain was continuous and non-radiating with no aggravating or relieving features. The vomiting was non-bilious and the amounts were small. There was no history of blood in the vomitus. Her past history included longstanding dyspepsia and an episode of haematemesis while holidaying overseas, which was never investigated.

On physical examination, the patient appeared unwell with tachycardia and mild dehydration. There was no pallor, jaundice or lymphadenopathy. Abdominal examination was unremarkable apart from mild epigastric tenderness. Her initial investigations included normal full blood count, liver and kidney function tests, inflammatory markers and serum amylase. An erect chest x-ray, plain x-ray of the abdomen and electrocardiography were reported as normal. Her symptoms failed to resolve despite parenteral opiates, nasogastric decompression of the stomach and intravenous fluids.

Urgent computed tomography of the abdomen revealed a large obstructing duodenal polyp in the second part of the duodenum (Fig 1). The polyp was confined to the duodenal lumen with no mural or perimural changes. No significant lymphadenopathy was noted, and the biliary tree and pancreas appeared normal. Subsequent gastroscopy revealed a large benign looking pedunculated polyp arising from the first part of the duodenum. The bulk of the polyp

was prolapsing and obstructing the second part of the duodenum. Polyp biopsies were reported as BGH. Further attempts at endoscopic snaring were unsuccessful owing to lack of adequate luminal space to deploy the snare.

The patient was managed with total parenteral nutrition and underwent a planned laparotomy via an upper midline incision. The findings revealed a large benign duodenal polyp impacting the second part of the duodenum. The pedicle base was transfixed with Prolene® sutures (Ethicon, Somerville, NJ, US) and the polyp removed through a duodenotomy. Her postoperative course was uneventful.

Polyp histology revealed a BGH characterised by disorganised lobules of Brunner's glands in the submucosa with cystically dilated ducts (Figs 2 and 3). There was surface mucosal ulceration due to the mechanical effect of the large size. There was no ectopic pancreatic or gastric tissue seen.

Discussion

Also referred to as Brunner's gland adenoma or Brunneroma, BGH is a rare benign tumour of the duodenum, featuring a proliferation of Brunner's glands in the submucosa. They represent about 5–10% of benign duodenal tumours¹ and have an estimated incidence of less than 0.01%.² Hamartoma designates an excessive focal overgrowth of mature normal cells and tissues in an organ, composed of identical cellular elements. The term 'Brunner's gland hamartoma' is favoured over 'Brunner's gland adenoma' as they show an admixture of normal tissues including Brunner's glands and ducts histologically. Adipose tissue, Paneth cells, mucus glands, pancreatic acini and lymphoid tissue usually without cellular atypia can also occur in BGH.^{1,2}

Brunner's glands are located in the submucosa of the duodenum. They secrete an alkaline fluid containing mucin, which protects the mucosa from the acidic stomach contents entering the duodenum.³ BGHs consist of an abnormal proliferation of Brunner's glands, along with a mixture of

Figure 1 Computed tomography showing obstructing duodenum polyp (arrow)

adipose tissue, fibrous septa, smooth muscle and other normal tissue components.⁴ They present typically as a single polypoid and pedunculated mass with a normal mucosal surface. The distribution of BGHs in the small bowel reflects that of Brunner's glands, with about 70% occurring in the

Figure 2 Hamartomatous polyp with superficial mucosal erosion composed of lobules of disorganised mucinous glands of Brunner's type (5x magnification, haematoxylin and eosin stain)

Figure 3 Hamartomatous lobules of Brunner glands with no atypia or sinister features (20x magnification, haematoxylin and eosin stain)

proximal duodenum and a decreasing frequency occurring throughout the rest of the duodenum.⁴

The aetiology of BGH is unclear although it is suggested that hyperacidity or *Helicobacter pylori* may provoke proliferation of these protective glands.⁵ In addition to alkaline mucus, Brunner's glands secrete urogastrone, which inhibits gastric acid secretion, and may have a protective role against peptic ulcer development. In a large study of 19,000 people, 5 out of 7 patients who were diagnosed with BGH were also found to have *H pylori* infection.⁶ However, the significance of this is unclear as the presence of *H pylori* is so commonplace.

BGHs are often asymptomatic and may be an incidental finding during endoscopy or imaging.⁷ BGHs may present acutely with gastrointestinal bleeding, bowel obstruction, or more insidiously with symptoms such as dyspepsia or anaemia.^{7,9} A study of 27 cases of BGH found that 37% presented with haemorrhage, 37% with bowel obstruction and the rest were incidental findings.⁴ In more unusual cases, BGH has led to intussusception,⁵ obstructive jaundice and pancreatitis.⁹

Removal of a BGH is usually by endoscopic resection unless it is particularly large or broad-based, as in the present case. There have been reported cases where malignancy was suspected so a more radical procedure such as a pancreaticoduodenectomy was performed to ensure wide enough clear margins.¹⁰ In many cases, the histological diagnosis of BGH can only be confirmed after complete resection of the polyp as biopsies obtained during endoscopy may not be deep enough to demonstrate the glandular proliferation in the sub-mucosal tissues.

BGHs are considered to be entirely benign tumours although there have been anecdotal reports of malignant foci in them.¹¹ There are no reported recurrences following removal of a BGH and the long-term prognosis is excellent.

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