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Case report

A 10 cm pedunculated duodenal Brunner gland hamartoma, case report and literature review

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ABSTRACT

Introduction: Brunner gland hamartoma is rare duodenal neoplasm. These benign lesions are usually presented by upper gastrointestinal bleeding and sometimes extend to cause intestinal obstruction.

Presentation of the case: We report a case of a 43-year-old male patient manifested with iron deficiency anemia. Upon investigations, computed topography (CT) scan found a dilated first part of the duodenum with presence of large pedunculated polyp. The histopathological examination revealed a submucosal lobular proliferation of duodenal Brunner's gland separated by a fine fibrous septum. No dysplastic signs were observed. Immunohistochemical studies confirmed the nature of the glands and reveled absence of *Helicobacter pylori* gastritis. Diagnosis was confirmed.

Discussion: Brunner glands hamartomas are rare tumors. They are commonly presented by upper GI bleeding and intestinal obstruction. The pathogenesis remains unclear. They are usually located in the first part (bulb) of the duodenum. Mucosal irritation and *Helicobacter pylori* infection are suggested causes. Different surgical and endoscopical modalities are applied in the management depending on the size and location of the mass. In our case, the tumor was removed by Endoscopic submucosal dissection.

Conclusion: Brunner gland hamartoma is a rare usually benign tumor. Presented clinically by upper GI bleeding and obstruction. Histopathologically Brunner gland Hamartoma characterized by lobular proliferation of Brunner gland associated with presence of other mature tissues. Although these tumors are benign it carries a minor risk of malignant transformation.

1. Introduction and importance

Primary intestinal neoplasms are rare and Brunner's gland hamartoma also referred as Brunner's gland adenoma or brunneroma is even rarer. These tumors are benign and represent 10.6 % of benign duodenal tumors [1,2]. These tumors had been found in 0.008 % in a single series of autopsies [1,2]. These lesions manifest clinically in most of cases by upper gastrointestinal bleeding and sometimes by intestinal obstruction. They are usually presented as pedunculated polyp and situated in the first part (bulb) of the duodenum. Endoscopic (surgical) resection is the treatment of choice in these polyps [3].

This work has been reported in line with the SCARE 2020 criteria [4].

2. Presentation of a case

A 43-year-old male patient presented with fatigue and intermittent melena for several months. The investigations revealed iron deficiency anemia (microcytic hypochromic anemia). His hemoglobin was 14 g/l MCV of 73.5 fl and MCH of 22.1 pg. The CT scan found a dilated first part of the duodenum with presence of large pedunculated polyp measuring 7 cm of largest dimension. Endoscopy of the upper digestive system confirmed the presence of a large pedunculated and eroded polyp arising from the duodenal bulb. The polyp was located in the anterior wall and had a wide base. Endoscopic submucosal dissection of the polyp was performed. The patient post endoscopic course was unremarkable.

3. Pathological examination

The gross endoscopic resection specimen showed a large duodenal polyp with a smooth homogenic surface and measuring 9.5 × 5 cm (Fig. 1). The specimen was fixed in 10 % formalin and routine hematoxylin-eosin-saffron sections were examined.

Microscopic examination revealed a polypoid lesion covered on the

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surface by intestinal type epithelium and focally by foveolar gastric epithelium. The submucosa composed of well differentiated homogenous proliferated lobules of Brunner glands (Fig. 2A). The glands were lined by cylindrical clear mucinous cells with basal semilunar hyperchromatic nuclei. The lobules were separated by fine fibrous septa. The lesion also contained bundles of smooth muscles fibers embedded within the lobules (Fig. 2B). No mitoses were observed. The adjacent connective tissue contained mild lymphocytic infiltration (peptic duodenitis). No other heterogenous tissues were noted within the lesion and no dysplastic changes had been observed. Immunohistochemical studies confirmed the smooth muscles fibers (desmin) separated the brunner gland lobules (AE1/AE3) (Fig. 2C). Brunner glands express MUC6 (Fig. 2D) and focally express MUC5AC (Fig. 2E). Brunner gland did not express MUC2 (Fig. 2F).

4. Discussion

Primary intestinal neoplasms are rare, accounting less than 1 % of the gastrointestinal tumors and benign duodenal tumors reaching almost 16 % [5,6]. Cruveilhier has described the first Brunner gland adenoma at the end of the 19th century [7] while Brunner's glands were first described by Brunner in 1688. These glands are situated in the duodenal submucosa. Most of these glands are seen in the first part of the duodenum and decreases towards its distal parts. The glands proportion also decreases with age as its quantity almost 50 % at birth and reduced to 35 % by 50 years [1]. the Brunner glands function is crucial for preventing ulcers as they secrete mucin and inhibit gastric acid secretions.

The exact pathogenesis of Brunner's gland adenoma is unknown one hypotheses is related to the chronic irritation of the local mucosa could stimulate the glands and undergo hyperplasia [8]. Another hypothesis suggest that *Helicobacter pylori* infection may play role in the pathogenesis [9]. A recent study showed that 71 % of Brunner's glands adenomas was found positive of *Helicobacter pylori* [10]. In our case no gastric biopsy was performed to detect *Helicobacter pylori* infection and it was not detected in the duodenal polyp. Although high prevalence of *Helicobacter pylori* infection in the general population, we cannot correlate this pathogenic relation as a cause due to the extreme rarity of Brunner's gland adenoma. Another suggested cause of these lesions is chronic pancreatitis as nodular hyperplasia of Brunner glands had been found in 75.5 % of patients with chronic pancreatitis [11]. Gastric foveolar metaplasia is often associated with Brunner gland hamartoma as in our case (Fig. 2E) and its considered as mucosal injury repair mechanism and promotes the occurrence of this lesion [12]. Currently the most approved pathogenetic hypothesis is to be more of dysmoembryoplatic lesion or hamartoma [13].

Most patients with Brunner's gland hamartoma are presented by gastrointestinal bleeding as demonstrated in our case and obstructive intestinal symptoms are reported. Levin et al. found 37 % of these tumors presented by ulcerations and chronic bleeding and 37 % presented by intestinal obstruction [14].

In a histopathological point of view the differnation between

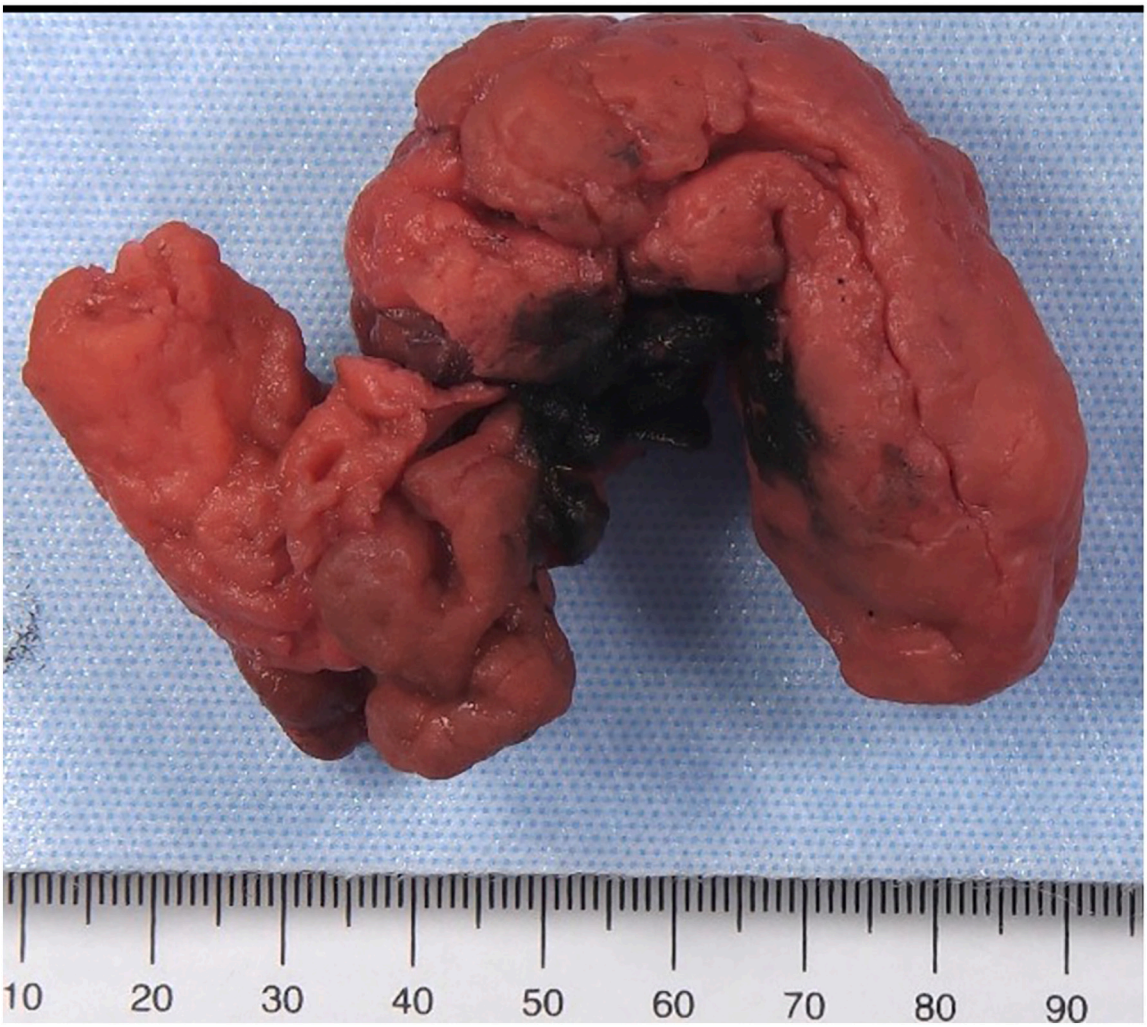

Fig. 1. illustrate the Grosse appearance of the large resectected duodenal polyp with a smooth surface.

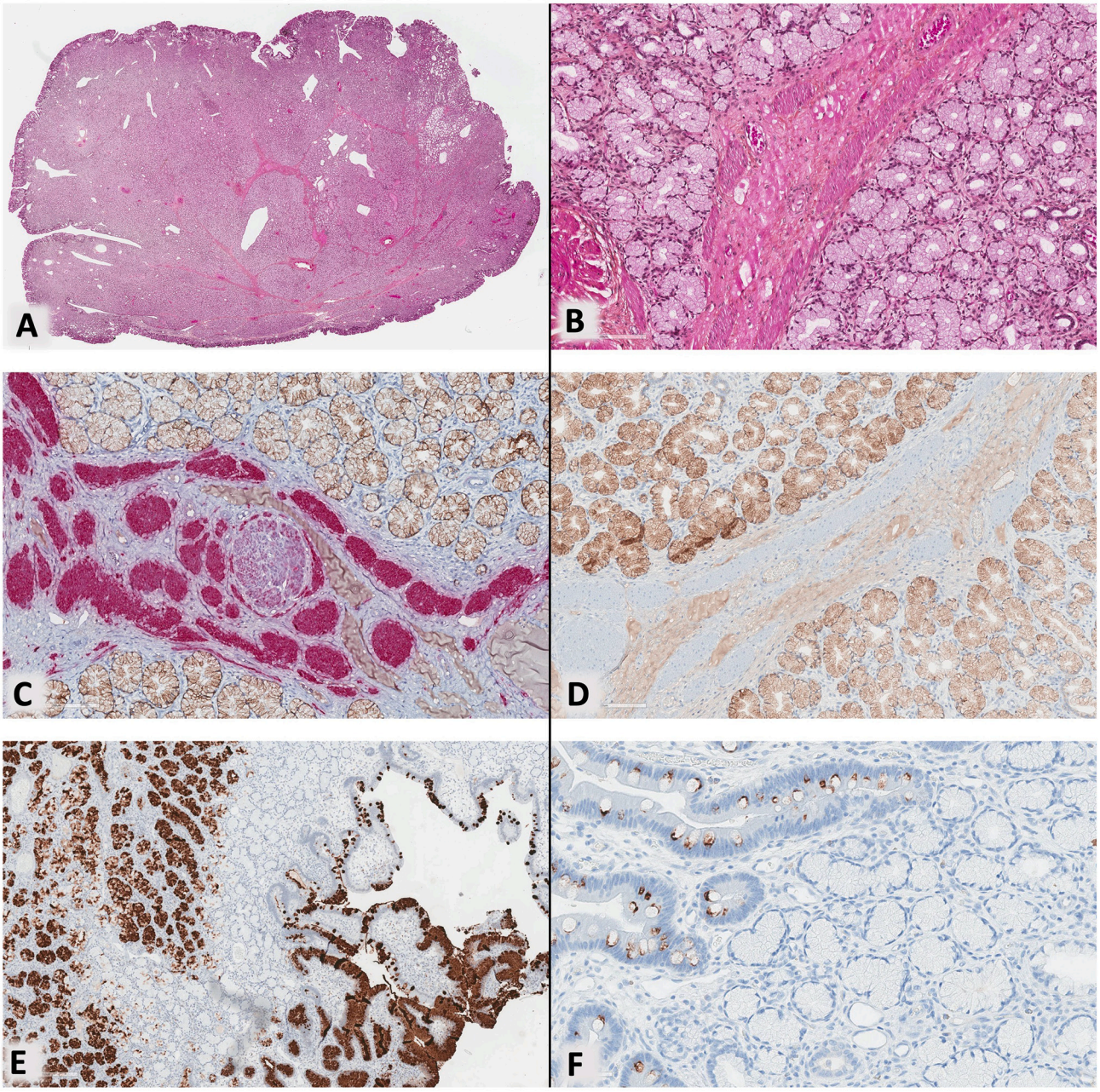

Fig. 2. (A) low power magnification of brunner gland hamartoma with the presence of diliated glands and smooth muscle fibers, (B) high power magnification illustrate the Brunner gland proliferation with mixture of smooth muscle fibers, (C) the Brunner glands proliferation with antiAE1/AE3 antibody (marron) and smooth muscle with anti desmin anti body (red), (D) Brunner glands express MUC6, (E) Brunner glands focally express MUC5AC note the expression on the gastric foveolar metaplasia on the surface, (F) Brunner gland did not express MUC2 note its expression by the intestinal goblet cells on the surface epithelium. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

Brunner's gland hyperplasia or hamartoma is often difficult [15]. However, it had been described those lesions smaller than 5 mm whether single or multiple are considered as hyperplasia and larger lesions of more than 5 mm as hamartomas [16]. In fact the term Brunner gland Hamartoma and Brunner gland adenoma are often used synonymously, making these lesions difficult to understand but it has been suggested that the presence of other mature tissues such as ciliated microcysts, adipose or muscular tissues within the lesion it might be considered more as hamartoma [17]. It is reported that Brunner's glands hamartomas are benign lesions but it had been associated with malignant and pre-malignant lesions such as epithelial dysplasia, duodenal adenocarcinoma and carcinoid tumors [18,19] prognosis of these tumors is favorable as recurrence after either endoscopic or surgical resection is rare [20,21]. Table 1 summarized the main clinical features of patients with Brunner's gland hamartoma that had been described in the literature.

5. Conclusion

Brunner gland hamartoma is a rare usually benign tumor. Presented clinically by upper GI bleeding and sometimes by intestinal obstruction. The pathogenesis remains unclear, but some hypotheses are suggested as mucosal irritation, *Helicobacter pylori* infection, chronic pancreatitis and mucosal injury. Histopathologically Brunner gland Hamartoma and Brunner gland adenoma used synonymously characterized by lobular proliferation of Brunner gland associated with presence of other mature

Table 1
Main clinical features of patients with Brunner's gland hamartoma: an overview of literature.

Author (year)	Sex	Age	Site	Size (CM)	Shape	Management
Gourtsoyiannis et al. (1990) [22]	Male	74	Descending duodenum	5	Pedunculated polyp	Surgical resection
Walden et al. (1998) [23]	Male	28	Duodenal bulb	4	Pedunculated polyp	Endoscopical resection
Hizawa et al. (2002) [24]	Female	62	Duodenal bulb	0.7	Broad based lesion	Endoscopical resection
	Female	71		1.8	Broad based lesion	
	Female	36		2	Pedunculated polyp	
	Male	63		1.5	Sessile polyp	
	Male	65		1.5	Sessile polyp	
	Male	34		2	Sessile polyp	
Tan et al. (2002) [25]	Male	70	Descending duodenum	2	Pedunculated polyp	transdeudenal approach Laparotomy
Gao et al. (2004) [13]	Male	32	Duodenal bulb	3.5	Pedunculated polyp	Surgical resection
Rocco et al. (2006) [26]	Female	58	Duodenal bulb	4	Pedunculated polyp	Endoscopical resection
Petersan et al. (2008) [27]	Female	56	Pylorus-Descending duodenum	8.5	Submucosal longtubular mass	Bilroth I procedure
Jung et al. (2013) [1]	Male	45	Pyloric ring	4.8	Pedunculated polyp	Endoscopical resection
Akaki et al. (2014) [12]	Male	26	Gastroduodenal junction	6.4	Pedunculated polyp	Distal gastrectomy
Martinez et al. (2014) [20]	Male	60	Duodenal bulb	4	Subepithelial mass	Surgical resection
Takeuchi et al. (2015) [28]	Female	52	Duodenal bulb	5	Pedunculated polyp	Laparoscopic partial duodenectomy
	Female	52		3.5		Laparoscopic partial duodenectomy
	Male	67		6		Laparoscopic tumor resection
Kostalas et al. (2016) [29]	Female	52	Duodenal bulb		Solid mass	Pancreatoduodenectomy
Peloso et al. (2017) [30]	Male	72	Descending duodenum	4	Broad based lesion	Surgical resection
Kitagawa et al. (2018) [31]	Female	64	Duodenal bulb	7	Pedunculated polyp	Endoscopical resection
Rana et al. (2019) [32]	Male	76	Descending duodenum	12	Pedunculated polyp	Endoscopical resection
Bakheet et al. (2020) [33]	Male	55	Duodenal bulb	3.2	Pedunculated polyp	Endoscopical resection
Naito et al. (2021) [34]	Female	69	Duodenal bulb	4	Submucosal longtubular mass	Surgical resection
Zhang et al. (2022) [35]	Male	53	Duodenal bulb	6	Submucosal oval mass	Endoscopical resection

tissues such as ciliated microcysts, adipose or muscular tissues. Although these tumors are benign it carries a minor risk of malignant transformation. Recurrence is rare after resection.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

Ethical approval for this study was obtained from the ethical committee of the department and the university ethical committee.

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Author contribution

Ziyad Alsugair, and Lisa Chassagne did the conception and design of the work, the data collection, and the data analysis and interpretation.
Pierre Marie Lavrut did the critical revision of the article.
Ziyad Alsugair did the final approval of the version to be published.

Guarantor

Ziyad Alsugair, Pierre Marie Lavrut and Lisa Chassagne.

Research registration number

None.

Declaration of competing interest

No conflicts of interest.

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