Annals of Medicine and Surgery 17 (2017) 43–49

Contents lists available at ScienceDirect

Annals of Medicine and Surgery

journal homepage: www.annalsjournal.com

Case report

Saving from unnecessary pancreaticoduodenectomy. Brunner's gland hamartoma: Case report on a rare duodenal lesion and exhaustive literature review

Andrea Peloso, MD PhD [a, b], Jacopo Viganò, MD PhD [a, b], Alessandro Vanoli, MD [c], Tommaso Dominioni, MD PhD [a, b], Sandro Zonta, MD PhD [a, b], Dario Bugada, MD [d], Carlo Maria Bianchi, MD [a, b], Francesco Calabrese, MD [a, b], Ilaria Benzoni, MD [a, b], Marcello Maestri, MD PhD [a, b], Paolo Dionigi, MD [a, b], Lorenzo Cobianchi, MD PhD [a, b, *]

[a] Department of Clinical Surgical Diagnostic and Paediatric Sciences, University of Pavia, Pavia, Italy
[b] IRCCS Policlinico San Matteo Foundation, General Surgery 1, Piazzale Golgi, 27100, Pavia, Italy
[c] IRCCS Policlinico San Matteo Foundation, University of Pavia, Department of Molecular Medicine, Piazzale Golgi, 27100, Pavia, Italy
[d] Department of Surgical Science, University of Parma, Via Gramsci 14, 43126, Parma, Italy

HIGHLIGHTS

- Brunneroma is an extremely uncommon benign tumour of the duodenum.
- The case is completely mimicking a malignant tumour of the duodenum.
- The report is good opportunity to take a stock of the situation from a clinical stand point.

ARTICLE INFO

Article history:
Received 13 January 2017
Received in revised form
24 March 2017
Accepted 25 March 2017

Keywords:
Brunner's gland hamartoma
Case report
EUS
Surgical overtreatment

ABSTRACT

Introduction: Brunner's gland hamartoma (BGH) is an infrequently encountered, benign, polypoid proliferation of Brunner's glands. Usually these lesions are asymptomatic, just only occasionally presenting with duodenal obstruction or bleeding signs and mimicking a tumoral lesion.
Case presentation: A 72-year-old male, referred for recurrent vomiting and epigastralgia, was investigated and all preoperative findings were suggestive of a tumour of the duodenum. During the scheduled pancreaticoduodenectomy a mass, resultant to a polyp, was palpatory felt inside the duodenum and then successfully and completely resected through a duodenotomy avoiding surgical overtreatment and connected postoperative morbidities. Histological analysis showed hyperplasia of Brunner's glands correspondent to a Brunner's gland hamartoma. BGH was undiagnosed before surgery, due to its particular sub-mucosal growth simulating an expanding process starting from the duodenum, and secondly due to unsuccessful biopsies performed during endoscopic procedure.
Conclusion: BGH is a rare lesion featuring, when symptomatic, obstructive or bleeding symptoms. Surgical treatment represents the gold standard approach in case of lesions that are technically impossible to remove endoscopically or in case of an undiagnosed lesion. Herein, we report a case of a patient presenting with a duodenal lesion mimicking, in all preoperative findings, a tumour of the duodenum. Duodenotomy and resection of the BGH provided a definitive cure avoiding surgical overtreatment. An intraoperative deep analysis of all surgical cases still remain crucial for a right therapeutic choice even in a new era for surgical technology. For similar intraoperative findings we recommend this technique.

* Corresponding author. Department of Clinical Surgical Diagnostic and Paediatric Sciences, University of Pavia, Pavia, Italy.
E-mail addresses: andreapeloso@hotmail.it (A. Peloso), j.vigano@smatteo.pv.it (J. Viganò), a.vanoli@smatteo.pv.it (A. Vanoli), t.dominioni@smatteo.pv.it (T. Dominioni), s.zonta@smatteo.pv.it (S. Zonta), dariobugada@gmail.com (D. Bugada), cm.bianchi@smatteo.pv.it (C.M. Bianchi), Francesco.calabrese01@ateneopavia.it (F. Calabrese), ilaria.benzoni@hotmail.it (I. Benzoni), mmaestri@smatteo.pv.it (M. Maestri), p.dionigi@smatteo.pv.it (P. Dionigi), lorenzo.cobianchi@unipv.it, l.cobianchi@smatteo.pv.it (L. Cobianchi).

http://dx.doi.org/10.1016/j.amsu.2017.03.034

1. Introduction

Brunner's gland hamartoma (BGH), also known as brunneroma or Brunner's gland adenoma, is an extremely uncommon benign tumour arising from alkaline-based mucin-secreting glands of the duodenum, with an estimated incidence in the population of around 0.008% [1]. These lesions are typically discovered incidentally, even though they can eventually lead, in case of increasing size, to obstructive or haemorrhagic symptoms such as: abdominal pain, nausea, dyspepsia, vomiting, intussusception, upper gastrointestinal bleeding and recurrent pancreatitis [2]. BGH therapeutic management is essentially based on endoscopic removal in case of peduncolated lesion or surgical resection for broad-based structures or in case of an unsuccessful endoscopic procedure. The current case report focuses on a Brunner's gland hamartoma causing intermittent duodenal obstruction, and reviews its clinical presentation, histopathological features and its therapy management.

1.1. Case presentation

A 72-year-old Caucasian male in moderately good general health conditions (medical history was positive for mild arterial hypertension, hepatitis C and previous laparoscopic appendectomy appendicitis and cholecystectomy, patient's BMI was 25) was referred to our department by his family physician due to a six-month history of strong epigastric discomfort unrelated with food intake, recurrent vomiting and epigastralgia non-responding to PPI administration. He denied medications, allergies or previous colonoscopy or esophagogastroduonenoscopy (EGDS). Physical examination revealed a soft diffusely tender abdomen, without palpable mass. Haematological examinations were regular with haemoglobin concentration of 14.3 gr/dl and a coagulation profile within normal limits, normal amylase, lipase, liver function studies and neoplastic markers (Ca19.9, CEA). Upon referral, EGDS was then performed to investigate eventual upper intestinal obstruction. On endoscopy he was found to have a broad-based lesion on the posterior-inferior wall of the second duodenal portion with no signs of active ulcer, particularly close to the pylorus. EGDS also revealed a normal oesophagus with hyperaemic gastric mucosa. The lesion, which was by-passable by the endoscopic instrument, deformed pylorus and duodenal bulb and the duodenum appeared normal until the passage D1-D2. Resection was not considered due to the broad-based nature of the lesion and secondly to its extension (judged around 35mm).

No signs of active bleeding were observed and the lesion was then unsuccessfully biopsied. In order to radiologically investigate the lesion, we performed a high-resolution contrast-enhanced computer tomography (CT) that confirmed the presence of a whorled 40 × 16 mm vegetant lesion characterized by inhomogeneous arterial contrast enhancement. This lesion was established to be very close to the pyloric area, occupying almost completely the first part of the duodenum without a real surgical cleavage plan of resection. No dilatation of common bile duct (CBD) or pancreatic duct was found. We considered mandatory a better characterization of the lesion, and, particularly, ascertain its depth of involvement within the duodenal layers, and for this reason, an endoscopic ultrasound (EUS) was completed. The EUS showed a sub-mucosal lesion of mixed echogenicity measuring around 4cm in diameter arising from D2-D3, with disomogeneous parenchymal distribution visible following contrast enhancing with Sonovue™. The EUS did not display, like the CT-scan previously, any clear planes between the mass and the head of the pancreas nor lymphnodes involvement. All the above preoperative findings led our group to a provisional diagnosis of duodenal tumour with a second-line diagnosis of tumour of the head of the pancreas (Fig. 1). For these reasons, 10 days after the referral, the patient was taken up for a laparotomy with a plan to perform Whipple's pancreaticoduodenectomy. After a midline laparotomy, the distal stomach and duodenum were carefully kocherized and a mass lesion was felt inside the duodenum, just after the pylorus on palpation. The lesion seemed to be moderately mobile at the duodenal bulb with hard parenchymal consistency. No adhesions suggestive of a chronic inflammation status were noticed during the entire operation. Due to these features Whipple's procedure was at the moment interrupted in order to deeply investigate intraoperatively the origin of the mass. Via a duodenotomy a large polyp occluding almost the entire duodenal lumen was observed (Fig. 2). The ampullar papilla was not involved. A very short and large stalk characterized the polyps' base, so we decided to excise it by using mechanical endo-GIA (Fig. 3). The duodenotomy was then closed via a double layer reconstruction and a close-suction drain was positioned close to it. There was no lymph node involvement. The procedure was carried out by an experienced senior surgeon in an academic high volume hospital/setting.

The post-operative period was characterized in third POD by signs of intestinal perforation: fever, abdominal pain and mild leukocytosis (15.2 × 10/l). A CT scan showed a small collection near the duodenal suture. For this reason the patient was conducted in operation theatre but at the laparotomy no perforative sites or collections were observed (Clavien Dindo Grade IIIb). The residual postoperative period was uneventful. On POD 5 the NGT was removed and he tolerated diet, on POD 6 the patient reported bowel movements and on POD 7 he was discharged. Histopathological findings finally showed that the resected polyp (2,8 × 2,8 × 1,7 cm) was a Brunner gland hamartoma with hyperplastic glands without histopathological evidence of atypia. Immunohistochemical profile was positive for Cit7, Cit20, MUC1, MUC6 and MUC5AC. Brunner glands appeared mixed to adipose tissue and lymphoid infiltrate. Some gastric metaplasia areas were observed in the overlying mucosal layer (MUC1+ and MUC5AC+) (Fig. 4).

2. Conclusion

Brunner glands are acinotubular glands located in the duodenal bulb, and the proximal and distal duodenum with a decreasing prevalence [3] (rarely Brunner glands are located into the jejunum [4]) and an anti-acid function related to the secretion of a specific mucus coating the duodenal epithelium. Described for the first time by Cruveilhier and Savioli, in 1835 [5] and 1876 [6] respectively, between 150 and 200 cases of this type of benign tumour counts have been documented in English literature [7,8] and approximately 5% of these were duodenal masses [1].

In 1934, Feyter (cited by Goldman [9]) a classification of the abnormal proliferation of Brunner's glands into three different subsets has been proposed: type 1, diffuse nodular hyperplasia, type 2 circumscribed nodular hyperplasia, and type 3 glandular adenoma [10,11]. Moreover, this classification of hyperplasia of Brunner glands seems to be potentially designed as hamartoma, principally due to its histological features such as lack of surfacing capsule, network combination of acini, smooth muscle, adipose tissue, Paneth cells and mucosal glands, and the lack of cellular atypia. Pathogenesis of BGH still needs to be completely explored but the three most quoted hypotheses that have been proposed are the hyperchlorhydria hypothesis, the pancreatitis-related hypothesis and the inflammation-based hypothesis.

Zangara et al. [11] suggested that the natural anti-acid function (and subsequent hyperchlorhydria) could promote hyperplasia of Brunner's glands. This theory has been also reported in 1985, with a