CLINICAL CASE

Endoscopic resection of a large Brunner’s gland hamartoma

Résection endoscopique d’une hyperplasie des glandes de Brunner

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Summary Brunner’s Gland Hamartoma (BGH) is a benign tumor of the duodenum that can lead to gastrointestinal bleeding and intestinal obstruction. Endoscopic resection has seldom been reported. We describe the case of a duodenal obstruction caused by a large BGH (6 cm \times 4 cm). We report a 57-year-old woman hospitalized for tarry stools, weight loss and anorexia. Endoscopy revealed a large BGH (6 cm \times 4 cm). Endoscopic ultrasound (EUS) revealed a submucosal duodenal tumor. In this paper, we report a case of large hyperplasia of BGH, successfully treated by endoscopic technique.

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Résumé Les hyperplasies des glandes de Brunner sont des lésions bénignes du duodénum qui sont à risque de complications (hémorragies et occlusions). L’exérèse endoscopique de ces lésions a très peu été publiée. Nous rapportons un cas d’obstruction duodénale par une hyperplasie des glandes de Brunner de 6 cm \times 4 cm chez une patiente de 57 ans, hospitalisée pour mélénas, anorexie et amaigrissement. L’échoendoscopie a mis en évidence une lésion sous-muqueuse duodénale. Dans cet article, nous décrivons une large hyperplasie des glandes de Brunner traitée avec succès par endoscopie.

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Introduction

Brunner’s Gland Hamartoma (BGH) is a benign tumor of the duodenum, which is a polypoid proliferation of Brunner’s glands [1,2]. Clinical presentation is variable, including gastrointestinal bleeding and intestinal obstruction. Those
lesions are usually diagnosed by endoscopy and usually require surgical excision [3]. Endoscopic resection has seldom been reported [4]. We describe a case of a duodenal obstruction with anemia caused by a large BGH, which was removed by endoscopic polypectomy.

Case report

A 57-year-old woman presented with abdominal pain, anorexia and nausea. She had anorexia for two months. She had lost eight kilos in six weeks and reported black stools. The patient denied use of either aspirin or NSAID and never smoked cigarettes. She has been suffering from non-insulin dependent diabetes for the last eighteen months, which was treated with metformine. She also has suffered with advanced Parkinson's disease since 1984 and she has been treated, since February 2003, with an intracerebral neurostimulator to control her symptoms. Her medications included Levodopa 400 mg per day and metformine 1500 mg twice a day.

On physical examination, the patient's vital signs were normal: her heart and lung exams were normal. The abdomen was not distended and was without mass. There was no melena or rectorrargia on rectal examination. Laboratory values were significant for a drop in hemoglobin from 120 to 73 g/l within three months. She had low serum iron concentration and low transferrin saturation. Endocrine markers were negative except for chromogranin A. Blood level of Chromogranin A was 241 µg/l (normal rate inferior to 86 µg/l). Octreoscan scintigraphy detected a loss of octreotide in the duodenum.

Upper endoscopy revealed a large pedunculated polyp, approximately $6 \times 4$ cm in size from the proximal duodenum (Fig. 1). The polyp had a lobulated surface and was smooth with small areas of ulceration and exudates. No sign of bleeding was seen. Multiple endoscopic biopsy specimens showed Brunner’s gland hyperplasia. Endoscopic ultrasound (EUS) revealed a submucosal duodenal tumor of mixed echogenicity without involvement of the muscularis propria. The lesion appeared as pedunculated submucosal tumor in the duodenal bulb on small bowel barium study (Fig. 2). Abdominal CT scan revealed a lobulated mass in the proximal duodenum without lymph node (Fig. 3).

The tumor was completely resected in two phases. During the first procedure, a double channel gastroscope (Fujinon EG450D) was used and two metallic clips (Olympus hemoclip) were applied on the stalk to decrease the blood flow. No sign of strangulation appeared on the polyp after application of both hemoclips. The use of the detachable snare was not possible since the length of the tumor was too long. Only a part of the polyp ($4 \times 3$ cm) was removed by electrosurgical snare polypectomy, without bleeding, so as to have a histological diagnosis of the tumor. This first resection was complicated by delayed hemorrhage: 24 h after the procedure, which required a blood transfusion. Three units of packed red blood cells were transfused with a resulting hemoglobin level of 8 gm/dl. Pantoprazole 40 mg was given intravenously every 12 h.

Histologically, there was no evidence of malignancy and the histological diagnosis was BGH. The surface epithelium consisted of normal duodenal mucosa with areas of focal ulceration. Elongated duct-like structures were lined by...
similar epithelium. The main body of the polyp contained several lobules of mature Brunner’s glands with areas of cystic dilatation (Fig. 4). The polyp was believed to represent a BGH.

A repeat upper endoscopy was done three days later. The lesion had no sign of active bleeding. Six days after the first endoscopy, the resection was completed. This second resection has been made in two steps: first, the stalk was snared with a detachable snare and then transacted using coagulation current ERBE Endocut® (Tubingen, Germany). No bleeding was observed during the procedure.

At endoscopic and EUS control one month later, there was no residual tumor. The patient remained stable without symptoms and with a hemoglobin level of 140 g/L during the follow-ups at one, six and 18 months thereafter.

Discussion

BGH is a benign tumor, accounting for less than 1% of small intestinal tumors, 5% of all duodenal tumors and 11% of all small bowel benign tumors [1]. Around 100 cases of symptomatic BGH have been reported in the English literature [3,5]. Brunner’s glands consist of submucosal mucin-secreting glands located exclusively in the duodenum [6] and BGH are generally located in the duodenal bulb (57%) [3,5], ranging from 0.7 to 12 cm in diameter with a mean of 4.0 cm [3,7]. Nearly all BGH described are pedunculated (88%) [3,5].

Histologically, Brunner’s glands are more consistent with hamartomas as opposed to neoplastic adenomas. They contain a mixture of ducts, acini, smooth muscle and adipose tissue [5]. A BGH has no cellular atypia and is generally considered as non-malignant potential [8,9]. Malignant potential has been describe in one single case [5]. If BGH do not cause symptoms, some authors consider that there is no indication for resection [10,11].

As many as half of all patients with Brunner’s gland adenomas have symptoms [12]. Clinical presentation is variable and non-specific. Obstruction and hemorrhage are the most clinically significant presentations [3,5,13]. Abdominal pain was associated in 51% of patients and symptoms of anemia were observed in 43% of patients. Eleven percent of patients had no pain, nausea, vomiting or symptoms of anemia, whereas 7% had all of these symptoms [5]. Patients with symptoms usually present in the fifth and sixth decade of life [3]. Endoscopy and radiographic techniques may be useful in the diagnosis of these lesions. Endoscopy usually reveals a submucosal mass and standard mucosal biopsies are therefore of little benefit. The differential diagnosis of a duodenal mass includes adenoma, ampullary neoplasms, carcinoid tumor, leiomyoma, lipoma, neurogenic tumor, myoepithelial tumor, hamartoma, prolapsed pyloric mucosa or antral polyp and aberrant pancreas [5,14].

EUS can be helpful in the diagnosis and clearly demonstrated the layer origin of the BGH, which is exclusively limited to the submucosal layer [7].

Surgical resection had been the traditional method of treatment for symptomatic BGH [3,15]. Some cases of patients with BGH who have undergone polypectomy are available in the literature [4,16]. No complications of endoscopic therapy have been described and no recurrence has ever been reported in patients treated by endoscopy. In this case, the first resection was done to obtain histology of the tumor and was followed by a hemorrhage. The second resection, with the use of a detachable snare, was not followed by any complication. The endoscopic resection of BGH should be attempted with detachable snare to decrease the risk of bleeding, as it is recommended for a tumor with a large stalk.

In conclusion, BGH is a rare but benign cause of submucosal tumor of the duodenum. BGH can be managed by endoscopic excision. The use of a detachable snare is recommended during the resection of large BGH to avoid the risk of gastrointestinal bleeding.

References