ABSTRACT

Introduction
Brunner’s gland hamartomas are very uncommon. They are small, benign lesions, frequently located in the bulb of duodenum. Usually found incidentally during routine esophago-gastro-duodenoscopies, these lesions are frequently asymptomatic, but some patients may present with symptoms of duodenal obstruction or hemorrhage secondary to ulceration. Surgical excision is required, especially if the lesion has large dimensions. Pathological examination is the most important in confirming the diagnosis.

Case presentation
We report a case of a 64-year-old female patient, who presented to the emergency room for vomiting partially digested food, without blood. Upper digestive endoscopy revealed a polypoid mass, with a 1 cm stalk, located in the duodenal bulb. The polyp was successfully endoscopically resected and the pathological diagnosis was of Brunner’s gland hamartoma. Histologically, this polyp consisted of the components of Brunner’s gland cells, as well as glandular, adipose and muscle cells.

Conclusions

ENDOSCOPIC MANAGEMENT OF A RARE CASE OF OBSTRUCTIVE GIANT DUODENAL BRUNNER’S GLAND HAMARTOMA

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Brunner’s gland hamartomas are rare duodenal tumors occurring in middle-aged patients that present either with gastrointestinal hemorrhage, obstructive symptoms, or as an incidental finding. Surgical or endoscopic excision is uncomplicated, and the long-term outcome is favorable. The particularity of our case is given by the fact that this lesion was very large, it caused symptoms such as vomiting and was managed solely by endoscopic resection.

**Key words:** giant hamartoma, Brunner’s gland, endoscopic resection.

**INTRODUCTION**

Brunner’s gland hamartoma is a rare tumor, a nodular hyperplasia of the normal Brunner’s gland with an unusual mixture of normal tissues, including ducts, adipose tissue and lymphoid tissue. It represents 10.6% of duodenal benign tumor.1

Brunner’s gland hamartomas are typically found in the duodenal bulb and in the second portion of the duodenum, proximal to the sphincter of Oddi.2

Their main physiological function consists in producing a mucus-rich, bicarbonate-containing alkaline secretion that, among other features, neutralizes the acidic content of chime and gastric acid.3 Although proliferative lesions of Brunner’s gland do not have yet an established nomenclature, and distinction between various lesions is arbitrary, it is important to identify this pathology as it is benign, because there are no documented cases of malignancy.4

Patients with Brunner’s gland hamartoma may be asymptomatic and the lesion is usually discovered incidentally.5 Among possible symptoms and complications caused by Brunner’s gland hamartoma, duodenal obstruction, intussusception, obstructive jaundice, pancreatitis and bleeding are included. At upper digestive endoscopy, Brunner’s gland hamartoma is a submucosal mass that often has a stalk, which points out that an endoscopic mucosal biopsy may often be inconclusive as the biopsy device does not penetrate the submucosa.6 Macroscopically, this lesion is usually pedunculated, polypoid and microscopically it is composed of normal Brunner glands combined with variable amounts of adipose, smooth muscle, and lymphoid tissue, as well as with sclerotic areas of Brunner glands in some cases.

**Treatment of Brunner’s gland hamartoma** consists of endoscopic polypectomy, which is more cost-effective and less invasive than surgery. The medical decision usually depends on location and size of the tumor, as well as on the patient’s symptoms and status.8,9

This article reports a case of giant Brunner’s gland hamartoma, and reviews briefly its clinical presentations, pathological features and therapy.

**CASE PRESENTATION**

A 64 year-old male patient was admitted to our hospital for epigastric pain and vomiting, symptomatology that began about a month before, progressing towards a worse clinical status.

The physical examination revealed epigastric pain on palpation. Laboratory investigations revealed mild hypochromic and microcytic anemia.

Abdominal ultrasound performed in the emergency room showed the stomach filled with food debris, raising the suspicion of gastric stasis. Upper digestive endoscopy was performed and gastric stasis, along with a stalked giant polyp located on the posterior wall of the duodenum, near the junction of its first and second portions, were revealed. The surface of the tumor was smooth, with mild erosions and ulcers. Multiple biopsy specimens were taken and interpreted as “mucosal mild-medium atypia”. Tumor markers CA 19.9 and CEA were within normal range.
Endoscopic resection of the polyp was suggested to the patient and after careful consideration, together with the surgical team, endoscopic polypectomy was chosen as the first option. The resection was preceded by submucosal injection of diluted 5 ml polygel with epinephrine 1:10,000 solution into the base of the polyp, followed by efficaciously removed polyp using electrosurgical snare. Diffuse bleeding occurred immediately after polypectomy, but efficient endoscopic hemostasis was performed by using hemoclips.

The resected specimen showed a lobulated, polypoid mass, measuring 5 cm × 4 cm, projecting into the duodenum. The stalk measured 1 cm in diameter and 1 cm in length (Figure 1,2).

The site of resection was closed with hemoclips (Figure 3) to prevent post-polypectomy bleeding and perforation.

After 48 hours post-polypectomy, the patient was allowed to resume feeding. On follow-up, there was no rebleeding and no residual lesion.

Pathological evaluation of Brunner’s gland hamartoma revealed: on gross examination, the tumor showed a light brown colored, firm, pedunculated, polypoid mass, measuring 5x4x2.5 cm. The surface was smooth, with erosions and ulceration on the surface. The cut surface was firm, yellowish and lobulated, with a few slit-like spaces. On microscopic examination, the tumor was covered by small intestinal mucosa and composed of lobules of proliferated Brunner glands with adipose tissue metaplasia; in addition, there was moderate infiltration with lymphocytes, with a few lymphoid follicles. Some of the ducts were cystically dilated. The covering mucosa was flattened and showed hyperemia, foci of hemorrhage and mild infiltration of lymphocytes and plasma cells. The hamartomatous components were not present in the stalk of these histological findings. Finally, a diagnosis of Brunner’s gland hamartoma was made. (Figures 4-9)

**DISCUSSION**

Brunner’s gland hamartoma has a rather unclear etiology. The average size is between 0.5 cm and 12 cm. The most frequent location is on the posterior wall of the duodenum, at the junction between its first and the second part. In rare cases, it
Figure 4. (H&E 50x) Tumor covered by small intestinal mucosa (thin blue arrow) and composed of lobules of proliferated Brunner’s glands (large thick arrow) separated by irregular bands.

Figure 5. (H&E 50x) Lobules of proliferated Brunner’s glands separated by irregular bands of fibromuscular stroma with moderate infiltration by lymphocytes.

Figure 6. (H&E 50x) Lobules of proliferated Brunner’s gland with fibrous bands and adipose tissue metaplasia.

Figure 7. (H&E 50x) Lobules of proliferated Brunner’s glands with ducts separated by irregular bands of fibromuscular stroma and lymphocytes with a few lymphoid follicles.

Figure 8. (H&E 200) Covering mucosa showing hyperemia and mild infiltration with lymphocytes.

Figure 9. (H&E 200) Brunner’s glands with dilated ducts and fibrous bands.
can be located in the second and third part of the duodenum. Brunner’s gland hamartomas are usually asymptomatic, benign incidental findings, discovered when performing upper gastric endoscopy or barium swallow. The most common symptoms are upper digestive hemorrhage and emesis. In the case of a giant Brunner’s gland hamartoma, the upper digestive hemorrhage may be exteriorized as hematemesis or melena; occasionally, this bleeding may be massive and very rarely fatal.

According to the endoscopic classification, these hamartomas can be sessile, pedunculated, polypoid tumor masses or submucosal tumors.

Differential diagnosis should be done with: malignant tumors, adenomatous polyps, ectopic pancreas, leiomyoma, GIST, neuroendocrine tumors, ampulla of Vater and pancreatic neoplasm due to duodenal invasion. The treatment for Brunner’s gland hamartomas is recommended in symptomatic cases or to prevent complications (such as bleeding, obstruction). It is still controversial whether asymptomatic Brunner’s gland hamartoma incidentally discovered needs surgical removal. Endoscopic resection is indicated for stalked polyps or for confirming the benign character of the lesion. Surgical treatment is proposed when there is a pathological proven malignant component, submucosal development or for those lesions difficult to resect by endoscopy. Brunner’s gland hamartomas are benign, but there are documented cases in the literature with dysplasia found in the covering intestinal mucosa. There have been no reports of malignant Brunner’s gland hamartomas.

**CONCLUSIONS**

Brunner’s gland hamartoma is a rare duodenal tumor. Symptomatology is present in large duodenal tumors. An alternative to surgical resection is the endoscopic resection; pre-resection histological diagnosis is not always easy.

There are no reported recurrences following Brunner’s gland hamartoma removal and the long-term prognosis is excellent.

**REFERENCES**