

Brunner's gland hamartoma: a case report and review of literature



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BACKGROUND: *Brunner's hamartoma is a quite rare neof ormation whose origin is not completely clear.*

METHODS: *A case of a 69-year-old man with recurrent epigastric pain and an endoscopic finding of a duodenal mass is reported.*

RESULTS: *The complete removal of hamartoma can give a complete remission of symptoms.*

CONCLUSIONS: *Hamartoma can be only diagnosed by endoscopy and histological exam; its therapy is always surgical, also in order to avoid possible malignant degeneration.*

KEY WORDS: Brunner's hamartoma, endoscopy, Duodenal mass, Histological exam.

Introduction

The duodenal tumours represent 14 to 25 % of small bowel tumours and, among them, only about 5 % of benign lesions are Brunner's glands hamartomas¹. The first Brunner's glands hamartoma was described in 1876 by Salvioli. Up to today a little more than 100 cases have been reported. The preoperative diagnosis is difficult, mainly because of the unspecific symptoms as well as of the few cases described in the literature.

The following case may add some interesting suggestions to the clinical management of this rare disease.

Case Report

J.V., 69 year-old man, had been complaining of recurring epigastric pain for seven years, with increase few days before admission. He denied any other symptoms. The deep palpation in the epi-mesogastric region evoked pain. A barium enema of stomach and duodenum made before admission showed thick gastric plicas in

antrum and ipertonic and deformed duodenal cap with irregular edges. An endoscopy made 3 months before admission showed a polypoid formation localized in the duodenal cap, and, after a biopsy, the specimen was described as a hyperplastic polyp at histology. During the admission the patient underwent a chest X-ray and an electrocardiogram, the results of which were normal, whereas a transabdominal ultrasound showed a liver with a small (1 cm. large) unechogen image in the VII segment. Blood chemistry were within normal limits. The endoscopy showed a large base polyp (about 3 cm. large) localized immediately in the post-pyloric region, rising from the back-upper wall of the duodenal cap (Fig. 1), which was impossible to remove at that time because of its dimensions and localization.

Therefore, due to the increasing of the symptoms, the patient was referred to surgery. A xifo-umbilical median laparotomy was made, through the antrum the polyp was localized in the first part of the duodenum, immediately behind the pylorus. It presented an irregular shape and soft-elastic consistence. A pre-pyloric gastrotomy was performed, a mandarin-shaped polyp was found exposed in the gastric cavity through the pylorus and then removed with a loop silk suture placed at its base. The histopathological examination showed a polypoid sessile formation with a large attaching base (3x2x1.5 cm.) and a friable consistence, grey colour and irregular surface (Fig. 2); the neof ormation had a surface covered with a duodenal mucosa with a lot of Brunner's glands in all

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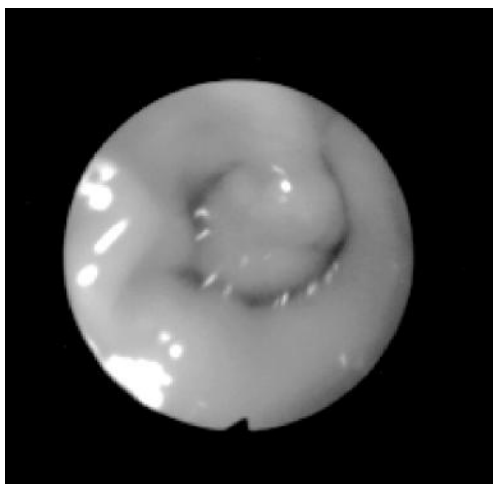


Fig. 1: Endoscopic vision of polypoid formation of Brunner's hamartoma.

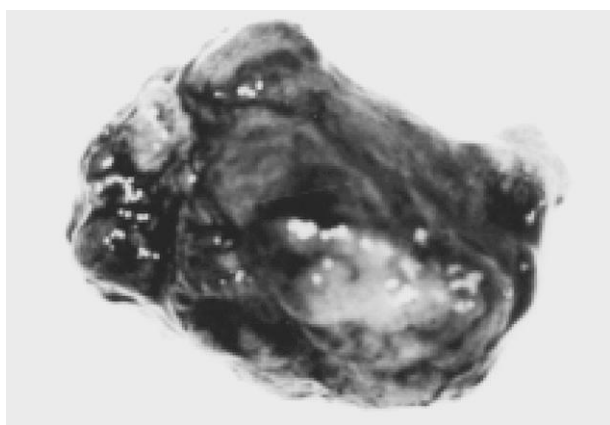


Fig. 2: Excised neof ormation.

sub-mucosa portion until the attaching base, these glands were clustered in large formation separated by smooth muscular cells rising from muscolaris mucosa. Moreover, a lot of enlarged cystic formation covered with a one layer cubic-cylindrical epithelium was observed (Fig. 3). There was no abnormal cells mitosis. Histopathological diagnosis of Brunner's glands hamartoma was made. The postoperative course was uneventful and the patient was discharged on the seventh postoperative day in good general conditions.

Conclusions

The nature of Brunner's glands hamartoma is still controversial, and some authors consider it as a simple hyperplasia of the glands, secondary to an overstimulation by a local irritation from gastric hypersecretion or sympathetic hyperactivity.

The prevailing opinion about the aetiology of these lesions is that they are hamartomas. Even though these tumours are benign, the literature reports a proven case

of cancer developing from Brunner's hyperplasia and a case associated with two *foci* of microcarcinoids. Brunner's gland hyperplasia is often associated with chronic renal failure, chronic pancreatitis and peptic ulcer disease, and has been known to regress, in some cases, after effective treatment of gastric hyperacidity². It is noteworthy that hamartomas are commonly associated with a generalized polyposis syndrome, going-ahead with an examination of the entire gastrointestinal tract³. Nevertheless, the hamartomatosis characteristics of this tumour are strengthened by the presence of ductular and acinar formation which is unlikely to be found in hyperplastic or neoplastic processes⁴⁻⁵, but some authors suspect the malignant power because of the finding, in some cases, of areas with epithelial dysplasia^{6,7}.

This pathology usually strikes patients between the fourth and the sixth decade of their lives⁷⁻¹¹; it can be discovered occasionally or it begins with dyspepsia and/or epigastric pain, as in our report, or haematemesis and/or obstruction. Naturally, the clinical picture is related to both the tumour localization and its dimensions, ranging from 2 cm. to more than 5 cm¹². The tumour can grow under the sub-mucosa layer and stretch the mucosa layer leading to superficial ulceration and bleeding, rarely intense⁴, simulating the clinical picture of a duodenal ulcer. Although barium series of the stomach and of the duodenum, endoscopy, with bioptic examination, still represents affordable diagnostic instruments¹³ to examine the neof ormation affecting the gastric-duodenal wall¹⁴. The endoscopic differential diagnosis should consider other benign tumours like lipoma, leiomyoma, and adenomatous polyp, as well as rare benign tumours and carcinoma⁴. The treatment is generally conservative in absence of symptoms. A revision of the literature shows that in selected situations a gastric-resection can be performed, mainly if the lesion is present in wide spread manner or when is not identified at the operation⁴, or to avoid the risk of a malignant transformation. The most characteristic aspect of this pathology is the absolute absence of specific symptoms and clinical signs. In a lot

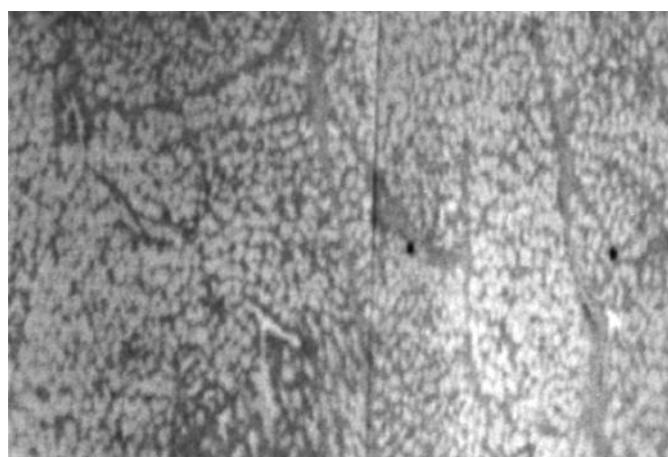


Fig. 3: Histological vision of operated piece.

TABLE I - Histological aspect found in Brunner's hamartoma in different cases

Authors	Histology
Iusco, Roncoroni <i>et al</i> 3	Brunner's glands hyperplasia with extension to the antro-pyloric region; gastric metaplasia of duodenum.
Gao, Zhu <i>et al</i> 6	Brunner's glands hyperplasia formed in lobules separated by intervening bands of fibrous and adipose tissue, ducts and well aggregated lymphoid.
Brookes, Manjunatha <i>et al</i> 7	complex and crowded architecture referable to Brunner's glands hamartoma; in deep layers some nuclear atypia with occasional mitotic figure; predominant lobules of relatively bland Brunner's glands; foci of obvious dysplasia and cytological changes.
Stolpman, Hunt <i>et al</i> 8	Histology referable to Brunner's gland hamartoma.
Kouraklis, Kostakis <i>et al</i> 4	Brunner's type without atypia coexisted with heterotopic pancreatic acini and ducts.
Coppola, Karl <i>et al</i> 9	Cells arranged in lobulated fashion and compartmentalized by fibromuscular septa and cystic dilatation of glands; columnar cells with vacuolated cytoplasm and basally located nuclei.
Bastounis, Pikoulis <i>et al</i> 10	Hyperplastic glands formed in lobules which were surrounded by bundles of well differentiated Brunner's gland with mucos-secreting epithelial cells.

of cases this pathology occurs with the haematemesis, but this sign is less specific too, so the diagnosis of Brunner's glands hamartoma can be made by exclusion of pathologies that frequently caused bleeding of the upper gastrointestinal tract. In this report the Authors demonstrate that, despite of the absence haematemesis and/or obstructive signs¹²⁻¹³, a good anamnesis together with endoscopy and upper gastrointestinal tract barium enema are sufficient for the diagnosis of epigastric hamartoma. Therefore the A.A emphasizes the importance of a careful analysis of symptoms and instrumental examination for a correct diagnosis and therapy¹⁵.

Riassunto

INTRODUZIONE: L'amartoma di Brunner è una neoformazione piuttosto rara la cui origine non è ancora ben definita.

METODO: Riportiamo un caso di 69 anni con dolore epigastrico ricorrente e riscontro endoscopico di una massa duodenale.

RISULTATI: La completa rimozione dell'amartoma può dare una completa remissione dei sintomi.

CONCLUSIONI: L'amartoma può essere diagnosticato solo tramite esame endoscopico ed istologico; la sua terapia è sempre di tipo chirurgico, anche allo scopo di evitare possibili degenerazioni maligne.

PAROLE CHIAVE: Amartoma di Brunner, Endoscopia, Massa duodenale, Esame istologico.

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