

Unexpected cause for duodenal obstruction: Brunner's gland hyperplasia

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Key words

Brunner gland • Hyperplasia • Malignancy • Adenoma • Duodenum

Summary

Brunner's gland hyperplasia is a rare benign lesion arising from the duodenum. It is often an incidental finding on endoscopy with the majority of patients being asymptomatic. It may also present with various symptoms depending on location and tumor size, such as gastrointestinal bleeding, obstruction and abdominal pain. We report an unusual case of large Brunner's gland hyperplasia in 72-years old man, admitted to hospital for epigastric pain, recurrent vomiting and significantly weight loss. Upper endoscopy showed an obstructive submucosis tumour of the bulb. Abdominal com-

puted tomography scan revealed a circumferential thickening and stenosing mass of the first part of the duodenum leading to severe obstruction. Though pre-operative biopsies were negative, imaging studies and endoscopy were strongly suggestive of malignancy and the patient underwent duodenocephalopancreatectomy.

Brunner's gland hyperplasia may have unusual presentation, mimicking malignancy. Therefore, extensive pre-operative evaluation, including repetitive tumor biopsies, is necessary to avoid radical surgical procedure.

Introduction

Brunner's glands are branched acinotubular glands normally found in the deep mucosal or submucosal layers of proximal duodenum and secrete alkaline-based mucus to protect the duodenal lining from gastric acid ¹. Brunner's gland hyperplasia, also known as Brunner's adenoma is a rare, benign, proliferative lesion characterized by lobules of Brunner's glands that are increased in both size and number ². We report an unusual case of Brunner's gland hyperplasia leading to severe duodenal obstruction mimicking a malignancy and requiring therefore radical surgical procedure.

Case report

A 72 year old male with no history, presented with epigastric pain and recurrent vomiting occurring tardively after meals during 12 months, associated with significantly weight loss.

Physical exam revealed abdominal distension and epigastric pain in palpation.

Esophagogastroduodenoscopy showed an obstructive

submucosal tumour of the bulb. Biopsies were small and negative for malignancy.

Abdominal computed tomography scan showed a slightly enhanced, circumferential thickening and stenosing 3 cm mass of the first part of the duodenum, abutting gallbladder and head of pancreas without loss of fat planes (Fig. 1).

Laboratory data showed low hemoglobin (9 g/dl) and albumin (31 g/l) levels. Tumor markers (Carcino-embryonic antigen and CA19-9) were within normal limits.

Fig. 1. Computed tomography of the abdomen shows circumferential thickening and stenosing mass of the first part of the duodenum (a) transverse cut and (b) coronal cut



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Patient underwent cephalic duodenopancreatectomy (Whipple's procedure) in our institute with clinical diagnosis of duodenal carcinoma and the specimen was sent for histopathological examination.

Gross examination noted circumferential thickening of the first part of the duodenum. When opened, mucosal surface exhibited 2,5 x 2 cm polypoid mass that was 1 cm from the ampulla. It was grey white on section with few cystic areas.

Histologically, tumor showed proliferating Brunner's glands in a lobular pattern, constituting about 75% of the thickness of the duodenal wall. They extend focally through the muscularis mucosae and are separated by delicate fibrous septa (Fig. 2) Some glands were dilated (Fig. 3). Cells were columnar with basally located round nuclei and bland neutral mucin-containing cytoplasm. No mitosis, necrosis or atypia were observed (Fig. 4).

Patient recovered well and remained symptom free at 12 months follow up.

Discussion

Brunner's gland hyperplasia is a benign lesion which most commonly encountered in the duodenal bulb ². There is no sex predilection and patients present in the fifth to sixth decades of life ³.

It is often an incidental finding on endoscopy with the majority of patients being asymptomatic, but it may be seen as one of the constellation of changes typical of peptic duodenitis or associated with end-stage renal disease and uremia. Depending on location and tumor size, Brunner's gland hyperplasia can result in dyspepsia, vomiting, gastrointestinal bleeding, obstruction and abdominal pain ^{3 4}.

The etiology of Brunner's gland hyperplasia is not known. It is hypothesized that excess gastric acid secretion or increased inflammation may lead to hyperplasia ². Polyps resulting from Brunner's gland hyperplasia are typically small (< 1 cm). Occasionally, they may be large in size with clinical manifestations of hemorrhage or obstruction, as in our case ⁵.

Imaging studies are of little help in the diagnosis ¹.

Endoscopically, Brunner's gland hyperplasia can be nodular or polypoid mimicking gastrointestinal stromal tumor, lymphoma, carcinoid or Peutz Jeghers polyp ³. It can also be diffuse with thickening of the duodenal wall and hence can be misdiagnosed as malignancy.

Even if endoscopy and endoscopic ultrasound can sometimes be helpful, definitive diagnosis requires pathologic examination ⁶.

Histologically, proliferating glands extend into the lamina propria and are separated by delicate fibrous septa. Cystically dilated glands have been reported, but this finding is relatively uncommon. The cells constituting the glands are cytologically bland with abundant neutral mucin cytoplasm and small, basally located nuclei with minimal or absent mitotic activity ².

Fig. 2. Proliferating Brunner's glands extend through the muscularis mucosae in a lobular pattern (Hematoxyllin Eosin x 100).

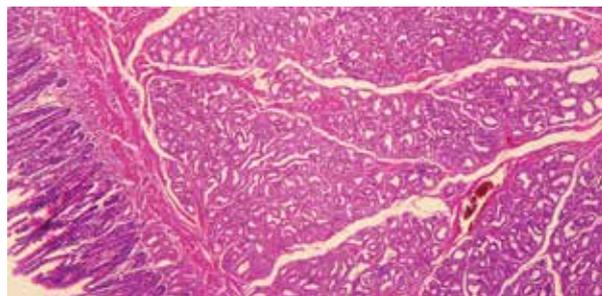


Fig. 3. Cystically dilated ducts and glands are interposed between small glands (Hematoxyllin Eosin x 200).

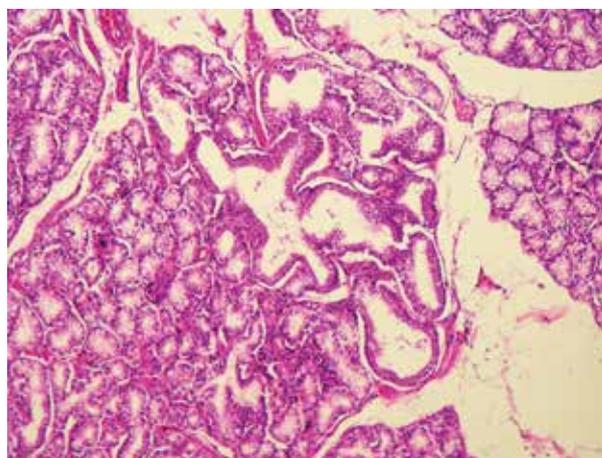
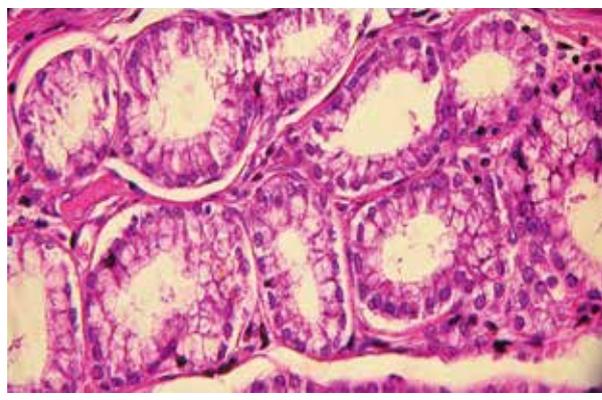


Fig. 4. High-power examination demonstrates cytologically bland neutral mucin-containing cells with basal nuclei (Hematoxyllin Eosin x 400).



The diagnostic criteria for Brunner's gland hyperplasia in endoscopically obtained biopsy specimens are subjective because Brunner's glands may be focally present in the lamina propria of the normal duodenum. Some authors require the presence of lobules of Brunner's glands within the mucosa in at least 50% of the length of a biopsy specimen to establish a diagnosis of hyperpla-

sia⁷. Furthermore, diagnosis in small biopsy specimens is difficult, as in our case, and a deeper samples would be more contributive. Exceptional cases of malignant transformation of Brunner's gland hyperplasia were reported⁸. These cases do not unequivocally demonstrate de novo neoplasia within Brunner's glands, as opposed to secondary involvement of Brunner's glands by dysplasia or carcinoma arising in the surface mucosa.

When Brunner's gland hyperplasia is symptomatic or leads to complications or when definite diagnosis is necessary, the treatment of choice is mass removal by endoscopic or surgical procedure⁹.

Strategies of resection suggest that size and pedunculation were important characteristics in determining amenability for endoscopic removal. This approach is less invasive and safer than surgery, but it can be limited by difficult anatomical sites¹.

Surgery is usually discussed for polyps 5 cm or larger. Even in these cases, cephalic duodeno-pancreatectomy is exceptionally made since it leads to high morbidity and mortality rates. Usually, surgical polypectomy, duodenal wedge resection, or partial gastrectomy extending to the duodenal bulb are proposed for Brunner's hyperplasia^{1 10}.

As in our case, when the tumor is discovered during an obstructive syndrome, radical surgery is often performed especially to eliminate malignancy^{1 3 5 9-11} (Tab. I). Some authors justified this attitude by the fact that consequences of leaving an undiagnosed pancreatic cancer are worse than the risk of undergoing duodenopancreatectomy^{10 11}.

In all cases, repeated and deeper duodenal biopsies could be useful to avoid "overtreatment".

Tab. I. Main clinico-pathological features of patients with Brunner's gland hyperplasia discovered during an obstructive syndrome.

Authors	Sex/age (years)	Symptoms	Abdominal CT	Esophagogastro duodenoscopy (EGD)	Initial Biopsy	Treatment	Macroscopic features
Duminda BS ¹	F/59	Acutely worsening oral intolerance + weight loss	Distended stomach with retention of contrast material	Pyloric channel narrowing + a clean-based antral ulcer	Negative for malignancy	Distal gastrectomy with gastrojejunostomy	2 cm × 2 cm polypoid mass
Sen R ⁵	M/42	Epigastric pain + recurrent vomiting	Circumferential thickening of the 2 nd DD	Nodular stricture at D1/D2 junction	Well differentiated adenocarcinoma.	Cephalic duodeno-pancreatectomy	Diffuse grey white area of 4 × 4 cm with cystic and hemorrhagic zones
Cheung, TT ⁵	M/70	Repeated vomiting + melena	Large tumour occupying the the 1 st and 2 nd DD	ND	ND	Cephalic duodeno-pancreatectomy	Well-encapsulated submucosal tumour of 10 × 8 × 6 cm
Lee WC ¹⁰	M/64	Dyspepsia, vomiting + weight loss	2.5 cm mass of the 2 nd DD with loss of fat plane beside the pancreas	Infiltrating and obstructive mass of the 2 ^{ed} DD	Chronically active duodenitis	Cephalic duodeno-pancreatectomy	2.5 cm-sized mass in the 2 nd DD
Lusco D ⁹	M/60	Belt-like upper abdominal pain	5.5 cm bulky mass of the 1 st and 2 nd DD + Ectasia of Wirsung duct	Large mass of the 1 st and 2 ^{ed} DD + impossible cannulation of the papilla	Aspecific duodenitis	Cephalic duodeno-pancreatectomy	A hard mass, vegetating in the 1 st and 2 nd DD lumen + intense peri-duodenitis.
Hwang IT ¹¹	M/44	Recurrent vomiting + epigastric pain	Dilatation of the CBD and the main pancreatic duct + wall thickening of proximal DD	Polypoid mass with mucosal swelling and nearly complete obstruction of the bulb	Non-specific duodenitis	Cephalic duodeno-pancreatectomy	Circumferential enlargement of the duodenal mucosa in the proximal DD
Our case	M/72	Epigastric pain, recurrent vomiting + weight loss	Circumferential thickening and stenosing 3 cm mass of the 1 st DD	Obstructive submucosal tumour of the bulb	Negative for malignancy	Cephalic duodeno-pancreatectomy	2,5 × 2 cm polypoid mass with cystic areas causing circumferential thickening of the 1 st DD

M: Male; F: Female, ND: Not done; DD: Duodenum; CBD: common bile duct.

Conclusion

Brunner's gland hyperplasia is a rare benign lesion arising from the duodenum. It may have an unusual presentation, mimicking malignancy. Thus, extensive pre-operative evaluation including repetitive and deep tumor specimen is necessary to avoid radical surgical procedure.

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