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 submucosal tumour of the bulb. Abdominal computed 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.

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. Esophagastroduodenoscopy 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 gall-bladder 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
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. There is no sex predilection and patients present in the fifth to sixth decades of life. 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.

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. 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.

Imaging studies are of little help in the diagnosis. Endoscopically, Brunner’s gland hyperplasia can be nodular or polypoid mimicking gastrointestinal stromal tumor, lymphoma, carcinoid or Peutz Jeghers polyp. 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.

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. 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 hyperplasia.
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 reported. 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 procedure.

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 sites.

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.

As in our case, when the tumor is discovered during an obstructive syndrome, radical surgery is often performed especially to eliminate malignancy (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.

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.

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

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.

References