Brunner Gland Cyst: Two Cases of a Rare Entity and Review of the Literature

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ABSTRACT

Objective: Brunner gland cysts are rare with only 14 cases reported in the literature. The term has been used to consolidate the following entities: Brunner’s gland cyst, cystic Brunner’s gland hamartoma, Brunner’s gland cystadenoma and mucocele of the Brunner gland.

Methods: We present the clinico-pathological features of two cases of Brunner gland cyst along with a review of the literature.

Results: Brunner gland cyst affects slightly more men than women. The age range is 30 to 72 years with an average of 55 years. The lesions range from 1 to 5 cm in size, the average being 2.3 cm in greatest dimension. The majority of lesions are found incidentally in patients with symptoms not specifically related to the lesion. They affect predominantly the first and second portion of duodenum. The majority of the lesions are single and pedunculated filling the submucosa. They can be unilocular or contain multiple cysts divided by fine septae. The epithelial lining is of columnar and clear cells with basally located nuclei.

Conclusion: Brunner gland cysts are benign lesions that are usually not diagnosed pre-operatively. The cases described here add awareness of this entity among pathologists and gastroenterologists allowing for a better recognition of this rare entity.

KEY WORDS: Duodenum; Brunner gland; Cystadenoma; Cyst; Hamartoma.

INTRODUCTION

Cystic lesions of the Brunner gland are rare. They have been referred to as Brunner’s gland cyst, cystic Brunner’s gland hamartoma, Brunner’s gland cystadenoma and mucocele of the Brunner gland. Recently these entities have been grouped under the term Brunner gland cyst. Even with these combined entities, there are only fourteen Brunner gland cysts reported in the English literature. We present two new cases of Brunner gland cyst and compared them to those previously reported. Their clinical presentation and similarities are summarized.

CASE PRESENTATIONS

A 69-year-old Hispanic male was referred by his primary care physician to our institution for further studies of a duodenal lesion that was discovered after a workup for acid reflux. Endoscopic ultrasound showed a 17x6 mm septated cyst that appeared to arise from the submucosa of the second portion of the duodenum (Figures 1 and 2). No other abnormalities were noted. The cyst was resected after lifting the lesion with saline and methylene blue.

The specimen consisted of an ovoid lesion with a 0.5 cm cystic cavity. Microscopic examination revealed a submucosal multicystic lesion lined by tall, columnar cells with round basally located nuclei (Figures 3 and 4). Most of the cells had abundant clear cytoplasm similar...
to those seen in the adjacent normal Brunner glands. In other areas, the lining cells were smaller with pink cytoplasm. The cysts were filled with a serous-like fluid. Necrosis, mitotic activity, and nuclear atypia were absent. The cells contained neutral, Periodic Acid Schiff (PAS) positive mucin, similar to the adjacent Brunner’s glands.

The second patient is a 52-year-old Caucasian man referred to our institution because of epigastric pain and a duodenal mass found at endoscopy. Another endoscopy was performed along with endosonography. An intramural (subepithelial) lesion was found in the second portion of the duodenum. The lesion appeared to originate from within the submucosa (layer 3). The possibilities of pancreatic rest or Brunner’s gland hyperplasia were considered. The lesion was hypoechoic, heterogeneous and multicystic that measured 14.5 mm x 10.9 mm. The outer margins were well defined. An intact interface was seen between the mass and the adjacent structures suggesting a lack of invasion. An endomucosal resection of the mass was performed.

Histologically, the multicystic lesion was located in the submucosa of the duodenum and was composed of tall cells with basally located round nuclei. The cytoplasm was clear and abundant. In areas, there were aggregates of glands by the single row of epithelial cells lining the cysts creating a nodular configuration. No mitosis, necrosis or atypia was observed.

**DISCUSSION**

Rankin, et al. reported the first cystadenoma of the duodenum in 1933. Varnholt, et al. were the first to group Brunner gland cyst, Brunner cyst, mucocele of Brunner gland, and cystic Brunner’s gland hamartoma as one entity. Later, Powers, et al. added Brunner gland cystadenoma to this group. There are fourteen cases reported in the English literature. Information from these cases and the current two is summarized in Table 1.

Brunner gland cysts are benign, as evidenced by the fact that the lesion described by Golan, et al. was present for 15 years without any long-term consequences. They are widely believed to be retention cysts that develop after obstruction of a larger duct of the Brunner gland outflow tract.

Based on this case and the well-described fourteen previously reported cases, the following conclusions can be made. The lesion can occur throughout the duodenum, specifically any place that contains Brunner glands. The large majority are single lesions with one patient reported to have two. Most of the lesions are pedunculated and located in the submucosa, but sessile lesions have been reported and one was transmural. While some are unilocular, others contain multiple cysts divided by fine septae. The cysts are lined by mucinous, columnar cells with basally located nuclei, resembling normal Brunner gland cells. Eosinophilic cells lining the cyst wall are also common. The case described by Chatelain, et al. showed ciliated columnar cells. Atypia is rare, and mitosis is only reported in one case.
<table>
<thead>
<tr>
<th>Year</th>
<th>Authors</th>
<th>Diagnosis</th>
<th>Age, Sex</th>
<th>Clinical presentation</th>
<th>Location</th>
<th>Size (cm)</th>
<th>Architecture</th>
<th>Depth</th>
<th># of cysts</th>
<th>Histology characteristics</th>
</tr>
</thead>
<tbody>
<tr>
<td>2015</td>
<td>Moul, et al. (current case)</td>
<td>Brunner gland cyst</td>
<td>69, M</td>
<td>Incidental finding in a patient with acid reflux</td>
<td>Second portion of duodenum</td>
<td>1.7 x 0.7 x 0.3</td>
<td>Sessile</td>
<td>Sub-mucosa</td>
<td>Single</td>
<td>Focal eosinophilic cells</td>
</tr>
<tr>
<td>2015</td>
<td>Moul, et al. (current case)</td>
<td>Brunner gland cyst</td>
<td>52, M</td>
<td>Epigastric pain</td>
<td>Second portion of duodenum</td>
<td>1.4 X 1.0</td>
<td>Polypoid</td>
<td>Sub-mucosa</td>
<td>Single</td>
<td>Focal eosinophilic cells</td>
</tr>
<tr>
<td>2011</td>
<td>Galiatsatos</td>
<td>Brunner gland cyst</td>
<td>72, M</td>
<td>Incidental</td>
<td>Distal end of the second part of the duodenum</td>
<td>2</td>
<td>Polypoid</td>
<td>Not specified</td>
<td>Not specified</td>
<td>Normal appearing duodenal mucosa and dilated lymphatic channels and capillaries (pictures not available for review)</td>
</tr>
<tr>
<td>2008</td>
<td>Powers, et al.</td>
<td>Brunner gland cyst</td>
<td>46, F</td>
<td>Dyspepsia and odynophagia</td>
<td>Two lesions located in the second part of the duodenum</td>
<td>1.8 and 2.2</td>
<td>Sessile</td>
<td>Sub-mucosa</td>
<td>Single</td>
<td>Cystic spaces lined by hyperplastic cells similar to Brunner glands cells.</td>
</tr>
<tr>
<td>2007</td>
<td>Varnholt, et al.</td>
<td>Brunner gland cyst</td>
<td>41, F</td>
<td>Incidental finding in a women being treated for H. pylori-associated gastritis.</td>
<td>Not specified</td>
<td>1.1 x 0.9 x 0.6</td>
<td>Sessile</td>
<td>Sub-mucosa</td>
<td>Single</td>
<td>Cyst lined by a simple cuboidal-to-columnar epithelium.</td>
</tr>
<tr>
<td>2003</td>
<td>Yamakawa, et al.</td>
<td>Cystic Brunner’s gland hamartoma</td>
<td>64, F</td>
<td>Epigastric pain</td>
<td>Descending duodenum (second portion)</td>
<td>2.4 x 1.1 x .5</td>
<td>Pedunculated</td>
<td>Sub-mucosa</td>
<td>Multiple</td>
<td>Multilocular cysts lined by columnar epithelium ; dilated ductal structures.</td>
</tr>
<tr>
<td>2002</td>
<td>Chatelain, et al.</td>
<td>Brunner gland hamartoma with pre-dominant adipose tissue and cysts</td>
<td>43, M</td>
<td>Two-day history of regurgitation.</td>
<td>Duodenal bulb (first portion)</td>
<td>3.5</td>
<td>Pedunculated</td>
<td>Sub-mucosa</td>
<td>Multiple, ciliated</td>
<td>Prominent mature adipose tissue, hyperplastic lobules of Brunner glands</td>
</tr>
<tr>
<td>1980</td>
<td>Fisher</td>
<td>Mucocele of Brunner gland</td>
<td>45, F</td>
<td>Right upper quadrant pain intermittently for two months</td>
<td>Not specified</td>
<td>Not specified</td>
<td>Not specified</td>
<td>Not specified</td>
<td>Sub-mucosa</td>
<td>Intact duodenal mucosa with prominently dilated glandular spaces, one being large and cystic.</td>
</tr>
<tr>
<td>1978</td>
<td>Golan J, et al.</td>
<td>Cystic Brunner’s gland hamartoma</td>
<td>64, M</td>
<td>Acute gastrointestinal bleeding; had a known duodenal polyp for 15 years</td>
<td>First part of the duodenum</td>
<td>5 x 4 x 2</td>
<td>Pedunculated</td>
<td>Sub-mucosa</td>
<td>Multiple</td>
<td>Cysts were lined by columnar and cuboidal epithelium. The surface epithelium was partly pyloric and partly duodenal.</td>
</tr>
</tbody>
</table>
Taura M, et al. 10 Brunner’s cyst 54, F
Nausea, vomiting, and epigastric pain for several days
Duodenal bulb (first portion) 1.5 Not specified Sub-mucosa Single
Lined by tall columnar cells with basal nuclei. Multinucleated cells were intermingled with the epithelial lining cells.

Wolk DP, et al. 11 Brunner’s gland cystadenoma 68, M
Presented with 20 lb weight loss and eructation.
Third portion of the duodenum, located on the posterior wall 3 x 3 Not specified Sub-mucosa Multiple
Multiple fluid-filled cystic spaces confined to the submucosa, lined by Brunner glands

Hately 12 Brunner’s gland cyst 54, M
Two year history of intermittent vomiting, now after every meal.
First part of the duodenum. 1.5 Sessile Sub-mucosa Single
Cyst lined by columnar epithelium and had a direct origin from one of the Brunner glands.

Rankin and Newell 1 Simple, multilocular cystadenoma 54, M
18 m history of pernicious anemia and ulcer-like dyspepsia.
- 2 Not specified Sub-mucosa Multiple
Cyst lined by cuboidal epithelial cells.

Table 1: Brunner gland cysts reported in the English literature: Patient characteristics, clinical presentation, diagnostic modality and lesion location, and histological characteristics.

There is no sex predilection; fifty-three percent were male. The age range was 30 to 72 years of age, with the average being 55 years. The lesions ranged from 1 to 5 cm in size, the average being 2.3 cm in greatest dimension. While increased size correlated with more clinical symptoms, the patient with the largest reported lesion was not symptomatic from the lesion but from an adjacent ulcer. Nonetheless, most clinical symptoms are regurgitation, vomiting, and epigastric pain. However, it may be discovered incidentally for unrelated symptoms. Most often, the lesions are located in either the first or second portion of the duodenum. This corresponds to the most common locations of the Brunner’s gland, which are mostly concentrated in the first portion and gradually decrease in number throughout the length of the duodenum.

The differential diagnosis on endoscopic imaging includes duplication cysts, lipomas, neuroendocrine tumors and Brunner gland hamartomas. Using endoscopic ultrasound it is possible to differentiate the echostructure (cystic, solid, hypo- or hyperechoic) and wall layer of involvement. The differential diagnosis of hypoechoic/anechoic duodenal lesions would be mainly duplication cysts, stromal cell tumors and neuroendocrine tumors. Our first patient underwent resection to ensure his was not a cystic neuroendocrine lesion. Histologically the main differential diagnosis is Brunner’s gland hamartoma, formerly known as Brunner’s gland adenoma. These lesions are admixtures of Brunner’s glands, ducts, adipose tissue and lymphoid tissue. While their ducts may be dilated, cystic lesions are not characteristic of this entity.

Brunner gland cysts are rare benign lesions of the duodenum. The treatment is surgical excision or polypectomy. Increased awareness of these lesions helps for a better recognition of this entity.

CONFLICTS OF INTEREST: None.

DISCLOSURES
No consent is required to our article publication referenced above.

REFERENCES


