CASE REPORT

Giant Brunneroma: Clip-assisted Polypectomy. A Case Report and Review of Literature

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ABSTRACT
Brunner glands are compound tubular submucosal glands typically found in the duodenal bulb. The most common benign tumors of the small intestine are adenoma, and 25% of these occur in the duodenum. Among the benign tumors of the duodenum, 5-10% arise from the Brunner glands. Most of the literature describes their presentations as ranging from benign, nonspecific, epigastric discomfort to obstruction and intestinal bleeding. A good percentage of large tumors (>2 cm) are surgically resected; however, there has been an advancement to remove them endoscopically. We present one of such case in which excision was done by clip assisted polypectomy. (J Dig Endosc 2012;3(1):16-18)

Keywords: Duodenal Hamartoma - Clip Assisted Polypectomy

Introduction
Brunner's gland hamartoma, is a rare, benign, proliferative lesion arising from the Brunner's glands of the duodenum, accounting for 10% of benign tumors of the duodenum. Usually asymptomatic and lesions are discovered incidentally. Fifty-seven percent of the tumors originate from the duodenal bulb and the incidence of the tumor decreases as the distance from the pyloric ring grows[1]. These lesions manifest occasionally as a rare cause of duodenal obstruction or upper gastrointestinal haemorrhage, and require surgical excision. Here we report a case of Brunner's gland hamartoma presenting as non cardiac chest pain, and review of literature.

Case History
A 42-year-old man with a 6 month history of vague intermittent retrosternal discomfort evaluated earlier by cardiologist. Symptoms were vague and intermittent treated symptomatically with no relief. No history of nausea or vomiting. He denied poor appetite, weight loss or altered bowel habits. He was otherwise healthy with no specific family or past medical history. Physical examination and laboratory evaluation revealed no abnormalities. Cardiac evaluation including ECG, ECHO and treadmill were normal. Upper GI endoscopy was performed which revealed a 4 x 3cm long pedunculated large polypoidal mass seen in the anterior wall of first part of duodenum entering into second part of duodenum(Figure 1).

Endoscopic clip assisted polypectomy was planned. The tumor was completely resected in single sitting. Using, regular gastroscope (Olympus GF 150 ) two metallic clips (Olympus hemoclip) were applied on the stalk to decrease the blood flow(Figure 1). Polyp (4cm×3cm) was removed by electrosurgical snare polypectomy, without bleeding and sent for histopathologic examination which confirmed a brunner gland hamartoma. Histologically, the presence of mature but disorganized tissue composed mainly of lobules of Brunner glands lined by cylindrical clear mucous cells were seen. The muscularis propria is split with proliferation in the lamina

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polyp located in the first part of the duodenum, measuring from 0.7 cm to 12 cm (average 4 cm), similar to our patient. Brunner’s gland hamartomas are mostly located in the duodenal bulb (57%). Rarely, they may be found in the second (27%) and third (5%) parts of the duodenum, the pyloric canal (5%), the fourth part of the duodenum, jejunum and proximal ileum[5]. Brunner’s gland hyperplasia is characterized by lobules of glands that are increased in both size and number, and are separated by thin fibrous septae. These lobules extend from the submucosa into the lamina propria and within the mucosa alone and constitute at least 50% of the length of the biopsy specimen. Brunner’s gland hamartoma consists of an admixture of fibromuscular and adipose tissue within and surrounding cystically dilated hyperplastic lobules of Brunner’s glands[6]. Brunner’s gland adenoma can be asymptomatic and are found incidentally. Symptomatic tumors can present as gastrointestinal bleed or obstruction. The clinical manifestations of the former are gastrointestinal hemorrhage, due to ulceration or erosion of the tumor. Obstructive tumors occur when hyperplasia diffuses or a single adenoma grows too large, causing epigastric bloating, discomfort, vomiting or weight loss. Duodenal intussusception has been rarely reported[7], probably because of the fixation of duodenum to the posterior abdominal wall. Hamartoma is a benign lesion, and very few cases of Brunner gland hamartoma have been reported in association with epithelial dysplasia, duodenal adenocarcinoma, and carcinoid tumors[8]. Prior to treatment, all patients diagnosed with duodenal polyps should undergo screening for colonic polyposis, because duodenal adenomas are common in patients with familial adenomatous polyposis (30% to 70%) and vice versa[9].

Upper gastrointestinal endoscopy with biopsy is the diagnostic method of choice. The differential diagnoses include polypoid adenoma of the superficial mucosal glands, adenoma of the islet cells, aberrant pancreatic tissue, lipoma, angioma, leiomyoma, gastrointestinal stromal tumor and malignant neoplasms like carcinoid, adenocarcinoma, lymphoma, leiomyosarcoma and metastasis. Endoscopic mucosal biopsy, however usually gave a negative result because the tumor are covered entirely with thick intact duodenal mucosa in the biopsy sites and the biopsy was often not deep enough to reach the submucosal tumor tissue[10]. Symptoms of our patient can be explained by an intermittent intussusception of polyp distally into the duodenum, as it disappeared after polypectomy. Brunner’s gland hamartoma of the duodenum can be removed endoscopically because it is usually benign. Malignant transformation has been reported rarely[11]. It has been suggested that endoscopic or surgical removal of symptomatic Brunner’s gland hamartoma can prevent the development of complications and endoscopic polypectomy represents the ideal approach depending on the site and size of Brunner’s gland hamartoma and the presence of a peduncle[12]. We performed clip assisted polypectomy as earlier described by YY Chen et al[13]. Very few such cases reported in literature. Open surgical excision is reserved for

Discussion

Brunner’s glands consist of submucosal mucin-secreting glands located exclusively in the duodenum. They extend from the pylorus distally for a variable distance, usually stopping at the first and second portions of the duodenum, and less often stopping at the third and fourth portions. Hamartomas are rare duodenal lesions and were first described by Cruveilhier at the end of the 19th century[2]. About 50% of patients complain of epigastric pain, gastrointestinal hemorrhage, or rarely intussusception; in the remaining cases, hamartoma is an incidental finding[3,4]. It usually presents as a pedunculated polyp located in the first part of the duodenum, measuring from 0.7 cm to 12 cm (average 4 cm), similar to our patient. Brunner’s gland hamartomas are mostly located in the duodenal bulb (57%). Rarely, they may be found in the second (27%) and third (5%) parts of the duodenum, the pyloric canal (5%), the fourth part of the duodenum, jejunum and proximal ileum[5]. Brunner’s gland hyperplasia is characterized by lobules of glands that are increased in both size and number, and are separated by thin fibrous septae. These lobules extend from the submucosa into the lamina propria and within the mucosa alone and constitute at least 50% of the length of the biopsy specimen. Brunner’s gland hamartoma consists of an admixture of fibromuscular and adipose tissue within and surrounding cystically dilated hyperplastic lobules of Brunner’s glands[6]. Brunner’s gland adenoma can be asymptomatic and are found incidentally. Symptomatic tumors can present as gastrointestinal bleed or obstruction. The clinical manifestations of the former are gastrointestinal hemorrhage, due to ulceration or erosion of the tumor. Obstructive tumors occur when hyperplasia diffuses or a single adenoma grows too large, causing epigastric bloating, discomfort, vomiting or weight loss. Duodenal intussusception has been rarely reported[7], probably because of the fixation of duodenum to the posterior abdominal wall. Hamartoma is a benign lesion, and very few cases of Brunner gland hamartoma have been reported in association with epithelial dysplasia, duodenal adenocarcinoma, and carcinoid tumors[8]. Prior to treatment, all patients diagnosed with duodenal polyps should undergo screening for colonic polyposis, because duodenal adenomas are common in patients with familial adenomatous polyposis (30% to 70%) and vice versa[9].

Upper gastrointestinal endoscopy with biopsy is the diagnostic method of choice. The differential diagnoses include polypoid adenoma of the superficial mucosal glands, adenoma of the islet cells, aberrant pancreatic tissue, lipoma, angioma, leiomyoma, gastrointestinal stromal tumor and malignant neoplasms like carcinoid, adenocarcinoma, lymphoma, leiomyosarcoma and metastasis. Endoscopic mucosal biopsy, however usually gave a negative result because the tumor are covered entirely with thick intact duodenal mucosa in the biopsy sites and the biopsy was often not deep enough to reach the submucosal tumor tissue[10]. Symptoms of our patient can be explained by an intermittent intussusception of polyp distally into the duodenum, as it disappeared after polypectomy. Brunner’s gland hamartoma of the duodenum can be removed endoscopically because it is usually benign. Malignant transformation has been reported rarely[11]. It has been suggested that endoscopic or surgical removal of symptomatic Brunner’s gland hamartoma can prevent the development of complications and endoscopic polypectomy represents the ideal approach depending on the site and size of Brunner’s gland hamartoma and the presence of a peduncle[12]. We performed clip assisted polypectomy as earlier described by YY Chen et al[13]. Very few such cases reported in literature. Open surgical excision is reserved for
cases where snaring has failed or when tumor is too large. The outcome of this endoscopic procedure is usually excellent.

In conclusion, Brunner gland hamartoma is a rare but benign cause of submucosal tumor of the duodenum. It can be managed by endoscopic excision. The use of a detachable snare or haemoclip is recommended during the resection of large hamartoma to avoid the risk of gastrointestinal bleeding. Endoscopic excision is a safe and effective method of polyp removal with favourable outcome.

References