Case Report Open Access

Pyloric Giant Brunner's Gland Adenoma Mimicking Duodenal Carcinoma

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Abstract

Brunner's gland adenoma is a rare benign tumor with 5% of tumors arising from the pyloric region of the small intestine. It is usually asymptomatic, but sometimes presents with ambiguous symptoms like nausea, vomiting, bloating, abdominal discomfort and luminal obstruction thus mimicking malignant tumors as well as other benign tumors that leads the treating physician to misdiagnose the lesion. Here, we present a case of a 62-year-old male with a history of weight loss and vomiting since 4 months. Initial imaging studies that revealed a large mass in the duodenum and endoscopic findings that suggested a mass arising from the pylorus and obstructing the lumen led to the clinical diagnosis of duodenal carcinoma/lymphoma. However, the endoscopic biopsy did not show any features of malignancy. Following laparotomic excision and histopathological examination, a final diagnosis of Brunner's gland adenoma was made. To conclude, the ambiguous presentation of Brunner's gland adenoma as well as its size make it a pertinent lesion to include in the differential diagnosis of malignancies.

Keywords: Giant Brunner's gland adenoma; Pylorus; Duodenal carcinoma

Introduction

Brunner's gland adenoma, also known as Brunneromma or polypoidal hamartoma, is a rare benign proliferative lesion arising from the Brunner's glands of the duodenum, accounting for 10.6% of benign tumors of the duodenum [1]. In majority of cases, these lesions develop into a polypoid mass, usually pedunculated (88%) being 1-2 cm in size while a few cases reaching several centimeters such as the "Giant Brunner gland adenoma" have also been reported [2]. Occasionally they may develop symptoms due to their large size including bloating, nausea, bleeding and obstruction [3]. These tumors usually occur in the first part of duodenum, may be asymptomatic [4]. In rare cases, these giant adenomas may stimulate pancreatic or duodenal malignancy leading to a diagnostic and therapeutic challenge regarding their management [5]. It was first described by Brunner in 1688, these glands were originally thought to be "pancreatic secundarium", however in 1846, Middeldorp identified these duodenal glands as a separate entity [6]. These large adenomas are difficult to diagnose pre-operatively, role of endoscopic biopsy is also limited as these lesions are submucosal and may be missed by a punch biopsy [2].

Case Report

A 62-year-old male presented with history of 20 kilograms weight loss in a span of 4 months with anorexia and occasional bouts of vomiting. There was no history of backpain, fever, malena or jaundice. Physical examination was unremarkable except for a vague ill-defined abdominal mass. All routine blood investigations along with tumor marker CEA were within normal limits except for reduced hemoglobin 10.5 gm/dL (reference range 13-16 gm/dL), decreased packed cell volume 33% (reference range 39-54%) and increased ESR 27 mm/hr (0-20 mm/hr). Ultrasound abdomen suggested a right hypochondriac mass probably of colonic region. MRI of abdomen without contrast revealed a large polypoidal mass measuring 17.5 cm in 3rd part of duodenum extending into the jejunum. Endoscopy revealed a large growth in the pyloric region and medial wall of D2 completely occluding the lumen. The possibility of carcinoma duodenum/lymphoma was considered.

Biopsy from the polyp showed near normal duodenal mucosa with underlying Brunner's glands and superficial muscle layer.

At laparotomy, duodenum was grossly dilated, with a spindle shaped large growth arising at the pylorus extending distally into 3rd part of duodenum and the proximal part of jejunum. Other findings were puckered stomach near the pylorus, slightly dilated jejunum, edematous stomach wall and edematous distended gall bladder.

The specimen submitted for histopathological examination, (Figure 1) grossly showed a 13×10×7 cm, large ovoid, intussuscepted tumor mass covered by mucosa, Cut surface was grey white to pale yellow lobulated with myxoid change (Figure 1).

Microscopy revealed gastroduodenal junctional type of epithelium with hyperplastic Brunner's glands in the submucosa separated by thick fibrous bands. Few dilated ducts and aggregates of lymphocytes were



Figure 1: Large mass measuring 13×10×7 cm.

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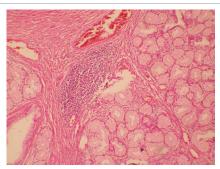


Figure 2: Brunner's glands separated by fibrous septa with dilated glands and lymphocyte aggregates.

also seen. The diagnosis of giant Brunner's gland adenoma was made (Figure 2).

The patient was followed up, one month later upper GI endoscopy showed a patent pylorus and duodenum without any mass.

Discussion

Brunner's glands consist of submucosal mucin-secreting glands located exclusively in the duodenum. They extend from pylorus distally for a variable distance, usually at the first and second portion of the duodenum and less frequently stopping at the third and fourth portion [7].

Curveilheir in 1835, and Savioli in 1876, described the first cases of "adenoma" of the Brunner gland [1,6]. Feyrter defined three types of Brunner's gland hyperplasia, type 1 diffuse nodular hyperplasia in which multiple sessile projections are seen throughout the duodenum, type 2 circumscribed nodular hyperplasia in which sessile projections are limited to duodenal bulb and type 3 glandular adenoma (adenomatous hyperplasia) a single polypoidal lesion [6,8]. The Brunner's gland adenoma is the rarest of three types of Brunner's gland hyperplasia or hamartoma [5]. Hyperplasia of Brunner glands with more than one centimeter was described as Brunner gland adenoma [6]. The largest reported lesion being a 12 cm hamartoma that caused intussusception in a 15 year old patient [9]. In our case the growth measured 17.5 cm on MRI scan and was 13 cm macroscopically showing no malignant transformation. To our knowledge, this is the largest reported case so far in the literature.

Brunner's gland adenoma commonly occurs in 5^{th} and 6^{th} decades of life with no gender predominance. Its clinical presentation include GI bleeding, obstructive symptoms such as vomiting and epigastric bloatedness, and rarely intussusception and diarrhea, possibly due to duodenal motor disturbances [10]. The distribution of Brunner's gland hamartomas is duodenal bulb (57%), the second (27%) and third (5%) portions of duodenum, the pyloric channel (5%), jejunum (2%) and proximal ileum 2% [11].

Useful diagnostic tools of Brunner's gland adenoma are barium contrast studies, abdominal CT and endoscopy [8, 12], however sensitivity of endoscopy is 72-89 % and occasionally, endoscopy is not useful in making the diagnosis [12]. Endoscopic punch biopsy usually gives a negative result, because the tumor is almost entirely covered with thick intact duodenal mucosa in the biopsy sites and the biopsy is often not deep enough to reach the submucosal tumor [2,8,12]. The giant variety is diagnostically challenging due to its large size. Most importantly these lesions may mimic pancreaticoduodenal malignancy on imaging [5].

Pathomorphological features of Brunner's gland are characterized by the presence of non-dysplastic, lobulated Brunner's glands. Its hyperplasia is divided into diffuse hyperplasia, nodular hyperplasia and adenomatous hyperplasia with or without erosion or ulcer. It is a tumor without malignant predisposition [1]. The differential diagnosis includes duplication cyst, leiomyoma, leiomyosarcoma, adenoma or adenocarcinoma, lymphoma, carcinoid tumors, heterotopic pancreatic or gastric tissue or gastrointestinal stromal tumor [13].

The treatment of Brunner's gland adenoma is excision of tumor, though asymptomatic patients can be treated conservatively. Endoscopic snare polypectomy is preferred treatment of small adenoma. Endoscopic resection is less invasive and most cost effective. Excision via open surgery and duodenectomy is reserved for larger and difficult tumors [4].

Conclusion

To summarize, we report a case of a giant Brunner's gland adenoma, one of the largest reported in literature, which presented as an intussusception in the pylorus, a rare localization. This tumor is included in the differential diagnosis of pancreatic duodenal malignancy.

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