Pyloric stenosis due to brunneroma masquerading as peptic ulcer complication

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Received – 04 July 2018 Initial Review – 31 July 2018 Accepted – 30 August 2018

ABSTRACT

A 42-year-male presented to the surgical emergency with features of severe gastric outlet obstruction. On investigation, upper gastrointestinal series showed features suggestive of a near complete gastric outlet obstruction. Endoscopy revealed multiple antral ulcers with near complete pyloric stenosis, features suggestive of complications arising out of antral ulcers. Biopsy report revealed Brunner's gland hyperplasia. A unique aspect about the case is the finding of antral ulcers associated with Brunner's gland hyperplasia. The patient underwent laparoscopic distal gastrectomy with gastrojejunostomy. A high index of suspicion of Brunner cell hyperplasia in case of the patient presenting with features suggestive of pyloric stenosis is the crux of diagnosis in view of the malignant potential of Brunner cell hyperplasia.

Key words: *Antral ulcers, Brunner's gland hyperplasia, Distal gastrectomy, Gastric outlet obstruction*

Brunner's glands are branched acinotubular glands normally found in the deep mucosal or submucosal layers of the proximal duodenum and secrete alkaline based mucus to protect the duodenal lining from the gastric acid [1]. Brunner's gland hyperplasia also known as Brunner's adenoma or Brunneroma, accounts for 10.6% of benign tumors in the duodenum and is often the incidental finding on endoscopy with a majority of the patient being asymptomatic [1,2]. Mostly asymptomatic Brunner's gland hyperplasia can present as gastrointestinal (GI) bleeding, obstruction, and abdominal pain in symptomatic cases [1].

Brunner's gland hyperplasia most commonly present at duodenal bulb (57% of cases), becomes rarer as we go further away from the duodenal bulb [2]. In spite of low propensity for the malignant transformation, Brunner's gland hamartoma can be confused with other malignancies, including duodenal adenocarcinoma or pancreatic head cancer [3,4]. We report an unusual case of Brunner's gland hyperplasia in the pylorus leading to severe gastric outlet obstruction requiring surgical intervention.

CASE REPORT

A 42-year-male presented to the surgical emergency with the complaints of epigastric pain and recurrent vomiting for 12 months. The patient was in follow-up at our surgery outpatient department for his complaints for 8 months. Initially, he used to vomit food, but gradually he used to vomit water also, after 30 min of intake.

Physical examination revealed visible peristalsis and tenderness in the epigastric region with exaggerated bowel sounds. His vital signs were normal and so were his blood parameters hemoglobin 15.1 g/dl (13.8–17.7), total leukocyte count 10,000 cells/cumm (4000–11000), random blood sugar 99 g/dl (70–140), and total bilirubin 1 mg/dl (0.2–1.2): Direct 0.4 mg/dl (0.1–0.3), indirect 0.6 (0.2–1.1), serum alkaline phosphatase 96 U/L (50–130), and serum amylase 43 U/L (30–110).

Upper GI series showed distension of stomach with retention of contrast material most suggestive of a near complete gastric outlet obstruction (Fig. 1). Upper GI endoscopy was done twice at an interval of 2 months. The first report revealed erythema and ulcers present in antrum near the pyloric opening and pyloric channel causing pyloric narrowing. The scope was negotiable with maneuvering, a biopsy was taken. The second endoscopic report revealed diffuse mucosal erythema with mosaic ulcer pattern with partially healed ulcers in the antropyloric region causing luminal stenosis of pyloric opening.

Ultrasound abdomen showed pyloric wall thickening which was circumferential with luminal narrowing. Contrast-enhanced computed tomography (CECT) abdomen revealed distended stomach with fluid contrast levels, thickening of folds of the stomach with circumferential asymmetrical mural thickening involving the antropyloric region, having a maximum thickness of approximately 6 mm with few subcentimetric lymph nodes along the greater curvature of the stomach.

Histopathological examination of the biopsy sample showed focal surface erosion. Subepithelial tissue showed moderate mononuclear inflammatory infiltrate with many eosinophils. Brunner's gland appeared hyperplastic and focally abutted the surface (Fig. 2). There was no evidence of malignancy in the section examined. Histological features were suggestive of Brunner's gland hyperplasia.

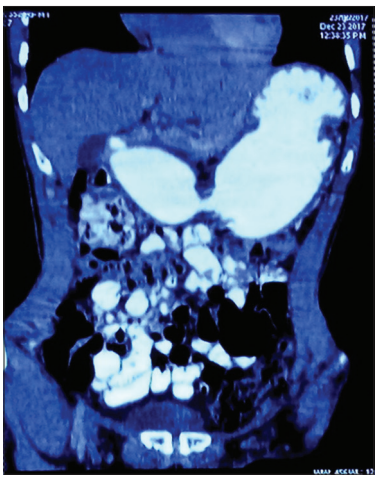

Figure 1: Near complete gastric outlet obstruction

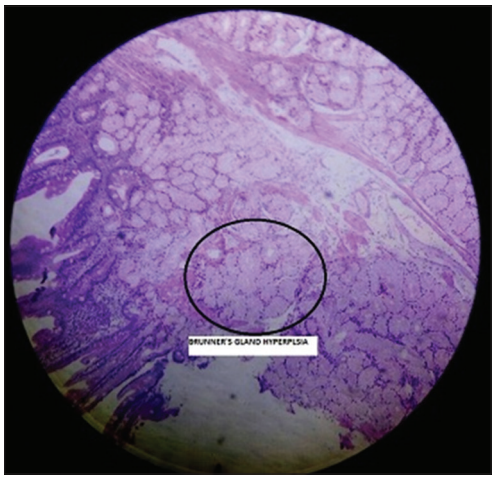

Figure 2: Circle showing Brunner gland hyperplasia

The patient underwent laparoscopic distal gastrectomy with gastrojejunostomy. Histopathological examination of the specimen confirmed Brunner's gland hyperplasia. The patient was discharged on the 7th post-operative day. Follow-up of the patient was done after 6 weeks, in which repeat study with contrast revealed no mass lesion in the duodenum and showed a normal transition.

DISCUSSION

Brunner's gland is submucosal mucin-secreting acinar glands of the duodenum which secretes mucus, pepsinogen, and urogastrone as a response to stimulation by gastric acid, to protect the duodenal mucosa [5]. Histologically, they are branched acinotubular glands covered with cuboidal cells in the ducts and cuboidal to columnar cells in the gland [3].

The incidence of benign tumor of the duodenum is 0.008% in patients at autopsy [6]. Brunner's gland hyperplasia is usually found in the 5th and 6th decades, and its size can range from 0.5 to 12 cm [7]. It is most often found in the duodenum bulb (57%), where it appears as a mucosal protrusion or polyp ranging from 0.5 cm to 1.5 cm in size. The incidence decreases as we go away from the duodenal bulb; the second part of the duodenum (27%), the third part (7%); and rarely at the sites such as pylorus (5%), jejunum (2%), and terminal ileum (2%) [2].

The pathogenesis for the development of Brunner's gland hyperplasia remains unknown [1]. Initially, it was thought that gastric hyperacidity leads to the hyperplasia. In contrast, 20% of cases are reported to have hypoacidity [5]. Other conditions associated with an increased incidence of Brunner's gland hyperplasia are *Helicobacter pylori* infection, uremia, and chronic pancreatitis [8-10]. According to the recent studies, hyperacidity of exocrine modulating factors such as vagus nerve and intestinal mucous membrane factor is the cause of Brunner's gland hyperplasia [2].

In our case, pancreatitis was ruled out on CECT abdomen, and uremia was not considered on view of normal kidney function test. *H. pylori* was negative in both rapid urease test and histological examination of the antral biopsy specimen. Serum gastrin level was within the normal range.

Majority of the Brunner's gland hyperplasia are asymptomatic and are diagnosed incidentally; the clinical manifestations depend on the size, type, and location of hyperplasia, which include dyspepsia, vomiting, epigastric pain, obstruction, GI bleeding, jaundice, and pancreatitis [3]. Levine *et al*. described clinical features in three categories including asymptomatic that are diagnosed incidentally (11%), patient with upper GI bleeding (40–50%), and those with obstructive symptoms (50%) [11]. In this case, gastric outlet obstruction related symptoms including epigastric pain, recurrent vomiting, and poor oral intake were present.

Brunner's gland hyperplasia can be classified into three groups; polyploid type, mass-forming type, and the circumferential infiltrate type. The obstructive symptoms are caused by large polypoid type or mass-forming type and are rarely caused by circumferential infiltrative type [12]. On the contrary, in the present case, the gastric outlet obstruction was caused by circumferential type involving the antropyloric region.

Although endoscopy and radiological examination can aid in diagnosis, definitive diagnosis requires tissue pathology [4]. In barium meal, findings are non-specific, and Brunner's gland hyperplasia appears as sessile or pedunculated filling defect [13]. Endoscopy though aid in diagnosis, has a sensitivity of 72–84%, the reason being some biopsies to be non-diagnostic as lesion are submucosal and may be missed on punch biopsy [1]. There are many pathological conditions for differential diagnosis, which may present as epigastric pain, recurrent vomiting, poor oral intake, and GI bleeding including duplication cysts, leiomyoma, leiomyosarcoma, adenoma or adenocarcinoma, lymphoma, GI stromal tumors, aberrant pancreatic tissue, and chronic pancreatitis [4]. The clinical picture and radiology can be non-specific at times to differentiate these conditions, therefore, arises the need of histopathology which still remains the gold standard for diagnosing this entity.

Conservative treatment with proton-pump inhibitor and antacids are sufficient for treatment of Brunner's gland hyperplasia when the patient is asymptomatic [12]. If the patient is symptomatic, endoscopic resection, laparoscopic, or laparotomy

may be required [14]. If the lesion is pedunculated and <5 cm in size, then endoscopic resection is done. In case of lesions, >5 cm laparoscopic or open surgical excision is preferred [15]. In our case, the size of the lesion was 6 cm in length, so we planned for surgical intervention.

One unique aspect of our case is the finding of an antral ulcer that may have been associated with Brunner's gland hyperplasia [16]. Very few cases have been reported of a concurrent antral ulcer and Brunner's gland hyperplasia [16]. Gastric outlet obstruction leading to upstream mucosal damage may be the cause of peptic ulcer, gastritis, and esophagitis. Fuse *et al.* have found in their study a negative correlation between the distance of the peptic ulcer from pylorus and thickness of Brunner's gland [17]. Another possible relationship between peptic ulcer and Brunner's gland hyperplasia is that both have an association with *H. pylori* [10]. The patient, in this case, tested negative for *H. pylori*. Presence of Brunner's gland hyperplasia in the region of the antropylorus leading to severe gastric outlet obstruction also makes our case distinct.

The patient underwent laparoscopic distal gastrectomy with gastrojejunostomy. Histopathological examination of the resected specimen confirmed it to be Brunner's gland hyperplasia. Post-operative hospital stay was uneventful.

CONCLUSION

We presented an unusual case of Brunner's gland hyperplasia that leads to severe gastric outlet obstruction requiring surgical intervention. Our case report highlights that in the evaluation of patients with gastric outlet obstruction, a benign pathology, i.e., Brunner's gland hyperplasia should be kept in the differential diagnosis. Hence, a combined clinical and radiological workup and histopathological analysis are needed to give a correct diagnosis.

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Funding: None; Conflict of Interest: None Stated.

How to cite this article: Kumar S, Alam MS, Akbal S, Choudhary TK. Pyloric stenosis due to brunneroma masquerading as peptic ulcer complication. Indian J Case Reports. 2018;4(5):347-349.

Doi: 10.32677/IJCR.2018.v04.i05.003