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A Rare Case of Brunneroma Presenting as Gastric Perforation with Review of Literature

Authors
Divya Rabindranath[1], Rana K Sherwani[2], Kafil Akhtar[3], Murad Ahmad[4], Azka Anees Khan[5], Shehtaj Khan[6], Senthil P[7]
[1]Junior Resident, Department of Pathology, J.N. Medical College, Aligarh
Email: *divy30@hotmail.com*
[2]Professor, M.D, Department of Pathology, J.N. Medical College, Aligarh
Email: *drranaksherwani@gmail.com*
[3]Associate Professor, M.D., Department of Pathology, J.N. Medical College, Aligarh
Email: *drkafilakhtar@gmail.com*
[4]Senior Resident, M.D., Department of Pathology, J.N.Medical College, Aligarh
Email: *drmuradahmad@gmail.com*
[5]Junior Resident, Department of Pathology, J.N. Medical College, Aligarh
Email: *azka.asquare.anees@gmail.com*
[6]Assistant Professor, M.S., Department of Surgery, J.N. Medical College, Aligarh
Email: *shehtaj@hotmail.com*
[7]Junior Resident, Department of Pathology, J.N. Medical College, Aligarh
Email: *senthiljune07@gmail.com*
Corresponding Author
Dr. Divya Rabindranath
Dept of pathology, J.N. Medical College, A.M.U, Aligarh-202002.India. Ph: +917895683197
Email: *divy30@hotmail.com*

ABSTRACT

INTRODUCTION: *Brunner`s gland adenomas are rare benign tumors usually located in the bulb of the duodenum. Very rarely, heterotopic Brunner`s gland tissue tumors have also been reported at remote locations from the duodenum, eg. pylorus, pancreas and jejunum.*

CASE: *We report a case of Brunneroma in a patient presenting with a prepyloric perforation with no mass lesion seen on surgery. The diagnosis was established on histopathological examination of the perforation margins.*

DISCUSSION: *Brunneromas are usually asymptomatic. If symptomatic, they commonly present with features of obstruction or GI bleeding. Grossly, they are usually seen as 1-2 cm pedunculated polyps; however, cases of giant Brunneromas have also been reported. To our knowledge, this is the first reported case of a Brunneroma presenting with a perforation located in the prepyloric region, and with no mass lesion identified on surgery.*

KEYWORDS: *Brunneroma, perforation, rare, prepyloric.*

INTRODUCTION

Benign tumors of the duodenum are very rare, with an incidence of 0.008% in a single series of 215,000 autopsies. [1] Brunner's gland adenomas (also known as Brunneroma/Brunner`s gland hamartoma) are rare benign tumors of the duodenum. They constitute 10.6% of the duodenal tumors. [2] They are usually small in size and asymptomatic, however, if large they may cause GI bleeding and obstruction. Usually, these lesions develop into a polypoid mass, 1-2 cm in size and pedunculated in 88% of cases. Few cases reaching several centimetres such as the "giant BGA" have also been reported. We report a rare case of Brunneroma which presented with features of perforation peritonitis, and was located in the prepyloric region. The main purpose of this case report was to highlight that Brunneroma can also present with unusual presentations such as perforation peritonitis, as in our case. Hence, the surgeon must consider a diagnosis of Brunneroma in such patients, once more common conditions are ruled out.

CASE PRESENTATION

A 35 year old Indian male patient presented to the emergency department with complaints of fever, pain and distension of abdomen for 1 day. Pain was associated with vomiting. There was no history of melena, hemetemesis, jaundice or diarrhea. On examination, tenderness was present all over the abdomen along with guarding and rigidity. X ray abdomen (erect) revealed gas under the diaphragm (Fig.1). On ultrasonography of the abdomen, dilated bowel loops were seen with free fluid in inter bowel spaces and peritonitis. The patient was operated for a diagnosis of prepyloric gastric perforation peritonitis. Exploratory laprotomy with primary closure of perforation and omentopexy was performed. A 1X1 cm perforation in anterior wall of stomach was discovered and repaired. No mass lesions were seen. The perforation margin was excised and sent for histopathological examination, which showed partial atrophy of villi with ulcerated mucosa, which is chronically inflammed and a benign proliferation of Brunner`s glands were seen along with adipose tissue, fibrocollagenous tissue and mild lymphocytic infiltrate (Fig.2a &2b). A diagnosis of Brunneroma was established on histopathology.

Endoscopic examination was not performed in this patient because of the unusual presentation with features of perforation peritonitis.

The patient recovered well after the surgery and is doing well.

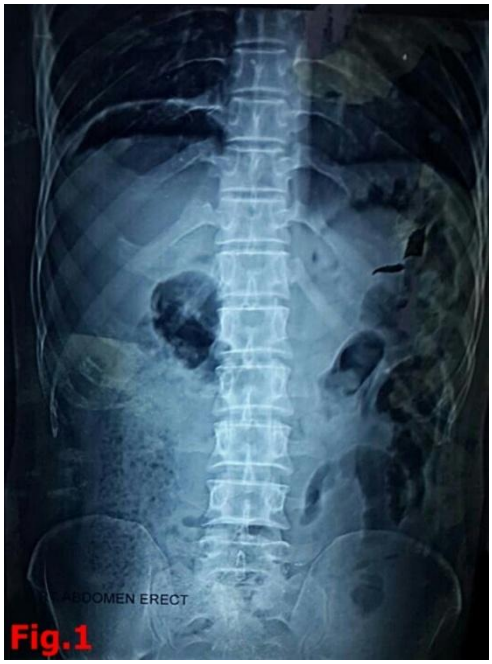

Fig.1: X ray abdomen (erect) showing gas under the diaphragm.

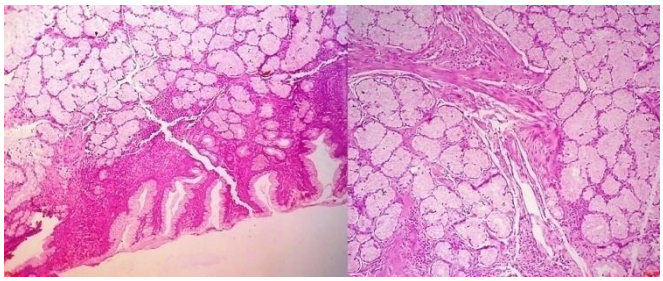

Fig.2a: Photomicrograph showing partial atrophy of villi and ulcerated mucosa with chronic inflammation and underlying brunner's gland proliferation (Hematoxylin & Eosin, X10x)

Fig.2b: Photomicrograph shows the benign proliferation of brunner's glands with fibrocollagenous tissue and mild lymphocytic infiltration (Hematoxyin & Eosin, x40x)

DISCUSSION

Brunner's glands are mucosal and submucosal glands seen in the first part of the duodenum and gradually decreasing in the second and third portions of the duodenum. They are infrequently seen in the pylorus and jejunum. Their mucinous secretions protect the duodenal epithelium by buffering the acidic chyme entering the duodenum from the stomach.

Brunneroma was first described by Curveilheir in 1835. Since then, many cases have been reported .They are described as benign proliferations of the Brunner`s glands. In 1934, Feyrter classified the abnormal glandular proliferation into three types-types 1, type 2, and type 3. [3] Type 1 has diffuse nodular hyperplasia, in which multiple sessile projections are found throughout the duodenum. Type 2 has circumscribed nodular hyperplasia limited to the duodenal bulb. Type 3 has glandular adenoma with polypoid lesions.

Levine et al. reported that 70% of Brunner`s gland adenoma are located in the first portion of the duodenum and predominantly affects the fifth or sixth decade of life with equal gender distribution.[4] Most cases are asymptomatic or are incidentally detected during imaging studies or esophago-gastro-duodenoscopy, or present with nonspecific symptoms like nausea, abdominal discomfort or pain.[2,4] The most common clinical presentations are gastrointestinal bleeding (37%) and obstructive symptoms (37%). GI bleeding usually manifests as chronic iron deficiency anaemia. Erosion or ulceration of the tumor surface would lead to melena or hematemesis. Rare presentations have also been described such as intestinal obstruction caused by intussusceptions of the duodenal wall by hamartoma migration, pancreatitis and bile duct obstruction and diarrhea owing to duodenal motor disturbances.[5] To our knowledge, no known cases of Brunneroma presenting as prepyloric perforation have been reported earlier.

Although Brunneroma is most commonly benign, few cases of malignant transformation have been reported previously. [6]

The pre operative diagnosis of these lesions is difficult. Large adenomas are at a particular risk of being overdiagnosed as carcinomas leading to surgical overtreatment. Small-bowel radiological studies show a smooth-walled filling defect in the duodenum. [7] Localized tumors can present as pedunculated or sessile filling defects with a sharp border, typical of submucosal lesions. CT is useful only for confirming the absence of extra-luminal extension of a tumor. In the majority of cases, a combination of endoscopy with biopsy and duodenography leads to the correct diagnosis. However, both have some limitations when used alone: duodenography has a reliability of 60% with a 20% rate of false negatives, and 10% of uncertain interpretations. [8] Esophagogastroduod-enoscopy has a sensitivity ranging from 72-89%. Intraoperative biopsy has a sensitivity of 83-92%, but it can lead to serious complications like pancreatitis, pancreatic fistula or bleeding. [9] Additionally, Brunneroma is a submucosal lesion and may be missed by a punch biopsy. On barium examination of the upper digestive tract, these lesions produce a characteristic "cobblestone" appearance or a well defined filling defect. [10] Endoscopic ultrasound has been proposed as the best diagnostic modality to differentiate Brunner gland adenoma from other duodenal submucosal tumors. On EUS, Brunneroma is seen as a submucosal mass in the fourth echolayer with hypoechoic pseudopodia. [11] The final diagnosis, however, can only be confirmed only after surgery and histopathology.

The usual differential diagnosis of a Brunner gland adenoma by endoscopy include: neurogenic tumor, leiomyoma, lipoma, carcinoid tumor, enlarged lymph node, aberrant pancreas, prolapsed pyloric mucosa, and some malignancies (duodenal adenocarcinoma, leiomyosarcoma, gastrointestinal stromal tumor etc).[1] Brunneromas should be considered in the differential diagnosis of mass lesions at the ampulla, once more common lesions are excluded. These are, however, conditions which should be kept in mind when the patient presents with a

conventional presentation of mass lesion detected in the duodenum. Since our patient presented with perforation peritonitis, a diagnosis of Brunneroma was not suspected preoperatively and the correct diagnosis was established only on histopathology.

Exact etiology and pathogenesis of Brunneroma is not known. Many mechanisms have been proposed to explain its pathogenesis. Brunner's glands have an "antiacid" function so it has been suggested that an increased acid secretion could stimulate the Brunner`s glands to undergo hyperplasia.[6] However, Spellberg et al., did not find regression of these lesions with acid secretion inhibitors.[12] In a recent study, H. pylori infection was found in 5 out of 7 (71%) cases of Brunneroma.[13] However, the extreme rarity of Brunneroma and the high prevalence of H. pylori infection in the general population suggest that some additional factors might play a role in the pathogenesis of these lesions. Other suggested mechanisms include proliferation in response to local irritation or excessive parasympathetic activity. [4] Another significant association that has been described in patients of Brunner`s gland adenoma is a history of chronic pancreatitis. Stolte et al, found diffuse gland hyperplasia in up to 76% of their patients in a series of 74 pancreaticoduodenotomies for this condition.[14] The significance of this is as yet unexplained, but the authors suggested that the exocrine deficiencies seen in chronic pancreatitis may stimulate an adaptive response of the Brunner's glands. Another hypothesis suggests that this lesion is of inflammatory origin due to the presence of a dense inflammatory cell infiltration.[15] However, since lymphocytes are normally present in the submucosa of the intestinal tract; the presence of inflammatory foci in the Brunneroma is not sufficient to sustain this "inflammatory hypothesis". The hypothesis that is most acceptable at the present time remains that Brunneroma is a duodenal dysembryoplastic lesion or hamartoma. [16]

Microscopically, Brunner's gland adenomas are characterized by the presence of diffuse or nodular hyperplasia of non-dysplastic Brunner's glands mixed with normal tissues, including ducts, adipose tissue, and lymphoid tissue. This unusual admixture of normal tissues supports the hypothesis that these lesions develop as a hamartoma or nodular hyperplasia rather than a true neoplasm.

The ideal approach for asymptomatic cases remains controversial. [5] Most researchers agree that a conservative approach is ideal for these cases. However, few are of the opinion that endoscopic excision should be performed in order to prevent complications. In symptomatic cases, surgical or endoscopic resection is indicated. It has been proposed that endoscopic polypectomy in the duodenum carries a higher complication rate than colonic polypectomies in terms of bleeding, due to its proximity to anatomically important structures. Other factors leading to the potential poor outcome of endoscopic resection are poor visualization within the duodenum, and the risk of peristalsis carrying the resected polyp distally and out of reach. The size of the polyp also limits the use of endoscopic resection, with tumors larger than 4 cm usually requiring an open approach. [17] In such cases, an open surgical approach is advocated.

Most of the Brunneromas are benign and have a good prognosis. [4]

TAKE HOME MESSAGES

- Brunneroma is a rare tumor of the duodenum which is usually asymptomatic or presents with vague symptoms like abdominal discomfort and nausea.
- If symptomatic, they usually present with features of GI hemorrhage or obstruction.
- Our case report highlights an extremely rare presentation of these lesions- as a prepyloric perforation with no mass lesions identified on surgery.
- Though the most common location of Brunneroma is in the duodenal bulb, it can also present at remote sites such as the stomach.

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