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Brunner Gland Hamartoma – A Rare Cause Of Iron Deficiency Anemia

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Abstract
Brunner gland hamartoma is a rare tumor arising from the duodenum. This tumor is usually asymptomatic and detected incidentally during radiological or endoscopic evaluation. The review of literature revealed few rare cases of Brunner gland hamartoma which presented with symptoms like iron deficiency anemia, intussusception and acute pancreatitis with bleeding. We report here a case of Brunner gland hamartoma which occurred in a 71-year-old lady who presented with iron deficiency anemia.

Keywords: Brunner gland, hamartoma, iron deficiency anemia

Introduction

Benign tumors of the small intestine are very rare and Brunner gland hamartoma (BGH) constitutes about 10% of duodenal tumors. It was first described by Cruveilhier in 1835 and Salvioli in 1876 [1]. And since then, less than 200 cases have been reported in the literature. BGH are usually asymptomatic and detected incidentally. But cases presenting with features of iron deficiency anaemia, acute pancreatitis and intussusception are reported in the literature.[2,3,4] We report here a case of BGH in a 71year old lady who presented with features of iron deficiency anaemia

Case Report

A 71-year-old lady presented with tiredness and pallor. On physical examination she was anemic. The routine blood investigations showed a hemoglobin of 5.7gm% and packed cell volume of 16.6%. Serum iron was12 microgram/dl, serum ferritin level was 21.6ng/dl and total iron binding capacity was 323. Stool occult blood was positive. The peripheral smear showed features of microcytic hypochromic anemia. The laboratory investigation findings were consistent with a diagnosis of iron deficiency anemia. She was investigated to find out the cause for iron deficiency.

Ultrasound of abdomen showed a large pedunculated polypoidal intraluminal lesion measuring 4.6x2.6cm arising from the proximal duodenum (D1). The lesion showed heterogenous enhancement with internal hypo enhancing areas. There were no internal calcifications, perilesional fat stranding or regional lymphadenopathy.

Endoscopy showed a large polypoidal lesion with a thick stalk arising from the inferior wall of D1-D2 junction (Fig 1) Multiple biopsies were taken and pathological evaluation showed only duodenal mucosa with regenerative atypia and Brunner gland hyperplasia.

She was further evaluated with contrast enhanced Computed Tomography (CECT) of abdomen(Fig 2) which showed a large heterogenous polypoid lesion

with a stalk arising from the proximal duodenum and the lesion occupying the first and second part of duodenum

Considering its large size she underwent surgical resection of the lesion. A duodenotomy was done at second part of duodenum and lesion was excised by division at base of the stalk. Duodenum was closed transversely. She had an uneventful recovery.

The specimen consisted of a polypoidal lesion with a stalk measuring 6x 4.5x3cm. Cut section showed a pale white lobulated lesion with tiny cystic spaces. Microscopically, the lesion showed lobules of hyperplastic Brunner glands (Fig 3a) admixed with cystic spaces lined by ciliated epithelium. (Fig 3b) The overlying duodenal mucosa showed ulceration and granulation tissue. The morphological features were diagnostic of Brunner gland hamartoma.

Discussion

Brunner's glands are branched acinotubular mucus glands located in the submucosa of the duodenum mainly proximal to ampulla of Vater. It secretes an alkaline fluid containing mucin, which protects the duodenal mucosa from the acidic stomach contents. In addition, in response to the presence of acid in duodenum, these glands secrete pepsinogen and urogastrone which inhibit gastric acid secretion.

Brunner gland hamartoma is a rare benign tumor formed from a proliferation of Brunner glands. This was reported by Cruveilheir in 1835 and Savioli in 1876. In 1934 Feyrter described proliferation of Brunner glands into three types: type 1- diffuse nodular hyperplasia, type 2-circumscribed nodular hyperplasia and type 3- glandular adenoma. The American institute of Radiologic pathology uses the term Brunner gland hyperplasia for lesions less than 5mm and Brunner gland hamartoma for those greater than 5mm[5].

Brunner gland hamartoma is also referred as Brunner gland adenoma or Brunneroma. The term 'Brunner's gland hamartoma' is favoured over 'Brunner's gland adenoma' because it lacks features of a neoplastic lesion like dysplasia, encapsulation and is characterised histologically by a mixture of Brunner glands, ducts, adipose tissue, Paneth cells, pancreatic acini and lymphoid tissue without cellular atypia.

The aetiology of BGH is not clear. It has been postulated that hyperchlorhydria could stimulate hyperplasia of Brunner glands. The role of local irritation due to gastric chyme, antral hormones and vagal stimuli are also reported in literature.[6] Similarly few studies demonstrated the role of Helicobacter pylori in the development of Brunner gland hamartoma.[7,8] H.pylori was identified in 60-70% of BGH.

Brunner gland hamartomas are usually asymptomatic and may be detected as an incidental finding during endoscopy or imaging. But this lesion can present with gastrointestinal bleeding , bowel obstruction or more insidiously with symptoms of dyspepsia or anaemia.

Levine et al (1995) studied 27 cases of BGH and showed that these tumours presented predominantly in the fifth and sixth decades[9]. They also reported that the clinical presentations were gastrointestinal haemorrhage (37%), obstructive symptoms (37%) and the rest were detected incidentally. There were reports of cases which presented with iron deficiency anaemia, acute pancreatitis and intussusception.

Endoscopic ultrasonography is helpful in detecting submucosal lesions. Computed tomography(CT) is useful in evaluating the extent of lesion.[5] Routine endoscopic biopsies may not be diagnostic in BGH as in this case, since Brunner glands are located mainly in the submucosa. Though different modalities of investigations are available, surgical exploration is required for definitive diagnosis.

The differential diagnosis of BGH includes adenomatous polyps, lipoma, leiomyoma, gastrointestinal stromal tumour, neuroendocrine tumour, pancreatic and ampullary neoplasms. Pathological evaluation of excised lesion is mandatory for a definite diagnosis.

Management by endoscopic polypectomy or limited resection is appropriate in most of the cases. Wide excision of a doubtful Brunner gland hamartoma is advisable to confirm diagnosis and to avoid possible complications like obstruction and bleeding. Extensive surgical resections including pancreatoduodenectomy is warranted in case of large lesions and when malignancy cannot be ruled out[10]

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Fig 1. Endoscopic image showing polypoid lesion in duodenum with active ooze

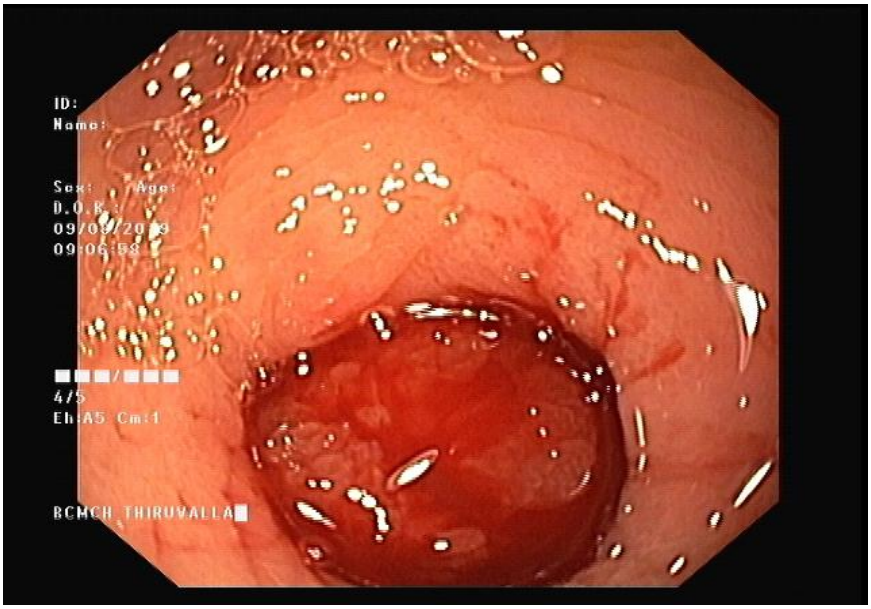

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Fig 2.CECT showing polypoid lesion in second part of duodenum.

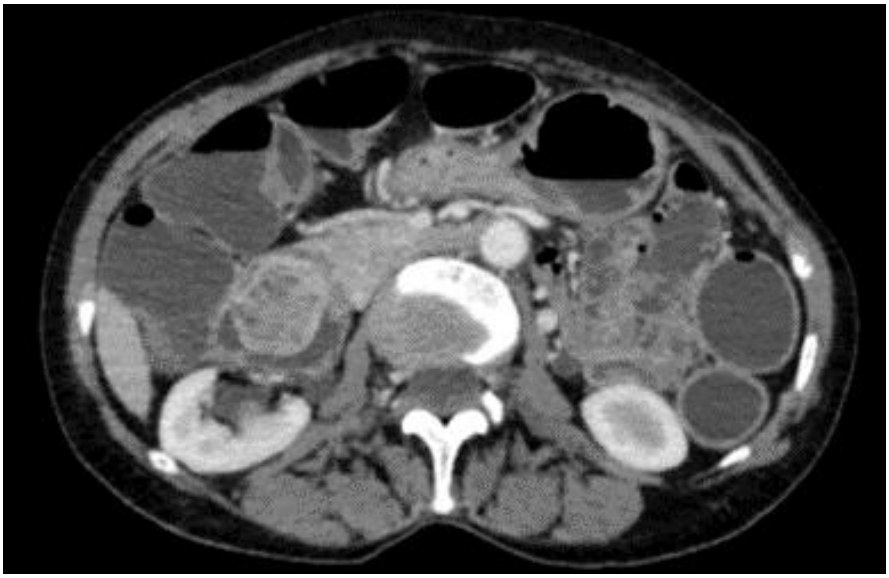

Fig 3a Photomicrograph showing lobules of Brunner glands in the mucosa of Duodenum (Haematoxylin & Eosin X20)

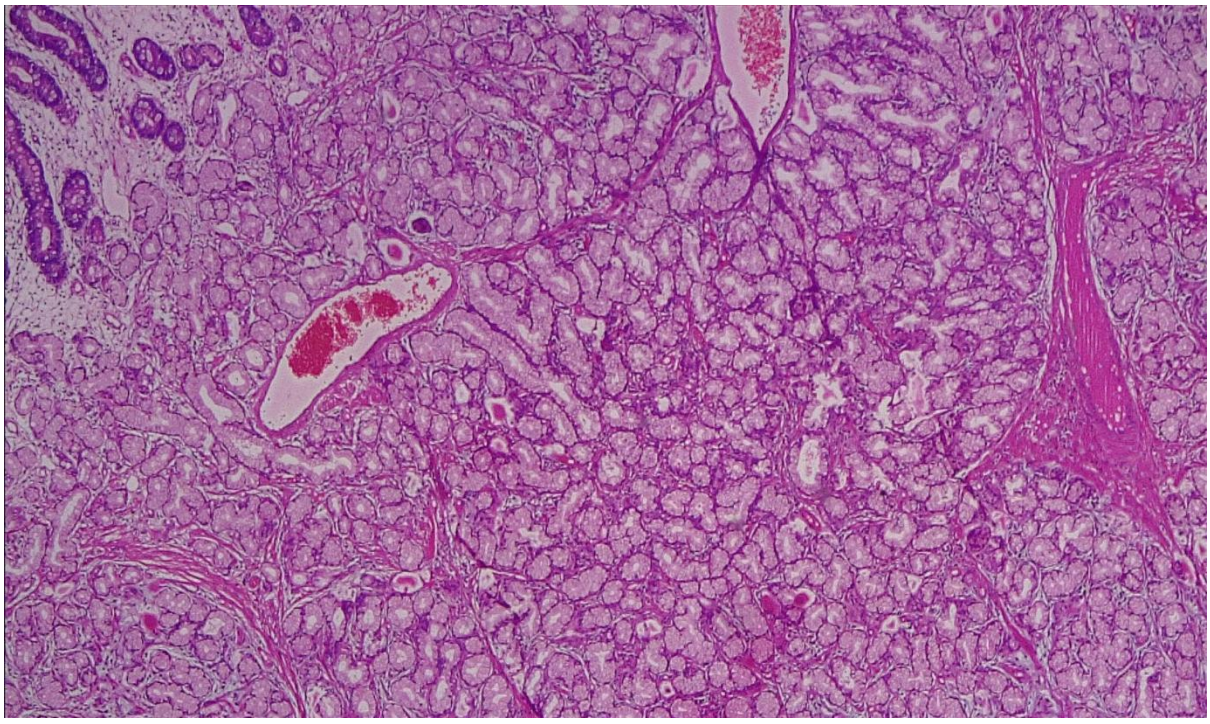

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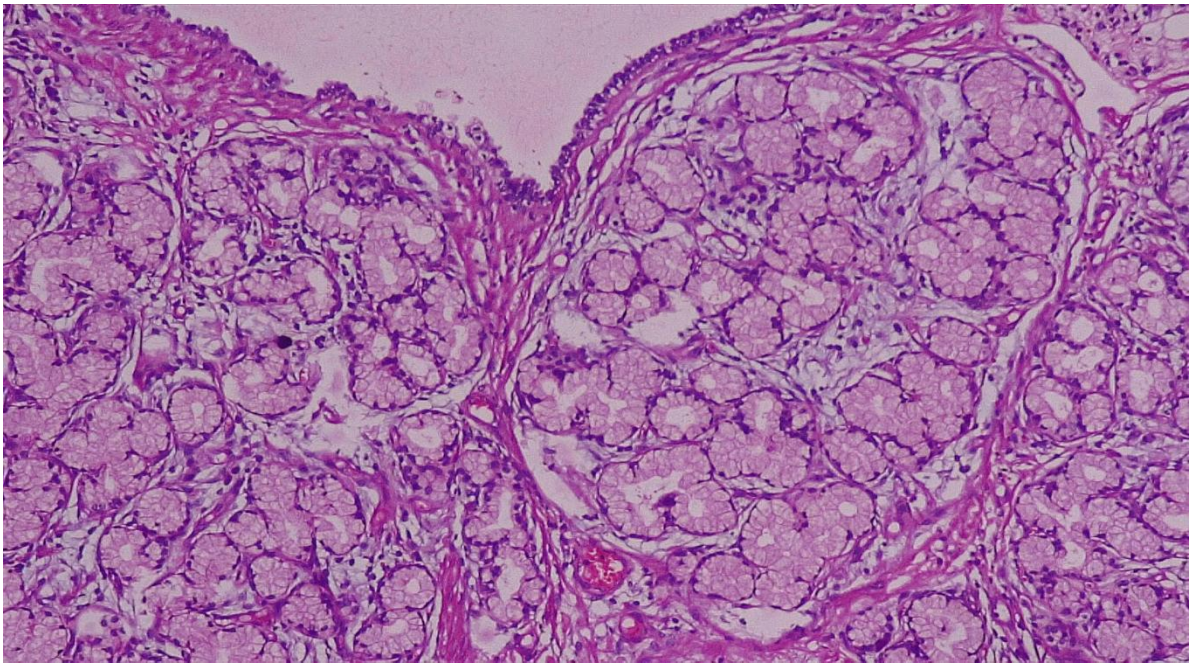
Fig 3b Photomicrograph showing Brunner glands with part of a cyst lined by ciliated epithelium (Haematoxylin & Eosin X40)



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