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Case Report:

Brunner Gland Hamartoma: A Rare Incidental Finding on Autopsy

Authors
Shushan Shweta Jayker, Assistant Professor,
Sharmila Prakash Surhonne, Professor,
Thirupathur Rajaram, Professor and Head,
Department of Pathology, Rajarajeswari Medical College, Bangalore, India.

Address for Correspondence
Dr.Shushan Shweta Jayker,
Assistant Professor,
Department of Pathology,
Rajarajeswari Medical College,
Mysore Road, Kambipura,
Kengeri, Bengaluru-560074,
India.
E-mail: shwetajayker@gmail.com

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Abstract: Brunner gland hamartoma (BGH), is a very rare benign tumor of the duodenum. It is usually asymptomatic and detected incidentally by endoscopy or other imaging modality. But, the definitive diagnosis is only by histopathological examination. We report an autopsy case in which a duodenal mass was found incidentally which was then confirmed histologically as Brunner gland hamartoma and the cause of death in this case was cerebral haemorrhage.
Key Words: Brunner gland hamartoma; Benign tumor; Duodenal mass.

Introduction:
Primary neoplasms of the small intestine are extremely rare comprising of 5% of all alimentary tumors.[1] Brunner gland hamartoma also known as Brunner Gland Adenoma or Brunneroma is a rare benign proliferative lesion of the Brunner Glands accounting for 10.6% of the benign tumors of duodenum.[2] BGH has an estimated incidence of <0.01% and less than 200 cases have been reported in the literature. It is seen in 5th or 6th decades of life. Curveilhier described the first case of BGH in 1835. Symptomatic patients can present with a variety of clinical manifestations such as gastrointestinal bleeding , abdominal pain and occasionally obstruction or intussusception. They present as a single polypoidal, sessile / pedunculated mass in the duodenum.[3]

Case Report:
A 27 years old male suddenly became unconscious and was declared as 'dead on arrival'in the hospital. He had no significant past history. Autopsy was performed. There were no significant external findings on autopsy. Internal examination revealed congested and oedematous brain with multiple pinpoint haemorrhages in the cerebrum and cerebellum diffusely. The liver showed nutmeg appearance on cut surface. Spleen was congested. Lungs and kidneys were unremarkable. Second part of duodenum with attached pancreas, on cutting open, showed a well-delineated tumor measuring 3×3 cms with solid, grey-white and nodular appearance. Microscopy showed a tumor beneath the muscularis mucosae composed of lobules of Brunner glands separated by fibrous tissue. Foci of smooth muscle bundles and adipose tissue were also seen (Figure 1). No dysplasia was seen in the sections studied. Based on the above histopatological findings a diagnosis of Brunner gland hamartoma was made.

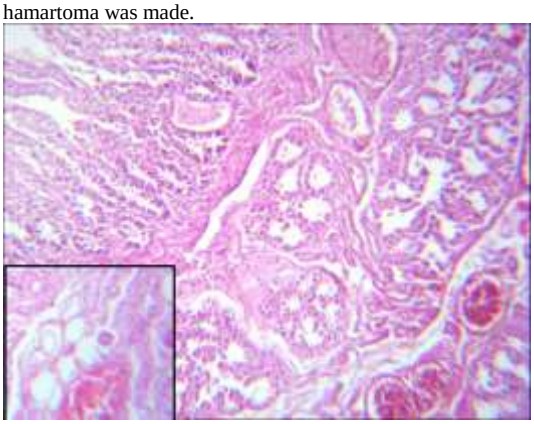

Figure 1: Duodenal mucosa with submucosal Brunner gland proliferation. Inset showing fibroadipose tissue and a congested blood vessel (H & E 40X)

Discussion:
Brunner glands were first described by Brunner in 1688. They are branched acinotubular glands in the submucosa of

the duodenum. They are located mainly in the duodenal bulb , proximal duodenum, and, with a progressive decrease in number and size in the distal duodenum.[4] Ectopic locations include pylorus and jejunum. With age, they constitute a smaller portion of duodenal area, from an estimated 55% in infancy to 35% at 50 years of age. Functionally, they secrete an alkaline fluid composed of mucin, which exerts a physiologic anti-acid function by coating the duodenal epithelium, protecting it from the acid chyme of stomach. Furthermore in response to the presence of acid in the duodenum, these glands secrete pepsinogen and urogastrone which inhibit gastric acid secretion. Thus, they play a significant role in duodenal resistance to ulcer formation.[5]

Brunner gland hamartoma are rare duodenal tumors, occurring in middle age with no gender prevalence.[6] Since its first description by Curveilhier in 1835 and Salvioli in 1876, less than 200 cases have been reported in the world medical literature. They account for 10.6% of benign duodenal tumors and occurred in about 0.008% of individuals in a single series of 215,000 autopsies.[5]

Brunner's gland hamartomas are mostly located in the duodenal bulb (57%). Rarely they may be found in the second (27%), third (5%) parts of the duodenum, the pyloric canal (5%), the fourth part of duodenum, jejunum and proximal ileum.[1]

In 1934, Feyrter categorized the abnormal proliferation of Brunner gland into the following types: type 1, diffuse nodular hyperplasia, in which multiple sessile projections are found through the duodenum; type 2, circumscribed nodular hyperplasia, in which sessile projections are limited to duodenal bulb; type 3, glandular adenoma, in which there are polypoid tumor likes projections. It remains a debate whether these three presentations are the result of same pathologic process.[4] Differentiating hyperplasia from normal Brunner gland is difficult. The diagnostic criteria for hyperplasia in endoscopically obtained specimens require the presence of lobules of Brunner gland within the mucosa in at least 50% length of biopsy specimen. Rarely, they may grow into pedunculated polyp and can produce symptoms. These lesions consist of admixture of fibromuscular and adipose tissue within and surrounding hyperplastic lobules of Brunner glands including heterotropic pancreatic tissue and ducts. Many authors refer these lesions are BGH because of their large size (>2 cm) and presence of combination of both mesenchymal and epithelial elements.[3,7] They are always benign, though a few cases have been reported in association with epithelial dysplasia, adenocacinoma and carcinoid tumors.[5] Increased expression of P_{53} antigens has been demonstrated in hamartomatous lesions by the use of Immunohistochemistry.[6]

The exact pathogenesis of Brunner gland hamartoma remains unclear. Hyperchlorhydria has been thought to stimulate the proliferation of Brunner glands. Associated *Helicobacter pylori* infection is commonly seen in patients of BGH. But their exact role is not clear. Another theory suggests that these glands are hyperplastic reactive proliferation to inflammation which is not well supported as lymphocytes are normally present in the submucosa of entire gastrointerstinal tract. Association with peptic ulcer disease, chronic renal failure and chronic pancreatitis has also been described.[3,5] At present, the most accredited pathogenetic hypothesis remains that BGA is a duodenal dysembryoplastic lesion or hamartoma.[8]

Clinical diagnosis of Brunner gland hamartoma is not always easy at present. Investigations by imaging modalities such as Barium meal, USG, CT and MRI are capable of localizing the tumor. The differential diagnosis includes gastrointestinal stromal tumor, lipoma, carcinoid tumor, lymphoma, vascular tumor, aberrant pancreatic tissue, adenocarcinoma, adenomyomatous polyp, prolapsed pyloric mucosa, ampullary neoplasm and even a foreign object. Definite diagnosis can be obtained only by histopathological examination.[5,9]

Conclusion:

BGH are rare benign lesions of duodenum, usually found incidentally. In our case it is still rarer as it was found in autopsy examination. Incidental findings in medicolegal autopsies, though do not influence the cause of death, yet gain a prime importance in the histopathological report which contributes to the academic and research development.

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