CASE REPORT

Brunner's gland hamartoma with dysplasia, presenting as multiple duodenal polyps: An unexplored entity with literature review

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ABSTRACT

Brunner's gland hamartomas (BGHs) are uncommon lesions of duodenum which show hyperplasia of these glands along with smooth muscle bundles, adipose tissue and lymphoid aggregates. These are usually benign, solitary, pedunculated, polypoidal lesions. Dysplastic changes in BGH are extremely rare and even rarer is the multiplicity of this lesion. We hereby report an index case of BGH showing features of high-grade dysplasia, presenting as multiple duodenal polyps.

KEY WORDS: Brunner's gland hamartoma, Brunner's gland hyperplasia, duodenal polyps, dysplasia

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INTRODUCTION

Brunner's glands, first described by Brunner in 1688, are submucosal acinotubular glands of duodenum. They protect the duodenal epithelium from the acidic chyme of the stomach by secreting an alkaline fluid containing mucin. Nomenclature for proliferation of Brunner's glands remains a matter of debate with various terminologies given to this condition like Brunner's gland hyperplasia, Brunner's gland hamartoma (BGH), and Brunner's gland adenoma.BGH is the preferred term for those usually solitary lesions where there is presence of adipose tissue, smooth muscle bundles and lymphoid tissue along with the hyperplasia of Brunner's glands, usually without atypia.[1] Lesions greater than 1cm, showing microscopic features of hyperplasia of Brunner's glands, were initially called Brunner's gland adenoma.[2] Though most of the literature describe BGH as completely benign, dysplastic changes are extremely rare, reported in only one case.[3] We hereby report a case of duodenal hamartoma with foci of high-grade dysplasia, which presented as duodenal polyposis with a brief review of literature.

CASE REPORT

A 22-year-old Hindu male reported to the Department of Gastroenterology of our institute with complaints of intermittent bilious vomiting, pain abdomen and burning sensation in empty stomach since 4 months. The previous reports that he carried revealed that he is a case duodenal polyposis and 1 year back he had presented with similar complaints, for which he had undergone duodenotomy and polyp excision along with gastrojejunostomy elsewhere. He was symptom free after this surgery for about 8 months. The aforementioned symptoms appeared again 4 months back for which he visited our institution. Clinical examination and routine blood tests were normal. The CT scan of abdomen showed extensive multiple enhancing polypoidal lesions in duodenum with near complete luminal narrowing along with jejunojejunal intussusception. Evidence of gastrojejunostomy was seen with passage of oral contrast into the jejunal loops. All other abdominal and pelvic organs appeared normal. No retroperitoneal lymphadenopathy or intra-abdominal collection was seen. The distal large and small bowel loops were normal in caliber, without any obvious wall thickening. Upper GI endoscopy was done which showed multiple polyps in the duodenum [Figure 1a]. Biopsy taken from one of the polyps revealed duodenal mucosa with Brunner's gland hyperplasia and foci of high-grade dysplasia. Keeping all these findings in

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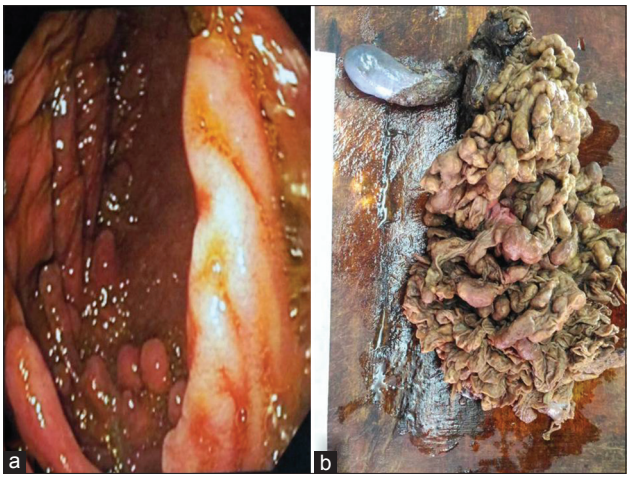

Figure 1: (a) Upper GI endoscopy showing multiple polyps in duodenum. (b) Pancreaticoduodenectomy specimen showing multiple small and large, pedunculated, and sessile polyps in duodenum

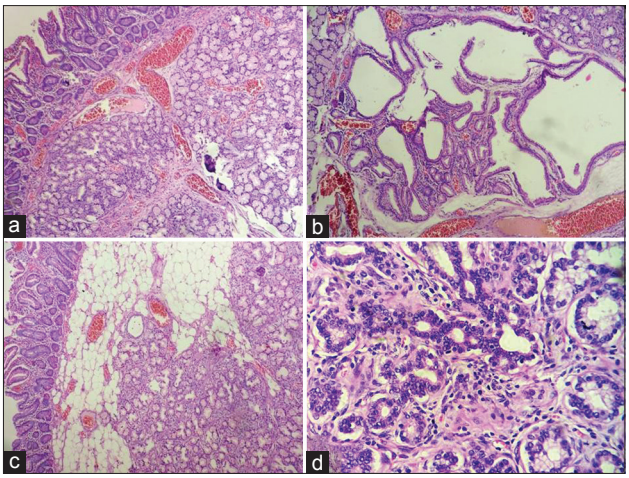

Figure 2: (a) Lobules of hyperplastic Brunner's glands, separated by smooth muscle bundles and fibrous bands and the overlying unremarkable duodenal mucosa (H and E, ×100). (b) Cystically dilated Brunner's glands (H and E, ×100). (c) Adipose tissue admixed with lobules of Brunner's glands (H and E, ×100). (d) Nuclear crowding and overlapping, loss of nuclear polarity, hyperchromasia, and prominent nucleolus seen in the dysplastic glands (H and E, ×200)

view, pancreaticoduodenectomy procedure was carried out, with removal of station 8,12,16 and 17 lymphnodes.

The gross we received was 30 × 15 × 7 cm and contained a portion of duodenum, part of pancreas, gall bladder, bile duct and jejunum. The lumen of duodenum showed multiple polyps (average 90 in number), both small and large, pedunculated and sessile [Figure 1b]. The largest pedunculated polyp was measuring 2.2 × 2 cm, with the stalk measuring 3.5 × 1 cm and the smallest sessile polyp was measuring 0.5 × 0.5 × 0.5 cm. Cut sections of the pancreas, gall bladder and bile duct were unremarkable. Histopathological examination of the duodenal polyps revealed normal duodenal lining epithelium with focal ulceration and hemorrhage. The lamina propria and submucosa showed marked hyperplasia of the Brunner's glands, some of which were cystically dilated [Figure 2a and b]. The Brunner's glands were arranged in lobulated manner, surrounded by adipose tissue [Figure 2c], hyperplastic smooth muscle bundles, dilated, congested blood vessels and lymphoid aggregates. Some of the Brunner's glands also showed features of high-grade dysplasia with nuclear crowding and overlapping, loss of nuclear polarity, hyperchromasia, and prominent nucleolus [Figure 2d]. Sections from the jejunojejunal intussusceptions showed ulcerated lining epithelium, edema and transmural inflammation. Microsections from gall bladder, common bile duct and pancreas were unremarkable. Evidence of reactive hyperplasia was seen in all 21 lymphnodes retrieved. With these findings, a final diagnosis of "Multiple Brunner's gland hamartoma of duodenum with focal high grade dysplasia" was made.

DISCUSSION

BGHs are rare benign duodenal lesions with around 150 cases reported in literature till date.[4] The first BGH was reported by Cruveilhier in 1835.[5] They vary in size, are solitary and pedunculated in most of the cases.[2] The most common age of presentation has been described to be between 40 and 60 years, without any sex predilection.[4] Though it is difficult to establish a pathologic link, the role of *Helicobacter pylori* infection in the pathogenesis of BGH cannot be denied.[2] In most of the cases, the patients of BGH have nonspecific complaints of nausea and abdominal fullness with pain. Bowel obstruction (intussusception), gastrointestinal bleeding and anemia are the less frequent but more worrisome symptoms.

Most BGH are benign, although cases displaying features of focal dysplasia and even carcinoma arising from Brunner's glands have also been reported.[3,6] After extensive search of literature for Brunner's gland hamartoma/hyperplasia with features of dysplasia, we found very few reported cases similar to our study. Of all the reviewed cases of BGH with dysplasia, our patient was the youngest. In most of the cases, the chief complaints were abdominal pain and vomiting similar to our study which in addition showed a peculiar feature of intussusception. Out of all the cases reviewed for BGH/hyperplasia with dysplasia, only the case presented by Brookes *et al.*[3] showed presence of mesenchymal hyperplasia similar to our case, but it was solitary. The studies of Chabib *et al.*,[7] Kim *et al.*,[8] and Fujimaki *et al.*[9] showed dysplastic foci among hyperplastic Brunner's glands, but those were cases of Brunner's gland hyperplasia/adenoma and not true hamartoma [Table 1]. The study by van Rooij *et al.*[10] showed a case of multiple duodenal hamartomas in a 43-year-old male, similar to our study, but lacked the evidence of dysplasia found in our case.

CONCLUSION

BGHs are usually solitary, pedunculated lesions of aged individuals, the diagnosis of which is based on combined features of Brunner's gland hyperplasia, mesenchymal hyperplasia and lymphoid tissue hyperplasia. Multiplicity, showing this combined

Table 1: Brunner's gland hamartoma/adenoma with dysplasia

Case	*Age (years)/Sex*	*Number of lesions*	*Size (cm)*	*Presentation*	*Treatment*
Brookes *et al*.[3]	79/Male	1	2	Melena	Polypectomy
Chabib *et al*.[7]	60/Female	1	3 × 1	Dyspepsia, vomiting	Polypectomy
Kim *et al*.[8]	60/Male	1	1 × 0.8	Anal bleeding	Polypectomy
Fujimaki *et al*.[9]	43/Male	1	2.2 × 2		Mucosal resection
Our study	22/Male	Multiple	2.2 × 2 (largest)	Vomiting, pain abdomen	Pancreaticoduodenectomy

morphology is extremely unusual. Dysplastic changes are still rarer, seen mostly in Brunner's gland hyperplasia/adenoma. Only one case of dysplasia in hamartoma is reported similar to our study till date, but it was not multiple in nature like the present case.

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Conflicts of interest

There are no conflicts of interest.

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