

CASE REPORT

Cystic Brunner's gland hamartoma in the duodenum: A case report

Beom Jin Park, Min Ju Kim, Jeong Hyeon Lee, Sung-Soo Park, Deuk Jae Sung, Sung Bum Cho

Beom Jin Park, Min Ju Kim, Deuk Jae Sung, Sung Bum Cho, Department of Radiology, Korea University, College of Medicine, Anam Hospital, 126-1, 5-Ka Anam-Dong, Sung buk-ku, Seoul 136-705, South Korea

Jeong Hyeon Lee, Department of Pathology, Korea University, College of Medicine, Anam Hospital, 126-1, 5-Ka Anam-Dong, Sung buk-ku, Seoul 136-705, South Korea

Sung-Soo Park, Department of Surgery, Korea University, College of Medicine, Anam Hospital, 126-1, 5-Ka Anam-Dong, Sung buk-ku, Seoul 136-705, South Korea

Author contributions: Park BJ and Kim MJ interpreted the data and wrote manuscript; Lee JH, Park SS, Sung DJ, and Cho SB collected the data and edited the manuscript.

Correspondence to: Min Ju Kim, MD, Department of Radiology, Korea University, College of Medicine, Anam Hospital, 126-1, 5-Ka Anam-Dong, Sungbuk-ku, Seoul 136-705, South Korea. dr.minjukim@gmail.com

Telephone: +82-2-9205578 Fax: +82-2-9203796

Received: August 6, 2009 Revised: September 11, 2009

Accepted: September 18, 2009

Published online: October 21, 2009

World J Gastroenterol 2009; 15(39): 4980-4983 Available from: URL: <http://www.wjgnet.com/1007-9327/15/4980.asp>
DOI: <http://dx.doi.org/10.3748/wjg.15.4980>

INTRODUCTION

Brunner's gland hamartoma is an infrequently encountered benign tumor in the proximal duodenum, accounting for approximately 5% of all duodenal masses^[1,2]. Fifty-seven percent of the tumors originate from the duodenal bulb and the incidence of the tumor decreases as the distance from the pyloric ring grows^[3].

Cystic lesions of the Brunner's glands are exceedingly rare. Only 12 cases have been documented in the literature under various terms such as Brunner's gland cyst, cystadenoma, mucocoele, and cystic Brunner's gland hamartoma^[4]. There has been no literature depicting predominant cystic lesions of Brunner's gland hamartoma in imaging modalities, except for two cases that were found at endoscopic ultrasonography. Although mild dilatation of the ducts may be present in Brunner's gland lesions, it is only detected in histologic test^[5,6].

Herein, we introduce a rare cystic Brunner's gland hamartoma with a long stalk arising from the duodenal third portion found on multidetector-row computed tomography (MDCT), magnetic resonance imaging (MRI), and a modified small bowel series, together with pathologic correlation.

CASE REPORT

A 30-year-old man with a 3-d history of aggravated episodes of nausea, vomiting, and epigastric pain was referred by a private clinic. He was healthy with no specific family or past medical history. Physical examination and laboratory evaluation revealed no abnormalities.

Upper tract endoscopy was not performed because endoscopic results reported by the private clinic were within the normal limits. MDCT showed a 4 cm × 4 cm polypoid mass with multifocal internal low densities in the proximal jejunum just below the Treitz ligament (Figure 1A), and the lesion's long stalk arising from the third portion of the duodenum (Figure 1B). Multifocal high-signal intensities on T2-weighted MRI reflected cystic components (Figure 1C), which were identical to the low densities on

Abstract

Cystic Brunner's gland hamartoma in the duodenum is exceedingly rare, although microscopic examination may sometimes reveal a Brunner's gland hamartoma containing dilated ducts in the duodenum. We present a case of large cystic Brunner's gland hamartoma in the duodenum with a long stalk, which is described in light of multidetector-row computed tomography, magnetic resonance imaging, and a modified small bowel series, together with pathologic correlation and differential diagnosis.

© 2009 The WJG Press and Baishideng. All rights reserved.

Key words: Brunner's gland; Hamartoma; Duodenum; Multidetector-row computed tomography; Magnetic resonance imaging

Peer reviewers: Dr. Serdar Karakose, Professor, Department of Radiology, Meram Medical Faculty, Selcuk University, Konya 42080, Turkey; Kazuo Chijiwa, Profesor, Department of Surgery 1, Miyazaki University School of Medicine, Miyazaki, 889-1692, Japan

Park BJ, Kim MJ, Lee JH, Park SS, Sung DJ, Cho SB. Cystic Brunner's gland hamartoma in the duodenum: A case report.

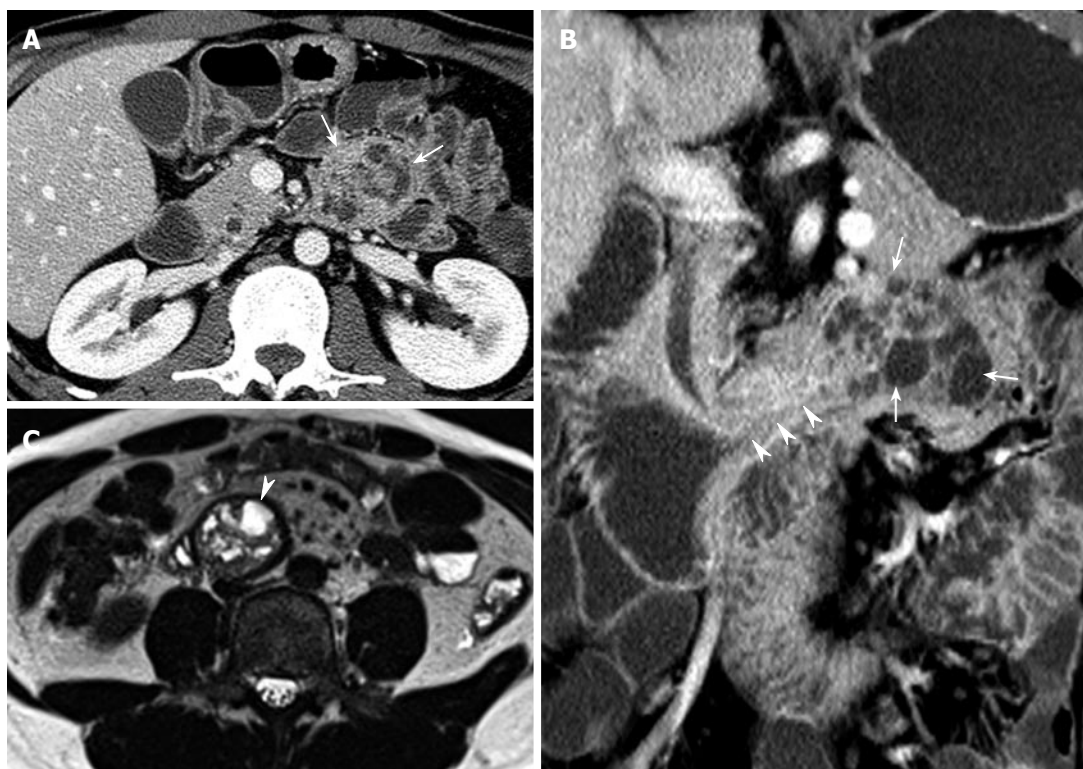


Figure 1 CT and MRI findings. A: Axial portal venous-phase CT scan showing an intraluminal round mass (arrows) with internal multifocal low densities in the proximal jejunum; B: Coronal reformatted portal venous-phase CT demonstrating an identical configuration to the modified small bowel series showing a long stalk (arrowheads) and a well-defined mass with multifocal cystic lesions (arrows); C: A large, well-defined multi-chambered cystic mass showing high signal intensities (arrowhead) with its moved location in the third portion of the duodenum on axial T2-weighted images.

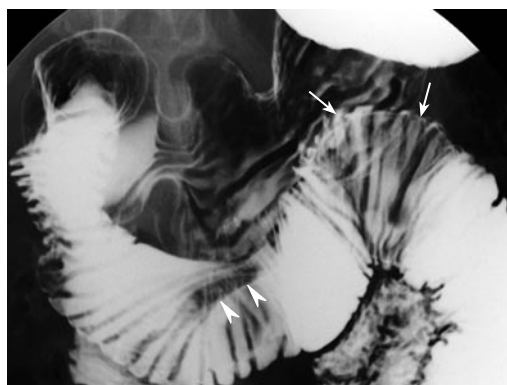


Figure 2 Modified small bowel series findings. A well-demarcated round filling defect (arrows) at the proximal jejunum just below the Treitz ligament and a tubular filling defect with a long stalk (arrowheads) originating from the proximal third portion of the duodenum.

MDCT (Figure 1A and B). The lesion's location moved to the third portion of the duodenum on MRI (Figure 1C). On the modified small bowel series, a mobile smooth lobular filling defect with a long stalk was visible in the proximal jejunum (Figure 2).

A pedunculated, well circumscribed mass was removed at laparoscopic enterotomy of the proximal jejunum, and the residual long remnant stalk was removed by post-operative endoscopy. The cut surface of the resected specimen revealed an approximately 4 cm × 3 cm whitish ovoid mass with multiple internal cystic spaces (Figure 3A). Histological examination of the specimen found that the

main body of the polyp contained a lobular collection of mature Brunner's glands with large multifocal areas of glandular cystic dilation (Figure 3B and C). There was no evidence of malignancy, and the lesion was diagnosed as a pedunculated cystic Brunner's gland hamartoma.

DISCUSSION

The etiology of Brunner's gland hamartoma has not been clearly elucidated. Brunner's gland hamartoma represents 5%-10% of all small bowel tumors and occurs most commonly in the fifth and sixth decades of life, with no gender or race predominance^[7]. Brunner's gland hamartoma is most commonly located on the posterior wall of the duodenum near the junction of the first and second portions. In a series of 27 patients described by Levine *et al*^[8], Brunner's gland hamartoma was located in the duodenal bulb, the second and third portions of the duodenum of 70%, 26%, and 4% of the cases, respectively. In the present case, the long stalk base was located in the proximal third portion of the duodenum.

Although Brunner's gland hamartoma is incidentally found, Levine *et al*^[8] reported that the most common presentations in symptomatic patients are gastrointestinal hemorrhage (37%) and obstructive symptoms (37%). Rarer presentations have been found in conjunction with recurrent pancreatitis^[9], obstructive jaundice^[10], and biliary fistula^[11]. Brunner's gland hamartoma is usually non-dysplastic and entirely benign. However, at least one recent report is available on a Brunner's gland hamartoma

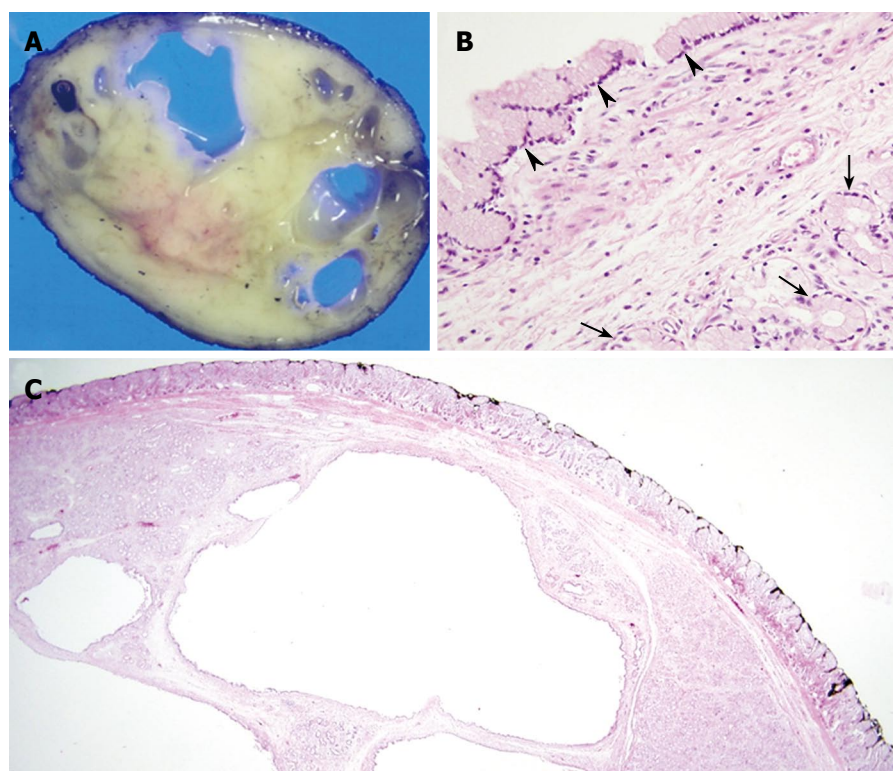


Figure 3 Histologic findings of the specimen. A: Cut surface of the gross specimen showing a multiloculated cystic appearance filled with mucin-like material; B: Aggregated glands (arrows) are lined with cuboidal to columnar cells with abundant pale cytoplasm and a basally-located oval nucleus, resembling a normal Brunner's gland in histopathologic findings of cystic Brunner's gland hamartoma (HE, $\times 200$) while the large cystic dilated gland showing the same lining (arrowheads) as a normal Brunner's gland; C: Cystic Brunner's gland hamartoma showing a well-demarcated nodular lesion, composed of a lobular collection of tubuloalveolar glands separated by fibrous septa, beneath the muscularis mucosa, in the low power field ($\times 12.5$), while some glands showing cystic dilatation.

with malignant potential^[12]. Brunner's gland hamartoma of the duodenum can be removed endoscopically because it is usually benign. However, we performed laparoscopic enterotomy because endoscopic results reported by the private clinic were within the normal limits. It has been suggested that endoscopic or surgical removal of Brunner's gland hamartoma can prevent the development of complications and endoscopic polypectomy represents the ideal approach depending on the site and size of Brunner's gland hamartoma and the presence of a peduncle^[13]. In the present case, considering its size and distal location, the endoscopist was reluctant to perform the procedure, hence laparoscopic enterotomy was performed as our next treatment option.

Patel *et al*^[14] have reported seven cases of Brunner's gland hamartoma on CT. In that report, two lesions with homogenous enhancement correlated with histologic findings of solid glandular proliferation, and the remainder were found with heterogeneous enhancement and hypodensities. The latter cases revealed smooth muscle proliferation and small cystic dilatation of the Brunner's glands. Up to now, to our knowledge, no study has described predominant cystic lesions of Brunner's gland hamartoma found on imaging modalities such as MDCT and MRI, except for two cases that were found on endoscopic ultrasonography^[5,6]. Endoscopic ultrasonography provides additional information about the internal components of the duodenal mass. Imaging modalities such as MDCT, MRI, and modified small bowel series were useful for tissue characterization and localization of the mass in our case, because the large pedunculated mass of the jejunum was difficult to approach by endoscopic ultrasonography.

In the present case, large cyst lesions in a pedunculated mass were revealed by characteristic multifocal high-signal

intensities on T2-weighted MRI images, and large hypodensities on enhanced MDCT correlated with large cystic dilatation of the glands. Mobility due to its long stalk was confirmed by a modified small bowel series and positional change during serial imaging studies.

Diagnosis of Brunner's gland hamartoma is not always easy at present because radiological findings are often nonspecific^[15]. CT is useful only for confirming the absence of extra-luminal extension of a tumor^[16]. Radiologic findings of Brunner's gland hamartoma are extremely similar to those of several other lesions, including adenomatous polyps, lymphoma, carcinoid tumors, and mesenchymal tumors such as gastrointestinal stromal tumor, leiomyoma, and neurogenic tumor. Since duodenal tumors depicted in a cystic configuration through radiologic modalities are rare, the recognition of a cystic component in a solitary duodenal mass may be useful for preoperative diagnosis of Brunner's gland hamartoma.

In conclusion, cystic configuration of a solitary duodenal mass should be generally considered the clue to the diagnosis of Brunner's gland hamartoma.

REFERENCES

- 1 Botsford TW, Crowe P, Crocker DW. Tumors of the small intestine. A review of experience with 115 cases including a report of a rare case of malignant hemangio-endothelioma. *Am J Surg* 1962; **103**: 358-365
- 2 Ghazi A, Ferstenberg H, Shinya H. Endoscopic gastro-duodenal polypectomy. *Ann Surg* 1984; **200**: 175-180
- 3 Silverman L, Waugh JM, Huizenga KA, Harrison EG, Jr. Large adenomatous polyp of Brunner's glands. *Am J Clin Pathol* 1961; **36**: 438-443
- 4 Silverman L, Waugh JM, Huizenga KA, Harrison EG Jr. Large adenomatous polyp of Brunner's glands. *Am J Clin*

- Pathol* 1961; **36**: 438-443
- 5 **Yamakawa M**, Murata I, Yamao T, Kawai K, Kohno S. Cystic Brunner's gland hamartoma. *Gastrointest Endosc* 2003; **57**: 919
 - 6 **Changchien CS**, Hsu CC, Hu TH. Endosonographic appearances of Brunner's gland hamartomas. *J Clin Ultrasound* 2001; **29**: 243-246
 - 7 **Peison B**, Benisch B. Brunner's gland adenoma of the duodenal bulb. *Am J Gastroenterol* 1982; **77**: 276-278
 - 8 **Levine JA**, Burgart LJ, Batts KP, Wang KK. Brunner's gland hamartomas: clinical presentation and pathological features of 27 cases. *Am J Gastroenterol* 1995; **90**: 290-294
 - 9 **Scholz HG**. [Recurrent acute pancreatitis, a complication of brunneromas (authors transl)] *Leber Magen Darm* 1976; **6**: 300-302
 - 10 **Skellenger ME**, Kinner BM, Jordan PH Jr. Brunner's gland hamartomas can mimic carcinoma of the head of the pancreas. *Surg Gynecol Obstet* 1983; **156**: 774-776
 - 11 **Hedges AR**. Hamartoma of Brunner's gland causing pyloric obstruction and a biliary fistula. Case report. *Acta Chir Scand* 1988; **154**: 475-476
 - 12 **Brookes MJ**, Manjunatha S, Allen CA, Cox M. Malignant potential in a Brunner's gland hamartoma. *Postgrad Med J* 2003; **79**: 416-417
 - 13 **Rocco A**, Borriello P, Compare D, De Colibus P, Pica L, Iacono A, Nardone G. Large Brunner's gland adenoma: case report and literature review. *World J Gastroenterol* 2006; **12**: 1966-1968
 - 14 **Patel ND**, Levy AD, Mehrotra AK, Sobin LH. Brunner's gland hyperplasia and hamartoma: imaging features with clinicopathologic correlation. *AJR Am J Roentgenol* 2006; **187**: 715-722
 - 15 **Gao YP**, Zhu JS, Zheng WJ. Brunner's gland adenoma of duodenum: a case report and literature review. *World J Gastroenterol* 2004; **10**: 2616-2617
 - 16 **Merine D**, Jones B, Ghahremani GG, Hamilton SR, Bayless TM. Hyperplasia of Brunner glands: the spectrum of its radiographic manifestations. *Gastrointest Radiol* 1991; **16**: 104-108

S- Editor Wang YR L- Editor Wang XL E- Editor Ma WH