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Brunner’s gland hamartoma of the duodenum is a rare, benign and usually asymptomatic lesion that manifests itself only following hemorrhage or obstructive complications. Approximately 200 cases have been reported in the English literature, and only 5 have presented with intussusception since the first description by Cruveilhier in 1835. We hereby report another rare case of a 53-year-old woman who presented with obstructive gastrointestinal symptoms. Abdominal CT showed intussusception from the antrum of the stomach to the proximal jejunum. Open surgical polypectomy through a longitudinal duodenotomy was successfully performed. Pathological examination of the resected tissue confirmed a giant Brunner’s gland hamartoma. It is worthwhile to remember that duodenal intussusception can occur when there is Brunner’s gland hamartoma.

Key words: Brunner’s glands hamartoma, duodenum, intussusception

Benign tumors of the duodenum are rare. They are reported in 0.008% of the patients at autopsy, and Brunner’s gland hamartoma comprises only 10.6% of these lesions.1 Most of these lesions present as polypoid masses 1-2 cm in size.2 The majority of cases are asymptomatic and occasionally cause upper gastrointestinal hemorrhage or obstruction.3 Duodenal intussusceptions, probably because of the fixation of the duodenum to the posterior abdominal wall, induced by Brunner’s gland hamartomas are extremely rare. Only 5 patients have been reported since the first description by Cruveilhier in 1835,4 which caused intussusception with a lethal outcome.5-8 We report a case of giant Brunner’s gland hamartoma in a patient who presented with intussusception from the antrum of the stomach to the proximal jejunum.

Case Report

A 53-year-old woman presented with severe epigastralgia and abdominal fullness for 3 days, which had taken place 8 months prior to this admission. At that time, gastroduodenoscopy revealed erythematous patches on the antrum, and mild nodular mucosa at the pyloric ring. No abnormality was found in the duodenum. Biopsy of the gastric mucosa revealed chronic active gastritis with Helicobacter pylori infection. She remained symptom-free for 8 months after this episode. Intermittent, dull upper abdominal pain of 3 days duration prompted her to attend our emergency department. Her physical examination revealed only mild epigastric tenderness. Her laboratory data included hemoglobin of 11.6 g/dL and mean corpuscular volume of 82.1 fl. Gastroduodenoscopy disclosed mild hyperemia of the gastric mucosa with a clear mucus lake. There was a large submucosal lesion at the duodenum that was moderately firm in consistency; tiny ulcers were seen on the surface of the post-bulbar section, extending to the 2nd portion of the duodenum (Fig 1). Vigorous bowel peristalsis was observed in this area. The bulb and distal 2nd portion were intact. Abdominal computed...
tomography (CT) showed intussusception from the antrum of the stomach to the proximal jejunum (Fig 2). The leading point of the intussusception was a 2.2-cm heterogeneous soft tissue lesion. No enlarged lymph nodes were found in the entire abdomen; no biliary dilation was found. A barium study disclosed a large and long submucosal mass filling the lumen without obstruction (Fig 3).

During laparotomy, it was seen that the intussusception had been reduced spontaneously. Polypectomy through a longitudinal duodenotomy was performed. There was a submucosal lesion composed of histologically normal Brunner’s glands in a nodular pattern, measuring 5.5 × 4.0 × 3.2 cm, with attenuated duodenal mucosa lining the surface (Fig 4). A diagnosis of Brunner’s hamartoma was made. No evidence of malignancy was found. A follow-up gastroduodenoscopy 4 months later showed no recurrence.

**Discussion**

Brunner’s gland hamartoma of the duodenum is rare. It is predominant in 5th or 6th decade of life, with equal gender distribution. Hyperplasia refers to multiple lesions of less than 1 cm, and hamartoma refers to lesion of larger. Feyrer’s classified 3 types of Brunner’s gland hyperplasia: diffuse nodular hyperplasia, occupying most of the duodenum; circumscribed nodular hyperplasia, the most common type mainly present in the duodenal bulb; and adenomatous hyperplasia, which may be sessile or pedunculated. In most cases, the lesion appears as a polyoid mass 1-2 cm in diameter. The most common...

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**Fig 1.** Gastroduodenoscopy showed a large submucosal lesion (arrow), moderately firm in consistency, and tiny ulcers on the surface of the post-bulbar area.

**Fig 2.** Computed tomography (CT) of the abdomen showed intussusception from the antrum of the stomach to the proximal jejunum. The leading point (white arrow) was a heterogeneous soft tissue lesion.

**Fig 3.** The upper gastrointestinal barium study exhibited abnormal position of the duodenal 2nd and 3rd portions (elevated, black arrows), with a large and long submucosal mass filling the lumen without obstruction.

**Fig 4.** Photomicrograph of hyperplastic well-differentiated Brunner’s glands (arrow) forming lobules surrounded by bundles of fibromuscular and connective tissues. (Hematoxylin & eosin, original magnification, 400 x).
location of the lesion is the posterior wall of the duodenum near the junction of its first and second portions. Clinically, such a lesion is usually asymptomatic, manifesting itself only following hemorrhage or obstructive complications. Duodenal intussusception at the site of a Brunner’s gland hamartoma is extremely rare. To the best of our knowledge, only 5 cases have been reported in the literature.

The most accredited pathogenetic hypothesis for this lesion is that it is a duodenal hamartoma or dysembryoplastic lesion. Nodular hyperplasia of the normal Brunner’s gland is an admixture of normal tissues, including ducts, adipose tissue, and lymphoid tissue. Brunner’s gland hamartoma is considered a benign tumor, although malignant degenerative change has rarely been described. In our patient, the pathology disclosed nodular hyperplasia of the Brunner’s gland without malignant degenerative change, which is consistent with previous reports.

Diagnosis of Brunner’s gland hamartoma is difficult. In most cases, it is noticed incidentally on gastroendoscopy or imaging studies for other disorders. Traditional endoscopy with pinch biopsies is usually negative because the tumor is almost entirely covered with thick, intact duodenal mucosa, and the biopsy is often not deep enough to reach the submucosal tumor tissue.

Removal of asymptomatic Brunner’s gland hamartomas could prevent complications such as hemorrhage, anemia, obstruction, and intussusception. The choice of the procedure, such as endoscopic polypectomy, Whipple’s operation, or biliary or digestive bypass, is determined by the functional status, size of the tumor, and presence of complications. Endoscopic polypectomy is an ideal approach when the tumor is small or pedunculated. Surgical excision is reserved for cases where snaring has failed or when the tumor is too large. For our patient, we chose open duodenotomy to remove the tumor for the purpose of relieving the obstruction, preventing hemorrhagic complications, and also because the tumor was large.

Our patient presented with gastrointestinal signs of the duodenal obstruction including abdominal distension. Gastroendoscopy showed a large submucosal lesion (5.5 × 4.0 × 3.2 cm) of moderately firm consistency, with tiny ulcers on the surface of the post-bulbar area, to the 2nd portion of the duodenum. Her intussusception had reduced itself prior to surgery. Hence, we did not find evidence of intussusception from the antrum of the stomach to the proximal jejunum during laparotomy, as was seen on the initial CT scan.

In conclusion, giant Brunner’s gland hamartoma presenting with intussusception is extremely rare. The diagnosis should be confirmed histologically. Surgical resection is indicated for large tumors; it would also prevent future complications such as intussusception and hemorrhage. Brunner’s gland hamartoma should be ruled out in all cases of intussusception.

References

十二指腸布朗氏缺陷瘤造成的腸套疊：病例報告

曾建仁 郭進榮 溫義輝

十二指腸布朗氏缺陷瘤是一種罕見的良性腫瘤。大部分沒有症狀，偶爾以出血或阻塞表現。到目前為止西方文獻大約有兩百個案例，其中因布朗氏缺陷瘤造成的腸套疊只有五例。我們報告一位53歲女性，電腦斷層發現從遠端胃到近端迴腸發生腸套疊，開刀後病理報告是十二指腸布朗氏缺陷瘤。我們認爲十二指腸出現腸套疊時，十二指腸布朗氏缺陷瘤應該列為鑑別診斷之一。