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BRUNNER'S GLAND ADENOMA

— Case report, and review of etiopathogenesis and clinical features —

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Benign proliferative changes of the Brunner's gland of the duodenum are a rare phenomenon, with less than 120 cases reported in the English literature. The first case was described by Cruveilhier in 1835: a lethal tumor causing intussusception histologically proven to be an adenoma of the Brunner's glands.[1] The vast majority of Brunner's glands tumors show histologically benign proliferation in the form of adenomatous hyperplasia, but two cases of malignant growth have been reported in the literature.[2] Improvements in radiologic techniques and particularly the introduction of fiberoptic endoscopy have accounted for an earlier diagnosis and treatment of these duodenual tumors, which may manifest non-specific clinical symptoms. A case of Brunner's gland adenoma is herein reported and the etiology, clinical manifestations, diagnosis, and treatment of this rather rare entity are reviewed and discussed.

CASE REPORT

A 51-year-old white male was admitted to the medical service on 12/9/83 because of a four week-history of diarrhea associated with upper abdominal pain partially relieved by antacids. The patient's past history was significant for multiple level laminectomies for arachnoiditis and an endoscopically demonstrated gastric ulcer treated with antacids for several years with good symptomatic control. His physical examination, was unremarkable, except for a mild epigastric and right upper quadrant tenderness to deep palpation. Blood studies, biochemical laboratory data including amylase and liver function tests, and urinalysis on admission were all within normal limits. On the second day after admission his LFT's increased with a total bilirubin of 2.6, alkaline phosphatase of 140, LDH447, SGOT 575, SGPT 650 and they all returned to normal a week later. Upper endoscopy on 12/20/83 showed findings consistent with duodenitis and the patient was discharged on cimetidine therapy.

Approximately a month later the patient was re-admitted for worsening symptoms of right upper quadrant and epigastric colicky pain. An ultrasound of the upper abdomen showed stones within the gallbaldder, which had a thickened wall. A HIDA scan revealed normal filling and delayed emptying of the gallbladder. Two days after re-admission, the patient had an episode of tarry stools. An ERCP was attempted on 1/23/84 and showed a small sessile duodenal polyp in the proximity of the papilla of Vater, rendering impossible the cannulation of the biliary and pancreatic ducts. The biopsy of the duodenal polyp was non-diagnostic. The patient was then referred to the surgical service and the following day he was taken to the operating room and his abdomen was explored through a right subcostal incision. A cholecystectomy was carried

out and an intraoperative cholangiogram showed no filling defects and good flow of dye into the duodenum. A longitudinal incision was performed in the second portion of the duodenum and a 1 cm in diameter sessile polyp was excised. The duodenotomy was closed in two layers in a transverse fashion. Pathology reports revealed chronic cholecystitis with cholelithiasis and a Brunner's gland adenoma of the duodenum.

The patient's postoperative course was uneventful and he has remained asymptomatic and is doing well 1 ½ years after surgery.

DISCUSSION

Benign duodenal tumors are rare: their incidence in the general population is 0.008% according to Nasio's series of 215,000 autopsies [3] and they represent 4 to 25% of benign tumors of the small intestine.[4] Brunner's gland adenomas constitute only 10.6% of benign tumors of the duodenum[5]. They are usually found in the bulb and less often in the post-bulbar portion[6~7] as in the case we described. Their size may range from few millimeters to several centimeters[1,5,8,9] They are most frequently seen in males 40 to 60 years of age.[1,2]

The clinical presentation of a Brunner's gland adenoma is absolutely nonspecific. Presenting symptoms include postprandial abdominal pain, epigastric discomfort, vague dyspepsia, bloating and occasionally nausea or even vomiting in case of obstructing lesions[2,3,10]. Ulcer-like pain with pyrosis or watery diarrhea have also been described.[1] The most common symptom is probably gastrointestinal bleeding from ulceration or erosion of the tumor, manifested as melena or anemia with its related sequelae[1,3,5]. Fatal hemorrhages have been rarely reported.[1,11] If the tumor is located near the papilla of Vater can cause biliary obstruction with jaundice, [2], and if pedunculated, can produce duodenal intussusception with ileus.[12,13] Approximately 50% of patients are completely symptom free.[14] It is apparent that the presenting symptoms of Brunner's gland adenoma mimic those of many other more common gastorintestinal disorders. In our case all the symptoms were initially thought to be related to cholelithiasis until the episode of melena prompted further diagnostic investigation.

Upper gastrointestinal series and endoscopy are the cornerstones of diagnosis, the latter presenting also interesting therapeutic possibilities. On routine barium studies, or hypotonic duodenography, Brunneroma can be manifested as a discrete duodenal filling defect ("vacuole sign") while a diffuse Brunner's glands hyperplasia can cause a cobblestone pattern in the first and second portions of the duodenum.[3,15] The differential diagnosis of these smooth-walled filling defects in the duodenum include a variety of lesions; adenoma of the superficial duodenal mucous glands, leiomyoma, lipoma, neurogenic tumors, carcinoid, aberrant pancreatic tissue, prolapsed pyloric mucosa, antegrade intussusception of a pedunculated antral polyp[6,7] Upper gastrointestinal endoscopy represents an effective method of both diagnosing and treating these tumors. After the first case described by Alper in 1973,[16] several reports of endoscopic removal of Brunneroma have been published in the recent literature.[17,18] This method appears to be particularly indicated in case of pedunculated lesions, where it could represent adequate treatment, with pathological identification of a lesion that only exceptionally has been reported to possess malignant potential. In case of lesions which are sessile or located in of the proximity the papilla of Vater an endoscopic biopsy can be inadequate due to the submucosal location of the lesion or potentially dangerous leading to complications of perforation or hemorrhage.[5]

Farkas [1] described the following three types of pathological changes of Brunner's glands:

Type I : diffuse, nodular hyperplasia where numerous sessile projections are found throughout the duodenum.

Type II : circumscribed, nodular hyperplasia where sessile projections are limited to the first portion of the duodenum.

Type Ⅲ : glandular adenoma, constituted by polypoid tumor-like projections.

Spellberg and Vucelic[4] argued that all three Farkas forms are different expressions of the same process and never represent a premalignant condition. However, malignant tumors involving Brunner's gland have been reported[19]. In our case the location of the nodule deep into the muscularis mucosa, the surrounding fibrous capsule, and the sharp demarcation from the surrounding submucosa led to the diagnosis of adenoma rather than hyperplasia.

Interesting pathogenetic mechanisma have been postulated to explain the occurrence of these hyperplastic growths. Hyperchlorhydria has been proposed as a cause of hyperplasis of the Brunner's glands[1] by investigators who accept the protective role of these glands for the duodenal mucosa. Other authors have observed a correlation between chronic pancreatitis[20] or even severe kidney disease[1] and Brunner's glands hyperplasia. Spellberg and Vucelic[4] postulated two possible mechanisms to explain the relationship between Brunner's gland hyperplasia and gastric hyperacidity: an obstructing lesion in the duodenum may stimulate gastric hypersecretion, or hyperplasia could be the reaction of Brunner's glands to hyperchlorhydria. The latter mechanism appears to be more likely although the duodenal lesions, once present usually do not regress after the H_2-receptor blocking treatment. It is of interest that this theory is supported by the fact that the enteric hormone enterogastrone, an inhibitor of gastric acid secretion, is produced by Brunner's glands[20]. Stolte et al.[20] found diffuse Brunner's gland hyperplasia in 75.7% of 74 specimens of duodenopancreatectomies performed for chronic pancreatitis. No correlation was found between the degree of Brunner's gland hyperplasia and degree of scarring of the exocrine pancreas or duodenal wall, the inflammatory infiltration of duodenal mucosa, duration of the disease, alcohol consumption, history of gallstones or ulcers. Brunner's gland hyperplasia in the presence of chronic pancreatitis is probably an adaptive reaction to the exocrine insufficiency of the pancreas.

The management of Brunner's gland adenoma is complete removal of the lesion and pathological examination, always necessary to define the nature of the lesion and exclude malignancy. In case of pedunculated lesions, the endoscopic snare-cautery technique offers the advantages of reduction in hospital costs and potential complications and risks of abdominal surgery and can represent a definitive treatment. In case of diffuse hyperplasia a biopsy should define the nature of the lesion and conservative management directed towards symptoms is usually sufficient. Formal complete surgical excision of the lesion through a duodenotomy remains the standard treatment if endoscopic removal is not possible. It is associated with minimal morbidity, offers the advantage of the total removal of the lesion and prevents complications related to obstruction and hemorrhage.

SUMMARY

Brunner's glands of the duodenum rarely develop proliferative lesions, occasionally in association with gastric hyperchlorhydria or chronic pancreatitis. The clinical presentation can vary from vague upper abdominal symptoms with dyspepsia and nausea to diarrhea, jaundice, obstruction and gastrointestinal bleeding. The diagnosis is usually made by radiological studies followed by upper endoscopy which can also provide definitive treatment. At times surgery is necessary for adequate removal of these lesions, as in the case herein described.

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— **Abstract** —

Brunner씨 선종

— 임상증례 및 임상상적 문헌고찰 —

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십이장에 있는 Brunner씨 선의 양성증식성 병변은 장당히 희유한 변화이다. 지금까지 영문으로 보고된 증례는 대체로 120예를 넘지 않는다.

1835년 Cruveilhier에 의해 처음 증례가 보고되었는 바 대부분의 증례는 양성종양 또는 양성증식성병변으로 나타나나 이 때까지 2예의 악성종양도 보고 되었다.

방사선진단법의 개선과 fibropic endoscopy의 이용으로 십이장의 종양성 병변의 조기진단과 치료에 많은 진전을 보았으며, 따라서 저자는 비교적 희유한 십이장에 생긴 Brunner씨 선에서 발생한 양성종양을 경험하였기에 이 때까지 보고된 문헌을 고찰하고 그 원인, 임상상적 소견, 진단 및 치료에 대한 경험예를 보고하는 바이다.

환자는 51세된 남자로서, 1983년 12월 9일 상복부 동통과 4주간의 설사를 주소로 하여 본 병원 내과에 입원하여 1984년 1월 23일 ERCP에 의해 십이장에 polyp가 있는 것을 확인하고 외과에서 적출하여 병리조직학적으로 Brunner씨 선에서 발생한 양성선종으로 진단하였다. 본 환자는 그 후 경과가 양호하여 수술후 1년 반까지 아무런 증상 없이 잘 지내고 있음이 확인되었다.