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Brief Report

Extreme adenomatous hyperplasia of Brunner's glands in the proximal jejunum

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Adenomatous hyperplasia arising in relation to Brunner's glands is rare. It is most frequently seen in the duodenal bulb and seldom below the papilla of Vater. Symptoms, if present, are non-specific and the diagnosis, suspected by radiological examination, is proven by histological examination of tissue obtained by laparotomy or endoscopy. The case is reported of a 43-yr-old female admitted to the hospital because of serious melena originating from an extreme adenomatous hyperplasia of Brunner's glands. The tumour measured 6 × 5 × 4 cm and was located in the proximal part of the jejunum; such a tumour has never previously been reported at this site. Neth J Med 1988;32:20–26.

Key words: Brunner's gland; Brunner's gland adenoma; Melena; Benign tumour of the duodenum and the jejunum

Introduction

In a previous issue of this *Journal*, the primary tumours of the small intestine received attention [1]. The differential diagnosis of these conditions includes, among others, the Brunner's gland adenoma. The first description of a Brunner's gland adenoma was published by Cruveilhier in 1835; it had caused the death of a 29-yr-old woman by intussusception and intestinal obstruction [2]. Tumours of the duodenum arising in relation to the glands of Brunner are rare, although recently various cases have been reported [3–7]. The total number of cases published since Cruveilhier's description is 120 cases in the English literature [8] and 73 cases in the Japanese literature [9]. The age of the patients reported in the literature varied

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Fig. 1. Barium-meal examination showing filling defect (arrows) in the proximal jejunum.

Fig. 2. The stalked tumour is removed by jejunotomy.

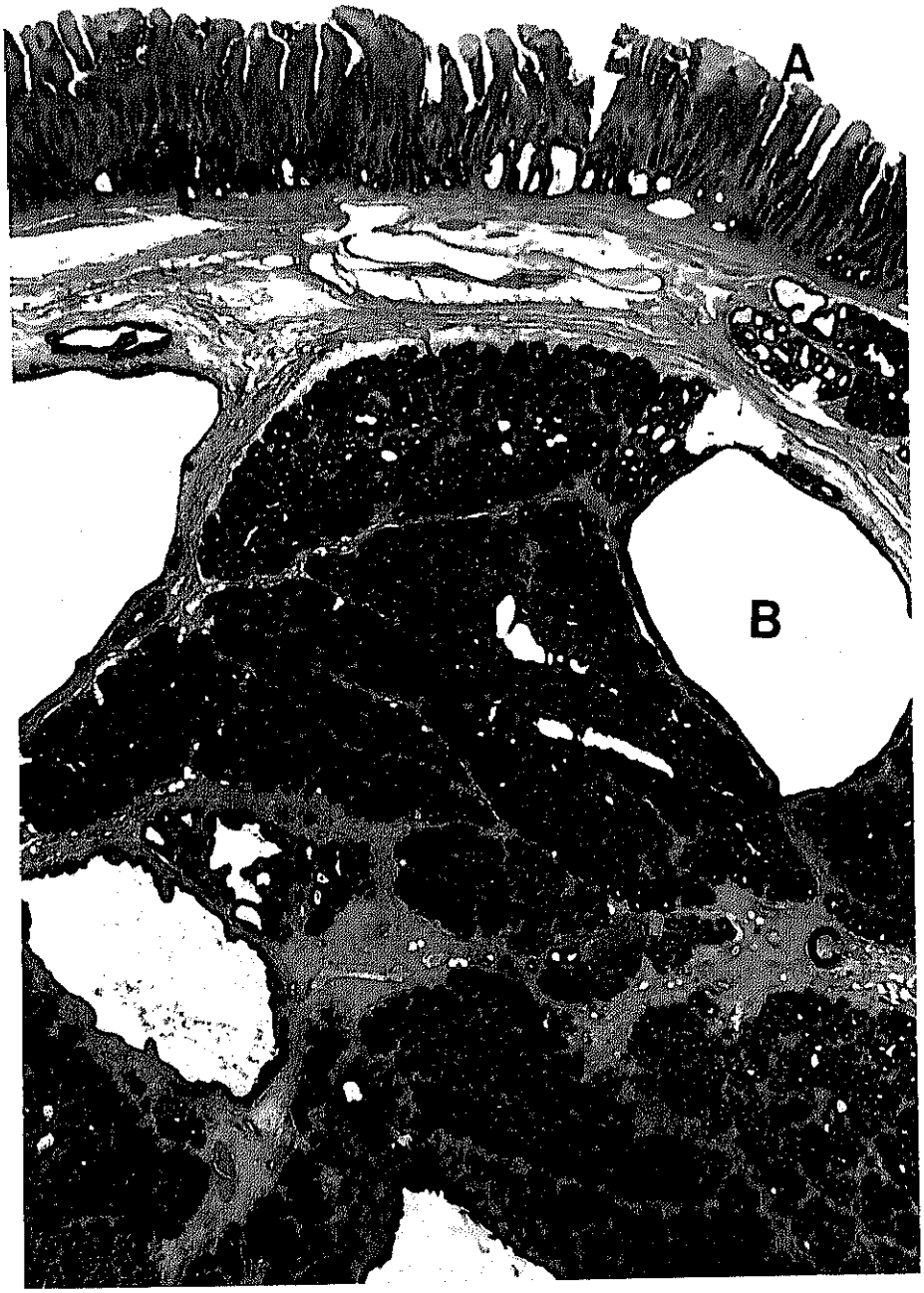

Fig. 3. Histological picture. A = jejunal mucosa; B = cyst; C = fibrous septum.

between 11 months and 80 yr, with a peak incidence between 40 and 60 yr. Males were affected almost twice as frequently as females [10].

Case Report

A 43-yr-old white woman was admitted to our hospital because of gastrointestinal bleeding. Two days earlier, she had suddenly experienced anorexia, nausea and upper abdominal discomfort, not related to food intake. After an attack of itching abdominal pain, she had passed a large tarry stool, and this occurred again the following day. Physical examination was unremarkable except for melena in the rectum. Blood studies revealed the following results: ESR 34 mm after 1 h, haemoglobin 4.7 mmol/l, haematocrit 0.24 l/l, MCV 92 fl, MCH 1850 amol, MCHC 20.1 mmol/l and a white blood cell count of 8.7×10^9 with normal differentiation. Liver and renal functions were unremarkable. Coagulation tests were normal.

Fibreoptic endoscopy and barium meal examination of the oesophagus, stomach and colon performed in the days following admission did not reveal the source of bleeding, nor did ultrasound examination. In the meantime, the haemoglobin stabilised after transfusion at 8 mmol/l and for 7 days the patient was free of complaints without signs of gastrointestinal bleeding. On the 8th day after admission, she again passed a large tarry stool followed by a fall in haemoglobin to 6.5 mmol/l.

Radiographic examination of the duodenum and the jejunum according to Sellink finally revealed a large, mobile, smooth filling defect (6 cm diameter) just distal to the duodenal–jejunal transition. The radiographic appearance was benign and a diagnosis of leiomyoma was assumed (Fig. 1).

Subsequently, the patient underwent operation by means of a longitudinal jejunotomy. A mandarin-sized soft stalked tumour was found just distal to the flexura duodeno-jejunalis and could be removed in toto (Fig. 2). Macroscopic inspection showed a tumour of $6 \times 5 \times 4$ cm (Fig. 3). On microscopic examination the tumour was found to be covered by mucosa which showed small defects with ulcerations and haemorrhages. Brunner's glands were found in the lower parts of the mucosa. The crypts of these Brunner glands extended into the submucosa separated from each other by fibrous septa. The ducts showed a variable degree of cyst formation. The tumour lacked encapsulation or infiltrative growth. There were no other signs of malignancy. The final histological diagnosis was hyperplasia of Brunner's glands in the jejunum (Fig. 3).

The patient made an uneventful recovery and was discharged on the 7th postoperative day. She was well when seen a few months later.

Discussion

Brunner's glands are located in the submucosa and deeper parts of the mucosa of the duodenum and most numerous just distal to the pylorus, gradually diminishing in number aborally in the duodenum. The finding of Brunner's glands distal to the

papilla of Vater is unusual. Rarely, Brunner's glands have been found just proximal of the pylorus and, among juveniles, in the proximal part of the jejunum [11]. Brunner's glands secrete an alkaline mucoid fluid, which appears to act as a protective mechanism against the action of gastric acid [12]. The anatomical distribution is consistent with this explanation.

Brunner's gland tumours are found almost exclusively in the first part of the duodenum. In 46 patients reviewed by De Castella, only one had a tumour which originated distal to the papilla of Vater [13]. Their size varies from 1 to 12 cm, although the majority are 2–4 cm in diameter [13–15]. Microscopically, the tumour consists of a proliferation of Brunner's gland elements surrounded by fibrous septa on a stalk of variable length, which is caused by traction of the intestinal wall. There is a variable degree of cyst formation. There is no definite demarcation of the adjoining normal Brunner's glands [13].

There is only one report of a malignant change in Brunner's glands [16], but malignant changes in a Brunner's gland adenoma have never been proven. The clinical manifestations may be nausea, vomiting, postprandial complaints, upper abdominal discomfort, intussusception, intestinal obstruction and watery stools. Gastrointestinal bleeding occurred in approximately 40% of the cases reviewed by De Castella [13]. In 1928, Golden described the characteristic radiological findings: nodular hypertrophy of Brunner's glands was seen as multiple small filling defects, resulting in a so-called "Swiss cheese" or cobblestone appearance [17]. Usually, the tumour has a sessile or pedunculated polypoid aspect. The differential diagnosis includes leiomyoma, lipoma, villous adenoma, neurogenic tumour, haemangioma, carcinoid, aberrant pancreas tissue, prolapsed pyloric mucosa, antegrade intussusception of a pedunculated antral polyp or pseudopolypous duodenitis [18].

Nowadays, endoscopy is a useful procedure for confirming the diagnosis if the tumour can be reached and if the biopsy is sufficiently deep [19]. Therapy, most often consisting of surgical excision, is indicated when the patient is symptomatic. Endoscopic removal has been described and is possible for the smaller stalked tumours [20,21].

Compared to findings in the literature, the size of the tumour in our case ($6 \times 5 \times 4$ cm) was notable. Its localization in the proximal jejunum is unique.

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