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Rare Case of a Giant Hamartomous Polyp of Brunner's Gland

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ABSTRACT

During an investigation of the upper digestive tract of a 61-year-old female patient mainly complaining of hematemesis and melena, a peduculated polyp was discovered in the duodenum, and was surgically removed. Histologically, the polyp was recognized as a hamartomous polyp of Brunner's gland. Concerning the cases reported as so-called "Brunner's gland adenoma" it seems that there is a considerable difference in the histological findings between Japanese cases and Western experiences, and this point is discussed in this paper, referring mainly to our case and other cases reported in Japan.

Benign tumors are rarely formed in the duodenum, and among these there is a high incidence of Brunner's adenoma. Particularly, in the past 5 years, reports on Brunner's adenoma have been increasing in Japan, and the number of cases reported in Japan is said to be nearly equal to the total number of cases reported in Europe and America. As one of the causes, the endoscope is used extensively in Japan, so that the chance of discovering swelling changes of the duodenum is very high. However, comparing the literature within Japan and that from abroad, it seems that the definition of Brunner's adenoma itself is somewhat different in Japan as that in Europe, and is considered to be the cause of the differences in a number of reports. Recently the authors have experienced a case of a huge polypoid hamartoma of Brunner's gland, and its clinical progress is reported in this paper together with the investigations into similar cases reported so far in Japan.

CASE REPORT

A 61-year-old housewife. Nothing notable in her family history or past history. Chief complaints were sudden hematemesis and melena, accompanied by a slight vertigo. The patient was admitted to a local hospital. At the time of hospitalization, the RBC was 268 $\times$ 10^4, and hematocrit was 26.2%, and the blood pressure was 110/60 mmHg. Due to a state of shock, the upper digestive tract was investigated while performing 2000 ml of blood transfusion. A pedunculated polypoid lesion was discovered first by fluoroscopy from the bulb of the duodenum to the third portion (Fig. 1), and a stalk and polyp

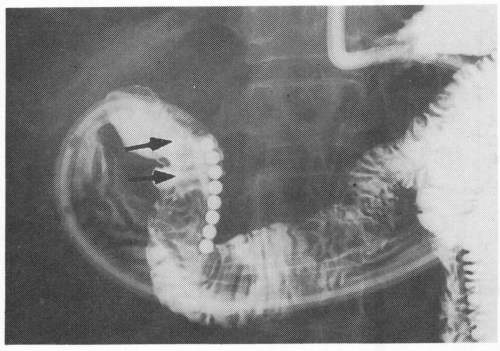

Fig. 1. A double contrast hypotonic duodenography showing a long gourd shaped pedunculated (arrow) polyp in the duodenum.

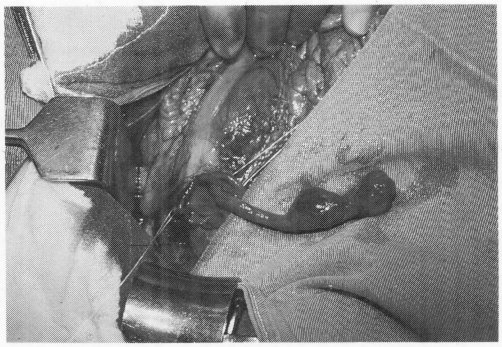

Fig. 2. Polyp pulled out of the duodenum bulb.

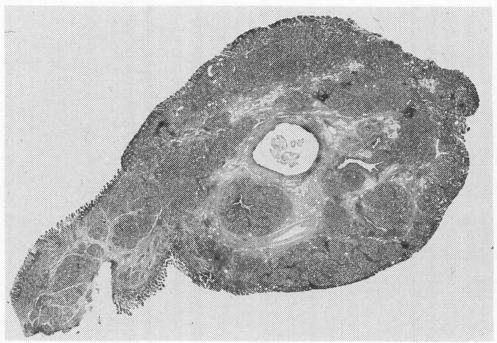

Fig. 3. Whole section of the polyp surgically excised showing lobular structures with cystic space.

were then recognized endoscopically. Oozing from the surface of the polyp was seen (Fig. 2). There were no signs of bleeding on the mucosa of the esophagus and stomach. The massive bleeding was considered to be due to the polyp, and an endoscopic polypectomy was unsuccessfully attempted. Hence, it was decided to remove it surgically. An incision of about 3 cm was cut in the second portion of the duodenum, and the polyp was pulled out. The stalk adhered to the posterior wall at the lower end of the terminal portion of the duodenum, and it was ligated and cut off at its root. Figure 2 shows the polyp pulled out of the incised portion of the duodenum. The polyp measured 40 mm by 25 mm, and the stalk was 30 mm long, the end of the polyp reaching the third portion of the duodenum. Figure 3 shows the whole longitudinal section of the polyp. Histopathological findings are shown in Fig. 4, 5. Most of the polyp was occupied by an overgrowth of Brunner's gland accompanied partly by a cyst, while

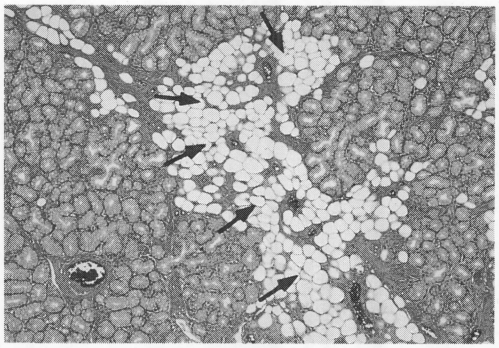

Fig. 4. Low power microscopic view of the polyp showing hyperplasia of normal Brunner's glands riddled with smooth muscle layer. Many fat cells are also seen (arrow)

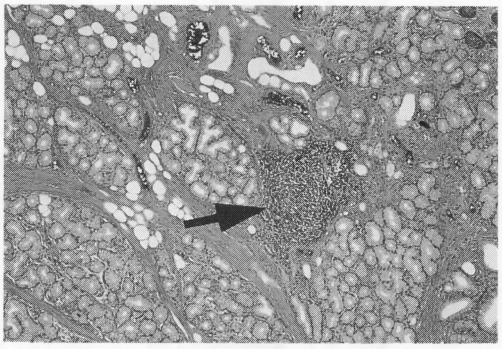

Fig. 5. Arrow showing lymph apparatous in the Brunner's glands.

smooth muscle fibers, adipose tissue, and lymph apparatus were recognized among the overgrowth. Paneth cells were also scattered about in the tissues of Brunner's gland, proliferating in a nodal form. These findings were thought to suggest clearly that this tumor is a hamartoma. The levels of gastrin, glucagon, insulin, and somatostatin in the serum of this patient, were all normal.

DISCUSSION

Various diagnostic names have been given to the pathological nature of the proliferative lesion of Brunner's gland, such as adenoma, hyperplasia, and hamartoma. This is a confusion, derived from the tumor's characteristic that its boundary from the normal tissues is not clear despite of an obvious formation of nodules, and that the cells forming Brunner's gland are free from polymorphism.

Feyster (1934)[3] investigated 2,800 cases of duodena, and discovered nodules originating from Brunner's gland in 36 cases, and classified them into (1) diffuse nodular hyperplasia, (2) circumscribed nodular hyperplasia, and (3) Brunner's gland adenoma. Types (1) and (2) can be distinguished by whether the nodules are multiple or single, but both are small nodules derived from hyperplasia of brunner's gland and totaled 34 cases out of 36. By contrast, there were 2 cases of a large polyp with stalk, which were identified as "adenoma" to be distinguished from types (1) and (2). Ever since, most of the cases reported as Brunner's gland adenoma in Europe and America are tumors with a stalk with a minimum of 2 cm length. In Japan, on the other hand, 143 cases had been reported as "Brunner's gland adenoma" by the end of 1985, but their descriptions are considerably different from the European definition. For example, in the allegedly first report in Japan by Sekiguchi (1927)[11], although the tumor length was 2.5 cm, it was considered to belong to type (2) of Feyster from both the gross and pathological findings. In the subsequent reports in Japan, of less than 2 cm in length which account for more than 60% of all are considered to belong to the category of hyperplasia estimating from the descriptions in the reports. It seems the custom was established in Japan to call adenoma all proliferative lesions of Brunner's gland including the cases histologically recognized as hyperplasia. This is regarded as one of the causes of the high incidence of Brunner's gland adenoma reported in Japan. Recently, there is a tendency of classifying the masses derived from Brunner's gland either as hyperplasia or as hamartoma, instead of adenoma, from the standpoint that they are not true neoplasms[4,9,13].

In this case, Brunner's gland hamartoma is characterized histologically, by the presence of adipose tissue, smooth muscle tissues, and ductal structures, in addition to overgrowth of Brunner's gland[8], and all these conditions were satisfied in the authors present experience (Figs. 3, 4, 5).

Sakata et al[10] first reported Brunner's gland adenoma in Japan, with a clear histological description, as hamartoma, and only few reports followed them[5-7]. Bleeding is a serious complication of so-called Brunner's gland adenoma, as we have experienced. Of the 119 cases reported in Japan, together with a description of symptoms, as summarized in Table I, hematemesis or melena was seen in 27%, while the incidence rate is as high as 45 to 50% in Europe[1,12]. Generally, bleeding tends to increase as the size of the mass derived from Brunner's gland becomes larger. Accordingly, the low incidence in Japan seems to be because most of the cases are actually hyperplastic nodules. Incidentally, of the 7 cases with large nodules larger than 6 cm, hematemesis or melena was noted in 6 cases. As for malignant changes of Brunner's gland adenoma, only one case has been reported so far in the world[2], but this is due to hyperplasia, and a malignant change from hamartoma is not known so far. Therefore, in the case of a giant hamartomous polyp, those with severe symptoms, such as bleeding and abdominalgia seem to be an indication for resection. As for the present case, an endoscopic resection was preferably desired, but since it was difficult to apply the snare on the mass, a surgical resection had inevitably to be resorted to.

Table 1. Chief complaints (119 cases)

abdominal pain	45 cases	(38%)
abdominal fullsensation	44	(37)
Bleeding	27	(27)
Nausea, Vomiting	8	(6)
Others	14	(12)
No complaint	19	(16)

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