

Adenomyoma of the Jejunum—A Rare Cause of Gastrointestinal Bleeding

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Adenomyoma of the small intestine is rare. It occurs mostly in the periampullary region or ileum. The common presentations are intussusception and intestinal or biliary obstruction, depending on the location. To our knowledge, gastrointestinal (GI) bleeding from a jejunal adenomyoma has not been reported previously. We present a 74-year-old female patient who suffered intermittent tarry stool passage for 1 month. Initial upper GI endoscopy, colonoscopy and computed tomography failed to find the bleeder. A papilla-like tumor with central depression and active bleeding in the proximal jejunum was found by push enteroscopy. Exploratory laparotomy showed a submucosal nodule about 1.5 cm in size located about 20 cm distal to the Treitz ligament. Wedge resection was carried out. Pathologic examination revealed that the tumor was composed of some cystic exocrine-type ducts and bundles of smooth muscle, indicating adenomyoma. The patient was symptom-free following operation. [*J Chin Med Assoc* 2008;71(2):96–99]

Key Words: adenomyoma, enteroscopy, gastrointestinal bleeding, jejunum

Introduction

Adenomyoma of the gastrointestinal (GI) tract is a rare benign lesion. It arises from pancreatic metaplasia or a displaced pancreatic anlage during embryogenesis. The term *ectopic pancreas* is used if the differentiation is into normal pancreas. If the differentiation is into various abnormally arranged pancreatic elements, various terms are used including adenomyoma, myoepithelial hamartoma, adenomyosis, and foregut choristoma.¹ Adenomyoma occurs mainly in the stomach.² The small bowel is the second most frequent location, usually in the periampullary area or ileum.¹ Jejunal adenomyoma is truly rare, and only 5 adult cases have previously been reported in the English literature.^{3–5} The most common clinical presentations of adenomyoma in the small bowel are intussusception and intestinal or biliary obstruction, depending on the location of the lesions.^{1,3–7} To our knowledge, GI bleeding from a jejunal adenomyoma has not been reported previously. We report a case of jejunal adenomyoma with

small intestinal bleeding that was diagnosed by push enteroscopy and surgical intervention.

Case Report

A 74-year-old woman was admitted because of intermittent tarry stool passage and progressive dizziness for 1 month. She denied any systemic disease, history of operation, or abuse of drugs or alcohol. No bowel habit change or body weight loss could be detected. Initially, she was admitted to a local hospital. Because of the negative findings of upper GI endoscopy, colonoscopy and abdominal computed tomography (CT), she was referred to our hospital.

On admission, she had a pulse rate of 82/minute, blood pressure of 138/96 mmHg, and respiratory rate of 20/minute. Head and neck examinations were normal except for pale conjunctiva. She did not have petechiae or lymphadenopathy. The abdomen was soft without tenderness, shifting dullness or palpable mass.



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E-mail: khilai@vghks.gov.tw • Received: June 5, 2007 • Accepted: September 28, 2007

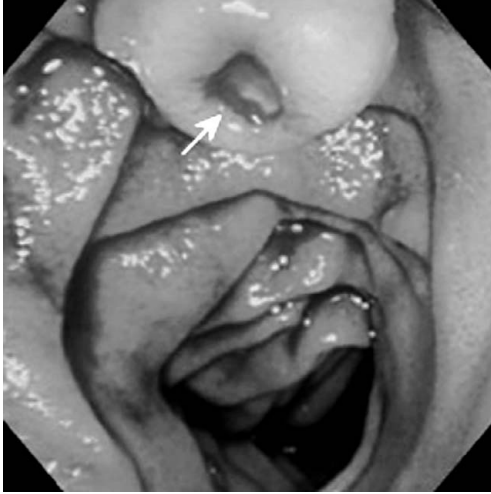


Figure 1. On enteroscopy, a papilla-like nodule mimicking a submucosal tumor with a central depression (arrow) was observed occupying the proximal jejunum.

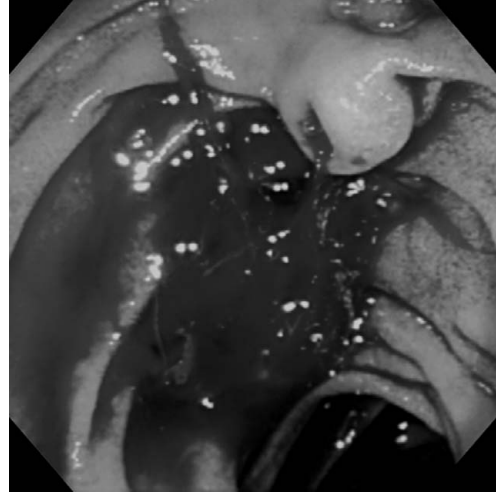


Figure 2. Active bleeding from the central depression of the tumor was seen during enteroscopy.

Rectal examination demonstrated the presence of maroon-colored liquid stool. The hemogram showed hemoglobin 8.4 g/dL and hematocrit 25.2%. Liver and renal function were normal. No coagulopathy or thrombocytopenia was found.

Upper GI endoscopy was repeated but failed to find the bleeder. Upper GI barium meal study led to the suspicion of a diverticulum occupying the duodenal second portion. Small intestinal bleeding was suspected due to the clinical presentation. Push enteroscopy was then performed, which disclosed a juxtapapillary diverticulum at the duodenal second portion and a papilla-like nodule about 1.5 cm in diameter with normal overriding mucosa and a central depression at the proximal jejunum (Figure 1). Active bleeding from the central depression was also noted (Figure 2). A stromal tumor of the jejunum was suspected initially.

The patient was referred for emergent operation. Exploratory laparotomy revealed a 1.5-cm nodule occupying the proximal jejunum about 20 cm distal to the Treitz ligament (Figure 3). Wedge resection of the tumor was carried out. Pathologic examination showed that the tumor was composed of some cystic exocrine-type ducts and bundles of smooth muscle without pancreatic acinar component, indicating an adenomyoma (Figure 4). The patient has remained symptom-free after operation.

Discussion

Reviewing the literature, 5 adult cases of jejunal adenomyoma have previously been reported.^{3-5,8} Olmsted et al presented 2 cases of jejunal adenomyoma without



Figure 3. Exploratory laparotomy showed a bleeding nodule at the proximal jejunum (arrow).

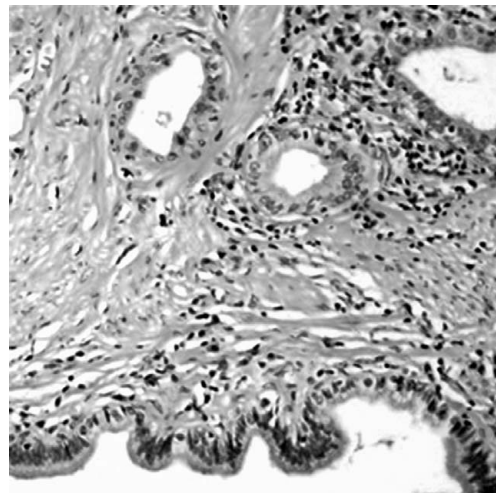


Figure 4. Pathologic studies revealed that the tumor was composed of focal cystic dilatation of exocrine-type ducts and bands of smooth muscle without pancreatic acinar component over the submucosal area.

Table 1. Cases of jejunal adenomyoma in the literature

Study	Age (yr)/Sex	Size (cm)	Location	Symptoms
Clarke ³	64/male	1.0	Pm + Ss	Incidental finding, necropsy
Lee et al ⁴	18/male	4.5	Sm + Pm	Intussusception
Park et al ⁵	63/male	1.3	Sm + Pm	Incidental finding
This case	74/female	1.5	Sm + Pm	Jejunal bleeding

Pm = proper muscle; Ss = subserosa; Sm = submucosa.

detailing clinical data in 1987.⁸ The details of 4 other cases including ours are shown in Table 1.³⁻⁵ Three were male and older than 60 years. Lesion size ranged from 1 cm to 4.5 cm. The location of adenomyoma was mainly in the submucosa and proper muscle. The clinical presentations were intussusception in 1 case, asymptomatic in 2 cases (found accidentally by necropsy and surgical resection for colon cancer) and 1 small intestinal bleeding in our case. In a retrospective report, the nonmalignant etiologies of small intestinal bleeding were mainly from benign stromal tumor, Meckel's diverticulum and angiodysplasia.⁹ Bleeding from ectopic pancreas only occurred in 3.9% (3/76) of patients.⁹ Jejunal adenomyoma with bleeding has not been reported previously. However, it should be considered a cause of small intestinal bleeding in adults.

Adenomyoma of the small intestine is composed of abnormal glandular formations lined with tall columnar epithelium and surrounded by smooth muscle.¹⁻⁶ Sometimes, the glands show cystic changes like the findings in this case.⁴ Pathologically, adenomyoma and ectopic pancreas are considered as variants of the same process.^{1,4} Adenomyoma differs from ectopic pancreas in that the acinar and islet-like tissues are not present. However, their clinical presentations are similar. Joo et al presented a case of jejunal ectopic pancreas with massive bleeding in 2001.¹⁰ This case was also the first report of jejunal bleeding from an ectopic pancreas. Usually, adenomyoma or ectopic pancreas in the GI tract is mostly asymptomatic, but they are capable of producing symptoms such as GI bleeding.

The diagnosis of small intestinal bleeding is not easy because of the limitation of imaging studies. Angiography or surgical laparotomy seems to be effective for diagnosis in a limited number of patients according to a retrospective report.⁹ However, currently, there are some reports advocating capsule endoscopy and double-balloon enteroscopy for the management of obscure GI bleeding after upper and lower endoscopy.¹¹⁻¹⁴ Capsule endoscopy may be helpful to determine the nature of occult intestinal bleeding, but this method is only for diagnosis and does not provide any therapy

for active bleeding.^{11,12} On the other hand, the traditional push enteroscopic diagnostic rate is only around 7.8%.⁹ The effectiveness of double-balloon enteroscopy in diagnosing small intestinal lesions is not well documented.^{13,14} In addition, the procedure is not very popular, which may explain why endoscopic findings of adenomyoma in the small intestine have rarely been mentioned before. In our case, the gross appearance of the adenomyoma was a papilla-like submucosal nodule with a central depression mimicking a submucosal tumor (Figures 1 and 2). It was difficult to differentiate from a stromal tumor because both of them were located in the submucosa and proper muscle. Final diagnosis depended on histologic study.

Surgical treatment seems to be the only effective management for this disorder.⁹ Laparoscopic or exploratory laparotomy combined with partial enterectomy has some good results not only for treatment but also for diagnosis.^{1-4,6,7} Simple wedge resection is also effective, as in our case. Conservative treatment has a high risk of failure if there is massive GI bleeding. Experience with endoscopic treatment has not been reported; it may be effective for stopping bleeding but difficult to exclude potential malignancy.

In summary, adenomyoma of the jejunum is a rare benign lesion of the GI tract. In this report, we described GI bleeding as a result of this disorder. Enteroscopy may be helpful in aiding diagnosis. Surgical treatment is needed for successful management. The final diagnosis relies on pathologic study.

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