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A Small Jejunal Angioleiomyoma Detected by Double-Balloon Enteroscopy: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Male, 42
Final Diagnosis: Jejunal angioleiomyoma
Symptoms: Melena
Medication: —
Clinical Procedure: Double balloon enteroscopy
Specialty: Gastroenterology and Hepatology

Objective: Rare disease
Background: Angioleiomyoma in the small intestine is a rare cause of gastrointestinal bleeding. Only 7 cases of angioleiomyoma in the small intestine were reported in the English literature, with 4 of them causing gastrointestinal bleeding. The diagnosis of angioleiomyomas in the small intestine before surgery is difficult.
Case Report: We report the case of a 42-year-old man with recurrent melena who underwent repeated esophagogastroduodenoscopy and colonoscopy, without positive finding. During a double-balloon enteroscopy, an elevated lesion with a diameter of 6 mm was found in the jejunum. The lesion was resected laparoscopically assisted with double-balloon enteroscopy. A microscopic examination showed fibric membrane of the mass and numerous vascular channels surrounded by proliferated smooth muscle. There were exudative fibrin and many thrombi formed by red blood cells. Immunohistochemistry was positive for SMA and CD34. A pathological diagnosis of jejunal angioleiomyoma with thrombus was established. During a 5-year follow-up, there was no further gastrointestinal bleeding.
Conclusions: The gastroenterologists should consider angioleiomyoma in the small intestine when assessing obscure gastrointestinal bleeding.

MeSH Keywords: Angiomyoma • Double-Balloon Enteroscopy • Gastrointestinal Hemorrhage • Intestine, Small

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/910884>

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Background

Among all the causes of gastrointestinal bleeding, benign tumors account for less than 1% of cases. Angioleiomyoma is a rare benign tumor in the small intestine, which can lead to bleeding or intussusception. Some authors suggested that these tumors might be vascular malformations, while others suggested that they could be a specific type of hamartoma. Three theories have been proposed for the origin of angioleiomyomas: from aberrant undifferentiated mesenchyme, from smooth muscle in the wall of blood vessels, or from both [1]. Angioleiomyomas have been classified into solid, cavernous, and venous histological types. These tumors are slow-growing, firm, grayish-white, round-to-oval nodules. The histological characteristics of angioleiomyoma are benign smooth muscle tumor with prominent abnormal vascular channels ranging from dilated capillaries to thick-walled arteries and veins with slit-like lumens. Degenerative changes can be found in these lesions, such as thrombosis, hyalinization, and dystrophic calcification. Arterial supply and venous drainage disturbances, inherent to such a mixed vascular and muscular proliferation, can cause intratumoral necrosis and mucosal ulceration, which in turn are responsible for intestinal bleeding [2]. Management is usually surgical resection of the affected bowel segment. Identifying a small angioleiomyoma in the small intestine before surgery is often difficult. Here, we report a case of jejunal angioleiomyoma detected by double-balloon enteroscopy (DBE).

Case Report

A 42-year-old male patient presented with a 9-month history of recurrent melena. There was no hematemesis or abdominal pain. He did not have any known comorbidities. His hemodynamic status was stable and the physical examination was unremarkable. Laboratory tests revealed hemoglobin 12.0 g/dL and hematocrit 36.0%. The patient's clotting profile was normal. A fecal occult blood test was positive. The results of repeated esophagogastroduodenoscopy and colonoscopy were normal. There was no abnormality on the abdominal CT scanning. He underwent DBE via the antegrade approach, which demonstrated a small elevated lesion with apical depression and bleeding in the middle part of the jejunum (Figure 1). Endoscopic biopsy showed chronic mucosal inflammation. The patient underwent laparoscopy, but the lesion could not be found during laparoscopy because of its small size and intraluminal growth pattern. An intraoperative DBE was performed and the lesion was located by the light of the enteroscope, which was found at 2 meters from the incisor. The lesion was resected and the anastomosis was hand-sewn. The procedure lasted about 180 min. The patient had an unremarkable postoperative recovery. Macroscopic pathological examination showed a submucosal grey and red mass

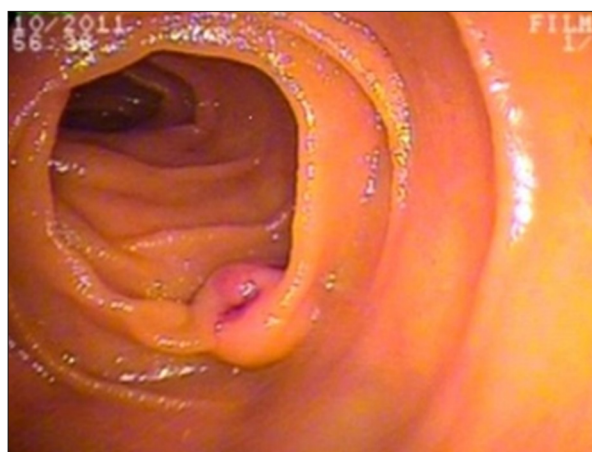


Figure 1. A small elevated lesion with apical depression and bleeding in the middle part of the jejunum.

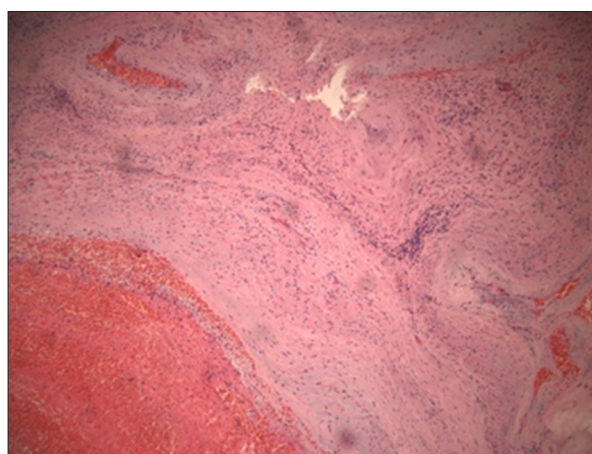


Figure 2. Vascular channels surrounded by proliferated smooth muscle.

with a diameter of 6 mm. Microscopic examination showed fibric membrane of the mass and many vascular channels surrounded by proliferated smooth muscle (Figure 2). There were exudative fibrin and many thrombi formed by red blood cells. Immunohistochemistry was negative for CD117, DOG-1, and S-100, but positive for SMA and CD34. A pathological diagnosis of jejunal angioleiomyoma with thrombus was established. During a 5-year follow-up, there was no further gastrointestinal bleeding and the patient did not undergo further enteroscopy or relevant imaging examination.

Discussion

Angioleiomyoma was first described in 1937 by Stout [3]. The incidence is highest in people age 30–60 years, and this disorder is more frequent in females than in males [4]. Angioleiomyomas commonly affect the skin and subcutaneous tissue of the lower extremities. Angioleiomyomas can occur in many unusual sites,

including the oral cavity, atrium, scrotum, and broad ligaments of the female genital tract, but rarely affect the small intestine. To the best of our knowledge, only 7 cases of angioleiomyoma in the small intestine were reported in the English literature, with 4 of them causing gastrointestinal bleeding [5–9]. This is the first reported case in the English literature, which was found with DBE and then resected laparoscopically assisted with DBE. Angioleiomyomas are benign tumors originating from smooth muscle cells of arterial or venous walls. The diagnosis of angioleiomyomas in the small intestine before surgery is difficult. Esophagogastroduodenoscopy and colonoscopy are usually used to find the causes of gastrointestinal bleeding, and angioleiomyoma in the small intestine is rarely diagnosed preoperatively. CT and MRI can be performed to detect gastrointestinal angioleiomyomas, but these methods can only find large lesions. Mesenteric angiography is helpful in diagnosing gastrointestinal angioleiomyomas. Definitive diagnosis can be made by immunohistochemical tests. Immunohistochemistry is important for the differentiation of angioleiomyomas from other neoplasia of soft tissue, mostly mesenchymal lesions with a predominance of fusiform cells [10]. Smooth-muscle actin corresponds to the alpha fraction of the actin chain and it is a specific immunomarker of smooth muscle. In the present case, the neoplastic cells showed positive immunoreactions to actin. CD34 is a transmembrane protein that is broadly expressed

by vascular endothelium, and the endothelial cells presented immunoreactivity against anti-CD34 antibody. Using DBE, we found an angioleiomyoma with a diameter of only 6 mm, and the diameter of another reported angioleiomyoma diagnosed by DBE in the small intestine was 5 mm [7]. It seems that DBE can detect angioleiomyomas in the small intestine during the early stage of these lesions better than other diagnostic methods. Traditionally, angioleiomyomas in the small intestine are treated with surgical resection or vascular intervention. The present case shows that a small angioleiomyoma in the small intestine can be managed successfully by minimally invasive surgery assisted with DBE.

Conclusions

Angioleiomyomas, although rare in the small intestine, should be considered in the differential diagnosis of obscure gastrointestinal bleeding. DBE is a useful tool for detecting angioleiomyomas in the small intestine and may play a key role in the surgical treatment of small lesions.

Conflicts of interest

None.

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