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Small intestine angioleiomyoma as a rare cause of perforation: A case report and literature review

Angioleiomyoma-related bowel perforation

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Abstract

BACKGROUND

Angioleiomyoma is a rare and benign stromal tumor typically found in subcutaneous tissue. It rarely occurs in the gastrointestinal tract. Among the reported cases, the most common complication was gastrointestinal bleeding. Perforation has only been reported as a complication in the last few decades.

CASE SUMMARY

This case report detailed the discovery of intestinal angioleiomyoma in a 47-year-old male presenting with abdominal pain that had persisted for 3 d. After suspecting hollow organ perforation, surgical intervention involving intestinal resection and anastomosis was performed.

CONCLUSION

The report underscores the significance of early surgical intervention in effectively treating angioleiomyoma while emphasizing the pivotal role of timely and appropriate measures for favorable outcomes.

Key Words: Angioleiomyoma; Intestinal perforation; Abdomen; Acute; Diarrhea; Case report

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Core Tip: Here we report the first case of angioleiomyoma in the small intestine with a complication of perforation. This type of complication is extremely rare as the last published report of a perforation complication related to angioleiomyoma was published 30 years ago in Russian.

INTRODUCTION

Angioleiomyomas are a vascular subtype of leiomyomas, categorized as benign smooth muscle tumors. The tumor itself is characterized by inclusion of vessel and smooth muscle cells[1]. Angioleiomyomas mostly arise from the wall of a vein. The majority of cases occur at 30 years to 60 years of age, with female predominance and location in a lower extremity[2]. Although, angioleiomyomas have been found in different organs[3-5]. Within the gastrointestinal tract, occurrences of angioleiomyomas are exceptionally rare[6]. This case report presents the inaugural instance documented in the English literature and details a patient who exhibited hollow organ perforation attributable to angioleiomyoma.

CASE PRESENTATION

Chief complaints

A 47-year-old male presented with lower abdominal pain and diarrhea.

History of present illness

The patient reported that his symptoms started 3 d prior to presentation, with both gradually progressing over that period. He also reported having a fever that would go up to 39 °C for 1 wk.

History of past illness

The patient had a medical history of controlled hypertension for many years.

Personal and family history

The patient's personal and family history was unremarkable.

Physical examination

The physical examination revealed the following: Body temperature, 36.7 °C; heart rate, 105 beats per min; respiratory rate, 18 breaths per min; and blood pressure, 125/80

mmHg. The skin and mucous membranes were free of yellow staining, rash, bleeding spots, liver palms, and spider nevus. Chest breathing was normal. However, the tenderness over the low abdomen and rebounding pain was mentioned.

Laboratory examinations

Laboratory tests indicated elevated C-reactive protein (335 mg/L; normal range: < 5 mg/L) and increased creatinine (1.81 mg/dL; normal range: 0.64-1.27 mg/dL).

Imaging examinations

Computed tomography (CT) showed pneumoperitoneum and a mass over the pelvic area (Figure 1A and 1B).

FINAL DIAGNOSIS

Ileum angioleiomyoma with complications of perforation and pneumoperitoneum.

TREATMENT

We initially suspected a perforation associated with sigmoid colon cancer. Therefore, surgical intervention was initiated approximately 7 h after admission. An exploratory laparotomy revealed a tumor, measuring 8 cm × 5 cm, located 70 cm from the ileocecal valve. It was surrounded by an abscess formation (Figure 2A and 2B). A thorough examination of the stomach, intestine, and colon did not reveal additional perforation. The tumor was excised followed by end-to-end anastomosis.

OUTCOME AND FOLLOW-UP

Postoperatively, the patient was transferred to the ward for continued care and was discharged 2 wk after the operation. The patient returned to our clinic for follow-up after 2 wk without complaint of abdominal pain, nausea nor vomiting. The immunohistochemical analysis of the resected specimen corroborated the diagnosis of angioleiomyoma.

DISCUSSION

Leiomyomas are most frequently located in the uterine myometrium (95.0%), followed by the skin (3.0%) and the gastrointestinal tract (1.5%)[6]. In 1969, the World Health Organization defined leiomyomas as well-circumscribed benign tumors composed of bundles of mature smooth muscle cells. The World Health Organization classification includes three main groups: Solid leiomyoma; vascular leiomyoma (angioleiomyoma); and epithelioid leiomyoma (leiomyoblastoma)[7]. Angioleiomyoma, initially described by Stout[8] in 1937, commonly affects the skin and subcutaneous tissue of the lower extremities. Its presence in the gastrointestinal tract is exceptionally rare. Angioleiomyomas in the gastrointestinal tract occur predominantly in the jejunum (44%), followed by the ileum (37%) and the duodenum (19%).

Four distinct subtypes of angioleiomyoma have been identified[2]. The first type is capillary or solid angioleiomyomas. They are characterized by a rich stratification of smooth muscle cells surrounding a few slit-like vascular channels. This subtype is the most common. The second type is venous angioleiomyomas, which is distinguished by more vascular channels and walls that are thicker compared to capillary angioleiomyomas. The third type is cavernous angioleiomyomas, and this subtype features widened vascular channels surrounded by a thin layer of smooth muscle cells. The fourth type is combined capillary and venous angioleiomyomas[9, 10].

Our patient was diagnosed with cavernous angioleiomyoma based on histological findings. Sections showed ileal tissue with a well-circumscribed submucosal tumor composed of proliferative spindle smooth muscle cells bearing brightly eosinophilic cytoplasm and arranged in fascicles, punctuated by variable-sized vascular channels. The vessels were irregularly dilated with attenuated walls. There was also a lack of a thick muscular wall (Figure 3A-C).

The clinical presentation of angioleiomyoma is diverse. Uncomplicated cases are typically asymptomatic. However, a limited number of reported cases exhibit rare clinical presentations. Therefore, it is challenging to establish comprehensive clinical features and

complication rates. Only 11 cases of gastrointestinal angioleiomyoma have been documented in the literature. Table 1 compares these cases with our own.

Gastrointestinal bleeding is the most commonly reported complication associated with angioleiomyoma. Past cases have reported other complications, such as intussusception and prolapse. Notably, our case represents the first reported instance of angioleiomyoma-related perforation in the English literature.

In our case, preoperative imaging, specifically CT, revealed a lesion located in the pelvic region accompanied by pneumoperitoneum. We initially suspected malignancy-related hollow organ perforation. Despite the absence of tarry or bloody stool reported by the patient, CT indicated a tumor of 9.5 cm × 6.5 cm × 6.4 cm in size that was surrounded by abscess.

Surgical resection of the affected bowel segment is the typical treatment to manage the angioleiomyoma and any accompanying complications. Resection is typically followed by side-to-side anastomosis, which was also performed in our case. Given the initial suspicion of hollow organ perforation, a comprehensive inspection of the entire abdomen was essential to identify potential perforations caused by other etiologies. This thorough examination was crucial to ensure an accurate diagnosis and appropriate intervention.

CONCLUSION

In summary, small bowel angioleiomyoma is a rare occurrence. Perforation is also a rare complication related to angioleiomyoma. Nonetheless, early surgical intervention plays a crucial role in the effective treatment of angioleiomyoma. Timely and appropriate surgical measures are pivotal for a favorable outcome in such cases.

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