

Cavernous hemangioma of the mesorectum involving the rectum: A rare case report

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Case report

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Abstract

Background: Gastrointestinal hemangiomas are very rare and are even rarer in the mesentery. It is not known whether they originate in the bowel wall or the mesentery currently. Some studies have described mesenteric hemangiomas that originate in the bowel wall and extend into the mesentery. We herein describe a case of cavernous hemangioma of the mesorectum.

Case presentation: A 31-year-old male presented with hematochezia and sensation of rectal tenesmus. Both the rectal MRI and contrast-enhanced CT scan of the whole abdomen indicated rectal wall thickening, marked dilated and tortuous vessels around the rectum. In addition, an anomalous vein had arisen from the superior mesenteric vein, extending down to the mesorectum. The patient underwent laparoscopic surgical resection of the mesorectal lesion and the involved portions of the rectum. The surgical samples underwent pathological analysis, and a diagnosis of mesenteric cavernous hemangioma was confirmed. Seven days after surgery, the patient was discharged without postoperative complications.

Conclusion: Mesenteric hemangiomas are extremely rare tumors. In our present case, an anomalous vein had arisen from the superior mesenteric vein, extending down to the mesorectum. This findings suggest that the mesenteric hemangioma more likely originated in the mesentery.

Background

Hemangiomas are benign proliferating vascular malformations, characterized by abnormal proliferation of blood vessels and mesenchymal tissue. They may be congenital or appear soon after birth(1). Hemangiomas are often solitary lesions. The presence of multiple hemangiomas or those that extensively involve an organ or body part are termed hemangiomatosis(2). Hemangiomas in the gastrointestinal system are very rare, accounting for only 0.05% of intestinal tumors(3, 4) and are even rarer in the mesentery; fewer than twenty have been reported.

Based on their pathology, hemangiomas are classified into three categories: capillary, cavernous, and mixed type, but the majority of mesenteric hemangiomas reported thus far are of the cavernous type. No definite gender predominance has been identified. Patients can present at any age, but mesenteric hemangiomas are more common in young adults often in the third decade of life(2).

It is not clear whether mesenteric hemangiomas originate in the bowel wall or in the mesentery. Some reported cases have been thought to originate in the bowel wall and extend into the mesentery(5–12). In previous reports, hemangiomas have been mostly located in the small intestine mesentery(5, 6, 8–12).

Case Presentation

A 31-year-old male developed a sensation of rectal tenesmus one month before admission. He consulted the local hospital and proctitis was considered. The symptoms did not improve significantly after

treatment. One week later, hematochezia suddenly occurred, resulting in blood loss of about 100 ml. The bleeding resolved without treatment. The patient was admitted to our hospital.

His physical examination was unremarkable except for the digital rectal examination, which identified a soft mass, about 3cm × 3 cm in size, that grossly bled on exam. The Complete blood count did not reveal anemia. A total colonoscopy revealed a discoid protuberance of mucosa at the rectum 7 cm from the anus, and erythema of the mucosal surface. The subsequent rectal biopsy indicated chronic inflammatory changes of the mucosa. A contrast-enhanced CT scan of the whole abdomen revealed rectal wall thickening, marked dilatation, and tortuous vessels around the rectum with scattered calcifications (Fig. 1A). There was an anomalous vein that arose from the superior mesenteric vein and extended down to the mesorectum (Fig. 1B). The rectal MRI showed wall thickening of the involved portion of the rectum. A high signal intensity lesion was seen on T2-weighted sequences and was more clearly depicted with fat suppression (Fig. 2). Hemangioma of the mesorectum was considered after a multidisciplinary team discussion.

Three days after admission, the patient underwent laparoscopic surgical resection of the involved portions of the rectum and mesorectum, as well as a coloanal anastomosis. Surgical samples were taken and sent for pathological examination. The pathology viewings indicated a dark red area in the mesentery adjacent to the peritoneal reflection; the lesion was spongy and there was blood outflow from the incision. The immunohistochemical results indicated CD34-positive results, whereas the lymphatic endothelial marker D2-40 was negative. The lesion was diagnosed as a cavernous hemangioma of the mesentery. The patient had no postoperative complications following laparoscopic surgical resection and returned home on postoperative day seven.

Discussion

Hemangiomas are a benign proliferative congenital vascular malformation that originate from the embryonic sequestrations of mesodermal tissue and can be found in any organ. They have an incidence of approximately 2%-3%(2, 4). While intestinal hemangiomas are well-known and have previously been described in the literature, mesenteric hemangiomas represent one of the rarest sites for hemangiomas. Mesenteric hemangiomas may originate either from the bowel wall and the mesentery, or the mesentery alone(7). We herein report a case of a mesenteric hemangioma that presented with an anomalous vein that arose from the superior mesenteric vein and extended down to the mesorectum. These findings suggest that, in this case, the mesenteric hemangioma originated in the mesentery and ultimately involved the intestinal wall.

Clinical presentations and diagnoses of mesenteric hemangiomas depend on their size, localization, and intestinal wall involvement. Smaller tumors are often asymptomatic whereas symptomatic masses tend to be large at the time of diagnosis. Larger hemangiomas located in the mesentery can cause intraperitoneal hemorrhage and can often result in life-threatening conditions. When mesenteric hemangiomas involve the bowel wall, they may result in intraluminal bleeding. Therefore, the clinical

manifestations are hematemesis or melena. Long-term chronic occult blood loss is an important cause of anemia.

Currently, radiological evaluation plays an important role in the noninvasive examination of abdominal diseases, including mesenteric hemangioma. Imaging examinations, such as CT and MRI, cannot be used to unequivocally diagnose mesenteric hemangiomas, but they do allow a more precise evaluation of the size, vascularization, and possible involvement of adjacent structures of the tumor(7). Calcifications were seen scattered throughout the lesion on CT. Some studies suggested that this was due to degenerative changes as a consequence of the thrombosis within the sinuses, caused by perivascular inflammation and stasis of blood flow(4, 11). Other studies have shown that calcifications may ultimately form phleboliths, which represent an important diagnostic feature, seen in 26 to 50% of affected adult patients(4). Phleboliths are usually signal voided on both T1 and T2 weighted images, and thrombosed vessels show serpiginous structures with high signal intensity on MRI(11). An important imaging finding in our case suggested that a mesenteric hemangioma originated from the mesentery rather than the bowel wall, which is different from some reported cases. A contrast-enhanced portal venous phase CT scan showed an anomalous vein arising from the superior mesenteric vein and extending down to the mesorectum. Image examination also revealed transmural thickening of the wall of the involved bowel loops. On CT hemangiomas may be homogeneous or heterogeneous with possible cystic structures. The heterogeneity of hemangiomas may be due to intralesional degeneration consisting of hemorrhage, fibrosis, or calcification(2). On MRI, hemangiomas usually have an iso- or hypo-intense signal on T1-weighted images. On T2-weighted images, hemangiomas typically show high signal intensity and are more clearly depicted with fat suppression. Degenerative changes, such as fibrosis, result in a heterogeneous signal intensity on T2-weighted images. Intratumoral hemorrhage may show hypointense signal on T2-weighted images, making hemangiomas hard to differentiate from other solid mesenchymal tumors, such as fibromas or leiomyomas (4). Characteristic discontinuous peripheral nodular progressive enhancement patterns can be seen in some mesenteric hemangiomas. However, enhancement may be heterogeneous or minimal or may be present only on delayed phases likely due to internal fibrotic changes(2).

A definitive diagnosis of cavernous hemangioma of the mesentery depends on histopathological examination. Because of the high risk of hemorrhage, biopsy of clinically suspected hemangiomas is not recommended. Histologically, cavernous hemangiomas are composed of dilated vessels of varying sizes, which are lined with flattened endothelial cells with little fibrous connective tissue between the vascular channels. The vessel lumen often shows thrombosis and contains a large number of erythrocytes(9, 13, 14). To avoid the risk of bleeding during biopsy, we used immunohistochemistry to diagnose our patient. The results were positive for CD34, but were negative for the lymphatic endothelial marker D2-40. CD34 is expressed not only in vascular endothelial cells, but also in lymphatic endothelial cells, while D2-40 is only expressed in lymphatic endothelial cells. Combined with vascular tumor suggested by histology, the diagnosis of cavernous hemangioma was made. In addition, CD31 is vascular endothelium-specific, and is often as a diagnostic marker of hemangioma(5, 9).

The optimal treatment of mesenteric hemangiomas is surgical resection of the mesenteric lesion and the involved intestinal segments (3, 8). Recurrence after complete resection are rare. Other nonoperative techniques, such as low-dose radiation therapy, cryotherapy, brachytherapy, sclerotherapy or interventional angiography, often result in symptom recurrence and are therefore only temporary solutions(3, 4).

Conclusions

Mesenteric hemangiomas are extremely rare tumors, and it is not known whether they originate in the bowel wall or the mesentery. Some studies have described mesenteric hemangiomas that originate in the bowel wall and extend into the mesentery. In this current case, the mesenteric hemangioma more likely originated in the mesentery. We used CT imaging for diagnosis and to evaluate the size, vascularization and possible involvement of adjacent structures of the mesenteric hemangioma. We surgically resected the mesenteric lesions and affected intestinal segments. Recurrences after complete resection are rare, and we are confident that our patient will not experience a recurrence of this tumor.

Declarations

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Authors' contributions

WB designed the report. ZY wrote the paper and provided and analyzed the pictures of CT and MRI. All authors read and approved the final manuscript.

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Consent for publication

Written informed consent for publication was obtained from the participant

Competing interests

The authors declare that they have no competing interests

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Figures

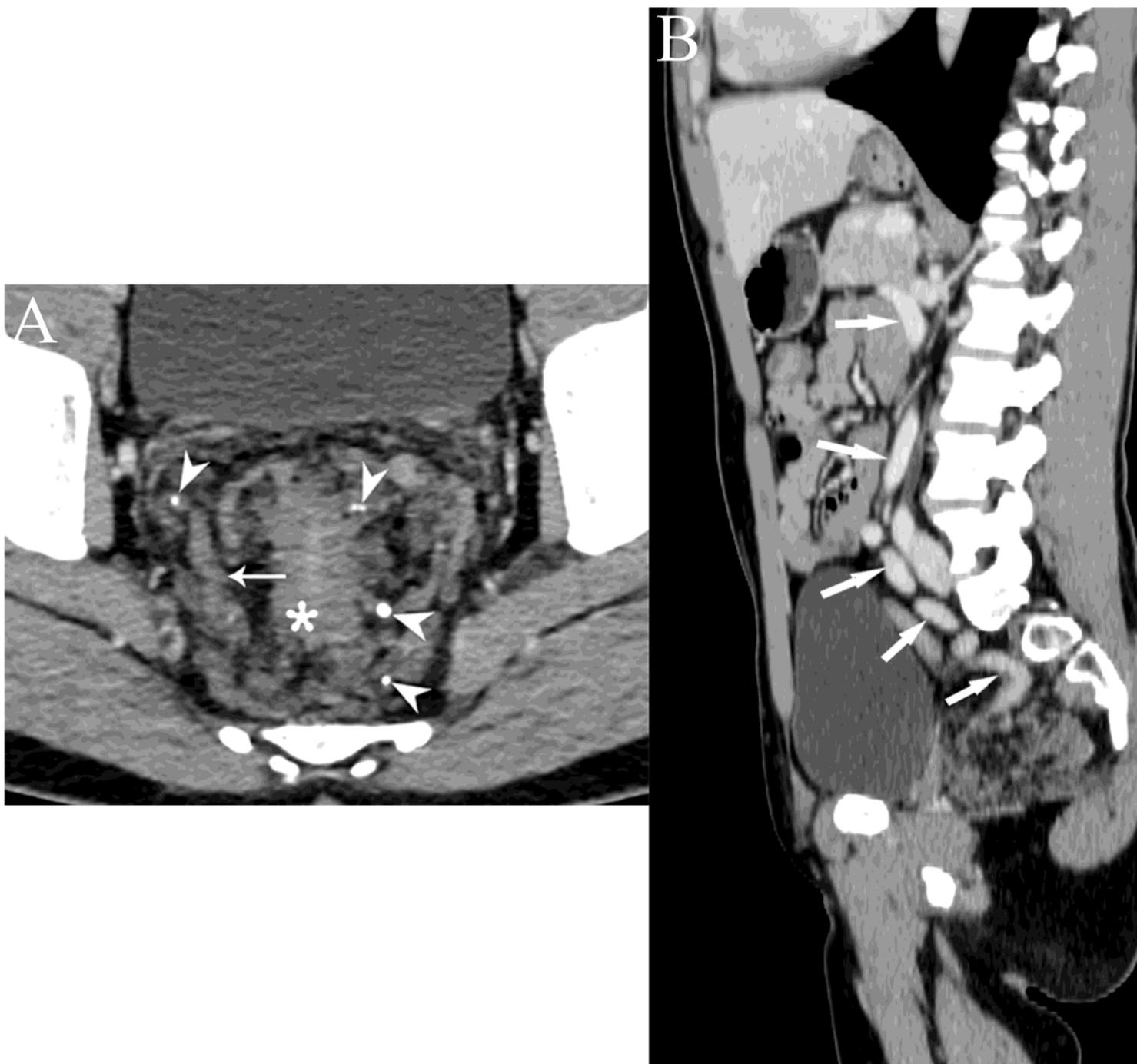


Figure 1

Contrast-enhanced portal venous phase CT scan. (A) axial view: CT showing wall thickening of rectum (asterisk), dilatation and tortuous vessels around the rectum (arrow) with scattered calcifications (arrowhead). (B) Sagittal view: There is an anomalous vein that arose from the superior mesenteric vein and extended down to the mesorectum (arrow).

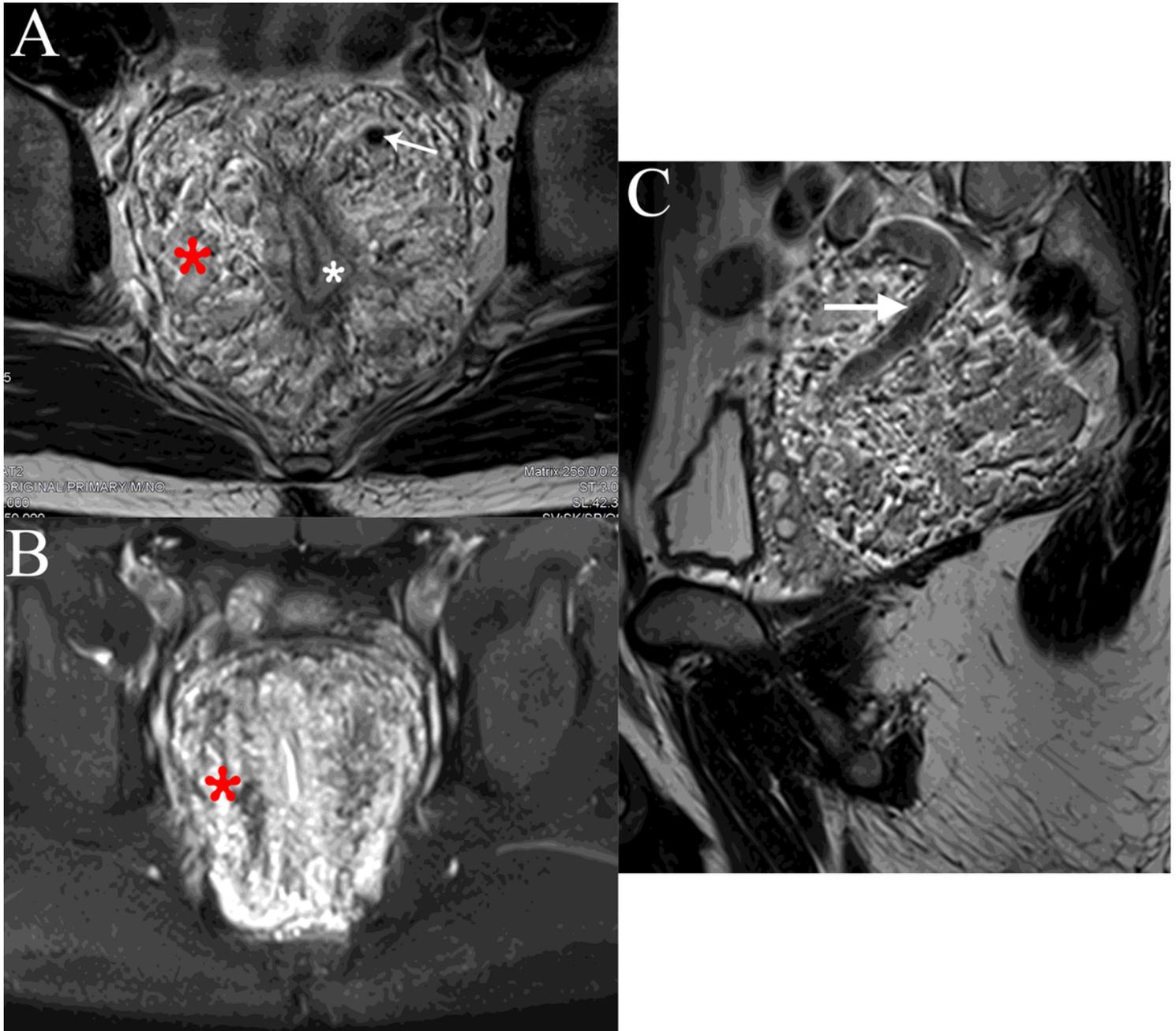


Figure 2

Rectal MRI. (A) Axial T2-weighted images showing wall thickening of the rectum (white asterisk), heterogeneous high signal intensity of the lesion (red asterisk) and black signal vessel (flowing void effect) (arrow). (B) Axial fat-suppressed T2-weighted images with the high signal intensity lesion more clearly depicted (red asterisk). (C) Sagittal T2-weighted images showing the anomalous vessel arising from the superior mesenteric vein (arrow).