Clinical Case Report

Medicine®

OPEN

Inferior mesenteric arteriovenous fistula with nonpulsatile abdominal mass

A case report and a mini-review

Ling Cheng, MD[a], Ruifeng Zhao, MD[a], Di Guo, MD[a], Kailin Cai, MD[b], Kaifang Zou, MD[a], Jun Yang, MD[c,*], Liangru Zhu, MD[a,*]

Abstract

Rationale: Inferior mesenteric arteriovenous fistula (IMAVF) is a rare condition principally characterized by portal hypertension and ischemic bowel disease. Up to now, only 30 cases have been reported. Presented here is an IMAVF patient with nonpulsatile abdominal mass as the main manifestation.

Patient concerns: A 62-year-old Chinese male who complained of abdominal discomfort for a month was admitted to our hospital. Physical examination revealed a hard and hardly mobile mass.

Diagnoses: Space-occupying lesions were first suspected but endoscopy did not reveal any masses. The computed tomography angiography exhibited no definite boundary between the inferior mesenteric artery and vein. The patient was diagnosed with IMAVF.

Interventions: The treatment of IMAVF mainly includes intra-arterial embolization and surgery. In our case, fistulas were complex and the patient had symptoms of colon ischemia, so we suggested a surgical resection instead of embolization. And the postoperative biopsy also confirmed the diagnosis.

Outcomes: After surgery, gastrointestinal symptoms disappeared and the patient began to gain weight gradually. During the follow-up, colonoscopy showed that the anastomotic astium and colonic mucosa were normal.

Lessons: Analysis of the case showed that computed tomography angiography is an important auxiliary examination for establishing the diagnosis of IMAVF and surgery is an effective treatment.

Abbreviations: CTA = computed tomography angiography, IMAVF = inferior mesenteric arteriovenous fistula.

Keywords: abdominal mass, arteriovenous fistula, inferior mesenteric artery

1. Introduction

Inferior mesenteric arteriovenous fistula (IMAVF) can be congenital or acquired. Congenital IMAVF originates from undifferentiated embryonic vessels that fail to regress and lead to a communication between the arterial and venous system.[1,2] The acquired IMAVF results from previous trauma or surgery. There exists an abnormal shunt between the inferior mesenteric artery and vein, and the main manifestations include portal hypertension and ischemic bowel disease. This condition is extremely rare and, to date, only 30 cases have been reported worldwide.[1,3–5] The disease is hard to be differentiated from other conditions of digestive system. This report described a less common form of the disease. Our study showed that computed tomography angiography (CTA) is an essential tool for the diagnosis of IMAVF. The disease can be effectively treated by embolization and surgical resection and the prognosis is good. Timely treatment can avoid gastrointestinal bleeding and other serious complications. We hope that this case report will help clinicians raise awareness of the disease.

2. Case report

A 62-year-old Chinese male who complained of abdominal discomfort for a month was admitted to our hospital. The main symptoms were abdominal distention, diarrhea, fever, loss of appetite, and weight loss of 10 kg. He did not have abdominal pain, nausea, vomiting, and gastrointestinal bleeding. Physical examination revealed a hard and hardly mobile mass of about 15×9 cm, without tenderness, located in the lower left quadrant. No other positive findings except paleness were noted. The patient had a history of 30 years of hypertension, which was well controlled by oral medication. He had not been subjected to surgery or trauma. At the local hospital, ultrasound-guided fine needle biopsy suggested chronic inflammatory lesions. The gastrointestinal symptoms were not significantly improved after

Editor: N/A.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

The authors have no funding and conflicts of interest to disclose.

[a] Division of Gastroenterology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, [b] Department of Gastrointestinal Surgery, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, China, [c] Department of Urology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, China.

[*] Correspondence: Jun Yang, Department of Urology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, 1277 Jiefang Avenue, Wuhan 430022, China (e-mail: jimyoung_39@126.com), Liangru Zhu, Division of Gastroenterology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, 1277 Jiefang Avenue, Wuhan 430022, China (e-mail: zhuliangru05@163.com).

Medicine (2017) 96:48(e8717)

Received: 12 September 2017 / Accepted: 12 October 2017

http://dx.doi.org/10.1097/MD.0000000000008717

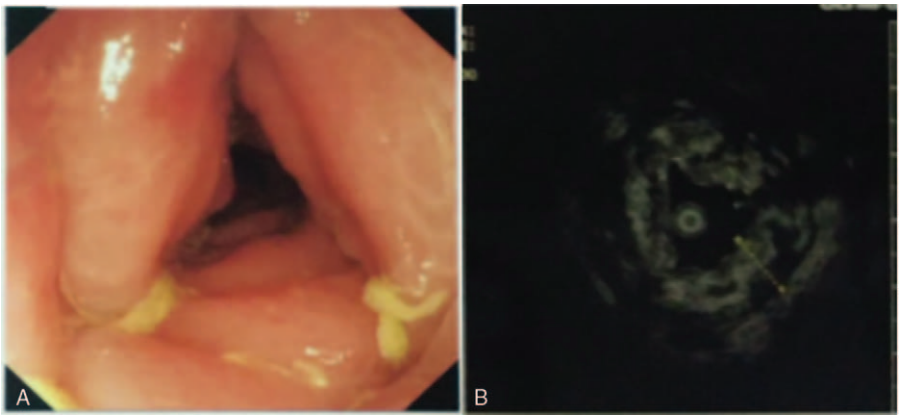

Figure 1. Endoscopy findings in the colon: (A) colonoscopy image and (B) endoscopic ultrasonography image.

antibiotic therapy. For definite diagnosis, the patient was referred to our hospital.

The laboratory tests yielded the following findings: The peripheral blood hemoglobin was 116 g/L (normal range 130–175 g/L), platelet 357 G/L (normal range 125–350 G/L), erythrocyte sedimentation rate 25 mm/h (normally under 15 mm/h), CA-125 320.1 U/mL (normally <35 U/mL), prostate-specific antigen 5.38 μg/L (normally <4 μg/L), and ferritin 302.8 μg/L (normal range 21.8–275 μg/L). C-reactive protein was within normal range and fecal occult blood test was weakly positive. Stool culture was negative. Gastric endoscopy was roughly normal. Colonoscopy showed mucosal edema, luminal stenosis, and deformation in the sigmoid colon (Fig. 1A). Abdominal ultrasonography revealed an irregularly shaped slightly higher echo mass of about 21.4 × 7.9 cm at the lower abdomen of the surrounding part of the intestinal tubes. This mass was believed to be solid, and its nature was unknown. In the abdominal cavity, there were dark areas suggestive of free liquid, one of them being about 2.6 cm in the anteroposterior diameter. Endoscopic ultrasonography in sigmoid colon and rectum suggested significant thickening of the whole layer of the intestinal wall, mucosal layer, with submucosal boundary blurred, and inherent muscular layer visible (Fig. 1B). Endoscopic colon biopsy showed chronic inflammatory lesions. Lower abdomen contrast-enhanced computed tomography exhibited edema of rectum and left semicolon and thickening of the colon wall. The mesenteric and peritoneal fat spaces and omentum became nebulous. In the arterial phase, there was a significantly enhanced nodule of about 10 mm in diameter at the splenic flexure of colon. Positron emission tomography-computed tomography suggested edema, thickening, luminal stenosis, and deformation of the colonic wall, with increased metabolism. CTA demonstrated that the inferior mesenteric vein was tortuous and dilated, and there was a tumor-like expansion of about 1.5 × 1.4 cm in the left lower quadrant (Fig. 2). The inferior mesenteric vein was close to the branch of adjacent inferior mesenteric artery, but the boundary was hazy (Fig. 2). The patient was diagnosed with IMAVF and was laparotomically treated. Intraoperatively, the bowel segment supplied by the inferior mesenteric blood vessels was found to have ischemic changes, the affected bowel wall becoming thick, pale, and stiff (Fig. 3A and B). The corresponding mesentery diffusely thickened and appendices epiploicae hardened, with extensive saponification spots (Fig. 3C). There existed yellow and clear ascites in the abdominal cavity (Fig. 3D). We performed a ligation of the inferior mesenteric blood vessels near the roots. The left hemicolon, sigmoid colon, upper rectum, and

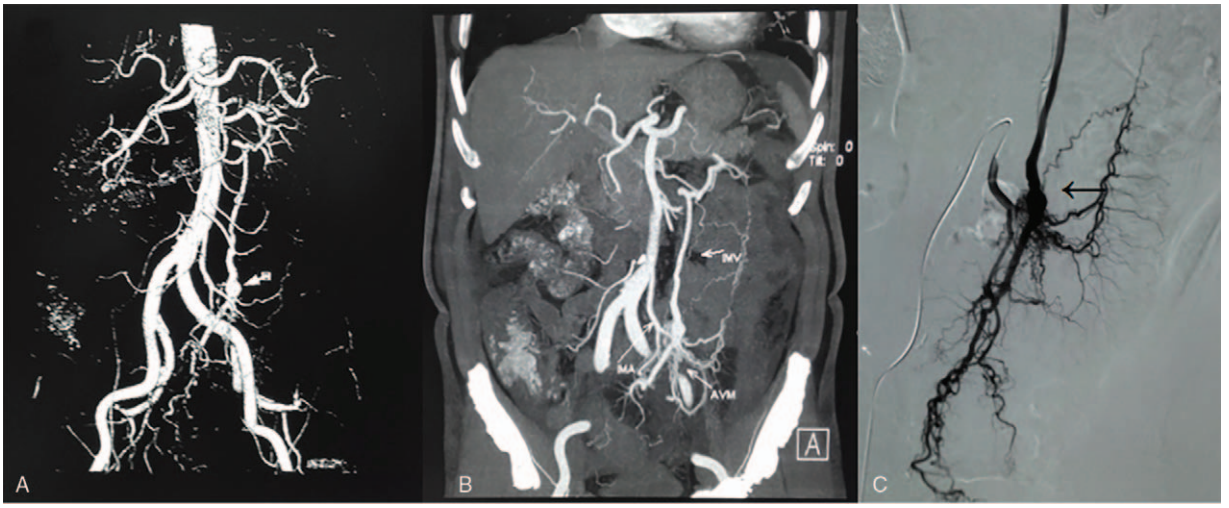

Figure 2. Computed tomography angiography findings of mesenteric vessels: (A) 3-dimensional reconstruction image, (B) coronal plane, and (C) arteriovenous malformation.

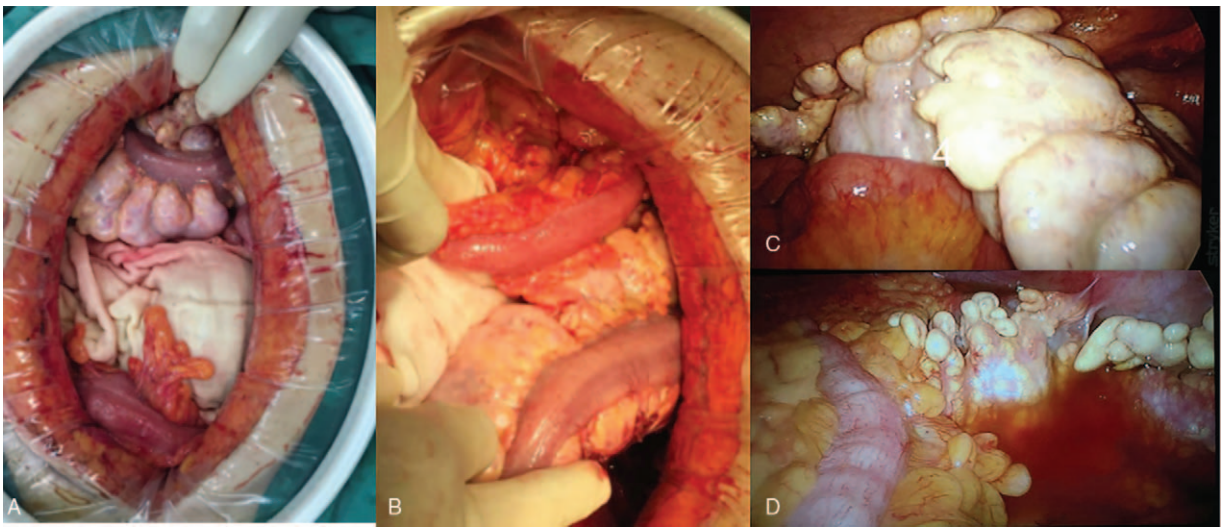

Figure 3. Image of exploratory laparotomy: (A, B) the affected bowel, (C) saponification spots, and (D) ascites.

corresponding mesentery were then resected. The transverse colon was anastomosed with the rectum. Pathological diagnosis included mucosal edema, mesentery thickening, which were in line with the clinical diagnosis of IMAVF and granulomatous chronic inflammation (Fig. 4). After surgery, gastrointestinal symptoms disappeared and the patient gained weight gradually. Eight months later, colonoscopy showed that the anastomotic astium and colonic mucosa were normal (Fig. 5).

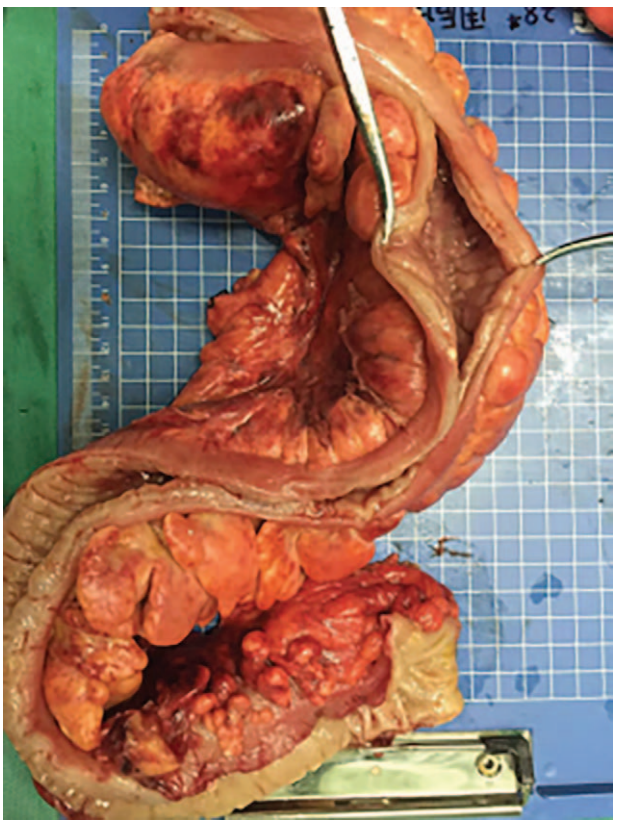

Figure 4. Surgical sample of left hemicolon, sigmoid colon, and part of rectum.

3. Discussion

IMAVF is extremely rare. It is usually secondary to previous operation, trauma, neoplasms, rupture of an arterial aneurysm into an adjacent vein, and, in some cases, idiopathic.[4,6] Our literature search found 17 congenital cases and 13 acquired ones. This case was idiopathic and its etiology was unknown. The average age of all cases was 58 years and male patients accounted for 67.7%.

Two major clinical manifestations of IMAVF are portal hypertension and ischemic bowel disease. Splenomegaly, ascites, and esophageal varicosity are symptoms typical of portal hypertension. Capuano et al[7] reported a 76-year-old woman with IMAVF who mainly presented with ascites. Ischemic bowel disease tends to cause abdominal pain, diarrhea, gastrointestinal hemorrhage, and abdominal masses.[8] According to the reports, 18 patients had abdominal pain and 16 patients had gastrointestinal bleeding symptom. In this case, the main symptom of this

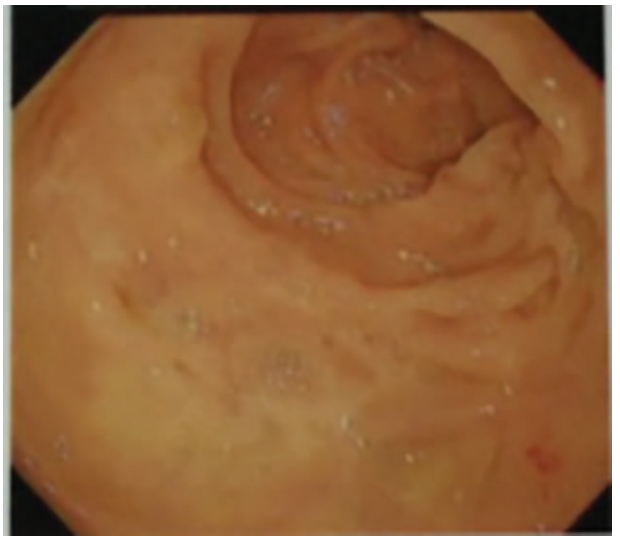

Figure 5. Colonoscopy after operation.

patient was abdominal mass. This symptom is rare and usually leads to misdiagnosis and missed diagnosis.

Because of its low incidence, there are no generally accepted diagnostic criteria at present. Angiography can help establish the diagnosis of IMAVF and typically shows early contrast filling of the involved vein during the arterial phase.[4] It can provide reliable diagnostic information in mesenteric ischemia due to its higher spatial resolution and faster acquisition time, allowing relatively accurate assessment of the peripheral visceral branches. In this patient, CTA demonstrated a local tumor-like dilated inferior mesenteric vein in the left lower abdomen. There was no definite boundary between the inferior mesenteric artery and vein and this is a typical sign of IMAVF.

The treatment of IMAVF mainly includes intra-arterial embolization and surgery. Embolization is an effective management that can avoid surgical resection. Hendy et al[3] reported a male case of IMAVF presenting with ischemic proctosigmoiditis who was treated by radiological embolization and his symptoms were improved shortly after the treatment. Brucher et al[9] described a patient with poorly differentiated intramucosal sigmoid adenocarcinoma who was treated by percutaneous endovascular arterial embolization. Embolization is less invasive and relatively safer but is associated with organ ischemia or recurrence especially if more than 1 feeding vessel are involved.[1] And the migration of embolization material is likely when the IMAVF diameter is >8 mm and flow rate is high.[10] Surgery may be more suitable for these patients. Akgun[11] reported a case of arteriovenous malformation treated with total colectomy and terminal ileostomy and the patient's recovery was uneventful. In total, 14 cases received surgical treatment, 11 embolization, and 4 combination therapy. Only 1 case refused to receive any treatment. In our case, fistulas were complex and the patient had symptoms of colon ischemia, so we suggested a surgical resection instead of embolization. After surgery, the abdominal symptoms gradually disappeared. Further colonoscopy showed that the anastomotic astium and colon mucosa were normal. Follow-up showed that the patient was generally in good condition without abdominal symptoms.

4. Conclusions

IMAVF is a very rare disease that may cause ischemic bowel disease. Nonpulsatile abdominal mass is a less common manifestation of the disease. CTA is an important diagnostic tool and surgery is an effective treatment. Timely management can avoid severe complications, such as gastrointestinal hemorrhage. With abdominal masses, clinicians should include IMAVF as a diagnostic possibility.

References

[1] Athanasiou A, Michalinos A, Alexandrou A, et al. Inferior mesenteric arteriovenous fistula: case report and world-literature review. World J Gastroenterol 2014;20:8298–303.
[2] Metcalf DR, Nivatvongs S, Andrews JC. Ischemic colitis: an unusual case of inferior mesenteric arteriovenous fistula causing venous hypertension. Report of a case. Dis Colon Rectum 2008;51:1422–4.
[3] Hendy P, Cheng EH, Livsey R, et al. Curative embolization of an inferior mesenteric arteriovenous fistula causing ischaemic proctosigmoiditis. ANZ J Surg 2015.
[4] Faghihi Langroudi T, Shabestari AA, Pourghorban R, et al. Idiopathic inferior mesenteric arteriovenous fistula: a rare cause of pulsatile abdominal mass. Indian J Surg 2015;77(suppl 1):84–6.
[5] Jeon DO, Park JS, Kim JE, et al. A case of traumatic inferior mesenteric arteriovenous fistula. Korean J Gastroenterol 2013;62:296–300.
[6] Justaniah AI, Molgaard C, Flacke S, et al. Congenital inferior mesenteric arteriovenous malformation presenting with ischemic colitis: endovascular treatment. J Vasc Interv Radiol 2013;24:1761–3.
[7] Capuano G, Pomponi D, Iaccarino V, et al. An unusual case of ascites. Dig Liver Dis 2004;36:628–31.
[8] Okada K, Furusyo N, Sawayama Y, et al. Inferior mesenteric arteriovenous fistula eight years after sigmoidectomy. Intern Med (Tokyo, Japan) 2002;41:543–8.
[9] Brucher N, Moskovitch G, Otal P, et al. Inferior mesenteric arteriovenous fistula treated by percutaneous arterial embolization: a breathtaking story!. Diagn Interv Imaging 2014;95:85–6.
[10] Turkvatan A, Ozdemir Akdur P, Akdogan M, et al. Inferior mesenteric arteriovenous fistula with ischemic colitis: multidetector computed tomographic angiography for diagnosis. Turk J Gastroenterol 2009; 20:67–70.
[11] Akgun V, Sari S, Verim S, et al. Arteriovenous malformation of the inferior mesenteric artery in a patient with ischaemic colitis. BMJ Case Rep 2013;Epub ahead of print.