

## รายงานการตรวจพบการเกิดภาวะหลอดเลือดดำและหลอดเลือดแดงซูพีเรียมีเซนเทอรีเชื่อมต่อกันในผู้ป่วยหลอดเลือดดำในช่องท้องอุดตัน

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### บทคัดย่อ

ภาวะหลอดเลือดดำและหลอดเลือดแดงซูพีเรียมีเซนเทอรีเชื่อมต่อกัน เป็นภาวะที่พบได้น้อย ไม่มีลักษณะจำเพาะของการแสดงโรค อาการของโรคจะขึ้นกับขนาดและตำแหน่งของความผิดปกติของหลอดเลือดที่เกิดขึ้น การรักษาโดยการผ่าตัดเป็นวิธีดั้งเดิมที่ใช้กันแพร่หลาย แต่ในปัจจุบันการรักษาโดยผ่านการสวนหลอดเลือดกำลังได้รับการยอมรับมากขึ้น ผู้นิพนธ์ได้รายงานการรักษาผู้ป่วยภาวะหลอดเลือดดำในช่องท้องอุดตันที่มีอาการปวดท้องรุนแรง และตรวจพบมีภาวะหลอดเลือดดำและหลอดเลือดแดงซูพีเรียเชื่อมต่อกันร่วมด้วย

**คำสำคัญ:** ภาวะหลอดเลือดดำและหลอดเลือดแดงซูพีเรียมีเซนเทอรีเชื่อมต่อกัน, ภาวะหลอดเลือดดำในช่องท้องอุดตัน, การรักษาโดยการใส่สายสวนหลอดเลือด

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# Superior mesenteric arteriovenous fistula with previous mesenteric vein thrombosis: case report

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## Abstract

Superior mesenteric arteriovenous fistula (SMAVF) is a rare vascular disorder. The clinical presentation of SMAVF is atypical, depending on the size and location of the fistula. Potential treatment of mesenteric AVFs is surgical correction of the AVFs with or without bowel resection. Another choice is percutaneous endovascular treatment. We present an unusual case of a 54-year-old man with crampy abdominal pain, initially thought to be ischemic bowel from mesenteric vein thrombosis. Further workup revealed multiple SMAVFs.

**Keywords:** superior mesenteric arteriovenous fistula, mesenteric vein thrombosis, endovascular treatment

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## Introduction

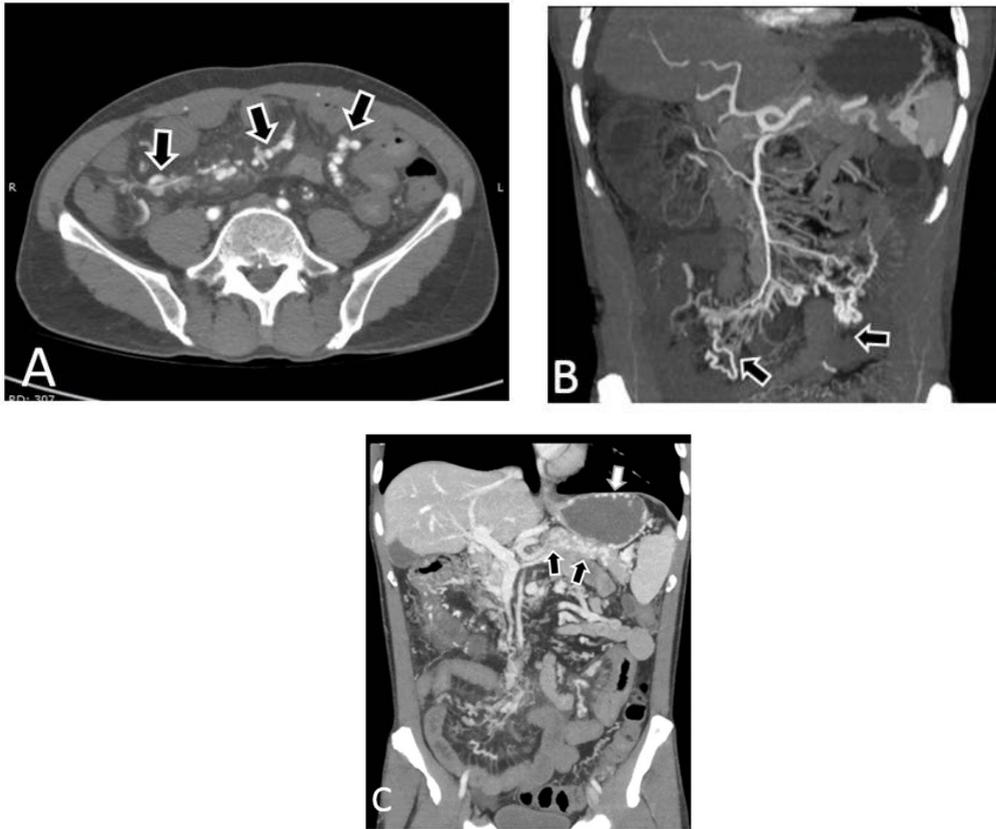
Superior mesenteric arteriovenous fistula (SMAVF) is a rare vascular disorder. The clinical presentation of SMAVF is atypical, depending on the size and location of the fistula. Patients may be asymptomatic or may present with crampy abdominal pain, portal hypertension, liver impairment, and even gastrointestinal bleeding. We present an unusual case of a 54-year-old man with crampy abdominal pain, initially thought to be ischemic bowel from mesenteric vein thrombosis. Further workup revealed multiple SMAVFs.

## Case Report

A 54-year-old man presented with postprandial left lower crampy abdominal pain for 2 months with watery diarrhea. The patient was treated by regional hospital as enteritis and readmitted in hospital many times but the clinical was not improved so he was referred to the tertiary hospital. Before this, the patient has history of right leg deep venous thrombosis 2 years ago and took vitamin K antagonist for 3 months without any investigation of thrombophilia cause or repeated Doppler ultrasonography.

The patient could not recall previous similar symptoms and had no history of previous abdominal surgery or trauma. Abdominal examination revealed normal bowel sound, no abnormal abdominal bruit, tenderness in the left lower quadrant without abdominal guarding. The digital rectal examination was unremarkable.

Computerized Tomography Venography (CTV) and Computerized Tomography Arteriography (CTA) of the abdomen were done which demonstrated the presence of small thrombus in proximal part of SMV, cavernous transformation around the porta hepatitis, and chronic thrombosis with small in size of right portal and splenic vein. Early filling of SMV with tortuous of venous structure in small bowel mesentery and ascending mesocolon, was suspected for arteriovenous fistula secondary to SMV thrombosis. Other CT findings were diffuse bowel wall edema, Multiple gastric varices and less enhancement at distal small bowel loop and ascending colon, which suggested bowel ischemia without evidence of bowel necrosis. Another CT finding is atrophy of right lobe liver which was likely due to chronic right portal vein thrombosis (Figure 1).



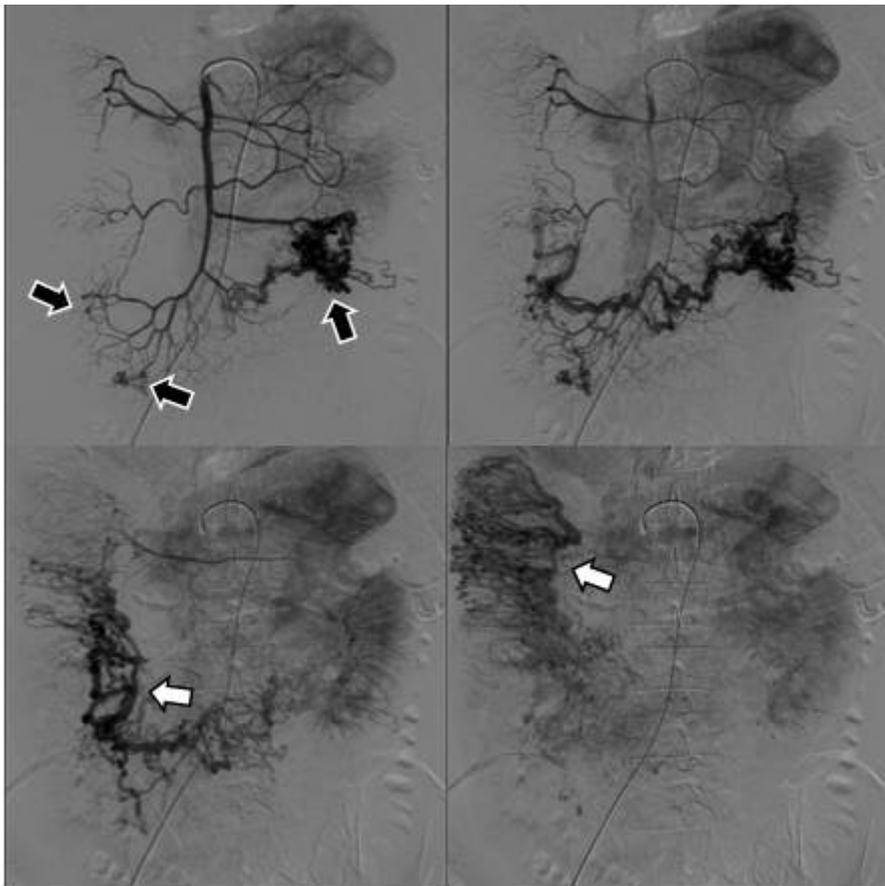
**Figure 1** CT scan arterial phase axial view (A), arterial phase coronal MIP reformatted (B), and portovenous phase coronal view (C) showed early opacification of the mesenteric vein on arterial phase with abnormal tortuous course (arrow in A and B). Small size of splenic vein with abnormal luminal irregularity is demonstrated, suspected of splenic vein thrombosis with partial recanalization (arrow in C). Diffuse bowel wall thickening is also demonstrated. Multiple gastric varices, secondary to splenic vein thrombosis is seen (white arrow in C).

After an informed consent was obtained, the patient was performed conventional angiography with 5Fr. Cobra catheter (Terumo, Japan) was placed in the origin of SMA to confirm the diagnosis and to provide more information about the fistula. The study revealed normal patency along the SMA. Dilated tortuous early

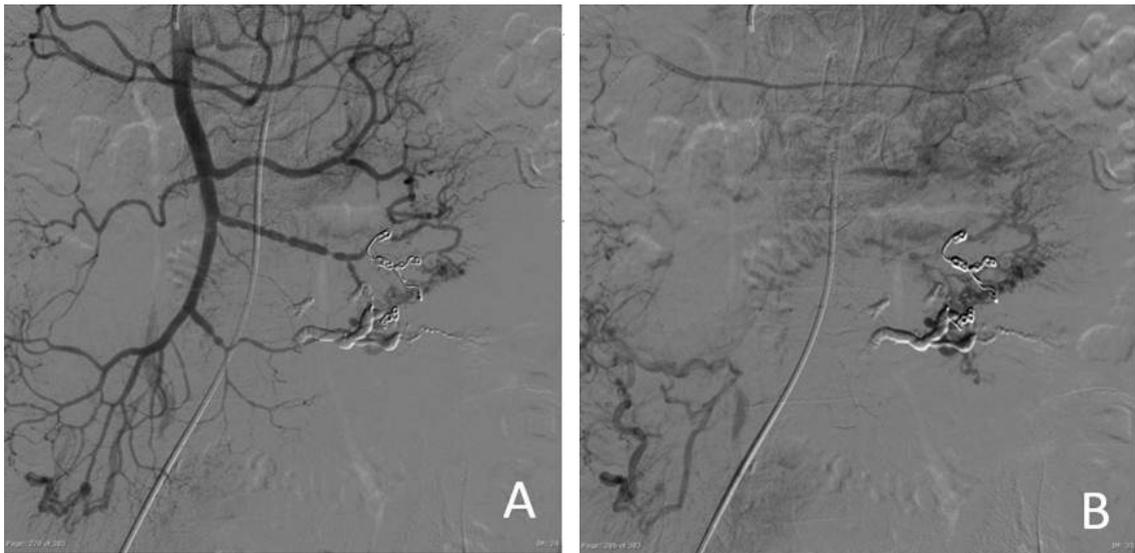
draining veins are noted from the jejunal and ileocolic branches (Figure 2). We decided to treat as endovascular technique first because the fistula was quite small. Therefore, super selection into small feeders from jejunal branch was performed using a 2.8 Fr Renegade microcatheter (Boston, Ireland). Multiple embolization with 5 mm x 5.5 mm

VortX-18 coils (Boston, Ireland) and 4 mm x 3.7 mm VorTX Daimond-18 coil (Boston, Ireland) were done. Post embolization showed a decreasing in number of early draining veins from jejunal branch but the

residual mesenteric shunts still presented (Figure 3). The patient was returned to bed and started oral diet two days after the procedure.



**Figure 2** SMA angiogram reveals mild hypertrophy of jejunal branch and iliocolic branches of SMA with adjacent dilated tortuous early draining veins (black arrows), suggestive of mesenteric AV fistula. These veins are draining into the dilated collateral veins along ascending colon on delayed phase (white arrows).



**Figure 3** SMA angiogram after glue and coils embolization at the jejunal branch of SMA (early (A) and delayed (B) phases) shows significant decrease AV shunting at the jejunal branch of SMA.

One week later the clumpy abdominal pain after meal occurred again so the reintervention was done. The second angiography showed occlusion of jejunal branches from previous coil embolization. Decreased amount of shunt from previous procedure and decreased collateral veins along the right side colon are demonstrated but the residual mesenteric shunt still remained. Superselection into the small jejunal branch was performed again. The embolization with 20% concentration glue was done. Post embolization showed decreased total occlusion of early draining veins from jejunal branch, however, minimal residual shunt still remained. Therefore, in order to reduce chance of bowel ischemia, we stopped the procedure and observed

the patient's clinical. On the next day, the patient clinical was improved. He could take oral diet without pain for 2 weeks. We attempted to improve his nutritional status by enteral and additional parenteral nutrition. Unfortunately, crampy abdominal pain after meal recurred. We decided to perform diagnostic laparotomy and laparoscopic partial small bowel resection by using the coil as a marker to identify pathologic segment.

Two weeks later, the patient was discharged in good condition. He was also put on anticoagulation therapy with warfarin due to his newly diagnosis of protein C deficiency syndrome. The dose was adjusted to achieve an international normalized ratio of 2 to 3. The patient came back to the

hospital for a checkup in one month. Upper gastrointestinal endoscopy showed slight gastric varices and dilated superficial gastric vein. The patient was doing well on follow-up visits after every 6 months. We also followed up the patient for the next 2 years, he has been doing well. He has continuously taking the warfarin and wearing gradual compressive stocking to treat his underlying disease and prevent post thrombotic syndrome.

## Discussion

Since the SMAVF was first reported by Movitz<sup>1</sup>, few cases have been reported in the world. SMAVF can be either congenital<sup>2</sup> or acquired. Most common causes for acquired SMAVF are abdominal surgery and abdominal trauma such as after bowel resection, gastrectomy or pancreatectomy<sup>3-6</sup> iatrogenic SMAVF typically result from ligation of both artery and vein together during bowel resection or ligature placement in the mesentery without the precise localization of the bleeding point. The most common site are ileocolic artery which sutured material was found near the site of the fistula in several cases<sup>7</sup>.

The presentations are vary such as abdominal pain, diarrhea caused by bowel ischemia<sup>8</sup>. Portal hypertension from the long standing high flow shunt<sup>9</sup>, resulting in esophageal varices, upper gastrointestinal tract hemorrhage or ascites<sup>10</sup>.

In this case, the patient's history of mesenteric vein thrombosis misled us to a diagnosis of bowel ischemia from extensive mesenteric thrombosis without significant peritonitis. Therefore, we started the treatment with anticoagulant, fluid resuscitation and close monitoring. After the CT scan was reviewed and the result showed early filling of SMV with tortuous of venous structure in small bowel mesentery and ascending mesocolon, we then asked the patient more about his past history and found that before this illness he could not eat as much as before because the severe clampy abdominal pain would occur 15-30 min after meal. This information suggested the clinical of steal phenomenon from arteriovenous shunt.

Potential treatment of mesenteric AVFs is surgical correction of the AVFs with or without bowel resection. Another choice is percutaneous endovascular embolization of the feeding artery<sup>11</sup>.

Recently, percutaneous endovascular treatment has been increasingly performed, using covered stents or embolization<sup>12-16</sup>. We decided to perform endovascular treatment first because we did the diagnostic angiography to identify the precisely fistula location and also the fistula's size. After we have identified that the fistula were quite small and the risks of distal migration of embolic material at fistula's site was low, therefore, we performed the embolization.

After the second attempt, the partial success with embolization due to multiple feeding vessels of AVFs was observed. We decided on laparoscopic segmental resection of bowel segment using the coil as a marker of the pathologic segment. The pathological report showed mixture of markedly abnormal arteries and veins including all layer of intestine.

We have reviewed the report about the patient with mesenteric thrombosis associated with mesenteric fistula<sup>17</sup>. This phenomenon may be due to new vessel formation within the mesenteric vein thrombosis and active lysis of the thrombus exposing the organizing vessels to the distal superior mesenteric veins, as same as the pathogenesis of cavernous sinus fistula<sup>18-19</sup>. Nevertheless, we still cannot clearly summarize that this case is the sequel of mesenteric thrombosis or the congenital AVM.

In conclusion, the patients' SMAVF clinical symptoms are often nonspecific and physical examination may not reveal the presence of abdominal bruit. Food fear symptom may be presented not only in patient with chronic mesenteric artery disease but also in patient with SMAVF. Potential treatment of mesenteric AVFs is surgical correction of the AVFs with or without bowel resection. Another choice is percutaneous endovascular embolization which is increasingly performed nowadays.

## References

1. Movitz D. Postoperative arteriovenous aneurysm in mesentery after small bowel resection. *JAMA* 1960;173:42-4.
2. Takehara H, Komi N, Hino M. Congenital arteriovenous fistula of the superior mesenteric vessels. *J Pediatr Surg* 1988;23:1029-31.
3. Weinstein D, Altshuler A, Belinki A, et al. Superior mesenteric artery to superior mesenteric vein arteriovenous fistula presenting as abdominal pain and gastrointestinal bleeding 3 years after an abdominal gunshot wound: Report of a case and review of the literature. *J Trauma* 2009;66:13-6.
4. Wu CG, Li YD, Li MH. Post-traumatic superior mesenteric arteriovenous fistula: Endovascular treatment with a covered stent. *J Vasc Surg* 2008;47:654-6.
5. Bratton CF, Hamid A, Selby JB, et al. Case report: gastrointestinal hemorrhage caused by a pancreas transplant arteriovenous fistula with large pseudoaneurysm 9 years after transplantation. *Transplantation Proc* 2011;43(10):4039-43.
6. Deitrick J. Traumatic superior mesenteric artery-portal fistula. *Ann Vasc Surg* 1990;4:72-6.
7. Kato S, Nakagawa T, Kobayashi H, et al. Superior mesenteric arteriovenous fistula: Report of a case and review of the literature. *Surgery Today* 1993;23:73-7.

8. An T, Zhou S, Song J, et al. Massive gastrointestinal bleeding secondary to superior mesenteric arteriovenous fistula. *Am J Gastroenterol* 2013;108: 1662-5.
9. Bettenworth D, Rijcken E, Muller KM, et al. Rare cause of upper gastrointestinal bleeding in a 27-year-old male patient. *Gut* 2012;61:1367.
10. Nie L, Luo XF, Li X. Gastrointestinal bleeding caused by extrahepatic arterioportal fistula associated with portal vein thrombosis. *World J Gastroenterol* 2012;18:6501-3.
11. Lee S, Chung J, Ahn B, et al. Inferior mesenteric arteriovenous fistula. *Ann Surg Treat Res* 2017;93(4):225-8.
12. Shintani T, Mitsuoka H, Masuda M. Transcatheter coil embolization of an iatrogenic superior mesenteric arteriovenous fistula: Report of a case. *Surgery Today* 2011;41:556-9.
13. Yeo KK, Dawson DL, Brooks JL, et al. Percutaneous treatment of a large superior mesenteric artery pseudoaneurysm and arteriovenous fistula: A case report. *J Vasc Surg* 2008;48:730-4.
14. Liu L, Yu Y, Jin X, et al. Management of superior mesenteric arteriovenous fistula after small bowel resection 20 years previously: Endovascular treatment. *Can J Gastroenterol* 2012;26:864-5.
15. Langroudi T, Shabestari A, Pourghorban R, et al. Idiopathic inferior mesenteric arteriovenous fistula: A rare cause of pulsatile abdominal mass. *Indian J Surg.* 2015;77:84-86.
16. Wang C, Zhu W, Guo GH, et al. Superior mesenteric arteriovenous fistula presenting as gastrointestinal bleeding: case report and literature review. *Rev Esp Enferm Dig* 2016;108(8):503-7.
17. Matsunaga N, Hayashi K, Mori H, et al. Mesenteric arteriovenous shunt associated with thrombosis of the portal venous system: case report. *Cardiovasc Intervent Radiol* 1998;11(5):281-4.
18. Houser O, Campbell J, Campbell R, et al. Arteriovenous malformation affecting the transverse dural sinus: acquired lesion. *Mayo Clin Proc* 1979; 54(10):651-61.
19. Debrun GM, Viñuela F, Fox AJ, et al. Indications for treatment and classification of 132 carotid-cavernous fistulas. *J Neurosurg* 1988;22:285-9.