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Superior mesenteric arteriovenous fistula presenting as gastrointestinal bleeding: Case report and literature review

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ABSTRACT
Superior mesenteric arteriovenous fistula (SMAVF) is a rare vascular disorder usually following penetrating abdominal trauma or gastrointestinal surgery. Percutaneous endovascular treatment such as embolization, has been widely used to treat this disease. We report a patient, who was presented with melena at the onset of his symptoms, then an acute hematemesis in shock. A SMAVF was diagnosed on an angiogram after a large mesenteric vein was seen on CT. The patient had a successful emergency endoscopic variceal ligation (EVL) to stop bleeding. Then the patient received fistula embolization with covered stent.

Keywords: Superior mesenteric arteriovenous fistula (SMAVF) endoscopic variceal ligation (EVL) Covered stent

INTRODUCTION
Superior mesenteric arteriovenous fistula (SMAVF) is uncommon vascular deformity that is easily misdiagnosed. The clinical presentation of SMAVF is atypical, which depends on the size and location of the fistula. Patients may be asymptomatic or
may present with crampy abdominal pain with or without diarrhea, portal hypertension, liver impairment, and even gastrointestinal bleeding (1). SMAVF may cause severe portal hypertension (2), which leads to upper and lower gastrointestinal bleeding, especially esophageal varices bleeding (EVB), which is not common, however is the most dangerous and high mortality.

We present an unusual case of a 34-year-old man presented with melena, initially thought to be secondary to bleeding varices from cirrhosis related to hepatitis B virus (HBV) infection. Further workup revealed a large SMAVF as the cause of severe portal hypertension and gastroesophageal varices.

CASE REPORT
A 34-year-old man presented with melena, without complaints of chronic abdominal pain. Other significant past medical history included the repair of colonic laceration or injury 2 years before presentation and transfusion history. At the time of admission, his pulse rate was 71 beats/min, and his arterial blood pressure was 104/62 mmHg. His physical examination revealed marked anemic face and shifting dullness. The patient’s medical history was not remarkable for chronic disease, drug use, smoking, or alcohol use. The patient also had HBV infection, which was incidentally discovered during his hospitalization; HBV antigens, HBeAb and HBcAb antibodies were positive on serological tests (HBsAg >250 IU/ml, HBeAb = 0.01 S/CO, HBcAb = 12.93 S/CO and HBV-DNA 2.19e+004 IU/ml). The laboratory tests disclosed the following values: Decreased red blood cells (3.28 × 10^12/L), hemoglobin (87 g/L) and albumin (32.4 g/L) levels suggested anemia; elevated prothrombin time (PT 15.1 s), activated partial thromboplastin time (APTT 45.6 s), direct bilirubin (9.9 umol/L) levels suggested coagulation impairment, while other blood tests, such as white blood cells, platelet, transaminase (aspartate aminotransferase/alanine aminotransferase), alkaline phosphatase, total bilirubin, creatinine and alpha fetoprotein (AFP ) remained within normal ranges.

Work up started with gastroscopy. Multiple severe esophageal and duodenal descendent varices and portal hypertensive gastropathy were found. Full
colonoscopy revealed old blood, no fresh blood. However, no source of bleeding was identified. Therefore, further examinations were taken, such as hypotonic contrast X ray examination, computerized tomography venography (CTV) and computerized tomography arteriography (CTA) of the abdomen, which demonstrated the presence of a large aneurysm filled by the superior mesenteric artery (SMA), draining into the superior mesenteric vein (SMV). Other CT findings were the presence of ascites, hydrothorax and gallstone (Fig 1). These findings were consistent with those expected for an SMAVF.

A surgical consult was requested by the primary service for potential surgical fistula ligation; however, the patient was considered a poor surgical candidate because of his history of previous abdominal surgery with possible extensive adhesions. Although existed the risk of small bowel ischemia, combined with the therapeutic schedule which was discussed with patient, endovascular embolization of the fistula was considered. Depending on the examination of CT we primally decided to use balloon catheter to occlude the fistula. The patient was brought to the angiography suite, and a selective diagnostic superior mesenteric arteriogram was performed via a right femoral approach. This confirmed the presence of an SMAVF. The fistula was tortuous and measured 17 mm in diameter. Portal vein was obviously enlarged and measured 49mm in diameter (Fig 2) The fistula was so large that we fred that balloon catheter was embolized in unexpected areas. Therefore the operation was aborted. Considering that the patient’s condition was steady, the following procedure was aborted.

The patient remained stable for 7 days after the digital subtraction angiography (DSA), at which point he developed another episode of massive hematemesis which reported 1500 ml of blood loss with hypovolemic shock and a blood pressure of 80/40 mmHg. Point of care Endoscopic variceal ligation (EVL) was taken emergently (3A). After the EVL, the gastrointestinal bleeding stopped. When the patient’s general condition was steady the covered-stent graft in treatment of SMAVF. After the procedure, the patient indicated symptomatic improvement and 7 days later, he was discharged in good condition. He was also put on anticoagulation therapy with
warfarin, and 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’s time. Upper gastrointestinal endoscopy showed slight esophageal descendent varices, which was lightened more than onset of his symptoms. CT scan was without any sign of SMAVF (3B 4). Since then, patient had been doing well on follow-up visits after every 6 months.

DISCUSSION
SMAVF, which is an abnormal communication between the SMA and SMV, usually occurs as a result of trauma or abdominal surgery. Since the SMAVF was reported by Movitz (3), a few reports have been publicized in the world. SMAVF can be either congenital or acquired. Intestinal surgery and abdominal trauma are the most common reasons for acquired SMAVF (1). Iatrogenic SMAVF typically result from mass ligation during bowel resection or ligature placement in the mesentery without the precise localization of the bleeding point (4). Some cases of SMAVF remain asymptomatic for long periods and then develop a sudden onset of symptoms. There was a report that the time interval between surgery and clinical onset of SMAVF can be as long as 25 years (4). Our patient had a history of colonic laceration or injury reparation received 2 years ago.

Patients with SMAVF may present with abdominal pain, diarrhea caused by bowel ischemia from this shunting (1). The high-flow shunt led to portal hypertension (5), resulting in esophageal varices (EV), moderate ascites and so on (6). EV is hardly diagnosed at an early stage of the disease as many patients to be asymptomatic. However, to bleeding, which is one of the leading causes of death in patients with SMAVF. To date, 7 such cases have been reported. (Table ). Two patients suffered from abdominal pain earlier than gastrointestinal bleeding (2,7). Therefore nonspecific presentation, even asymptomatic in initial stage, is apt to misdiagnose. In our case, the patient’s examination of HBV infection misled us to a diagnosis of cirrhosis at the beginning. The most common physical finding is the presence of an
abdominal bruit or thrill. However, this can be easily missed.

There are several methods to visualize an SMAVF. Abdominal ultrasonic scanning, CT or MRI usually revealed vascular deformity firstly (2). DSA is the gold standard to define the exact anatomic location and extent of mesenteric vessel involvement, however, this procedure is typically used only for treatment. So CTA and CTV are widely used in clinical.

Surgical treatment is the most common approach and generally consists of ligation and division of the fistula, with or without removal of the affected organ (1). However, most of those patients had the history of previous abdominal surgery. The surgical repair would be extremely difficult and dangerous because of expected adhesions after the former laparotomy. Recently, percutaneous endovascular treatment of such AVFs has been increasingly performed, using covered stents or embolization (8-11). The angiographic technique used in our patient used covered stents. However, if the patient is in shock for esophageal variceal bleeding (EVB), we think that hemostasis through endoscopy is the most effective. EVL as the preferred treatment was used in our patient. It created an opportunity for further treatment.

In conclusion, the present case illustrates 3 important points: Because of the patients’ SMAVF clinical symptoms being often nonspecific and physical examination may not reveal the presence of abdominal bruit. SMAVF needs to be considered in portal hypertension and has had previous abdominal surgery) n patients who may not tolerate operative intervention, endovascular embolization can be safely performed with excellent results) the patient present with acute gastrointestinal bleeding, especially EVB, it is to be contained with endoscopic treatment.
Table  Patients with superior mesenteric arteriovenous fistula combined with gastrointestinal bleeding

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Fig 1 A and B CTA demonstrated the early filling and the huge diameter of the SMV (black arrow) reflected the SMA-SMV fistula. C Three-dimensional CT reconstruction demonstrated an SMA-SMV fistula (white arrow) CTA computed tomography angiography; SMA superior mesenteric artery SMV superior mesenteric vein.

2 Selective arterial angiography of the visceral arteries depicted a high flow fistula between the SMA (white arrow) and the SMV (black arrow) with massive engorgement of the portal system.
Fig 3 A EVL: Gastroscopy demonstrated severe esophageal varices. Red vein thrombosis (black arrow) was seen on the surface of gastric varicose vein. B The elastic band was successfully released by multiple band ligator and the bleeding was subsequently stopped. C Gastroscopy (1 month later) showed the absence of severe gastric varicose vein EVL Endoscopic variceal ligation

Fig 4 Four days after treatment A CTA depicted the stent inside the SMA (black arrow), the fistula disappeared. B Three-dimensional CT reconstruction demonstrated the above stent (white arrow) CTA computed tomography angiography;
SMA superior mesenteric arter.