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Splenic Arteriovenous Fistula Complicated by Severe Diarrhea: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Male, 36-year-old
Final Diagnosis: Splenic arteriovenous fistula
Symptoms: Chronic diarrhea • hematemesis
Medication: —
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology

Objective: Unusual clinical course

Background: Splenic arteriovenous fistula is a relatively rare disease. Patients are often admitted to the hospital with gastrointestinal symptoms. It is easy to misdiagnose due to the difficulty of confirming diagnosis only by routine examination.

Case Report: Our patient was critically ill, with an initial diagnosis of severe diarrhea with retroperitoneal hematoma before being referred to our hospital. Upon admission, the diagnosis of splenic arteriovenous fistula was made by computed tomography angiography. This patient with SAVF was successfully cured by distal splenic artery endovascular embolization therapy.

Conclusions: Clinicians should consider SAVF in the differential diagnosis of patients with severe diarrhea with uncommon causes. Endovascular embolization therapy needs to be considered *vs.* conventional surgical ligation by open surgery in terms of operation risks and outcomes, along with subsequent exploratory laparotomy.

MeSH Keywords: Arteriovenous Fistula • Diarrhea • Endovascular Procedures • Hypertension, Portal • Splenectomy

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Background

Splenic arteriovenous fistula is a rare disease in clinical practice. Patients are often admitted to the hospital with gastrointestinal symptoms. It is easy to misdiagnose due to the difficulty of confirming diagnosis only by routine examination and due to the lack of pertinent data. About 100 cases have been reported in the world medical literature, the first of which was reported based on an autopsy [1,2]. Currently, the diagnosis can be confirmed by the less invasive procedure of digital subtraction angiography (DSA) [3]. An increasing number of reports show that endovascular treatment can achieve good results [4]. This article reports a case of splenic arteriovenous fistula successfully cured by endovascular therapy, with the aim of summarizing the etiology, main symptoms, diagnosis, and treatment options.

Case Report

A 36-year-old man was admitted to the hospital due to "diarrhea for 2 months, aggravated for 1 day." In March 2019, the patient developed diarrhea about 10 times/day, 5 days after being kicked in the middle abdomen. Then, the diarrhea aggravated after symptomatic control in a local clinic. After being referred to another local hospital, colonoscopy showed "colon edema, colitis". However, the diarrhea continued to aggravate after treatment of colitis. A month later, symptoms of abdominal and low back pain, accompanied by nausea and vomiting of gastric fluid, developed, after which he was referred to the affiliated hospital of a local medical university. CT then showed "thoracic, abdominal and pelvic cavity effusion" and about 800 ml hemorrhagic ascites was drained. Considering the critical situation, laparotomy was performed the next day, showing "extensive retroperitoneal hematoma, active bleeding in the abdominal aorta". The symptoms of bloating were significantly relieved after vascular repair, but there was still the symptom of diarrhea 10 times/day with bloody peritoneal drainage. Several days later, hematemesis developed, with gastroscopy showing "esophageal varices". The patient was referred to our hospital for further diagnosis and treatment. We found a soft abdomen with no tenderness, but with systolic jet-like murmur heard on the left middle abdomen with active bowel sounds. He had a history of chronic systematic hypertension and splenectomy 10 years ago due to trauma.

The results of laboratory tests showed white blood cell (WBC) count 12.98×10^9/L (4–10×10^9/L), platelet count 485×10^9/L (86–303×10^9/L), C-reactive protein (CRP) 5.27 mg/dl (0–3 mg/L), total bilirubin 47.1 μmol/L (3.4–17. 1 μmol/L), direct bilirubin 27.6 (normal less than 5.1 μmol/L), and procalcitonin 8.92 ng/ml (normal less than 2 ng/ml). Other test results were within normal range.

The spleen arteriovenous fistula was suggested by abdomen contrast-enhanced abdominal computed tomography with 3D image reconstruction (Figures 1, 2). Later, celiac angiography confirmed this diagnosis (Figure 3).

Treatment

The distal splenic artery was embolized during celiac angiography (Figure 4). An abdominal CT examination 3 days after the operation showed that the intestinal edema was significantly decreased (Figures 5, 6) and ascites was significantly reduced. The frequency of diarrhea on the 5th day after the operation was about 4 times/day. Drainage fluid was not seen in the abdominal drainage bag. At follow-up 2 months later, the general condition of this patient was good and the diarrhea and hematemesis had not recurred.

Discussion

The splenic arteriovenous fistula (SAVF) was first described by German surgeon and pathologist Carl Wegger in 1886 [5] and it consists of congenital and acquired forms. In a wide range of reports, congenital type accounts for 20%, while acquired type accounts for 80%. In the latter form, most cases are related to trauma, splenectomy, and splenic aneurysm rupture [6], and fewer are associated with fungal emboli, malignant lymphoma, and pancreatectomy [7–9]. Among them, the incidence of splenectomy is more insidious; onset occurs about 5–20 years after splenectomy, with the longest being 23 years [10].

Clinical presentation depends on the location and size of the fistula. A patient with a small orifice can be without clinical symptoms [10], while patients with large orifices often have portal hypertension, esophageal varices, hematemesis, and diarrhea. Repeated treatment failure can lead to cirrhosis, hepatic encephalopathy, and portal vein thrombosis. As color Doppler ultrasound and abdominal CT scan often fail to detect splenic arteriovenous fistula, abdominal enhanced CT can only assist in diagnosis, and digital subtraction angiography (DSA) must be performed for diagnosis confirmation [3,11], which increases the difficulty of treatment.

Our literature search found that the misdiagnosis rate is high, and most patients have symptoms such as gastrointestinal bleeding, abdominal pain, and diarrhea. Relevant accessory examinations often misleadingly suggest gastrointestinal diseases. Treatment of GI diseases worsens the condition, and long-term, ineffective, repeated treatment can impair patient quality of life.

Traditional treatment of splenic arteriovenous fistula is mainly performed by surgery, including splenectomy and fistula

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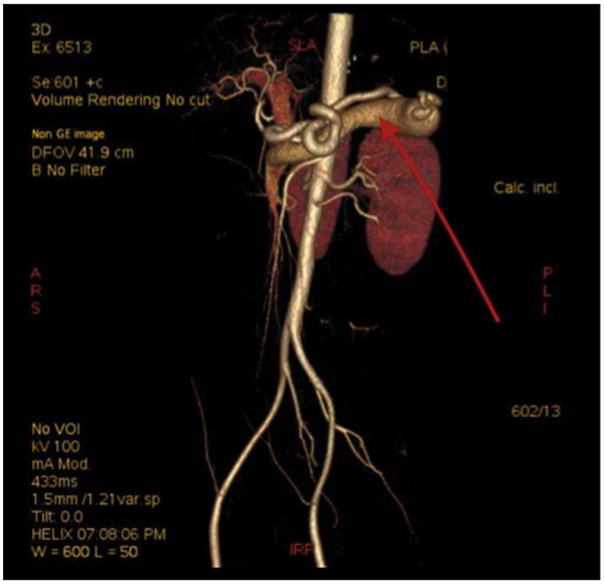

Figure 1. Contrast-enhanced abdominal computed tomography reconstruction shows dilated splenic vein, tortuous splenic vein, and splenic arteriovenous fistula.

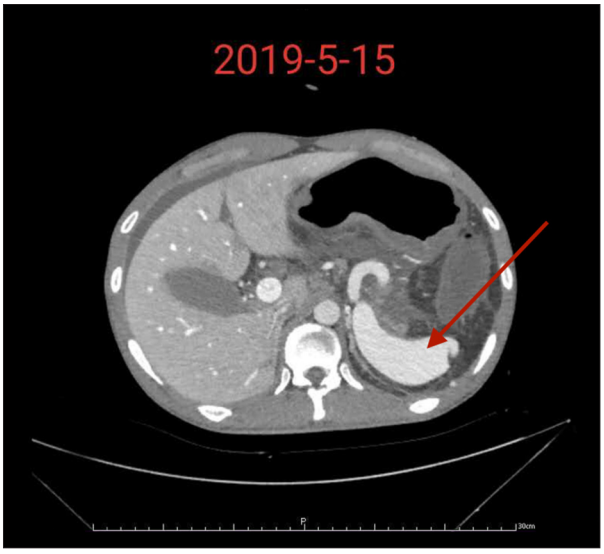

Figure 2. Tortuous splenic vein indicated by arrow in contrast-enhanced CT.

ligation or resection. However, some patients have a special position of the fistula and poor general condition, thus limiting the surgical options and making the operation difficult and high risk [12,13]. In contrast, endovascular treatment has advantages of minor degree of trauma, rapid postoperative recovery, good prognosis, and ability to significantly improve quality of life, making it the preferred method for treatment of splenic arteriovenous fistula [5,10,11]. According to the literature, the mortality rate of conventional surgical methods is as high as 7.1%, but no deaths have been reported with endovascular treatment [14].

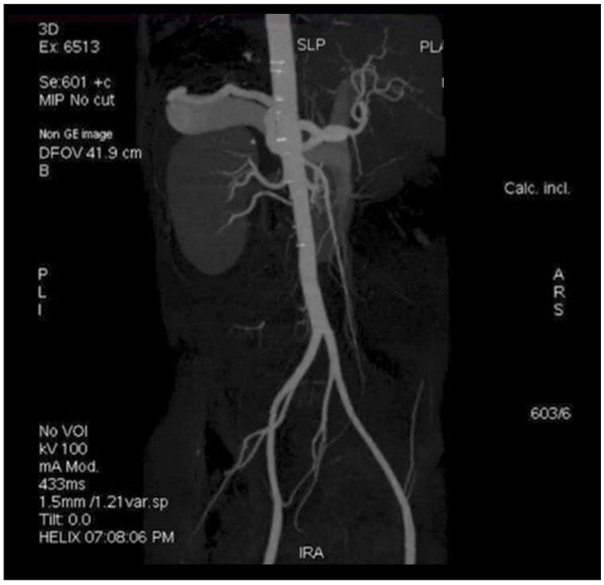

Figure 3. Splenic arteriovenous fistula shown by celiac angiography.

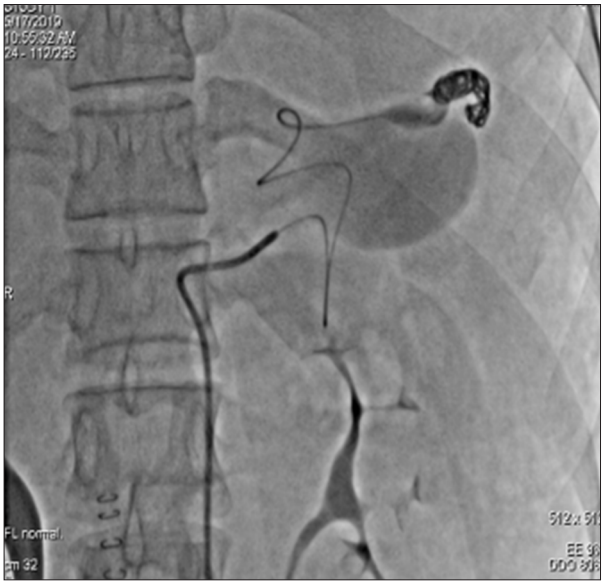

Figure 4. Endovascular treatment using a 7-F balloon to block the splenic artery, and 3-F microcatheter carefully inserted into the distal end of the splenic artery for embolization.

Pathophysiology related to diarrhea and other symptoms in this patient

Splenic arteriovenous fistula and splenic artery aneurysm formed after splenectomy resulted in portal hypertension and portal system compromise, which then led to intestinal wall edema and ischemia due to an engorged colic vein. Later, the blunt trauma to his abdomen caused the aneurysm rupture,

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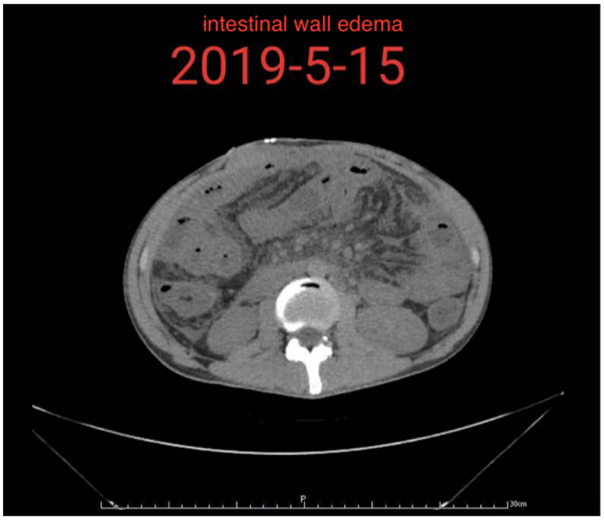

Figure 5. Intestinal wall edema caused by portal hypertension.

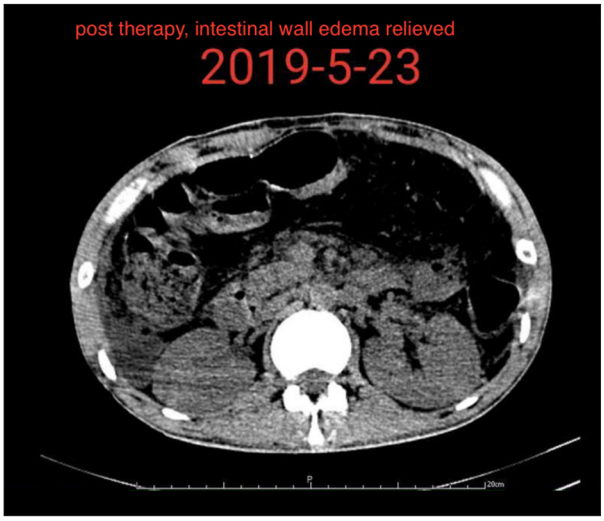

Figure 6. Intestinal wall edema relieved after splenic artery endovascular embolization therapy.

leading to retroperitoneal hematoma, further irritating and compromising the already dysfunctional intestine. Consequently, severe diarrhea and bloody ascites developed.

Diagnostic evaluation of the diarrhea in this patient

Before further investigation, the patient had the clear eliciting factor of abdominal trauma. In addition, he had a history of splenectomy. The situation in which the chronic diarrhea did not improve after initial symptomatic control, and emerging manifestations such as abdominal and low back pain, indicated the culprit was an underlying organic disorder.

An abdominal CT scan confirmed intestinal wall edema, suggesting the direct cause of his diarrhea. Subsequently, the bloody ascites and abdominal murmur further indicated there was a root cause indirectly leading to his severe diarrhea, which may be related to a late complication of the splenectomy that he underwent 10 years ago. Then, he was diagnosed as having splenic arteriovenous fistula by contrast-enhanced CT scan and celiac angiography. Subsequent treatment by splenic artery endovascular embolization was carried out, after which his intractable diarrhea, intestinal wall edema, and ascites were significantly improved. Hence, we finally determined that the ultimate cause was a splenic arteriovenous fistula.

Conclusions

Splenic arteriovenous fistula is a rare cause of portal hypertension. There are few relevant reports. Most clinicians are relatively unaware of it and are liable to make a misdiagnosis.

Therefore, in a patient without chronic liver disease who manifests symptoms of portal hypertension such as hematemesis and intractable diarrhea, it is important to consider splenic arteriovenous fistula as a differential diagnosis [12].

Institution where work was done

This work was done in the General Hospital of the People's Liberation Army, Beijing, P.R. China.

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Conflict of interest

None.

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