Int J Clin Exp Med 2018;11(8):8752-8756
www.ijcem.com /ISSN:1940-5901/IJCEM0066748

Case Report

Transjugular intrahepatic portosystemic shunt combined interventional embolization for splenic artery aneurysm with splenic arteriovenous fistula

Zhaobin Yan[1], Qinsheng Zhang[1], Fuliang He[2], Xian Li[1], Zhendong Yue[2], Lei Wang[2], Mingyan Li[1], Zhenhua Li[1], Shijie Zhang[3]

[1]Department of Gastroenterology, Henan Provincial Hospital of Traditional Chinese Medicine, The Second Affiliated Hospital of Henan University of Traditional Chinese Medicine, Zhengzhou 450000, Henan Province, China; [2]Department of Interventional Therapy, Beijing Shijitan Hospital, Capital Medical University, Beijing 100038, China; [3]Department of Oncology, The First Affiliated Hospital, Guangxi University of Traditional Chinese Medicine, Guangxi, China

Received October 7, 2017; Accepted April 9, 2018; Epub August 15, 2018; Published August 30, 2018

Abstract: Splenic arteriovenous fistula (SAVF) with splenic artery aneurysm (SAA) is an extremely rare cause of portal hypertension. Here, We report a case of 43-year-old man (no evidence of liver disease) with sudden melena, hematemesis and massive ascites because of portal hypertension, which caused by SAVF with SAA. The paitent cured with the treatment of transjugular intrahepatic portosystemic shunt (TIPS) combined interventional embolization, which demonstrates that TIPS combined interventional embolization may be a good way for SAVF with SAA.

Keywords: Splenic arteriovenous fistula, splenic artery aneurysm, portal hypertension, endovascular embolization, transjugular intrahepatic portosystemic shunt

Introduction

Splenic arteriovenous fistula (SAVF) with splenic artery aneurysm (SAA) is an extremely rare cause of portal hypertension [1]. and can be cured by open surgery and embolization [2-4]. However, there have been no reports on the occurrence of relapse after endovascular embolization. Here, we report a case of how to cure SAVF with SAA which recurrent after endovascular embolization.

Case report

A 43-year-old man was transferred from a local hospital to our institution in December 2015 because of uncontrolled gastrointestinal bleeding managed conservatively with, endoscopic hemostasis and blood transfusion. Due to chronic renal disease, surgery was not an option of treatment. His past medical history included a renal transplant 13 years earlier, recurrent diarrhea one month ago, and no history of cirrhosis.

On admission, his temperature was 36.4°C, blood pressure was 119/85 mmHg, pulse rate was 84 beats per minute, and respiration rate was 20 breath/min. His symptoms included melena, diarrhea, vomiting, and slight abdominal pain. There were no other physical signs except positive shifting dullness. The results of laboratory tests were as follows: red blood cell count 3.34 × 10^9/L, hemoglobin 94 g/L, platelet 288 × 10^9/L, white blood cell count 5.45 × 10^9/L, blood urea nitrogen 27.65 mmol/L, and creatinine 934 μmol/L.

A contrast-enhanced computed tomography (CT) scan of the abdomen revealed that alarge splenic artery aneurysmal expansion between the splenic artery and the dilated splenic vein, and both the splenic artery and the widened portal vein were filled with contrast medium in the arterial phase. The patient also had moderate ascites (**Figure 1**).

Under local anesthesia and using the Seldinger technique, a catheter was successfully inserted

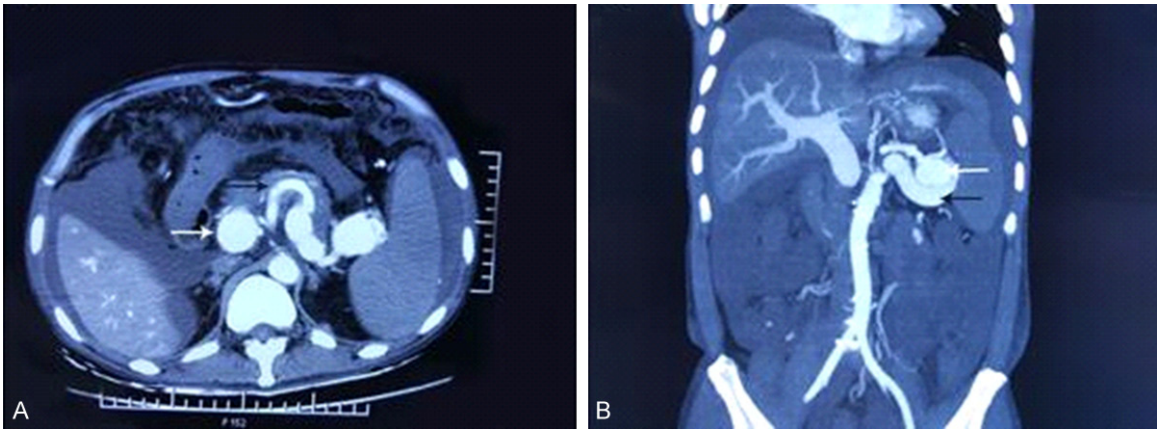

Figure 1. Contrast-enhanced abdominal CT. A: In the arterial phase, splenic artery (black arrowhead) and the widened portal vein (white arrowhead) are filling contrast-medium. B: A coronal computed tomography angiography (CTA) demonstrating the splenic artery aneurysmbetween the splenic artery (black arrowhead) and the splenic vein (white arrowhead).

into the right femoral artery. Selective celiac and splenic artery angiography showed a large splenic artery aneurysmal expansion, a dilated splenic vein and an enlarged portal vein in the early arterial phase. Combined with the patient's history,the patient was diagnosed with SAVF (**Figure 2A**).

We successfully deployed six coils (MicroPlex-18 20 mm/50 cm) at the sites of the SAA using a micro-catheter. Two coils (MWCE-35-14-8-NESTER, Cook Medical), one coils (MWCE-35-14-10-NESTER, Cook Medical), and three coils (MWCE-35-14-12-NESTER, Cook Medical) were successively used for embolization at the distal splenic artery through the catheter. Splenic arteriography was performed which showed that there is still a contrast agent through the splenic vein in the late arterial phase. We then used gel foam for embolization, and splenic arteriography showed that the contrast medium from the SAA had flowed into the splenic vein in the late stage (**Figure 2B**). Following endovascular treatment, the patient recovered well and was discharged without clinical symptom.

Approximately two months later, the patient returned to our hospital with melena and massive ascites. Splenic artery angiography showed that, the splenic artery was occluded, and the contrast medium flowed through the left gastric artery and left gastro-omental artery into the splenic artery and SAA to the portal vein (**Figure 2C**). However, there was no space for further embolization. Therefore, we decided to perform TIPS with variceal embolization. Five coils (MWCE-18-14-6-NESTER, Cook Medical), and two coils (MicroP18 10 mm/30 cm com) were successively used for embolization of the varicose veins through a micro-catheter. Two stents (Fluency Plus 10 × 80 mm) were implanted in the shunt channel after balloon dilation (**Figure 2D**). During this treatment, the pressure in the portal vein dropped from 58 cm H_2O to 22 cm H_2O. The patient was free of abdominal symptoms at the 7 months follow-up, and the CT scan and color ultrasound showed unremarkable results. He will be followed up annually.

Discussion

SAA is an abnormally dilated splenic artery, measuring more than 1 cm in diameter [5]. The most frequent comorbidities among SAA patients have included atherosclerosis, cirrhotic portal hypertension, multiparity, pancreatitis, and fibromuscular dysplasia [6-8]. In the present case, the etiology of the SAA was unknown, because of no associated factor was identified from the patient's history or imageological examination.

Splenic arteriovenous fistula (SAVF) can be congenital or acquired. congenital SAVF are intrasplenic and hemangiomatous, and acquired SAVF are secondary to traumatic, rupture of splenic artery aneurysm into corresponding splenic vein, post-splenectomy, post-gastrectomy, mycotic infection or Pancreatitis [9-14]. In our case, after exclusion of other causes, the SAVF was considered secondary to erosion of the SAA.

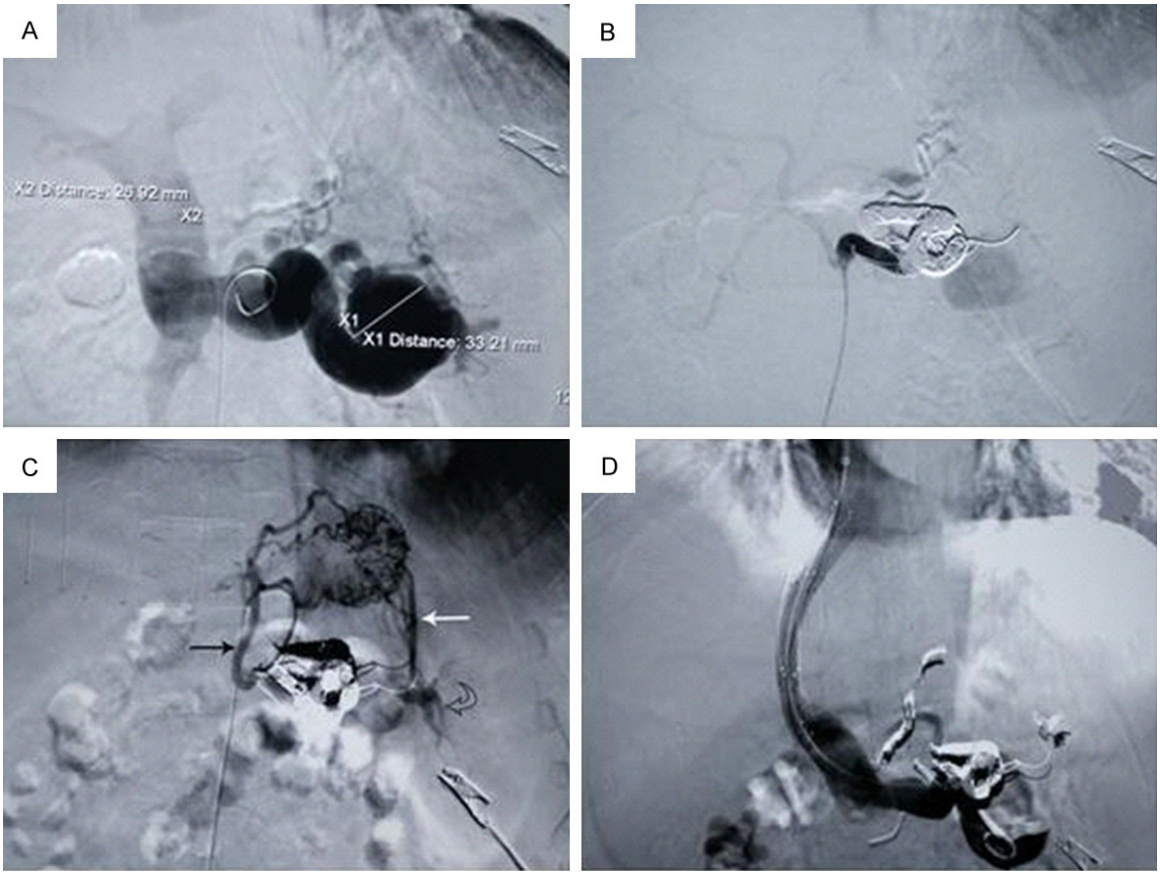

Figure 2. Digital subtraction angiography. A: The SAA has a diameter of 33 mm and the portal vein is approximately 27 mm; B: After coil embolization, the SAA is developing in the late stage; C: The contrast medium flowing through the left gastric artery (black arrowhead) and left gastro-omental artery (white arrowhead) into the splenic artery (curved arrow); D: After Transjugular intrahepatic portosystemic shunt, stent patency and the gastric varices disappeared.

SAVF with SAA is rare diseases in the clinic causing portal hypertension [15]. It is usually diagnosed in patients with upper abdominal pain, gastrointestinal bleeding, ascites, diarrhea or splenomegaly [16, 17]. However, it can be completely cured by traditional surgery and interventional treatment [4, 18]. Due to high surgical risk, interventional therapy has become the main treatment for SAVF [19, 20].

Here, we describe the first case of an SAVF treated by TIPS after interventional embolization in a patient with melena and massive ascites. A timely diagnosis is very important before the treatment. Doppler ultrasound, CT scanning, and splenic arteriography have been used to diagnose SAVF. In our patient, CT scanning demonstrated SAVF, and the diagnosis was confirmed by splenic artery arteriography.

In this case, we used sufficient coils to embolize the splenic artery aneurysmal expansion and splenic artery. However, after two months, the patient melena developed and massive ascites. Unfortunately, there are no literature reports on melena and massive ascites following endovascular embolization for SAVF with SAA. The main cause of portal hypertension in this patient may have been in complete embolization of SAVF with SAA or in complete embolization of the distal splenic artery. Therefore, the left gastric artery and left gastro-omental artery flowed through the splenic artery and SAVF with SAA into the portal vein. Thus melena and massive ascites reappeared and it was not possible to perform embolization again. Therefore, we decided to carry out TIPS with gastric variceal embolization as rescue therapy. At the 10-month follow-up visit, the patient showed no symptoms and stent patency.

In order to complete embolization, the following steps is necessary. Frist, the patient must be undergo embolization of the proximal splenic

artery (between the splenic artery aneurysm and the spleen); and then embolization of the SAA and main trunk of the splenic artery to ensure that there is no blood flow. If these steps are followed, we believe that satisfactory outcome can be achieved.

In conclusion, we report the first case of SAVF with SAA treated with endovascular embolization which developed after treatment with TIPS.

Disclosure of conflict of interest

None.

Address correspondence to: Shijie Zhang, Department of Oncology, The First Affiliated Hospital, Guangxi University of Traditional Chinese Medicine, No. 89-9 Dongge Road, Nanning, Guangxi, China. Tel: +86-14795592324; E-mail: 328075698@qq.com

References

[1] Rajiv M, Deepak S, Rajesh G and Sadasivan S. Splenic artery aneurysm with arteriovenous fistula as a cause of portal hypertension. Indian J Gastroenterol 2004; 23: 36-37.

[2] Yadav R, Tiwari MK, Mathur RM and Verma AK. Unusually giant splenic artery and vein aneurysm with arteriovenous fistula with hypersplenism in a nulliparous woman. Interact Cardiovasc Thorac Surg 2009; 8: 384-386.

[3] Azar CR, Al-Kutoubi AO and Mourad FH. A short case of a splenic arteriovenous fistula coexisting with portal hypertension secondary to hepatitis C: radiologic diagnosis and treatment. J Med Imaging Radiat Oncol 2010; 54: 134-136.

[4] Jiang R, Ding X, Jian W, Jiang J, Hu S and Zhang Z. Combined endovascular embolization and open surgery for splenic artery aneurysm with arteriovenous fistula. Ann Vasc Surg 2016; 30: 311.

[5] Uy P, Francisco DM, Trivedi A, O'Loughlin M and Wu GY. Vascular diseases of the spleen: a review. J Clin Transl Hepatol 2017; 5: 152-164.

[6] Abbas MA, Stone WM, Fowl RJ, Gloviczki P, Oldenburg WA, Pairolero PC, Hallett JW, Bower TC, Panneton JM and Cherry KJ. Splenic artery aneurysms: two decades experience at Mayo clinic. Ann Vasc Surg 2002; 16: 442-449.

[7] Siablis D, Papathanassiou ZG, Karnabatidis D, Christeas N, Katsanos K and Vagianos C. Splenic arteriovenous fistula and sudden onset of portal hypertension as complications of a ruptured splenic artery aneurysm: successful treatment with transcatheter arterial embolization. A case study and review of the literature. World J Gastroenterol 2006; 12: 4264-4266.

[8] Unlu C, van den Heuvel DA, Leeuwis JW and de Vries JP. Ruptured aneurysm of the splenic artery associated with fibromuscular dysplasia. Ann Vasc Surg 2014; 28: 1715-1799.

[9] Schmidt JH, Howard RJ, Herrera MA and Hawkins IF. Splenic arteriovenous fistula with portal hypertension, ascites, and diarrhea. South Med J 1988; 81: 670-672.

[10] Siablis D, Papathanassiou ZG, Karnabatidis D, Christeas N, Katsanos K and Vagianos C. Splenic arteriovenous fistula and sudden onset of portal hypertension as complications of a ruptured splenic artery aneurysm: successful treatment with transcatheter arterial embolization. A case study and review of the literature. World J Gastroenterol 2006; 12: 4264-4266.

[11] Yadav R, Tiwari MK, Mathur RM and Verma AK. Unusually giant splenic artery and vein aneurysm with arteriovenous fistula with hypersplenism in a nulliparous woman. Interact Cardiovasc Thorac Surg 2009; 8: 384-386.

[12] Saddekni S, Anis KH, Hegazi AA, Hamed MF and Abdel AA. Traumatic complex splenic arteriovenous fistula causing prehepatic portal hypertension and variceal bleeding: the importance of the diagnosis for the endovascular treatment approach. Vasc Endovascular Surg 2014; 48: 180-185.

[13] Azar CR, Al-Kutoubi AO and Mourad FH. A short case of a splenic arteriovenous fistula coexisting with portal hypertension secondary to hepatitis C: radiologic diagnosis and treatment. J Med Imaging Radiat Oncol 2010; 54: 134-136.

[14] Buchholz RR. Arteriovenous fistula of the splenic vessels; report of a case following splenectomy. Ann Surg 1959; 149: 590-592.

[15] Chen B, Tang CW, Zhang CL, Cao JW, Wei B and Li X. Melena-associated regional portal hypertension caused by splenic arteriovenous fistula. World J Gastroenterol 2012; 18: 1996-1998.

[16] McClary RD, Finelli DS, Croker B and Davis GL. Portal hypertension secondary to a spontaneous splenic arteriovenous fistula: case report and review of the literature. Am J Gastroenterol 1986; 81: 572-575.

[17] Yadav R, Tiwari MK, Mathur RM and Verma AK. Unusually giant splenic artery and vein aneurysm with arteriovenous fistula with hypersplenism in a nulliparous woman. Interact Cardiovasc Thorac Surg 2009; 8: 384-386.

[18] Saddekni S, Anis KH, Hegazi AA, Hamed MF and Abdel AA. Traumatic complex splenic arteriovenous fistula causing prehepatic portal hy-

pertension and variceal bleeding: the importance of the diagnosis for the endovascular treatment approach. Vasc Endovascular Surg 2014; 48: 180-185.

[19] Law ST, Wong CK, Chow KC and Loo KT. Splenic arteriovenous fistula: unusual cause of portal hypertension complicated with gastric variceal bleeding. J Dig Dis 2012; 13: 549-552.

[20] Madsen MA, Frevert S, Madsen PL and Eiberg JP. Splenic arteriovenous fistula treated with percutaneous transarterial embolization. Eur J Vasc Endovasc Surg 2008; 36: 562-564.