Melena-associated regional portal hypertension caused by splenic arteriovenous fistula

Bin Chen, Cheng-Wei Tang, Chun-Le Zhang, Jia-Wei Cao, Bo Wei, Xiao Li

Bin Chen, Cheng-Wei Tang, Chun-Le Zhang, Jia-Wei Cao, Bo Wei, Xiao Li, Department of Gastroenterology, West China Hospital, Sichuan University, Chengdu 610041, Sichuan Province, China

Author contributions: Chen B, Tang CW, Zhang CL, Wei B and Li X managed the patient; Chen B, Zhang CL, Cao JW and Li X wrote the report.

Supported by The National Natural Science Foundation of China, No. 81171444

Correspondence to: Xiao Li, Professor, Department of Gastroenterology, West China Hospital, Sichuan University, No. 37 Guoxue Lane, Chengdu 610041, Sichuan Province, China. simonlixiao@gmail.com

Telephone: +86-28-85422385 Fax: +86-28-88092354

Received: October 9, 2011 Revised: January 4, 2012

Accepted: February 26, 2012

Published online: April 28, 2012

Abstract

Regional portal hypertension is a rare cause of upper gastrointestinal bleeding. We reported an extremely rare case in which regional portal hypertension was associated with both the splenic arteriovenous fistula and chronic pancreatitis. In June 2010, our patient, a 41-year-old man, was admitted to a local hospital due to a sudden melena and dizziness without haematemesis and jaundice. He was managed conservatively, with fluid support and blood transfusion. No melena occurred again, and the symptom of dizziness was reduced. With the melena of unknown causes, he was referred to our institution for further diagnosis and treatment. On admission, his temperature was 36.7 °C, pulse was 85 times/min, respiration was 19 times/min, and blood pressure was 124/76 mmHg. Splenomegaly was found in physical examination. The liver was not palpable and no signs of jaundice were observed. The patient had a history of alcohol abuse, but no history of liver disease, trauma and surgery. Five years ago, the patient had an acute pancreatitis which occurred for five times. The details of laboratory tests were as follows: red blood cell count was 2.5 × 10^12/L, haematocrit was 9.7%, white blood cell count was 11.5 × 10^9/L, and platelet count was 100 × 10^9/L. The details of liver function tests were as follows: total bilirubin was 0.9 mg/dL, direct bilirubin was 0.2 mg/dL, alanine aminotransferase was 22 U/L, aspartate aminotransferase was 34 U/L, alkaline phosphatase was 83 U/L, γ-glutamyl transferase was 17 U/L, total protein was 9.2 g/L, albumin was 5.2 g/L, and serum creatinine was 1.1 mg/dL. The details of coagulation function tests were as follows: prothrombin time was 12.0 s, and activated partial thromboplastin time was 38.4 s. The results of blood gas analysis were as follows: pH was 7.39, pCO2 was 34.2 mmHg, and pO2 was 94.6 mmHg. The results of faecal occult blood test were negative. The results of esophagogastroduodenoscopy were normal. The results of upper gastrointestinal series were normal. The results of upper gastrointestinal endoscopy were normal. The results of upper gastrointestinal endoscopy with biopsy were normal. The results of abdominal ultrasound were normal. The results of abdominal CT scan were normal. The results of upper gastrointestinal angiography showed a splenic arteriovenous fistula and a portal venous system. The splenic arteriovenous fistula was successfully occluded through transcatheter arterial embolization. At the 12-mo follow-up, our patient was in good condition.

© 2012 Baishideng. All rights reserved.

Key words: Regional portal hypertension; Splenic arteriovenous fistula; Pancreatitis; Transcatheter arterial embolization; Upper gastrointestinal bleeding

Peer reviewer: Dr. Andrea De Gottardi, Hepatology-Inselspital, Freiburg Strasse, Berne 3010, Switzerland

INTRODUCTION

Regional portal hypertension is a rare cause of upper gastrointestinal bleeding, with pancreatitis disease being the most frequently reported cause in the literature [1,2]. Splenic arteriovenous fistulas are also rare. Until now, there are approximately 126 reported cases of splenic arteriovenous fistula in the database of PubMed. Herein, we reported an extremely rare case in which regional portal hypertension was associated with both the splenic arteriovenous fistula and chronic pancreatitis.

CASE REPORT

A 41-year-old man was admitted to a local hospital in June 2010 due to a sudden melena and dizziness without haematemesis and jaundice. He was managed conservatively, with fluid support and blood transfusion. No melena occurred again, and the symptom of dizziness was reduced. With the melena of unknown causes, he was referred to our institution for further diagnosis and treatment. On admission, his temperature was 36.7 °C, pulse was 85 times/min, respiration was 19 times/min, and blood pressure was 124/76 mmHg. Splenomegaly was found in physical examination. The liver was not palpable and no signs of jaundice were observed. The patient had a history of alcohol abuse, but no history of liver disease, trauma and surgery. Five years ago, the patient had an acute pancreatitis which occurred for five times. The details of laboratory tests were as follows: red blood cell count was 2.5 × 10^12/L, haematocrit was 9.7%, white blood cell count was 11.5 × 10^9/L, and platelet count was 100 × 10^9/L. The details of liver function tests were as follows: total bilirubin was 0.9 mg/dL, direct bilirubin was 0.2 mg/dL, alanine aminotransferase was 22 U/L, aspartate aminotransferase was 34 U/L, alkaline phosphatase was 83 U/L, γ-glutamyl transferase was 17 U/L, total protein was 9.2 g/L, albumin was 5.2 g/L, and serum creatinine was 1.1 mg/dL. The details of coagulation function tests were as follows: prothrombin time was 12.0 s, and activated partial thromboplastin time was 38.4 s. The results of blood gas analysis were as follows: pH was 7.39, pCO2 was 34.2 mmHg, and pO2 was 94.6 mmHg. The results of faecal occult blood test were negative. The results of esophagogastroduodenoscopy were normal. The results of upper gastrointestinal series were normal. The results of upper gastrointestinal endoscopy were normal. The results of upper gastrointestinal endoscopy with biopsy were normal. The results of abdominal ultrasound were normal. The results of abdominal CT scan were normal. The results of upper gastrointestinal angiography showed a splenic arteriovenous fistula and a portal venous system. The splenic arteriovenous fistula was successfully occluded through transcatheter arterial embolization. At the 12-mo follow-up, our patient was in good condition.
cell 4.13 × 1012/L, hemoglobin 119 g/L, platelet 246 × 109/L, white blood cell 6.90 × 109/L, total bilirubin 13.3 μmol/L, albumin 42.9 g/L, creatinine (CREA) 572.9 μ mol /L, glucose (GLU) 0 9.53 mmol/L, GLU120 21.75 mmol/L, prothrombin time 12.0 s, and alpha-fetoprotein 1.36 ng/mL. Tests for HBsAg, HBeAg, anti-HBe, anti-HBc, anti-HCV and anti-HBs were all negative, except for anti-HBs. Gastroscopy revealed severe gastric varices and non-atrophic gastritis with bile reflux. Unenhanced computed tomography (CT) scanning of upper abdomen showed an oval cystic low-density lesion located in the area of splenic hilum, which was easily misdiagnosed as pancreatic pseudocyst on unenhanced CT (Figure 1A) and as pseudoaneurysm on contrast-enhanced scan. The lesion was significantly enhanced on the arterial phase of contrast-enhanced scan (Figure 1B). And on the portal phase of contrast-enhanced CT scan, this lesion was further enhanced and collateral vessels could be shown at the fundus of the stomach (Figure 1C). Additionally, thrombosis in the splenic vein was also visible on the portal phase of contrast-enhanced CT scan. Biopsy of the liver was not performed. The current diagnosis was regional portal hypertension considered to be related to chronic pancreatitis. Under local anesthesia, a percutaneous transjugular approach was used to estimate the portal venous pressure; the portal pressure was normal and an increased pressure of splenic vein was 23.5 mmHg (Figure 1D). Through a percutaneous femoral approach, splenic artery angiography was performed, showing a smaller splenic artery, splenic vein aneurysmal expansion, and esophageal and gastric varices, suggesting the formation of splenic arteriovenous fistula (Figure 1E).

With splenic artery being extremely tortuous, it was difficult for conventional catheter to track the orifice of the fistula through target vessels, therefore embolization of the fistula was performed using a micro-catheter. Postoperative angiography revealed that splenic arteriovenous fistula had been totally occluded (Figure 1F).

**DISCUSSION**

Regional portal hypertension, also known as sinistral or left-sided portal hypertension, is a rare cause of upper gastrointestinal bleeding, with pancreatitis disease being the most frequently reported cause in the literature[1,2]. Splenic arteriovenous fistula is a rare but potentially curable cause of portal hypertension. The fistula may be congenital or acquired. It is thought that they arise from rupture of a splenic artery aneurysm or after abdominal trauma, surgery or pancreatitis[3]. To our best knowledge, splenic arteriovenous fistulas related to pancreatitis have been rarely reported and its pathogenesis remains unclear[4]. In this case, the fistula arose from pancreatitis, and regional portal hypertension was caused by splenic arteriovenous fistula and splenic vein thrombosis, with the former playing a major role. As for regional portal hypertension, the key point is to make the correct diagnosis as soon as possible, because it is curable. It should be considered in the presence of gastrointestinal bleeding with normal liver function tests and splenomegaly. Surgical excision of splenic arteriovenous fistula is technically difficult, and is often unsuccessful because of the remote location of the lesion, presence of numerous portal collaterals, and adhesion. Interventional radiologic technique, such as transcatheter arterial embolization, has

**Figure 1** The main findings of enhanced computed tomography and angiography. A: An oval cystic low-density lesion located in the area of splenic hilum; B: The lesion was enhanced significantly on the arterial phase; C: Significantly enhanced on the portal phase of contrast-enhanced scan; D: A normal portal pressure and an increased pressure of splenic vein of 23.5 mmHg; E: Splenic arteriovenous fistula manifested by angiography; F: Splenic arteriovenous fistula being totally occluded after embolization.
been demonstrated to be a safe and effective alternative to surgery. The splenic arteriovenous fistula in our patient was successfully occluded through transcatheter arterial embolization. At the 12-mo follow-up, this patient was found in good condition.

REFERENCES

1 Köklü S, Coban S, Yüksel O, Arhan M. Left-sided portal hypertension. *Dig Dis Sci* 2007; 52: 1141-1149
2 Sakorafas GH, Sarr MG, Farley DR, Farnell MB. The significance of sinistral portal hypertension complicating chronic pancreatitis. *Am J Surg* 2000; 179: 129-133
3 Vanhoenacker FM, Op de Beeck B, De Schepper AM, Salgado R, Snoeckx A, Parizel PM. Vascular disease of the spleen. *Semin Ultrasound CT MR* 2007; 28: 35-51
4 Raat H, Stockx L, De Meester X, Van Steenbergen W, Marchal G. Percutaneous embolization of a splenic arteriovenous fistula related to acute necrotizing pancreatitis. *Eur Radiol* 1999; 9: 753
5 Hung CF, Tseng JH, Lui KW, Wan YL, Tsai CC, Shem CH, Wu CS. Intractable oesophageal variceal bleeding caused by splenic arteriovenous fistula: treatment by transcatheter arterial embolization. *Postgrad Med J* 1999; 75: 355-357

S- Editor Gou SX  L- Editor Ma JY  E- Editor Zhang DN