Intractable oesophageal variceal bleeding caused by splenic arteriovenous fistula: treatment by transcatheter arterial embolization

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Summary

We describe a rare case of splenic arteriovenous fistula and venous aneurysm which developed after splenectomy in a 40-year-old woman who presented with epigastralgia, watery diarrhoea, repeated haematemesis and melaena caused by hyperkinetic status of the portal system and bleeding of oesophageal varices. It was diagnosed by computed tomography and angiography, and obliterated with giant Gianturco steel coils.

Keywords: splenic arteriovenous fistula; gastrointestinal bleeding; transcatheter arterial embolization

Splenic arteriovenous fistula (SAVF) and splenic venous aneurysm after splenectomy are extremely rare. To our knowledge, only five cases have been reported in the English literature.¹⁻⁵ SAVF is unusual but curable. It may cause portal hypertension. The hyperkinetic state of flow through SAVF may cause gastrointestinal bleeding, diarrhoea, ascites, or life-threatening heart failure. Elimination of SAVF by transcatheter arterial embolization has been advocated as an alternative to surgical resection. We present a patient with SAVF who presented unusually with epigastralgia associated with variceal bleeding.

Case report

A 40-year-old woman complained of intermittent epigastric pain associated with watery diarrhoea, haematemesis and melaena. She had undergone splenectomy 17 years earlier due to splenic rupture after a motor vehicle accident. Endoscopy performed at a local hospital revealed variceal bleeding in the lower third of the oesophagus. She was transferred to our hospital. Endoscopic variceal ligation was performed several times within a month.

A grade II systolic murmur was heard over the left lower chest. The abdomen was soft and tender. There was no ascites. Liver was not palpable. Laboratory examination showed haemoglobin 10.5 g/dl, haematocrit 29.1%, leucocytes $11.6 \times 10^9/l$ and platelets $76 \times 10^9/l$. Liver function tests were normal. Computed tomography (CT) scan revealed an engorged splenic artery, 1 cm in diameter, and a fusiform dilated splenic vein, $5 \times 6 \times 9$ cm in size (figure 1). A fistula between the splenic artery and vein was tentatively diagnosed.

Selective celiac angiography and superselective splenic arteriography with frontal and bilateral oblique projections were performed. Angiograms revealed a fistula between the tortuous, dilated splenic artery and aneurysm-like splenic vein (figure 2). There was early opacification of the splenic venous aneurysm with a 'turbo-whirl' pattern as well as hepatofugal opacification of the superior and inferior mesenteric veins (figure 2). A tentative diagnosis of splenic arteriovenous fistula and splenic venous aneurysm with hyperkinetic portal hypertension was made.

Because of continuing melaena, embolization of the splenic artery was performed using eight giant Gianturco steel coils. Initially, a coil with a diameter of 10 mm and a length of 10 cm was used because the splenic artery measured around 10 mm in diameter on CT images. Subsequently, a smaller coil (5 mm $\times$ 5 cm) was used to occlude the lumen of the 10 mm $\times$ 10 cm coil. This was not successful because of hyperkinetic flow, and the smaller coil floated into the aneurysm. Finally, for further thorough embolization, six larger coils (10 cm $\times$ 15 mm) were deployed into the splenic artery to establish a complete meshwork. After the embolization by coils, Gelfoam pledgets were added to the coil tangle for further closure of the fistula (figure 3). The bruit in the left lower chest was absent after embolization. The

Figure 1 Serial CT images (from left to right) show an engorged splenic artery (white arrows) which is contiguous to the strongly enhanced mass or aneurysm (A) and dilated splenic vein (v). Note the accessory spleen or splenosis (white arrowheads)

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Figure 2 Celiac angiogram shows the venous aneurysm (black arrowheads) and early opacification of the dilated portal venous system (white arrows). Hepatofugal opacification of superior mesenteric vein (small black arrows) and inferior mesenteric vein (large black arrows) was noted

Figure 3 (A) Radiograph shows the giant Gianturco steel coils (arrowheads) deployed into the splenic artery to create a meshwork. (B) Follow-up angiogram reveals the stasis of contrast medium in the splenic artery with occlusion of the arteriovenous fistula. The aneurysm was no longer visualized. Note the smaller coil floated into the aneurysm.

patient was finally discharged without any symptoms.

Discussion

Fistula between the splenic artery and vein are rare. The first case was reported by Wiegert in 1886.⁶ Only 42 patients with SAVF have been reported in the English literature. The majority of these occurred after rupture of a splenic

Learning points

- SAVF should be suspected in cases of portal hypertension with normal liver function, audible abdominal bruit, and a traumatic or surgical history
- characteristic angiographic findings of SAVF include a tortuous splenic artery, early filling of the splenic vein during the arterial phase, dense opacification of splenoportal venous system and aneurysm-like splenic vein
- transcatheter arterial embolization is a safe and effective alternative to surgery in the management of SAVF

artery aneurysm (29 cases). Other less common causes included splenectomy ($n = 5$), gun-shot ($n = 3$), congenital origin ($n = 3$), mycotic infection ($n = 1$) and splenoportography ($n = 1$).⁶⁻⁹ The communication of artery and vein usually remains asymptomatic for a long time.

The main clinical manifestations are upper abdominal pain, diarrhoea, oesophageal variceal bleeding and abdominal bruit. The bruit, as noted in this patient, can be either machinery or systolic murmur. It is a diagnostic hallmark and is present in only one-third of patients.⁶ It may be heard over the epigastrium, left upper abdomen, left lower chest or left flank. These clinical manifestations may disappear after the development of collateral venous channels. Gastrointestinal bleeding may be caused by an increased portal pressure and hepatoportal sclerosis.¹⁰ High blood flow through the central splenic shunt may lead to development of nontransmural small bowel ischaemia and mesenteric steal syndrome.

Arteriovenous fistula after surgery most probably occurs when vessels are ligated with transfixion sutures or *en masse* ligation of arteries and veins. Occasionally, penetrating injury of vessels by needles, wires, or pins, may also be the cause of fistulae. SAVF may be avoided by individual ligation of the splenic artery and vein in cases of splenectomy.³

SAVF must be confirmed by selective celiac or splenic arteriography. An elongated, tortuous splenic artery, early filling of the splenic vein during the arterial phase, dense opacification of the splenoportal venous system and an aneurysm-like splenic vein are the characteristic findings of SAVF. If the splenic vein is grossly dilated, the contrast medium may fill in with a 'turbo-whirl' pattern. Angiography should be carried out in all patients with portal hypertension, normal liver function, audible abdominal bruit, and traumatic or surgical history.

In most instances, elimination of SAVF is recommended, even in asymptomatic patients, to prevent complications. Surgical excision of SAVF is technically difficult and is sometimes unsuccessful because of the remote location of the lesion, presence of numerous portal collaterals and adhesion. Interventional radiologic techniques, such as transcatheter occlusion of vessels, offer alternatives to surgery. Oblitera-

tion of SAVF with Gianturco coils creates a permanent thrombosis. Coils reduce the flow and induce clotting by providing an increased surface for thrombocytic aggregation. The SAVF in our patient was successfully occluded

by eight giant Gianturco coils deployed through a catheter. Transcatheter arterial embolization is now emerging as a safe and effective alternative to surgery in the management of SAVF.

- 1 Buckhottz RP. Arterial venous fistula of the splenic vessels. *Ann Surg* 1958;149:590-2.
- 2 Sethi GK, Gonzales GC. Splenic arteriovenous fistula: diagnosis and management. *J Kansas Med Soc* 1973;74:224-5.
- 3 Hyde GL. Post-splenectomy arteriovenous fistula causing portal hypertension. *J Kentucky Med Soc* 1979;77:113-5.
- 4 Gudmundsen TE, Lie M, Ostensen H. Splenic arteriovenous fistula. *Acta Chir Scand* 1988;154:603-4.
- 5 Keller FS, Rosch J, Dotter CT. Bleeding from esophageal varices exacerbated by splenic arterial-venous fistula: complete transcatheter obliterative therapy. *Cardiovasc Intervent Radiol* 1980;3:97-102.
- 6 Van Way CW, Crane JM, Riddell DH, Foster JH. Arteriovenous fistula in the portal circulation. *Surgery* 1971;70:876-90.
- 7 Bredfeldt JE, O'Laughlin JC. Portal hypertension secondary to a congenital splenic arteriovenous fistula. *J Clin Gastroenterol* 1980;2:355-8.
- 8 Vauthey JN, Tomczak RJ, Helmberger T, et al. The arterioportal fistula syndrome: clinicopathologic features, diagnosis and therapy. *Gastroenterology* 1997;113:1390-401.
- 9 Pietri J, Remond A, Reix T, Abet D, Sevestre H, Sevestre MA. Arterioportal fistulas: twelve cases. *Ann Vasc Surg* 1990;4:533-9.
- 10 Pasternak BM, Cohen H. Arteriovenous fistula and forward hypertension in the portal circulation. *Angiology* 1978;29:367-73.

Postpericardiotomy syndrome following temporary and permanent transvenous pacing

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Summary

The postpericardiotomy syndrome may occur as a complication of temporary and permanent pacing. Physicians involved in procedures which may be complicated by this condition therefore need to be aware of its diagnosis and management.

Keywords: postpericardiotomy syndrome; cardiac pacing

Postpericardiotomy syndrome is recognised as one of the commonest complications following cardiac surgery, with an incidence of between 10-50% being reported.¹ Its description following the insertion of a transvenous pacemaker, however, is relatively rare. We present a case of temporary and subsequent permanent transvenous pacing resulting in the postpericardiotomy syndrome.

Case report

A 68-year-old man was admitted with a history of presyncopal and syncopal symptoms. There was a previous history of atrial fibrillation, transient ischaemic attacks (for which he was anticoagulated with warfarin) and ischaemic heart disease. On examination, he had a heart rate of 35 beats/min, the electrocardiograph (ECG) showing slow atrial fibrillation. A temporary pacing wire was inserted via the right internal jugular vein and subsequently required repositioning on two occasions due to failure to capture. Two hours following repositioning he initially experienced left shoulder pain, later associated with intermittent neck and chest pains and a temperature of 37.8°C.

A VVI permanent pacemaker was inserted (via the left subclavian route) and temporary wire removed 5 days following admission. This was complicated by dull central chest pain following the procedure. A pneumothorax was excluded and a small posterior pericardial effusion was demonstrated on echocardiography. He continued to experience chest pain and an intermittent pyrexia. A swelling at the right side of his neck (site of temporary pacing) was shown by Doppler ultrasonography to be due to a thrombosis involving the internal jugular vein. The erythrocyte sedimentation rate (ESR) was elevated at 115 mm/h. He was discharged 11 days later following resolution of his symptoms and pyrexia, although the ESR remained elevated at 100 mm/h.

At re-admission, 3 weeks later, he gave a history of acute pleuritic chest discomfort and dyspnoea. On examination, he had a temperature of 38°C, a pericardial friction rub and reduced air entry at the left pulmonary base. Chest radiography revealed cardiomegaly and a small left pleural effusion. The white cell count (WCC) was $13 \times 10^9/l$ (neutrophilia) and cardiac enzymes normal. Serial blood cultures remained sterile. The ECG revealed ventricular pacing on a background of atrial fibrillation. At echocardiography, a 3.5 cm pericardial effusion was identified with mild mitral regurgitation and good left ventricular function. The ESR was 107 mm/h, C-reactive protein 251 mg/l (normal <10 mg/l), complement C3/C4 levels normal and autoantibodies (including antineutrophil cytoplasmic antibody and anticardiolipin IgG and IgM) negative. Right heart catheterization revealed moderate pulmonary hypertension (pulmonary artery pressure

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