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CASE REPORT

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Endovascular treatment of giant splenic aneurysm that developed after liver transplantation

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Abstract Splenic artery aneurysms (SAAs) are not uncommon in patients with portal hypertension. They are usually diagnosed in preliminary examinations prior to orthotopic liver transplantation (OLT) and are treated surgically at the time of transplantation. In our case, the patient developed a giant SAA after liver transplantation. This was detected incidentally upon routine ultrasound follow-up, and the diagnosis was confirmed on magnetic resonance (MR) angiography. The patient was treated by endovascular embolization because it is believed that this minimally invasive approach is beneficial in an immunocompromised patient following OLT. After coil embolization, to achieve complete and immediate blood flow exclusion of the sac, it was decided to inject some glue (*N*-butyl-2-cyanoacrylate) directly into the aneurysm. The aneurysm was successfully obliterated. To the best of our knowledge the use of cyanoacrylate glue in an SSA has never been reported.

Keywords Portal hypertension
Liver transplantation
Splenic artery aneurysm
Endovascular embolization
Cyanoacrylate glue · Post-OLT complication

Introduction

Splenic artery aneurysm (SAA) is the third most common intra-abdominal aneurysm after infrarenal aortic aneurysm and iliac artery aneurysm. Stanley and Fry reported an incidence of SAA ranging from 0.8% in unselected autopsy population to 7.1% in patients with portal hypertension and cirrhosis [1]. The reported rupture rate of SAA in the unselected population is 3–10%, with a related mortality of 10–25% [1, 2]. There is a significantly higher mortality rate following rupture in patients with cirrhosis [3, 4]. In the current literature only two cases of SAA after orthotopic liver transplantation (OLT) have been reported [3, 5]. The indications for treatment are controversial and include: the presence of symptoms, increasing size, and a diameter of 2 cm [6]. Surgery remains the definitive treatment, but percutaneous embolization could be an effective and less invasive treatment in selected cases.

Case report

A 56-year-old male patient with hepatitis B and C-related cirrhosis underwent OLT from a cadaveric donor in December 1993. His post-operative recovery was uncomplicated, and immunosuppressive therapy commenced (cyclosporine, azathioprine, and prednisolone) on the first post-operative day. From 1993 to 2000 routine follow-up investigations, including biochemical evaluation of liver, kidney, and pancreatic function and serum cyclosporine, α_1-antitrypsin, and ferritin levels, were normal, except for a low level of α_1-antitrypsin. Regular clinical examination, annual colour-Doppler ultrasound of the graft, and liver biopsy, were also performed and found to be normal.

In December 2000 a routine liver biopsy was complicated by the development of severe abdominal pain. Ultrasound examination of the abdomen demonstrated a dilatation of the splenic artery (Fig. 1a). Magnetic resonance (MR) angiography was performed, which showed a 4-cm SAA containing an eccentric thrombus (Fig. 1b) and significant tortuosity of the splenic artery (Fig. 1c). None of these features had been present in pre-operative examinations. Following informed consent, the patient underwent transfemoral digital subtraction angiography (DSA) that confirmed the MR findings. The tortuosity of the splenic artery prevented distal catheterization by the transfemoral approach and, therefore, a left axillary approach was used. The splenic artery was then selectively catheterized with a 5 F head-hunter catheter (Terumo, Tokyo, Japan) advanced over a 0.035 hydrophilic wire (Terumo).

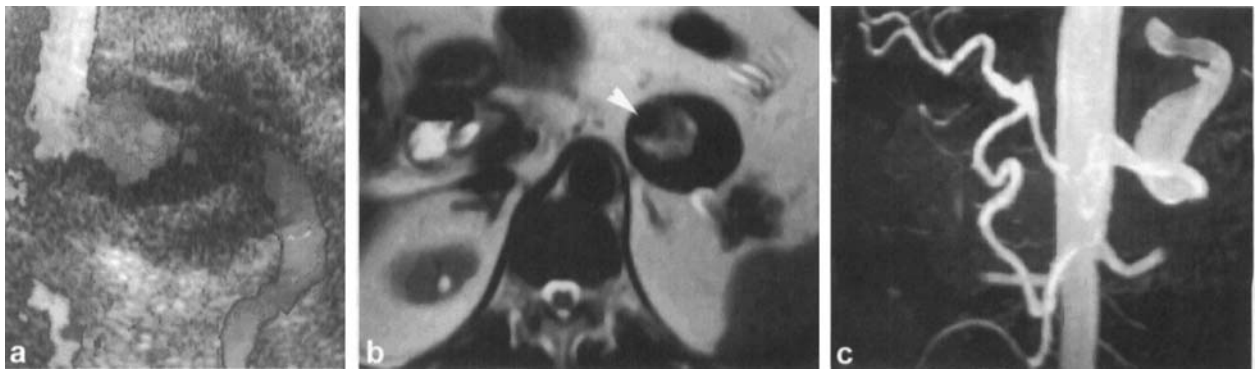
Gianturco coils of 6 and 8 mm in diameter and 10 cm in length (Cook, Bloomington, Ind. USA) were then deployed both proximally and distally to the neck of the aneurysm in order to exclude the aneurysm sac. Confirmation DSA and follow-up CT angiography after 1 month showed persistent flow into the aneurysm sac, and some flow in the distal part of the artery was also noted. A second embolization was therefore attempted, again via a transaxillary approach, with larger coils (9 mm–6 cm and 15 mm–10 cm). During the procedure a reduction in flow was seen, but filling of the aneurysm was still present (Fig. 2a). To achieve total occlusion of the SAA, we instilled 0.7 cc of *N*-butyl-2-cyanoacrylate (Embucrilate, Histoacryl, Braun, Melsungen, Germany), mixed with 0.7 cc of iophendylate (Lipiodol, Laboratoires Guerbet, France), into the aneurysm. The addition of iophendylate was required to delay the polymerization time of the cyanoacrylate glue. Completion DSA demonstrated total occlusion of the splenic artery with complete absence of flow into the aneurysm (Fig. 2b). Colour-Doppler examination after 24 h showed no flow into the sac, and CT angiography at 1 and 6 months confirmed complete occlusion of the SAA (Fig. 3). The patient is currently asymptomatic and well.

Fig. 1 a Colour-Doppler ultrasound following liver transplantation shows partially thrombosed aneurysm of the splenic artery. The lesion is confirmed by MR angiography, which shows the large amount of eccentric thrombus within the aneurysm (**b**) and significant tortuosity of the splenic artery (**c**)

Discussion

SAAs have been reported in 0.1% of autopsy cases and in 0.8% of unselected angiograms [1]. They are more common in women, particularly those with a history of multiple pregnancies [1, 6]. This is thought to be related to a combination of haemodynamic and hormonal effects on the arterial wall. The incidence of SAAs is higher in patients with liver cirrhosis and portal hypertension, ranging from 7 to 13% [1, 7]. The reasons for this are unclear, and numerous factors have been implicated, including α_1-antitrypsin deficiency [8] and haemodynamic changes. The haemodynamic changes associated with cirrhosis are likely to play an important role. It has been widely shown that increased flow and high blood pressure can produce elongation and overall dilatation of the splenic artery [9]. In patients with cirrhosis and portal hypertension, both splenic artery blood flow and pressure are increased. Splenic artery pressure rises as a consequence of increased portal vein pressure. The formation of arteriovenous shunts and collateral vessels results in compensatory changes in cardiac output that contribute to the increased splenic and splanchnic blood flow. This is compounded by splanchnic vasodilatation secondary to hyperglucagonaemia and vascular changes caused by other hormone changes, such as those that "feminize" male cirrhosis patients [9].

The reasons for the development of SAA after OLT are unknown, as there are only two cases reported in the literature [3, 5]. They are unlikely to be related to alterations in both splenic artery flow and pressure following OLT. In fact, a restoration of normal splenic blood flow after transplantation was well demonstrated by Bolognesi et al., who measured splenic Doppler impedance in patients pre-OLT and post-OLT [10]. Splenic artery pressure also appears to return to normal values at 6–8 months after OLT [11]. In our case, no



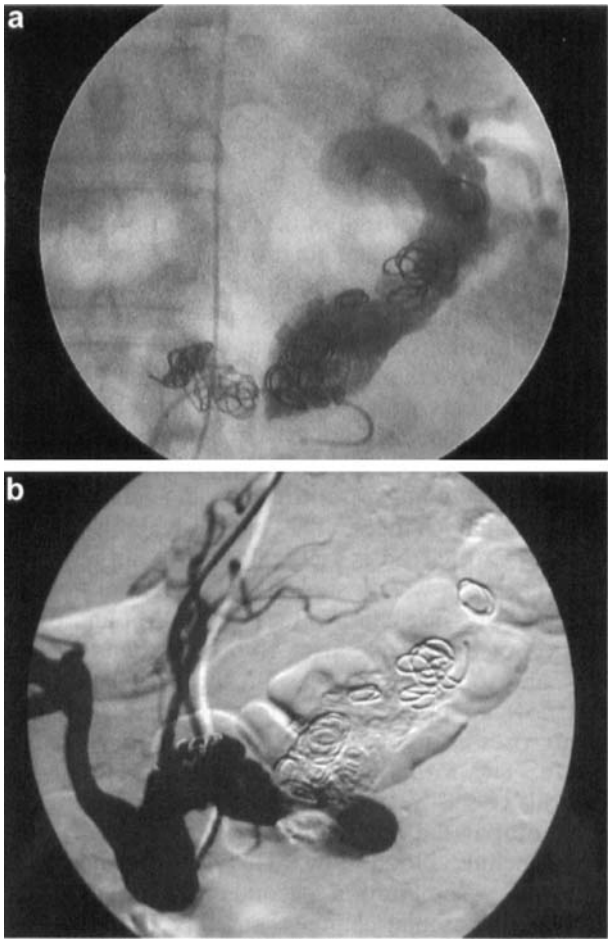


Fig. 2a, b Selective angiography of the splenic artery. **a** Placement of coils alone shows presence of residual flow in the artery. **b** After injection of 0.7 cc *N*-butyl-2-cyanoacrylate directly within the sac complete exclusion of the aneurysm is seen

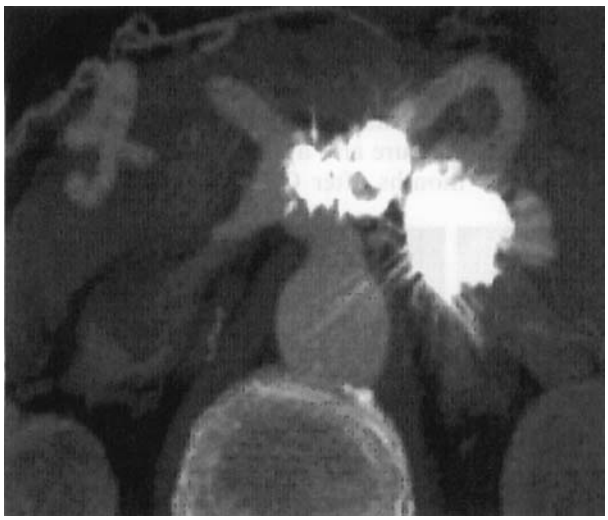


Fig. 3 Follow-up CT angiography at 6 months confirms complete exclusion of the SAA

clear aetiological factors were identified, although the patient was noted to have low serum α_1-antitrypsin.

The aneurysm arose from the medial third of the splenic artery, whereas most originate from the distal third of the vessel [1, 5, 6]. Although some aneurysms may present with evidence of rupture or symptoms of local mass effect, they can also be incidental findings. In these cases, the presence of a large aneurysm of the main trunk of the splenic artery is an indication for elective treatment. Most authors agree that rupture is most likely when the aneurysms achieve 2 cm in size [2, 6]. Surgery has also been recommended if the SAA is adjacent to the coeliac artery or is located in the middle-to-distal third of the splenic artery [6].

The procedure involves exclusion of the sac, by proximal and distal ligation of the splenic artery (to prevent back-filling of the aneurysm from the abundantly supplied spleen) or excision of the aneurysm with or without splenectomy [6, 12]. Although the procedure is effective, the mortality after such operations is often high and, furthermore, splenectomy renders the immunosuppressed patient at risk of severe infection [13]. For these reasons, a minimally invasive technique that preserves the spleen may be preferred. Trans-catheter percutaneous embolization of the splenic artery has been successfully performed [14, 15]. However, repeated embolization may be required, as in this case.

In the first procedure we chose 6–10 and 8–10 Gianturco coils (steel spring coils with attached thrombogenic Dacron strands), with curve memory and good hoop strength, to begin a coil nest. These coils are highly stable and more likely to conform to the vessel than larger coils, and feature a more predictable shape once released. The coils were precisely placed proximally and distally to the aneurysm in order to prevent both antegrade and retrograde filling of the aneurysm sac, the latter due to splenic re-vascularization via the pancreatic arteries. Slow flow was seen in the aneurysm at the end of the procedure, and, instead of deploying further coils, we considered that the SAA would eventually thrombose over the next few days. Indeed, on the basis of our previous experiences in the treatment of aneurysms, and in accordance with the literature [12], we initially prefer to treat such lesions without releasing a great number of coils (or other embolic material), as this often fails to provide better results and, instead, may increase the risk of both immediate and/or delayed material displacement as well as the risk of artery rupture. Generally, the release of a small number of coils proximally and distally to the aneurysm neck results in complete aneurysm exclusion over the next few days [12]. However, in a few cases a second embolization (sometimes directly within the sac) may be required. A second embolization does not generally present increased difficulties unless the aneurysm is supplied from a retrograde flow through the

distal coils (occasionally a delayed migration of one or more coils may occur, resulting in the re-opening of the vessel). In such a case, the release of embolic agents distally into the artery may be restricted, due to the catheter's inability to pass through the proximal coil "wall". When even the use of a micro-catheter is unsuccessful in this situation, the second embolization cannot be performed, and the patient must undergo surgery. However, this technical impediment is less likely to happen in big vessels (such as the splenic artery) than in the smaller ones, where it may be more difficult to find a path in the coil "wall" through which to push the catheter.

Probably due to the giant dimension of the aneurysm, total thrombosis of the sac did not occur in our patient, and a second embolization was performed, initially with larger-diameter coils, with some being released directly into the aneurysm sac. This carried the potential risk of arterial wall rupture secondary to increasing sac pressure during and after release of the coils. We agree with Uflaker that occlusion of the splenic artery itself is preferable and safer in comparison to aneurismal cavity embolization [15, 16]. However, when coils fail to occlude the artery either distally or proximally to the aneurysm, embolization of the sac may be attempted.

Complete occlusion of the splenic artery was achieved by the instillation of 0.7 cc of *N*-butyl-2-cyanoacrylate mixed with 0.7 cc of iophendylate (ratio 1:1). The instillation of this occluding agent, used most commonly for the embolization of cerebral arteriovenous malformations as well as carotid and cerebral aneurysms, has never been reported in the literature for the treatment of SAA [17]. In addition to occluding the main vessel, the *N*-butyl-2-cyanoacrylate mixture may also be useful in such aneurysms that can no longer accommodate further coils, or in preventing delayed migration of micro-coils.

However, migration of the cyanoacrylate from the injection site remains a realistic problem. For this reason this agent must only be handled by experienced and skilled interventional radiologists and should be used mainly when coil embolization has resulted in only a reduction of blood flow within the splenic aneurysm. In such cases, cyanoacrylate is likely to ensure complete aneurysm occlusion and, furthermore, is much less likely to damage the non-target vessels because of the reduced flow, which deters migration. This complication also limits the use of other particulate embolic agents (e.g. gelfoam or polyvinyl alcohol), which may enter the distal splenic circulation and cause splenic infarction or abscess formation [14]. A more recent report described the use of a stent graft to exclude a SAA [18], and this would avoid the risks of embolization. However, as in our case, the use of this technique may be limited, as the tortuosity and large diameter of the splenic artery may preclude stent graft deployment.

In conclusion, endovascular techniques could be considered an attractive alternative to surgery for the treatment of SAA in patients following OLT. The procedure could also provide temporary control of bleeding prior to surgery. Embolization with cyanoacrylate is a valid option for SAA, which is difficult to treat by coil embolization alone [19]. The aetiology of post-OLT SAAs is unclear, and regular ultrasound surveillance of the splenic and splanchnic vessels after transplantation is recommended.

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