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BRIEF REPORT

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Splenic artery aneurysm and orthotopic liver transplantation

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Abstract Splenic artery aneurysms are a rare but potentially fatal complication after liver transplantation. We report three cases presenting in a 12-month period in adult patients who underwent transplantation for chronic liver disease. Doppler ultrasound of the splenic artery should be performed in all patients with cirrhosis and portal hypertension who are being assessed for liver transplantation. The aneurysm can be ligated at the time of transplantation.

Key words Liver transplantation, splenic artery aneurysm · Splenic artery aneurysm, liver transplantation · Portal hypertension, splenic artery aneurysm · Alpha-1-antitrypsin deficiency, splenic artery aneurysm

Introduction

Splenic artery aneurysms are the third most common intra-abdominal aneurysm and account for 60% of all splanchnic artery aneurysms. The incidence of splenic artery aneurysm in patients with cirrhosis and portal hypertension is between 7% and 20% [3, 13, 18], and in those with chronic liver disease who undergo liver transplantation it is 10% [1]. Splenic artery aneurysm rupture occurs in 3%–10% of cases [3, 18, 20] with a mortality of 25%. Following liver transplantation, alteration in splanchnic blood flow, surgical trauma, weakening of the aneurysm wall and systemic hypertension may predispose to rupture [2].

Case reports

Case 1

A 36-year-old man developed cryptogenic cirrhosis at 3 years and procto-colitis at 7 years of age and was diagnosed as having primary sclerosing cholangitis at 30 years. He developed progressive liver failure and was referred for transplantation. Pre-transplant, an aorto-portogram demonstrated a large splenic artery aneurysm and porto-systemic collaterals. At subsequent orthotopic liver transplantation (OLT) he was unable to tolerate cross clamping of the inferior vena cava (IVC). He had a systemic vascular resistance (SVR) of 250 dynes/cm-5/s-2 despite a cardiac output of 12 l/min and was placed on veno-venous bypass. He was found to have a partial portal vein thrombosis with severe calcification of the posterior wall and an 8-cm splenic artery aneurysm associated with a large arterio-venous fistula (AVF). The splenic artery and AVF were ligated and the aneurysm excised. His post-operative recovery was complicated by severe rejection, which responded to three doses of 1 g methylprednisolone and conversion to tacrolimus, as well as haemodialysis for deteriorating renal function. Laparotomy was performed following an intra-abdominal bleed, but no specific bleeding site was identified. Post-operatively he developed a pancreatico-cutaneous fistula that settled with conservative management that included octreotide (Sandostatin, Sandoz). The patient was discharged 9 weeks post-transplant with normal liver and renal function on tacrolimus.

Case 2

A 63-year-old man with alcoholic liver disease, hepatocellular carcinoma and severe portal hypertension underwent transplantation using veno-venous bypass. He underwent preoperative chemo-embolisation of the tumour as well as a standard OLT apart from a thrombectomy for a localised portal vein thrombosis. He made a good recovery until the 6th post-operative day when he became hypotensive and bled 1700 ml from the left intraperitoneal drain. At laparotomy the liver was healthy but there was copious blood and a clot in the retroperitoneum and perisplenic region. A 6-cm splenic artery aneurysm, which had ruptured posteriorly, was found and transfixed from within the aneurysm sac proximally and distally. Six days later he rebled, and at laparotomy a small ar-

terial bleeding vessel was ligated in the lesser sac. Following this he made an uncomplicated recovery and was discharged on cyclosporin, azathioprine and prednisolone.

Case 3

A 22-year-old man with alpha-1-antitrypsin deficiency received an orthotopic liver transplant in Australia in 1991. He presented to our unit 3 years later with jaundice, abdominal pain and fevers. Liver function tests revealed an obstructive pattern. Abdominal ultrasound and percutaneous transhepatic cholangiogram demonstrated an anastomotic stricture, hepatic duct stones and obstructed Roux loop. An angiogram was performed and showed a large, tortuous splenic artery but no other abnormality. The hepatico-jejunostomy was revised and stones removed. At operation, multiple splenic artery aneurysms were noted of 3–4 cm in diameter. Twelve days later the patient was readmitted with right upper quadrant pain and fevers. A perihepatic collection was drained percutaneously but failed to resolve. Laparotomy was performed and the collection drained. Two splenic artery aneurysms were ligated with transfixion of feeding vessels. He was discharged on cyclosporin, azathioprine and prednisolone and is well 1 year later.

Discussion

Splenic artery aneurys m has been associated with cirrhosis and portal hypertension [1, 11, 14], and the most common sites are the distal third or branches of the splenic artery [20]. The majority of splenic artery aneurysms are asymptomatic and less than 20 mm in diameter [18]. Rupture appears to be more likely if the aneurysm is greater than 20 mm in size [3]. Two descriptions of five patients with chronic liver disease and ruptured splenic artery aneurysms reported no survivors [9, 13], presumably because of a delay in the diagnosis or decompensation of the cirrhotic liver. It has been suggested that the increased splenic arterial blood supply is a result of the hyperdynamic circulation of cirrhotic patients with portal hypertension [9] and is an important factor in their development [2, 9]. Increased collagen lysis following laparotomy [19] and an increase in splenic arterial blood flow after liver transplantation [1] have also been suggested as predisposing factors for rupture. Ultrasound has been recommended for patients being assessed for liver transplantation [2], but specific views of the splenic artery must be obtained. Ultrasound examination requires an experienced ultrasonographer [11] and, in view of our recent experience, all patients undergoing assessment for liver transplantation should be evaluated for splenic artery aneurysm. Selective angiography may be helpful in defining the aneurysm and excluding multiple aneurysms, which occur in 20% of cases. Some centres [11] have recommended coeliac angiography in all patients prior to liver transplantation; however, with careful Doppler ultrasonography this is unnecessary.

All splenic artery aneurysms, regardless of size [21], should be ligated or excised in this population because of the increased risk of rupture. If recognised prior to transplantation, radiological intervention to thrombose the aneurysm with coil embolisation, should be considered. There is a risk of splenic infarction and splenectomy may be required. Splenectomy in patients with cirrhosis and portal hypertension prior to transplantation poses the risk of splenic vein thrombosis with extension into the portal vein. At the time of liver transplantation, the aneurysm should be ligated proximally and distally, and bleeding points should be underrun from within the sac. Splenectomy should only be performed for aneurysms of the very distal splenic artery because of the increased risk of severe infections in immunosuppressed patients [16] and of splenic vein thrombosis. Despite adequate control of the splenic artery aneurysm, subsequent laparotomy was required in two of our patients for bleeding from the lesser sac and/or pancreas. This occurred in case 1 because the aneurysm was embedded in pancreatic tissue and difficult dissection resulted in damage to the superior border of the pancreas. This patient had also developed a splenic AVF, which is a recognised complication of ruptured splenic artery aneurysm [6, 7, 22]. The presence of the AVF explains the low SVR and the unexpected inability of a young patient to tolerate cross clamping of the IVC and portal vein without veno-venous bypass. Splenic AVF has not previously been described as a problem during liver transplantation, but it has been reported as a cause of hepatic hypoperfusion following liver transplantation [10].

Alpha-1-antitrypsin deficiency has also been associated with splenic artery aneurysms. Including our cases, 3 of the 13 patients reported with splenic artery aneurysms who underwent OLT had alpha-1-antitrypsin deficiency [1, 2, 8, 11]. Ruptured splenic artery aneurysm has also been reported in a patient with cirrhosis secondary to alpha-1-antitrypsin deficiency [17], as well as in a patient with iliac artery dissection [4]. Alpha-1-antitrypsin content in the aortic wall has been found to be significantly lower in patients with multiple infra-renal aneurysms and ruptured abdominal aortic aneurysms [5]. Alpha-1-antitrypsin deficiency has also been associated with rupture of a middle colic artery aneurysm in a patient with multiple visceral aneurysms [12] and with rupture of an intra-cranial aneurysm and spontaneous dissection of the internal carotid artery [15]. Alpha-1-antitrypsin is important for maintaining the integrity of blood vessel walls by inhibiting proteolytic enzymes. Degradation of the arterial wall is thought to occur because of an imbalance between proteolytic enzymes (e.g. elastase) and their inhibitors (e.g. alpha-1-antitrypsin), which predisposes to aneurysm formation [15]. Alpha-1-antitrypsin deficiency appears to be a risk factor for the development of aneurysms and particularly for splenic artery aneurysms in patients with cirrhosis and portal hypertension. These patients must be carefully screened for aneurysms prior to liver transplantation.

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