CASE REPORT

Hepatic arterial embolization for massive bleeding from an intrahepatic artery pseudoaneurysm using *N*-butyl-2-cyano-acrylate after living donor liver transplantation

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Keywords

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Introduction

Bleeding from a hepatic artery pseudoaneurysm (HA PsA) is a life-threatening but rare complication following liver transplantation [1]. Most of the cases occur at the surgical anastomosis site of the extrahepatic portion of the HA. Surgical resection, covered stenting, and coil embolization are possible ways of managing extrahepatic artery PsA [2]. However, using coil embolization to treat peripheral intrahepatic artery PsA may be unsuccessful if there is failure of the coil to embolize the bleeding vessel resulting from a difficult selective catheterization of the PsA sac which prevents back-filling of the PsA from more distal arterial collaterals. Inadvertently, coil embolization may also cause necrosis or atrophy of the involved arterial segment.

The authors report a postliving donor liver transplant female recipient who developed massive subcapsular

Summary

Parenchymal pseudoaneurysm of the hepatic arteries with massive intraperitoneal bleeding is rare but a serious life-threatening complication when it occurs following liver transplantation. We report a case of an adult postliving donor liver transplant recipient who developed massive subcapsular bleeding combined with massive right pleural effusion from ruptured multiple small intrahepatic arteries, which developed from a pseudoaneurysm that was treated by hepatic arterial embolization. Successful embolization was performed via a percutaneous trans-catheter approach by depositing 20–25% *N*-butyl-2-cyanoacrylate (NBCA) through the multiple small intrahepatic arteries into the pseudoaneurysm. Complete occlusion of the feeding arteries and pseudoaneurysm cavity resulted to immediate cessation of bleeding. There was no re-bleeding; and normal liver graft function was noted postembolization. Hepatic arterial embolization with NBCA can be used as treatment for postliver transplant peripheral intrahepatic artery pseudoaneurysm bleeding.

> bleeding combined with massive right pleural effusion from ruptured multiple peripheral intrahepatic arteries fed by a HA PsA and was successfully treated by transcatheter approach depositing 20–25% *N*-butyl-2-cyanoacrylate (NBCA). The clinical presentation and management resulting in full patient recovery are described.

Case report

A 67-year-old Chinese female (height: 154 cm; weight: 46 kg) with hepatitis C virus-related end-stage liver disease (Child score: 10; Model for end-stage liver disease score: 15) underwent living donor liver transplantation with a right-lobe liver graft without the main middle hepatic vein trunk (graft weight: 514 g; graft weight-to-standard liver volume: 51.8%; graft-weight-to-recipient weight ratio: 1.11) donated by her daughter. Her preoperative imaging

modalities showed liver cirrhosis with one early enhancing tumor (2 cm) in segment 4 of the liver. There was unremarkable finding in the preoperative imaging of the donor's liver. The intraoperative course was, likewise, unremarkable (cold ischemia time: 60 min; warm ischemia time: 53 min; anhepatic phase: 69 min; total operative time: 565 min). The intraoperative blood loss was approximately 3000 ml. She was transfused 2272 g of leukocytefree packed red cells and 10 units of fresh frozen plasma.

On the first postoperative day, the patient developed unstable hemodynamics with arterial blood pressures that ranged from 60 to 90 mmHg and profuse bleeding was noted from the drainage tubes. Emergency laparotomy revealed a huge subcapsular hematoma, 10×8 cm in size, which was not present at the time of wound closure. There was rupture of the hematoma along the raw surface of the liver graft with active bleeding. Adequate hemostasis was achieved after application of Surgicel® (Johnson & Johnson, New Brunswick, NJ, USA) and initial packing with laparotomy pads. The bleeding abated and the abdomen was closed after removing the laparotomy pads.

Forty-five days after transplant, the patient was exhibiting massive ascites and progressive shortness of breath. The ascites culture showed heavy growth of *Acinetobacter lwoffii*. On further septic work-up, the computed tomography angiography study of the newly transplanted liver graft and abdomen showed a 1.13-cm PsA in the periphery of the liver graft parenchyma to its right with multiple small right hepatic arterial collaterals (Fig. 1a). A 14-cm hematoma in the subcapsular region of the liver graft to its right was also seen. With an impression of ruptured HA PsA, the patient was subsequently referred for emergency interventional radiologic procedure(s).

Prophylactic dose of ciprofloxacin (400 mg intravenous q12 h) was given prior to intervention. Using Seldinger technique, the right femoral artery was punctured and a 4-French sheath was placed into the aorta through the right femoral approach. Through the sheath, a 4-French catheter was advanced into the celiac artery for celiac

angiography. Celiac arteriography showed a PsA over the distal portion of the HA with multiple small twig collaterals. A micro-catheter (Progreat® 2.8-French; Terumo, Tokyo, Japan) was used to access the PsA. The peripheral location of the PsA with its small caliber and spastic appearance of the distal HA twigs made it impossible to access the PsA sac. Embolization was performed using 25% mixture of NBCA (Histoacryl Blue; Braun, Melsungen, Germany) and iodized oil (Lipiodol; Laboratoire Guerbet, Roissy, France) via flow guidance into the predominantly supplied branches of the right HA until blood flow into the PsA was almost completely impeded. Immediate on-table postembolization celiac angiography showed residual PsA with its blood supply coming from other small feeding branches of the right HA not previously identified. A repeat embolization was performed using 20% mixture of NBCA and iodized oil via flow guidance to these small feeding branches of the right HA to occlude the residual PsA. A second postembolization celiac angiography showed total occlusion of the branches of the supplying arteries of the PsA. There was almost no effect on the normal liver parenchymal enhancement after the two embolization procedures (Fig. 2).

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The immediate postembolization clinical course was unremarkable. Because *Acinetobacter* sp. and other Gramnegative organisms may have predisposed the patient to PsA (mycotic aneurysm), the same dose of ciprofloxacin was continued until the 14th postprocedure day. The patient complained only of mild right upper abdominal discomfort after hepatic arterial embolization (HAE). This was effectively relieved by routine orally administered analgesics. The shortness of breath was gradually relieved and patient activity improved. The liver enzymes showed good, functioning liver graft.

One month postprocedure, the follow-up computed tomography angiography did not show any infarction in the liver graft and there was a marked decrease in the size of the hematoma (Fig. 1b). At 6 months post-HAE, there was no recurrence of bleeding or PsA.

Figure 1 (a) Pre-embolization computed tomography angiography (CTA) showing an intrahepatic arterial psuedoaneurysm (arrow) with multiple small intrahepatic arteries supplement. (b) Follow-up CTA in 1 month after hepatic artery embolization showed total occlusion of the pseudoaneurysm without infarction in the liver graft.





Figure 2 (a) Pre- and (b) postembolization celiac arteriogram showing occlusion of supplement hepatic artery branches of pseudoaneurysm with minimal obliteration of normal liver parenchyma.

Discussion

Hepatic artery PsAs are uncommon and occur in 0.4% of patients after liver transplantation [3]. HA PsA are classified into intra- and extrahepatic PsAs. Their etiology, clinical course, and radiologic features differ [4]. Surgical resection with extra-anatomical autologous vascular bypass, trans-catheter endoluminal embolization, stent placement (stent-graft) and re-transplantation are approaches to manage extrahepatic PsAs [2].

Different treatment options for intrahepatic PsA have been proposed. Intrahepatic PsA can be managed nonoperatively by observation and imaging follow-up with judicious treatment of any associated infection [4]. There is high mortality (50%) associated with HA PsA reported in the literature and its combination with abdominal infection is a poor prognostic factor [4,5]. Nonoperative management is most likely applicable to asymptomatic, small, and nonenlarging intrahepatic PsA. Large symptomatic PsA require procedural intervention. Intrahepatic PsA can be treated by surgical ligation of the involved intrahepatic artery, transplant hepatic segmentectomy, and re-transplantation [4]. Recently, endoluminal trans-catheter embolization [4] and/or direct percutaneous trans-needle embolization (coil or fibrinogen) has been described to treat intrahepatic PsA [2,6].

In postliver transplant recipients, re-operation for surgical correction is a complicated procedure especially in the early post-transplant period. The use of minimal invasive therapy in the aforementioned patients with progressively enlarging intrahepatic artery PsA provides lesser invasive procedure(s) as compared with a laparotomy to treat the PsA [4]. In cases of PsA, an ideal approach is to perform selective micro-catheterization of the sac and deploy coils to pack the lesion. If the involved HA branch is peripheral and relatively dispensable and the operator is unable to selectively catheterize the PsA sac, embolizing the involved arterial branch can be performed. However, in transplanted livers, necrosis or atrophy of the involved segment(s) may occur. Therefore, the inability to selectively catheterize the PsA sac to prevent back-filling of the PsA from more distal arterial collaterals may cause embolization failure. Hence, embolizing an arterial branch with its potential consequence(s) (i.e. necrosis or atrophy) should be weighed against its benefits.

Direct percutaneous image-guided trans-hepatic puncture for coil embolization of intrahepatic arterial aneurysms have been rarely described [6–12]. In the case presented, a percutaneous image-guided trans-hepatic puncture will be time-consuming and a high-risk procedure as it may violate the pleura owing to the high location of the PsA. Further, the lack of liver parenchyma to buttress the procedure is a potential risk for coil migration or fibrinogen leaking into the intraperitoneal cavity.

Recent studies have shown that trans-catheter embolization for peripheral PsA with NBCA is safe and effective [13,14]. In this patient, 20% and 25% NBCA were used as embolizers. The authors applied the technique of introducing an initial 25% NBCA into the peripheral intrahepatic artery via inflow guidance instead of the usual proximal coils embolization as it is easier for the NBCA to reach peripheral branches with lesser complications. Once the predominant PsA was obliterated, the smaller PsA appeared. For this reason, operators should perform a "scout angiogram" to make sure that the initial procedure to obliterate the main PsA has not unmasked any other PsA. If this occurs, a repeated HAE with 20% NBCA is given for the smaller branches. The NBCA mixture has different viscosity to liquid contrast medium and the polymerization time of NBCA is very short. The key to successful HAE using this method is that an optimal concentration of NBCA be approximated according to blood flow velocity and vessel diameter. In this case, it took 30 min to achieve the desired satisfactory vessel

occlusion using the concentration(s) of NBCA given. Postprocedural adverse effects were minor. Low-grade fever and right upper quadrant pain were relieved by oral administration of routine antipyretics and analgesics.

In conclusion, conservative (nonoperative) management is most likely applicable to asymptomatic, small, and nonenlarging intrahepatic PsA. Large progressive symptomatic PsA and those with rupture require procedural intervention. In the early postliving donor liver transplant period vis-a-vis with high-risk re-laparotomy, the role of trans-catheter HAE should be considered in HA PsA. HAE using NBCA is a quick, safe, and effective method of controlling peripheral intrahepatic artery PsA with preservation of the normal liver function.

Authorship

HYO and YFC: designed and performed the case, and wrote the paper. AMC, SHW, and CLC: transplantation surgery in the case. CYY, TLH, TYC, LLT: experts in intervention radiology and contributed ideas for the case.

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