

Case Report

Arterioportal fistula with hepatic artery pseudoaneurysm: A rare complication of percutaneous transhepatic biliary drainage

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ABSTRACT

We describe a rare complication of percutaneous transhepatic biliary drainage in a 51-year-old female with recurrent hilar cholangiocarcinoma. The patient presented with blood-mixed pericatheter leak of bile, 1 month after placement of a 10F ring biliary internal-external drainage catheter through the left-sided biliary radical. Color Doppler and triphasic computed tomography angiography revealed a pseudoaneurysm arising from segment 3 branch of the left hepatic artery and an arterioportal fistula immediately distal to the pseudoaneurysm. Endovascular coil embolization of the hepatic artery proximal to the pseudoaneurysm was done which resulted in occlusion of the pseudoaneurysm and fistula as well as clinical improvement in the symptoms.

Keywords: Klatskin tumor, Percutaneous transhepatic biliary drainage, Pseudoaneurysm, Arterioportal fistula

INTRODUCTION

Hepatic arterial injury can be seen in approximately 1.3–8% of percutaneous transhepatic biliary drainage (PTBD) manifesting as active extravasation, arteriocholedochal fistula, pseudoaneurysm, or arterioportal shunt.^[1-5] Endovascular embolization is the preferred modality of management while surgery is indicated for fistula recurrence or embolization failure.^[6] We report a case of hepatic arterial injury post-PTBD manifesting as hepatic arterial pseudoaneurysm with arterioportal fistula (APF) managed by endovascular coil embolization.

CASE REPORT

A 51-year-old female was referred with recurrent hilar cholangiocarcinoma of size 4 × 4 cm at hepatic hilum on magnetic resonance imaging with IHBRD after palliative hepaticoduodenostomy and external beam radiation therapy. PTBD was performed with US-guided puncture using a Neff set (Cook Medical system, Bloomington, IN) and a 10F ring biliary catheter was placed on the left side. One month later, the patient presented with blood-mixed pericatheter leak of bile. Ultrasonography revealed a well-defined anechoic lesion in adjacent to the ring biliary catheter showing pulsatile flow on color Doppler. Computed tomography (CT) angiography showed that a well-defined contrast filled outpouching was seen in segment 3 arising from the left hepatic artery measuring 9.2 × 8.2 × 13.3 mm suggestive of pseudoaneurysm. Furthermore, APF was seen as opacification of the left portal vein in arterial phase [Figure 1].

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Figure 1: (a) Axial CT angiography image showing early opacification of segment 3 portal venous branch (thick arrow) and adjacent hepatic artery branch (thin arrow) suggestive of arteriportal fistula. (b) Sagittal reconstructed CT image on arterial phase showing a well-defined contrast filled outpouching in segment 3 arising inferiorly from the left hepatic artery of size (curved arrow) confirming the diagnosis of pseudoaneurysm.

Endovascular coil embolization of the hepatic artery proximal to the pseudoaneurysm was planned. Celiac trunk was catheterized using 5F Simmons catheter (Cook, Indiana, USA) through right femoral artery access. Selective catheterization of the left hepatic artery was done using 2.7F 130 cm microcatheter (Progreat; Terumo Medical, Tokyo). Selective angiograms showed the presence of pseudoaneurysm of segment 3 hepatic arterial branch [Figure 2a] and immediate opacification of portal vein branches [Figure 2b] suggestive of APF. A pair of 2 × 2 0.018 inch Hilal microcoils (Cook, Bloomington, IN) were deployed in segment 3 hepatic artery proximal to the pseudoaneurysm [Figure 2c]. Post-embolization check angiogram of the left hepatic artery did not show any further contrast opacification of portal venous branch [Figure 2d]. Clinically, change in the color of pericatheter leak was noted from serosanguineous to straw colored few hours after the procedure which also showed reduction in volume over 24 h. On 2 weeks follow-up, pericatheter leak had completely subsided with improvement in hemoglobin level.

DISCUSSION

Persistence of sanguineous output through the biliary drainage (hemobilia) catheter should alert the radiologists toward a possibility of arterial injury. Other clinical features include systemic hypotension, perihepatic hematoma, hematemesis, or melena. Risk factors associated with hepatic arterial injury are left-sided PTBD, benign stricture, perihepatic ascites, platelet count $<50,000/\text{mm}^3$, and international normalized ratio >1.5 .^[7]

APF is a rare manifestation of arterial injury with only few case reported in the literature.^[8] Small, peripheral, and intrahepatic fistula without features of portal hypertension, as in the present case, is classified as type 1 APF.^[9] They usually resolve spontaneously and need transarterial embolization if symptomatic or unresolved after a month.

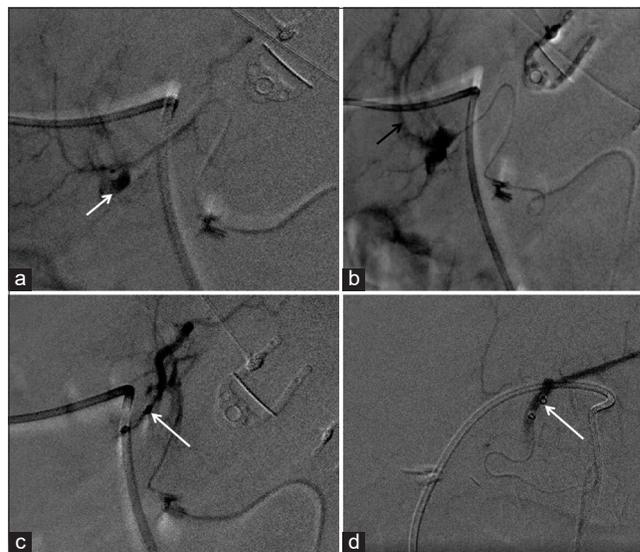


Figure 2: Digital subtraction angiography translateral images (a) Contrast-filled outpouching (white arrow) from segment 3 branch of hepatic artery (pseudoaneurysm), (b) immediate opacification of portal vein (black arrow) suggesting arteriportal fistula, post coiling translateral (c) and anteroposterior (d) run images showing coils (long white arrows in c,d) *in situ* with non opacification of pseudoaneurysm and portal vein.

Large central APFs (type 2) or congenital APF (type 3) should be immediately treated. CT angiography is required for diagnosis as well as to assess hemodynamics, delineate shunt location, and estimate the size of the feeding artery. Early filling of the portal vein on the arterial phase and enlargement of both hepatic arteries and portal veins as a result of high flow in the fistula, portal arterialization, and portal hypertension are suggestive of arteriportal fistula.^[10] For obtaining hemostasis for hepatic arterial injury occurring after biliary interventions, transcatheter embolization has been established as an alternative to surgery.^[11-13] Possible complications after transarterial embolization for hepatic arterial injury include hepatic ischemia, infarction, and abscesses.^[11] Embolization can be done by coil, glue-Lipiodol mixture, and a combination of both or Amplatzer occlusion device for larger lesions.^[12] Choice of the embolic agent depends on the number of feeding artery, accessibility of fistula location, size of the fistula, and flow rate across it.^[14] Coil is the most commonly used embolic agent in small symptomatic fistulae and should be placed as superselectively as possible, particularly in cases of liver transplant where the flow dynamics is reversed. Multiple coils and coils in combination with glue (N-butyl cyanoacrylate) might be needed to achieve better packing and closure of larger fistulae. Coil migration can occur in large fistula (more than 8 mm) with high arteriportal flow rate which might lead to partial or complete portal vein thrombosis. In such circumstances, larger coils (with oversizing around 20–30% as measured

on pre-deployment angiogram) may be used as scaffolding deployed by side branch technique where initial end of the coil should be placed in one of the side branches. This is followed by either packing by smaller coils or administration of glue with or without packing coils.^[15] Mechanical or electrolytic detachable coils can be used for controlled release of coil to avoid coil migration. Amplatzer vascular plugs (St. Jude Medical, Plymouth, MN) or detachable balloon can be used for larger arterioportal fistula using single embolizing device in one step; however, need for relatively straighter course with constant diameter of target vessel and large sheath for delivery restricts its use in many cases, particularly in patients with chronic liver disease.^[16] Since thrombosis by coils and vascular plug depends on coagulation profile of the patient for thrombosis, the use of glue-Lipiodol with or without coils is preferred in patients with coagulopathy. In APE, where flow rate is high, lower cyanoacrylate/Lipiodol ratio of 1:1 or 1:2 may be used to prevent non-target distal penetration.^[17] In cases of multiple small arterial feeders supplying the fistula not amenable for subselective catheterization, injection of collagen-thrombin mixture (D-Stat) can be used to occlude the portal venous outflow of the APE.^[18]

Teaching points

- In case of hemobilia, monitoring of vitals, general condition of the patient, output monitoring (volume and color), and regular follow-up are needed. In cases of unstable vitals, emergent laboratory investigations and evaluation of hepatobiliary axis by ultrasound color Doppler with or without CT angiography might be needed. In cases of persistent hemobilia observed as appearance of clots in drainage catheter, cholangiogram should be done to exclude venous communication and if needed, catheter repositioning
- Possibility of pseudoaneurysm, arterioportal fistula with erosion into catheter tract, and tumor bleed should be considered, if catheter is positioned correctly. Pseudoaneurysm and arterioportal fistula may warrant endovascular embolization by coil, glue-Lipiodol, detachable balloon, or Amplatzer vascular plug as per architecture of the lesion
- Careful pre-procedure evaluation of biliary and vascular anatomy and correction of any underlying coagulopathy is needed to minimize the incidence of such complications.

MCQs

1. In a patient with persistent hemobilia and pericatheter serosanguinous leak a month after PTBD, the sequence of imaging modalities in evaluation of underlying etiology is:

- a. CT angiography
- b. USG and color Doppler > cholangiography > CT angiography
- c. Cholangiography > diagnostic angiography
- d. Diagnostic angiography

Answer Key: b

2. What would be the management in this case had there been arterioportal fistula without hemobilia/pericatheter serosanguinous leak, pseudoaneurysm, or features of portal hypertension?
 - a. Endovascular coil embolization
 - b. Close monitoring, counseling for warning signs, and follow-up
 - c. Prophylactic beta-blockers
 - d. Percutaneous glue-Lipiodol administration

Answer Key: b

3. Best embolic agent for symptomatic small low-flow peripheral arterioportal fistula with single arterial feeder is:
 - a. Glue-Lipiodol
 - b. Coil
 - c. Vascular plug
 - d. Gelfoam

Answer Key: b

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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