Delayed bilhemia complicating percutaneous transhepatic biliary drainage: Successful treatment with primary coil embolization

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ABSTRACT

Bilhemia is very rare and serious complication of percutaneous transhepatic biliary drainage (PBD). Bile leakage occurs into the bloodstream through a fistula between the biliary tree and the hepatic venous system. We report a case of a 45-year-old woman with bilhemia complicated by PBD. She was successfully treated with primary coil embolization of biliovenous fistula tract. In the follow-up, bilirubin values dramatically regressed and returned to its normal limits. Rapid increase in total and direct bilirubin values after PBD without biliary tree dilatation almost always suggest biliovenous fistula. It is more likely that biliovenous fistulas will develop in catheters that are removed before the time of the tract maturation. Symptomatic bilhemia should be treated as soon as possible to prevent major complications like bile pulmonary embolism and biliary sepsis.

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Introduction

Transhepatic biliary drainage (TBD) is a commonly used method in obstructive biliary tract diseases to ensure effective bile drainage [1]. There are several well-known complications of TBD like subcapsular-parenchymal bleeding, hemobilia, pseudoaneurysm, cholangitis, and sepsis [2,3]. Patients without intrahepatic bile duct dilatation had significantly higher complication rates compared to patients with dilated bile ducts [4]. Bilhemia is a rare but serious complication also can be mortal [5,6]. Bile leakage occurs into the bloodstream through a fistula between the biliary tree and the hepatic venous system.

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Case report

A 45-year-old woman with recent hepatic duct injury during laparoscopic cholecystectomy complained of right upper quadrant pain due to biliary leakage into the peritoneal recess. She had just mild jaundice. Total bilirubin was 5.61 mg/dL (normal 0.3-1.2 mg/dL) and direct bilirubin was 3.26 gm/dL (normal 0-0.2 mg/dL). The patient was referred for biliary drainage. Sonographically there was no dilatation of biliary system and no sign of chronic liver disease. After successful fluoroscopy-guided biliary drainage procedure with successful right lobe access, bilirubin values almost regressed to normal limits (total was 1.73 mg/dL and direct was 0.80 mg/dL). Unfortunately the drainage catheter was displaced spontaneously during patient movement. Bilirubin values peaked at the same day (total bilirubin 13.12 mg/dL, direct bilirubin 8.5 mg/dL) and increased in the following days (total bilirubin 35.08 mg/dL and direct bilirubin 18.80 mg/dL). Despite this marked increase in bilirubin values, there was no dilatation of biliary system and perihepatic and intraabdominal biliary leakage. This suggested that there could be a high-volume biliovenous fistula. Biliary drainage was difficult due to lack of biliary dilatation.

After several unsuccessful attempts, puncture of the right lobar bile duct was performed. Multiple cholangiographies showed a large fistula with high flow between the right hepatic vein and the right anterior lobar duct and also total occlusion of common bile duct due to iatrogenic injury (Fig. 1).

Fistula was superselectively catheterized with 2.7 F microcatheter (Progreat, Terumo, Tokyo, Japan) (Fig. 2) and primary coil embolization was performed with pushable coils (Azur, Terumo Medical, Somerset, NJ) (Fig. 3). Subsequent cholangiogram showed total occlusion of fistula (Fig. 3). An external drainage catheter was inserted after fistula closure. A dramatic decrease in bilirubin values was observed in hours (Fig. 4). To our knowledge, there is no other case in the
literature like ours that bilhemia occurs after displacement of biliary drainage catheter before tract maturation.

**Discussion**

Biliary system and vascular structures are adjacent structures. There may be some communications between these systems mainly due to trauma and interventional procedures. Biliovenous fistulae due to liver biopsies have been reported, with a frequency between 0.02% and 0.1%. The most common finding in biliovenous fistula during TBD is hemobilia with a frequency between 7% [7] and 13% [8]. Bilhemia is much less common than hemobilia. Normally, bile duct pressure is lower than venous and also arterial pressure. Bilhemia is unusual presentation which occurs when the biliary system pressure exceeds the venous system pressure. There is any data on the frequency of bilhemia in the literature. The aim should be to prevent the passage of the bile into the systemic circulation. To provide this, sphincterotomy and stent placement in distal pathologies or externally biliary drainage in proximal pathologies is treatment options. The majority of biliovenous fistulae close spontaneously. Optimal time for tract maturation is not
well established. Hatjidakis and et al reported that 2 weeks suffices for the majority of patients used transhepatic access to develop a mature tract [9]. In our opinion, tract maturation is very important to reduce biliovenous fistula. Successful primary coil embolization has been presented in the failure of spontaneous closure in this case report.

In conclusion, biliovenous fistula leads to bilhemia is very rare complication of percutaneous transhepatic biliary drainage. Fistula can be silent in the presence of a catheter. Removal or displacement of drainage catheter before tract maturation can lead severe bilhemia as in our case. Rapid increase in total and direct bilirubin values after percutaneous transhepatic biliary drainage without biliary tree dilatation almost always suggest fistula. Symptomatic bilhemia should be treated as soon as possible to prevent major complications. The first treatment option is to put external drainage catheter to keep the bile out of circulation. In case of showing fistula, tract occlusion should be tried. Primary coil embolization of fistula is an effective treatment option.

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