



Arterio-Biliary Fistula as a Rare Life-Threatening Complication of Transjugular Intrahepatic Portosystemic Shunt: A Case Report

경정맥 간내 문맥 정맥 단락술 후 드물게 발생하는 동맥-담관루: 증례 보고

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A 46-year-old male with alcoholic liver cirrhosis underwent a transjugular intrahepatic portosystemic shunt (TIPS) for refractory ascites. On the 9th day after the procedure, he presented with melena and decreasing hemoglobin levels. Hemobilia due to fistula formation between the right intrahepatic bile duct and right hepatic artery was suspected on computed tomography. Angiography revealed a fistula of the small branches of the hepatic segmental arteries, and right intrahepatic bile duct was confirmed; embolization was successfully performed with a coil for the eighth segmental hepatic artery, a glue-lipiodol mixture for the fifth segmental hepatic artery, and gelfoam slurry for the right anterior hepatic artery. However, 2 days after embolization, the patient died owing to aggravated disseminated intravascular coagulopathy. When gastrointestinal bleeding occurs after TIPS, careful evaluation is immediately required, and hemobilia should be considered.

Index terms Portosystemic Shunt; Portasystemic Shunt, Transjugular Intrahepatic; Hemobilia; Biliary Fistula

INTRODUCTION

Transjugular intrahepatic portosystemic shunt (TIPS) involves the creation of a con-

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duit from the hepatic vein to the portal vein. It is performed to reduce portal hypertension that is not responsive to medical or endoscopic therapy (1, 2). Indications for TIPS include variceal bleeding, refractory ascites, hepatorenal syndrome, gastric antral vascular ectasia, Budd Chiari syndrome, and refractory hepatic hydrothorax (3). Although TIPS is effective in reducing portal hypertension, the mortality rate after TIPS has been reported to range from 3% to 33% (1, 4). Potential complications of TIPS are acute liver failure, hepatic encephalopathy, hemorrhage, biliary injury, injury to surrounding organs, TIPS thrombosis, TIPS dysfunction, and stent migration (2). Arterio-biliary fistula leading to persistent bleeding, which requires embolization, has rarely been reported as a complication after TIPS (5, 6).

Herein, we report a case involving arterio-biliary fistula as a complication after TIPS, which was performed to control refractory ascites in a patient with alcoholic liver cirrhosis. We focused on the clinical course and angiographic findings. Successful embolization was performed on the hepatic artery for an arterio-biliary fistula; however, the patient died due to ongoing disseminated intravascular coagulopathy (DIC). Case reports on arterio-biliary fistula after TIPS are extremely rare in the English literature; therefore, we report the case we encountered.

CASE REPORT

A 46-year-old male with alcoholic liver cirrhosis visited our hospital for evaluation of dyspnea and abdominal distension for one month. He was prescribed diuretics at the local clinic for his symptoms; however, his symptoms did not improve. In laboratory tests, his hemoglobin (Hb) was 11.8 g/dL (normal range: 14–17 g/dL), total bilirubin was 6.65 mg/dL (normal range: 0.3–1.2 mg/dL), prothrombin time-international normalized ratio (PT-INR) was 1.36 (normal range: 0.8–1.2), and serum albumin was 2.6 g/dL (normal range: 4.0–5.3 g/dL). The Child Pugh score was C11, and the model for end-stage liver disease (MELD) score was 17. Initial abdominal CT showed cirrhotic features of the liver, and there were multiple collateral vessel including esophageal, paraesophageal, and paraumbilical varices and gastrosplenic shunt, and a large amount of ascites in the abdominal cavity. He was treated with furosemide (40 mg) and spironolactone (100 mg) once a day, and repeated paracentesis for a large volume of ascites. For the first two weeks after hospitalization, 2.5 L of ascites was drained per week, and from the 15th day after hospitalization, 1 L of ascites was drained every day. However, even on the 20th day of hospitalization, the amount of ascites did not decrease significantly, and the patient's symptoms of dyspnea and abdominal distension persisted. TIPS was planned as a further treatment method for intractable ascites.

After the selection of the right hepatic vein, the right posterior portal vein was punctured using a Colapinto needle (Cook Medical, Bloomington, IN, USA) on the third attempt with targeting guidewire at the right hepatic artery under US guidance. A portosystemic shunt was ballooned with 4 mm × 4 cm and 6 mm × 10 cm mustang balloon catheters (Boston Scientific, Marlborough, MA, USA). Subsequently, a 10 mm × 10 cm Covera Plus self-expandable stent graft (BARD, Tempe, AZ, USA) was placed between the right hepatic vein and the right posterior portal vein. After stent insertion, the portocaval pressure gradient decreased from 41 to 20 mm Hg. Final venography revealed patent shunt flow and no findings suggesting

complications (Fig. 1A).

After the procedure, there were no immediate complications, but PT-INR and MELD scores were aggravated to 2.01 and 22, respectively. On the 6th day after the procedure, the patient complained of melena. The source of bleeding was unclear during gastroscopy and colonoscopy. Doppler ultrasonography was performed to evaluate TIPS patency. The shunt was patent, and there were no other specific findings. One unit of red blood cells (RBCs) was transfused every day; however, the Hb level continuously decreased (from 8.3 g/dL to 7.3 g/dL, normal range: 12–17 g/dL) until the 8th day after the procedure.

On the 9th day after the procedure, massive hematochezia occurred, and hypotension (systolic and diastolic blood pressure: 82/48 mm Hg) was observed. In laboratory tests, the Hb level was 3.0 g/dL and PT-INR was 5.15. CT angiography was performed to evaluate the bleeding focus. Massive contrast extravasation to the common bile duct and second part of the duodenum was noted, and life threatening fistula formation between the right intrahepatic duct and the right hepatic artery was highly suspected on CT angiography (Fig. 1B). Emergency embolization for a suspected arterio-biliary fistula was requested.

Hepatic arteriography in the early arterial phase showed contrast extravasation to the common bile duct, confirming an arterio-biliary fistula, but a direct fistula between the right hepatic artery and the right intrahepatic bile duct was not definite. Selective arteriography revealed active bleeding at multiple small branches of the eighth segmental hepatic artery and multiple small branches of the fifth segmental hepatic artery, leading to an arterio-biliary fistula. Coil embolization of the eighth segmental hepatic artery using 4 mm × 15 cm and 5 mm × 15 cm interlocking detachable coils (Boston Scientific, Marlborough, MA, USA), embolization of the fifth segmental hepatic artery using glue (N-butyl cyanoacrylate; Histoacryl, B. Braun, Melsungen, Germany)-Lipiodol (Guerbet, Aulnay-sous-Bois, France) 1:3 mixture, and embolization of the right anterior hepatic artery using gelfoam slurry (EG gel S PLUS 350–560 μm, Engain, Seungnam, Korea) were performed and the arterio-biliary fistula disap-

Fig. 1. A 46-year-old male patient with arterio-biliary fistula after transjugular intrahepatic portosystemic shunt.

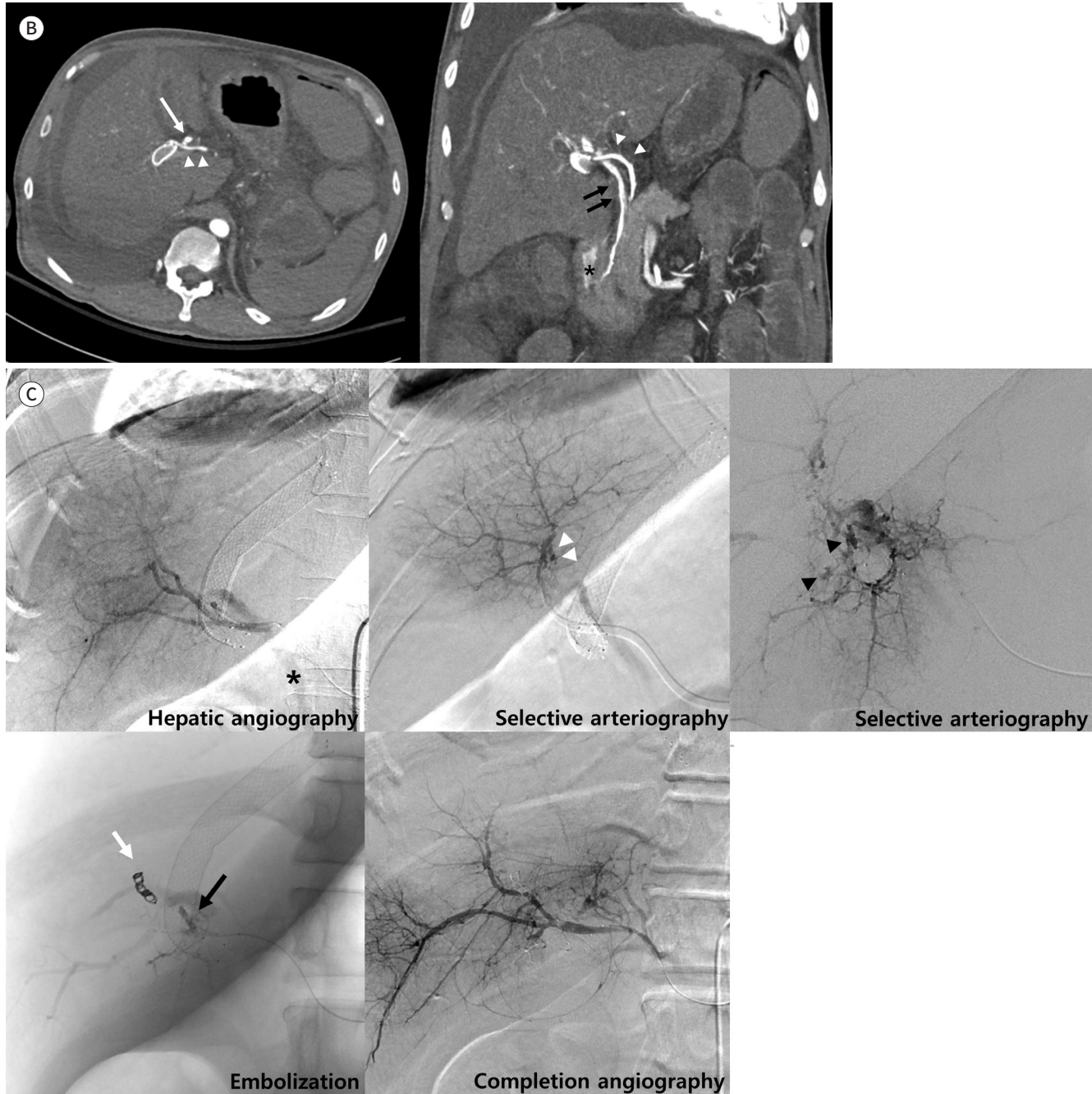
A. Under US guidance, the right portal vein was punctured using a Colapinto needle (white arrowheads) in the right hepatic vein. Fluoroscopy shows a Colapinto needle with a transjugular intrahepatic portosystemic shunt sheath (black arrowheads) in the right hepatic vein and a guiding microcatheter (white arrow) in the right hepatic artery. The placement of a 10 mm × 10 cm covered stent (Covera Plus) (asterisk) was performed at the shunt, showing patent shunt flow.



Fig. 1. A 46-year-old male patient with arterio-biliary fistula after transjugular intrahepatic portosystemic shunt.

B. Arterial phase of CT angiogram shows possibility of fistula between the right hepatic artery (arrowheads) and right intrahepatic bile duct (white arrow) in axial image and massive contrast media extravasation from the right hepatic artery (arrowheads) to the common bile duct (black arrows) and the second part of the duodenum (asterisk) in coronal image.

C. Hepatic arteriography in the early arterial phase shows contrast extravasation to the common bile duct (asterisk) confirming an arterio-biliary fistula, but a direct fistula between the right hepatic artery and the right intrahepatic bile duct was not observed. Selective arteriography reveals active bleeding at multiple small branches of the eighth segmental hepatic artery (white arrowheads) and multiple small branches of the fifth segmental hepatic artery (black arrowheads), leading to an arterio-biliary fistula. Coil embolization of the eighth segmental hepatic artery using an interlocking detachable coil (white arrow) and embolization of the fifth segmental hepatic artery using a glue-lipiodol mixture (black arrow) was performed, and the arterio-biliary fistula disappeared after the procedure.



peared after the procedure (Fig. 1C).

Unfortunately, massive transfusion was administrated after embolization, and the follow-up Hb and PT-INR were 5.0 g/dL and 8.1, respectively. The patient died 11 days after the TIPS procedure because of ongoing DIC.

Approval from the hospital's Institutional Review Board was obtained, and the requirement for obtaining patient consent was waived owing to its retrospective observational nature (IRB No. 2021-03-007).

DISCUSSION

TIPS is known to be a safe and effective procedure; however, it can result in mortality, as in our case. Liver and/or multiorgan failure (54.5%), septic shock (13.6%), and gastrointestinal bleeding (4.5%) have been reported as causes of mortality after TIPS (4). The reported factors that can predict early mortality after TIPS include older age (> 60 years), coagulopathy (prothrombin time greater than 17 seconds), hyperbilirubinemia (total bilirubin > 3 mg/dL), and renal insufficiency (serum creatinine > 1.7 mg/dL) (1, 4). In cases involving MELD scores of 18 or more, a significantly low three-month survival rate was reported (7). In general, elective TIPS should be avoided in patients with MELD scores \geq 25, and alternative treatment strategies should be considered (7). In this regard, our patient had a marginal MELD score of 17 and PT of 17.1 seconds.

Early complications that can occur within a month after TIPS include hematoma, acute hepatic encephalopathy, acute hepatic failure, biliary complications, migration of the stent, and early acute occlusion (2). Even 3 months after TIPS, delayed complications such as recurrent symptoms due to portal hypertension, infection, and hernia incarceration may occur (2). Cases involving arterio-biliary fistula after TIPS have rarely been reported (5, 6). Kaswala et al. (6) reported a case involving successful embolization of an arterio-biliary fistula after TIPS using gelfoam on the right hepatic artery. Menzel et al. (5) reported an arterio-biliary fistula case-related stent dislocation after TIPS, for which successful embolization was performed using a coil on the branch of the accessory right hepatic artery where the fistula was formed.

Embolization is the treatment of choice for arterio-biliary fistulas after TIPS (5, 8). When TIPS is performed, the portal vein flow diverts from hepatic portal perfusion into the systemic circulation. Therefore, arterial embolization after TIPS in a highly selective manner is required for hepatic perfusion (9). Stefańczyk et al. (8) suggested soft and biodegradable materials (thrombin or histoacrylic glue) as an optimal material for embolization because of concerns about coil migration or chronic infection in the coil.

In this case, TIPS was successfully performed without demonstrable injury to the hepatic artery or biliary system during the procedure. The patient was symptom-free for six days after the TIPS procedure. According to the literature, accidental puncture of the bile duct or hepatic artery could be observed in up to 26% of TIPS procedures, and intrahepatic bile duct injury may cause hemobilia, but may be asymptomatic (10). A subsequent inflammatory reaction after TIPS might have resulted in erosion of the bile duct and hepatic artery (5). After TIPS, the patient's liver function and coagulopathy continued to deteriorate, resulting in massive hemobilia, arterio-biliary fistula, and DIC. If there are symptoms of gastrointestinal

bleeding after TIPS, evaluation using endoscopy and CT angiography should be performed at an appropriate time, and treatment through embolization should be initiated especially in patients with underlying liver cirrhosis accompanied by coagulopathy.

In conclusion, arterio-biliary fistula is a life-threatening complication that can occur after TIPS. It is necessary to evaluate the presence or absence of coagulopathy and preserve hepatic function before performing TIPS. If there is gastrointestinal bleeding, careful evaluation should be performed, considering the possibility of iatrogenic hemobilia.

Author Contributions

Conceptualization, K.L.M., P.J.W.; data curation, K.M., H.H.I., W.J.Y.; investigation, K.J.S., K.L.M.; methodology, K.L.M., K.H.M.; project administration, K.L.M., K.H.M.; resources, K.J.S., K.L.M.; supervision, K.L.M.; visualization, K.L.M., K.H.M.; writing—original draft, K.J.S., K.L.M.; and writing—review & editing, K.L.M., P.J.W.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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경경정맥 간내 문맥 정맥 단락술 후 드물게 발생하는 동맥-담관루: 증례 보고

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46세 남자 환자가 알코올성 간경화와 동반된 지속되는 복수로 인해 경경정맥 간내 문맥 정맥 단락술을 시행 받았다. 시술 후 9일째 흑색변 및 헤모글로빈 수치 감소로 혈관조영 컴퓨터 전산화단층촬영을 시행하였으며 혈액담즙증 및 우간동맥과 우간내담관 사이의 동맥-담관루가 의심되었다. 혈관조영술에서 분절간동맥의 작은 분지와 우간내담관의 동맥-담관루가 확인되었다. 분절8 간동맥에 대해 코일로, 분절5 간동맥에 대해 글루-리피오돌 혼합물로, 우전 간동맥에 대해 젤폼으로 색전술을 시행하였고, 시술 후 혈관조영술에서 동맥-담관루는 소실되었다. 그러나 색전술 시행 2일 후 환자는 진행되는 파종성 혈관 내 응고로 인해 사망하였다. 경경정맥 간내 문맥 정맥 단락술 이후 위장관 출혈이 있을 경우 혈액담즙증의 가능성을 반드시 고려하여 가능한 빠른 시기에 적절한 조치가 필요할 것이다.

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