



A Case of Biliary Cast Syndrome After Endoscopic and Percutaneous Management of Common Bile Duct Stone

총담관 결석 환자에서 내시경적, 경피적 담관 시술 후 발생한 담관 원주 증후군

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Biliary cast syndrome is an unusual complication in patients who have previously undergone liver transplantation. It occurs in approximately 5%–18% of such patients. Rare cases of biliary cast syndrome in patients without liver transplants have also been reported. The pathogenesis of biliary cast has not been clearly identified, although etiologic factors including post-transplant bile duct damage, ischemia, biliary infection, and presence of a post-operative biliary drainage tube have been proposed. Here we present a case of biliary cast that developed in a 49-year-old male who underwent a non-liver surgery after endoscopic and percutaneous management of common bile duct stone.

Index terms Bile Ducts; Cholelithiasis; ERCP; PTBD

INTRODUCTION

Biliary cast syndrome, an unusual complication in patients who have undergone liver transplantation, is defined as having sludge forming a circumference along the bile duct (1). It is known to occur in about 5%–18% of patients who have received orthotopic liver transplantation (2). Rare cases of biliary cast syndrome in patients without liver transplants have also been reported (3, 4). The exact pathogenesis of biliary cast is unknown. It is more likely to occur in situations of post-transplant bile duct damage, ischemia, biliary infection, and post-operative biliary drainage tube (5, 6). Although treatment of choice for biliary cast has not been established yet, treatments with surgical endoscopic retrograde cholangiopancreatography (ERCP) and percutaneous cholangiography with cast extraction have been reported (4, 5). Here we present a case of biliary

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cast developed in a 49-year-old male with a non-liver surgery after endoscopic and percutaneous managements of common bile duct (CBD) stone with a review of the literature.

CASE REPORT

A 49-year-old male was admitted to the local emergency room (ER) with right upper abdominal pain that occurred after excessive drinking a day earlier. He was a heavy alcoholic. There was no family history. Enhanced abdominal CT scan showed a distal CBD stone and acute cholecystitis with multiple gallbladder stones. Endoscopic biliary sphincterotomy was performed followed by stone removal (Fig. 1A). After that, his jaundice persisted. His serum bilirubin and alkaline phosphatase levels continued to increase. Follow-up CT scan showed persist dilated CBD, dilated both intrahepatic bile ducts, and thrombus in the left portal vein (Fig. 1B). After 3 days, percutaneous transhepatic biliary drainage (PTBD) was performed and drainage catheter interposition was performed. Follow-up cholangiography revealed a long intraluminal filling defect (Fig. 1C). A large amount of stone debris was removed using a stone basket. After that, about a month after the initial symptom, the patient was transferred to the emergency center of our hospital. At the time of the visit to the ER, vital signs were normal and jaundice was observed in the sclera and skin. Laboratory data showed bilirubin of 6.22 mg/dL (reference range: 0.2–1.3 mg/dL), alanine aminotransferase of 107 IU/L (reference range: 13–69 IU/L), aspartate aminotransferase of 101 IU/L (reference range: 15–46 IU/L), alkaline phosphatase of 613 IU/L (reference range: 42–140 IU/L), γ -glutamyl transferase of 261 IU/L (reference range: 5–55 IU/L). Hepatitis c was confirmed in the patient with viral test and hepatitis C virus RNA positive were 4462906 IU/mL in quantitative tests (real-time quantitative polymerase chain reaction).

Three days after hospitalization, a follow-up cholangiography was performed through an 8F drainage catheter that was previously inserted. On cholangiography, a smooth cast like material, filling of the extrahepatic duct extending into both intrahepatic ducts, and marked ductal irregularity were identified. The patient was diagnosed with biliary cast syndrome and the cast was extracted using a basket through the PTBD catheter (Fig. 1D). The cast was composed of inflammatory exudates with pigmented material (Fig. 1E). The pathology report was negative for malignancy. The patient was discharged with PTBD after conservative treatment.

After a month, the patient visited the emergency center again for abdominal pain and melena. He was diagnosed with cholecystitis and cystic artery pseudoaneurysm (Fig. 1F). He was discharged after cholecystectomy and CBD repair. Over the next two years, there had been intermittent episodes of abdominal pain and general weakness. Each time he was admitted, he received ERCP or PTBD for recurrent biliary cast. Finally, he received liver transplantation. The patient was symptom-free at one-month follow-up.

This case report was exempt from the institutional review board standards (IRB No. KUGH 2021-06-017).

DISCUSSION

Biliary cast syndrome is a rare condition, that is almost always related to liver transplanta-

Fig. 1. A 49-year-old male with biliary cast syndrome after endoscopic and percutaneous management of CBD stones.

A. Abdominal CT and endoscopic retrograde cholangiopancreatography show dilatation of the bile duct with CBD stones (arrows).

B. Pre-contrast (left) and portal phase (right) CT images show portal vein thrombosis and periportal edema in the liver.

CBD = common bile duct



tion. Cast can be present inside or outside the liver. It can cause cholangitis due to bile duct obstruction, bile duct stenosis, bile duct expansion, and micro abscess in the liver (7). Biliary cast syndrome refers to the development of a dark stone like material within the biliary ductal system that takes the physical shape of the bile duct. Biliary cast syndrome is known as a complication of orthotopic livers transplantation. It is rare in patients without receiving liver transplantation. Such rare cases have been reported worldwide (3, 4, 8). Chemical analysis of the cast has shown that its primary compositional element is bilirubin along with collagen, bile acid, and/or cholesterol (1, 9). The mechanism of development of a biliary cast is not clearly understood yet. Etiological mechanisms including acute cellular rejection, prolongation of cold ischemic time, infection, biliary drainage tubes, and biliary obstruction have been proposed.

The diagnosis of biliary cast syndrome is usually based on elevated liver-associated enzyme, showing biliary dilatation and obstruction on imaging examination such as ERCP, PTBD, and CT (6, 8). The diagnosis should be suspected when jaundice or cholangitis is asso-

Fig. 1. A 49-year-old male with biliary cast syndrome after endoscopic and percutaneous management of CBD stones.

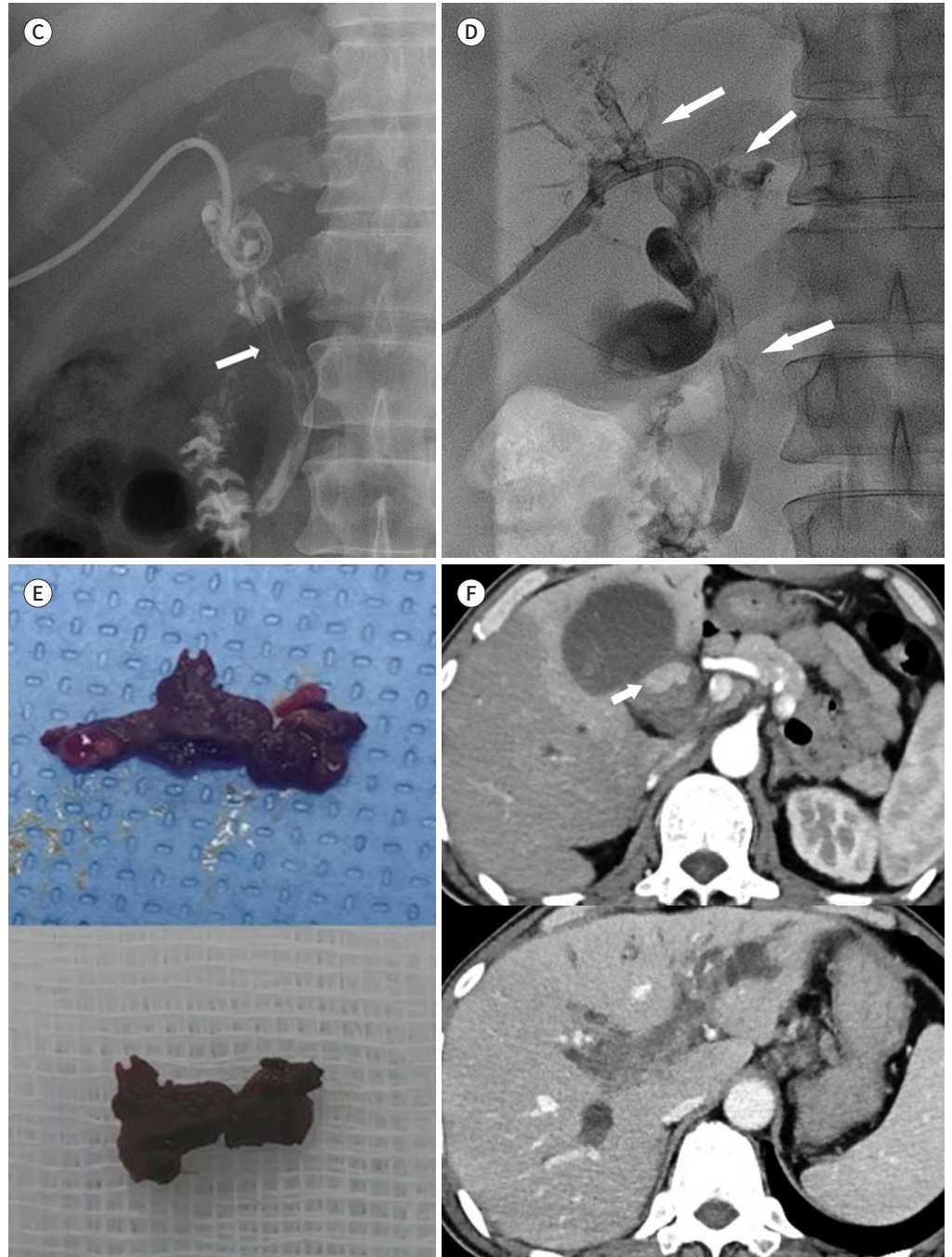
C. Follow-up cholangiography shows ductal irregularity of the CBD with long intraluminal filling defect (arrow).

D. Cholangiography through percutaneous transhepatic biliary drainage show large elongated filling defect (arrows) of both intrahepatic and CBDs.

E. Fragments of the biliary cast after removal are shown.

F. Contrast-enhanced abdominal CT shows a pseudoaneurysm (arrow) in the distended gallbladder and progression of dilation of intrahepatic ducts with hemobilia.

CBD = common bile duct



ciated with dilated ducts on abdominal imaging studies. Imaging studies may present with cast like filling defect in dilated bile duct.

There is no formal standard management protocol to remove biliary casts, although surgery and endoscopic or percutaneous procedures have been attempted. In the case of endoscopic or transhepatic procedures, it is often necessary to perform multiple repetitive procedures. Successful treatment involved cholesterol clearance of biliary casts, and there was no clinical, laboratory or radiographic evidence of recurrent cast (5, 10).

In the present case, a patient with hepatitis C virus and a history of heavy alcohol drinking was diagnosed with bile duct stones due to upper right abdominal pain and jaundice. It is difficult to explain the occurrence of biliary cast syndrome due to causative factors such as ischemia of bile duct. Portal vein thrombosis was observed in this patient. Portal vein thrombosis might trigger bile duct ischemia and lead to extrahepatic cast formation and bile duct stricture. A few domestic cases of biliary cast syndrome without liver transplantation such as our case have been reported after ERCP, after cardiopulmonary resuscitation and head trauma patients (3, 4). ERCP or PTBD should be the primary method for diagnosis and treatment of biliary cast syndrome. However, liver transplant surgery might be considered to be the final treatment method as in the present patient. In conclusion, our case is a good example of biliary cast syndrome, showing that endoscopic and percutaneous managements can induce ischemic change, resulting in cast formation.

Author Contributions

Conceptualization, all authors; data curation, P.J.C.; formal analysis, all authors; investigation, P.J.C.; methodology, P.J.G.; project administration, P.J.G.; resources, all authors; supervision, P.J.G.; validation, P.J.G.; visualization, P.J.C.; writing—original draft, P.J.C.; and writing—review & editing, P.J.G.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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총담관 결석 환자에서 내시경적, 경피적 담관 시술 후 발생한 담관 원주 증후군

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담관 원주 증후군은 간이식을 받은 환자들에게서 발생하는 흔치 않은 합병증이다. 간이식을 받은 환자의 약 5%~18%에서 발생한다. 간이식을 받지 않은 환자에게서 드물게 담관 원주 증후군이 발생하는 것으로 보고되었다. 이식 후 담관 손상, 허혈, 담도 감염, 수술 후 거치된 담도 배액관 등이 병리요인으로 제안되었으나 담도 원주의 병리현상은 아직 명확히 규명되지 않았다. 이에 저자들은 간이식을 받지 않은 49세 남성에게서 총담관 결석에 대해 내시경과 경피적 담관 조영 시술 후에 발생한 담관 원주 증후군의 사례를 소개한다.

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