## **ORIGINAL ARTICLE**

## Biliary cast syndrome in Roux-en-Y hepaticojejunostomy anatomy



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Biliary cast syndrome represents a rare adverse event after liver transplantation, reported to occur in 4% to 18% of transplant patients. Biliary casts are defined as black, hardened material molded to the bile ducts, formed from fragments of biliary epithelium mixed with bile components. Although the pathogenesis is not clearly understood, biliary casts are believed to be the result of mucosal necrosis following ischemia. This may lead to additional adverse events including biliary obstruction causing jaundice and cholangitis. Biliary casts may be defined on MRCP, where it can be seen as white hyperintense material in the bile ducts on T<sub>1</sub>-weighted sequences and confirmed during ERCP.

We describe a case of a 63-year-old woman with metabolic dysfunction-associated steatohepatitis cirrhosis who underwent an orthotopic liver transplantation from a donation after circulatory death. Her postoperative course was complicated by a bile leak with dehiscence of the biliary anastomosis (Fig. 1), necessitating return to the operating room with creation of a Roux-en-Y hepaticojejunostomy 12 days after her transplantation. Two months later, the patient was found to have elevated liver function tests, with a total bilirubin of 2.6 mg/dL and alkaline phosphatase of 374 U/L. She underwent a single-balloon enteroscopyassisted ERCP that revealed diffuse irregularities of the common hepatic duct, left main hepatic duct, right main hepatic duct, as well as secondary intrahepatic branches (Fig. 2; Video 1, available online at www.videogie.org). The biliary tree was swept with an 8.5-mm and 11.5-mm balloon with removal of sludge. After sweep and dilation of the left and right hepatic ducts with a 6-mm balloon

dilator, two 7F stents were placed into the left and right hepatic ducts, respectively. The patient had subsequent improvement in her liver enzymes tests.

Two months later, a repeat ERCP was performed, with the previously placed stents seen at the hepaticojejunostomy anastomosis. After removal of the first stent, a grasping forceps was used to remove what was thought to be the second biliary stent but was found to be an extremely large biliary cast (Figs. 3-5; Video 1) measuring 6 cm in length. Upon cast removal, there was evidence of a widely patent hepaticojejunostomy allowing for direct insertion of the enteroscope into the hepatic duct (Fig. 6; Video 1). Cholangiogram following balloon sweep revealed a severely dilated common hepatic duct, left main hepatic duct, and right main hepatic duct. The irregularities seen on prior cholangiogram had resolved with removal of the biliary cast.

Biliary cast syndrome following liver transplantation is associated with increased morbidity and mortality as well as graft rejection. As such, it is important to treat biliary casts if suspected. Paik et al reported successful removal of casts in 95% of patients; however, relapse occurred in over half of these patients. Persistent biliary strictures have also been associated with relapse frequency. 1,4

Treatment of biliary cast syndrome is primarily with ERCP with biliary sphincterotomy and balloon extraction of the cast.<sup>5</sup> In this case, the Roux-en-Y hepaticojejunostomy anatomy necessitated the use of a single-balloon enteroscope (with the balloon overtube) with which endoscopic techniques and devices are significantly limited. Fortunately, the cast was able to be grasped

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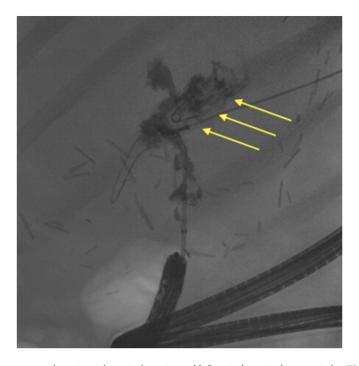
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Figure 1. Cholangioscopy images of the biliary anastomosis on index ERCP, with evidence of dehiscence.



**Figure 2.** Diffuse irregularities of the common hepatic, right main hepatic, and left main hepatic ducts on index ERCP during balloon occlusion cholangiogram (*yellow arrows*).

with a forceps and removed in its entirety. In cases in which ERCP fails to remove the cast, removal may be attempted percutaneously. Due to the high rate of relapse in these patients, multiple ERCP procedures may also be needed for successful treatment of biliary casts. Gor et al reported a median number of 3 procedures for successful endoscopic treatment. Finally, when endoscopic or percutaneous interventions fail, repeat liver transplant may be required.

## DISCLOSURE

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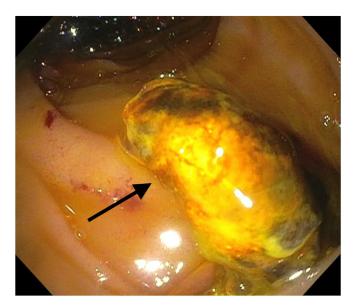


Figure 3. Biliary cast initially thought to be a plastic stent protruding from the hepaticojejunostomy (black arrow).



Figure 5. Biliary cast upon removal.



Figure 4. Large biliary cast removed with a grasping forceps.



Figure 6. Widely patent hepaticojeunostomy after biliary cast removal.

## REFERENCES

- 1. Gor NV, Levy RM, Ahn J, et al. Biliary cast syndrome following liver transplantation: predictive factors and clinical outcomes. Liver Transpl 2008;14:
- 2. Lemmers A, Pezzullo M, Hadefi A, et al. Biliary cast syndrome after liver transplantation: a cholangiographic evolution study. J Gastroenterol Hepatol 2021;36:1366-77.
- 3. Mazzarelli C, Vigano R, Vangeli M, et al. An atypical presentation of biliary cast syndrome (BCS). Dig Liver Dis 2021;53:1050.
- 4. Paik WH, Lee SH, Ryu JK, et al. Long-term clinical outcomes of biliary cast syndrome in liver transplant recipients. Liver Transpl 2013;19: 275-82.
- 5. Pfau PR, Kochman ML, Lewis JD, et al. Endoscopic management of postoperative biliary complications in orthotopic liver transplantation. Gastrointest Endosc 2000;52:55-63.