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Single-session percutaneous alcohol sclerotherapy for the treatment of isolated draining intrahepatic biliary duct in a five-month-old liver transplant recipient

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ABSTRACT
The present paper describes the first reported successful use of a single session of percutaneous alcohol sclerotherapy for the treatment of an excluded segmental biliary duct in a five-month-old left lateral liver recipient. A concomitant stricture of the hepaticojejunostomy was also successfully treated using standard percutaneous balloon dilatation. The patient has remained in good general condition for the next 26 months.

Key words: • *interventional radiology* • *complication* • *stenosis* • *liver transplantation*

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Liver transplantation has become an accepted treatment for children with end-stage liver disease. Biliary complications occur in 20%–40% of patients after pediatric liver transplantation (1, 2). Frequent complications include biliary stricture, bile leakage, biliary stones, and biloma. An isolated biliary duct is rare and may be found in cases of arterial insufficiency with occlusive intrahepatic biliary strictures, or may be the result of accidental surgical ligation in split liver recipients (missing bile ducts). The present paper describes the first reported successful use of a single session of percutaneous alcohol sclerotherapy for the treatment of an isolated draining intrahepatic biliary duct.

Case report

A five-month-old child affected by end stage liver disease resulting from neonatal sclerosing cholangitis received an AB0-compatible orthotopic liver transplant in our institution, using a deceased donor split liver graft of liver segments II and III. The graft was implanted using the piggy-back technique. One bilioenteric anastomosis was performed. The early postoperative course was uneventful with good graft Doppler ultrasonography findings. After two weeks, we observed a slow increase in the level of serum enzymes associated with cholestasis: aspartate transaminase, 133 U/L; alanine transaminase, 137 U/L; γ-glutamyl transpeptidase, 703 U/L; total serum bilirubin, 1.84 mg/dL; direct bilirubin, 1.21 mg/dL; and alkaline phosphatase, 1426 U/L. The liver ultrasonography showed minimal biliary tree dilatation in segment II with normal Doppler findings. A percutaneous liver biopsy showed mechanical cholestasis and no evidence of rejection. Percutaneous transhepatic cholangiography was performed and, with ultrasonographic guidance, a 20 G needle was advanced into the segment II bile duct. The cholangiogram showed the segment II the bile duct; however, the segment III bile duct and bowel loop were not detected (Fig. 1). The segment II bile duct was catheterized, but we were not able to obtain the bowel loop using a hydrophilic guide wire. An external catheter was inserted and left to external gravity drainage.

Despite the presence of the external catheter in the bile duct, we did not observe a significant improvement in liver function tests during the following two weeks. A second cholangiography was performed after a period of external decompression to reduce possible inflammation. However, we were still unable to visualize and catheterize the bowel loop, and the segment III bile duct remained undetected. An external catheter was inserted again and left to external gravity drainage. We then considered an invasive percutaneous recanalization of the anastomosis. We performed a computed tomography (CT) scan with multiplanar reconstruction to precisely locate the adherence of the bowel loop with the liver surface and to evaluate the distance between the distal tip

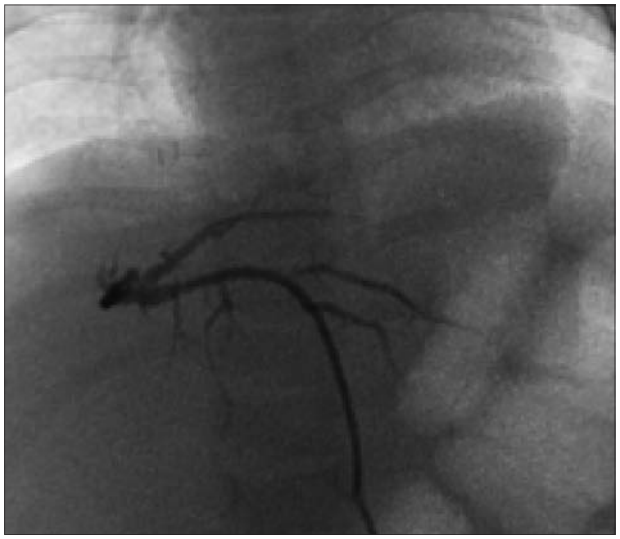

Figure 1. A percutaneous transhepatic cholangiogram shows the segment II bile duct. The segment III bile duct and bowel loop were not detected. An external catheter was inserted.

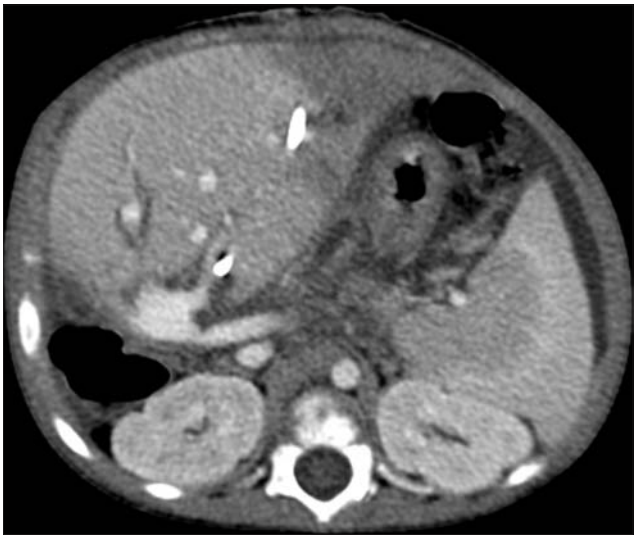

Figure 2. A multidetector CT scan was obtained during the portal venous phase. The axial image shows a significant distance between the distal tip of the external biliary catheter and the bowel loop at the hepatic hilum.

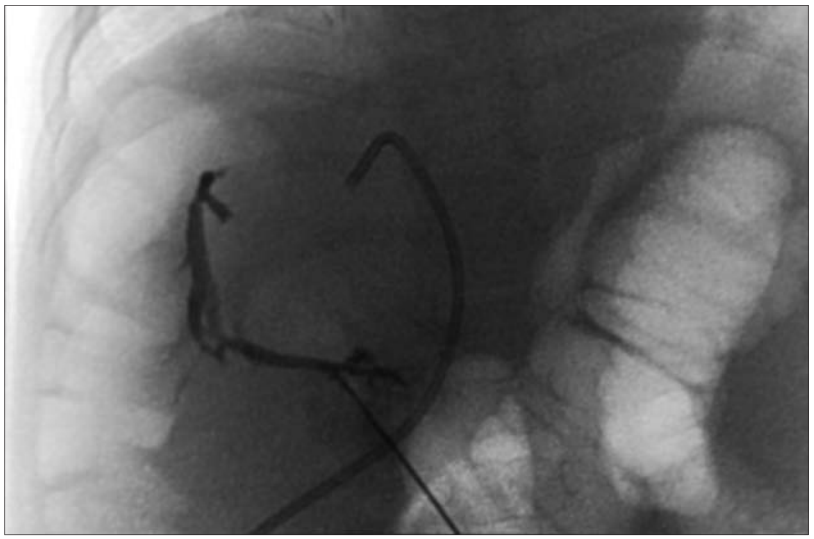

Figure 3. A percutaneous transhepatic cholangiogram of the segment III bile duct reveals a severe focal anastomotic stricture. The stricture was crossed, and an external/internal biliary catheter was inserted.

of the biliary catheter and the bowel loop in order to visualize the potential puncture site. The CT scan showed a significant distance between the tip of the catheter and the bowel loop (Fig. 2), excluding any possible percutaneous recanalization. The minimal dilatation of the segment III bile duct was also appreciable. The possibility that a bile duct might have been missed during harvesting of the graft was raised.

Percutaneous transhepatic cholangiography of the segment III bile duct was performed. The cholangiogram revealed a severe stricture at the hepaticojejunostomy site (Fig. 3). The stricture was crossed, and a transanastomotic biliary catheter was inserted. The liver function tests showed rapid improvement: aspartate transaminase, 75 U/L; alanine transaminase, 82 U/L; γ-glutamyl transpeptidase, 179 U/L; total serum bilirubin, 0.70 mg/dL; direct bilirubin, 0.37 mg/dL; and alkaline phosphatase, 563 U/L. CT volumetry of the graft revealed an estimated volume of 371 cm^3, the patient's weight was 7.3 kg, and a graft weight-to-recipient weight ratio of 5% was calculated.

In view of the high risk and technical difficulty associated with surgical revision of both bile ducts in a five-month-old patient, sclerotherapy of the excluded drained bile duct was considered as a possible treatment, in addition to conventional balloon cholangioplasty of the stenotic anastomosis. Given the large graft volume calculated by CT volumetry and the weight of the recipient, we concluded that inducing atrophic changes in 50% of the liver mass would not put the patient at risk for liver failure. The patient received prophylactic intravenous antibiotics. The procedure was performed under monitored anesthesia care with spontaneous respiration and additional local anesthesia. The biliary catheter was exchanged for a 6 F sheath. A cholangiogram revealed the excluded segment II bile duct, confirming no communication with the segment III bile duct and no leak of contrast from the resection margin. Alcohol sclerotherapy of the excluded bile duct was performed using selective injection of 2 mL of 95% alcohol mixed with iodinated contrast and subsequent aspiration 2 min later through a 5 F hydrophilic catheter (Fig. 4). The transhepatic tract was closed using Gelfoam sponge inserted during the sheath removal.

The procedure was well tolerated; the patient had no fever or leukocytosis in the following days. The liver function tests performed two days after alcohol sclerotherapy were substantially unchanged. Liver ultrasonography performed the day after the

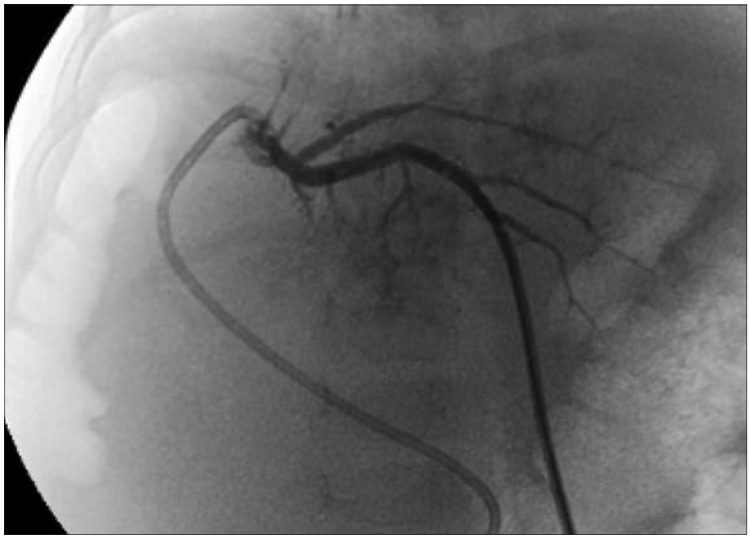

Figure 4. Alcohol sclerotherapy of the excluded bile duct was performed using selective injection and subsequent aspiration of 95% alcohol mixed with contrast.

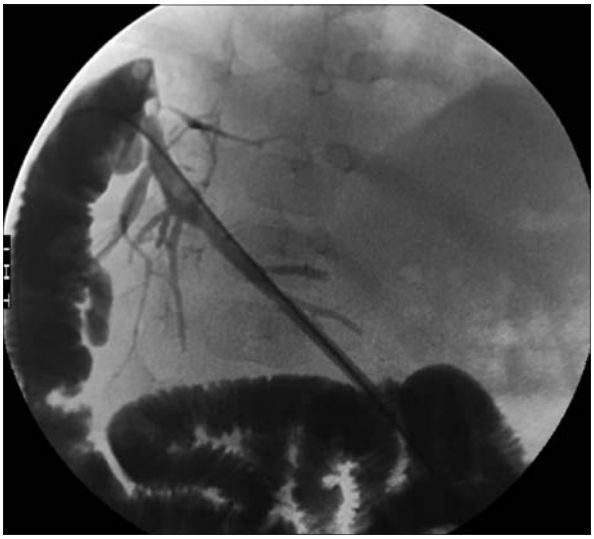

Figure 5. A cholangiogram of the segment III bile duct shows a patent biliary anastomosis after five sessions of percutaneous cholangioplasty. The catheter was removed.

procedure and one week later showed no intrahepatic bilomas or abscesses in the treated parenchymal segment. The patient remained stable during the following days, and we performed the first session of balloon dilatation of the stenotic biliary anastomosis of the segment III bile duct two weeks later. Five sessions of cholangioplasty were performed over four months. A final cholangiography, performed through a sheath, revealed patent biliary anastomosis (Fig. 5). The catheter was then removed. The patient has been without a percutaneous biliary catheter for 26 months and is in good general condition, with liver function tests showing aspartate transaminase, 56 U/L; alanine transaminase, 78 U/L; γ-glutamyl transpeptidase, 43 U/L; total serum bilirubin, 0.41 79 mg/dL; direct bilirubin, 0.16 mg/dL; and alkaline phosphatase, 420 U/L. The most recent liver ultrasonography showed minimal dilatation of the segment II bile duct with surrounding parenchymal atrophy and no bile duct dilatation in segment III. A protocol liver biopsy performed in segment III after 22 months showed no mechanical cholestasis.

Discussion

Bile duct exclusion is a potential complication of split liver transplantation; however, the incidence is reduced by an analysis of bile duct anatomy at graft procurement, backtable work, and intraoperative cholangiography during the transplantation procedure. In 10%–20% of livers, the division of the ducts into segments II and III occurs to the right of the falciform ligament (1, 2). Failure to identify two separate ducts when the graft is harvested may lead to inadvertent "orphaned duct syndrome," with the absence of anastomosis of the overlooked orphaned duct or ligation of the duct with the creation of an isolated bile duct (1, 2). The natural evolution of the isolated bile duct is toward spontaneous parenchymal fibrosis and slow self-amputation. Treatment becomes necessary in cases of isolated duct with leakage and/or external fistula when the natural evolution of the process cannot develop because continuous bile drainage is present and hepatocytes continue to produce bile. A few cases where ethanol injection therapy was used to treat an isolated bile duct with leakage and/or a biliary-cutaneous fistula have been described in adult patients (3–5). Recently, the successful treatment of a draining isolated intrahepatic biliary duct using alcohol sclerotherapy in a pediatric liver transplant recipient has been reported (6). However, we used a different technique in our case. In the case reported by Liu et al. (6), alcohol sclerotherapy was performed in four different sessions with injection of 0.6 mL of absolute alcohol and subsequent aspiration through a catheter, with a progressive decrease in bile output. The biliary drainage was removed 19 weeks after the initial injection. In contrast, we treated the excluded bile duct in a single session of alcohol sclerotherapy. We closed the parenchymal tract using Gelfoam sponge inserted during removal of the sheath to increase the effect of residual alcohol in the bile duct epithelium and to avoid perihepatic leakage. Our technique, performed in a single session, proved to be as effective and safe as the procedure described by Liu et al. (6), which required four different sessions. Alcohol destroys the bile duct epithelial cells by causing cell membrane lysis and protein denaturation. Alcohol can permeate the parenchyma and induce hepatocyte degeneration, but preservation of the hepatic blood supply should prevent clinical liver necrosis and abscess formation. Finally, alcohol causes complete destruction of the treated biliary duct, with no development of interlobar biliary communication, resulting in hypotrophy of the surrounding parenchyma (7). Possible complications of the procedure are sclerosis of the communicating bile ducts, peritoneal irritation if bile leaks are present, and abscess formation. To reduce the possibility of complications, it is necessary to use cholangiography to exclude peritoneal bile leaks and communication of the treated duct with other intrahepatic bile ducts. Furthermore, prophylactic intravenous antibiotics should be used before and after the procedure to reduce the possibility of abscess formation. An external

decompression of biliary ducts for a period of time prior to sclerotherapy is also recommended. Our patient did not have fever or leukocytosis, but when present, fever and leukocytosis should resolve prior to initiating a therapy that causes cellular destruction. An overlooked leaking duct at the cut-edge of a split liver transplant may be amenable to alcohol sclerotherapy by injection through a sheath and concurrent balloon occlusion to prevent peritoneal spillage. This treatment protocol could increase the usefulness of sclerotherapy.

Prior to performing alcohol sclerotherapy, the transplant team must weigh the risks associated with sacrificing a portion of the liver parenchyma against the risks associated with complex surgical revision involving, as in our patient, two separate biliary anastomoses and a high risk for stricture recurrence. The advisability of destroying a large volume of liver parenchyma can be assessed by estimating the remaining functional liver volume based on the graft weight-to-recipient weight ratio calculated from CT or magnetic resonance volumetric reconstruction data.

We attempted to reduce the minimum radiation dose by using lead protection under the patient's body by limiting the fluoroscopy time and by using a minimum dose or no magnification. The total radiation dose in each procedure was less than 2 Gy, which is the dose considered to be the threshold for adverse effects such as transient erythema and depilation (8). A dose higher than 12 Gy puts the patient at risk for dermal necrosis (8). The irradiation risk in the present case owed primarily to several sessions of balloon dilatation. Nevertheless, any unnecessary additional irradiation should be avoided in a small child, emphasizing the importance of single-session alcohol sclerotherapy.

In conclusion, a single selective intrahepatic infusion of 95% alcohol was used to successfully treat a large draining isolated intrahepatic biliary duct without complications. The percutaneous approach should be considered as an alternative to redo surgery for a large draining isolated intrahepatic biliary duct in patients who are at high-risk for surgical revision. Further studies in more patients are required to confirm our findings.

Conflict of interest disclosure

All authors declared no conflicts of interest.

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