Case Report

DOI: 10.5582/irdr.2019.01081

Two-staged biliary reconstruction with temporary complete external biliary drainage as a bailout procedure in a pediatric patient after difficult living donor liver retransplantation

Hidekazu Yamamoto*, Yasuhiko Sugawara, Sho Ibuki, Keita Shimata, Yasuko Narita, Taizo Hibi

Department of Pediatric Surgery and Transplantation, Kumamoto University Graduate School of Medical Sciences, Kumamoto, Japan.

Summary **Biliary leakage at the site of the hepaticojejunostomy after liver transplantation is a life-threatening complication. We herein present the case of a 7-year-old girl who underwent complete external biliary drainage during difficult living donor liver retransplantation as a bailout procedure. The patient had undergone duct-to-duct biliary reconstruction in the initial living donor liver transplantation. In the retransplantation, Roux-en-Y (RY) reconstruction was planned but abandoned due to the critical condition in the operation. As an alternative procedure, the patient underwent complete external drainage using a 6Fr drainage tube with cuff. Five months after retransplantation when the nutrition status and physical strength of a patient recovered fully, RY hepaticojejunostomy was successfully performed. This is a case report of two-staged biliary reconstruction with temporary complete external biliary drainage used in pediatric liver retransplantation, which was performed after some months not a few days. It is a safe and feasible alternative when primary anastomosis is deemed to carry a high risk of bile leakage in cases of difficult liver transplantation in critically ill patients.**

Keywords: Living donor liver transplantation, two-staged biliary reconstruction, Roux-en-Y hepaticojejunostomy

1. Introduction

Retransplantation, an extended operation time, severe malnutrition, and hepatic artery thrombosis have been reported as risk factors of biliary leakage, which may lead to fatal consequences (*1-4*). In addition, Roux-en-Y (RY) hepaticojejunostomy in retransplantation cases may require extensive adhesiolysis, putting the patient at an increased risk of bile leakage (*5*). Delayed biliary reconstruction after a few days from liver transplantation (LT) were reported to avoid initial biliary anastomosis in the literature (*6-9*). However, the patient's condition doesn't always recover fully to allow us to perform biliary reconstruction for a few days. We herein report a patient who underwent successful two-staged biliary reconstruction after five months from retransplantation.

2. Case Report

The patient was a 5-year-old girl (height 98 cm and weight 14 kg). She underwent living donor liver transplantation (LDLT) for primary sclerosing cholangitis (PSC) with a left lateral segment graft donated from her 40-year-old father. Duct-to-duct (DD) reconstruction was performed. However, she suffered two episodes of biopsy-proven acute cellular rejection at three and nine months after LDLT. Her serum bilirubin level increased gradually two years after transplantation, and endoscopic retrograde cholangiopancreatography showed strictures of the intrahepatic biliary tract compatible with recurrent PSC. Her condition eventually progressed to decompensated liver cirrhosis. Her pediatric end-stage liver disease (PELD) score was 43.

**Address correspondence to:*
Dr. Hidekazu Yamamoto, Department of Transplantation and Pediatric Surgery, Postgraduate School of Medical Science, Kumamoto University Hospital, 1-1-1, Honjo, Chuo-ku, Kumamoto 860-8556, Japan.
E-mail: yamahide@kuh.kumamoto-u.ac.jp

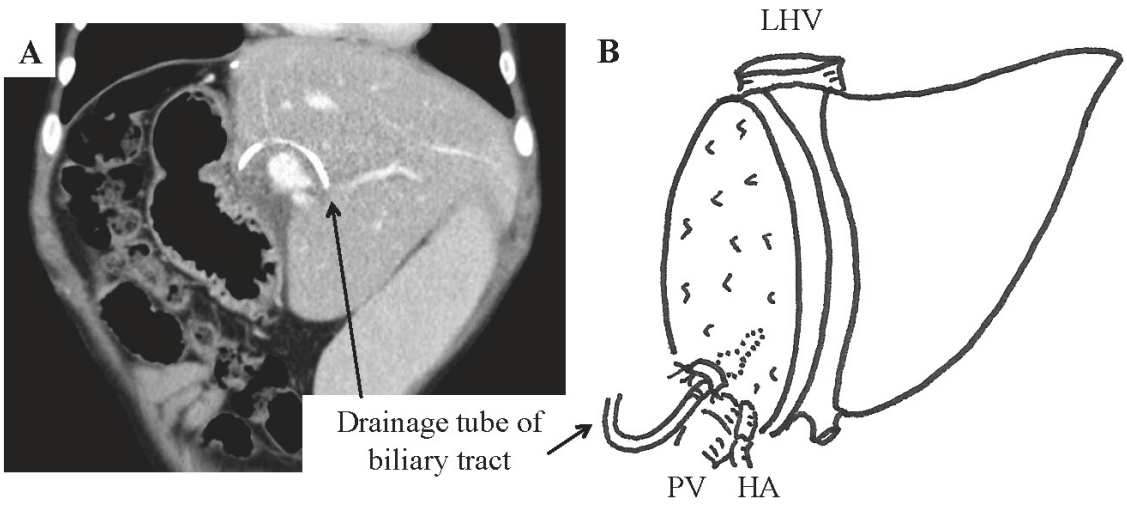

Figure 1. Computed tomography scan and schematic diagram of the lateral liver graft. (A) Temporary complete external biliary tube into the lateral liver graft. **(B)** Similarly, external biliary tube into the lateral liver graft.

She underwent retransplantation with a left lateral liver segment donated by her mother. Laparotomy revealed massive ascites and dense adhesion between the upper jejunal loops and the cirrhotic liver. The primary liver graft was explanted six hours after the skin incision. The new graft was placed, and hepatic and portal veins were reconstructed, after which the graft was successfully reperfused (warm and cold ischemic time, 37 and 176 minutes, respectively). Microvascular hepatic arterial reconstruction followed. At this time, 8 hours had passed since the skin incision, and the estimated blood loss was over 6,000 g. The total urine output was only 50 ml, suggesting significant intravascular volume depletion, and her hemodynamics were unstable despite the use of catecholamine.

The native common bile duct was severely injured during primary liver graft removal, and DD re-reconstruction was impossible. In addition, the intestine was diffusely edematous and was unsuitable for biliary reconstruction. We decided to perform external biliary drainage instead of primary anastomosis. A 6-Fr drainage tube was placed in the left hepatic duct of the liver graft, measuring 3 mm in diameter (Figure 1). The orifice was closed using interrupted sutures of 5-0 prolene and the tube was fixed to the bile duct with a single tie. The tube was passed through the abdominal wall as an external biliary drainage route.

After the operation, she required an intensive-care unit stay of one month, including two weeks on machine ventilation. Enteral nutrition and gait rehabilitation were started on the postoperative day 5 and 31, respectively. Her general condition eventually recovered thereafter. She and her family were given detailed instructions to safely manage the external biliary drainage tube at home, and she was discharged three months after retransplantation with an Eastern Cooperative Oncology Group Performance Status (ECOG PS) of 3. Our transplant coordinators maintained close contact with the patient and her family to make sure that there were no tube-related complications. At five months after retransplantation, she was confirmed to have fully recovered from her impaired nutritional status with a weight gain of 2 kg and an ECOG PS of 1. She underwent elective hepaticojejunostomy with an uneventful postoperative course and was discharged 25 days after the operation. Since then, she has been doing well for eight years without biliary complication or signs of recurrent PSC and liver function test has been within normal level.

3. Discussion

We herein report the first pediatric patient who underwent two-staged biliary reconstruction after some months from reLDLT. Several centers have previously reported the usefulness of delayed biliary reconstruction after LT (*6-8*). Komorowski *et al*. (*7*) reported that perihepatic packing and temporary abdominal closure with delayed biliary reconstruction were viable options for massive uncontrollable bleeding and bowel edema during LT. DiNorcia *et al*. (*9*) reported on 150 adult patients including retransplantation (29.3%) with postreperfusion hemodynamic instability and coagulopathy during LT which required damage control. Eighty-four of the 150 underwent delayed biliary reconstruction. In both reports, as well in others, the interval between LT and biliary reconstruction was approximately two days, during which hemostasis was achieved with stable vital signs and the general condition of the recipient was deemed fit for relaparotomy (*6,7,9*). However, our patient was considered too sick to bring back to the operating room at only a few days after reLDLT, as demonstrated by her one-month stay in the intensive-care unit. We therefore decided to postpone biliary reconstruction until her nutritional status and physical

strength had fully recovered. To minimize the risk of tube-related complications, such as accidental tube dislodgment and cholangitis, which in her case was considered higher than in adults, she and her family were given strict instructions from the nurses regarding tube management at home. Our transplant coordinators were also available 24 hours a day, 7 days a week, in case of emergency and made frequent phone calls to make sure that the patient was safe. Such meticulous multidisciplinary management enabled us to safely perform two-staged biliary reconstruction in a pediatric patient.

Indications for two-staged biliary reconstruction should be based on variable factors, including the preoperative condition (*e.g.*, high PELD/MELD, ECOG PS, nutritional status, retransplantation) and difficulty of the operation (*e.g.*, duration, estimated blood loss, hemodynamic instability requiring high doses of vasopressive agent, anuria, level of metabolic acidosis, significant bowel edema, quality of the liver graft). The timing of the biliary reconstruction should also be determined on a case-by-case basis. Complete functional recovery with a good nutritional condition is essential for successful staged surgery. In LDLT, the graft functional recovery tends to be slower than that after deceased donor LT, so the timing of relaparotomy should be set later. Although there is no general rule regarding the appropriate timing of staged biliary reconstruction in critically ill patients, it may be worthwhile to wait several months. In fact, the feasibility of two-staged pancreaticojejunostomy at three months after pancreaticoduodenectomy for patients who are at a high risk of pancreatic fistula has been reported (*10*). In the present case, staged biliary reconstruction was delayed up to five months after retransplantation because of the patient's poor nutritional and functional status. Despite this delay, this surgical procedure was safely performed five months after retransplantation without severe adhesion.

In conclusion, two-staged biliary reconstruction with temporary complete external biliary drainage is a safe and feasible alternative in difficult LT in critically ill patients.

References

1. Luthold SC, Kaseje N, Jannot AS, Mentha G, Majno P, Toso C, Belli DC, McLin VA, Wildhaber BE. Risk factors for early and late biliary complications in pediatric liver transplantation. Pediatr Transplant. 2014; 18:822-830.
2. Laurence JM, Sapisochin G, DeAngelis M, *et al*. Biliary complications in pediatric liver transplantation: Incidence and management over a decade. Liver Transpl. 2015; 21:1082-1090.
3. Antolovic D, Koch M, Galindo L, Wolff S, Music E, Kienle P, Schemmer P, Friess H, Schmidt J, Buchler MW, Weitz J. Hepaticojejunostomy--analysis of risk factors for postoperative bile leaks and surgical complications. J Gastrointest Surg. 2007; 11:555-561.
4. Greif F, Bronsther OL, Van Thiel DH, Casavilla A, Iwatsuki S, Tzakis A, Todo S, Fung JJ, Starzl TE. The incidence, timing, and management of biliary tract complications after orthotopic liver transplantation. Ann Surg. 1994; 219:40-45.
5. Pungpapong S, Alvarez S, Hellinger WC, *et al*. Peritonitis after liver transplantation: Incidence, risk factors, microbiology profiles, and outcome. Liver Transpl. 2006; 12:1244-1252.
6. Gilbo N, Mirabella S, Strignano P, Ricchiuti A, Lupo F, Giono I, Sanna C, Fop F, Salizzoni M. External biliary fistula in orthotopic liver transplantation. Transplant Proc. 2009; 41:1316-1318.
7. Komorowski AL, Li WF, Millan CA, Huang TS, Yong CC, Lin TS, Lin TL, Jawan B, Wang CC, Chen CL. Temporary abdominal closure and delayed biliary reconstruction due to massive bleeding in patients undergoing liver transplantation: An old trick in a new indication. J Hepatobiliary Pancreat Sci. 2016; 23:118-124.
8. Kim J, Zimmerman MA, Lerret SM, Scott JP, Voulgarelis S, Fons RA, Vitola BE, Telega GW, Hoffman GM, Berens RJ, Hong JC. Staged biliary reconstruction after liver transplantation: A novel surgical strategy for high acuity pediatric transplant recipients. Surgery. 2019; 165:323-328.
9. DiNorcia J, Lee MK, Harlander-Locke MP, Xia V, Kaldas FM, Zarrinpar A, Farmer DG, Yersiz H, Hiatt JR, Busuttil RW, Agopian VG. Damage Control as a Strategy to Manage Postreperfusion Hemodynamic Instability and Coagulopathy in Liver Transplant. JAMA Surg. 2015; 150:1066-1072.
10. Hasegawa K, Kokudo N, Sano K, Seyama Y, Aoki T, Ikeda M, Hashimoto T, Beck Y, Imamura H, Sugawara Y, Makuuchi M. Two-stage pancreatojejunostomy in pancreaticoduodenectomy: A retrospective analysis of short-term results. Am J Surg. 2008; 196:3-10.

(Received June 19, 2019; Revised August 27, 2019; Accepted August 31, 2019)