Case Report

A 72-year-old man affected by cholangiocarcinoma (Klatskin 3a) who underwent right hepatectomy with a biliodigestive anastomosis (Roux-en-Y), was referred to our Department to perform a 1-year follow-up MRI scan.

MRI showed intrahepatic biliary dilation caused by stenosed anastomosis. Multidisciplinary board decided for percutaneous bilioplasty. PTBD was positioned through left access.

A month later, biliary drainage revision was necessary because of bile leakage through cutaneous access of PTBD. Conservative treatment was chosen; therefore, replacement of biliary drainage with higher French scaling was successfully performed. PTBD was removed 3 months later after a normal cholangiogram.

Within 8 days from PTBD removal, biliary leakage of 150 cc/day re-appeared, compelling the placement of a new PTC under US guidance through left biliary system was performed.

Cholangiography showed a bilo-cutaneous fistula at the entry site of previous PTBD [Figure 1].

The point of leakage was then reached through biliary tree with a 0.035" hydrophilic guidewire (Radifocus® Guidewire M Standard type, Terumo, Tokyo, Japan) and a multipurpose 4F vascular catheter (Cordis, Fremont, California); a mixture of Lipiodol (Guerbet, Villepinte, France) and N-butyl-Cyanocrylate (Glubran 2, GEM, Italy) was then injected [Figure 2].
The final cholangiography demonstrated complete obliteration of the fistula and no more bile leakage was observed [Figure 3A and B].

Patient was discharged after 3 days and no recurrence was documented during the 3 months follow-up.

**Discussion**

Biliary fistula and bile leakage are complications that may occur during hepato-biliary surgery (both open and laparoscopic) and percutaneous biliary interventions.[1‑3]

Variations in biliary anatomy can increase the risk of ductal injuries.

Adequate pre-operative imaging evaluation (CT and/or MRCP) may reduce the incidence of bile duct injury with occurrence of leakage.[4,5]

Damaged bile ducts (isolated or communicating with the main biliary tree) may leak directly into the abdomen from the time of intervention or develop fistulas through thoracic wall to the skin, along percutaneous transhepatic drainage (PTBD).[1]

Acute injuries may not be detected immediately during procedures, enduring silently, resolve spontaneously or they can reveal as bile peritonitis, prolonged time of wound healing and lately with malabsorption.[6,7]

The high rate of morbidity and mortality related to surgical reoperation in such delicate patients making endoscopic and interventional radiology techniques the best choice treatments.[8‑10]

Development of cutaneous biliary leakage is a well-known and frequent complication that occurs in patients who keep PTBD catheters in place for a long time. The occurrence of secondary obstructions to bile discharge (e.g., debris, malignant stenosis) represent a constant cause of bile spreading in the space between the catheter and surrounding tissues.

Usually this condition is solved by removing the cause of obstruction, but in many cases bile leakage persist or, more often, no clear evidence of a possible cause is found, with a good antegrade bile drainage to duodenum at percutaneous cholangiography (PTC).

In other conditions a normal cholangiogram would suggest PTBD removal and patient discharge but in the presence of a cutaneous leakage the first option is the insertion of a larger PTBD with more discomfort and longer time of hospitalization for the patient.

Different materials have already been studied and tested to seal the biliary tract, in particular fibrin,[11] ethanol,[12] and NBCA.[1,2,13‑16]

These studies, based on case reports or small series, demonstrated that the latter is safer and more effective.

Due to its efficacy on biliary tree, in addition on its established role as embolic agent in different endovascular procedures,[17‑19] we decided to use NBCA to treat our patient.

We have found no literature cases in which the glue was used to seal a sub-cutaneous route; in conclusion we find that NBCA can be successfully employed in this field in order to improve patient comfort and shorten hospital stay.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have...
given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

References
1. Vu DN, Strub WM, Nguyen PM. Biliary duct ablation with N-butyl cyanoacrylate. J Vasc Interv Radiol 2006;17:63-9.  
2. Görich J, Riilinger N, Sokiranski R, Siech M, Vogel J, Wikström M, et al. Percutaneous transhepatic embolization of bile duct fistulas. J Vasc Interv Radiol 1996;7:435-8.  
3. Seewald S, Groth S, Sriram PVJ, Xikun H, Akaraviputh T, Mendoza G, et al. Endoscopic treatment of biliary leakage with n-butyl-2-cyanoacrylate. Gastrointest Endosc 2002;56:916-9.  
4. Sharif K, de Ville de Goyet J. Bile duct of Luschka leading to bile leak after cholecystectomy—visiting the biliary anatomy. J Pediatr Surg 2003;38:E21-3.  
5. Minutoli F, Naso S, Visalli C, Ianneli D, Silipigni S, Pitrone A, et al. A new variant of cholecystohepatic duct: MR cholangiography demonstration. Surg Radiol Anat 2015;37:539-41.  
6. Smith AC, Schapiro RH, Kelsey PB, Warshaw AL. Successful treatment of nonhealing biliary-cutaneous fistulas with biliary stents. Gastroenterology 1986;90:764-9.  
7. Lichtenstein S, Moorman DW, Malatesta JQ, Martin MF. The role of hepatic resection in the management of bile duct injuries following laparoscopic cholecystectomy. Am Surg 2000;66:372-6; discussion 377.  
8. Ryan ME, Geenen JE, Lehman GA, Aliperti G, Freeman ML, Silverman WB, et al. Percutaneous transhepatic embolization of bile duct fistulas. J Vasc Interv Radiol 1996;7:435-8.  
9. Lauterio A, Slim A, Aseni P, Giacomoni A, Di Sandro S, Corso R, et al. Percutaneous transhepatic bile duct ablation with n-Butyl cyanoacrylate in the treatment of a biliary complication after split liver transplantation. J Transplant 2009;2009:1-3.  
10. Carrafiello G, Piacentino F, Ierardi A, Cardim L. Percutaneous transhepatic embolization of biliary leakage with N-butyl cyanoacrylate. Indian J Radiol Imaging 2012;22:19.  
11. Mauri G, Pescatori LC, Mattiuz C, Poretti D, Pecorini V, Melchiorre F, et al. Ethanol injection therapy of an isolated bile duct associated with a biliary-cutaneous fistula. J Gastroenterol Hepatol 2002;17:807-10.  
12. Kuran S, Disibeyaz S, Parlak E, Arhan M, Kacar S, Sahin B. Biliocutaneous fistula following alveolar hydatid disease surgery treated successfully with percutaneous cyanoacrylate. Dig Dis Sci 2006;51:18-20.  
13. Wajswol E, Jazmati T, Contractor S, Kumar A. Portal vein embolization utilizing N-Butyl cyanoacrylate for contralateral lobe hypertrophy prior to liver resection: A systematic review and meta-analysis. Cardiovasc Intervent Radiol 2018;41:1302-12.  
14. Caloggero S, Catanzariti F, Stagno A, Silipigni S, Bottari A. Use of a mixture of lipiodol and cyanoacrylate in percutaneous embolization treatment of symptomatic renal Angiomyolipomas: Our experience. Radiol Case Rep 2019;14:343-7.  
15. Kim PH, Tsauo J, Shin JH, Yun S-C. Transcatheter arterial embolization of gastrointestinal bleeding with N-butyl cyanoacrylate: A systematic review and meta-analysis of safety and efficacy. J Vasc Interv Radiol 2017;28:522-531.e5.