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# **Combined Bile Duct Ablation and Fistulous Tract Embolization Using N-Butyl Cyanoacrylate to** Manage a Biliary-Cutaneous Fistula Following Percutaneous Transhepatic Biliary Drainage (PTBD) for Hilar Cholangiocarcinoma

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#### Abstract

#### **Keywords**

- bile leak
- ► biliary-cutaneous fistula
- cholangiocarcinoma
- N-Butyl cyanoacrylate
- percutaneous transhepatic biliary drainage

Biliary fistula and bile leak are known complications following hepatobiliary surgery, trauma, and percutaneous biliary interventions. In the case of an isolated biliary system with a prolonged indwelling percutaneous transhepatic biliary drainage (PTBD) catheter, a biliary-cutaneous fistula (BCF) may develop after catheter blockage or its accidental slippage. Due to the absence of internal drainage, secreted bile flows through the matured PTBD tract to form a fistula. If left untreated, chronic BCF will result in malabsorption, infection, and delayed wound healing. Here, we report a case of left-sided BCF following prolonged PTBD for Bismuth type II cholangiocarcinoma (metastatic disease), which was initially managed by bile duct ablation using N-butyl cyanoacrylate. The patient further needed fistulous tract embolization to obliterate the BCF.

## **Case Report**

A 41-year-old male initially presented to the surgical gastroenterology outpatient department (OPD) with complaints of jaundice for 15 days. On further evaluation, he was diagnosed with Bismuth type II hilar cholangiocarcinoma with nodal and pulmonary metastases. Initially, he had undergone right percutaneous transhepatic biliary drainage (PTBD) for hyperbilirubinemia. Left PTBD was performed after 10 days since patient

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developed cholangitis due to infected left biliary system (Fig. 1). Subsequently, the right PTBD was internalized, and metallic stenting was also done. However, the left PTBD (8 French pigtail) was kept on external drainage since stricture could not be negotiated despite multiple attempts.

The patient was started on chemotherapy on a daycare basis and was on regular follow-ups in the interventional radiology clinic. Five months after the PTBD, the patient presented to the OPD with complaints of persistent biliary

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**Fig. 1** (A) Axial computed tomography (CT) image shows ill-defined hypoenhancing hilar mass (black arrow) with atrophied left lobe (white arrow). (B) Axial CT section at lower thoracic level reveals multiple well-defined metastatic nodules (black circle). (C) Clinical image shows bilious output within the colostomy bag.

leakage from the left PTBD site for 20 days after the accidental removal of the left PTBD tube. The patient did not give consent for reinsertion of the left PTBD catheter. Alternatively, the biliary fistula was managed temporarily with a colostomy bag at the site of the bile leak (**-Fig. 1C**). The daily output was 150 to 200 mL even after 1 month. Output was greenish and non-purulent. The patient was afebrile and had a total bilirubin of 2.2 mg/dL, total leukocyte counts of 7200/ mm,<sup>1</sup> and a hemoglobin level of 9.1 g/dL. Ultrasonography (USG) and contrast-enhanced computed tomography (CT) showed nondilated right-sided bile ducts and mildly dilated left-sided bile ducts with an atrophied left lobe of the liver. No perihepatic or local site collection was found.

Given the persistent biliary-cutaneous fistula (BCF), gastroenterology consultation was taken for endoscopic hepaticogastrostomy. However, the endoscopist found it unsuitable for hepaticogastrostomy because of the nonapposed gastric wall and atrophied left lobe. Therefore, percutaneous embolization of the fistulous tract was planned. Nevertheless, the fistulous tract could not be cannulated despite repeated attempts. Thus, percutaneous left-sided bile duct ablation using N-butyl cyanoacrylate (NBCA) was considered, given an atrophied left lobe and isolated left-sided biliary system.

### **Percutaneous Bile Duct Ablation**

The patient received 1 g of intravenous ceftriaxone 1 hour before the procedure. Segment 3 bile duct was accessed under the USG guidance using a 21G micropuncture set. Cholangiogram revealed left-sided bile ducts not communicating with the right-sided biliary tree (**-Fig. 2A, B**). Then, a 6F vascular sheath was placed within the segment 3 duct over a 0.035" stiff guidewire, and a 5F Kumpe (KMP) access catheter was inserted within the sheath into the biliary tree. Bile ducts were then irrigated with normal saline before glue injection.

Note that 3 mL of 25% glue solution was made by mixing 0.75 mL of NBCA and 2.25 mL of lipiodol. The sheath was retracted from the biliary system, and the bile ducts were then embolized using the glue solution. KMP catheter was



**Fig. 2** (A) Segment 3 biliary access was obtained using a micropuncture set; cholangiogram shows isolated left biliary system. (B) Fluoroscopy and (C, D) computed tomography (CT) images reveal glue cast (black arrow) within the left-sided biliary tree. Right-sided biliary metallic stent is seen in situ.

slowly withdrawn to the skin surface, filling the access tract with glue.

Immediate postprocedure noncontrast CT showed the left-sided bile ducts filled with glue cast (**-Fig. 2C, D**). The patient received oral antibiotic therapy for 1 week. Bile output through the fistula was reduced to 50 to 60 mL/day.

#### **Fistulous Tract Embolization**

Given the continuous output from the fistulous opening, the second intervention session was planned after 10 days. Fortunately, we could cannulate the fistula this time using a 23G plastic cannula. Fistulogram showed a linear tract communicating with the left-sided biliary tree without any ramification or collection (**-Fig. 3A**). A 0.035" J-tipped



**Fig. 3** (A) Fistulogram after cannulation of external opening shows linear fistulous tract communicating with the left-sided biliary tree. (B) An 0.035" hydrophilic guidewire (white dash arrow) was inserted through the tract, followed by insertion of a 5F Kumpe (KMP) catheter. (C) Postembolization fluoroscopy image reveals linear glue cast along the fistulous tract (open black arrows).

hydrophilic guidewire was inserted into the bile duct through the fistulous tract (**-Fig. 3B**). A 5F KMP catheter was inserted and flushed with a 5% dextrose solution. The fistulous tract was then embolized using 2 mL of 50% gluelipiodol mixture while slowly withdrawing the KMP catheter to the skin surface (**-Fig. 3C**).

No further leakage was observed. The patient did not show any features of cholangitis or further increase in bilirubin levels till the latest follow-up at 2 months.

#### Discussion

Although the surgical resection of the hepatic region associated with the fistula remains the best reliable treatment, the high rate of morbidity and mortality related to surgery, especially in comorbid patients (metastatic cholangiocarcinoma in our case), make nonoperative interventional management a preferred option.<sup>2</sup>

Embolization of the fistulous tract using fibrin glue, gelfoam, metallic coils, and NBCA glue has been described previously with very low related morbidity and mortality.<sup>3</sup> Bae et al successfully treated seven cases of biliary fistula using NBCA. While most of the cases were due to ruptured hepatic abscesses following tumor ablation, one patient had developed a biliary fistula after pancreatitis.<sup>1</sup> Bottari et al<sup>4</sup> and Alberto et al<sup>5</sup> performed successful obliteration of a BCF using glue. Furthermore, Ierardi et al demonstrated successful treatment of a BCF following the removal of a PTBD catheter using an Amplatzer vascular plug.<sup>6</sup>

When a fistula persists, obliteration of the contributing bile duct remains an alternative option. Different embolic agents used for this purpose include fibrin, acetic acid, ethanol, and NBCA. Several case reports and series have demonstrated successful closure of biliary leakage by ablation of bile ducts using ethanol.<sup>3,7–9</sup> However, ethanol ablation is not without risk. Ethanol causes irreversible cellular damage; hence, it is not advisable to use ethanol when there is a communication of contributing bile duct with the remaining biliary tree. On the contrary, glue is less toxic, and isolated bile duct ablation can

be performed even in communicating biliary trees, unlike ethanol. However, unintentional spillage of glue into nontarget ducts may occur.<sup>3</sup> Vu et al reported the utility of bile duct ablation using NBCA to treat six postsurgical bile leak cases. While four patients only required a single session of glue injection, the remaining two needed a second embolization session with coils and glue.<sup>3</sup> Bile duct ablation using glue has been shown to be effective for isolated biliary dilatation after split liver transplantation.<sup>10</sup>

Segmental portal vein embolization with or without bile duct ablation is also effective in controlling post-hepatectomy bile leak. However, it has not been described for BCF in patient with hilar cholangiocarcinoma.<sup>7,11</sup> In patients with hilar block, endoscopic hepaticogastrostomy remains an alternative option; however, it is a technically challenging procedure requiring an expert endoscopist. Furthermore, several anatomical factors, such as left lobe atrophy, tumor infiltration of the gastric wall at the puncture site, and isolated right intrahepatic bile duct obstruction, may limit its creation. Therefore, endoscopic hepaticogastrostomy may not always be possible.<sup>12</sup>

In our case, bile duct ablation by glue was performed first due to the inability to cannulate the fistula. Persistent low output demanded a second treatment when the fistulous tract was embolized. Bile duct ablation using glue in malignant obstruction has not been described previously. It makes our case report unique.

#### Conclusion

Spontaneous closure of the BCF usually occurs in the early post-PTBD period; however, if the persistent leak is seen because of delayed accidental removal of the catheter, then embolization of the tract with or without associated bile duct embolization is the treatment of choice.

Ethical Approval The internal ethical committee approved this article. Availability of Data and Materials

The data sets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

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Conflict of Interest None declared.

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