

# Post-traumatic biliopleural fistula treated with endoscopic retrograde cholangiopancreatography: a case report

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## Abstract

Thoracobiliary fistulae are a rare disorder caused by iatrogenic injury or trauma. Because these fistulae are uncommon, no standard of care has been established, but non-operative management (NOM) is often advised. As a result, endoscopic retrograde cholangiopancreatography (ERCP) appears to be playing a growing role in the treatment of select cases. A 42-year-old male patient

involved in a street fight developed a biliopleural fistula as a result of an abdominal penetrating injury. Even in the lack of a clearly defined site of biliary leak, NOM and ERCP have proven to be safe and effective therapy in this case.

## Introduction

Biliopleural fistula (BPF) is a communication between the biliary system and the pleural cavity. Thoracobiliary fistulae are a rare condition most commonly resulting from iatrogenic injuries or trauma.<sup>1</sup> Laceration of the diaphragm and hepatic injury with bile spillage consequent to a penetrating thoraco-abdominal trauma are indeed the ideal prerequisites for the development of a BPF.<sup>2</sup> To date, no standard of care for this condition has been established, nevertheless, non-operative management has been proposed as the first-line approach to BPF with the use of ERCP in selected cases.<sup>3,4</sup> However, WSES-AAST guidelines stated the general principles about the diagnostic assessment of a hemodynamically stable patient with a biliary tree trauma: a CT scan should be performed at symptoms onset and a magnetic resonance cholangiopancreatography (MRCP) should be performed after 48 hours in order to properly identify the leakage.<sup>5</sup> According to these guidelines an early ERCP should be attempted after MRCP. ERCP is indeed a both diagnostic and therapeutic procedure by which a biliary tree injury can be detected and treated by sphincterotomy or stenting of the biliary duct.<sup>6</sup> Recently we faced the case of a delayed-onset biliopleural fistula following an abdominal penetrating injury in a male patient. In this case, the diagnostic assessment was unable to identify an evident site of biliary leak, but ERCP has proven to be effective in achieving a reduction of fistula flow rate and the subsequent healing of the patient.

## Case Report

A 42-year-old man was admitted in March 2022 to our emergency department after a street fight. Past medical history was unremarkable. On admission the patient was hemodynamically stable; physical examination showed a penetrating stab wound of 1.0×0.5 cm in the right upper abdominal quadrant right below the costal margin. Blood tests showed a white blood cell count of 8.9×10<sup>9</sup>/L, a hemoglobin of 14.3 g/dL, a CRP of 1.62 mg/dL, a total bilirubin of 0.8 mg/dL, a GGT of 46 IU/L, an AST of 45 IU/L and an ALT of 65 IU/L. Extend Focused Sonography Assessment for Trauma (e-FAST) was performed without identifying free fluid or air both in the abdominal and pleural cavities. An additional II-level diagnostic imaging was performed with a CT scan, showing a small amount of perihepatic fluid without detectable blushing

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Ethics approval: this case report does not include any interaction or intervention with human subjects and does not include any access to identifiable private information and no ethical approval was needed.

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and no signs of pneumothorax, pleural effusion, or diaphragm laceration. Therefore, the patient was admitted to our General Surgery department for observation and an antibiotic prophylaxis was provided. Three days later, the patient complained of dyspnea and sharp chest pain especially when breathing deeply. At the time of symptom onset blood pressure was 130/90 mmHg, heart rate was 94 beats per minute, temperature was 37.5°C, and oxygen saturation was 93% on room air with a respiratory rate of 20 breaths per minute. Blood test showed a white blood cell count of  $11.4 \times 10^9/L$ , a CRP of 3.84 mg/dL, a total bilirubin of 1.0 mg/dL, a GGT of 186 IU/L, an AST of 102 IU/L, and an ALT of 120 IU/L. A thoracoabdominal CT scan was performed, revealing a significant right pleural effusion associated with right lung atelectasis (Figure 1). A chest tube was then placed revealing an unexpected bilious output of 500 mL. Immediately upon drainage, the patient reported full resolution of dyspnea. In the following days, a constant bilious output of about 200 mL per day has been collected. An MRCP was performed six days after trauma to identify the presence of any biliary leak: neither biliary tract dilation nor biliary tree injury was found. MRCP also confirmed the presence of right pleural effusion without diaphragm laceration and a small amount of perihepatic effusion (Figure 2).

An ERCP was then performed. Intra-procedural cholangiography confirmed the absence of a detectable bile ducts injury. Therefore, we assumed that the biliary leak was minimal so, even in the absence of a definite leaking site, we decided to lower endoluminal pressure in the biliary tree in order to reduce the fistula's bile supply. Then a sphincterotomy was promptly performed and a plastic stent was placed in the common bile duct. The day after the biliary output from the chest tube dropped down and the patient complained of no more chest pain. Post-procedural blood tests did not show WBC, CRP, or lipase elevation. Two days after the ERCP the chest tube output was serous with a flow rate of 30 mL/24 hours. A thoracoabdominal CT scan was performed showing the almost complete resolution of the residual pleural effusion and a reduction of the residual abdominal perihepatic free fluid (Figure 3A). The chest tube was then removed. The patient was discharged three days after the ERCP. An outpatient follow-up CT scan was scheduled after 2 weeks that revealed a complete resolution of pleural effusion and the plastic stent still in place in the common bile duct (Figure 3B).

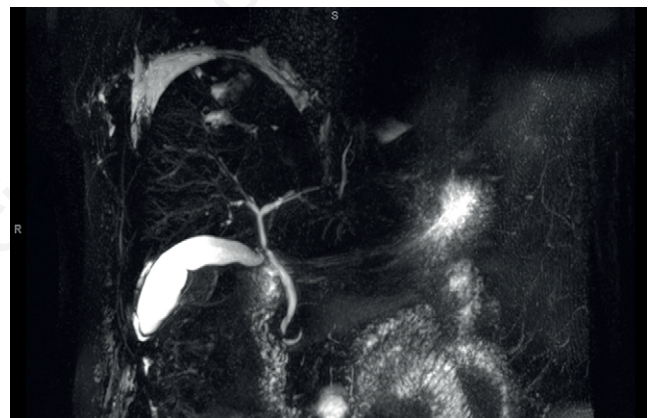
Four months later the biliary stent was removed without any following relapse of BPF. At the end of the treatment pathway, the patient reported his satisfaction with the early resolution of the dyspnea after the chest tube placement and the early discharge with a prompt return to daily activities provided by using a minimally invasive technique to treat his condition.

## Discussion

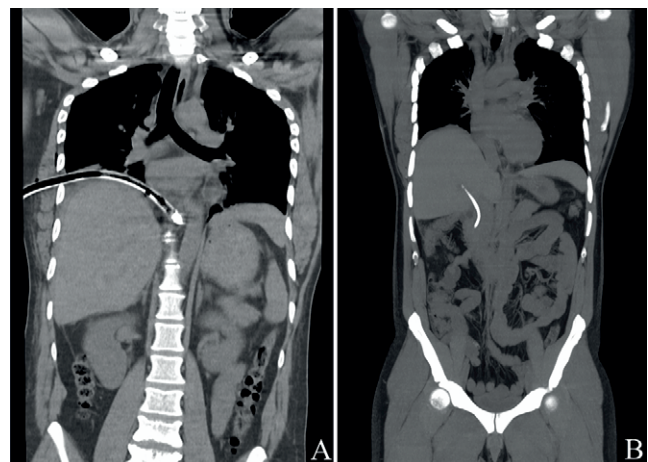
BPF is a communication between the biliary tree and the pleural cavity. Trauma-related biliopleural fistulae are a rare occurrence following thoracoabdominal injuries that can lead to potentially life-threatening complications due to the risk of sepsis related to both thoracic and abdominal bile collections (*i.e.*, pleural empyema, broncho-biliary fistula, bilomas, bilhemia, and choleperitoneum).<sup>7</sup> For this reason timely diagnosis and prompt management are crucial to improve the patient's survival rate. Scientific literature about BPF treatment is primarily based on case reports and case series either about post-traumatic fistulae or about fistulae of other etiologies (*e.g.*, echinococcosis, hepatic abscesses, iatrogenic injuries).<sup>8</sup> This lack of studies, the rarity of the condition, and the variety of possible clinical presentations made it difficult to establish a consensus algorithm for the treatment of BPF.<sup>4,8</sup> The



**Figure 1.** CT scan performed at the onset of symptoms showed perihepatic free fluid and right pleural effusion.



**Figure 2.** 3D reconstruction of MR T2 weighted sequences.



**Figure 3.** A) CT scan performed 2 days after ERCP showing an almost complete resolution of the pleural effusion and a small amount of residual perihepatic free fluid; the chest tube is still in place. B) CT scan performed 2 weeks after ERCP showing complete resolution of pleural effusion and the plastic stent still in place in the common bile duct.

first approach to BPF has historically been a thoracotomy with pleural decortication that reported a 97% success rate.<sup>9</sup> Many cases of operative management (OM) had been reported in the existing literature and, to date, OM is still an option in the management of hemodynamically unstable patients, wide diaphragmatic defects, or BPF associated with both chest and abdominal injuries due to trauma or sequelae of the fistula.<sup>2,7,10</sup> The choice between OM and NOM is a key factor in treating a BPF. Although WSES guidelines about the management of liver trauma and extra-biliary tree injuries are not focused on the treatment of BPF, they state that NOM is the best choice for a hemodynamically stable patient: in this case, a proper diagnostic assessment with MRCP and ERCP to identify the biliary leak is mandatory before proceeding with any treatment.<sup>5,11</sup> Minimally invasive management of BPF has been proposed since the early 2000s.<sup>8,12</sup> More recently, Marcos-Ramirez *et al.* proposed a standardization for the treatment of BPF starting with conservative management and then, in case of persistent fistula, proceeding with minimally invasive endoscopic treatment prior to surgery.<sup>4</sup> In our experience we faced the late onset of a BPF in a hemodynamically stable patient so we decided to treat the patient conservatively. Several authors unanimously agree on the use of thoracic drainage as the first-step approach to soothe dyspnea and prevent thoracic complications onset.<sup>4,8,9</sup> According to this, we proceeded with the placement of a chest tube. Nevertheless, the persistence of biliary output in the following days requested further diagnostic and therapeutic procedures. An assessment with MRCP was first performed and then the patient underwent ERCP. However, in our case, neither MRCP nor ERCP were able to detect the site of the injury. Anyway, we decided to proceed with sphincterotomy and common bile duct stenting. The release of the sphincter of Oddi can indeed determine a significant reduction of the pressure gradient at this site.<sup>13</sup> This procedure, along with the common bile duct stenting, enhances the physiological bile flow and reduces the pressure inside the biliary tree promoting the healing of the fistula. After ERCP was performed we obtained resolution of the BPF in 24 hours as expected according to other literature data.<sup>14</sup> However, even though ERCP is a minimally invasive procedure, it is not free from complications that are mostly represented by cholangitis, pancreatitis, and stent migration.<sup>15</sup> None of that occurred in this case and the patient was early discharged without the onset of short and long-term complications. The use of a minimally invasive technique not only supports a fast recovery but also can reduce trauma and treatment-related morbidity and mortality.<sup>2</sup> According to literature, OM is indeed related to a longer hospital stay and a high risk of local and systemic complications (*i.e.*, biloma, surgical site infections, sepsis).<sup>7,11</sup> For this reason if NOM fails or surgical exploration is needed, a minimally invasive surgical approach (thoracoscopy and/or laparoscopy) should be performed to ensure a proper exploration, the identification of the biliary leak, irrigation, and drainage.<sup>11,16</sup> A limitation of laparoscopy is the lack of possibility to carry out an exploration of the chest that seems to be mandatory in case of a bronchial involvement, byliptisis, or large diaphragm defects.<sup>2,7</sup> In our experience ERCP was successful and there was no need to resort to OM. However, in a step-up approach view, a laparoscopic exploration to perform irrigation and drainage would have been the next step according to liver trauma WSES guidelines.<sup>11</sup> Thus, the use of ERCP has proven to be both effective and safe in the treatment of BPF. Moreover, our experience seems to be consistent with other reported cases.<sup>2,4,10</sup> As a matter of fact, the evidence provided by a case report is weak and this represents a limit to our conclusion, but the uncommon presentation of this condition makes it difficult to set up higher-quality studies about BPF due to its uncommon presentation.

## Conclusions

Post-traumatic BPF is a rare and potentially life-threatening condition because of the possible development of further complications. To date, there is no consensus about the standard of care for this kind of fistulae. In the past, surgery represented the first approach in the treatment of BPF but today, after the development of minimally invasive endoscopic and radiological techniques, there seems to be agreement about non-operative management as the first-line approach. This conclusion is based on the existing literature data that are represented mostly by case reports and case series. Our paper can be an addition to these reports to improve the understanding of the management of BPF. Therefore, according to literature, conservative management and ERCP should always be attempted before considering more invasive strategies, especially in the absence of any detectable biliary leakage. Anyway, to confirm these conclusions, further evaluations are still necessary.

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