

# Biliopleural fistula with cholethorax

## *A rare complication of percutaneous transhepatic biliary drainage*

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### ABSTRACT

يؤدي الناسور الصفراوي الجنبى إلى تجمع الصفراء في الصدر وهو اختلاط نادر الحدوث للتصريف الصفراوي عبر الكبد. ويكون العلاج المثالي لهذا الاختلاط بالعلاج التحفظي حيث يتوقف هذا الناسور تلقائياً عند معظم المرضى ولكن في بعض الحالات المعقدة يتطلب الأمر تدخلاً جراحياً. قد نوجز هنا حالة مسجلة لمريض يبلغ من العمر 35 سنة مُصاب بناسور صفراوي جنبى مع وجود الصفراء في جوف الصدر. وبعد إجراء التصوير الصفراوي عبر الكبد، شكَا المريض من ضيق في التنفس ووجود ألم صدري في الجهة اليمنى مع سعال بعد نزع قسطرة التصريف. وقد أظهرت صورة الصدر الإشعاعية وجود انصباب جنبى شديد وتم التأكد من التشخيص عندما تم سحب سوائل خضراء داكنة اللون من الجوف الجنبى حيث كان تركيز البيلوروبين فيها عالياً. وتم علاج الحالة مبدئياً بوضع أنبوب في الصدر للتصريف بشكل ناجح، كما وأجري للمريض تنظيف بالفيديو للصدر، وسوف تتم مناقشة طرق معالجة الناسور الصفراوي الجنبى، وكذلك مناقشة المقالات الطبية المتعلقة بذلك.

Biliopleural fistula leading to cholethorax formation is a rare complication of percutaneous transhepatic cholangiography (PTC). Ideally it should be treated conservatively as it closes spontaneously in most patients. However, in complicated cases it may require surgical intervention. We report a 35-year-old male with right biliopleural fistula with cholethorax following PTC. The patient complained of shortness of breath, right pleuritic chest pain, and cough after removal of PTC catheter. Chest radiograph showed an extensive right pleural effusion. The diagnosis was confirmed by drainage of dark green pleural fluid with high bilirubin content. The initial treatment with chest tube and drainage was unsuccessful. He underwent video assisted thoracoscopic intervention. Diagnostic modalities and management options for biliopleural fistula are discussed along with a review of literature.

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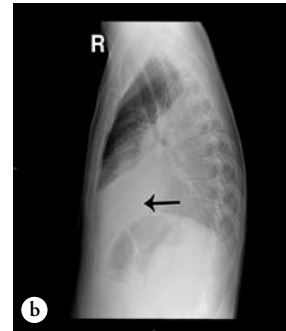
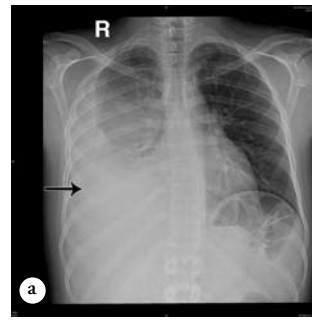
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**B**iliopleural fistula and the formation of bilious pleural effusion or cholethorax is a rare complication of percutaneous transhepatic cholangiography (PTC).<sup>1,2</sup> It occurs when the pleural cavity is traversed during the percutaneous procedure to gain access to the biliary tract. The fistula usually closes spontaneously. However, in complicated cases it may require surgical intervention.<sup>3,4</sup> Here, a rare case of biliopleural fistula with cholethorax after PTC is presented in which the treatment was initially conservative followed by video assisted thoracoscopic intervention. Our objective in presenting this case is to increase the awareness of physicians and interventional radiologists of this rare complication that may lead to significant morbidity and even mortality. A brief review of the literature is also presented.

**Case Report** A 35-year-old patient presented to Emergency Department (ED) complaining of right upper quadrant abdominal pain associated with yellowish discoloration of sclera, dark urine, and pale stool since 4 months. One year ago, he underwent cholecystectomy complicated by complete injury of the common hepatic duct, for which end-to-side hepaticojejunostomy was performed at that time. Physical examination revealed jaundiced skin and sclera with mild tenderness in the right upper quadrant of the abdomen. Laboratory values were as follows: white blood cell count (WBC)  $9.5 \times 10^9/L$ , hemoglobin 13.2g/dL, platelet count  $236 \times 10^9/L$ , aspartate aminotransferase (AST) 135 U/L, alanine aminotransferase (ALT) 176 U/L alkaline phosphatase (ALP) 436 U/L, total bilirubin 221  $\mu\text{mol/L}$ , direct bilirubin 186  $\mu\text{mol/L}$ , and a normal coagulation profile. Ultrasound (US) abdomen revealed marked intrahepatic biliary dilatation.

Patient underwent PTC under US guidance with insertion of an external biliary drainage catheter at the right mid-axillary line. Percutaneous transhepatic cholangiography showed severe stricture at the anastomotic site of previous hepaticojejunostomy. Percutaneous dilatation was unsuccessful (Figure 1). Three weeks later, after improvement of liver function

test (LFT), patient underwent surgical revision of the hepaticojejunostomy. Postoperative PTC on day 5 after surgery showed a free flow of contrast from the biliary tract to the roux loop of jejunum. Percutaneous transhepatic cholangiography tube was clamped on day 6 and removed on day 9 after the surgery. The patient was asymptomatic with almost a normal LFT and he was discharged the following day. Seven days after discharge from the hospital, he returned to the ED complaining of right sided pleuritic chest pain, shortness of breath, and cough. Physical examination revealed a temperature of 38.2°C, and diminished breath sounds in the right side of the chest. Laboratory studies revealed WBC of  $12.9 \times 10^9/L$  with a normal LFT. Chest radiograph showed an extensive right pleural effusion (Figure 2). Pigtail catheter was inserted immediately in the right pleural space under US guidance with drainage of 1345 ml dark green fluid (bile) (Figure 3) followed immediately by insertion of a 28F chest tube connected to underwater seal suction. Biochemical analysis of pleural fluids revealed: total bilirubin 287  $\mu\text{mol/L}$ , and direct bilirubin 198  $\mu\text{mol/L}$ . Chest pain and shortness of breath were relieved after drainage of large amount of bilious pleural fluid. Hepatobiliary scintigraphy was carried out on day 5 after admission showed prompt hepatic uptake and excretion of radioisotope into the intestine without delay and no evidence of radioisotope in the right hemithorax (Figure 4). Based on the clinical findings and a pleural fluid/serum ratio of total bilirubin more than one, diagnosis of biliopleural fistula with cholethorax was made.<sup>2</sup> Cultures of the pleural fluid was positive for *Escherichia coli* which was sensitive to Cefuroxime. Antibiotics (Cefuroxime) were administered and drainage maintained. Initially the drainage was 150 to 200 ml/day then it diminished over 11 days after admission. Patient continued to have spikes of fever. He underwent CT scan chest which showed significant right pleural effusion with collapse of the lower lobe of right lung (Figure 5). He underwent video assisted thoracoscopic evacuation of the pleural



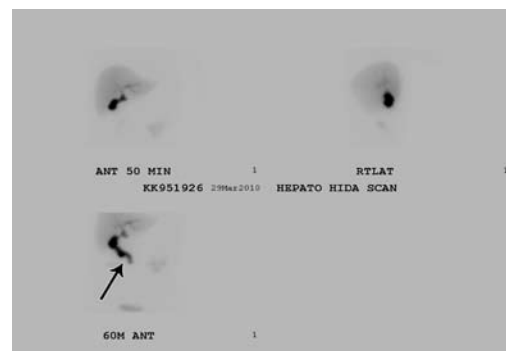
**Figure 2** - Chest radiographs showing a) frontal extensive pleural effusion (arrow) and b) lateral extensive pleural effusion (arrow).



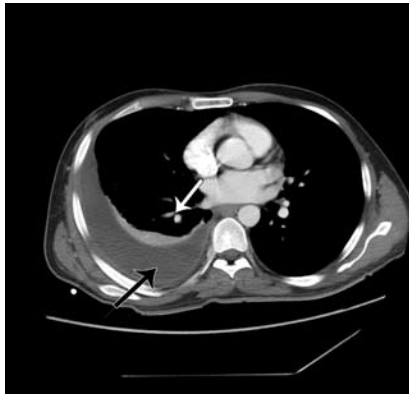
**Figure 3** - Thoracentesis fluid demonstrating dark green color (bile).



**Figure 1** - Image of the percutaneous transhepatic cholangiography showing dilated intrahepatic bile ducts with stricture at the site of previous hepaticojejunostomy.



**Figure 4** - Hepatobiliary scintigraphy showed excretion of the radioisotope to the roux loop of jejunum (arrow) with no evidence of radioisotope in the right hemithorax.



**Figure 5** - A computed tomography scan showing right pleural effusion (black arrow) with mild adjacent collapse of the lower lobe of right lung (white arrow).

effusion, decortications, and insertion of 2 chest tubes. This was followed by complete cessation of drainage and a normal chest radiograph within one week. Chest tubes were removed and patient was discharged. During the follow up in the outpatient department 4 months after discharge from the hospital, the patient remains asymptomatic with a normal chest radiograph and laboratory values.

**Discussion.** Biliopleural fistula is a rare complication following PTC. It has also been reported after percutaneous transhepatic cholangioscopy,<sup>5</sup> percutaneous transhepatic gallbladder drainage,<sup>6</sup> percutaneous radio-frequency ablation of metastatic colon cancer in the liver,<sup>7</sup> transarterial chemoembolization in a patient with large HCC,<sup>8</sup> and blunt and gunshot thoraco-abdominal trauma.<sup>9,10</sup> Percutaneous transhepatic cholangiography with biliary drainage is used in patients with obstructive jaundice where ERCP is inappropriate or has been unsuccessful. It involves the percutaneous cannulation and placement of drainage catheter to decompress the obstructed biliary tract. The potential for pleurobiliary fistula and cholethorax formation exists because the pleural cavity can be traversed during the procedure of PTC to gain access to the biliary tract. Elevated pressure gradient in the biliary tract, could drive the bile to leak back into the pleural cavity.<sup>5</sup> The likelihood of fistula formation between the biliary tract and pleural cavity increases with the duration of catheter in place.<sup>2</sup> In this patient who presented with obstructive jaundice, the PTC catheter remained for more than 4 weeks in place which may have contributed to fistula and cholethorax formation. Early diagnosis of biliopleural fistula can reduce complications which may require surgery.<sup>2</sup> It should be suspected in any patient developing right sided pleural effusion after PTC. Patients usually present

with right sided pleuritic chest pain and shortness of breath with or without fever. Fluid (bile) collection in the pleural cavity can be identified by chest radiograph, ultrasound, CT, and radionuclide scan.<sup>3</sup> Thoracentesis will confirm the diagnosis by demonstrating the bile in the pleural fluid with bilirubin level higher than that in the serum.<sup>2</sup> In this patient the drained fluid was dark green (bile) with bilirubin level of 287  $\mu\text{mol/L}$ . Essential steps in the management of biliopleural fistula and cholethorax include rapid thoracentesis, analgesia, correction of the underlying cause of the fistula, and treatment of infective complications.<sup>3</sup> Early endoscopic or percutaneous biliary drainage is important for successful management of biliopleural fistula.<sup>2</sup> Endoscopic drainage was precluded in our patient because of the previously performed end-to-side hepaticojejunostomy. Furthermore, in this patient hepatobiliary scintigraphy showed excretion of the radioisotope to the intestine without delay and there was no evidence of radioisotope in the right hemithorax. This indicated no obstruction at the hepaticojejunostomy site, and a possibility for spontaneous closure of the biliopleural fistula. Drainage of pleural cavity can be carried out conservatively by thoracostomy drainage tube. However, it may warrant surgical intervention (open or video-assisted thoracic surgery) in complicated cases.<sup>4</sup> In this patient, the initial attempts to drain the pleural effusion with pigtail catheter and chest tube were unsuccessful. The patient then underwent video assisted thoracic surgery with drainage of the pleural effusion, pleural decortications and insertion of 2 chest tubes. The fistula healed in 2 weeks.

In conclusion, biliopleural fistula with cholethorax should be suspected in a patient who develop right sided pleural effusion after PTC. This complication should initially be treated conservatively. Surgical treatment should be offered if the conservative treatment has failed.

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