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Treatment of Biliary-Pleural Fistula as a Severe Complication after Percutaneous Transhepatic Biliary Drainage

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Abstract

Introduction: Percutaneous Transhepatic Biliary Drainage (PTBD) is the most commonly used interventional treatment in patients with biliary obstruction. Biliary-Pleural Fistula (BPF) and the formation of bilious pleural effusion is a rare and severe complication of PTBD. We report the successful treatment experience of 2 patients with severe complications of BPF after PTBD. We hope to give reminders to the interventional radiologists and reference in treatment for such cases.

Case Report: BPF and the formation of bilious pleural effusion occurred in two patients when the biliary drainage tube was removed after PTBD. Thoracic drainage is the first choice as a conservatively therapy for BPF, and the biliary puncture and drainage were performed. Through thoracentesis to drain bile-like pleural effusion, the existence of BPF and the formation of bilious pleural effusion can be observed. When we performed re-PTBD, the patients were eventually cured.

Conclusion: Taking another form of biliary drainage as soon as possible is the most important factor in the successful management of BPF. Accurately judging the lower boundary of the right pleural cavity before PTBD or puncturing the left bile duct can avoid the occurrence of BPF to the greatest extent.

Keywords: Biliary pleural fistula; Bilious pleural effusion; Percutaneous transhepatic biliary drainage; Complications; Interventional radiology

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Introduction

Biliary obstruction can have various benign and malignant etiologies. The most common etiology is choledhocholithiasis or gallstone. Pancreatic adenocarcinoma and cholangiocarcinoma are the main causes of Malignant Biliary Obstruction (MBO). Because obstructive jaundice may be the first manifestation of the underlying disease, it is difficult to determine early diagnosis, so that approximately 70% of MBO patients cannot be surgically removed at the time of diagnosis. Quickly relieve the symptoms of biliary obstruction, improve liver function and nutritional status are the primary problems for prolonging the patient survival [1,2].

Currently, the Percutaneous Transhepatic Biliary Drainage (PTBD) has become widely used for either temporary or definitive relief of MBO, which permits biliary tree decompression and recovers physiological bile flow. It resulted in better therapeutic success rate, and lower incidence of overall complications, such as intraperitoneal bile leak, 30 day mortality, sepsis and duodenal perforation [3]. The incidence of pleural injury was 0.5% during PTBD placement [4]. Biliary-Pleural Fistula (BPF) and the formation of bilious pleural effusion is a rare and severe complication of PTBD. We have encountered two clinical cases, both of which recovered after re-percutaneous liver puncture and biliary decompression and drainage. We hope to be able to increase the awareness and treatment ability of interventional radiologists for this serious complication through the successful treatment experience of two patients.

Case Series

Case 1

A 55-year-old female was diagnosed with gastric cancer and underwent subtotal gastrectomy in other hospital one year ago. The pathological diagnosis after surgery was adenocarcinoma. She

Figure 1: A) The enhanced CT scan of the upper abdomen revealed nodular enhancement lesions (white arrowhead) in the ampulla at the lower end of the common bile duct, and the pancreatic duct was slightly dilated. B) During PTBD operation, the puncture point was the 9-10 intercostal space on the right anterior axillary line. Cholangiography showed stenosis and truncation of the lower end of the common bile duct, dilated biliary system. C) A bare biliary stent (white arrowhead) was implanted in the common bile duct and the biliary drainage tube was removed. D) CT scan of the chest showed a large amount of pleural effusion with right lower lung atelectasis. E) CT scan showed that the abdominal effusion increased, especially the perihepatic effusion. The liver parenchyma was compressed and shifted inward (white arrowhead), and the hilar bile duct was slightly dilated. F) The right lower hepatic bile duct was punctured in the intercostal space below the original puncture point. It can be shown that the contrast agent flowing into the right pleural cavity through the bile duct and intrahepatic puncture tract of the original puncture (white arrowhead) on the cholangiography, and the BPF is confirmed.

received five courses of intravenous chemotherapy after surgical resection. She was referred to the hospital with yellowish sclera and urine associated with white clay-like stool, and biliary obstruction was considered for clinical diagnosis. Laboratory values were as follows: Total Bilirubin (TBIL) 236.8 μmol/L, Direct Bilirubin (DBIL) 192.4 μmol/L, Carcinoembryonic Antigen (CEA) 192.4 μmol/L, Carbohydrate Antigen 19-9 (CA19-9) >1000 U/ml. The enhanced CT scan of the upper abdomen revealed nodular enhancement lesions in the ampulla at the lower end of the common bile duct, the upper common bile duct and intrahepatic bile duct branches were dilated, and the pancreatic duct was slightly dilated (Figure 1A). According to CT imaging findings and tumor markers, it is judged that obstructive jaundice may cause by ampullary carcinoma. There is not any absolute contraindication to interventional therapy after admission to the hospital to complete relevant examinations. PTBD was carried out to reduce biliary pressure and improve liver function. The puncture point was the 9-10 intercostal space on the right anterior axillary line (Figure 1B). Five days after the operation, the patient's stool turned yellow, and the blood biochemistry was recorded: TBIL 94.5 μmol/L; DBIL 79.4 μmol/L.

Seven days after PTBD, the general condition of the patient improved considerably, and then the biliary stent implantation was performed (Figure 1C). During the process of retaining the guide wire and removing the drainage tube to exchange the biliary stent conveyor, the patient complained of palpitation, chest tightness and discomfort. Electrocardiogram monitoring showed that heart rate was 120 beats/min, blood pressure was 130/85 mmHg, and no special treatment was granted. Finally, the common bile duct stent implantation was successfully completed, and the biliary drainage tube was removed.

On the 4th day after biliary stent implantation, the patient relapsed with palpitation and chest tightness, with a heart rate of 145 beats/min. Emergency laboratory studies revealed: TBIL 83.96 μmol/L, DBIL 72.63 μmol/L, WBC 24.39 × 10^9/L with 95.2% neutrophils. CT scan of the chest and abdomen showed a large amount of pleural effusion with right lower lung atelectasis, and a small amount of ascites, part of the intrahepatic bile duct had air accumulation without dilation, indicating that the biliary stent was unobstructed (Figure 1D). Right pleural puncture tube drainage was given urgently, and the dark green pleural effusion was extracted. Combined with the physical properties of the pleural effusion and the appearance after the removal of biliary drainage, the biliary pleural effusion was considered. Routine examination of pleural effusion: Brown, nucleated cell counts 30 × 10^6/L, a large number of impurities can be viewed under the microscope. After about 1,000 ml of dark green pleural fluid was drained daily, the patient's symptoms of palpitation and chest tightness were significantly relieved.

Despite anti-infective treatment has been given, the patient still has intermittent fever, with the highest temperature of 39.3°C. On the 8th day after the removal of the biliary drainage tube, the patient suddenly experienced severe abdominal pain. The emergency CT scan showed that the right pleural effusion and right lower lung atelectasis were significantly improved compared with 4 days ago, but the abdominal effusion increased, especially the perihepatic effusion increased. The liver parenchyma was compressed and shifted inward, and the hilar bile duct was slightly dilated (Figure 1E). She was given abdominal puncture and drainage again, and the dark brown ascites were extracted. *Klebsiella pneumoniae* was cultured from ascites. Imipenem and cilastatin sodium anti-infective treatment was replaced based on drug sensitivity test.

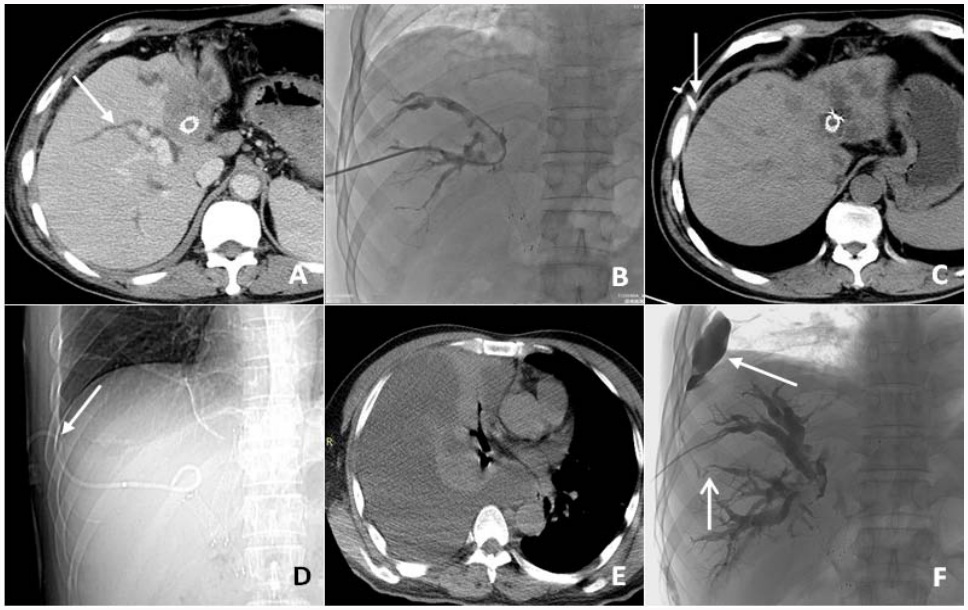

Figure 2: A) Contrast-enhanced CT of the abdomen revealed that malignant tumors in the left lobe and dilatation of the intrahepatic bile duct (white arrowhead). B) During PTBD operation, the puncture point was the 8-9 intercostal space on the right anterior axillary line. The original biliary stent occlusion can be seen on the cholangiography image. The biliary drainage tube is smoothly inserted into the right dilated intrahepatic bile duct. C, D) CT scan and film of the abdomen showed that the biliary drainage tube passed through the pleural cavity (white arrowhead). E) CT scan showed a moderate accumulation of fluid density in the right pleural cavity, and some lung tissues to the lower lobe of the right lung had consolidated and atelectasis. F) During re-PTBD operation, It can be seen that the contrast agent flow into the right pleural cavity (white arrowhead) through the punctured bile duct and the original intrahepatic puncture tract (white arrow) on the cholangiography. The BPF was confirmed.

Following consideration of the presence of biliary pleural and peritoneal effusion, the intrahepatic bile duct began to expand, indicating that biliary obstruction may still exist. PTBD was performed again on the 22nd day after the removal of biliary drainage tube. The right lower hepatic bile duct was punctured in the intercostal space below the original puncture point. The cholangiography after drainage showed that the biliary tract was blocked in the stent, and the common bile duct and intrahepatic bile duct were dilated. It can be shown that the contrast agent flowing into the right pleural cavity through the bile duct and intrahepatic puncture tract of the original puncture, and the BPF is confirmed (Figure 1F). Because the pressure of the biliary system decreased after re-puncture and drainage, we did not block the fistula. Five days after re-PTBD, the symptoms of abdominal pain and fever disappeared completely, and then the patient was discharged with a drainage tube.

Case 2

A 51-year-old man was diagnosed with primary liver cancer and liver cirrhosis after hepatitis B infection 34 months ago. He underwent PTBD due to obstructive jaundice 5 months ago. Intraoperative cholangiography showed hilar bile duct stenosis. Transcatheter intraluminal radiofrequency ablation and balloon catheter dilatation were performed in the stricture. A bare metal stent was placed in the left and right hepatic duct stricture respectively. The clinical symptoms of the patient improved after the operation.

He complained fever for 3 days with a maximum body temperature of 38.6°C. Laboratory values were as follows: WBC 2.97 × 10^9/L with 73.1% neutrophils, C-reactive protein level 66.99 mg/L, TBIL 169.3 μmol/L, DBIL 112.0 μmol/L. Contrast-enhanced CT of the abdomen revealed: malignant tumors in the left lobe and hilar of the liver with tumor thrombus in the left portal vein and dilatation of the intrahepatic bile duct (Figure 2A). The clinical diagnosis was malignant obstructive jaundice, and PTBD was performed to reduce jaundice (Figure 2B). On the 3rd day after the operation, the drainage tube drained out bright red bloody liquid, and biliary hemorrhage was considered. The patient's vital signs and hemodynamics are stable. Under fluoroscopy, the biliary drainage tube has slipped outward about 1 cm, and the drainage tube is adjusted. On the 8th day after PTBD, CT scan of the abdomen showed that the biliary drainage tube passed through the pleural cavity (Figure 2C, 2D). On the 34th day after PTBD, the patient suddenly developed severe abdominal pain after sneezing. The drainage tube slipped and shifted outward, and flushing the biliary drainage tube is not clear. Through the drainage tube for cholangiography, fluoroscopy showed that the initial biliary drainage tube fell off and moved away from the intrahepatic bile duct, trying to adjust the drainage tube into the correct location failed. So that the biliary drainage tube had to be removed finally. After that laparoscopy exploration was performed under general anesthesia. Laparoscopy showed severe adhesions in the abdominal cavity and no bile leakage at the puncture point on the liver surface. After the operation, the patient had no chills and fever, and only left a mild right upper abdominal pain.

The patient felt dyspnea and suffocation on the 3rd day after the biliary drainage tube was removed. Physical examination showed wheezes in the left lung and low breath sounds in the right lung. Chest CT scan showed a moderate accumulation of fluid density in the right pleural cavity, and some lung tissues to the lower lobe of the right lung had consolidated and atelectasis (Figure 2E). Drainage was placed on the right thoracic cavity, and the yellow-brown pleural effusion was drawn out. Laboratory values of pleural effusion as follows: Appearance: Yellow, transparency: Turbid, protein: Positive, WBC: 1.010 10^9/L; red blood cell: 0.016 × 10^{12}/L; nucleated cell number: 1.010 × 10^9/L. Biochemical studies of pleural fluid revealed: Chlorine 106.1 mmol/L, total protein 15.4 g/L, lactate dehydrogenase

491 U/L, adenosine deaminase 2; glucose 8.5 mmol/L. A week later, examine the abdomen with color Doppler ultrasound showed that the outer bile duct of the left lobe of the liver was dilated, the widest was about 4.1 mm, the inner diameter of the right lobe of the bile duct was about 4.1 mm to 5.3 mm, and the inner diameter of the upper part of the common bile duct was about 10 mm. Re-examination of chest CT revealed that there was still a large amount of pleural effusion in the right pleural cavity with atelectasis of the right lung. Consider BPF and biliary pleural effusion due to persistent biliary obstruction. In order to mitigate the biliary obstruction, PTBD was performed again on the 17th day after the drainage tube was removed. Percutaneous puncture into the other branch of the right hepatic bile duct. The cholangiography showed that the right hepatic duct in the hilar area was obstructed and the stent in the common bile duct was occluded. It can be seen that the contrast agent flow into the right pleural cavity through the punctured bile duct and the original intrahepatic puncture tract. The BPF was confirmed (Figure 2F). No treatment for fistula tract closure. 17 days after re-PTBD, the patient's general condition improved significantly, the right pleural effusion disappeared, and the liver function recovered.

Discussion

Biliary-pleural fistula is pathological communications between the biliary tract and the pleural space [5]. It was reported as a complication after percutaneous liver biopsy as early as 1970 [6]. In recent years, there have been reports of BPF or bronchobiliary fistula after radiofrequency ablation of liver tumors near the diaphragm [7-9]. BPF formations and resulting bilious pleuritis is a recognized complication following percutaneous hepatic and biliary intervention. It is a rare and severe complication after PTBD, because the patient is in distress, with pain in the lower right part of the chest, high fever, irritating cough, Acute Respiratory Distress Syndrome (ARDS) and a persistent or delayed onset right pleural effusion, etc. The clinical manifestations resemble recurrent pneumonia. Rapid accumulation of bilious pleural effusion may cause dyspnea, hypoxemia, pleuritic chest pain, respiratory failure, and bacterial empyema [4,5,10]. Bile has a corrosive effect upon the lung and pleural space and persistent flow can cause severe lung damage, such as pleural fibrosis, pneumonitis, and pulmonary fibrosis. When BPF is accompanied by infection, appropriate diagnosis and prompt treatment are extremely important.

PTBD is a safe and well-established technique for treating obstructive jaundice or biliary leakage, where ERCP is inappropriate or has been unsuccessful [11,12]. The most common post-interventional complications included the falling out of the drain from the biliary ducts and sub capsular or perihepatic leakage after PTBD [13]. Because the liver is anatomically surrounded by the right thoracic cavity across the diaphragm, PTBD may involve penetration of the thoracic cavity. Lung injury is easily avoided under video fluoroscopy and ultrasonography during PTBD. Nevertheless, Starting at the xyphoid process, pleural reflections descend across the costochondral junction of the 8th rib down to the 10th rib. Posteriorly, the reflections end at the level of L1 [11]. PTBD is typically performed at the 9th to 11th intercostal space. It may cause the puncture tract to pass through the pleural cavity and diaphragm at the same time. While the right costophrenic angle is deeper, especially when the diaphragmatic crura have a mutation, it can be located between the liver and the diaphragm and the right abdominal wall. When puncturing the right liver bile duct, the puncture tract often passes through the pleural cavity and the diaphragm. If the puncture point is selected below the 10th rib and anterior to the midaxillary line, the chance of crossing the pleura will be minimized. Dealing with asthenic or athletic patients, the dimensions of the pleural reflections can easily be underestimated leading to an accidental transpleural approach. Fluoroscopy scans in inspiration are not difficult to obtain and can help for planning the percutaneous approach to avoid traversing the pleural cavity [11]. Bile leakages into the pleural space resulting in a biliary pleural fistula occur due to the negative pressure in the pleural space, especially when the sinus is not completely closed [14]. In some cases, even if the bile duct and the pleural cavity are not penetrated during PTBD, bile that has accumulated around the liver can enter the pleural space through congenital diaphragmatic defects or connective tissue sheaths of the mediastinum [4].

Common causes of BPF include the following possibilities, such as complete biliary obstruction, placement of a catheter between the 9th and 10th ribs of the mid-axillary line, and prolonged drainage preceded by fistula formation [15]. The primary factor resulting in fistula formation related to duration of catheter placement. Fistula formation often occurs within 3 wk of catheter placement after PTBD [12,16]. The fistulas will eventually close if there is no associated infection, malignancy or distal obstruction. When biliary obstruction is present, biliary decompression is mandatory to facilitate closure of the fistulous tract. Endoscopic therapies or re-percutaneous puncture of the biliary tract to relieve biliary obstruction and achieve drainage can obviate the need for high-risk surgery [10]. Early institutions of another form of biliary drainage appear to be the single most important factor in the successful management of BPF [15].

Once a biliary pleural fistula occurs, thoracic drainage is the first choice as a conservatively therapy. However, because of the high pressure in the bile duct and the negative pressure in the pleural cavity, bile will still enter the thoracic cavity through the sinus tract. Chest drainage ceases or dramatically decreases once an alternative route of biliary decompression is available. For patients who have previously performed end-to-side hepatojejunostomy, endoscopic drainage was precluded. Only by re-puncturing the bile duct for decompression can be the sinus tract be gradually closed and the bile thoracic fistula can be recovered. After re-puncturing the bile duct, it can be seen that the contrast agent flow into the pleural space and gathers through the original puncture sinus. When a new bile duct drainage tube is inserted, the biliary obstruction is relieved, and the original puncture sinus tract will gradually close after the bile duct pressure is reduced, thereby curing the biliary pleural fistula. There are also some authors who report that other treatments are used, such as the repair of diaphragmatic defects through video-assisted thoracoscopy, Fistula closure with the greater omentum, and the use of gelatin foam pledget or NBCA to block the intrahepatic tissue tract [11,14,17,18].

Two of our patients had biliary-pleural fistulas after the biliary drainage catheter was removed, and biliary pleural effusion appeared. Thoracic puncture extracts greenish dark or straw-colored bilious pleural effusion combined with a compatible history of injury to the biliary tract, BPF should be suspected. After re-puncturing the bile duct, it can be seen that the contrast agent flow into the pleural space and gathers through the original puncture sinus. It has confirmed the existence of BPF due to extubation. When a new bile duct drainage tube is inserted, the biliary obstruction is relieved, and the original puncture sinus tract will gradually close after the bile duct pressure is

reduced. Thereby curing the BPF finally.

Conclusion

PTBD is the most used interventional treatment in patients with biliary obstruction. Biliary-Pleural Fistula (BPF) and the formation of bilious pleural effusion is a rare and severe complication of PTBD. Thoracic drainage is the first choice as a conservatively therapy for BPF. It helps the diagnosis of BPF, which the greenish dark or straw-colored bilious pleural effusion combined with a compatible history of extubation. Once the diagnosis of BPF is considered, taking another form of biliary drainage as soon as possible is the most important factor in the successful management of BPF. The original puncture sinus tract will gradually close, and the BPF will be cured. In order to avoid the serious complication of PTBD, the extent of the pleural space must be evaluated by CT or MRCP prior to PTBD to avoid transthoracic drainage, or choose to puncture and drain the left bile duct. Reducing the placement time of the drainage tube is another key factor in decreasing the incidence of BPF.

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