

Percutaneous Treatment of a Bronchobiliary Fistula Caused by Cholelithiasis: Case Report¹

Jae Soo Kim, M.D., Jin Jong You, M.D.

Bronchobiliary fistulae are rare disorders, with inflammatory diseases of the liver, trauma, previous surgery and biliary obstruction being frequent causative factors. Endoscopic or transhepatic biliary drainage has been used successfully to avoid surgical treatment. We describe a case of a bronchobiliary fistula in a 78-year-old man with biliary obstruction caused by impacted calculi. Without surgical or endoscopic intervention, fistulae were treated by percutaneous transhepatic biliary drainage and removal of calculi, in conjunction with balloon sphincteroplasty.

Index words : Bile ducts, calculi
Fistula, biliary
Fistula, pulmonary
Interventional procedures

The development of a bronchobiliary fistula (BBF) manifests as bilioptysis (the presence of bile in sputum), and this is an unusual and serious complication of liver and biliary diseases. There are many conditions can give rise to the development of such a communication (1 - 4). Biliary lithiasis is one of these, and it is perhaps the one most amenable to percutaneous management. Endoscopic or percutaneous procedures have been used successfully to avoid surgical exploration for this disorder (1 - 3, 5, 6). However, few reports exist on the percutaneous management of BBF due to cholelithiasis. We describe here a case where BBF was secondary to an obstruction caused by impacted common bile duct stones, and this was successfully treated with percutaneous biliary drainage and stone removal with balloon sphinc-

teroplasty (7). This is the first reported case of the removal of calculi, in conjunction with balloon sphincteroplasty, for the percutaneous management of BBF due to cholelithiasis.

Case Report

A 78-year-old man was referred to us from the department of pulmonary diseases for the percutaneous management of BBF due to cholelithiasis. He was originally admitted to the hospital with paroxysmal bilioptysis. His past history revealed a healed pulmonary tuberculosis that happened about 50 years ago, and he had a chronic irritable cough and yellowish sputum for one year. There was no previous abdominal surgery or trauma. On the initial examination, the patient had no clinical cholangitis or jaundice secondary to the impacted calculus. On admission, a chest radiograph showed infiltration in the right middle lobe of the lung. The laboratory studies revealed a mild leukocytosis, a normal serum bilirubin value, and the high value of total bilirubin in the sputum sample (18 mg/dl). The CT showed diffuse

¹Department of Diagnostic Radiology, GyeongSang National University Hospital

Received April 12, 2004 ; Accepted August 17, 2004

Address reprint requests to : Jin Jong You, M.D., Department of Diagnostic Radiology, GyeongSang National University Hospital
90 Chilam-dong, Chinju city, KyeongNam 660-702, Republic of Korea.

Tel. 82-55-750-8211 Fax. 82-55-758-1568

E-mail: jjyou@nongae.gsnu.ac.kr

mild pleural thickening within the areas of infiltration in the middle lobe of the right lung (Fig. 1A), and biliary dilatations secondary to the common bile duct stones were also noted. Because of the patient's advanced age and medical contraindications, percutaneous transhepatic cholangiography and biliary drainage were performed. The cholangiogram demonstrated opacification of the bronchial tree in the right middle lobe of the lung (Fig. 1B), and there were at least three calculi with obstruction of the distal common bile duct secondary to the impacted calculus (Fig. 1C). To decompress the biliary system, percutaneous transhepatic biliary drainage was performed via a right hepatic duct, which dramatically improved the patient's violent cough. The stones took three days to go into the duodenum after the biliary drainage, and the biliary catheter was then exchanged for a modified, commercially available 8-F long vascular sheath (Terumo, Tokyo, Japan), which was modified into a J-shape to better fit the form of patient's biliary tree. We performed sphincteroplasty with a balloon catheter (18mm in diameter, 4 cm in length) (Medi-Tech/Boston Scientific, Galway, Ireland). We then used an angiographic balloon catheter (12 mm in diameter, 4 cm in length) (Medi-Tech/Boston Scientific, Galway, Ireland) to push the stones into the duodenal lumen. During the dilatation, the mild hemobilia that usually occurs in this procedure spontaneously resolved. After the calculi were pushed into the duodenum, a biliary drainage catheter was placed with its distal end in the common hepatic duct. Four days after the calculi removal, a cholangiogram displayed the complete obliteration of the BBF and the disappearance of the calculi with an adequate passage of the contrast material into the duodenum (Fig. 1D).

All the calculi were successfully pushed into the duodenum, and this resulted in the restoration of normal bile flow. The patient was hospitalized for a total of 13 days. The mild leukocytosis and violent biliptysis resolved following the biliary drainage. No significant complications such as gall stone ileus, pancreatitis, or sepsis occurred during the patient's hospitalization. No recurrent biliary obstruction or biliptysis has been observed during the two months follow-up period.

Discussion

BBF are specific clinical conditions in which the leaked bile penetrates the diaphragm and enter the bronchial tree. This condition may develop after liver

surgery, trauma, hydatid disease and choledocholithiasis, or from other causes of a biliary obstruction. The pathogenesis of BBF that's caused by bile duct obstruction probably involves a local inflammatory process (cholangitis) from the high pressure within the bile ducts; this is followed by a liver abscess or biloma development, and there is rupture into the pleural space and lung (3, 5). The presence or absence of pleural adhesions could determine if there is the appearance of a bronchobiliary fistula or a pleurobiliary fistula (3, 4). Our patient also showed mild pleural thickenings due to old pulmonary tuberculosis, which was suggestive of the presence of pleural adhesion involving the formation of a BBF.

Treatment options for BBF have traditionally been surgical (4, 8). Because for some patients surgical exploration can be difficult or contraindicated, there are various endoscopic or transhepatic biliary procedures that have been developed to avoid surgery-related complications (1 - 3, 5, 6, 9). However, very little information exists on the percutaneous management of BBF due to cholelithiasis. To the best of our knowledge, this is the first report describing the percutaneous transhepatic biliary drainage and removal of calculi, in conjunction with balloon sphincteroplasty for BBF due to cholelithiasis, with no surgical or endoscopic intervention. Endoscopic methods have been used in selected cases to seal off the fistula by reducing the intrabiliary pressure (2). Endoscopic procedures are not universally available, and these procedures are usually technically impossible in those patients having an altered anatomy of the upper gastrointestinal tract, or when the calculi are larger than 2cm. In addition, endoscopic sphincterotomy may be associated with significant complications, including the reported morbidity and mortality of 7% and 1.4%, respectively (7). Therefore, BBF due to cholelithiasis is perhaps one of the conditions most amenable to percutaneous management.

The common treatment strategy for patients with biliary fistulae and obstruction involves the re-establishment of biliary drainage into the duodenum, and this allows the high intrabiliary pressure to be released (2). The basal pressure of the sphincter is 5 - 10 mmHg greater than the common bile duct pressure (10), whereas the intrathoracic pressure is lower than the intraabdominal pressure. In the formation of a BBF, these pressure gradients probably favor bile flow into the chest. Therefore, an adequate treatment to relieve the biliary obstruction should lead to the preferential flow of bile

back into the duodenum. For this reason, a balloon sphincteroplasty and stone extraction procedure were performed. There was the restoration of the physiological bile flow, which was shown on the follow-up cholangiogram. In our patient, we did not embolize the fistulous tract because the fistula closed spontaneously following the removal of the stone.

The combined application of balloon sphincteroplasty and stone removal was successful in this patient, and we consider it considered as being safer and more effective than surgery or endoscopic treatment for patients

with choledocholithiasis (7). Our successful management of a BBF due to cholelithiasis was based on our prior experience with the percutaneous management of stone removal as a primary treatment modality for cholelithiasis. As for the technical aspect, we used a modified technique for the percutaneous removal of calculi with a balloon, as employed by Berkman et al. (7). The technique used by Berkman et al. was not so easy to perform due to the difficult passage of the fully distended balloon catheter through the dilated ampulla. When employing our method, a partial decompression

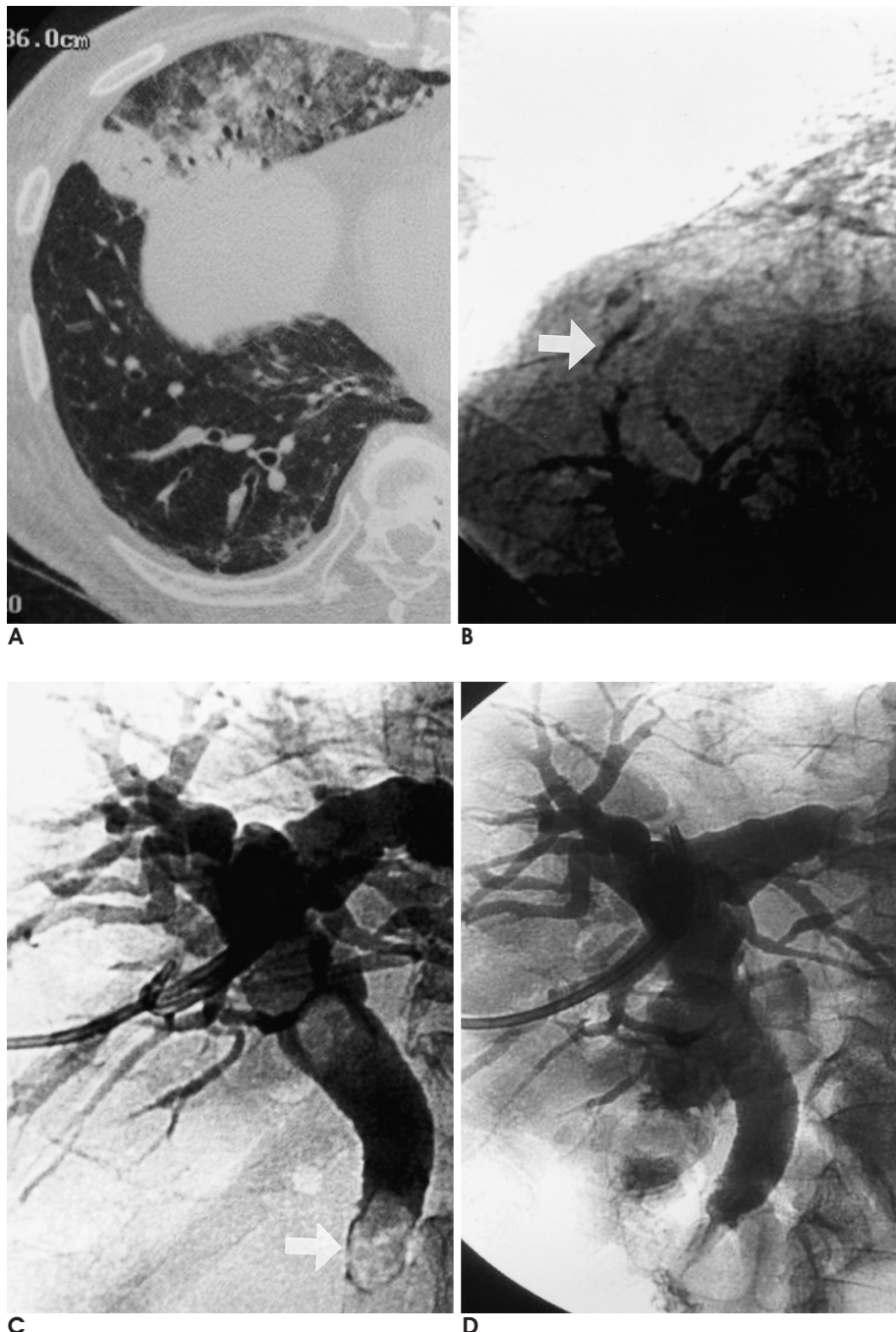


Fig. 1. 78-year-old man with paroxysmal biliptysis had a bronchobiliary fistula due to cholelithiasis. It was successfully managed by percutaneous transhepatic biliary drainage and removal of calculi in conjunction with balloon sphincteroplasty.

A. CT scan in lung window setting reveals infiltration in the right middle lobe.

B. Cholangiogram showing the biliary fistula (arrow) originating from the right intrahepatic duct and running to the right bronchial tree.

C. Cholangiogram showing three stones in the common bile duct with impaction of the distal stone (arrow).

D. Four days after the stones were removed, the final cholangiogram obtained through biliary drainage catheter shows no fistulous communication in the biliary tree and no stone in the common bile duct.

maneuver of the balloon during the pushing of the stone into duodenum and the use of a modified, J-shaped, 8-F vascular sheath were the major strong points. This simple modification gives us the ability to perform the procedure safely and rapidly as the primary treatment modality for cholelithiasis.

In conclusion, percutaneous drainage of bile and percutaneous transhepatic removal of calculi, in conjunction with balloon sphincteroplasty, was an effective management technique for BBF with choledocholithiasis.

References

1. Schwartz ML, Coyle MJ, Aldrete JS, Keller FS. Bronchobiliary fistula: complete percutaneous treatment with biliary drainage and stricture dilation. *Radiology* 1988;168:751-752
2. Memis A, Oran I, Parildar M. Use of histoacryl and a covered nitinol stent to treat a bronchobiliary fistula. *J Vasc Interv Radiol* 2000;11:1337-1340
3. Moreira VF, Arocena C, Cruz F, Alvarez M, San Roman AL. Bronchobiliary fistula secondary to biliary lithiasis. Treatment by endoscopic sphincterotomy. *Dig Dis Sci* 1994;39:1994-1999
4. Adams HD. Pleurobiliary and bronchobiliary fistulas. *J Thorac Surg* 1955;30:255-264
5. Al-Mezem SS, Al-Jahdali HH. Chronic cough due to bronchobiliary fistula. *Respiration* 1999;66:473-476
6. D 'Altorio RA, McAllister JD, Sestric GB, Cichon PJ. Hepatopulmonary fistula: treatment with biliary metallic endoprosthesis. *Am J Gastroenterol* 1992;87:784-786
7. Berkman WA, Bishop AF, Palagallo GL, Cashman MD. Transhepatic balloon dilatation of the distal common bile duct and ampulla of Vater for removal of calculi. *Radiology* 1988;167:453-455
8. Warren KW, Christophi C, Armendariz R, Basu S. Surgical treatment of bronchobiliary fistulas. *Surg Gynecol Obstet* 1983;157:351-356
9. Feld R, Wechsler RJ, Bonn J. Biliary-pleural fistulas without biliary obstruction: percutaneous catheter management. *AJR Am J Roentgenol* 1997;169:381-393
10. Csendes A, Kruse A, Funch-Jensen P, Oster MJ, Ornscholt J, Amdrup E. Pressure measurements in the biliary and pancreatic duct systems in controls and in patients with gallstones, previous cholecystectomy, or common bile duct stones. *Gastroenterology* 1979;77:1203-1210

2004;51:441 - 444

