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 that 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 treatment 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 sphincteroplasty (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. Without surgical or endoscopic intervention, fistulae were treated by percutaneous transhepatic biliary drainage and removal of calculi, in conjunction with balloon sphincteroplasty. 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 sphincteroplasty (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.
bronchial tree. This condition may develop after liver sepsis occurred during the patient’s hospitalization. No significant mild leukocytosis and violent biliopysis 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 biliopysis 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.

Treatement 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 2 cm. 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 choledolithiasis. 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

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**Fig. 1.** 78-year-old man with paroxysmal bilioptysis 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 choledochoolithiasis.

References