

# The Use of Endoscopy to Treat Bronchobiliary Fistula Caused by Choledocholithiasis

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**Bronchobiliary fistula is a rare disorder consisting of the formation of a passageway between the biliary ducts and the bronchial tree. We describe an 87-year-old woman who had a bronchobiliary fistula secondary to obstruction caused by multiple common bile duct stones. The diagnosis was confirmed by the presence of bile in the sputum and transhepatic cholangiography. Successful stone extraction with balloon and basket techniques and endoscopic sphincterotomy decompressed the biliary tract and was followed by healing of the bronchobiliary fistula without surgical intervention, which has heretofore been considered the only successful means of treatment.**

**B**ronchobiliary fistula is a rare disorder associated with significant morbidity. We describe an elderly woman suffering from multiple medical ailments who had a bronchobiliary fistula resulting from biliary obstruction due to multiple calculi in the biliary tract. Although surgical repair is currently accepted as the treatment of choice, endoscopic sphincterotomy and removal of biliary calculi resulted in healing of the bronchobiliary fistula in this case.

## Case Report

An 87-year-old white woman sought treatment because of a 3-mo history of intermittent fever, chills, and progressive lethargy. She complained of diffuse abdominal pain, most severe in the epigastrium, and subsequently developed right-sided pleuritic chest pain. She had a 2-wk history of a cough productive of dark yellow sputum but denied hemoptysis, melena, and hematochezia. Her history included hypertension, congestive heart failure, atrial fibrillation, pneumonia, diverticulosis, severe osteoporosis with compression fractures, and recurrent urinary tract infections. Eight

months earlier, she had been operated on for bilateral hip fractures complicated by postoperative pulmonary emboli. During that hospitalization she had an isolated episode of cholangitis and was found to have choledocholithiasis. However, only supportive treatment was initiated at that time because of her poor physical condition.

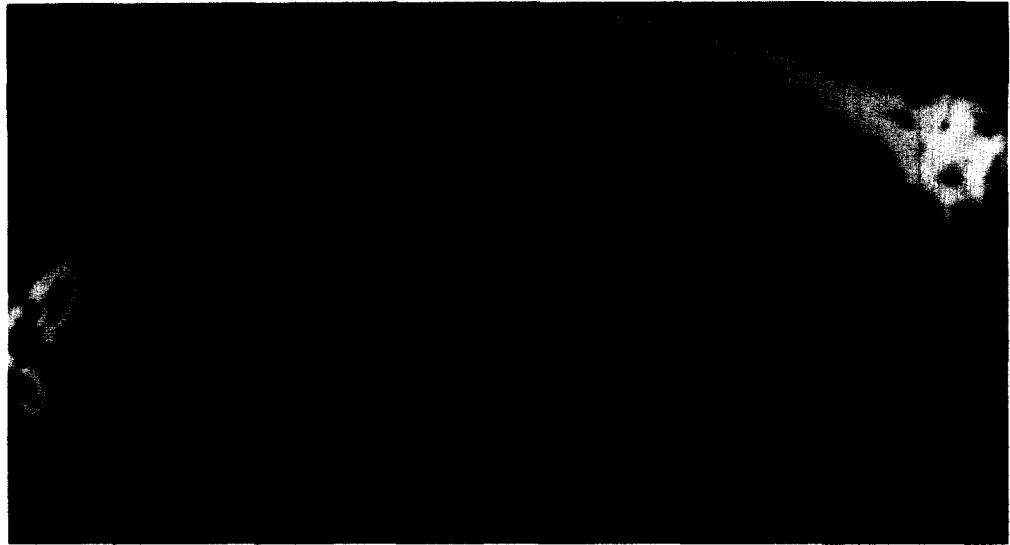
On physical examination, she was anicteric, normotensive, and febrile to 100.6°F. There were bilateral diminished breath sounds and coarse right lower lobe rales. The abdomen was distended with diffuse tenderness, most intense in the right upper quadrant. Rectal examination showed no occult blood.

The sputum was positive for bile. Results of laboratory studies included white blood cell count, 19,100/ $\mu$ L; hematocrit, 29.4%; serum glutamic oxaloacetic transaminase, 47 IU/L; serum alkaline phosphatase, 1827 IU/L; serum glutamic-pyruvic transaminase, 22 IU/L; and  $\gamma$ -glutamyl transferase, 769 IU/L. Total bilirubin was 2.0 mg/dl. Urine and blood cultures were negative. Sputum cultures grew *Klebsiella oxytoca*.

An anteroposterior chest roentgenogram showed a right lower lobe medial consolidation with bilateral pleural effusions. A transhepatic cholangiogram demonstrated reflux of contrast into the right pulmonary architecture. Computed tomography showed air and calculi in the biliary ducts. In addition, there was a small subphrenic collection consistent with the presence of an abscess (Figure 1).

The patient underwent endoscopic sphincterotomy, and multiple (>10) common bile duct stones were seen (Figure 2); most of the stones passed spontaneously. Other stones were extracted by balloon technique. A nasobiliary catheter as described by Cotton et al. (1) was placed to prevent obstruction and to permit repeated visualization of the biliary tree. The nasobiliary catheter was perfused with normal saline for 36 h, and a fluoroscopic study through the catheter demonstrated approximately 3 more stones in the common

**Figure 1.** Computed tomographic scan showing air in the markedly dilated biliary tree, a confluent density between the right liver parenchyma and the right lower base of the lung, a small calculus in the gallbladder, and at least one large, dense calculus in the common bile duct.



bile and hepatic ducts. In two additional endoscopic procedures within 3 days, using both Dormia baskets and balloon technique, the remaining stones were removed.

The patient was morbidly ill after the first procedure, in a state characterized by confusion and extreme lethargy, but

she improved dramatically after the final procedure. Her cough resolved, and within 1 wk she was discharged. She regained strength, and within 3 wk was able to live by herself. Four months after discharge, her serum alkaline phosphatase was 159 IU/L and  $\gamma$ -glutamyl transpeptidase was 289 IU/L. Lactate dehydrogenase and bilirubin levels and white blood cell count were normal. A chest roentgenogram was normal except for minimal right pleural thickening. Two years after her hospitalization she continues to do well.

### Discussion

A bronchobiliary fistula is a communication with a primary opening in the biliary tract and a second opening in the bronchial tree. The presence of bile in the sputum, "biloptysis," is suggested by bitter taste and a cough that produces yellow sputum. Laboratory proof of bile in the sputum is pathognomonic of a bronchobiliary fistula. Bronchoscopy is not necessary. A computed tomographic scan or magnetic resonance imaging study will help define the presence and location of a commonly associated subphrenic abscess. When the diagnosis is in doubt, a technetium 99m-di-isopropyl iminodiacetic acid (DISIDA) scan is the imaging test of choice (2).

The propensity of echinococcal and amoebic organisms to rupture into adjacent viscera once they have invaded the hepatic parenchyma accounts for the relatively high incidence of bronchobiliary fistulae in geographic areas where these organisms are endemic. Worldwide, these organisms are the most common cause of bronchobiliary fistula formation (3). A bronchobiliary fistula may rarely present as a congenital anomaly (4) or as a consequence of thoracoabdominal trauma (5).

In the Western hemisphere, obstructive biliary tract disease is the predominant cause of bronchobiliary fistula (6). However, it is exceedingly rare in view of



**Figure 2.** Photograph of multiple common bile duct stones at the time of the first endoscopic retrograde cholangiopancreatography. Some stones measured >2 cm in diameter.

the common occurrence of biliary obstruction in general and choledocholithiasis in particular. Nearly all biliary fistulae either are spontaneous or occur as a result of traumatic surgical procedures. Spontaneous fistula formation occurs most frequently in association with cholelithiasis, and the secondary openings occur most commonly in the duodenum, colon, stomach, or abdominal wall. For example, traumatic fistulae, those that result from biliary strictures, usually require surgical repair (7). Fifteen cases of bronchobiliary fistulae resulting from choledocholithiasis have been reported in the literature, and all of these patients either died or were treated surgically (8-11). The formation of a biliary fistula often results in decompression of the biliary tract.

The cause of the fistula must be ascertained in order to plan treatment. Cropper et al. (8) have suggested that medical management alone is rarely successful in the treatment of a bronchobiliary fistula. Recent observers have chosen a surgical approach in the treatment of bronchobiliary fistulae, including drainage of all (subphrenic) abscesses. Warren et al. (6) have further advocated closure of the diaphragmatic defect and excision of the fistulous tract. However, treatment recommendations were not based on the etiology of the fistula. In contrast, as early as 1928, Morton and Phillips (12) reported that fistulae between the biliary and bronchial trees would heal spontaneously if the biliary tract obstruction was corrected.

Biliary fistulae are an infrequent but well-recognized complication of biliary tract surgery. Del Olmo et al. reported a series of patients who developed postoperative external biliary fistulae that were successfully treated by endoscopic sphincterotomy (9). Others have reported successful treatment of biliary cutaneous fistulae by endoscopically placed nasobiliary tubes or endoprotheses (10,11). Endoscopic sphincterotomy has also been reported to be useful in the treatment of a biliary enteric fistula (13). These reports, as well as the present case, demonstrate that definitive treatment should be directed toward the underlying cause of the fistula formation. This can be accomplished by nonsurgical endoscopic management (14).

The applications of endoscopic sphincterotomy have broadened so that it is considered by many to be safer than and as effective as surgery in patients with choledocholithiasis after cholecystectomy (15). Furthermore, it is a viable therapeutic alternative in the elderly or compromised patient whose gallbladder remains in place (16,17). In our patient, medical illnesses (congestive heart failure, pneumonia, and cholangitis) precluded surgical intervention. The use of endoscopy and sphincterotomy with stone extraction in this case demonstrates the value of nonsurgical

measures in the treatment of bronchobiliary fistula associated with choledocholithiasis, illustrating the principle that decompression of the common duct allows a biliary fistula to heal spontaneously.

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