Bronchobiliary Fistula Detected by Cholescintigraphy

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We present a case of a bronchobiliary fistula initially detected by hepatobiliary scintigraphy. The patient developed bilioptysis 18 mo after undergoing a right hepatic lobectomy and resection of the common bile duct for cholangiocarcinoma. The procedure was complicated by the development of a subphrenic abscess that required percutaneous biliary drainage.

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Bronchobiliary fistula is a rare disorder, which is characterized by bilioptysis that may be congenital in origin, but which is more commonly associated with trauma, surgery, or with hepatic abscess and/or parasitic liver disease. Although surgery has been the conventional treatment, recent evidence suggests that percutaneous drainage may also be a valid therapeutic option (*1*). We present a case of bile extravasation associated with a bronchobiliary fistula detected by cholescintigraphy in a patient with bilioptysis. The physiologic nature of the hepatobiliary scan enabled the detection, localization, and the differentiation of the bronchobiliary fistula from bile reflux, the more common cause of bile emesis.

CASE REPORT

A 38-yr-old white woman presented complaining of "coughing up bile" 18 mo after having undergone a right hepatic lobectomy with resection of the common bile duct and creation of a Roux-en-Y left hepatico-jejunostomy. Pathologic examination of the resected specimens revealed the presence of mucinous adenocarcinoma with involvement of the liver capsule by local extension. Her postoperative course was complicated by a subphrenic abscess, which necessitated percutaneous drainage. Immediately after the procedure, the patient developed a right pneumothorax and a chest tube was placed. She received two courses of chemotherapy (5 FU) and 5940 total rads of local radiotherapy.

She remained asymptomatic for ~1 mo, after which time the symptoms recurred. She re-presented to her physician who witnessed the patient coughing up bilious material. Decreased breath sounds and dullness to percussion were noted at the right lung base. Her abdomen was slightly distended and shifting dullness could be elicited.

Laboratory values on admission showed an elevated total bilirubin of 2.0 mg/dl (normal: 0.0–1.2), with a conjugated fraction of 0.7 mg/dl (0.0–0.1). The GGTP measured 449 U/l (0–40) and the alkaline phosphatase measured 630 U/l (35–125). The transaminase, LDH, and CPK levels were all within the normal range. She had an elevated white blood cell count of 27.4 K/μl, with 92% polymorphonuclear cells, 7% band cells, and 1% lymphocytes on a manual leukocyte differential count. Multiple electrolyte abnormalities were also noted with a sodium level of 123 mmol/l (133–143), a chloride level of 89 mmol/l (97–107), a CO_2 content of 21 mmol/l (24–32), a glucose level of 120 mg/dl (70–110), a blood urea nitrogen of 24 mg/dl (10–20), and a creatinine level of 0.5 mg/dl (0.6–1.0).

Her chest radiograph revealed an elevated right hemidiaphragm and an infiltrate at the base of the right lung consistent with either a subpulmonic effusion or a subphrenic abscess. An abdominal radiograph showed mild ascites and possible gastric outlet obstruction. A hepatobiliary scan, performed with 5 mCi of technetium 99m-DISIDA, in order to assess the integrity of the patient's hepatobiliary system, revealed a perihepatic collection of bile with fistulous tracking through the diaphragm to the trachea (Fig. 1 A–C). Percutaneous transhepatic cholangiography (PTC) confirmed the presence of a collection of bile posterior to the liver with fistulous tracking through the lung and demonstrated its source to be a leak from the anastomotic site (Fig. 1D). A percutaneous biliary drainage catheter was placed and put on suction.

The etiology of this bronchobiliary fistula is unclear but may be related to the percutaneous drainage of the subphrenic abscess that the patient developed postoperatively. Immediately after the percutaneous drainage, the patient developed a right pneumothorax, indicating that the pleural space and perhaps the lung had been traversed before the needle pierced the diaphragm, allowing the perihepatic fluid collection to enter the pleural space. A drainage catheter was placed into the left hepatic duct percutaneously and advanced through the ductal system and then through the leak in the anastomotic site until it reached the collection of bile, at which point it was attached to suction. A few days later, a second catheter was percutaneously introduced into the left hepatic duct and advanced across the anastomotic site to end in the jejunum and act as a stent.

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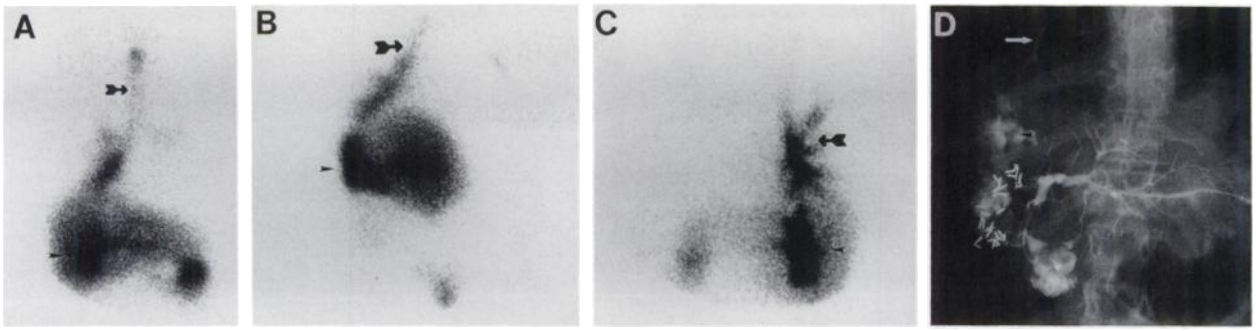

FIGURE 1
(A) Hepatobiliary scan. Anterior projection (60 min). An abnormal collection of radioactivity is seen overlying the right lobe of the liver (arrowhead) that tracts up over the dome of the liver and enters the bronchial tree (arrow). (B) Hepatobiliary scan. Right lateral projection (60 min). An abnormal collection of radioactivity is seen posterior to the right lobe of the liver (arrowhead) that tracts up over the dome of the liver and enters the bronchial tree (arrow). (C) Hepatobiliary scan. Posterior projection (85 min). An abnormal collection or radioactivity is seen overlying the right lobe of the liver (arrowhead) that tracts up over the dome of the liver and enters the bronchial tree (arrow). (D) Percutaneous transhepatic cholangiography. Percutaneous transhepatic cholangiogram performed through the left hepatic duct confirms the presence of the bronchobiliary fistula.

DISCUSSION

Although bronchobiliary fistula was first described as a complication of hydatid disease of the liver (*2*) and is most often associated with parasitic disease worldwide, in America it more commonly occurs in association with trauma, surgery, or hepatic abscess formation (*3–6*). A combination of hepatic infection (hydatid disease, amebic, pyogenic) and biliary obstruction is usually present in the nontraumatic cases. Biliary obstruction may result in cholangitis with the subsequent development of an intrahepatic abscess which progressively ruptures into the right pleural space. Early diagnosis and treatment is essential due to the high associated morbidity and mortality. Therapy usually consists of a combination of biliary drainage and treatment of the underlying condition.

In rare cases, bronchobiliary fistula may be of congenital origin, usually involving the left biliary radicals and the right main stem bronchus (*7,8*). Congenital bronchobiliary fistula may be due to duplication of the upper gastrointestinal tract or union of an anomalous bronchial bud with an anomalous bile duct (*9*).

Patients often present with bile stained sputum (bilioptysis), which is pathognomonic, and may have fever, dyspnea, cholangitis, jaundice, and sepsis. The chest radiograph usually shows opacification of the right lung base due to pleural effusion and/or bronchiolitis.

Computed tomography (CT)and ultrasonography (US) are often non-diagnostic. Bronchoscopy and bronchography usually fail to delineate the fistula. If access exists through an external component of the fistula, a pressure contrast injection (fistulogram) may be performed. However, most bronchobiliary fistulas are entirely internal. Percutaneous transhepatic cholangiography or endoscopic retrograde cholangiography (ERCP) are invasive procedures that may fail to detect small fistulas. Cholescintigraphy can evaluate the integrity of the hepatobiliary system and detect space occupying lesions such as bilomas. It can also determine whether or not the latter communicate with the biliary system (*10*), which is not always possible with CT or US, but which has important prognostic and therapeutic implications. The "physiologic" route of bile flow is demonstrated using cholescintigraphy, including the presence or absence of bile leaks, fistulas, and obstruction. In contradistinction, cholangiography is a "non-physiologic" study that requires a pressurized injection of contrast material into the biliary tree, access to which may be difficult in patients with blunt trauma.

Cholescintigraphy has been found to be a sensitive noninvasive technique for the detection and localization of bile leaks (*11–18*). The case presented here illustrates the important role that hepatobiliary scintigraphy plays in the detection and localization of bile leaks. Our experience was similar to other previous reports, in that CT was not especially helpful in making the diagnosis. Cholescintigraphy, however, identified the fluid as bile and demonstrated the bronchobiliary fistula.

Interventional angiography has played an increasing role in the percutaneous treatment of abscesses, fluid collections, and fistulas that previously required surgery. In this era of DRGs, this is an important advantage with respect to cost-effective treatment and patient morbidity. Finally, cholescintigraphy may be used in order to evaluate the effectiveness of therapy, in this case the closure of the fistula and bile leak. Important principles of therapy include the relief of obstruction and resolution of any collections by percutaneous drainage.

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