Case Report

Bronchobiliary fistula complicating open cholecystectomy

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We report a case of a bronchobiliary fistula following an intrahepatic abscess as a result of a bile leak after an elective open cholecystectomy.

CASE REPORT A 64 year old woman with history of scleroderma had an elective open cholecystectomy. One week later she developed a subhepatic bile collection. A subhepatic drain was inserted but she developed a persistent bile fistula. Endoscopy revealed a leak from the cystic duct. A 10 French gauge stent was inserted into the common bile duct beyond the level of the cystic duct.

The bile leak resolved, but two months following discharge she developed right upper quadrant pain, rigors and bilioptysis (a cough productive of bile). Chest radiograph showed a right sided pleural effusion and CT scan demonstrated a large abscess in the right lobe of the liver (Fig).

Percutaneous drainage was unsuccessful so a laparotomy was performed. A large area of the right lobe of the liver (most of segments VII and VIII) was found to be necrotic, and a defect was noted in the dome of the diaphragm, which appeared to communicate with the right chest cavity. The necrotic parenchyma was excised and two large sump drains were inserted, one into



Contrast CT scan demonstrating large subcapsular hepatic abscess in segments VII and VIII.

the abscess cavity and the other into the defect in the diaphragm.

Post operatively she developed an air and bile leak. A subsequent ERCP was performed and the plastic biliary stent removed from the bile duct. There was a suspicion of contrast in the bronchial tree on this examination. A further CT scan demonstrated a pleural effusion, diaphragmatic disruption and a collection under the diaphragm consistent with bronchobiliary fistula. Her bilioptysis settled when the drain from the defect in the diaphragm was connected to an underwater seal. The air and bile leak slowly diminished allowing removal of the drains. She remained well at follow up two months later.

DISCUSSION

Bronchobiliary fistula is a rare complication of disease of the biliary tract. Peacock first highlighted the complication in a patient with a hydatid cyst in 1850.¹ It is most frequently associated with necrotizing hepatic infections, such as amoebiasis and echinococcosis.² Other causes include subphrenic abscess, thoracoabdominal trauma including iatrogenic percutaneous biliary injury³ and suppurative complications of biliary tract obstruction. The two major aetiological factors are, therefore, infection (subphrenic or intrahepatic abscess) and mechanical bile duct obstruction with the

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latter being more common in Western countries.^{2, 4} In this case there was no evidence of biliary obstruction but the patient did have an intrahepatic abscess. The coexistence of scleroderma may have contributed to poor healing resulting in leakage from the cystic duct and subsequent abscess formation.

Presentation of a bronchobiliary fistula may range from chronic episodes of bilioptysis to a severe bronchopneumonitis. Bilioptysis is pathognomonic of bronchobiliary fistulae and, in one review of non-traumatic bronchobiliary fistulae, was present in all the patients studied.⁴ The triad of symptoms of right upper quadrant pain, fever and bilioptysis, as noted in this case, was reported in 11 of the 16 patients reviewed by Gugenheim et al.⁴

Bile stained sputum can be analysed for bilirubin concentration and in true bilioptysis it may contain up to 8 mg/dL of bilirubin. Chest X-ray findings of a right pleural effusion and a raised hemidiaphragm, as seen in this patient, are common in this condition. The finding of a diaphragmatic disruption secondary to an abscess or laceration at the liver dome with associated right pleural effusion in a patient with bilioptysis strongly suggests a bronchobiliary fistula.⁵ Cholangiography, either percutaneously or endoscopically, should be performed.⁶ Large amounts of contrast may be required at ERCP to demonstrate the fistula and the detail may not be as clear as with percutaneous imaging.⁷ Isotope scanning with HIDA may also be used,⁸ but bronchoscopy is generally unhelpful.⁴

Drainage of any abscess and relief of bile duct obstruction are the two key principles of management. Drainage of an abscess may be carried out either percutaneously or surgically. This case required surgical drainage as the necrotic material was too viscous to drain percutaneously. In patients with associated biliary tract obstruction the priority of management is towards decompression of the bile duct by means of a stent or a surgical procedure such as a Roux-en-Y hepaticodochojejunostomy.

Bronchobiliary fistulation is a rare complication of biliary disease. Diagnosis with CT scan and cholangiography followed by drainage of abscesses and biliary decompression are the key elements in the management of this potentially life threatening condition.

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