Case report

Usefulness of Tc99m-mebrofenin Hepatobiliary Scintigraphy and Single Photon Emission Computed Tomography/Computed Tomography in the Diagnosis of Bronchobiliary Fistula

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Abstract

Bronchobiliary fistula (BBF), a rare complication of liver disease, is an abnormal communication between the biliary tract and bronchial tree. BBF may occur as a consequence of local liver infections such as hydatid or amebic disease, pyogenic liver abscess or trauma to the liver, obstruction of biliary tract, and tumor. As such management of liver disease with BBF is very difficult and often associated with a high rate of morbidity and mortality. Therefore, timely diagnosis of BBF is imperative. Hepatobiliary scintigraphy along with hybrid single photon emission computed tomography/computed tomography using Tc99m-mebrofenin is a very useful noninvasive imaging modality, in the diagnosis of BBF.

Keywords: Amoebic liver abscess, bronchobiliary fistula, single photon emission computed tomography/computed tomography, Tc99m-mebrofenin hepatobiliary scintigraphy

<u>Introduction</u>

Bronchobiliary fistula (BBF) represents a rare benign clinical entity with a significant mortality risk. BBF can result from congenital etiologies^[1] or more often from acquired etiologies.^[2-4] Bile stained sputum, bile presence in the pleural effusions, or even jaundice should raise concerns for BBF in a patient with liver disease. The expectoration of bile (biliptysis) is a cardinal feature of BBF, and its presence should lead to prompt diagnosis and intervention.^[3] The single photon emission computed tomography (SPECT)/CT in a Tc99m-mebrofenin

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hepatobiliary scintigraphy is a very useful noninvasive imaging modality in the diagnostic workup for detecting BBF. We are reporting a diagnosed case of amebic liver abscess presenting with fever, chest pain, and cough with bilious sputum highly suspicious for BBF in whom hepatobiliary scintigraphy helped to detect BBF.

Case Report

A 40-year-old male patient presented with a history of fever and right-sided abdominal pain for 2 months, followed by cough with expectoration and right-sided chest pain for 20 days. His liver function tests were deranged with total and conjugated bilirubin being

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3 and 2 mg/dL, alkaline phosphatase 789 g/L, aspartate transaminase 32.12 U/L, and glutamic pyruvic transaminase 32.9 U/L, respectively. The ultrasonography (USG) of abdomen showed multiple large oval to round shape echogenic lesions in the right lobe of the liver characteristic of amebic liver abscess. Patient underwent pigtail drainage from liver abscess, and serology was positive for IgG antibody by ELISA test confirming amebic disease.

The contrast-enhanced CT (CECT) of abdomen showed multiple hypodense liver lesions in the right lobe of liver, largest measuring ~5 cm × 6 cm in segment VIII extending up to the subcapsular location with a large collection measuring 8.4 cm × 9.2 cm × 9.2 cm in right subphrenic space with air fluid level. CECT of chest showed minimal right side pleural effusion with adjacent patchy consolidation in posterior basal segment of lower lobe of the right lung. The endoscopic retrograde cholangiopancreatography (ERCP) showed contrast leak from the right biliary system with subsequent collection in right subphrenic space. Patient was subjected to biliary sphincterotomy and stenting in biliary system. CECT of chest and abdomen and ERCP did not give any clue of communication between biliary system and bronchial tree, in spite of patient complaining of cough with copious greenish expectoration. Tc99m-mebrofenin hepatobiliary scintigraphy was performed. Initial dynamic images of abdomen [Figure 1a] acquired immediately after intravenous injection of 4 mCi of Tc99m-mebrofenin showed adequate hepatocyte uptake followed by excretion of the tracer into bowel within 15 min of radiotracer injection. A streak of tracer passing cranially across the right lobe of the liver on the right side of the chest and increasing intensity with time was also

noticed. Subsequent static images of thoracic and abdomen regions [Figure 1b] showed collection of tracer in the right subphrenic space with faint tracer activity in thoracic region indicating BBF. However, dynamic and static images were not able to exactly localize any tract between bronchial segment of lung and biliary system. Hybrid SPECT/CT of thoracic and upper abdomen [Figure 2] showed exact fistulous tract between right lower lobe bronchus and biliary system with localization of tracer to a large right subdiaphragmatic collection. This information was really helpful in the decision-making and further management of the patient.

Discussion

There are two major factors responsible for the formation of BBFs:^[5,6] first being the formation of biloma that resides underneath the diaphragm, resulting from liver/diaphragm injuries, tumor, postoperative biliary stenosis, etc.^[7] and second factor is spreading of a hydatid liver cyst or amebic liver abscess to the adjacent lung or pleural space. Both these factors lead to the collection of bile underneath diaphragm, which can erode tissues and reach the pleural space, bronchus, or both resulting in the fistula formation. Similarly, in our case, amebic liver abscess adjacent to the lower lobe of the right lung lead to the formation of BBF.

The diagnosis of BBF is commonly made clinically based on the high index of suspicion due to the presence of bile in the sputum (biloptysis). The plain chest X-ray, abdominal CT, and USG may be used for the detection of BBF; however, most of the times these imaging modalities fail to demonstrate the fistulous tract. The transhepatic cholangiography or ERCP and magnetic

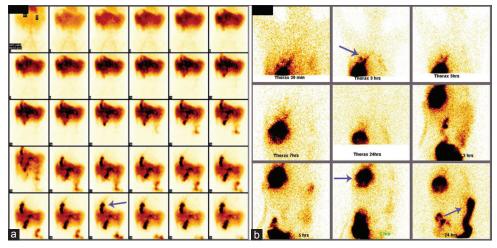


Figure 1: Tc99m-mebrofenin hepatobiliary dynamic images (a) showing adequate hepatocyte uptake and excretion of tracer into the bowel. In addition, a streak of tracer activity (arrow) is passing cranially across the right lobe of the liver on the right side of the chest and subsequent static images of thoracic and abdominal regions (b) show collection of tracer activity (arrows) in the right side of the body and excretion of tracer (arrow) into the large intestine

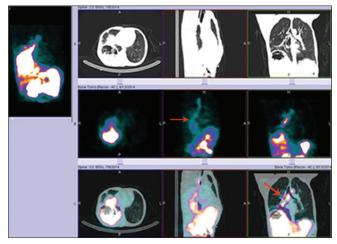


Figure 2: Hybrid single photon emission computed tomography/ computed tomography images of thoracic and abdominal regions showing localization of tracer in the right subdiaphragmatic large collection with exact visualization of communication between right lower lobe bronchus and biliary system (arrows)

resonance cholangiography helpful in demonstrating fistulous tract, but these are invasive modalities and also fail to demonstrate the fistulous tract.^[8]

Tc99m-mebrofenin hepatobiliary scintigraphy is a noninvasive, safe, simple imaging modality that can show the presence, location, and extent of bile leak. This imaging modality can demonstrate the communication between the biliary tree and an intra-abdominal or intrathoracic fluid collection which is helpful in identifying active biliary leakage. The absence of biliary leak on hepatobiliary scintigraphy indicates that the leak has either stopped or is so slow that it will likely resolve spontaneously, and no aggressive therapy is warranted. Furthermore, this study can be repeated and is useful in evaluating the response to treatment. [9,10] The hepatobiliary scintigraphy provides important information regarding the patency of common bile duct, which is very useful, as common bile duct obstruction is a major cause of BBF and its presence is a hindrance to successful recovery in these patients.[11] Similarly, in our patient, radiological imaging and other modalities failed to detect BBF. Hepatobiliary scintigraphy with SPECT/CT not only detected abnormal communication between biliary tree and bronchial system in our patient but also provided information about liver function and patency of bilioenteric system, which is essential in decision-making during the management of these patients. The hepatobiliary scintigraphy has been also successfully used to diagnose even small BBF with minimal risk to the patient.

Conclusion

Hepatobiliary scintigraphy with SPECT/CT is a very useful diagnostic tool to confirm the clinically suspected BBF, which is helpful in early intervention and treatment in such type of clinical condition.

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Conflicts of interest

There are no conflicts of interest.

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