

Case report

Role of Cholescintigraphy with Single-Photon Emission Computerized Tomography-Computed Tomography in Detecting Bronchobiliary Fistula: Unusual Complication of a Common Disease

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Abstract

Bronchobiliary fistula (BBF) is an abnormal communication between the biliary tree and airway. A patient usually presents with cough and bilioptysis, and at times, it poses diagnostic and therapeutic challenge. This case demonstrates the usefulness of cholescintigraphy with single-photon emission computerized tomography in diagnosing BBF in case of hydatid cyst of the liver.

Keywords: Bronchobiliary fistula, single-photon emission computerized tomography-computed tomography, scintigraphy, technetium-99m N-(3-bromo-2,4,6-trimethylacetanilide)iminodiacetic acid

Introduction

Bronchobiliary fistula (BBF) was first described by Peacock in 1850.^[1] Bilioptysis is one of the cardinal symptoms. Therefore, the diagnosis is based on symptoms as well as clinical history. Congenital malformations are a rare etiology for BBF; however, acquired etiologies are more common. Pathogenesis involves increased local pressure or infectious condition resulting from infected hydatid cyst, amebic liver abscess, liver trauma, or neoplasm.^[2,3] The disease may lead to deferent

pulmonary complications, such as recurrent chemical or bacterial pneumonitis, mediastinitis, bronchiolitis, or bronchiectasis.^[4] Timely, accurate diagnosis and treatment are critical to the successful outcome. Herein, we describe a rare case of cyst-biliary communication secondary to a hydatid cyst of the liver, associated with BBF and diagnosed by hepatobiliary scintigraphy and exact localization by single-photon emission computerized tomography-computed tomography (SPECT-CT). The functional nature of hepatobiliary scintigraphy enabled the diagnosis and the extent of BBF.

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Case Report

A 25-year-old male who presented with bilioptysis in the Department of Surgical Gastroenterology was

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referred to the Department of Nuclear Medicine for cholescintigraphy. He gave a history of jaundice, for which he was diagnosed as a case of hydatid cyst with choledocholithiasis. He was initially managed with endoscopic retrograde cholangiopancreatography (ERCP), common bile duct (CBD) clearance, and stenting of the bile duct elsewhere. He developed recurrent cholangitis due to stent block and later was managed surgically with open choledochotomy and extraction of hydatid cyst and T-tube drainage in other hospital. However, the stent was not removed. The patient remained asymptomatic for 4 years. Later, he developed recurrent episodes of fever and cough which was bile stained and was referred to our Center for Management. On examination, he had good body built and good performance status. Chest examination revealed crepitations in the right basal area. Abdominal examination was unremarkable except that a healed scar mark of the previous surgery. He was referred for hepatobiliary scintigraphy in view of high suspicion of BBF. Dynamic [Figure 1]

and static [Figure 2] hepatobiliary scintigraphy by 185 MBq technetium-99m N-(3-bromo-2,4,6-trimethylacetanilide)iminodiacetic acid (99mTc-mebrofenin) and SPECT depicted tracer accumulation in the right lung [Figure 3]. Contrast-enhanced CT thorax and abdomen revealed misplaced stent with coiled tip in the right subdiaphragmatic region with small collection along in vicinity of the coiled stent tip, suggestive of pus. A linear defect in mid-portion of the right dome of the diaphragm was also noticed along with peripheral bronchus of anterolateral segment of the right lower lobe of the lung. Multiple calculi were also noted in the gallbladder. He was taken up for surgery and underwent dismantling of the biliobronchial fistula, closure of the diaphragmatic rent, open cholecystectomy, CBD excision, and Roux-en-Y hepaticojejunostomy. Operative picture [Figure 4a] reveals rent in the diaphragm. Operative specimen [Figure 4b] reveals displaced stent with granulation tissue. Postoperative course was uneventful. After 2½ years of follow-up, he is asymptomatic.

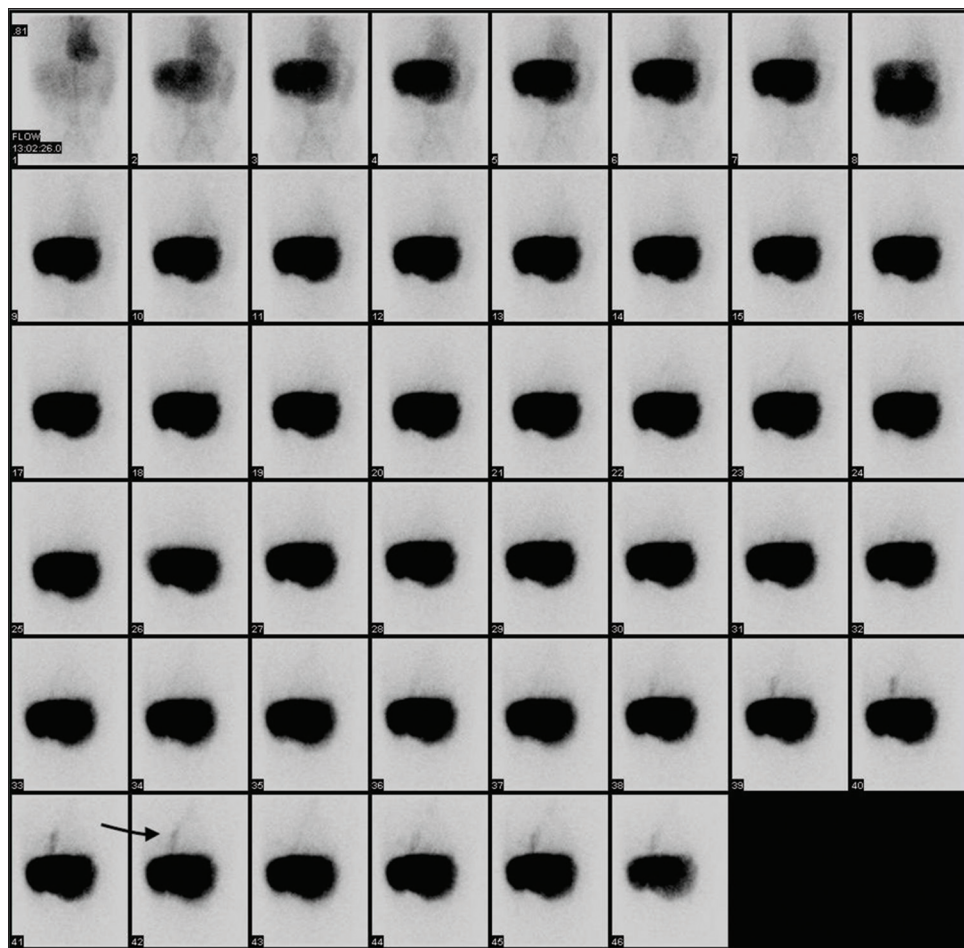


Figure 1: Hepatobiliary scintigraphy was performed with Tc-99 m N-(3-bromo-2,4,6 trimethylphenylcarbamoylmethyl)iminodiacetic acid (mebrofenin). Serial images (1 min/frame for 46 frames) reveal good hepatocyte extraction with linear area of increased tracer uptake in the right hemithorax extending from the superior surface of the liver (arrow) visualized in frame 36–46

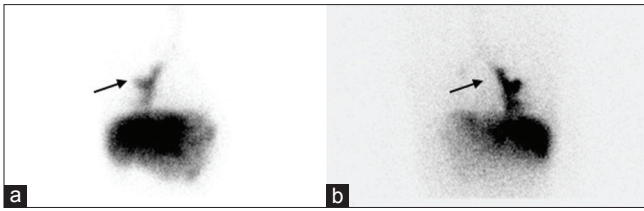


Figure 2: Delayed anterior (a) and posterior (b) static images confirm the early findings. Tracer was ascending from the superolateral surface of the liver up to the left hemithorax (arrow)

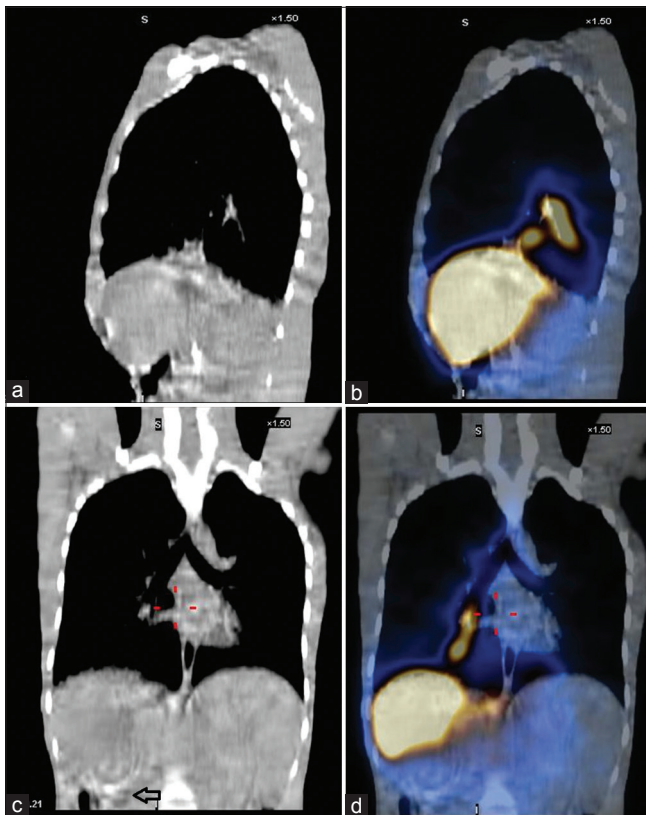


Figure 3: Single-photon emission computerized tomography-computed tomography (b and d) fused with low-dose computed tomography (a and c) images reveal increase tracer uptake extending from the superolateral surface of the liver up to the right lower lobe bronchus. Misplaced stent is noted on the computed tomography images in Figure C (arrow)

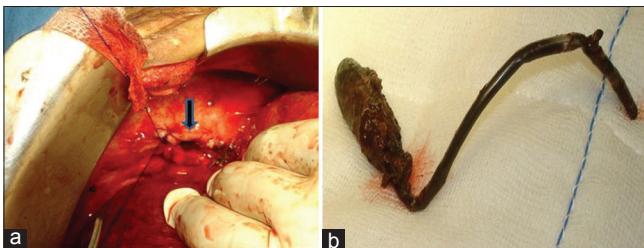


Figure 4: Operative image (a) reveals rent in the diaphragm (solid arrow). Operative specimen (b) reveals removed stent and gallbladder

Discussion

In the present case, we demonstrate the role of hepatobiliary scintigraphy with SPECT in localizing

the BBF. The likely cause of the BBF in this case was penetration of the displaced stent through the diaphragm into the bronchial tree.

For the diagnosis of BBF, the most frequently used modalities in the investigative armamentarium are CT, ERCP, magnetic resonance cholangiography, and hepatobiliary scintigraphy as used in our case. Percutaneous transhepatic cholangiography, bronchoscopy, and bronchography may also be used to confirm the diagnosis.^[5]

CT incidentally may show indirect evidence of BBF, such as subphrenic fluid collection, discontinuity of the diaphragm, bronchiectasis, atelectasis, or pleural effusion.^[6] In our case, CT and ^{99m}Tc-BrIDA both revealed the BBF. Displaced stent could be visualized on the low-dose CT of the hepatobiliary scintigraphy. Thus, demonstrating the additional advantage of SPECT-CT over conventional imaging also validated by studies by Damle *et al.*^[7]

Hepatobiliary scintigraphy is noninvasive, investigation to assess anatomy, and function of the biliary tree, the site of any bile collection and provides useful information for treatment and planning of the BBF.^[8] It can be used effectively to diagnose trivial BBF and can be repeated after treatment without posing any risk.^[9-11]

Conclusion

Bronchobiliary fistula (BFF) is associated with high mortality rate and requires a well-planned management strategy, hence carries the importance of early detection. Hepatobiliary scintigraphy with additional use of SPECT-CT stands as a robust modality in the accurate diagnosis and may help treatment planning and follow-up of BFFs.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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