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 (BFF) 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 BFF 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

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 cholecintigraphy. 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.
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.\(^\text{[5]}\)

CT incidentally may show indirect evidence of BBF, such as subphrenic fluid collection, discontinuity of the diaphragm, bronchiectasis, atelectasis, or pleural effusion.\(^\text{[6]}\) In our case, CT and 99mTc-BiDa 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 \textit{et al}.\(^\text{[7]}\)

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.\(^\text{[8]}\) It can be used effectively to diagnose trivial BBF and can be repeated after treatment without posing any risk.\(^\text{[9-11]}\)

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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Conflicts of interest
There are no conflicts of interest.

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