

# Hepatobiliary scintigraphy with SPET in the diagnosis of bronchobiliary fistula due to a hydatid cyst

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#### **Abstract**

In this report, we present the application of hepatobiliary scintigraphy using Tc-99m mebrofenin in the diagnosis of bronchobiliary fistula caused by a liver hydatid cyst, which penetrated the diaphragm. Hepatobiliary scintigraphy noticeably depicted the leakage of the tracer from the biliary system of the liver to the bronchial tree. Hepatobiliary scintigraphy stands as a robust modality in the accurate diagnosis and treatment planning of bronchobiliary fistulas.

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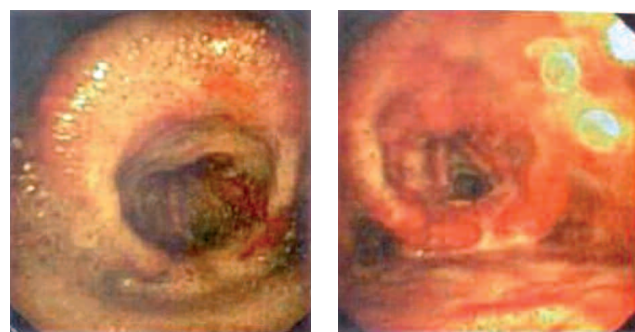
#### **Introduction**

**B**ronchobiliary fistula (BBF) is very rare and usually refers to the right bronchi [1]. Bronchobiliary fistula is defined as the abnormal communication between the biliary tract and the bronchial tree [2]. The first case was reported by Peacock in 1850 [3]. The presence of biliptysis or bile in sputum is its main clinical manifestation, crucial for proper diagnosis [4]. Unlike congenital BBF, which has a prevalence of  $<1 \times 10^6$ , acquired BBF commonly develops as a result of local pressure and/or infection, such as from a hydatid cyst, an amebic abscess of the liver, trauma, obstruction of the biliary tract, or neoplasm [5]. The disease can have different pulmonary complications, such as recurrent chemical or bacterial pneumonitis, mediastinitis, bronchiolitis, or bronchiectasis. Timely, accurate diagnosis and treatment are critical to the patients' outcome [3]. The prognosis for patients with malignancy is poor but patients with infectious etiologies after appropriate therapy have good prognosis [3]. Although biliptysis is characteristic of BBF, imaging techniques are needed to confirm the diagnosis, to delineate the site of interconnection, to diagnose the underlying disease and for treatment, planning and follow-up [6].

Herein, we describe a rare case of bile extravasation secondary to a hydatid cyst of the liver, associated with BBF and diagnosed by hepatobiliary scintigraphy. The functional nature of hepatobiliary scintigraphy enabled the diagnosis of both the cause and the extent of BBF.

#### **Case report**

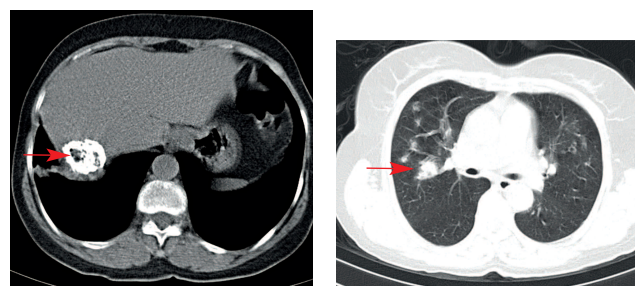
A 62 years old housekeeper woman presented with persistent cough and copious expectoration of greenish material containing biliary secretions (biliptysis). An endoscopic examination revealed bilious material in the bronchi (Figure 1), and computed tomography (CT) showed a large cystic mass with multiple small cysts inside (daughter cysts) at the base of the dome of the liver (Figure 2). Dynamic and static hepatobiliary scintigraphy by 185MBq technetium-99m N-(3-bromo-2,4,6-trimethylacetanilide) iminodiacetic acid (<sup>99m</sup>Tc-mebrofenin) and single photon emission tomography (SPET) depicted tracer accumulation in the right lung (Figure 3), while acquisition showed better delineation of the lesion (Figure 4). There was no indication of another cyst elsewhere in the body. The patient often communicated with unattended dogs. The BBF, which



VC Carina LMB



**Figure 1.** Bronchoscopy demonstrated severe bile secretion and edematous mucosa in both main bronchi.



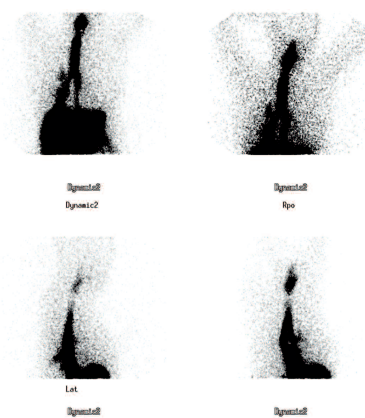
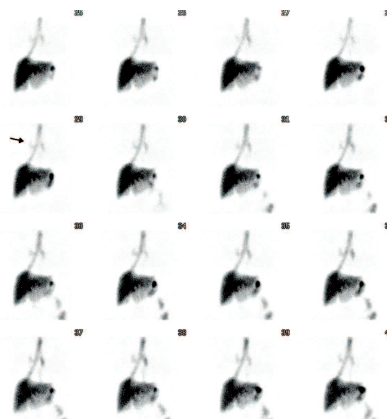
**Figure 2.** A and B: Computed tomography scan of the fistula from the right lobe of the liver entering into the right lung.

communicated with the right bronchial tree was confirmed and corrected by surgical intervention. Over the following two years, the patient was readmitted three times and given conservative treatment.

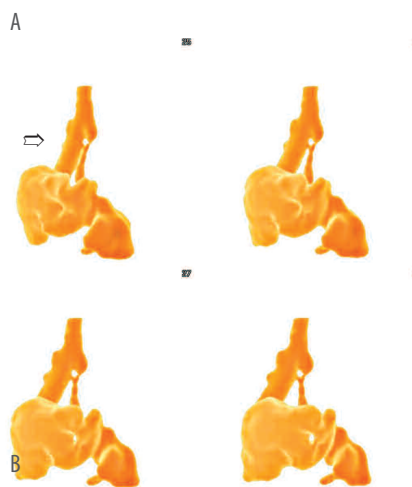
### Discussion

In the presented case, it was likely that penetration of the diaphragm developed through its invasion by the subdiaphragmatic hydatid cyst.

Computed tomography (CT), endoscopic retrograde cholangiopancreatography (ERCP), magnetic resonance cholangiography, and hepatobiliary scintigraphy are the most frequently used imaging modalities in diagnosing BBF. Percutaneous transhepatic cholangiography (PTC), bronchoscopy, and bronchography may also be used to confirm diagnosis [7].



**Figure 3.** A and B: Hepato-biliary scintigraphy. Dynamic and static imaging depicted an abnormal collection of radioactivity overlying the right lobe of the liver, tracking up over the dome of the liver and communicating with the right bronchial tree, where was accumulated over the next six hours. Serial images showed good hepatocyte uptake and excretion of the tracer into the gut.



**Figure 4.** Acquisition of the SPET images demonstrated better delineation of the fistula.

Computed tomography incidentally may show indirect evidence of BBF, such as subphrenic fluid collection, discontinuity of the diaphragm, bronchiectasis, atelectasis, or pleural effusion [8]. In the present case, CT revealed a large complex cystic mass with air-fluid levels crossing the right diaphragm [6]. Bronchoscopy cannot accurately locate BBF, especially when the BBF tracks through the diaphragm. Percutaneous transhepatic cholangiography is an invasive technique, which should be used only in patients when external percutaneous biliary fistulization has already been diagnosed [9]. Endoscopic RCP is an excellent technique for detecting BBF and for visualizing the effect of treatment of the involved biliary ducts, but is more invasive and may have significant complications compared with hepatobiliary scintigraphy [6, 10].



Hepatobiliary scintigraphy is noninvasive, shows the anatomy and function of the biliary tree, the site of any bile collection and provides information about treatment planning of the BBF [11].

Treatment of the BBF can be endoscopic, percutaneous or surgical [11].

Hepatobiliary scintigraphy has been used effectively to diagnose even trivial BBF without posing any risk to the patient and can be performed repeatedly after therapy to assess whether the fistula was closed [6]. Therefore, hepatobiliary scintigraphy seems to be the diagnostic modality of choice for BBF.

In conclusion, our case showed that hepatobiliary scintigraphy with <sup>99m</sup>Tc-mebrofenin diagnosed a hydatid cyst causing BBF and suggesting treatment planning.

The authors declare that they have no conflicts of interest.

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Cornelis Dusart, Copperplate. Applying hot cups to relieve pain.