

Case Report

## Integrated Imaging in a Bronchobiliary Fistula after Radiofrequency Ablation Treatment

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### Abstract

The bronchobiliary fistula (BBF) consists of an abnormal communication between the bronchial tree and the biliary tract that might arise as a result of an iatrogenic complication of a radiofrequency ablation treatment of liver metastases. A 74-year-old patient, who had been treated with radiofrequency ablation (RFA) for liver metastases, came to our attention with green sputum and bitter saliva. This sign was strongly suspect for a BBF. To reach a correct diagnosis of a pathological interconnection between the biliary tree and bronchial branches, several laboratory tests and imaging studies have been performed including Radiography, Contrast-Enhanced Computed Tomography (CECT) and Magnetic Resonance Cholangiography (MRC). The imaging techniques, especially the CECT and the MRC images have allowed a definitive diagnosis of the BBF as a result of a complication of RFA of liver metastases, demonstrating the pathological anatomical leak between the bronchial tree and the biliary tract.

**Keywords:** Bilioptysis; Bronchobiliary Fistula; Radiofrequency Ablation Therapy; Diagnostic Imaging Anatomy

### Abbreviations

BBF: Bronchobiliary Fistula;

RFA: Radiofrequency Ablation;

CECT: Contrast-Enhanced Computed Tomography;

MRC: Magnetic Resonance Cholangiography

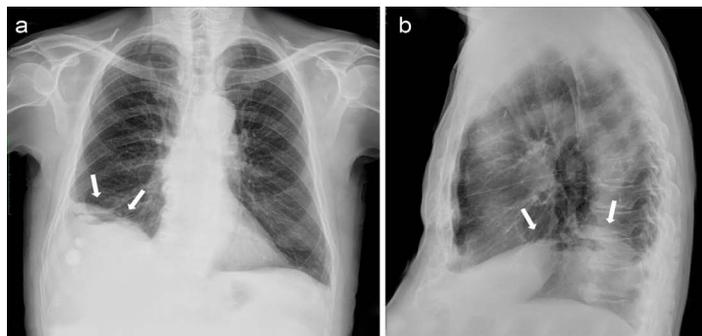
## Introduction

The BBF consists of a pathological interconnection between the biliary tract and bronchial tree, which was first reported by Peacock in 1850 [1]. Unlike the congenital form, the acquired BBF can result from liver infections (hydatid cyst) or abscess (echinococcal, amoebic or pyogenic), trauma, biliary tract obstruction, iatrogenic diseases (liver resection, RFA, bile duct stricture, irradiation, thoracic drainage, etc.) [2], or neoplasm such as the hepatocellular carcinoma [3]. Furthermore a rare case of acquired BBF has been described as result of hepatic endometriosis [4].

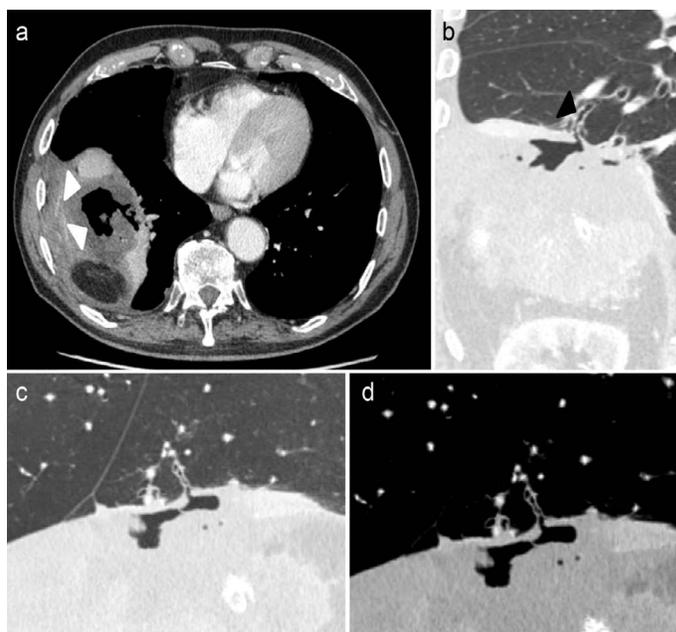
## Case report

A 74-year-old Caucasian man was admitted to the first Department of Pneumology of our Hospital with a history of persistent cough, green sputum and bitter saliva which increased especially in the supine position. He underwent surgical treatment for a prostatic carcinoma in the 2004 and, two years later, left-sided hemi-hepatectomy for hepatocellular carcinoma. After that, hepatic metastases were showed and then treated by chemoembolization technique. Because occurrence of vascular stenosis, this therapy was suspended and the patient was candidate for RFA. After about six months after several cycles of RFA, the patient arrived to our Department referring cough, greenish sputum and bitter saliva. He had no fever or jaundice and at the physical examination of the chest and the abdomen abnormal findings were not present. Inspection of the sputum showed green and fetid material, concluding for a clinical diagnosis of biliptysis and a BBF was suspected. The microbiological examination of the sputum was negative for bacteria. Laboratory studies revealed an increase of alkaline phosphatase, gamma-glutamyltranspeptidase (GGT), erythrocyte sedimentation rate (ESR) and C-reactive protein. At first, a chest Radiography showed a mixed and heterogeneous opacity at the base of the right lung (Figure 1). A subsequent CECT of the thorax and abdomen demonstrated a consolidation of the right lower lung lobe and a heterogeneous hypodense hepatic lesion with necrotic degeneration areas and bubbles gas inside, localized at the VII hepatic segment. The upper portion of the necrotic lesion appeared abnormally connected to the basal bronchial tree of the right lower lobe of the lung by a pathological leakage (Figure 2). These findings led to the strong suspicion of BBF. The MRI images and the MRC study confirmed the consolidation of the lower right lung parenchyma and clearly revealed the abnormal communication between the bronchial tree and the hepatic formation sided in the VII segment. The hepatic lesion appeared very heterogeneous because the presence of necrotic degeneration areas post-RFA and air bubbles inside for the communication with the bronchial tree (Figure 3). Flexible bronchoscopy showed bile-like material in the basal-posterior segment (B10) of the right lower bronchus and

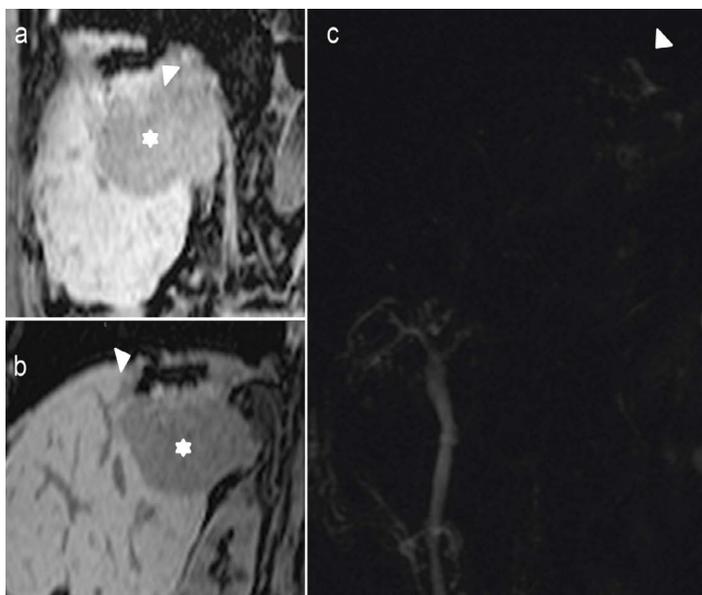
the next biochemical study of the bronchial aspiration confirmed the presence of biliary material (Figure 4). The patient underwent to a right lower lobe resection by video-assisted thoracoscopic surgery (VATS) in another Hospital, for removal the bronchobiliary fistula. He reached a prompt improvement in symptoms after treatment without any complications.



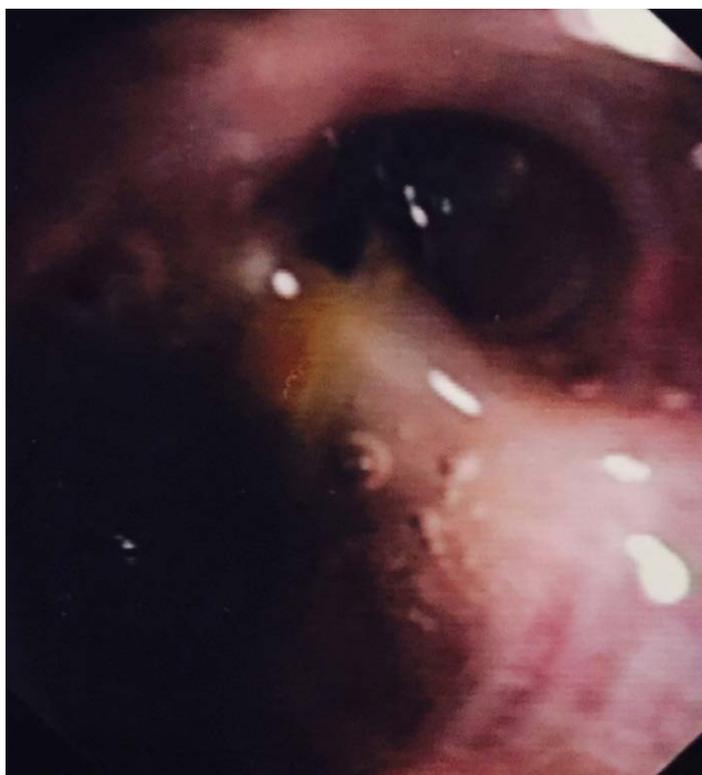
**Figure 1.** Chest-X-ray shows a pathological elevation of the right hemi-diaphragm associated with a mixed and heterogeneous opacity sided at the base of the right lung (white arrows).



**Figure 2.** The axial CECT image of the thorax and abdomen, demonstrates the abnormal presence of air within the necrotic lesion localized at the VII segment of liver (a, white arrowhead); in the coronal (b, black arrowhead) and in the sagittal (c, d) multiplanar reconstruction images a pathological communication between the bronchial tree and the biliary tract is documented, with a strong suspicion for a broncho-biliary fistula.



**Figure 3.** The coronal and sagittal TSE T2 weighted Breath Hold images (a, b) demonstrate the pathological leakage between the bronchial tree of the right lower lung base and the apex of the VII segment of liver (white arrowhead) where a heterogeneous hypo-intense mass is appreciated (white star). The Magnetic Resonance Cholangiogram (c) demonstrates the continuity between the biliary system and the bronchial branches showing the exact site of the fluid leakage (white arrowhead).



**Figure 4.** Flexible bronchoscopy shows bile-like material in the bas-

al-posterior segment (B10) of the right lower bronchus.

## Discussion

Bilioptysis represents the pathognomonic sign of BBF's clinical diagnosis. Respiratory symptoms, such as irritating cough, fever (36%) and jaundice (20%), are other rare clinical features of BBF, but abdominal pain results a very uncommon associated symptom (14%) [3]. At laboratory examination, elevated levels of white cell blood count, C-reactive protein (CRP) or direct and total bilirubin in case of bile duct obstruction could be noticed. The microbiological analysis of sputum can reveal the presence of *E.coli*, *Klebsiella* spp., *Pseudomonas aerug.*, *Enterococcus* spp. and *Enterobacter cloacae* [2].

Since Peacock's first report [1], the local infections of liver were the most frequent cause of acquired BBF, but according to updated data [3], the primitive neoplasms (32%) and metastases (13%) of the liver have become the most common.

A BBF as a complication of RFA is extremely rare, with only seven cases reported worldwide [5]. RFA represents an increasing treatment of hepatic metastatic tumors as an alternative or adjunct to surgical resection, associated with a complication rates of 9% [6]. A BBF after RFA may be caused by scars' formation that, obstructing the biliary ducts, could result in a biloma and in a subsequent abscess [2]. By increasing, the abscess can gradually erode the diaphragmatic profile, breaking in the lung parenchyma and in the bronchial tree. A BBF post-RFA treatment can occur also as a result of a thermal injury if the hepatic lesion is localized in proximity to the lung base [7].

The BBF, if diagnosed late, may cause significant complications, including chemical and bacterial pneumonitis or mediastinitis, with a morbidity and mortality rate up to 12.2% [8].

The imaging tests are required to define the biliary anatomy and to demonstrate the diagnosis of the pathological communication between biliary ducts and the bronchial tree. In literature, the diagnostic methods to verify the clinical suspicion vary greatly from bronchoscopy, bronchogram, CT, MRC and percutaneous (PTC) or endoscopic retrograde cholangiography (ERCP) [2,9]. CT usually is performed as the first imaging technique, even if, in literature, is not considered the most sensitive method [2]. ERCP or PTC have the advantage of demonstrating the biliary anatomy and making the biliary tree decompression or can repair the ductal damage [10]. However, because these procedures are invasive and can lead to serious complications compared with imaging techniques such as MRC [11], they can represent the first therapeutic approach for BBF.

MRC is a non-invasive and safe imaging technique which doesn't need contrast media. When it is associated with conventional MRI, it becomes a very useful imaging tool in the diagnosis of BBF, demonstrating the biliary anatomy and the

presence of the abnormal bronchobiliary communication. This multiplanar imaging technique can provide information for the subsequent treatment planning, whether endoscopic, percutaneous, or surgical [12]. Recently an emerging imaging technique, the Contrast-Enhanced Magnetic Resonance (CE-MRC) using hepatobiliary contrast agents, is demonstrating high ability to visualize non-dilated bile ducts and biliary leaks [8,9].

In our case, especially the CECT and the MRI associated with the MRC images have allowed to demonstrate the pathological anatomical interconnection between the basal bronchial tree and the biliary tract at the VII hepatic segment, concluding for a BBF as a iatrogenic complication of a RFA for liver metastases. MRC has provided high advantage in the evaluation of the anatomy of the biliary tree and, through the use of various sequences and acquisition plans, has proved to be a helpful study technique in the diagnosis of the BBF.

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