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Bronchobiliary Fistula, a Late Complication of Liver Surgery[☆]



Fístula biliobronquial, una complicación tardía de la cirugía hepática

To the Editor,

Bronchobiliary fistula is an abnormal communication between the bile duct and the bronchial tree. In 1850, Peacock described the first case in a 20-year-old woman with hepatic echinococcosis.¹ It is a rare entity,² and few cases have been reported in the literature. Morbidity and mortality are high, and the diagnosis is essentially clinical, guided by the pathognomonic presence of biliptysis, or bile in the sputum.³ Bile has a corrosive effect on the lung and pleural space.

We report the case of a 57-year-old man who presented in the emergency room with a 3-day history of fever 38.5 °C and bilious expectoration. Significant history included sigma

adenocarcinoma with liver metastases, for which he received neoadjuvant chemotherapy with irinotecan and cetuximab. Portal vein embolization and surgical resection of the liver metastasis (right hepatectomy, section of segment 4 A, and left lobe), cholecystectomy and splenectomy were subsequently performed. He received single-agent cetuximab until sigmoidectomy with colorectal anastomosis, which was performed 8 months before he presented in the emergency room. At that time, the sigma cancer was in remission (CEA 3.9 ng/ml), and the patient was being treated with cetuximab every 15 days. On physical examination, arterial blood pressure was 130/80 mmHg, heart rate 115 bpm, temperature 37.5 °C, and oxygen saturation 89%. Poor general condition, conjunctival icterus, reduced breath sounds in the right field with crackles, and hepatomegaly of 2 finger breadths. Clinical laboratory tests showed significant leukocytosis (25 790/μl) with neutrophilia (80%). Chest X-ray revealed consolidation in the right lower lobe. Given the likelihood of a biliopulmonary fistula as a late complication of liver surgery, a computed tomography (CT) of the chest-abdomen was requested, which showed parenchymal

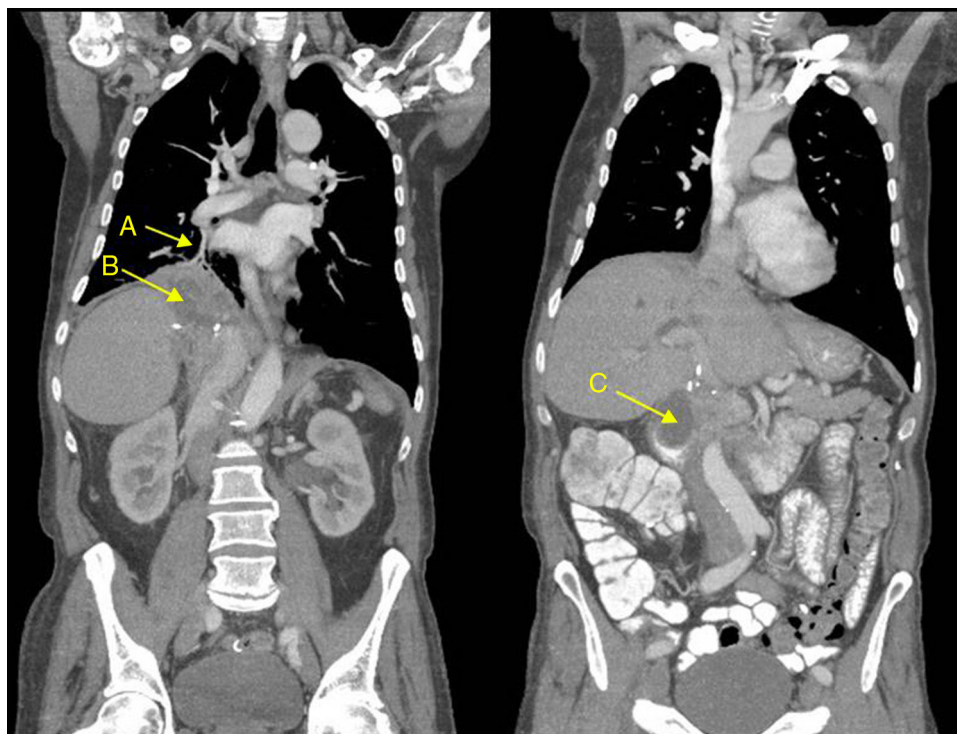


Fig. 1. Multiplanar coronal CT reconstruction slices with intravenous contrast medium. (A) Fistulous tract between the biliary tree and the bronchus of the right lower lobe. (B) Right subphrenic collection. (C) Subhepatic collection.

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involvement in the right lower lobe due to a fistula between the right lower lung and the hepatobiliary and subphrenic space. Both the CT and the nuclear magnetic resonance cholangiography showed a large collection extending craniocaudally from the right hemithorax to the subhepatic region, measuring 16 cm. This was composed of a multiseptated subphrenic collection (7.5×12 cm) connecting with a subhepatic collection (4.5×4.5 cm) (bilioma) (Fig. 1).

Empirical antibiotic therapy began with a 12-day course of meropenem. Percutaneous cholangiography was performed, with placement of double external percutaneous biliary drainage in segments II and III. The procedure was incident-free.

Although the patient was initially transferred to the intensive care unit, his clinical and radiological course were very favorable with resolution of fever on day 3, improvement of respiratory failure, and resolution of radiological infiltrate. At discharge, he was prescribed a third-generation cephalosporin (cefixime) for 1 week. The follow-up chest-abdomen CT 11 days after discharge showed reduced involvement of the right lower lung, with some small residual image along the length of the fistula and reduced subphrenic collection (6.2×3.67 cm).

Bronchobiliary fistulas can be congenital or acquired. Acquired fistula occur due to 3 mechanisms:

1. Fistula due to trauma is the most frequent, caused by injuries penetrating the lung, diaphragm and liver. Bile duct surgery is included in this category, particularly laparoscopic interventions. In our case, the fistula was a secondary complication of previous liver surgery.
2. Fistula due to liver disease, the most common causes being hydatid cyst and amebic liver abscess.
3. Fistula due to bile duct obstruction.

Recommended diagnostic procedures for bronchobiliary fistulas are percutaneous transhepatic cholangiography,⁴ bronchoscopy, 3-dimensional CT reconstruction,⁵ and cholescintigraphy.⁴ Magnetic resonance imaging with contrast medium contributes functional data. There is no consensus regarding the therapeutic management of these fistulas; a review of the literature suggests reserving surgery for fistulas that do not respond to conservative treatment or for complicated cases.

References

1. Carrillo Muñoz A, Sánchez Valdez T, Gil Rojas N, Navarro Reynoso F, Núñez-Pérez Redondo C, Cícero Sabido R. Un caso raro de fistula biliobronquial. Revisión bibliográfica. *Rev Med Hosp Gen Mex.* 2013;76:47–51.
2. Matsumoto T, Otsuka K, Kaihara S, Tomii K. Biliary pneumonia due to the presence of a bronchobiliary fistula. *Intern Med.* 2017;56:1451–2.
3. Eryigit H, Oztas S, Urek S, Olgac G, Kurutepe M, Kutlu CA. Management of acquired bronchobiliary fistula: 3 case reports and a literature review. *J Cardiothorac Surg.* 2007;2:52.
4. Loinaz C, Hernández T, Mitjavila M, Martín J, Ochando F, Madariaga ML, et al. Biliobronchial fistula after liver surgery for giant hydatid cyst. *HPB Surg.* 2011;2011:347654.
5. Martínez-Velado E, Palomar-Rodríguez LM, Olivo Esteban JR. Bronchobiliary fistula. *Rev Esp Enferm Dig.* 2012;104:210–1.

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Leser-Trélat Sign Secondary to Thymic Carcinoma



Signo de Leser-Trélat secundario a carcinoma tímico

Dear Editor:

A 64-year-old woman presented with left hemithorax pain, cough and weight loss of approximately 20 kg over a 4-month period. She had a history of smoking (40 packs/year) and chronic hypertension. On physical examination, she was eupneic on room air, with normal pulmonary auscultation, and presented left supraclavicular lymph-node enlargement. Multiple brownish warty plaques with verrucous texture were present on the patient's skin; they predominated in the anterior trunk (Fig. 1A), with onset about 6 months previously. The patient also reported mild signs and symptoms of dysphonia, dysphagia and hoarseness. Blood tests revealed mild anemia. Other laboratory data were unremarkable.

Chest computed tomography showed an irregular mass in the anterior mediastinum, in close contact with the aortic arch, with heterogeneous contrast enhancement. The mass infiltrate the left paratracheal space, through the aortopulmonary window. The left hemidiaphragm was elevated, probably due to a phrenic nerve injury. Lymph node enlargement was observed, predominantly in the left supraclavicular region, with necrotic centers (Fig. 1C and D). A biopsy of the supraclavicular lymph node with immunohistochemical study revealed a poorly differentiated malignant neoplasm compatible with thymic carcinoma. The skin lesions were

characterized as seborrheic keratoses. Given these features, a diagnosis of Leser-Trélat sign was made. The patient was referred for treatment of thymic carcinoma. Her condition worsened, and she died 2 months later.

Seborrheic keratoses are benign dermatological lesions characterized by proliferation of immature keratinocytes, which develop normally and gradually in some patients, especially those in the fifth and sixth decades of life. They present in well-defined, rounded or ovoid shapes and they are hyperpigmented, brownish or blackish with raised, verrucous and wrinkled surfaces. Preferred locations are the trunk, extremities, face and neck.¹

Leser-Trélat sign refers to the sudden onset and rapid growth in number and size of multiple lesions of seborrheic keratoses, sometimes associated with pruritus, which precede, succeed or occur concomitantly with a neoplasm, whether hidden or known.^{2,3} About 20% of patients present associated acanthosis nigricans.⁴ Classically, the sign is related to adenocarcinomas, especially those of the gastrointestinal tract and breast, but also those of the lung, kidney, liver, pancreas, ovary, uterus and prostate, as well as lymphoproliferative diseases, among others.^{2–4} The pathophysiological mechanism is not completely understood, but the sign is believed to be caused by cytokine stimuli, growth or humoral factors produced by or in response to the tumor.⁵ Some authors have also reported associations with benign conditions, such as pregnancy and some benign tumors. Histopathological findings are similar to those of usual seborrheic keratosis. No specific treatment is available for the lesions,⁴ but regression occurs with treatment of the underlying disease in some cases.³