Revista de PATOLOGÍA RESPIRATORIA

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CLINICAL NOTE

Fortuitous bilothorax: a picture is worth a thousand words

Biliotorax fortuito: una imagen vale más que mil palabras

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Abstract

We present a 91-year-old male was admitted in Clinical Hospital of Valladolid (Spain), due to respiratory infection and acute respiratory failure. Given his torpid evolution and increased oxygen requirements, the new X-ray showed submassive right pleural effusion. Thus, we carried out diagnostic thoracentesis and placed a endothoracic drainage. The greenish turbid pleural fluid (PF) was polymorphonuclear exudate, establishing the diagnosis of bilothorax by the presence of bilious pleural effusion (PF/Serum bilirubin > 1). The thoraco-abdominal CT scan showed a subphrenic abscess with an interruption in right diaphragmatic dome, accompanied by collections around the holed gallbladder. Subsequently, it was placed a biliary drainage and antibiotherapy was adjusted to the growth of C. albicans and E. faecalis in PF, with favorable recovery after surgery. It is essential to make a detailed review of patients' complementary tests, owing to the X-ray was suspicious of subphrenic abscess with hydroaerial level, but it was erroneously classified as situs inversus with levocardia.

Keywords: *Bilothorax. Thoracentesis. Pleural effusion.*

Resumen

Presentamos el caso de un varón de 91 años que fue ingresado en el Hospital Clínico Universitario de Valladolid (España), debido a una infección respiratoria e insuficiencia respiratoria aguda. Dado la tórpida evolución e incremento de las necesidades de oxígeno, una nueva radiografía mostró la aparición de derrame pleural submasivo derecho. Tras esto, se llevó a cabo una toracocentesis y se colocó un drenaje endotorácico. El líquido pleural (LP) verdoso y turbio fue un exudado polimorfonuclear, y permitió establecer el diagnóstico de biliotorax dado la presencia de bilis (cociente bilirrubina LP/ bilirrubina sérica > 1). El TAC mostró un absceso subfrénico con una solución de continuidad en la cúpula diafragmática, acompañado de colecciones alrededor de la vesícula perforada. Posteriormente, se colocó un drenaje biliar y se pautó antibioterapia ajustada al crecimiento de C. albicans y E. facealis en el LP, con una favorable recuperación del paciente tras ser intervenido. Es fundamental realizar una minuciosa revisión de las pruebas complementarias de los pacientes, dado que la radiografía de ingreso ya era sospecha de absceso subfrénico con nivel hidroaéreo, pero erróneamente se clasificó de situs inversus con levocardia.

Palabras clave: *Biliotórax. Toracentesis. Derrame pleural.*

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Date of reception: 31-08-2024
Date of acceptance: 18-11-2024
DOI: 10.24875/RPR.24000040

Available online: 20-03-2025
Rev Pat Resp. (ahead of print)
www.revistadepatologiarespiratoria.org

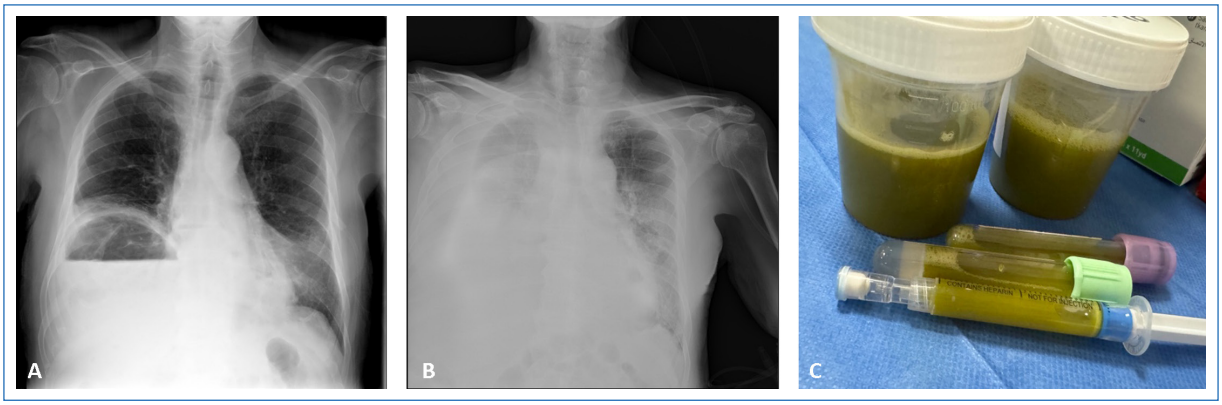

Figure 1. A: postero-anterior chest radiograph upon patient admission showing a right subphrenic air-fluid level (compatible with a possible subphrenic abscess) and infiltrate in the left lower lobe. **B:** portable antero-posterior radiograph taken 7 days after admission showing moderate right pleural effusion and bilateral infiltrates. **C:** right pleural fluid obtained after thoracentesis with macroscopic greenish characteristics.

We present a 90-year-old male who was admitted to the University Clinical Hospital of Valladolid (Spain) due to a respiratory infection and acute respiratory failure. He had no significant medical history except for a hospitalization two years prior for acute lithiasic cholecystitis with a perivesicular collection and choledocholithiasis, which required an endoscopic retrograde cholangiopancreatography and sphincterotomy. The clinical presentation began with fever, dyspnea progressing to moderate on exertion (grade 2-3/4 mMRC), greenish expectoration, and semi-liquid stools, without pathological products, for 4 days. He showed no improvement with azithromycin. The initial X-ray was classified as potential *situs inversus* with levocardia (Fig. 1A), and the laboratory results showed a mixed alkalosis. Consequently, empirical treatment with meropenem and linezolid was initiated.

During hospitalization, the patient experienced several episodes of bronchospasm that required an increase in oxygen requirements. There was a good response to intravenous glucocorticoids, bronchodilators with short-acting beta-agonists, and antimuscarinics. A follow-up chest X-ray revealed the presence of alveolar infiltrate in the left lower lobe and moderate right pleural effusion, suggesting possible cardiac decompensation due to elevated pro-BNP levels. Consequently, a diuretic treatment with furosemide was initiated.

On the 7th day of admission, due to the absence of clinical, analytical, and radiological improvement, as well as an increase in the pleural effusion (PE) (Fig. 1B), a diagnostic thoracentesis and endothoracic drainage were requested. The initial thoracic ultrasound revealed an anechoic pleural fluid chamber of 10 cm in the right 7th intercostal space (posterior axillary line, R4-R6 ultrasound field), causing compression of the surrounding lung parenchyma ('jellyfish sign'). Additionally, a hypoechoic fluid collection of 6 cm was observed in the ipsilateral subphrenic recess with debris inside (Fig. 2A).

Subsequently, diagnostic ultrasound-guided thoracentesis was performed under local anesthesia, obtaining macroscopically greenish turbid pleural fluid (Fig. 1C). Rapid gasometer determination did not suggest infection. However, due to the atypical characteristics and the patient's history, a pigtail drain was inserted using a Safe-T-Centesis device, and an urgent thoraco-abdominal CT scan was requested to identify the origin.

Biochemical analysis of the pleural fluid revealed the following results: pH 7.38; glucose 223 mg/dL, proteins 2 g/dL, LDH 12,956 U/L, adenosine deaminase 72 U/L, amylase 97 U/L, leukocytes 7,181 /µL, with 1% mononuclear cel/mm and 99% polymorphonuclear cel/mm. Bilirubin levels were elevated, with total bilirubin (B) at 2.85 mg/dL and direct bilirubin at 2.05 mg/dL, consistent with a polymorphonuclear-predominant exudate biliothorax (pleural fluid/serum bilirubin ratio > 1). Microbiological analysis indicated the growth of gram-positive cocci (streptococcal type) with 10^3 CFU Enterococcus faecalis multisensitive and 10^3 CFU Candida Albicans multisensitive. Treatment was adjusted to piperacillin/tazobactam and fluconazole.

The thoraco-abdominal CT scan revealed a right pleural effusion with passive atelectasis and a properly positioned endothoracic drain. Additionally, there was abundant fluid in the subphrenic recess, along with a 6 mm defect in continuity observed in the right

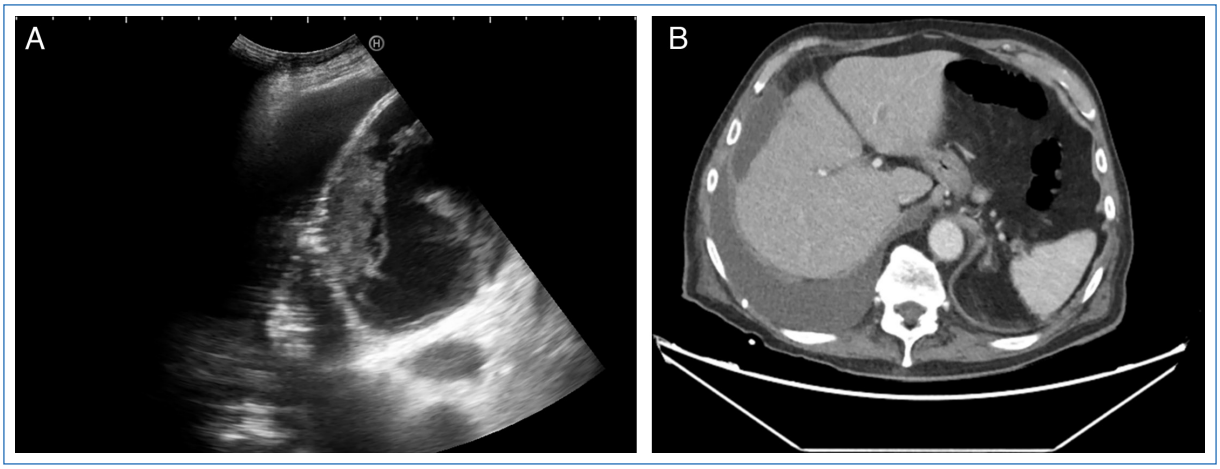

Figure 2. A: thoracic ultrasound showing anechoic pleural effusion and a subphrenic collection with debris inside. **B:** axial cut of the thoraco-abdominal CT scan showing right pleural effusion with a collection in the recess and a defect in the diaphragmatic dome.

diaphragmatic dome, communicating with the ipsilateral pleural effusion. As well, multiple collections were identified around the gallbladder, with a perforation on its anterior-superior aspect. Furthermore, one of the collections was found to communicate with the fluid in the subphrenic recess (Fig. 2B).

Following the described findings, the Interventional Radiology Service placed a drain in the right subdiaphragmatic collection and performed a cholecystostomy to evacuate the biliary content. The patient was evaluated by the General Surgery service, which recommended initial conservative management, but since gradual clinical improvement was observed, a cholecystectomy with abscess drainage were carried out, confirming the presence of a 1 cm fistula in the diaphragmatic dome. It was possible to reduce oxygen needs and remove previously placed drains due to the absence of drainage; after 10 days of surgery without further incidents, the patient was discharged from the hospital.

In conclusion, it is crucial to meticulously review all complementary tests from the start. The patient's admission radiograph was highly indicative of a subphrenic abscess due to the presence of an air-fluid level, yet it was erroneously classified as *situs inversus* with levocardia. Biliothorax, is a rare cause of pleural effusion, typically attributed to a biliopleural fistula or rupture of a subphrenic abscess secondary to gastrointestinal tract infection. Delays in diagnosis and treatment can pose life-threatening risks due to the development of sepsis and severe pleural complications, attributed to the intense inflammation caused by alkaline bile salts in the pleural space[1–7].

Funding

None.

Conflicts of interest

None.

Ethical considerations

Protection of humans and animals. The authors declare that no experiments involving humans or animals were conducted for this research.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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