

# Bilioptysis associated with alcohol hepatitis without evidence of bronchobiliary fistula: A rare case report

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**Associate Editor:** Coenraad F Koegelenberg

## Abstract

The presence of bilirubin in the sputum is uncommon but, when present, is most commonly associated with the presence of bronchobiliary fistula, which could be associated with a number of underlying conditions. However, the finding of bilioptysis without bronchobiliary fistula is uncommon, with one associated mechanism postulated to involve increased capillary membrane permeability. This case report describes a patient presenting with bilioptysis while being medically managed with prednisolone for severe alcoholic hepatitis. The patient developed hospital-acquired pneumonia during her hospitalization associated with bilioptysis, resulting in progressive respiratory failure requiring ventilatory support. Alcohol-related pulmonary dysfunction alters pulmonary immune processes, leading to increased susceptibility to pulmonary infection and disrupting the basal alveolar epithelial membrane, thus increasing permeability. This patient's findings were in the absence of a bronchobiliary or broncho-pleural fistula, and we hypothesize that increased capillary membrane permeability was contributory to the bilioptysis in this case.

## KEYWORDS

alcoholic hepatitis, bilioptysis

## INTRODUCTION

Bilioptysis is an uncommon clinical finding, with less than 50 cases reported in the literature. Most commonly, this finding has been associated with bronchobiliary fistula that can occur as a periprocedural complication, such as following surgery<sup>1,2</sup> or procedural management of hepatocellular carcinoma.<sup>3-5</sup> Other bronchobiliary fistula cases have been described as associated with trauma,<sup>6,7</sup> liver abscess<sup>8</sup> or echinococcosis.<sup>9-11</sup> Cases of bilioptysis unassociated with bronchobiliary fistula are infrequently reported in the literature; often, the mechanism is attributed to capillary endothelial permeability injury that allows for bilirubin, a high-molecular-weight protein, to cross the endothelial membrane. We describe the case of a 36-year-old patient with severe alcoholic hepatitis presenting with bilioptysis in the absence of a bronchobiliary fistula.

## CASE REPORT

A 36-year-old female patient with a history of substantial alcohol consumption was referred to a tertiary care hospital in Calgary, Alberta, Canada with a 6-week history of progressive jaundice and increasing abdominal girth. Her last reported alcohol consumption was 6 weeks prior; she had abstained from consuming alcohol consumption due to general malaise.

Past medical history was significant for well-controlled asthma. She was an active smoker, approximately half a pack per day, with a cumulative 15-pack year smoking history. She reported binge-drinking hard liquor, up to 20oz at a time, at least several times weekly. She endorsed partaking in recreational inhaled cannabis use biweekly. The patient was adopted and unaware of her family history.

Her vital signs at the time of presentation were: temperature 36.3°C (97.3°F), heart rate 106 beats/min, blood



**FIGURE 1** Orange-coloured sputum with elevated bilirubin

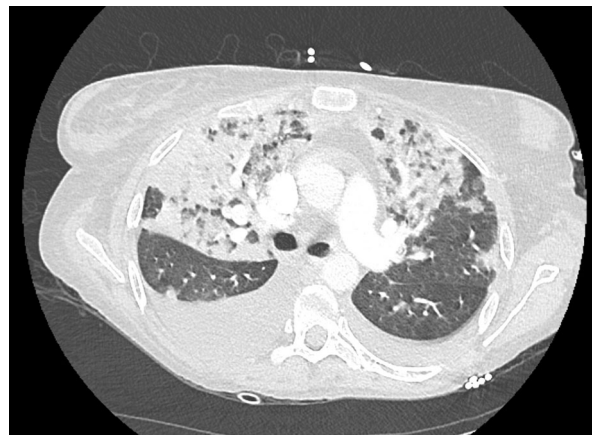
pressure 93/54 mmHg and oxygen saturation 100% on room air. She was in no acute distress but visibly jaundiced with scleral icterus. She was alert and oriented in three spheres and not encephalopathic. Asterixis was not present. Cardiovascular exam was unremarkable. Pulmonary exam revealed mild wheeze, which the patient indicated was consistent with her baseline in the context of being affected by asthma. She had stigmata of liver disease including spider nevi and ascites. Notable laboratory investigations included a total bilirubin level of 415  $\mu\text{mol/L}$  and INR of 3.1, equating to a Maddrey's discriminant function score of 116 points. She was admitted to the General Internal Medicine Service with a diagnosis of alcoholic hepatitis.

The patient underwent additional investigations to exclude sepsis and was then initiated on prednisolone for alcoholic hepatitis.

On Day 15 of admission, she developed hypoxemia requiring 1 L/min of supplemental oxygen. Due to radiographic pneumonia, she was initiated on levofloxacin 750 mg po q24h. COVID NAT was negative.

On Day 16, she developed copious amounts of orange-coloured sputum (Figure 1). Computed tomography (CT) pulmonary angiography study showed no pulmonary embolism, but extensive bilateral airspace opacities, radiographically compatible with pneumonia (Figure 2). Her antimicrobial regimen was switched to piperacillin-tazobactam 4.5 g IV q6h to treat hospital-acquired pneumonia empirically. Her prednisolone medication was discontinued. The laboratory analysed the sputum specimen for the presence of bilirubin, which was 161  $\mu\text{mol/L}$  (serum total bilirubin was 175  $\mu\text{mol/L}$  on that day), and she was confirmed to have biliptysis. The case was discussed with gastroenterology-therapeutics and general surgery consultants, who advised evaluation for bronchobiliary fistula.

On Day 17, an enhanced CT abdomen and pelvis scan showed no anatomical suspicion of bronchobiliary fistula. A hepatobiliary iminodiacetic acid scan (HIDA) scan was then pursued, showing that biliary accumulation and clearance of tracer was also unremarkable. There was no evidence of a connection between the thoracic cavity and the biliary



**FIGURE 2** Computed tomography lung image demonstrating bilateral pleural effusions and dense upper lobe consolidation

system or the liver on the static images and SPECT-CT, and no specific activity within the lungs corresponding to biliary activity. Later that evening, the patient was transferred to the intensive care unit (ICU) for intubation and mechanical ventilation due to progressive hypoxemia respiratory failure.

On Day 23, transthoracic echocardiogram showed mild pulmonary hypertension and no valvular or left ventricular abnormalities. On Day 27, a right-sided chest tube was placed for pleural effusion drainage; the pleural effusion did not have bilious features on visual inspection, and per light's criteria, was categorized as a transudative effusion (fluid lactate dehydrogenase 114 U/L and fluid protein 11 g/L). She had a protracted course in the ICU. Her alcoholic hepatitis showed gradual improvement. On Day 49, the patient was stable to be transferred out of ICU to the General Internal Medicine ward.

She was discharged home on Day 56 of admission. Six months post-discharge from the hospital, she received outpatient follow-up by Hepatology, who indicated that the patient had evidence of underlying liver cirrhosis. Total bilirubin level at that time was 25  $\mu\text{mol/L}$ , within normal range. The patient was documented to have maintained ongoing abstinence from alcohol and was back to full-time employment.

## DISCUSSION

The unusual presentation of bile in the airway secretions is most commonly associated with bronchobiliary fistula, which may be present due to trauma, procedural complications, or hepatic infection. The literature describing biliptysis in the context of liver dysfunction is limited. Choong et al. describe a case of biliptysis related to decompensated liver cirrhosis and acute pulmonary edema from transfusion-associated circulatory overload.<sup>12</sup> In an abstract presented by Gandhi et al.,<sup>13</sup> their group documented a single case of biliptysis related to fulminant liver failure, multi-organ failure and systemic inflammation, which compromised the capillary membrane, leading to increased permeability.

Our patient did not have bronchobiliary fistula identified on CT imaging or HIDA scan. We postulate that the clinical finding of bilioptysis for this individual was associated with systemic inflammation due to alcoholic hepatitis with the superimposed development of hospital-acquired pneumonia and acute respiratory distress syndrome, leading to increased capillary-alveolar membrane permeability causing bilirubin leakage. Acute respiratory distress syndrome is a diffuse lung injury that involves a disruption of the pulmonary alveolar membrane resulting in excess interstitial and alveolar fluids. Alcohol use disorder predisposes the likelihood of developing acute respiratory distress syndrome as high as 43% compared to 22% of patients without alcohol use disorder.<sup>14</sup> Alcohol-related pulmonary dysfunction has been demonstrated to impact pulmonary immune processes, thus increasing susceptibility to infections such as pneumonia and disrupting the basal alveolar epithelial membrane increasing permeability. This pulmonary dysfunction increases the risk of leakage of large molecules, such as bilirubin, and diminishes alveolar liquid clearance, resulting in bilious sputum.<sup>14,15</sup> In addition to this patient's hospital-acquired pneumonia, bilirubin-associated chemical lung injury could have also contributed to her respiratory failure.

#### AUTHOR CONTRIBUTION

Jennifer Ngo and Mathew Wenger completed the conception, the acquisition, analysis and interpretation of data for the work. Jennifer Ngo and Mathew Wenger drafted the manuscript and prepared the images. Alex Chee revised the manuscript. All authors agreed on the final manuscript.

#### ACKNOWLEDGEMENT

We acknowledge Ms. Tanya Kurich MN NP for her contribution to conception of this work and for image acquisition.

#### CONFLICT OF INTEREST

None declared.

#### DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

#### ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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**How to cite this article:** Ngo J, Wenger M, Chee A. Bilioptysis associated with alcohol hepatitis without evidence of bronchobiliary fistula: A rare case report. *Respirology Case Reports.* 2022;10:e01028. <https://doi.org/10.1002/rcr2.1028>