Case Report

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Unexpected color appearance of pleural fluid: a bilothorax case

Plevra sıvısının beklenmedik renkte görünmesi: bilotoraks olgusu

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Abstract

Bilothorax, or cholethorax, is a rare cause of exudative pleural effusion characterized by the presence of bile in the pleural space. Recognizing this condition is essential to prevent severe complications such as empyema and acute respiratory distress syndrome. This report presents a 70-year-old male patient who developed right-sided bilothorax following multiple biliary tract interventions. The patient presented with symptoms of jaundice, fever, and abdominal pain. Clinical and radiological evaluations revealed right-sided pleural effusion, and thoracentesis yielded dark yellow-green fluid. The diagnosis of bilothorax was confirmed by a pleural fluid/serum total bilirubin ratio >1.0. Early intervention was performed using a cystofix to drain the pleural fluid. However, despite all interventions, the sepsis condition could not be controlled, and the patient unfortunately passed away. Bilothorax is generally associated with hepatobiliary procedures and is mostly observed on the right side due to anatomical proximity. Diagnosis requires a high index of suspicion, especially in patients with relevant clinical histories and characteristic pleural fluid appearance. Rapid thoracentesis and pleural fluid analysis are crucial for diagnosis. Treatment typically involves pleural drainage and early administration of broad-spectrum antibiotics. In conclusion, bilothorax is a life-threatening condition requiring urgent diagnosis and intervention. This case highlights the importance of recognizing this rare condition and the necessity for early aggressive management in patients with a history of hepatobiliary procedures.

Keywords: Bilothorax, unilateral pleural effusion, biliopleural fistula.

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Öz

Bilotoraks, plevral boşlukta safra varlığı ile karakterize, eksüdatif plevral efüzyonun nadir bir nedenidir. Bu durumun tanınması ampiyem ve akut solunum sıkıntısı sendromu gibi ciddi komplikasyonları önlemek için önemlidir. Bu raporda, birçok safra yolu müdahalesi sonrasında sağ taraflı bilotoraks gelişen 70 yaşındaki bir erkek hasta sunulmaktadır. Hasta sarılık, ateş ve karın ağrısı şikayetleri ile başvurdu. Klinik ve radyolojik değerlendirme ile sağ taraflı plevral efüzyon görüldü ve torasentez ile koyu sarı-yeşil sıvı elde edildi plevral mayii/serum total bilirubin oranının >1,0 olması ile bilotoraks tanısı konuldu. Erken müdehale ie sistofix ile plevral mayi drenajı sağlandı. Ancak tüm müdehalelere rağmen sepsis tablosunun önüne geçilemeyerek hastamız kaybedildi. Bilotoraks, genellikle hepatobiliyer prosedürlerle ilişkilidir ve anatomik yakınlık nedeniyle çoğunlukla sağ tarafta görülür. Tanı, özellikle ilgili klinik öykülere ve karakteristik plevral sıvı görünümüne sahip hastalarda yüksek şüphe gerektirir. Tanı için hızlı torasentez ve plevral sıvı analizi kritik öneme sahiptir. Tedavi genellikle plevral drenaj ve geniş spektrumlu antibiyotiklerin erken uygulanmasını içerir. Sonuç olarak Bilotoraks, acil tanı ve müdahale gerektiren hayatı tehdit eden bir durumdur. Bu olgu, ilgili hepatobiliyer öyküye sahip hastalarda bu nadir durumu tanımanın ve erken agresif yönetimin önemini vurgulamaktadır.

Anahtar kelimeler: Bilotoraks, tek taraflı plevral efüzyon, biliyoplevral fistül.

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Introduction

The presence of bile in the pleural space is termed bilothorax or cholethorax, which is one of the rare causes of exudative pleural effusion. It is crucial to recognize it in order to make a proper differential diagnosis and to prevent complications such as empyema and acute respiratory distress syndrome [1, 2].

Bilothorax is primarily caused by congenital factors, iatrogenic injury to biliary tract, diaphragmatic ruptures, hepatic abscesses, and thoracentesis can be helpful to diagnose it [1, 3]. Therefore, bilothorax should be considered in cases of pleural effusion in patients with a history of liver and bile duct-associated procedures [4].

With educational purposes aimed at highlighting its diagnostic challenges, we present the case of a 70-year-old male patient who developed right-sided bilothorax following numerous biliary tract interventions, with a suspected diagnosis of primary sclerosing cholangitis.

Case report

The patient underwent evaluation following a consultation requested from the relevant clinic. This 70-year-old male, who has been monitored for jaundice at the Gastroenterology Department, exhibited right-sided pleural effusion on posteroanterior chest radiography and thoracic computed tomography (CT). The medical history revealed that he underwent coronary bypass surgery ten years ago, he has been consistently treated for coronary artery disease and hypertension. He ceased smoking 30 years ago after smoking a few cigarettes daily for 20 years. He has no prior history of lung disease.

Cholelithiasis in common bile duct and gallbladder was diagnosed through magnetic resonance cholangiopancreatography (MRCP) one month ago. Consequently, the stone was removed, and biliary stenting was performed during Endoscopic Retrograde Cholangiopancreatography (ERCP). Shortly after the procedures, the patient was discharged. Fifteen days later, he began experiencing jaundice, fever, and abdominal pain prompting a repeat MRCP and ERCP. Piperacillin tazobactam treatment was started with infectious diseases consultation. Cholangiocarcinoma was suspected due to the occurrence of primary sclerosing cholangitis. When a 44x30 mm irregular lesion was detected in the main ducts, a percutaneous transhepatic cholangiography and percutaneous biliary drainage was performed at the interventional radiology department. Three days later, t-tube cholangiography and biliary dilatation were performed as well. However, the bile drainage was significantly low. Two days after from these procedures, the patient began complaining of shortness of breath.

On inspection, the patient presented icteric findings. On auscultation of his chest, the left hemithorax appeared normal, but the lower right side of the lung exhibited diminished sound and dullness upon percussion. His oxygen saturation was 95% (with the support of 4 liters/minute of oxygen). Other vitals findings were within normal limits. Abnormal laboratory findings revealed as an elevation of CRP (248 mg/L), leukocytosis (WBC count: 20.170/mm^3), and a low hemoglobin level (9.2 gr/dl). Increased density consistent with right-sided pleural effusion was observed on chest radiography (Figure 1). On thoracic CT, a unilateral moderate pleural effusion approximately 6 cm in size was identified on the right side. Additionally, the catheter passage line was observed adjacent to the right hemi-diaphragm (Figure 2). Dark yellow-green colored 500 mL fluid drained during thoracentesis (Figure 3).

Considering the macroscopic appearance of the pleural fluid, the analysis of bilirubin levels was added to the routine laboratory testing for diagnostic approach to pleural effusions. The laboratory test results are summarized in Table 1.

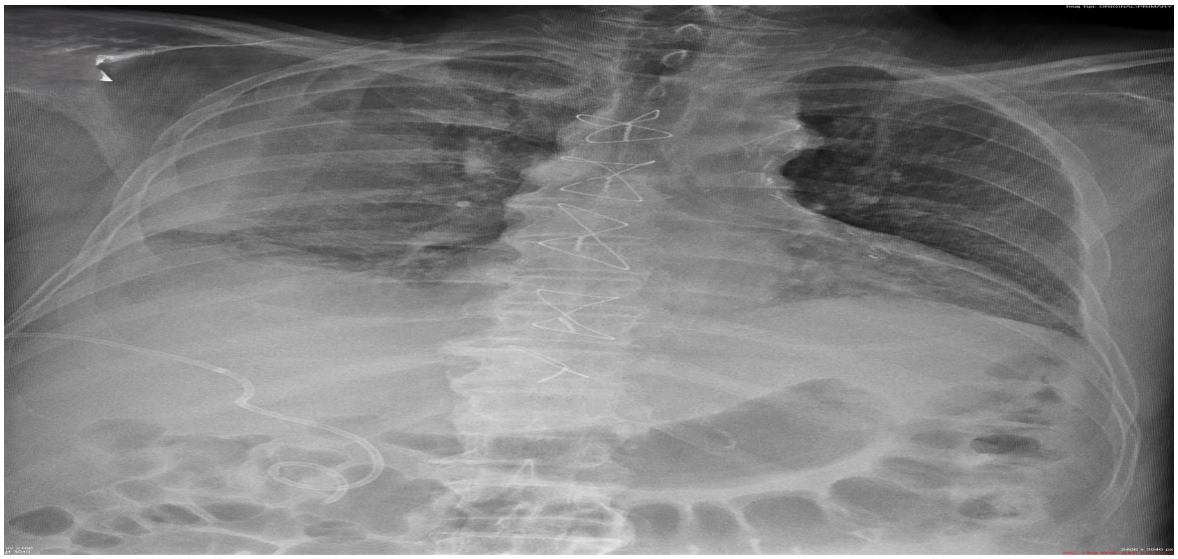

Figure 1. Posterior-anterior chest radiography

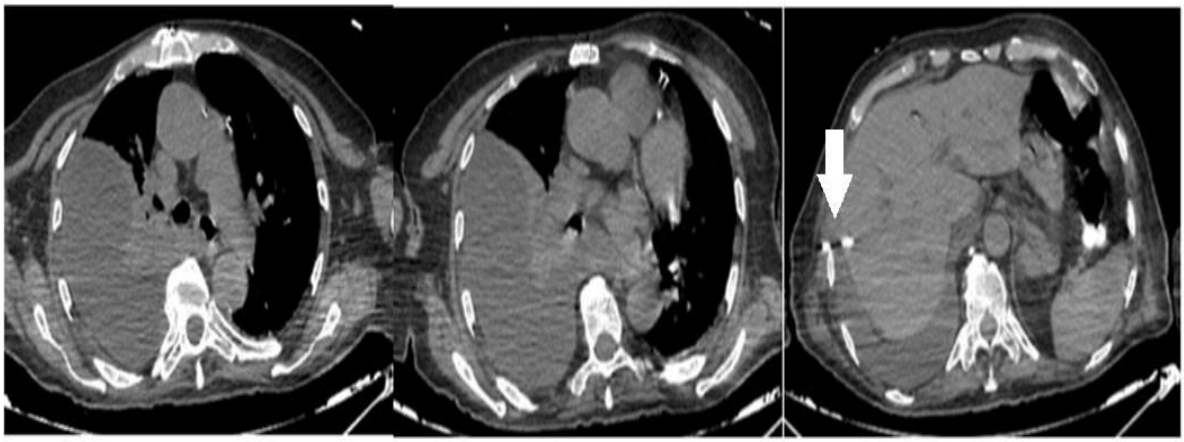

Figure 2. Computed tomography

Arrow is pointing to: hepatobiliary katater

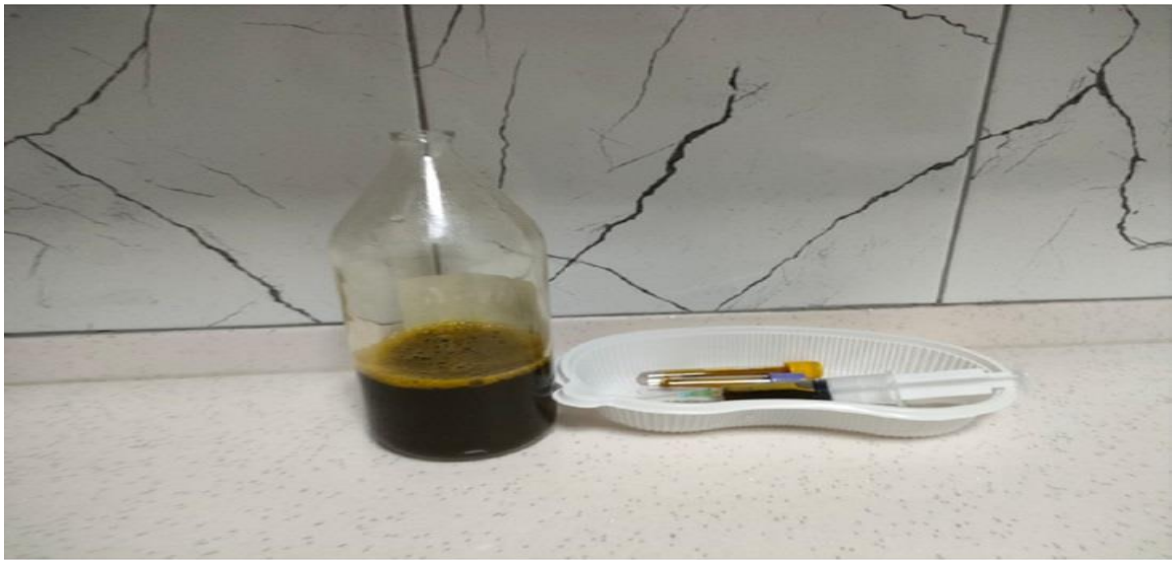

Figure 3. Thoracentesis fluid

Table 1. Laboratory results of blood serum and pleural fluid

Parameters	Pleural Fluid	Blood Serum	Pleura/Blood Serum Ratio
LDH (U/L)	688	189	3.64
Total Protein (g/L)	13.5	55.8	0.24
Albumin (g/L)	12.9	24.8	11.9
Glucose (mg/dL)	6	116	
Bilirubin (mg/dL)	53.45	5.94	8.99
pH	6.92		
Polymorphonuclear Leukocyte (%)	97.1		
Mononuclear Cells (%)	2.9		

The results revealed an exudative pleural fluid with an elevated percentage of polymorphonuclear cells, and the ratio of pleural fluid to serum bilirubin was higher than 1. These findings were favored the diagnosis of bilothorax. Tube thoracostomy was performed using 14F catheter by the thoracic surgeons. The patient had been given piperasilin-tazobactom during 7 days. But acute phase reactants had increased and the Infectious Disease Department changed antibiotics, and they started setazitim. Cystofix drain was monitored daily by thoracic surgeons. There was no drainage from thoracic sistofix. The patient's condition worsened and his symptoms had showed that sepsis. The patient was transferred to the intensive care unit for close monitoring. Meropenem treatment was started. Two days later Klepsiella pneumonia and Enterococcus Faecium growth was seen in the pleura culture. According to culture sensitivity, the meropenem treatment dose was increased and amikacin was added to the treatment. Bile flow through the cystofix stopped. However, there was no response to the patient's sepsis condition. Despite all the interventions, the patient suffered cardiopulmonary arrest days later and died.

Discussion

Bilothorax is a life-threatening condition as bile can increase the susceptibility to infection in the pleural space [5].

Diseases occur after penetrating or non-penetrating trauma. Biliopleural fistula is a rare condition that may occur. It can also be seen as bronchobiliary fistula after these diseases [4]. It should be always in differentials in patients who have pleural effusions. Even though patients usually present with pleuritic chest pain, respiratory distress, and a history of hepatobiliary procedures, few cases have been described with a spontaneous onset [5, 6]. Same as what occurred in our case, the localization is right-sided most of the time due to its anatomical nearity to the biliary tract, however, there are different cases that have been reported isolated left-sided and bilaterally [2, 5, 7]. As a rare incident, one case has reported a left-sided presentation of bilothorax due to cholangiocarcinoma which can occur because of a natural transition of the bile through the esophageal and aortic hiatuses [8]. Patients' medical history and chest radiographies should be key to suspect bilothorax for diagnosis and prevent further complications. Thoracentesis can be a part of the diagnosis seeing the green and viscous fluid. Additionally, after the tube insertion during the interventional procedures, realizing a low drainage of fluid than expected might be a cue as well. Observation of a pleural effusion should be followed by diagnostic thoracentesis with measurement of pleural total bilirubin. Visually, the pleural fluid will appear green-black, with an exudative profile on fluid studies. In various case reports, pleural-to-serum total bilirubin (P/S) ratio >1.0 has been reported as an indicator of the presence of bilothorax [6].

Prompt recognition of bilothorax is important as bile is a potent chemo irritant that can lead to a severe inflammatory response, such as acute respiratory distress syndrome [9]. In addition, given that bile is a medium conducive for bacterial growth, bilothorax places patients at higher risk for the development of bacterial empyema, especially those with chronic liver disease and

cirrhosis given their immunocompromised state [6]. Infections such as empyema can increase mortality 4-fold in patients with cirrhosis [10, 11].

Our case had a presentation of respiratory distress, a history of jaundice and fever, and multiple interventions to the hepatobiliary tract including ERCP, MRCP and, percutaneous transhepatic biliary drainage (PTBD). Bilothorax was suspected due to the development of pleural effusion in the case with known condition. The pleural fluid was drained immediately upon suspicion of bilothorax, the infection could not be controlled despite all treatment attempts. Unfortunately, our patient died due to sepsis.

There is no existing extensive experience regarding the best way of treatment but successful management requires a rapid and accurate diagnosis. Several reports describe conservative management by chest tube insertion and pleural drainage as a successful and appropriate way of solving the problem [12].

Additionally early administration of broad-spectrum antibiotics to prevent future infection is recommended as well [13].

Informed consent: Written informed consent was obtained from the patient.

Authors contributions: G.A.E. constructed the main idea and hypothesis of the study. A.R.K and K.F.T. developed the theory and arranged/edited the material and method section. N.Y. has done the evaluation of the data in the Results section. Discussion section of the article was written by A.R.K., K.F.T., N.Y., G.A.E reviewed, corrected and approved. In addition, all authors discussed the entire study and approved the final version.

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