

Biliopleural Fistula Due to a Rare Liver Hydatid Cyst

Nadir Görülen Karaciğer Hidatik Kistine Bağlı Bilioplevral Fistül

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Öz

Hidatik kist (HK) hastalığı Echinococcus tenyalarının larvalarının neden olduğu, fekal oral yolla bulaşan paraziter bir enfeksiyondür. İnsan vücudunda en sık karaciğerde paraziter enfeksiyona neden olmaktadır. İkinci sıklıkta yerleştikleri organ ise akciğerlerdir. Karaciğer HK'e bağlı Bilioplevral fistül (BPF) nadir görülen bir durumdur. Hastadaki plevral efüzyonun yeşil renkte ve Plevra sıvısı/serum bilirubin oranının >1,0 üzerinde olması biliotoraksı desteklemektedir. Tedavide biliyer drenaj ve diyafragma tamiri yapılır. 53 yaşında bayan hasta. Karın ağrısı şikayeti ve halsizlik şikayetleri ile hastanemize başvurdu. 8 yıl önce karaciğer HK'i nedeniyle opere edilmişti. Yapılan tetkikler sonucunda bilioplevral fistül(BPF) tespit edildi. BPF sonucunda pnömotoraks ve biliotoraks gelişen hastaya drenaj ve cerrahi tedavi uygulandı.

Anahtar Kelimeler: Ekinokok, bilioplevral fistül, karaciğer hidatik kisti

Abstract

Hydatid cyst (HC) disease is a fecal orally transmitted parasitic infection caused by the larvae of Echinococcus tapeworms. In the human body, they cause parasitic infections most commonly in the liver. The second most common organ is the lungs. Biliopleural fistula (BPF) due to liver HC is a rare condition. Green pleural effusion and the pleural fluid/serum bilirubin ratio being >1.0 are supportive indicators of biliothorax. Treatment involves biliary drainage and diaphragm repair. A 53-year-old female patient. She consulted our hospital with complaints of abdominal pain and asthenia. She had undergone an operation for liver HC 8 years ago. Biliopleural fistula (BPF) was detected after examinations. Drainage and surgical treatment were performed on the patient who developed pneumothorax and biliothorax as a result of BPF.

Key words: Echinococcus, biliopleural fistula, hepatic hydatid cyst

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INTRODUCTION

Hydatid cyst (HC) disease is a parasitic infection caused by the larvae of *Echinococcus* tapeworms. It is transmitted to humans by the fecal-oral route (1). HC disease was first reported by Thebesius in the 17th century and stated to be caused by the dogs of Icelandic fishermen (2,3). It is a common disease in countries focusing on husbandry such as the Mediterranean, Middle Eastern, and South American countries (4). There are 4 forms of *Echinococcus* that can cause disease in humans. Mostly, *Echinococcus granulosus* causes parasitic infections in humans (4,5). To complete their life cycle, they need 2 different hosts, namely the main host and middle host. Animals such as wolves, dogs, and foxes are the main hosts, and the cysts in the intestines of these animals can reach their adult form. In middle hosts such as humans, sheep, and cattle, the larva form of the cyst causes HC disease (5,6).

A significant proportion of embryos entering the human body cause parasitic infection most commonly in the liver since they get stuck in the liver sinusoids acting as a filter. Embryos smaller than 0.3 mm in diameter can pass through liver sinusoids. These embryos arrive at the right valve via the hepatic vein and inferior vena cava (IVC) and reach the lungs, the second most commonly infected organ (6). Bilioleural fistula (BPF) is a rare condition. Generally, liver hydatid cyst develops as a secondary complication to cholelithiasis, choledocholithiasis, malignancy, and surgery (7).

CASE

A 53-year-old female patient. She consulted our hospital with complaints of abdominal pain and asthenia. She had undergone an operation for liver HC 8 years ago. Liver enzymes (SGPT; 53,4U/L,GGT;124 U/L)were high, Crp;73,28 mg/l and the Indirect Hemagglutination Test (IHA) was positive. She was admitted to our hospital with a prediagnosis of colodecolythisis, after 'biliary sludge' was detected in the biliary tracts in the abdominal ultrasonography. The patient underwent endoscopic retrograde cholangiopancreatography (ERCP), and the cyst membrane was detected in the biliary tracts, and the membrane was removed by ERCP.

The patient, who found to be developed pneumonia and pleural effusion symptoms during follow-ups, underwent pneumonia treatment. The patient had chest pain, cough, and shortness of breath. There were rales during inspiration on lung auscultation.

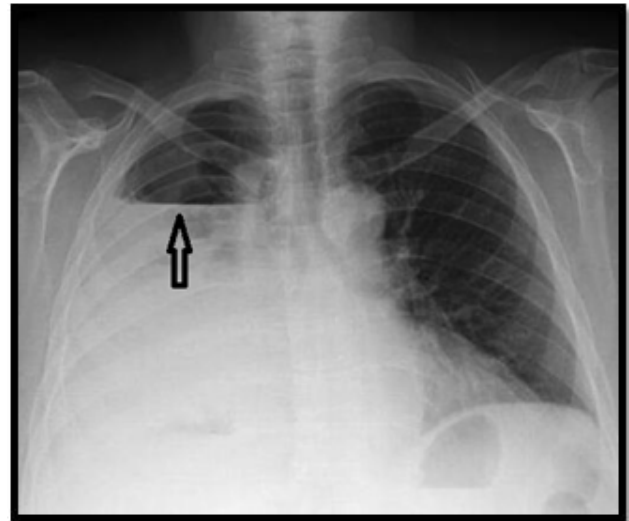


Figure 1. On the patient's posterior-anterior radiograph; pneumothorax line and air-fluid level

Pneumothorax was detected in the patient whose breathing is also deteriorated (Figure 1). The patient underwent closed underwater drainage and chest tube treatment, and it was observed that the fluid drained from the thorax of the patient contained bile. Green pleural effusion and pleural fluid/serum bilirubin ratio >1.0 were supportive for biliothorax. The patient was diagnosed with a bilioleural fistula. When the vital symptoms of the patient, whose general condition was impaired, became stable, she was taken into operation. The patient underwent right thoracotomy. It was observed that both the visceral and parietal pleura of the patient were thickened. It

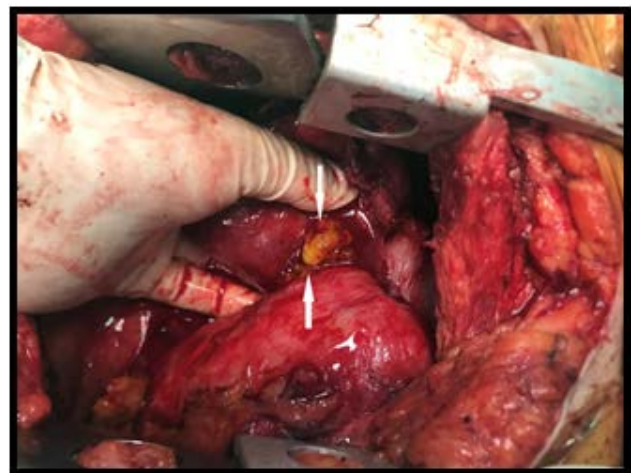


Figure 2. There was a fistula opening into the pleura in the lateral lower lobe of the lung and bile fluid was coming from this area.

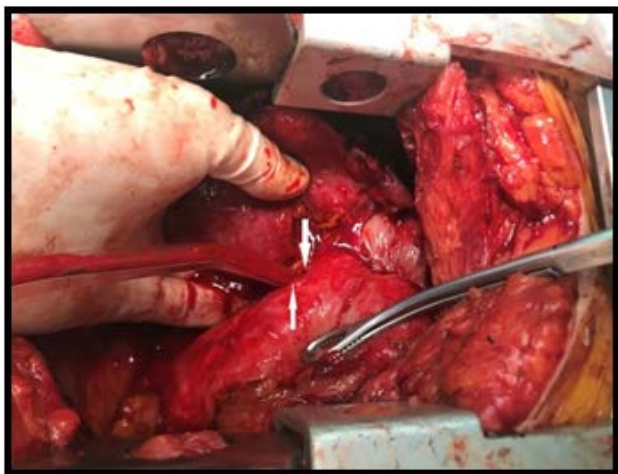


Figure 3. In the diaphragmatic dome, another fistula opening bile fluid was observed.

was found that the patient developed entrapped lung. It was observed that there was a fistula opening to the pleura in the lateral of the lower lobe of the lung and bile fluid was coming from this region (Figure 2).

The thickened visceral pleura of the patient was peeled off. The lungs were expanded. In the diaphragmatic dome, another fistula opening bile fluid was observed (Figure 3). The cystic membrane, which can be seen from the fistula opening in the diaphragm, was removed. Fistula in the diaphragm and lower lobe were repaired with primary sutures. Lung expansion was provided (Figure 4).

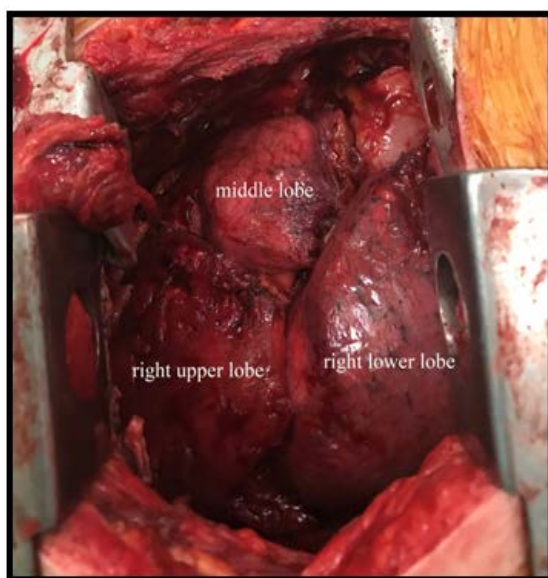


Figure 4. Lung expansion was provided



Figure 5. Posteroanterior chest radiography 2 months after

The patient, whose drainage was found to be stopped during postoperative follow-ups, was discharged after her general condition was improved. The general condition of the patient was good in the 2-month follow-up after discharge (Figure 5).

Diagnosis

Radiological and serological tests are used in the diagnosis of HC. Peripheral blood eosinophilia, increased leukocytosis, and sedimentation rate can be seen in ruptured cysts. However, the symptoms are nonspecific. The diagnostic value of Casoni and Weinberg tests - different serological tests - is around 52% (8). The positivity rate of the Indirect Hemagglutination Test (IHA) method indicating the presence of *Echinococcus granulosus* antibodies, was found to be 78.3% (9). The result of the IHA test was also positive in the case of our patient.

Serological tests were found to be more sensitive in liver cysts. This rate varies between 85% and 98% in liver cysts and 50-60% in pulmonary cysts (6). Our patient had a primer liver hydatid cyst. Since our patient also had symptoms of pneumonia, leukocytosis and increased sedimentation were evaluated as nonspecific symptoms. There are publications in the literature reporting that over 90% of the diagnoses can be made with posteroanterior chest PA radiography and thoracic computerized tomography (CT) (8). Ultrasonography can also be used for liver HCs (5). In our patient, the perforating of the cyst and the occurrence of biliopleural fistula caused pleural effusion and pneumothorax. ERCP is the most used method for the diagnosis of BPF (7). We also used ERCP for our patient. Cyst membranes were found in ERCP, and they were removed.

The dark green color of the pleural effusion and the pleural fluid/serum bilirubin ratio being >1.0 support the diagnosis of biliopleural fistula (10). The pleural effusion in our patient was dark green and the pleural fluid/serum bilirubin ratio was above 1.0.

Treatment

The main treatment of lung HC is surgery. Surgical treatment is recommended as soon as possible to prevent the risk of HC rupture(11). However, medical treatment is recommended for patients with concomitant diseases that cannot be surgically treated and patients with too many cysts that cannot be surgically removed. Unfortunately, oral treatment with albendazole or mebendazole has not been successful in this case (12).

Different tissue features of the liver and lungs may lead to different development of HC disease. Percutaneous treatment used for the livers is not suitable for the lungs (13). While the liver has a solid structure, the lung has a soft structure and contains air. Percutaneous treatment in the lung may cause infection with the communication from a residual bronchogenic cyst (13).

There is no standard surgical technique for the treatment of BPF due to HC. Biliary fistula is still a common complication after surgery. Treatment is basically based on the drainage of the pleural biliary fluid and surgically closing the fistula. In our patient, we first used a chest tube to treat pneumothorax and pleural effusion. When the general condition of the patient has improved, we peeled the visceral pleura thickened by right thoracotomy and closed the fistula openings in the lung parenchyma and diaphragm. ERCP is used both as a diagnosis and treatment method in biliopleural fistula (14). In our patient, cysts were found after ERCP and they were removed. Unfortunately, the fistula could not be closed. After pneumothorax symptom was also detected in the patient, firstly a chest tube was inserted into the patient, and drainage procedure was performed. When the general condition of the patient improved, the patient underwent right thoracotomy.

DISCUSSION

HC causes disease most commonly in the liver (70%) and secondly in the lung 20% (6). In the literature, the coexistence rate of lung HC and liver HC is reported at 10-20% (11). HCs may rupture due to tension due to growth in size, trauma, or surgery. Rupture can be intraperitoneal or intrapleural. They can also cause damage to the biliary tracts, causing

rupture of the intrabiliary areas. Different rates of biliary fistula are reported in the literature 2.5-26.4% (10,15). Intrathoracic spread is at 0.6-16%. The addition of intrapleural negative pressure to the chemical effect of bile content is one of the factors leading to the occurrence of biliopleural fistula(7). It has been reported that the most common cause of BPF is tumors with a rate of 32.3% (2,6).

The green color of the pleural fluid and the ratio of pleural fluid bilirubin to serum bilirubin being greater than 1 are strong evidence of biliothorax (16). In thoracoabdominal traumas, biliary calculus perforation and pleural fluid (Biliothorax) in cholecystitis may occur, aside from liver hydatid cysts (16). Biliary fistula may develop from the biliary tracts to the pleura, after bile duct obstruction, or after cholecystectomy. Treatment involves biliary drainage and diaphragm repair.

Outcome And Follow-Up

Liver HC is one of the rare causes of BPF. In the examinations of our patient, dark green pleural effusion and the pleural fluid/serum bilirubin ratio being >1.0 were evaluated in favor of BPF. BPF treatments are biliary drainage and fistula repair. We first performed drainage with a chest tube for our patient. Then, by applying right thoracotomy, the fistula in the parenchyma and diaphragm was repaired, and the thickened visceral pleura that prevented lung expansion was peeled off. In the postoperative period, rapid improvement in her general condition was observed. The general condition of our patient is very good in the 2 months after the operation.

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