Cumhuriyet Tıp Derg 2011; 33: 361-364
Cumhuriyet Med J 2011; 33: 361-364

Case report-Olgu sunumu

Bronchobiliary fistula: A rare case

Bronkobilier fistül: Nadir görülen bir olgu

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Abstract

Bronchobiliary fistula (BBF), which often presents itself with yellow sputum, is an abnormal communication between the bronchial system and biliary tree. BBF is a serious complication of hydatid cyst of the liver. It is a complication associated with a high mortality rate and requires a well-planned management strategy. We reported a case of BBF which occurred after surgery for hepatic hydatid cyst and was diagnosed by magnetic resonance cholangiography.

Keywords: Bronchobiliary, cyst, magnetic resonance cholangiography.

Özet

Bronkobiliyer fistül genellikle kendini sarı renkli balgam ile gösteren bronş ve bilier sistem arasında bir bağlantı oluşmasıdır. Bronkobilier fistül karaciğer kist hidatiğinin ciddi bir komplikasyonudur. Komplikasyonları yüksek mortalite oranına neden olduğundan dolayı iyi bir tanı ve tedavi stratejisi izlenmelidir. Biz karaciğer kist hidatik operasyonundan sonra bronkobiliyer fistül gelişen ve manyetik rezonans kolanjiografi ile tanı koyduğumuz vakayı sunduk.

Anahtar sözcükler: Bronkobilier, kist, manyetik rezonans kolanjiografi

Geliş tarihi/Received: June 20, 2011; **Kabul tarihi/Accepted:** September 13, 2011

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Introduction

Bronchobiliary fistula, (BBF) which often presents with yellow sputum, is an abnormal communication between the bronchial system and biliary tree [1]. BBF has also been reported as a congenital malformation; however, in most cases, it occurs following liver pathologies particularly parasitic infections, pancreatitis, cholecystitis, subdiaphragmatic abscess, thoracoabdominal trauma and strictures of the biliary tract secondary to postoperative obstruction of biliary system [2, 3]. In 1850, Peacock described the first case of BBF in a patient presenting with liver hydatidosis. The incidence of BBF has been reported as 0.6 to 0.9% in the literature [4, 5]. BBF is a serious complication associated with a high mortality and morbidity rate and requires a well-planned management strategy [6].

In this case report we described a case of BBF which occurred after surgery for hepatic hydatid cyst and was diagnosed by magnetic resonance cholangiography.

Case report

Seventy-one year-old female patient was admitted to our clinic with cough which did not respond to medical treatment after three consecutive months, about 400 cc of daily yellow sputum history, and right side chest pain.

The patient had undergone cystotomy operation for hydatid disease of the liver five years ago. Physical examination revealed that her breathing sounds were decreased at the right hemithorax under the scapula, abdominal tenderness at the right upper quadrant and other systems were normal. Computerized tomography scan showed necrotic areas and consolidated areas with air bronchograms on the diaphragmatic site of lower lobe of the right lung. Interlobar septal thickening was observed in the parenchyma adjacent to the lesion. Nodular pleural thickening of the anterior wall of the lower lobe of the right lung was observed, within the plane of the investigated liver sections, the calcifications in the right lobe of liver and fluid collections (compatible with bilioma) were detected (Figure 1a and b).

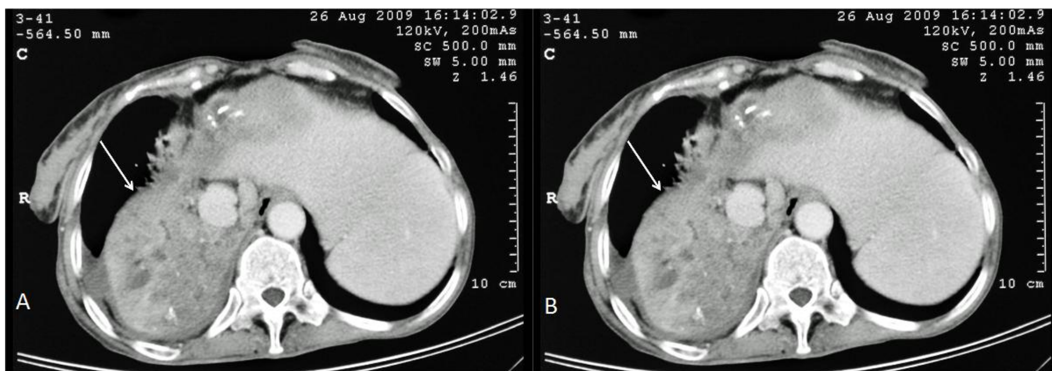

Figure 1a. Computerized tomography of the thorax revealing necrotic areas and consolidated areas with air bronchograms on the diaphragmatic site of lower lobe of the right lung. Nodular pleural thickening of the anterior wall and 1b. liver calcifications and fluid collections (bilioma compatible image).

Yellow secretion at the entrance of trachea and a contraction due to deformation of the entrances of the lower lobe basal segment were observed during the fiberoptic bronchoscopy and density of the yellow sputum was increased at the lower lobe (Figure 2)

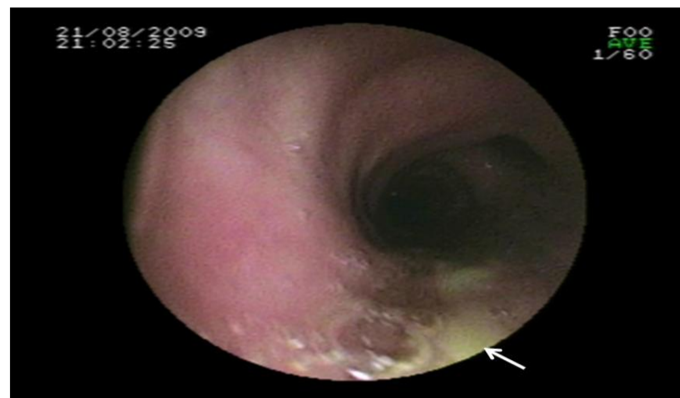

Figure 2. Yellowish bile secretion in bronchoscopic imaging at the lower lobe basal segments of the right lung

Conjugated and total bilirubin levels were determined in the bronchial lavage fluid (0.12 mg/dL and 2.02 mg/dL respectively). Conjugated and total bilirubin levels were also determined in serum (0,44mg/dL and 3,3mg/ dL respectively). MR cholangiography was performed for definitive diagnosis which showed loculated fluid collections in the subcapsular area of the right liver lobe that was also extending to the thorax and dilated intrahepatic bile ducts. Thoracic sections revealed a consolidated area with mucus plugs on the diaphragmatic site of lower lobe of the right lung. (Figures 3a and b) The patient was consulted to the thoracic surgery clinic. Fistulotomy and right lower lobe segmentectomy were planned.

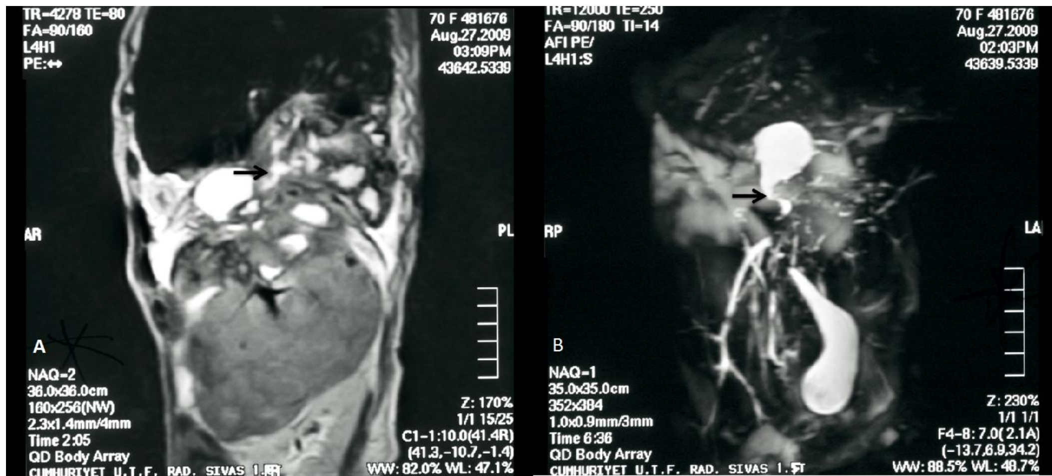

Figure 3a. Sagittal T2A MR and 3b. MR Cholangiography showed fistula tract between dilated intrahepatic bile ducts in liver dome and dilated bronchi at the lower lobe of right lung

Discussion

BBF occurs following liver pathologies particularly parasitic infections, pancreatitis, cholecystitis, subdiaphragmatic abscess, thoracoabdominal trauma and strictures of the biliary tract secondary to postoperative obstruction of biliary system [2, 3]. Although hydatid disease is still the leading cause of BBF, various other reasons are responsible for 20% of the reported cases [1]. Our case had undergone cystotomy about five years ago, due to hydatid disease of the liver. BBFs show itself as yellow colored bilious cough. Bile behaves as a strong irritant when present outside the biliary channel and gastrointestinal system. An irritating cough and expectoration of yellow sputum, termed bilioptysis are the typical presentation of BBF [1]. This patient was admitted to our clinic with cough which did not respond to medical treatment after three consecutive months and about 400 cc of daily yellow sputum history in concordance with the previous literature. Significant increase in bilirubin levels in sputum is important for early diagnosis of BBF and indicates direct communication between the biliary system and bronchial tree [1]. In our case bilirubin levels were high in bronchial lavage samples. Radiology reveals patch densities dominantly in the right lower lobe; however, these patches may also be present throughout the right lung and even in the left side [4, 5, 7]. The cases can best be defined by performing both computerized tomography of thorax and upper abdominal tomography. Verifying persistent air in cyst pouch in the computerized tomography sections of liver is important for diagnosis [8, 9]. Computerized tomography scan of our case showed necrotic areas and consolidated areas with air bronchograms on the diaphragmatic site of lower lobe of the right lung, liver calcifications in the right lobe and fluid collections (compatible with bilioma). Endoscopic retrograde cholangiopancreatography (ERCP), MR cholangiography and bronchography are used to support the radiological diagnoses in these cases. MR cholangiography findings of

loculated fluid collections in the subcapsular area of the right liver lobe that was also extending to the thorax and dilated intrahepatic bile ducts support the suspicion of bronchobiliary fistula. Fiberoptic bronchoscopy is used to demonstrate the yellow-colored mucus (bilioptosi) in these cases [1]. Fiberoptic bronchoscopy showed yellow secretion at the entrance of trachea and density of the yellow sputum was increased at the lower lobe in our case. Treatment method is usually surgery in these cases. Transthoracic approach is recommended in patients with bronchobiliary fistulas. Also endoscopic sphincterotomy is recommended in order to facilitate the flow of bile [10]. Various anatomical resections to the lower lobe of the right lung were performed in patients who were ruptured to thorax [5]. Fistulotomy, diaphragmatic repair, segmentectomy or lobectomy is a good treatment of choice for these cases. Treatment of BBF requires a well-planned management strategy as the mortality and morbidity is high due to this complication. BBF developed after surgery for liver hydatid cyst in this case. Fistulotomy and lower lobe segmentectomy of right lung was planned for the patient.

In conclusion, hydatid cyst is still a common problem in our country. Especially in endemic areas, BBF should be considered in patients having refractory cough and yellow sputum at the same time.

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