

Thoracobiliary Fistula of Calcified Hydatid Cyst of the Liver

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Abstract

Thoracobiliary fistula is a rare complication of hydatid cyst disease of the liver especially in calcified form. Given the rarity and high mortality of such fistula, it is not surprising that there is no consensus about surgical treatment of this condition. In the present paper we report two cases of thoracobiliary fistula of complicated calcified hydatid cyst of the liver. A 64-year-old man with cough and biliary sputum and a 66-year-old man with cough and right biliary pleural effusion (pleurobiliary fistula). The patients underwent left hepatectomy with cholecystectomy and T-tube choledochal drainage in one patient and successful decortication of the lung, and cystectomy with excision of calcified pericystectomy of the liver in another patient. Pathologic examinations revealed calcified hydatid cysts of the liver. Because of poor prognosis of thoracobiliary fistula, radical surgical intervention is recommended.

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Keywords • Echinococcosis • liver • calcification • biliary fistula

Introduction

H ydatid disease is an old topic and many cases have been reported from the Mediterranean area, Australia, New Zealand, and Middle East. In some areas including Iran hydatid disease is endemic and remains a major public health problem. Rupture of Echinococcal cysts leads to notable increase in mortality.^{1,2} Thoracobiliary fistula - as an uncommon condition - is one of the serious complications of liver hydatid cysts.³

Calcified hydatid cyst of the liver can be complicated by thoracobiliary fistula with the same mechanism of non-calcified liver hydatid disease. Toole and colleagues reported the first case of hepatic extension of hydatid cyst from Greece.⁴ Posiotis and colleagues recommended that large cysts (greater than 5 cm), even if they are calcified, should be treated because the parasite is usually viable and the cyst will continue to grow although a portion of its wall is necrotic and calcified.⁵

Liver Echinococcosis with thoracobiliary fistula has been reported frequently in the literature, however, its calcified form has been reported sporadically. In this article we report two rare cases of calcified liver hydatid cyst that produced bronchobiliary and pleurobiliary fistulae.

Case Reports

Patient One

On May 1996, a 64-year-old man (employee) was admitted

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to the Thoracic Surgery ward at Mottahary Hospital affiliated to Urmia University of Medical Sciences. The diagnosis of Thoracobiliary fistula was confirmed by biliptysis (presence of bile in the sputum).

The patient complained of cough, right chest pain, fever, and biliary purulent sputum for two months. His white blood cell (WBC) count was $9800/\text{mm}^3$ with normal eosinophil count. Casoni test was negative. Chest radiography and abdominal ultrasonography revealed a large ($9 \times 8 \text{ cm}$) calcified hydatid cyst in the left lobe of the liver with a fistulous tract to the inferior lobe of the right lung (figure 1).



Figure 1: Calcified Hydatid cyst of the left lobe of the liver with fistulous tract to inferior pulmonary lobe and haustrations of colon below the right diaphragm.

Bronchoscopy showed purulent and biliary sputum in right bronchus. Analysis of bronchial washing and sputum were positive for bilirubin. Bacteriologic examination of sputum was positive for pseudomonas.

After administration of broad-spectrum antibiotics, the patient was scheduled for surgery. After posterolateral thoracotomy and disconnection of thoracobiliary fistula, the diaphragm was opened. The left hepatic lobe was occupied by a large calcified hydatid cyst with no more viable healthy tissue. Left liver lobectomy was performed. After cholecystectomy and exploration of choledochus and T-tube drainage, subdiaphragmatic drainage was performed (figure 2). Pathologic examination revealed calcified hydatid cyst of the liver with multiple gelatinous and brown membranous tissue and severe cyst wall calcification in the left lobe. There was a little hepatic tissue around the calcified hydatid cyst. The patient was discharged 15 days later and T-tube was extracted 34 days after operation. Two-year follow-up of the patient was satisfactory.



Figure 2: T-tube cholangiography shows normal diameter of the choledochus and right hepatic duct with normal drainage of contrast into the duodenum. Left hepatic duct was closed because of the left lobectomy of the liver.

Patient Two

On 21st of March 2008, a 67-year-old man (farmer), was admitted to Thoracic Surgical Ward of our center, with a 10-day history of right thoracic pain and cough. He had recurrent episodes of right thoracic and right upper hypochondriac discomfort and cough in the past three years. He was referred to our center with closed tube drainage on the right hemithorax and with the diagnosis of empyema.

On physical examination he was looking ill and febrile. On abdominal examination he had only right hypochondriac pain. His cardiovascular examination was not remarkable.

The color of secretions in the collecting bottle was purulent biliary with a daily $500-700^{\text{cc}}$ drainage. No bacterial organism was found in microbiological testing of the pleural effusion. The results of fasting blood glucose, erythrocyte sedimentation rate, serum creatinine, and urine analysis were normal. The hemoglobin level was 11 g/dl. Analysis of pleural effusion for bilirubin was positive. Eosinophil count was 5% with $\text{WBC}=11500/\text{mm}^3$. Serologic test for hydatid cyst was negative. The chest radiography showed right pleural effusion with basal infiltration of the right lung. Chest radiography and abdominal ultrasonography revealed a large calcified liver cyst ($10 \times 8.9 \text{ cm}$), almost occupying the whole right lobe of the liver. Other parts of abdomen, spleen, pancreas, and kidneys were normal.

The patient was prepared and underwent surgery using right posterolateral thoracotomy through 8th intercostals space. After decortication and investigation of right pleural space,

there was not any fistula to the lung. Fistula of the liver on diaphragm to pleural space was opened and calcification of compressed biliary contents was almost totally excised (figure 3). On excision of the calcified part, hemorrhage was severe, but it was controlled. Between 1 to 2 cm of calcification of the depth of cyst was adhered to inferior cava and superior hepatic veins, which remained intact. Biliary fistulae were closed partially by suturing with Vicryl. Partially calcified liver cavity and bare area of the liver drained separately through sub diaphragm. In macroscopic pathologic examination there was multiple creamy brownish membranous tissue and severe cyst wall calcification. In microscopic examination there was brown yellowish laminated membrane with destruction of germinative layer and scolices, and severe fibrosis and calcification of pericyst layer, which confirmed calcified liver hydatid cyst. One-year follow-up of the patient was satisfactory.



Figure 3: Calcified Hydatid cyst of the right lobe of the liver through opened pleurobiliary fistulous tract are shown after right thoracotomy and decortication of the lung.

Discussion

As a complication of hydatid cyst disease of the liver, bronchobiliary fistula is a rare condition, which can be manifested as biliptysis. The disease represents with chronic cough, right shoulder pain, fever, and bile stained sputum. Abdominal signs are less remarkable.⁶ For unknown reasons, certain hepatic hydatid cysts stop growing and regress, degenerate, and die. The cyst becomes an amorphous mass and eventually undergoes calcification. These cysts are not only very old but incapable of producing biologically active cysts. Many factors may promote the intrathoracic development of a hepatic cyst. The pressure gradient between pleural and abdominal cavities favors a thoracic direction. Compromised perfusion of the diaphragm secondary to inflammation around the cyst, and the action of bile on the

diaphragm, lung, and pleura are another factor to produce sinus tract communication between the biliary and the bronchial trees or pleural spaces.⁷ Judd believes that, calcified cysts - irrespective of their sites and locations- are best left alone if they are clinically silent.⁸ Ian Mc Conchie reported successful treatment of a case of bronchobiliary fistula with calcified hydatid cyst of liver.⁹ He treated hepatopulmonary fistula by long posterolateral right thoracotomy, disconnection of fistula to pulmonary tissue, complete excision of calcified adventitial wall of hydatid liver cyst and suturing of biliary openings and parenchymal liver cyst wall with sub diaphragmatic drainage.

Tocchi and co-workers studied 31 patients with thoracobiliary fistula of liver hydatid cyst in a 30-year period. They did not mention how many of them had thoracobiliary fistula with calcified hepatic echinococcosis. They noted that radical surgery was more effective in thoracobiliary fistulae of hepatic echinococcosis. Total or partial pericystectomy was preferred to hepatic resection because it avoided unnecessary loss of liver parenchyma. They also noted that conservative treatment of calcified liver hydatid cysts with thoracobiliary fistula was complicated postoperatively by infection and recurrence of disease. The efficacy of radical surgery has been confirmed by the absence of postoperative infective complications or thoracobiliary fistula recurrence.^{10,11}

In many cases, bronchobiliary disease will be progressive and severe. So definitive surgery for thoracobiliary fistula entails excision of the fistula tract. And if pulmonary tissue has been destroyed by necrotizing pneumonia, this may entail a wide resection, segmentectomy, or lobectomy to remove all the damaged pulmonary tissue and decortication. Early surgical intervention has been advocated to avoid respiratory complications.^{12,13} Greenen and colleagues noted that after sphincterectomy or T-tube drainage, mean basal sphincter of Oddi pressure decrease to 1 mmHg, favoring the prograde flow of bile into the duodenum rather than into the fistula tract. In most patients this procedure is indicated to prevent bile stasis or biliary obstruction.¹⁴

Conclusion

Thoracobiliary fistula from large calcified hydatid cyst of the liver has been rarely reported in the literature. It can produce complications similar to any other forms of thoracobiliary fistulae such as non-calcified hydatid cysts of the liver. Physicians should be aware of the diagnosis and

treatment of such complications in the endemic areas. There is no consensus about the surgical treatment of calcified hydatid cysts of the liver. Complicated calcified hydatid cysts of the liver need treatment similar to non-calcified hydatid cysts. Surgical approach to these patients is more aggressive than uncomplicated hydatid cysts of the liver.

Conflict of Interest: None declared

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