INTRATHORACIC RUPTURE OF HEPATIC HYDATID CYST- A RARE COMPLICATION OF HYDATID DISEASE

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PRESENTATION OF CASE

Intrathoracic rupture with resultant pleurobiliary and/or bronchobiliary fistula is a rare but potentially life threatening complication of hepatic hydatid cyst. We report a case of a trans-diaphragmatic rupture of hepatic hydatid cyst resulting in hydatidothorax and biliothorax. CECT abdomen and serology test were consistent with hydatid disease. Patient developed sudden respiratory distress during the hospital stay. Chest examination revealed decreased breath sound on the right side of hemithorax. Chest X-ray and USG showed massive pleural effusion on the right side. Tube thoracostomy was done and 1.5 litre of thick biliopurulent fluid was drained. A repeat CECT chest revealed collapsed right lung with rent in the diaphragm. Decortication was done. In post-operative period bile leak from tube thoracostomy continued, for which ERCP was done which revealed common bile duct obstruction by membranes of hydatid cyst. Evacuation of the membranes, sphincterotomy and stent placement was done by ERCP. Biliary leak subsided completely after 2 days and patient was discharged on 15th postoperative day without any drain. One stage operation through thoracotomy only, is successful in most of the cases as it facilitates simultaneous evacuation of both pleural and hepatic hydatid cysts.

DIFFERENTIAL DIAGNOSIS

Hepatic hydatid cyst rupturing in to pleural space can mimic amoebic liver abscess.

CLINICAL DIAGNOSIS

Biliothorax is a rare clinical entity associated with biliary tree disease. Biliary contamination in the thorax can occur due to transdiaphragmatic leak of bile. It may present as pleurobiliary fistula or brochobiliary fistula due to erosion of lung parenchyma or the bronchial tree.[1] Bile in the pleural fluid or expectoration of bile is pathognomonic of thoracobiliary fistula.[2]

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We report a case of spontaneous transdiaphragmatic rupture of large hepatic hydatid cyst resulting in massive biliothorax and hydatidothorax.

A clinical diagnosis of spontaneous rupture of hydatid cyst liver in to pleural space through diaphragm was made.

PATHOLOGICAL DISCUSSION

The most commonly involved organ in hydatid disease is liver, followed by lung. Hydatid disease or Echinococcosis is a worldwide parasitic zoonosis. The eggs of the Tapeworm Echinococcus granulosus excreted by carnivores may infect various species of natural intermediate host animals such as sheep, cattle, deer and accidentally humans. Hydatid cysts of E. granulosus primarily form in the liver or in the lungs, but may rarely be found in different organs.[3] The parasite can settle in any organ or tissue in the human body. Rupture of the hydatid cyst may be spontaneous, iatrogenic or traumatic.[4] Spontaneous trans-diaphragmatic rupture of a hepatic hydatid cyst into the pleural cavity is unusual serious complication. We report a case of a giant hepatic hydatid cyst which ruptured spontaneously into the right intrapleural space resulting in pleurobiliary fistula.

DISCUSSION OF MANAGEMENT

A 40-year-old male, labourer admitted in medicine department of GMCH with abdominal pain and distension, vomiting with fever of 102^0 F for two and a half months and jaundice for one month. On examination patient systolic blood pressure 70 mmHg with saturation of 95%, heart rate 85/minute and body temperature 38^0 C. On chest examination, breath sounds were normal with normal bilateral equal air entry. Clinically abdomen was distended and with tender hepatomegaly. Laboratory findings were as follows: HB 9.6 g/dl, WBC 43,000/mm^3 with neutrophil of 95, serum bilirubin 2.0mg % [ref range 0.2-1.0], serum albumin 2.2mg/dl [ref. 3.8 -5.5], positive indirect hemagglutination test for E. granulosus. Chest X-ray was normal with mild blunting of CP angle probably reactive effusion (fig 1). On USG abdomen, liver size was 23 cm and in right lobe of liver there was a large cystic lesion of 16x16 cm size with internal echoes and multiple small cystic lesion within it. CECT abdomen showed hepatomegaly with multiseptated cystic lesion, calcific specks along wall and along septation in right lobe of liver with its subcapsular rupture and multiseptated collection in subcapsular region –s/o hydatid cyst (fig 2). Albendazole 400 mg twice daily was started. During the course of hospital, on 8th day patient suddenly developed dyspnoea, chest pain, increase abdominal distension,

tenderness and coarse crepitus with decreased air entry on right hemithorax. Chest X-ray revealed opaque right hemithorax (fig 3). Right side thoracostomy was done and intercostal tube drain was put. ICD drained 1500 ml of thick biliopurulent fluid and patient's respiratory symptoms were relieved (fig 4, 5).

Fluid send for culture sensitivity and broad-spectrum antibiotics were started. Post thoracostomy USG showed large subdiaphragmatic collection with 1.5 cm of rent in right dome of diaphragm. Surgery consultation was done in view of transdiaphragmatic ruptured hydatid cyst into the right intrapleural cavity and patient was transferred to surgery ward. A repeat CECT chest revealed right empyema thorax and collapsed right lung (fig 6).

Right VATS followed by thoracotomy was done with single lung anaesthesia. Intraoperatively whole of the right pleural cavity was full of glistening white hydatid membrane and bile (fig 7). The pleural cavity hydatid cysts were evacuated by VATS but owing to dense adhesions and for decortication, procedure was converted into open thoracotomy. Pleural cavity was irrigated with warm saline and betadine solution. No cyst was seen in the lung parenchyma. Bile leak from a 2 cm rent in the diaphragm with friable margins could be identified which was closed with 2-0 Vicryl interrupted suture. Due to poor general condition of the patient, exploration of liver hydatid through diaphragm or additional laparotomy was deferred as patient was not maintaining saturation and chest was closed over apical and basal chest drains.

Post operatively the ICD was draining 500 to 600 ml bile per day with persistent jaundice. ERCP done on post op day 10 revealed, multiple filling defects in the form of linear and round opacities in the common bile duct. Sphincterotomy was done and few green muddy membranes, causing distal biliary obstruction were removed from common bile duct and a stent was placed it. Post ERCP, ICD output subsided completely after two days and jaundice disappeared. Patient was discharged after removal of ICD tubes on 15 post op day.

Hydatid disease presents as hydatid cysts primarily in the liver and lungs. Although cysts may be asymptomatic for many years, they become symptomatic due to expansion, rupture, and/or pyogenic infection.[3,5,6] Intrathoracic rupture of the hepatic hydatid cyst with pleurobiliary and bronchobiliary fistula is a rare and potentially life threatening complication of hepatic hydatid disease. Cyst erosion is associated with pericystic inflammation. Adhesion formation determines whether the rupture is confined to lung parenchyma or the free pleural space, or both. Bronchobiliary fistula leads to hemoptysis, bilioptysis and cyst expectoration. The clinical presentation is predominately pulmonary, with abdominal symptoms being less frequent.[7,8] Cough, expectoration, and dyspnea are present in 30% of cases. Jaundice implies cyst rupture into a bile duct with biliary obstruction. In chest x ray lower lobe opacity or pleural effusion is seen. Abdominal ultrasound is necessary to show the morphology of the liver and possible biliary tree obstruction or stones. Computed tomography aids in differentiating the relationship among the cyst, blood vessels, and bile ducts, and identifying other possible sites and assisting in the choice of surgical approach.[7-10] On CT, it can be easily differentiated from other mass lesions due to its typical three-layered structure and the ring-like calcification. Various serological tests available for the diagnosis, screening and postoperative follow up for recurrence are: hydatid immunoelectrophoresis, enzyme linked immunosorbent assay, latex agglutination and IHA test. In our case the CT scan findings were suggesting possibility of liver hydatid cyst and a positive serology test confirmed the diagnosis.

Role of bronchoscopy is limited role as visualization via small tract is impossible, however it may show bile, daughter vesicles, or fragments of cyst membrane, bronchial tree and help in assessment of the severity of inflammatory process, not done in this case.

Various factors which lead to the intrathoracic rupture of the hepatic hydatid cyst are: the pressure gradient between the pleural and abdominal cavities favors a thoracic direction; compression and ischemia of the diaphragm secondary to inflammation about the cyst; and the chemical action by bile on the diaphragm, lung, and pleura.[7,8] Four types of thoracobiliary fistulae have been described in litrature.[11] Type I: direct fistulization of the cyst into the bronchi with small, I-A, or large fistula, I-B; Type II: intrapulmonary parenchymal collection without fistula, II-A, or with large fistula, II-B; Type III: intrapleural encysted collection without, III-A, or with bronchitic fistula, III-B, or parietal fistula, III-C; and Type IV: rupture into the pleural cavity with pleural effusion (hydatidothorax), IV-A, or a secondary pleural hydatidosis, IV-B. Hyponatremia and hypoproteinemia are usually related to bile leakage and should be corrected preoperatively. Hypoxemia is frequently noted. Postoperative results are directly related to correction of fluid and electrolyte imbalance, aspiration of the bronchi, respiratory physiotherapy, and antibiotic agents. Thoracotomy offers adequate simultaneous access to both the chest and hepatic lesions.[8,10,12,13] Other authors use only laparotomy to treat this disease.[7,9,14] Some authors have recommended endoscopic sphincterotomy for relief of biliary obstruction.[15] ERCP was also done in our case, for biliary obstruction and bile drainage in chest tubes postoperatively. This technique may be a reasonable alternative to laparotomy.

Mortality rate of this disease is high, varying from 9% to 43%.[7,8,14] Except for type IV-B and rarely in type IV-A, medical adjuvant treatment is never required for a ruptured intrathoracic hepatic hydatid cyst. It is indicated when dissemination is confirmed and total resection of the cysts is not possible. Albendazole 400 mg twice daily for 6 months is the current recommended therapy when indicated. In endemic areas, prophylactic measures should always be taken but surgery remains the treatment of choice in large hepatic hydatid cysts.

FINAL DIAGNOSIS

This case demonstrates that early surgical intervention should be offered in large hepatic hydatid cyst as potentially life threatening complication like thoracobiliary fistula can result due to its transdiaphragmatic rupture into the intrapleural space espescially with coexistent distal biliary obstruction. One stage operation through thoracotomy is mostly successful to address both thoracic and hepatic lesions.

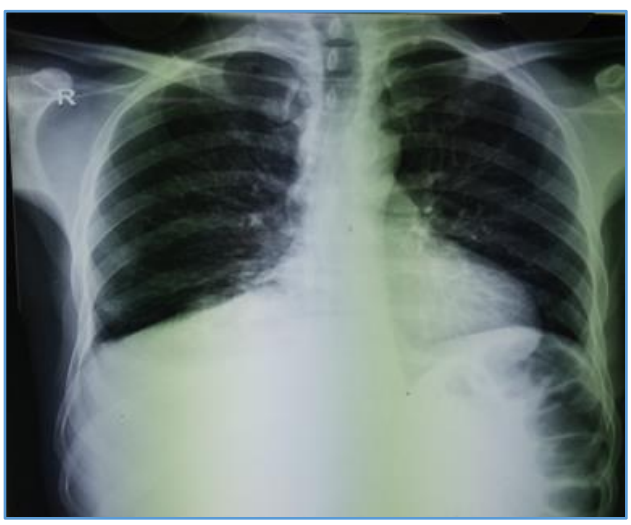

Figure 1. Chest X-Ray on Admission; Reactive Right Side Effusion

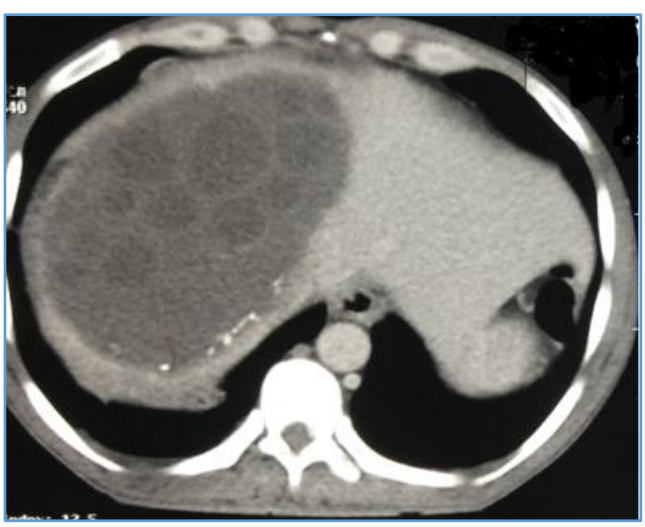

Figure 2. CT Abdomen Hydatid CYST Liver with Calcified Wall

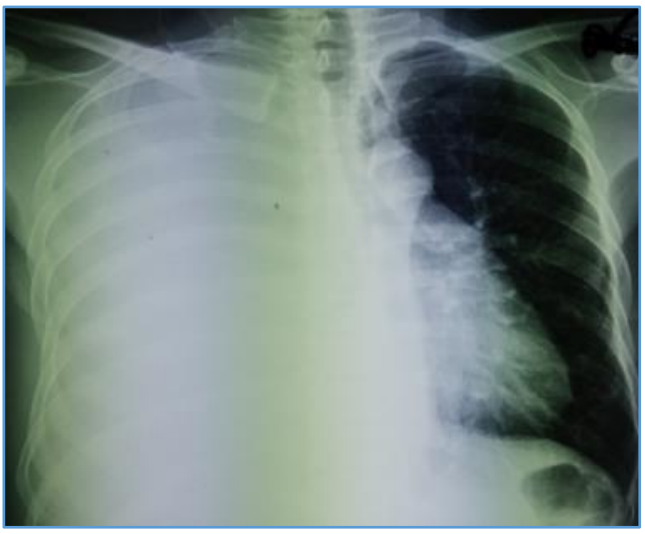

Figure 3. Right Side Massive Pleural Effusion

Figure 4. ICD Draining Bile

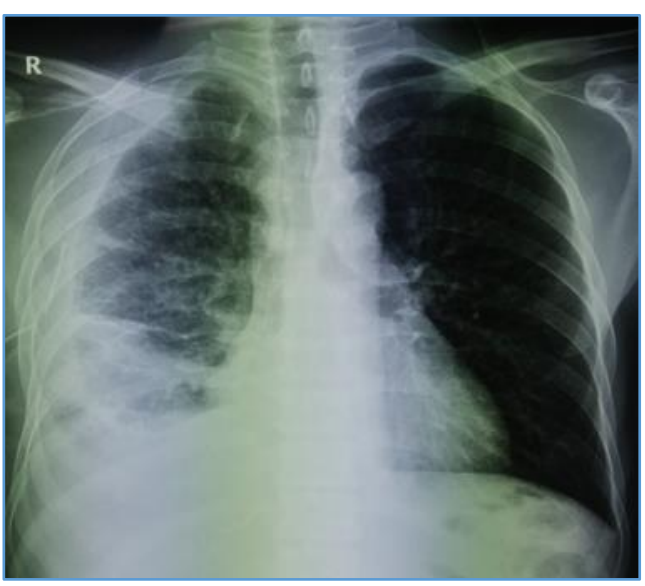

Figure 5. Post ICD x-ray Showing Lung Expansion with Residual Effusion

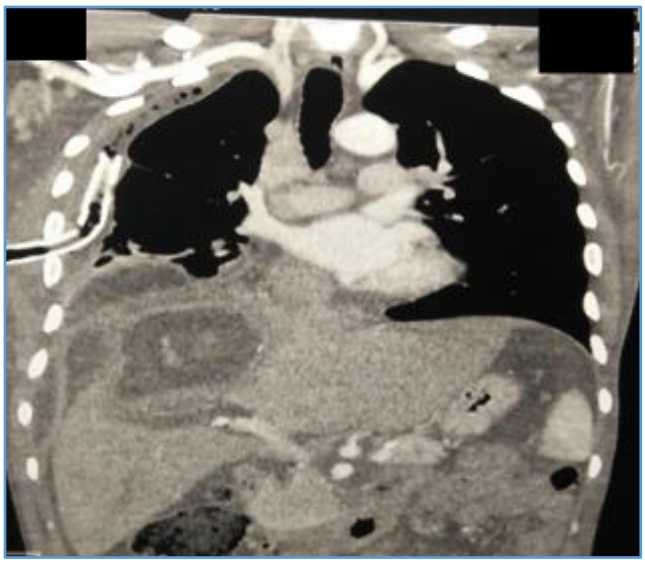

Figure 6. CT Chest; Rupture Hydatid Cyst into the Pleural Space with Diaphragmatic Rent

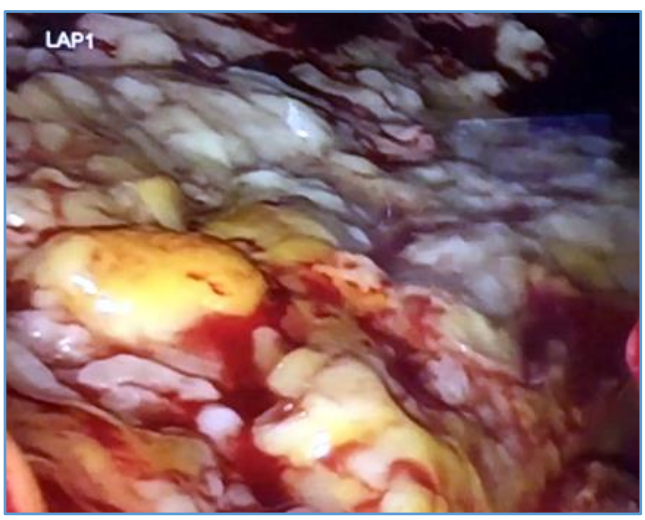

Figure 7. Bile Stained Hydatid Cyst Membranes Covering Whole of Lung Parenchyma

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