

Transdiaphragmatic rupture of hydatid cyst: A rare case report

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ABSTRACT

Hydatid disease is a major zoonotic disease worldwide. It remains a serious health problem in developing countries and in countries where animal husbandry is common. Here, we report the case of a 40-year-old male who presented with chief complaints of pain in the right upper quadrant of the abdomen and swelling of the right side lower chest for 2 days. He mentioned intermittent episodes of pain within the past 7 days, the frequency and severity of which is increasing in the past 2 days. Chest X-ray suggestive of opacity of the right lower hemithorax with blunting of cardiophrenic angle seems like pleural effusion with opacity in the right upper lobe but on ultrasound, cystic area with multiple septations found in right lung parenchyma likely suggestive of hydatid cyst.

Key words: Echinococcosis, Hydatid cyst, Zoonotic disease

Hydatid disease is a major zoonotic disease that remains a serious health problem in developing countries and in countries where animal husbandry is common. Among six recognized species of echinococcosis and four being of public health concern, *Echinococcus granulosus* causing cystic echinococcosis (CE) is the most common followed by *Echinococcus multilocularis* causing alveolar echinococcosis and *Echinococcus vogeli* and *Echinococcus oligarthrus* causing polycystic echinococcosis [1]. CE is the most common presentation in humans, contributing to more than 95% of the estimated 2–3 million global cases [2]. A definitive host for *E. granulosus* is a dog (or other carnivores), which harbors adult worms in their small bowel and releases the eggs in feces. Excreted ova are ingested by an intermediate host (most commonly sheep) and in the intestine of an intermediate host, ova develop into a hexacanth embryo and reach the liver through the portal circulation. Embryos in the liver develop into cysts. Humans are accidental hosts and become part of this lifecycle through contact with a definitive host or the ingestion of water or vegetables contaminated with echinococcal ova [3]. The initial growth of a cyst is usually asymptomatic until symptoms are caused by the cyst's space-occupying mass effect, mechanical obstruction, rupture, or secondary allergic reaction [4]. Anaphylactic reactions are rare presentations of ruptured hydatid cysts [5]. Rupture may occur spontaneously, following trauma, due to increased abdominal pressure which can also be iatrogenic. Diagnosis of a ruptured hydatid cyst is difficult and hydatid cyst

may be misdiagnosed as another disease, which delays correct treatment. Rupturing of cysts predisposes to infections and allergic reactions and is associated with poor outcomes. Imaging technology including ultrasonography, computed tomography (CT), magnetic resonance imaging, and serological tests could be done for diagnosis. Definitive diagnosis is made by aspiration of the cyst content to find protoscoleces. An aspirating cyst may increase the risk of anaphylactic reaction due to spillage of cyst content and rupture of the cyst. After cystectomy or surgical drainage of the cyst, cyst fluid should be sent for protoscoleces examination to confirm the diagnosis.

CASE REPORT

A 40-year-old male, driver by occupation, presented with chief complaints of pain in the right upper quadrant of the abdomen and swelling of the right side of the lower chest for 2 days. He mentioned intermittent episodes of pain within the past 7 days, the frequency and severity of which is increased in the past 2 days. The pain worsens with breathing and sitting position. The patient observed swelling of the right lower chest for the past 2 days which was associated with pain and dyspnea. There is no history of fever, yellowish discoloration, or itching of the overlying skin. There is no history of cough, exertion, or any lifting of any heavyweight. The patient had a past history of recurrent hydatid cyst liver in 2000 and 2014, hydatid cyst spleen in 2014, the patient underwent left lobe hepatectomy in 2000, and splenectomy with cholecystectomy with cystopericystectomy liver in 2014. The patient was treated with tablet albendazole 400 mg twice a day

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for 1 month, in 2000 and 2014. The patient also had a history of tubercular pleural effusion in 2012, for which the patient was taken antitubercular treatment for 6 months. He was a reformed smoker and alcoholic.

The patient was calm, conscious oriented to time, place, and person on examination. His vitals on presentation were blood pressure 130/80 mmHg, pulse rate 82/min, respiratory rate 22/min, and saturation 97% on room air and were clinically afebrile. Physical examination of the chest revealed bulging of the right infra-mammary and infra-axillary area with no overlying redness, itching, or local rise of temperature present. On auscultation, air entry was decreased in the right infra-axillary, infra-mammary, and infra-scapular areas. On per abdomen examination, tenderness was present in the right upper quadrant with an enlarged liver, palpable 4 cm below the costal margin in midclavicular line along with the presence of a scar mark from previous surgeries. The rest of the examination was unremarkable.

Routine investigation related to complete blood count, renal function test, and liver function test was within normal limits. Chest X-ray was suggestive of opacity of the right lower hemithorax with blunting of cardiophrenic angle which seems like pleural effusion with opacity in the right upper lobe but on ultrasonography (USG), cystic area with multiple septations was found in the right lung parenchyma likely suggestive of hydatid cyst (Fig. 1a). USG whole abdomen done suggestive of 4×5 cm, ill-defined cystic lesion with multiple septations present in the right liver lobe, and another 4×3 cm hyperechoic area with the presence of peripheral calcification.

Contrast-enhancement CT chest and whole abdomen done were suggestive of transdiaphragmatic rupture of the liver hydatid cyst. A 4.5×5.2×3.4 cm cyst was present in the right lobe of the liver extending to the right hemithorax, and an 11×0.8×13.3 cm multicystic lesion present extending up to the pre-vertebral space at T9–T10 level. Multiple small cysts were also present in the right lung along the pleural border and the right lobe of the liver (Figs. 1b and 2).

The patient was managed surgically and the hydatid cyst liver along with the remnant of the ruptured cyst was excised. The patient was operated on single lung ventilation to prevent contralateral lung contamination and for easier removal of the cyst from a static lung. The diaphragmatic rupture was restored. The sample was sent for serological testing and a diagnosis of hydatidosis was confirmed. The patient was discharged on tablet albendazole 400 mg twice a day for 4 weeks and tablet praziquantel 2400 mg twice a week for 2 weeks.

DISCUSSION

A hydatid cyst contains parasite, endocyst, and exocyst. The endocyst is an elastic hyaline membrane that contains fluid and daughter colices while the pericyst is a thick fibrous layer formed due to host reaction. Pericyst in the lung does not develop well enough that of liver, and hence, risk of rupture is more in the case of pulmonary hydatid cysts [6]. Hydatid cysts are commonly found in the liver followed by the lungs. In most of the patients,

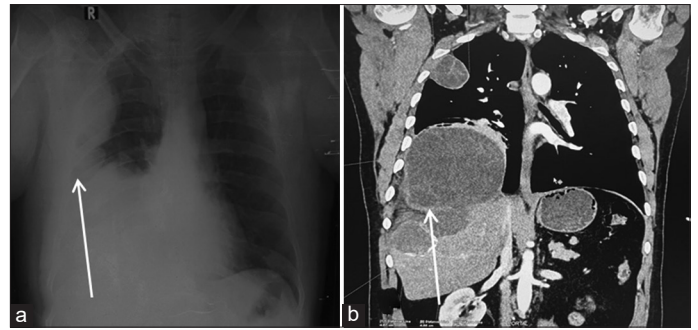


Figure 1: (a) Chest X-ray showing blunting of cardiophrenic and costophrenic angle with opacity in right lung; (b) computed tomography showing transdiaphragmatic herniation of hydatid cyst

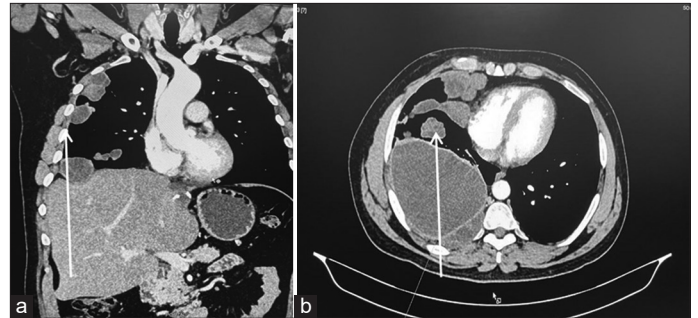


Figure 2: Computed tomography scan showing multiple hydatid cyst along the pleural border along with herniated hepatic cyst

a single cyst is found which may remain asymptomatic for years. The symptoms depend on the size and site of the cyst. They may be discovered incidentally or during radiological investigations. Cysts may involve any organ and can also erode the bone.

Liver hydatid cysts may remain asymptomatic, and symptomatic patients may present with pain abdomen or a palpable mass in the right upper quadrant. If it involves a biliary tree, it may mimic cholangitis, cholelithiasis, or biliary obstruction and can lead to obstructive jaundice. Rupture of hydatid cyst may present as fever, pruritus, urticaria, eosinophilia, or severe allergic reactions. Liver hydatid cysts may rupture into the peritoneum, biliary system, retroperitoneal, and into the lungs transdiaphragmatically. They can lead to hydatid cysts in the lung or bronchobiliary fistula by accessing the bronchial tree through transdiaphragmatic transmission. Dome cysts in the liver that has become excessively large can erode the diaphragm muscles and perforate the thorax. The complications with the diaphragmatic involvement of the liver cysts and intrathoracic propagation are reported at a rate of 0.6–16% [7].

These complications many of which are seen in the cysts located in the liver dome can generally originate from factors including spontaneous, traumatic, and increase of intra-abdominal pressure. The pressure gradient between the thorax and abdomen cavity, diaphragmatic ischemia secondary to compression, inflammation in the cyst membrane, and the chemical effect of the gallbladder on the diaphragm are the other predisposing factors [7]. If the liver dome cysts become a perforated thorax, they may lead to pleural effusion, empyema, or parenchymal destruction. If it is ruptured into the bronchus, it will present as a productive cough

with salty sputum, hydatid fluid, fragments of cyst membrane, and hemoptysis. The opening of the liver hydatid cyst to the pleural cavity or bronchus is the cause of high mortality (9–43%) and morbidity [8].

It is observed that most of the time whenever there is transdiaphragmatic herniation of the liver hydatid cysts, it remains intact in the liver and gets ruptured in the lung. Rupturing of cysts predisposes to infection which is a serious problem and difficult to manage. The gallbladder has a corrosive effect on the lungs and pleural area. An indication of the presence of bile in the pleural liquid and with the analysis of biloptysis and bilirubin levels is pathognomonic for the bronchobiliary fistula [7]. In the bronchobiliary fistula where the mortality is already high, the surgical treatment must be considered only when no response is given with conservative approaches [7]. Diagnosis can be made radiologically and serologically.

Medical management is advised for all uncomplicated cysts to reduce the cyst size and in case of complicated cysts, surgery is the mainstay of treatment but albendazole should be given preoperative and post-operatively. Adjunctive chemotherapy before and after surgery reduces the risk of reoccurrence by inactivating protoscolices and reducing the tension of the cyst by reducing size for the easy removal of the cyst [6]. For uncomplicated cysts, carcinoembryonic antigen grade I to III, percutaneous aspiration with the infusion of scolicalid agents, and respiration (puncture, aspiration, injection, and reaspiration [PAIR]) are recommended instead of surgery. A minimum 15 min of contact with the scolicalid agent is required to kill protoscolices. PAIR is contraindicated in superficially located cysts due to the risk of rupture and cysts having multiple septations. Treatment of CE depends on the size, site, clinical manifestations, and ultrasonographic staging of the hydatid cyst. Surgery remains the mainstay of treatment. The basis of surgical therapy is the removal of parasites, which prevents contamination of surrounding tissues with maximum preservation of the involved organ. In uncomplicated cysts, a refusal for surgery, inoperable cysts, pregnancy, and patients with CE, CE1, CE2, and CE3 PAIR can be done. Keeping in mind spillage or contamination during the procedure, it is avoided in superficial cysts and impending rupture cysts. Pre- and post-operative chemotherapy with albendazole (15 mg/kg/day) in two divided doses should be given at least 2 days before surgery and up to 4 weeks after the surgery as it will help in reduce the size of cyst and kill the protoscolices that's eases the removal cyst and decreases recurrence. Praziquantel 50 mg/kg daily for 2 weeks

should be given to hasten the death of protoscolices. Response to treatment is best assessed by serial radiological imaging.

CONCLUSION

Hydatid cyst is a common parasitic disease in developing countries like India. The liver and lungs are the most common organs to be involved. Manifestations of which may vary according to the size, site, and associated complications. The most feared complications are cyst rupture or cyst infection. Transdiaphragmatic rupture of liver hydatid cyst is very rare. It may lead to bronchobiliary or bronchocystic fistula which is associated with a poorer prognosis. Treatment of such complicated cysts is mainly surgical. One thing we need to emphasize on, people believe that once they get operated, the disease is eliminated but in the case of hydatid cysts in spite of removing cysts, the risk of recurrence is high as in our case, the patient was operated on twice but no follow-up after that and later on, the patient presented with complications, so serial radiological imaging along with the chemotherapy should be advised to the patients and to be followed up with the same.

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