

Secondary pleural hydatidosis: Complication of intrapulmonary echinococcosis

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ABSTRACT

Hydatid disease has a wide geographic distribution around the world. In human, the liver is the most commonly affected organ, followed by the lungs. Intrathoracic extrapulmonary locations are generally the mediastinum, pleura, pericardium and chest wall. Pleural involvement usually follows the rupture of a pulmonary or hepatic cyst inside the pleural space causing secondary pleural hydatidosis. We report four cases of patients who were referred to our hospital for management of pleural hydatid disease as a complication of intrapulmonary echinococcosis.

KEY WORDS: Complicated hydatid cyst, lung, pleura

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INTRODUCTION

Echinococcosis or hydatid cystic disease is caused by larvae of the tapeworm *Echinococcus*. It is an endemic infection which has a wide geographic distribution. Its incidence in Tunisia is still high; the disease reportedly affects 15 per 10,000 inhabitants. Humans may be infected incidentally as intermediate hosts in the parasite's life cycle by the accidental consumption of the soil, water or the food that has been contaminated by the fecal matter of an infected dog.^[1]

The most commonly affected organs are the liver and the lungs.^[2] Pleural involvement is rare and usually follows the rupture of a pulmonary or hepatic cyst inside the pleural space.^[3] The purpose of this study was to exhibit some characteristics of secondary infestation of the pleura as a complication of intrapulmonary echinococcosis.

PATIENTS AND METHODS

We retrospectively reviewed the medical records and the chest X-ray (CXR) of the patients who had been hospitalized

for pulmonary hydatid disease at the center for Chest Diseases of Hedi Chaker hospital, Sfax, Tunisia, between January 1993 and December 2012. We identified those patients whose CXR demonstrated pleural abnormalities. Data were collected from the medical records of patients in the archives and analyzed in terms of age, gender, clinical and radiological features, cyst localization, diagnostic tools, operative techniques, complications, morbidity and mortality.

CASE REPORTS

Case 1

A 38-year-old male patient resident of Sidi Bouzid (Tunisia) was admitted to hospital with cough, left thoracic pain and increasing dyspnoea of two months duration. He was a tobacco smoker (3 pack-years). Physical examination revealed the dullness to the percussion and diminished breath sounds over the left lower lung field. A CXR [Figure 1] showed a large left-sided hydropneumothorax with significant shift of midline structures. Electrocardiographic data were normal. Laboratory studies revealed hemoglobin of 13.8 g/dL, total leucocytes count of 16400 cells/mm³, erythrocyte sedimentation rate (ESR): 47 mm/hr. Arterial blood gas values, on room air, revealed hypoxemia (arterial oxygen pressure PaO₂: 76 mmHg). Thoracic computed tomographic (CT) scan revealed large left-sided hydropneumothorax due to ruptured hydatid cysts [Figure 2]. Surgical intervention was indicated. The patient underwent resection of the pulmonary cyst without postoperative chemotherapy instituted. There

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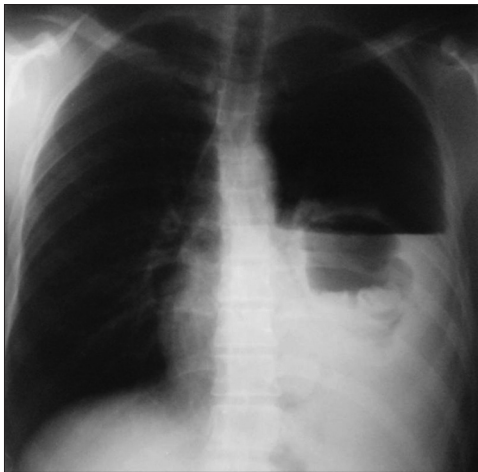


Figure 1: Chest X-ray: Large left-sided hydropneumothorax with significant shift of midline structures

were no complications in the postoperative period and no recurrence of the disease during a follow-up of 6 months.

Case 2

A 68-year-old male patient was admitted with a 5-month history of cough and chest pain. In the past story of the patient, there was a concept of contact with carnivores and a sheep. Physical examination revealed decreased breath sounds over the all the right lung and scattered rhonchi were heard in his lungs bilaterally. The admission chest radiograph was initially interpreted as showing right plentiful pleural effusion, a right lower lobe consolidation and a left oval cystic lesion. At the abdominal ultrasound, the appearance of the liver, spleen, gall bladder and pelvic organs was normal. Investigations revealed a normal complete blood count, an elevated ESR, and a negative indirect hemagglutination test. Arterial blood gas values, on room air, revealed very hypoxemia (PaO_2 : 61 mmHg). A right posterolateral thoracotomy was performed for the patient. Surgical treatment consisted on removing the pleural fluid, the cystic membrane and total pericystectomy. There was a septic shock in the postoperative period which had evolved with antibiotic and inotropic support. The antihelminthic agent albendazole (10 mg/kg) was administered daily for three months postoperatively. There was no recurrence of the disease during a follow-up of six months.

Case 3

A 68-year-old female patient presented with a history of cough, right chest pain and low abundance hemoptysis since 40 days. Physical examination revealed tachypnoea with respiratory rate of 24/min, tachycardia with heart rate of 110/min, and the blood pressure was 130/70 mm Hg. There was no cyanosis and temperature was 37°C. Chest examination revealed diminished movements over right hemithorax with decreased breath sounds in the right infra scapular, infra axillary and mammary area. Cardiovascular examination was unremarkable except for tachycardia.



Figure 2: CT: Large left-sided hydropneumothorax due to ruptured hydatid cysts

Per abdominal examination revealed no organomegaly. Investigations revealed hemoglobin of 12.1 g/dL, total leucocytes count of 8400 cells/mm³. ESR: 110 mm/hr. Electrocardiogram showed sinus tachycardia. Emergency chest radiograph showed a left oval cystic lesion and a right-sided hydropneumothorax with significant shift of midline structures. Ultrasound abdomen was normal. CT thorax showed cavitory lesion measuring 8.5 × 8 cm, involving the right lower lobe. Folded membrane like structure was seen with in the cavity. All lesions were associated with pleural effusion. CT features suggested ruptured hydatid cyst on the pleura with significant shift of midline structures. The rest of lung fields showed left oval cystic lesion. Patient was referred to surgeon for consideration of surgical intervention. The decortication procedure was performed with cystotomy. Patient was discharged on seventh day after the surgery. Albendazole was started at the dose of 10 mg/kg as a postoperative prophylactic measure to prevent recurrence.

Case 4

A 53-year-old male patient presented to the medical emergency with a 9-day history of cough and right pleuritic chest pain which increased with inspiration and cough. Physical examination revealed tachypnoea with respiratory rate of 23/min. Blood pressure was 130/70 mm Hg. Temperature was 39°C. The chest examination revealed diminished movements over the right hemithorax with decreased breath sounds and persistent crackles. The CXR showed uniform opacity in the lower part of the right hemithorax with a concave upwards upper border [Figure 3]. CT of the chest showed cavitory lesion measuring 7.5 × 7 cm, involving the right lower lobe surrounded by parenchymal condensation and right pleural effusion. Folded membrane like structure was seen within the cavity [Figure 4]. CT of the abdomen showed a residual calcified lesion in the right liver lobe. Investigations revealed hemoglobin of 12.3 g/dL, total leucocytes count of 12,300 cells/mm³. The hydatid antibody level was raised at 1/2560 (indirect hemagglutination test). Electrocardiogram was normal. The patient was posted



Figure 3: Chest X-ray: Uniform opacity in the lower part of the right hemithorax with a concave upwards upper border

for decortication and right posterolateral thoracotomy was done. Parietal pleura was thickened >1 cm and visceral pleura was approximately 4 mm. We removed the pleural fluid and performed visceral and parietal pleural decortication. A ruptured hydatid endocyst was found adhered to the thickened pleura overlying the right lower lobe and was removed. A bronchopleural fistula was seen originating from floor of probable site of ruptured cyst cavity in the right upper lobe of lung which was repaired. On long-term follow-up, the patient remained healthy and there was no recurrence.

DISCUSSION

Cystic hydatid disease, known in times of Galen, is an anthroponosis produced in the intermediate hosts by larvae of the parasite platyhelminth *Echinococcus granulosus*.^[4] It appears to be more common in younger individuals. Hydatid disease commonly occurs in the first three or four decades of life.^[5] The larvae most often involves the liver (55-70%) followed by the lungs (18-35%); the two organs can be affected simultaneously in about 5-13% of cases.^[6] However, any organ may be involved.^[7] Echinococcal infestation of the pleura or chest wall has been reported to occur in 0.9-7.4% of patients with hydatid disease.^[3,7] It may be primarily caused by direct hematological or lymphatic larval seeding or secondarily as a late complication of surgery or chest tube insertion during the therapy for thoracic hydatid disease.^[3,7] Pleural manifestations of hydatid disease may also occur as a complication of intrapulmonary echinococcosis without parasitic pleural infestation.^[3] The increase in the cyst size, factors such as trauma and cough that cause an increase in the intrathoracic pressure may predispose patients to cyst rupture formation.^[8] The incidence, diagnostic characteristics and the patient outcomes associated with this condition are not well understood. The clinical appearance is variable for the perforated hydatid cysts and dependent on the characteristic of the perforation. It varies from non-specific to acute and life-threatening conditions such as anaphylactic shock.^[3] No

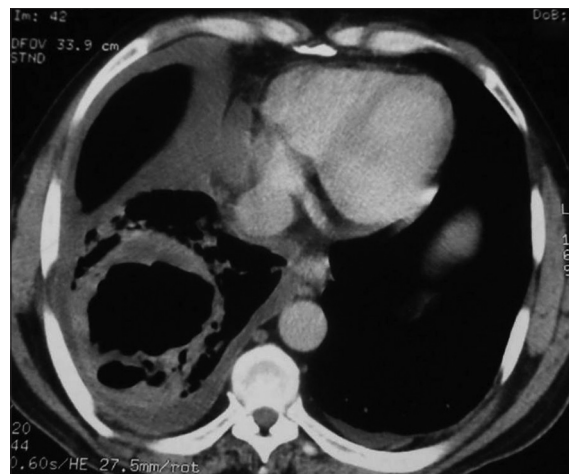


Figure 4: CT: Cavitory lesion measuring 7.5 × 7 cm, involving right lower lobe surrounded by parenchymal condensation and right pleural effusion

one from our patients had this complication. The symptoms depend on the size and site of the lesion and the accessibility of the organ involved for clinical examination.

Radiographic findings can vary from simple pleural effusion to complicated empyema, pneumothorax, or hydropneumothorax.^[9] Two of our patients had hydropneumothorax. In this instance, the transfer of air into the pleural space is facilitated by the connection and attachment of the laminated and germinative membrane of the cyst to adjacent peripheral bronchioles. The diagnosis can be made by combination of clinical presentation, radiographic studies, serological tests and exposure. Different serological tests are being carried out for the diagnosis, screening and post-operative follow-up for recurrence. These include, enzyme-linked immunosorbent assay (ELISA), latex agglutination and indirect hemagglutination test and an immunoblot assay using lentil-lectin purified glycol proteins and it has >99% specificity and the test is highly sensitive.

Although recent reports suggest medical therapy with albendazole and percutaneous treatment with needle aspiration of the cyst.^[10] Surgery remains the treatment of choice for patients with complicated pulmonary hydatid cyst. Surgical intervention consists on a pleurectomy in addition to cyst removal. All of our patients underwent surgery. Medical treatment may be applied in inoperable cases or after surgery to avoid a relapse as an adjunctive therapy. No recurrence of the disease was seen during a follow-up of six months in our study.

CONCLUSION

The probability of hydatid cystic disease should always be considered in cases in which the patient has an undiagnosed pleural effusion and empyema in countries where hydatid disease is endemic. In this area, the prevention of hydatidosis is more important than treatment.

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