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Case report

Primary pleuropulmonary echinococcosis – Varied manifestations

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ABSTRACT

Echinococcosis commonly known as "hydatid disease" is seen worldwide and is caused by eccinococcus larvae. It mainly affects liver (70%) followed by lung involvement (20%) in adults. In the thoracic cavity, it mostly affects lung parenchyma but pleural involvement is well documented. We present three cases of hydatid disease affecting pleuro-pulmonary system as primary site of involvement without hepatic involvement.

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1. Introduction

Hydatid disease has a worldwide presence and cases are commonly seen in Australia, South East Asia, the Mediterranean countries, the Middle East, the southern part of South America, Iceland, New Zealand, and southern parts of Africa.[1] China in Central Asia is also an endemic area.[2] It is a parasitic disease caused by tapeworms of the genus *Echinococcus*. Definitive hosts are dogs and sheep and humans are incidental hosts and get infected by incidental contamination of food and water by parasitic eggs. More than one million people are infected at any point of time worldwide.[3]

Echinococcus granulosus (*E. granulosus*) causes cystic echinococcosis and *Echinococcus multilocularis* causes alveolar echinococcosis. Infection with *E. granulosus* results in the development of one or several unilocular hydatid cysts that in humans develop mainly in the liver (70%), but also lungs (20%) and 10% of cysts can occur almost anywhere in the body (e.g., brain, body musculature, wall of the heart, kidneys, orbit of the eye, marrow cavity of bones). *E. multilocularis* metacestodes is a series of small, interconnected cysts, growing as a metastasising lesion almost exclusively in the liver (98%–100%), but in the later phase of infection distant metastases anywhere in the body may occur.[4]

Suspicion of the disease still remains a challenge for the clinician as it can have most varied and bizarre presentation. Imaging techniques like Xray/ computed tomography/ magnetic resonance imaging are the main diagnostic tools and serology supports the diagnosis. Surgical intervention along with prolonged benzimidazoles (most commonly albendazole) is curative in most of the cases.[5]

1.1. Case 1: Pleural hydatid disease in isolation

A 31 yrs old male, resident of Bihar, grocer by occupation presented with progressively increasing breathlessness, cough and significant weight loss for six months and fever for past three months. His X-ray chest done showed large left sided effusion with loculations. Pleural fluid analysis was non contributory towards diagnosis as it was neutrophilic exudates with normal ADA. Aerobic culture grew E. coli for which he was given appropriate antibiotics. His effusion persisted and surgical opinion taken. Serology for echinococcus was positive, and computed tomography (CT) scan thorax showed multiseptated effusion with a possibility of hydatid disease of pleural space. Video assisted thoracoscopic (VAT) decortication was done and many cystic structures were removed which further confirmed hydatid disease of the pleural space on histopathology. There was no simultaneous involvement of liver, lung or any other organ indicating primary haematogenous involvement of pleural space (Fig. 1).

1.2. Case 2: Hydatid lung cyst

A 41 yrs old, male, resident of Delhi, presented with two months history of right sided chest pain and fever for one month. Patient had no other chronic illness. He was started on antibiotics but when he didn't improve he was put on anti tubercular treatment. He continued to be symptomatic and a bronchoscopy was done. It showed a white cystic lesion in the posterior segment of right lower lobe. A clinical possibility of any benign lesion with a

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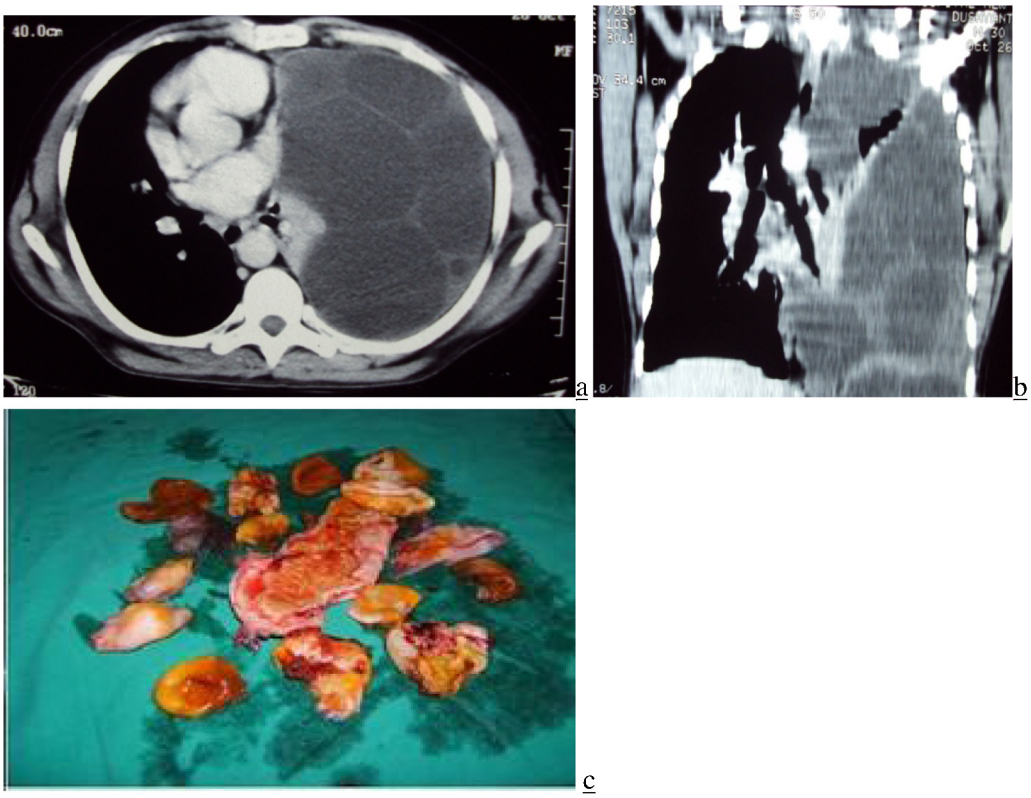

Fig. 1. Case 1: (a,b) CT scan of Chest showing Hydatid disease of pleural space and involvement of pleura; (c) Cystic structures post surgery.

possibility of hydatid disease was put forward. Biopsy was not done and right lower lobectomy was done and a final diagnosis of hydatid disease was made on histopathological examination of the respected segment (Fig. 2).

1.3. Case 3: Left lower lobe cavitary disease

Our third patient was a sixty year old female, housewife, presented with ten day history of fever, left sided chest pain and productive cough. X-ray chest showed a left lower zone cavity lesion with air fluid level which was confirmed on CT thorax. Sputum wet mount showed hooklets and hydatid serology was positive (Fig. 3).

2. Discussion

Hydatid disease is mainly a disease of liver but at times it poses a challenge to pulmonologists as well. Lungs and other thoracic structures being in close vicinity of the liver and receiving blood after it passes through liver venous system can act as second stop for eccinococcus larvae. Lungs are the second most common site involved in hydatid disease in adults and most common site of involvement in children.[6,7] Parasitic larvae enter circulation from duodenum and first filter is venous circulation of the liver and ones that are not filtered lodge in other organs.[6] Out of all patients with lung cysts, around 20–40% also have liver cysts but most of them have isolated lung involvement.[8] Pulmonary hydatid disease affects the right lung in ~60% of cases, and 30% of them have multiple pulmonary cysts, 20% cysts are bilateral and 60% are located in the lower lobes.[8] Pulmonary echinococcosis can follow intrathoracic rupture of a cyst of the liver or primary lodging of the larvae.[9] Within the chest, hydatid disease can also primarily involve the pleural cavity, mediastinum and chest wall.[10]

Our first case is unique as primary pleural involvement is not only uncommon but equally difficult to prove and cure. It needs surgical treatment for sure as is the case with most of the cases of ecinococcosis. In a study by Aribas et al, 1/3rd patients in ten year duration had pleuraopericardial involvement.[9] Pleural thickening followed by empyema as in our case were the two most common presentations.

All cases mimicked tuberculosis as they had similar clinical picture of fever and weight loss with other non specific symptoms as chest pain. All patients were on anti- tubercular drugs to begin with but definitive diagnosis was achieved when they did not improve. So it is a big tubercular mimic for Indian population. Our second case was usual with right sided lower lobe cystic lesion but was treated as tuberculosis and diagnosis was delayed by several weeks in view of lack of suspicion. Bronchoscopy helped in suspecting echinococcal disease. Our third case was again unusual as it involved only left lower lobe in isolation. Lesion mimicked a lung abscess and simple sputum examination established the diagnosis.

Pathogenetic explanations for hepatic omission appear possible: a passage of oncospheres through hepatic sinuses without causing disease, a passage via lymphatic vessels or via portocaval anastomoses and the vascular passage in a retrograde fashion explains lung and pleural involvement in isolation though extrahepatic manifestation of alveolar echinococcosis without apparent liver involvement is rare.[11] However, alveolar echinococcosis should be taken into account among other differentials. Pulmonologists should be aware of the thoracic cavity manifestation of the disease and keep it as a differential diagnoses even in cases of extra hepatic lesions without liver involvement.

All these presentations again emphasize the fact that hydatid disease can involve lungs in multiple and bizarre ways and surgical intervention is the most definitive way of treatment.[12] Cystic echinococcosis is the most common parasitic disease of the lung

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