Case Report

A Rare Case of Hydatoptysis

V. Arun Raja[1], G. Sindhu[1], B. Sreevidya[1], Rashmi Patnayak[2], Vishnu Priya Bukya[2], S. Settipalli[3], S. Kadiyala[3] and Alladi Mohan[1]

Division of Pulmonary, Critical Care and Sleep Medicine, Departments of Medicine[1], Pathology[2] and Radiodiagnosis[3], Sri Venkateswara Institute of Medical Sciences, Tirupati (Andhra Pradesh), India

Abstract

Hydatid disease, caused by *Echinococcus granulosus* usually affects liver. Pulmonary hydatidosis deserves special mention because isolated involvement of lungs is rare and patients with isolated pulmonary hydatidosis often remain asymptomatic. We report the case of a 24-year-old male who presented with vomiting, pain in the epigastric region, left upper abdomen and chest that was followed by high-grade fever of one day duration following a binge of beer drinking. Chest radiograph and computed tomography of chest revealed well defined cavitatory lesion with wall calcification and air fluid level in the lingular segment of left lung with left minimal pleural effusion. Sputum cytopathological examination revealed echinococcus hooklets. The patient was diagnosed to have ruptured hydatid cyst in lung. The present case highlights the importance of sputum cytopathological examination in confirming the diagnosis in patients with clinically suspected ruptured pulmonary hydatid cyst. This case also brings to light the uncommon occurrence of pulmonary aspergillosis along with hydatid disease of lung. This association in an immunocompetent individual has rarely been reported. **[Indian J Chest Dis Allied Sci 2018;60:83-85]**

Key words: Echinococcosis, Lung, Hydatoptysis.

Introduction

Human echinococcosis also called as hydatid disease is a cestode infestation caused by a parasite belonging to the genus *Echinococcus*. It is a major zoonotic disease with public health importance. This disease is endemic in many parts of the World, namely, Central Asia, Australia, South America and Eastern Africa.[1] Among the four recognised species, *Echinococcus granulosus* is the most common causative agent in humans, and causes cystic echinococcosis. Professions which make a person work in close contact with animals are highly susceptible to develop cystic echinococcosis. Commonly, cystic echinococcosis affects the liver and less commonly the lung. Isolated involvement of lung is unusual and is seen in only 10% to 30% of the cases.[2] Most of the cases remain undiagnosed as patients are asymptomatic. Here we report a case of pulmonary hydatidosis in which the complication of the disease *per se* led us to the diagnosis.

Case Report

A 24-year-old male, resident of Tirupati, driver by occupation presented to the emergency room with chief complaints of pain in the epigastric region, left upper abdomen and chest of one day duration. A day before admission, he drank about 360mL of beer at mid-noon, an hour later he had an episode of vomiting – non bilious, and non-blood stained after which he developed severe sharp, stabbing pain in the epigastric region, left upper abdomen and left side of the chest that worsened with deep breaths and was not relieved on bending forward. He rated the pain as 6 on a scale of 0 to 10. He went to a local clinic, where, he was prescribed oral pantoprazole 40mg daily and oral paracetamol 650mg thrice-daily which did not result in any symptomatic improvement. The next day, he had three episodes of vomiting and reported to Surgical Gastroenterology out-patient service of our hospital and was advised to undergo abdominal ultrasonography. On the same day, he visited the emergency room at 11:30 p.m. with high grade sudden onset of fever without chills or rigors. He also experienced an increase in intensity of the chest pain along with productive cough which was mucopurulent, moderate in quantity without haemoptysis. There was no other significant history suggestive of any other organ system involvement. He had been previously healthy with no co-morbid conditions and took no medications or had no known allergies. He had no exposure to animals or contact with sick persons. There was no history of recent travel. He was a non-smoker. He drank about 200 – 360 mL of beer for about five days a week (CAGE score = 2)[3] and did not use any illicit drugs. There was no significant family history.

[Received: June 28, 2017; accepted after revision: April 17, 2018]

Correspondence and reprint requests: Dr Alladi Mohan, Chief, Division of Pulmonary, Critical Care Medicine and Professor and Head, Department of Medicine, Sri Venkateswara Institute of Medical Sciences, Tirupati-517 507 (Andhra Pradesh), India; E-mail: alladimohan@rediffmail.com

On general physical examination he was febrile (temperature of 101 °F), pulse rate 120/min respirations 34 breaths/min. Blood pressure was 120/70 mmHg and oxygen saturation (measured by pulse oximetry) while he was breathing room air was 89%. Rest of the physical examination was normal. Respiratory system examination was suggestive of left-sided pneumonitis. Rest of the systemic examination was unremarkable. Acute pancreatitis was ruled out by computed tomography (CT) of the abdomen and serum amylase levels (45 IU/L) which were within normal limits. Electrocardiogram (ECG) showed normal sinus rhythm with no significant ST-T changes or regional wall motion abnormalities. The left ventricular ejection fraction was 61% on two-dimensional echocardiography. Neutrophilic leucocytosis was evident (neutrophils 83%). Chest radiograph revealed an air-fluid level in the left lower zone with obliteration of left cardiophrenic angle with obscuration of the left dome of the diaphragm and left heart border (Figure 1). A diagnosis of left lower lobe lung abscess/necrotising pneumonia was made and the patient was initiated on intravenous piperacillin-tazobactam (4.5g thrice-daily). Non-contrast CT (NCCT) of the chest showed a well-defined cavitatory lesion with wall calcification and air-fluid level in the lingular segment of the left lung with minimal left-sided pleural effusion (Figure 2A). The differential diagnosis at this point in time included (i) lung abscess; (ii) resolving pneumonia with cavity; (iii) infected bronchogenic cyst; (iv) hydatid cyst; (v) loculated empyema; (vi) cystic bronchogenic carcinoma, among others. Gram staining of the sputum showed plenty of pus cells. Pyogenic culture of the sputum showed no growth, and sputum smear for acid-fast bacilli was negative (twice). Sputum cytology

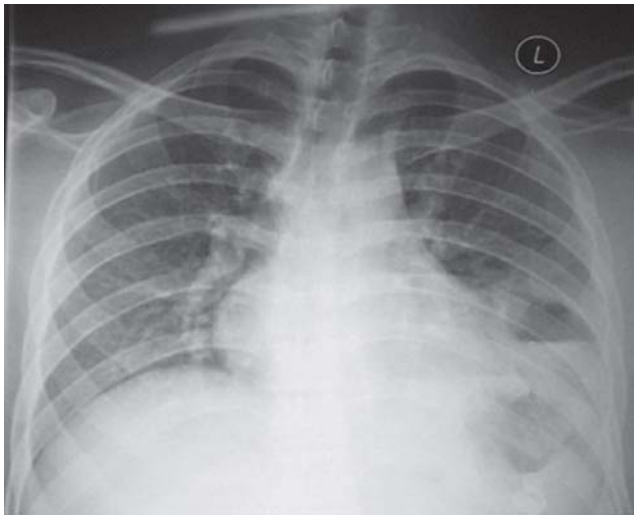

Figure 1. Chest radiograph (postero-anterior view) showing homogeneous opacity in the left mid- and lower-zones with an air-fluid level. Silhouetting of left cardiac border, obliteration of left dome of the diaphragm and left costophrenic angle are also seen.

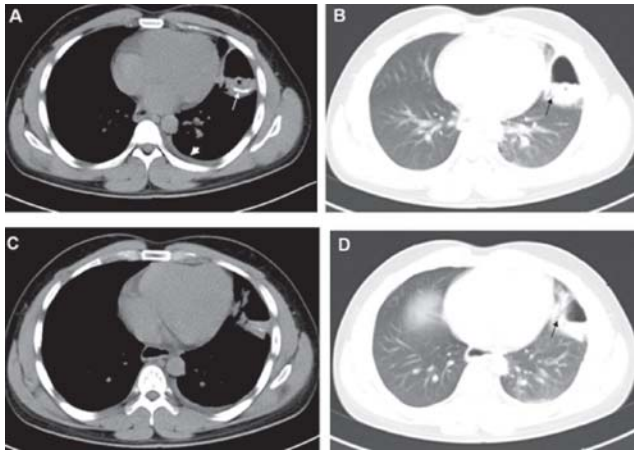

Figure 2. Non-contrast computed tomography of chest (A, C) (medisastinal window), showing a thin-walled cystic lesion with air-fluid level in the lingular segment of the left upper lobe. Left-sided pleural effusion (arrow head) and focal calcification in the posterior wall (white arrow) are also seen and (B, D) (lung window) showing consolidation in the adjacent lung parenchyma (black arrow).

revealed multiple hooklets and scolices suggestive of hydatid cyst. Another incidental finding was thin slender hyphal elements of *Aspergillus* on sputum cytopathological as well as potassium hydroxide (KOH) mount examination (Figure 3).

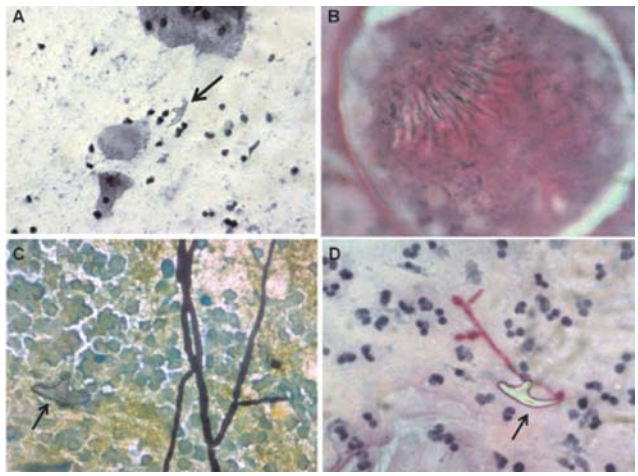

Figure 3. Photomicrograph of sputum smear showing (A) benign squamous cells, hooklet (black arrow) in a background of inflammatory cells (Papanicolou × 200); (B) scolex of *Echinococcus* with row of hooklets (Periodic acid Schiff × 400); (C) hooklet of *Echinococcus* (black arrow), slender, branched, septate fungal hyphae of *Aspergillus* (Gomori methanamine silver × 200); and (D) hooklet of *Echinococcus* (black arrow), *Aspergillus hyphae* (Periodic acid Schiff × 200).

The patient was diagnosed to have spontaneous communicating ruptured hydatid cyst of upper lobe of the left lung with fungal colonisation with *Aspergillus*. The patient was started on oral albendazole 10mg/kg/day in two divided doses for 21 days. Patient gradually improved symptomatically and was

discharged in a clinically stable condition 10 days after admission and was instructed to return for follow-up after two weeks. However, the patient did not return to follow-up.

Discussion

Humans are accidental and dead-end hosts for the cestode *Echinococcus* and get infected due to faeco-oral contamination. Several mechanisms have been postulated to explain how the pathogen reaches the lungs. Humans acquire infection following ingestion of *Echinococcus* eggs. Following gastric, enteric digestion of the eggs, hexacanth embryos are released. These attach to and subsequently penetrate the intestinal wall and reach the liver through portal circulation. Embryos <0.3mm pass through the hepatic sinusoids, traverse through the hepatic vein, inferior vanacava, right heart and reach the lungs. Sometimes, the embryos may pass through the intestinal lymphatics, bypassing the liver, enter the thoracic duct and reach the lungs. Rarely, pulmonary involvement can occur following inhalation of air containing *Echinococcus* eggs. Embryos can also traverse the portosystemic anastamosis in the space of Retzius and reach the lung. Very rarely trans-diaphragmatic dissemination by the formation of biliary-bronchus fistula may also occur. Most patients remain asymptomatic, some may develop cough due to compression of the surrounding structures by the expanding cyst, few develop chest pain and fever due to cyst rupture, others can present with haemoptysis as the cyst erodes the bronchus and a vessel and very few expectorate the contents of the cyst due to communicating cyst rupture which occurs as the endocyst rips apart and a rent in the pericyst communicates with the bronchus which causes expulsion of the cyst contents.[4] The expectoration of hooklets in sputum is termed *hydatoptysi*s. Another most important complication of cyst rupture is anaphylaxis that occurs due to the spread of antigenic hydatid fluid in the tissues.

In our patient, thin slender hyphal elements of *Aspergillus* were evident on sputum cytopathological as well as KOH mount examination. We considered this to be suggestive of *Aspergillus* colonisation. Available evidence suggests that isolation of *Aspergillus* from a single sputum specimen does not confirm it as the aetiological pathogen. Repeated isolation of identical *Aspergillus* species and demonstration of anti-*Aspergillus* antibodies and/or *Aspergillus* antigens in the sera are required to establish the aetiological role of the isolate.[5] A clinical case-definition scoring system has been proposed for interpreting the significance of *Aspergillus* spp isolated in culture from the lower respiratory tract secretions; a score of ≤0.3 being suggestive of colonisation.[6] Further, *Aspergillus* colonisation is frequently reported to occur in the diseased cavities of tuberculosis, sarcoidosis and malignancy; but *Aspergillus* colonisation of hydatid cyst is uncommon. In a large retrospective analysis of 100 consecutive cases of hydatid cysts, colonisation by *Aspergillus* sp. was seen in two cases both of whom were immunocompetent.[7] The present case documents such a rare occurrence. Deterioration of the local defence mechanisms is perhaps the most appropriate explanation for *Aspergillus* colonisation.

In conclusion, diagnosis of pulmonary hydatid disease rests on clinical, radiological, and serological examinations. Our experience with this case suggests that sputum examination for hydatoptysis must also be included in the routine work-up of any adult presenting with chest pain and a cyst in the lung. The present case also highlights the point that hydatid disease of the lung presenting with chest pain is atypical and should be considered in the differential diagnosis of chest pain in young individuals, without conventional risk factors of atherosclerosis.

References

1. Wahlers K, Menezes CN, Wong ML, Zeyhle E, Ahmed ME, Ocaido M, *et al*. Cystic echinococcosis in sub-Saharan Africa. *Lancet Infect Dis* 2012;12:871–80.
2. Mandal S, Mandal MD. Human cystic echinococcosis: epidemiologic, zoonotic, clinical, diagnostic and therapeutic aspects. *Asian Pac J Trop Med* 2012;5:253–60.
3. Ewing JA. Detecting alcoholism. The CAGE questionnaire. *JAMA* 1984;252:1905–7.
4. Pedrosa I, Saíz A, Arrazola J, Ferreirós J, Pedrosa CS. Hydatid disease: radiologic and pathologic features and complications. *Radiographics* 2000;20:795–817.
5. Tashiro T, Izumikawa K, Tashiro M, Takazono T, Morinaga Y, Yamamoto K, *et al*. Diagnostic significance of *Aspergillus* species isolated from respiratory samples in an adult pneumology ward. *Med Mycol* 2011;49:581–7.
6. Greub G, Bille J. Aspergillus species isolated from clinical specimens: suggested clinical and microbiological criteria to determine significance. *Clin Microbiol Infect* 1998;4:710–6.
7. Bal A, Bagai M, Mohan H, Dalal U. Aspergilloma in a pulmonary hydatid cyst: a case report. *Mycoses* 2008;5:357–9.