

## Case Report

# “Necklace in the lung:” Multilocularis hydatid cyst mimicking left-sided massive pleural effusion

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### ABSTRACT

Massive pleural effusions are commonly caused by malignancy, parapneumonic effusion, and tuberculosis. Parasitic infections are rare causes of massive pleural effusion. Echinococcosis or hydatid disease is a major public health problem in poor hygienic environments. The liver and lungs are the most frequently involved organs. Pulmonary disease appears to be more common in younger individuals. *Echinococcus multilocularis* causes alveolar echinococcosis, which accounts for <5% of all cases of hydatid liver disease and less frequently lung disease. Here, we present an unusual case of multilocular pulmonary hydatid cysts mimicking massive pleural effusion in a 25-year-old young male.

**KEY WORDS:** Computed tomography thorax, *Echinococcus multilocularis*, hydatid cyst, pleural effusion

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### INTRODUCTION

Pleural effusions which involve the entire hemithorax or more than two-thirds of hemithorax are designated as massive effusions. Massive pleural effusion is commonly caused by malignancy and has an incidence of 11.2%–12% of all the pleural effusions.<sup>[1]</sup> The etiology and occurrence of massive pleural fluid collection have remained largely undetermined and therefore not known. Hydatid cyst mimicking as massive effusion is very uncommon. Hydatid disease in humans occurs by infection with the larval stage of the tapeworm *Echinococcus granulosus* (the dog tapeworm) or *Echinococcus multilocularis* (the fox tapeworm). Humans acquire the infection by ingesting the eggs due to poor hygiene practices or contaminated food and water. Lungs are usually less common site of disease which occurs most frequently in the liver. Pulmonary hydatid cysts are more commonly seen in children but can also be seen in adults.<sup>[2]</sup> Single cysts are more common (70%–80%), with unilateral multiple cysts being found in 10%–15% of cases and bilateral cysts in

6%–13%.<sup>[3]</sup> Here, we report an unusual case of pulmonary hydatid cyst masquerading as massive pleural effusion in a young male.

### CASE REPORT

A 25-year-old unmarried male laborer presented with fever, dry cough, exertion dyspnea, and left-sided chest pain for the last 6 months. The fever was of low grade and relieved by antipyretics. The cough was gradually progressive and lacked any associated expectoration. The left-sided chest pain was localized and dull-aching and responded favorably to analgesics. His medical history was unremarkable for major illness or surgery as was his personal history negative for any addiction. His general examination was within normal limits. Chest inspection revealed right-sided tracheal shifting while palpation additionally demonstrated shifting of the apex beat to the right side. A stony dull note was appreciated on percussion

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**How to cite this article:** Bajpai J, Jain A, Kar A, Kant S, Bajaj DK. “Necklace in the lung:” Multilocularis hydatid cyst mimicking left-sided massive pleural effusion. Lung India 2019;36:550-2.

Access this article online	
<b>Quick Response Code:</b> 	<b>Website:</b> www.lungindia.com
	<b>DOI:</b> 10.4103/lungindia.lungindia_76_19

over the left hemithorax along with decreased air entry. As such, a Stony dull note is indicative of pleural effusion.

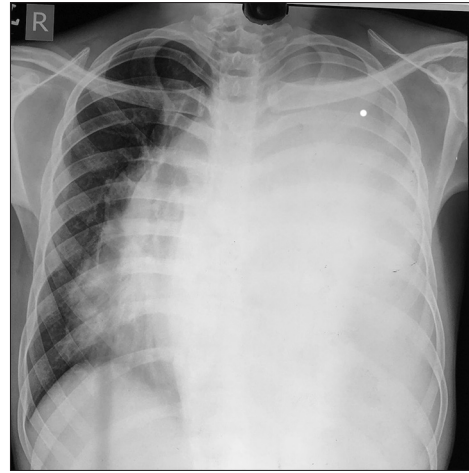
Routine blood investigations were within normal limits, and the chest X-ray revealed a homogeneous opacity left hemithorax with contralateral shifting of the mediastinum [Figure 1]. Chest X-ray posteroanterior view and examination findings were suggestive of massive pleural effusion. Differential diagnoses of massive pleural effusion are bronchogenic carcinoma, metastatic malignancy, mesothelioma, tuberculosis, pulmonary embolism, etc.<sup>[4]</sup> Thoracocentesis was performed under aseptic precaution, which revealed a purulent aspirate. A provisional diagnosis of left-sided empyema was made, and empirical antibiotic therapy was initiated. Presence of dyspnea, mediastinal shifting to opposite hemithorax, and aspiration of pus all mandated intercostal drainage (ICD) was aptly performed. The microscopic examination of pus (Gram stain and AFB) was negative, while cultures also were sterile for any organisms. Neither was any abnormal parasitic cyst reported.

The patient's dyspnea responded well and the output from the ICD became minimal without considerable change in the appearance of the chest X-ray. A computed tomography (CT) thorax was done which showed a single, large rounded cyst in the left lung surrounded by multiple small, rounded cysts that appeared like a necklace [Figure 2]. ICD was clamped and taken out. An ultrasound-guided thoracocentesis revealed a clear aspirate which on microscopic examination revealed hooklets of *Echinococcus* [Figure 3]. A diagnosis of left-sided multiple hydatid cysts with superadded infection was entertained. The diagnosis was further confirmed by the demonstration of raised serum IgG for *Echinococcus*. The patient was started on albendazole 15 mg/kg/day twice daily and referred to a cardiothoracic surgeon for removal of the cyst.

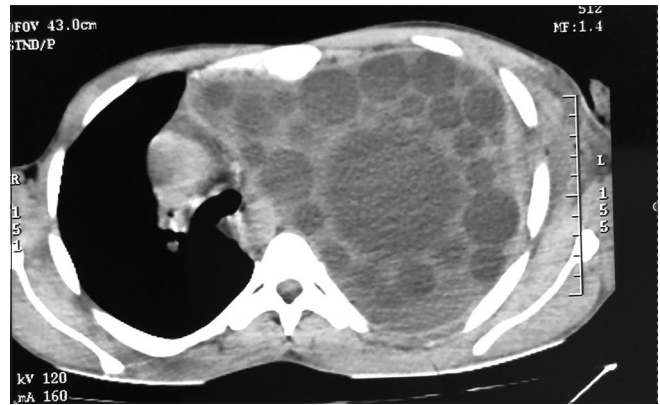
## DISCUSSION

Pleural effusions are a common medical problem with >50 recognized causes, including disease local to the pleura or underlying lung, systemic conditions, organ dysfunction, and drugs. Effusions that involve the entire hemithorax or at least two-thirds of it are designated as massive. Massive pleural fluid collection can be caused by a wide range of clinical conditions that can be classified into malignant and nonmalignant causes.

Assessment of mediastinum in massive effusion cases is very important. In the Spanish study by Porcel and Vives, the three most frequent etiologies in patients with massive effusion were malignancy (59%), parapneumonic effusion (23%), and tuberculosis (10%).<sup>[5]</sup> Pleural effusion due to parasitic infection is hence uncommon.



**Figure 1:** Chest X-ray revealed a homogenous opacity in left hemithorax with contralateral shifting of the mediastinum



**Figure 2:** Computed tomography thorax mediastinum window showed a single, large rounded cyst in left lung surrounded by multiple small, rounded cysts appeared like a necklace



**Figure 3:** Clear aspirate on microscopic examination revealed hooklets of *Echinococcus* (black arrow) in hydatid sand

Human echinococcosis, also known as hydatid disease, is a zoonotic disease caused by the larval stage (metacestode) of the parasite. *Echinococcus* is most commonly reported in

pastoral communities of South America, Australia, and the Mediterranean.<sup>[6]</sup> Of the four species recognized to cause hydatid disease, *E. granulosus* is the most common species to cause the human disease. Although *E. multilocularis* is rare, it is the most virulent species and causes alveolar echinococcosis.<sup>[7]</sup> Hydatid disease involves the lung via various mechanisms. Human receives the infection by ingesting the eggs mixed with uncooked vegetables, fruits, and drinking water or by handling the soil and dirt. Pleural involvement with hydatid disease can occur via rupture of hepatic, splenic, and pulmonary cyst into the pleural space, or on rare occasions, the pleura may be primarily involved by the slowly enlarging cyst. Pleural empyema may be as a result of complication of enlarged hydatid cyst.

The investigation of choice in pulmonary hydatid cysts is CT or magnetic resonance imaging of the chest. CT typically shows an uncomplicated cyst with a high-attenuation wall and a low-density content. Hydatid-specific IgG is the most sensitive and specific serological test. The diagnosis of pleural echinococcosis is established by the demonstration of echinococcal scolices with hooklets in the pleural fluid. The treatment of choice in the pulmonary hydatid cyst is surgical resection. Although the Percutaneous Aspiration, Injection of cysticidal agent, and Re-aspiration using radiographic guidance (PAIR) method is being routinely used in cases of hydatid liver disease, the WHO currently recommends that PAIR should not be used in case of pulmonary cysts.<sup>[8]</sup> Surgery should be coupled with albendazole (10–15 mg/kg/day) administration in two doses from 4 days before to at least 1 month after surgery. Antihelminthic therapy before surgery has shown to reduce the risk of recurrence by 3.5 times.<sup>[9]</sup>

## CONCLUSION

Pulmonary echinococcosis may rarely present as massive pleural effusion, and this diagnosis can be kept on differential diagnosis in endemic areas.

Chest CT is a useful adjunctive diagnostic maneuver and helps in pinning down the underlying etiology.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

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