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PRIMARY PLEURAL HYDATIDOSIS REVEALED BY ACUTE RESPIRATORY DISTRESS: A RARE EXTRAPULMONARY MANIFESTATION OF ECHINOCOCCOSIS

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AUTHOR AND AFFILIATION

Joud Boutaleb[1], Kaoutar Imrani[1], Sarah loubaris[1], Hafsa Riache[1], Abdelmajid Elkabbaj[2], Outhmane Maalbi[2], Itimad Nassar[1].

[1] Central Radiology Department, Ibn Sina University Hospital, Mohammed V University, Rabat, Morocco.
[2] Reanimation department Mohammed V University, Rabat, Morocco .

Corresponding author: Joud Boutaleb

ABSTRACT

Pleural hydatidosis is a rare manifestation of echinococcosis, most often secondary to the rupture of a hepatic or pulmonary cyst into the pleural cavity. It provokes a significant inflammatory response, frequently leading to acute respiratory compromise. The main pathophysiological mechanisms include pleural effusion, floating hydatid membranes, and compressive cystic expansion. Diagnosis relies heavily on imaging, particularly chest CT, which can delineate the extent of both pleural and hepatic involvement.

We report a representative case of compressive pleural hydatidosis with concomitant hepatic localization, revealed by acute respiratory distress and managed through antiparasitic therapy, non-invasive ventilation, and surgical intervention. This case underscores the importance of rapid recognition and a multidisciplinary approach to avoid severe complications in advanced hydatid disease.

KEYWORDS

Hydatid disease ; Pleural hydatidosis , Extrapulmonary echinococcosis , Thoracic CT imaging

MAIN ARTICLE

Introduction

Hydatid disease (HD) is a cosmopolitan zoonosis, highly prevalent in Mediterranean countries [1]. It results from the tissue development of the larval form (hydatid) of the tapeworm Echinococcus granulosus, whose adult stage parasitizes the small intestine of canines. Humans, as accidental intermediate hosts—along side herbivores—acquire the infection through ingestion of food contaminated with parasite eggs [2].

Pleural hydatidosis is an extremely rare condition. In most reported cases, it is secondary to ruptured or protruding peripheral pulmonary hydatid cysts into the pleural cavity. Primary pleural hydatid cysts, however, are classified as extrapulmonary intrathoracic forms, potentially resulting from lymphatic or hematogenous dissemination [3]

Patient and observation

We report the case of a 37-year-old male patient with no significant past medical history, residing in a rural area with regular exposure to dogs and herbivorous livestock. He was admitted to the emergency department for acute respiratory distress.

On initial clinical evaluation, the patient was conscious but agitated, with a Glasgow Coma Score of 15 and no focal neurological deficits. Respiratory assessment revealed a respiratory rate of 30 breaths per minute, oxygen saturation of 88% on room air, which improved to 95% under high-flow oxygen via non-rebreather mask. The patient exhibited signs of respiratory distress, including intercostal retractions. Pulmonary auscultation revealed diminished breath sounds at the right lung base. Cardiovascular examination showed a heart rate of 110 bpm, blood pressure of 130/70 mmHg, no signs of peripheral hypoperfusion, and sinus tachycardia on ECG. The patient was afebrile, with supple thighs and calves, and no signs suggestive of deep vein thrombosis.

Initial management included semi-upright positioning, high-flow oxygen therapy at 15 L/min via a non-rebreather mask, placement of a peripheral intravenous line, and initiation of laboratory investigations (CBC, PT/aPTT, CRP, procalcitonin, serum electrolytes). Chest radiography was inconclusive, and a thoracic computed tomography (CT) scan was promptly performed. Given the clinical suspicion of hydatid disease, hydatid serology was also systematically performed.

Thoracic computed tomography (CT) revealed a large, loculated hydro-aeric pleural effusion measuring approximately 77 × 190 × 87 mm, associated with a band of passive atelectasis causing left lung collapse (Figure 1). Within the pleural collection, multiple daughter cystic vesicles were identified (Figure 2), (Figure 3) consistent with pleural hydatid material.

An adjunctive ultrasound examination confirmed the presence of daughter cysts within the pleural cavity, supporting the diagnosis of pleural hydatidosis (Figure 4).

Abdominal imaging slices demonstrated a large, simple cyst located in liver segments VIII and VII, consistent with a hydatid cyst classified as Gharbi type I (Figure 5)

The patient was admitted to the intensive care unit for further stabilization. Non-invasive ventilation (NIV) was initiated in multiple sessions to improve oxygenation. Medical therapy with albendazole was started at a dose of 15 mg/kg/day. Once stabilized, the patient was transferred to the thoracic surgery department for planned surgical excision of the cyst.

Discussion

After the liver, the thorax is the second most common site affected by hydatid disease. Within the thoracic cavity, the intrapulmonary form is predominant; however, several atypical intrathoracic localizations have been described, including pleural, mediastinal, pericardial, and diaphragmatic forms. These extrapulmonary locations often present significant diagnostic and therapeutic challenges, particularly due to their rarity and the nonspecific nature of clinical manifestations. (4)

Pleural hydatid cysts, whether primary or secondary, are rare. The primary form—without associated lung involvement—is exceedingly uncommon and is presumed to result from hematogenous or lymphatic dissemination of E. granulosus larvae to the pleural space. The pleura, especially the parietal layer, is thought to be relatively resistant to cyst implantation, making such cases exceptional. Conversely, secondary pleural involvement is more common and typically arises from rupture of a subpleural pulmonary cyst into the pleural space, which may lead to hydropneumothorax, empyema, or pleural dissemination. (5)

Pathophysiologically, the development of a cyst in the pleura can be facilitated by the permeable nature of the pleural layers, which allow diffusion of water, electrolytes, and nutrients that support cyst growth. The pleural environment, although not as vascularized as parenchymal tissues, can sustain slow cystic expansion over time, often remaining clinically silent until significant pleural reaction or compression occurs. (6)

Clinically, primary pleural hydatid disease may mimic more common causes of pleural effusion. Symptoms are nonspecific and include progressive dyspnea, dull chest pain, cough, and occasionally constitutional symptoms such as fatigue or low-grade fever. In large effusions or cysts exerting mass effect, mediastinal shift or lung compression may occur. However, up to 15% of patients may be entirely asymptomatic, with diagnosis made incidentally on imaging. (7)

Radiological imaging plays a central role in the diagnosis and management of pleural hydatid disease, especially given its rarity and frequent diagnostic ambiguity. Chest radiography is often the initial investigation, typically revealing a homogeneous opacity, air-fluid level, or mediastinal shift in large cysts or associated effusions. (8) However, computed tomography (CT) remains the modality of choice, offering precise localization, characterization, and surgical planning. CT typically demonstrates a well-defined,

water-attenuation cystic lesion within the pleural cavity or along fissures, occasionally containing daughter cysts arranged in a rosette or wheel-spoke pattern. Ruptured cysts may show floating membranes (the classic "water-lily sign"), air-fluid levels, or adjacent pleural thickening. MRI, while less commonly employed, provides superior soft tissue contrast and is especially useful in assessing complex mediastinal or diaphragmatic involvement; cysts appear hypointense on T1 and hyperintense on T2-weighted images, with possible rim enhancement if inflammation is present. (7) Ultrasound may assist in detecting diaphragmatic transgression or guiding interventions, although percutaneous aspiration is generally contraindicated due to the risk of spillage and anaphylaxis. Radiological differential diagnoses include empyema, pleural neoplasms (mesothelioma or metastases), tuberculous pleuritis, lymphangiomas, or post-traumatic collections. Clues favoring hydatid disease include the presence of daughter cysts, non-enhancing cystic content, floating membranes, and epidemiological context. A thorough radiological assessment is thus essential to avoid misdiagnosis, guide surgical approach, and prevent potentially severe complications. (8)

Serologic testing, though not pathognomonic, remains supportive in diagnosis. A combination of tests—indirect hemagglutination, ELISA, and Western blot—enhances diagnostic yield. However, false negatives may occur, particularly in cases of isolated extrapulmonary disease, necessitating reliance on imaging and surgical findings for definitive diagnosis. (9)

From a therapeutic standpoint, surgery remains the cornerstone of management. The objective is complete excision of the cyst while preventing intraoperative spillage and secondary dissemination. In cases of primary pleural cysts, careful dissection is needed to avoid injury to adjacent structures, including the lung, diaphragm, and mediastinum. Postoperative medical therapy involves albendazole at a dose of 10–15 mg/kg/day for one month. For non-operable patients, the same dosage is administered for 4-week cycles, repeated at least three times with 2-week intervals between each cycle [10].

Conclusion

Primary pleural hydatidosis is an exceptionally rare manifestation of echinococcosis that poses diagnostic and therapeutic challenges. Prompt imaging, especially CT, combined with serology and clinical suspicion, is essential for diagnosis. Surgical excision remains the mainstay of treatment, complemented by antiparasitic therapy to prevent recurrence.

FIGURES :

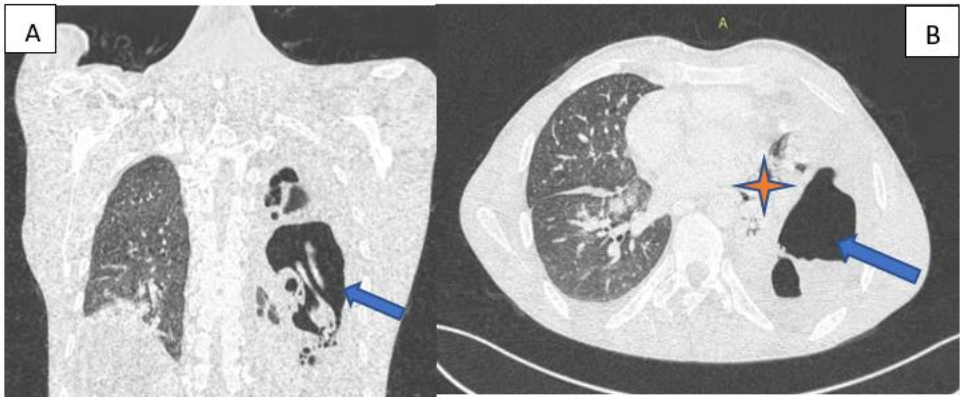

Figure 1 : Coronal (A) and Axial (B) chest CT image showing a large loculated hydro-aeric pleural effusion (measuring 77 × 190 × 87 mm) occupying the left pleural cavity (blue arrow), associated with a band of passive atelectasis causing near-complete collapse of the adjacent left lung (orange star).

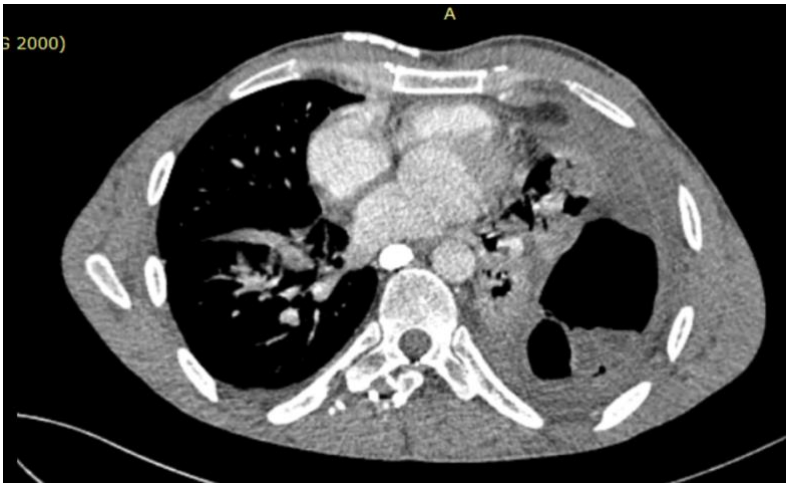

Figure 2 : axial (CT) mediastinal window showing a large loculated hydro-aeric pleural effusion occupying the left pleural cavity.

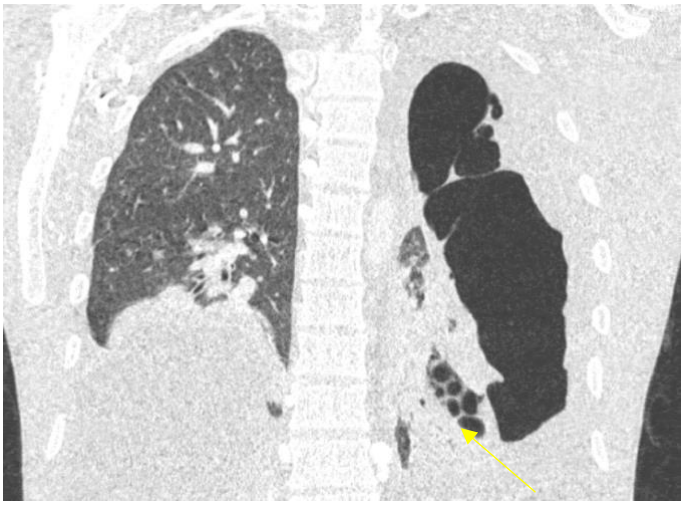

Figure 3: Coronal CT slice demonstrating multiple daughter cystic vesicles within the pleural fluid collection (yellow arrow)

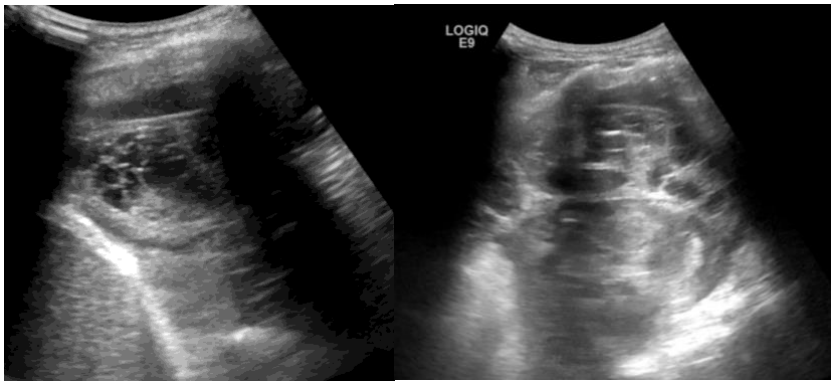

Figure 4: *Thoracic ultrasound image confirming the presence of multiple daughter cysts within the pleural space, supporting the diagnosis of pleural hydatid disease .*

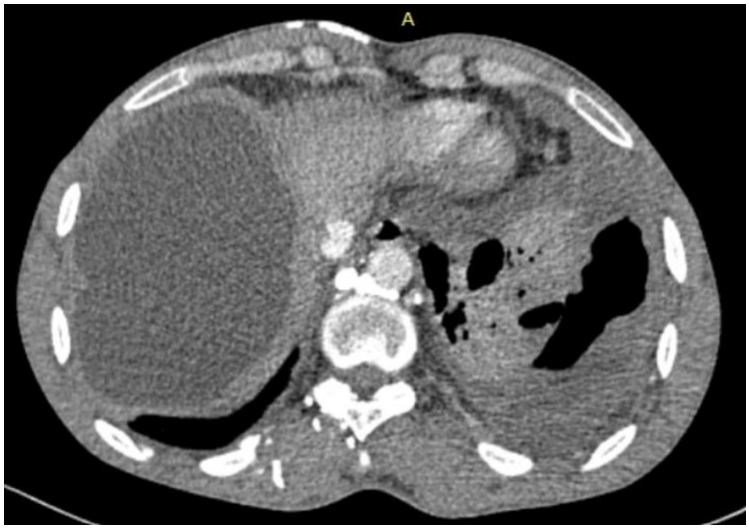

Figure 5 : Abdominal CT image showing a large, simple cystic lesion located in hepatic segments VII and VIII, classified as Gharbi type I hydatid cyst, consistent with a unilocular fluid-filled cyst without internal septations or daughter cysts.

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