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Case Report

# Imaging of primitive pleural hydatidosis in children: A case report ☆

*Salma Marrakchi, MD[a,*], Ouiam Taibi, MD[a], Sara Ez-zaky, MD[a], Moustaine El Mamoune, MD[b], Bouanane Rania, MD[a], Nazik Allali, PhD[a], Latifa Chat, PhD[a], Siham El Haddad, PhD[a]*

[a] *Paediatric Radiology Department, Ibn Sina University Hospital Center, Rabat, Morocco*
[b] *Central Laboratory of Parasitology and Mycology, Ibn Sina University Hospital Center, Rabat, Morocco*

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ABSTRACT

Hydatidosis, caused by the larval form of the parasite *Echinococcus granulosus*, is a rare condition, especially in pediatric patients, with pleural involvement being exceedingly uncommon. We report a case of primary pleural hydatidosis in a 9-year-old child, emphasizing the importance of various imaging techniques in establishing an accurate diagnosis.



## Introduction

Hydatidosis is a parasitic infection caused by the larval form of *Echinococcus granulosus*, typically found in dogs and humans. Although the disease is endemic in many regions, pleural involvement is extremely rare, particularly in pediatric populations. In this report, we present a unique case of primary pleural hydatidosis in a pediatric patient, highlighting the importance of various imaging modalities in achieving an accurate diagnosis.

## Case report

We report the case of a 9-year-old boy with no significant medical history who had early childhood exposure to dogs. He presented with persistent left thoracic pain and chest tightness over the past 3 months, without dyspnea or other respiratory symptoms. The symptoms occurred in the context of apyrexia, with overall preservation of his general condition. Clinical examination revealed signs consistent with left pleural effusion.

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* Corresponding author.
E-mail address: Marrakchi.salma@gmail.com (S. Marrakchi).

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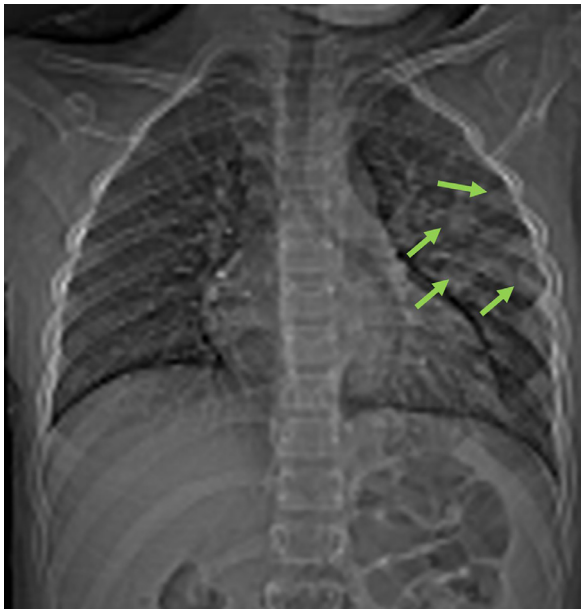

**Fig. 1 – Frontal chest X-ray demonstrating multiple homogeneous rounded opacities in the left hemithorax (arrows).**

The frontal chest X-ray revealed multiple homogeneous rounded opacities in the left hemithorax (Fig. 1). Subsequent thoracic CT scans demonstrated multiple cystic formations within the left pleura, characterized by oval and round shapes, thin walls, and lack of enhancement after contrast injection. A subtle mass effect on the adjacent lung parenchyma was exerted by these formations. Apart from passive atelectasis adjacent to the cysts, the lung parenchyma showed no abnormalities.

Ideally, a pleural ultrasound and thoracic MRI should have been performed to further characterize these formations. However, the ultrasound images were not retrievable, and the MRI was not conducted due to issues with availability and cost. Therefore, we relied on the clinical context and the CT scan findings to suggest the diagnosis of pleural hydatid cysts classified as Type I (Fig. 2).

The blood work results showed hypereosinophilia (10%). The diagnosis was further supported by positive ELISA Eg and indirect agglutination serologies. Additional investigations, including brain CT, echocardiography, and abdominal ultrasonography, revealed no abnormalities, confirming the primary nature of pleural hydatidosis. Surgery revealed multiple cystic formations in the left parietal pleura. The cystectomy was performed without complications, and the specimens were sent to the parasitology laboratory. Macroscopic examination showed no proliferative membrane, while microscopic investigation of the intracystic fluid identified viable scolexes (Fig. 3).

## Discussion

Hydatidosis is a helminthic zoonotic disease that remains endemic in several countries, particularly in the Mediterranean region. It is caused by the larval form of *E granulosus* developing in the body [1]. Humans are accidental hosts [2]. Transmission to humans usually occurs through the ingestion of parasite eggs present in contaminated food or water, or through direct contact with the definitive host [1].

Although hydatid cysts can potentially develop anywhere in the body, the liver is the most commonly affected organ, accounting for approximately 75% of cases, followed by the lungs, which are involved in 20% to 40% of cases [3,4]. Pleural hydatidosis is extremely rare, representing only 1.3% of thoracic cases [1]. Primary pleural hydatidosis, constituting less than 1% of all hydatidosis cases, is classified under extrapulmonary intrathoracic cysts. These cysts may also be found in locations such as the parietal pleura, mediastinum, pericardium, diaphragm, fissures, and chest wall, typically spreading via lymphatic or hematogenous routes [3,4].

In children, echinococcosis predominantly affects young males [5], leading to significant morbidity and mortality in endemic regions [6]. Unlike in adults, the lungs are the primary site of involvement in the pediatric population [7]. However, there has been limited research into pediatric hydatid cysts.

The main cause of pleural hydatidosis typically arises from the rupture of a liver or lung cyst [8,9]. Initially, primary pleural hydatidosis was disputed by Dévé, who claimed it did not exist [10]. Nevertheless, subsequent case reports have documented its occurrence [11]. In our case, no other lesions indicative of hydatidosis were found, leading us to the diagnosis of primary pleural hydatidosis.

Pleural hydatid cysts can remain asymptomatic for a long time or present with nonspecific clinical symptoms, similar to other pleural-pulmonary diseases, such as chest pain, dyspnea, and dry cough. In rare instances, signs of mediastinal compression may appear. Establishing a definitive diagnosis is challenging due to the absence of specific clinical, radiological, and biological markers [1,11].

Imaging plays a crucial role in establishing a definitive diagnosis. In primary pleural hydatidosis, chest X-rays typically show a homogeneous pleural opacity with well-defined, water-like characteristics. Occasionally, cystic formations with peripheral calcifications, although rare, may guide the diagnosis. However, in our patient, the chest X-ray findings were nonspecific.

Thoracic ultrasound has an estimated specificity of 96% for diagnosing pleural hydatidosis and is also valuable for assessing disease extent and monitoring [14]. It typically reveals rounded or oval anechoic multiloculated formations or hydroaerial images in cases where the hydatid cyst has ruptured. It often demonstrates the characteristic double-layered membrane within the pericyst, a pathognomonic sign of hydatid cysts.

Computed tomography (CT) plays a key role in confirming pleural involvement, typically revealing a well-defined fluid mass within the pleural space without enhancement after contrast injection. CT is more sensitive and specific than earlier imaging modalities in diagnosing pleural hydatidosis [11], as observed in our patient.

Magnetic resonance imaging (MRI) can be beneficial when cysts do not display characteristic features on ultrasound or CT. It provides a clearer anatomical localization of the cyst and its relationship with adjacent organs.

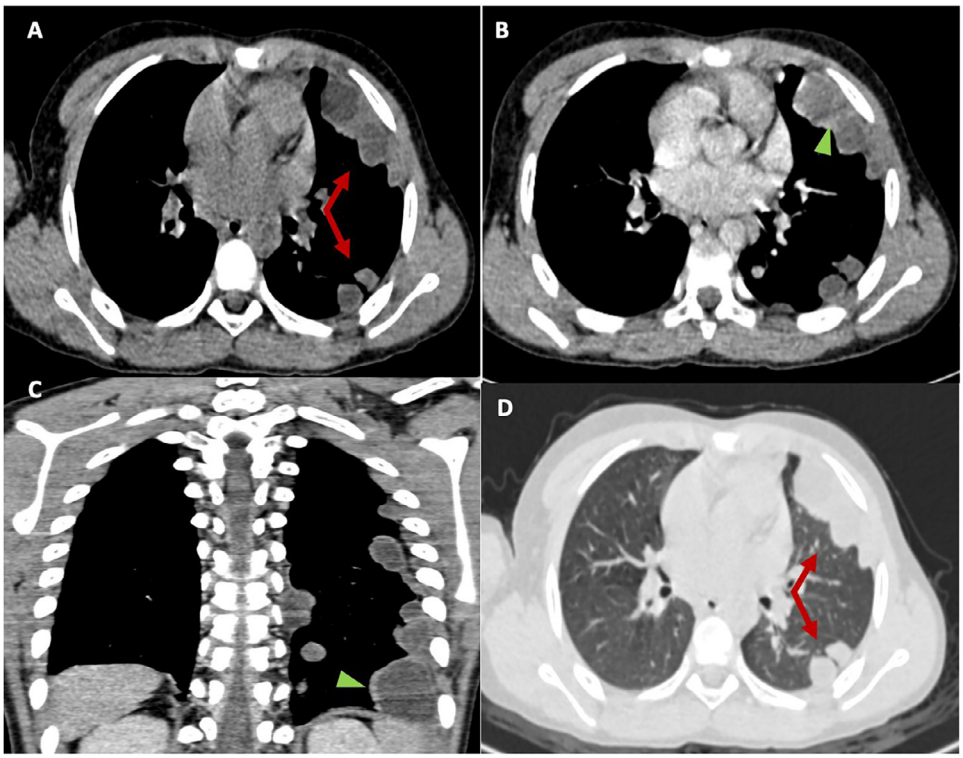


**Fig. 2 – Thoracic CT scan in axial view with mediastinal window before (A) and after (B) contrast injection, coronal view with mediastinal window postcontrast (C), and axial view with parenchymal window. The images reveal multiple cystic formations in the left pleura, characterized by oval and round shapes, thin walls, and lack of enhancement postcontrast. Note the subtle passive atelectasis caused by the largest cysts on the adjacent lung parenchyma (arrowheads).**

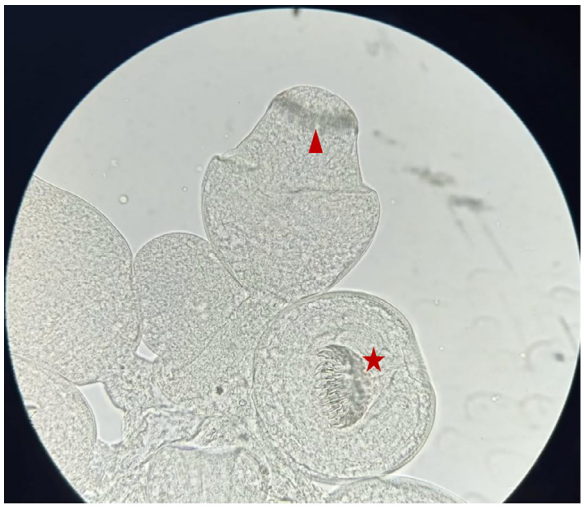

**Fig. 3 – Parasitological analysis of the cyst fluid demonstrating viable scolices: evaginated scolex (arrowhead) and invaginated scolex (asterisk).**

The classification of hydatid cysts is based on their imaging features [4]. Type I cysts are simple, while type II cysts present as smaller cysts within a larger 'mother' cyst. Type III cysts are entirely calcified, and type IV cysts are complicated, often involving rupture and/or superinfection.

Hypereosinophilia is typically absent in cases of intrathoracic hydatid disease. Immunological tests such as IgG ELISA, indirect hemagglutination, and Western Blot may provide supportive evidence, although their sensitivity is approximately 60%. Therefore, a combination of biological tests and radiological imaging is recommended for an accurate diagnosis [1,12,13].

In the absence of cyst rupture, cyst puncture is strictly contraindicated, which is why cytological or pathological diagnosis is usually reserved for postsurgical excision of the cyst [1].

Differential diagnoses may include pulmonary tuberculosis, pneumonia, and lung abscess, all of which can present with similar symptoms. Imaging plays a crucial role in distinguishing these conditions. Additionally, laboratory tests and serological analyses can help differentiate hydatidosis from other infectious and inflammatory diseases [4].

Definitive diagnosis is typically established through visualization of the hydatid membrane and/or daughter vesicles or through parasitological analysis of the surgical specimen, which involves observing hydatid material such as scolices, hooks, and related structures [13,14].

The primary treatment for pleural hydatid disease is surgical removal [15]. Medical intervention is used when surgery is not feasible or as an adjunct to surgical therapy [16]. For patients who are not suitable candidates for surgery, the PAIR (puncture, aspiration, injection, re-aspiration) technique may serve as an alternative treatment strategy [17].

## Conclusion

Pleural hydatidosis is extremely rare, particularly among pediatric patients, and can result in significant morbidity and mortality. Imaging plays a key role in its diagnosis. Due to the rarity of this condition, it is essential for radiologists to be aware of its existence and consider it, especially in regions where echinococcal infection is endemic.

## Ethics approval

This is a case report, based on the editor Guidelines-Ethics Approval and Informed Consent Statements: Ethics committee/IRB approval is often not required.

## Guarantor of submission

The corresponding author is the guarantor of submission.

## Patient consent

Written informed consent was obtained from the legal authorized representative (LAR) of the patient for the publication of this case report.

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