

An Unusual Location of Hydatid Cyst in the Pleural Space in a Child

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Received: May 22, 2023

Published: September 13, 2023

Abstract

Hydatid disease is a zoonotic parasitic disease caused by *Echinococcus granulosus*. The most common site is the liver, followed by the lung. Pleural involvement is rare; it could be primary or secondary. Imaging plays an important role in the diagnosis. We present the case of a 9-year-old patient who was previously operated on for a left lung hydatid cyst 4 years ago. She consults in the emergency room for a cough, chest pain, and expectoration, all evolving in the context of apyrexia. Her chest x-ray revealed multiple round opacities of the left pulmonary field. A CT scan showed several pleural cystic lesions, some of which contained septa and others embedded at the cost diaphragmatic recess, pushing down the diaphragm and exerting a mass effect on the spleen. No pulmonary involvement was noted. The diagnosis of pleural hydatid cysts was suspected on imaging and confirmed later by surgery and histological analysis.

Keywords: Pleural hydatidosis; *Echinococcus granulosus*; Hydatid cyst

Introduction

Hydatid disease, also known as hydatidosis, is an endemic parasitic zoonotic disease in some developing countries and constitutes a real public health problem [1]. It is more prevalent in sheep-raising areas [2].

The lung is the second most common site of involvement after the liver, but it is the most common in children. Pleural involvement is rare and presents in only 1.3% of thoracic locations [3,4].

Clinically it may be asymptomatic or cause cough, chest pain, fever, dyspnea, or hemoptysis. [3].

We report the case of secondary pleural hydatidosis in a 9-year-old girl who was previously operated on for a left lung hydatid cyst 4 years ago.

Case Report

A 9-year-old girl who was previously operated on for a left lung hydatid cyst 4 years ago, consults for a cough, chest pain, and expectoration that have been progressing for a month without fever. The patient was from a rural area. Her physical examination revealed that she was in respiratory distress and had decreased vocal fremitus.

The chest x-ray showed multiple well-circumscribed, rounded opacities of water tonality with no air-fluid levels occupying the left pulmonary field. No plural effusion was noted (**Figure 1**).

A thoracic and abdominal CT-scan revealed multiple cystic lesions along the costal pleura, without signs of rupture, some of which contained septa, exerting a mass effect on the chest wall, the diaphragm, and the spleen (**Figure 2**). No other similar le-

sions were found in the rest of the examination besides a small and partially calcified hepatic cyst (not shown). The blood count was normal, but the hydatid serology was positive. The patient underwent surgery, and histological examination confirmed the presence of non-ruptured pleural hydatid cysts. The outcome was good, and she has been prescribed Albendazole in the post-operative period, and she remained asymptomatic in the follow-up visits.



Figure 1: CT scout showing multiple round opacities of the left hemi thorax with water tonality.

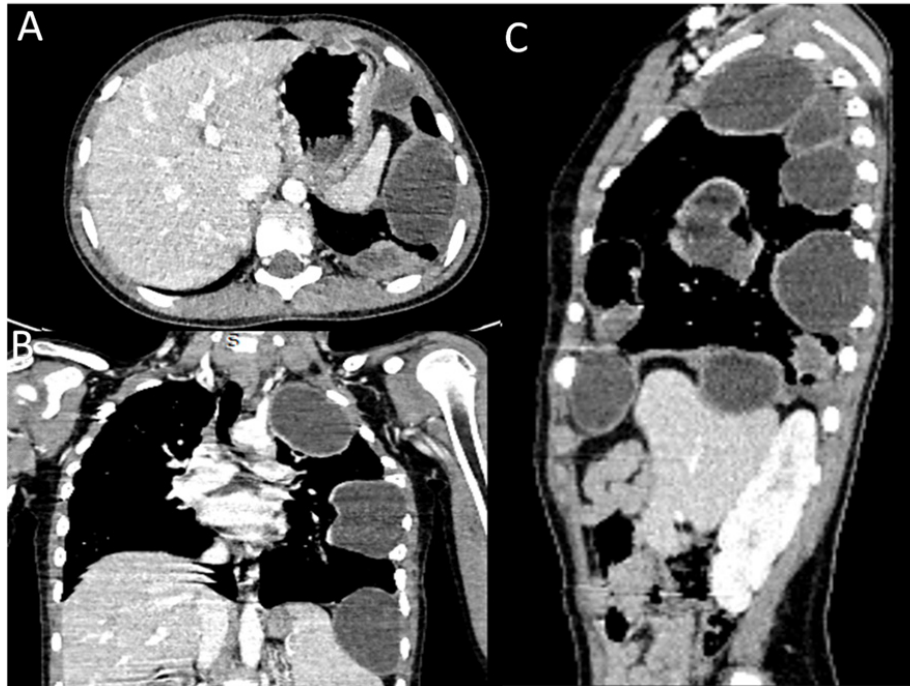


Figure 2: A axial, B coronal, and C sagittal enhanced chest CT demonstrating multiple hypodense cystic lesions along the left costal pleura with mass effect on the spleen.

Discussion

Human echinococcosis is a parasitic zoonotic disease caused by the genus *Echinococcus* and distributed worldwide. Carnivores are definitive hosts, while humans are accidental intermediate hosts. They may be infected either through the ingestion of contaminated food or fluids or through direct contact with the contaminated fur of a definitive host [5].

Intrathoracic extrapulmonary hydatid disease is very rare, but it can involve the pleural space, the extrapleural region, the pleural fissure, the chest wall, the mediastinum, the pericardium, the myocardium, and the diaphragm.

Primary involvement of the pleura, including the pleural fissure, is due to direct larval infestation by hematogenous or lymphatic spread. Secondary pleural involvement may occur after the spilling of the cyst contents, after the rupture of pulmonary or liver cysts into the pleural space, as a late complication of surgical treatment, or after diagnostic percutaneous transthoracic aspiration of pleural hydatid cysts [6].

Hydatid cysts can be asymptomatic or present symptoms related to compression of adjacent organs. The most feared complication is rupture, which could lead to anaphylactic shock and even death [6]. Clinical symptoms may include fever, dyspnea, chest pain, cough, hemoptysis, and sweating [7]. According to Sarkar M et al., hemoptysis is more prevalent in adult patients; in contrast, it is usually absent in pediatric patients [2].

Secondary pleural hydatidosis is an uncommon complication that may manifest in three different ways: hydatidothorax, hydatid pleural granulomatosis, and pleural hydatid grafting [7]. Our case is a peculiar example of this last form.

Pneumothorax is rare and can be associated with pleural rupture of pulmonary or hepatic hydatid cysts. Intact cysts can cause pleural effusion. However, the rupture of infected cysts is responsible for empyema [7].

Hydatid cysts in the pleural cavity can be found in both the

subpleural region and fissures, and most of the cysts are attached with a thin pedicle to the visceral pleura [8].

On chest radiography, cysts appear as a well-defined and homogenous pleural opacity of water tonality, revealing acute angles with pulmonary parenchyma [9]. Peripheral calcification may also exist [4]. CT and MRI allow a more precise study of the cysts and confirm their liquid nature. They can be unilocular or multilocular [8]. It also permits the detection of an associated pleural effusion, pneumothorax, or empyema [6].

On CT, the collapsed membranes in the pleural fluid produce the so-called "serpent sign" [6,7]. Regardless, MRI is preferred for demonstrating the detached membranes, as they appear hypointense in T2-weighted images [6].

Even though the pleural layers don't have blood vessels, calcium, potassium, chloride, water, and urea can pass through the cyst membrane and cause a hydatid cyst to form and grow [8]. The differential diagnosis of pleural hydatid cyst should include empyema, bronchogenic cyst, pleuropericardial cyst, enteric cyst, dermoid cyst, thymic cyst, lymphangioma, pleural metastases, lymphoma, mesothelioma, and synovial sarcoma [4,8].

Hyper eosinophilia is generally absent in intrathoracic hydatid disease [4]. Serological tests such as enzyme linked immunosorbent assays (ELISA) have a very high sensibility. However, its specificity is low at 61% [8,10]. All cases of confirmed Hydatid disease in Zaman K et al.'s study were seropositive for anti-Hydatid IgG antibodies, indicating that serological tests are important in the diagnosis [2].

The treatment is surgical and is based on the excision or enucleation of the cyst, followed by a histopathological examination [5]. Medical treatment using albendazole, mebendazole, or the combination of albendazole and praziquantel is considered a complement to surgical treatment but can be used in patients who can't be operated on, with recurrent hydatidosis, or in the case of a small, asymptomatic, and calcified cyst [6,7].

Puncture, aspiration, injection, and re-aspiration (PAIR) is another alternative for non-surgical patients but can be associated with allergic reactions. PAIR may be combined with albendazole to prevent secondary hydatidosis [7].

Conclusion

Pleural space is a rare location for hydatid disease. The possibility of secondary pleural hydatidosis should be considered in patients with a previous history of liver or lung hydatidosis. The diagnosis is based on clinical symptoms, radiological findings, and supportive serological evidence.

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