Case Report

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Loculated pleural effusion caused by a primary plural hydatosis

Vahan Moradians[1]; Zahra Mozooni[2]; Ali Babazadeh[3]; Leyla Bahadorizadeh[2,3]*; Mahshad Esfandiari[3]

[1]Department of Pulmonology and Critical Care, Iran University of Medical Sciences (IUMS), Tehran, Iran.

[2]Antimicrobial Resistance Research Center, Institute of Immunology and Infectious Diseases, Iran. 3Department of internal medicine, Iran University of Medical Sciences, Tehran, Iran.

[3]Department of internal medicine, Iran University of Medical Sciences, Tehran, Iran.

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***Corresponding Author:** Leyla Bahadorizadeh, Department of Pulmonology and Critical Care, Iran University of Medical Sciences (IUMS), Tehran, Iran. Tel: +9864352474. Fax: +9866509073.
E-mail: L.bahadorizadeh@yahoo.com

Abstract

Background: Echinococcosis is a parasitic infection caused by echinococcus granulosus larvae. It primarily affects the liver but can be found in other organs as well. Symptoms of pulmonary cystic echinococcosis include cough, chest pain, dyspnea, and hemoptysis. Pleural invasion as a primary location for hydatosis is extremely rare.

Objective: The case presented involves a patient with primary pleural hydatid cyst involvement, which posed a diagnostic challenge due to unusual radiological manifestations.

Case Report: A 70-year-old man who was a farmer and had habitual contact with dogs, presented with symptoms of progressive exertional dyspnea, fatigue, productive cough, weight loss, and pleuritic chest pain. Physical examination revealed decreased lung sounds and coarse crackles in the left hemithorax. Laboratory data showed elevated white cell count (Eosinophil count 1600/µl), high C-reactive protein, elevated erythrocyte sedimentation rate, and high serum lactate dehydrogenase and ferritin levels. CT scan revealed lobulated pleural effusion. Thoracentesis was performed and the pleural fluid was semi-clear and yellowish. Further Abdominal ultrasound was performed, which did not show evidence of liver and spleen involvement, also evaluation led to a diagnosis of primary pleural hydatid cyst involvement based on positive hydatid cyst serology tests.

Main Measures: Treatment with Albendazole 400 mg BD "15 mg/kg/day in 2 divided doses" started for 6 months, also the patient was a candidate for a Video-assisted thoracic surgery which, he did not consent to, then follow-up CT scan showed resolution of consolidation with fibrotic bands remaining. Pleural hydatid cysts are typically secondary to lung or liver cysts.

Discussion and conclusion: Lung involvement can occur through larval or hematogenous spread. Primary pleural hydatidosis is very rare, occurring in less than 1% of cases, in this case, no other lesions were found, confirming the diagnosis of primary pleural hydatidosis. Therefore, in the differential diagnosis of all patients who live in an endemic area of echinococcosis and present with pleural effusion, the reason of which is not determined in the investigations, we should consider hydatid cyst.

Keywords: Hydatid cysts, Echinococcosis, hemithorax.

Introduction

Echinococcosis is a parasite infection caused by larval stage of the echinococcus granuloses [1, 2]. It involves definitive and intermediate host in their life cycle [3]. they have three developmental states:

1. The adult tapeworm lives in the definitive host.
2. Eggs are in the environment
3. The meta cestode lives in intermediate host [4,5].

Definitive host ingests Meta cestodes and the mature tape worms release the eggs, which hatch into Meta cestodes, which infest the liver, lung, muscles, and other organs of the intermediate host like humans [6]. Hydatid cysts may be found in almost any site of the body, either from primary inoculation or via secondary spread [7]. The liver is affected in approximately two-thirds of patients, the lungs in approximately 25 percent, and other organs including the brain, muscle, kidneys, bone, heart, and pancreas in a small proportion of patients [8]. The most common symptoms of pulmonary

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cystic echinococcosis (CE) include cough (53 to62 percent), chest pain (49 to 91 percent), dyspnea (10 to 70 percent), and hemoptysis (12 to 21 percent [9]. Less frequent symptoms include malaise, nausea and vomiting, and thoracic deformations [7]. Pleural invasion as a primary location in hydatosis is extremely rare and has not been reported yet in Iran. Here in this case, we present a patient with primary pleural hydatid cyst involvement and diagnostic challenge because of the unusual radiological manifestation in Spiral Thoracic computed tomography scan (CT-Scan).

Case Report

An Iranian 70 years old male, known case of HTN, that was controlled on diet and valsartan 80 mg daily, presented to the emergency ward with the history of 2-month-long progressive exertional dyspnea and fatigue. He had been suffering from productive cough without hemoptysis. There was significant weight loss of about 20 kg within the past 2 years due to anorexia and vague abdominal pain in left upper quadrant with non-documented frequent fever, without complaint of night sweating. He experienced pleuritic chest pain in his left hemithorax with no radiation, orthopnea, or paroxysmal nocturnal dyspnea. He was a farmer and had habitual contact with dogs. He lived in an apartment with his wife. He smoked 50 pack/years and was a chronic oral opium user but did not use illicit drugs or alcohol. There was no remarkable family or allergy history. He denied any suspicious contact with the same symptomatic person.

Up on physical examination, He looked ill, and the temperature was 37.1, blood pressure 130/90 mm Hg, heart rate 92 per minutes and respiratory rate 16 per minutes. Oxygen saturation was 93% on room air.

Chest examination revealed decreased lung sounds in left hemithorax with inspiratory coarse crackles in base of left lung. The rest of the systemic physical examinations were otherwise unremarkable. Laboratory data, the white cell count was 13000 per cubic millimeter (reference range: 4500-11000, with 73% neutrophils, 18% lymphocytes and 9% Eosinophils). The hemoglobin level was 11.1 gr per deciliter (reference range: 13-17) and the platelet count was 148,000 per cubic millimeter (reference range: 150,000-400,000). The reverse transcription-polymerase chain reaction (RT-PCR) test for covid 19 was negative, qualitative C-reactive protein titer was high (3+), erythrocyte sedimentation rate was 84 millimeters per hour (reference range: 0-13), serum lactate dehydrogenase was1478 unit per liter (reference range: 140-280) and serum ferritin was 850 micrograms per liter (reference range:20-300). Other laboratory results were otherwise unremarkable. Spiral Thoracic computed tomography scan (CT-Scan) without contrast was done and it showed, loculated pleural effusion was obvious in figure1.

Ultrasound guided thoracentesis was done and 400cc pleural effusion was drained. The PE was semi clear and yellowish in

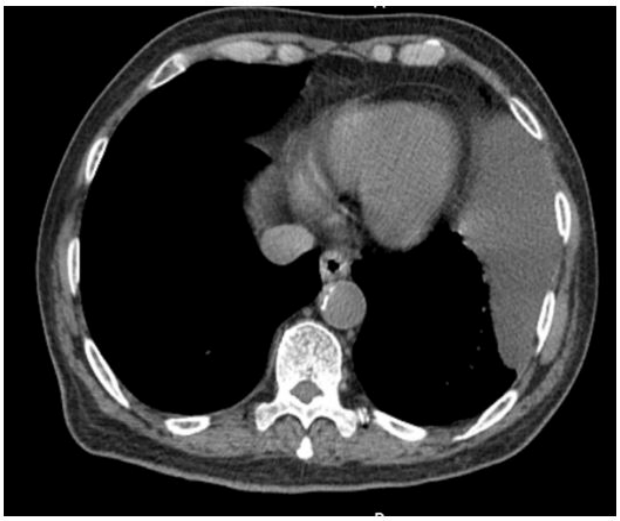

Figure 1: *Loculated pleural effusion was obvious with maximum thickness 5 mm in inferolateral aspect of left hemithorax.*

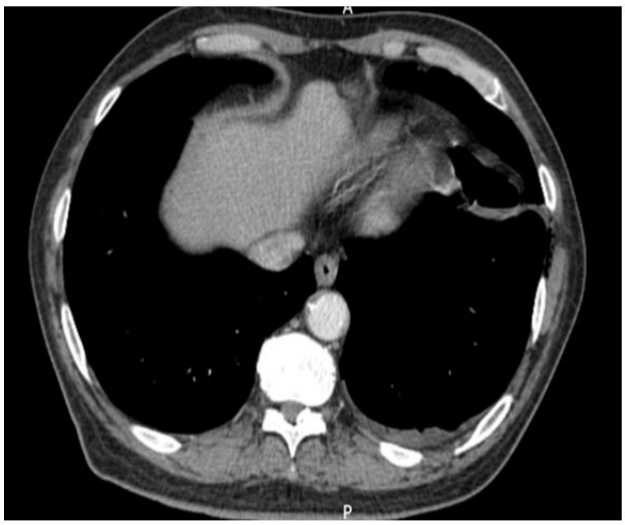

Figure 2: *The chest CT after treatment demonstrated consolidation had been resolved and only fibrotic bands remained at the site of the lesion.*

color with a total protein concentration of 1g/dL (7.5 g/dL in serum) and a lactate dehydrogenase level of 10 IU/mL (421 IU/mL in serum). It contained 0-1/ml WBC and 10/ml RBC and glucose level was 19 mg/dl. Pleural culture was negative and cytologic study of pleural effusion was negative for malignancy.

pleural effusion analysis was not indicative of human body fluids results. Therefore, reevaluation started, and hydatid cyst serology tests were performed because of its endemic distribution in Iran. Hydatid cyst IgG antibody was positive. Therefore, pleural fluid analysis was sent to investigate hydatid cyst antibody, which was positive, so Albendazole 400 mg BD “15 mg/kg/day in 2 divided doses” started for 3 months according to Hydatid Cyst diagnosis. The patient was a candidate for Video Assisted Thoracoscopic Surgery (VATS), but he did not consent to do it. Abdominopelvic sonography was unremarkable for liver cyst or mass and completely was normal. The chest CT after treatment demonstrated consolidation had

been resolved and only fibrotic bands remained at the site of the lesion (Figure2).

Discussion

Originally, we described a case of primary pleural hydatid cyst that presented with chronic pulmonary symptoms and plural effusion. (Symptoms of pleural hydatidosis are similar to those found in pleural effusions such as dyspnea, mediastinal shift and reduction in lung volume. Even so, up to 15% of cases can be asymptomatic [10, 11]. However, these can also be present alongside chest pain and other signs of cardiac or vascular involvement or compression [12]. Hydatid disease is also a rare cause of recurrent acute pulmonary embolism. This complication may develop after invasion of the cardiovascular system or direct invasion of the inferior vena cava [13].

Our clinical suspicion of hydatid disease was based on pleural fluid analysis, which was inconsistent with body fluids and could not be justified by any diagnosis. Because of his occupation and living in endemic region (disease is very common in Middle Eastern countries, including Iran), the antibody against Echinococcus was checked by ELISA method from the patient's pleural fluid and blood, which was positive, so our diagnosis was confirmed. (Imaging methods and serological tests are useful diagnostic tools in this disease [14].

CT scan of hydatid cysts with low attenuation round lesions and no contrast enhancement is diagnostic. Calcification of the cyst wall called "eggshell calcification" is best seen on unenhanced CT [14]. Other radiological signs for hydatid disease include presence of daughter cysts, water-lily sign (for ruptured cysts) and serpent sign (ruptured and completely drained cysts) [15].

(Serological tests include immunoelectrophoresis and ELISA. Immunoelectrophoresis is a more sensitive test for antihydatid antibodies, but ELISA is more specific [16].

(Other tests such as skin tests, complement fixation, blood eosinophil count and indirect hemagglutination tests can be used, but must be interpreted carefully as they have a tendency towards false-positive results [17].

(Pleural hydatid cysts are usually secondary to peripheral lung cysts that rupture or herniate into the pleural cavity or, less commonly, transdiaphragmatic contamination from cysts located in the right upper lobe of the liver [18].

Lung involvement are usually caused by transphrenic spread of larvae through the hepatic sinusoids or by haematogenous spread of metacestodes following rupture of eggs in the stomach. After digestion of outer capsule of the egg, the freed embryo enters the portal vein through duodenal mucosa and locates and grows in the hepatic capillaries. Some of them pass through the capillaries and become lodged in the lungs and other organs, which may also result from trans lymphatic spread [14,19,20].

(Primary pleural hydatidosis, which is classified as extrapulmonary intrathoracic cysts, is very rare and occurs in less than 1% of cases [21].

It has been shown that the replacement of hydatid cysts in unusual locations might be linked to the genotypes of the parasite and possibly its intra-genotypic characteristics and occurs through hematogenous or lymphatic spread [22].

(Primary pleural hydatidosis appears as a solitary pleural hydatid cyst or as a parasitic pleural effusion [23].

In our case, no parenchymal lesions of the lung or other parts of the chest were found except pleural effusion. Abdominal and pelvic ultrasound showed no evidence of involvement. For this reason, according to the positive serology of blood and pleural fluid, the diagnosis of our patient was primary pleural hydatidosis.

(Frequent, primary pleural hydatidosis has been presented as an exceptional situation for hepatic hydatidosis [3, 24].

Primary development of a hydatid cyst within the parietal pleural structures is possible and can later lead to hydatid pleural effusion. The histological structure of the cystic membranes allows the passage of calcium, magnesium, water, urea as well as other nutritional substances that may pass through by diffusion and favor the development of the cyst [1]. (The treatment strategy for hydatid disease is surgery combined with pharmacotherapy with benzimidazoles (mebendazole or albendazole). Puncture, aspiration, injection and reaspiration is effective in liver cysts, and surgical options in pulmonary cysts, depending on the location of the lesion, include enucleation, wedge resection, segmentectomy, lobectomy and pulmonectomy. After Surgery, for recurrence and primary treatment, a long follow-up is needed [14, 25, 26].

(In the case of primary pleural lesions, due to the rarity of the disease, there is no definite treatment strategy, but according to the literature, Video-assisted thoracoscopic surgery (VATS) and medical treatment with albendazole is recommended.)

In our patient, despite the fact that VATS was not performed, there was no recurrence after two months of stopping the treatment.

Conclusion

The development of hydatid cysts in unusual body locations presents a major diagnostic dilemma, and may mimic other entities, making it difficult to distinguish from other diseases. Moreover, the lack of specific radiological findings makes it difficult to differentiate the disease in such cases. Therefore, in any endemic area, if there is clinical doubt, the diagnosis of hydatid disease should be considered and the necessary investigations should be performed. Furthermore, in cases of uncommon hydatid disease, evaluating other locations should be taken into consideration for any infestations of hydatidosis

Declarations

Conflict of Interest: The authors declare that there is no conflict of interests.

Consent to participate: All authors have read and approved the final version of the manuscript for publication.

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