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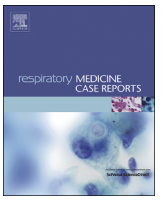

Case Report

Hydatid pulmonary embolism underlying cardiac hydatid cysts – A case report

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ABSTRACT

Cystic hydatidosis is an endemic parasitic disease with usual localization in liver and lungs. Rarely it localizes in uncommon sites, the right ventricle being an exceptional localization.

We present an extremely rare case of a young man with hydatid pulmonary embolism complicating right-ventricle hydatid cysts. Echocardiography, CT pulmonary angiogram and MR-angiography were performed for the diagnostic evaluation. Our patient did not undergo surgery. He was discharged on a regimen of albendazole, and is still being followed-up.

Hydatid disease rarely presents with pulmonary embolism. It has uncharacteristic clinical features, requiring particular diagnosis and therapy.

1. Introduction

Cystic echinococcosis (CE), or cystic hydatidosis, is a anthropozoonotic parasitic disease caused by the larval stages of the cestode *Echinococcus granulosus*. It has a cosmopolitan distribution and it is highly endemic in pastoral communities, particularly in Mediterranean countries [1].

75% of hydatid cysts are found in the liver, 15% in the lungs, and 10% in other locations. The cardiovascular system is involved in less than 2% of the cases, making it a potentially lethal condition [2]. The right ventricle localization is exceptional and may be revealed by cyst rupture.

Hydatid pulmonary embolism is a very rare complication.

We present an exceptional case of a young man who has developed hydatic pulmonary embolism secondary to right ventricule hydatid cysts.

2. Case presentation

Twenty-three years old man, without history of illness nor contact with dogs, admitted to the Department of Pneumology and Phtiseology for daily episodes of moderate hemoptysis, with increasing shortness of breath, which had started approximately 15 days prior to admission.

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Anamnesis revealed that the patient had similar episodes throughout the previous year, associated with dyspnea, all evolving in a context of uncounted weight loss. The clinical examination was without particularities. Oxygen saturation level was at 95%.

Biological tests showed eosinophilia with 670 elements/mm^3 and a normal hemoglobin level at 14,5 g/dL. The platelets count and the C reactive protein were within the normal range. The D-dimer test was normal.

The chest X-ray showed regular nodular opacities, in both lung fields.

On the first line of investigations, a Computed Tomography Pulmonary Angiography (CTPA) was performed. It showed multiple echinococcal cysts in both lung parenchyma, containing floating membranes for most of them (Fig. 1), with intra-arterial cysts extending along the segmental pulmonary arteries of the left upper lobe and the right upper and middle lobes, that caused luminal widening consistent with emboli (Fig. 2).

It also showed 02 heterogeneous cystic images containing air in the left upper lobe (Fig. 3); they are hydatid cysts fistulized in the bronchi.

The CTPA also revealed typical univesicular hydatid cysts in the right ventricle, measuring 18 × 17 mm in size for the biggest one (Fig. 4).

Signs of pulmonary hypertension were observed: Enlarged pulmonary trunk, measuring 38 mm in diameter, and a main pulmonary artery to ascending aorta ratio >1 (Fig. 5).

A two-dimensional echocardiography showed a dilated right ventricle containing a small cystic mass.

No cysts in pericardial cavity were found. There was significant pulmonary hypertension, with a dilated main pulmonary artery, without any proximal cysts.

The suspected diagnosis was of right cardio-pulmonary hydatid cysts complicated with hydatid pulmonary emboli. Hydatid serology was then carried out and the level of antiechinococcal antibodies was found to be positive.

Thoracic MR Angiography confirmed the cystic nature of the cardio-pulmonary lesions and masses described above. They appeared as round, T1-hypointense and T2-hyperintense structures (Figs. 6 and 7), with no hyperintense signal in DWI and no contrast enhancement. Floating membranes within cysts appeared hypointense in T2-weighted sequences (Fig. 6). The cystic structure of the emboli was also shown in MR images.

The positive hydatid serology, the imaging findings and the prevalence of hydatid cysts in our country led to the diagnosis of a pulmonary embolism complicating cardiac hydatid cysts.

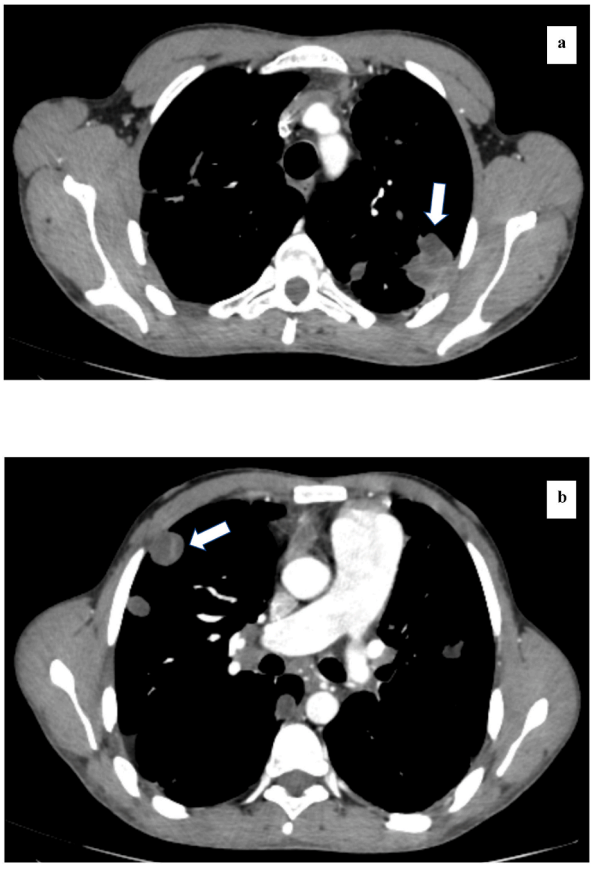

Fig. 1. (a, b). Axial post-contrast CT scan images: Pulmonary nodules compatible with echinococcal cysts in both lung parenchyma, containing floating membranes (arrow).

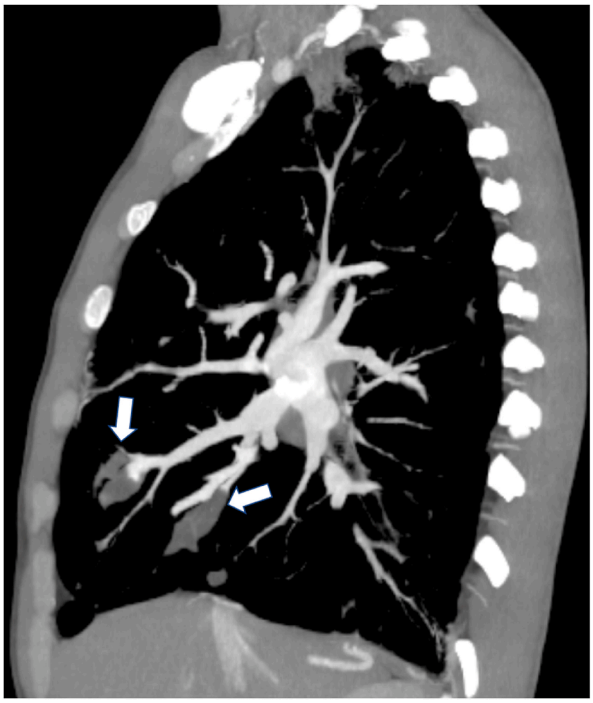

Fig. 2. Sagittal post-contrast CT scan MIP image: Intra-arterial defects corresponding to echinococcal cysts extending along the segmental pulmonary arteries of the middle lobe (arrows).

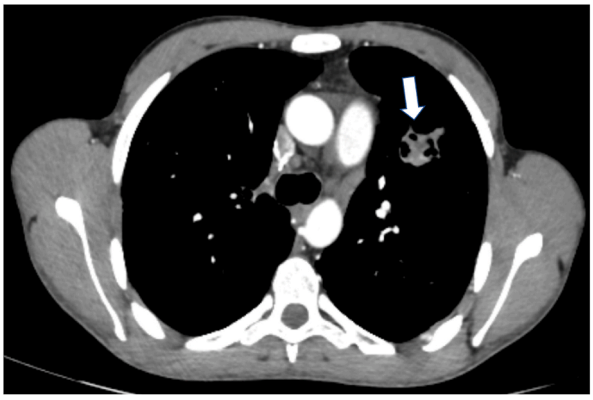

Fig. 3. Axial post-contrast CT scan image: Heterogeneous pulmonary cystic image containing air in the left upper lobe (arrow).

A cerebral CT-scan and an abdominal sonography were also performed in search of other hydatid localizations. They were found to be normal.

Our patient did not undergo surgery, as the cysts were located in distal branches of the pulmonary arteries. He was discharged on a regimen of albendazole, 15 mg/kg per day, for 28 days with a therapeutic window of 14 days, for a duration of 06 months. He was also put on furosemide and spironolactone to treat his pulmonary hypertension.

The patient will be in close follow-up every month in the Department of Pneumology and Phtiseology, and a second CT-scan will be performed by the end of the treatment duration.

3. Discussion

Cystic hydatid disease is considered a public health problem in undeveloped and developing countries [1].

Humans may be infected incidentally as intermediate hosts in the parasite's life cycle. The parasites enter the portal circulation through the intestinal mucosa and can reach any part of the body; most of them get trapped in the liver while the rest disseminate to other organs, developing into hydatid cysts.

Heart is rarely involved with less than 2% of all cases. The cyst is mostly located in the left ventricle (60%) followed by septal location [3]. Right ventricular, atrial and pericardial involvements have also been reported [4].

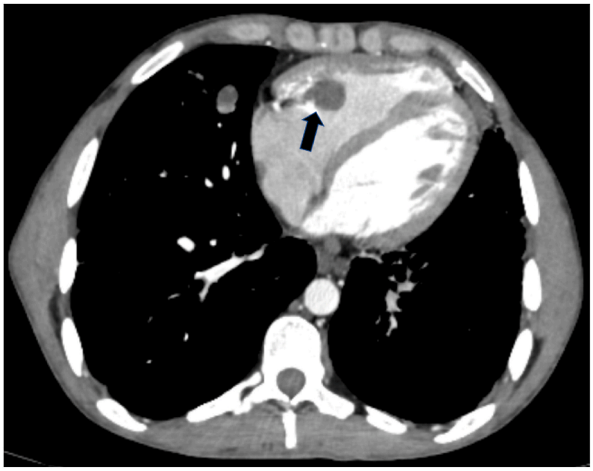

Fig. 4. Axial enhanced CT scan image: Univesicular hydatid cyst in the right ventricle (arrow).

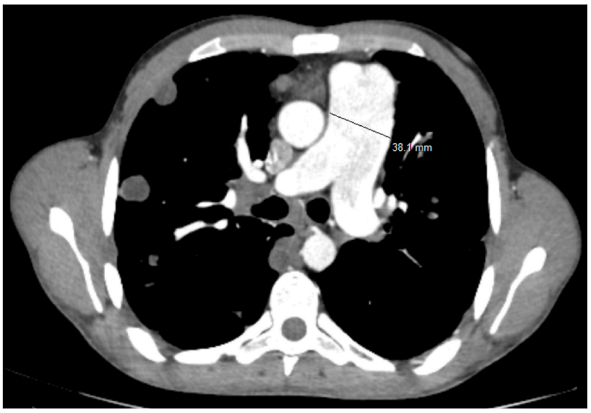

Fig. 5. Axial post-contrast CT scan image: Enlarged pulmonary trunk – Main pulmonary artery to ascending aorta ratio >1.

Hydatid pulmonary embolism is an exceptional but a potentially life-threatening complication [5]. In case of cardiac localization, it can occur as a result of a ruptured cyst of the right cardiac chambers and a migration of daughter vesicles to the pulmonary arteries [5,6].

Surgical and autopsy findings reveal that the embolism is caused by daughter cysts or vesicles that mechanically block blood flow, and that there are no blood clots or added thrombosis [5].

Hydatid pulmonary embolism can be classified in three groups according to the clinical picture: acute fatal cases; subacute pulmonary hypertension; chronic pulmonary hypertensive cases. The latter being the most frequent clinical form, punctuated by acute embolic episodes [7].

The pulmonary artery obstruction caused by slow-growing cysts may remain asymptomatic, as the pulmonary perfusion is maintained via the bronchial arteries. Excessive growth in size of these cysts may lead to total obstruction of the pulmonary arteries [8].

Clinical manifestations of hydatid pulmonary embolism are not specific. A clinical presentation similar to acute pulmonary thromboembolism with coughing, hemoptysis, dyspnea and chest pain is usually seen [8]. Chronic HPE can appear as pulmonary hypertension. Anaphylactic shock may develop due to leakage of the hydatid cyst fluid.

In the absence of a medical history of a visceral hydatid cyst, it is unlikely to think of a HPE as a first-line diagnosis. It can be suspected by the presence of anti-echinococcal antibodies or eosinophilia in blood tests. The diagnostic investigation is based essentially on the imaging findings.

Two-dimensional echocardiography is the imaging procedure of choice for detecting hydatid cysts in the heart and pericardium, but it rarely allows direct visualization of the pulmonary emboli. Features that may be suggestive of acute pulmonary embolism are right ventricular dysfunction and flattening or dyskinesis of the interventricular septum [6].

The Computed Tomography Pulmonary Angiography (CTPA) and Magnetic Resonance Angiography are the cornerstone of the diagnosis.

The CTPA shows the topography of hydatid cysts, the obstruction of the pulmonary arteries and the repercussions on the heart. Hydatid cysts are represented by round or oval masses, with well-defined borders and hypodense content relative to the capsule. The fluid is of variable attenuation, depending on the amount of proteinaceous debris.

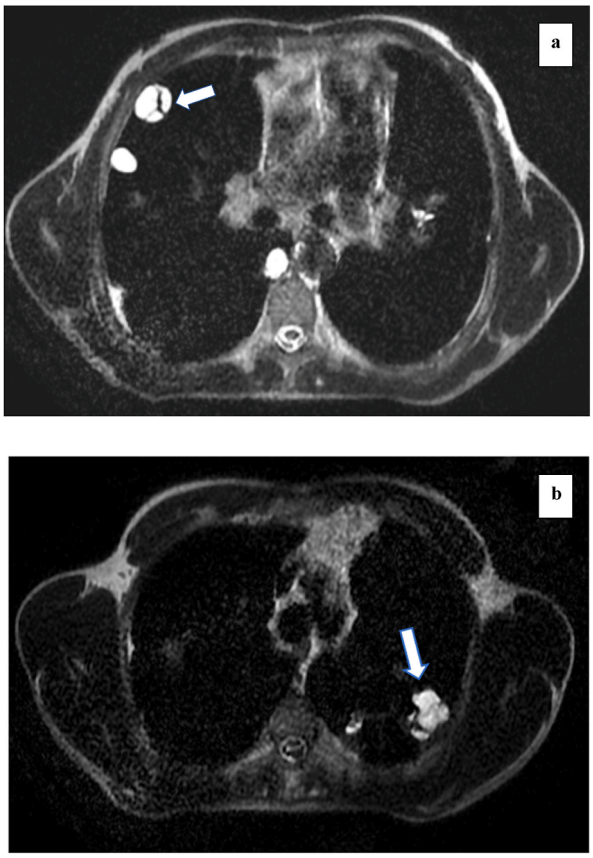

Fig. 6. (a, b). Axial MRI, T2-weighted images: Round hyperintense structure, containing hypointense floating membranes, compatible with a pulmonary hydatid cysts (arrow).

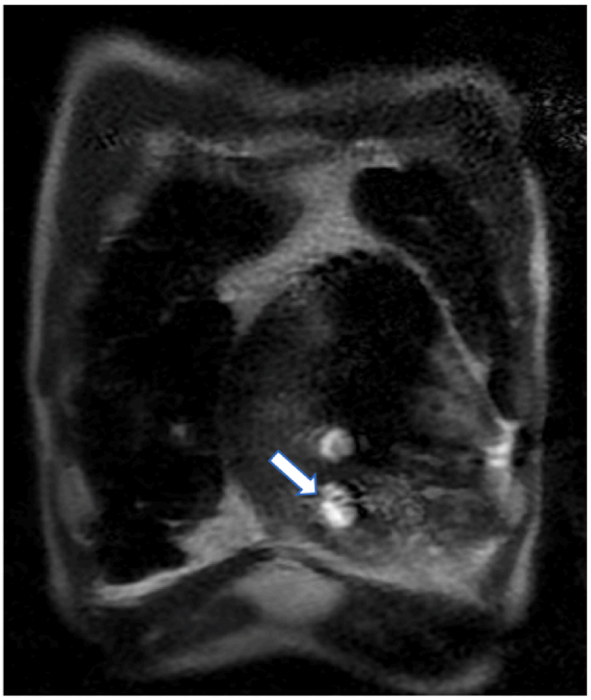

Fig. 7. Coronal MRI, T2-weighted image: Round hyperintense structure in the right ventricle, compatible with a cardiac hydatid cyst (arrow).

Hydatid emboli appear as homogenous hypodense cystic structures causing focal widening of the lumen of arteries, not enhancing with contrast, resulting in filling defects within the pulmonary vasculature [9]. They can be occlusive or non-occlusive, the latter is seen with a thin stream of contract adjacent to the embolus.

MRI displays the cystic nature of lesions better than Computed Tomography [9]. They appear as hypointense in T1-weighted sequences, hyperintense in T2-weighted sequences. The cyst capsule is T2-hypointense (hypointense rim sign), T1-isointense and shows mild contrast enhancement. The folded membranes within cysts are T2-hypointense [10]. In addition, MRI is described to be more advantageous than spiral CT-scan for examination of the heart and the great vessels, because, with images in multiple phases, it gives a more complete anatomic picture [11].

The additional performance of conventional pulmonary angiography is unnecessary.

There are no clear guidelines when it comes to the treatment of this pathology.

Although the use of antihelminthic drugs, such as mebendazole and albendazole, have shown promising results in the treatment of hydatid disease, surgical management is the first-line treatment for cardiac hydatid disease and most cases of pulmonary arterial hydatidosis. Median sternotomy with cardiopulmonary bypass must be the surgical approach of choice in the treatment of cardiac hydatid cysts [12] and the removal of echinococcal material from proximal pulmonary arteries. It can also allow a simultaneous treatment of lung cysts.

Drug therapy helps to reduce a secondary seeding or a postoperative recurrence and is also advocated for patients with recurrent hydatidosis or who are contraindicated for surgical treatment [7].

4. Conclusion

Hydatid pulmonary embolism underlying cardiac hydatid cysts is an exceptional complication, that can be potentially life-threatening. CTPA and MR Angiography are the keys of the diagnosis. Early diagnosis with imaging studies and treatment are the main means of preventing complications.

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Availability of data and materials

The data sets are generated on the data system of the CHU Hassan II of Fez, including the biological data and the interventional report.

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Author's contribution

ZE is the corresponding author, she participated in the organization and writing of the article and studying the case with AK.
Professor NE, MH, BA and YL supervised working and validated the figures.
RB and YA contributed in clinical examination, treatment and follow up of the patient.
Professor and chief of department of radiology MM read and allowed the article for publication.

Declaration of competing interest

The authors declare that they have no competing interests.

List of abbreviations

CE	Cystic echinococcosis
CH	Cystic hydatidosis
LUL	Left upper lobe
HPE	Hydatid pulmonary embolism
CT	Computerized Tomography
CTPA	Computed Tomography Pulmonary Angiography
MRI	Magnetic Resonance Imaging
DWI	Diffusion-weighted imaging
MIP	Maximum intensity projection

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