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Case Report

Thoracic Hydatid Pulmonary Embolismic

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Abstract

Thoracic hydatid disease is a cosmopolitan anthropozoonosis that essentially prevails in the Middle East, South America and the Mediterranean countries. Its prevalence ranges between 1 and 10% and the arterial localization is rare, it does not exceed 2% of all thoracic hydatid cysts. The diagnosis should however be brought up, especially in endemic area of this zoonosis. The diagnosis has become easier with chest imaging. We report a case of hydatid pulmonary embolism secondary to the migration of hydatid cysts into the pulmonary arteries.

Keywords: *Hydatid Pulmonary Embolism; Thoracic Hydatid Disease*

Abbreviations

HC: Hydatid Cysts; CTPA: Computed Tomography Pulmonary Angiogram; CRP: C-Reactive Protein; TTE: Transthoracic Echocardiography; IVC: Inferior Vena Cava; PHT: Pulmonary Hypertension; WHO: World Health Organization

Introduction

Thoracic hydatid disease is a cosmopolitan anthropozoonosis that essentially prevails in the Middle East, South America and the Mediterranean countries. Its prevalence ranges between 1 and 10% [1] and the arterial localization is rare, it does not exceed 2% of all thoracic hydatid cysts [2]. The migration of hydatid cysts (HC) through pulmonary arteries is often acute or subacute. In rare cases, the embolization can cause chronic lung hydatid disease with post embolic precapillary pulmonary hypertension. The therapeutic management of this condition is difficult and the short and long term outcome can be dreadful with high risk of mortality [3]. We report a case of hydatid pulmonary embolism secondary to the migration of hydatid cysts into the pulmonary arteries.

Observation

A 30 years old male patient, living in a rural area, presented to the emergency department with an acute dyspnea. He had undergone a pericystectomy for a hydatid cyst of the right lung 5 years ago. He also presented with cough, low abundance hemoptysis and feeling feverish. There were no pulmonary embolism risk factors (Geneva score at 5). The clinical examination found the patient's temperature at 39.5°C, rhonchus on pulmonary auscultation and signs of right heart failure. There were no signs of thrombophlebitis of the lower limbs. The chest X-ray showed bilateral multiple round opacities, an elevated right diaphragmatic dome and a cardiomegaly with a cardiothoracic index at 0.66 cm (Figure 1). The Computed Tomography Pulmonary Angiogram (CTPA) found a voluminous intraluminal

defect inside the trunk of the right pulmonary artery measuring about 43 mm extending largely to its division branches: the upper lobar, the middle lobar and inferior lobar arteries (Figure 2). The left lobar arteries: superior and inferior were nearly completely obstructed by a thrombus which extended to the segmental branches. Multiple scattered cystic formations have been identified in both lungs with a cardiomegaly at the expense of the right cavities associated with a right pleural effusion of low abundance. On lower images, the chest CT showed a calcified cystic lesion of the liver.

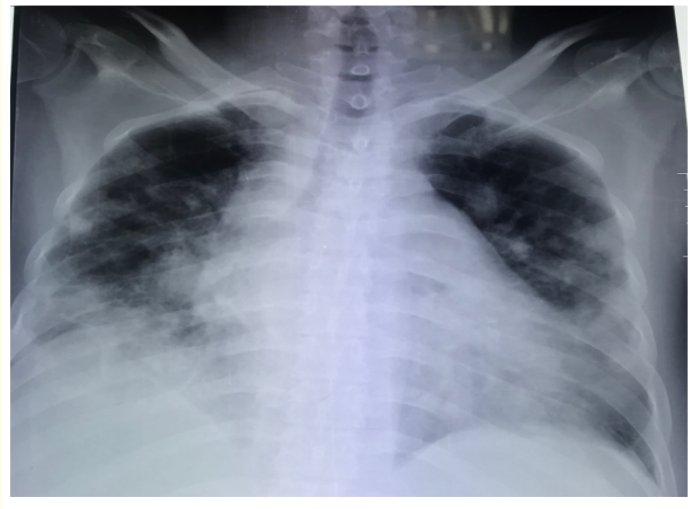

Figure 1: *Appearance of balloon release with cardiomegaly.*

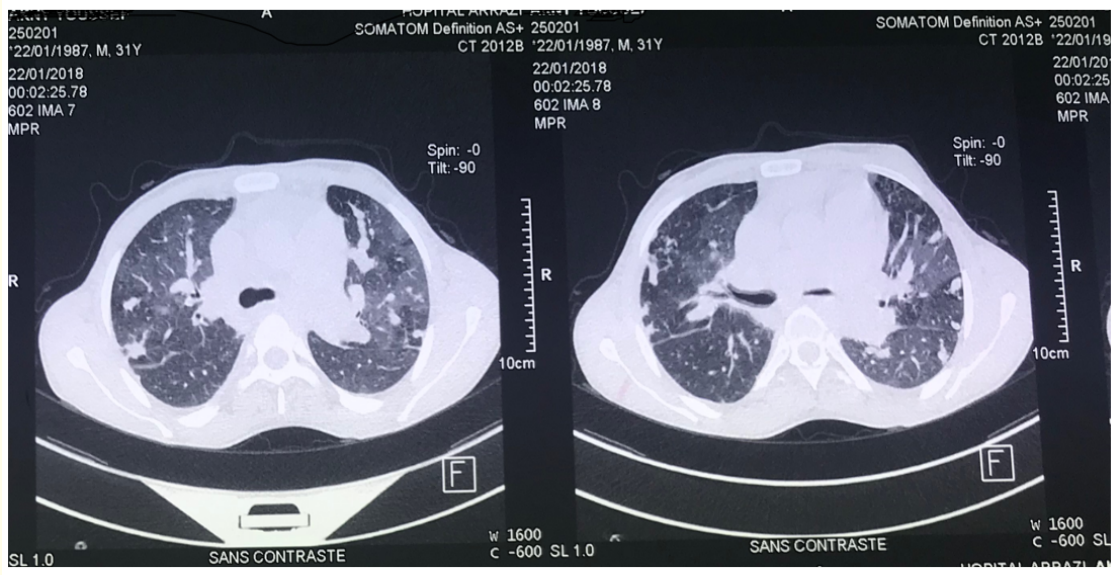

Figure 2a: *Chest CT, parenchymatous window showing a multiple lung hydatidosis.*

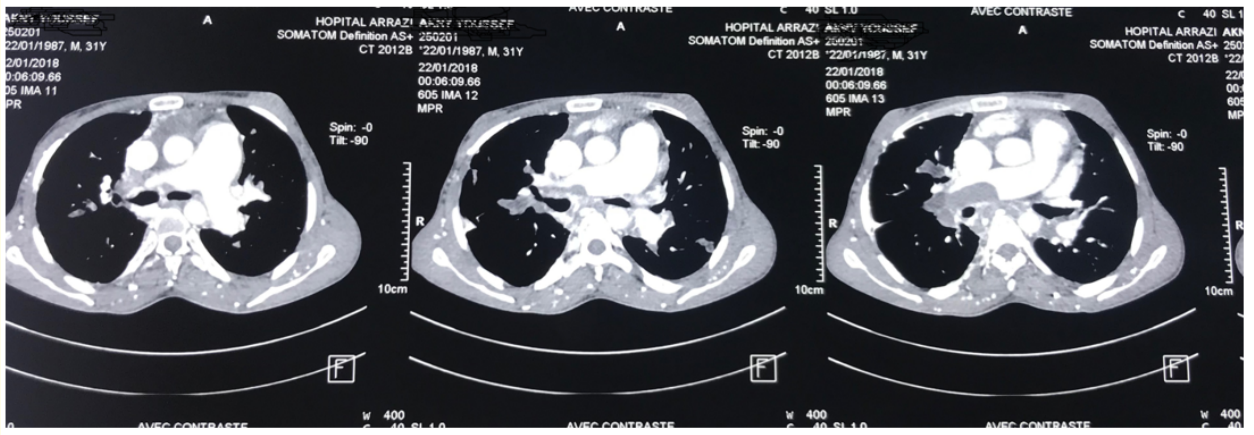

Figure 2b: *Chest CT; mediastinal window showing bilateral massive pulmonary embolism.*

The complete blood count found a hyperleukocytosis made of predominantly neutrophils (16,360 elements/mm^3) without hypereosinophilia. The C-reactive protein (CRP) was at 255 mg/L. The patient was put under noninvasive ventilatory support and antibiotics: amoxicillin-clavulanic acid associated with an aminoglycoside. The transthoracic echocardiography (TTE) did not find any intracardiac hydatid cyst but showed signs of acute pulmonary heart. The final diagnosis was a multiple hydatid disease (lung and liver) with a pulmonary arterial localization. The hydatid origin of these intra-arterial lesions was retained because of their purely cystic appearance on the computed tomography and the origin of hydatid emboli in the pulmonary artery was primitive. This hypothesis is supported by the absence of cyst in the heart chambers and the presence of calcified cyst of the liver.

The patient developed an acute respiratory distress with unconsciousness requiring his transfer to the intensive care where he was intubated and ventilated without improvement. The patient died the following day.

Discussion

Multiple pulmonary hydatid represents 5.1% of pulmonary presentations; the usual starting point of hydatid emboli is the heart via the pulmonary arteries; more rarely it can be primitive following a massive or repeated infestation from the liver, it is exceptionally bronchogenic [4,5]. Cardiovascular localizations of Hydatid cysts are estimated at 0.02% to 2% of the hydatid disease [6]. The hydatid pulmonary embolism is a common complication when there is a HC of the right heart but is rare complication of the liver's HC [7]. The fistulization of a liver's HC in the inferior vena cava (IVC) causes the migration of daughter vesicles in the venous return to the right atrium, the right ventricle and the pulmonary artery [8]. The other mentioned but less probable pathophysiological hypothesis to explain this condition is that the parasite manages to cross the arterial wall through previous small lesions of the intima, aneurysms or nutritional vessels [3]. This mechanism was retained for our patient because the transthoracic echocardiography did not reveal any intracardiac HC in addition to the existence of a calcified cyst of the left liver. The clinical presentation of hydatid pulmonary embolism is not specific and therefore is often wrongly considered as a blood clot. The hydatid origin of pulmonary embolism is suspected based on the patient's medical history, the clinical presentation and the positivity of the hydatid serology [3]:

- An acute presentation with a high risk of sudden death by massive pulmonary embolism or fatal anaphylactic shock. The diagnosis is often done after the autopsy which would explain the scarcity of reported similar cases to our patient's.

- Subacute pulmonary embolism and pulmonary hypertension (PHT) that can be responsible for death during the following year.
- Chronic PHT complicating an acute or subacute hydatid pulmonary embolism which had developed favorably.

The erosion of the intima by the cyst is responsible for thinning of the vessel wall with the formation of hydatid aneurysms which increases the risk of hematogenous dissemination and exposes to massive hemoptysis by rupturing into a bronchi [9]. Circulating hydatid debris can also cause thrombosis of the branches of the pulmonary artery and therefore give a clinical presentation of pulmonary embolism [7]. The discovery of hydatid emboli in the pulmonary artery can also be fortuitous when performing the assessment of the lesions of the hydatid disease. Thoracic imaging is, in this context, the examination of choice to confirm the diagnosis.

On the CT scan, the non-complicated intra-arterial HC shows as a mass of liquid density with well-defined limits and an enhancement of its wall after contrast injection [10,11]. The transthoracic sonography or better yet, the transesophageal echocardiography is useful for the analysis of cardiac and pulmonary artery hydatid with an excellent sensitivity and specificity. It also assesses the impact of pulmonary embolism on the cardiac function [12].

The treatment of hydatid pulmonary embolism is surgical in the absence of any contraindications [13]. The extraction of daughter vesicles occurs at the level of the pulmonary artery and its proximal branches and requires a cardiopulmonary bypass. Mortality depends on the severity of PHT and the parenchymal hydatid dissemination. PHT, chronic pulmonary heart and miliary hydatid are associated to a high mortality rate [14,15]. This surgery could not be performed in our patient because of the extension of the hydatid embolus to the distal branches of the pulmonary artery, the presence of bilateral emboli and multiple intra-parenchymal locations.

The medical treatment is indicated in addition to surgery or alone in case of chronic emboli, important dissemination across the pulmonary arterial bed or surgical contraindication (high surgical risk patients) [7]. Albendazole is the most commonly used molecule and its prescription is done following two therapeutic regimens. The first one includes a monthly 15 days of treatment at 10 mg/kg daily [16]. The second one seems to be more effective; it involves the continuous administration of albendazole for three months at a dosage of 10 to 12 mg/kg per day and is the one recommended by the World health Organization (WHO) [16]. The purpose of medical treatment is to reduce the size of cysts and the viability of the parasites, thereby decreasing the hydatid dissemination and recurrence. However, the response to treatment is inconsistent and its effectiveness can be difficult to distinguish from the natural evolution of the disease. The duration of the anti-parasite treatment is not codified. It is usually three to six months and is often extended if multiple active lesions. The role of anticoagulation remains controversial in the literature and deserves to be further evaluated. Most often it is implemented systematically, prior to the confirmation of the hydatid nature of pulmonary embolism, while other authors do not see its interest and instead adopt the anti-parasite treatment [5,9]. The best management of hydatid disease remains its prevention. It is based on individual prophylactic measures such as: avoidance of contact with dogs, systematic hand washing, especially in children, thorough washing of fruits and vegetables. Collective measures are also needed, such as improvement of the conditions of slaughter, the veterinary control of meat, especially in the rural areas and the control of stray dogs.

Conclusion

The hydatid pulmonary embolism is a rare and serious complication of hepatic hydatid disease. The diagnosis should however be brought up, especially in endemic area of this zoonosis. The diagnosis has become easier with chest imaging. The treatment is surgical ideally but often only medical treatment can be initiated given the multiplicity of lesions. The hydatid pulmonary embolism can cause serious acute life-threatening complications. It can also progress to chronic pulmonary heart and chronic respiratory insufficiency making its prognosis reserved. The prevention remains the best way to fight against this disease.

Bibliography

1. Budke CM., *et al*. "A systematic review of the literature oncystic echinococcosis frequency worldwide and Its associated clinical manifestations". *American Journal of Tropical Medicine and Hygiene* 88 (2013): 1011-1027.
2. Rada IO. "Ischemic syndrome of the lower limb produced by ahydatic cyst". *Journal of Cardiovascular Surgery* 27 (1986): 282-285.
3. S Maiouak., *et al*. "Pulmonary hydatid disease: report of three cases". *Revue de Pneumologie Clinique* 72.2 (2016): 152-155.
4. Morar R and Feldman C. "Pulmonary echinococcosis". *European Respiratory Journal* 21 (2003): 1069-1077.
5. Afif H., *et al*. "Hydatidose pulmonaire multiple en lâcher de ballon avec localisation cardiaque". *Revue des Maladies Respiratoires* 17 (2000): 697-698.
6. Aribas OK., *et al*. "Embolisation of hydatid cysts in the pulmonary artery presenting with haemoptysis". *Netherlands Journal of Medicine* 65 (2007).
7. M Serraj., *et al*. "Embolie pulmonaire d'origine hydatique: une complication rare du kyste hydatique du foie". *Revue des Maladies Respiratoires* 30.3 (2013): 215-221.
8. Ben M'Rad S., *et al*. "Embolie pulmonaire d'origine hydatique". *La Presse Médicale* 27 (1998): 205-207.
9. Talmoudi T., *et al*. "L'embolie pulmonaire hydatique. Étude de deux observations personnelles et revue de la littérature". *Annales de Chirurgie* 34 (1980): 245-250.
10. Menassa-Moussa L., *et al*. "Une hydatidose diagnostiquée à l'occasion d'une embolie pulmonaire". *Journal des Maladies Vasculaires* 34 (2009): 354-357.
11. Yagüe D., *et al*. "Bilateral hydatid cyst of pulmonary arteries: MR and CT findings". *European Radiology* 8 (1998): 1170-1172.
12. Karantanas AH., *et al*. "Echinococcus of the pulmonary artery: CT, MRI and MRA findings". *Computerized Medical Imaging and Graphics* 24 (2000): 265-267.
13. Ben Hamda K., *et al*. "Eighteen year experience with echinococcosis of the heart: clinical and echocardiographic features in 14 patients". *International Journal of Cardiology* 91 (2003): 145-151.
14. Lioulias A., *et al*. "Acute pulmonary embolism due to multiple hydatid cysts. Case report". *European Journal of Cardio-Thoracic Surgery* 20 (2001): 197-199.
15. Chemchik H., *et al*. "Hepatic hydatid cyst complicated of hydatid pulmonary embolism". *Annales Françaises d'Anesthésie et de Réanimation* 29 (2010): 658-666.
16. Buz S., *et al*. "Severe chronic pulmonary hypertension Caused by pulmonary embolism of hydatid cysts". *Annals of Thoracic Surgery* 84 (2007): 2108-2110.

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