Annals of Case Reports & Reviews

Case Report

doi: 10.39127/2574-5747/ACRR:1000344

LAASRI K, et al. Annal Cas Rep Rev: ACRR-344

An Unusual Cause of Pulmonary Embolism: Hydatid Cyst

Khadija LAASRI*, Meriem ZHIM, Salma MARRAKCHI, Ittimade NASSAR, Nabil MOATASSIM BILLAH

Radiology Department, Ibn Sina University Hospital, Mohammed V University, Rabat, Morocco

***Corresponding Author:** Khadija LAASRI, Radiology Department, Ibn Sina University Hospital, Mohammed V University, Rabat, Morocco. Email: Kadijalaasri93@gmail.com

Citation: LAASRI K, ZHIM M, MARRAKCHI S, NASSAR I, BILLAH NM (2023) An Unusual Cause of Pulmonary Embolism: Hydatid Cyst. Annal Cas Rep Rev: 344.

Received Date: January 06, 2022; **Accepted Date:** February 02, 2023; **Published Date:** February 07, 2023

Abstract

Hydatid pulmonary embolism is extremely rare but serious condition, due to rupture of a hydatid cyst in the venous circulation returning from the liver or rupture of an intracardiachydatid cyst of the right heart, or, more rarely, originating from a direct involvement of the pulmonary artery by a pulmonary hydatid cyst. The aim of this case is to report a rare entity of pulmonary embolism caused by a direct invasion of the pulmonary artery by a hydatid cyst in a 46-year-old woman with no history of hydatid cyst, and describe its appearance on the CT pulmonary angiography. To our knowledge, this is the second case of inaugural hydatid pulmonary arterial embolism found on CT scan establishing the diagnosis of the disease in a patient who had no other location of hydatid cyst. Therapeutic management is non-consensus and the prognosis is poor.

Keywords: *Pulmonary embolism, Hydatid cyst, Pulmonary artery, CT angiography*

Introduction

Hydatidosis is an infectious disease that is endemic in Morocco, and it is caused by the cestodeEchinococcusgranulosus. Although it can affect any part of the body, the liver (75%) and lungs (15%) are the most usually affected organs.Arterial localization of hydatidosis is extremely rare, with less than 2% of all hydatid cysts in the thorax (1, 2). The diagnosis is essentially based on imaging particularly, on computed tomography (CT) and magnetic resonance imaging (MRI), and the prognosis is poor. (3) Therapeutic management is non-consensus and the prognosis is poor (4, 5). We present a case of a rare entity of pulmonary embolism caused by a direct invasion of the pulmonary artery by a hydatid cyst in a 46-year-old woman with no history of hydatid cyst, and describe its appearance on the CT pulmonary angiography. To our knowledge, this is the second case of inaugural hydatid pulmonary arterial embolism found on CT scan establishing the diagnosis of the disease in a patient who had no other location of hydatid cyst (6).

Case report

A 46-year-old woman living in a rural area, without significant medical or surgical history, presented to our radiology unit for an evaluation of gradually worsening dyspnea. No predisposing factor responsible for thromboembolic disease was found. Physical examination was unremarkableand there was no deep vein thrombosis in the lower extremities. The electrocardiogram revealed sinus tachycardia with no axis deviation nor T wave or ST segment abnormalities. The D-dimer test was normal. CT pulmonary angiography was performed, revealing multiple cystic emboli in the segmental pulmonary arteries causing vessel enlargement (Figure 1). The results of a transthoracic echography revealed no evidence of cardiac cysts or pulmonary hypertension. Abdominal ultrasonography and CT scan were normal.Initial laboratory analysis was normal. The hydatid serology was positive.

The diagnosis of a pulmonary embolic cyst was made based on the clinical history, imaging findings, and the fact that hydatic cysts are still endemic in our country.

Due to the high risk for an anaphylactic reaction,pulmonary embolectomy was not decided as a therapeutic option. The patient was treated with albendazole. The patient became asymptomatic during the third month of the medical therapy, and a follow-up CT scan performed 10 months later shows a clear decrease in the majority of intra-arterial lesions (figure 2).

Figure:

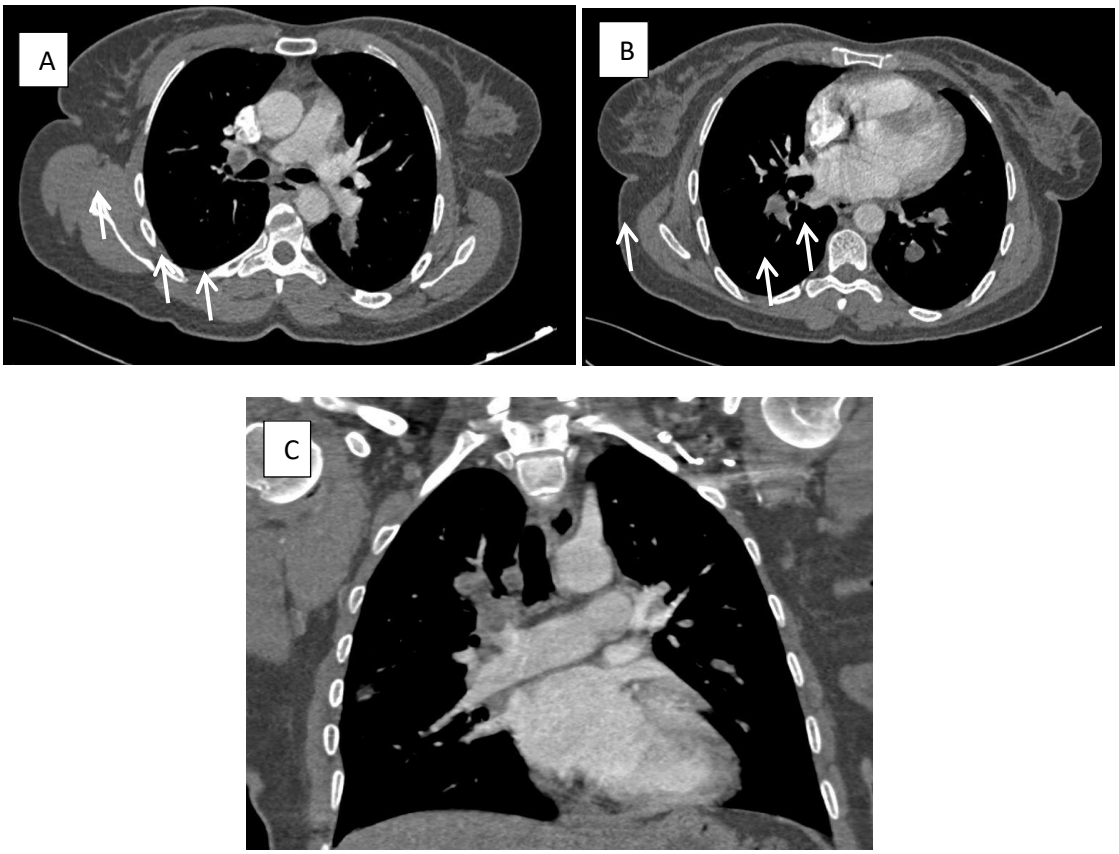

Figure 1: CT pulmonary angiography images (A, B: axial sections, C: coronal reformatted image), shows a bilateral enhancement defect of the segmental arteries with the presence of well-limited cystic lesions compatible with hydatid vesicles. (white arrows)

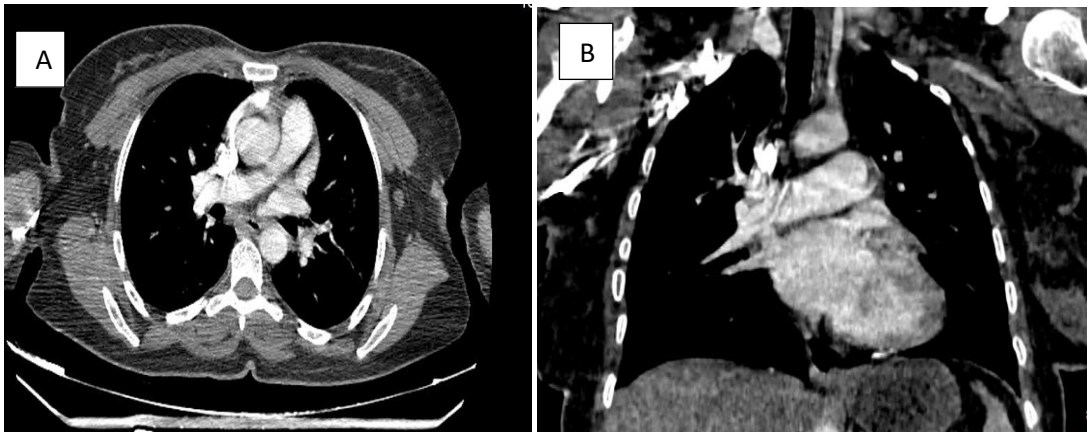

Figure 2: ControlCT pulmonaryangiography(A: axial sections, B: coronal reformatted image), after treatment shows a clear decrease in the majority of intra-arterial lesions.

Discussion

Hydatic disease is a zoonotic disease caused by the echinococcus parasite, that can affect any part of the human body, although the liver (50%-77%), and lungs (15%-47%) are the most commonly affected organs. Arterial localization of hydatidosis is extremely rare, with less than 2% of all hydatid cysts in the thorax (1, 2). It can occur either as a result of hepatic or abdominal cyst rupturing into the hepatic veins or the inferior vena cava (7), or directly from the ruptured cyst in the right cardiac chambers (2), or, more rarely, originating from a direct involvement of the pulmonary artery by a pulmonary hydatid cyst, as was the case in our observation.

Clinical manifestations of the hydatic pulmonary embolism are nonspecific and it can mimics that of thromboembolic disease. It may cause hemoptysis due to invasion of vascular structures such as bronchial and pulmonary arteries, chest pain, dyspnea, cough, and/or an anaphylactoid reaction (8). Hydatid pulmonary embolism has been classified based on the clinical presentation in three groups because of obstruction of pulmonary blood flow: acute fatal, subacute with pulmonary hypertension and death in less than one

year, or resulting in chronic pulmonary hypertension (2, 3). Our case differs from those previously described lacking any signs of pulmonary hypertension, and it seems possible that their pulmonary perfusion was maintained via the bronchial arteries (9).

The diagnosis is essentially based on imaging particularly, mainly CT angiography and magnetic resonance imaging (MRI). They shows the topography of hydatid cysts, the obstruction of pulmonary artery, its degree, and cardiac repercussion (4, 10). On CT angiography, uncomplicated intra-arterial KH presents as a well-defined mass of fluid density with a rim enhancement after contrast injection (4, 11, 12). On MRI the appearance is characteristic, it appears as a hypointense oval lesion in T1-weighted sequence and hyperintense in T2-weighted sequence, sometimes a hypointense peripheral rim on T2 corresponding to the pericyst, a dense fibrous reaction capsule is visualized; this aspect is considered characteristic of the parasitic nature (4,13,14).

Other causes of intraluminal filling defects, such as tumor emboli, bland pulmonary thromboemboli, and pulmonary artery sarcoma, should be considered in the differential diagnosis on CT (14). This entity can be distinguished from pulmonary thromboembolism and primary or secondary pulmonary arterial tumors by the cystic nature of the pulmonary artery filling deficiencies and the lack of enhancement (15).

After confirmation of the diagnosis of hydatid pulmonary embolism by imaging, the search for the emboligenic site is necessary(16). Abdominal ultrasound allows to localize the hydatid cysts and to analyze their anatomical relationships with the IVC and the hepatic veins. Transthoracic echocardiography allows to evaluate the impact on cardiac function, to look for abnormal shunts, and especially to eliminate a cardiac localization (17). Echocardiography is the investigative procedure of choice for studying cardiopericardialhydatidosis, and also allows to detect pulmonary hypertension. This usually prompts further evaluation with contrast-enhanced CT or pulmonary magnetic resonance (MR) angiography (14). Transesophageal echocardiography may detect massive emboli in the main pulmonary arteries (18, 19, 21).

Serologic tests are useful to confirm infection (14). There is no established therapeutic consensus (4, 10). Although surgery is the treatment of choice (20, 15), that consists in the removal of intra-arterial cysts when the patient's condition allows it, often only medical treatment can be instituted given the multiplicity of lesions or whene surgical intervention involves a high rate of morbidity or mortality (21). hence the importance of focusing on preventative treatment (2). In our patient, medical treatment resulted in a significant clinical and radiological improvement (17).

The prognosis of intra-arterial pulmonary hydatid cyst is poor given the risk of acute fatal complications such as anaphylactic shock and vascular rupture as well as of chronic progression to corpulmonale and respiratory failure (2).

Conclusion

Hydatid pulmonary embolism is rare but remains an etiology that needs to be considered and kept in mind, especially in endemic areas, as regards nonthrombotic embolism. CT angiography and/or MRI angiography can show pathognomonic features leading to a correct diagnosis.

References

1. Maiouak, S., Zaghba, N., Benjelloun, H., &Yassine, N. (2016). L'hydatidose artérielle pulmonaire : à propos de trois cas. Revue de Pneumologie Clinique, 72(2), 152–155. doi:10.1016/j.pneumo.2015.09.008
2. Serraj, M., Smahi, M., Kamaoui, I., El Houari, A., Sahnoune, F., Ouadnouni, Y., … Benjelloun, M. C. (2013). Embolie pulmonaire d'origine hydatique : une complication rare du kyste hydatique du foie. Revue Des Maladies Respiratoires, 30(3), 215–221. doi:10.1016/j.rmr.2012.06.004
3. Karima Bouytse, Bouchra Daher, JihaneAchrane, Mohammed Aharmim, Jamal-Eddine Bourkadi. Embolie pulmonaire hydatique: rare complication du kyste hydatique du poumon: à propos d´un cas. PAMJ - ClinicalMedicine. 2021;6:28. [doi: 10.11604/pamj-cm.2021.6.28.25637]
4. Serraj, M., Smahi, M., Kamaoui, I., El Houari, A., Sahnoune, F., Ouadnouni, Y., B. Amaraa , M. El Biazea , S. Tizniti, Benjelloun, M. C,. (2013). Embolie pulmonaire d'origine hydatique : une complication rare du kyste hydatique du foie. Revue Des Maladies Respiratoires, 30(3), 215–221. doi:10.1016/j.rmr.2012.06.004
5. Maiouak, S., Zaghba, N., Benjelloun, H., &Yassine, N. (2016). L'hydatidose artérielle pulmonaire : à propos de trois cas. Revue de Pneumologie Clinique, 72(2), 152–155. doi:10.1016/j.pneumo.2015.09.008
6. Menassa-Moussa, L., Braidy, C., Riachy, M., Tabet, G., Smayra, T., Haddad-Zebouni, S., …Aoun, N. (2009). Une hydatidose diagnostiquée à l'occasion d'une embolie pulmonaire. Journal Des Maladies Vasculaires, 34(5), 354–357. doi:10.1016/j.jmv.2009.04.004
7. El HadjSidi, C., Mgarrech, I., Alimi, F., &Tarmiz, A. (2020). Four pathways for hydatid pulmonary embolism. Journal of CardiacSurgery. doi:10.1111/jocs.14754
8. Akgun, V., Battal, B., Karaman, B., Ors, F., Deniz, O., &Daku, A. (2011). Pulmonary artery embolism due to a ruptured hepatic hydatid cyst: clinical and radiologic imaging findings. Emergency Radiology, 18(5), 437–439. doi:10.1007/s10140-011-0953-8
9. O. K. Aribas, F. Kanat, E. Turk-Aribas, I. Erayman, and T. Yuksek, "Embolisation of hydatid cysts in the pulmonary artery presenting with haemoptysis," The Netherlands Journal of Medicine, vol. 65, no. 3, pp. 109–111, 2007.
10. Zaimi, S., Bellamlih, H., &Nassar, I. (2020). Hydatid pulmonary embolism: An exceptional complication of hydatid disease. Radiology Case Reports, 15(5), 545–547. doi:10.1016/j.radcr.2020.01.015
11. Menassa-Moussa L, Braidy C, Riachy M, et al. Une hydatidose diagnostiquée à l'occasion d'une embolie pulmonaire. J Mal Vasc 2009; 34:354—7.
12. Yagüe D, Lozano MP, Lample C, et al. Bilateral hydatid cyst of pulmonary arteries: MR and CT findings. EurRadiol 1998; 8:1170—2

13. Karantanas AH, Bitsios G, Karaiskou E. Echinococcus of the pulmonary artery: CT, MRI and MRA findings. Comput Med Imaging Graph 2000;24:265—7.
14. Pena, E., Dennie, C., Franquet, T., &Milroy, C. (2012). Nonthrombotic Pulmonary Embolism: A Radiological Perspective. Seminars in Ultrasound, CT and MRI, 33(6), 522–534. doi:10.1053/j.sult.2012.05.002
15. Saeedan MB, Aljohani IM, Alghofaily KA, Loutfi S, Ghosh S. Thoracic hydatid disease: A radiologic review of unusual cases. World J Clin Cases 2020; 8(7): 1203-1212 [PMID: 32337194 DOI: 10.12998/wjcc.v8.i7.1203]
16. Bousnina, S., Racil, H., Maghraoui, O., Marniche, K., Ben Mrad, S., Ghedira, H., … Chabbou, A. (2005). L'embolie pulmonaire hydatique. Revue de Pneumologie Clinique, 61(1), 31–36. doi:10.1016/s0761-8417(05)84779-0
17. Boubbou, M., Zakraoui, M., Belhoussine, F. Z., Kamaoui, I., Maaroufi, M., Sqalli, N. H., &Tizniti, S. (2012). Embolie pulmonaire hydatique. Feuillets de Radiologie, 52(1), 2–6. doi:10.1016/j.frad.2011.12.002
18. Buz, S., Knosalla, C., Mulahasanovic, S., Meyer, R., &Hetzer, R. (2007). Severe Chronic Pulmonary Hypertension Caused by Pulmonary Embolism of Hydatid Cysts. The Annals of ThoracicSurgery, 84(6), 2108–2110. doi:10.1016/j.athoracsur.2007.04.
19. Lahdhili, H. (2002). Acute pulmonary embolism due to the rupture of a right ventricle hydatic cyst. European Journal of Cardio-Thoracic Surgery, 22(3), 462–464. doi:10.1016/s1010-7940(02)00360-3
20. Namn, Y., &Maldjian, P. D. (2013). Hydatid cyst embolization to the pulmonary artery: CT and MR features. Emergency Radiology, 20(6), 565–568. doi:10.1007/s10140-013-1130-z
21. Kardaras, F., Kardara, D., Tselikos, D., Tsoukas, A., Exadactylos, N., Anagnostopoulou, M., … Anthopoulos, L. (1996). Fifteen year surveillance of echinococcal heart disease from a referral hospital in Greece. EuropeanHeart Journal, 17(8), 1265–1270. doi:10.1093/oxfordjournals.eurhea