IDCases 26 (2021) e01251

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcr

Case report

A rare combination of cardiac and pulmonary cyst and review of the literature

Taamallah Karima[a,*], Slim Chenik[b], Wafa Fehri[a]

[a] Department of Cardiology, Military Hospital of Tunis, Tunisia
[b] Department of Cardiac Surgery, Military Hospital of Tunis, Tunisia

ARTICLE INFO

Article history:
Received 5 January 2021
Received in revised form 8 August 2021
Accepted 8 August 2021

Keywords:
Cardiac hydatid cyst
Pulmonary hydatid cyst
Cardiopulmonary by-pass
Hydatid cyst

ABSTRACT

Hydatid disease remains an important public health problem in endemic areas. We report a rare case of intramyocardial hydatid cyst of the right atrium along with a pulmonary hydatid cyst in a 16-year-old girl who was admitted to our hospital because of chest pain with recurrent episodes of fainting. One-stage surgery by median sternotomy under cardiopulmonary bypass was performed with excision of the hydatid cyst in the right atrium followed by the removal of the pulmonary hydatid cyst in the same session. Her postoperative recovery was uneventful. Based on this case, we emphasize, the rare combination of cardiac and pulmonary hydatid cyst. Another aspect that makes this case interesting is the location of the hydatid cyst at the right atrium, which is very rare. To the best of our knowledge, our case is the first to describe the combination of a hydatid cyst of the right atrium and the lung.

Background

Hydatidosis is a human parasitic disease caused by larval stages of Echinococcus granulosus [1], it is a significant health problem worldwide, and endemic in countries where farm animals are raised. The liver and lungs are the most frequently involved organs [2]. Cardiac hydatid cyst is uncommon, it may present asymptomatic for years or experience minor symptoms, but potentially fatal disease [3]. The combination of cardiac and pulmonary hydatid cyst is exceptional. Early surgery is the treatment of choice of cardiac hydatid cyst even in asymptomatic patients [4].

Hence, we report a case of a young girl with cardiac and pulmonary hydatid cysts, which were surgically removed in a single-stage surgical session. The two particularities of this case are the combination of both intracardiac and pulmonary hydatid disease and the location of the cardiac cyst at the right atrium which is very rare. To the best of our knowledge, our case is the first to describe the combination of a hydatid cyst of the right atrium and the lung.

Case report

A 16-year-old young girl, without previous medical history, was admitted to our hospital with chest pain, fainting, breathlessness, and low-grade fever in the last three months. Physical examination and electrocardiogram were normal. The chest radiography showed a well-defined lesion in the right lower zone of the lung, and cardiomegaly (Fig. 1-A). Microcytic hypochromic anemia with hemoglobin level at 9 g/dl and a high CRP level at 26 mg/l were noted. Transthoracic echocardiography showed a large, well-defined intramyocardial, multicystic heterogeneous mass, with a well-contrasted capsule, measuring 3.7 × 3.4 cm located at the lateral wall of the right atrium, suggestive of hydatid cyst. This mass reaches the anterior tricuspid valvular and caused deformation of the tricuspid annular with a moderate tricuspid insufficiency (Fig. 1-B,C). A serology for Echinococcus granulosus, with enzyme-linked immunosorbent assay (ELISA) was positive for echinococcal infection. In the search for another organ involvement, a complete radiological assessment was carried out. Ultrasonography of the abdomen ruled out hydatid disease of the liver. The chest computed tomography (CT) revealed a cystic lesion with an air-fluid level, suggesting a hydatid cyst, measuring 4.6 × 3.4 × 3 cm in the subpleural region of the right lower lobe of the lung and an intramyocardial mass located at the right atrium, these results were confirmed by Cardiac magnetic resonance imaging (MRI). Albendazole was started and the patient was referred to a cardiac surgeon for resection of the cardiac and the pulmonary cysts. She underwent a single-stage surgical session, via a median sternotomy under cardiopulmonary bypass (CPB) (Fig. 1-E). Histopathological and microbiological examinations were consistent for Echinococcus granulosus. The patient's postoperative recovery was uneventful, A transthoracic

* Corresponding author.
E-mail address: karima.taamallah@fmt.utm.tn (T. Karima).

http://dx.doi.org/10.1016/j.idcr.2021.e01251

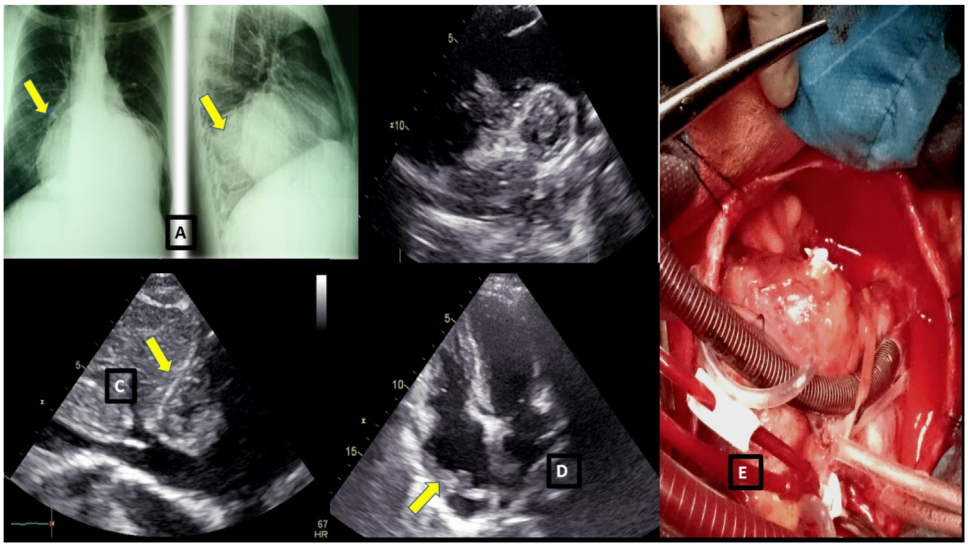

Fig. 1. A: Chest radiography view showing a well-defined lesion in the right lower zone of the lung, and a cardiomegaly. **B**: Transthoracic echocardiography: Well-defined intramyocardial, multicystic heterogeneous mass, with a well-contrasted capsule, measuring 3.7 × 3.4 cm localized at to the lateral wall of the right atrium. This mass reaches the anterior tricuspid valvular. **C:** The mass (arrow) extends to the roof of the right atrium and exerts no compressive effect on the inferior venae cavae. **D**: Transthoracic echocardiograpphy performed in post-operatively, apical 4-chamber view, persistence at the level of the roof of the right atrium of a hyperechoic image (blue arrow) corresponding to the pericardial patch reconstruction of the right atrium, pericardial effusion at the right atrium. E: Intraoperative photograph of intramyocardial right atrium hydatid cyst.

echocardiography was performed post-operatively to control results of surgery, which showed vacuity of the right atrium (Fig. 1-D). The patient was discharged on Albendazole therapy for five years. The patient was asymptomatic without recurrence of hydatid cyst in the four years follow-up period.

Discussion

Hydatidosis is a zoonosis transmitted from domestic and wild members of the canine family as definitive hosts and a variety of wild and domestic animals, usually sheep, as intermediate hosts. Human beings are only incidental intermediate hosts [2].

The liver (70 %) and lungs (25 %) are the most affected organs. Hydatid cyst of the heart is an uncommon lesion seen only in 0.2–2 of the cases [1]. The common location of cardiac hydatid cysts is the left ventricle followed by the right ventricles, hydatid cyst of the right atrium is very rare seen only in 4% of the cases [1], which is a particularity of our case. The association between cardiac and lung hydatid cyst is also extremely rare, very few cases were published reporting this combination [1,4–8]. Table 1 summarizes the clinical and paraclinical particularities of the reported cases. The diagnosis of hydatidosis is difficult because of the long latency between infection and manifestations of the disease. Clinical presentation of cardiac hydatid cyst is variable. The usual

Table 1
Clinical, paraclinical, and treatment of patients reported in the literature.

Authors/year/country	Age/ gender	Clinical presentation	Cardiac location	Lung location	Other location	Surgical treatment	Follow up
Atalay et al. [4] Turkey	48 /M	Cough Dyspnea chest pain.	Right ventricle	bilateral pulmonary hydatic cysts	no	Same session excision of lung and heart cyst	–
Kaskar et al. [7] India	14 /F	Dyspnea Syncope 7months[a]	Right ventricle	Right lower lobe	Extension to the pulmonary artery	Same session excision of lung and heart cyst	3 months uneventful
Khattabi et al. [6] Morocco	21/M	Hemoptysis 2years[a]	Right ventricle (Infundibular pulmonary)	multiple bilateral cysts of different sizes	Extension to the pulmonary artery	Excision of the cardiac cyst Lung cyst: Medical treatment	9 months uneventful
Martín-Izquierdo and Martín-Trenor [8] Spain	12/M	Chest pain Caught Hemoptysis fever	Right ventricle	multiple bilateral cysts of different sizes		excision of lung and heart cyst separately (6 months)	–
Seth et al. [1] India	38/F	hemoptysis chest pain, breathlessness low- grade fever	Apex of the ventricle	Right lower lobe		Same session excision of lung and heart cyst	Eight months uneventful
Yilmazer et al. [2] Turkey	5years/ M	cough and chest pain 2 months[a]	Left ventricle (posterior wall)	Right middle, upper and left upper lobe	Liver spleen	No available information	–
Our case	16/F	Chest pain, fainting, fever, dyspnea 3months	Right atrium	Right lower lobe		Same session excision of lung and heart cyst	Four years uneventful

M: male, F: Female.
[a] Interval time between the onset of the symptoms and the diagnosis of cardiac hydatid cyst.

manifestations reported are dyspnea, chest pain, palpitations, and syncopal episodes [2]. Hydatid cysts of the heart can result in serious cardiovascular complications which can be lethal, they must be operated on as soon as the diagnosis is established, even in asymptomatic patients, to avoid these complications [9]. Pulmonary hydatid disease affects the right lung in around 60 % of cases, 30 % exhibit multiple pulmonary cysts, 20 % bilateral cysts and 60 % are located in lower lobes [10] which is the case of our patient. Most lung cysts are incidentally diagnosed on chest radiographs [10,11]. Our patient had hydatid disease of the lung and right atrium along, making this case, the first presentation of this combination.

Echocardiography remains the most appropriate non-invasiveness diagnostic tool. It could detect the location of the cyst and its relation to cardiac and vascular structures and give an accurate diagnosis of cardiac echinococcosis [12]. In our case, echocardiography was found to be sufficient for the diagnosis of cardiac echinococcosis, which also supported recent studies reporting that echocardiography was sufficient in 94 % of the cases with cardiac involvement [2], computed tomography and magnetic resonance imaging are other valuable diagnostic tools, MRI can give precious knowledge on both the lesion morphology and its relation to other cardiac and extracardiac structures and thereby can guide the surgery [5].

Serological tests are positive in only about 50 % of patients and thus limited diagnostic accuracy [1]. In our case, the ELISA test was positive which reinforced the diagnosis. In our case, CPB was used to remove the cardiac hydatid cyst, it is required for the removal of the cyst, and recommended if there is no safe way [4]. It is important to consider the location, number, and size of the cysts when choosing the operative approach and deciding whether to use CPB or to perform surgery on the beating heart [4,13]. The operative management for concomitant pulmonary hydatid cyst can be done in the same setting or a different setting. In our case, it was done in the same setting via a median sternotomy. According to some authors, one-stage operation compared to a two-stage approach via a median sternotomy have a lot of advantages in the case of cardiac and lung cyst hydatid [4]. Finally, patients should take 400 mg of albendazole or mebendazole twice a day for five years, after surgery to prevent recurrence and the drawbacks of additional surgery, which was conducted in our patient [13].

Conclusion

A cardiac hydatid cyst associated with a pulmonary hydatid cyst is a rare entity. Radiological investigations, especially echocardiography and CT scan are the mainstay of diagnosis. One stage surgery remains to be the treatment modality of choice.

Declaration of Competing Interest

The authors report no declarations of interest.

Funding

There was no funding for the presented work.

Ethics approval and consent to participate

This case report was approved by the ethics committee of the Military hospital, Tunis, Tunisia. Patient gave written consent to use his clinical and paraclinical information to prepare this article. The patient gave permission to publish her case, as well as relevant related workup and diagnostic images, presented in the medical literature.

Authors contribution

Taamallah Karima: Manuscript redaction.
Wafa Fehri: Final manuscript approval.
Slim Chenik: Final manuscript approval Conception, he is the surgeon who operate on the
Patient.

References

[1] Seth HS, Mishra P, Khandekar JV, Raut C, Mohapatra CKR, Ammannaya GKK. A concomitant intramyocardial and pulmonary hydatid cyst: a rare case report. Braz J Cardiovasc Surg 2017;32:138–40, doi:http://dx.doi.org/10.21470/1678-9741-2016-0046.
[2] Yilmazer MM, Devrim I, Tavli V. Cardiac hydatid cyst associated with multiple organ involvement. Acta Cardiol 2011;66:87–8, doi:http://dx.doi.org/10.1080/ac.66.1.2064973.
[3] Daş T, Özer M, Yaşmur G, Yildirim M, Özgün A, Demirel H. Hydatid disease involved in the heart, liver, and kidney that caused sudden death: case report. Am J Forensic Med Pathol 2015;36:265–7, doi:http://dx.doi.org/10.1097/PAF.0000000000000200.
[4] Atalay A, Salih OK, Gezer S, Göçen U, Yaliniz H, Keklik V, et al. Simultaneous heart and bilateral lung hydatid cyst operated in a single session. Heart Lung Circ 2013;22:682–4, doi:http://dx.doi.org/10.1016/j.hlc.2012.11.014.
[5] Yılmaz R, Akpınar YE, Bayramoglu Z, Ozyavuz HI, Unal OF, Dursun M. Magnetic resonance imaging characteristics of cardiac hydatid cyst. Clin Imaging 2018;51:202–8, doi:http://dx.doi.org/10.1016/j.clinimag.2018.05.016.
[6] El Khattabi W, Afif H, Berrada Z, Rhissassi J, Aichane A, Bouayad Z. Hydatidose pulmonaire multiple avec localisation cardiovasculaire. Rev Mal Respir 2011;28:686–90, doi:http://dx.doi.org/10.1016/j.rmr.2010.10.037.
[7] Kaskar A, Shetty V, Shetty D. Chronic pulmonary thromboembolism due to intracardiac and pulmonary hydatidosis. Asian Cardiovasc Thorac Ann 2020;28:610–2, doi:http://dx.doi.org/10.1177/0218492320957918.
[8] Martín-Izquierdo M, Martín-Trenor A. Hidatidosis simulando un tumor cardíaco con metástasis pulmonares. Cir Cir 2016;84:318–23, doi:http://dx.doi.org/10.1016/j.circir.2015.03.003.
[9] Tuncer E, Tas SG, Mataraci I, Tuncer A, Donmez AA, Aksut M, et al. Surgical treatment of cardiac hydatid disease in 13 patients. Tex Heart Inst J 2010;37:189–93.
[10] Morar R, Feldman C. Pulmonary echinococcosis. Eur Respir J 2003;21:1069–77, doi:http://dx.doi.org/10.1183/09031936.03.00108403.
[11] Rawat S, Kumar R, Raja JSR, SK T. Pulmonary hydatid cyst: review of literature. Fam Med Prim Care Rev 2019;8:2774–8.
[12] Prati G, Gatti G, Belgrano M, Pinamonti B, Rauber E, Gripshi F, et al. Disseminated echinococcosis: follow your heart. J Cardiovasc Med 2016;17:e146–8, doi:http://dx.doi.org/10.2459/JCM.0000000000000389.
[13] Noaman H, Rawaf S, Majeed A, Salmasi AM. Hydatid cyst of the heart. Angiology 2017;68:765–8, doi:http://dx.doi.org/10.1177/0003319717690093.