Case report

Uncommon Infection of Pericardial Hydatid Cyst Imitating Cardiac rhabdomyosarcoma: A Challenging Diagnosis and the Role of Nuclear Magnetic Resonance Imaging

Abstract

Cardiac hydatid cyst (HC) is a rare localisation of hydatidosis**.** The main preoperative diagnostic tools are serology, Transthoracic Echocardiography (TTE),Cardiac Tomography (CT) and Nuclear Magnetic Resonance Imaging (MRI)**.**MRI is the key examination for diagnosing cardiac masses. It enables anatomical relationships to be studied and a reliable etiological orientation to be made. We report a challenging case of pericardial hydatid cyst associated to a rare infectionto *Brevundimonasnasdae*. In our case, the HC presented as a cardiac masse, with an atypical appearance on echocardiography suggestive of a rabdomyoscracoma. The MRI rectified the diagnosis, which was subsequently confirmedon operative finding, and on anatomo-pathological examination**.**

Keywords: Cardiac Hydatid Cyst, Nuclear Magnetic Resonance Imaging,*Brevundimonasnasdae*

INTRODUCTION

The cardiac hydatid cyst is a rare manifestation of Echinococcosis infection. The main characteristics of hydatid cyst of the heart are clinical polymorphism, clinical latency and severity of complications. Moreover, those complications are often the revealing mode of this condition, and can be life-threatening. The most common localization is the myocardium of the left ventricle.In addition, other locations may be found, and we cite the following in order of frequency, the right ventricle, the left and right atrium, the pericardium, the interventricular septum, and pulmonary artery.

To date, The Bacterial coinfection of cardiac hydatid cyst has not been reported in the literature.

In addition, the Infections caused by *Brevundimonas* species are rare in humans and are mainly nosocomial bacteremia in immunocompromised patients. [1]There are very few cases of *Brevidimonas*bacteriesinfections and furthermore, the isolation of *BrevidimonasNasadae* in cardiac pericardial fluid or hydatid cyst has never been described.

Here we describe the first clinical case of the co-infection of a Pericardial Hydatid Cyst *BrevidimonasNasadae*, and we strongly support the role of MRI in this challenging diagnosis.

CASE REPORT

A 25 years old, Moroccan man, with a history of liversurgery of hydatid cyst 2 yearsearlier, and dyspnea evolving six months before his admission. He was referred to the emergencydepartment for an acuteatypical chest pain associated to a NYHA stage II dyspnea,fever and a preserved general condition. The initial examination finds a conscious patient with stable hemodynamic and respiratory parameters, body temperatureat 38°C, normal cardiac and respiratory auscultation,and clinical examination of the abdomen revealed no significant abnormalities.

The Electrocardiogram (EKG) findsa regular sinus rhythm and inverted T waves in septo-apico -lateral and inferior leads**.(Figure 1)**

TEE showsan echogenic and heterogenous mass measuring 67x 37 mm on the apex of the heart covering the apical parts of the ventricules and seems to be having a doubtful lineary limit (red arrows) with the other parts of the endomyocardiumparticularly the septum evocating a rhabdomyosarcoma. There was no mitro-aortic valve disease,no vegetation images or pericardial effusion. **(Figure 2)**

We realized thoraco-abdominal Computed tomography (CT) with cardiac protocol, to enhance theetiological diagnosis through density analysis and to assess the mass extension. CT rules out the diagnosis of rhabdomyosarcoma and retained the diagnosis of an isolated cardiac hydatid cyst without other signs of hepatic recurrence,and absence of malignancysigns: no distant metastasis, no mediastinal invasion, no rapid growth, or hemorrhagicpericardial effusion.

These conflicting findings forced us to perform aMRI for better characterization. Cardiac MRI revealed aPolylobed, compartmented cystic mass in favor of a pericardial hydatid cyst, with scalloping of cardiac muscle (interventricular septum and right ventricular wall).**(Figure 3)**

Hydatid serology was positive.The routine laboratory was normal especially Blood count, C-reactive protein (CRP), and Liver function tests.

The therapeutic management was based on a combinedsurgical and medical strategy.

The operating procedure revealed the presence of a multi-lobed mass in front of the apex of the right and left ventricles. A direct opening of the mass was done with discharge of a greenish liquid after aspiration and washing with physiological serum, capitonnagethen closure of the neo cavity.**(Figure 4)**

The histology findings confirmed the diagnosis of hydatid cyst. Microbial culture of the aspiration fluid revealed the presence of a non-fermentative gram-negative bacterium called*Brevundimonasnasdae*, which was resistant to cephtazidime and sensitive to meropenem.

Paralelly, medical treatment was started and the patient was treated with Albendazole15 mg /kg /day divided into 2 doses during meals for 6 months with 15-day windows every two months and prednisolone 1mg/Kg for one week. Post-operatively, the patient was treated with albendazole and meropenem to treat the co-existing bacterial infection.

The post-operative follow up reveals no complications and our patient was discharged.

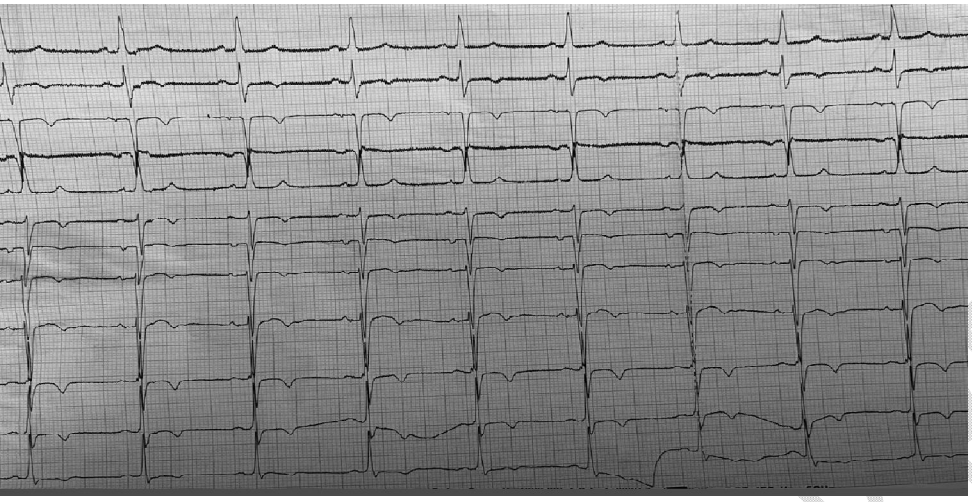

Figure 1: EKG showing a regular sinus rhythm and inverted T waves in septo-apico -lateral and inferior leads

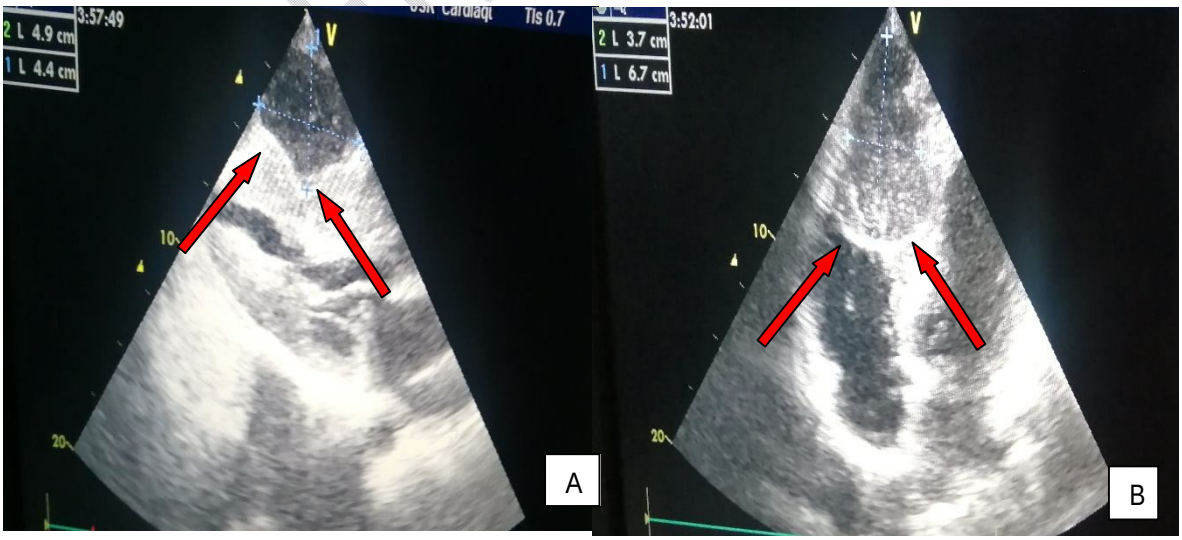

Figure 2: TTEs showing an echogenic mass in the apical part of the interventricular septum covering the apex of the ventricles with pericardial extension as visualised at A) the apical 4-chamber view and B) parasternal long-axis section centered on the right cavities

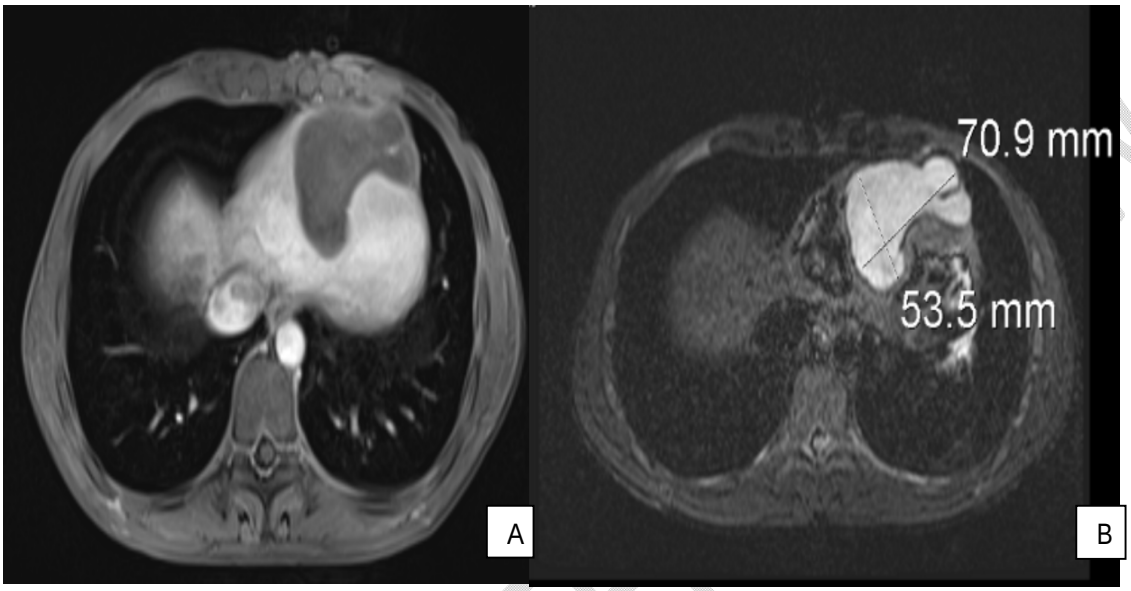

Figure 3: MRI showing a well-circumscribed polylobed pericardial cystic mass with liquid signal in T1 hyposignal (A) and hyper signal T2(B), segmented with discrete contrast of the wall and the septum.

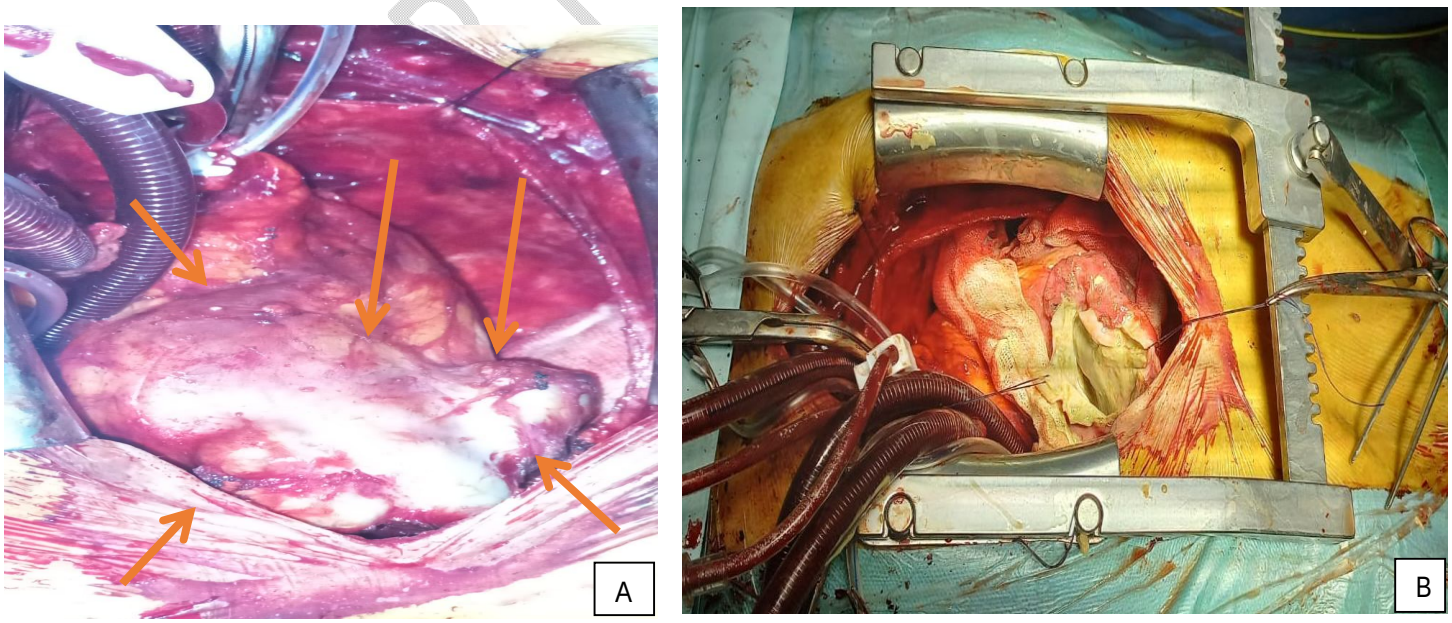

Figure 4: Surgical findings as visualized showing :(A) Multi-lobed mass in front of the apex of the right and left ventricles encapsulated and distinct from surrounding structures as indicated by the orange

arrows. (B): A direct opening of the mass.

DISCUSSION

Cardiac hydatidosis is one of the rarest localizations, even in endemic areas. Out of all visceral localizations, the incidence of cardiac hydatic cyst is the lowest, ranging from 0.5% to 2% [2]. Myocardial involvement of the left ventricle is the most frequent, and accounts for 55 to 60% of all cardiac localizations. While that of the right ventricle is found in 15% of cases,the left atrium 8%, the right atrium 3 to 4%, and the interventricular septum 7 to 9% [3]. The pericardial localization, as in our case, is exceptional and always considered secondary[4].

The clinical presentation of cardiac hydatid cyst is highly nonspecific.The exposure to dogs and sheep, the endemic context or personal history of other hydatid localizations should evoke the diagnosis.

Trans-thoracic echocardiography is the first-linenon-invasiveimaging examination. TTE allows the cardiologist to approach characteristics of a cardiac mass, describing the topography, the echogenic abnormalities and its hemodynamic impact. Thus, in the context of a hydatid cyst: a hypoechoic lesion transcribing a fluid nature with a fine parietal wall suggestive of a cystic lesion. [5] The image of membrane detachment or small vesicles is strongly suggestive of the diagnosis, but is rarely observed.

As in our case, this exam method may fail to identify the nature of the cardiac mass due to several factors: the specificity and sensitivity of this imaging method compared to computed tomography and magnetic resoning, In addition to other limitations, including operator dependency, a restricted field of view in heavy-bodied patients and limited views of the left ventricular apex and right heart chambers.[6]

The ESC 2022 guidelines suggest that computed tomography is a useful diagnostic tool,and magnetic resonance is considered as an imaging method of choice to distinguish cysts from solid tumors among other diagnosis: thrombus, vegetations, etc.

Hydatid cysts generally appearsin Computed Tomography (CT) as masse of liquid density, well defined, rounded, thin-walled and unaffected by contrast. It is usually univesicular, and exceptionally multivesicular. Well-visualized parietal calcifications are inconstant but suggestive.[7,8]The radiological appearance depends on the hydatid cyst stage and according to Gharbi's classification, it represents stage IV.

Moreover, CT contributes to the assessment of disease extension through thoraco-abdominal acquisition in search of multivisceral localization.

MRI ismore specificand gives detailed morphology signs of the cardiac hydatid cystand its relation to other cardiac and extra cardiac structures. Hydatic cyst usually appears as an oval or spherical lesion that is hypo intense on T1-weighted images, and with a signal intensity superior or equal to cerebrospinal fluid on T2-weighted images. In addition, the presence of peripheral contrast is crucial to distinguish HC from other cardiac masses.[9]

Another particularity of our case is the co-infection of hydatid cyst and a *Brevundimonasnasdae*. To our knowledge the occurrence of this association is unusual and has not been previously reported.

Previously serious infections with *Brevundimonasspp* especially *Brevundimonasvesicularis* and *Brevundimonasdiminuta* with include four instances of septicaemia (8%), two of endocarditis (4%), one of septic arthritis (2%) and one of meningitis (2%). Other conditions include instances of two cases of tonsillitis (2%), two of liver abscess (2%) and two of botryomycosis (2%).Reports of cases of co-infection with *Brevundimonasspp* and other bacteria were rare [10]. To our knowledge, there has been no cardiac or pericardial infection or coinfection with the hydatid cyst described.

Since there are no controlled trials of antimicrobial therapy for *Brevundimonas*agents especially *Brevundimonasnasdae* infections in humans, treatment must be guided by the results of in vitro susceptibility testing of isolates. Cephalosporins, penicillins, carbapenems or aminoglycoside antibiotics were given to treatpatients and these were mostly successful.[10]

Conclusion

The co-infection of Hydatic Cyst is an usual and to our knowledge we report the first case in literature with this association. The choice of anti-microbial therapy treating both infection was challenging. The morality of our case is to demonstrate the important role of MRI in the diagnostic process of cardiac mass and to reduce the mortality by raising the alarm: “simple hydatic cyst case infecting the heart is a rare entity but the co-infection is possible and might be lethal”.

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