Case report

An extremely rare case of Cardiac Hydatid Cyst Co-infected with Brevundimonas nasdae: Diagnostic Challenges and Management"

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 *Brevundimonas nasdae*clinically revealed by chronic dyspnea and atypical acute chest pain evolving in a febrile context.In our case, the HC presented as a cardiac masse, with an atypical appearance on echocardiography suggestive of a rhabdomyosarcoma. The MRI rectified the diagnosis, which was subsequently confirmedon operative finding, and on anatomo-pathological examination**.** The Patient was successfully managed after multidisciplinary treatment including combinedsurgical, antibiotics and antiparasitic treatments.

Keywords: Cardiac Hydatid Cyst, Nuclear Magnetic Resonance Imaging,*Brevundimonas nasdae*

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 Bacterialco-infection 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 Brevundimonas bacterial infectionsand furthermore, the isolation of Brevundimonas *Nasadae* 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 withBrevundimonas *Nasadae*, and we strongly support the role of MRI in this challenging diagnosis.

CASE REPORT

Clinical Presentation

A 25 years old, Moroccan man, with a history of liversurgery of hydatid cyst two years previously, and dyspnea evolving six months before his admission. He was referred to the emergencydepartment for an acuteatypical chest pain associated with***NYHA*** stage II dyspnea,fever and a preserved general condition.

Diagnostic Evaluation

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 endomyocardium particularly 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 especiallyhigh sensitivity troponins,Complete Blood Count, C-reactive protein (CRP), and Liver function tests.**(Figure 4)**

Therapeutic management involved a combination of surgical and medical strategies.

Surgical Management

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 5)**

Medical management

Paralely,Medical treatment was started and the patient was treated with Albendazole 15 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.

Postoperative Course

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*Brevundimonas nasdae*, which was resistant to ceftazidime and sensitive to meropenem.

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.

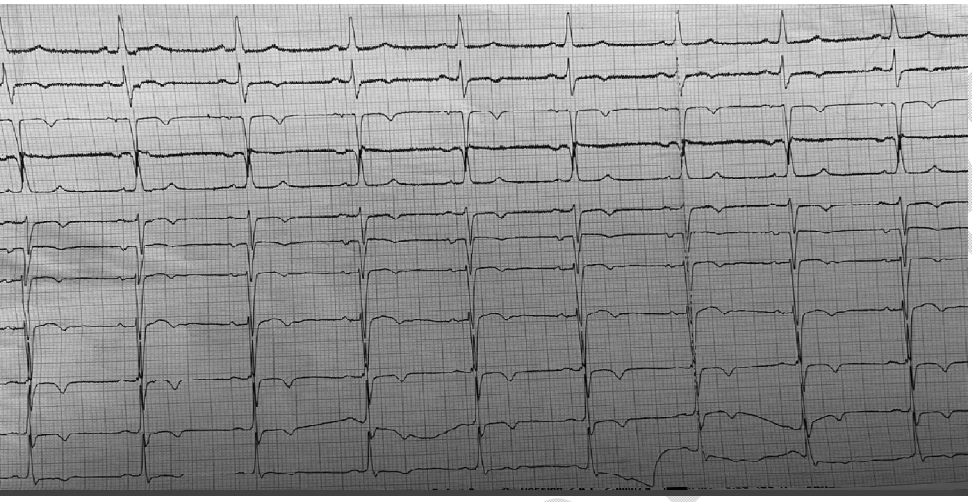

Figure 1: EKG showing a regular sinus rhythm and inverted T waves in septo-apico -lateral and inferior leads

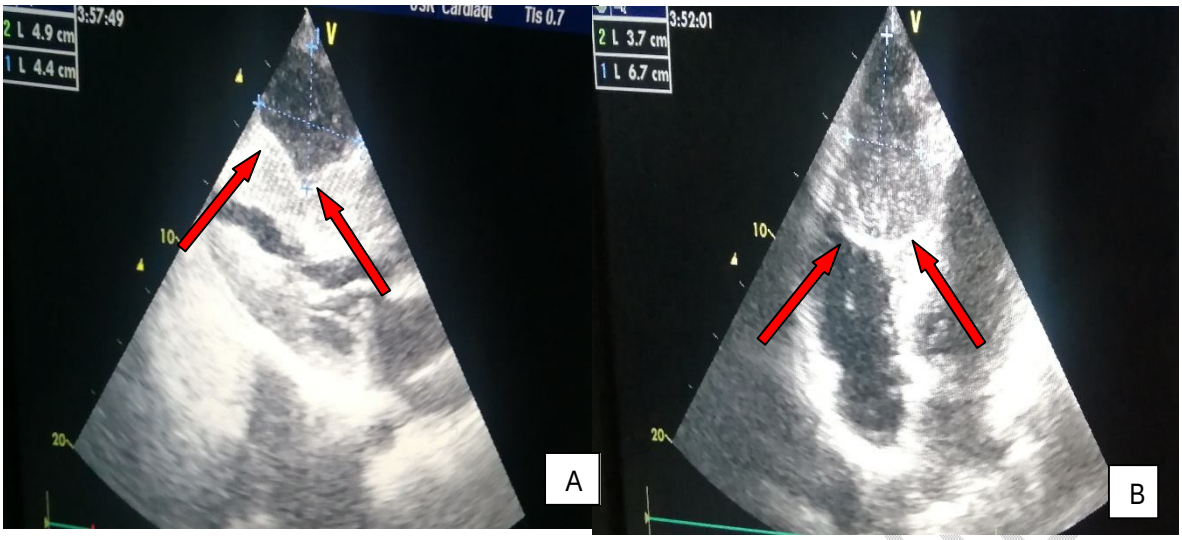

Figure 2: TTEs showing an echogenic mass in the apical part of the interventricular septum covering the apex of the ventricles with pericardial extension as visualised at A) the apical 4-chamber view and B) parasternal long-axis section centered on the right cavities

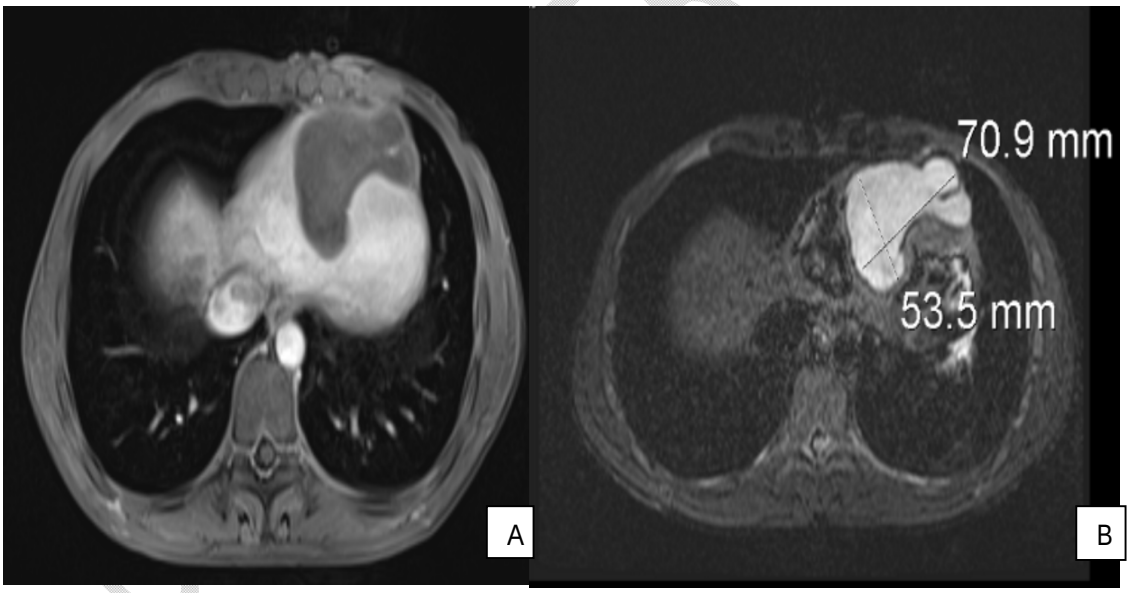

Figure 3: MRI showing a well-circumscribed polylobed pericardial cystic mass with liquid signal in T1 hyposignal (A) and hyper signal T2(B), segmented with discrete contrast of the wall and the septum.

Biological analysis	**Values**

Complete Blood Count (CBC)	Hemoglobin (Hb): 13g/dL White Blood Cells (WBCs): 9000 cells/µL Platelets (Plt): 150,000 platelets/µL Eosinophils: 5 cells/µL
Hydatid cyst serology by ELISA	IgG: 2 OD units(Positive)
CRP (C-reactive protein)	CRP: 5mg/L
Troponin T (TnT)	TnT:0.01 ng/ml (normal <0.04ng/ml)
Hepatic tests	Alanine Aminotransferase(ALT): 7 U/L Aspartate Aminotransferase (AST): 10 U/L Alkaline Phosphatase (ALP): 44 U/L

Figure 4: Summary Table of the Biological diagnostic tools.

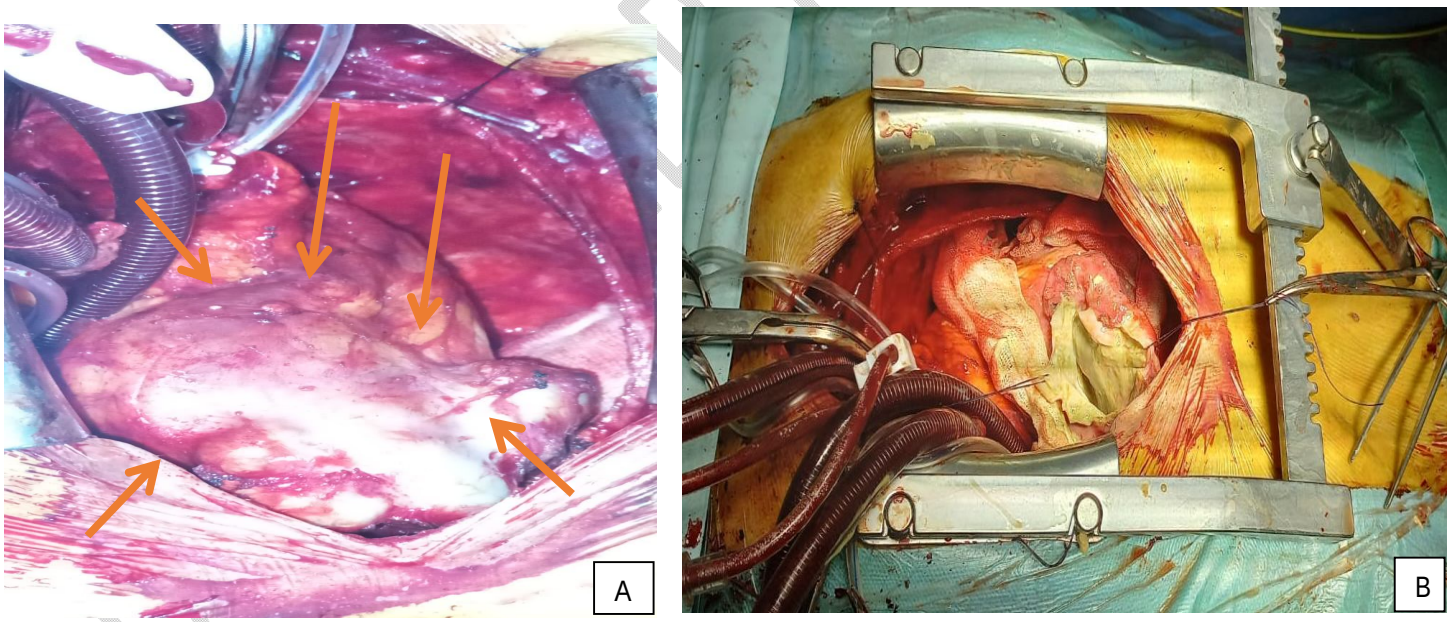

Figure 5: Surgical findings as visualized showing :(A) Multi-lobed mass in front of the apex of the right and left ventricles encapsulated and distinct from surrounding structures as indicated by the orange arrows. (B): A direct opening of the mass.

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.

While MRI clearly demonstrates the exact location of the cyst and its relationship with adjacent structures. On T1-weighted and T2-weighted MR images, a hydatid cyst is typically observed as a hypointense and hyperintense lesion, respectively. On T2-weighted images, the pericyst also appears as a low-signal intensity. Other specific indications that may help in the diagnosis of cardiac hydatidosis include the existence of daughter cysts and membrane detachment.[9,10]

According to the Adapted from of the 2010 World Health Organization Informal Working Group on Echinococcosis (WHO-IWGE) International classification of ultrasound images incystic echinococcosis (CE) and the WHO-IWGE Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans the cyst Type of our case is CE4; and according to the expert consensus, only simple surveillance is recommended. [11, 12]

However, given the cardiac location, management is different. Due to the high risk of associated complications, cardiac hydatid cysts should be removed surgically, even in asymptomatic patients.

During the operation, measures should be taken to prevent perioperative embolization of a germinative membrane. Surgical excision under cardiopulmonary bypass is the treatment of choice.[13]

Surgical R0 resection should be followed by a chemotherapy with albendazole (10–15 mg/kg body weight per day) or mebendazole (40–50 mg/kg body weight per day).[12]

A review of the literature studying the clinical and microbiological features of superinfected hydatid cysts found 37 cases out of 503 patients with hydatid cysts. The Gram-negative bacilli reported was Escherichia coli, Acinetobacter baumannii,Aeromonas hydrophila , Klebsiella oxytoca and Pseudomonas aeruginosa . They represented 21% of superinfections, mainly in hepatic hydatid cysts. [14]

Another particularity of our case is the co-infection of hydatid cyst with *Brevundimonas nasdae* as Gram –negative bacilli.To our knowledge, the occurrence of this association is unusual and has not been previously reported.

Previously serious infections with *Brevundimonas spp* especially *Brevundimonas vesicularis* and *Brevundimonas diminuta*~~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 *Brevundimonas spp* and other bacteria were rare [15]. As far as we know,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 *Brevundimonas nasdae* 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.[15]

Conclusion

The co-infection of Hydatic Cyst with Brevundimonas nasdaeis unusual and to our knowledge, we report the first case in literature with this association. The morality of our case is to demonstrate the important role of MRI in the diagnostic process of cardiac masse and the choice of anti-microbial therapy treating both infection was challenging.In short,to reduce the mortality we must sound the alarm:“simple hydatic cyst case infecting the heart is a rare entity but the co-infection with rare bacteriais possible and might be lethal”.

Disclaimer (Artificial intelligence)

Option 1:

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc.) and text-to-image generators have been used during the writing or editing of this manuscript.

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