



Unusual Clinical Presentation of a Giant Left Ventricle Hydatid Cyst

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ABSTRACT

A 39-year-old woman was hospitalized in our center due to chest and left shoulder pain. Having a history of tamponade and tuberculosis, she was under treatment for the previous two months. Echocardiography, chest CT and MRI documented intramyocardial and pericardial hydatid cyst which was later confirmed by further pathological studies. Later, the cyst was removed surgically.

Learning objective

Perforated hydatid cyst can be presented as granulomatous pericarditis. Hydatid cyst should be taken into consideration in the differential diagnosis of all cystic masses in all anatomic locations, especially with rare localizations and when they occur in endemic areas.

Introduction

Hydatid disease (HD) may affect almost any part of the body. However, the involved organs mostly include the liver (75%) and the lungs (15%).¹ Hydatid cysts rarely present in the pericardial cavity. Among radiographic techniques, CT imaging and MRI can acceptably detect hydatid cysts.^{2,3} Yet, hydatid cysts with unusual localizations may cause serious difficulties in differentiating the diagnosis. Therefore, in this study we report a case with misdiagnosed pericardial hydatid cyst.

Case report

The patient, a 39-year-old woman, was admitted to our center with intermittent retrosternal chest discomfort, dyspnea of exertion (DOE) and pain in the left shoulder from 3 days ago. With a history of tamponade during her pregnancy period, she had previously undergone subxiphoid drainage followed by a pericardiectomy procedure due to her persistent DOE in which there was no evidence of pericardial cyst; however the histopathologic findings of the pericardial sample reported granulomatous changes. Therefore, an empirical therapy of anti-

tuberculosis had been initiated for 2 months. Despite the mentioned interventions, her DOE lingered on and she was referred to our center. Physical examination did not reveal any further abnormal findings. Her blood pressure was 110/70 mmHg and heart rate was 85 beats/min. Complete blood count revealed mild anemia with hemoglobin: 10.3 g/dl and erythrocyte sedimentation rate (ESR): 43 mm/h. Liver and kidney function test results were within normal limits. The posteroanterior chest x-ray film demonstrated a spherical mass located at the left contour of mediastinum. Echocardiography and CT investigations were performed. Echocardiography demonstrated a multiple echo-free space cystic formation in posterolateral wall of left ventricle (LV) with no particle on it suggestive for intramyocardial cyst (Figure 1. A,B). Multiple pericardial cysts at posterior aspect of left chamber without obvious calcification measured about 28×27 mm and 36 × 36 mm were seen on CT. Cardiac MR imaging (dynamic MR imaging with gadolinium) was also performed for better evaluation. On T1 weighted and true-FISP MR images, there was a large multilobulated and septated cystic mass in the posterolateral wall of LV with size about 32.8× cm, 42.98× cm and 21.4× cm that was similar to intramyocardial and pericardial hydatid cyst (Figure 2. A,B). Based on previous data (granulomatous pericarditis) and epidemiologic evidence and typical radiologic findings for hydatid cyst, empiric treatment for TB in addition to hydatid cyst was initiated pre-operatively. Simultaneously, we started to investigate previous data for culture results

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for TB which turned out to be negative after 2 months. Finally she underwent cardiac surgery under general anesthesia and standard median sternotomy. After decortication and releasing the strong adhering bands and standard single cava cannulation the cardiopulmonary bypass (CPB) was started and cardiac arrest was induced. The apex was completely attached to the lateral wall of left hemithorax and lungs, which was hardly detached. A large cystic lesion was observed on posterolateral wall of left ventricle. After covering the surrounding tissue with silver nitrate solution soaked sponge, an incision was made and 3 large hydatid cysts and some daughter cysts were evacuated. The wall of the cyst was unroofed and the region was washed with silver nitrate solution. The patient was weaned from CPB off without inotropic support. There was no evidence of residual cyst in intraoperative trans-esophageal echocardiography. Early postoperative period was uneventful. Surgical view and gross pathology of hydatid cysts have been demonstrated in Figures 3.A and 3.B, respectively. Histopathological examination

findings confirmed the diagnosis. The rationale for starting Albendazol before surgery was lowering the intracystic pressure, preventing cyst rupture during surgery and preventing the seeding of daughter cysts. Throughout the follow-up period, we planned to treat the patient for a longer course and followed her up with IgG ELISA levels which were negative until 3 months from the treatment onset.

Discussion

HD is caused by echinococcus granulosus which is a tape worm and is still among common health problems due to its endemic status in some parts of the world.⁴ HD may locate in any parts of the body mostly in liver, lungs and brain. Some unusual sites for HD involvement include heart, pericardium, kidney, intraperitoneum, retroperitoneum, bone, soft tissue, and breast.^{1,5} Pericardial hydatid cysts are typically developed at the right costophrenic angle. Cardiac hydatid cysts mostly occur in the ventricular septum. However in this case the cysts were located in

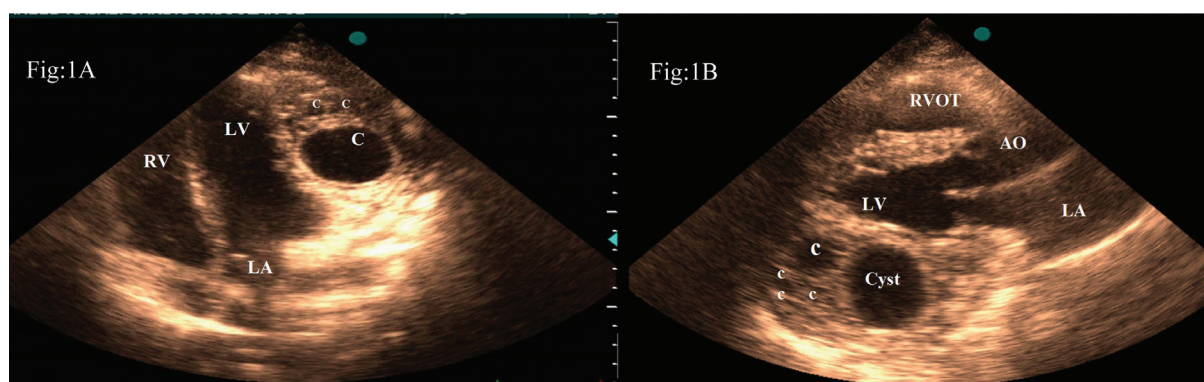


Figure 1. A) Transthoracic echocardiography in 4 chamber view showing multiple septated intramyocardial cysts. B) Transthoracic echocardiography in parasternal long axis view in the posterior wall of the left ventricle showing multiple intramyocardial cysts with honeycomb pattern.

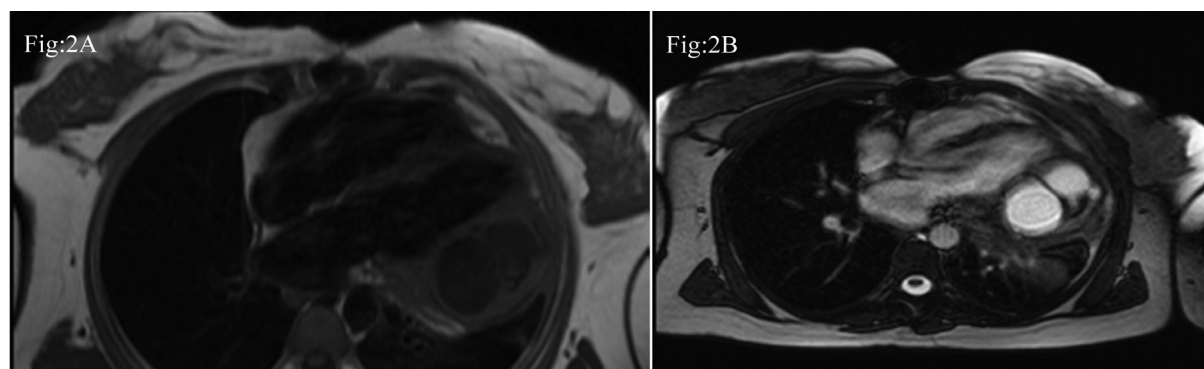
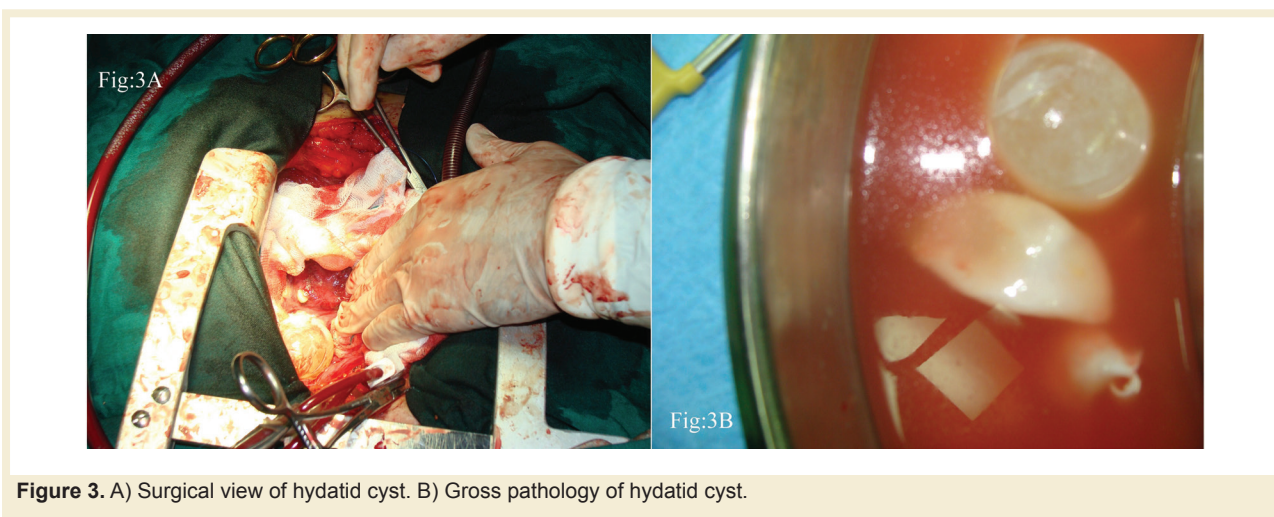


Figure 2. A) Cardiac MR view of cystic lesion. B) Cardiac MR view of cystic lesion after enhancement.



the posterolateral aspect of the left ventricle. Symptoms vary depending on size, location and involvement of neighboring structures. Some studies similar to our study reported symptoms of chest pain, shoulder pain, dyspnea and persistent cough in patients with pericardial hydatid cyst.^{2,6} There are different methods in order to investigate cardiopericardial hydatosis and echocardiography is the procedure of choice in this matter.² However, Sakarya *et al.* and Thameur *et al.* have suggested that CT scan is better than echocardiography considering its ability to distinguish solid from liquid tumors and also CT scan is an effective and reliable tool for the surgeons which can provide the exact site of the abnormality.^{2,7} In our case, CT scan was performed and multiple pericardial cysts at posterior aspect of left chamber without obvious calcification were revealed. Similar to the studied of Desnos *et al.* and sakarya *et al.*, cardiac MR imaging (dynamic MR imaging with gadolinium) was also performed.^{2,8} A large multilobulated and septated cystic mass in the posterolateral wall of LV, similar to intramyocardial and pericardial hydatid cyst was detected in MR findings. However, similar to the statement of Engin *et al.*, hydatid cysts with unusual localization may cause serious difficulties in order to differentiate the diagnosis.¹ Diagnosis of hydatid cyst mainly depends on radiologic stage it is presented at. In our case, the first presentation was with cyst rupture leaving no radiologic specification. Rupture of the cyst may explain pericardial effusion, and tamponade while secondary reactive changes in histopathology of the pericardium can explain the granulomatous changes in first episodes of the disease. Hence, after her first surgery, granulomatous changes were misleading and misdiagnosis of TB was made because of its high prevalence. However, after observing hydatid cyst and as TB occurrence in association with hydatid cyst is very rare, we also searched for granulomatous pericarditis and we found out that rupture of hydatid cyst can interestingly cause granulomatous pericarditis.

Nevertheless, due to modern imaging techniques and laboratory tests, misdiagnosed HD seems unacceptable and in order to prevent the misjudgment of HD with rare localization the combination of clinical history, imaging findings and serologic test results should be collected and evaluated.

Although the myocardial cystectomy may be performed without cardiac arrest or even CPB, we believed our approach would be safer due to the history of a previous pericardiectomy and cyst location.

Conclusion

In endemic areas, HD should be taken into consideration as a differential diagnosis of all cysts in all anatomic locations and even rare conditions such as pericardial tamponade. Consequently, a combination of clinical history, imaging findings, and serologic test results should be utilized in order to prevent any misdiagnosis and its related complications.

Ethical issues: This study was reviewed and confirmed by the ethics committee of Rajaei Cardiovascular Medical & Research Center.

Conflict of interests: The authors declare no conflicts of interest.

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