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Recurrent cardiac hydatid cysts with pericardial protrusion; a rare case presentation of *Echinococcus granulosus*

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Abstract

Hydatid cyst (HC) is a rare pathology mostly found in less developed, livestock-raising countries. HC can be found anywhere in the body, however it is a rare finding in the heart, and its recurrence in the heart and pericardium is very rare. A 54-year-old woman who had undergone surgery for the removal of HC in another country 11 years before presented with shortness of breath and heaviness in the chest. The examinations and computerized tomography (CT) scan revealed a large mass measuring 60×50 mm in the pericardium cavity originating from the right atrial wall. Infection with *Echinococcus granulosus* was confirmed with serological enzyme-linked immunosorbent assay (ELISA) test and the patient underwent cardiac surgery. The patient underwent cardiopulmonary bypass, nevertheless did not suffer cardiac arrest, and aortic cross-clamping was not performed for her. The removed mass was filled with daughter cysts. The endocysts were removed and intramural cysts were drained. Following the surgery, the patient received treatment with albendazole for six months. Cardiac hydatid cyst recurrence is a rare but possible incident, and annual follow-ups and echocardiography are recommended.

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Introduction

Echinococcus granulosus was first described in the 17th century (1) and reported in 1836. The first operation for cardiac echinococcosis was performed approximately one hundred years later in 1932 (2). Hydatid cyst (HC) continues to be endemic in rural areas and livestock-raising communities in developing or underdeveloped countries (3). HC is a parasitic infection of humans caused by the cestode (tapeworm) *E. granulosus* and lives in dogs' guts, and humans are accidental hosts who become infested if they consume vegetables or water contaminated with the parasite ova from dog stool (2).

Echinococcus granulosus is one of the few infections that are primarily diagnosed serologically using indirect hemagglutination tests and enzyme-linked immunosorbent assay (ELISA) as the most widely-used methods for the detection of anti-*Echinococcus* antibodies immunoglobulin G (IgG) (4). Cardiac involvement has been reported in only 0.5-2% of cases (1,5- 8), and cardiac HC recurrence is rare in patients (9-11). The complete removal of the cyst and its entire layers is recommended while making sure to avoid injury to the heart. Precaution must be taken to avoid contamination of the surrounding area with fluid from the cyst, therefore the chances of recurrence and dissemination are minimized (5,12).

Key point
Although a recurrent cardiac hydatid cyst is rare, annual follow-ups and echocardiographyare recommended after hydatid cyst surgery. If cardiac hydatid cyst recurrence is diagnosed, surgical removal is recommended.
The complete removal of the cyst and its entire layers is recommended while making sure to avoid injury to the heart. Precaution must be taken to avoid contamination of the surrounding area with fluid from the cyst, therefore the chances of recurrence and dissemination will be minimized.

The treatment of hydatid cysts anywhere in the body, even in asymptomatic cases, is surgical removal (2,5,6,10,13), because there is always a risk of rupture into the bloodstream and its spread throughout the body, leading to malignant hydatidosis and

risk of anaphylactic shock and death (2,6,13). In cases where surgery cannot be performed or the cysts are multiple, small, and scattered, long-term treatment with albendazole is recommended (9,14).

Case Report

A 54-year-old woman who had undergone surgery for the removal of cardiac HC in another country 11 years before (no information was available on the location of cardiac involvement and type of surgery performed) presented with shortness of breath and heaviness in the chest for the past six months. No abnormal findings were detected in the clinical examinations. Her biochemical tests were normal and cardiac enzymes were also in the normal range. The serological ELISA test for HC was performed on the patient with a positive result for *E. granulosus*. The heart rhythm was normal with no symptoms of ischemia. The two-dimensional echocardiography showed a mass effect on the right atrium (RA), and a large mass anterior to RA about 60 × 50 mm with some pressure effect on the RA anterior wall and invasion of the RA free wall was reported.

Multilocular cystic lesions in the right pericardium measuring about 70 × 67 mm with compression to the RA compatible with hydatid cysts was reported in the computerized tomography (CT) scan (Figures 1 and 2).

The coronary arteries were deemed normal in the angiography. Abdominal and pelvic ultrasounds were normal for other brain lesions. The patient became a candidate for cardiac surgery once the examinations were completed. Administration of albendazole tablets started for the patient prior to surgery. A mid sternotomy was performed on the patient and excessive adhesion was released. A tense sickly mass measuring approximately 11 cm in length and 4cm in width was detected on the right side of the pericardial cavity with calcification in some parts.

The cyst was a well-formed intramyocardial cyst attached to the RA. The patient underwent cardiopulmonary bypass by cannulation of the RA and aorta to reduce the heart volume and reduce the risk of rupture during the procedure, but there was no cardiac arrest, and aortic cross-clamping was not performed and the heart was kept beating. Hypertonic saline-impregnated gauze was placed around the cyst. Due to the high endo-tension on the cyst, injection of hypertonic saline serum into the cyst was not initially possible. Therefore, a double-lumen syringe was used to suction out part of the cyst fluid, and the hypertonic saline serum was injected into the cyst through the other lumen. Then, part of the cyst wall was opened, showing hundreds of daughter cysts within, waste suctioned and drained (Figures 3 and 4).

Two suction machines must be available in the operating room, because there is a possibility of lumen blockage caused by the small cysts. After complete drainage, the cyst cavity was cleaned multiple times with hypertonic saline serum, and the lateral wall of the cyst was removed

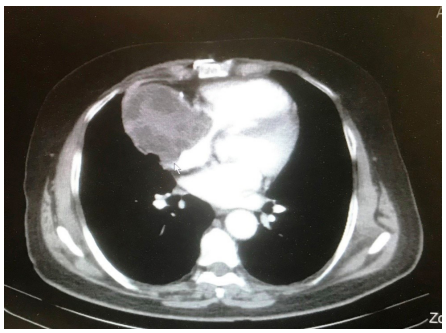

Figure 1. Computerized tomography (CT) scan of the chest showing the mass filled with hydatid cysts

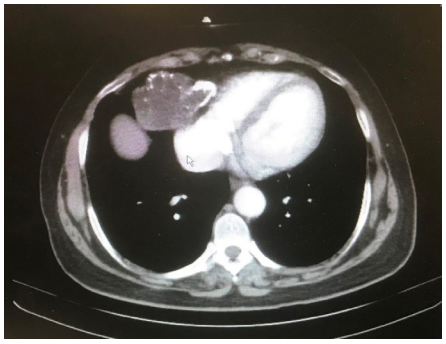

Figure 2. Computerized tomography (CT) scan of the chest showing calcification in parts of the mass wall filled with hydatid cysts.

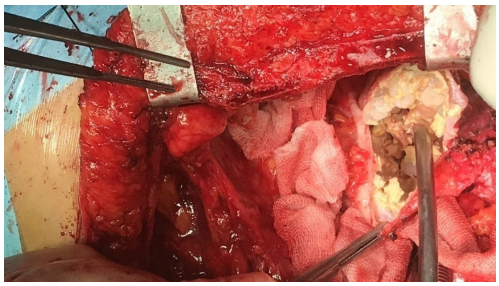

Figure 3. Multiple daughter cysts inside the hydatid cyst.

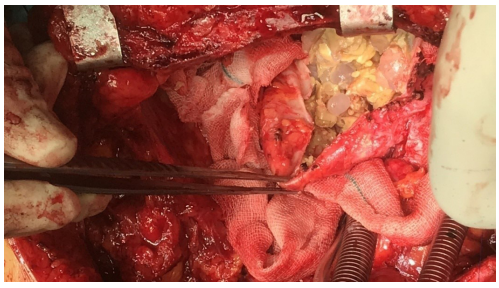

Figure 4. Draining the cardiac hydatid cyst by surgery.

completely. The wall on the RA was fully overlapping the right atrial wall and could not be removed. Therefore, the wall was shaved and the germinal layer of the cyst was removed. At the end of the surgery, the cardiopulmonary bypass was removed without difficulty and the arterial and venous cannulas were also removed and surgery was thus completed.

Pharmacotherapy with albendazole continued and the patient was discharged in a good general condition.

Discussion

Although cardiac hydatid cyst is rare, accounting for 0.5-2% of the cases of HC (10), a high index of suspicion must be kept by physicians among patients living in rural areas and developing countries (3,5). The symptoms of cardiac HC are nonspecific and it is diagnosed with echocardiography or CT scans (5,15,16) and is confirmed with serological tests. HC can have potentially life-threatening complications (9,10,17) and must be surgically removed (2,5,6,10,13). Recurrent cardiac hydatid cysts are rare, but a few cases have been reported (9-11,16).

Cardiac involvement is often asymptomatic, but can present as cardiac tamponade, embolization, chest pain, and heaviness in the chest (18). If a patient is diagnosed with HC, other organs must also be checked for HC (15), and a CT scan is recommended for the localization of the site and assessing the other organs (15,19). The complete removal of the cyst and all its layers is recommended while making sure to avoid injury to the heart (which is why cardiopulmonary bypass was performed on our patient). Precaution must be taken to avoid contamination of the surrounding area with fluid from the cyst, so that the chances of recurrence and dissemination are minimized (5,12).

Although recurrent cardiac HC is rare (9-11), our patient had symptomatic recurrence after 13 years. Therefore, it is recommended that patients with cardiac HC undergo annual cardiac imaging and follow-ups to treat any recurrence with surgery at its initial stages.

Conclusion

Although cardiac hydatid cysts are not very common, they should can considered as a life-threatening pathology. Complete removal and efforts to prevent their dissemination are essential for cardiac hydatid cysts. If these cysts are not completely removed or their fluid leaks, recurrence is possible even after ten years. The recurrence of cardiac HC is rare; however, follow-ups and annual echocardiography are recommended after HC surgery. If a cardiac HC is diagnosed, surgical removal is recommended.

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Authors' contribution

Conceptualization: MBM.
Methodology: RBM.
Validation: HG.
Formal analysis: HG.
Investigation: SAM.
Resources: MH.
Data curation: SAM.
Writing–original draft: HG.
Writing–review and editing: ZAA.
Visualization: RBM.
Supervision: MBM.
Project administration: ZAA.
Funding Acquisition: RT.

Conflicts of interest

None of the authors has any conflict of interest.

Ethical issues

This case report was conducted in accordance with the World Medical Association Declaration of Helsinki. Written informed consent was obtained from the patient for publication of this report. All ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by the authors.

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