Cardiac Echinococcosis

— Recurrent Intramyocardial-Extracardiac Hydatid Cysts With Pericardial Protrusion —

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A 26-year-old male patient was diagnosed with an isolated recurrent intramyocardial—extracardiac hydatid cyst with pericardial protrusion after being admitted with chest pain and palpitation. He had undergone surgical resection of an intramyocardial pericardial hydatid cyst without cardiopulmonary bypass 10 years earlier. In the current admission, the results from transthoracic and transesophageal echocardiography and multislice computed tomography were confirmed by serological and histopathological tests. The cyst was excised under cardiopulmonary bypass, and the patient was treated postoperatively with albendazole for 9 months. His clinical status improved postoperatively and he was asymptomatic without signs of recurrence as determined by echocardiography. (Circ J 2008; 72: 1718–1720)

Key Words: Cardiac hydatic cysts; Echinococcosis; Recurrence

Echinococcosis is an infection with the canine tapeworm Echinococcus granulosus (E. granulosus) and is associated with various sheep- and cattle-raising areas of the world. Typically in humans, cysts form in the liver (60% of cases) and lung (20–30% of cases; more commonly in children). Although most often found in the liver and lung, hydatid cysts can occur in any organ or tissue and if ruptured, there is dissemination of scolices (an immature stage) via the blood stream. Cardiac involvement of echinococcosis is rare, and occurs in approximately 2% of all patients, typically localized to the left or right ventricle.

Although cardiac echinococcosis is rare, localization to the myocardium may lead to life-threatening complications, including cyst rupture, anaphylactic shock, tamponade, pulmonary, intracerebral or peripheral arterial embolism, acute coronary syndrome, arrhythmias and infection, any of which require aggressive treatment.

We present a rare case of recurrent intramyocardial—extracardiac hydatid cyst with pericardial protrusion that was surgically removed.

Case Report

A 26-year-old man was admitted with chest pain and palpitations, which had started approximately 6 months before admission. He had undergone surgical resection 10 years earlier of an intramyocardial hydatid cyst without cardiopulmonary bypass. Physical examination did not reveal any abnormal findings: his lungs were normal on auscultation, no cardiac murmur or gallop rhythm was noted, and biochemical laboratory test results were within normal limits. Myocardial-specific enzyme values were within the normal range.

The patient’s chest X-ray was normal, except for the sternum suture, and the ECG showed normal sinus rhythm with T-wave inversion in leads V1–6, consistent with ischemia. Apical 2-chamber transthoracic echocardiography showed a multivesicular cystic mass on the left lateral ventricular wall in the pericardial sac. The cyst, measuring 6×4×5 cm, was localized to the inferoposterior wall of the left ventricle and protruded toward the pericardium (Fig 1A); this finding was confirmed by tranesophageal echocardiography (TEE). We did not detect any additional visceral localization of the cyst on abdominal ultrasonography. The patient was examined further by multislice computed tomography (CT) of the chest in order to determine the exact location, size and number of disseminated hydatid cysts; a 5.7×4.8 cm round cystic mass with well-defined contours was located next to the left ventricle (Fig 1B). The cyst had a germinative membrane that did not allow communication between the mass and the cardiac chambers or extrinsic structures. Additionally, in view of the clinical suspicion of coronary artery disease, coronary angiography was performed, and was determined to be normal.

Because of the previous history of hydatidosis, serologic tests (indirect hemagglutination tests) were performed. The results were positive for E. granulosus, and there was an accompanying eosinophilia. Because the clinical, radiologic, and serologic findings were suspicious for pericardial hydatid cyst, the patient underwent surgery for diagnostic and therapeutic purposes.

Surgical intervention confirmed an intramyocardial—extracardiac hydatid cyst with pericardial protrusion (Fig 2A). Cystectomy was performed under conventional cardiopulmonary bypass. The cystic lesion was found to be encapsulated by a well-formed intramyocardium and to be attached to the left ventricle. The cyst was first sterilized with hyper-
**Fig 1.** (A) Transthoracic echocardiography (apical 2-chamber long-axis view) shows multiple hydatid cysts (arrows) in the apex of the left ventricle (LV). (B) Computed tomographic view shows multivesicular hydatid cyst with pericardial protrusion (arrow).

**Fig 2.** (A) Peri-operative imaging shows the left ventricle and its relation to the multivesicular cystic mass in the apex. (B) The vesicles that were drained from the left ventricular apex.

**Fig 3.** (A) Cavity remaining after intramyocardial cleaning. (B) Vesicles and membranes excised from the intramyocardial-extracardiac tissues.
tonic saline solution, punctured and its contents drained (Fig 2B). Following excision, the apex of the ventricle had a 4×5 cm cavity (Fig 3A), which was closed by obliteration without evidence of damage to cardiac structures (Fig 3B). Postoperatively, the patient was treated with albendazole (50 mg/kg daily) for 9 months. The T-wave inversions on ECG continued after operation, but 6 months later, the patient was asymptomatic and did not show any signs of recurrence as determined by echocardiography.

**Discussion**

Hydatid cysts are a parasitic disease caused by the larval form of *E. granulosus*, and most commonly occurs in the liver and lungs. Cardiac echinococcosis is rare, representing only 0.5–2% of all cases and recurrence after treatment is uncommon. Chest pain, palpitations and dyspnea are the primary symptoms associated with cardiac echinococcosis and patients with cardiac hydatid disease must undergo surgery because of the potentially life-threatening complications.

The chest pain may imitate angina pectoris, but more often suggests a non-coronary origin. In young patients from endemic areas, especially when the ECG shows a T-wave change, a diagnosis of cardiac hydatid cyst should be considered. In the present case, based on the T-wave change and the patient’s history of a previous hydatid cyst operation, we suspected a recurrent pericardial or intramyocardial cyst. The ECG changes mimicked coronary ischemia, but there were no enzymatic changes, so to rule out the possibility of coronary artery disease, we performed coronary angiography, which revealed normal arterial anatomy. Furthermore, all serologic tests for hydatidosis were positive. Taken together, the serologic tests, TEE and CT findings led us a precise diagnosis of an intramyocardial-extracardiac hydatid cyst with pericardial protrusion.

Treatment of hydatidosis begins with antiparasitic medication, but frequently complete surgical removal of the cyst is required. Because improvement may occur very late, albendazole therapy should be continued even when no change is present during the first year of treatment. Despite objective mass regression and encouraging immunological changes, the decision to withdraw albendazole remains difficult. More accurate methods of measuring the viability of the parasitic lesion are needed to more effectively treat patients. Surgical cyst removal in patients with multiple small recurrent cysts should be considered because of the high risk of associated complications, including rupture, tamponade and anaphylactic shock. Furthermore, our findings emphasize the need for thorough and frequent re-evaluation to detect new hydatid cysts in the heart and other target organs.

In conclusion, although hydatid disease of the heart is very rare, our case report shows that cyst recurrence should be considered and systematic investigation of other target organs be performed, especially in patients who live in endemic areas.

**References**