Case Report

Cardiac Echinococcosis with Negative Serologies: A Report of Two Cases

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Cardiac involvement in hydatid cyst disease is uncommon, occurring in approximately 2% of hydatid cases. Two cases of cardiac hydatid cysts with negative serologic tests are reported herein. In Case 1, the patient underwent surgery to remove cysts from the brain and 3 years later for cyst removal from the breast. In both cases the diagnosis was established by transthoracic two-dimensional echocardiography and then confirmed by surgery and histological examination. These cases are of particular interest because of the rarity of cardiac localisation as a new site of the hydatid cyst after one or more previous surgeries for hydatid cyst removal, and stress the need for frequent reevaluation to detect new hydatid cyst formation in the heart and elsewhere caused by the Echinococcosis organism.

Keywords. Hydatid cyst; Heart; Echinococcosis granulosus

Introduction

Echinococcosis is a significant health problem in sheep and cattle-raising areas that usually involves liver and lungs, but cardiac involvement is not common. It can occur as part of a systemic infection or as an isolated event and may have classic manifestations, but unusual sites of involvement may cause misdiagnosis. Serological techniques for the diagnosis of the disease are well established and, together with imaging techniques, are the most important diagnostic tools. Serologic false negative result is an important factor which may cause the diagnosis to be missed. In such instances, the vigilance of physicians plays a significant role in diagnosis of the disease.

Case 1

A 73-year-old lady was admitted complaining of chest pain and exertional dyspnea for 1 year which started to increase in severity. She had a history for hydatid cyst of liver 10 years ago. Physical examination revealed a heart rate of 120 bpm, a blood pressure of 140/85 mmHg and a temperature of 37.5°C. There was no dyspnea or chest pain at rest. Total leucocyte count was 13,300/mm³ (7% eosinophils). Left bundle branch block was diagnosed in the posterior part of the interventricular septum along with a moderate pericardial effusion, trivial aortic insufficiency and trivial mitral regurgitation. Serologic tests including ELISA and immunoelectrophoresis were negative. ECG findings were consistent with inferolateral wall ischaemia.

The echocardiogram showed a 3.3 cm × 3.8 cm cystic mass in the posterior wall of left ventricle. In transthoracic two-dimensional echocardiography the ejection fraction was 60% and there was a trivial mitral regurgitation. After induction of anaesthesia transthoracic echocardiography was done. The patient was operated on using cardiopulmonary bypass through a mid-sternotomy approach. A whitish cystic mass was noted to be over the posterior wall of left ventricle. To reduce the risk of spillage of the cystic fluid, the cyst was aspirated before any manipulation (Fig. 1). The cyst was excised and the cavity was sterilised with sodium chloride solution (Fig. 2).

Case 2

A 37-year-old lady presented with a 2-year history of palpitation. She underwent craniotomy 10 years ago for a hydatid cyst of the left breast. Physical examination revealed a temperature of 38°C, heart rate of 130 bpm, blood pressure of 100/75 mmHg and fine crepitations in both lungs. Total leucocyte count was 16,300/mm³ (5% eosinophils). Left bundle branch block was diagnosed in 12-Lead ECG. A chest X-ray showed cardiomegaly and echocardiography pinpointed a 3.4 cm × 3.4 cm cystic mass in the posterior part of the interventricular septum along with a moderate pericardial effusion, trivial aortic insufficiency and trivial mitral regurgitation. Serologic tests including ELISA and immunoelectrophoresis were negative. The patient was then referred to cardiac surgery ward for excision of the cyst. The cyst was removed successfully using cardiopulmonary bypass through a mid-sternotomy approach. There were no post-operative complications.
Figure 1. The large cystic mass (whitish area) indicated by arrow seen on the posterior wall of the left ventricle. Aspiration of the cystic fluid avoids cystic fluid spillage.

Figure 2. Removal of the cyst using an ovary clamp following aspiration of the cystic fluid.

and both patients were discharged home without any symptoms. Post-operatively they continued on medical treatment which had been started 2 weeks before the operation. They are currently being followed and report no complications.

Discussion
Echinococcosis is a zoonotic infection of major health importance in many parts of the world such as the Middle East, the Mediterranean countries, South America and South Africa.1,2 The parasites enter the portal vein and reach the liver where 60-70% of them are retained. The lungs are involved in 15-25% of cases. Other organs are usually involved in 10-15% of instances.3,4 Cardiac hydatid cysts are found in fewer than 2% of cases of hydatidosis.5,6 The larva reach the myocardium through the coronary circulation. The left ventricular wall is the most frequent site which gets involved in the heart.7 Cardiac hydatid disease can mimic left ventricular aneurysm and atrial myxoma. The cysts may give rise to symptoms such as pericardial pain, dyspnoea and palpitation.

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Imaging plays an important role in preoperative diagnosis, surgical planning and follow up of cases. In our cases, echocardiography was the most useful means used to detect the lesion. However, the diagnosis was confirmed post-operatively by demonstration of scolex and hooklets in the cyst fluid. Besides imaging, there are some serologic tests such as: hydatid immunoelectrophoresis, ELISA, latex agglutination and indirect haemagglutination (IHA) test which are used in diagnosis, screening and post-operative follow up for recurrence. ELISA is 80–100% sensitive and 88–96% specific for liver cyst infection but less sensitive for lung (50–56%) or other organ involvement (25–56%). Positive predictive value and negative predictive value for ELISA test have been reported 100% and 95%, respectively. Therefore, only positive hydatid serology is valuable and negative serologic test does not exclude the diagnosis. Laboratory errors and serologic false negative results may cause the diagnosis to be missed. Eosinophilia is expected in patients with parasitic infections but it is not always seen. The treatment of hydatid cysts is principally surgical. Removing the cyst should be performed using cardiopulmonary bypass. Extracorporeal circulation and cross-clamping both aorta and pulmonary artery prevent the drastic embolic complication into the systemic or pulmonary circulation and also provide direct visualisation of the hydatid cyst and the cardiac structures. However, pre-operative and post-operative 1-month courses of albendazole and 2 weeks of praziquantel should be considered in order to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall (thus reducing the risk of spillage during surgery) and reduce the recurrence rate post-operatively. To sum up, Echinococcus granulosus can affect any organ in the body and a high suspicion of this disease is justified in any cystic neoplasm of any organ, especially in endemic regions. Cardiologists and cardiac surgeons must be alert to the possibility of cardiac involvement by hydatid cyst disease even if the serologic tests are negative.

References