A case of cardiac hydatidosis: role for transesophageal echocardiography in evaluating bilateral pulmonary nodules

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Abstract

Echinococcusis is a zoonotic disease, which is endemic in sheep-raising areas such as Iran. Cardiac involvement of the hydatidosis is rare and mostly asymptomatic, but it could lead to lethal complications. Thus, early diagnosis with accurate treatment would be life-saving. Here we report a 17-year-old female with nonspecific pulmonary presentations and a positive history of pulmonary hydatid cysts. Transesophageal echocardiography showed multiple cardiac hydatid cysts in the right ventricle. Patient underwent the bypass surgery to remove cardiac cysts. Postoperatively patient was on Albendazole and Praziquantel for two years. In a two-year-follow up, the patient had no complications.
Keywords: *cardiac surgery, hydatid cyst, transesophageal echocardiography, pulmonary nodule*

Rezumat

Un caz de hidatidoză cardiacă: rolul ecocardiografiei transesofagiene în evaluarea nodulilor pulmonari bilaterali

Echinococcoza este o zoonoză endemică în zonele de creștere a oilor cum este Iran. Afectarea cardiacă a hidatidozei este rară și de cele mai multe ori asimptomatică, dar poate duce la complicații letale. Astfel, diagnosticul precoce și tratamentul corect sunt salvatoare de viață. Prezentăm cazul unei femei de 17 ani, cu acuze respiratorii nespecifice și un istoric de chiste hidatice pulmonare. Ecocardiografia transesofagiană a evidențiat multiple chisturi hidatice cardiace în ventriculul drept. Pacienta a fost supusă chirurgiei cardiace deschise pentru îndepărtarea chisturilor și a primit postoperator, timp de 2 ani, tratament cu Albendazol și Praziquantel. Pe o perioadă de urmărire de 2 ani pacienta nu a prezentat complicații.
Cuvinte-cheie: *chirurgie cardiacă, chisturi hidatice, ecocardiografie transesofagiană, nodul pulmonar*

Introduction

Hydatid disease is an orthozoonotic disease of larval stage of *Echinococcus granulosus* which lives in dogs, rodents, sheep and some other animals. Human is an incidental intermediate host, so this parasitic infection is prevalent in many sheep raising regions. Considering diverse presentations of the disease and its ability to invade various organs, hydatidosis should be always put on differential diagnosis in endemic regions[1].

E. granulosis could reach many parts of body, mostly liver and lungs. It could also locate less frequently in bones, muscles, brain and heart[2]. Cardiac involvement is rare and is reported to involve as much as 0.5 to 2% of the cases[3]. Silent progression of the disease usually results in late diagnosis and hence lethal complications related to ruptured cysts or postoperative complications are encountered in majority of patients. Therefore early diagnosis, prompt and accurate treatment and proper follow-up are mandatory steps in management of this disease.

Hydatid cyst could be diagnosed via many clinical, radiological and laboratory tests. So far, it has been stated that echocardiography is the most sensitive imaging test for cardiac hydatid cysts[4]. In this report we illustrate how transesophageal echocardiography helped to identify etiology of bilateral pulmonary nodules and helped diagnosis and proper surgical treatment of cardiac hydatid cyst.

Case presentation

A 17-year-old female presented with complaints of chronic productive cough, dyspnea, intermittent hemoptysis and right pleuritic chest pain to Masih Daneshvari Medical center, Tehran, Iran. She also had significant weight loss, headache and dizziness. Her symptoms started one year before presentation to our medical center and, despite outpatient evaluation and treatment, they were aggravated. Physical examination on first visit showed a pulse rate of 120 beats/minute, a respiratory rate of 18 beats/minute, a blood pressure of 100/60 mmHg and an oral temperature of 37.1 ^{0}C. She had diffuse wheezing in both lungs. Heart auscultation revealed a systolic murmur. Hyper dynamic right ventricle was evident on palpation. No other positive finding was noted on examination.

Chest radiography revealed multiple bilateral lung nodules and cysts (Figure 1). In laboratory tests mild anemia (Hb=10.8 g/dL), mild eosinophilia (6%), high ESR (61 mm/hr) and negative ANCA were detected. Computed tomography (CT) scan demonstrated multiple bilateral cystic lesions with different size and well-defined border. The lesions were encountered mostly in sub-pleural region with some excavation and internal floating membrane. Brain, pelvic and abdominal CTs were normal. Transesophageal echocardiography (TEE) was done and showed cystic masses (at least two cysts with size of 11X9 mm and 17X20 mm) in right side of interventricular

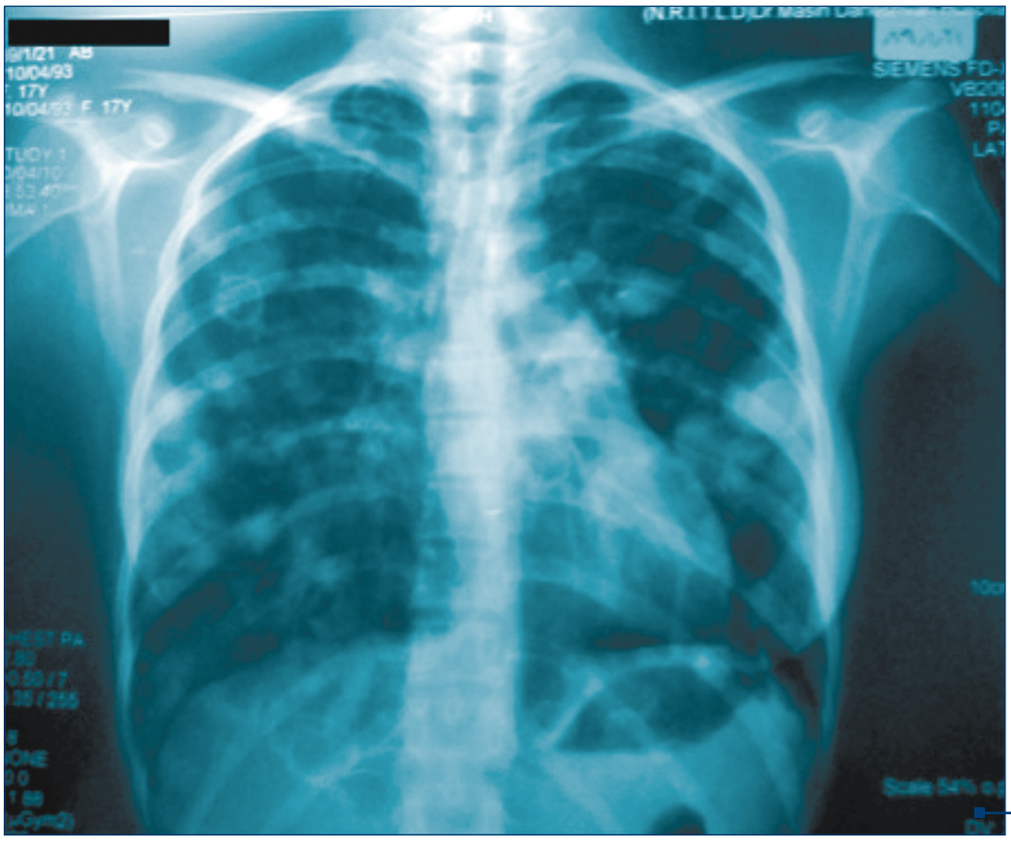

Figure 1. ***Chest X-ray demonstrating multiple bilateral lung nodules and cysts***

septum highly suggestive for cardiac hydatid cyst. Careful review of patient's history revealed a positive history of pulmonary hydatidosis more than a year ago, which was treated medically by oral Albendazole.

Due the severity of the symptoms the patient underwent open-heart surgery to remove the ventricular masses. Using cardiopulmonary bypass and midline sternotomy, the right atrium and interatrial septum were opened for decompression. Then, right ventricle was opened just inferior to the pulmonary artery. Multiple cysts were removed and sent for pathologic evaluations. Adjacent areas were irrigated and then sterilized by silver nitrate. Atria, ventricles and septum were closed in 2 layers; then heart was shocked and patient got off the pump. She was transferred to intensive care unit and was extubated on the same day. Pathologic examination of the specimen confirmed the diagnosis of hydatid cyst. Cytopathologic evaluation of the cyst's fluid was negative for malignancy.

In control echocardiography no cyst was seen. After the surgery, medical treatment was started with Albendazole (400 mg b.i.d) and Praziquantel (2400 mg monthly) for at least 2 years. During a two-year follow-up, the patient had no complaint and in control chest radiographies hydatid cysts were reduced in size.

Discussion

Hydatidosis is a parasitic disease caused by *Echinococcus* tapeworms specially *E. granulosis* and *E. multilocularis*. Definitive hosts pass eggs through their feces. Intermediate hosts such as sheep, goats and humans get infected by ingesting eggs from contaminated food or water. Humans are incidental hosts and the larval stage of *Echinococcus* tapeworms causes the disease.

After egg ingestion, freed embryos pass through intestinal mucosa and enter the blood. By blood circulation they transfer to any part of the body, mostly liver and lungs. They could also seed in heart, brain, bones, kidneys, muscles and soft tissues.

In hydatidosis, cardiac involvement occurs in less than 2% of all cases and may be due to hematogenous spread or ruptured lung hydatid cysts[2]. Affected areas are the left ventricle (50-60% of cases), interventricular septum (10-20%), right ventricle (5-15%), pericardium (10-15%), and right or left atrium (5-8%)[5].

Cardiac hydatidosis causes a variety of signs and symptoms. Presentations are related to size, location, number and integrity of the cardiac cysts[4, 6]. In most cases cardiac involvement is asymptomatic and the silent progression results in complications. Cardiac hydatid cysts complications are ischemia, arrhythmia, conduction anomalies, ventricular aneurysms, septic emboli, cardiac tamponade, valvular dysfunctions and pericardial effusion[7]. By early diagnosis and proper treatment, these complications could be prevented.

Diagnosis is based on clinical features, imaging studies and laboratory tests. Chest radiography, CT scan and Magnetic Resonance Imaging (MRI) are imaging modalities that can help us to find and localize the lesions. Our patient had nonspecific symptoms and signs. Chest X-ray and CT scan showed multiple bilateral pulmonary nodular and cystic lesions. This radiographic picture suggests many differential diagnoses such as Wegener's granulomatosis, metastases and infections (nocardia, pneumocystis jirovecii, mycobacterium

complex, tuberculosis etc). Although in the course of hydatid cyst, primary origin of the disease is usually the lungs, in our case, owing to the radiologic findings, septic emboli from heart chambers was one of the possibilities. Hence, we seek for a potential source in the heart by transesophageal echocardiography. Thus far, it has been stated that echocardiography is the most sensitive test for cardiac hydatidosis[4]. Transesophageal echocardiography versus transthoracic echocardiography provides more details about heart lesions and is more accurate to determine chambers pathology[8]. In our patient TEE revealed presence of multiple cysts in the right ventricle which suggested hydatid cysts. Abdomen, brain and pelvic CT scans were performed to evaluate presence of the disease in other organs and showed no involvement.

There are medical and surgical treatments for hydatidosis. The choice treatment for cardiac hydatidosis is surgical excision[6]. Surgery is a life-saving treatment and could prevent fatal complications like parasitic emboli. Albendazole or Mebendazole alone or along with Praziquantel is an effective supplemental medical therapy after the surgery to reduce the risk of recurrence and clear the pulmonary lesions[9]. Medical treatment without surgery would eradicate cysts as much as 30% of the cases[10]. The shortcoming of this single therapeutic approach was also evident in our case since despite Albendazole therapy for more than one year; pulmonary lesions of our patient were still present and resulted in cardiac involvement.

Various surgical methods could be implemented depending on the fragility of the cysts and their location[11]. Here, another advantage of the TEE comes to play role in the defining the most proper therapeutic approach. In pericardial or epicardial attachment of the cysts, surgery could be performed on the beating heart. On the other hand, for the intracavitary lesions cardiopulmonary bypass should be considered[11]. This is while Rossouw et al.[12] reported a case of right ventricle hydatid cyst that was treated with normothermic cardiopulmonary bypass in an empty beating heart.

In our patient and according to TEE findings, cysts were intracavitary and the bypass surgery for cysts removal was planned. Then the patient was put on medical therapy to prevent recurrence and reduce pulmonary cysts. Serial control echocardiographies after the surgery were planned and were all negative for any new cardiac involvement.

Surgery for cardiac hydatidosis may result in postoperative complications such as myocardial tearing, atrioventricular block, ventricular arrhythmias, and sudden death[13]. Moreover, surgical excision is associated with increased risk of cyst rupture and successful surgery is highly dependent on surgeon's expertise in this field. Di Bello and colleagues[14] have reported four sudden deaths after the operation; but a two-year follow-up of our patient showed no complications and control TEEs as well as chest radiographies have shown that the pulmonary cysts were reduced in size and number.

In conclusion, in endemic areas for *echinococcosis*, presence of bilateral lung lesions that are accompanied by cardiac symptoms should suspect clinician to a cardiac hydatid cyst. In this setting, TEE is a valuable diagnostic tool to rule out the disease and help defining a proper treatment strategy. We recommend that right side of the heart and pulmonary arteries should be checked specifically. After confirmation of the diagnosis, surgery with postoperative chemoprophylaxis is life-saving and improves patient's prognosis. ■

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