A rare presentation of cystic echinococcosis: aortic involvement

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Abstract

Cystic echinococcosis is an endemic parasitic infestation caused by the larval stage of Echinococcus granulosus. Although infestation of any part of human body can occur, isolated cardiac involvement is uncommon. We present a case of isolated hydatidosis involving the ascending aorta.

Keywords: Aorta • Cystic echinococcosis • Parasitic infestation

CASE REPORT

A 53-year old male was referred to our clinic for the evaluation of recurrent arterial embolic events. He had undergone an operation for massive pericardial fluid 10 years earlier. He had femoral artery emboli accompanied by brachial artery emboli 12 months ago and was diagnosed with cerebral artery emboli 5 months ago. Brachial and femoral surgical embolectomy was performed for these events previously. He had no cerebrovascular sequelae. His physical examination was unremarkable with palpable pulses. His biochemical and haematological findings were within the normal range. The chest radiography showed no evidence of anomalies. The electrocardiogram and transthoracic echocardiography were both normal. There was a cystic lesion with irregular contours in the tomography images which showed a partially occluded ascending aorta (Fig. 1). This was considered as the embolic origin so we decided to perform a surgical resection. The operation was performed using cardiopulmonary bypass. We used right subclavian artery for arterial cannulation. Through a median sternotomy, the ascending aorta was transected just distal to the sinotubular junction. Aortic lumen was opened longitudinally and an intramural cauliflower-like solid mass 3–4 cm in diameter resembling a hydatid cyst was observed in the lateral wall of the ascending aorta, 4–5 cm distal to the aortic valve (Fig. 2). The surgical field was covered with iodine-soaked sponges to prevent possible seeding. The mass and the surrounding aortic tissue were removed and a 26-mm prosthetic Dacron graft was anastomosed to the distal aorta by antegrade cerebral perfusion technique. Thereafter, the other end of the graft was sutured to the proximal aorta just above the aortic commissures. During surgery cross-clamp time was 20 min, cardiopulmonary bypass (CPB) time was 30 min and total surgery time was 80 min. We performed the operation without using circulatory arrest. During CPB, the temperature was 32°C. Pathological analysis of resected specimen revealed the hydatid disease. After the uneventful postoperative course, the patient was discharged on mebendazole therapy for 1 year (50 mg/kg/day). He was asymptomatic at 2-year follow-up physical examination.

DISCUSSION

Hydatid disease is a parasitic infestation seen endemically in sheep-raising regions of the world such as South America, South Europe, Africa, Turkey, Australia, New Zealand and India due to Echinococcus granulosus. Humans are infected by ingesting foods contaminated with larvae of Echinococcus granulosus which cross the intestinal mucosa and enter portal circulation and subsequently become intermediate hosts. Hydatid cysts mostly affect the liver (55–70%) and lung (17–35%). Nevertheless, some larvae may pass these filtering organs and reach the systemic circulation. Cardiac involvement is rare (0.5–2%) and usually accompanied by the involvement of other organs [1]. There are no statistical data regarding aortic involvement. The arterial wall is the exceptional site for hydatid cysts even in endemic countries. Aortic involvement may be primary as an intramural form due to intimal tear or vaso vasorum or secondary due to local invasion from adjacent organs or embolism from other locations. Gerber et al. reported a case with aortic hydatid involvement and aortic pseudoaneurysm of the ascending aorta 11 years after a ventricular cyst hydatid surgery [2]. Hydatid cysts involve descending and abdominal parts of the aorta more commonly than ascending part [3]. However, why the hydatid cysts penetrate descending part of aorta rather than ascending part remains unknown.

Life-threatening complications such as anaphylactic shock, false aneurysm, systemic emboli, arterial occlusion may emerge...
because of the aortic disease. Accordingly, our case presented with multiple embolic events. A negative serodiagnostic assay does not exclude the diagnosis of echinococcosis, because up to 50% of infected individuals may have negative serology [3]. Digital subtraction angiography, ultrasound, computed tomography and magnetic resonance imaging, which can detect the exact relationship of the cyst with the aorta are the current recommended diagnostic tools for aortic wall hydatid disease. In our case, computed tomography revealed the mass lesion partially occluding the aorta. An aortic cystic echinococcosis may not be diagnosed preoperatively even in endemic areas, unless it is specifically considered as a possibility. Moreover, diagnosis may be confirmed intraoperatively or later histopathologically in an exceptional location. Rarity of this disease precludes a definitive surgical technique for arterial hydatid disease. However, resection coupled with graft interposition has been preferred over patch repair. By the use of partial resection and patch repair, fatal bleeding in one and false aneurysm in another case were reported [4]. However, resection with graft interposition has been successful in all reported cases [5]. Rupture of the cyst and subsequent dissemination of the infection should be cautiously avoided during surgery. Medical therapy with albendazole or mebendazole after surgery may help in the complete eradication of the cysts.

Aortic wall involvement due to a hydatid cyst, causing multiple embolism, is very rare. In spite of this unusual manifestation, diagnosis of a hydatid cyst should be kept in mind in endemic areas. It can be treated surgically with graft interposition.

Conflict of interest: none declared.

REFERENCES


eComment. Evidence-based diagnosis and treatment of intraaortic cystic echinococcosis

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We read with great interest the case report by Tosya et al. on isolated hydatidosis involving the ascending aorta [1]. Cystic hydatid disease still represents a neglected illness, despite being highly endemic in a myriad of livestock-raising regions worldwide, and this case presentation makes evident how diverse the localization and clinical manifestations of this infection can be. With this comment, our aim is to extend the discussion by highlighting several important issues.

The described patient initially presented with recurrent arterial embolic events, which is an unusual manifestation of this disease. Hydatid embolism usually stems from spontaneous or traumatic rupture of the hydatid cyst, with only a handful of case reports describing such events in the literature [2]. Still, histopathologic evaluation of the embolus can be pursued, which in some instances may reveal blood clots around hydatid membranes and guide clinicians towards a final diagnosis [2]. Unless it is specifically considered as a possibility, a preoperative diagnosis of echinococcosis in locations other than the lungs and liver is cumbersome, even in endemic areas of the world. Although the diagnosis is usually performed with serologic tests, the authors rightly state that up to 50% of infected individuals may present with negative serology. It is important to highlight this point, as large numbers of clinicians still seek serologic confirmation of their working diagnosis.

Although significant efforts have been made to improve this percentage by using synthetic peptides, recombinant proteins and combinations of defined antigens, it must be noted that only 60-80% of infected individuals become seropositive [3]. Furthermore, 10-15% of serological examinations yield negative results due to the wall thickness of the individual hydatid cysts. By contrast, false-positive serological findings can also be found in 10-15% of patients, primarily as a result of cross-reactions with other parasitic infections, but also in non-active disease stages, patients with malignancy or during pregnancy [3]. Evidence-based treatment in cases like this is lacking, even if surgical management is obviously associated with a better outcome of patients. The authors state that rupture of the cyst and ensuing dissemination of the infection should be carefully avoided during surgery [1]. According to Volpe et al., a pivotal step required to avoid systemic dissemination is the accurate surgical resection of parasitic cyst, with aortic reconstruction as the treatment of choice whenever possible [4]. As the rate of disease recurrence after surgical treatment can be high, the treatment of cystic echinococcosis with cardiac and aortic involvement should be a combination of surgical intervention and chemotherapy. Anti-helmintic medication should be given during operation or in the postoperative period, since their administration...