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CASE REPORT

An Incidental Finding of Heart Echinococcosis in a Patient with Infective Endocarditis: a Case Report

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Echinococcosis is a cosmopolitan zoonotic parasitic disease caused by infection with the larval stage of tapeworms from the *Echinococcus genus*, most commonly *Echinococcus granulosus*. According to WHO, more than 1 million people are affected by hydatid disease at any time.[1] About 10% of the annual cases are not officially diagnosed.[2] In humans, the disease is characterized by development of three-layered cysts. The cysts develop primarily in the liver and the lungs, but can also affect any other organ due to the spreading of the oncospheres. Cardiac involvement is very uncommon - only about 0.01–2% of all cases.[4,5] In most cases, the cysts develop asymptomatically, but heart cysts could manifest with chest pain, dyspnea, cough, hemophtisis and can complicate with rupture. Diagnosis is based on a number of imaging techniques and positive serological tests. Treatment for cardiac localization is almost exclusively surgical.

We present a case of an incidental finding of an echinococcal cyst in the left atrium (rarest possible localization of heart echinococcosis) in a patient, admitted for infective endocarditis.

INTRODUCTION

Echinococcosis is a cosmopolitan zoonotic parasitic disease caused by infection with the larval stage of tapeworms from the *Echinococcus genus*, most commonly *Echinococcus granulosus*. Definitive hosts of the parasite are usually canid carnivores (dogs, wolves, foxes, etc.), whereas herbivores (e.g. sheep, goats, etc.) and humans serve as intermediate hosts and are infected by oral ingestion of the parasite's eggs.[1,2]

According to WHO, more than 1 million people are affected by hydatid disease at any time.[1] In humans the disease is characterized by the development of three-layered cysts. The cysts primarily develop in the liver and the lungs because of their barrier functions, but can also affect any other organ due to the spreading of the oncospheres from the intestines via the bloodstream. Cardiac involvement is very uncommon - only about 0.01–2% of all cases: 55-75% in the left ventricle, specifically in its myocardium, 15-25% in the right ventricle and 10-14% equally distributed between the right and left atrium.[4,5]

Diagnosis is based on ultrasonography (US), plain X-ray films and computed tomography (CT) findings, as well as positive serological tests.

We present a case of an incidental finding of an echinococcal cyst in the left atrium (rarest possible localization of heart echinococcosis) in a patient, admitted for infective endocarditis.

CASE REPORT

A 77-year-old farmer was admitted to St George University Hospital, Plovdiv with a 10-day history of persistent fever as high as 40°C, accompanied by fatigue and loss of appetite. Earlier that month the patient underwent debridement in another facility due to a phlegmon of the right foot.

The past medical history of the patient included a long list of cardiovascular diseases such as rheumatic heart disease as adolescent, mitral valve stenosis combined with regurgitation, permanent atrial fibrillation, long term hypertension and consecutively congestive heart failure. Three years prior to the current hospitalization a mitral valve replacement was performed and a biological valve was implanted.

Echocardiographical exams were performed 5 and 4 months before the current hospitalization as the patient was seeking medical attention for worsening heart failure. They revealed no abnormal masses inside any of the heart chambers, but showed an enlargement of left atrium - from 4.7 cm to 7.2 cm over a period of one month (we leave a room for certain doubt whether the enlargement is as dramatic as it would seem, considering that the patient had had mitral valve stenosis and regurgitation for most of his life, as well as chronic atrial fibrillation for years, all of which would lead to a potentially much larger left atrium cavity than 4.7cm as it was measured initially), pulmonary hypertension, enlargement of right heart chambers, lowered amplitude of opening of the biological mitral prosthesis with calsinosis and a stenotic blood flow through it. Ejection fraction was measured respectively 63% and 54%. No thrombotic lesions of the mitral valve were registered at that time.

Ten days prior the patient was hospitalized for a necrotic purulent wound on his right foot which was assessed as phlegmon and incision and debridement were carried out. According to the relatives, the fever that was present during the hospitalization, persisited after the discharge from the hospital.

The status upon admission to the Clinic of Cardiology in St George University Hospital showed pathological findings, consistent with chronic heart failure, chronic atrial fibrillation, tricuspid and aortic regurgitation. Mild anemia and slightly elevated markers of inflammation were also found.

Transthoracic echocardiography confirmed a severily dilated left atrium - 7.07 cm (anterior posterior dimension), with volume of around 450 ml, indexed to body surface area volume ~ 266 ml/m^2 (with normal values below 4 cm, <58 ml and <28 ml/m^2 respectively); visualized spontaneously increased echogenicity, pulmonary hypertension, ejection fraction ~ 53%, grade I aortic regurgitation, grade III tricuspid regurgitation, stenotic blood flow through the prosthetic mitral valve (mitral valve opening ~ 1cm), thrombosis of the same valve and a vegetation of the anterior mitral leaflet – 15x9 mm, the latter indicating endocarditis. What aroused our immense interest was a round, mobile, pedunculated formation, 3.51x3.14 mm, attached to the upper part of the left atrium that could not be explained as a manifestation of infective endocarditis. The finding was further examined with transesophageal echocardiography ***(Fig. 1)****. Due to its very specific morphology – including the pathognomonic "water-lily sign", indicating detachment of the endocyst membrane, we deduced that it is very likely an echinococcal cyst. The patient tested positive with ELISA IgG for hydatidosis (47.98 U/ml; where <10 U/ml is negative, 10-15 U/ml – gray zone and >15 U/ml*

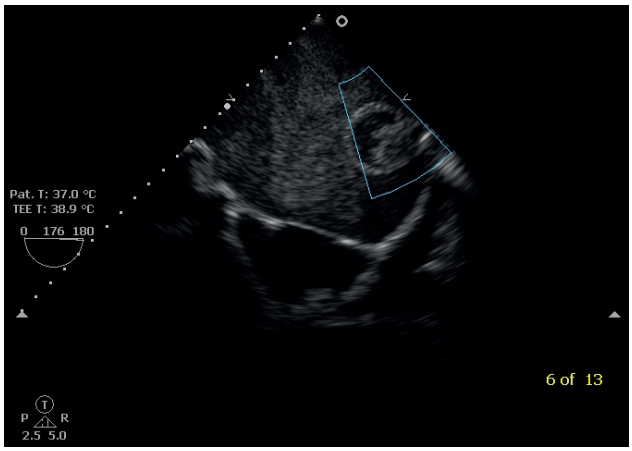

Figure 1. Echnococcal cyst in LA.

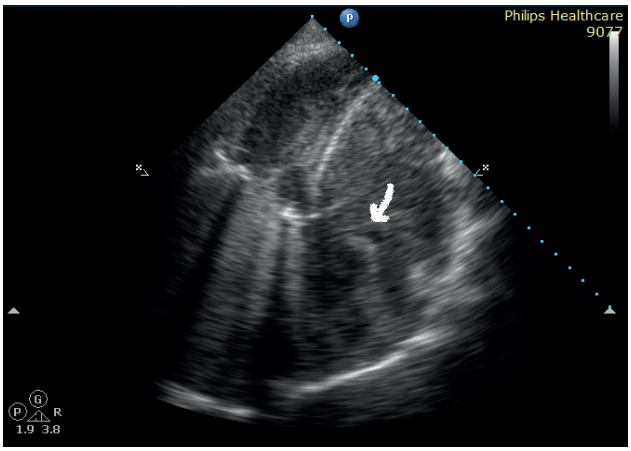

Figure 2. Ruptured cyst in LA.

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is positive). As in most cases with hydatic disease, eosinophil count was not elevated. Echinococcal cysts in other locations were not visualized using sonography. Computed tomography was planned, but not performed due to technical issues.

Diagnosis infective endocarditis (IE) was established based on 2 major and 2 minor Duke criteria - echographic visualization of thrombosis and vegetations of the prosthetic valve, 2 consecutive isolations of a coagulase-negative staphylococcus (CoNS), persistent fever and the presence of a prosthetic mitral valve as a predisposing factor. A most likely entrance door was the phlegmon the patient was treated for 10 days before the examination. Treatment with vancomycin 2x1g i.v., according to the antibiogram was started. Treatment for the concomitant heart conditions was also administered - torasemide, metildigoxin and acenocoumarol.

Consultation with a cardiac surgeon was sought and it was concluded that intervention for removal of the cyst would be considered after the completion of the endocarditis treatment. The consulting parasitologist advised against the use of preoperative chemotherapy.

On the 15th day of treatment for the infective endocarditis, to which the patient was responding well, his condition suddenly deteriorated. He showed signs of hemodynamic instability and emergency treatment with dopamine, urbason, adrenalin and hydration was started. Echocardiography showed signs of cyst rupture ***(Fig. 2)****. Death occurred despite reanimation efforts. Autopsy was refused by the relatives*.

DISCUSSION

In most cases echinococcal cysts develop asymptomatically, which delays diagnosis and treatment, therefore leading to a higher risk of complications. Clinically, heart localization could manifest with chest pain, dyspnea, cough, hemophtisis etc. The presence of the cyst may cause left or right ventricle outflow obstruction, stenosis or regurgitation of the valves, contractility failure, rhythm and conductivity disorders and congestive heart failure.[3]

The rupture of the cyst is associated with a very high mortality rate, on account of the occurrence of anaphylactic shock, intracardiac perforation, pulmonary, cerebral and coronary embolism.[6]

Differential diagnosis was made with cardiac tumors, parietal thrombosis, and mitral valve thrombosis, given the patient's history of valvular disease and the presence of an implanted biological valve.

The preferred treatment of heart echinococcosis is surgical. Chemotherapy using albendazole and/or mebendazole should be avoided before surgery as it leads to weakening of the cyst wall and a higher risk of cyst rupture. Depending on the localization of the cyst either an off-pump procedure (in cases of pericardium involvement) or cardiopulmonary bypass is used. Due to the specific location, in cases of heart echinococcosis it is rarely possible to perform pericystectomy with full removal of the fibrous capsule or partial organ resection as it is done in other organs.[5,7] After removal, the content of the removed cyst is microscopically examined to determine if the cyst contains alive protoscoleces, which increase the risk of secondary echinococcosis.[8,9] Recurrences have been reported despite adequate intraoperative prophylaxis – ablastics and the use of germicides.[5,10]

Postoperative chemotherapy with antihelminthic drugs is advised as it is shown to decrease recurrence significantly.[5,6] It is done with either mebendazole or albendazole, with preference given to the latter.[11] Dosage is adjusted to body weight (10–15 mg/kg for albendazole and 40–50 mg/kg for mebendazole), however there is no consensus as to how long the treatment should be.[9]

CONCLUSIONS

The lethal outcome in this case was a result of two treatable conditions, both with a very high-mortality risk. Lack of endocarditis would have sped up the surgical intervention, thus leading to a potentially better outcome. On the other hand, it is very difficult to determine when the cyst would have been diagnosed if it wasn't for the endocarditis and rupture might have been its first manifestation. A very important question that this case poses is whether more flexible decisions should be made regarding timing of surgery in patients with IE and potentially lethal co-existing conditions.

CONFLICT OF INTEREST

The authors have no conflict of interest to declare.

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Случайная находка сердечного эхинококкоза у пациента с инфекционным эндокардитом: клинический случай

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Эхинококкоз является космополитной зоонозной паразитарной болезнью, вызываемой инфекцией личиночной стадии ленточных червей рода Echinococcus, чаще всего Echinococcus granulosus. По данным Всемирной организации здравоохранения свыше одного миллиона человек поражено гидатидной болезнью на каждый конкретный момент времени.[1] Около 10% ежегодных случаев не являются официально диагностированными.[2] У людей болезнь характеризуется развитием трёхслойных кист. Эти кисты развиваются главным образом в печени и лёгких, но могут быть поражены и любые другие органы ввиду распространения онкосфер. Поражение сердца встречается сравнительно редко – приблизительно в 0.01 – 2 % из случаев.[4,5] В большинстве случаев кисты развиваются асимптоматично, но сердечные кисты могут сопровождаться болью в груди, диспноэ, кашлем, гемофтизом и ухудшаться при разрыве. Диагноз основывается на данных ряда исследований с применением образной диагностики, а также положительных серологических исследованиях. Лечение эхинококкоза с сердечной локализацией в основном хирургическое.

Нами представлен случай случайной находки эхинококковой кисты в левом предсердии (возможно наиболее редкая локализация сердечного эхинококкоза) у пациента, поступившего с диагнозом инфекционного эндокардита.

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