CASE REPORT

HYDATID CYST IN A RARE SITE; THE LEFT VENTRICLE

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Echinococcosis is a disease that has been affecting human beings since decades and is primarily a zoonotic disease. The larvae of genus Echinococcus causes this disease. The most common sites for these cysts are the lungs and liver. Cardiac hydatidosis is a rare phenomenon and the incidence of such cases is about 1.3%. The case presented is of a 15-year-old boy who would have shortness of breath on exertion and acute right leg pain. To diagnose cardiac variant of the cystic disease, computed topography (CT) and echocardiography were done. Doppler studies confirmed acute limb ischemia. The treatment modality was an open-heart surgery and bilateral embolectomy of the popliteal artery. The patient was discharged to home with albendazole chemotherapy. The study gives an insight into an unusual presentation and successful treatment of cardiac hydatid cyst as well as the associated complications that can be encountered.

Keywords: Echinococcosis; Cardiac hydatidosis; Embolectomy; Left ventricle

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INTRODUCTION

Echinococcosis is a disease that is affecting human beings since decades and is primarily a zoonotic disease. The larvae of genus Echinococcus causes this disease and is also called hydatid disease or hydatidosis.[1] Symptoms occur when echinococcal cyst enlarges and affect the involved organ due to space occupying effect. Most common variant of hydatid cyst has daughter cysts within parent cyst.[2] Most common sites for these cysts are the lungs with incidence of about 25% and the liver in about 65% of reported cases. Rarely, the cyst can involve the heart occurring in about 1.3% of the reported cases[3–5]. The left ventricle is the most commonly involved site in cardiac variant of the disease (55–60% of cases).[1]

Left ventricle and in particular the free wall of left ventricle is most commonly involved. Most common presenting symptoms are chest pain, shortness of breath and palpitations however majority of the patients are asymptomatic.[6] Solitary pericardial hydatid cyst is unusual. The cysts are notorious for its slow growth and has tendency to grow towards a weak myocardial wall particularly the endocardium or epicardium. Moreover, ventricular cyst is usually located sub-epicardially.[7] The most specific and sensitive tools used to diagnose cardiac hydatidosis are CT scan chest and echocardiography.[3]

To treat such a case, things that needs to be taken into evaluation should be the general health of the patient, along with the location of the cyst, clinical manifestation and size. However, for definitive treatment surgery is the standard of care followed by chemotherapy with albendazole.[2] Recently, hydatidosis of chest wall has been reported[8] but there is no documented case of cardiac hydatidosis involving left ventricle in Pakistan till date.

CASE REPORT

The patient who was a 15-year-old boy was referred to the outpatient clinic with chief complaint of shortness of breath on exertion. Cardiac auscultation showed sinus rhythm without a murmur. He had high blood pressure of 162/119 mmHg and heart rate of 119/min on his first appointment. Previously, he had liver hydatidosis and underwent surgery for its removal a year back. With that background in check, an echocardiogram was requested. The report showed a 3×3 cm cystic lesion in left ventricular cavity and was attached to its antero-lateral wall suggestive of hydatid cyst (Figure-1).

To rule out cysts in remote sites a CT scan chest and abdomen were ordered which confirmed a 3.6×3.2×3.6 cm cyst attached to left ventricle extending into the ventricular cavity (Figure-2, 3). In this case FNAC and serology were not done due to cyst location. The mystery in this case was the right limb pain with gangrenous toes for which a Doppler ultrasound of lower extremities was requested (Figure-7). It reported bilateral stenosis of popliteal arteries and occluded right posterior tibial and dorsalis pedis arteries. The imaging findings of the case were discussed among cardiac surgeons and radiologists and cardiac echinococcosis involving left ventricular cavity was suggested which urged prompt surgical intervention.

Figure-1: Echocardiogram shows hydatid cyst inside left ventricular cavity

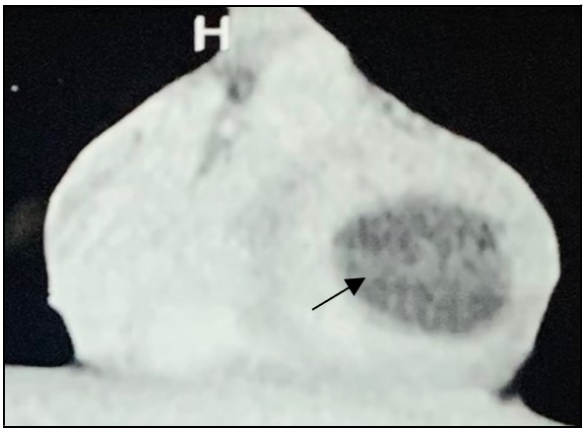

Figure-2: CT scan of chest (coronal section) shows left ventricle hydatid cyst.

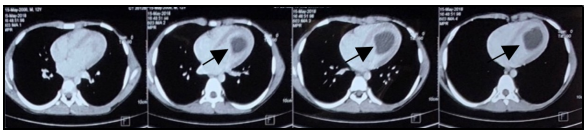

Figure-3: CT scan of chest (transverse section) shows left ventricular cyst

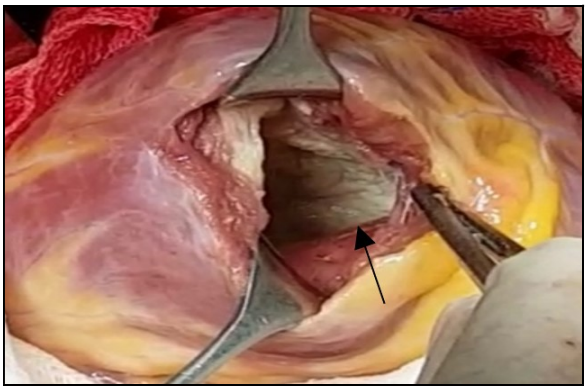

Figure-4: Cyst in-situ. Excision of cyst from ventricular myocardium.

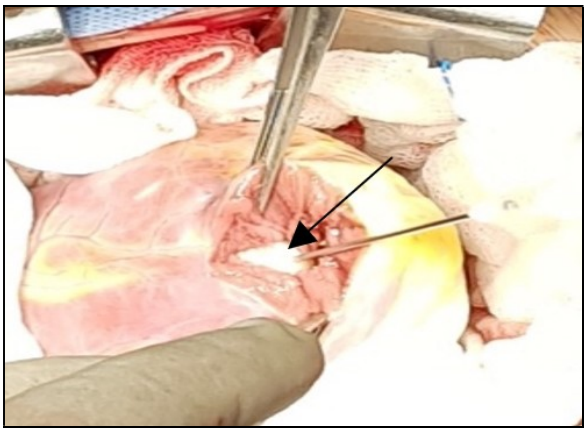

Figure-5: Left ventricular opening shows cyst wall

Figure-6: Hydatid cyst removed from left ventricle

Figure-7: Gangrenous toes secondary to acute limb ischemia from a suspected hydatid cyst embolus

Figure-8: CT Angiogram lower limbs show absent blood flow below knee

The family was counselled and an open-heart surgery with bilateral embolectomies were planned for which the patient was admitted. During the operation the cyst was found in the sub-endocardial layer of left ventricle in its lateral wall. It was injected with hypertonic saline multiple times after which the cyst was removed (Figure-4, 5 & 6). Moreover, bilateral embolectomies of the popliteal artery via femoral artery were also performed. The patient had five days stay in surgical intensive care upon successful removal of the hydatid cyst. A post-operative echocardiogram was done which reported normal biventricular function, however, due to prolapse of anterior leaflet of mitral valve, moderate mitral regurgitation was reported. The next challenge in this case was the right gangrenous foot for which help was called from the orthopaedic team. Distal pulses were still absent and hence orthopaedic surgeons decided to do a CT angiography of the lower limbs to decide level of amputation. The angiogram showed complete right popliteal artery occlusion with no visible direct or collateral flow and hence a through knee amputation of the right limb was performed (Figure-8). The patient was discharged to home with albendazole chemotherapy and a follow up review was planned after a month. Approval for this case report was obtained from the Ethics Committee of Northwest General Hospital and Research Centre. Informed consent has been obtained. Confidentiality of this patient was not breached at any stage during this study.

DISCUSSION

Presentation of hydatidosis in heart is very rare and usually asymptomatic. Symptoms occur in only 1 out of 10 patients.[9] Manifestations of hydatidosis in heart has a presentation similar to malignancies such as atrial myxoma and angiosarcoma as well as cytoskeletal pathologies such as ventricular aneurysms.[5] Most frequent site is the left ventricular wall and the transit of larvae to the myocardium is via coronary circulation as it is evident from this case. In chronological order, the areas involved are as such; 60% in left ventricle, 10% in right ventricle, 7% in pericardium, 6% in left atrial appendage and pulmonary artery each and about 4% in interventricular septum.[10] The most devastating complication of such a cyst is rupture which can cause cardiac tamponade or anaphylactic shock. It can also cause pulmonary and systemic embolus and can also compress coronary arteries.[11] An interesting finding in this case was the sub-endocardial attachment of the cyst which is contrary to the general rule. Most hydatid cysts in the left ventricle are located in the sub- epicardial site.[7] Additionally, the cyst has extended into the ventricular cavity and almost filling it. The case is in accord with a similar case of an intracavitary hydatid cyst in a child which required urgent surgical cystectomy, hence, emphasizing the need to urgent surgical removal of hydatid cysts in ventricular cavity.[12] As described previously echocardiography is sensitive in the diagnosis of cardiac cysts, a more sensitive tool as CT scan is often required for more accurate size and site determination as well as to rule out hepatic and pulmonary hydatid disease. The cardiac hydatid cyst should be considered as a surgical emergency and must be operated as soon as possible keeping in mind its disastrous consequences. Hence, surgery is the principal method of treatment which is followed by chemotherapy with albendazole.[2] To reduce spillage and chances of developing complications certain agents such as hypertonic saline, iodine and ethanol,

methylene blue are injected.[11] The major concern in this case was the acute right limb ischemia which we encountered. Another study matches this case of acute limb ischemia secondary to cardiac hydatid cyst and emphasizes the use of an urgent doppler or duplex scan's in prompt management to save the limb as well as suggesting that the limb ischemia can be denoted to an embolus from the parent cyst and causing disease sequalae.[3] However, in this case, the limb ischemia was beyond hope and an amputation was considered in patient's best interest. Concrete evidence to support that the embolus was from pre-existing cardiac cyst is lacking but we can say it with confidence, keeping in view the disease course, that it was likely a daughter cyst embolus responsible for the acute limb ischemic changes in this case.

CONCLUSION

The reason this case is unique is that left ventricular hydatid cyst is not reported in literature of our country. It is also unique for its extension into left ventricular cavity and associated acute limb ischemia. It is thus important to rule out cardiac echinococcosis while encountering hydatidosis in the liver or the lungs which can ensure timely management and prevent complications.

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