Clinical Case Report

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Echinococcosis in left ventricle: a case report

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Abstract

Introduction: Echinococcosis, also called hydatid disease, is a common parasitic infection of the liver. However, echinococcus lesions rarely involve the heart, especially in children.

Patient concerns: An 8-year-old child from grazing areas of northwest China was referred to our hospital for the complaint of inpersistent precordial chest pain and left upper quadrant pain for 3 years. Palpation showed hepatomegaly, abdominal palpable mass while inspection abdominal distension. Routine blood tests were within the normal ranges.

Diagnosis: Combining the life history in pasture area, imaging features and serology results, it was consistent with the diagnosis of cardiac echinococcosis.

Interventions: Surgery was performed to evacuate cyst liquid and remove the internal capsule of the cyst.

Outcomes: There was no cystic lesion in heart on ultrasound and her physical condition improved significantly after the surgery. The patient died of hepatic hydatid cyst rupture due to refusing high-risk surgical treatment and other treatment.

Lessons: We presented a rare case of cystic echinococcosis involving left ventricle in a child, and surgery is an alternative and effective therapy for this lesion due to the cyst rupture or leakage that can result in anaphylaxis. The typical imaging features of the cardiac echinococcosis on cardiac magnetic resonance are presented. Patient prognosis relies on proper treatment of all lesions.

Abbreviations: AE = alveolar echinococcosis, CE = cystic echinococcosis, CMR = cardiac magnetic resonance, CT = computed tomography, MRI = magnetic resonance imaging, US = ultrasound, WHO = World Health Organization.

Keywords: computed tomography, echinococcosis, hydatid disease, magnetic resonance imaging, parasitic infection

1. Introduction

Echinococcosis is a zoonosis, a disease of animals that affects humans. Human echinococcosis (also called hydatidosis, or hydatid disease) is caused by larval forms of the genus Echinococcus. *Echinococcus granulosus* causes cystic echinococcosis (CE), the form most frequently encountered; *Echinococcus multilocularis* causes alveolar echinococcosis (AE); *Echinococcus vogeli* causes polycystic echinococcosis; and *Echinococcus oligarthrus* is an extremely rare cause of human echinococcosis.[1] The liver is the most frequent location of echinococcosis, while the lung is the second most common location.[2] Echinococcosis can also be present in other organs although it is rare. According to World Health Organization (WHO) data, the incidence of cardiac echinococcosis was approximately about 0.03% to 1.1% in all cases of Echinococcosis.[2] Cardiac echinococcal cysts of the heart often require surgical treatment due to the risk of rupture and anaphylaxis. Although the mortality of cardiac echinococcosis is low, the morbidity of which is high.[3] Thus, accurate diagnosis of cardiac echinococcosis is critically important for effective treatment. Herein, we report a very rare case with left ventricular cardiac echinococcal cysts accurately diagnosed with cardiac magnetic resonance (CMR). The utility of CMR in the evaluation of this condition is discussed.

2. Case report

An 8-year-old child from grazing areas of northwest China was referred to our hospital for the complaint of nonpersistent precordial chest pain and left upper quadrant pain for 3 years. She had no palpitation and nausea. Palpation showed hepatomegaly, abdominal palpable mass while inspection abdominal distension. Cardiac auscultation showed sinus rhythm with a heart rate of 72 bpm without heart murmur. Her blood pressure was 96/53 mm Hg. Routine blood tests were within the normal ranges.

Ultrasound (US) examination demonstrated an echo-free lesion measuring 4×3.5 cm in the left ventricle with well-defined border. Serologic IgG test was positive for Echinococcosis. CMR was performed to characterize the lesion. CMR images revealed a cystic lesion adhering to and infiltrating the left ventricular anterolateral wall on T2-weighted images (Fig. 1). The cystic mass had water content inside with relatively thick wall. The wall had two layers ("double-line sign") with the inner layer being hypodense signal intensity and the outer layer being isodense signal intensity, which produced by the endocyst (inner layer) and the pericyst (outer layer). After administration of gadolinium, the cystic wall had minimal enhancement (Fig. 1A–C). Despite the left ventricle was deformed and the ventricular cavity became

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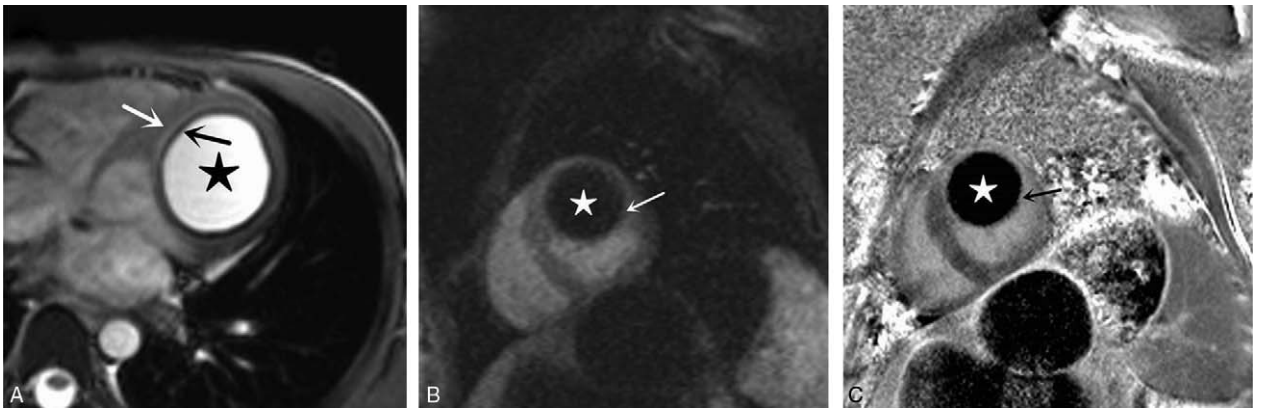

Figure 1. Cardiac magnetic resonance imaging shows the imaging features of the left ventricular echinococcosis (asterisk). (A) A cystic lesion with liquid content and visible cyst wall with "double line sign" (black and white arrows); (B, C) Two chamber view of left ventricle depicts the cyst wall with early contrast enhancement and minimal late gadolinium enhancement (arrow).

smaller due to mass effect of the cyst, the global left ventricular function is preserved within the normal range.

A noncontrast computed tomography (CT) was performed for the abdomen due to abdominal pain. Two large low-density cysts were detected. Additionally, one slightly lower density mass and one patchy lesion with minimal calcification were found. Within the scan range, a cyst with thick wall was noted in left ventricle, with the internal part of the wall being a slightly high-density ring consistent with calcium deposition (Fig. 2A, B).

After a multidisciplinary team discussion with hepatobiliary surgeons, cardiovascular surgeons and radiologists based on the imaging findings of the lesion in the left ventricle and liver, diagnosis of echinococcosis involving the heart and liver was suggested.[4] It was concluded that the hydatic cyst in the left ventricular wall was extremely likely to rupture, and heart surgery was needed to remove the lesion.[5] During the surgery, hypertonic saline was injected in the cardiac cyst several times after cyst liquid was evacuated, and then the internal capsule of the cyst was removed. Diagnosis of cardiac echinococcosis was confirmed by histologic examination. The lesions in the liver were treated with albendazole, and the radical surgical removal of the echinococcosis in the liver was planned 3 months after the heart procedure.

The present study was approved by the Ethics Committee of the West China Hospital of Sichuan University, and the patient information was anonymized and deidentified before analysis. Informed consent has been obtained.

3. Discussion

Echinococcosis, caused by the larvae of the genus echinococcus in humans, is a rare infection. Among the recognized echinococcus genus, *E. granulosus* and *E. multilocularis* are of most clinical importance, which cause CE and AE in humans, respectively. The most commonly affected organ is the liver (70%) and followed by

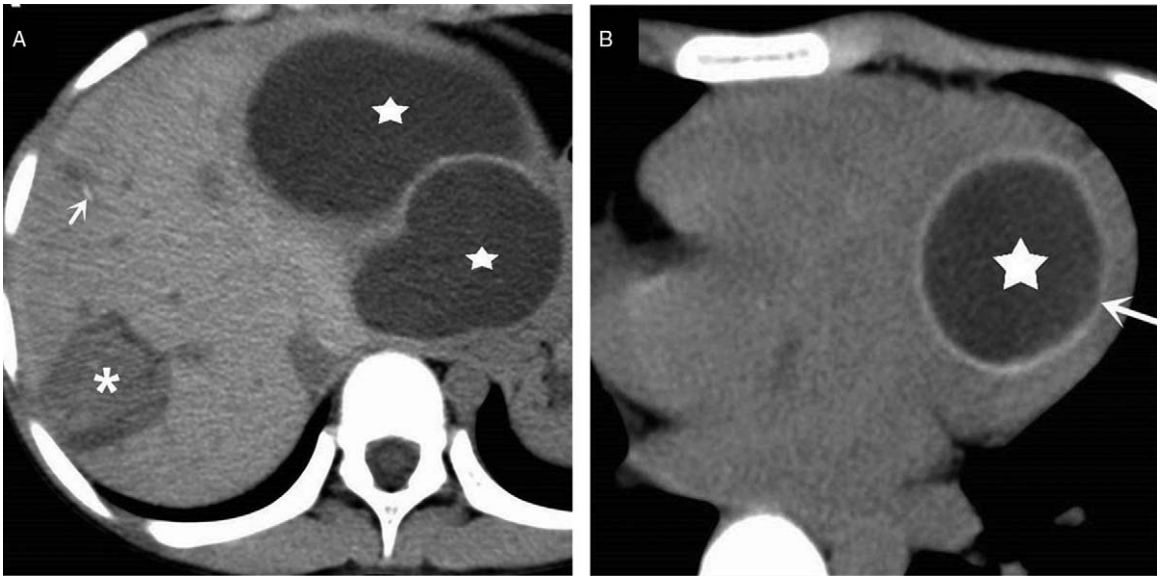

Figure 2. A noncontrast computed tomography imaging shows the imaging features of the abdomen and the left ventricular echinococcosis (asterisk). (A) Two cystic lesions (asterisk) with thin walls, and two slightly lower density lesions, one of which contained heterogenous solid (light asterisk) and another had minimal calcification (arrow). (B) Scanned range shows a cystic lesion in left ventricle, which contained an attenuation density with slightly high-density ring (arrow) due to calcium deposition.

the lungs (20%), while other organs are less frequently involved. Cardiac echinococcosis involved approximately in only 0.03% to 1.1% in all cases of Echinococcosis.[1] In all cases with cardiac involvement, the left ventricle is the most frequently affected site (55–60% of cases), followed by the right ventricle (10%), pericardium (7%), pulmonary artery (6%), and left atrial appendage (6%) and involvement of the interventricular septum is reported to be rare (4% of cases).[6]

According to the reached consensus on the diagnosis and treatment,[7] CE and AE differ observably in their behavior and the characteristic of imaging. AE tends to be tumor-like mass with irregular margins and heterogeneous contents with varied attenuation, such as calcifications, necrosis, and parasitic tissue, mainly in the liver.[8] The imaging features in other organs are substantially the same as those in the liver; however, there is no report about involving heart of alveolar echinococcosis. While the imaging feature of CE is a well-defined cystic or multicystic mass containing liquid or septa and some with calcification of the cyst wall, matrix, or both, mainly in the liver or lung.[9] Most cases of cardiac cystic echinococcosis were occurred on adult patients,[10–17] while only 19 cases were on children.[18–22] For the 19 reported cases in children, there were 8 cases of cardiac echinococcosis involving left ventricle. In the 19 cases, 6 cases were associated with extracardiac system involvement, while heart were the sole target organ in 13 cases.[22]

According to literature, 15 cases of cardiac echinococcosis reported in children had imaging examinations, of which only one case had magnetic resonance imaging (MRI) and CT studies.[19] In literature, the cardiac hydatid cysts mostly characterized multiple, thin walls, and more to invade the myocardium or pericardial.[16] CMR revealed a multiple septa hydatid cysts with calcification in apex of heart and local thickening of pericardium.[14] Chest CT showed well-defined cystic or multicystic mass with calcification of the cyst wall, internal septa, or both, which were characteristic imaging features for cystic echinococcosis.[17] Fiengo et al[19] reported a 11-year-old girl with large lesion of left ventricular-free wall with a hypointense rim on T2-weighted MRI scans, while other organs had no cysts lesion. In our case, CMR revealed a uniloculated hydatid lesion from the myocardium of left ventricular antero-lateral wall that is hypointense on T1-weighted images and hyperintense on T2-weighted images, with the inner layer being hypodense ring and the outer layer being isodense signal intensity on T2-weighted MRI scans, which demonstrated by T2-weighted MRI representing type CE1 cysts following the classification of the WHO.[1] Besides, in abdominal CT scan, two large low-density cysts and two slightly lower heterogeneous contents mass with minimal calcification were seen in liver.

Surgical treatment is preferred for cardiac hydatid disease even for asymptomatic cases because rupture of the cystic lesion may causes anaphylaxis and metastatic disease.[23] To decrease the risk of recurrence, antihydatid drugs are essential as supplemental medical therapy.[24] In addition, these patients should be followed up against recurrent cyst using serologic and US examination in the first 5 postoperative years.[25] In our case, the surgical excision was performed to remove the cardiac cyst under cardiopulmonary bypass. After consultation with pediatric surgeon, surgical removal of the hepatic cystic lesions was recommended 3 months after the cardiac surgery, and albendazole therapy should be given at the same time. However, about 2 weeks after discharge, the patient was readmitted to our institution due to abdominal pain and high heart rate (220 bpm). Bedside ultrasound in insensitive care unit (ICU) suspected hepatic hydatid rupture. After consultation with pediatric surgeon and communication with the patient's family, the surgery is planned. Through careful consideration, the family refused surgical treatment due to high risk of the procedure and asked to be discharged without any further treatment. Tefera et al[26] reported a case with a large hydatic cyst within the interventricular septum in a 9-year-old girl, who underwent surgery under cardiopulmonary bypass. There was no cyst in other organs or tissues. The patient recovered well after the surgery. To be concluded, the location, number, and size of the cysts should be considered for choosing appropriate treatment, such as staging operation or combined operation at the same time.

Early detection of cardiac echinococcosis is often difficult, for there is often no marked symptoms before the cyst is large enough to compress adjacent structures or to rupture resulting in infection.[1] Common symptoms of CE include chest pain, palpitations, and dyspnea, which are not specific. In clinical practice, we should combine the life history in pasture area, imaging features, and serology results. For the diagnosis of cardiac echinococcosis, noninvasive imaging such as CT, MRI, and US plays a critical role in making the diagnosis.[27] There is no routine blood tests specific for echinococcosis; however, serology may be used as a first-line test and plays an important role for the correct diagnosis of CE.[28]

US is a current reliable method for diagnosing cystic lesion and a classification for evaluating and staging of CE in liver.[29] However, it is necessary to do CT or MRI scanning to find additional information on the accurate location of lesion and relation of the lesion with extracardiac structures, such as liver, lung, bone, muscle, and for detecting calcified cysts.[30] MRI may have some advantages over CT detecting the soft tissue for surgical removing of the cysts and is also superior in identifying stage-defining features of cysts seen on US.[29] Antibody assays are useful to confirm the presumptive radiologic diagnosis, although some patients with CE do not demonstrate a detectable immune response.[28]

In conclusion, in patients with left ventricular cystic lesion and grazing areas life history, cardiac echinococcosis should be considered. CMR is a useful and important tool for the diagnosis and presurgical evaluation.

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