

## Short Communication

# Uncommon Localizations of Echinococcosis: Primary Cerebral and Cardiac Hydatid Cyst

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## Abstract

Hydatid cyst disease is endemic, occurs following accidental ingestion of eggs released from the intestine of the definitive hosts and commonly invades the liver and the lungs. A man with a large cyst intracranial, with symptoms of intracranial hypertension and a solitary cardiac cyst is presented. The cyst was removed after suitable craniotomy. A review of the literature is presented.

## ABBREVIATIONS

CT: Computed Tomography; MRI: Magnetic Resonance Imaging; 3DTTE: Three-Dimensional Transthoracic Echocardiogram

## INTRODUCTION

There are six types of Echinococcus causing hydatid cyst: *E. granulosus*, *E. multilocularis*, *E. vogeli*, *E. oligarthrus*, *E. shiquicus* and *E. felidis*. *E. granulosus* is the most common species in humans [1]. Hydatid cyst disease is endemic in the Middle East and Mediterranean countries in addition to South America, North Africa and Australia [2,3]. Its incidence varies globally with highest rates in Kenya (220 cases per 100,000 persons) and other Middle East regions [3].

It occurs in humans following accidental ingestion of eggs released from the intestine of the definitive hosts (canines) [4]. Hydatid cysts most commonly invade the liver (60%–80%) and the lungs (20%–25%), but they can be located in the all parts of the body [5,6], presenting in the brain (0.4-1%) and in the cardiac muscle (0.02-1.1%) infrequently [5].

## MATERIALS

A man of 33 year-old presents who is complaining of a blunt headache and symptoms of increased intracranial pressure such as nausea and vomiting. He was in a good general state of health, fully conscious and oriented and his neurological examination revealed mild hemiparesis on the right side.

His cranial CT revealed an intracranial cyst (Figure 1) that although the lesion embedded within the dominant hemisphere, he was intact in speech. Hydatid disease was suspected, several

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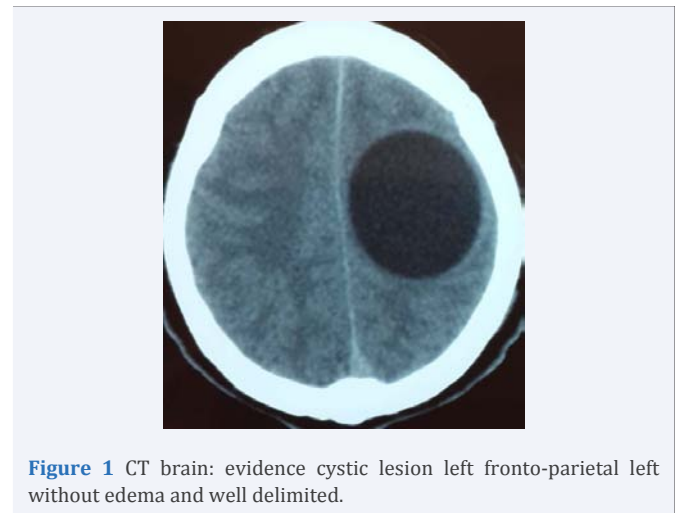
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## Keywords

- Hydatid cyst
- Echinococcus
- Brain
- Cardiac

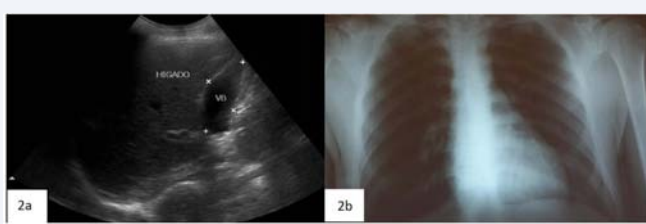


**Figure 1** CT brain: evidence cystic lesion left fronto-parietal left without edema and well delimited.

imaging studies were carried out, which were negative (Figure 2a,2b). At the same time samples were taken for Ig-ELISA serology for hydatidosis.

He was operated on by a left fronto-parietal craniotomy and the cyst was removed (Figure 3) by the Dowling- Orlando technique with the aid of gravity without rupture. The postoperative period was uneventful with resolution of hemiparesis.

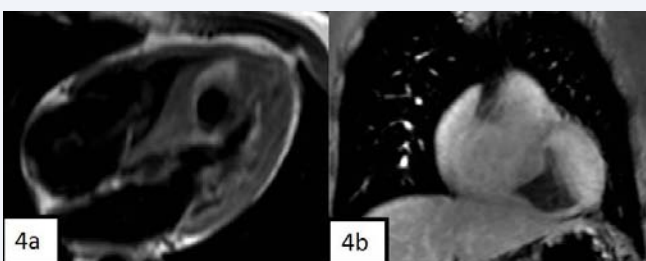
Cardiac evaluation done postoperatively showed no evidence of patent ductus arteriosus or patent foramen ovale although surprising evidence of a cardiac cystic lesion, he underwent a cardiac MRI (Figure 4a,4b) where a cystic lesion of thin wall is observed at the level of the interventricular septum. He never had heart symptoms and studies of cardiac function within



**Figure 2** (a) - Abdominal ultrasound where no cystic lesions are present in the liver. (b) - Chest X-ray where no cystic lesions are evident.



**Figure 3** Hydatid cyst obtained by craniotomy according to the Dowling's technique.



**Figure 4** Cardiac MRI (a) - In T1, hyposignal lesion with a thin wall at the level of the interventricular septum. (b) - Find does not enhance after contrast, compatible with hydatid cyst.

normal ranges, which is why it was decided to observe this lesion.

Histopathological examination confirmed as the primary hydatid cyst, as well as serology, confirming *Echinococcus granulosus*. Medical therapy was completed by albendazole.

## DISCUSSION

*Echinococcus granulosus* is a parasite frequently isolated and is responsible for cystic lesions, whereas *E. multilocularis* is more involved in visceral lesions and is called alveolar echinococcosis. *E. granulosus* is mainly kept in a dog-sheep-dog cycle, however several other domestic animals may be involved, including goats, pigs, horses, cattle, camels and yaks. Humans are accidental intermediate hosts and are not capable of transmitting disease [7].

Hydatid cysts do not have a specific symptom. When they

reach larger sizes, they cause mass effect to the involved organ. The growth speed of the cysts varies according to the involved organ. In the past, hydatid cysts were thought to be slow-growing lesions. Many studies reported that hydatid cysts grow in human beings by approximately 1 cm in a year, whereas others reported that they grow by approximately 4–5 cm a year [6].

Cerebral echinococcosis is observed in 0.5–3% of patients with echinococcosis and is mostly seen in children and young adults [1,2], this high incidence in children is probably related to patent ductus arteriosus. There is a male preponderance, with a male: female ratio of 1.5:1. The most common species diagnosed in the brain are *E. granulosus* (97.1%) and *E. multilocularis* (2.9%) [1].

In the brain, hydatid cysts are usually confined to the supratentorial compartment; in the region supplied by middle cerebral artery being the parietal lobe is especially involved [8].

Clinical signs and symptoms in patients with cerebral echinococcosis are headache, increased intracranial pressure, papilledema (63%), optic atrophy, nausea, vomiting, cranial nerve palsy, seizure (24%), focal neurological findings, cognitive deficit, ataxia, speech disorder, visual disturbances, head swelling, difficulty in swallowing, and chorea. The mortality rate is 10% for cerebral echinococcosis. However, the mortality rate is higher in patients with multiple lesions than in those with a single lesion (13 vs. 7%) [1].

Non complicated or non infected lesions demonstrated smooth, well-defined, thin-walled, spherical, homogeneous appearance, without calcification, peripheral o edema or contrast enhancement which had inner density/intensity similar to CSF on CT and MRI [9].

MRI spectroscopy helps in the noninvasive diagnosis of the hydatid cyst, peaks at 2.4 ppm have been found as a noninvasive marker of cestodal cysts. Alanine and acetate peaks have also been reported. Creatine is usually absent, unlike cysticercal cysts [10].

Intracranial hydatid cysts may be classified as primary or secondary. The primary cysts are formed as a result of direct infestation of the larvae in the brain without demonstrable involvement liver or lungs. The primary cysts are fertile as they contain scolices and brood capsules, hence rupture of primary cyst can result in recurrence. The secondary multiple cysts results from spontaneous, traumatic or surgical ruptura of the primary intracranial hydatid cyst and they lack brood capsule and scolices. The secondary intracranial hydatid cysts are, therefore, infertile and the risk of recurrence after their rupture is negligible [2,8].

Long-term follow-up confirms that intracranial hydatid cysts should always be surgically removed without rupture; the outcome remains excellent in these cases. Wide scalp flap and dissection of the atrophic cortex, lowering the head of the operating table to benefit from gravity and the use of warm saline between the hydatid cyst and brain parenchyma to deliver the hydatid cyst is the most frequently used method, known as Dowling's technique, with an 88 % success rate reported [11]. There have been other techniques proposed, such as Arana-Rodriguez's technique, but these are not frequently implemented [8].

On the other hand, the heart is less often affected by echinococcosis compared to other organs. According to World Health Organisation (WHO) data, the heart accounts for approximately 0.03% to 1.1% of all hydatid cyst cases [12].

There is no consensus on how cysts spread to the heart. Cysts involving the left side are believed to reach that region via coronary circulation. Cysts that pass through the liver reach the right heart via the venous system and infect the right atrium, right ventricle and interatrial septum. In systemic circulation, some larvae may pass into the coronary circulation and settle into a subepicardial site of the left ventricle and interventricular septum. This may explain why extracavitary echinococcosis is seen mostly in the left ventricle [12].

Cardiac hydatidosis usually presents with nonspecific symptoms such as chest pain, palpitations, and dyspnea, our patient was asymptomatic. Diagnosis of cardiac involvement is mainly established by echocardiography as it is a noninvasive test to demonstrate cysts.

Contrast-enhanced CT and MRI remains the primary diagnostic modality, three-dimensional transthoracic echocardiogram may be the next step to corroborate the diagnosis of cardiac hydatidosis [13].

Finally, the gold-standard drugs to administer in adjuvant therapy are benzimidazoles [14], as the Albendazole with 10-15 mg/kg/day dosage, for at least 1 to 6 months [5,14]. It is effective in sterilizing the cyst, decreasing the risk of anaphylaxis and reducing the recurrence rate. Corticosteroids may help control perilesional edema, while anticonvulsants are used prophylactically [15].

## CONCLUSION

Hydatidosis is a very rare disease in the brain and heart. A high index of suspicion is therefore required in endemic areas despite the availability of advanced imaging techniques.

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