

## ECHINOCOCCOSIS CEREBRI: REPORT OF TWO LESS COMMON CASES

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**Summary.** Cerebral hydatid disease is very rare and occurs in about 2% of all echinococcosis cases. We present two less common cases of echinococcosis and the problems related to differential diagnosis. The diagnosis was based on clinical findings CT scanning, MR imaging, ultasonography and IgG ELISA test..

**Key words:** Cerebral echinococcosis, multiple hydatid cysts

### Introduction

Echinococcosis is a widespread parasitic disease caused by the larva of *Taeniae chinococcus*. Hydatid disease is endemic in the Middle East, Mediterranean countries, South America, North Africa and Australia (1,2). The dog is the primary definitive host of the adult tapeworm. Intermediate hosts for the larval stage include sheep and man. Ova are excreted in dog feces and contaminate herbage eaten by sheep. Man is infected either by eating food contaminated by ova, or by direct contact with infected dogs (3).

Unlike *echinococcus multilocularis* which produces invasive lesions of firm consistency, full of connective tissue and jelly-like material, *echinococcus granulosus* produces cystic lesion(4). *Echinococcus granulosus* is manifested in humans as the intermediate host by the development of cysts in the liver, lungs, heart and brain. Cerebral hydatid disease is very rare and occurs in about 2% of all echinococcosis cases (3,5,6). The most common sites of involvement are the cerebral parenchima and subarachnoid spaces after secondary involvement. CT density of the cyst is similar to CSF, and there is little surrounding edema (2,3). The brain is infected because the liver and the lungs as primary sites fail to act as a barrier. Despite the improvement in hygiene measures and the benefits of prevention, echinococcosis remains medical problem all over the world. Montenegro with a population of 650,000 has implemented extensive health education programmes in recent decades but echinococcosis appears as an unsolved medical and social problem. According to the latest statistical data from the period of 40 years (1951 – 1991) 1965 patients were operated due to the *echinococcus granulosus* in various organs; in liver 34%, lungs 35% and in other organs less than 10% (6).

### First case report

A 16-year-old farmer boy was admitted at Department of Neurosurgery in December 2000 with a history of severe headache, vomiting and drowsiness. For two years he had periodically had dull headaches. When he played football he noticed his right leg was clumsy. Once he had an epileptic fit, which was recognized by local neuropsychiatrist as a generalized tonic clonic seizure. On admission he appeared to be confused, drowsy and disoriented in time. On neurological examination a mild right spastic hemiparesis was found. CT of brain revealed large cystic change in the left temporal, parietal and occipital lobes without pericystic oedema. Three *echinococcus* cysts were completely independent of each other, and in contact in some sites (Figs. 1 and 2).

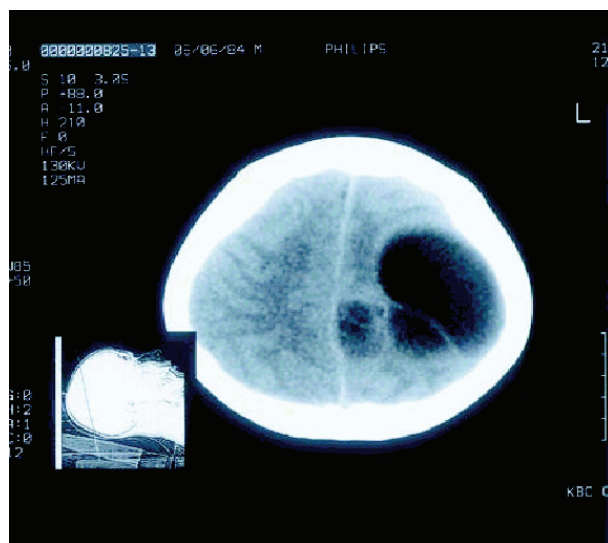


Fig. 1. CT scan of three independent hydatid cysts in brain

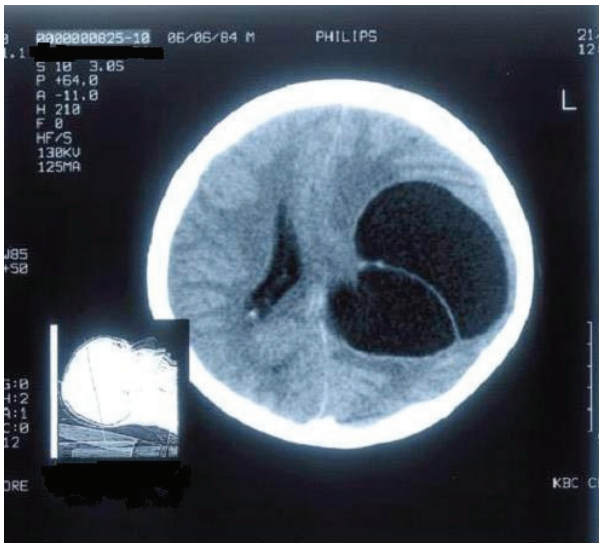


Fig. 2. CT scan of brain- lower level

Ultrasonography, CT scanning, and X ray of other organs (lung, liver, kidney etc) did not show any cystic formation. IgG ELISA test was negative. Mild eosinophilia was found. The patient was operated by using the classic manoeuvre by Dowling and Orlando (7) with lowering the position of head. During operation we found three independent hydatid cysts (size of 11cm, 5cm and 4cm), with separate ecto and endo cysts, which we removed totally without rupture. Hystopathological examination confirmed hydatid cysts. The postoperative course was good. The patient was commenced on antiepileptic medication and followed up 8 years after surgery, but there was no recurrence (Fig. 3). We checked the other organs but the results were normal.

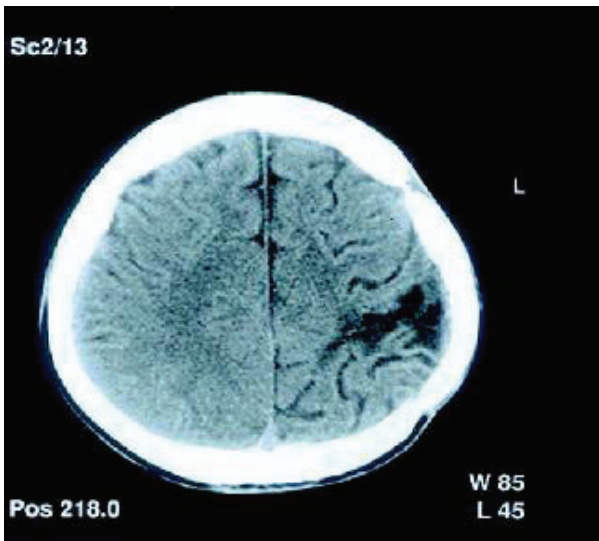


Fig. 3. CT scan finding after surgical treatment

**Second case report**

A 22 year old man was admitted with symptoms of embolic stroke, moderate right hemiparesis and expres-

sive aphasia. Four days before admission he experienced TIA, with monocular blindness and dysphasia. Three years before he had been operated on for liver hydatid cyst. On examination: echocardiography -cyst in the left atrium was located; lung CT scan - multiple hydatid cysts in the mediastinum and left apex (Fig. 4); brain MRI and CT scan - cerebral infarct in the distribution of middle cerebral artery (Fig. 5). The serodiagnosis of hydatid disease by indirect hemagglutination test was positive at 1/310 titer. The patient was presented to thoracic surgeon and cardiovascular surgeon, but he and his family refused any kind of operation.

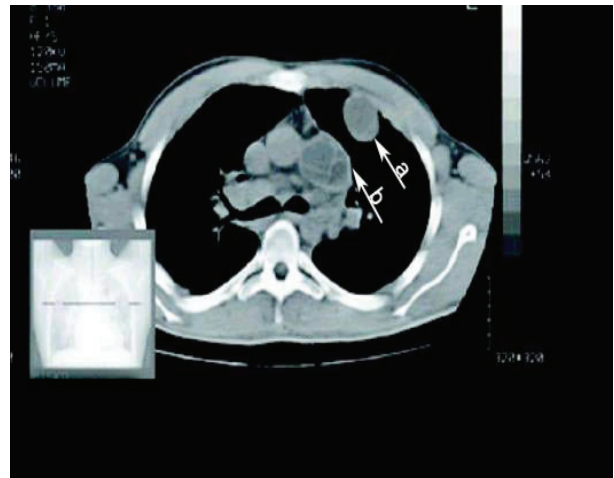


Fig. 4. Lung CT scan- multiple hydatid cysts (arrow)

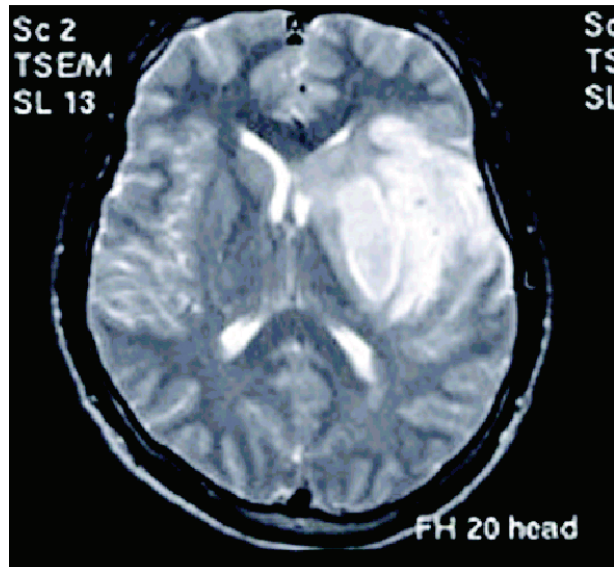


Fig. 5. Brain MRI- cerebral infarct in the distribution of middle cerebral artery

**Discussion**

The clinical picture of cerebral hydatid disease is similar to those of other space occupying lesions, with increased intracranial pressure and focal neurological symptoms; the latter may be worse due to the large size

of cysts or due to interference with pathway of CSF flow. Hydatid cysts of brain are usually single primary and large and are more frequent in children and adolescent than in adults. Lunardi et al. (8) assumed that the occurrence of primary cerebral hydatid cysts in children might imply a communication between the right and left side of the heart. These researchers also reported two cases of children with a single cerebral hydatid cyst that contained a daughter and that ruptured during operation. The primary cysts are formed as a result of direct infestation of the larvae in the brain without demonstrable involvement of other organs. In primary multiple cysts, each cyst has a separate pericyst with brood capsule scolices and these originate from multiple larvae affecting brain crossing gastrointestinal tract, liver, lung and right side of heart without affecting them. The primary cysts are fertile as they contain scolices and brood capsules, hence rupture of primary cyst can result in recurrence. Nurchi et al (9), while reviewing the literature, found only eleven reported cases of primary multiple hydatid cysts.

The secondary multiple cysts result from spontaneous, traumatic or surgical rupture of the primary intracranial hydatid cyst and they lack brood capsules and scolices. Turgut M. Benli K. Eryilmaz M. (10) present an extremely rare case of secondary multiple cerebral echinococcosis caused by presumed intracerebral and arterial embolism of cardiac hydatidosis in a 7-year-old girl. The first manifestations were symptoms of raised intracranial pressure. Before the primary ruptured echinococcosis cyst was detected in the myocardium of the left ventricle, the patient underwent nine operations for hydatid embolism affecting the brain and femoral artery and was treated with concurrent mebendazole therapy. The com-

bined therapy would not have been successful without removal of intracardiac hydatid cyst.

Treatment of cerebral hydatid cysts is surgical removal of the intact cyst in toto, without rupture to prevent recurrence and allergic reaction. Adjunctive medical treatment with Albendazole therapy may be used in daily dose of 15mg/kg x 28 days, taken with fatty meal to enhance absorption (same dose for pediatrics) given for 3 month, and repeated if necessary.

In our first case the main symptoms were headache, vomiting, mild right spastic hemiparesis and epileptic fit. Diagnosis was made on the basis of CT scan with a finding of three completely independent echinococcus cysts. Other differential diagnoses included a single cerebral cyst, which contained daughter cysts as well as other space occupying lesions (arachnoid, ependymal and other cystic formation in the brain) (4, 6, 8, 11). Some types of brain tumors with cystic formation made the differential diagnosis more difficult. There have been rapid developments in the surgery of these cysts in an effort to prevent post-operative complications. In our case by using a Dowling technique we were able to evacuate cysts without complications. Our second case was a young adult with symptoms of embolic stroke and hydatid cysts in the cardiac left atrium, lung and mediastinum.

## Conclusion

Hydatid cysts of the brain affect children and young people. Although it is a very rare disease, it must be taken into consideration in areas of low level of social and economic development. Sometimes echinococcosis appears with less common clinical course, which makes the differential diagnosis more difficult.

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## EHINOKOZA MOZGA: PRIKAZ DVA RIJETKA SLUČAJA

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*Kratak sadržaj: Moždana ehinokokoza je rijetko oboljenje i javlja se kod 2% svih slučajeva ehinokokoze. Mi prikazujemo dva ređa slučaja i diferencijalno dijagnostičke probleme u vezi sa njima. Dijagnoza se bazira na CT skenu, MRI nalazu, ultrasonografiji i IgG ELIZA testu.*

*Ključne reči: Moždana ehinokokoza, multiple hidatidne ciste*