Case Report

# Unusual presentation of echinococcal cysts

***Farukh Qureshi,*** *FRCS(Edin), FEBU,* ***Nabeel Maalbared,*** *MBBS, Facharzt.*

## ABSTRACT

We report a case of echinococcal disease affecting the brain and the kidney. The case is unusual because of multiplicity of the intracerebral lesion and sparing of the liver and lungs. The intracerebral lesion was presented with epileptic convulsions. This was successfully treated surgically. Subsequently renal echinococcal cysts were identified. These were treated by partial nephrectomy due to the location of the cysts. The patient is disease free at 8 years.

**Saudi Med J 2003; Vol. 24 (7): 781-782**

Echinococcal cyst disease is well known in Australia, North and East Africa, Middle East, Turkey, Southern Europe and South America.[1,2] Human hydatid cysts are caused by the larval form of *Echinococcus granulosus* (*E.granulosus*) and *Echinococcus multilocularis (E.multilocularis).* Multiple cysts are not rare in *E.granulosus* infections though they are usually unilocular as distinct from the multilocularis, externally budding cysts of *E.multilocularis*.[3] The tapeworm lives in the small intestine of the dog and other canids, which is the definitive host for the parasite. It is here that it lays eggs, which are then excreted into the lumen of the bowel and passed in the feces. Ingestion of these eggs infects both humans and sheep. After ingestion the eggs hatch and the free oncospheres penetrate the intestinal mucosa of the host to reach the liver via portal circulation. Embryos that succeed in passing through the hepatic and pulmonary filtering systems reach the brain and kidneys via the systemic circulation. Although hydatid cysts can develop practically at any site in the body, approximately 70-80% of cysts are located in the liver and the lung. Both renal and cerebral hydatids are uncommon presentations of echinococcal disease and occur in 2-3% of cases.[4] The cyst is composed of 2 layers: the outer elastic lamina and the inner germinal membrane. The germinal membrane produces the brood capsules, which may become detached to form hydatid sand.

Case Report. A 36-year-old male from a sheep rearing community was admitted with acute onset of epileptic convulsions. Neurological examination did not reveal any deficit. Subsequent investigation by computerized tomography (CT) scan revealed multiple cysts in the frontal lobe **(Figure 1)**. These were non-enhancing of cerebrospinal fluid density with minimal surrounding edema. A provisional diagnosis of hydatid disease was made. Albendazole was given prior to surgery and continued postoperatively at 800 mgs per day for 28 days. Craniotomy was performed. Following cyst aspiration and intracystic injection of hypertonic saline, the cysts were excised completely and confirmed by histopathology.

In the post-operative period, CT scan of the chest and abdomen was arranged as part of screening for other site involvement. This revealed multi-loculated cysts at the upper pole of the left kidney **(Figure 2)**. These cysts were parenchymal with no evidence of communication to the collecting system. At operation the cysts were confined to the upper pole deforming the outline of the left kidney. The entire kidney was mobilized on its pedicle and packed with iodine soaked gauze. Partial nephrectomy was carried out. The cysts were not exposed at any time during the procedure. The diagnosis was confirmed by histopathology. Post-operative recovery was uneventful and the patient went home after 10 days.

From the Department of Urology (Qureshi), King Fahd Armed Forces Hospital, Department of Neurosurgery (Maalbared), King Abdul-Aziz Hospital & Oncology Center, Jeddah, *Kingdom of Saudi Arabia.*

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Address correspondence and reprint request to: Dr. Farukh Qureshi, King Fahd Armed Forces Hospital, PO Box 9862, Jeddah 21159, *Kingdom of Saudi Arabia.* Tel. +966 (2) 6653000. Fax. +966 (2) 6652469. E-mail: farukhqsa@hotmail.com

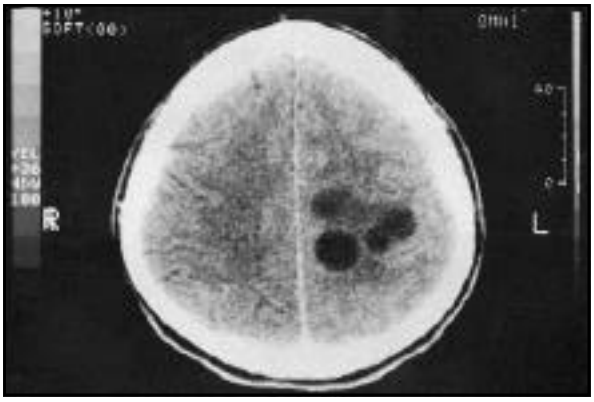

Figure 1 - Computerized tomography scan of the brain showing multiple cysts in the cerebrum.

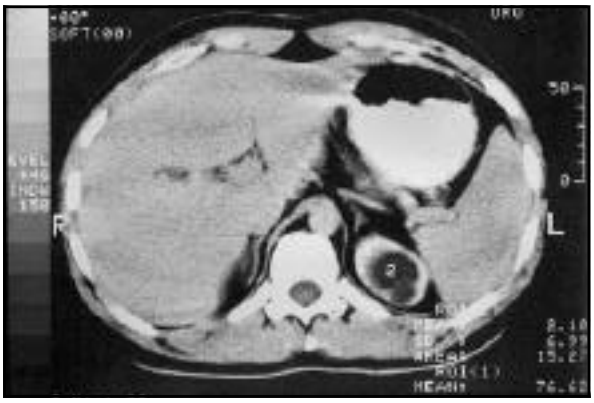

Figure 2 - Computerized tomography scan of the abdomen showing multiple left renal cysts.

Discussion. Cerebral echinococcal disease is usually supratentorial and often involves the middle cerebral artery due to the embolic nature of the infestation.[5-7] Patients present with hemiparesis, speech disorders or epileptic seizures. On the other hand renal hydatids may be completely asymptomatic and found incidentally. Eosinophillia is usually due to rupture of the cyst and is a non-specific finding. Skin testing (intradermal skin testing) is considered to be unreliable due to the low sensitivity and specificity.[5,6,8] Serological tests (indirect hemaglutinin test and enzyme linked immunoassay test) are helpful in the diagnosis but negative serology does not rule out echinococcal disease. Only hydatiduria is pathognomonic for echinococcal disease with renal involvement.[1,9] Echinococcal cysts of the kidney are easy to identify on ultrasonography if the contents are of low echogenicity. Typical CT findings include a non-enhancing cyst with minimal edema. Atypical appearances include irregularity of cyst wall contour, enhancement of surrounding rim, heterogeneity of cyst contents, surrounding edema and globular as opposed to curvilinear calcification.[10,11] Computerized tomography and magnetic resonance imaging are more sensitive in differentiation of solid renal tumors and simple or complicated renal cysts. Open surgical removal of the cyst remains the treatment of choice. Percutaneous aspiration of the renal hydatid cyst has been described in the literature,[12] but carries the risk of anaphylactic shock and dissemination of daughter cysts. Marsupialisation is only recommended in patients with a solitary kidney. In these cases the cyst has to be aspirated and the cavity to be filled by a scolicidal agent for example hypertonic saline, 0.5% silver nitrate, 1% iodine or 95% alcohol. Many of these agents may be dangerous if there is communication between the cyst and vital structures. In our report hypertonic saline was used for the cerebral hydatid with minimal side effects. Larvicidal agents such as mebendazole and albendazole may reduce the fertility of the cyst especially when a cyst ruptures. They are not useful as the sole treatment of cerebral cyst.[3] Medical treatment should also be given pre and post-operatively with marsupialisation and percutaneous management of hydatid cysts. Higher response rates have been achieved with the use of albendazole due to its active metabolites (albendazole sulfoxide) as it can reach higher and more effective concentrations in the serum, cystic fluid and the cyst wall.[12]

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