

SHORT REPORT

Multiple Infected Cerebral Hydatid Cysts Treated with Albendazole

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Humans acquire echinococcosis by ingesting viable parasite eggs with their food. The parasite eggs are distributed via local environmental contamination by the feces of tapeworm-infected canines. Once in the intestinal tract, the eggs hatch to form oncospheres that penetrate the mucosa and enter the circulation. Oncospheres then encyst in host viscera, developing over time to form mature larva cysts. The hydatid cysts of *Echinococcus granulosus* tend to form in the liver or lungs but may be found in any organ of the body, including the bones, heart and brain (1).

Multiple intracerebral hydatidosis is a rare disease. Brain involvement is seen in 2% of all *E. granulosus* infestations. In many cases, a cerebral hydatid cyst is associated with a hepatic cyst. The diagnosis is established clinically and by serology and imaging techniques (2). When the presence of cysts is combined with an appropriate residential or travel history or an occupation such as sheep raising in areas where the parasites are endemic, there are reasonable grounds to suspect infection. Cerebral hydatid cysts are usually single, spherical and unilocular (3,4). A round, cystic lesion is detected on CT in hydatid disease (5). Cerebral hydatid cysts should be removed without rupture (6). Nevertheless, only half of these cysts can be removed totally unruptured (7). Medical treatment may be required in some risky cases (8). In such cases, all patients

have to receive mebendazole therapy (2). In the literature, to date, there are very few papers reporting such multiple cerebral hydatid cysts treated with only albendazole (9,10). Therefore, we present this rare and interesting case.

Case Report

A 65-year-old male-farmer was admitted to our department due to complaints of epileptic attacks and right-sided weakness. Two years before, he had been operated on for liver hydatid cyst (Figure 1). His neurological examination revealed right-sided hemiparesia. Other systemic radiological examinations were normal for hydatid disease. However, cranial MRI (0.5 T) revealed 3 isointense cystic lesions, 0.5-2 cm in diameter, with contrasted ring and perilesional edema in his left parietal lobe (Figure 2a). The serodiagnosis of hydatid disease by indirect hemagglutination test (Laboratories Fumouze, Levallois-Perret, France) was positive at 1/320 titer. The most likely diagnosis was infected cerebral hydatid cyst and surgery was planned. However, the patient refused the operation and therefore he was treated with albendazole in 6 cycles of 400 mg twice a day for 4 weeks, followed by a 2-week rest period without therapy. On MRI (1.5 T) performed 18 months later, interestingly all of the cystic masses had totally disappeared (Figure 2b).

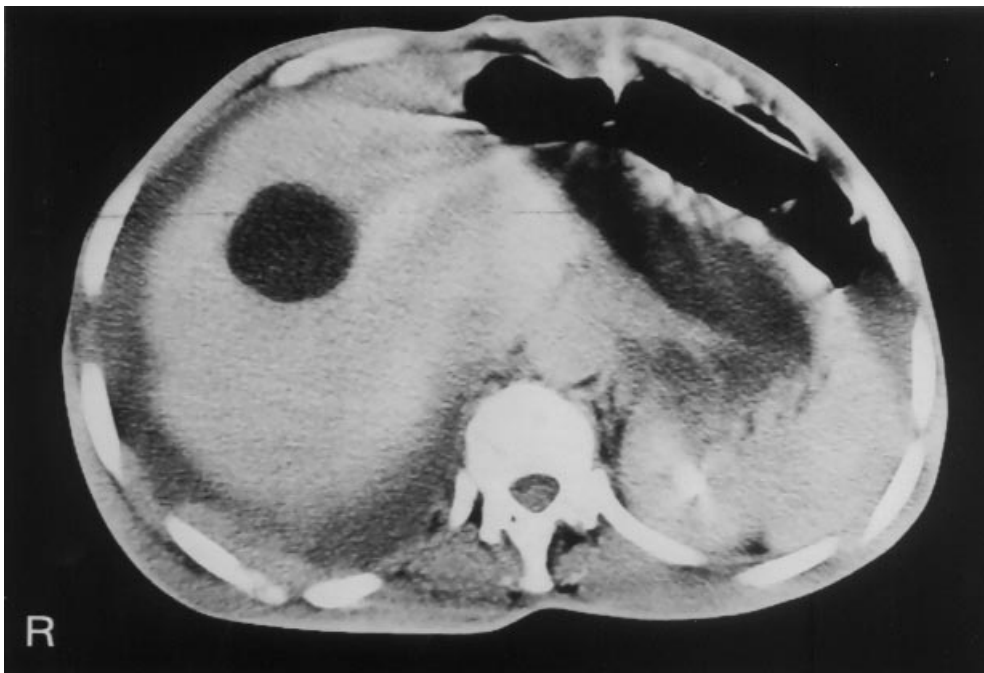


Figure 1. A hydatid cyst, 4 cm in diameter, is seen in the patient's liver on CT.

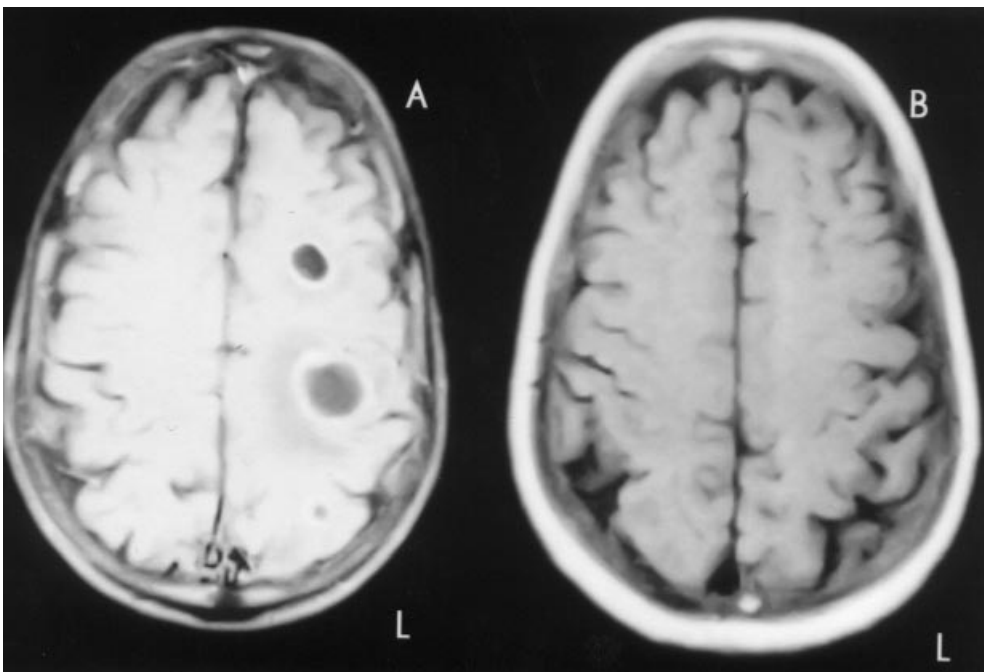


Figure 2a. Axial cranial MRI (0.5 T) shows 3 isointense cystic lesions, 0.3-2 cm in diameter, with contrasted ring and perilesional edema, in the left parietal lobe.

Figure 2b. Cystic masses are not seen on control MRI (1.5 T) performed 18 months later.

Multiple intracranial hydatidosis is a rare disease, with serious neurological manifestations, high recurrence and a mortality rate comparable sometimes to malignant disease (9). Infected cerebral hydatid cysts, however, have been reported very rarely so far (10). *Echinococcus granulosus* infestation of the central nervous system may be primary or secondary and has been estimated to be very low (2%). After the ingestion of contaminated food, hexacanth embryos migrate by the portal system to the liver and later to the lungs and other tissues and may even metastasize to the central nervous system. The cysts are almost always found in the cerebral hemisphere.

They favor the region supplied by the middle cerebral artery, especially the parietal lobe (11). Sharply demarcated, spherical and intraparenchymal cysts may reach a large size, causing neurological symptoms. Despite hydatid cysts of the brain usually being single (4), they may also be multiple (3). Infected hydatid cysts may contain an isointense or hyperintense cavity and may be covered with a contrasted ring-shaped zone and perifocal edema on T1-weighted MRI (12). According to the literature and the patient's data, infected hydatid cysts was the most likely diagnosis in the presented case. It is generally accepted that patients with this disease rarely

exhibit focal neurological deficit unless herniation occurs (13). Our patient, however, was symptomatic because of the infected nature of the cysts. In the literature, a report advocates the treatment of such a multiple intracranial hydatid cyst disease with medication. Albendazole was found to have favorable effects in patients with severe, inoperable hydatid disease, although the degree of response varied (8). Nowak et al. (12) reported that following 2 months of oral albendazole administration a clear reduction in the size and number of hydatid lesions in the brain might be revealed.

Although mebendazole is effective in hydatid disease (6), hydatid cysts should be removed without rupture and all patients must receive mebendazole at pre- and postoperative periods for 8 weeks (2,14). In our case, all cysts were treated with albendazole therapy without surgical intervention.

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References

1. King CH. Cestodes (Tapeworms). (Eds. Mandell GL, et al). Principles and Practice of Infectious Diseases. Fifth ed. Churchill Livingstone. Philadelphia 2000, pp. 2956-2970.
2. al-Bassam A, Hassab H, al-Olayet Y, et al. Hydatid disease of the liver in children. Annals of Tropical Paediatrics 19: 191-196, 1999.
3. Baysefer A, Erdogan E, Gonul E, et al. Primary multiple cerebral hydatid cysts: case report with CT and MRI study. Minimally Invasive Neurosurgery 41: 35-37, 1998.
4. Ramsey PG, Plorde JJ. Cestode infections. pp. 825-829 in Braunwald, E. Ed. Harrison's Principles of Internal Medicine. McGraw-Hill, New York, 1987, pp. 825-829.
5. Ozgen T, Erbenli A, Bertan V, et al. The use of computerized tomography in the diagnosis of cerebral hydatid cysts. Journal of Neurosurgery 50: 339-342, 1979.
6. Ersahin Y, Mutluer S, Guzelbag E. Intracranial hydatid cysts in children. Neurosurgery 33: 219-225, 1993.
7. Taratuto AL, Venturiello SM. Echinococcosis. Brain Pathology 7: 673-679, 1997.
8. Todorov T, Vutova K, Petkov D, et al. Albendazole treatment of multiple cerebral hydatid cysts. A case report. Trans R Soc Trop Med Hyg 82: 150-152, 1988.
9. Al Zain TJ, Al-Witry SH, Khalili HM, et al. Multiple intracranial hydatidosis. Acta Neurochirurgica 144 : 1179-1185, 2002.
10. Erongun U, Ozkal E, Acar O, et al. Multiple and infected cerebral hydatid cysts. Case report. Neurosurgical Review 17: 77-81, 1994.
11. Hattingen J, Uerlings A, Jaussi R. Alveolar echinococcosis of the liver with lymphogenous, pulmonary and cerebral metastasis. Cerebral alveolar hydatidosis: case report. Surgical Neurology 45: 575-581, 1996.
12. Nowak DA, Lucking MO, Bohmer RH, et al. Isolated intracerebral cystic echinococcosis. Unusual presentation of a rare disease. Deutsche Medizinische Wochenschrift 127: 1638-1642, 2002.
13. Carcassonne M, Aubrespy P, Dor V. Hydatid cysts in childhood. Progress in Pediatric Surgery 5: 1-35, 1973.
14. Todorov T, Vutova K, Mechkov G, et al. Experience in the chemotherapy of severe, inoperable echinococcosis in man. Infection 20: 19-24, 1992.