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CASE REPORT

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Cerebral Alveolar Echinococcosis Mimicking Primary Brain Tumor

We present a case of cerebral infestation by *Echinococcus multilocularis* mimicking an infiltrative primary brain tumor. A heavily calcified mass invading the midbrain enhanced in a cauliflower-like fashion with small peripheral nodules present on MR imaging. Perfusion-weighted MR imaging revealed low relative cerebral blood volume within the calcified lesion and peripheral hyperemia. Single-voxel proton MR spectroscopy with an echo time of 135 milliseconds was normal.

Two species of the genus *Echinococcus* are the most clinically relevant forms: the cystic echinococcosis (hydatid cyst) caused by *Echinococcus granulosus* (EG) and alveolar echinococcosis (AE) caused by *E. multilocularis* (EM). Human AE is an uncommon zoonotic infestation caused by intrahepatic growth of the EM parasitic larvae. The metacestode spreads from the inevitably involved liver to other organs such as the lungs and brain and bone by infiltration and metastasis. Cerebral metastasis is very rare, being reported in only 1% of patients.¹⁻³ Cerebral infestation by EM and EG differ: AE occurs in adults who live in rural areas, whereas hydatid disease affects mostly children. EG infestation is usually self-limited, whereas EM infestation may appear as an infiltrative neoplasm in both the liver and brain, which makes total surgical removal difficult.⁴⁻⁹ This report presents findings of brain CT and MR examinations, in addition to perfusion-weighted MR (pMRI) and proton MR spectroscopy imaging findings of cerebral AE.

Case Report

A 26-year-old woman was referred to our hospital with a complaint of headache lasting for 2 months. Neurologic examination revealed confusion, anisocoria, and right hemiparesis. The Babinsky sign was present on the right side. Routine blood tests revealed mild leukocytosis with a white blood cell count of 14,600.

She had a long history of a liver cystic disease, for which she had been surgically treated in 1992 and 1998 at an outside institution; however, neither written information about her medical history nor a report of pathologic examination about her liver disease was available. Abdominal CT performed in our hospital, however, revealed irregular cysts at the capsular surface near the right hepatic lobectomy margin (not shown).

Initial cranial CT showed a calcified mass in the left thalamus with profound edema (Fig 1). Conventional MR imaging demonstrated left thalamic mass infiltrating the surrounding tissue and down to the midbrain. The lesion was isointense to cerebral white matter with nodular low-signal-intensity areas on T1-weighted images. T2-weighted images revealed a heterogeneous low-intensity mass with peripheral high signal intensity consistent with edema (Fig 2A, -B). Calcification on CT corresponded to nodular hypointense areas in the lesion on both T1- and T2-weighted images. Irregular cauliflower-like enhancement of the nodules and an enhancing rim all around the mass were observed (Fig 2C, -D).

Perfusion MR imaging revealed low relative cerebral blood vol-

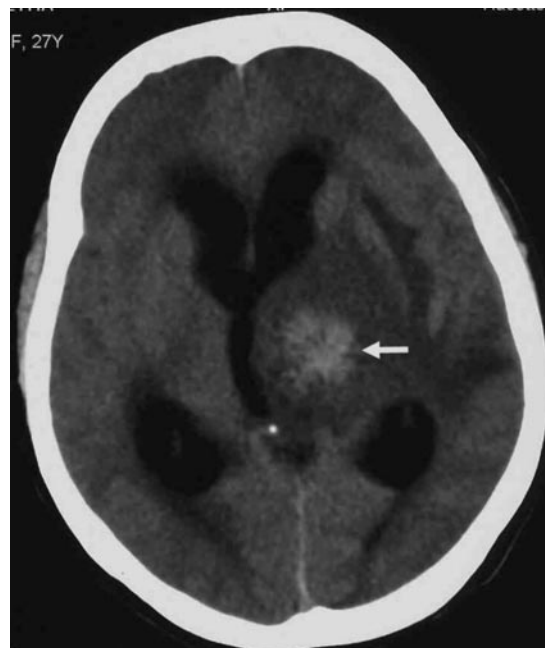


Fig 1. Cranial CT shows left thalamic calcified mass (arrow) with profound edema.

ume (rCBV) within the lesion, whereas higher values were obtained from peripheral areas of the lesion, most probably representing inflammation, later demonstrated on pathologic specimen (Fig 3). Baseline artifacts were present in MR spectroscopy (TR/TE, 3000/135; single voxel; voxel size, 15 × 15 × 15 mm) because of heterogeneity of the lesion with massive calcification. A voxel obtained at the periphery, which consisted of both enhancing and nonenhancing portions of the mass, however, showed normal *N*-acetylaspartate/creatine (1.10) and choline/creatine (0.71) ratios, which suggested that the lesion was of non-neoplastic origin.

The lesion was solid, multilocular, and white-colored macroscopically and was grossly and totally removed during surgery. The patient was discharged as neurologically intact except for slight right homonymous hemianopsia.

Histologic examination revealed a multilobulated lesion composed of multiple cysts characterized by a thick laminated-noncellular outer layer exhibiting focal discontinuities. Cuticle was periodic acid-Schiff (PAS) positive, and a granular layer was present. Amorphous necrotic debris was surrounded by attenuated gliosis with reactive astroglial proliferation. Epithelioid and giant cells containing multiple granulomas of different size were detected. Dense lymphoplasmacytic cell infiltrate rich in eosinophilic leukocytes was observed in the surrounding brain. Morphologic findings revealed degenerative AE cysts with reabsorption (Fig 4).

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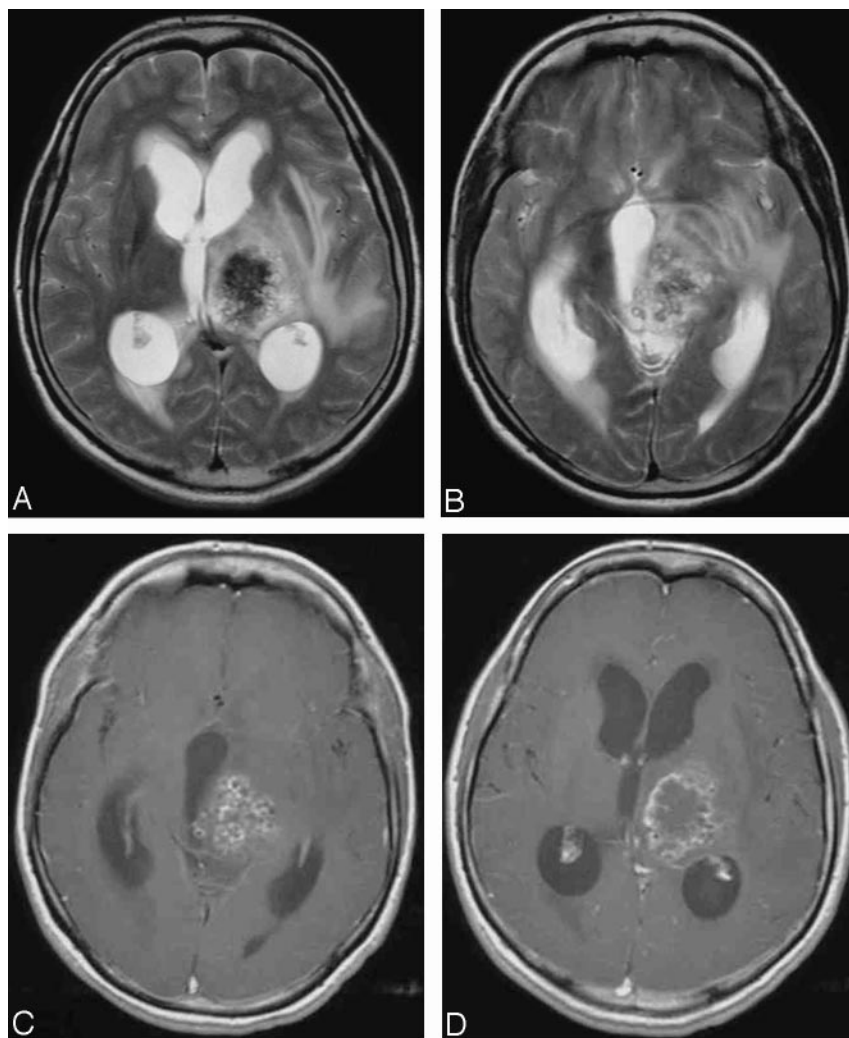


Fig 2. Axial T2-weighted imaging (TR/TE, 3080/93 milliseconds) shows calcified mass with microcysts extending between the left thalamus (A) and the midbrain (B). Axial T1-weighted postcontrast imaging (TR/TE, 650/9 milliseconds) reveals irregular enhancement of the nodules in a cauliflower-like pattern (C) and an enhancing rim around the mass (D).

served in the present case. Although not always seen, contrast enhancement of the lesion has been proposed because of blood-brain barrier disruption caused by the inflammatory reaction.¹¹ Peripheral ringlike, heterogenous, nodular, and cauliflower-like enhancement patterns have all been reported in different presentations of the infestation.^{8-10,12} In our case, T1-weighted postcontrast imaging showed that the mass was enhancing irregularly in a cauliflower-like pattern with small enhancing nodules. In addition, there was a rimlike enhancement surrounding the mass. With these findings the mass was indistinguishable from an ependymoma especially in the absence of knowledge about the patient's history of liver disease. pMRI revealed low rCBV within the lesion with higher values in the periphery. The low rCBV might have resulted from attenuated gliosis and granulomas shown on histologic examination, which also give the concrete nature of the lesion observed in the operation. More peripheral areas with increased blood volume, on the other hand, might be related to surrounding parenchymal inflammation observed on histologic specimen (Fig 4).

Discussion

The cyst of AE grows by external budding of the germinal membrane with progressive infiltration of the surrounding tissue, and this feature differs from EG, which is usually self-limited.² Difficulty in diagnosis stems from resemblance of AE infestation to malignancy as a result invasion of adjacent structures, destructive tissue growth, and metastasis in distant organs. Lesions of AE have alveolar structures composed of numerous irregular cysts with a diameter between 1 and 20 mm and not sharply demarcated from surrounding tissue. Central cystic cavities can result from necrosis and liquefaction of the inner part. Irregularly thickened and partially calcified wall is present in most cases.¹ Clinical features of cerebral AE—including headache, seizure, increased intracranial pressure, and focal neurologic deficits, depending on the location of the lesion—are those of any space-occupying lesion.

There are just a few published reports of separate cases on CT and MR imaging features. Yet, to the best of our knowledge, there has been no report of pMRI or MR spectroscopy findings. On CT and MR imaging the lesions mainly appear as solid and semisolid or sometimes as a multilocular cystic mass.⁷⁻¹³ Disseminated intracerebral AE lesions mostly appear at the terminal phase of the disease.^{7,9,12} Calcification and surrounding edema are common findings of the lesions as ob-

rounding parenchymal inflammation observed on histologic specimen (Fig 4).

A few reports on proton MR spectroscopy in differential diagnosis of cystic cerebral lesions displayed resonances from lactate, succinate, and acetate, as well as pyruvate, in cestodal cysts.¹⁴⁻¹⁷ The mass we report, however, was solid, and MR spectroscopy of a voxel at the periphery of the lesion, which contained a partly enhancing component and accompanying reactive inflammatory edema, was sufficient to show its non-tumoral nature (a glial tumor or a metastasis), though it did not give an additional finding to suggest its infectious origin.

Our case differs from previously reported cases in that the epicenter of the mass was the left thalamus and it infiltrated the brain stem. Involvement of the deep gray matter, a feature of this case, has not been present in any previously reported cases. The locations of reported AE lesions are lobar and posterior fossae.^{7-9,11,12}

It is known that human parasitic zoonoses sometimes cause diagnostic and therapeutic problems. Despite newly developed neuroradiologic and serologic methods, these entities are rarely diagnosed preoperatively.^{16,18} Differential diagnosis includes tumors and infectious lesions such as tuberculosis and bacterial abscess. Because AE infestation simulates malignancy by its invasive nature in the brain, gliomas and metastases are also included in the differential diagnosis.

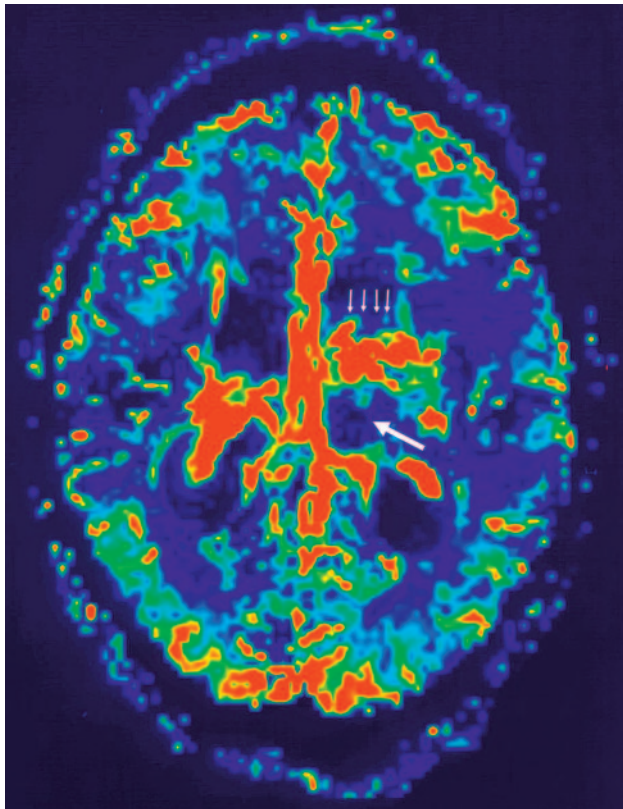


Fig 3. rCBV map of perfusion-weighted MR imaging (EPI [TR/TE, 1430/46 milliseconds]; NEX, 1; section thickness, 5 mm; intersection gap, 10 mm; mtx, 128 × 128) demonstrates low rCBV within the lesion (arrow) located at left thalamus with higher values peripherally (thin arrows).

Geographic prevalence, clinical history of hepatic involvement, and serologic tests are required for diagnosis. Intra-axial lesions imaging as grapelike multilocular cystic masses, cauliflower-like enhancing, or infiltrative heavily calcified masses with small nodules in patients with known liver disease should suggest the cerebral involvement of AE infestation. Moreover, the recently developed neuroradiologic tools such as pMRI and MR spectroscopy help to differentiate these cerebral space-occupying lesions from malignant tumors, which remain the main differential in our patient.

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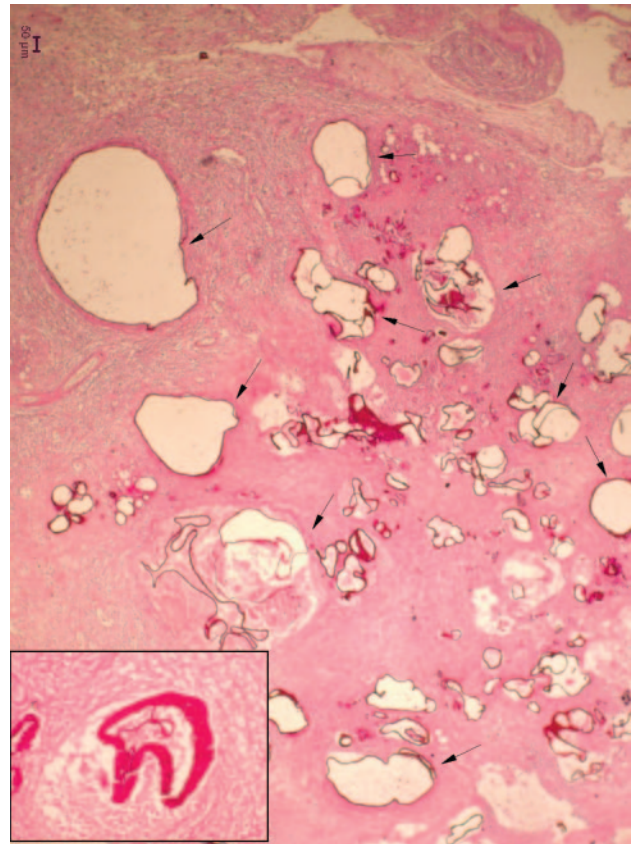


Fig 4. Multiple cysts (arrows) in the brain parenchyma surrounded by necrosis and giant cell granulomatous reaction (PAS, ×4). Inset, PAS-positive cuticular membranes (PAS, ×40).

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