Calcified Cerebral Hydatid Cyst Following Head Trauma: Case Report

Kafa Travması Sonrası Gelişen Kalsifiye Kist Hidatik: Olgu Sunumu

ABSTRACT

An extremely rare case of a calcified cerebral hydatid cyst is presented. The cyst was treated surgically at our department. The only symptom was generalized seizure. The calcified, non-enhancing and well-circumscribed characteristics of the lesion were detected on computed tomography (CT). The patient did not have any evidence of hydatid disease elsewhere in the body. The lesion was removed totally by surgery. The postoperative period was uneventful and seizures were not seen during 2-year follow up. This rare lesion is one of few cases presented in the literature where a complete neuroradiological and neuropathological evaluation was performed. In addition, this is only reported case growing at the surgical area following head trauma.

KEY WORDS: Brain, calcified hydatid cyst, computed tomography, head trauma.

ÖZ

Bu çalışmada, oldukça nadir görülen bir kalsifiye serebral kist hidatik olgusu sunulmaktadır. Bu olgu kliniğimizde cerrahi olarak tedavi edildi. Olguda tek semptom generalize nöbetti. Kalsifiye, kontrast tutmayan, keskin sınırlı lezyon, bilgisayarlı tomografide belirlendi. Hastanın vücudunun başka bir yerinde kist hidatiğe rastlanmadı. Lezyon total olarak çıkartıldı. Ameliyat sonrası dönem sorunsuzdu ve 2 yıllık takipte nöbet gözlenmedi. Bu nadir lezyon, literatürde tam olarak nöroradyolojik ve nöropatolojik açıdan değerlendirilen birkaç olgudan biridir. Buna ilave olarak bu olgu, kafa travması sonrası geçirilmiş operasyon alanında gelişmesi yönüyle de literatürdeki tek olgu olarak sunulmaktadır.

ANAHTAR SÖZCÜKLER: Beyin, bilgisayarlı tomografi, kafa travması, kalsifiye kist hidatik.

Ahmet MENKÜ¹
Ali KURTSOY¹
BÜLENT TÜCER¹
A. Candan DURAK²
Hidayet AKDEMİR¹

- Department of Neurosurgery, School of Medicine, Erciyes University, Kayseri, Turkey
- 2 Department of Radiology, School of Medicine, Erciyes University, Kayseri, Turkey

Received: 09.09.2003 Accepted: 18.02.2004

Correspondence Address:

Ahmet MENKÜ

Erciyes University, Medical School, Department of Neurosurgery, 38039 TR, Kayseri, Turkey

Phone : +90 352 4374938 Fax : +90 352 4378655 E-mail : menkua@hotmail.com

INTRODUCTION

Cerebral hydatidosis is reported very rarely in Western neurological articles, presumably because the worldwide distribution of the parasite is mainly limited to agricultural and sheep-raising communities.

Although hydatid disease of liver and lungs is quite common, involvement of brain is quite rare (involvement in only 2 percent of cases of hydatid disease). The frequency of calcified cerebral hydatid cyst is less than 1% of all cerebral hydatid cysts (3).

Cerebral hydatid cysts are usually distributed within the watershed zone of the middle cerebral arteries, often in the parietal lobe (11). The symptoms may be insidious in onset, with the cyst reaching massive size before the diagnosis is made. Seizures and signs of increased intracranial pressure may be present. A calcified cerebral hydatid cyst is generally inactive and only unusually causes seizures. The calcified cerebral lesion in our patient was diagnosed as a dead cerebral hydatid cyst according to the clinical and histopathological studies performed during the postoperative period.

CASE REPORT

A 35-year old man was admitted to our clinic for an epileptic attack. The patient was alert and oriented on neurological examination. He was describing generalized seizures two or three times a month for about two years despite anticonvulsant therapy. He had been operated on for traumatic intracerebral hemorrhage at another institution 22 years ago.

Unenhanced CT scan showed a well-circumscribed, 50X45 mm calcified lesion in the right parieto-occipital area (Figure 1), the same area as the previous surgery. The lesion did not enhance after contrast infusion. There was also evidence of a previous craniotomy, with a surgical defect in the right parieto-occipital bone (Figure 2). These findings suggested a calcified mass lesion or calcified intracerebral hemorrhage. Preoperative axial T1-weighted magnetic resonance imaging (MRI) scans showed right parieto-occipital intra-axial mass lesion which had sharp margins and mixed intensity (iso-hyperintense to gray matter) with a ring-like peripheral contrast enhancement

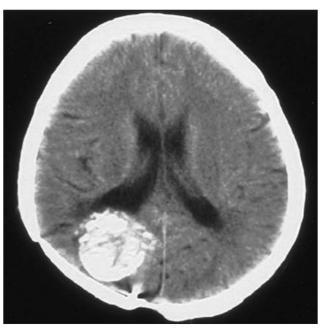


Figure 1: Unenhanced CT scan shows a well-circumscribed, 50X45 mm calcified lesion in the right parieto-occipital area

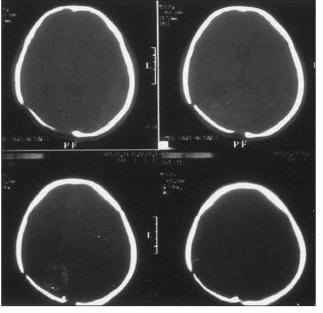


Figure 2: A CT scan reveals evidence of a previous craniotomy, with a surgical defect in the right parieto-occipital bone

(Figure 3A). Axial T2-weighted MRI scans showed marked peripheral hyperintensity and heterogenous (iso-hyper-hypointense) central intensity, sharp margins and mild peripheral edema (Figure 3B).

An operation was performed under endotracheal anesthesia in the supine position. The head was

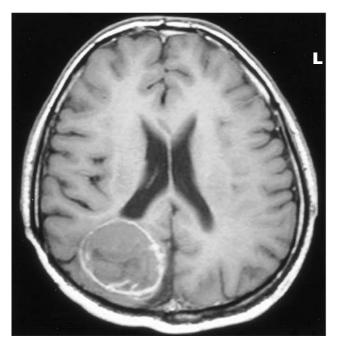


Figure 3A: Axial T1-weighted MRI scan shows right parieto-occipital intra-axial mass lesion.

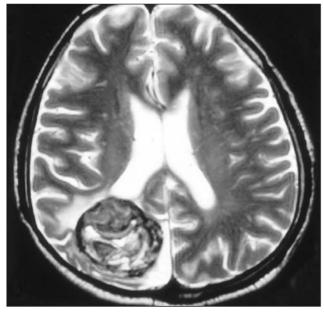


Figure 3B: Axial T2-weighted MRI scan shows marked peripheral hyperintensity and heterogenous central intensity, sharp margins and mild peripheral edema.

firmly fixed in a Mayfield headholder. Following the right parieto-occipital old scalp incision, an osteoplastic bone flap was made. The right parieto-occipital cortical approach was performed, using an operating microscope. The tumor was found to be 2-3 mm below the cortical surface. The tumor was

globular, hard, calcified and well-circumscribed. It was removed completely. The postoperative course was eventful. Postoperative CT confirmed total removal of the lesion (Figure 4).



Figure 4: Postoperative CT shows total removal of the lesion

Histopathological examination revealed no evidence of a tumor or calcified hemorrhage but a calcified hydatid cyst. An inner, nucleated, germinative layer, and an outer, opaque, non-nucleated layer were found. The outer non-nucleated layer had delicate laminations. Outside of this opaque layer, there was a host inflammatory reaction that produced a zone of fibroblasts, giant cells and mononuclear cells (Figure 5).

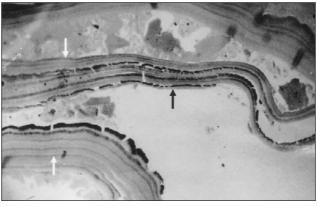


Figure 5: Acellular, hyaline laminations composing the lining of the cyst wall (white arrows) and the degenerative germinal layer (black arrow) are clearly seen (40X, HE).

Detailed investigation failed to reveal any evidence of hydatid disease elsewhere in the body. Full recovery took place one month later. The duration of follow-up has now been 2 years. Seizures or recurrence were not seen during follow-up period (Figure 6).

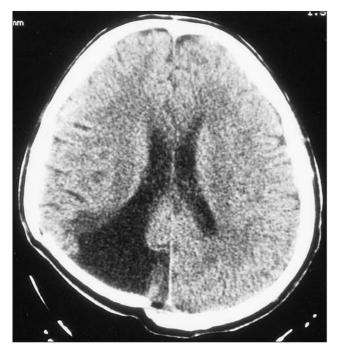


Figure 6: Postoperative CT scan reveals no recurrence after two years.

DISCUSSION

Calcified cerebral hydatid cyst is very rare. A medline search of articles published from post-CT era to 2003, using the subject headings "Calcified hydatid cyst and brain" revealed only three cases (1, 3, 4).

The relative frequency of cerebral hydatidosis in children is because of the preterm permeability of the capillary circulation of the lung in association with a relative frequency of head injury in childhood (7). Primary hydatid infection caused by embryos that have escaped hepatic and pulmonary barriers is generally single and fertile. On the other hand, secondary hydatidosis caused by scolices from ruptured fertile cysts is usually multiple and sterile (9). The parasite may have been ingested by the child with unboiled milk during the neonatal period . The communication between the right and left heart in childhood may allow the parasite to reach the brain (8). We believe that trauma or surgery may induce

the hydatid cyst to grow in the brain because of damage to the blood brain barrier.

It takes about six months from the time of initial implantation for a given cyst to become fertile. Some cysts never produce broad capsules; others may become sterile either through superimposed bacterial infection or through death and ensuing calcification (8). In our case, the cyst spontaneously regressed with calcification without treatment. CT scan may show broad calcification in the wall or septae. Although calcification of all cysts in liver is quite common (2, 5, 6), this is quite rare in the brain (12). A calcified cyst in the brain may be called a 'dead cerebral hydatid cyst' (12). Calcification indicates chronicity of the cyst which may not need to be removed. However, we excised the cyst as it was causing intractable seizures.

The present case is the first case that developed at the surgical area following head trauma to our knowledge.

The presence of a hydatid cyst in the brain without cysts in other organs has even now not been fully explained (10). Our case is therefore also a primary cerebral hydatid cyst, a condition rare in adults.

In conclusion, hydatid cyst should be included in the differential diagnosis when a calcified brain lesion is found in patients from an endemic echinococcosis area.

References

- Alvarez F, Blazquez MG, Oliver B, Manrique M: Calcified cerebral hydatid cyst. Surg Neurol 17:163-164, 1982
- Ayral MN: Kist hidatikler ve tedavi prensipleri, Türkiye'de Ekinokok Problemleri Simpozyumu Kitabı, 73-94, 1974
- Boles DM: Cerebral echinococciasis. Surg Neurol 16:280-282, 1981
- 4. Krajewski R, Stelmaslak Z: Cerebral hydatid cysts in childeren. Childs Nerv Syst 7:154-155, 1991
- Kune AG: Hydatid Disease, in Schwartz SI (ED): Maingot's Abdominal Operations, Norwalk, Appleton-Centuray-Crofts, Eight edition, Volume two pp 1605-1624, 1985
- 6. Lichtenberg FV: Pathology of Infectious Disease, New York, Raven Press, pp 331-335, 1991
- 7. Lombard P: Kystes hydatiques du cerveau. J Chir (Paris) 16:278-297, 1920
- Lunardi P, Missori P, Di Lorenzo N, Fortuna A: Cerebral hydatidosis in childhood: a retrospective survey with

- emphasis on long-term follow-up. Neurosurgery 29:515-517, 1991
- 9. Micheli F, Lehkuniec E, Giannaula R, Caputi E, Paradiso: Calcified cerebral hydatid cyst. Eur Neurol 27:1-4, 1987
- Paşaoğlu A, Tel E, Erbengi A: The frequency and classification of intracranial tumors. Anadolu Tip Dergisi 5:85-96, 1983
- 11. Roquer J, Escudero D, Guelar A, Herraiz J, Canas A, Maso E, Martinez J, Ferrer F: Calcified cerebral hydatid cyst in a patient with abdominal hydatidosis. Med Clin (Barc) 12:458-460, 1985
- 12. Yolaş C, Gündoğdu C, Yakut C: Ölmüş serebral kist hidatik (Bir olgu sunumu). Türk Nöroşirürji Dergisi 3:38-40, 1993