Surgical Neurology International

OPEN ACCESS

For entire Editorial Board visit : http://www.surgicalneurologyint.com

Editor-in-Chief: **James I. Ausman**, MD, PhD University of California, Los Angeles, CA, USA

Case Report

Alveolar hydatid cyst mimicking cerebellar metastatic tumor

Cagatay Ozdol, Ali Erdem Yildirim, Ergun Daglioglu, Denizhan Divanlioglu, Esra Erdem[1], Deniz Belen

Ankara Numune Education and Research Hospital, Neurosurgery Clinics, Ankara, Turkey, [1]Ankara University Medicine Faculty, Pathology Department, Ankara, Turkey

E-mail: *Cagatay Ozdol - drcagatayozdol@gmail.com; Ali Erdem Yildirim - alierdemyildirim@gmail.com; Ergun Daglioglu - ergundaglioglu@gmail.com; Denizhan Divanlioglu - ddivanlioglu@gmail.com; Esra Erdem - info@kesitpatoloji.com; Deniz Belen - denizbelen2000@yahoo.com

*Corresponding author

Received: 18 October 10 Accepted: 4 January 11 Published: 29 January 11

Surg Neurol Int 2011, 2:13

This article is available from: http://www.surgicalneurologyint.com/content/1/2/13

This article may be cited as:
Ozdol C, Yildirim AE, Daglioglu E, Divanlioglu D, Erdem E, Belen D. Alveolar hydatid cyst mimicking cerebellar metastatic tumor Surg Neurol Int 2011;2:13
Available FREE in open access from: http://www.surgicalneurologyint.com/text.asp? 2011/1/2/76281

Abstract

Background: Echinococcus multilocularis is a rare infestation in the world with a particularly increased incidence mainly in South America, Central Europe and Asia. Progression of alveolar Echinococcosis is more aggressive that can metastasize to lungs, brain and bones however brain involvement is usually rare with an incidence about 1%.

Case Description: We report a 23-year-old man with a cerebellar Echinococcosis multilocularis mimicking a metastatic cerebellar tumor. Suboccipital craniotomy was performed for gross total removal of the tumor. Histopathological specimens confirmed the diagnosis of Echinococcosis multilocularis.

Conclusion: Radical surgical excision should be recommended for single Echinococcosis multilocularis lesions particularly at infratentorial localization.

Key Words: Alveolar hydatid cysts, cerebellum, echinococcus, surgery, tumor

Access this article online

Website: www.surgicalneurologyint.com

DOI: 10.4103/2152-7806.76281

Quick Response Code:

INTRODUCTION

Hydatid disease is a parasitic infestation produced by Echinococcus granulosus (EG) and Echinococcus multilocularis (EM).[3] EM is a rare human infestation with a wide geographic distribution especially in regions like South America, Central Europe, Central and East Asia.[8,10] Echinococcosis is endemic for Turkey, particularly in Eastern and Northeastern Anatolian region.[2] The life cycle is completed when a fox or canine consumes a rodent that is infected with cysts. Human beings can be the accidental host for these parasites.[11] EM primarily affects the liver. This chronic infestation may spread hematogenously to produce metastatic foci in the distant organs. Involvement of the brain is rare, reported to be around 1%.[7,13] Alveolar hydatid cysts constitute 3% of cerebral hydatid cyst cases. Infratentorial involvement is considerably rare.[4] Here we report a 23-year-old man with cerebellar involvement of Echinococus multilocularis mimicking a metastatic cerebellar tumor.

CASE REPORT

A 23-year-old man presented with nausea, imbalance, occasional urinary and fecal incontinence and a severe headache for 1 month. The patient was a farmer with an unremarkable past experience for his relatives. Neurological examination was completely normal. Magnetic resonance imaging (MRI) demonstrated a left cerebellar mass lesion of 3×2×1.5 cm in size with marked peripheral contrast enhancement [Figure 1]. Computed tomography (CT) examination of the chest and abdomen

were performed for a primary origin. Multiple calcified mass lesions with lobulated contours were shown in the right upper lobe of lung, right liver and another solid tumor between right kidney and liver. Suboccipital craniotomy was performed and a left intracerebellar pale yellow mass was excised grossly as total. The tumor was almost avascular and it was easily dissected from the surrounding cerebellar tissues. Postoperative course was unremarkable without any neurological deficit. Histopathological examination revealed PAS (+) cuticular membrane with wide areas of necrosis and inflammation which were typical for EM [Figure 2]. Serological tests at the postoperative period confirmed the presence of EM with indirect hemagglutination test.

Albendazole (800 mg, bid, 3 cure, 28-day cycle followed by a 14-day albendazole-free interval) and cephotaxime (4 g, bid) were prescribed for postoperative treatment. A further operation was performed to resect the lesion in the lung a month after intracranial surgery. Postoperative early CT examination and MRI performed 6 months after surgery showed no recurrence.

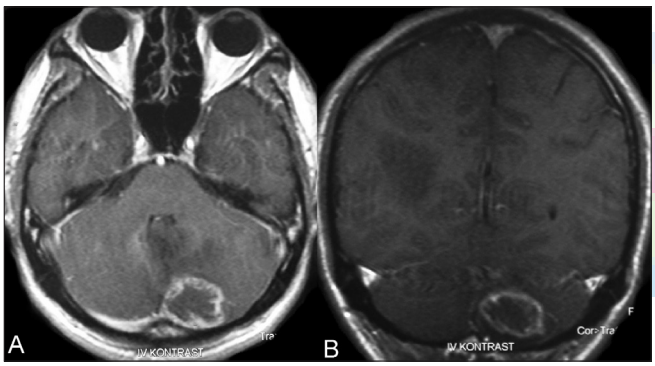

Figure 1: Gadolinium enhanced T1-weighted axial (A) and coronal (B) MR images demonstrate a left cerebellar lesion with peripheral enhancement.

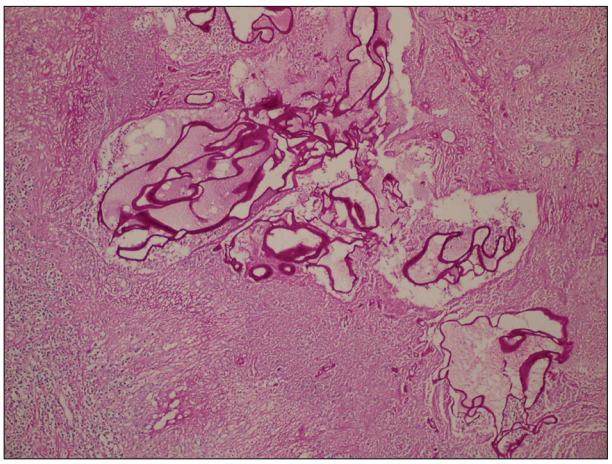

Figure 2: PAS (+) cuticular membrane pieces were prominent on the histopathological studies with wide necrotic areas and inflammation which were concordant with alveolar echinococcosis. (PAS (+)×100).

DISCUSSION

Alveolar hydatid disease is a human infestation caused by the larval stage of EM. The natural definitive host is the canine, fox and cats while the intermediate hosts are usually small rodents and sometimes humans. The metacestode form of EM has a tendency to disseminate into other organs than liver by local spreading or hematogenous metastasis.[5] Lung, heart, thyroid, brain, bone and spleen are main target organs.[6,13] EM cases are usually adults and clinical findings depend on the area and size of the lesion. The clinical presentation usually includes symptoms of increased intracranial pressure (headache, vomiting, motor weakness. etc.), seizures and cranial nerve findings.[14] Immunofluorescence, indirect hemagglutination tests, ELISA and immunoelectrophoresis with antigenic extracts of EM are usually supportive laboratory tests for a diagnostic accuracy.[9] A solid, semisolid or multilocular cystic mass is a common finding on CT and MRI of EM.[15,16] Calcification and perilesional edema are prominent.[1] Cerebral lesions are usually single or multiple. They are usually supratentorial and within the watershed zone of middle cerebral artery; however, infratentorial EM is extremely rare.[16] Altinors *et al*. studied 219 hydatid disease patients and reported only 11 infratentorial lesions with 2 EM localized in cerebellum.[4] EM involvement of brain and liver is similar to an infiltrative tumor. Tuberculosis, bacterial abscesses, fungal infections, invasive brain tumors and metastases should be considered in the differential diagnosis of these infections.[13] Differential diagnosis of EM lesions includes malign tumors due to local invasive pattern, tissue destruction during growth and metastases to distant organs. Histopathological examination is necessary for a definite diagnosis.

The case we presented here had a left cerebellar lesion presumed to be a metastatic tumor which is quite uncommon. On the other hand, lesions in the liver and lung were misinterpreted as a primary source of metastasis which was another diagnostic conflict. The diagnosis of EM was confirmed with further histopathological examination.

Radical excision should be performed for all accessible surgical lesions. Several cycles of postoperative albendazole chemotherapy are almost always required. Gamma knife radiosurgery may be an alternative to surgery for patients with a high risk of surgery and anesthesia.[12]

CONCLUSION

Alveolar echinococcus should be considered in the differential diagnosis of cerebral tumors, particularly in endemic areas. Morbidity and mortality of cerebellar

EM is higher than the supratentorial EM lesions due to high risk of complications. Cerebellar EM lesions should always be considered as surgical candidates due to herniation risk; however, surgical treatment for multiple cerebral lesions is usually palliative with alternative chemotherapeutics.

REFERENCES

1. Algros MP, Majo F, Bresson-Hadni S, Koch S, Godard J, Cattin F, et al. Intracerebral alveolar echinococcosis. Infection 2003;31:63-5.
2. Altıntaş N. Cystic and alveolar echinococcosis in Turkey. Ann Trop Med Parasitol 1998;92:637-42.
3. Altinors N, Kars Z, Cepoglu C, Gurses L, Sagbil S, Ariyurek M. CT findings and surgical treatment of double intracranial Ecchinococcal Cysts. Infection 1991;19:110-4.
4. Altinors N, Bavbek M, Caner HH, Erdogan B. Central nervous system hydatidosis in Turkey: a cooperative study and literature survey analysis of 458 cases. J Neurosurg 2000;93:1-8.
5. Aydınlı B, Aydın Ü, Yazıcı P, Öztürk G, Onbaş Ö, Polat KY. Alveolar Echinococcosis of Liver Presenting with Neurological Symptoms due to Brain Metastases with Simultaneous Lıng Metastasis:A Case Report. Turkiye Parazitol Derg 2008;32:371-4.
6. Canda MS, Canda T. The presentation of 47 cases and the echinococcus problem of Turkey (in Turkish) T Parazitol Derg (Acta Parasitologica Turcica) 1995;19:64-82.
7. Cataltepe O, Colak A, Ozcan OE, Ozgen T, Erbengi A. Intracranial hydatid cysts: experience with surgical treatment in 120 patients. Neurochirurgia (Stuttg) 1992;35:108-11.
8. Craig P. Echinococcus multilocularis. Curr Opin Infect Dis 2003;16:437-44.
9. Hakan T, Aker FV. A case report of fatal Echinococcosis. Ann Neurosurg 2001;1:14-9.
10. McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. Lancet 2003;36239:1295-304.
11. Pamir MN, Ozer AF, Keles GE, Tozun N, Gurmen N, Kullu S. Cerebral echinococcosis multilocularis: Case report. J Neursurg Sci 1991;35:161-4.
12. Schmid M, Pendl G, Samonigg H, Ranner G, Eustacchio S, Reisinger EC. Gamma knife radiosurgery and albendazole for cerebral alveolar hydatid disease. Clin Infect Dis 1998;26:1379-1382.
13. Senturk S, Oguz KK, Soylemezoglu F, Inci S. Cerebral alveolar echinoccosis mimicking primary brain tumor. AJNR Am J Neuroradiol 2006;27:420-2.
14. Takci E, Sengul G, Akar A, Uslu H, Alper F, Erdogan F, et al. Alveolar echinococcosis of the brain in five patients. J Clin Neurosci 2008;15:1105-9.
15. Tunaci M, Tunaci A, Engin G, Ozkorkmaz B, Ahishali B, Rozanes I. MRI of cerebral alveolar echinococcosis. Neuroradiology 1999;41:844-6.
16. Tüzün M, Altinörs N, Arda IS, Hekimoğlu B. Cerebral hydatid disease: CT and MR findings. Clin Imaging 2002;26:353-7.