ECRONICON OPEN ACCESS

EC NEUROLOGY
Case Report

Cerebral Hemorrhage Connected to Alveolar Echinococcosis: An Exceptional Case

Khalid Al Taha[1]*, Alda Rocca[1], Philippe Maeder[2,3] and Jocelyne Bloch[1,3]

[1]*Department of Clinical Neurosciences, Service of Neurosurgery, Lausanne University Hospital (CHUV), Lausanne, Switzerland*

[2]*Department of Radiology, Lausanne University Hospital (CHUV), Lausanne, Switzerland*

[3]*University of Lausanne (UniL), Lausanne, Switzerland*

***Corresponding Author:** Khalid Al Taha, Department of Clinical Neurosciences, Service of Neurosurgery, Lausanne University Hospital (CHUV), Lausanne, Switzerland.

Received: April 08, 2023; **Published:** April 25, 2023

Abstract

Alveolar echinococcosis, for which the human being is an accidental host, commonly affects the liver and may spread only in approximately 1% of patients to the brain. Switzerland is one of the endemic countries for this rare disease. In our report, we describe an atypical brain manifestation of this disease in a young female patient, who experienced a rapid neurological deterioration, due to an intraparenchymal hemorrhage.

Keywords: *Intracranial Hemorrhage; Cerebral Echinococcosis; Mycotic Aneurysm*

Introduction

Alveolar echinococcosis is a rare parasitic disease caused by *Echinococcosis multilocularis*. Its distribution is restricted to the northern hemisphere, particularly to regions of China, the Russian Federation and countries in continental Europe and North America [1,2,18,20,23,26]. Switzerland is an endemic country for this pathology [24,25].

Echinococcosis multilocularis is almost always found in liver (50 - 77%) and may spread to other organs, such as lungs (8.5 - 43%) and bones. Rarely implicated organs encompass the spleen, the spine, the middle ear, soft tissues and the brain (1 - 2%) [1,2,14,23].

The parasitic cycle of alveolar or multilocularis echinococcosis implicates foxes as final host. Rodents, dogs and human being are accidental and intermediate hosts. Their tenia, which grows in the small gut of the final host, contains eggs, which are released in intestine lumen and afterwards in the feces, which make the contamination of grass and food as fruits and vegetables. When the human being eats contaminated food, the helminth is released in the duodenum, and then crosses the intestine wall to target the liver. Hepatic lesions can cause liver necrosis. Invasion of the bile ducts and vessels can lead to severe complication, such as cholangitis, septic shock, portal thrombosis and hypertension [1,6,11,20,23,26]. Echinococcosis spread to the brain is very rare (only 1-2% of cases) [1,10,15-17,19,23,26]. These lesions can give aspecific symptoms connected to mass effect, with increased intracranial pressure, seizures or focal neurological impairment [2,5,7,9,10,12-18,20].

Here we present a rare case of a young woman with liver, lung and brain alveolar echinococcosis, who suddenly presented severe headaches and neurological deterioration, due to a cerebral intraparenchymal bleeding.

Case Report

A 41-years-old woman of Vietnamese origin, living in Switzerland for 20 years, presented, a non-painful icterus that was investigated with an ultrasound and an abdominal CT scanner, which demonstrated a voluminous mass in the left part of the liver. A biopsy of this lesion led to the diagnosis of *Echinococcosis multilocularis*. An abdominal MRI demonstrated a peritoneal and pulmonary dissemination, with biliary duct obstruction. For this reason, an endoscopic retrograde cholangio-pancreatography (ERCP) with sphincterotomy and biliary duct stenting was performed. The lesion was not surgically accessible. Therefore, a long-term treatment of albendazole was initiated. Three weeks later, the patient developed a postoperative large hepatic abscess, which was treated with a hepatic drainage. The fluid was positive for *Klebsiella Oxytoca, Enterobacter aerogenes* and *Candida albicans* and the patient was prescribed intravenous antibiotics (Piperacillin/Tazobactam). 3 months later, the patient underwent a follow up extended imaging assessment including a cerebral MRI, which showed a millimetric lesion in the left cuneus, interpreted as a cyst of *Echinococcosis multilocularis* (Figure A).

During the same month, she also underwent a new ERCP for a biliary duct stent occlusion, complicated by a septic choc, due to *Pseudomonas putida*.

4 months later, the patient was admitted for an elective hepatectomy. The day before surgery, she complained of a sudden severe headache. The clinical examination revealed a confused patient with left homonymous hemianopia. A cerebral MRI showed a massive right temporo-parieto-occipital intraparenchymal hemorrhage. The left cuneus cyst was almost entirely resolved (Figure B and D). A few hours later, the patient suddenly became comatose, with a Glasgow coma scale of 5/15. A right temporo-parieto-occipital craniotomy to drain the intraparenchymal hemorrhage was performed. During surgery, cystic lesions were observed in the antero-medial part of the surgical cavity and were left in place, in order to avoid cystic wall rupture and further dissemination (Figure F). The microbiological analysis of the excised hemorrhagic tissue detected no germs.

Two days after surgery, the patient suffered a focal seizure confirmed by an EEG, expressed by intermittent consciousness state fluctuation, A treatment by levetiracetam and lacosamide was initiated, and the patient remained seizure free thereafter.

A week later, the patient showed a good neurological recovery, with only mild left brachio-crural sensitive deficit and left homonymous hemianopia.

3 months later, the left hepatectomy and a cholecystectomy have been performed, without complication.

Nevertheless, the lifelong antifungal therapy was maintained.

Nine months after her initial neurosurgical intervention, the patient remained asymptomatic, no abnormality was identified upon neurological assessment except for a left homonymous hemianopia. The MRI showed no suspected residual cystic lesion nor vascular abnormality (Figure E).

Discussion

In our case, the patient presented with severe headache and rapid loss of consciousness. The cerebral MRI showed a large right-sided temporo-parieto-occipital intraparenchymal hemorrhage (Figure B and D). During the surgical evacuation of the hemorrhage, some cysts were visualized in the cavity.

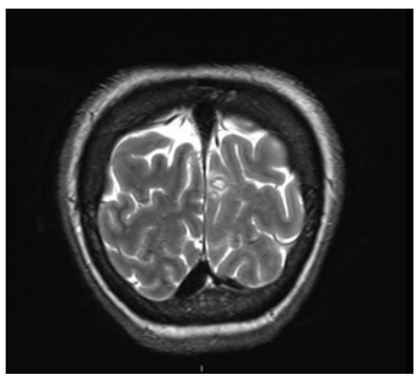

Figure A: *First brain MRI. T2-weighted coronal sequence showing a lesion in the left cuneus with perilesional edema, which is compatible with an alveolar echinococcosis cyst.*

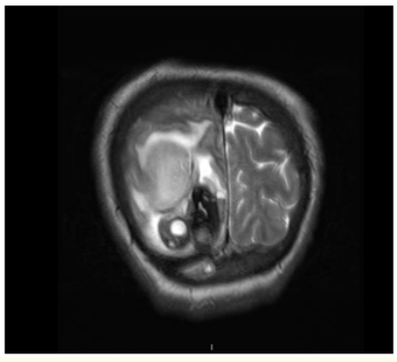

Figure B: *Preoperative brain MRI. T2-weighted coronal sequence showing a right-sided temporo-parieto-occipital intraparenchymal hemorrhage. The previous cystic lesion in the left cuneus is no more visible.*

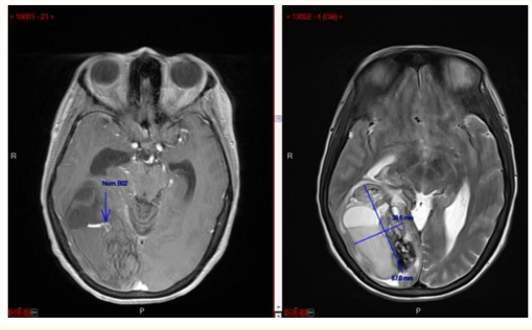

Figure C and D: *Preoperative brain MRI.*

Figure C: *T1-weighted contrast enhanced axial sequence, showing a nodular structure inside the hemorrhage, associated with contrast product extravasation.*

Figure D: *T2-weighted axial sequence, showing the volume of the temporo-parieto-occipital intraparenchymal hemorrhage.*

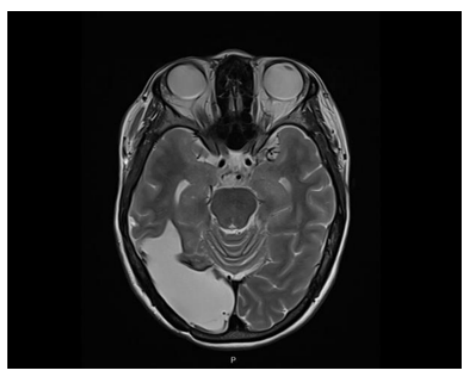

Figure E: *Post-operative T2-weighted axial sequences, 9 months after surgery, showing the left temporo-parieto-occipital resection cavity, without any suspected residual cystic lesion.*

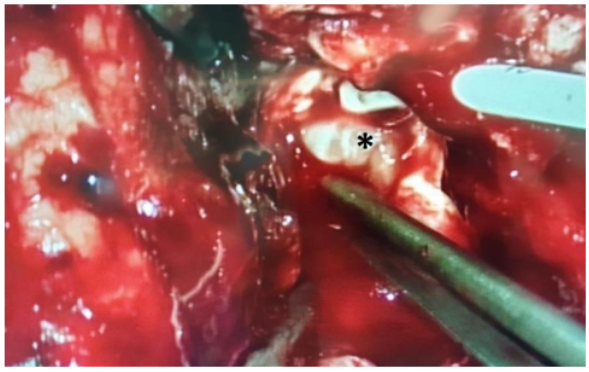

Figure F: *Microscopic image taken intraoperatively of the cystic lesions. (*) in the antero-medial part of the surgical cavity. The cysts were left in place, in order to avoid dissemination.*

Because of their avascular nature, these cysts usually don't bleed. The initial lesion of alveolar echinococcosis in fact consists of the larva, with a surrounding double membrane. The inner membrane produces secondary avascular cysts, which communicate with each other and give to the lesion its alveolar appearance. These cysts are surrounded by a fibrotic and granulomatous tissue. Cytotoxic effects of the vesicular fluid filling the cysts result in necrosis of the surrounding structures. It leads to the formation of a cavity filled with gelatinous magma, containing only necrotic tissue and foci of calcification [4,8,21,22].

A bleeding event associated with this pathology is a very rare entity. And to our knowledge, there is only one case described in the German literature [3]. One physiopathological hypothesis could be that a ruptured mycotic cerebral aneurysm could have caused the temporo-parieto-occipital intraparenchymal hemorrhage. Since no other germ was detected after microbiological analysis of the surgically excised tissues, a mycotic cerebral aneurysm due to echinococcosis is possible. Knowing that our patient presented a disseminated alveo-

lar echinococcosis (mainly in the left side of the liver and in the upper lobe of the left lung) and a patent foramen ovale, septic emboli may have been sent from the lung to the brain, because of the patent foramen ovale, causing the formation of a mycotic aneurysm. The brain MRI support our hypothesis, since it shows a small nodular enhanced structure within the hematoma evoking an aneurysm, with contrast extravasation after Gadolinium injection (Figure C). On the other hand, no vascular lesion is clearly seen in the T2-weight sequences, probably because mycotic aneurysms have no rapid flow and, after their rupture, a vasospasm can immediately occur (Figure B and D).

We realize that this explanation is just a hypothesis, based on indirect observations, since no histological nor radiological finding firmly confirmed our theory.

However, it is important to recognize this possible rare clinical manifestation for a disease that is becoming more endemic in Europe.

Conclusion

This case report shows a very rare and atypical presentation of brain alveolar echinococcosis, with a large intraparenchymal hemorrhage, probably due to a mycotic aneurysm, originating from a lung septic emboli, which ruptured and causing the hemorrhage.

After an emergency surgical evacuation, the neurological outcome was good.

In patients with disseminated multilocularis echinococcosis, brain imaging can be considered early during the course of the disease, since early diagnosis of brain spread is important and allows a better management.

Acknowledgments

All authors certify that they have NO affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

Conflict of Interest

None.

Patient Consent

The participant has consented to the submission of the case report to the journal.

Bibliography

1. Algros M-P., *et al*. "Intracerebral Alveolar Echinococcosis". *Infection* 31 (2003): 63-65.
2. Atlan Guneri., *et al*. "A case of alveolar echinococcosis presenting as cerebral and spinal intradural metastasis". *The Eurasian Journal of Medicine* 48 (2016): 149-152.
3. Assad F and Lins E. "Mykotisches Aneurysma der Arteria cerebri media bei Echinokokkenbefall". *Minim Invasive Neurosurg* 27.3 (1984): 89-92.
4. Aydin Y., *et al*. "Alveolar hydatid disease of the brain. Report of four cases". *Journal of Neurosurgery* 65.1 (1986): 115-119.
5. Batçik OE., *et al*. "Cerebral alveolar echinococcosis concomitant with liver and lung lesion in a young adult patient: Case Report and Literature Review". *Türkiye Parazitoloji Dergisi* 40.3 (2016): 169-171.

6. Bresson-Hadni S., *et al*. "A twenty-year history of alveolar echinococcosis: analysis of a series of 117 patients from eastern France". *European Journal of Gastroenterology and Hepatology* 12 (2000): 327-336.

7. Bulakci M., *et al*. "Disseminated alveolar hydatid disease resembling a metastatic malignancy: a diagnostic challenge-a report of two cases". *Radiology* (2014): 638375.

8. Ci-Peng Jiang., *et al*. "Liver alveolar echinococcosis in China: Clinical aspect with relative basic research". *World Journal of Gastroenterology* 11.30 (2005): 4611-4617.

9. Kantarci M Ogul and H Bayraktutan U. "Intracerebral alveolar echinococcosis". *Headache* 52 (2012): 1296-1297.

10. Kvascevicius Robertas., *et al*. "Fatal liver and lung alveolar echinococcosis with newly developed neurologic symptoms due to the brain involvement". *Journal of Surgery* 2 (2016): e83-e88.

11. Liu Yue Han., *et al*. "Continuous albendazole therapy in alveolar echinococcosis: long-term follow-up observation of 20 cases". *Transactions of The Royal Society of Tropical Medicine and Hygiene* 103.8 (2009): 768-778.

12. Llanes DV Erasmo Gonzalo., *et al*. "Echinococcosis presenting as an otogenic brain abscess: An unusual lesion of the middle ear cleft and temporal lobe". *Auris Nasus Larynx* 35 (2008): 115-120.

13. Lu Ma., *et al*. "An echinococcosis multilocularis presenting as a giant anterior cranial basal tumor". *Neurological Sciences* 30 (2009): 247-249.

14. Ogul Hayri., *et al*. "Primary echinococcosis presenting with simultaneous cerebral and craniofacial involvement". *The Journal of Craniofacial Surgery* 29.7 (2018): 4.

15. Okur Aylin MD., *et al*. "Magnetic resonance spectroscopy and magnetic resonance imaging findings of the intracerebral alveolar echinococcosis". *Journal of Craniofacial Surgery* 25 (2014): 1352-1353.

16. Ozdemir Nuriye Guzin., *et al*. "Echinococcosis alveolaris: Presenting as a cerebral metastasis". *Turkish Neurosurgery* 22.4 (2012): 448-451.

17. Ozdol Cagatay., *et al*. "Alveolar hydatid cyst mimicking cerebellar metastatic tumor". *Surgical Neurology International* 2 (2011): 13.

18. Piotin M., *et al*. "Disseminated intracerebral alveolar echinococcosis: CT and MRI". *Neuroradiology* 39 (1997): 431-433.

19. Senturk S., *et al*. "Cerebral alveolar echinococcosis mimicking primary brain tumor". *American Journal of Neuroradiology* 27 (2006): 420-422.

20. Stauga Sabine and Schmiedel Stefan. "Inoperable cerebral alveolar echinococcoisis controlled with high dosage of Albendazole adjusted with monitoring of blood levels". *Journal of Travel Medicine* 19.3 (2012): 198-201.

21. Takci Erhan., *et al*. "Alveolar echinococcosis of the brain in five patients". *Journal of Clinical Neuroscience* 15 (2008): 1105-1109.

22. Taxy Jerome., *et al*. "Echinococcosis: Unexpected Occurrence and the Diagnostic Contribution of Routine Histopathology". *The American Journal of Surgical Pathology* 41.1 (2017): 94-100.

23. Tünzün Meriç., *et al*. "Cerebral hydatid disease CT and MR findings". *Journal of Clinical Imagining* 26 (2002): 353-357.

24. Universität Zürich. Institut für Parasitologie (2000-2017) Surveillance of alveolar echinococcosis and control of Echinococcosis multilocularis in Switzerland (2019).

25. Wen Hao., *et al*. "Echinococcosis: Advances in the 21st Century". *Clinical Microbiology Reviews* 32.2 (2019): e00075-18.

26. Yibulayin Amina MA., *et al*. "Biological characteristics of 18F-FDG PET/CT imaging of cerebral alveolar echinococcosis". *Medicine* 97 (2018): 39.

Volume 15 Issue 5 May 2023