



Primary multiple cerebral hydatid cysts in an immunocompetent, low-risk patient



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ABSTRACT

Cystic echinococcosis is a zoonotic infection that occurs worldwide. Humans are infected through ingestion of parasite eggs in contaminated food, water or through direct contact with infected dogs, which are the definite host. Humans serve accidentally as intermediate host, and occurrences are common in children and young adults. Cystic echinococcosis is endemic in Mediterranean, South American, Middle Eastern, Central Asia, East Africa countries and Australia. The liver is the most frequently involved organ, followed by lungs. Hydatid cysts have been reported only in 2% of cases in the brain. Primary cerebral hydatid disease is a rare entity, but should be considered in the differential diagnosis of cerebral lesions.

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Introduction

Cystic echinococcosis is a zoonotic infection that occurs worldwide. It is produced by larval stage of the *Echinococcus* tapeworm (most commonly *Echinococcus granulosus*). The definite host of *Echinococcus* are various carnivores, the dog is the most common and the intermediate hosts are sheep, goats and cattle, which excrete the eggs in their feces. Humans are infected through ingestion of parasite eggs in contaminated food, water or through direct contact with infected dogs. Humans are accidental intermediate host, with most of occurrences in the children and young adults, who do not transmit the disease. Cystic echinococcosis is endemic in Mediterranean, South American, Middle Eastern, Central Asia, East Africa countries and Australia [1].

Although it is an uncommon parasitic infestation, it can be found in people who are in direct contact with canines and sheep, which form important links in life cycle of *Echinococcus*.

Larvae hatching from ingested eggs in the intestine enter the portal circulation, spreading to different tissues where they produce hydatid cysts [2]. The liver is the most common organ involved (77%), followed by lungs (43%) [3]. Hydatid cyst(s) have been reported in the brain in 2% of cases [4].

We describe the case of a 22-year-old Caucasian female. Multiple primary cerebral hydatid cysts, with obvious clinical symptoms and imaging findings, remains to our knowledge, an unusual case report from European countries.

Case report

A 22-year-old female presented in our hospital in 2018, with complaints of progressively worsening headaches and vomiting since the last 10 days. There were no significant findings on general physical and neurological examination. The patient's routine laboratory investigations were normal and she was treated for the diagnosis of upper dyspeptic syndrome. She presented again to the hospital after 11 days, where on physical examination she was conscious but aphasic. After 24 h she became stuporous and had a generalized tonic-clonic seizure.

On the MRI examination six supratentorial cerebral lesions were identified, described as intraparenchymal lesions of non-enhancing nature, with well-circumscribed borders and no perifocal edema, and with presence of thin septa, localized in the left frontal lobe (2.9 × 2.7 cm), in the left frontal-parietal lobe (4.6 × 3.8 cm), in the left temporo-parietal lobe (4.5 × 4.1 cm), in the right occipital lobe (3.1 × 2.7 cm) and two in the right frontal lobe (4.3 × 3.9 cm, 3.0 × 2.7 cm) [Fig. 1a–c].

Serological quantitative analysis, through detection of circulating hydatid antigen (CAG) in the serum by enzyme-linked immunosorbent assay (ELISA), confirmed the clinical suspicion. A titer of ≥ 1: 320 was considered as positive.

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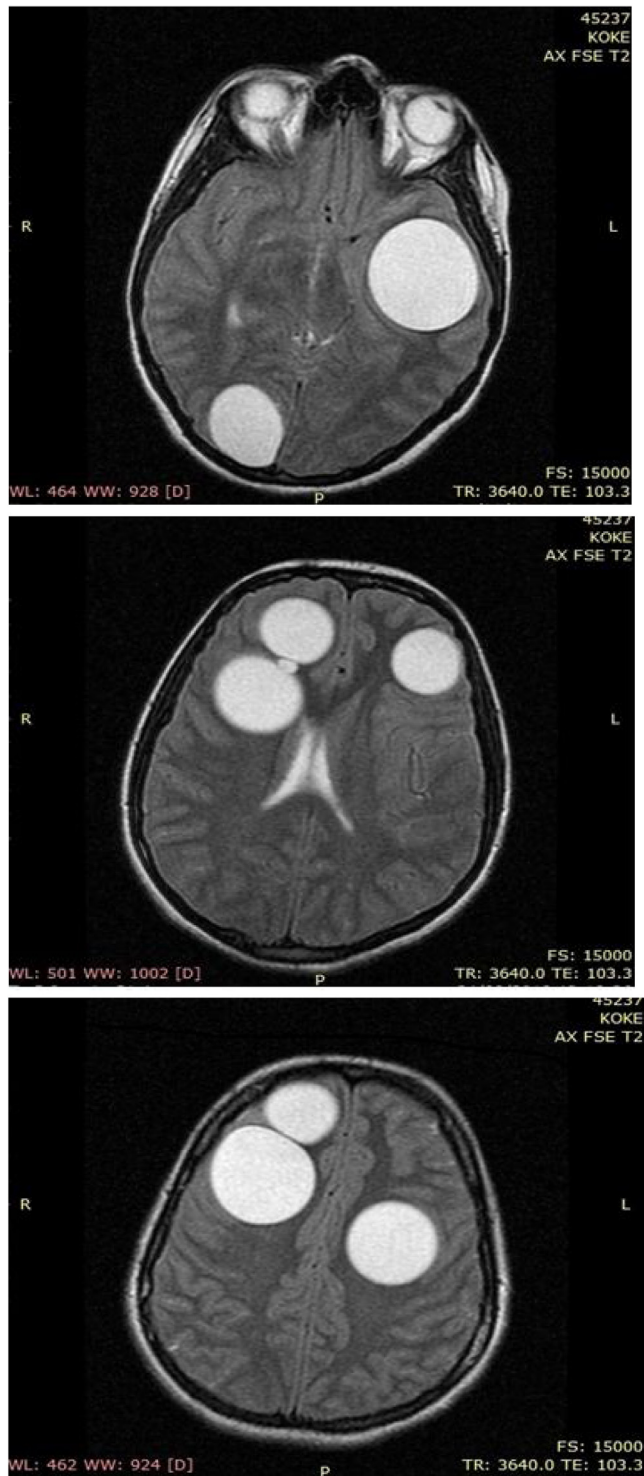


Fig. 1. a-c: Magnetic resonance imaging (MRI) in SAG SE T1, brain shows presence of six well defined non-enhancing cystic lesions in right and left cerebral hemisphere, and two of them in right frontal lobe with presence of thin septa.

The patient was living in urban zone, but she had a history of close contact with a pet dog during the previous two years. The thorax and abdomen were also examined with CT scans and no lesions were seen. Echocardiogram ruled out any cardiac disease. Eventually, she underwent surgery in another facility. To prevent recurrence, the patient was put on therapy with albendazole, 800

mg per day for three months, and anticonvulsants. Post-operative MRI showed encephalomalacic changes with no mass effect. No new cysts were detected in the brain or other organs, on the scheduled follow-up during the year post surgery.

Discussion

Cerebral echinococcosis is a rare manifestation as compared to the manifestations of other organs, for this reason there are few cases reported. Cerebral hydatid diseases are 2–3 times more common in children than in adults. The reason might be related to a patent ductus arteriosus in heart, allowing direct right-to-left shunting of blood flow in the heart, and thus evading the filtering by the lungs.

Cystic echinococcosis is endemic in Mediterranean Area, including Albania. Hydatid disease is one of the most common parasitic zoonosis in Albania. During 1959–1983, echinococcosis disease was evidenced in 1141 patients, including few cases with cerebral hydatid cysts. In a retrospective study on surgery case and autopsy findings, the incidence of human hydatid cysts was 2.05 per 100.000 inhabitants, for the period of 1958–1987. The fact that the occurrence has become almost a surgeon's exclusivity has recently raised doubts about its early detection and prompt conservative treatment [5].

The role of dogs in life cycle of *Echinococcus* in Albania was initially mentioned by Prokopić, but there are no local studies on the prevalence of canine *E. granulosus* [6]. Nevertheless, a recent study of Shukullari et al., detected no intestinal infection of *E. granulosus*, among 602 dogs involved in this study [7]. Despite this last data, the case that we present in this paper shows that hydatid disease is still present in our country, and its prevalence might be underestimated.

Brain hydatid cyst can be primary or secondary. Primary cerebral cystic hydatid cysts are unusual, resulting from direct infestation of the brain from the blood circulation after bypassing the others organs, such the liver and lungs, but without visceral involvement. Primary cyst usually is solitary and their rupture may result in recurrent hydatid cyst [8]. Primary multiple hydatid cysts are a rare pathological entity and only few case reports have been reported in the literature. Authors reviewing the literature have collected less than a score of reported cases of primary multiple cerebral hydatid cysts, during the period 1982–2008 [9].

It is important that after surgery and medical treatment, patients should be followed up regularly in order to recognize any possible recurrence [10]. This will prevent further dissemination and complications of the disease, as well as any early emergence of hydatid cysts in other organs where they can growth slowly (liver, lungs).

Conclusions

Our patient's case is important to report, because the primary multiple cranial hydatid cysts in adults are rare, both in literature, and even more in European countries. Hydatid disease is often neglected even in endemic areas or not early diagnosed till the lesions assumes an enormous size. It seems an dramatic and uncommon disease, but is a totally curable disease. Control and vaccination of the intermediate hosts is important to interrupt the transmissive cycle, and thus to prevent human infection.

Author statement

ER, EK and ER are the treating clinicians that have diagnosed, treated and followed-up the case. ER and GV wrote the manuscript and reviewed the literature. All authors have approved the final submitted version.

Declaration of Competing Interest

Nothing to disclose.

Acknowledgments

None.

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