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## CASE REPORT

Echinococcus Infestation of the Central Nervous System as the Primary and Solitary Manifestation of the Disease: Case Report and Literature Review

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## ABSTRACT

**Introduction:** Hydatid disease of the central nervous system, especially when there is no involvement of other organs, is a rare occurrence and it accounts for 2–3% of all the hydatid cyst cases that are recorded worldwide. Echinococcus is considered as an endemic disease, mainly encountered in sheep and cattle-raising regions, with a large proportion of patients living in Mediterranean countries, as well as Turkey and Syria. Pediatric neurosurgeons in nonendemic countries are facing a differential diagnostic challenge when a patient with hydatid cyst of the central nervous system is admitted to their department, mainly due to lack of awareness.

**Materials and Methods:** We report the case of an eleven years old patient, coming from a village, who was admitted to our hospital due to progressive hemiparesis, hemianopsia, headache and episodes of vomiting. A thorough laboratory and neuroimaging investigation was performed, which revealed the presence of a hydatid cyst at the right fronto-parietal region. An en-block total resection was performed and the patient neurological signs and symptoms were immediately relieved.

**Conclusions:** The clinical features related to hydatid cyst involving the central nervous system are largely nonspecific. Nevertheless, MRI has proved to be a very important diagnostic tool, significantly enhancing the diagnostic aid of neuroimaging and supporting our pre-operative diagnostic accuracy. Laboratory investigation with immunological tests may be indicative, but negative results should be interpreted with caution. En block removal of the cyst without interruption of its wall and avoidance of any spillage of content is our ultimate therapeutic goal. Infection with Echinococcus granulosus should be included in the differential diagnosis for non-specific neurologic symptoms such as progressive headache, especially in pediatric patients who are coming from regions which are considered as endemic for such parasitic diseases. In addition, the adjuvant therapy with albendazole is strongly indicated. The majority of misdiagnoses or delayed diagnoses may have resulted from confusing clinical features and atypical radiographic findings.

## Abbreviations

CT: computed tomography  
EEG: electroencephalography  
MRI: magnetic resonance imaging  
SE: spin echo  
DWI/ADC: diffusion weighted imaging/apparent diffusion coefficient  
FLAIR: fluid-attenuated inversion recovery  
3D BRAVO ISO: 3-dimensional brain volume  
T2\* GRE: gradient recalled echo  
FRFSE: fast relaxation fast spin echo  
FSPGR: fast spoiled gradient echo  
T1W: T1 weighted  
T2W: T2 weighted  
CSF: cerebrospinal fluid  
MEPs: motor evoked potentials  
SSEPs: somatosensory evoked potentials

## Introduction

Parasitic infection that involves the central nervous system, albeit rare in the countries of the Western world, continues to affect a significant amount of people that live in the developing world, the main reason being poverty and associated conditions. However, it seems that an evolving change in the geographic distribution of these diseases is currently in progress, and that should be investigated under the general term of globalization<sup>1</sup>. In general, hydatid disease, caused by the cestode *Echinococcus*, is well-known to be associated with a worldwide prevalence. Nevertheless, it is considered to be endemic in parts of Latin America, as well as Australia, Mediterranean countries, the Middle East, and India. All these areas share in common the fact that sheep and cattle raising are part of their daily living. The definitive hosts for *Echinococcus* are canines such as dogs, wolves, and foxes<sup>2</sup>.

Currently, the most common genus in humans is considered to be *Echinococcus granulosus*, which involuntarily serves as intermediate hosts through the ingestion of contaminated food. Viable parasite eggs form oncospheres in the human intestine. These penetrate the mucosa, which is followed by hematogenous spread to the liver (75%), lung (15%), and brain (2%)<sup>3,4</sup>. As far as the predilection of distribution of the hydatid cysts is investigated, the central nervous system, and the brain in particular, is considered to be an uncommon site of involvement. More precisely, brain involvement occurs in only 2% of human hydatid infestations<sup>1, 5-15</sup>. Among all of these affected patients, a definite

predilection for the pediatric population is universally recorded<sup>16-18</sup>. Regarding the distribution of hydatid cysts, they are most commonly described as distributed in the territory of the middle cerebral artery, although several cases have been described where cysts have been identified elsewhere. Namely, they have been recognized in an infratentorial location<sup>2,8</sup>, intraventricularly<sup>19</sup> and in the spine<sup>15</sup>. Although these cysts are usually recognized as solitary, unilocular lesions, multiple primary cerebral cysts have been reported<sup>13</sup>.

Cyst rupture is not a common occurrence, but cases of spontaneous rupture or associated with trauma have been reported<sup>1</sup>.

## Case description

We report the case of an eleven years old boy, generally fit and well, which is a Greek citizen, living at a village in the country. He was admitted to our hospital due to a recent onset headache, which was gradually worsening the previous days before his admission. Moreover, several episodes of vomiting were reported, albeit the level of consciousness was not impaired.

A detailed neurological examination was performed, which revealed bilateral papilledema, without any impairment of the ocular mobility. Moreover, a left-sided hemiparesis was noted, with the upper extremity being more severely affected than the lower one. The ipsilateral deep tendon reflexes were increased, indicating a lesion that was impairing the pyramidal tract. Finally, left-sided hemianopsia was detected, although it was neglected by the patient.

The patient underwent a CT scan on an emergent basis, which revealed an extensive mass lesion, involving the right fronto-parietal region, with low signal intensity characteristics, well-circumscribed, without perilesional edema (Figure 1). A midline shift to the contralateral side was noted and at the periphery of the lesion, a distinct area of increased signal density, adhered to the cyst wall, was noticed. After intravenous contrast injection, no additional imaging findings were noticed. Moreover, a transesophageal echocardiography was performed, in order to rule out heart involvement by the disease. This examination also proved to be negative. Moreover, in order to exclude the possibility of subclinical epilepsy, an EEG was performed, which did not reveal pathological findings

Figure 1



**Figure 1.** Preoperative CT scan, indicating the presence of a well-circumscribed, encapsulated mass, without perilesional edema, located in the right fronto-parietal region. A thin rim of hyperdense material is noticed in the wall of this lesion.

Our initial differential diagnosis included supratentorial low-grade astrocytoma with an accompanied mural nodule, or echinococcus infection of the central nervous system. Our laboratory investigation included measurement of antibodies (IgG) with the ELIZA method, against different parasites (*Taenia solium*, *Echinococcus granulosus*, *Entamoeba histolytica*, *Toxocara* spp) and against *Toxoplasma* spp (IgM, IgG) -all of them proved to be negative.

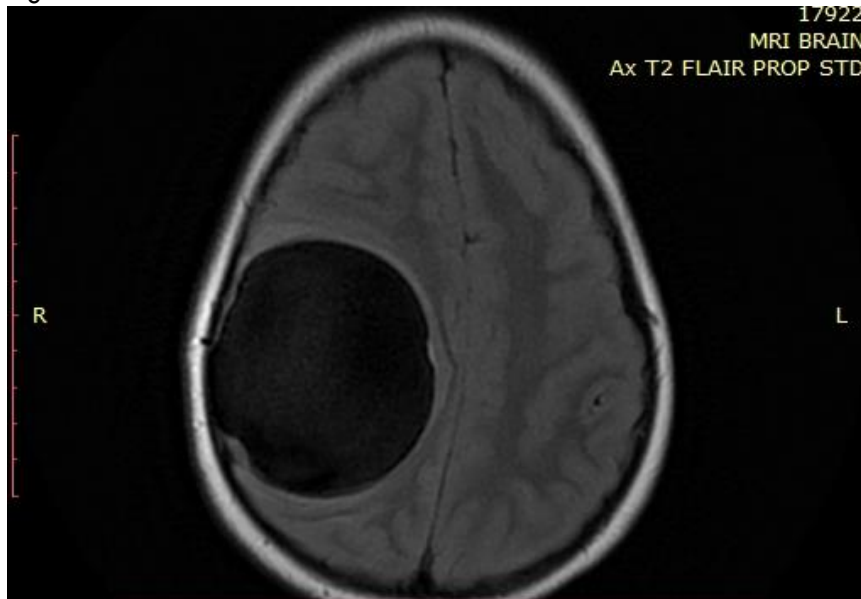
Subsequently, an MRI scan of the brain was performed, with and without the administration of an intravenous paramagnetic substance. The examination included SE, DWI/ADC, FLAIR, CUBE T2W, 3D BRAVO ISO, T2\* GRE, FRFSE and FSPGR sequences. It revealed a single, circular, large cystic lesion, located in the supratentorial brain compartment, more precisely in the right cerebral hemisphere. Its approximate dimensions were 6,3X7,3X7 cm (transverse, anteroposterior and rostro-caudal respectively). It was located in the right frontal and parietal lobe and it extended from the level of the right lateral ventricle to the supra-ventricular area and it was compressing the adjacent cerebral structures. More precisely, a compression effect was noticed on the ipsilateral island of Reil, basal ganglia and thalamus, as well

as on the corpus callosum. There was a degree of effacement of the ipsilateral sulci, along with contralateral displacement of the midbrain. The aforementioned mass lesion was compressing the right atrium of the lateral ventricle, with accompanying effacement of the ipsilateral occipital horn and simultaneous dilation of the central part (body) of the lateral ventricle. A moderate enlargement of the contralateral ventricular system at the level of the trigone of the occipital horn, along with mild periventricular edema, was recognized. This mass lesion was well-circumscribed, its margin was well – illustrated and it seemed to be encapsulated, harboring at several sites high signal intensity at T1W sequences.

It seemed to contain a liquid material, characterized by low signal intensity at T1W sequences, high signal intensity at T2W sequences (similar to that of CSF), without diminishment of diffusion and without contrast enhancement after intravenous administration of gadolinium. At the periphery of the lesion, we could recognize areas with thickening of the wall of the capsule, with protrusion into the cystic cavity. These are characterized as areas of intermediate-high signal intensity at T2W sequences, intermediate-low signal at T1W sequences, without diminishment of

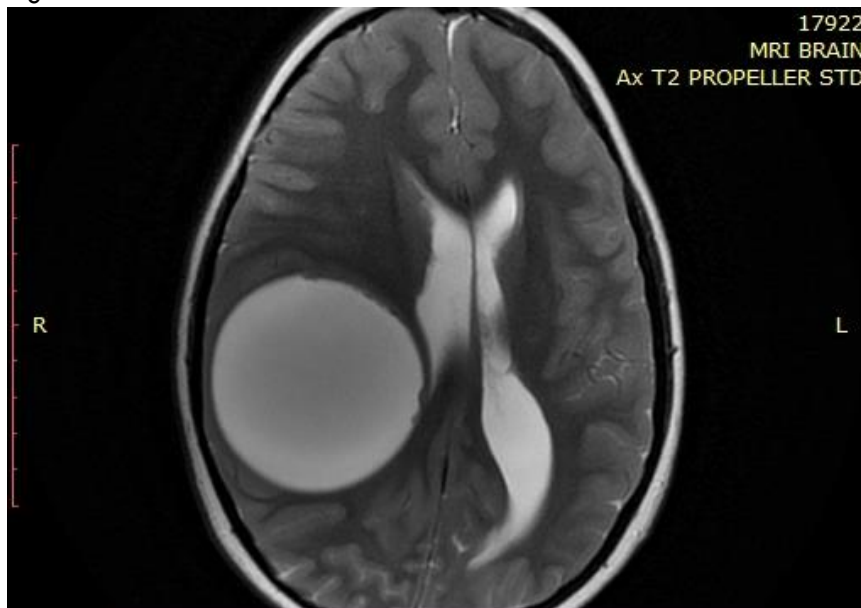
diffusion. Moreover, neither hemosiderin deposits, nor enhancement after intravenous contrast administration was mentioned (Figures 2-5).

Figure 2



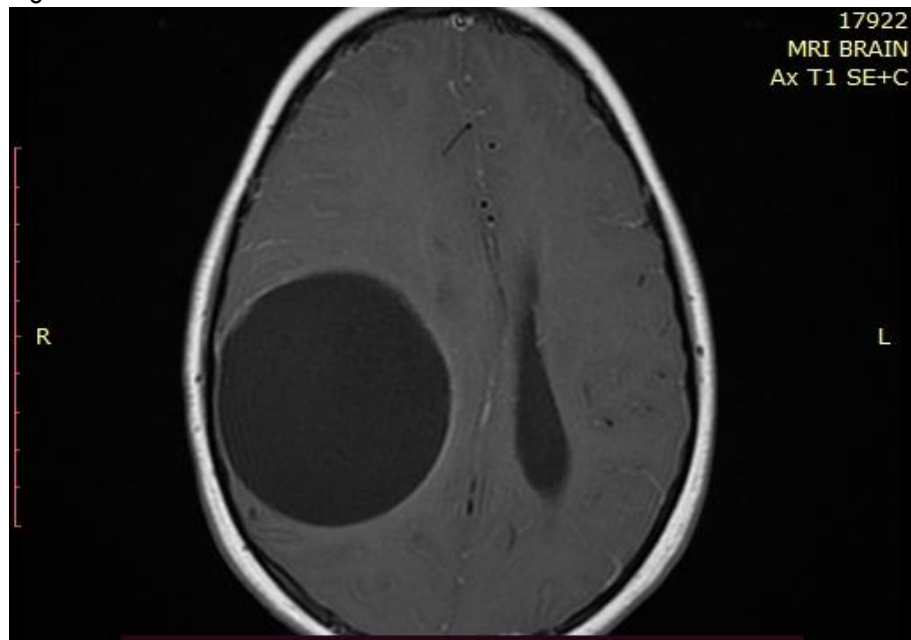
**Figure 2.** Preoperative MRI scan, axial T2 FLAIR sequence, exhibiting the compression of the mass to the nearby brain structures.

Figure 3



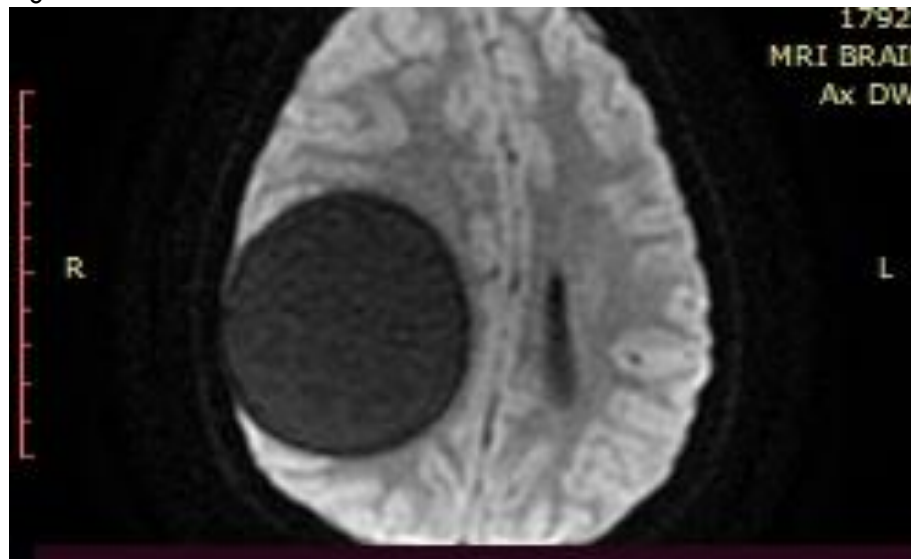
**Figure 3.** Preoperative MRI scan, axial T2 propeller sequence, indicating the high signal intensity of the cystic fluid, as well as the intermediate signal intensity of the thickened wall of the capsule.

Figure 4



**Figure 4.** Preoperative MRI scan, axial T1 Weighted sequence, after the intravenous administration of gadolinium. No significant enhancement of the wall of the cavity is evident.

Figure 5



**Figure 5.** Preoperative MRI scan, Diffusion-Weighted Image, indicating no restriction of the diffusion signal of the offending lesion.

As far as the MRI was unable to definitively determine what was the diagnosis of the underlying lesion, we performed an ultrasound of the abdomen, in order to exclude the possibility of the existence of other lesions occupying the patient liver or other organs, as it could be expected in the case of an Echinococcus infection. This examination did not reveal any relevant findings and we continued our diagnostic evaluation with a computed tomography of the chest and abdomen, without and

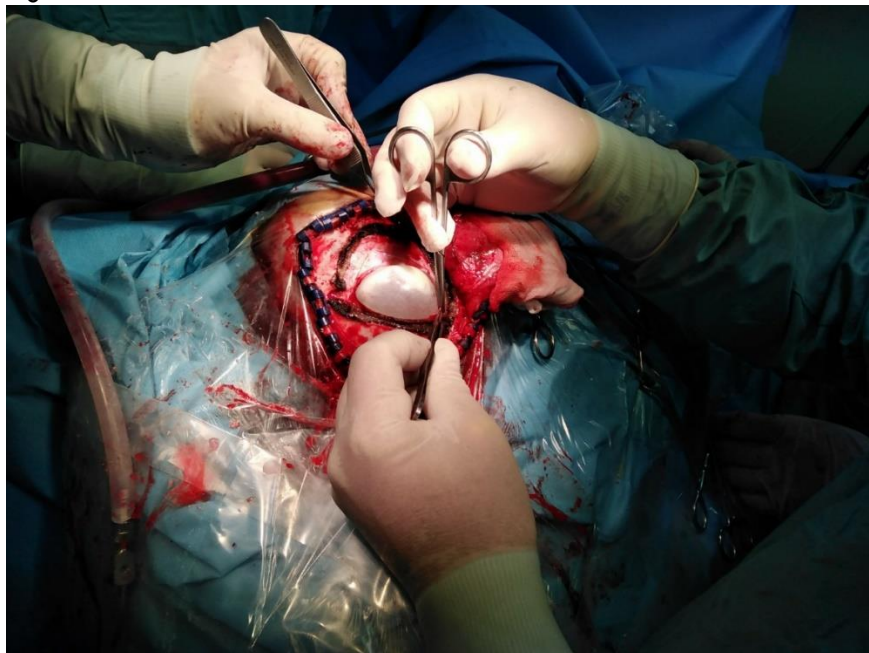
with intravenous contrast administration. These studies did not add any other relevant findings.

The patient underwent an operation in order to excise the offending lesion. A right fronto-temporo-parietal skin incision and a frontoparietal craniotomy was performed. Under intra-operative ultrasound guidance, the cystic lesion was recognized, the dura was opened, a limited cortical incision was performed and the offending pathology was revealed. The cortical-cystic margin

was intraoperatively developed using blunt dissection and an attempt was performed toward an en block removal of the cyst. Intraoperatively, neurophysiological monitoring was performed (MEP, SSEP's) and strip and grid electrodes were utilized in order to identify the location of the motor cortex and if there was any vicinity of this brain region with the offending pathology. As soon as no vicinity of the lesion with the primary motor cortex

was recognized, we continued on our operation in order to excise the pathological entity. The cyst was totally removed without being accidentally opened and after its resection, the cyst wall was incised. It was filled with fluid that was not lucent (seemed to be proteinaceous) and, regarding the cyst wall, our intraoperative findings were consistent with hydatid cysts (Figure 6-8).

Figure 6



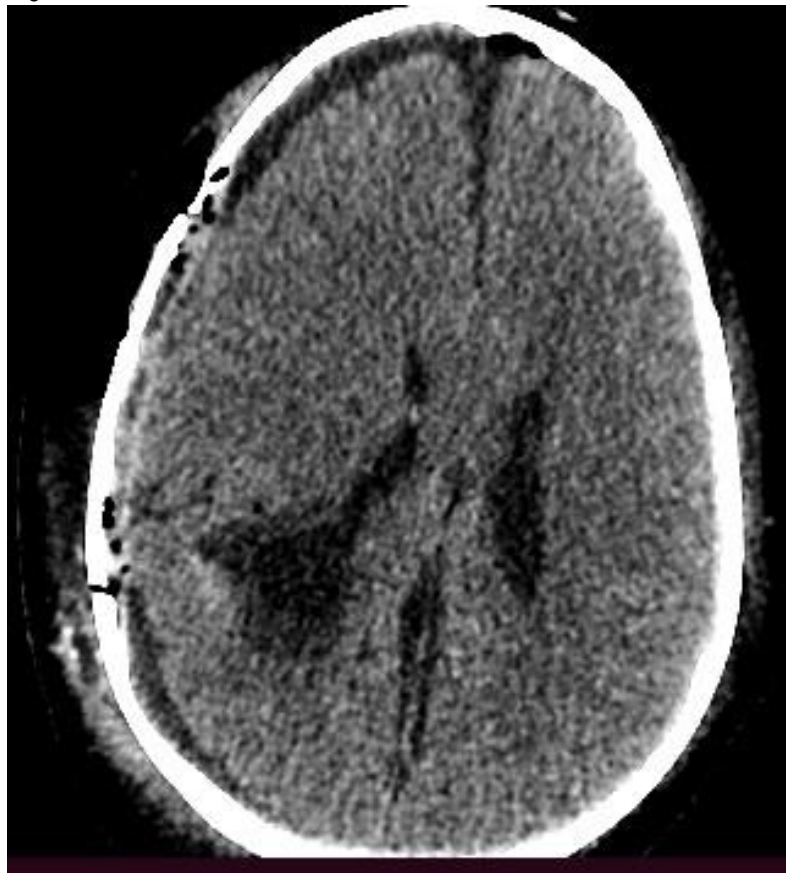
**Figure 6.** Intra-operative photograph, indicating the presence of the cystic lesion which is well-encapsulated.

Figure 7



**Figure 7.** Intra-operative photograph, after complete resection of the cyst. This icon indicates the existence of hydatid cysts at the wall of the extirpated cyst.

Figure 8



**Figure 8.** Post-operative CT scan (first post-operative day). It indicates complete extirpation of the cyst, without any adverse post-operative sequelae.

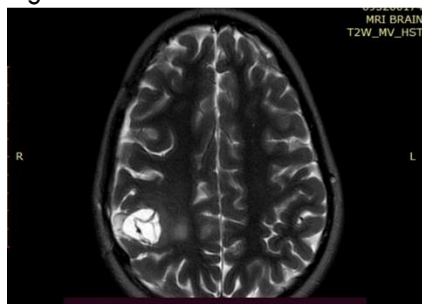
The whole specimen (cystic fluid and wall of the lesion) was sent for microbiologic and histopathological examination, which verified the existence of hydatid cysts, that is Echinococcus infection of the central nervous system. We examined the cystic fluid for antibodies against Echinococcus granulosus, but the result was negative.

The patient recovered immediately after the operation and no neurological deficits were reported. On the contrary, the papilledema

subsided, as well as the contralateral hemiparesis and the associated hemianopsia.

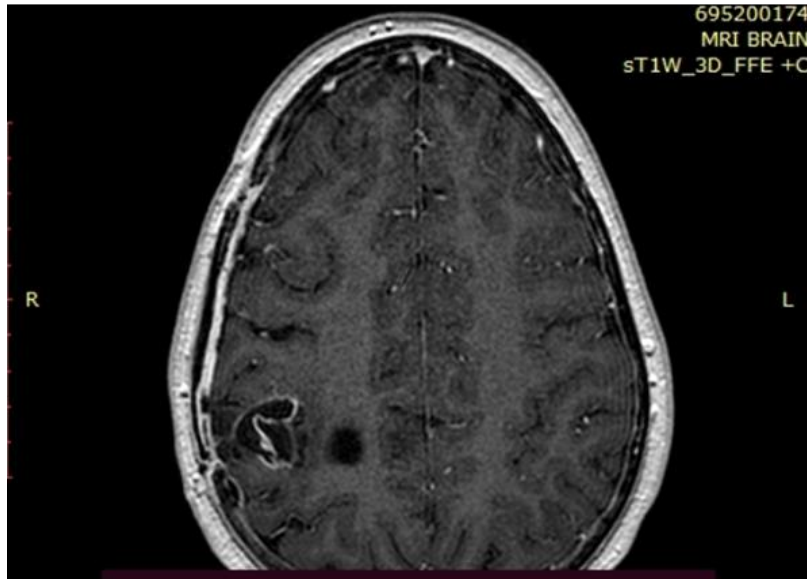
After the verification of the Echinococcus infection, the patient is on anti-parasitic therapy for three months and a post-operative MRI was performed three months after the operation, in order to verify the completeness of the resection. This MRI scan verified the completeness of the previous resection (Figure 9,10).

Figure 9



**Figure 9.** Post-operative MRI scan, T2 W, depicting the completeness of the resection of cystic lesion. The remaining fluid cavity is fulfilled with CSF and a fibrous diaphragm traverse it.

Figure 10



**Figure 10.** Post-operative MRI scan, T1 W, 3D, after intravenous injection of gadolinium. Same findings as previously. The absence of enhancement of the wall of the remaining cystic cavity verifies the absence of Echinococcus infection.

## Discussion

Hydatid disease constitutes a complex medical and economic problem that mainly affects the developing countries<sup>20</sup>. More precisely, Echinococcosis is rarely encountered in countries of the Western World, although it remains a public health issue in endemic areas that is difficult to be managed<sup>21-23</sup>. Echinococcosis is one of the most common parasitic infections involving humans, and is equally distributed among both sexes according to the literature, it is presumed that the younger age groups are more severely affected<sup>24</sup>. The definite hosts of Echinococcus are a variety of carnivores, and dogs are the most common host. Sheep, cattle, and human beings can act as the intermediate host. The transmission routes of echinococcosis are through contacting with definite host or consuming contaminated water or food. At the initial stage of contamination, the eggs of Echinococcus lose their enveloping layer in the stomach, and the embryos are released. After that, they pass through the wall of the gut into the portal system and they are transferred to the liver. Most larvae would be entrapped and encysted in the liver. However, a small subpopulation of larvae could potentially be entrapped to the lungs, or alternatively enter the systemic circulation. This could happen if they circumvent the capillary filter of the liver and lungs. Another, albeit, more rare possibility, includes the scenario that some larvae reach the brain or spinal cord. Brain echinococcosis seldom occurs in an

independent fashion, as is most commonly associated with liver and lung infections. When cerebral hydatid cysts are considered, a comprehensive literature review suggests that the majority of cases involve the supratentorial compartment, whereas a minority are encountered infratentorially<sup>25</sup>. More specifically, they are located within the cerebral hemispheres, although anecdotal cases exist with atypical locations, such as thalamus or ventricle<sup>26</sup>. When cases with CNS involvement are studied, solitary lesions were most commonly referred, and only a small percentage of patients were suffering from multiple cyst involvement<sup>27,28</sup>.

Regarding the neurological signs associated with patients harboring echinococcosis, they resemble that which are attributed to elevated intracranial pressure due to a space-occupying intracranial lesion of any kind. Apart from that, these lesions are slowly growing and this may be responsible for the delayed onset of the offending symptomatology. The aforementioned issues render a detailed medical record mandatory, and this should be supplemented with an auxiliary examination. There are several points that deserve special attention: (1) the patients can exhibit a variety of clinical and neurological manifestations, from being completely asymptomatic, or presenting with elevated intracranial pressure, epilepsy, and local neurological deficits. (2) imaging examinations should be executed, in order to aid in the



differential diagnosis of these lesions<sup>29</sup>. A CT scan usually indicates the presence of a spherical and well-circumscribed, smooth, thin walled, homogeneous cystic lesion containing a fluid with a density similar to the CSF<sup>5,7</sup>. The cyst is non-enhancing after contrast administration and not accompanied by perilesional brain edema. In the majority of cases, midline shift to the contralateral side and ventricular compression is observed, as is the case with our patient. Based on that imaging characteristics, differential diagnosis includes cystic tumors, pencephalic cysts, brain abscess, or arachnoid cysts<sup>29</sup>. Nevertheless, MRI is considered to be the imaging modality of choice in order to verify our diagnosis and to obtain all the necessary information in order to make our surgical plan<sup>30</sup>. The classic MRI appearance of cerebral echinococcosis consists of a spherical or oval-shaped lesion, which is characterized by well-defined borders, it is well-circumscribed and surrounded by a thin-walled capsule. It is filled with fluid that is homogeneous and its signal intensity characteristics are similar to that of CSF<sup>31</sup>. The cyst wall is seen as a rim of low signal intensity on T1- and T2-weighted images, whereas perilesional edema may be detected, although it is rarely present. (3) In order to establish the diagnosis, a positive hydatid immunological examination is helpful, although it could not be considered as a prerequisite requirement. This means that the diagnosis could not be doubted only because a negative result is obtained besides, this was the case to our patient.

Based on the aforementioned data, we would like to underline the fact that the diagnosis of echinococcosis should be supported by a constellation of data, including a detailed patient history, a comprehensive neurological evaluation, a thorough radiographic evaluation (CT and MRI), along with a relevant laboratory investigation. This is due to the fact that neither the clinical investigation nor the neuroimaging results could independently be considered as pathognomonic. Because of that, there should be good feedback between the reporting clinician, the neuroradiologist and the laboratory team. Moreover, misinterpretation of the results is more frequently observed in cases of brain involvement, mainly due to the existence of the blood-brain barrier, as well as in cases of immunocompromised patients, where the laboratory results may be impaired. Children are included in this category, and this may be the reason of the negative

immunological results that were obtained from our patient.

There is no published study to report cases of echinococcosis that were adequately treated only with pharmacological therapy, without any surgical intervention. Current guidelines support surgery as the preferred therapeutic option. Another important issue is that the removal of the unruptured cyst should be performed in an un-block fashion. If inadvertent cyst rupture exists, cystic contents may enter the subarachnoid space, resulting in a widespread dissemination of scolices. This would lead to severe inflammation, and secondarily, to anaphylactic reactions. This is presumed to be caused by the cystic content which contains numerous antigens. Pharmacological therapy is considered to serve as an adjunct for therapy, and it mainly based on albendazole. Nevertheless, extensive data are not available regarding the pharmacological treatment of hydatidosis of the central nervous system. For example, there is lack of definitive evidence centered on drug penetration across both the blood-brain barrier and the membrane of hydatid cyst.

It would be important to state why this case report is remarkable and the information provided is relevant, because all what is presented is aligned with current state of the art regarding this pathology. The highlights of this specific case are centered on two separate distinguishing features. The fact that the only manifestation of the disease was the space-occupying lesion of the brain constitutes a rare manifestation of the disease, which deserves special notification. The other peculiar characteristic of this individual case is related with the discrepancy between the clinical and neurological findings of the patient, which were moderate in nature, and the dimensions and the anatomical location of the offending lesion. Based on the imaging findings, the expected clinical condition of the patient should be devastating, a hypothesis that is diametrically opposed with the initial clinical presentation of the patient. According to our opinion, this is intimately related to the gradual expansion of the dimensions of the cyst, which allowed for the brain compliance to compensate for the increasing intracranial volume. This is a fact that should be taken into consideration and renders this case report worthy of publication.

**Conclusion**

Hydatid disease is a worldwide zoonosis produced by ingesting the larval stage of the echinococcus tapeworm. Although it is rarely encountered in the Western World community, it continues to be an important health and social-economic issue in developing countries, mainly due to its widespread endemic nature. Although the combination of medical history and CT imaging findings is usually adequate in order to establish the diagnosis, there are cases which could represent main diagnostic dilemmas, especially when the laboratory investigation is non-diagnostic. We would like to draw attention of the medical community to the fact that the diagnosis of cerebral hydatid cyst, even when it is not accompanied by

other organs involvement, should always be considered. This is especially true in every case where an atypical CT and MRI imaging is encountered, even in individuals who do not come from endemic areas and their medical history is not indicative of such a diagnosis.

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