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Primary Multiple Cerebral Hydatid Cyst in 8 Year-Old Girl: A Rare Cause of Childhood Seizure

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Abstract

Background: 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. Multiple cerebral hydatid cysts are very rare with only a few reports in the literature. Case Description: We present the case of an 8-year-old girl who presented with focal seizures, hemiparesis, headache, vomiting and bilateral optic atrophy. Diagnostic workup was performed, and magnetic resonance imaging revealed multiple intracranial cysts predominantly in the right frontal region with significant mass effect. A total of 11 intracranial cysts were removed surgically, and the child recovered uneventfully. Conclusion: Neurosurgeons should keep hydatidosis in the list of differentials when evaluating patients with cystic diseases of the brain. Although the removal of such cysts is challenging, outcomes are excellent when cysts are evacuated without rupture and patients show complete resolution of symptoms.

Keywords

Hydatid Cysts, Multiple Hydatic Cysts, Childhood Seizures

1. Introduction

Hydatid disease (echinococcosis) is a worldwide zoonosis produced by the larval

stage of the Echinococcus tapeworm [1]. This is an endemic manifestation that is more frequently found in Mediterranean countries, the Middle East, South America, and Australia, and this entity affects particularly those involved with sheep and cattle rising [2].

Hydatid cysts occur most commonly in the lung and liver with intracranial involvement reported in 0.5% - 3% of all hydatid disease [3] [4] Cerebral hydatidosis is thus a rare disease entity and represents 0.05% of all intracranial mass lesions in the developed world. Primary cysts are often solitary, spherical, unilocular lesions, and can be surgically excised [5] Multiple cysts are very rare with only a few reports in the current literature [3] [6]-[13].

Surgical excision of multiple cysts can be very challenging as these are often disseminated and involve vital structures. Here, we report the case of an 8-year-old girl with 11 intracranial cysts, revelated by epileptic attack, which were successfully removed surgically and also review the literature with an emphasis on diagnosis and management of patients with multiple cerebral cysts.

2. Patient and Observation

This 8-year-old girl, from a rural area, previously healthy, was admitted to the emergency department for an epileptic attack, she described seven episodes of partial seizure secondary generalized.

Neurological examination revealed postictal confusion, hemiparesis and bilateral papilledema. Headache and vomiting were not experienced. An anticonvulsant therapy was administrated. The patient's routine laboratory investigations were normal. A magnetic resonance imaging (MRI) brain (**Figure 1**) was

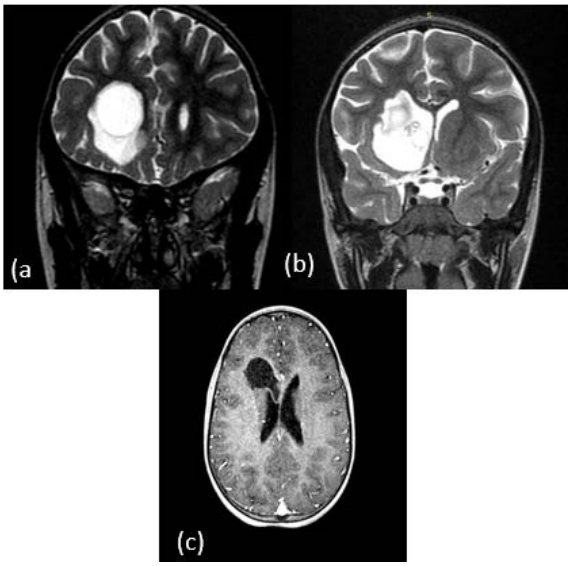

Figure 1. (a) (b): Coronal T2-weighted MRI of the brain showing the right frontal hyperintense multiple intracranial multivesicular cysts. (c): Post-contrast axial T1-weighted MRI shows homogenous low signal intensity of the cyst with mild marginal contrast enhancement.

performed which showed multiple intracranial cysts predominantly in the right frontal region without significant mass effect with calcifications, no surrounding edema, and no contrast enhancement were seen. Preoperative radiological diagnosis of cerebral hydatid cyst was suggested. The patient's chest X-ray, ultrasound abdomen, and eosinophil counts were normal.

The family refused the intervention then lost follow-up. Nine months later, the patient was readmitted to the emergency department for the management of absence seizures. Neurological examination revealed Glasgow Coma Scale score of 8 and left hemiplegia.

A computed tomography (CT) scan (Figure 2) of the head showed hydrocephalus with multiple intracranial hydatid cysts in the right frontal.

Based on the cranial imaging findings and the patient history, the diagnosis of recurrent cerebral hydatid disease with multiple cysts of the brain was suggested.

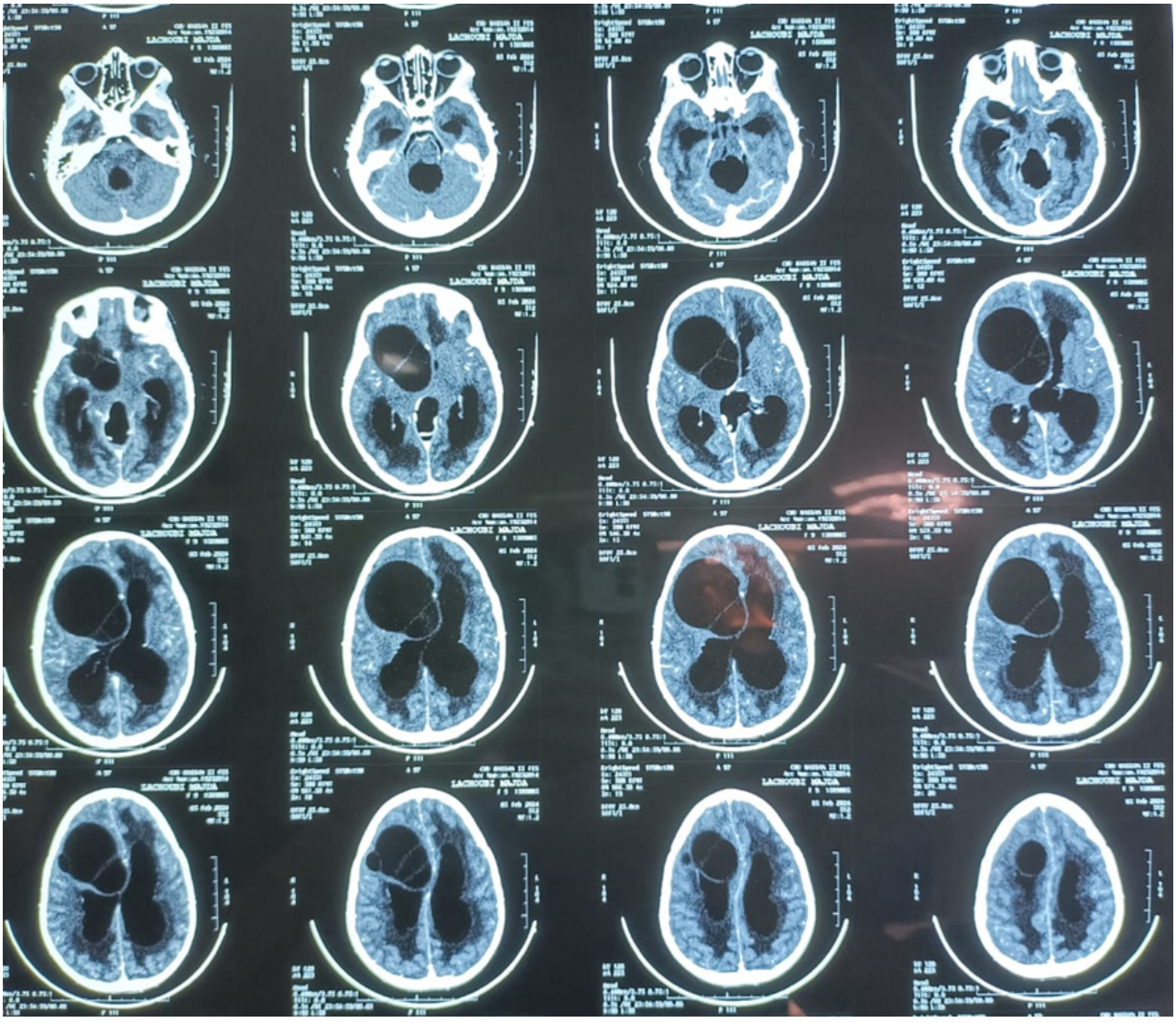

Figure 2. CT scan showing axial image of multiple intracranial multivesicular cysts in the right frontal region with hydrocephalus causing mass effect and midline shift to the left. There is no peri-lesional oedema.

Upon surgical exploration, using the technique of hydrodissection, as described by Arana-Iniguez and San Julian, the cysts were totally extracted without rupture. The surgical areas were cleaned with sodium chloride. The removed material was sent to the Parasitology Laboratory for analysis.

The macroscopic analysis of the surgical specimen revealed spherical cysts with whitish walls (**Figure 3**). Several pearly white translucent daughter cysts of variable sizes, with clear fluid and membranous structures, were observed inside 8 cysts.

Histopathologic examination of the specimen confirmed the diagnosis of hydatid cyst by visualizing the laminated membrane of the cyst wall.

The patient's postoperative course was uneventful; the patient's neurological status remained good. Postoperative CT (**Figure 4**) confirmed total removal of the lesion our patient was treated with albendazole. She stayed in the intensive care unit for two days after the operation and was then transferred to the Neurosurgery Department. There is without post-operative complication. Therefore, she was discharged with long-term prescription of oral albendazole at 400 mg/day given in cycles of 28 days interrupted by a 15-day period with a routine monitoring of blood counts and liver function tests (in view of the potential hepatic and hematological adverse effects of albendazole).

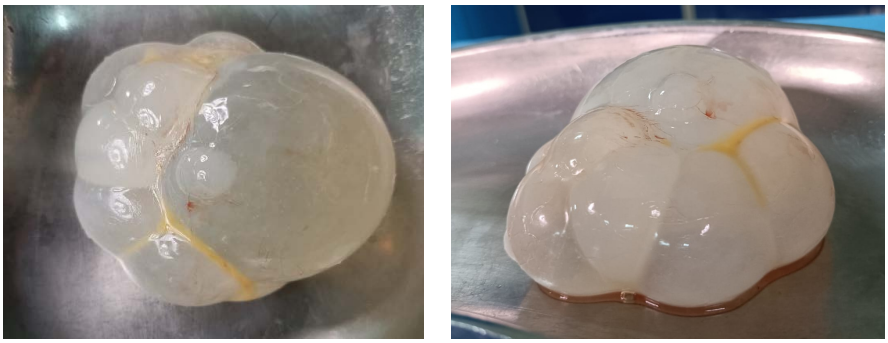

Figure 3. Macroscopic findings of extracted hydatid cysts. Whitish cysts filled by multiple variable-sized daughter cysts.

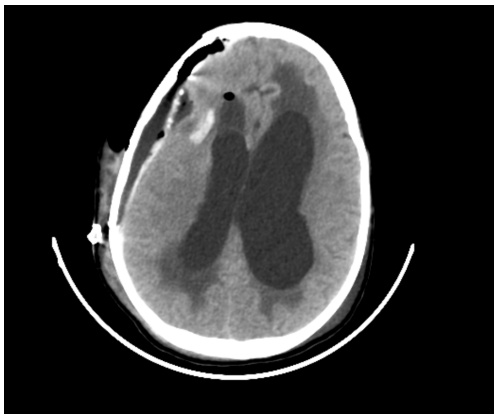

Figure 4. Post-operative CT scan of the lesion shows a large void. A small haematoma is also seen.

3. Discussion

Primary intracranial hydatid cyst is generally solitary, whereas secondary cysts are multiple. The cerebral hydatid cyst is secondary to the development of the Echinococcus Granulosis parasite in the intracranial. Normally the dog is the definitive host, but the cycle can be broken, and man can be an accidental host [14].

Hydatidosis is currently considered as an endemic zoonotic disease in Morocco. A total of 23,512 human cases were noticed by the Ministry of Health in Morocco during the periods 1980-1992 and 2003-2008 [15].

Hydatid disease occurs by infection with the larval stage of the tapeworm Echinococcus granulosus. Humans become infected by ingestion of eggs from the dog hydatid tapeworm due to poor hygiene practices or contaminated food and water [16].

Humans acquire the disease mostly during childhood. The liver (50% - 77%) and the lung (8.5% - 43%) are the organs most commonly involved. The remaining lesions may involve any organ in the body, including cerebral involvement in approximately 2% of patients infected with the parasite [1]. They may reach a considerable size before the patient becomes symptomatic. There is no consensus on the growth rate of the hydatid cyst of the brain and has been variably reported between 1.5 - 10 cm per year [17].

Cerebral cystic echinococcosis is most commonly seen in children and young adults (approximately 50% - 70%) [1]. El-Shamam *et al.* [8] and Lunardi *et al.* [9], reported that cerebral hydatid cysts are commonly seen in children especially in males and young adults. Beskonaklí *et al.* [10] reported a reasonable explanation for this, as young male children were more closely occupied with animals than girls or adults and were not as aware of the importance of hygienic principles.

Hydatid cysts are generally located in the territory of the middle cerebral artery and posterior fossa, or infratentorial lesions are very rare [3] [7].

The neurological examination is very polymorphic and can evolve from an asymptomatic picture to coma (behavioral disorder, motor deficit, cranial pair damage, consciousness disorder) [18] [19]. The cranial CT scan shows a rounded, well-limited image with a fluid density close to that of the CSF, homogeneous without peripheral edema [20] [21] [22]. The hydatid cyst may take on contrast in a peripheral ring with perilesional oedema when it is infected [23] [24].

Brain hydatidosis can be primary or secondary. The direct invasion of the brain by the larvae, without affecting other tissues, represents the primary form. In our case, cysts contain brood capsules released from the germinal membrane and protoscolices. Primary cysts are characterized by high fertility, and their rupture may be responsible for recurrent forms. Secondary cysts result from a fertile primary cyst, which has ruptured following trauma or during surgery or spontaneously. These cysts lack germinal membranes and are infertile. In case of their ruptures, the recurrence is exceptional [25].

Both CT and MRI scan have been reported to be useful in the correct preo-

perative diagnosis of cerebral hydatid cysts [6] [7] [10] [12]. The lesions appear as hypodense, intraparenchymal circular lesions without perifocal edema, and a hyperdense rim on noncontrast CT scans [1] [12] [13]. However, MRI is superior to CT scan as it shows greater soft tissue detail and can define the anatomical location of the lesion relative to sulci and ventricles, which aid in operative planning [12]. On MRI, the cyst gives hypointense signals on T1-weighted images, and hyperintense signals on T2-weighted images, and the cyst wall gives hypointense signals on both T1- and T2-weighted images [4]. Calcification of the cyst wall is rare and reported in about 1% of the cases [2]. The presence of daughter cysts is considered pathognomonic [2]. Other differentials for cystic lesions include cystic tumors, pyogenic brain abscess, and cystic lesions such as porencephalic cysts or arachnoid cyst [7] [12]. These can be differentiated from hydatid cysts as cystic tumors have an enhancing mural nodule and tumor edges, and the central necrosis of pyogenic abscess is almost always accompanied by peripheral edema with contrast enhancing margins. Similarly, unlike hydatid cysts, porencephalic and arachnoid cysts are not spherical and are not surrounded by brain parenchyma [2]. Moreover, arachnoid cysts are always extra-axial, and porencephalic cysts have a rim of gliotic white matter that is, easily observable on MRI [2].

Serological tests have traditionally had low sensitivity and specificity in diagnosing intracerebral hydatid cysts [2] [4]. Serological tests were normal in our patient as well, and thus we believe that serology has limited use in the diagnosis and postoperative follow-up of intracerebral cysts. However, serological tests and especially the newer enzyme-linked immunosorbent assays maybe more useful in diagnosing hydatid disease involving multiple organs [4].

The treatment of hydatid cyst is exclusively surgical. It is paramount to remove the cyst without rupture in order to avoid complications such as spread of parasites, recurrence, and anaphylactic reaction [15]. The most common surgical approach is that described by Arana-Iniguez and San Julian and consists of forced expulsion of the cyst by hydrodissection introducing hypertonic saline solution under and around the cyst [5].

Intraoperative cyst rupture is a possible event associated with a potentially fatal anaphylactic reaction and a high rate of cyst recurrence [26] [27]. On the other hand, seizures, subdural effusion, porencephalic cyst, hemorrhage, pneumocephalus, hydrocephalus, and focal neurological deficits are the main reported postoperative complications that sometimes need further intervention [28]. Medical therapy is also important. Albendazole is currently the elective medication; it is effective in sterilizing the cyst, decreasing the risk of anaphylaxis, and reducing the recurrence rate [29] [30]. Corticosteroids may help control perilesional edema, while anticonvulsants are used prophylactically [4].

4. Conclusion

Neurosurgeons should keep hydatidosis in the list of differentials when evaluat-

ing patients with cystic diseases of the brain. An MRI can greatly aid in diagnosis and surgical planning. The goal of surgery should be total excision of intact cyst which should be followed by antihelminthic treatment to avoid recurrence. Surgical outcomes are excellent, and patients show complete resolution of symptoms after complete excision of the cyst.

Declarations

Patient was informed and his consent was obtained before submission of this case report.

Approval of Ethic committee of Teaching hospital of Fes has been obtained.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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