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Case Report

Primary cerebral hydatid cyst with orbital extension: A case report ☆

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ABSTRACT

Hydatid cyst, which is caused by the cestode echinococcus granulosus, is endemic in many countries around the world. Despite the frequency of this disease, no previous case of cerebral hydatid cyst with orbital extension has been reported, demonstrating the scarcity of this presentation. Cerebral hydatid cyst may be primary or secondary. Given to the nonspecifity of the clinical symptoms, cross sectional imaging, including CT and MRI, play an important role in the diagnosis by ruling out differential diagnoses, which are represented essentially by neoplastic and nonneoplastic intraparenchymal cysts as well as brain abscesses. Surgery remains the cornerstone treatment for cerebral and orbital hydatid cyst. In this article, we present the imaging findings of a patient with a primary cerebral hydatid cyst with orbital extension, and also the radiological features that could eliminate other differential diagnoses.

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Introduction

Cerebral hydatid cyst with orbital extension is very rare, and no previous case have been found on bibliographic databases nowadays. Hydatid cyst, which is caused by the cestode echinococcus granulosus, is endemic in many regions worldwide, especially around the Mediterranean basin [1,2]. However, given to the frequency of international travels, hydatic cyst should come to our mind when a brain cystic lesion is encountered. Given to the nonspecificity of clinical symptoms, the diagnosis can be challenging. However, clinical history along with cross sectional imaging can help to narrow down the differential diagnosis list [3]. Concerning the treatment, surgical removal of the cyst is the procedure of choice [3,4]. This case report presents a patient who have been diagnosed with parenchymal hydatid cyst of the brain extending to the orbit.

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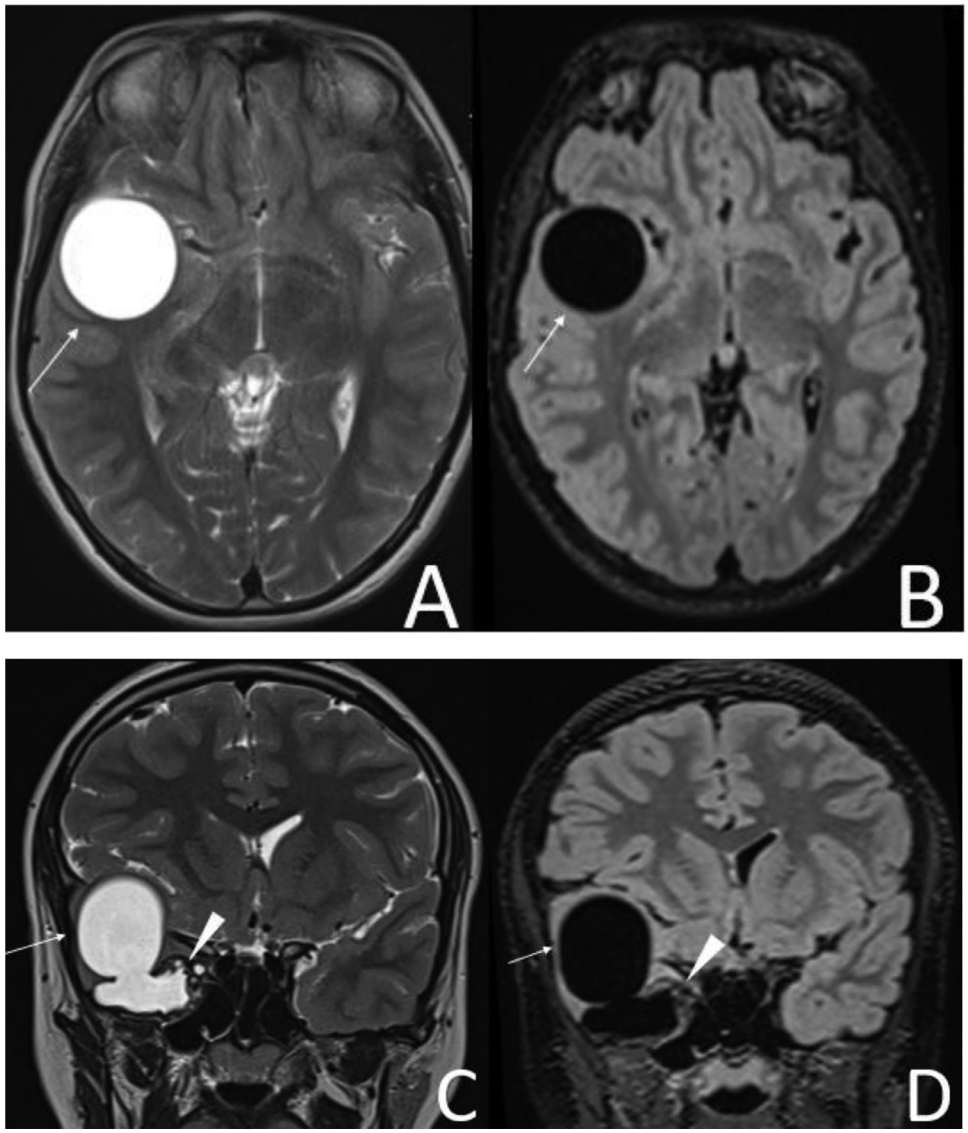

Fig. 1 – MRI T2 weighted and Flair images on axial (A and B) and coronal view (C and D) showing a well-defined cyst in the temporal lobe (white arrow). Images with coronal view (C and D) demonstrate also a multivesicular cyst (white head arrow).

Case report

A 16year Moroccan man, living in rural area, presented to our hospital for exophthalmia, ptosis and chemosis of the right eye. No other pathologies have been observed in the neurologic and systemic examinations. On ophthalmologic examination, there was a limitation in ocular motility. Routine blood tests were unremarkable. The patient underwent cerebral and orbital CT scan and MRI, which showed a cystic mass lesion developed at the temporal lobe of the brain parenchyma with well-defined borders (Fig. 1A and B). No evidence of peripheral edema, enhancement or diffusion restriction have been noted. This cyst extends to the extra axial space in the temporal fossa, and to the orbit across the superior orbital fissure which is enlarged. It also makes an impression on the posterior wall of the ethmoidal bone. At this level the mass presents a multivesicular aspect (Figs. 1C, D and 2). Then, it extends to the intra orbital intraconal space, pushing the eyeball forward. The optic nerve is also pushed upward. This mass extends also across the inferior orbital fissure to the inferior temporal fossa, and makes an impression on the posterior wall of the maxillary bone. Superiorly and medially, it presents a contact with the internal carotid and the right cavernous sinus which is repressed. The mass also exerts a pressure on the inferior and lateral rectus muscles, resulting in their displacement. The dimension of the cyst was $57 \times 40 \times 51$ mm.

Based on the lifestyle of the patient (living in a rural area) and on the imaging features, a hydatid cyst has been suspected. Therefore, additional imaging has been realized, including CT of abdomen and thorax, which didn't show any cystic lesion in the liver and the thoracic parenchyma. Few weeks later, a serologic test ELISA (enzyme linked immunosor-

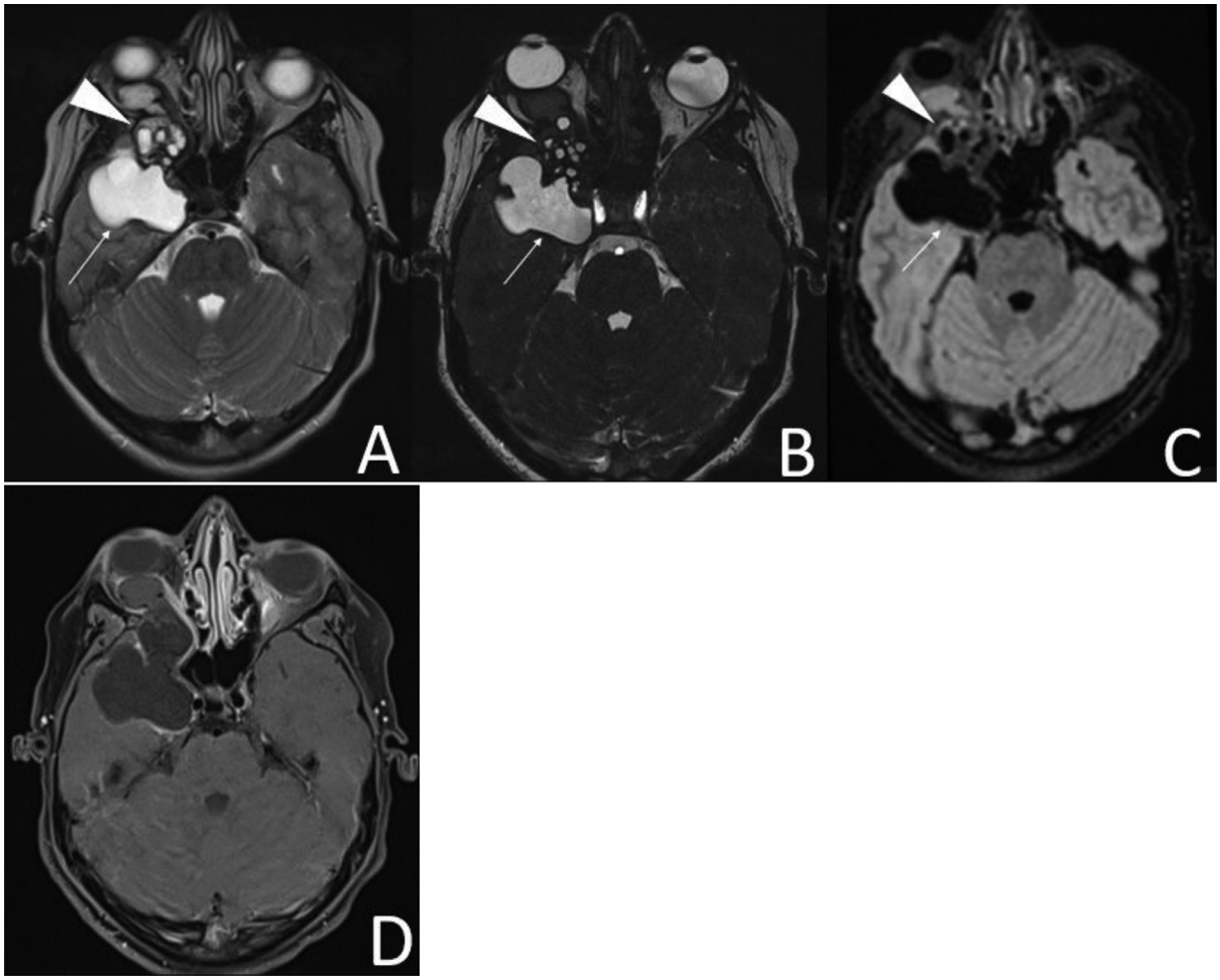

Fig. 2 – MRI images: (A-C) axial T2 weighted image and axial CISS sequence show a high signal intensity whereas axial FLAIR shows a low signal intensity related to an intraparenchymal cyst located in the temporal lobe (white arrow), extending forward to the orbit where it presents as a multivesicular cyst (white head arrow). (D) Axial T1 weighted image show no enhancement after contrast administration.

bent assay) for the diagnosis of hydatid cyst has been performed, and was positive. Given to the noncompliance and the refusal of the patient's family for surgery, the preoperative CT scan was only performed a few months later, showing an increase in cerebral hydatid size, with a density similar to that of cerebrospinal fluid. CT scan also showed lysis of the sphenoidal bone, adjacent to the mass (Fig. 3). Given to these findings, a surgery has been performed. The histopathology of the pearly white cystic formation removed during surgery revealed hydatid proligerous membranes with scoleces, which was consistent with a hydatid cyst. The symptoms of the patient improved and the neurological examination was normal after surgery.

Discussion

Hydatid cyst, caused by ingestion of echinococcus granulosus, represent a common parasitic infection. Human are involved during the larval stage of the disease, and represent an intermediate host. On the other hand, dogs and wild animals, like foxes, are the final host of the adult worm [4]. Contaminated vegetables and fruits contain ova than can enter human intestine. Afterwards, larvae can pass the intestine and spread via the portal and systemic circulation to organs [4]. The liver (75%) and the lung (15%) represent the main sites involved [3]. Cerebral hydatid cyst with orbital extension is very rare, and no case have been reported in the literature nowadays. Cerebral hydatid cyst is an uncommon site of infection, even in regions in which the infection is endemic [5], and may be seen in 2% of cases only [3]. On the other hand, 1%-2% of cases are represented by orbital hydatid cyst [4]. Therefore, the association of both is even rarer. Almost all orbital hydatid cyst is uniloculated. Thus, multiloculated are even rarer [6]. Cerebral hydatid cyst may be primary or secondary. Constitution of primary cerebral hydatid cyst is explained by the direct infestation of the larvae in the cerebral parenchyma, and don't involve other organs. In some contemporary series, systemic hydatid disease has been reported in only 18% of cerebral hydatid cyst [3]. Primary cerebral cyst is generally solitary and unilocular, with a thin wall [7].

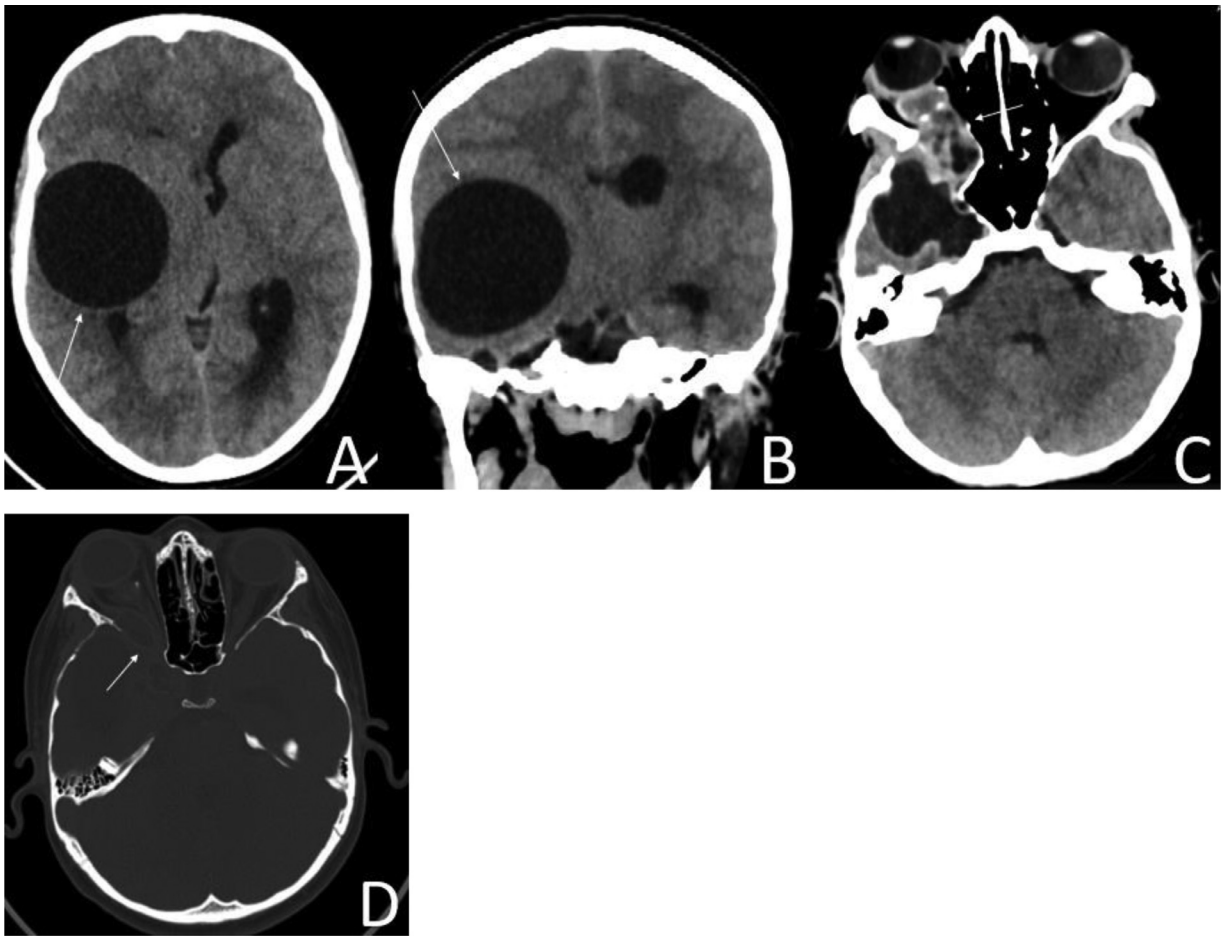

Fig. 3 – CT scan images with soft tissue window demonstrating an increase in size of the cyst with subfalcine and uncal herniation (A and B), and also a multivesicular cyst in the orbit (C). The bone window also shows sphenoidal bone lysis (D).

Commonly, symptoms associated with the cerebral hydatid cyst include classical symptoms of increased intracranial pressure (headache, nausea, vomiting, and papilledema) or focal neurologic deficit related to local compressive effects [3,7]. The main symptoms of orbital cyst are due to compression of adjacent anatomical structures [4]. Commonly, exophthalmia represents the reason for consultation [8], like in our case. When the diagnosis is delayed, reduction of visual acuity along with ophthalmoplegia, papillary hyperemia and optic atrophy may be seen [8,9].

Cross sectional imaging of cerebral hydatid cyst may show well defined borders with an internal content identical to cerebrospinal fluid. The lesion presents a thin wall with no pronounced enhancement, and in most of cases, don't show perilesional edema [10]. They are principally encountered in the cerebral hemisphere within the territory of the middle cerebral artery, as shown in our case [3]. On the other part, imaging of orbital hydatid cyst present similar features [4,5,9]. Frequently, the lesion is localized behind the globe, resulting in proptosis. It also occurs mostly in the intraconal space [4]. The polycystic form of orbital hydatid cyst seems as intimately packed multiple cyst. Only 5% of orbital hydatid cysts may show this appearance [9].

Treatment can be either medical or surgical, and many options can be used. However, surgery remain the cornerstone treatment for both, cerebral and orbital hydatid cyst [7,8]. Removal of the cyst represent the curative treatment. Intact extraction rate of an intracranial hydatid cyst accounts for 60%-70% of cases [3]. Preoperative medical treatment can be effective, resulting in sterilization of the cyst and in tension reduction [7]. It should be treated at an early stage to withdraw any functional impairment. The accuracy of the diagnosis is essential and define the prognosis. A misdiagnosis can generate several consequences like rupture of the cyst which can lead to spread of infection and subsequent anaphylactic reaction [5].

Differential diagnosis

Primary cerebral hydatid cyst with orbital extension should be distinguished from other cerebral cystic masses, whether neoplastic or not.

Supra tentorial non-neoplastic intra parenchymal cyst

Porencephalic cyst and arachnoid cyst should be differentiated from hydatid cyst. Contrary to hydatid cyst, porencephalic cyst and arachnoid cyst aren't completely enveloped by brain parenchyma and don't present a circular shape. Arachnoid cyst is extra axial and the adjacent

brain parenchyma may be deformed. Porencephalic cyst follow brain injuries, and are lined by gliotic tissue, that could easily be identified on MRI [11]. Neuroglial cyst represent also differential diagnosis and it may be challenging to differentiated them. However, they are often unilocular, and in our case, the intraconal part of our mass is multilocular.

Supra tentorial neoplastic intraparenchymal cyst

Pilocytic astrocytoma are astrocytic glioma with well-defined borders. These tumors can take several radiologic appearances, but in most cases, they present as a large cystic mass with vivid enhancement of the mural nodule. In our case, no mural nodule has been described. The other tumors, including glioma and metastases, often present with surrounding edema and infiltration of adjacent tissues [12].

Cerebral abscess

Pyogenic abscess can present as a cyst-like lesion with central necrotic area. However, high signal on DWI sequence is commonly present centrally, representing true restricted diffusion. Perilesional edema is also often seen in addition to an intense rim enhancement following contrast injection. The satellite lesions are also common [11–13]. Clinical and laboratory investigations can also help to differentiate them.

Conclusion

Hydatid cyst can occur as primary lesion in the brain, and extend to the orbital space. CT and MRI in combination are useful in the diagnosis of cerebral and orbital hydatid cyst. When we have a multilocular cyst involving both, brain parenchyma and orbit, the diagnosis of hydatid cyst, even if it's uncommon, should come to our mind, especially when the patient live in an endemic country. The absence of perilesional edema and vivid enhancement could help to exclude a lot of differential diagnoses.

Patient consent

Informed consent for publication was obtained from patient.

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