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Late Recurrence of Brain and Orbital Hydatid Cyst: A Case Report and Literature Review

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Abstract

Background: Hydatidosis is a zoonotic disease endemic in some parts of the world including Morocco. The cyst is commonly encountered in the liver, lungs, heart muscle and central nervous system. **Case Description:** A 16-year-old-male with a history of intra orbital hydatid cyst presented with a progressive worsening of the vision on the right eye as well as proptosis and headache. Indeed, he had been operated 13 years ago for a right intra-orbital cyst and received Albendazole for an unknown duration. The MRI showed an intra-axial cystic mass extending from the right temporal lobe to the eyeball through the superior orbital fissure, suggesting a hydatid cyst. The patient benefited from total excision of the cyst despite a perioperative rupture. **Conclusion:** Hydatid cyst can recur even many years after the first episode. Relapse is common, especially if a rupture occurred during previous surgery. Medical treatment with a long follow-up is recommended.

Keywords

Hydatid Cyst, Recurrence, Brain and Orbital Cyst

1. Introduction

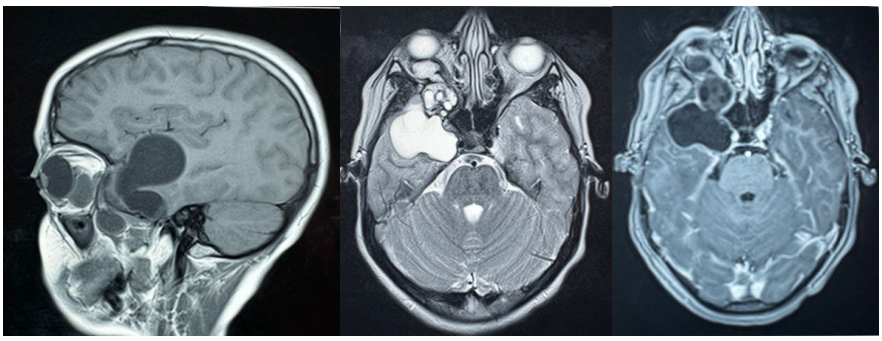
Hydatidosis is a zoonotic disease caused by a tape worm. This tape worm is found in canids, which are their definitive host but also in sheep, cattle, camel horses and goats as their intermediate hosts. Humans are affected only as accidental host. Hydatidosis is common in sheep-rearing countries where there is a tight and constant dog-sheep contact [1]. Certain human activities, like feeding dogs with the viscera of home-butchered sheep facilitates the worm cycle and its transmission [2]. It is an endemic disease in some part of the world, including Middle East countries, China, South America and Africa. Despite some progress in the control

of this zoonosis, it constitutes an emerging and re-emerging disease. In USA it is diagnosed in immigrants of countries where the disease is highly endemic [2]. In Morocco latest epidemiologic data reports an incidence of 5.2/100,000 inhabitants [1]. Hydatid cyst involves liver and lungs, but other localizations have been reported, namely heart muscle, vertebrae and central nervous system. Central nervous system involvement represents 2% of patients [3] [4]. Primary brain hydatid cyst is rare, as well as the intra orbital localization, moreover, the association of the two lesions is rarer. Management of intra-cranial lesions is the cautious surgical excision to avoid cyst's premature rupture and medical treatment is mandatory. Here we present the case of a 16-year-old male with recurrent hydatid disease involving cerebral and intra orbital localization. We, therefore emphasize the importance of long-term medical follow-up to reduce the risk of recurrence.

2. Case Study

A 16-year-old male living in the countryside in central Morocco was seen by an ophthalmologist for proptosis. The patient had been operated when he was three years old for intra orbital hydatid cyst then was lost to follow-up. He was put on Albendazole after his surgery for an unknown duration and neither the MRI nor the surgical report are available. The initial treatment of this patient is unclear. He consulted few months ago as he presented a progressive worsening of the vision on the right eye with proptosis and headaches. He denied any vomiting, seizures or speech difficulties. His family reports he used to play with stray dogs.

Upon physical examination, he was well-oriented, no signs of sensory or motor deficit. He had normal extra ocular movements. The proptosis was obvious on the right eye, there was no bruit on auscultation. The right eye was not inflamed, and it was not deviated. His visual acuity was 4/10 on the right eye and 10/10 on the left eye. The fundoscopy revealed a grade 2 papilledema on the right eye the left being normal. All the lab tests were normal with thyroid hormones in the normal range.



(A) (B) (C)

Figure 1. Cerebral MRI A/T1 hypo intense lesion B/T2 Hyper intense lesion C/T1 contrast enhancement of cysts wall.

The patient had a cerebral and orbit magnetic resonance image (MRI) which

showed a well defined lobulated cystic lesion measuring about 65 × 46 × 30 mm at the right temporal lobe extending through the orbital fissure with multiple small cysts. It appeared hypo intense on T1, hyper intense in T2 and had a thin wall that enhanced with contrast. There was a mild effect on the brain, distorted right optic nerve, no edema, no hydrocephalus (**Figures 1(A)-(C)**). The CT scan showed erosion of medial wall of sphenoid's lesser wing as well as optic canal (**Figure 2**). We did not perform any other radiographic assessment to rule out any other sites of hytadid cyst localization because the patient had no other complaints.

Figure 2. CT scan erosion of medial wall of sphenoid's lesser wing and optic canal.

An extended cranio-orbito-zygomatic approach was used to reach the temporal and intra orbital lesion. We opened the lateral wall of the orbit as well as the temporal fossa. After opening the dura, the temporal lobe appeared flat, relaxed. We performed a corticectomy of 50 mm of the temporal lobe corresponding to three-fourth of the larger diameter of the cyst. We gradually removed the gliotic parenchyma above the cyst using bipolar forceps. Cyst delivery was performed using the Dowling's technique of hydro dissection starting from the temporal lesion. We followed the wall of the cyst to reach the intra orbital portion. As we progressed the space became increasingly narrow, and it was hard to have control on the distal portion of the cyst. The cystic wall which has become irregular ruptured at the end of the procedure allowing the content to spill out. We immediately flushed the surgical site with 3% hydrogen peroxide, followed by isotonic saline and proceeded with cystic wall excision. Upon histological examination it was a hydatid cyst.

The post operative course was unremarkable, and the patient was discharged on post operative day 5 with a prescription of albendazole for 6 months. The patient was seen month later as an outpatient, his proptosis had completely regressed, and he was doing fine. Unfortunately, he did not show up for the following checkup.

3. Discussion

Hydatid disease is caused by Echinococcus granulosus. It is a tape worm which

has been described since 1901 [3]. It is endemic in some regions of the world including Morocco. The last report goes back to 2008 where the incidence was found to be 5.2/100,000 inhabitants [1]. The hydatid cyst is most common in rural places especially among children and young adults [1] [3]. Patients get infected through water and food contaminated by dog feces which contain Taenia echinococcus rings.

Hydatid cysts are more common in the liver and lungs and are encountered in only 2% - 3% in the central nervous system [5]. Common localizations includes frontal, temporal and occipital lobes [3]. However orbital hydatic cyst is rarer even in endemic area [6]. Infratemporal lesions are usually associated with swelling in maxillofacial region, but proptosis is rarely described [7]. Our case associated an orbital and infratemporal localization with proptosis and no maxillofacial swelling.

Symptoms are mainly related to the raised intra cranial pressure however some cysts can reach a very large size before having any clinical symptoms. The cyst growth rate in brain is about 1 cm/year but might be faster in children [5] [8]. Seizures are a less common presentation with reports ranging from 8% - 22% [8].

The mainstay for management of cerebral hydatid cyst is surgical excision. The goal is to remove the cyst intact without spillage of the content. Many techniques have been described such as puncture-aspiration-injection-re aspiration but are mainly used for hepatic cysts. Dowling's technique is mostly used for central nervous system localization. Traditional approach is the Dowling's technique with hydro dissection which allows safe dissection without further damage to surrounding parenchyma. We performed an extended cranio-orbito-zygomatic approach as we had to reach both the temporal and the orbital lesion. The Dowling's technique reduces the risk of cystic wall rupture, however when cysts extend through a narrow space as in the orbital fissure premature rupture is hard to avoid. Nonetheless, no studies have linked cyst location with the risk of rupture [9]. It might be possible that due to the fact it was a second surgery, and we did not have any report of the previous surgery, there might be more tissue adherence increasing premature rupture of the cyst. It is common admitted that cysts wall as well as scolices contained in the cyst must be removed entirely to prevent any recrudescence. In the event of inadvertent rupture, the surgical site can be irrigated with hypertonic saline or ringer lactate to prevent the dissemination of cyst content [9]. We flushed surgical site after the cyst rupture with 3% hydrogen peroxide because it is an effective scolicidal solution used when performing the puncture-aspiration-injection-re aspiration technique. Even though it is not described in the literature, we assumed that small amount of this solution is not going to be harmful compared to the risk of a recurrence. Fertile scolices are contained in the cyst and may contaminate the adjacent brain leading to recurrence [9] [10]. Moreover, premature rupture can cause an anaphylactic shock with dramatic consequences. Mortality is increased among patients with recurrence, half of them will die within 2 years mainly by systemic hydatidosis [10]. Signs of recurrence usually appear in

4 - 12 months after the first surgery. Recurrent cysts are more fragile and easily break up during further surgeries creating a vicious circle of recurrence and spillage of scolices.

Medical treatment is mandatory mostly after cysts rupture. Moreover it is commonly used when there is multiple organ involvement, multiple central nervous system lesions, recurrence, pre-operative volume reduction and post operative [5]. Albendazole is the best therapeutic option compared to other medications such as Mebendazole. The efficacy of treatment depends on size, age, calcification and fibrosis of the cysts, thereby young cysts are more accessible to the drugs. Albendazole's average dosage is 10 mg/Kg/day with monitoring of blood cell counts and liver enzymes. The treatment is 3 - 6 months and the follow up must be long enough to detect any relapse.

Hydatid disease remains a public health issue mainly in countries where it is endemic. Morocco is the third most affected country in North Africa with rural population more concerned and less aware of the disease [11]. The average cost for surgical management is estimated at 1350 € [1] added to this cost is reduced quality of life, morbidity and loss of income.

4. Conclusion

Hydatid disease is a zoonosis endemic in many countries around the world and remain a public health issue with a huge cost for health system. More frequently, in rural areas, humans are only accidental hosts. The rate is higher in sheep rearing areas with human contamination favored by some human practices. Central nervous system as well as orbital localization are rare, management is challenging, and rupture risk elevated. Dowling's technique is the most common approach with reduced risk of rupture. However, operate on a recurrence could increase the risk of per operative rupture. Cyst's rupture increases risk of recurrence and long-term treatment must include chemotherapy with Albendazole. Practicians should emphasize on patient education and importance of long-term follow-up to avoid recurrence. Governments in endemic areas should establish policy to regulate sheep farming practices to help people avoid risky behavior.

Declaration of Patient Consent

The author certify that they have obtained all appropriate patient's guardian consent form. They have given their consent for the images and clinical information to be reported in the journal. Moreover, we gave them the guaranty that neither the name nor the initials will be published to preserve the identity of the patient.

Conflicts of Interest

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

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