

A Rare Case of Orbital Hydatid Cyst

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Abstract: *Background:* Cystic echinococcosis, also known as hydatid disease, which is mostly caused by larval stage of *Echinococcus granulosus*. The orbital localization is rare and uncommon. Preoperative diagnosis is important to prevent from complications. *Case information:* Here we report the case of a child admitted for chronic proptosis whose investigations concluded with an orbital hydatid cyst. *Result:* We describe the case of an 8-year-old child, who consulted in our ophthalmic emergency department for a painful proptosis of the right eye with a progressive decrease in visual acuity and chronic fatigue that had been evolving for 6 months. Due to this highly evocative lesion of an orbital hydatid cyst, we carried out with an ELISA and Western Blot hydatid serology which ended negative. Faced with this orbital involvement, we performed an orbital MRI which objectified two right T2-hypersignal eye cystic lesions, well-limited, exerting a mass effect on the optic nerve and responsible of a grade 2 exophthalmos. We also performed a biological assessment including hemogram, renal and hepatic function that had been normal. The patient remained under medical treatment with albendazole before and after surgery. *Conclusion:* Orbital involvement, although rare, should not be ignored especially when it comes to chronic proptosis with or without a visual impact in children living around dogs or breeders or in deplorable hygienic conditions.

Keywords: Echinococcus granulosus, Hydatid Cyst, Proptosis, Surgery

1. Introduction

Hydatid cyst is the larval stage of a dog tapeworm named *Echinococcus granulosus*. It has a worldwide distribution mostly in developing countries. The cysts are commonly found in liver and lungs but other localizations must not be excluded [1, 2]. The orbital localization is rare and uncommon. Preoperative diagnosis is important to prevent from complications.

We present a rare case of an orbital hydatid cyst treated by surgery.

Purpose: To report a rare case of orbital alveolar echinococcosis in Morocco.

2. Case Report

We describe the case of an 8-year-old child, who consulted in our ophthalmic emergency department for a painful proptosis of the right eye with a progressive decrease in visual acuity and chronic fatigue that had been evolving for 6 months.

His best visual acuity was limited to 6/10 in the right eye. Slit lamp examination showed an irreducible proptosis without any thrill associated to a lower inflammatory chemosis. Lateral movements of the eye were restricted (Figure 1).



Figure 1. Irreducible exophthalmos with a lower inflammatory chemosis.

The examination of the anterior segment showed no abnormalities, the ocular fundus revealed a stage 2 papilloedema with a vascular tortuosity.

The left eye examination was normal with visual acuity preserved at 10/10.

The rest of the physical examination was strictly normal.

Faced with this orbital involvement, we performed an orbital MRI which objectified two right T2-hypersignal eye cystic lesions, well-limited, exerting a mass effect on the optic nerve and responsible of a grade 2 exophthalmos (figure 2).

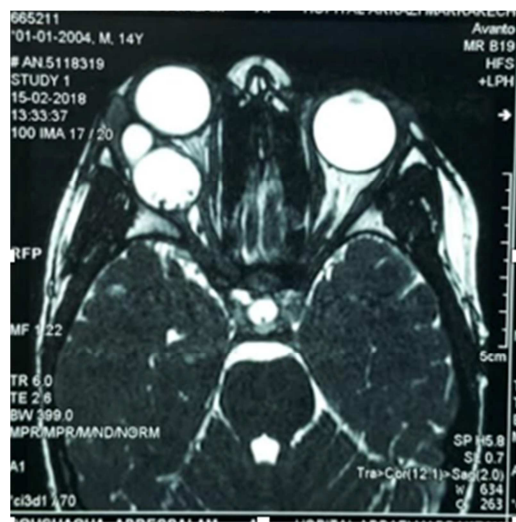


Figure 2. Eye cystic lesions, well-limited, exerting a mass effect on the optic nerve and responsible of a grade 2 exophthalmos.

Due to this highly evocative lesion of an orbital hydatid cyst, we carried out with an ELISA and Western Blot hydatid serology which ended negative. We also performed a biological assessment including hemogram, renal and hepatic function that had been normal.

As part of the extension assessment and given the rare location of this hydatid cyst, the patient had a chest X-ray and abdominal ultrasound but showed no other localization.

The child underwent surgery performing an inferolateral orbital margin incision approach. Histopathology confirmed

our primary diagnosis.

The patient remained under medical treatment with albendazole before and after surgery.

He was seen again in consultation after 1, 2 and 3 months of treatment, his visual acuity went up after correction to 8/10, the proptosis disappeared and the inflammatory signs dried up.

3. Discussion

Echinococcosis is an infection that is caused by the genus, is distributed mainly in temperate regions, such as some parts of Europe and Asia (especially Mediterranean area, Middle East, central Asia and China). Australia, several parts of America, and north and east Africa are also endemic for the tapeworm [3, 4].

Echinococcus granulosus is a cestode whose life cycle involves dogs and other canids, as definitive hosts for the intestinal tapeworm, as well as domestic and wild ungulates as intermediate hosts for the tissue-invading metacestode (larval) stage. The metacestode (echinococcal cyst) is a fluid-filled, spherical, unilocular cyst that consists of an inner germinal layer of cells supported by a characteristic acidophilic-staining, acellular, laminated membrane of variable thickness [5]. Each cyst is surrounded by a host-produced layer of granulomatous adventitial reaction. Small vesicles called brood capsules bud internally from the germinal layer and produce multiple protoscolices by asexual division. In humans, the slowly growing hydatid cysts can attain a volume of several liters and contain many thousands of protoscolices. With time, internal septations and daughter cysts can form, disrupting the unilocular pattern typical of the young echinococcal cysts.

Orbital hydatidosis is a rare zoonosis caused by the larval stage of *Echinococcus granulosus*. There are 4 important species of this genus which are of concern for public health: 1-Cystic echinococcosis, the most common type, is caused by *Echinococcus granulosus*. 2-Alveolar echinococcosis is caused by *E. multilocularis*.

Rarely, *Echinococcus vogeli* (3) and *Echinococcus oligarthrus* (4) cause polycystic echinococcosis [5, 6]. It is primary echinococcosis, and is generally unilateral and left [7]. Patients are mostly children. It is uncommon to find other simultaneous hydatid cysts anywhere in the body [8]. Unilateral proptosis is the most common clinical finding in intraorbital hydatid cyst cases [9].

The most frequent clinical findings are exophthalmos, chemosis, lid oedema, visual impairment, and restriction of extraocular motility [10].

On CT, the orbital hydatid cyst is typically seen as a unilateral, non-enhancing homogeneous cyst with low density, similar to visualisation of the vitreous body. On MR imaging, the cyst shows isointense signalling on T1- and T2-weighted images. In both CT and MR imaging, peripheral rim contrast is seen after the injection of a contrast medium [11]. However, it is difficult to distinguish hydatid cysts from other mass lesions of the orbit via radiological imaging

methods. Therefore, other cystic mass lesions like abscesses, mucocele, intraorbital haematomas, lacrimal tumours or cysts, and lymphangiomas must be considered in the differential diagnosis [12, 13]. Although serological tests are used for diagnosis of hydatid cyst, serological tests are usually negative in cases of orbital involvement. Therefore, radiological imaging methods are valuable in the diagnosis of orbital hydatid cysts [14].

Treatment of orbital hydatidosis is exclusively surgical. It must be as early as possible to avoid any functional damage. The incision must consider the location and size of the cyst. The most important complication in surgical treatment is rupture of the cyst during excision, which can cause a relapse. However, complete extirpation of the cyst without rupture is almost impossible. Recurrences are exceptional even in case of release of the cyst content because of the absence of a peritoneum in the orbit. Albendazole treatment is useful, especially if begun 14–28 days before surgery and is used as an adjunctive therapy to surgery [15]. After surgical excision, the diagnosis of orbital alveolar echinococcosis was confirmed by pathologic evaluation. Physicians should maintain a high index of suspicion for hydatidosis when approaching a patient with proptosis due to several important standpoints especially in endemic area: rupture of hydatid cyst during operation may

cause anaphylactic shock, dissemination, and implantation of scolices. These complications can be more serious in orbit in comparison with other parts of the body. Therefore, preoperative diagnosis and complete excision of the intact cyst are important. Because of limited space in the orbital cavity, mass effect of slowly growing cyst may include the optic nerve involvement and impaired vision, making early diagnosis and removal of the cyst of utmost importance in this aspect [16, 17].

4. Conclusion

The diagnosis of orbital hydatid cyst should not be misunderstood in case of unilateral exophthalmos. The main treatment remains the prevention based on the careful hand washing and extensive washing of fruit and vegetables especially in developing countries.

Conflicts of Interest

The authors declare having no conflicts of interest

References

- [1] Vuitton DA, Brunetti E. Cystic and alveolar echinococcosis: fraternal twins both in search of optimal treatment. In: Sing A, ed. *Zoonoses e Infections Affecting Humans and Animals*. Berlin: Springer Netherlands; 2015: 715e747.
- [2] Grosso G, Gruttadauria S, Biondi A, Marventano S, Mistretta A. Worldwide epidemiology of liver hydatidosis including the Mediterranean area. *World J Gastroenterol*. 2012; 18 (13): 1425e1437.
- [3] Otero-Abad B, Torgerson PR. A systematic review of the epidemiology of echinococcosis in domestic and wild animals. *PLoS Negl Trop Dis*. 2013; 7 (6): e2249.
- [4] Shoaee S, Rezvanizadeh M, Haghighi M, Yousefi H. Epidemiological, clinical and paraclinical study of hydatid cysts in three educational medical centers in 10 years. *Nov Biomed*. 2016; 4 (1): 28e33.
- [5] Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev*. 2004; 17 (1): 107e135.
- [6] Eckert J, Gemmell MA, Meslin FX, Pawlowski ZS. WHO/OIE Manual on Echinococcosis in Humans and Animals: A Public Health Problem of Global Concern. Paris, France: WHO/OIE; 2001: 20e71.
- [7] Basset D, D'hermis F, Leport et coll. Localisation orbitaire d'un kystehydatique chez un enfant surinamien. *Bull Soc Fr Parasit* 1989; 7: 241-4.
- [8] Moro P, Schantz PM. Echinococcosis: a review. *Int J Infect Dis*. 2009; 13 (2): 125e133.
- [9] Benazzou, S., Arkha, Y., Derraz, S., El Ouahabi, A., & El Khamlichi, A. (2010). Orbital hydatid cyst: review of 10 cases. *Journal of Cranio-Maxillofacial Surgery*, 38 (4), 274-278.
- [10] Öztekin, P. S., Yilmaz, B. K., Gokharman, F. D., & Koşar, P. N. (2014). Primary orbital hydatid cyst: computed tomography and magnetic resonance imaging findings. *Singapore medical journal*, 55 (11), e184.
- [11] Gomez Morales, A., Croxatto, J. O., Crovetto, L., & Ebner, R. (1988). Hydatid cysts of the orbit: a review of 35 cases. *Ophthalmology*, 95 (8), 1027-1032.
- [12] Turgut, A. T., Turgut, M., & Koşar, U. (2004). Hydatidosis of the orbit in Turkey: results from review of the literature 1963–2001. *International ophthalmology*, 25 (4), 193-200.
- [13] Torun, F., Tuna, H., Bozkurt, M., & Deda, H. (2004). Orbital Hydatid Cyst: Case Report. *Türk Nöroşirürji Dergisi*, 14, 184-7.
- [14] Gökçek, C., Gökçek, A., Bayar, M. A., Tanrikulu, S., & Buharali, Z. (1997). Orbital hydatid cyst: CT and MRI. *Neuroradiology*, 39 (7), 512-515.
- [15] Berradi, S., Hafidi, Z., Lezrek, O., Lezrek, M., & Daoudi, R. (2014). Orbital hydatid cyst. *QJM*, 108 (4), 343–344.
- [16] Turgut AT, Turgut M, Kosar U. Hydatidosis of the orbit in Turkey: results from review of the literature 1963-2001. *Int Ophthalmol*. 2004; 25 (4): 193e200.
- [17] Betharia SM, Pushker N, Sharma V, Avinash M, Kashyap S. Disseminated hydatid disease involving orbit, spleen, lung and liver. *Int J Ophthalmol*. 2002; 216 (4): 300e304.