

Primary Intra-Orbital Hydatid Cyst as A Cause of Unilateral Exophthalmos: A Case Report

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ABSTRACT

Introduction: Intra-orbital hydatid cyst is a very rare pathological entity that affects children and the young adults. Hydatid cysts rarely appear isolated in the orbital cavity without involvement of other organs. Most of these are situated in the supéro-lateral and supéro-medial angles of the orbit. Inferiorly located cysts are very uncommon.

Case report: We report a case of 5 years male patient who presented with left eye exophthalmos, and was subsequently diagnosed with left intra-orbital hydatid cyst, treated surgically using a combined endoscopic and external approach.

Discussion: Intra-orbital hydatid cysts are a very rare occurrence. Clinical presentation of intra-orbital hydatid cyst is dominated by proptosis and a decrease in visual acuity. Complete surgical excision is difficult, and evolution is generally better when the treatment is early before the installation of irreversible optic atrophy.

Conclusion: Although very uncommon, the intra-orbital hydatid cyst must be evoked in endemic countries. Clinical and imaging characteristics should be used to further confirm the suspected diagnosis.

Keywords: Orbital cyst; Hydatid cyst; Intra-orbital hydatid cyst; Hydatidosis

Introduction

Orbital hydatid cysts are a rare localization of the *Echinococcus granulosus* parasite, whose hosts are sheep and dogs. Morocco is an endemic country where hydatidosis is still rife^{1,2}. In 1 to 2% of cases, the parasite is localized in the orbit^{1,3}. The main clinical sign of orbital cysts is exophthalmos. The contribution of imaging, ultrasound and especially CT imaging, is essential for pre-operative diagnosis. Furthermore, serology is insufficient and treatment is essentially based on surgery

Case Report

We report the case of a 5 years old male patient without a medical history, who presented to our department for the

appearance of isolated exophthalmos of the left eye over a three-month period, with no other associated signs, in particular no decrease of visual acuity, and no sino-nasal symptoms (**Figure 1**).



Figure 1. Preoperative image of the patient showing left exophthalmos

Clinical examination found a prominent left eye exophthalmos. Nasal endoscopy discovered no abnormalities, and the ophthalmological examination showed stage 2 papillary oedema in the left eye.

The craniofacial CT-scan (**Figure 2**) showed grade 3 exophthalmos with a thickening and infiltration of the left palpebral soft tissues, with evidence of a medial extra conical collection of mild density, measuring 31 x 22 mm and extending over 21 mm, with a mass effect on the superior rectus muscle, which was tumefied and compressed. It also had a mass effect on the ipsilateral optic nerve, which was discreetly swollen

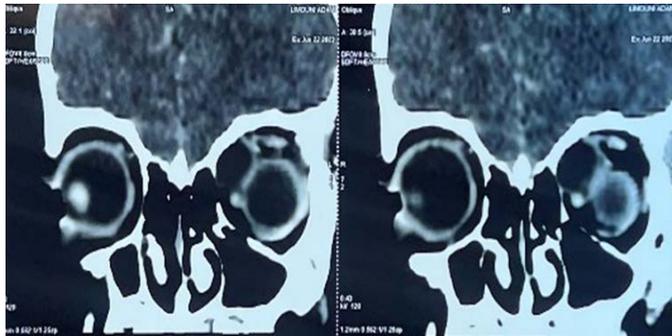


Figure 2. Coronal ct scan images showing the hypodense mass and its effect on the globe

The orbital MRI showed an oval, well-limited left intra-orbital process, with regular contours and extra-conical development, in hypo-signal in T1-weighted images, and in homogenous hyper-signal in T2 weighted images, not enhanced after gadolinium injection, and measuring 31 x 17 mm extended over 17mm, evoking a neuro-fibroma or a schwannoma (**Figure 3**).

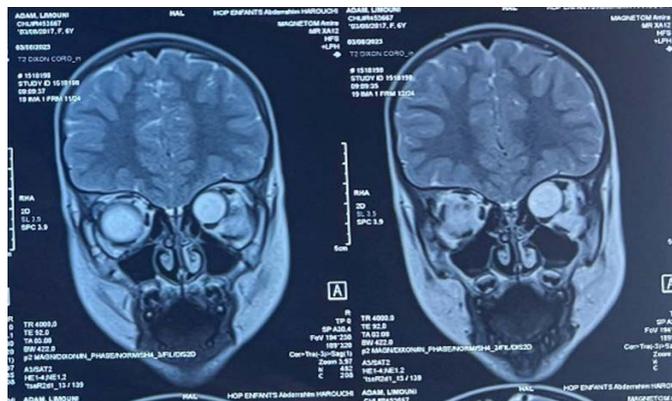


Figure 3. t2-weighted coronal images of the orbital mri scan showing the hyperintense mass located in the superior and medial part of the left orbit

The case file was discussed with the ophthalmologists, and the decision was to preform surgical excision of the mass. The surgery was performed by a senior ENT surgeon, and a combined approach was used, involving both endoscopic surgery and an external approach. Through endoscopic exploration of the left nasal fossa, the lamina papyracea was opened, thus discovering the mass, which was then dissected anteriorly and medially.

An external approach was then used, by way of an eyebrow incision. The mass was discovered in the orbit, and was adherent to the superior oblique muscle and the optic nerve posteriorly. During the intervention, the mass was ruptured, revealing that it was in fact a hydatid cyst, with the exteriorization of the outer pericyst. Because of its adherence to the optic nerve and to the superior oblique muscle, the excision of the cyst in totality was

deemed too risky. We decided to hollow out the cyst as much as possible. Pathological examination of the excision tissues confirmed the diagnosis of a hydatid cyst of the orbit.

Immediate post-operative follow-up found a paresis of the superior oblique muscle, without diminution of the visual acuity. The patient underwent adapted physiotherapy, and had recuperated completely after a month. An abdominal ultrasound was performed, but didn't find any sign of hydatid cysts in the liver. Albendazole was prescribed after the surgery for a period of 3 weeks, to diminish the risk of a relapse.

The follow-up examination, at one month after surgery, found a significant reduction of the exophthalmos, with a correct function of the superior oblique muscle. The aesthetic outcome was also very good (**Figure 4**). Subsequent follow-ups over a six-month period found no clinical sign of a local recurrence.

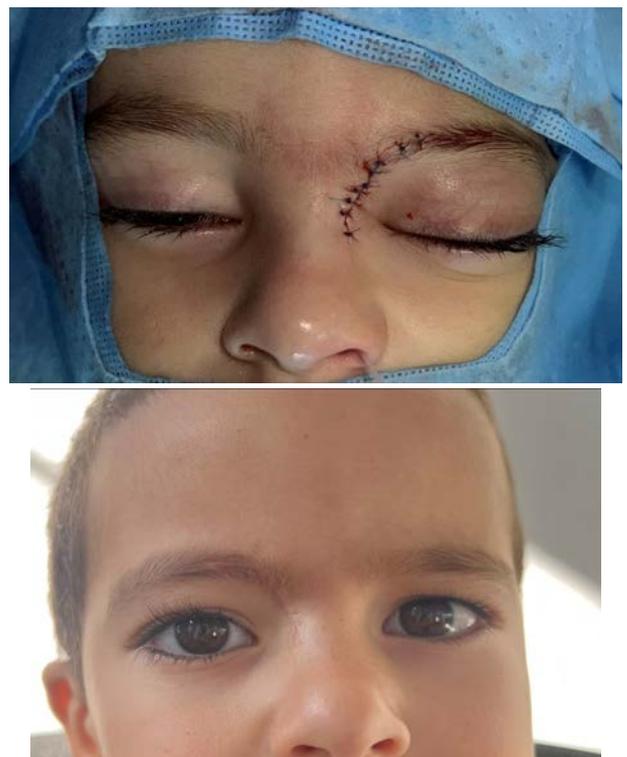


Figure 4. Image of the patient post-operatively (left) and after one month of surgery (right) showing a remarkable regression after treatment

Discussion

Hydatid cysts are most commonly located in the liver (60%-70%) and the lungs (20%)^{4,5}. The incidence of intra-orbital hydatid disease is extremely low, and accounts for 1 to 2% of all hydatid cysts⁴⁻⁸.

The symptoms include progressive exophthalmos with or without pain, disturbance in ocular motility, visual deterioration, and chemosis^{6,7,9}. Orbital involvement is usually unilateral, without right or left dominance^{4,5}. Typically, an orbital hydatid cyst is unilateral and can occur with or without other localizations of hydatid cysts^{7,9}. It is usually localized in the superior part of the orbit, either medially or laterally, and more often than not affects the motility of the ocular globe^{1,8,10-13}.

On CT, the orbital hydatid cyst is typically seen as a unilocular, well-defined, non-enhancing homogeneous cyst with low density, similar to the aspect of the vitreous body^{1,14-16}. MRI examination is especially useful to rule out other possible

cysts of the orbit. The hydatid cyst appears as a well-contoured lesion which had a low signal on T1-weighted images, and a high signal on T2-weighted signals^{1,14}. In both CT and MR imaging, peripheral rim enhancement is seen after the injection of a contrast product^{6,17}.

Surgery is the primary treatment in these cases^{5,10}. Complete excision is the treatment of choice, but in case of intra-operative rupture of the cyst, abundant irrigation with saline solution and hydrogen peroxide should be used to minimize the risk of a recurrence^{18,19}. Anthelmintic treatment is an essential component of the management of these cases, and should ideally be started 2 to 4 weeks before surgery, or as an adjuvant to surgery, to diminish the risk of recurrence^{1,8,18}.

Conclusion

Although it is exceptional, this unusual location of hydatidosis is an important entity, due to its repercussions, mainly functional. Thus, it should always be thought of in endemic countries. Preventive measures on the modes of contamination and general hygiene measures are primordial in these cases, and are the basis for the eradication of this disease.

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