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Case Report

Primary Intraorbital Hydatid Cyst: Case Reports

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A B S T R A C T

Background: Hydatid cyst is a chronic helminthic parasitic disease.It is endemic in Morocco.The orbital involvement of isolated hydatid cysts is a very rare pathological entity, affecting children and young adults living in rural areas.

Materials and Methods: These are 3 cases report of primary intraorbital hydatid cysts , collected over a 6-year period from 2019-2024 in the maxillofacial surgery department at CHU Ibn Rochd in Casablanca.

Discussion: Intra orbital hydatid cysts are very uncommun. Symptoms include progressive exophthalmos with or without pain, disturbance of monocular motility, deterioration of vision up to and including blindness, and inflammatory chemosis.Surgery is imperative, although complementary medical treatment is useful in most cases to prevent relapse.

Keywords: Hydatidosis; Hydatid cyst; Intra orbital cyst; Unilateral exophtalmos

Introduction

Hydatid cyst is a chronic parasitic disease caused by the cyst-forming tapeworm Echinococcus granulosis. It is endemic in several parts of the world, including our country (Morocco), the Middle East, India , South America, Turkey and southern Europe. Humans are accidental hosts through ingestion of viable eggs. The intra-orbital hydatid cyst is a very rare pathological entity, affecting youngsters living in rural areas. In most cases, the diagnosis is made by imaging (ultrasound, CT and MRI). Treatment is based essentially on surgery, usually combined with medical therapy. Our case series inclunding 3 cases of primary intraorbital hydatid cysts collected over a 6-year period in our department is the largest of its kind, with a wide range of clinical presentations.

Case Reports

All our patients are female and reside in a rural area.

All our patients underwent paraclinical examinations in search of another location and no abnormalities were found.

Case Report 1

A 13-year-old adolescent presented with progressive, chronic (4 years) and non-pulsatile unilateral proptosis of the right eye. On clinical examination, a 20 mm non-axial right exophthalmos was noted; palpation revealed a deep, non-pulsatile, slightly tender, firm superior-internal orbital mass with decreased visual acuity without chemosis or oculomotor disturbance.Computed tomography revealed a 26 x 42 mm isodense retrobulbar cystic mass at the level of the superior-internal angle of the right orbit, with a thick calcified wall pushing the optic nerve backwards.

Hydatidosis serology was positive.

The patient underwent exeresis of the cyst by internal paracanthal obitotomy, followed by treatment with albendazole 200 mg daily for 6 weeks. Complete irrigation of the orbit

with H2O2 hydrogen peroxide was performed following cyst rupture. Clinical evolution was favorable (disappearance of exophthalmos and return of vision).

Case Report 2

67-year-old woman with asthma and type II diabetes presented with chronic ocular pain and headaches (8 months) followed by the appearance of chemosis, exophthalmos and rapidly progressive visual deterioration. Clinical examination revealed an irreducible, non-tender, non-thrilling superolateral mass responsible for axial exophthalmos, chemosis, cleft eyelid, corneal dystrophy, ophthalmoplegia, abolished photo-motor reflex and blindness **(Figure 1)**.

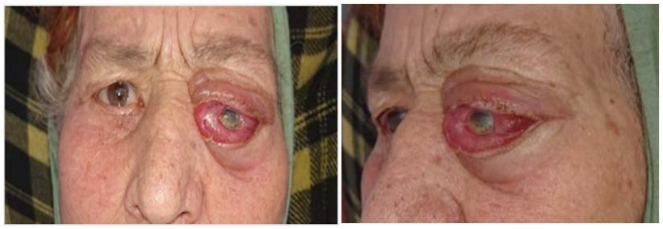

Figure 1: Photos of an elderly woman with an orbital hydatid cyst revealed by chronic exophthalmos, inflammatory chemosis and blindness.

Imaging (CT and MRI) revealed a 35 x 28 x 27 mm left extraconical cystic formation pushing back a deformed globe, the muscular cone and the lacrimal gland with grade III exophthalmos and optic nerve stretch **(Figures 2 and 3)**.

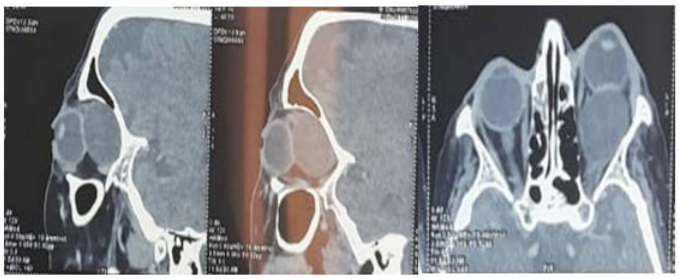

Figure 2: Orbital CT scan showing a well-limited extra conical formation with regular contours and non-enhanced liquid density after injection of contrast medium measuring 30 x 26 mm extended over the eye.

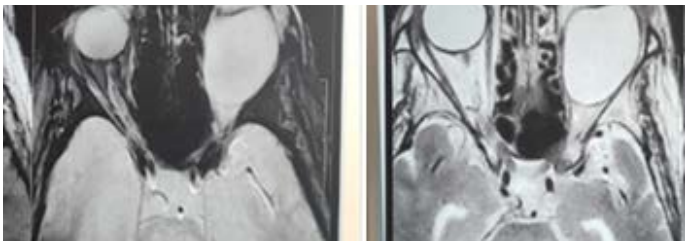

Figure 3: Orbital MRI : Extra conical intraorbital cystic formation with thin and regular wall in hyposignal on all sequences not enhanced after gadolinium injection measuring 35x28x27 mm, the content is in T1 hyposignal, T2 hypersignal disappearing on the FLAIR sequence without restriction of dissidence or enhancement after injection of gadolinium this mass exerts a mass effect on the intraorbital structures (the eyeball, the superior rectus muscle and the optic nerve).

Hydatidosis serology was positive. Enucleation of the cyst via the superolateral approach with abundant lavage with hypertonic saline solution following cyst rupture **(Figure 4)**.

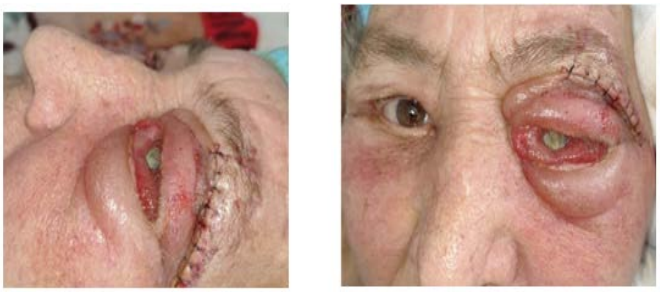

Figure 4: Immediate post-operative photos after enucleation by left lateral orbitotomy.

As a preventive measure, the patient received albendazole for 6 months.The clinical evolution was marked by the disappearance of exophthalmos and chemosis **(Figure 5)**.

Figure 5 : Ilage of the patient after two months of surgery showing a remarquable regression of exophtalmos et chemosis after the treatment.

Case Report 3

43-year-old woman with no particular pathological or trauma history, presenting with rapidly progressive left exophthalmos evolving for one month. Clinical examination revealed painless, irreducible, non-pulsatile, non-axial exophthalmos without thrill, with limited globe elevation, decreased visual acuity, exotropia and papilledema.

Imaging (CT and MRI) revealed a cystic lesion of the superior-internal angle of the left orbit, measuring 29x 20 x 17 mm, at the thin-walled level of the medial rectus muscle, pushing the eyeball and nerve outwards and causing grade I exophthalmos.

Hydatid serology was negative.Enucleation of the cyst was carried out via the internal paracanthal route, without breaking the cyst wall **(Figure 6)**.

Figure 6: Photo of cyst delivery via internal paracanthal approach.

Post-operative follow-up was favourable (disappearance of exophthalmos, good ocular motility and return of good visual acuity).

Discussion

It is well known that hydatid cysts affect the liver and lungs in 50-70% and 20-30% of cases respectively; however, they can appear in any part of the human body[1,2]. The orbit represents a

rare (1%) but not exceptional location for these cysts in endemic countries such as Morocco[3,4]. Commonly, orbital hydatid cysts are primary and unilateral, manifesting clinically as exophthalmos of insidious, non-pulsatile onset, with no palpable thrill; often non-axial and painless[5].

It is frequently diagnosed early in children, due to the limited space in the orbit[6]. In addition to exophthalmos, the mass effect resulting from cyst enlargement leads to periorbital pain, chemosis, restriction of extraocular motility, compressive optic neuropathy and optic atrophy, which may lead to reduced visual acuity or even blindness[7].

Until today, no case of bilateral orbital hydatid cyst has been reported. The cyst is most often located in the left orbit.

Orbital hydatid cysts tend to involve retrobulbar tissues, either inside the muscular cone or outside in the upper angles of the orbit. Inferior location remains exceptional.

Diagnosis is made in most cases by imaging (ultrasound, CT and MRI), but MRI represents the best paraclinical examination as it can rule out other lesions[8-10].

Infact : A CT scan reveals a round or oval lesion, hypodense, homogeneous, with regular boundaries and denser contours (capable of taking moderate contrast).MRI provides a better analysis of the cyst, which appears hypointense in T1 and hyperintense in T2, and the wall is enhanced after injection of gadolinium.

The serological tests used in difficult cases - enzyme-linked immunosorbent assay (ELISA) or Western Blot - can only affirm the diagnosis.Consequently, a negative test does not rule out the diagnosis.

Confirmation relies on histological study and/or direct identification of Echinococcosis granulosus protoscolae or hooks in cyst aspirates, but it should be noted that the clear appearance of cystic contents found intraoperatively is highly suggestive. However, in the absence of an epidemiological context, negative serology or inconclusive imaging, other differential diagnoses must be eliminated.

Several differential diagnoses may be evoked, including: a reworked cavernous angioma, a mucocele, a dermoid cyst, a colobomatous cyst, an epidermoid cyst and a post-traumatic hematoma.

In the absence of enucleation, the hydatid cyst will progressively form a thick, adherent shell with no cleavage plane with surrounding tissues, making complete dissection difficult.

Extracranial or transcranial approaches can be used to excise orbital hydatid cysts.

Depending on the location of the cyst, there are several approaches, including rhinotomy (ideally in the case of an inferointernal cyst), orbitotomy (lateral or paracanthal, or medial anterior supra superciliary).

Because of its complexity and thin wall, the orbital hydatid cyst often ruptures, causing severe anaphylaxis.It is therefore advisable to administer albendazole 2 weeks to 1 month before surgery, as an adjunctive treatment to reduce the risk of relapse[1]. Scholastic agents (e.g. 15% hypertonic saline, 0.5% silver nitrate, 30% hydrogen peroxide, 95% ethanol) can be instilled into the cyst immediately prior to surgery, or at the time of dissection of the mass and orbital fat above the cyst head, using absorbent cotton soaked in one of these solutions, or at the time of cyst rupture to prevent further spread or anaphylactic reaction (although direct mortality from echinococcosis is almost nil).

Indeed, the scolices present in the surgical field will be destroyed by osmotic desiccation.

Not forgetting the anaesthetist, who will need to be rapidly informed in order to take precautionary measures such as antihistamines and/or corticoids.

Finally, orbital hydatid cysts have a good prognosis if treated early[12].

The evolution is generally marked by the progressive disappearance of functional signs.

Recommandation

1. Orbital hydatid cysts should be considered as a differential disease in anyone presenting with unilateral proptosis and living in livestock-raising areas[13].
2. Adjunctive treatment by albendazole is preconised because complete excision of the lesion is difficult, and may lead to cyst rupture and subsequent complications.
3. The best outcome is achieved when the disease is treated early, before irreversible optic atrophy sets in.

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