



ECHINOCOCCOSIS: A RARE CAUSE OF PROPTOSIS IN INDIA

Ophthalmology

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ABSTRACT

Hydatid disease is a parasitic infestation caused by larvae of Echinococcus granulosus worm, commonly causing liver and lung involvement in the form of cysts with scolices. The disease is prevalent in patients having animal contact like sheep and dogs. Humans are accidental hosts, so larvae entering the circulation can lodge anywhere and form cysts. This case discusses one such case with formation of hydatid cyst in the left orbit without involvement of any other organ. **Purpose:** The purpose of this article is to report and discuss a case of proptosis, the diagnosis of which is a rare condition of orbital hydatid disease. We hereby discuss the approach to investigation, diagnosis and management of this case of proptosis.

KEYWORDS

Orbital Hydatid disease, hydatid cyst, echinococcus granulosus, proptosis.

INTRODUCTION

The incidence of orbital hydatid disease is less than 1%. Isolated orbital Hydatid cyst with no other systemic involvement is an extremely rare occurrence. For patients presenting with unilateral proptosis with or without diminution of vision, diplopia, and restriction of extraocular movements, a suspicion of orbital mass should be made.⁽¹⁾ Radiological investigations such as CT and MRI can be useful to diagnose such cases. Cystic lesions of the orbit include dermoid, abscess, haemangiomas, hydatid, lacrimal gland cyst, mucocele, etc.⁽²⁾ Orbital hydatid is more commonly unilateral at the superior location, with a predilection to left orbit.^(3,4)

Case Report

A 61 year old female presented with left sided protrusion of eyeball and inferior displacement of the eye since 6 months, which was painless and gradually progressive in nature. Patient denied any other ocular complaints. She had no complaint of diplopia. There was no history of fever with cough or weight loss. She had no history of any other systemic comorbidities such as thyroid disease. She denied history of Koch's or Koch's contact. She however did give history of contact with pet dog. On examination, she had left-sided proptosis with no lagophthalmos. Exophthalmometry was 17mm in the right eye and 20mm in the left eye showing a 3mm difference between the two eyes. There was 6mm of inferior dystopia associated with restriction of extraocular movements (-2) superiorly. There was orbital fullness however no specific mass was palpable. Orbital rim was palpable, with finger insinuation negative. Fundus examination was normal. USG orbit was suggestive of anechoic cystic lesion superior to the globe with small echogenic content within the cyst without vascularity. An MRI brain with orbital cuts was done which showed a low signal intensity cystic lesion with peripheral rim enhancement superior to the left globe on T1 weighted imaging with high signal intensity on T2 weighted imaging. A diagnosis of suspected hydatid cyst was made based on these findings. Systemic examination, Xray of the chest and USG abdomen was done to look for systemic involvement and it revealed no signs of hydatid disease at any other site. A CT orbit scan was done to look at the bony margins of the orbit which showed superior orbital wall scalloping with thickness of the superior orbital wall plate just 1.2mm. Surgical excision of the cyst was done by superior orbitotomy approach with a lid skin crease incision. Lid skin crease incision was given. Cyst isolated and fluid aspirated. Hypertonic saline injection locally as scolicidal agent. The shrunken cyst was then excised and Aspirated fluid was sent for cytology. Excised cyst was sent for Histopathology confirmation. Fluid cytology revealed proteinaceous material. Cyst wall was three-layered with scolices of the organism. Post-operatively, symptoms of proptosis and dystopia resolved immediately. Extraocular movements were full and free. A simple course of oral albendazole was given for 4 weeks along with wound care and topical antibiotics postoperative oral antibiotics. Upon follow up, the patient has no fresh ocular complaints till date. Apart from being a diagnostic challenge, this case was a test of surgical skills pertaining to the location of the cyst, which was superior to the superior rectus muscle with scalloping of the superior orbital wall with a minimum thickness of 1.2 mm.

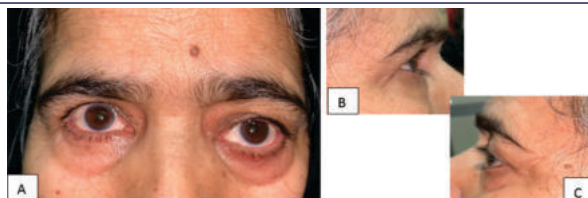


Figure 1: left eye proptosis with inferior dystopia seen on frontal(A) and lateral view (B right eye and C left eye). Orbital fullness appreciated on lateral view compared to the right eye.

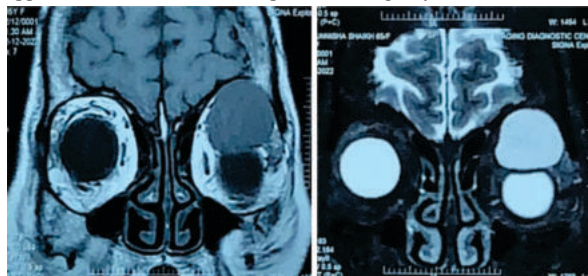


Figure 2: Coronal T-1 weighted image(left) showing cystic lesion with low signal intensity Superior to the left globe. Coronal T-2 weighted image(right) showing High signal intensity cystic lesion.

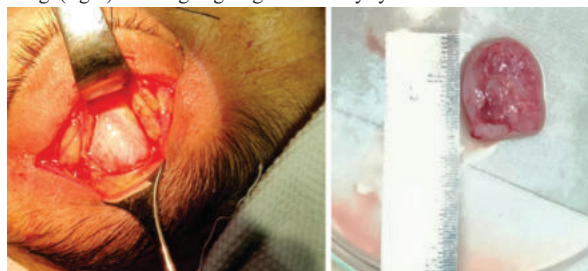


Figure 3: cyst wall visible through superior orbitotomy approach. (left) Cyst after excision(right)

DISCUSSION

On MRI well defined T1 hypointense T2 hyperintense cyst-differentials include⁽²⁾ Mucocele, teratomas, hydatid cyst and Cysticercosis

Pahwa S et. Al⁽²⁾, discussed various intra-orbital cystic lesions and the radiological findings- Parasitic cysts include the cysticercosis and hydatid cysts.

The hydatid cysts can be unilocular or multilocular, with fluid signal intensity contents which are T1 hypointense T2 hyperintense on MRI. Hydatid disease is the most common parasitic infestation in orbit. The extraocular muscles being the most common site show T1 hypointense cyst with eccentric scolex on MRI.

In a study conducted by F Limaïem et. al. in 2010⁽³⁾ A 74 year old farmer with proptosis, loss of vision with exposure keratitis and chemosis.

Diagnosed with orbital hydatidosis. They managed with cyst excision by frontoparietal craniotomy approach and post operative albendazole therapy. Similar to our case their symptoms resolved quickly.

In a similar study conducted by Taghipour M et. al. in 2017⁽⁴⁾, A 36 year old male developed right sided proptosis with disc edema. MRI revealed cystic lesion in retro-orbital fossa with pressure effect and medial displacement of optic nerve. Complete cyst excision was done for this patient.

A case reported by Al-Muala et al⁽⁵⁾ a 42 year old female had swelling and restriction of eye movements in the right eye. She was diagnosed radiologically and underwent cyst excision by lateral rhinotomy approach. Patient's symptoms resolved after the management.

Kahveci et al⁽⁶⁾ used the PAIR technique (Puncture-aspiration-injection-reaspiration) for the management of inferolateral presentation of an isolated orbital hydatid cyst. Similar to our case this patient didn't have any signs of liver cysts or other systemic involvement.

CONCLUSION

As an ophthalmologist, such a rare presentation of a systemic disease poses a great challenge since the diagnosis cannot be confirmed until after surgical removal of the cyst. Preoperative diagnosis is important to rule out systemic disease and the chances of intraoperative dissemination due to spillage and rupture of the cyst. In conclusion, in a case of unilateral proptosis diagnosed as orbital cyst, with or without systemic signs and symptoms, orbital hydatid disease should be considered as a differential diagnosis and care to be taken to prevent further dissemination with measures such as use of intraoperative scolicidal agents and post operative albendazole and steroids.

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