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Case Report

Intra Orbital Hydatid Cyst - An Unusual Presentation of a Common Disease

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Abstract

Hydatid cyst disease is a common presentation of a parasitic infestation by a tapeworm of genus *Echinococcus*. Hydatid cyst may present anywhere in the body but it is extremely rare for it to be isolated to intra orbital region. Literature is riddled with cases and discussion of multiple hydatid cysts within the thorax and abdominal cavity. There are only handful of reported cases of intraorbital hydatid cysts and most are within the Asian Subcontinent. We herein report a rare case of primary intra orbital hydatid cyst on a 12 year old otherwise healthy young patient. The patient presented with proptosis and mild visual acuity loss, with limited medial eye movements due to the presence of cyst in that region. This is one of very few cases reported worldwide of such a presentation and it is the first case noted and managed at our institution. Clinical diagnosis was confirmed by CT scan imaging. Patient was managed Preoperatively by two weeks of oral Albendazole. Surgical management of the patient included external fronto-ethmoidectomy to access the intra orbital cavity and total excision of the cyst. Post-operatively patient continued oral Albendazole for further three months and her recovery was uneventful. Albendazole has been widely described as the best therapeutic choice for hydatid cyst in the literature.

Keywords: Case Report; *Echinococcus*; Hydatid Cyst; Intraorbital Hydatid Cyst; Albendazole; Hypochloride Solution

Introduction

Hydatid disease is a zoonotic infection caused by a tapeworm of the genus *Echinococcus. Echinococcus* life cycle involves only 2 hosts, one definitive (canine) and the other intermediate (sheep). Humans act as an accidental intermediate hosts. Theoretically Echinococcosis can involve any organ but in literature, the liver is most common organ infected followed by the lung.

Case Report

A 12y female presented to ophthalmology department with slowly progressing, non-painful, proptosis of the left eye noted over two months duration. Initial Ophthalmology examination revealed a sluggish eye movement on the medial side of the left eye. Patient had lateral proptosis of the left eye with reduced visual acuity of the affected eye. Social history was unremarkable.

A CT scan was ordered, which revealed a large retro-bulbar, double density mass with ground glass content within the matrix. The mass had an enhancing capsule and was pushing into the ethmoids posterior-medially. There was no bony erosion noted and the lamina papyracea was intact. A Diagnosis of Intraorbital Hydatid cyst was made and patient was Referred to our ENT department for surgical removal of the Hydatid cyst.

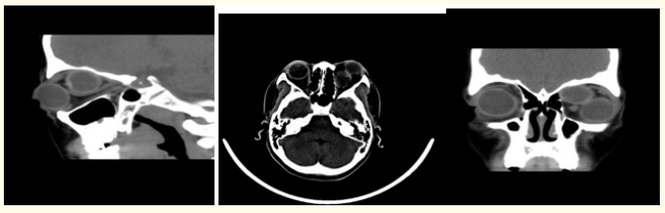


Figure 1: Sagittal, axial and coronal cuts of CT scan revealing laterally displaced eye as well as a ring enhancing cyst (note the similarity in density with the globe).

ENT management

ENT examination was as above notable clinical finding importance of patient from ENT perspective was that no nasal mass or nasal discharge on presentation. Further examination of CT scan showed clear sinuses. The retrobulbar cystic mass appeared to be along the postero-medial aspect of the orbit, causing infero-lateral displacement of the globe. The mass was clearly noted to be behind the medial rectus and encroaching on the posterior ethmoid sinuses. Lamina papyracea was of normal thickness and was intact.

Based on CT scan findings a clinical diagnosis of intraorbital hydatid cyst was made.

Patient received initial treatment of oral albendazole (anti helminthic) 400 mg twice daily for 2 weeks leading to surgery.

Surgical management: A Left external fronto-ethmoidectomy approach was used to expose and excise the relatively large hydatid cyst encapsulated within the orbital cavity. The cyst unfortunately burst during the dissection but its egg shell like white capsule (germinal layer) was removed in its entirety and prolonged washout of the operative site with sodium hypochlorite (Milton Solution) was done to prevent seeding. An irrigation drain was placed at the site before closure.

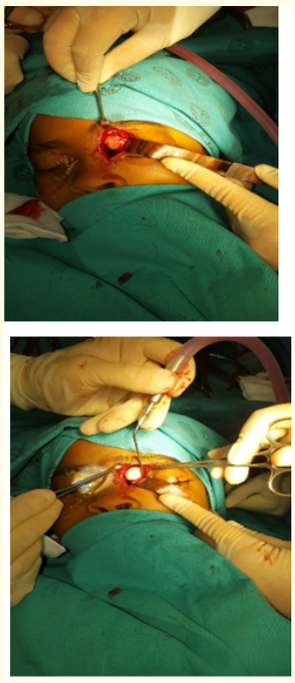


Figure 2: External approach revealing the cyst before enucleation (The second picture depicts the germinal layer of the hydatid cyst).

Patient received daily saline irrigation via the drain placed in the operative area. The drain and sutures were removed on day 5. Patient was discharged home on day 7 post operation period. She was further placed on oral Albendazole for 3 months post discharge in accordance to hydatid cyst treatment protocols.

Histological confirmation of a hydatid cyst was obtained from NHLS pathology lab of the specimen sent as follows.

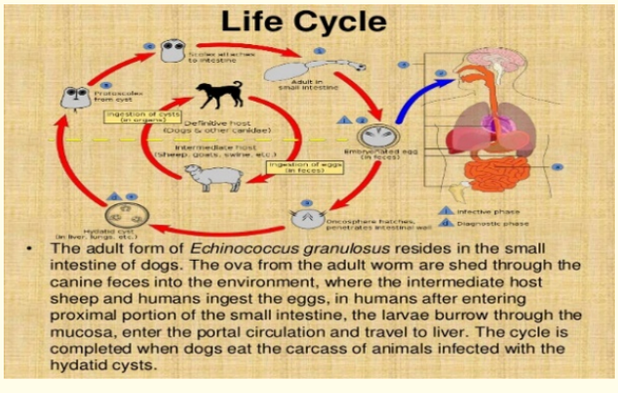
Numerous fragments of laminated cyst wall. Focal regions demonstrates a germinative layer (endocyst) with accompanying degenerate scolices. Refractile hooklets are noted within the scolices. Surrounding fibrous host tissue is not present.

Pathological DX = orbital, excision hydatid cyst.

There has been no recorded recurrence of the disease clinically and radiologically (CT PNS and orbit) at two years. Her pyroptotic left eye has returned to its socket and the eye movement as well as the visual acuity has normalised.

Discussion

Hydatid disease is a parasitic infestation by a tapeworm of genus *Echinococcus*. In humans symptoms are caused by the larval cestodes of tapeworms [1,3]. Humans are accidental intermediary hosts and are infected Via the faeco-oral spread, particularly when in contact with dogs that are infected with *Echinococcus* and shed tapeworm eggs in their faeces [1,4]. Hydatid cyst commonly affects the liver in 60 -70% of cases. Lung infections contribute to around 25% of cases [6]. Primary Hydatid cyst of the orbit is extremely rare and only a handful of cases have been reported in literature mainly around the Asian Subcontinent [2].



Figure

There are four known species of *Echinococcus* that can cause disease, three of which are of medical importance in humans [1].

Echinococcus granulosus, is the most common strain and causes cystic echinococcosis.

Echinococcus multilocularis, causes alveolar *Echinococcus*. It is rare but is the most virulent-*Echinococcus vogeli* is the rarest species.

Southern parts of Africa is considered intensive endemic area for hydatid cysts because of the population of the two hosts, sheep and dog.

Clinical signs and symptoms of cystic *E. granulosus* varies as it slowly enlarges. Visceral *E. granulosus* can present with an abdominal mass or pain due to pressure effects of enlarging cysts. Morbidity from this disease is secondary to free rupture of the cyst (with or without anaphylaxis), infection or dysfunction of the affected organ.

The disease is rarely diagnosed during childhood or adolescence unless there is a rupture of the cyst and anaphylaxis sequelae that follow a ruptured cyst. Presentation to hospital is earlier if the brain or eye is involved, due to the limited space within the bony cavity of the cranium and the orbit.

Diagnosis is often made clinically in conjunction with CT scan imaging. Ultrasound imaging is not of much value in diagnosis of intraorbital hydatid cysts [2]. Blood work may reveal some eosinophilia but this is not diagnostic of this particular form of disease.

Treatment of the intraorbital hydatidosis requires both medical and surgical management. Medical management with oral Albendazole aims to reduce cyst size prior to surgery as well as sterilise cysts in case of accidental spillage of cyst contents [5-7]. The use of Albendazole in the therapy of pre- and post- operative, intraorbital hydatid cyst is not well documented as compared to visceral hydatid organ involvement. We, however opted to treat in accordance to visceral hydatid organ involvement, which recommends Oral dose of Albendazole of 400 Mg 12 hourly, two weeks prior to surgery. We opted to treat post operatively for three months due to the rupture of the germinal layer during the intraoperative period. Recommended guidelines are up to 6 months postoperatively [5].

Surgical management aims to eradicate Hydatid cysts with complete excision of the cyst [6]. Intraoperative irrigation with a hypertonic or sclerosing agent such as Miltons solution will assist in eradicating any local spread and seeding [6]. Medical treatment with anthelmintic agent such as Albendazole for a prolonged period post operatively will ensure that any residual or subclinical systemic disease is eradicated.

Due to rarity of this particular presentation of the disease, a good degree of suspicion is required to make the diagnosis. A positive history of living in endemic area with exposure to contaminated food or water by definitive hosts will ensure that correct diagnosis is made.

Conclusion

Hydatid cyst of the orbit is a rare occurrence even in intensely endemic areas [2]. A high degree of suspicion must be present in addition to the positive history of living in endemic areas where the two definitive hosts of the Echinococci live freely. Once diagnosed, treatment consists of both medical and surgical management. Medical management aims to reduce the size of cysts preoperatively and to ensure eradication of systemic residual helminths post operatively. Surgical management aims to eradicate the cyst that is causing the clinical presentation. For cysts that present in small but bony cavities, our departments treatment of choice is Surgical excision of the cyst followed by irrigation of the affected area and a prolonged course oral Albendazole.

Prevention is of utmost importance and can be implemented by education. Education needs to include that of proper hygiene, adequate cleansing and avoidance of eating raw contaminated food where the hosts roam freely.

We also second the recommendation that pet dogs in endemic areas be treated with praziquantel [1] periodically.

Acknowledgement

Dr Claudio Favara.

Disclosure

No personal or Financial Gain for the author from this presentation. This case was submitted and accepted as a poster presentation to the South African ENT society's Annual conference in Port Elizabeth, South Africa.

Consent

Informed consent was obtained in writing from both the patient and her parents. Family agreed to this case being pictured and potentially discussed at Various ENT meetings and conferences. No incentives offered to the patient and family for the informed consent.

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