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Case Report

Unusual presentations of extraocular cysticercosis: A clinical challenge to the ophthalmologists

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Introduction

Cysticercus cellulosae, the larval form of the cestode, *Taenia solium*, affects human eye in 13–46% of larva infested patients. Ocular cysticercosis occurs in all ethnic groups regardless of dietary habits and is endemic in tropical areas, such as sub-Saharan Africa, India, East Asia, Mexico and Latin America.¹ The human becomes an accidental intermediate host by ingesting food or water contaminated with eggs or autoinfection.

Ocular cysticercosis has varied presentations and may be extraocular (subconjunctival, extraocular muscles or orbital tissues) or intraocular (vitreous, subretinal space, or anterior chamber). Extraocular cysticercosis is reported more

commonly from India.² We report two cases of extraocular cysticercosis in young having unusual presentations leading to a wrong diagnosis.

Case reports

Case 1

A 32 year old male patient was transferred in from peripheral hospital with complaints of redness, acute pain and swelling right eye of 04 days duration. He also complained of double vision on looking up. There was no history of recent trauma, diminution of vision, bleeding or discharge from right eye. There was also no history of fever, sinusitis, nodular swelling elsewhere over body, vomiting, headache or seizures. He gave past history that something entered lower eyelid of right eye while he was chopping meat 02 years back and has been having mild pain, redness and lower lid swelling off and on since then, which used to subside in about five days with topical drops. General physical and systemic examinations were unremarkable. There were no palpable subcutaneous nodules or enlarged regional lymph nodes.

Ocular examination revealed Distant Visual Acuity (DVA) of 6/6 and Intra Ocular Pressure of 12 mmHg in both eyes. The right eye showed lower lid oedema and on retracting lower eyelid a subconjunctival swelling in inferior fornix with congested conjunctival vessels over and around swelling was noticed. The subconjunctival swelling was approximately 12 × 8 mm, smooth, oval, tense cystic, partially mobile, non-reducible, tender and pinkish with central yellowish

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discolouration suggestive of an abscess (Fig. 1). Rest of the anterior & posterior segment of both eyes was normal. There was no proptosis of right eye. The ocular movements revealed restriction in elevation in right eye (Fig. 1). The differential diagnosis of Subconjunctival abscess or Orbital Pseudotumor was made. He was started on systemic and topical antibiotics.

Routine laboratory investigations and radiograph of Paranasal Sinuses were normal. Computerized Tomographic Scan (CT Scan) and Ultrasonography (USG) of Orbit showed features suggestive of Right myositis of Inferior Rectus with in continuity a focal lesion in lower fornix showing area of liquefaction (Fig. 2). CT Scan head was normal. Subsequently swelling increased with increasing pain and redness of right eye. Repeat USG of right eye showed increased focal area of liquefaction and inflamed swelling of Right Inferior Rectus (RIR). At surgery pus was drained out and a thin walled, creamy-white cyst of 10×6 mm extruded out spontaneously. Histopathological examination (HPE) confirmed the diagnosis of Extraocular Cysticercosis (Fig. 3). No organism was isolated from pus. Stool examination revealed no segment or egg of Taenia. He was treated with oral Prednisolone 60 mg per day and oral Albendazole 400 mg BID for 4 weeks.

After 6 weeks there was partial resolution of the restricted ocular motility with diplopia in extremes of upgaze (Fig. 4). USG of orbit revealed decreased thickening of RIR. Six months

later he still has minimal ocular motility restriction with no diplopia.

Case 2

A 24 year old male patient, presented with history of something entering left eye late in evening while he was trying to put a nail in the wall. He noticed redness, foreign body sensation and swelling in the upper fornix in the left eye and reported to hospital on subsequent day. General physical and systemic examination revealed no abnormality. DVA in both eyes was 6/6. After retracting the upper eyelid in left eye, a smooth, pinkish, oval, subconjunctival swelling of approximately $8 \text{ mm} \times 6 \text{ mm}$ was observed 8 mm from limbus at 10 O' clock. The swelling was tense, loosely adherent to the sclera, non-reducible, and was mildly tender. The conjunctival vessels over and around swelling were mildly congested. No other abnormality was detected in the eye or ocular adnexa. Fundus was normal. The right eye was normal. Ocular movements were full. A provisional diagnosis of subconjunctival foreign body granuloma was made.

He was started on topical Moxifloxacin (0.5%) + Dexamethasone (0.1%) eye drops 6 hourly, Moisol eye drops 6 hourly and Flurbiprofen (0.03%) eye drops 6 hourly. As no response was seen, a USG of the left eye was done (Fig. 5). CT Scan brain was normal. An excision biopsy was carried out through upper fornix conjunctival incision and a cystic mass was expressed

Fig. 1 – Subconjunctival cyst at inferior fornix abutting eyeball with restricted ocular motility in up gaze in right eye.

Fig. 2 – CT Scan of orbit – the arrow showing Right inferior rectus myositis with diffuse thickening of inferior rectus including its tendinous insertion and in continuity a focal lesion in lower fornix which is isodense with inferior rectus with focal area of liquefaction on coronal cut (left) and sagittal view (right).

Fig. 3 – HPE (H & E stain, $\times 40$) showing cysticercus larvae with scolex (annotated by red arrow), hooklets (annotated by black arrow), suckers (annotated by green arrow) and membranous body with tubules encased in a corrugated cuticular layer.

out in intact condition which was grossly 6×4 mm in size, creamy-white in colour, with a milky white spot in the centre which indicated the position of scolex. The diagnosis of Subconjunctival Cysticercosis was established by HPE. Patient was reviewed regularly and is asymptomatic.

Discussion

The clinical manifestations and therapy of orbital cysticercosis depend on the location, size, relation to adjacent structures and stage of evolution of the cyst i.e. inflammatory response.

The common presenting features of myocysticercosis are restricted ocular motility, diplopia, recurrent pain and redness as exemplified by our first case and it needs to be differentiated from myositic form of idiopathic inflammation of the orbit. Any of the extraocular muscles can be involved but superior rectus has been reported to be more common. The sterile abscess in our case was probably caused by inflammation induced by leakage or release of antigens into the surrounding tissue from the cyst. All cases dramatically improve on treatment with Albendazole and steroid, however surgical excision of cyst may be required in few cases.^{2,3} In our first case long history of disease resulted in residual ocular motility restriction.

Subconjunctival cysticercosis usually presents as a painful, yellowish, nodular subconjunctival mass with surrounding conjunctival congestion. However small subconjunctival cysticercosis may not invite any complaints as it was in second case due to which it came to the notice of the patient after foreign body fell into his left eye. Excisional biopsy is recommended for subconjunctival cysticercosis, as cysticidal drugs have little effect owing to poor drug penetration.⁴

Fig. 4 – Residual motility restriction in up gaze in right eye after 06 weeks of therapy.

Fig. 5 – USG of left eye – the arrow showing a subconjunctival cyst in the medial aspect of the eyeball with clear contents and a central echogenic scolex attached to the inner wall of cyst.

The ultrasonography findings of hanging drop sign (Fig. 5) may be missed in the orbit and subconjunctival location as the cyst may provoke acute inflammation thereby masking the classical sonographic features as was in our first case leading to a wrong diagnosis. Inability to detect scolex on CT scan was again because of severe myositis with focal liquefaction. Higher resolution scans with 1 mm cuts and spiral CT scans with image reconstruction could be used for better visualization of scolex.

In a retrospective case series by Rath et al., the cyst in 80.7% of orbital and subconjunctival cysticercosis is usually in relation to extraocular muscle.² The cyst attached to the muscle sheath induces an inflammatory reaction and because of its constant motility it comes to lie in subconjunctival space as subconjunctival swelling or erodes through the conjunctiva resulting in spontaneous extrusion.^{3,5} As the cyst may be adherent to the adjacent muscle, care must be taken not to damage the extraocular muscle during dissection.

A high index of suspicion of cysticercosis should be entertained for a subconjunctival swelling or myositis in endemic areas to ensure timely diagnosis, treatment and decrease ocular morbidity. It would be wise to investigate all such cases with ultrasonography and CT scan. Improving

sanitation and health awareness are the only ways to prevent cysticercosis.

Conflicts of interest

The authors have none to declare.

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