CASE REPORT

ORBITAL CYSTICERCOSIS MASQUERADING AS INTERNUCLEAR OPHTHALMOPLEGIA
Nita Shanbhag¹, Taran Wanage², Shalakha Sobti³, Neha Vyas⁴, Nupur Bhatt⁵

HOW TO CITE THIS ARTICLE:

ABSTRACT: A Muslim lady, 24 year old presented with complaints of headache, orbital pain and right eye adduction deficit mimicking Internuclear Ophthalmoplegia. However absence of diplopia in primary or left gaze made us look for another cause of ocular pain. A Type II Duane Syndrome was ruled out as there were no alterations in palpebral aperture on horizontal gazes. A painful swelling in right upper lid corner a week later made us suspect lacrimal gland inflammation. USG B scan revealed cysticercosis cyst with hyperdense scolex, around the lacrimal gland. USG did not show any EOM involvement. CT scan was done to rule out neuro-cysticercosis or any other cause of adduction deficit. Patient was successfully treated with Antihelminthics and Steroids. Orbital cysticercosis with Adduction deficit masquerading as internuclear ophthalmoplegia without muscle involvement is rare, hence been sent for publication.

KEYWORDS: Orbital Cysticercosis, Internuclear Ophthalmoplegia, Lacrimal gland, Duanes syndrome.

CASE REPORT: INTRODUCTION: Tapeworms of the genus Taenia can cause two different human diseases, Taeniasis and Cysticercosis. Taeniasis is an intestinal infection caused by the adult T. Solium and T. saginata. Cysticercosis, the most common ocular platyhelminth infestation in humans, is caused by encystment of the larvae (cysticercus cellulosea) of the tapeworm Taenia solium. Humans are the intermediate hosts in the life cycle. Cysticercosis is acquired by ingestion of infective Cysticerci in undercooked pork or ingestion of eggs of Taenia Solium in contaminated water, food, vegetables or regurgitation of eggs from the small intestine.[1] (Figure 1).

CASE REPORT: A Muslim lady 24 year old came with right orbital pain since 15 days. There were no signs of inflammation, no congestion. BCVA was 6/6 for both eyes. Anterior segment findings were normal. Pupil was normal size reacting to light and fundus was normal, intraocular pressure was 17mmhg. Extra ocular movements showed an adduction deficit in the right eye, of which the patient was unaware. She did not complain of diplopia. Our first thought was a Duane’s type II syndrome however there were no palpebral aperture changes on abduction or adduction.

To think this to be internuclear-opthalmoplegia, the lack of diplopia was contradictory to a third nerve involvement. Patient was referred for neurological evaluation. Patient on next visit presented with tender swelling in the lacrimal gland area with conjunctival congestion, all other findings remaining status quo. (Figure 2) (Figure 3) (Table 1). The patient was now sent for radiological evaluation. A USG B Scan showed a cyst with hyperdense echo in its center. (Figure 4) A diagnosis of cysticercosis around the lacrimal gland was done. A CT scan brain was done to rule out neuro-cysticercosis or any other cause of adduction deficit. (Figure 5)
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Patient was treated with antihelminthics and steroids as the site was extraocular with no intraocular involvement. Albendazole was chosen over Praziquantel Albendazole 400mg twice a day for one month, along with oral Prednisolone one mg per kg of body weight was preferred treatment. Steroids were gradually tapered. Patient responded to above treatment with marked reduction in swelling, inflammatory signs and adduction deficit. (Figure 6) (Table 1)

DISCUSSION & REVIEW OF LITERATURE: Cysticercus cellulosae, the larval form of the pork tapeworm Taenia solium, is the causative organism of cysticercosis, in which humans are the intermediate hosts in the life cycle. Cysticercus cellulosae may become encysted in various bodily tissues, usually the eyes, central nervous system, and subcutaneous tissues (Figure 1). Acquired strabismus, diplopia, recurrent redness, and painful proptosis are some of the clinical signs in patients with orbital cysticercosis. Diagnosis of cysticercosis is based mainly on orbital imaging because of its highly specific appearance. Medical therapy is the main stay of treatment.

Taeniasis and Cysticercosis occur where sanitary conditions are poor and where raw and undercooked contaminated pork and beef are routinely consumed. It may also occur on ingestion of unwashed vegetables contaminated by cysticercus cysts.

Ocular and adnexal cysticercosis represents 13% to 46% of systemic disease. Ocular cysticercosis can involve any part of the eye, 4% involve the eyelid or orbit, 20% involve the subconjunctival space, 8% involve the anterior segment, and 68% involve the posterior segment (subretinal and intravitreal,[2] although vitreous, subretinal and subconjunctival cysticercosis have been reported most often, the orbit is occasionally the site of cyst lodgement.[3-4] Ocular involvement is usually unilateral but bilateral involvement may occur in cases of disseminated cysticercosis.[5-6] in the eyelids it presents as a subcutaneous, painless, mobile mass with varying degrees of mechanical ptosis. In Conjunctiva it presents as painless or painful yellowish, nodular mass with congestion.[7] Cysticercosis of extraocular muscle usually presents as recurrent pain, redness, ocular motility restriction, diplopia and ptosis.[8] any muscle may be involved with greater predilection for superior and lateral rectus.[9]

The diagnosis of myocysticercosis is based on clinical, serologic, and radiological findings. Diagnosis is made by stool examination and finding the eggs of proglottids of the worm. Serological tests used for the specific diagnosis of cysticercosis are indirect hemagglutination, indirect immunofluorescence, and immunoelectrophoresis such as ELISA. High resolution Ultrasonography (USG), computed tomography (CT) and Magnetic Resonance Imaging (MRI) help in detection of the orbital cyst.

B-scan ocular ultrasonography reveals a well-defined cystic lesion with clear contents and a hyperechoic area suggestive of a scolex. Typically, A-scan USG shows high amplitude spikes corresponding to the cyst wall and scolex. The scolex shows a high amplitude spike due to presence of calcareous corpuscles.[10] CT scanning of the orbits show a hypodense mass with a central hyperdensity suggestive of the scolex. Usually, a solitary cyst with wall enhancement is observed. Adjacent soft-tissue inflammation may be present. The scolex may not be visible if the cyst is dead or ruptured and has surrounding inflammation. Concurrent neurocysticercosis may be present and should be excluded.
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MRI may reveal a hypointense cystic lesion and hyperintense scolex within the extraocular muscle or elsewhere in the brain.\textsuperscript{[11-12]} A complete blood count count may reveal eosinophilia.\textsuperscript{[13]}

Gross examination of the live cyst may reveals a globular, elongated or oval, milky white cyst varying from 0.5 to 0.3cm in diameter with translucent wall and a white, opaque, dot-like area at one point, indicating the position of the scolex. It may transilluminates against light. On sectioning a live cyst, clear fluid is released, and a small 2–3mm nodule is seen on the inner wall of the capsule suggestive of an invaginated scolex with birefringent hooklets.\textsuperscript{[14]}

Albendazole and praziquantel are the larvicidal drugs used in the treatment of cysticercosis in human. Albendazole has largely supplanted praziquantel because of slightly greater cure rates, decreased cost and increased availability. It has also been observed that patients not responding to praziquantel show a response to albendazole. (Table 2). It is of utmost importance to rule out intraocular and central nervous system involvement. Dying cysticercus releases its toxin and incites severe inflammatory reaction leading to vitritis and may lead to blindness. Prophylactic and concomitant administration of corticosteroids is recommended to avert an inflammatory response.that usually occurs 2-5 days after initiation of therapy. Neurocysticercosis has been treated successfully with oral albendazole and with praziquantel.

Surgical removal of the Cysticercus is the recommended treatment for subconjunctival space cysticercosis when adjacent to a muscle, great care needs to be taken as there could be an extension. If the cyst is adherent to the adjacent muscle, excision may be difficult. If the cyst wall is opened, the content should be aspirated and the area irrigated with hypertonic saline. Patient in whom, the cyst is opened during surgery, probably should be treated medically.

Subconjunctival abscess formation may occur due to release of toxins, which require drainage and removal of the cyst. Resolution of the cyst may take from a few days to months depending on the density of the surrounding inflammation. Based on the individual’s response to medical therapy, another course of medication may be required.

Serial B-scan ocular ultrasonography or CT scanning of the orbit helps to follow the resolution of the cyst, which is recognized by the disappearance of the scolex.

Cysticercosis can be prevented through practicing good hygiene measures, such as washing hands frequently, washing raw vegetables and fruits well before consumption to prevent fecal-oral transmission and avoiding consumption of raw or undercooked pork and other meat.

In conclusion orbital cysticercosis can have variety of presentation depending upon the site of lodgement and may mimic various neurological disorders putting the physician on the wrong track. A basic knowledge and understanding of the disease coupled with high index of suspicion could help us reach an early clinical diagnosis. USG B scan and CT scan can easily confirm the diagnosis. We are presenting this case, as despite the lack of involvement of extraocular muscles, there can be a mechanical restriction of ocular movements, mimicking nerve palsy, in our case an internuclear ophthalmoplegia.

The adduction deficit, improved because the cysticercus migrated sub-conjunctivally releasing the mechanical restriction. The patient was treated with systemic steroids and Albendazole (Table 2). If the lesion is extraocular localized to a particular site, surgical excision can be done alone depicting the characteristic histopathological findings. (Figure 7)
Medical line of management completely absorbed the cyst and was chosen as it would resolve any other possible cysticerci elsewhere in the body. However these drugs should be avoided in intraocular lesions as their use may excite an inflammatory reaction within the eye and can lead to irreversible damage to the delicate structures of the eye.

<table>
<thead>
<tr>
<th>Ocular Examination</th>
<th>First Visit</th>
<th>Second Visit</th>
<th>Third Visit</th>
</tr>
</thead>
<tbody>
<tr>
<td>BCVA</td>
<td>6/6 - No Diplopia</td>
<td>Upper Lid Oedema</td>
<td>Normal</td>
</tr>
<tr>
<td>Ocular Adnexa</td>
<td>Normal</td>
<td>Clear</td>
<td>Clear and swelling markedly decreased in size</td>
</tr>
<tr>
<td>Conjunctiva/Sclera</td>
<td>Clear</td>
<td>Conjunctival congestion With a Cystic Swelling superotemporally in the lacrimal gland area.</td>
<td></td>
</tr>
<tr>
<td>Cornea</td>
<td>Clear</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Anterior Chamber</td>
<td>Well Formed</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Iris</td>
<td>Color Pattern Normal. No signs of Kps / Flare or Cells</td>
<td>Orthophoria in primary gaze with improvement in Adduction restriction</td>
<td></td>
</tr>
<tr>
<td>Pupil</td>
<td>Central circular reacting to light</td>
<td>Clear</td>
<td>Full and free</td>
</tr>
<tr>
<td>Lens</td>
<td>Clear</td>
<td>Clear</td>
<td>Clear</td>
</tr>
<tr>
<td>EOM</td>
<td>Orthophoria in primary gaze with Adduction restriction</td>
<td>Orthophoria in primary gaze with improvement in Adduction restriction</td>
<td></td>
</tr>
<tr>
<td>Fundus Examination</td>
<td>Normal</td>
<td></td>
<td></td>
</tr>
<tr>
<td>IOP on Aplanation</td>
<td>17 mm of Hg at each visit</td>
<td></td>
<td></td>
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</tbody>
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Table 1: Right eye ocular examination on subsequent visits, Left eye being Wnl

<table>
<thead>
<tr>
<th></th>
<th>Albendazole</th>
<th>Praziquantel</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mechanism of Action</td>
<td>Albendazole sulfoxide active element - Larvicidal</td>
<td>It induces rapid contraction of schistosomes by a specific effect on permeability of the cell membrane destroys Larvae as well as Adult worm</td>
</tr>
<tr>
<td>Dosage</td>
<td>15 mg / kg / day x 1 month 400 mg BID x 1 month better absorbed with Fatty Food</td>
<td>50 mg / kg / day x 14 days Effect enhanced by High Carbohydrate diet, Cimetidine &amp; Grape fruit juice</td>
</tr>
<tr>
<td>Side Effects</td>
<td>agranulocytosis, liver function abnormalities, balding, nausea, vomiting, dizziness, headache</td>
<td>Lesser</td>
</tr>
</tbody>
</table>
Drug Reactions

<table>
<thead>
<tr>
<th>Effects on Cysts</th>
<th>With antiseizure drug dilantin and carbamazine and corticosteroids</th>
</tr>
</thead>
<tbody>
<tr>
<td>Able to Eliminate 85% of Cysts</td>
<td>Able to Eliminate 75% of Cysts</td>
</tr>
<tr>
<td>Enhanced Potency as it reduces its elimination</td>
<td>Decreases blood levels reducing the potency</td>
</tr>
<tr>
<td>Concomitant corticosteroid usage 1 mg.kg / day</td>
<td></td>
</tr>
<tr>
<td>Cost</td>
<td>Lesser</td>
</tr>
<tr>
<td>Efficacy</td>
<td>More</td>
</tr>
<tr>
<td>Patients not responding to Praziquantel respond to Albendazole</td>
<td>May be given when non-responsive to Albendazole</td>
</tr>
</tbody>
</table>

Table 2: Antihelminthics for Cysticercosis.[15,16,17]

Fig.1: Life Cycle of Taenia Solium
Fig. 2: RE inflammatory swelling in lacrimal gland area

Fig. 3: EOMs in all cardial gazes depicting RE

Fig. 4: B Scan USG showing Cysticercosis cyst with scolex around
Fig. 5: CT scan showing cyst of cysticercosis around lacrimal gland

Fig. 6: Resolution of cyst and adduction deficit Post treatment

Fig. 7: Histopathology of cysticercosis
REFERENCES:
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