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CASE REPORT

Isolated Cysticercosis of Inferior Rectus Muscle Presenting With Eccentric Proptosis – Case Report and Treatment Review

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ABSTRACT

Intraocular infections by Cysticercus Cellulosae larvae are often found as part of a generalized systemic infestation. Reports of orbital adnexal cysticercosis are uncommon, despite the high incidence of brain and ocular involvement, with isolated infestation of extraocular muscle being exceedingly rare. We at our institute report a rare case of isolated right inferior rectus muscle cysticercosis which presented with unilateral eccentric proptosis and restriction of upward gaze.

Key Words: Inferior Rectus Palsy, Cysticercosis, Orbital Cysticercosis.

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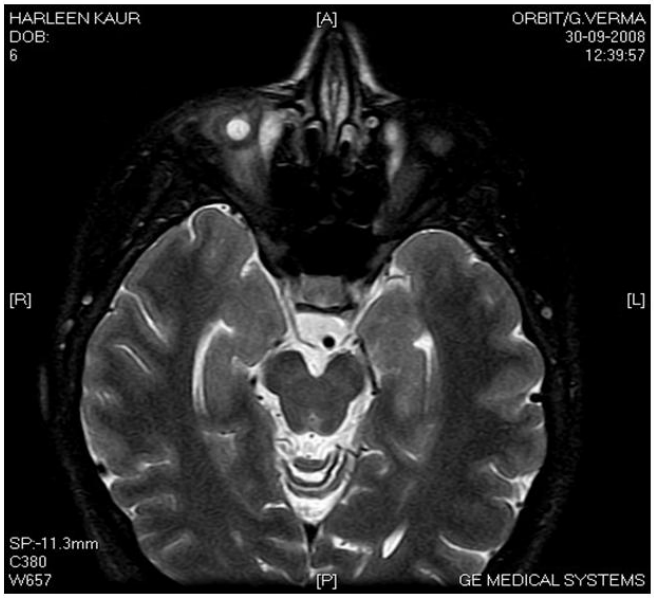
Introduction

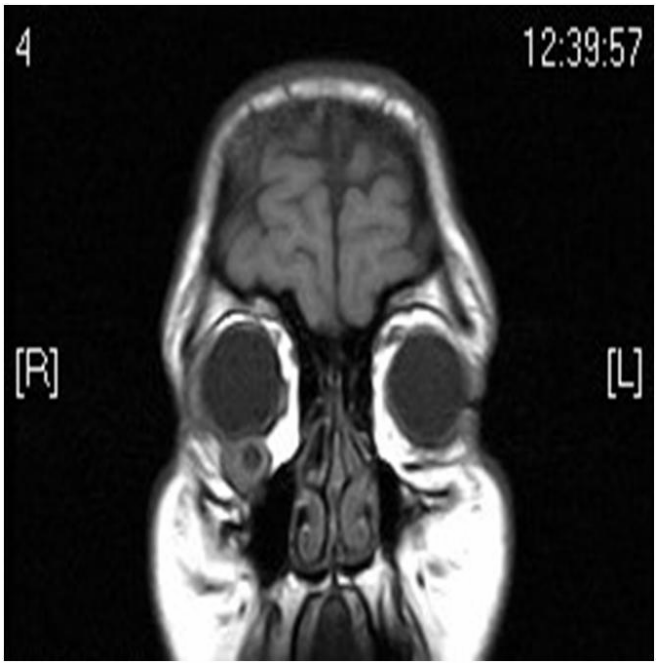
Cysticercosis is caused by larval form of Taenia Solium. The commonest pattern of systemic involvement is seen as neurocysticercosis which appears as a space occupying intracranial lesion. Intraocular infections by Cysticercus cellulosae are more common compared to ocular adnexal involvement and are often found as part of a generalized systemic infestation. Isolated ocular involvement is rare with isolated orbital adnexal involvement an occasional occurrence in clinical practice[1]. In our clinical practice we came across a patient who presented with isolated cysticercosis of inferior rectus muscle and discuss management options in such patients.

Case Report

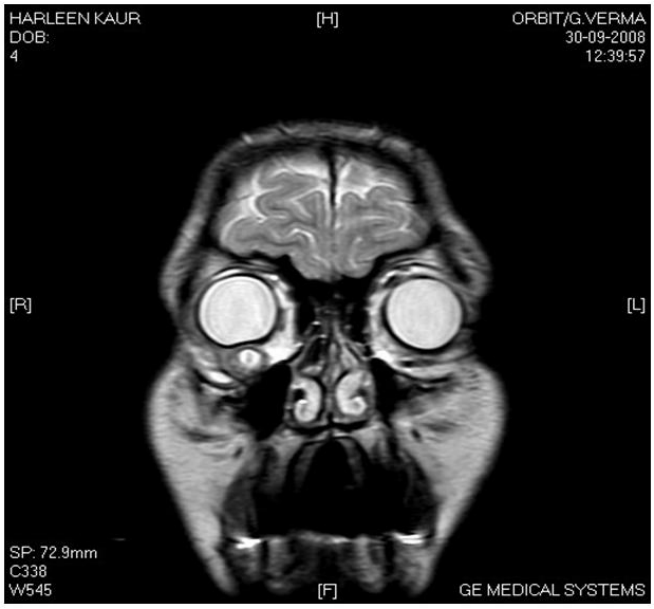
A 16 year old female presented in our Out Patient Department with chief complaints of mild unilateral eccentric proptosis and restriction of upward gaze in right eye. Her vision and fundus examination were normal and there was no pain during extraocular movements, with restriction of upward gaze in right eye. There were no signs of inflammation in conjunctiva. All routine blood investigations were normal. A provisional diagnosis of orbital tumor was made and MRI orbit with brain was suggested. MRI orbit revealed a cystic lesion in the belly of inferior rectus muscle in the right eye. It was hypointense in T1 as shown in [Table/Fig 1] and hyperintense on T2 as shown in [Table/Fig 2] weighted images. A tiny eccentric hypointense speck suggestive of scolex was seen as shown in [Table/Fig 3]. A diagnosis of cysticercosis was made and since fundus examination and MRI showed no intraocular cysts patient was treated with Albendazole (15 mg/ $kg/_{day}$)along with oral prednisolone (1 to 1.5 mg /$kg/_{day}$) for 4 weeks. Patient gradually improved and upward gaze palsy improved . Her proptosis also disappeared on subsequent follow-ups.



(Table/Fig 1) Axial T2 weighted image showing cystic lesion in inferior rectus muscle



(Table/Fig 2) Coronal T1 weighted image showing cystic lesion in right inferior rectus muscle belly



(Table/Fig 3) Coronal T2 weighted image showing hyper intense cyst with eccentric hypointense speck suggestive of scolex

Discussion

Life Cycle

Taenia Solium (along with its larval form cysticercosis cellulose) is most common species causing cysticercosis in humans. Taxonomically, taeniasis (infection by the adult worm) must be differentiated from the cysticercosis (infection by the larvae). However, patients may harbor both taeniasis and cysticercosis. Taeniasis is an intestinal infection caused by consumption of the adult worm through undercooked pork. It occurs in non vegetarians and is not associated with ocular disease[2]. In cysticercosis, the humans act as an intermediate host following the consumption of eggs in contaminated food or water and can occur even in vegetarians. After ingestion, the eggs hatch and mature to larvae, which are carried by mesenteric vessels to various parts of the body, where they are filtered through subcutaneous and intramuscular tissues, with preference for the brain and eyes [3] .

Demographic Features

Ocular disease is reported to occur in a significant number of cases of cysticercosis . Ocular cysticercosis is caused by the growth of the larvae of Taenia solium with in the ocular tissues. Soemmering was the first to report the cyst in the eye, in 1830 and Schott extracted the cyst

from the anterior chamber in 1836. Since then there have been many reports of ocular cysticercosis published in the literature. The cysts may be located in descending order of frequency, subretinal (35%), vitreous(22%), conjunctiva (22%), anterior segment (5%) and orbit(1%)[4]. There may be geographic difference in the incidence of location of ocular cysticercosis;for example, involvement of conjunctiva is most commonly reported in India[5]. Despite the high incidence of brain and ocular involvement reports of orbital adnexal cysticercosis (1%) are uncommon with isolated infestation of extraocular muscle being exceedingly rare.

The clinical manifestations of ocular cysticercosis, like elsewhere in the body are determined by the location, size and by the host's immune status and inflammatory reactions. It is believed that the viable cysts, evoke little inflammatory response, while the degenerating cyst rapidly increases in size due to osmotic regulation and causes compression of the surrounding structures [6].

Clinical Presentation

The 3 main symptoms at presentation were periocular swelling (38%), proptosis (24%), and ptosis (14%) with a median duration of 2 (range 0-24) months. The 3 main signs at presentation included ocular motility restriction (64.3%), proptosis (44.4%), and diplopia (36.8%). The cyst locations in the decreasing order of frequency were anterior orbit (69%), subconjunctival space (24.6%), posterior orbit (5.8%), and the eyelid (0.6%). In all, 80.7% of patients had cysts in relation to an extraocular muscle. The superior rectus (33.3%) was the most commonly involved extraocular muscle [7].

Investigations

The diagnosis of cysticercosis can be presumptively established by the presence of small calcific densities on roentgenogram of the skull or extremities. Other tests such as electroencephalography, CT scan, or arteriography may confirm space occupying intraparenchymal brain lesions. Eosinophilia may be present in disseminated disease. Serological testing is often inconclusive, more than 50% of patients with neurocysticercosis having no quantifiable antibody response [8]. Spinal fluid analysis with radioimmunoassay may reveal detectable titres. The diagnosis of non-calcified orbital cysticercosis may be elusive until the cyst is surgically excised.. Diagnosis of ocular cysticercosis is usually accomplished by direct ophthalamoscopic demonstration of the larval worm. Ultrasound, CT and MRI are other modalities for establishing the diagnosis. MRI is however the best tool for intraocular cysticercosis and may confirm presence of coexistant neuro-cysticercosis. MRI imaging is the best method of assessing patients with presumed orbital cysticercosis . If routine sequences do not show the intraocular cyst clearly, a high resolution CISS sequence may be used [2]. A high degree of suspicion and use of correct sequence with thin slices is necessary to reach the diagnosis.

Medical Management

Medical treatment is known to cause severe ocular complications, which may lead to loss of the eye. However these days orbital adnexal disease in the absence of intraocular disease is being treated medically, with Albendazole (15 mg/ kg-/$_{day}$)along with oral prednisolone (1 to 1.5 mg /kg/$_{day}$ for 4 weeks). The therapeutic efficacy of this drug for extraocular muscle cysticercosis has been reported to be good [9].With administration of this drug spontaneous extrusion of cyst from the subconjunctival location has been reported [10].

Role of surgery

The best course is the removal of the cyst by surgery. As with intraocular cysticercosis, it would be better that the orbital cysts are removed/ extrude out than be killed inside. However, surgery for orbital myocysticercosis is fraught with complications as it may damage important orbital structures because the cyst is adherent to the surrounding structures from inflammatory reaction, and is best avoided. Corticosteroids and anti-inflammatory drugs are given to suppress the inflammation. We have noticed that the cysticerci of the extraocular muscles travel forward, come to lie in a subconjunctival location and then extrude out spontaneously. We have adopted a policy to wait and watch in these cases. We avoid giving steroids and antihelminthics as these suppress inflammation and delay the movement of the cyst outward, and hence, its extrusion

The classical description of orbital cysticercosis is that of an anterior orbital mass with a severe inflammatory reaction. The patients experience considerable pain accompanied by oedema, chemosis, ptosis, optic

neuritis, and restriction of extraocular movements [1]. However as our case points out these patients can present with variety of symptoms such as painless eccentric proptosis and restriction of upward gaze. MR imaging is the investigation of choice in such cases. In the absence of intraocular disease medical therapy results in clinical improvement and resolution of all symptoms.

Conclusions

Ocular cysticercosis is common in conjunction with neurocysticercosis with isolated ocular involvement a rare clinical presentation. Orbital adnexal involvement is a rare event with only few cases reported worldwide. Isolated involvement of extraocular muscles is a rare phenomenon and can present in unusual ways as in our case. Hence possibility of orbital cysticercosis should be kept in mind in patients presenting with proptosis and isolated extraocular muscle palasy. In the absence of intraocular disease medical therapy results in resolution of symptoms and occasional spontaneous extrusion of cyst from the subconjunctival location.

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