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Case Report

Extraocular cysticercosis - atypical involvement of Medial Rectus muscle: A case report

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ABSTRACT

Cysticercosis, caused by the larval form of the pork tapeworm Taenia solium is potentially harmful disease with a variable clinical manifestation. The most commonly involved sites include eye, brain, bladder wall, and heart. Ocular cysticercosis can be extraocular or intraocular and may present with varied clinical symptoms. We report the case of extraocular cysticercosis in a 20 year old female presented with right exotropia, head tilting and painful right eye. CT-scan of right orbit showed bulky medial rectus with multiple ring enhancing lesions within the muscle, suggestive of cysticercosis. It becomes important to report this case because of the relative rarity of the condition, unusual site of the cyst i.e atypical involvement of medial rectus and the young age of the patient.

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1. Introduction

Cysticercus cellulosae, the larval form of the pork tapeworm Taenia solium, is the causative organism of cysticercosis. Humans act as the intermediate host in its life cycle. The larva gets encysted in different tissues of the body, commonly the eye, central nervous system and subcutaneous tissues. The eye is a prime location for development of cysticercosis due to its rich vascularization. Incidence is common in the tropical areas, particularly the developing countries and listed as one of the neglected tropical diseases.[1,2]

Cysticercosis is a parasitic infection of humans by Cysticercus cellulosae, the larval form of Taenia solium, commonly known as the pork tapeworm. It infects humans by ingestion of undercooked meat. When humans ingest T. solium eggs by feaco - oral route, they become accidental intermediate host. The oncospheres (primary larvae) penetrate the intestinal mucosa and enter the circulatory system. Hematogenous spread to neural, muscular, and ocular tissues occurs. Within these tissues, the oncospheres develop into secondary larvae (ie, the cysticerci). Ocular and orbital cysticercosis has varied presentations depending upon the site of involvement, number of lesion and the host immune response. Acquired strabismus, diplopia, recurrent redness, and painful proptosis are some of the clinical signs in patients with orbital cysticercosis. Ocular cysticercosis may be extraocular (in the subconjunctival or orbital tissues) or intraocular (in the vitreous, subretinal space or anterior chamber).[3–5] One or more extraocular muscles may be simultaneously involved, although a propensity for involvement of the superior muscle complex and the lateral rectus muscles has been reported.[6–9]

2. Case Report

A 20 year - old Indian female presented with right-sided esotropia, head tilting and redness of right eye. On evaluation of the patient, her vision was 6/12 (R/E) and 6/6 (L/E), Right-eye esotropia; restricted ocular motility laterally, head tilting, right conjunctiva was congested, inflammation and tenderness of supra-temporal part of right orbit. All her other ocular parameters were normal in both her eyes. On systemic examination, her BP = 120/70mmhg,

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pulse = 84 bpm, patient was well oriented with no h/o seizures, bilaterally chest was clear, heart sounds – S1 and S2 were normal and abdominal cavity was normal and soft with normal bowel sounds. She did not give any h/o HTN, TB, T2DM or any other systemic illness. The patient was provisionally diagnosed to have Right lacrimal gland mass/inflammation and was advised CT-scan of orbit and brain and routine blood investigations viz.- CBC + ESR, RBS and Sreum Creatinine. She was empirically started on Tab. Cefpodoxime (200mg) and Tab. Indomethacine (75mg).

Fig. 1:

Her serial axial and coronal sections of orbit were studied with and without contrast media and it revealed – bulky right medial rectus muscle with multiple ring-enhancing lesions within the muscle – possibility of cysticercosis. Her NECT brain did not reveal anything significant.

The patient was subsequently started on – Tab. Albendazole (400mg twice daily after meal for 1 month) and Tab. Prednisolone (1mg/kg/day, started at 50mg/day and tapered over a period of 1 month). There was marked improvement of the clinical signs on this therapy. There was marked reduction in inflammation, improvement in ocular motility, and reduction in pain.

A repeat CT-scan of orbit was done again after the completion of the therapy. The CT-scan showed marked reduction in size of the lesion. There was also marked reduction in the inflammatory response of the surrounding tissues. There were no associated systemic symptoms associated with the treatment regimen.

3. Discussion

Cysticercosis is a condition where the larva of Taenia solium, known as Cystercercus cellulose gets enlodged in various tissues of the body. Humans act as intermediate host in the life cycle of Taenia solium. Ocular cysticercosis occurs quite frequently due to its rich lymphatics and vasculature. Due to the presence of the encysted larva, an immunologic reaction with fairly intense inflammatory signs and symptoms may be produced, leading to compression of surrounding tissues. Ocular cysticercosis may be presented as an extraocular (in the subconjunctival or orbital tissues) or intraocular (in the vitreous, subretinal space, or anterior chamber) infestation.[3–5]

Cysticercosis affects an estimated 50 million people worldwide. Ocular cysticercosis is endemic in tropical areas, such as sub-Saharan Africa, India, and East Asia. The reported incidence of ocular involvement varies from 10-30% in endemic areas.[10–16] In India, both intraocular cysticercosis and extraocular cysticercosis is observed with almost equal frequency. Amongst the extraocular tissues, extraocular muscle form is the commonest type. Lodgment of cysts in the subconjunctival space is another common site, followed by the eyelid, optic nerve and retro-orbital space. One or more extraocular muscles may be simultaneously involved, although a propensity for involvement of the superior muscle complex (33%) and the lateral rectus muscles has been reported. In our case, the patient however presented with atypical involvement of medial rectus.[6–9]

The clinical signs and symptoms of extraocular cysticercosis is mainly due to the inflammatory reaction to the larva and due to the space-occupying lesion that it produces, leading to the compression of surrounding structures. Cysticercosis of extraocular muscle usually presents as recurrent pain, redness, ocular motility restriction, diplopia, proptosis and ptosis. Duane Retraction Syndrome or Brown Syndrome may be observed, depending on the muscle group involvement. Blapharoptosis may be seen in case of severe inflammation of the superior muscle complex.

Table 1: Clinical presentations of patients with extraocular muscle cysticercosis[17]

Presentation	Percentage %
Restricted ocular motility	88%
Inflammatory signs	70%
Diplopia	53%
Ptosis	35%
Strabismus in the primary position	26%
Proptosis	16%

In the studies, it has been found that extraocular cysticercosis is a common condition in the tropics which affects the younger age group, with median age of presentation at 13 years.[18]

Our patient was a 20 years old female who presented with right-sided esotropia, head tilting and redness of right eye. On evaluation of the patient, her vision was 6/12 (R/E) and 6/6 (L/E), Right-eye esotropia; restricted ocular motility laterally, head tilting, right conjunctiva was congested, inflammation and tenderness of supra-temporal part of right orbit. The features were due to inflammatory constriction of the medial rectus muscle due to the immunological reaction to the cysticercus cysts. The patient was initially diagnosed

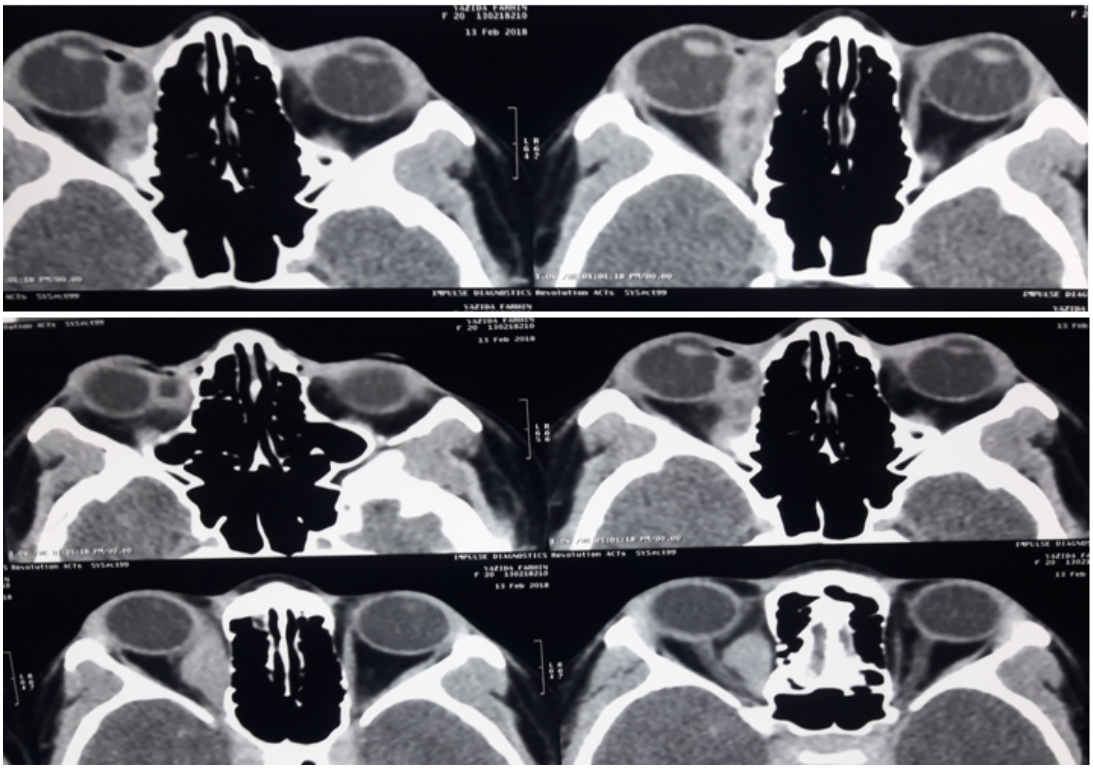

Fig. 2: CT-orbit

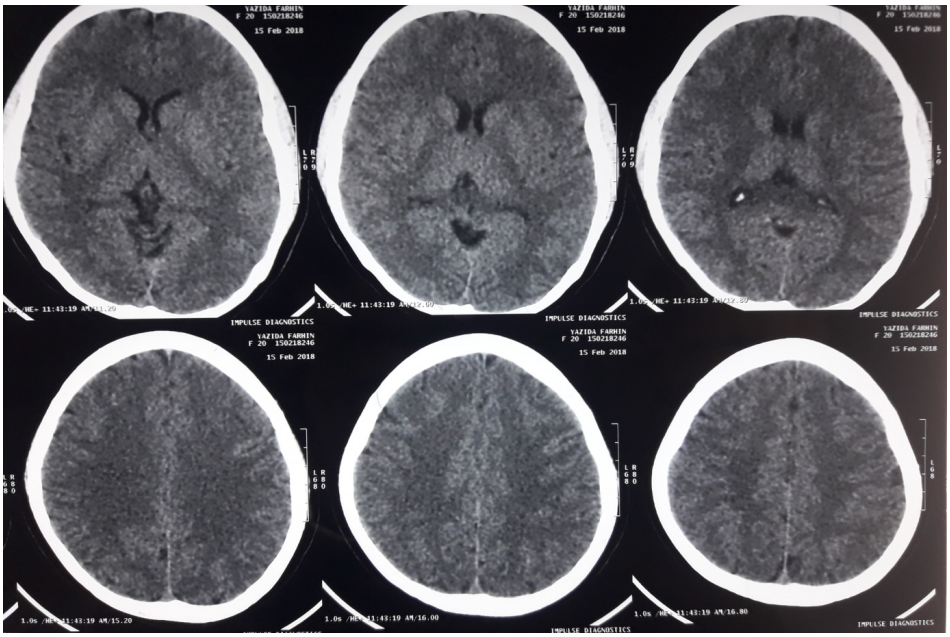

Fig. 3: CT-Brain

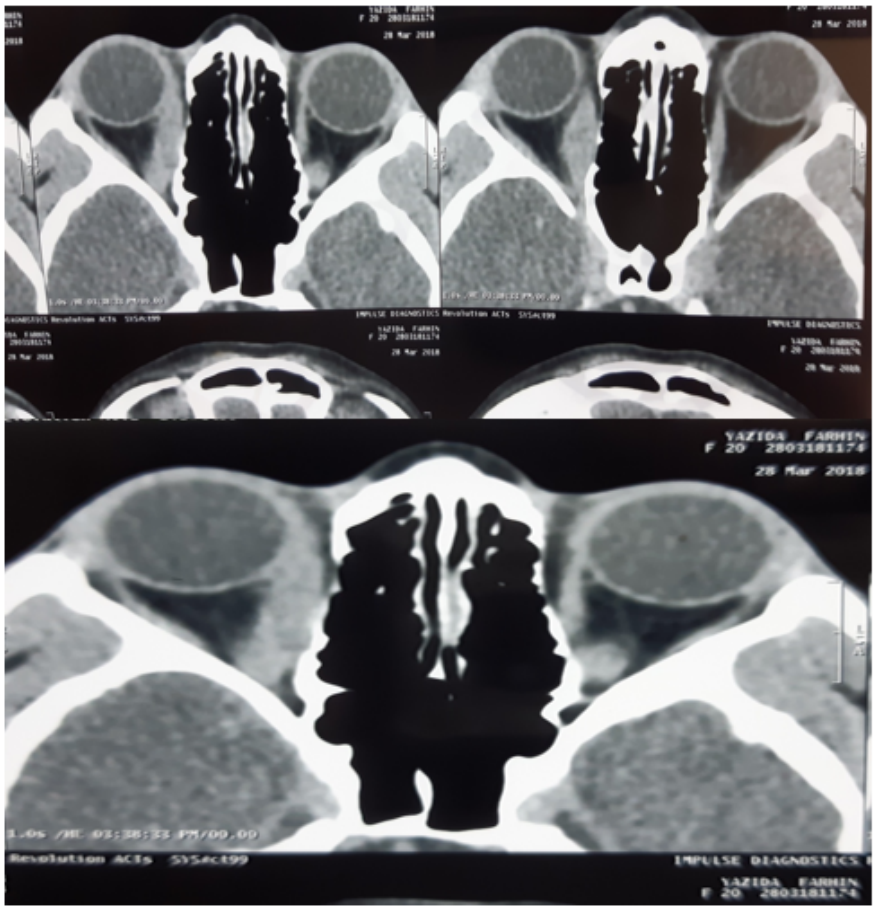

Fig. 4:

with Right lacrimal gland inflammation.

The important imaging techniques used include – computerized tomography (CT) scan of Orbit and contact B-scan Ultrasonography of the orbit. It has been found that CT scan of orbit is a reliable and cost-effective technique with high degree of sensitivity and specificity. CT scan shows a characteristic hypodense mass with central hyperdensity which is suggestive of scolex. Usually, a solitary cyst with wall enhancement is observed. The CT-orbit also reveals any inflammation or compression of the surrounding structures due to the cyst. The scolex is not visible in case ruptured cyst, where there will be intense surrounding inflammation. Any calcification of old cysts can also be visualized. A CT-scan of brain should also be done to exclude concurrent neurocysticercosis.

Contact B-scan ultrasonography reveals a well definedcystic lesion with clear contents and a hyperechoic area suggestive of scolex. This can be observed either intraocularly or within the extraocular soft tissues. In case of intraocular cysticercosis, any other associated ocular pathology can also be observed. Studies have shown that sensitivity of USG B-scan for extraocular cysticercosis to be as high as 83%.

Other imaging modalities for cysticercosis include MRI orbit which reveals hypointense cystic lesion and hyperintense scolex.

Our patient was advised – CT-orbit and CT-brain. Her serial axial and coronal sections of orbit were studied with and without contrast media and it revealed – bulky right medial rectus muscle with multiple ring-enhancing lesions within the muscle – possibility of cysticercosis. Her NECT brain did not reveal anything significant.

Various laboratory techniques have been devised which include – serum ELISA for anticysticercal antibody – however its specificity is only 50% for extraocular cysticercosis. A CBC count may reveal eosinophilia. FNAC of the cyst may be done to confirm the diagnosis.

In our patient; CBC, RBS and Serum Creatinine levels were checked as other tests were not available. Her CBC revealed, marked eosinophilia (differential count=10%) and marginally raised ESR value (25 mmAEFH). All other parameters were within normal range.

Although for intraocular cysts surgical excision is mandatory, the treatment of extraocular muscle

cysticercosis has undergone a radical change with the successful results of medical alternatives to surgical excision of the cysts. Anthelminthics (eg, albendazole, praziquantel) and oral corticosteroids have been found to be effective in extraocular disease. The standard regimen for treatment of extraocular cysticercosis is a 4-week regimen of oral albendazole (15 mg/kg/d) in conjunction with oral steroids (1.5 mg/kg/d) in a tapering dose over a 1-month period.

Our patient, who was 45kg in weight, was subsequently started on – Tab. Albendazole (400mg twice daily after meal for 1 month) and Tab. Prednisolone (1mg/kg/day, started at 50mg/day and tapered over a period of 1 month). The treatment regimen showed marked improvement in the condition of the patient.

4. Conclusion

Extra-ocular cysticercosis is a common clinical condition in the developing world. It typically affects young individuals and has a wide spectrum of clinical manifestations. Of the affected extra-ocular structures, the superior extra-ocular muscle complex is the most common, though other groups may also be affected. Both B-scan ultrasonography and computed tomography scan are useful in confirming the diagnosis. Medical therapy for the condition is now the treatment of choice for this condition and has now largely replaced surgical excision.

5. Source of Funding

None.

6. Conflict of Interest

None.

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