CLINICAL STUDY

Medical management of orbital myocysticercosis: a pilot study

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Abstract

Purpose **To evaluate the efficacy of albendazole in the management of orbital myocysticercosis.**

Method **Twenty-one consecutive patients diagnosed as having orbital myocysticercosis by ultrasonography, supported by computed tomography (CT)/magnetic resonance imaging (MRI), were included in the study. All patients received oral albendazole at a dosage of 30 mg/kg for 15 days with a low-dose steroid cover (5–10 mg per day). The patients were followed on day 2, at 2 weeks, 1 month, 3 months, 6 months and 9 months, and finally at 1 year. Orbital sonography was performed at 2 weeks, 3 months, 6 months, 9 months and 1 year. CT scan was performed at 6 months and 1 year.**

Results **Orbital sonography revealed a well-defined cystic lesion with clear contents and a hyperechoic area suggestive of a scolex in all the patients. CT or MRI provided additional supportive evidence. The size of the cysts measured before treatment ranged from 6.2 to 13.4 mm (mean 11.4 mm). Medial rectus was involved in 10 cases, superior rectus in 7 cases and lateral rectus in 4 cases. Serial ultrasonography revealed a gradual reduction in the cyst size in 20 patients. A mild obscuration of the cyst wall, followed by collapse of the cyst cavity and obscuration of the scolex, were progressively seen as the cyst reduced in size. Complete resolution of the cyst was seen in all cases at 6 months. A CT scan performed at 1 year supported the ultrasonographic findings. No systemic side effects were noted.**

Conclusion **Oral albendazole appears to be highly efficacious in the management of orbital myocysticercosis.**

Key words Albendazole, *Cysticercus cellulosae*, Orbital myocysticercosis

Cysticercus cellulosae, the larval stage of *Taenia solium*, is usually found in pig. Man becomes infested by ingesting *T. solium* eggs from contaminated soil or food. The commonest pattern of systemic involvement is neurocysticercosis, which presents as an intracranial space-occupying lesion. The larval form may enter the eye through the choroidal circulation and migrate into the subretinal space or enter the vitreous cavity.[1–3] Diagnosis of intraocular cysticercosis is easy because of its visibility.[2] Ocular adenexal involvement in cysticercosis is exceptional.[3,4] Diagnosis is based mainly on orbital imaging (ultrasonography, computed topography) and serology. Availability of high-resolution orbital ultrasonography has led to more effective detection and follow-up of these lesions. Medical therapy with praziquantel or albendazole has been found to be effective in the management of neurocysticercosis.[5,6] However, reports about usage of these drugs in the management of orbital cysticercosis are scarce.[7] This prospective study was carried out to evaluate the efficacy of albendazole in the management of orbital myocysticercosis.

Patients and methods

Twenty-one consecutive patients diagnosed as having orbital myocysticercosis by ultrasonography, supported by computed tomography (CT)/magnetic resonance imaging (MRI), were included in the study. Investigation comprised a detailed clinical history including dietary history, best corrected Snellen acuity, anterior and posterior segment evaluation, evaluation of ocular motility and Hertel's exophthalmometry. Ultrasonography of the orbit was performed with a Nidek US 3000 Echoscan machine, using A- and B-scan imaging. Imaging with CT or MRI was also carried out. An ELISA for cysticercosis was performed in all patients.

Treatment

All patients received oral albendazole at a dosage of 30 mg/kg for 15 days. A low-dose steroid cover (5–10 mg prednisolone per day as a single dose after breakfast depending on the patient's weight) was given throughout this time to suppress any acute inflammatory reaction secondary to a possible release of toxins

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following death of the cyst. In patients who had received previous treatment with steroids, albendazole treatment was deferred until 1 month after stoppage of the steroids.

Follow-up

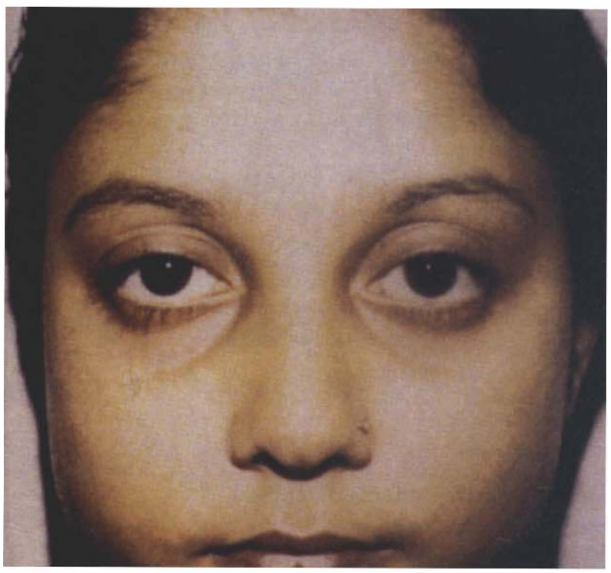
Patients were followed on day 2 after starting therapy, at 2 weeks, 1 month, 3 months, 6 months, 9 months and at 1 year. A detailed investigation was performed at each visit. Ultrasonography was repeated at 2 weeks, 3 months, 6 months, 9 months and finally at 1 year. A CT scan was performed at 6 months and 1 year.

Observations

The study included 11 males and 10 females aged 3–40 years. The commonest mode of presentation was proptosis (14 patients; Fig. 1a), followed by ocular motility disorders (4 patients; Fig. 1b), ptosis (2 patients) and an acute orbital cellulitis (Fig. 1c). Many patients had recurrent episodes of acute redness and inflammation.

Thirteen patients were not vegetarians while 8 were vegetarians. Fourteen patients had received at least one course of steroids, 2 of whom were still receiving steroids at the time of presentation. Orbital ultrasonography revealed a well-defined cystic lesion with clear contents and an eccentric hyperechoic area suggestive of a scolex in all the patients (Fig. 2a). CT or MRI provided additional supportive evidence (Fig. 2b).

The size of the cysts measured on ultrasonography ranged from 6.2 to 13.4 mm (mean 11.4 mm). The medial rectus was involved in 10 cases, the superior rectus in 7 and the lateral rectus in 4 cases. No cysts were found in the inferior rectus or the oblique muscles. No case



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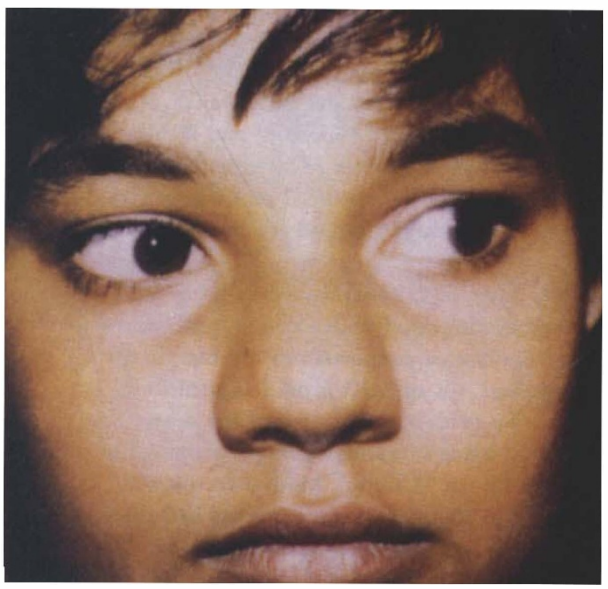

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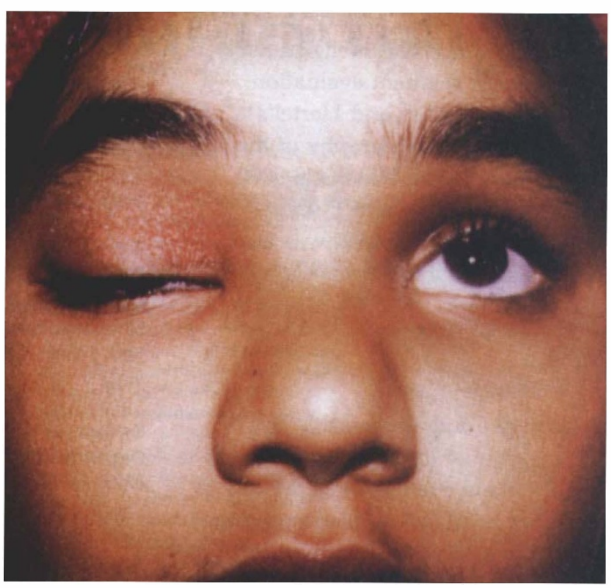

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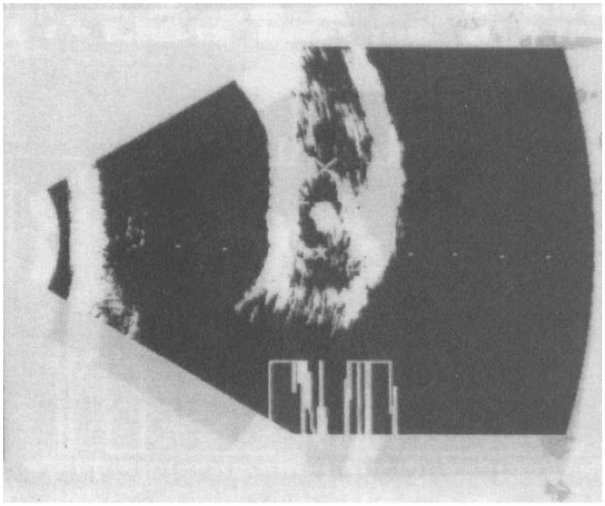
Fig. 1. *Clinical presentations in orbital myocysticercosis. (a) Proptosis; (b) ocular motility disturbance; (c) acute orbital cellulitis.*



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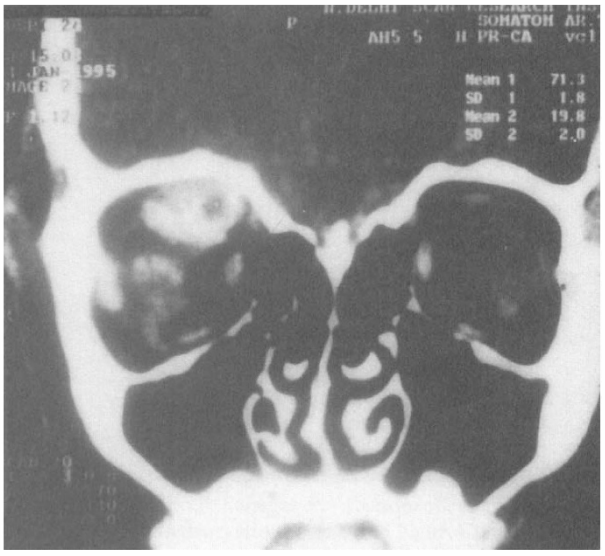


(b)

Fig. 2. *(a) Orbital B-scan ultrasonography demonstrating a cysticercus. (b) CT scan of the orbit showing a right superior rectus cysticercus.*

showed any evidence of any cysticercosis anywhere. Serology for cysticercosis was negative in all cases except one.

One patient presented with acute swelling and conjunctival chemosis and periorbital oedema 1 day after starting the treatment.

Ocular evaluation after 2 weeks of therapy showed a slight improvement in the clinical signs. Ultrasonography revealed a mild obscuration of the cyst wall in places (Fig. 3a). The scolex was clearly defined in 18 cases. A decrease in the size of the cyst was recorded in 20 cases. Follow-up at 1 month revealed a significant symptomatic improvement in all but one of the patients and a reduction in clinical signs. B-scan sonography now revealed a marked decrease in the size of the cyst and obliteration of the capsule (Fig. 3b). The scolex could not be detected in 20 patients, whilst in 1 patient a well-defined scolex was still present. The following description pertains to the 20 patients who showed a response.

The follow-up visit at 3 months revealed complete resolution of symptoms in all patients except 1, although some patients did complain of diplopia in extreme gaze. Ultrasonography in these patients now revealed a collapsed cyst with obliteration of the scolex and the clear area. Increased signals were received from the involved muscle, suggestive of inflammation (Fig. 3c).

Sonographic evaluation at 6 months did not reveal any cysts in the muscle. Areas corresponding to the site of the cysts now revealed increased signal suggestive of infiltration and inflammation (Fig. 3d). Sonographic follow-up at 9 months revealed only diffuse echoes from the involved muscles which persisted at 1 year (Fig. 3e). A CT scan performed in these 20 patients after 1 year revealed complete resolution of the lesion, although mild thickening of the muscle was often seen. In one patient the cyst persisted with no increase in size at all stages of evaluation. No systemic side effects of the therapy were noted.

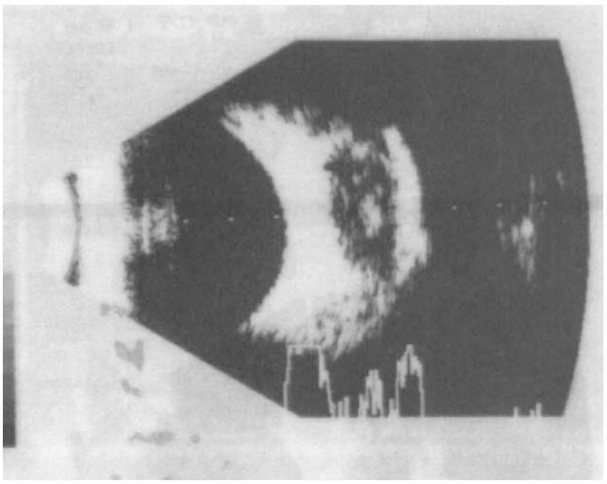
Discussion

Orbital myocysticercosis is a relatively uncommon condition in the west but appears to be endemic in Mexico, Africa, Eastern Europe, South-East Asia, and Central and South America. A review of the literature reported orbital involvement in only 1% of cases.[2,7] Medical management has been tried extensively in cases of neurocysticercosis, but its application to orbital cysticercosis remains to be proven.[5,6]

Praziquantel and albendazole have been shown to be highly effective in therapy of neurocysticercosis. Albendazole, a broad spectrum antihelmintic that acts by blocking glucose uptake of the parasite and depleting its glycogen stores, has been reported to have an elimination rate of 80% as compared with 67% for praziquantel.[5] Albendazole has also been shown to be effective in cases not responsive to praziquantel. Response to medical therapy was reported 3 months after initiation of the therapy.

Adverse reactions seen with these drugs are due not to the drug *per se* but to a strong inflammatory response secondary to the release of toxins following the death of the parasite. Simultaneous use of systemic low-dose corticosteroids and non-steroidal anti-inflammatory agents has been advocated to suppress this response. Medical treatment has no role in the management of intraocular cysticerci due to the potential sight-threatening side effects of toxin release following death of the parasite.

We attempted albendazole therapy in myocysticercosis in view of the rich blood supply of the extraocular muscles, ensuring availability of the drug. A double-masked controlled study would not have been



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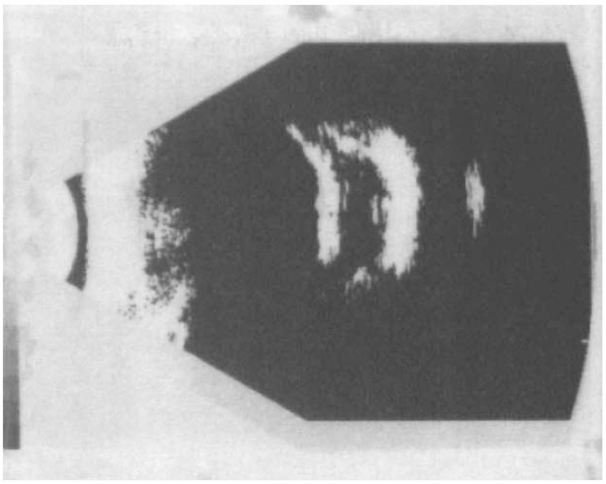

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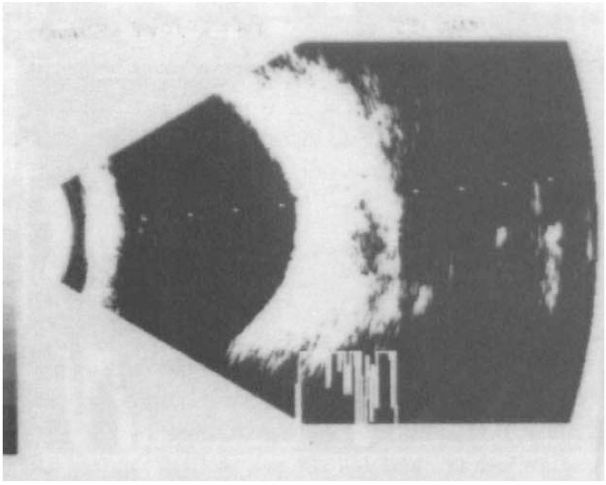

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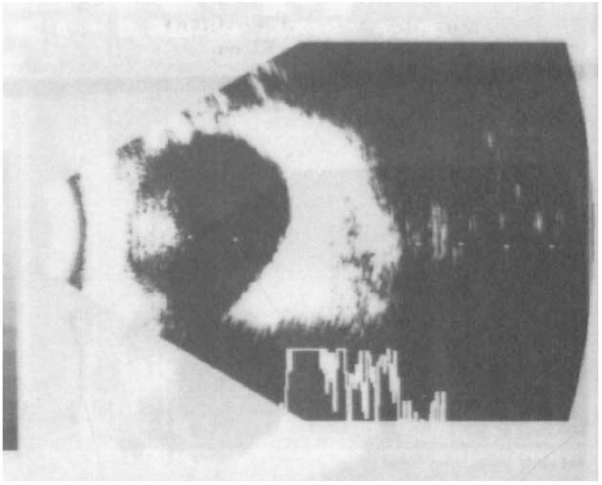

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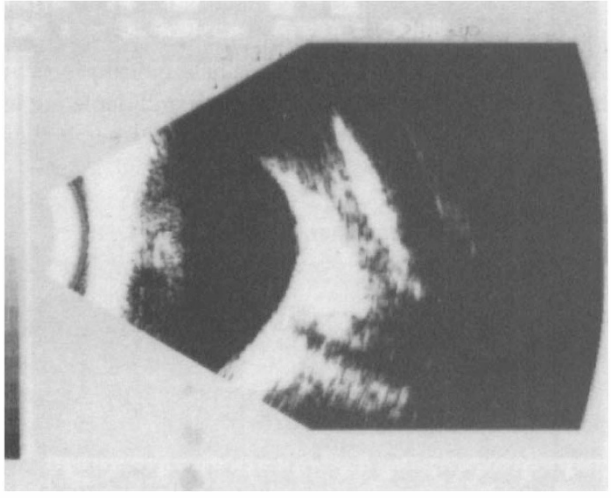

(e)

Fig. 3. *(a) Orbital B-scan ultrasonography 2 weeks following therapy, showing mild obscuration of the cyst wall in places. (b) Orbital B-scan at 1 month of follow-up demonstrating a marked decrease in cyst size and obscuration of the cyst wall. (c) Orbital B-scan revealing a collapsed cyst with obliteration of the scolex and clear area. (d) Orbital B-scan 6 months following therapy showing increased acoustic signals in the area corresponding to the cyst. (e) Orbital B-scan 9 months following therapy showing resolution of the cyst.*

ethical as preliminary results suggested that the therapy was highly effective. Albendazole was the drug of choice as it has a higher rate of parenchymal cyst removal in neurocysticercosis. We followed a therapeutic regimen of 30 mg/kg body weight for 15 days, together with a low dose of steroid 5–10 mg (depending on the body weight) to suppress the associated inflammatory response.

The results of the study were highly gratifying, showing a cyst elimination rate of up to 95.2%. Our cyst elimination rates are comparable to those reported in a previous series and may be attributed to a high drug availability.[6]

Orbital sonography was used as the primary investigation for diagnosis and follow-up as it was cost-effective and provided a precise location of the cyst and its surroundings. CT was used to confirm the diagnosis. Serology was positive in only one of the patients, confirming our earlier observation that ELISA is not valuable in the diagnosis or orbital myocysticercosis as we obtained negative serology in 7 surgically treated and histologically confirmed cases.

Destruction of the cyst was evident on ultrasonography, suggested by a gradual regression in cyst size, loss of the scolex, collapse of the cyst wall, increasing echoes from the lumen of the cyst and increased echoes from the surrounding tissues suggestive of inflammation. A similar regression pattern was described by Zenteno *et al*.[8] in cerebral cysticercosis.

It is imperative to have a high index of suspicion for the diagnosis of orbital myocysticercosis in patients from endemic areas with subacute proptosis, ocular motility disorders and episodes of inflammation. Oral albendazole therapy seems to be highly efficacious in the management of the condition.

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