CASE REPORT

Multiple brain parenchymal neurocysticercosis with extraocular muscle cysticercosis affecting levator palpebral superioris and superior rectus complex: an unusual association

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SUMMARY

An 8-year-old girl presented to the neurology department with a complaint of insidious onset of left-sided ptosis and restricted elevation of the left eye. A CT scan orbit and brain revealed a ring-enhancing lesion in the levator palpebral superioris (LPS) and superior rectus (SR) muscle complex of the left eye and left parietal and right temporal region. She was started on steroid, followed by albendazole with improvement. The LPS/SR complex is the least common site of involvement among extraocular muscles in ocular cysticercosis. Specially, with brain neurocysticercosis (NCC), it is extremely rare. We report an unusual association of multiple brain NCC with ocular cysticercosis involving LPS and SR muscle.

BACKGROUND

Cysticercosis, the commonest parasitic disease of the nervous system, endemic in Asia, Latin America and Africa,^{1–3} is caused by *Cysticercus cellulosae*, the larval form of the cestode, *Taenia solium*. Other than the brain, the skeletal muscle, subcutaneous tissue and eyes could be affected. Among orbital cysticercosis, extraocular muscle (EOM) involvement is commonest but involvement of the levator palpebral superioris (LPS) and superior rectus (SR) muscle complex is extremely rare.^{4–5} Moreover, brain neurocysticercosis (NCC) and involvement of the LPS+SR complex is a rare association. We report an unusual association of brain NCC with ocular cysticercosis involving the LPS and SR complex.

CASE PRESENTATION

An 8-year-old girl presented with progressively increasing drooping of the left eyelid, double vision and headache for 1 month. Diplopia was for near objects only and aggravated on looking upwards (figure 1A–D). There was no history of associated dimness of vision, painful ocular movements, retro-orbital pain, tearing or redness or proptosis. Headache was mild to moderate in intensity and was occasionally associated with nausea and vomiting. There was no history of loss of consciousness and seizure. Examination revealed left-sided ptosis with restriction of elevation of the left eye (figure 1). Maximum levator function of the left eye was 7 mm, while in the right eye it was 13 mm. Ophthalmoscopy

was normal. The rest of the neurological and systemic examinations were normal.

INVESTIGATIONS

The contrast-enhanced CT (CECT) orbit showed a well-defined, heterogeneously enhancing round to oval soft tissue attenuation lesion measuring approx. 2×1.5 cm×1.2 cm with central area of hypodensity involving the left SR/LPS complex (figure 2). The lesion was abutting the MR muscle, posterior part of the eyeball and superior surface of the optic nerve. It was causing mild axial proptosis of the left eyeball. Mild stranding of the perilesional fat plane is seen. The CECT head revealed a ring-enhancing lesion with a hypodense core. An eccentric hyperdense dot was seen in the left parietal (figure 3A) and right temporal region with grade I perifocal oedema (figure 3B). ELISA for serum antibodies (IgG) against cysticercus was positive. Consent for lumbar puncture was not given.

DIFFERENTIAL DIAGNOSIS

The differentials for this patient could be other cystic lesions of the EOM, like *Echinococcus granulosus* (hydatid cyst), cystic tumours of EOM or myositis of EOM with cystic changes. A hydatid cyst lacks a scolex and does not usually demonstrate intense rim enhancement.

TREATMENT

With a diagnosis of brain NCC with ocular cysticercosis, the child was started with oral dexamethasone 12 mg/day in three divided doses to be tapered in 6 weeks. Cysticidal treatment was started on day 7 with albendazole 300 mg/day, which was continued for 28 days.

OUTCOME AND FOLLOW-UP

Within 10 days of treatment, ptosis of the left upper eyelid markedly improved and ophthalmoplegia improved after 14 days. Patient is still on tapering doses of corticosteroids.

DISCUSSION

In 1830, Semmering reported the first interesting case of ocular cysticercosis. Larva was extracted and demonstrated by Schott in 1836.⁶ In total, 13–46% of infected patients may have ocular or adnexal involvement,² but involvement as low as 1.5–4.8% has been reported by Kaliaperumal *et al*⁷

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Figure 1 (A–D) Photograph showing left-sided ptosis with restriction of elevation of the left eye.

in their series. Children and young adults are mostly affected by an equal sex ratio.⁴ Posterior location is common in the West,⁷ but in our subcontinent, ocular adnexa is the commonest site.⁸ EOM involvement, the commonest, is followed by subconjunctival eyelid, optic nerve, retro-orbital space and lachrymal gland in decreasing order.⁹ Among EOMs, SR, lateral rectus, medial

rectus and the superior oblique muscles are frequently affected, and the presentation depends on the site, size, relation to adjacent structures and the developmental stage of the cyst. Involvement of the LPS/SR complex is quite rare.^{4–5} The most encountered symptoms are restricted ocular motility with diplopia, recurrent pain and redness.¹⁰ Patients may present with proptosis, subconjunctival cyst, atypical optic neuritis, papilloedema, lid nodule, and subretinal and intravitreal cysts and ptosis.^{9–10} With EOM muscle involvement, simultaneous brain involvement is reported in 16% of cases,⁸ but involvement of the LPS/SR complex with brain NCC is scantily reported.⁵ CT/MRI are imaging modalities of choice for diagnosing myocysticercosis involving EOM, while intraocular ones are easily diagnosed by ophthalmoscopy.² Ultrasound and CT have equal ability to detect the scolex.^{2–9} CT may be preferable as head CT can identify NCC.¹¹ As our patient was having headache, we went in for CT. Albendazole with steroids, and excisional biopsy have been studied as a treatment modality. Surgical intervention is not recommended¹² as it could result in injury to EOM. Moreover, surgical excision is technically demanding due to the presence of neurovascular structures in the posterior orbit, degenerating cyst having amorphous consistency and probable attachment of cysts to orbital structures.¹³ Postoperative restrictive myopathy may be a complication of surgical excision due to fibrotic response.¹³ Medical therapy with albendazole and oral steroid is the treatment of choice for the EOM myocysticercosis.^{2–13–15} Its efficacy has been proven in retrospective and non-randomised prospective studies.^{2–11–13–15} Both prednisolone and dexamethasone have been studied. We preferred dexamethasone over prednisolone due to the presence of brain parenchymal NCC. Cyst elimination rates greater than 90% and ocular motility improvement were achieved between 0.5 and 35 months in various studies.^{9–14–15} Some have reported ocular motility limitation after albendazole treatment.^{2–12–16} This may be due to delay in medical treatment and lack of steroids. Steroids, being anti-inflammatory, lessen inflammation from dying cysticerci and

Figure 2 Contrast-enhanced CT orbit showing a well-defined, heterogeneously enhancing round to oval soft tissue attenuation lesion measuring approx. 2×1.5 cm×1.2 cm with central area of hypodensity involving the left superior rectus/levator palpebrae superioris complex. The lesion is abutting the medial rectus muscle, posterior part of the eyeball and superior surface of the optic nerve. It is causing mild axial proptosis of the left eyeball. Mild stranding of the perilesional fat plane is seen.

Figure 3 (A) Contrast-enhanced CT (CECT) of the head showing a ring-enhancing lesion with a hypodense core and an eccentric hyperdense dot in the left parietal cortex. (B) CECT of the head showing a ring-enhancing lesion with a hypodense core and an eccentric hyperdense dot in the right temporal region with perifocal oedema.

thus help in reducing chronic fibrotic sequelae.⁴ Albendazole efficacy in LPS+SR myocysticercosis is most likely similar to extraocular myocysticercosis as a group.² In this case, steroids with albendazole were given, and significant improvement was achieved after 28 days. Steroids with albendazole were also helpful in alleviating symptoms of NCC.

Learning points

- Myocysticercosis could be a cause of acquired ptosis in endemic areas.
- Brain parenchymal involvement may present with cysticercosis of the extraocular muscles.
- Treatment has to be individualised as patients with multiple neurocysticercosis may deteriorate with albendazole.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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