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AN UNUSUAL PRESENTATION OF DISSEMINATED CYSTICERCOSIS

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ABSTRACT
A 48 years old male presented with multiple subcutaneous swellings, recurrent episodes of seizures and headache over six years. The swellings were present over chest, upper limbs, lower limbs, neck and were gradually increasing in size. Ultrasonography revealed well-defined cystic lesion with clear contents and a hyperechoic area suggestive of a scolex in the extraocular muscles of both eyes and in the psoas muscle in abdomen. CT scan orbit showed hypodense mass with a central hyperdensity lesion suggestive of the scolex in the extraocular muscles. Magnetic resonance imaging (MRI) brain showed multiple discrete hypoluscent lesions with surrounding enhanced whitish ring .Histo pathology examination (HPE) showed ectocyst and endocyst, confirming the diagnosis of cysticercosis. The patient was treated with Phenytoin sodium and prednisolone, and albendazole. The patient showed gradual regression of swellings over limbs and subconjunctival space over 6-8 weeks. *Cysticercus cellulosae*, the larval form of the pork tapeworm *Taenia solium*, is the causative organism of cysticercosis. Human act as intermediate host in the life cycle of *T. solium*. *C. cellulosae* may become encysted in various body tissues, usually eyes, brain and subcutaneous tissues, resulting in disseminated cysticercosis. We report an unusual case of disseminated cysticercosis, with simultaneous involvement of the brain, eyes, muscles and subcutaneous tissues. Disseminated cysticercosis is an uncommon manifestation of a common disease. Awareness about the extent of the disease should guide the physician to get a complete work up of the patient before starting the treatment.

CASE PRESENTATION

A 48 years old male had complained of multiple subcutaneous swellings, recurrent episodes of seizures and headache over six years. The swellings were noticed over chest, upper limbs, lower limbs, neck and were gradually increasing in size. There was history of generalised tonic clonic seizures 5-6 times associated with one episode of loss of consciousness in last 7 years. There was no history of loss of vision. There was history of eating pork occasionally. There was no history of chronic cough, chronic diarrhoea, weight loss, decreased appetite or any past history suggestive of diabetes, hypertension, tuberculosis and immunodeficiency.

On examination the subcutaneous swellings were discrete small (0.5-3 cm), round, firm, mobile, non-tender, non-pulsatile with no overlying skin changes (figure 1). A cystic swelling (6 mm in size) was noticed near insertion of superior rectus in both eyes (figure 2). Fundus was normal in both eyes.

Investigations

Blood investigations revealed haemoglobin of 15. 2g/dl, total leucocyte count of 10, 400 cells/cu.mm with polymorphs 53%, lymphocytes 39%, eosinophils 6% and

monocytes 2%. The erythrocyte sedimentation rate was 40 mm/h. Tests for human immunodeficiency virus 1 and 2, hepatitis B surface antigen and hepatitis C virus were negative.

B-scan ocular ultrasonography revealed a well-defined cystic lesion with clear contents and a hyperechoic area suggestive of a scolex in the extraocular muscles of both eyes (figure 3A). Ultrasonography of abdomen revealed a well-defined cystic lesion with clear contents and a hyperechoic area suggestive of a scolex in the psoas muscle (figure 3B). CT scan orbit showed hypodense cystic mass with a central hyperdensity lesion suggestive of the scolex in the extraocular muscles (figure 4). Magnetic resonance imaging (MRI) brain showed multiple discrete hypointense lesions with surrounding whitish ring (figure 5).

FNAC of the subcutaneous swelling over lower limb showed clear, acellular watery fluid. Histo pathology examination (HPE) showed multiple small cysts with eccentric nodule in bilateral psoas muscle, pectoral muscle and other skeletal muscles confirming the diagnosis of cysticercosis(figure 6).

Differential diagnosis

Although diagnosis of cysticercosis was of primary suspicion due to history of seizure and presence of cysts in the eyes, and was confirmed after the imaging, but milliary tuberculosis was one of the differential diagnosis as it can affect multiple organs but lack of characteristic finding in imaging and lack of granuloma in histopathology examination ruled out tuberculosis.

Treatment

As the patient had only extaocular involvement and there was no cyst intraocularly, or in vitrous cavity, there was no need for surgical removal of the cyst. We decided to treat the patient medically. The patient was treated with Phenytoin sodium 4 mg/kg body weight and prednisolone 1 mg/kg body weight, and both were started a week earlier to albendazole. Albendazole was given in dosage of 15 mg/kg body weight for 6 weeks. Phenytoin sodium was continued and predinisolone was tapered slowly after 4 weeks.

Outcome and follow-up

The patient showed gradual regression of swellings over limbs and subconjunctival space over 6-8 weeks.

Fig 1. Cystic swelling in skin (white arrow) over mastoid region.

Fig 2. Cystic swelling near insertion of superior rectus in both eyes.

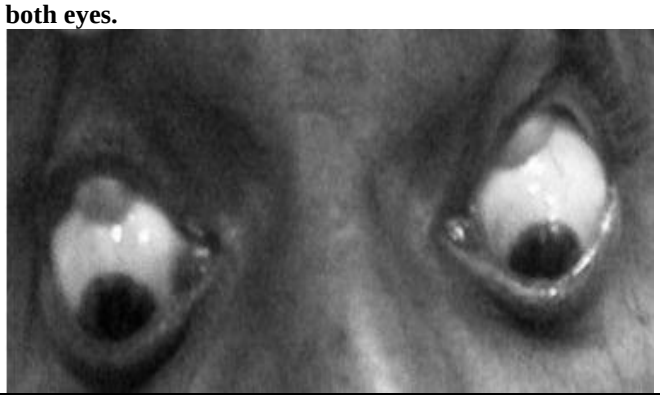


Fig 3(A). Ultra sonic B-scan showing well-defined cystic lesion (long arrow) in extraocular muscle with scolex (small arrow) inside the cyst. (B). Well-defined cystic lesion (long arrow) in psoas muscle with a scolex(small arrow) inside the cyst.

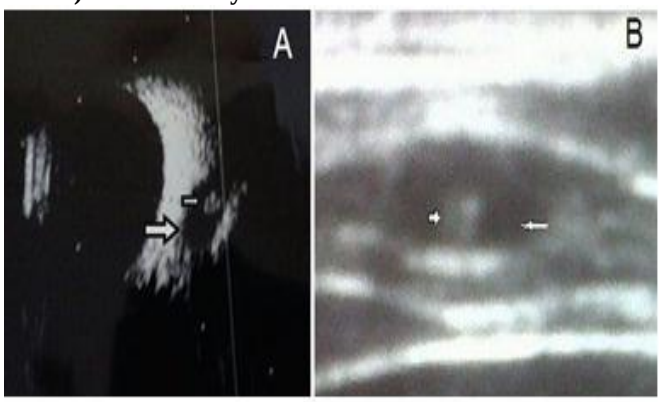


Fig 4. CT scan orbit showing cystic lesion (long arrow) with scolex inside (short arrow) above the eyeball.

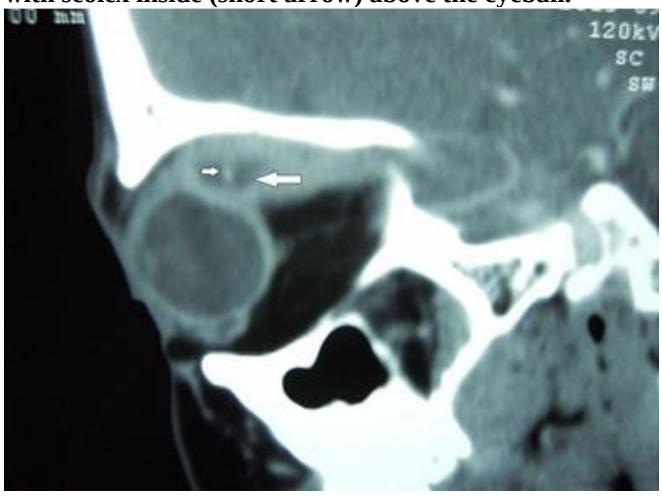


Fig 5. MRI brain showing multiple discrete hypo intense lesions with surrounding enhanced whitish ring.

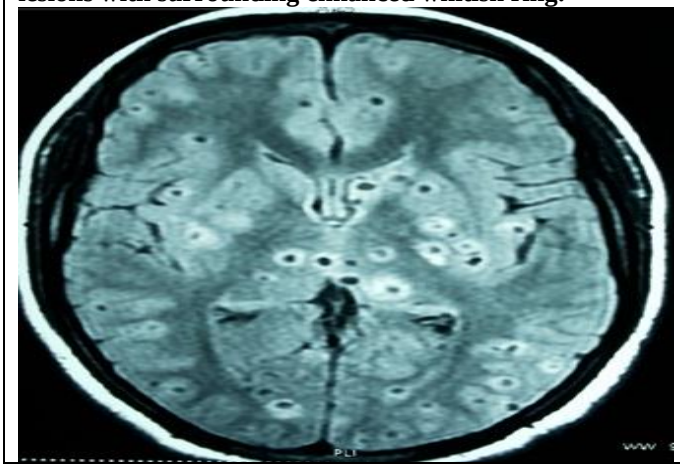
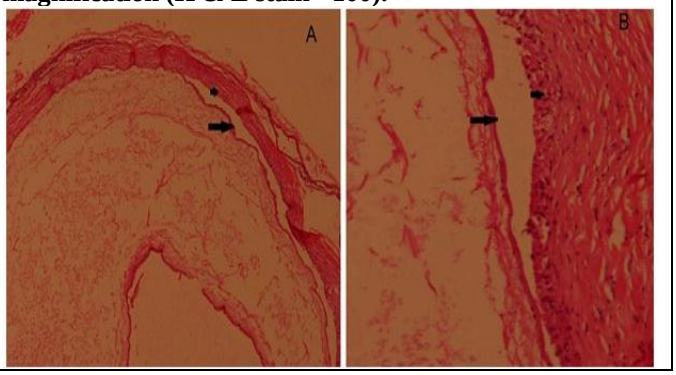


Fig 6. (A) Histology of skin biopsy showing ectocyst (short arrow) and endocyst (long arrow) of *Cysticercus* larva (H & E stain ×40). (B). Ectocyst (short arrow) and endocyst (long arrow) of the same slide under higher magnification (H & E stain ×100).



DISCUSSION

Cysticercosis is caused by encystment of the larvae (*Cysticercus cellulosae*) of the tapeworm *T. solium*, in tissues. Usually human is the definitive host and gets infested by the ingestion of undercooked pork meat containing resting stage larva (*C. cellulosae*) of the parasite. On ingestion these larvas mature into adult worms in the small intestine of human and shed their proglottids loaded with thousands of eggs in faeces, from where it is uptaken by pigs (intermediate host). In the pig's stomach and small intestine the egg shell dissolves and the larva hatches out, attaches to intestinal mucosa with hooklets, perforates it, and passes in hepatic-portal system and with blood carried to striated muscles or other tissues, where it develops into a resting stage larva. When food (usually unwashed/ under washed raw vegetables and fruits) or water contaminated with the parasite eggs is consumed by human they act as the intermediate host (hetero-infection). They may acquire infection (Cysticercosis) through external auto-infection or internal auto-infection (reverse peristalsis). The most common form of systemic involvement is neurocysticercosis [1]. The organs most commonly affected are subcutaneous tissues, skeletal muscles, the lungs, brain, eyes, liver and, occasionally, the heart. The main features of disseminated cysticercosis include intractable epilepsy, dementia, enlargement of muscles, subcutaneous and lingual nodules and a relative absence of focal neurological signs or obviously raised intracranial pressure, at least until late in the disease [2,3]. Pseudo-hypertrophy of the muscles is the most common presentation of disseminated cysticercosis, followed by palpable nodules and seizures [4].

Ocular and adnexal cysticercosis represents 13% to 46% of systemic disease [5]. Ocular cysticercosis can involve eyelid, orbit, sub-conjunctival space, anterior segment or posterior segment (sub-retinal and intra-vitreal). Conjunctival involvement is usually in the form of a painless or painful yellowish, nodular subconjunctival mass with surrounding conjunctival congestion. Cysticercosis of extraocular muscle usually presents as recurrent pain, redness, proptosis, ocular motility restriction, diplopia and ptosis [6, 7]. Optic nerve compression by the cyst may cause decreased vision, disc edema and painful ocular motility [8]. Central nervous system involvement with *T. solium* cysts, neurocysticercosis, is a pleomorphic disease whose clinical manifestations vary with the number, size, location and stages of *Cysticerci* as well as the intensity of the host's immune response [9]. Common manifestations include epilepsy, focal neurological signs, intracranial hypertension, cognitive decline, cerebellar ataxia, symptoms of hydrocephalus and psychiatric disorders. A set of diagnostic criteria based on neuroimaging studies, serological tests, clinical presentation and exposure history has been proposed by Del Brutto *et al*, [10]. CT and MRI remain the most effective means of diagnosis. Sensitivity of serological tests tends to be high for patients with multiple cysts (94%) but substantially lower for patients with a single cyst or calcified cysts (28%) [11]. Serological tests used for the specific diagnosis of cysticercosis are indirect hemagglutination, indirect immunofluorescence, and immunoelectrophoresis such as ELISA specific serology [12]. B-scan ocular ultrasonography reveals a well-defined cystic lesion with clear contents and a hyperechoic area suggestive of a scolex [13]. The scolex shows a high amplitude spike due to presence of calcareous corpuscles, [14]. Ocular ultrasonography is a useful tool for diagnosis and monitoring of the cyst during treatment. CT scan of the orbits is a reliable technique to help establish a diagnosis of ocular cysticercosis. Concurrent neurocysticercosis may be present and should be excluded [15]. CT is more sensitive than MRI in detecting small calcifications. However, MRI is more sensitive than CT as it identifies scolex and the cyst.

Management of disseminated cysticercosis is symptomatic (antiepileptic and steroids), surgical (removal of cysts and ventriculoperitoneal shunt) and with cysticidal drugs [15]. If intraocular cyst is detected (e.g. on indirect

ophthalmoscopy) cysticidal drugs are contraindicated as they can lead to sterile endophthalmitis. Surgical excision (e.g. vitrectomy with cyst aspiration and photocoagulation) is indicated as such cases. As there was no intraocular cyst in vitreous cavity in our case, we managed the case with medical management. It is always advised to use oral steroid throughout the treatment, starting at high dose (1mg/kg/day) 3 days before introducing cysticidal drug and tapering slowly over 4-6 weeks. This prevents or controls sterile inflammation which occurs during lysis of cyst. Anti-epileptics cover should be given in case of intracranial cysticercosis or history of seizure [9]. Modality of treatment, medical vs surgical, depends upon clinical response to medical management and site of involvement [15]. Cyst excision can be advisable in cases of sub-conjunctival, lacrimal gland or rarely lacrimal sac cysticercosis [16].

CONCLUSION

- Thorough clinical examination to be done if there are multiple sites of involvements in cysticercosis.
- Indirect ophthalmoscopy, ultrasonograpy of eye and orbit along with MRI or CT brain and orbit should be done in all cases of cysticercosis.
- Presence of intraocular cyst should be ruled out before starting cysticidal drugs.
- Neurology evaluation should be done in case of intra-cranial cyst or neurological symptoms, before starting cysticidal drugs.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

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