

## Submacular cysticercosis in two cases: Course and outcome

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Ocular manifestations due to cysticercosis may involve the vitreous cavity or subretinal space in the posterior segment of the eye. Management of subretinal cysticercosis is challenging, especially if it involves the submacular region. Removal of submacular cysticercosis (SMC) requires utmost care and competent expertise. In recent times, modern vitreo-retinal equipment has improved the outcome of SMC. On the contrary, untreated SMC run a high risk of spontaneous rupture, liberation of toxin from dying worm, and intense intraocular inflammation threatening vision eventually. In the present report, two cases of SMC were discussed who presented to us within a span of 1 week. We described the course of disease and final outcome in these two patients (one underwent surgical removal while other denied surgery) highlighting the urgent need of cyst removal in such scenarios.

**Key words:** Cysticercosis, pars-plana vitrectomy, scolex, submacular

Cysticercosis, caused by the encystment of the larvae of *Taenia Solium*, is one of the commonest helminthic infestations in endemic countries<sup>[1]</sup> and can manifest with both intraocular and extraocular involvement. We report two cases of unilateral submacular cysticercosis (SMC), presented to us 1 week apart. One of them underwent transvitreal SMC removal immediately and achieved stable vision. The other patient did not undergo surgery and landed up in total retinal detachment (RD) with poor visual outcome. This report describes the procedure of SMC removal in the first patient and also highlights the need of immediate surgical removal of SMC.

### Case Report

Examination of a 55-year-old male revealed a single undulating translucent cyst with an intra-cystic dense white lesion at submacular space [Fig. 1a and b] with surrounding area of

retinal pigment epithelium (RPE) alteration in the left eye (OS). Best-corrected visual acuity (BCVA) was 6/6 in the right eye (OD) and 3/60 in OS. Ultrasonography-B (USG-B) scan of OS confirmed the diagnosis of SMC. Optical coherence tomography (OCT) of OS [Fig. 1c] demonstrated a well-defined subretinal hyporeflective cyst with hyper-reflective wall. Fundus autofluorescence showed hyperautofluorescent area [Fig. 1d] corresponding to the SMC. Computed tomography (CT) scan of brain and orbit did not show concomitant neurocysticercosis or any extraocular involvement. He underwent 25-gauge pars-plana vitrectomy (PPV) through a temporal approach due to deep-seated eyes. After performing triamcinolone-assisted hyaloid removal, retinotomy was made over a suitable location ensuring cyst integrity. A 25-gauge silicone tip-assisted suction (STAS) induced delivery of the cyst from the submacular space into the vitreous cavity was completed in-toto (Video1: surgical video demonstrating key steps of SMC removal). The cyst was rapidly engulfed with the help of cutter thereafter. Low intensity endolaser was done around the retinotomy after fluid-air exchange. Silicone oil tamponade was performed as the patient belonged to a high altitude region, followed by a face-down position for 2 weeks. Postoperatively, BCVA remained at 1/60 due to subretinal fibrosis although retina was attached [Fig. 2a]. Silicone oil removal was performed after 3 months without any adverse events. At 6 months follow-up, his BCVA remained stable at 3/60; fundus [Fig. 2b] showed dense scar at the macular region.

Our second patient, a 35-year-old lady also presented with live SMC in OS [Fig. 3a] with BCVA of 6/24. Examination of OD was unremarkable. In view of co-existent but unrelated acute mastoiditis, clearance for ocular surgery could not be obtained immediately. In subsequent 4 weeks, a rapid increase in the size of the cyst was noted by fundus photography [Fig. 3a-c] and her BCVA declined to 6/60. After 4 weeks, although she recovered from the episode of acute mastoiditis, she refused to consent for ocular surgery despite explaining the urgency repeatedly. She was lost to follow-up thereafter. She came back after 6 months with severe painful diminution of vision in OS. Examination of OS revealed vision of light perception, raised intraocular pressure of 40 mm of Hg, epithelial corneal edema, mild anterior segment inflammation, and a leukocoria [Fig. 4a and b]. Dilated fundus examination and USG-B scan [Fig. 4c] revealed narrow-funnel RD. She was explained about the poor prognosis in OS and was managed conservatively.

### Discussion

Cysticercosis of the posterior segment of the eye can involve vitreous cavity or subretinal space.<sup>[2]</sup> It lodges in the subretinal space after entering the eye through choroidal circulation. It can

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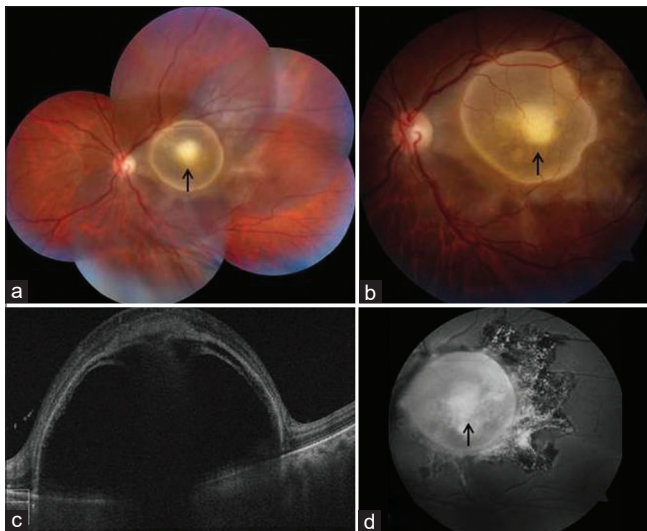
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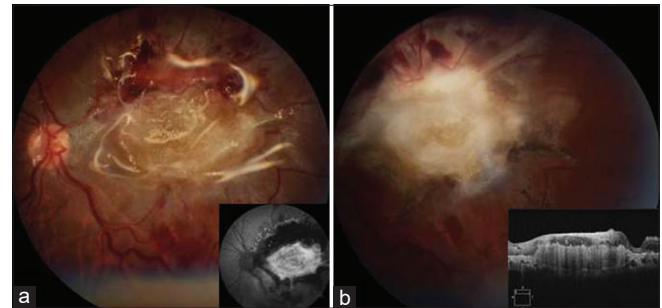
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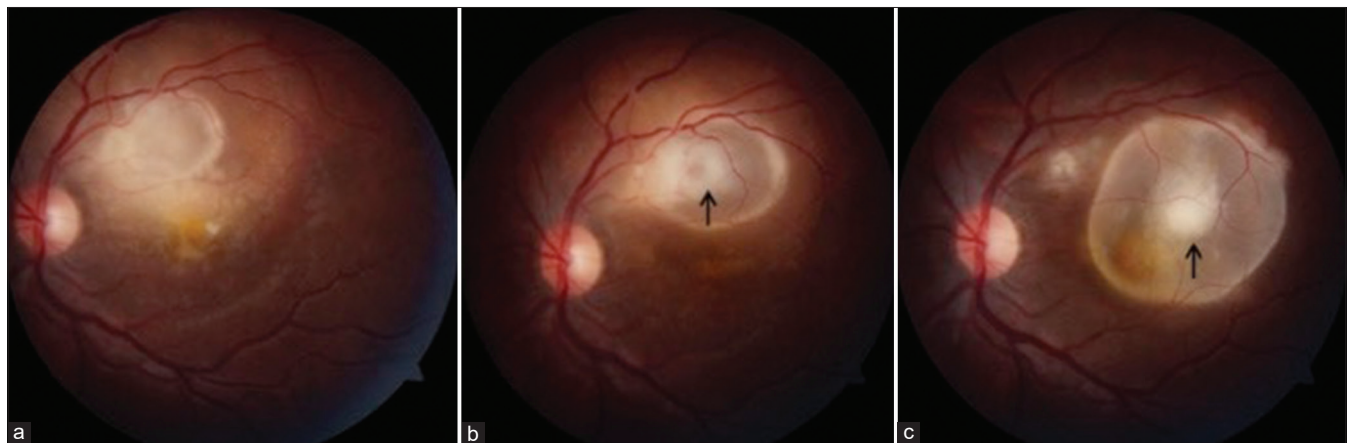


**Figure 1:** Montage (a) and posterior pole (b) fundus photo (FP) of left eye (OS) of first patient showing a submacular translucent cystic lesion suggestive of submacular cysticercosis (SMC) with a surrounding area of altered RPE. An intra-lesional white structure (black arrow) suggestive of scolex was also noted. OCT (c) showing a well-demarcated hyporeflective cystic lesion with hyper-reflective wall and a small area of neurosensory detachment nasally. Fundus AF (d) shows hyper-AF corresponding to the area of SMC with a surrounding area of stippled hypo and hyper-AF

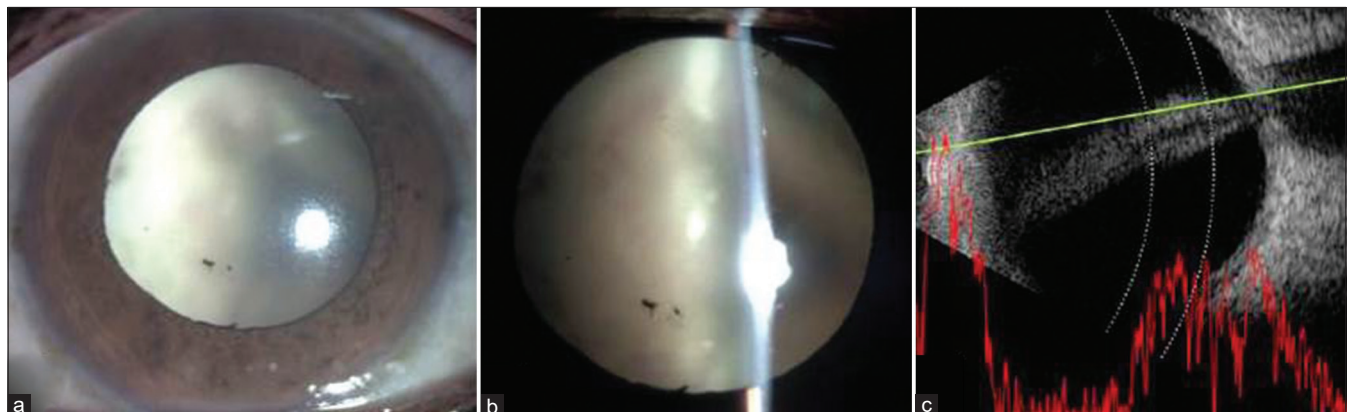
further migrate into the vitreous cavity through a retinal hole, which is generally obscured by an inflammatory chorioretinal scar.<sup>[3]</sup> Left eye predilection as seen in our cases have also been reported by few authors, probably due to more direct course of the left carotid artery from the aorta.<sup>[4,5]</sup> The submacular space is more predisposed due to high vascular supply.<sup>[6]</sup> Few authors at past reported good outcome of intraocular cysticercosis without surgical removal and advocated conservative treatment to avoid



**Figure 2:** (a) FP of OS after PPV and oil tamponade in first patient showing oil-filled globe with submacular fibrosis and few retinal hemorrhages along supero-temporal arcade. Inset shows AF image depicting central hyper-AF with surrounding hypo-AF. (b) FP of OS after silicone oil removal showing persistent submacular scar; inset shows OCT images depicting hyper-reflective subfoveal scar extending nasally



**Figure 3:** Serial FP (a-c) of OS of the second patient showing SMC at presentation (a); after two weeks (b) and after four weeks (c). Gradual increase in size of SMC was noted and intra-lesional white structure suggestive of scolex (black arrow) was also seen



**Figure 4:** Anterior segment photograph of OS of second patient in diffuse illumination (a) and slit-illumination (b) after six months showing diffuse corneal edema and leukocoria. USG-B scan (c) shows narrow funnel RD

risk of iatrogenic cyst rupture.<sup>[7,8]</sup> However, with advancement of vitreo-retinal surgeries in modern era, most surgeons advocate early surgical removal and demonstrated the safety as well.<sup>[3]</sup> Transscleral approach of cyst removal is advocated for cysts located anterior to equator, while transvitreal approach is employed for intravitreal and subretinal cysts posterior to the equator.<sup>[3]</sup> Although it is a standard practice to deliver the cyst in-toto, no significant difference in outcome was reported when the cysts were removed in-toto versus cysts engulfed rapidly in vitreous cavity with cutter.<sup>[3]</sup> However, complete wash out of all debris with balanced salt solution must be ensured to avoid any residual cyst-material and consequent perpetual inflammation. The other key step of surgery is ensuring complete removal of posterior hyaloid to avoid post-operative contraction of vitreous.

Antihelminthic drugs were not used in either of our two patients as sole medical treatment can lead to severe intraocular inflammation arising from the toxins of the dying worms. Spontaneously ruptured cysts can also incite intense inflammation as seen in our second patient. The submacular cysts carry a worse prognosis as compared to other subretinal locations, which can be attributed to physical damage of photoreceptors, underlying RPE atrophy or inflammatory effects from liberated toxins.<sup>[6,9]</sup> Poor functional outcome has also been reported in eyes with RD more than two quadrants and preoperative vision of <5/200.<sup>[3]</sup> In a large series of intraocular cysticercosis cases, retinal reattachment was seen in 86.6% cases and ambulatory vision (>5/200) was regained in 67.5% cases.<sup>[3]</sup>

## Conclusion

In conclusion, we demonstrated sequential follow-up of two patients with SMC, who presented simultaneously with almost identical features but had different outcomes.

High index of suspicion, early diagnosis, and timely intervention through precise surgical approach can achieve optimal outcome in SMC, although the final visual acuity may depend upon multiple factors.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

## References

1. Lech J. Ocular cysticercosis. *Am J Ophthalmol* 1949;32:523-48.
2. Wender JD, Rathinam SR, Shaw RE, Cunningham ET. Intraocular cysticercosis: Case series and comprehensive review of the literature. *Ocular Immunol Inflammation* 2011;19:240-5.
3. Sharma T, Sinha S, Shah N, Gopal L, Shanmugam MP, Bhende P, *et al.* Intraocular cysticercosis: Clinical characteristics and visual outcome after vitreoretinal surgery. *Ophthalmology* 2003;110:996-1004.
4. Atul K, Kumar TH, Mallika G, Sandip M. Socio-demographic trends in ocular cysticercosis. *Acta Ophthalmol Scand* 1995;73:438-41.
5. Malik SRK, Gupta AK, Choudhry S. Ocular cysticercosis. *Am J Ophthalmol* 1968;66:1168-71.
6. Kumar V, Surve A, Kumar P, Sharma A, Azad S. Submacular cysticercosis. *Eur J Ophthalmol*. 2019 April 7; 1120672119841542. doi:10.1177/1120672119841542.
7. Messner KH, Kammerer WS. Intraocular cysticercosis. *Arch Ophthalmol* 1979;97:1103-5.
8. Lim WK, Chee SP. Nonsurgical management of subretinal cysticercosis. *Retina* 2004;24:469-71.
9. Wani VB, Kumar N, Uboweja AK, Kazem MA. A case of submacular cysticercosis treated by pars plana vitrectomy in Kuwait. *Oman J Ophthalmol* 2014;7:144-6.