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Case report

"Pacman" invasion of the retina: Two cases of ophthalmomyiasis interna posterior

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ABSTRACT

Purpose: To discuss two striking cases of ophthalmomyiasis interna posterior, in which the larval stage of a botfly is found in the posterior segment.

Observations: In the first case, the subretinal maggot is alive and found to be migrating under the retina. The maggot was lasered in the office and killed. In the second case, a dead maggot was discovered in the subretinal space in a child, after it had caused significant subretinal scarring and permanent vision loss.

Conclusions and Importance: Ophthalmomyiasis is a rare condition that can often be unrecognized and result in permanent vision loss. Early diagnosis and photocoagulation of the larva (if alive) can halt progression of vision loss in these cases.

1. Introduction

Ophthalmomyiasis interna is invasion of the globe by larvae of any species of the botfly. Ophthalmomyiasis externa involves only the external ocular structures.[1] Ophthalmomyiasis interna is of two types-anterior (involving the anterior segment) or posterior (involving the subretinal space or vitreous cavity). *Dermatobia hominis,* endemic to tropical or subtropical areas, and *Oestrus ovis* (sheep bot fly) cause most cases of ophthalmomyiasis.[2] Ophthalmomyiasis externa is more common.[2] From late June to early September, the fly lays eggs directly on the guard hairs of the caribou.[2,3] Once deposited, eggs hatch into larvae that penetrate skin.[3] They move subcutaneously to reach the animal's dorsal region, where they cut breathing holes and are encased within granulomatous cysts, termed warbles. There they develop for 9–11 months; in May or June of the following year, they leave the animal and pupate on the ground.[3] Mated adult females are capable of long flights (≈900 km) in search of suitable hosts.[3]

The pathophysiology of human ophthalmomyiasis is not known. Eyebrows and eyelashes have been suggested as possible targets for oviposition.[3] Oviposition on human scalp hair has been achieved experimentally and could be the preferential site in humans.[3] An alternative explanation is transfer of the larvae directly from the guard hairs of the caribou to the human eye or skin through close contact with animal pelts. The parasite does not appear to complete its life cycle in humans.[1,2]

We present 2 cases of ophthalmomyiasis interna posterior. In both cases the larva was discovered in the subretinal space. The first case involves a maggot that was alive, and the second case involves a dead maggot in the subretinal space. Both cases involve significant scarring and retinal atrophy.

2. Findings

2.1. Case 1

A 55-year-old white male presented with complaints of "lines" in his vision, first noted the evening before. These lines doubled in number, causing blurred vision and photopsias. He reported working in mulch about 2 weeks prior to the onset of symptoms. His general medical health was unremarkable except for mild hypercholesterolemia.

Spectacle corrected visual acuity was 20/40 in the right eye and 20/30 in the left. Sensorimotor and anterior segment exams were within normal limits. Funduscopic exam of the right eye showed multiple circum-linear tracks that coursed throughout the posterior pole and into the periphery 360°. A subretinal maggot was appreciated superior-temporal to the fovea with associated mild subfoveal hemorrhage. During examination the maggot was noted to be alive and was actively migrating. Multimodal imaging, including fundus photography (Figs. 1 and 3), fluorescein angiography (FA) (Fig. 2), optical coherence tomography (OCT) (Fig. 5) and autofluorescence was performed. Once it was outside the foveal area, laser photocoagulation was performed and the maggot was killed (Fig. 4). The following laser settings were used-Argon yellow laser with a power of 350 mW, duration of 0.2 seconds and spot size of 200 μm. A total of 330 laser spots were placed to cover the entire maggot with a 100-μm border. The physician watched to confirm that maggot was no longer mobile.

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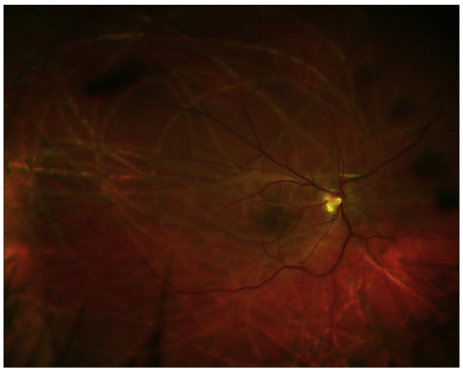

Fig. 1. Color fundus photograph (case 1) of maggot (superior to disc) heading towards the periphery. Multiple circumlinear tracks are seen of varying pigmentation. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

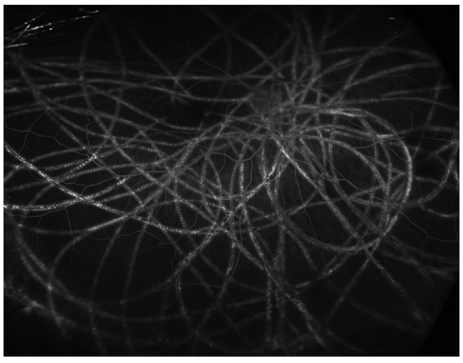

Fig. 2. Late phase fluorescein angiogram (case 1) clearly demonstrating the migratory tracks of the larva.

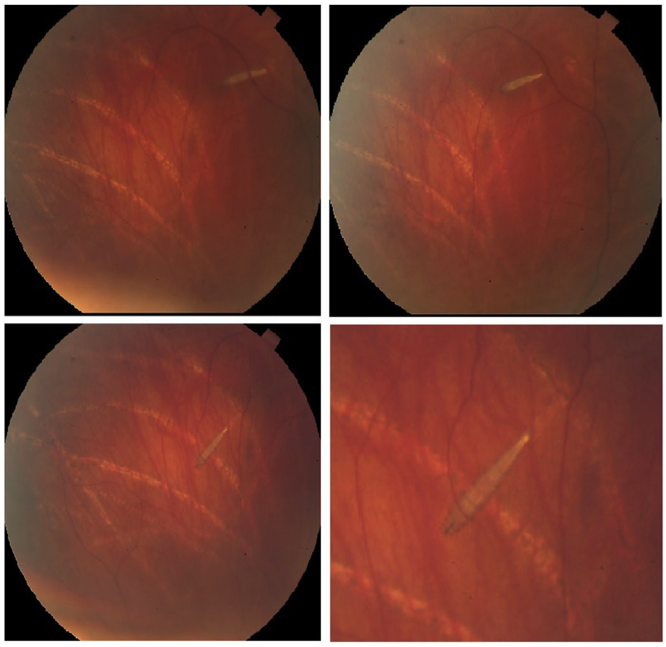

Fig. 3. 3a, 3b and 3c- Consecutive color fundus photographs (3a, 3b, 3c) in case 1 showing clear movement of the larva. The larva demonstrates fusiform shape with multiple striations. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

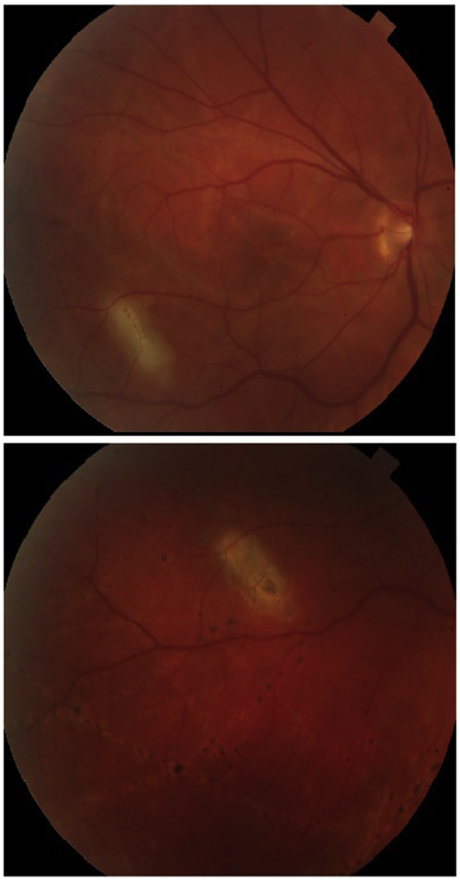

Fig. 4. 4a and 4b – Color fundus photograph of larva (case 1) immediately following laser photocoagulation and 6 months following laser showing skeletonization of the larva. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

The patient had a benign postoperative course and visual acuity 8 months later had returned to 20/20 with mild residual visual distortion. On OCT there were minimal changes noted in the ellipsoid zone under the fovea. Color fundus photography showed the ablated and skeletonized maggot and the tracks had pigmented.

2.2. Case 2

A 6 year-old Caucasian female presented with exotropia and decreased vision in her left eye for a few weeks and was referred for a possible retinal detachment. She could only see hand movements in the left eye, and the right eye was 20/20. An afferent pupillary defect was noted on examination and confrontation visual fields were severely diminished in the left eye. Intraocular pressure was normal. Ophthalmoscopy revealed significant subretinal linear scarring and diffuse retinal pigment epithelium (RPE) atrophy in the left eye. Retinal vessels were severely attenuated. A dead maggot was noted in the subretinal space along the equator in the superior-temporal quadrant, and was not moving (Fig. 6). Examination of the fellow eye was unremarkable. Follow-up exam showed that the dead larva had not changed its position. OCT of the macula showed significant RPE and ellipsoid zone loss (Fig. 7).

3. Discussion

Ophthalmomyiasis interna posterior is an extremely rare condition in which a maggot is found in the posterior segment. Maggots occasionally burrow through the conjunctiva and eye wall and enter the subretinal space or vitreous cavity.

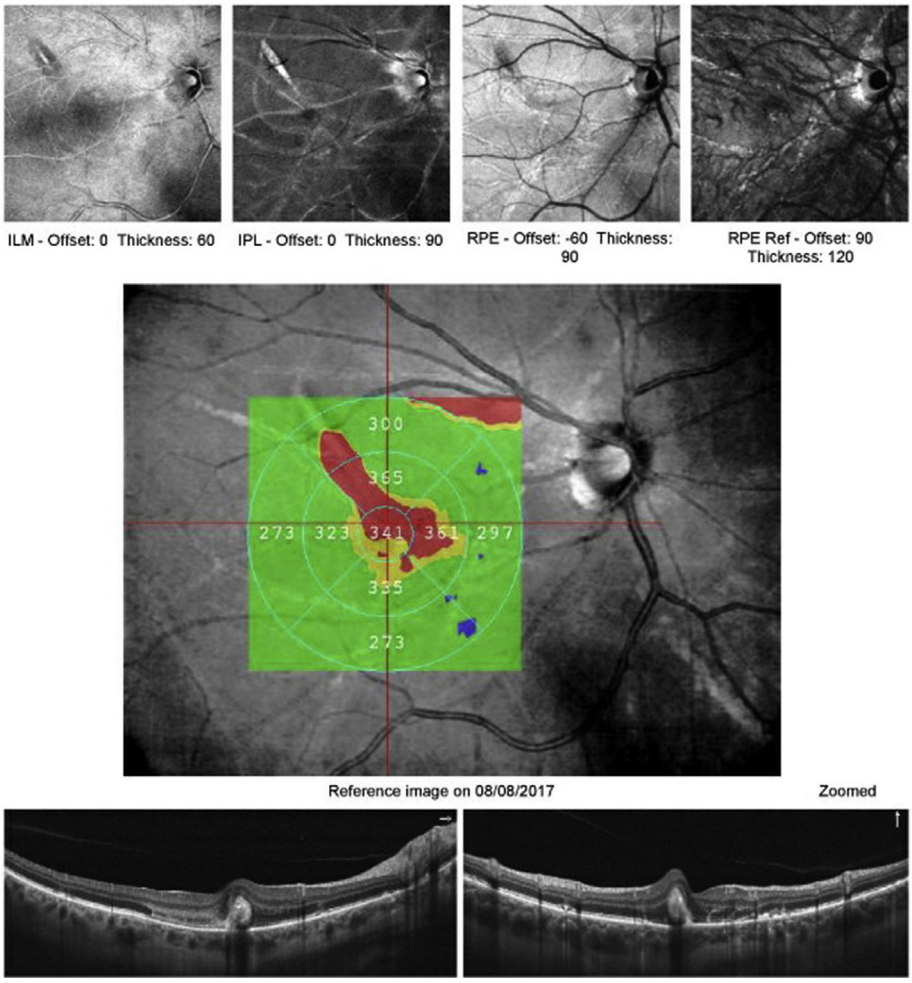

Fig. 5. OCT near infra-red image of larva (case 1) just exiting foveal region at 10 o'clock and an OCT B-scan showing significant loss of ellipsoid zone due to the migrating larva. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

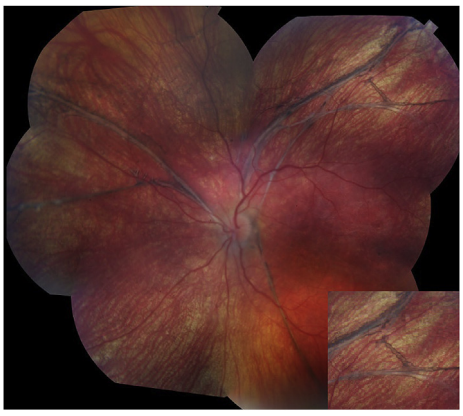

Fig. 6. (case 2)-Color fundus photo (montage) of left eye (6A) showing significant subretinal scarring and RPE dropout throughout the posterior pole. The dead maggot can be seen in the region of the equator in the superior-temporal quadrant. A magnified view of the dead maggot is shown (6B).. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

Although this condition is extremely rare, previous studies have reported that moderate or severe vision loss appeared to occur with both *Hypoderma* and *Cuterebra* infestations (53.3% vs 14.3%).[4] However, in their study, the difference in frequency was not statistically significant ($p = 0.08$).

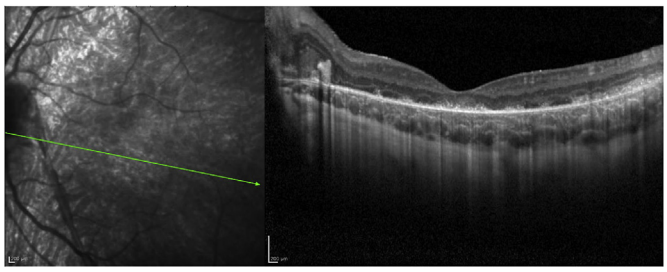

Fig. 7. OCT in case 2 confirmed significant RPE and ellipsoid zone atrophy in the macula.

There is no definitive proven treatment of a subretinal maggot, but some available options involve observation (if dead), or laser photocoagulation (if living) once it has migrated away from the macular region. If the maggot is found in the vitreous cavity, it can lead to significant inflammation and should be removed in-toto during vitrectomy.

Photocoagulation appeared to produce better outcomes (80% good outcomes) than no intervention (73.7% good outcomes) or surgical removal/vitrectomy (41.2% good outcomes).[3] Often times by the time patients seek treatment, damage to ocular structures usually has already occurred. Recent cases have been treated with photocoagulation or vitrectomy and intraocular steroid administration. Although no specific intervention is associated with better outcomes, given the difficulty in studying outcomes in this rare condition and the success of

this therapy for other foreign bodies in the eye, this course of action is reasonable. Ivermectin as medical therapy for ophthalmomyiasis interna has been reported to be effective as therapy and prophylaxis for bot fly infestation of livestock.[5] There is some evidence of benefit in human cases of ophthalmomyiasis as well.[6]

4. Conclusions

Although ophthalmomyiasis caused by botflies is rarely reported, diagnoses may increase as a result of increasing population in northern latitudes, encroachment into habitats with natural hosts like caribou, and better access to ophthalmologists. The treatment of choice for a maggot in the subretinal space involves laser photocoagulation (if alive) or observation (if dead). Vitrectomy with larva removal and co-administration of intraocular steroids may be an option if the larva has migrated into the vitreous cavity. Early diagnosis and prompt treatment is most likely to reduce the chances of vision loss, like in case 1. However, most cases are diagnosed late in the disease process and already have permanent vision impairment.

Patient consent

Written consent to publish this case has not been obtained. This report does not contain any personal identifying information.

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Conflicts of interest

None of the authors have any conflicts of interests in relationship to this case series.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ajoc.2019.100491.

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