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Case Series

The hidden threat by larval invasion of the eye: A case series of ocular myiasis

Padmashri VV[1], Bindu Malini Mudduveerappa[1]*, Kirthana Achar[1], Kavitha Bandravi[1], Soumya Harapanahalli Venkatesh[2], Amrutha Aravindakshan[1]

[1]Dept. of Ophthamology, JSS Medical College and Hospital, Mysore, Karnataka, India
[2]Dept. of Medical Retina and Ophthalmic Education, JSS Medical College and Hospital, Mysore, Karnataka, India

Abstract

Purpose: Ophthalmomyiasis is a zoonotic infestation of ocular or orbital tissue by fly larvae. Patients present with varied manifestation from mild symptoms to extensive involvement of ocular structures. Here we report three cases of ophthalmomyiasis, aiming for better understanding of clinical features and management of this condition.

Material and Methods: It is a retrospective case series where patients with ocular or orbital myiasis over past 2 years were included. After obtaining ethical committee clearance, detailed history on patient's demography like age, gender, occupation, associated systemic conditions and previous eye problems was noted. Patient's symptoms were documented and ophthalmic evaluation details were recorded which included best corrected visual acuity, anterior and posterior segment slit lamp findings. Data was also collected on medical and surgical management of the cases.

Results: We describe three cases of ocular myiasis. Two were females and one was a male patient. The age of the patients ranged between 20 to 90 years. Two cases lived in rural area and were involved in both agricultural activity and livestock rearing. Unilateral presentation was observed in all the three cases. Two patients were found to have external ophthalmomyiasis who underwent larval removal and subsequently received medical management. In the third case of orbital myiasis further investigation through biopsy confirmed squamous cell carcinoma. Due to delayed presentation and despite rigorous medical management, patient required exenteration.

Conclusion: Ophthalmomyiasis is a relatively uncommon condition that mimics conjunctivitis. Prompt intervention through proper history taking and meticulous examination, is essential to manage external ophthalmomyiasis effectively, thereby preventing its progression to internal and orbital myiasis.

Keywords: Maggots, sheep botfly, External ophthalmomyiasis, Internal ophthalmomyiasis, Orbital myiasis.

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1. Introduction

Ophthalmomyiasis, a zoonosis, is an infestation of ocular or orbital tissue by fly larvae.[1] It is a rare condition in humans and seen in people residing in rural areas who are living close to livestock.[1] Ophthalmomyiasis occurs in three forms namely external, internal and orbital ophthalmomyiasis. External ophthalmomyiasis is the most common form and the orbital type remains the most destructive form.[2] Here we describe infrequently encountered cases of external and orbital myiasis.

2. Case Presentation

2.1. History of case 1

A 20-year-old female student sought medical attention for sudden onset of left eye pain, redness, foreign body sensation and excessive watering that began a day prior. She had experienced similar episodes of redness in the past, which were previously treated as allergic conjunctivitis leading to her delay in presentation for a day. In addition to above information, she informed about a recent visit to the botanical garden.

Corresponding author: Bindu Malini Mudduveerappa
Email: bindumalini.mh@gmail.com

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2.2. History of case 2

A 42-year-old male farmer presented with a two-day history of intense left eye itching, redness, foreign body sensation and excessive watering to the outpatient department.

Neither patient had a history of ocular trauma or any significant systemic comorbidities.

2.3. Presentation of case 1 and 2

General and Systemic examination were unremarkable with no signs of immunosuppression. On ophthalmic examination, best corrected visual acuity (BCVA) in both eyes were 6/6, N6 in both the cases. Both had similar ocular findings. Detailed examination revealed mild eyelid edema with diffuse conjunctival congestion, excessive tearing and scanty mucoid discharge. Slit lamp examination of left eye showed 12-15 tiny spindle shaped worm like organisms about 1-2 mm in size with dark heads actively motile over ocular surface.(**Figure 1**)

Under topical anesthesia and slit lamp visualization, the larvae were carefully extracted using fine forceps with double eversion of upper eyelids. Patients reported immediate symptom relief post removal. The collected larvae were then sent to microbiology department in normal saline for analysis. Further assessment of anterior and posterior segment of the eye and extraocular movements were within normal limits.

Detailed ocular examination of right eye was within normal limits.

Microscopic examination highlighted a larva with a pair of sharp, dark brown oral hooks, a prominent cephalopharyngeal skeleton, 11 body segments adorned with tufts of brown hooks and two caudal tubercles each bearing ten curved spines. These features confirmed the identification of larva as the first instar of *Oestrus ovis*, Family *Oestridae*, commonly known as sheep botfly.(**Figure 2**)

Diagnosis of External Ophthalmomyiasis by *Oestrus Ovis* was made in both cases.

Patients were started on topical moxifloxacin with dexamethasone eye drops, one drop hourly for left eye and tapered gradually. Both were followed up every day for one week, later twice weekly for one month. On each follow up patients' signs and symptoms had improved and no other larva was found on detailed examination.

2.4. Presentation of case 3

An 86-year-old female who was bed ridden, lived alone came from a rural area with limited socioeconomic resource presented to ophthalmology out-patient department with complaints of bleeding mass in left eye from past 2 days. She also gave history of swelling since a month and gradual diminution of vision. She gave history of frequent visits to agricultural field previously. There was no history of trauma, previous ocular surgeries and systemic co morbidities. Patient was debilitated due to severe pain and intense itching and had multiple episodes of nausea and vomiting with poor appetite.

A comprehensive ophthalmic examination of left eye revealed no light perception, severe proptosis and a proliferative, fungating mass in the eyelids, infested with numerous live maggots. The ocular surface was compromised with loss of ocular adnexa, conjunctiva and a lustreless cornea. (**Figure 3**A,B). Extra ocular movements were limited in all the directions. Rest of the ocular details could not be appreciated. The fellow eye had an immature cataract with normal fundus findings and achieved a BCVA of 6/12, N8. Systemic examination was unremarkable. Immediate larvae extraction was done under topical anaesthesia and a tissue sample was taken for histopathological analysis and frozen section. Nasal endoscopy and otoscopy performed by and otolaryngologist showed no evidence of myiasis in paranasal sinuses, nasal cavity and auditory canal.

Patient was started on intravenous cefotaxime 1 gm twice daily and intravenous metronidazole 500mg thrice daily which was continued for 5 days. Computed tomography imaging revealed left orbital myiasis demonstrating soft tissue nodules within fat component, with fat stranding and bulky extra ocular muscles and no intracranial and sinus involvement.

Due to extensive maggot infestation of the left eye, patient was planned for further exploration under general anaesthesia. Frozen section report showed squamous cell carcinoma of the tissue. Extensive conjunctival and tenon's capsule tissue loss was noted intraoperatively. Extra ocular muscles were released to explore the orbit which revealed maggots in the orbital fat with tissue destruction extending up to the optic foramen. Patient's attendees were explained about the condition and consent was taken for lid sparing exenteration. All necrotic tissue and numerous live maggots around 30-40 were removed meticulously. (**Figure 4**) Left eyeball and periorbital tissue were sent for histopathological examination which confirmed frozen section findings. Betadine-soaked gauze wound packing was done. There was no regional lymphadenopathy. The systemic examination pertinent to squamous cell carcinoma showed no abnormalities.

Postoperatively wound was closely looked for infections and possibly missed larvae. On Day 1 and day 2, a couple of non-motile maggots were extracted. Patient recovered well and was discharged uneventfully. One week postoperatively, the socket was healthy and free of maggots, with visible granulation tissue. The lid wound was clean but two sutures had given away and required re-suturing. Patient was on oral and topical antibiotics with oral NSAIDs for 5 days. Due to financial constraints patient wasn't willing for ocular

prosthesis. She was followed up for 2 months regularly and eventually lost to follow-up.

The histopathological examination of the eyeball showed two irregular grey white areas within the cavity measuring 1.8×0.8 cm and 1.8×0.5 cm. The microscopic examination of above tissue revealed sheets of pleomorphic squamous epithelial cells with clusters having large nucleus, prominent nucleoli and mitosis. Intervening stroma showed inflammatory cell infiltrates and necrosis.

Macroscopic examination of maggot revealed fully grown third instar worms, measuring 12-13 mm in length. They had broad, smooth segmented bodies with cuticular spines. With these findings diagnosis of squamous cell carcinoma with orbital myiasis was made. Maggot was identified as *Chrysomya bezziana*, the old-world screwworm fly.

3. Results

The first two cases in the series were systemically immunocompetent, whereas the third patient had local tissue compromise. None of the patients had history of trauma. Two patients, diagnosed with external ophthalmomyiasis, were managed conservatively. In the third case, due to the extensive infestation, frozen section biopsy showing features of squamous cell carcinoma and no light perception, surgical exenteration was performed. On regular follow-ups for a month, patients remained asymptomatic throughout the course of observation.

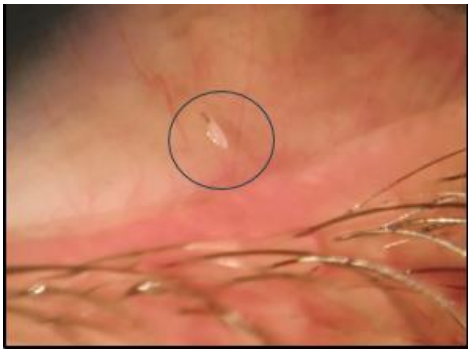

Figure 1: Slit lamp photo of case 1 showing larva (*Oestrus ovis*) over ocular surface.

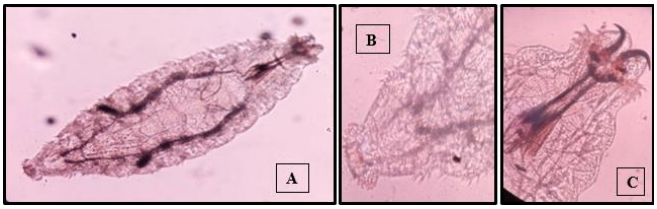

Figure 2: Microscopic picture with higher magnification (10x to 40x) of *Oestrus ovis* showing; **A**: Internal structure with an eleven segmented body; **B**: Foot end of *Oestrus ovis*; **C:** Head end of *Oestrus ovis* showing dark brown sharp oral hooks connected to cephalopharyngeal skeleton.

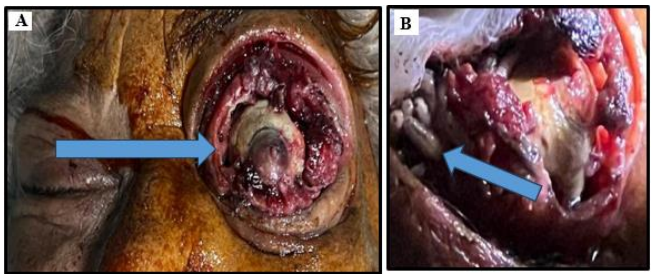

Figure 3: A: Photograph of the case 3 with orbital myiasis featuring lustreless cornea, loss of conjunctiva, proptosis and fungating mass of left eye. **B**: Image showing maggots infesting eye lid tissue and orbital tissues

Figure 4: Maggots collected in a container intraoperatively from case 3.

4. Discussion

Our study is a retrospective case series which included two cases of external ophthalmomyiasis and one cases of secondary orbital myiasis in an undiagnosed case of squamous cell carcinoma of eye. Two were female patients and one was a male patient. The age of the patients ranged between 20 to 90 years. Unilateral presentation was observed in all cases.

Ophthalmomyiasis is a rare and significant condition that can lead to vision threatening complications caused due to parasitic infestation of ocular and orbital tissue by fly larvae.[3] It accounts to less than 5 % of all human myiasis.[4] In accordance to site of ocular tissue infested by larvae, ophthalmomyiasis is classified as external, internal or orbital myiasis. Entomological classification of myiasis includes obligatory myiasis, facultative myiasis and accidental myiasis.[5] In obligatory myiasis, the parasite can only survive on warm-blooded live vertebrates and completes in life cycle in host. [5] In facultative myiasis, parasites infest on living tissue and are free living organisms.[5] In accidental myiasis, parasites are free living surviving on dead and decaying organic matter and when gravid female comes in contact with the open body cavities, they reproduce and infest the tissue.[5]

External ocular myiasis is the most common form and refers to superficial larval infestation of the periocular tissues, most commonly by sheep botfly (*Oestrus ovis*). Two cases of external ophthalmomyiasis in our study were infested with *Oestrus ovis*. Other common agents of the order *Diptera*

includes *Dermatobia hominis* (human botfly), *Hypoderma bovis* (Ox warble fly), and *Lucilia sericata* (green bottlefly).[6]

It manifests immediately after contact with the fly, as acute catarrhal conjunctivitis and presents with varied symptoms like redness, lacrimation, pain, itching and mucoid discharge along with foreign body sensation.[7,8] A case series by Sucilathangam et al reported 10 external ophthalmomyiasis cases with *Oestrus ovis*.[9] All patients in their series were farmers, had close contact with goat and sheep, unilateral involvement and immediate presentation with no previous history of allergic conjunctivitis.[9] Almost all studies had similar symptomatic presentation and our study was in accordance to these clinical findings except for the first patient in our study who had previous episodes of allergic conjunctivitis and was treated elsewhere, because of which she delayed in seeking medical help.

Oestrus ovis larvae is a spindle shaped organism,1-2 mm in length, white in color with dark heads, segmented body, photophobic and crawl rapidly to hide. Its larvae infest host (sheep, cattle and horse), as soon as birth.[10] In its normal life cycle, the adult female deposits larvae around the nostrils of hosts, to reach sinuses. Here they mature passing through three progressive developmental stages (instars).[10] The 3rd instar passes out of the host nostrils and pupates on the ground. Around 3 to 6 weeks later adult flies emerge from the pupa and live for approximately 1 month. Human serves as an accidental host when these botfly darts close to the eyes or nostrils and ejects a stream of larvae into the target area leading to external ocular myiasis. [10]

Larvae can penetrate the sclera, reach to the vitreous and sub retinal space causing Ophthalmomyiasis interna.[11,12] It presents with signs like uveitis, floaters, and atrophic and pigmented RPE tracts in multiple crisscross patterns along with fibro vascular proliferation leading to retinal detachment and blindness.[11,12] A case of internal ophthalmomyiasis in an 86 year old female patient was reported by Homer H Chiang et al.[13] She presented with a complaint of floaters and history of contact with lambs. Examination revealed subretinal tracks and larva suspended in vitreous which was extracted through pars plana vitrectomy.[13]

Orbital myiasis presents with marked swelling, foul smelling discharge, necrosis, maggots crawling in orbital tissue and wound with or without bone destruction, which is usually seen in severe cases.[14] Third case in our study had similar findings without bone destruction.

Ophthalmomyiasis is common in warm and humid tropical or subtropical zones, especially rural regions than urban areas. Various occupational activities such as shepherding, farming and veterinary activities have inherent risk of ophthalmomyiasis. Studies by Hira et al[15] and Masoodi et al[16] found that myiasis is an occupational hazard for individuals involved in farming and animal husbandry. Ophthalmomyiasis in urban centres is common among teachers, nurses and students who returned from endemic areas and is usually accidental.[17] Tamponi et al[18] and Cucera et al[19] both reported cases of external ophthalmomyiasis in tourists. In our study, two cases lived in rural area and were involved in farming and animal rearing activities. One case included a student who lived in urban region and had recently visited a botanical garden as a part of academic curriculum.

Other common risk factors included advanced age, poor social-economic status, illiteracy overcrowding, poor sanitation, livestock exposure and social isolation. Alcoholics, chronic debilitating diseases like diabetes may lead to poor hygiene maintenance. Patients with open wounds, necrotic tissue and malignancies are predisposed to ophthalmomyiasis more often.[14,17,20] Third case in our study had most of the above risk factors.

Imaging studies, such as contrast-enhanced computed tomography or magnetic resonance imaging of the orbit and brain, are valuable for determining the extent of infestation and ruling out intracranial involvement.[17,20,21] Similar imaging modality was used in our cases wherever required.

A number of approaches have been described for the removal of maggots like, mechanical removal with or without occlusion- suffocation approach wherein turpentine oil, liquid paraffin, petroleum jelly etc is applied to the affected area.[22] These agents block the spiracles of larvae forcing them to reach the surface for air.[22] Manual removal of larvae was done in our cases. Topical antibiotic-steroid combination and systemic antibiotics were used to prevent secondary bacterial infection. According to literature anti parasitic drugs like ivermectin in the dosage of 200µg/kg is effective for both prophylaxis and treatment of myiasis.[22,23] A case reported by Kumar et al in India had persistent symptoms despite extraction of all larva and topical medication. Patient's symptoms subsided only after administration of single dose of oral ivermectin 12mg.[24]

Exenteration with surgical debridement of necrotic tissue involved is indicated in cases with extensive disease to prevent tissue destruction and intracranial extension.[22] Cases of secondary myiasis in squamous cell carcinoma have been reported by many authors.[14,21,25] All of these reported cases underwent surgical management. The third case in our study was managed similarly.

Microscopic examination should be done to confirm the diagnosis and identifying the larvae.[22] Microscopic examination along with biopsy for histopathological examination of tissue should be done in cases with tissue destruction or loss to know about any predisposing factors. In our study organism were identified as *Oestrus ovis* in first two cases and *Chrysomya bezziana* in third case. Biopsy showed features of squamous cell carcinoma in third case.

All patients were closely monitored with regular follow-ups and they remained asymptomatic throughout the course

of observation. Follow up was done for a month as the duration of larval life cycle is one month.[10,17]

Prevention of human myiasis can be achieved by (a) ensuring adequate sanitation and proper waste disposal practices (b) Good wound care and regular dressings (c) use of mosquito nets and screens for windows (d) Using insecticide spray regularly especially in endemic areas (e) Raising awareness among general population and health care workers.[14]

5. Conclusion

Ophthalmomyiasis is a rare and under diagnosed condition.[17] It mimics infectious conjunctivitis and can also complicate ocular malignancies hence misleading the diagnosis.[14,17] Therefore detailed history, careful detailed ocular examination and prompt diagnosis with high suspicion is needed to prevent other visual complications like keratitis, iridocyclitis, and internal ophthalmomyiasis. Imaging studies are essential to exclude intracranial spread and adnexal involvement.[17] Due to underreporting and lack of epidemiological studies on ocular myiasis, estimation of disease burden is tough. Clinicians in endemic regions should be aware of the condition, most importantly in patients with risk factors. Future concerns on standardized guidelines on treatment combining both management of larval infestation and oncological condition is necessary. Controlling of fly population, educating the patients about good ocular hygiene and basic sanitation is important in preventing this disease. Following preventive measures and prompt diagnosis with appropriate management at right time helps in obtaining good visual prognosis.

6. Source of Funding

None.

7. Conflict of Interest

None.

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