

CASE REPORT

Orbital Soft Tissue Granuloma Caused by *Dirofilaria* couple: A rare clinical case report

ABSTRACT

Introduction:

Dirofilariasis is a ubiquitous zoonotic infection mainly involves the canine animals. Humans are accidental hosts and considered dead end of the parasite transmission.

Presentation of case:

This case report involves a rare presentation of orbital soft tissue granuloma caused by *Dirofilaria repens* species with involvement of both male and female worms in a single lesion. The clinical, radiological, and histopathological findings are presented providing insights into this rare condition.

Discussion:

Dirofilaria repens infections in humans typically involve as single worm. Presence of both male and female worm co-existing in a single lesion is an unusual presentation.

Conclusion:

Possibility of humans emerging as definitive host discussed.

Key words: Dirofilariasis, orbital granuloma, Histopathology, Definitive host.

INTRODUCTION

Dirofilariasis is ubiquitous zoonotic infection that primarily affects dogs and other wild carnivores. It is common in regions with warm climates. Common species of Dirofilariasis includes *D. repens* and *D. immitis*. *Dirofilaria repens* typically involves the subcutaneous tissue and 31% of documented cases present with nodules.^[1]

The parasite's life cycle relies on dogs as definitive hosts and mosquitoes (*Aedes*, *Culex*, and *Anopheles*) serves as intermediate hosts. Humans are accidental hosts and considered as dead end of the parasitic transmission.^[2] This report details a rare case of a soft tissue granuloma, caused by a pair of adult male and female Dirofilarial worms. Our findings suggest that humans could potentially be definitive hosts.

PRESENTATION OF CASE:

A 45-year-old male presented with painless swelling of the right eye lower lid. On examination an ill-defined, firm, mobile, and non-tender mass, approximately 2x2 cm in size, not attached to the bone or the overlying skin was present at the inferior orbital margin below the lower lid [Figure 1]. Extraocular movements were full. Anterior segment and posterior segment examination was within normal limits. Systemic examination was normal.



Figure 1: Preoperative picture showing ill-defined lesion below the right lower lid

Investigations Hb, total WBC, ESR, Peripheral smear, Absolute eosinophil count, chest Xray and Mantoux were normal. CT scan showed infraorbital soft tissue lesion seen in anteroinferior aspect of the right orbit bordering the globe and the inferior rectus muscle insertion [Fig. 2-a and b].

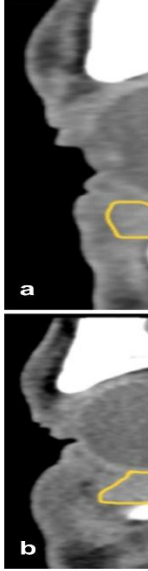
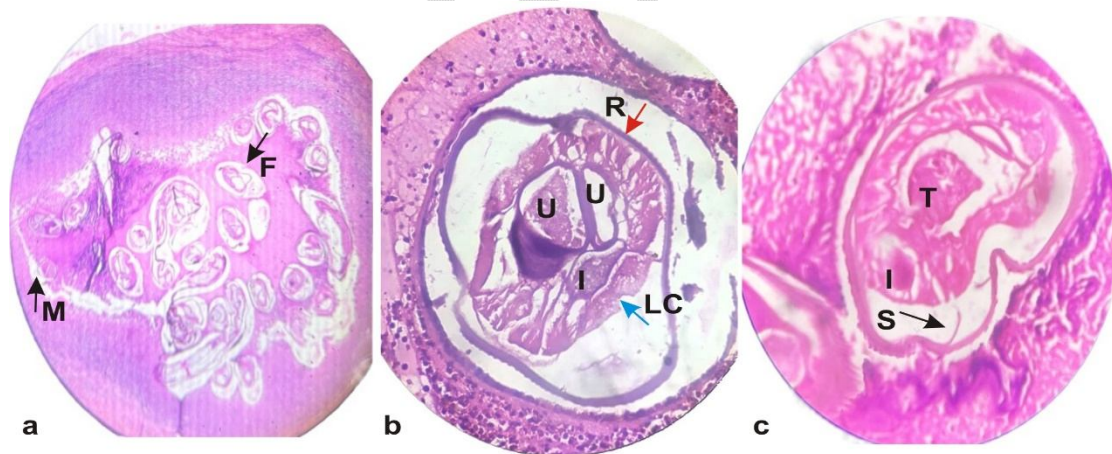


Figure 2: CT Orbit sagittal section[a]/coronal section(b) showing infraorbital soft tissue lesion in anteroinferior aspect of the right orbit bordering the globe and the inferior rectus muscle insertion.No calcification/ fat noted. Globe and intraconal space were normal

Excisional biopsy was followed by histopathological examination. On gross examination a single grey brown homogenous soft tissue fragment measuring 1.8*1.3*0.7cm noticed. HPE revealed loose hyperemic fibro-collagenous tissue with granuloma formation and dense inflammatory cell infiltrates encircling the multiple lobulate of dead worms belonging to Dirofilarial group [Fig. 3a].

Cross section of two worms revealed prominent multilayered cuticle suggestive of *D. repens* genera. Cross section of one worm had noticeable lateral cords, curved body with two uterine tracts and a digestive tract suggestive of female *D. repens* [Fig. 3b]. Cross section of another worm showed part of intestine, with granular testis and characteristic spicules suggestive of male *D. repens* [Fig. 3c]. Inflammatory cells comprising of lymphocytes, eosinophils, plasma cells and epithelioid cells were found encapsulating the worms.

Figure 3: Histopathological section (Hematoxylin and eosin) of the granuloma showing [a] multilobulate cut section of the dead *Dirofilaria* worms with surrounding dense inflammatory cells, M- cross section of male worm and F- cross section of female worm (under low magnification-10x). [b] Cross sectional view of the female worm showing the typical external cuticular ridges(R) (red arrow), lateral chords (blue arrow) and two uterine tracts(U) and intestine(I)(40x). [c] Cross sectional view of the male worm having granular testis(T), intestine(I) and characteristic spicules(S) (black arrow).(R-cuticular ridges, U-uterine tract, I-intestine, LC-lateral chords, T-testis, S-spicules)



Post-operatively patient did not have any complaints and recovered completely. He was referred to medicine department and treated with Ivermectin.

DISCUSSION

This case highlights the rare occurrence of orbital dirofilariasis granuloma. *Dirofilaria repens* (Spirurida, Onchocercidae) is a dioecious nematode. Worldwide 800 cases and in India 73 cases

have been reported.^[3] In humans, pulmonary dirofilariasis is caused by *D. immitis* and characterized by smooth cuticle. Extrapulmonary dirofilariasis, primarily attributed to the *D. repens* is characterized by longitudinal cuticular ridges and commonly found within subcutaneous tissues.^[4]

Humans serve as an atypical host, as infectious larvae are eradicated by the host's immune response.^[1] Despite common belief, Claudio et al. proposed, humans considered as accidental hosts, might exhibit a diminished immune response to the parasite and subcutaneous localization of the parasite could aid in its evasion of host defenses, facilitating its maturation.^[5] In our case patient presented with history of swelling of lower eyelid with no other features of inflammation. We hypothesize, reduced inflammation observed in our case could be attributed to the combination of factors mentioned earlier. Additionally, we believe diminished immunogenicity against the worms may have facilitated their sexual maturity, ultimately leading to granuloma formation.

Epidemiological data from India indicates that *Dirofilaria* infections typically involve a single worm encysted within a granuloma. Among 73 reported cases, only two patients were documented to have multiple worm infestations. Sekhar et al. (2000) and Sangit et al. (2012) described instances of paired adult female dirofilarial worms in a granuloma.^[3] Notably, none of the reports mention the coexistence of adult male and female *Dirofilaria*. This case represents the first documented occurrence of a matured adult male and female *Dirofilaria* found encysted in a subcutaneous nodule.

Ermakova et al., in a study involving 266 cases of human dirofilarial infestation, concludes humans serve as a biological dead end.^[6] However, a report by Ana Bakrac presents contrasting findings. Bakrac observed microfilariae in the bloodstream of a patient with a subcutaneous nodule caused by an adult female *Dirofilaria*. Author suggests the presence of microfilariae indicates likely presence of an adult male, even if not specifically detected, challenging the notion that humans are dead-end hosts. Bakrac concludes that if the simultaneous presence of sexually matured adult male and female dirofilarial worms can be demonstrated, then humans are likely definitive hosts.^[7] In our patient, sexually matured adult dirofilarial worms, males with testis and spicules, females with uterine cavities, were found coexisting in a soft tissue granuloma. This simultaneous presence of adult male and female worms within a single granulomatous nodule has not been histopathologically demonstrated previously, lending support to Ana Bakrac's proposition that humans may not be dead-end hosts but rather definitive hosts for this helminth.

CONCLUSION

This case report underscores the need for awareness of rare parasitic infestations such as dirofilariasis which can mimic as soft tissue orbital granulomas. Previous studies have reported

adult worms of same sex coexisting. We report first case of histopathologically confirmed adult male and female worms encysted in orbital soft tissue granuloma. This unique finding of dirofilarial couple encysted in a granuloma provides evidence, suggesting, humans may be emerging as a definitive host to *D. repens*. We advocate further research into the epidemiology, pathogenesis and reproductive cycle of human dirofilarial infestations.

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