

An unusual presentation of lacrimal gland dirofilariasis

Amit Sethi, Vani Puri¹, Natasha Dogra²

Dirofilariasis is a helminthic infection caused by filarial parasite of the genus *Dirofilaria*. It is a common zoonotic disease mostly affecting canines and uncommon in humans. It is endemic to Mediterranean countries, with few reports from other parts of the world. The clinical forms in humans are pulmonary, subcutaneous, and ocular. Ocular involvement is rare. We report a case of lacrimal gland dirofilariasis, an unusual ocular presentation which masquerades as a lacrimal gland tumor. Ours would be the third reported case in the world and first in India. This case was managed with excision of the lacrimal gland mass. Postoperatively the patient recovered completely and there has been no recurrence in the 1-year follow-up.

Key words: Dacryoadenitis, *Dirofilaria*, lacrimal gland

Dirofilariasis is a zoonotic helminthic infection caused by filarial parasite of the genus *Dirofilaria*,^[1] endemic in Mediterranean countries, i.e., Italy, France, Greece, and Croatia. It commonly affects canines, mostly dogs, wolves, coyotes, foxes, and cats, and is rarely seen in humans. It is transmitted to humans through mosquitoes, which are the vectors for *Dirofilaria*, and humans are the final host of this parasite.^[2] The clinical forms in humans are pulmonary, subcutaneous, and ocular. Ocular dirofilariasis is uncommon; however, there have been few reports from various parts of the world. It may present as periorbital, subconjunctival, orbital, or intraocular infection. We report a case of orbital dirofilariasis with involvement of lacrimal gland which, to our knowledge, is the first case of lacrimal gland dirofilariasis being reported in India and third in the world.

Case Report

A 33-year-old pilot, resident of Goa, presented with rapid onset swelling, redness, and inability to open the right eye for 1 day. There was no pain except for heaviness in the right eye. There were no other ocular or systemic

complaints. There was no history of drug intake, itching, or skin lesions in any other part of the body. There was swelling in the right lacrimal gland area which on palpation revealed a well-defined, firm, nontender mass, measuring 8 mm × 5 mm [Fig. 1a], with no associated lymphadenopathy. Best-corrected visual acuity was 6/6, N6 in both eyes. On slit lamp biomicroscopy of the right eye, there was generalized conjunctival congestion with chemosis [Fig. 1a]. Rest of the anterior and posterior segment was within normal limits in both eyes. Systemic examination was unremarkable. A clinical diagnosis of dacryoadenitis or pseudotumor lacrimal gland was made, and the patient was started on oral nonsteroidal anti-inflammatory drugs (NSAIDs). Meanwhile, the standard laboratory investigations, i.e., complete blood count, erythrocyte sedimentation rate, serum angiotensin converting enzyme, chest radiograph, Mantoux test, peripheral smear for microfilaria, stool examination for ova, and cyst, urine routine examination were carried out and all were within normal limits. Absolute eosinophil count was raised (798 cells/μl). Ultrasonography revealed a cystic swelling around the right lacrimal gland. The symptoms started regressing within a week of starting NSAIDs [Fig. 1b]; however, the patient was then lost to follow-up for the next 2 months. During this period, he was managed elsewhere, where magnetic resonance imaging of the orbit was done which showed a 5 mm × 4 mm cystic lesion in the lateral part of the right lacrimal gland [Fig. 2]. There he was started on tablet albendazole 400 mg with tapering dose of oral steroids for 4 weeks. Patient came back to us after 2 months of initial presentation with complaints of recurrence of symptoms. The mass had increased to 15 mm × 10 mm and was nontender and noninflammatory. Patient was advised excision biopsy. The mass was excised in toto giving an upper lid crease incision [Fig. 3a]. It was an encapsulated tumor with no adhesions to the underlying structure and measured 14 mm × 10 mm in size [Fig. 3b].

Histopathology demonstrated a parasite in the central part of the lacrimal gland surrounded by chronic inflammatory cells and significant eosinophils. The presence of thick cuticle, prominent internal cuticular ridges with lateral chords, heavy musculature, and a cavity with few partitions at places and filled with ova/larvae was morphologically matching a parasite of the genus *Dirofilaria*; most consistent with *Dirofilaria repens* [Fig. 3c].

The postoperative period was uneventful. The lesion resolved completely with no recurrence in 1-year follow-up.

Discussion

Dirofilariasis is a common zoonosis that rarely affects humans. Although considered endemic to Mediterranean countries reports have been published from other parts of the world

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Departments of Ophthalmology and ²Pathology, Indian Naval Hospital Jeevanti, Vasco da Gama, ¹Department of Ophthalmology, SMRC Hospital, Chicalim, Mormugao, Goa, India

Correspondence to: Dr. Amit Sethi, House No. 166A, NOFRA-1, Dabolim - 403 801, Goa, India. E-mail: amitsethi72@gmail.com

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Figure 1: (a) Patient on presentation – Swelling in the right lacrimal gland area with chemosis (arrow), (b) regression of symptoms with evident lacrimal gland mass (arrow)

as well.^[3] The clinical forms of dirofilariasis in humans are pulmonary, subcutaneous, ocular, and rarely infection of the cerebrum, testicles, female breast, invariably leading to an incorrect diagnosis of malignant tumor.^[4] Six out of 40 species of *Dirofilaria* are known to cause diseases in human which are: *Dirofilaria immitis*, *D. repens*, *Dirofilaria striata*, *Dirofilaria tenuis*, *Dirofilaria ursi* and *Dirofilaria spectans*.^[2] *D. immitis* is responsible for pulmonary dirofilariasis, whereas *D. repens* is responsible for subcutaneous and ocular dirofilariasis. Once the parasite enters blood circulation of definitive host, the adult female helminth, produces microfilaria that is typically found in the peripheral blood. The mosquito takes a blood meal of the infected host and ingests microfilaria. The larvae are injected into human tissues by the mosquito bite. The characteristic feature of *Dirofilaria* in humans is that the larvae do not survive after crossing the tissues. Those that manage to survive evolve into adult helminth which is reproductively inactive and cannot produce microfilariae. Hence, the infected human will never has filaremia and will always be noncontagious.

In the eye, *Dirofilaria* can be found subconjunctivally,^[5,6] periorbital (lids, orbit)^[3] and intraocularly-anterior chamber,^[7] vitreous.^[8] During the last decade, ocular dirofilariasis has increasingly been reported from countries previously considered nonendemic such as in parts of South Asia.^[9]

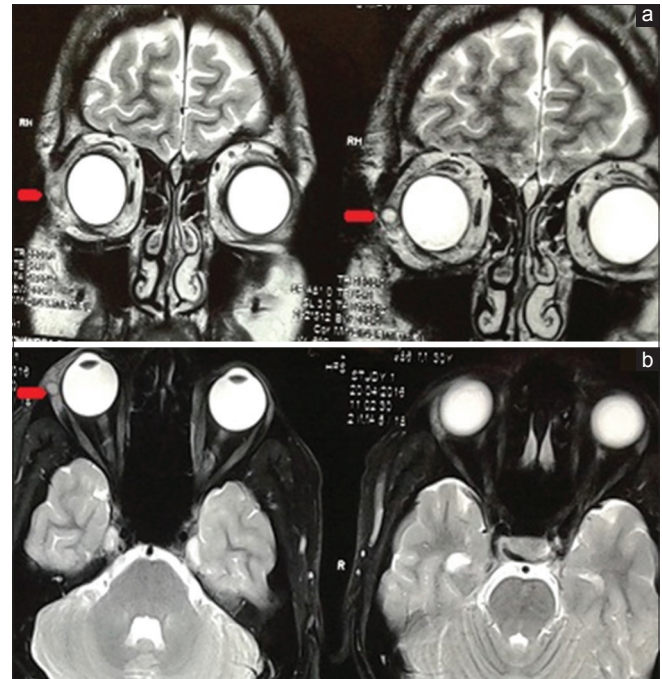


Figure 2: Magnetic resonance imaging showing a mass involving right lacrimal gland 5 mm × 4 mm (red arrows), (a) coronal view, (b) axial view

Periocular dirofilariasis usually presents as an inflammatory mass lesion and the differential diagnosis includes pseudotumor, dermoid cyst, and infectious abscess. The case reported here presented with lacrimal gland swelling with signs of local inflammation in the form of lid swelling, conjunctival congestion, and chemosis.

As *Dirofilaria* is reproductively inactive and there is no filaremia as in other filarial worms, antihelminthic drugs are not indicated. Surgical removal of the parasite or excision of the encapsulated mass with the parasite within is the only treatment for ocular dirofilariasis. No postexcision systemic workup or medication is required. In our case, we removed the lacrimal gland mass in toto which led to complete recovery.

In reviewing the literature, it is apparent that all ocular and periocular sites can be involved. Lacrimal gland dirofilariasis, to the best of our knowledge, has been reported twice, Hong Kong in 2004^[10] and Croatia in 2007.^[11] Our case would be the third in the world and first in India. There has been a previous case report from India in which the tissue around the lacrimal gland was involved.^[12]

Conclusion

Lacrimal gland dirofilariasis is a rare presentation which masquerades as a lacrimal gland tumor, and surgical removal of the parasite is the only appropriate management.

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



Figure 3: (a) Excision of mass through a lid crease incision, (b) encapsulated mass 14 mm × 10 mm, (c) histopathological section of the mass lesion with hematoxylin and eosin staining illustrating an adult female *Dirofilaria* with thick cuticle (arrow) and partitions in cavity at places filled with larvae (arrowhead)

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.

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