Sac over the sac - a rare case of subcutaneous dirofilariasis over the lacrimal sac area

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Abstract

Introduction: Dirofilariasis is a rare parasitic infestation in humans. Case: A 33-year-old female from the coastal area presented with a swelling over the right lacrimal sac area. It was clinically diagnosed as a lacrimal abscess and was incised under local anaesthesia. A live coiled worm was found and removed along with the cyst. Observations: The worm looked like a thin white thread measuring 101 mm in length. On microscopic examination, the anterior end was slightly tapered and had a rounded head. The H & E stained section of the worm revealed longitudinal ridges with transverse striations on the cuticle. Based on these findings, the worm was identified as dirofilaria repens. Conclusion: Dirofilariasis should be considered in the differential diagnosis of orbital region swelling and conjunctivitis. Surgical removal of the worm not only establishes the diagnosis but provides a definitive cure.

Keywords: lacrimal sac, cuticle, dirofilaria repens, cyst, dirofilariasis

Introduction
Human dirofilariasis is a zoonotic infection which is found world-wide. It is caused by D repens, D immitis, D tenuis and D ursi. Humans can become an accidental dead-end host. Transmission of the infection is by mosquitoes. In human infection, the development of the parasite is impaired and no microfilaria are produced. Although it is an uncommon parasite in humans, when found it is usually located in the sub-epithelial tissues of the eyelids, fingers, cheeks, breast, abdomen, and, very rarely, conjunctivae. Human ocular dirofilariasis is sporadically reported from different parts of the world (Pampiglione et al, 1995). In India, there are several reports of ocular dirofilariasis (Joseph et al, 1977; Sekar et al, 2000; Nadgir et al, 2001; Singh et al, 2010; Badhe et al, 1989). We here report a case of a subcutaneous dirofilarial sac over a lacrimal sac.

Case report
A 33-year-old female from Karwar, a coastal town of Karnataka State, India, presented with a swelling of two months duration over the right lacrimal sac. She gave no other history of ocular or medical problems. On examination, her visual acuity was 6/6 in both eyes. The eyelids of the affected eye was absolutely normal. Extra-ocular movements were full. The skin overlying the swelling was inflamed and tender. The swelling measured 8 x 6 mm. No other subcutaneous body swellings were noted. The examination of her left eye was normal. Routine laboratory examinations were within normal limits. Lacrimal syringing and dacryo-cystography were not attempted due to the presence of the acute inflammatory swelling over the lacrimal sac. The case...
was clinically diagnosed as a lacrimal abscess and was incised under local anesthesia. A live coiled worm was found and removed along with the cyst. Macroscopic examination revealed a white, thin and cylindrical worm measuring 101 mm in length. On microscopic examination, the anterior end of the worm was slightly tapered and had a rounded head. The H & E stained section of the worm revealed longitudinal ridges with transverse striations on the cuticle. The size of the worm and the location of the striations and ridges on the cuticle supported the diagnosis of dirofilariasis due to the female *Dirofilaria repens*.

The photograph of H & E stained section was sent to CDC, Atlanta by email (DPDX team, CGH DPDM DPDX. Web inquiries/whose mail ID: dpdx@cdc.gov) for confirmation, where the worm was confirmed to be *Dirofilaria repens*.

Repeated peripheral blood smear was negative for microfilaria and eosinophilia. Repeated stool and urine examinations did not reveal any parasite, ova or cysts.

Discussion

Human ocular dirofilariasis was first reported by Addario in 1885 from Milan, Italy, and was described as an infection by filaria conjunctivae (Jelnek et al, 1996). In India, it was first reported in Kerala State (Joseph et al, 1977). Dirofilariasis is accidentally transmitted to humans through the bite of Culex, Aedes and Anopheles mosquitoes. *D repens* is the most common causative agent of human dirofilariasis in India. However, few cases caused by *D immitis* have also been reported (Singh et al, 2010; Badhe et al, 1996). Human dirofilariasis is detected many years after the initial infection, when the worm dies and is enveloped in a foreign body granuloma. Surgical removal of the worm or the lesion is the treatment of choice. Most cases are diagnosed retrospectively, when the histopathological section of biopsy or excision material are viewed. There is no need for chemotherapy as microfilaria is extremely rare (Jelinek et al, 1996). The few reported cases of meningoencephalitis secondary to *D repens* microfilaria were treated with antihelminthic drugs like albendazole under the coverage of methylprednisolone and have shown a good response (Poppert et al, 2009).

The positive identification of the species may be possible after studying the morphological characteristics of the nematode. *D repens* has a long thin filariform appearance. The male are short and vary in length from 4 to 4.8 cm, while the female are longer and vary from 8 to 13 cm in length. The cephalic end is pointed while the caudal end has a rounded tail with a patent anus. It is unsheathed. *D repens* has a thick cuticle, transverse striations and a large number of external longitudinal ridges. *D immitis* can be differentiated from *D repens* by the absence of longitudinal ridges and transverse striations (Nadgir et al, 2001; Jelnek et al, 1996). Eosinophilia occurs in less than 15% cases with *D immitis* and rarely with *D repens* (Jariya et al,
1983). All these morphological features of *D repens* were present and repeated blood smear were negative for both microfilaria and eosinophilia in the case reported here. Six months after the surgical removal of the worm, the patient was completely relieved of the symptoms. In India, owing to the low prevalence rate, the PCR method and the standard antibody detecting tests are not available for the diagnosis of *D repens*.

Sub-conjunctival dirofilariasis was reported from Karwar, a coastal town in Karnataka State of India, a decade ago (Nadgir S et al, 2001). This implies that a recognized focus seems to exist in this region. The probable reasons for such a focus need to be analyzed. Human cases of dirofilariasis are probably under-reported because many of them remain undiagnosed or are unpublished. Increased awareness of this infection among clinicians treating this condition and development of the antibody screening system would improve the patient care and epidemiological survey.

**Conclusion**

Dirofilariasis should be considered in the differential diagnosis of cases with swellings over the lacrimal sac area, especially if patient is from an endemic area. Surgical removal of the worm is essential in establishing the accurate diagnosis.

**References**


