

## Live encysted *Thelazia callipaeda* presenting as a lump adjacent to the right lacrimal sac in a 42-year-old female: A rare case report

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*Thelazia callipaeda* or the oriental eye worm infects a wide variety of hosts including dogs, cats, foxes, rabbits, and humans through the *Drosophila* flies. We report here a case of a 42-year-old female who presented with pain, redness, and cystic swelling in the right lacrimal sac area for 8 months. Ocular examination was within normal limits. Excision biopsy of the cyst revealed a live worm. In humans, the worm is usually found in the conjunctival sac, lacrimal canaliculi, or the lacrimal sac leading to a disease which can be subclinical or symptomatic. Thus, we report a rare

presentation of thelaziasis, that is, periocular involvement. This case presentation is first of its kind in the Indian literature.

**Key words:** Human thelaziasis, oriental eyeworm, periocular thelaziasis, *Thelazia callipaeda*, zoonoses

In 1910, Railliet and Henry first described the arthropod-borne zoonosis by a nematode, *Thelazia callipaeda*.<sup>[1]</sup> In 1917, the first human case was reported by Stucky in China.<sup>[2]</sup> *T. callipaeda* is also known as the "Oriental eye worm" because it is mainly confined to South Asian countries.<sup>[3]</sup> The two important species causing infection in humans are *T. callipaeda* and *Thelazia californiensis*. *T. callipaeda* is prevalent in India, Thailand, China, Korea, Japan, Russia, Northern Europe, and Southern Italy. However, *T. californiensis* is exclusively seen in the United States.<sup>[4]</sup> The transmission is through the tear-seeking flies, *Phortica okadai* and *Phortica variegata*. It has been found that the *Musca domestica* (Common fly) is not a vector of *T. callipaeda*.<sup>[5]</sup>

### Case Report

A 42-year-old female, farmer and cattle breeder by occupation, presented with chief complaint of swelling, medial to the medial canthus of the right eye associated with mild pain, and redness for the past 8 months [Fig. 1a]. The patient complained of minimal watering from the right eye which was occasional with no history of discharge. On clinical examination, her best-corrected visual acuity in both eyes was 6/6 and N6. Slit-lamp biomicroscopy showed normal anterior segment with normal intraocular pressure. Fundus examination was within normal limits. Syringing showed a

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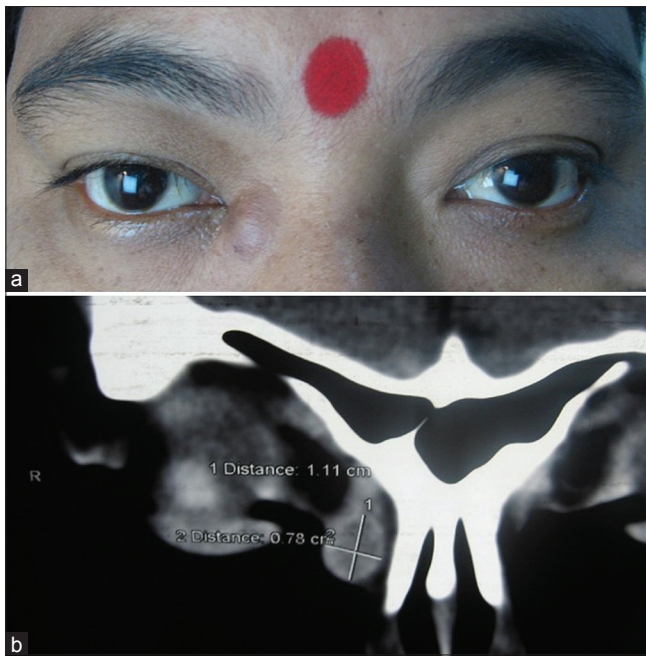
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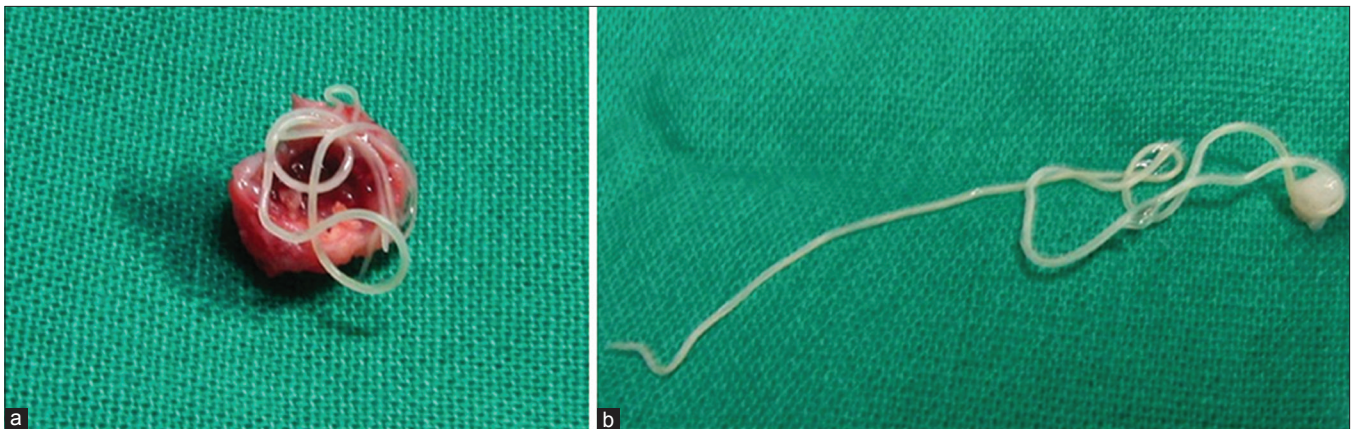
**Figure 1:** (a) Clinical photograph of the patient showing swelling medial to the right side medial canthus. (b) Computed tomography scan showing a mass of dimension 1.11 cm x 0.78 cm over the lacrimal sac area

patent lacrimal system. There was no associated history of any systemic illness.

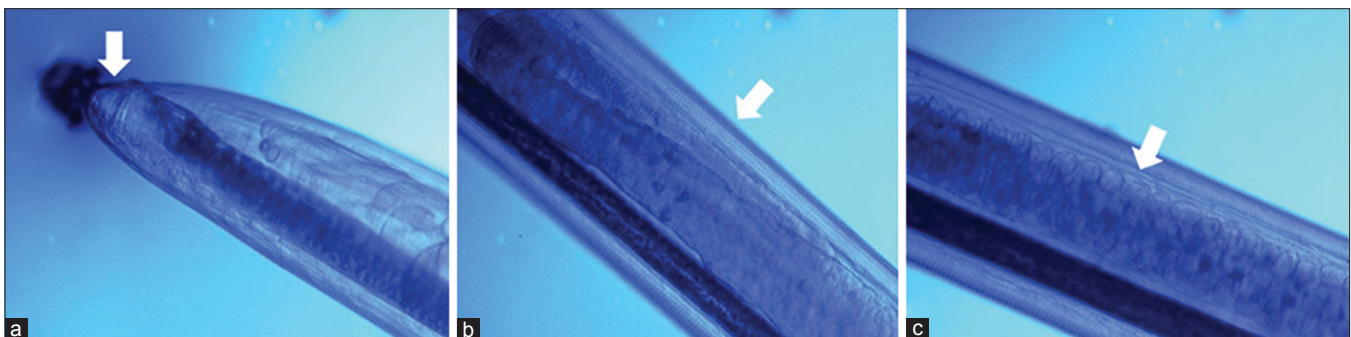
There was a 4 cm x 2 cm swelling over the right lacrimal sac area. On palpation, it was nontender and cystic in consistency with restricted mobility both sideways and vertically. The overlying skin had normal texture and was freely mobile. Thus, a differential diagnosis of dermoid, mucocele, or soft-tissue tumor was made. A computed tomography scan of orbit was advised to define the limits of swelling and to image the bony architecture and paranasal sinuses [Fig. 1b]. It showed a mass lesion of dimension 1.11 cm x 0.78 cm over the lacrimal sac area which was isodense with the surrounding soft tissue with smooth margins. Complete blood count including red blood cell count, white blood cell count, and differential leukocyte count were within normal limits.

Under local anesthesia, soft-tissue dissection was done and it was found that the cystic lesion was not adherent to the lacrimal sac. Excision biopsy of the cystic lesion was done and sent for histopathological examination. On exploration of the cyst, a creamy white thread-like live worm was found [Fig. 2a and b]. The worm showed active motility when transferred to a container containing normal saline and was video documented.

Histopathological examination of the cyst revealed granulomatous inflammation. Morphologically, the worm was



**Figure 2:** (a and b) Dissection of the cyst showing creamy white thread-like worm



**Figure 3:** (a) Microscopic view of the worm showing buccal opening (white arrow), (b) cuticular striation (white arrow), and (c) middle body segment filled with eggs (white arrow)

creamy white in color and 36 cm long and 1.75 mm wide in dimension. Microscopically, the worm had a buccal opening [Fig. 3a] with upper alimentary canal and cuticular striation all over the surface [Fig. 3b]. The middle body segment was filled with eggs [Fig. 3c]. The tail was blunted. Based on the morphological features, the worm was identified as adult female *T. callipaeda*.

The patient is under constant follow-up. In the recent visit, the incision site had completely healed with minimal scarring and no recurrence. The syringing was patent with normal tear meniscus height [Fig. 4].

## Discussion

Review of the Indian literature shows that 10 cases of ocular thelaziasis have been reported all over the country. All these cases are of ocular thelaziasis, but ours is the first case report describing the periocular thelaziasis.<sup>[1]</sup> Table 1 shows epidemiological and geographical distribution of human thelaziasis in India. Considering the global scenario, more than 250 cases of *T. callipaeda* have been reported in the literature. Maximum number of cases have been reported from China, Japan, India, Russia, Thailand, and Korea.<sup>[6]</sup>

The patient was a farmer and a cattle breeder by occupation, which is a predisposing factor for thelaziasis. Occupational history is important because the source of infection for thelaziasis is an infected animal (cows, horses, and dogs – definitive host) or human (accidental host) who harbors the first-stage larvae in the lacrimal secretions. The flies which are the intermediate host, feed on the infected lacrimal secretions, and ingest these larvae. These larvae then develop into infective third-stage larvae in the intermediate



**Figure 4:** Clinical photograph of the patient at 1-year follow-up showing healed incision with minimal scarring and no recurrence

**Table 1: Epidemiological and geographical distribution of human thelaziasis in India**

Region	Number of cases	Year of publication	Location of worms
Tamil Nadu <sup>[6]</sup>	1	1917	-
Manipur <sup>[1,7]</sup>	3	1993, 2015	Conjunctival sac
Assam <sup>[8-10]</sup>	4	1996, 2008, 2014	Conjunctival sac
Himachal Pradesh <sup>[11]</sup>	1	2006	Conjunctival sac
Karnataka <sup>[9]</sup>	1	2011	Conjunctival sac
Our case report, Assam	1	-	Lacrimal sac area

host. This stage is infective to humans. These infective larvae are transmitted to another susceptible host by the flies when it feeds on their lacrimal secretions which then develop into adult worms. Humans are considered accidental hosts.<sup>[7]</sup> Human thelaziasis usually presents with mild signs and symptoms such as foreign body sensation, epiphora, follicular hypertrophy, and less often with severe signs and symptoms such as keratitis, photophobia corneal opacities, ectropion, and secondary bacterial infections.<sup>[8]</sup>

The first case of thelaziasis was reported in India from Salem in 1948.<sup>[9]</sup> Subsequently, nine cases were reported from various parts of the country including Himachal Pradesh, Manipur, Assam, Kerala, and Karnataka. Maximum number of cases have been reported from the Northeastern part of the country, that is, seven cases, four from Assam and three from Manipur.<sup>[1,10-13]</sup> This could be attributed to the tropical climate with hilly terrain and longer monsoon period, which is favorable for the growth of arthropods, and subsequently, the vector-borne diseases.<sup>[14]</sup>

Preventive measures include use of bed nets, maintenance of personal hygiene, and keeping surroundings clean to control the vector population responsible for the transmission of infection. Since the main reservoir of *Thelazia* are the domestic animals, maintaining the hygiene and cleanliness of them would go a long way in controlling the spread within population of domestic animals which eventually become a source of infection to humans.

Even though 10 cases of thelaziasis have been reported in India, only one study is available regarding its epidemiology.<sup>[1,3,9-15]</sup> Thus, an epidemiological study should be undertaken to study the true picture of ocular thelaziasis in India.

## Conclusion

Ocular thelaziasis can manifest as a lacrimal sac mass.

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## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that her name 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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