# *Case Report*
# *Human Ocular Thelaziasis: A Case Report from Indonesia*

Revised SCARE guidelines.

SCARE Checklist

| Topic | Item | Checklist item description | Page Number |
|---|---|---|---|
| Title | 1 | The words “case report” should appear in the title. The title should also describe the area of focus (e.g. presentation, diagnosis, surgical technique or device or outcome). | 1 |
| Key Words | **2** | 3 to 6 key words that identify areas covered in this case report (include "case report" as one of the keywords). | 1 |
| Abstract | **3a** | Introduction — Describe what is unique or educational about the case (i.e. what does this work add to the surgical literature, and why is this important?). | 3 |
| | **3b** | Presenting complaint and investigations – describe the patient's main concerns and important clinical findings. | |
| | **3c** | The main diagnoses, therapeutics interventions, and outcomes. | |
| | **3d** | Conclusion — Describe the main lessons to “take-away” from this case study | |
| Introduction | **4** | Background – summarise what is unique or educational about the case. Give reference to the relevant surgical literature and current standard of care. The background should be referenced, and 1–2 paragraphs in length. | 3 |
| Patient Information | **5a** | Demographic details – include de-identified demographic details on patient age, sex, ethnicity, occupation. Where possible, include other useful pertinent information e.g. body mass index and hand dominance. | 3-4 |
| | **5b** | Presentation - describe the patient's presenting complaint (symptoms). Describe the patient's mode of presentation (brought in by ambulance or walked into Emergency room or referred by family physician). | |
| | **5c** | Past medical and surgical history, and relevant outcomes from interventions | |
| | **5d** | Other histories – Describe the patient's pharmacological history including allergies, psychosocial history (Drug, smoking, and if relevant, accommodation, walking aids), family history including relevant genetic information. | |
| Clinical Findings | **6** | Describe the relevant physical examination and other significant clinical findings. Include clinical photographs where relevant and where consent has been given. | 4 |
| Timeline | **7** | Inclusion of data which allows readers to establish the sequence and order of events in the patient's history and presentation (using a table or figure if this helps). Delay from presentation to intervention should be reported. | 4 |
| Diagnostic Assessment | **8a** | Diagnostic methods – describe all investigations taken to arrive at methods: physical exam, laboratory testing, radiological imaging, histopathology. | 4 |
| | **8b** | Diagnostic challenges – describe what was challenging about the diagnoses, where applicable, for example access, financial, cultural. | |
| | **8c** | Diagnostic reasoning – Describe the differential diagnoses and why they were considered. | |
| | **8d** | Prognostic characteristics when applicable (e.g. tumour staging or for certain genetic conditions). Include relevant radiological or histopathological images in this section. | |
| Therapeutic Intervention | **9a** | Pre-intervention considerations – if there were patient-specific optimisation measures taken prior to surgery or other intervention these should be included e.g. treating hypothermia/hypovolaemia/hypotension in a burns patient, Intensive care unit treatment for sepsis, dealing with anticoagulation/other medications, etc. | 4 |
| | **9b** | Interventions – describe the type(s) of intervention(s) deployed (pharmacologic, surgical, physiotherapy, psychological, preventive). Describe the reasoning behind this treatment offered. Describe any concurrent treatments (antibiotics, analgesia, anti-emetics, nil by mouth, Venous thrombo-embolism prophylaxis, etc). Medical devices should have manufacturer and model specifically mentioned. | |
| | **9c** | Intervention details – describe what was done and how. For surgery include details on; anaesthesia, patient position, use of tourniquet and other relevant equipment, prep used, sutures, devices, surgical stage (1 or 2 stage, etc). For pharmacological therapies include information on the formulation, dosage, strength, route, duration, etc. Include intra-operative photographs and/or video or relevant histopathology in this section. Degree of novelty for a surgical technique/device should be mentioned e.g. "first in human". | |
| | **9d** | Who performed the procedure - operator experience (position on the learning curve for the technique if established, specialisation and prior relevant training). For example, “junior resident with 3 years of specialised training” | |
| | **9e** | Changes – if there were any changes in the interventions, describe these details with the rationale. | |
| Follow-up and Outcomes | **10a** | Follow-up – describe 1) When the patients was followed up. 2) Where. 3) How (imaging, tests, scans, clinical examination, phone call), and 4) whether there were any specific post-operative instructions. Future surveillance requirements - e.g. imaging surveillance of endovascular aneurysm repair or clinical exam/ultrasound of regional lymph nodes for skin cancer. | 4 |
| | **10b** | Outcomes - Clinician assessed and (when appropriate) patient-reported outcomes (e.g. questionnaire details). Relevant photographs/radiological images should be provided e.g. 12 month follow-up. | |
| | **10c** | Intervention adherence/compliance - where relevant how well patient adhered to and tolerated their treatment. For example, post-operative advice (heavy lifting for abdominal surgery) or tolerance of chemotherapy and pharmacological agents | |
| | **10d** | Complications and adverse events – all complications and adverse or unanticipated events should be described in detail and ideally categorised in accordance with the Clavien-Dindo Classification. How they were prevented, diagnosed and managed. Blood loss, operative time, wound complications, re-exploration/revision surgery, 30-day post-op and long-term morbidity/ mortality may need to be specified. If there were no complications or adverse outcomes this should also be included. | |
| Discussion | **11a** | Strengths – describes the strengths of this case | 4-6 |
| | **11b** | Weaknesses and limitations in your approach to this case. For new techniques or implants - contraindications and alternatives, potential risks and possible complications if applied to a larger population. If relevant, has the case been reported to the relevant national agency or pharmaceutical company (e.g. an adverse reaction to a device) | |
| | **11c** | Discussion of the relevant literature, implications for clinical practice guidelines and any relevant hypothesis generation. | |
| | **11d** | The rationale for your conclusions. | |
| | **11e** | The primary “take-away” lessons from this case report. | |
| Patient Perspective | **12** | When appropriate the patient should share their perspective on the treatments they received. | 4 |
| Informed Consent | **13** | Did the patient give informed consent for publication? Please provide if requested by the journal/editor. If not given by the patient, explain why e.g. death of patient and consent provided by next of kin or if patient/family untraceable then document efforts to trace them and who within the hospital is acting as a guarantor of the case report. | 7 |
| Additional Information | **14** | Conflicts of Interest, sources of funding, institutional review board or ethical committee approval where required | 7 |

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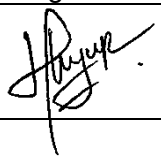
| Liem Meysie Kristi Harlimton | meysie92@gmail.com |  | |
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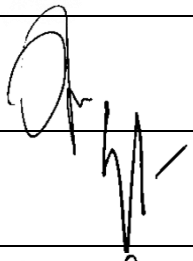
| Alif Reza | dr.alifreza@gmail.com |  | |
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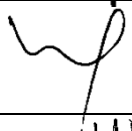
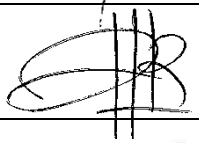
| Hasnah B. Eka | ekahasnah@yahoo.co.id |  | |
| Itzar Chaidir Islam | itzarislam@gmail.com |  | |
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# COVER LETTER

Date: 13th March 2022

To
The Editor,
**Case Reports in Ophthalmology**

I am enclosing herewith a manuscript entitled:

## *Case Report*

## *Human Ocular Thelaziasis: A Case Report from Indonesia*

For publication in Case Reports in Ophthalmology for possible evaluation. The aim of this paper is to report an uncommon occurrence of ocular thelaziasis in human. throughout our library search, this present case is the second ocular thelaziasis occurrence in the country where the worm resided in the anterior chamber, while Indonesia's first case occurred in North Sumatra reported in 1989 in a 10-month-old child. Consequently, this case also highlights the importance of parasite identification based on clinical presentation and parasitologic confirmation.

Submitted manuscript is Case Report.

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*Case Report*

# *Human Ocular Thelaziasis: A Case Report from Indonesia*

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Number of Figures: 3 (three) figures.

Word count: 1,946 words.

Keywords: Thelaziasis, *Thelazia callipaeda*, human ocular, Indonesia.

## Abstract

Thelaziasis is a parasitic disease caused by a nematode of genus Thelazia, which is rare in the world, including Indonesia. The definitive hosts for Thelazia are canids, felids, mustelids, and other mammals, while the vector is drosophila flies. Consequently, this study reported an uncommon occurrence of human ocular thelaziasis in Indonesia. Based on the patient's complaints and physical examination, we found a living worm that move actively in the anterior chamber, then documentation is carried out both during the examination at the polyclinic and in the operating room. The surgery was performed using topical anesthesia, clear corneal incision, and removing worm through the main port. Morphological examination from the parasitology laboratory showed that the worm was *Thelazia callipaeda* species. Following this intervention, the patient was given an oral anthelminthics drug, topical and oral antibiotics, topical steroid and surgical treatment. There was no recurrence or appearance of any other symptoms was reported in two months of follow-up.

## Introduction

The epidemiology of parasitic ocular diseases can be explained by the causative organism's habitat as well as the patient's habits and health status. Furthermore, an ocular examination may provide information about the underlying infection, and knowledge of the possibilities of travel-related pathology may explain the disease symptoms.(1) Thelaziasis is a parasitic condition caused by a nematode of the genus Thelazia, which infects the eyes of wild and domestic animals. The species associated with this disease include *Thelazia californiensis* and *Thelazia gulosa*, which have been reported in the United States, along with *Thelazia callipaeda*, which is common in Asia and affects humans.(2) Meanwhile, canids of domestic and wild origins are concerned as the primary definitive hosts for *Thelazia callipaeda*, though infections in mustelids, lagomorphs, and felids have been identified.(3) Thelazia californiensis infections have been detected in several mammals, mostly domestic and wild canids, as well as cervids, jackrabbits (Lepus californicus), bears, sheep, and felids. The intermediate vectors or hosts for these parasites are drosophilid flies, which consume lacrimal secretions (lacrimophagous). Fannia spp., including Fannia benjamini (canyon fly) and F. canicularis (lesser house fly), are the hosts of T. californiensis, while Phortica variegate and Phortica okadai are the primary intermediate vectors of T. callipaeda. The vectors feed on the conjunctival secretions of infected animals, thereby ingesting the parasite's first-stage larvae, which are produced from the eggshell and covered in sock-like membranes.(4) Subsequently, the parasites mature into the third larval or infective stage in 2–3 weeks while in the vector before migrating to the fly's trunk for transmission to a new host. These flies, which are active during the daytime, land on the eye area and release infective larvae onto the conjunctiva while feeding on lacrimal fluid.(5)

The first case of human thelaziasis was discovered in Asian countries, owing to the disease's spread in the former Soviet Union and Far East countries, including Korea, Nepal, China, Thailand, India and Europe.(3,6–10) This contrasts with Southeast Asian countries, such as Thailand, Myanmar, Vietnam, and Indonesia, where only a small number of ocular thelaziasis cases have been reported.(11,12)

This work has been reported in line with the improved SCARE checklist (Supplementary Material 1). The SCARE guidelines were published in 2016 and modified in 2018 to provide a structure for surgical reports.(13)

## Case Presentation

A 49-year-old man with the complaint of a moving worm in the left eye was referred to Hasanuddin University Hospital. The initial symptom was blurred vision, followed by redness and an itchy feeling in the left eye that began about 2 weeks later. A history of trauma, systemic disease, and previous ocular symptoms were denied. According to the information obtained, the patient was a farmer who resided in a district about 500 km from the capital city, where humans and animals coexisted, and no similar history had been reported in his neighbourhood.

Subsequently, the visual acuity (VA) of the right and left eyes were 20/20 and 1/60, while the intraocular pressure measured with non-contact tonometry obtained 11 and 10 mmHg, respectively. The slit-lamp examination, as shown in figure 1, revealed conjunctival hyperaemia, corneal and palpebral oedema, minimal lens opacification at the anterior capsule, and a living worm in the anterior chamber of the left eye. In addition, the assessment indicated that the right eye was normal, the funduscopic examination showed a normal posterior segment, and routine investigations, including chest X-rays and blood tests, were conducted to rule out systemic disorders.

The surgical was performed under topical and intracameral anesthesia of lidocaine, the worm then extracted from the patient's eye via a clear corneal incision made at the superior with a keratome blade. The injection of Ophthalmic Viscosurgery Devices (OVD) rouse positive pressure in the anterior chamber resulting the worm moved out through the main incision as shown in figure 2. The worm transferred immediately into the tissue container then identified in the Parasitology Laboratory of The Faculty of Medicine of Hasanuddin University and The University of Indonesia. This involved submerging the organism in a formalin solution, resulting in the appearance of a creamy-white colored worm measuring 13 mm long and 0.3 mm wide with both ends tapered. The species was identified as *Thelazia callipaeda* based on the morphology of the organism, particularly the width of the smooth and non-prominent cuticle striations, and the posterior ventral curving indicated that the nematode was male (figure 3).

Following this intervention, the patient was given levamisole 250 mg single dose, natrium diclofenac 50mg bid., ciprofloxacin 500 mg bid., topical antibiotic, and topical steroid. The post-operative inflammation subsided in one week, remaining a minor lesion on the corneal endothelium and minimal lens opacity at the anterior capsule. During the follow-up a month after

the surgery, the VA of the left eye slightly improved to 3/60. There was no recurrence or appearance of any other symptoms was reported in two months of follow-up.

## Discussion/Conclusion

Ocular parasitosis in human is a prevalent disease in certain areas, which depends on the habitat of the causative organism, vector of transmission, the host's habits, and environmental factors. Meanwhile, thelaziasis is caused by nematodes of the genus Thelazia (known as “eyeworms”), which parasitize the orbital cavity and related structures in birds and mammals, including humans, rodents, dogs, monkeys, cattle, deer, cats, pigs, foxes, horses, and camels. Thelazia parasites feed on the tears or ocular secretions of their host and are common in regions with poor hygiene and sanitation where humans live near animals.(14)

Thelaziasis report is still rare especially in Asia, throughout our library search, this present case is the second ocular thelaziasis occurrence in the country where the worm resided in the anterior chamber, while Indonesia's first case occurred in North Sumatra reported in 1989 in a 10-month-old child.(15) In this study, the patient was living and working as a farmer in a rural area and had a cat in his house, all of which may be related to this pathological finding. Several studies suggested a relationship between human ocular thelaziasis and rural settings, improper personal hygiene, low socio-economic status, and rearing livestock or parasitized animals, such as sheep, dogs, pigs, and cats, in the same environment where humans reside.(16–18)

Thelazia is a member of nematode family and has a length of 7-20 mm. The adult worms are creamy-white and thread-like with transversely striated cuticles. Their open mouth is hexagonally shaped, with a well-developed buccal cavity and a slightly short esophagus. Generally, male Thelazia species have lengths ranging between 5 - 12 mm and widths 0.3 - 0.4 mm, while the female is slightly above 10 mm to nearly 20 mm long with widths between 0.4 - 0.5 mm. Male nematodes can be distinguished macroscopically from females by the indentation on the posterior area, as they possess 6-10 pairs of precloacal papillae and 3-5 pairs of postcloacal papillae. Meanwhile, the female parasite is recognized by a genital opening known as the vulva, which has a short flap and is located in the anterior region near the esophageal-intestinal junction. The female's posterior end is rounded and blunt, with a pair of lateral papillae under the surface, and the anal canal is close to the tail tip. Therefore, the number of male pre- and post-cloacal papillae, as well as the position of the female vulva, are used to sex differentiation of Thelazia

species. In this study, the parasite species were identified as *Thelazia callipaeda* based on the morphology of the worm. (11,19,20)

Thelazia worms parasitize the orbital cavity and related structures of the eye, such as the conjunctival sac, nictitating membrane, as well as nasal and lacrimal ducts. Although a few cases of intraocular thelaziasis have been reported, this case discovered a worm in the anterior chamber, and similar cases have been reported in Pakistan and Nepal.(14,21,22) However, the process of the worm's entry into the anterior chamber is still unidentified, as the mouth of T. callipaeda lacks hooks or sharp spines, making tissue penetration unlikely. Although the route of entry into the eyes is still undetermined, some studies suggested the skin or the ingestion of untreated drinking water containing the larvae or embryonated eggs as a plausible means.(23) Despite various investigations, the ability of the larvae to penetrate human skin and travel along the bloodstream or the larvae and embryonated eggs to survive in the human digestive tract is still unexplained. However, previous studies proved that Thelazia parasites are transmitted by intermediate hosts that land on the eye region and discharge larvae into the conjunctiva.(9,24)

Worm extraction is the definitive treatment for alleviating the symptoms of infestation. Furthermore, the effectiveness of levamisole and ivermectin for similar infestations in Asia and Europe has been reported.(6,25) Studies also recommend irrigation with lugol's iodine or 2% –3% boric acid after removing the worms or when the parasites are suspected to be in the lacrimal tract.(26) For this patient, 5 mg/kgBW of levamisole was used to eradicate the parasite after the thelaziasis diagnosis was confirmed by parasitologist. During two months observation, the patient did not complain of recurrence and any other related symptoms.

As a conclusion, this case of ocular thelaziasis caused by *Thelazia callipaeda*, a rare form of parasitosis in Indonesia, was discovered in South Sulawesi. The definitive treatments for this condition are worm extraction and the administration of an anti-helminthic drug. Consequently, this case highlights the importance of parasite identification based on clinical presentation and parasitologic confirmation.

# Statements

## Acknowledgement

The author would like to thank Professor Agnes Kurniawan from the Department of Parasitology, University of Indonesia for the intensive discussion on parasite morphology. We also appreciate the team of nurses and staff of Hasanuddin University Hospital for their kind support.

## Statement of Ethics

All procedures performed in this study were in accordance with the ethical standards of the institutional and national research committee with the 1964 Helsinki Declaration standards. The study was reviewed and approved by The Ethics Committee of Medical Research, Faculty of Medicine, Hasanuddin University (No.108/UN.4.6.4.5.31/PP36/2022). Written informed consent was obtained from the patient for all medical examinations, treatments, and also publication of this case report including any accompanying images.

## Conflict of Interest Statement

The authors state that there was no conflict of interest in documenting this study.

## Funding Sources

This manuscript did not receive any funding.

## Author Contributions

**LMKH**: conception or design of the work, caring for the patient, performing follow-up after surgery, analysis, interpretation of data, and drafting the work. **SW:** parasite identification and laboratory analysis. **AR:** performing the surgical. **JS, HBE:** revising the work critically for important intellectual content. **ICI:** project administrator, drafting and revising the work critically for important intellectual content. **AMI:** conception or design of the work, perform surgical, revising the work critically for important intellectual content.

## Data availability statement

All data that support the findings of this study are included in this article.

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## Figure Legends

Fig. 1. Slit-lamp examination showed a live worm (arrow) in the anterior chamber and a hazy cornea (arrowhead).

Fig. 2.a. Surgical removal of the worm (arrow) from the anterior chamber.

Fig. 2.b. A clear corneal incision was made using keratome

Fig. 2.c. A small amount of lidocaine and viscoelastic material were inserted into the anterior chamber.

Fig. 2.d. The worm was extracted using microsurgery tweezers.

Fig. 2.e. Aspiration and irrigation to remove the ophthalmic viscosurgical devices.

Fig. 2.f. Device was extracted from the anterior chamber.

Fig. 3.a. The anterior part of the nematode has a tapered end, though the lips and buccal cavity were not visible. Cuticle stylization appeared smooth with narrow spaces.

Fig. 3.b. Posterior area of the adult male showing spicules (arrows), and the anal canal appears at the end of the tail. The curved posterior sections and long spicules are characteristic of adult male nematodes.

Fig. 3.c. Mid-section of the nematode with intestinal tube and transversally striated smooth cuticle.

Fig. 3.d. The appearance of adjacent stria in the cuticle is characteristic of *Thelazia callipaeda.*