

1 **Case Report**

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

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8
9 Short title: Human Ocular Thelaziasis: A Case Report from Indonesia

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19
20 Number of Tables: 0 (none).

21 Number of Figures: 2 (two) figures.

22 Word count: 1,946 words.

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

24 **Abstract**

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

37 **Introduction**

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

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

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

67

68 Case Presentation

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

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

83 The surgical was performed under topical and intracameral anesthesia of lidocaine, the worm
84 then extracted from the patient's eye via a clear corneal incision made at the superior with a
85 keratome blade. The injection of Ophthalmic Viscosurgery Devices (OVD) rouse positive pressure
86 in the anterior chamber resulting the worm moved out through the main incision as shown in
87 figure 2 figure 1. The worm transferred immediately into the tissue container then identified in
88 the Parasitology Laboratory of The Faculty of Medicine of Hasanuddin University and The
89 University of Indonesia. This involved submerging the organism in a formalin solution, resulting in
90 the appearance of a creamy-white colored worm measuring 13 mm long and 0.3 mm wide with
91 both ends tapered. The species was identified as *Thelazia callipaeda* based on the morphology of
92 the organism, particularly the width of the smooth and non-prominent cuticle striations, and the
93 posterior ventral curving indicated that the nematode was male (figure 3) (figure 2).

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

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

100

101 **Discussion/Conclusion**

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

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

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

129 species. In this study, the parasite species were identified as *Thelazia callipaeda* based on the
130 morphology of the worm. [11,19,20]

131 *Thelazia* worms parasitize the orbital cavity and related structures of the eye, such as the
132 conjunctival sac, nictitating membrane, as well as nasal and lacrimal ducts. Although a few cases
133 of intraocular thelaziasis have been reported, this case discovered a worm in the anterior
134 chamber, and similar cases have been reported in Pakistan and Nepal. [14,21,22] However, the
135 process of the worm's entry into the anterior chamber is still unidentified, as the mouth of *T.*
136 *callipaeda* lacks hooks or sharp spines, making tissue penetration unlikely. Although the route of
137 entry into the eyes is still undetermined, some studies suggested the skin or the ingestion of
138 untreated drinking water containing the larvae or embryonated eggs as a plausible means. [23]

139 Despite various investigations, the ability of the larvae to penetrate human skin and travel along
140 the bloodstream or the larvae and embryonated eggs to survive in the human digestive tract is
141 still unexplained. However, previous studies proved that *Thelazia* parasites are transmitted by
142 intermediate hosts that land on the eye region and discharge larvae into the conjunctiva. [9,24]

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

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

155 **Statements**

156 **Acknowledgement**

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

160 **Statement of Ethics**

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

167 **Conflict of Interest Statement**

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

169 **Funding Sources**

170 This manuscript did not receive any funding.

171 **Author Contributions**

172 **Liem Meysie Kristi Harlinton**: conception or design of the work, caring for the patient,
173 performing follow-up after surgery, analysis, interpretation of data, and drafting the work.
174 **Sitti Wahyuni**: parasite identification and laboratory analysis. **Alif Reza**: performing the surgical.
175 **Junaedi Sirajuddin** and **Hasnah B. Eka**: revising the work critically for important intellectual
176 content. **Itzar Chaidir Islam**: project administrator, drafting and revising the work critically for
177 important intellectual content. **Andi Muhammad Ichsan**: conception or design of the work,
178 perform surgical, revising the work critically for important intellectual content.

179 **Data availability statement**

180 All data that support the findings of this study are included in this article. **Further enquiries can**
181 **be directed to the corresponding author.**

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239

240 **Figure Legends**

241 **Fig. 1.a.** Surgical removal of the worm (arrow) from the anterior chamber.

242 **Fig. 1.b.** A clear corneal incision was made using keratome

243 **Fig. 1.c.** A small amount of lidocaine and viscoelastic material were inserted into the anterior
244 chamber.

245 **Fig. 1.d.** The worm was extracted using microsurgery tweezers.

246 **Fig. 1.e.** Aspiration and irrigation to remove the ophthalmic viscosurgical devices.

247 **Fig. 1.f.** Device was extracted from the anterior chamber.

248

249 **Fig. 2.a.** The anterior part of the nematode has a tapered end, though the lips and buccal cavity
250 were not visible. Cuticle stylization appeared smooth with narrow spaces.

251 **Fig. 2.b.** Posterior area of the adult male showing spicules (arrows), and the anal canal appears at
252 the end of the tail. The curved posterior sections and long spicules are characteristic of adult male
253 nematodes.

254 **Fig. 2.c.** Mid-section of the nematode with intestinal tube and transversally striated smooth
255 cuticle.

256 **Fig. 2.d.** The appearance of adjacent stria in the cuticle is characteristic of *Thelazia callipaeda*.

RESPONSE TO EDITOR

Dear Dr. Rita Czirjak
Editorial Office
'Case Reports in Ophthalmology'

Thank you very much for your kind information. Here is our point-by-point response:

- 1) Thank you for submitting your Response to Editor. However as per our Author guideline a Response to Reviewers is required so please provide your detailed response to reviewers into the relevant place in the online submission system as well as uploading your response to reviewers file.

Response:

We have revised it by adding the “response to reviewers” file and provide the detailed response to reviewers into the submission system. We also send response to editor files (20 and 27 April 2022) as the attached document.

If you have any further information. Please do not hesitate to inform us.

Best regards,

Andi Muhammad Ichsan

RESPONSE TO REVIEWER

Dear editor,

Thank you very much for your kind information. Here is our point-by-point response to the reviewers' comments:

Reviewer 1:

It look interesting. Cases with this parasite are limited in literature and in some area clinician must be careful. The paper is well structured. The authors have followed the journal rules and I suggest to accept this paper.

Response:

Thank you very much for your kind comment and support. It means a lot to us.

Reviewer 2:

I suggest reviewing the grammar. I suggest not using figure 1. Figure 2 and 3 are sufficient.

Response:

Thank you very much for your kind suggestions. We have made some improvements to the grammar errors through the "Goodlingua English editing service". Furthermore, according to your suggestion, we have removed figure 1 in the revised version of the manuscript. Some of the changes contained in the manuscript include:

1. We removed the "figure 1" legend from manuscript line 77 (page 4)
2. We changed the legend "figure 2" to "figure 1" on the line 86 (page 4)
3. We changed the legend "figure 3" to "figure 2" on the line 92 (page 4)
4. We change the figure legends on line 237-253 (page 10):
 - Fig. 1.a. Surgical removal of the worm (arrow) from the anterior chamber.
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81 investigations, including chest X-rays and blood tests, were conducted to rule out systemic
82 disorders.

83 The surgical was performed under topical and intracameral anesthesia of lidocaine, the worm
84 then extracted from the patient's eye via a clear corneal incision made at the superior with a
85 keratome blade. The injection of Ophthalmic Viscosurgery Devices (OVD) rouse positive pressure
86 in the anterior chamber resulting the worm moved out through the main incision as shown in
87 figure 2 figure 1. The worm transferred immediately into the tissue container then identified in
88 the Parasitology Laboratory of The Faculty of Medicine of Hasanuddin University and The
89 University of Indonesia. This involved submerging the organism in a formalin solution, resulting in
90 the appearance of a creamy-white colored worm measuring 13 mm long and 0.3 mm wide with
91 both ends tapered. The species was identified as *Thelazia callipaeda* based on the morphology of
92 the organism, particularly the width of the smooth and non-prominent cuticle striations, and the
93 posterior ventral curving indicated that the nematode was male (figure 3) (figure 2).

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

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

100

101 **Discussion/Conclusion**

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

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

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

129 species. In this study, the parasite species were identified as *Thelazia callipaeda* based on the
130 morphology of the worm. [11,19,20]

131 *Thelazia* worms parasitize the orbital cavity and related structures of the eye, such as the
132 conjunctival sac, nictitating membrane, as well as nasal and lacrimal ducts. Although a few cases
133 of intraocular thelaziasis have been reported, this case discovered a worm in the anterior
134 chamber, and similar cases have been reported in Pakistan and Nepal. [14,21,22] However, the
135 process of the worm's entry into the anterior chamber is still unidentified, as the mouth of *T.*
136 *callipaeda* lacks hooks or sharp spines, making tissue penetration unlikely. Although the route of
137 entry into the eyes is still undetermined, some studies suggested the skin or the ingestion of
138 untreated drinking water containing the larvae or embryonated eggs as a plausible means. [23]

139 Despite various investigations, the ability of the larvae to penetrate human skin and travel along
140 the bloodstream or the larvae and embryonated eggs to survive in the human digestive tract is
141 still unexplained. However, previous studies proved that *Thelazia* parasites are transmitted by
142 intermediate hosts that land on the eye region and discharge larvae into the conjunctiva. [9,24]

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

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

155 **Statements**

156 **Acknowledgement**

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

160 **Statement of Ethics**

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

167 **Conflict of Interest Statement**

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

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171 **Author Contributions**

172 **Liem Meysie Kristi Harlimton**: conception or design of the work, caring for the patient,
173 performing follow-up after surgery, analysis, interpretation of data, and drafting the work.
174 **Sitti Wahyuni**: parasite identification and laboratory analysis. **Alif Reza**: performing the surgical.
175 **Junaedi Sirajuddin** and **Hasnah B. Eka**: revising the work critically for important intellectual
176 content. **Itzar Chaidir Islam**: project administrator, drafting and revising the work critically for
177 important intellectual content. **Andi Muhammad Ichsan**: conception or design of the work,
178 perform surgical, revising the work critically for important intellectual content.

179 **Data availability statement**

180 All data that support the findings of this study are included in this article. **Further enquiries can**
181 **be directed to the corresponding author**

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239

240 **Figure Legends**

241 **Fig. 1.a.** Surgical removal of the worm (arrow) from the anterior chamber.

242 **Fig. 1.b.** A clear corneal incision was made using keratome

243 **Fig. 1.c.** A small amount of lidocaine and viscoelastic material were inserted into the anterior
244 chamber.

245 **Fig. 1.d.** The worm was extracted using microsurgery tweezers.

246 **Fig. 1.e.** Aspiration and irrigation to remove the ophthalmic viscosurgical devices.

247 **Fig. 1.f.** Device was extracted from the anterior chamber.

248

249 **Fig. 2.a.** The anterior part of the nematode has a tapered end, though the lips and buccal cavity
250 were not visible. Cuticle stylization appeared smooth with narrow spaces.

251 **Fig. 2.b.** Posterior area of the adult male showing spicules (arrows), and the anal canal appears at
252 the end of the tail. The curved posterior sections and long spicules are characteristic of adult male
253 nematodes.

254 **Fig. 2.c.** Mid-section of the nematode with intestinal tube and transversally striated smooth
255 cuticle.

256 **Fig. 2.d.** The appearance of adjacent stria in the cuticle is characteristic of *Thelazia callipaeda*.

Case Report

Human Ocular Thelaziasis: A Case Report from Indonesia

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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 variegata* 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, 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 fundoscopic 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 1. 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 2).

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]

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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

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Author Contributions

Liem Meysie Kristi Harlimton: conception or design of the work, caring for the patient, performing follow-up after surgery, analysis, interpretation of data, and drafting the work. **Sitti Wahyuni:** parasite identification and laboratory analysis. **Alif Reza:** performing the surgical. **Junaedi Sirajuddin** and **Hasnah B. Eka:** revising the work critically for important intellectual content. **Itzar Chaidir Islam:** project administrator, drafting and revising the work critically for important intellectual content. **Andi Muhammad Ichsan:** conception or design of the work, perform surgical, revising the work critically for important intellectual content.

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Figure Legends

Fig. 1.a. Surgical removal of the worm (arrow) from the anterior chamber.

Fig. 1.b. A clear corneal incision was made using keratome

Fig. 1.c. A small amount of lidocaine and viscoelastic material were inserted into the anterior chamber.

Fig. 1.d. The worm was extracted using microsurgery tweezers.

Fig. 1.e. Aspiration and irrigation to remove the ophthalmic viscosurgical devices.

Fig. 1.f. Device was extracted from the anterior chamber.

Fig. 2.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. 2.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. 2.c. Mid-section of the nematode with intestinal tube and transversally striated smooth cuticle.

Fig. 2.d. The appearance of adjacent stria in the cuticle is characteristic of *Thelazia callipaeda*.

COVER LETTER

Date: 28th April 2022

To
The Editor,
Case Reports in Ophthalmology

I am enclosing herewith a **revised version** of 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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With the submission of this manuscript, I would like to undertake that:

1. All authors of this paper have directly participated in the planning, execution, or analysis of this study;
2. All authors of this paper have read and approved the final version submitted;
3. The contents of this manuscript have not been copyrighted or published previously;
4. The contents of this manuscript are not now under consideration for publication elsewhere;
5. The contents of this manuscript will not be copyrighted, submitted, or published elsewhere, while acceptance by the Journal is under consideration;
6. There are no directly related manuscripts or abstracts, published or unpublished, by any authors of this paper;
7. My Institute's Department of Ophthalmology, Hasanuddin University, Makassar, Indonesia representative is fully aware of this submission.





