Case Report: Periorbital Filariasis Caused by *Brugia malayi*

Poonyawee Nunthanid,1 Kosol Roongruanchai,2 Sirichit Wongkamchai,2 and Patsharaporn T. Sarasombath2*

1Department of Ophthalmology, Damnoen Saduak Hospital, Ratchaburi, Thailand; 2Department of Parasitology, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand

Abstract. *Brugia malayi* is a lymphatic nematode that accounts for approximately 10% of lymphatic filariasis cases worldwide. It is endemic in several countries in South and Southeast Asia. In Thailand, *B. malayi* is endemic in the southern region. The extralymphatic presentation of *B. malayi* is rare. Here, we report the case of a woman residing in the central region of Thailand who presented with an erythematous periorbital nodule at the left medial canthal area caused by lymphatic filaria. A viable sexually mature filarial adult was removed from the lesion. The nematode species was identified as *B. malayi* by histology staining and DNA sequencing of the partial mitochondrial 12S ribosomal RNA (rRNA) gene. As far as we know, this is the first case report of *B. malayi* presenting with a periorbital nodule that has occurred in a disease non-endemic area of Thailand with possibly a zoonotic origin.

INTRODUCTION

*Brugia malayi* is a filarial nematode of the Onchocercidae family, which is one of the leading causes of lymphatic filariasis. It affects people in many tropical and subtropical areas, including in Thailand.1 Approximately 90% of human lymphatic filariasis cases in the world are caused by *Wuchereria bancrofti* (bancroftian filariasis), whereas the remainder are caused by *B. malayi* and *Brugia timori* (brugian filariasis).1,2,3 In Thailand, *B. malayi* is endemic in the southern region.4 Several genera of mosquitoes, including mosquito species from the genera *Aedes*, *Anopheles*, and *Mansonia*, can transmit brugian filariasis, depending on the *Brugia* strain and geographic area.4-6 *Mansonia* spp. has been reported to be a main vector of *B. malayi* in Thailand.2,4 Unlike *W. bancrofti*, another species of lymphatic filaria that is also endemic in Thailand, *B. malayi*, has natural animal reservoirs besides humans, including cats, dogs, monkeys, and pangolins.5,7 The extralymphatic presentation of *B. malayi* is rare, and only a few cases have been proven for the causative species.8-10 Most of the reported cases of extralymphatic filariasis were suspected to be of brugian origin, but in only a few of those cases was the causative species truly identified.8,9 Moreover, the presence of an adult stage of a sexually mature or gravid adult worm in the lesion is extremely unusual.2,3 Here, we report the first case of brugian filariasis, in which a viable adult worm was recovered in the periorbita. The causative filarial species was determined by histopathology and confirmed by DNA sequencing of the partial mitochondrial 12S rRNA (mt 12S rRNA) gene as *B. malayi*.

CASE REPORT

A 70-year-old woman presented with an erythematous periorbital nodule at the left medial canthal area that had persisted for 2 weeks. Ophthalmic examination disclosed a 1-cm inflamed solid nodule adhering to the superomedial wall of the left orbital rim. There was no history of migratory edema. No lymphadenopathy was observed. The patient denied any history of an underlying condition or eye trauma. She resided in Damnoen Saduak, Ratchaburi Province, Thailand, with no travel history outside the city. She was previously healthy and had a domestic dog pet at home. Her visual acuity was 6/6 for both eyes. Examination of her eyelids, extraocular muscles, and eyeballs was within normal limits. An orbital computed tomography (CT) scan was performed to exclude intraorbital lesion and determine the extent of the lesion. It revealed an 8.6 × 4.5 × 7.1-mm rim-enhancing extra-orbital hypodense nodule adjacent to the medial canthus of the left orbit (ante-ro lateral nasal region). No bone invasion was observed (Figure 1A and B). A complete blood count examination was not performed before surgery. An infected periorbital nodule was suspected. The patient received dicloxacillin 250 mg, four times a day for 2 weeks. A mass excision was planned after the inflammation had subsided. During the surgery, a thin-walled cyst was found attached to the anteronasal orbital rim of the left eye. Inside the cyst, a white thin long coiled nematode surrounded with some serous fluid was found and removed with forceps. The nematode was slender and still viable and approximately 9.5 cm long by 1 mm wide, as shown in Figure 2 and in the Supplemental Video 1. The nematode was sent to the Department of Parasitology, Faculty of Medicine Siriraj Hospital for identification. The histological section of the nematode stained with hematoxylin and eosin dye is shown in Figure 3. A sexually mature adult female most consistent with those of the *Brugia* species was identified. The section of the nematode revealed a nematode containing two uterine tubes filled with developing cells, a single intestine, and a few flat and broad muscle cells per quadrant. The nematode showed compatible features characteristic of a sexually mature female of the filarial species (Figure 3).

To further confirm the species of the filarial nematode, PCR was performed targeting the partial sequence of the mt 12S rRNA gene with the protocol previously described11,12 and cloned into the T-Vector pMD20 for sequencing. The mt 12S rRNA sequence from this case was submitted to the NCBI database under GenBank accession number MT584737. The 110 bp of the PCR product of the partial sequence of the mt 12S rRNA of this nematode was 100% identical to the reference sequences of *B. malayi* (GenBank accession numbers AF538716.1 and AJ544843.1).

After surgical removal of the worm, the patient was prescribed two tablets of albendazole (200 mg) twice daily for 7 days. The surgical wound was completely healed without secondary bacterial infection. The complete blood count,
peripheral blood smear, and stool examination after treatment were all within normal limits. Blood for microfilaria and serum filarial IgG4 were not performed in the study. No other abnormal findings were observed in this patient.

DISCUSSION

Filariasis is a disease caused by nematodes in the Filarioidea superfamily. Thailand is an endemic area of lymphatic filariasis caused by two major species of filaria: *W. bancrofti* and *B. malayi*. *Brugia malayi* is endemic in four provinces in the southern part of Thailand: Surat Thani, Nakhon Si Thammarat, Krabi, and Narathiwat provinces. The clinical presentations of lymphatic filariasis vary, ranging from asymptomatic to lymphangitis and lymphadema, whereas in bancroftian filariasis, enlargement of the scrotum can be observed. Unlike *W. bancrofti*, in which humans are the only definite host, *B. malayi* has a wide variety of animal reservoirs, including dogs, cats, monkeys, and pangolins. *Brugia malayi* has been observed in blood samples of cats in the brugian endemic areas of Thailand, including Nakhon Si Thammarat, Phatthalung, Pattani, Yala and Narathiwat, and Surat Thani. In addition, *B. malayi* was also reported in the disease non-endemic area in Thailand including Bangkok metropolitan region. Zoonotic filariasis is becoming a major concern in disease-endemic areas, with several case reports recognizing the symptoms with increasing frequency. This hinders the success of disease eradication in endemic areas, especially in the southern part of Thailand. Interestingly, the patient in the present case report resides in Ratchaburi Province in the central region of Thailand. Such an infection of *B. malayi* in this disease non-endemic area without prior travel history is unusual. The infection of *B. malayi* in dogs in Bangkok which is not far from Ratchaburi has been previously reported. Thus, a zoonotic nature of the infection in this patient is possible. Although intraocular filariasis is relatively common, a periorbital nodule caused by adult *B. malayi* has never been reported before. In addition, most orbital filariasis in East Asia as previously reported were caused by other filarial species, mainly *Dirofilaria* spp. and *W. bancrofti*. 

![Figure 1.](image1.png)  
*Figure 1.* (A) Contrast computed tomography (CT) (A: axial view, B: coronal view) of the patient’s orbit revealing an enhancing nodule with central hypodensity at the superomedial aspect of the left orbit (shown by the arrow). (B) Contrast CT (A: axial view, B: coronal view) of the patient’s orbit revealing an enhancing nodule with central hypodensity at the superomedial aspect of the left orbit (shown by the arrow). This figure appears in color at www.ajtmh.org.

![Figure 2.](image2.png)  
*Figure 2.* The threadlike, whitish, slender-shaped coiled nematode as shown was removed from the patient’s lesion. The size of the nematode was approximately 9.5 cm long and 1 mm wide. This figure appears in color at www.ajtmh.org.

![Figure 3.](image3.png)  
*Figure 3.* Section of the nematode revealing a nematode with a thin cuticle, two uterine tubes (U) containing developing eggs, and one intestine (I). It has a few, well-developed broad muscle cells per quadrant and prominent lateral cords (Lcs) with a few Lc nuclei (shown by the arrows). This figure appears in color at www.ajtmh.org.
Only a few extralymphatic filariasis cases in which the species identification has been confirmed as *B. malayi* have been previously reported. Moreover, none of these had the species identified by a molecular technique. In the case of extralymphatic filariasis discovered in this patient was a sexually mature but not gravid adult female. No lymphadenopathy was observed in this patient. Eosinophilia was not observed in this patient, consistent with the fact that only a certain number of patients with extralymphatic filariasis show elevated eosinophilia. In the case of extralymphatic filariasis, identification of the filarial species is extremely important for the diagnosis, treatment decision, and control strategy. Normally, the removal of an effected lesion without additional treatment, in the case of amicrofilaremia and a negative serum IgG4, is usually considered sufficient treatment for zoonotic filariasis. In *Brugia* filariasis, zoonotic transmission is possible; thus, eradication of the microfilaria in natural reservoirs is necessary for the control strategy. Exploration of the mosquito vectors and animal reservoirs in this patient’s hometown should be further performed. As far as we know, this is the first case report of *B. malayi* presented with a periorbital nodule in a disease non-endemic area with suspected zoonotic origin.

Received July 14, 2020. Accepted for publication August 9, 2020.

Published online September 21, 2020.

Note: Supplemental video appears at www.ajtmh.org.

Acknowledgments: We would like to thank Prof. Prapathip Eamsobhana, Department of Parasitology, and Assoc. Prof. Panita Sithinamsuwan, Department of Pathology, Faculty of Medicine Siriraj Hospital, Mahidol University, for their suggestions and assistances with this work.

Authors’ addresses: Poonyawee Nunthanid, Department of Ophthalmology, Damnoen Saduak Hospital, Ratchaburi, Thailand, E-mail: poonyawee.nd@hotmail.com. Kosol Roongruanchai, Sirichit Wongkamchai, and Patsharaporn T. Sarasombath, Department of Parasitology, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand, E-mails: kosol.run@mahidol.ac.th; sirichit.won@mahidol.ac.th; and patsharaporn.tec@mahidol.ac.th.

REFERENCES


