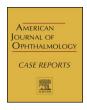
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American Journal of Ophthalmology Case Reports

journal homepage: www.elsevier.com/locate/ajoc



Case report

A new worm infiltrating the human cornea: A report of three cases



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ARTICLE INFO

Keywords: Parasite Nematode Ocular Uveitis Cornea

Stroma

ABSTRACT

 ${\it Purpose:}\ \ {\it To\ characterize\ a\ new\ species\ of\ parasitic\ nematode\ that\ triggers\ uveit is.}$

Observations: Three previously healthy, relatively young people each contracted a corneal stromal nematode that, upon surgical removal and examination, did not match any known nematodes. Clinical ocular findings included corneal opacification, visible corneal worms, conjunctival injection, and uveitis.

Conclusions and Importance: The three cases presented here represent a previously undescribed parasitic infection of the cornea by an unidentified nematode. These findings may represent a previously unrecognized zoonotic infection from wildlife sources and potentially a newly documented nematode requiring description. Future clinical findings regarding this newly described nematode are needed to further develop our understanding of the disease.

1. Introduction

Ocular parasites-including protozoa, nematodes, cestodes, and trematodes-are well-documented, and ocular parasitosis has been found to be significantly more common in regions with favorable environmental factors and poor sanitary conditions. 1-3 In these regions, ocular parasitosis can be endemic in the canine and feline populations, as well as in a range of wildlife species including other mammals or birds, providing a breeding ground from which arthropod vectors can transmit parasites to humans.2 However, it is unusual to find a live worm in intraocular structures. Nematode parasites do not usually proliferate within their definitive hosts, but rather grow, molt, mature as dioecious adults in specific anatomical sites, mate, and then produce eggs, larvae or microfilariae. 4 During this life cycle, worms can migrate to different locations within the body, including the eye¹⁻⁴; migration takes place via blood borne carriage or through tissue to the eye or adjacent structures. 1–3^{,5} The eye's immune privilege may allow further growth and development relative to other tissues, 6-11 and helminth parasites can infect the conjunctiva, eyelid, and intraocular cavities. 1-3 A diverse assemblage of zoo parasitic nematodes have been documented in ocular infections in people, and involve both fully developed nematodes or larval stages: for example, zoonotic species of Onchocerca have been documented to involve the cornea (probably Onchocerca cervicalis)^{2,12,14} and the anterior chamber.^{2,13,14} The following is a report of three patients from the southwestern Pacific island of Saipan, in the Mariana Islands, who presented with corneal stromal nematodes between 1997 and 2009. We believe these nematodes to be of a previously undescribed species.

2. Findings

2.1. Case 1

Two weeks prior to presenting to an ophthalmologist in March 1997 on Saipan, the patient, a healthy 29-year-old Chamorro male without prior ocular, medical, or surgical history, had seen an optometrist for photophobia of 2 weeks duration in his left eye. He reported having traveled to Honolulu, Hawaii two weeks prior to development of his symptoms, but had not been outside either the Hawaiian Islands or the Mariana Islands recently. He was placed on prednisolone acetate $1\%\ q\ 1$ hour, and homatropine $5\%\ TID$.

On examination by the ophthalmologist, his best corrected visual

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acuity with plano lenses was RE 20/20, LE 20/25+. External exam, pupils, and motility were normal, and intraocular pressure was 10 mmHg in both eyes. Slit lamp examination of the right eye was unremarkable.

The left eye showed areas of peripheral and mid-peripheral corneal opacification. There was no injection. There was a 1.5 mm long translucent motile worm located approximately $\frac{2}{3}$ depth within the midperiphery of the corneal stroma (Supplemental Fig. 1). The movement of the worm was primarily undulating, and its speed through the cornea was not fast enough to note any forward or backward movement. The worm appeared to be photophobic, contracting to light from the slit lamp beam.

The anterior chamber showed rare cells and no flare. Dilated fundus exam was normal for both eyes, without evidence of vitreous cell, posterior segment parasites, or chorio-retinal tracks. The prednisolone dose was decreased to every 2 hours, and the homatropine to once daily. Complete blood count was normal with no eosinophilia. Liver function tests were also normal. Stool examination showed no mature or larval parasites or eggs.

Over the ensuing weeks, the worm was noted to traverse the midperipheral cornea, moving as far as 2–3 mm per day, but often remaining in the same area of the cornea: 3–4 mm from the limbus. There was no change during this period in the visual acuity, corneal opacity, or anterior inflammation. It had been hoped that the worm would move far enough to the periphery to allow a direct cut-down over the worm, in order to remove the worm with minimal refractive effect of the incision.

After 2 weeks of observation, surgical intervention was considered. Removal of the worm was attempted with a slit lamp, as visualization under the operating microscope was impaired. A 22-degree SuperBlade[™] was used to place a 2 mm horizontal incision ²/₃ depth into the cornea directly over the worm, at the 5:00 mid-periphery. As the incision was made, it was noted that the intracorneal worm was moving vigorously away from the incision site. Viscous lidocaine was placed into the incision, and the worm stopped moving. However, the light reflexes from the corneal stroma and the incision made it difficult to distinguish the worm from the corneal stroma. Fluorescein dye was placed, which did not significantly highlight the worm. After multiple attempts to grasp and remove what may have been the worm through the incision, the decision was made to cease further manipulations. The cornea and anterior chamber were carefully examined, and it was noted that the parasite was not visible in either, so it was presumed to be present in the area of the incision. The partial thickness incision was left unsutured. Antibiotic ointment and a patch were placed, and the pa-

On postoperative day 1, visual acuity decreased to 20/40. There was 3 + conjunctival injection. The horizontal incision site at 5:00 was well approximated, and at 3:00 in the mid-periphery, the live worm was visible. The patient and his wife were highly distressed. The decision was made to attempt to kill the worm.

An argon laser was used through an Abraham lens, at a spot size of $100~\mu m$, and duration of 0.1 msec. Beginning at 80~mW, the power was titrated up to 400~mW, focusing treatment on the ends of the worm. A total of 35 spots were placed along the length of the worm, and at the end of the treatment, the worm had ceased moving. The following day, the worm had moved to the 2:00~mid-periphery. It was motile and continued to contract when exposed to the slit lamp beam.

The patient was referred to a corneal specialist for further evaluation and management. The worm was measured at the slit lamp to be approximately 1500 μ m long, at $\frac{2}{3}$ depth into the paracentral cornea.

It was considered that destroying the worm by cryoablation or photoablation might incite a severe immunologic reaction, and for this reason, the decision was made to proceed with surgical removal. Because topography showed that the previous vertical incision had resulted in astigmatism, it was elected to use an astigmatically neutral lamellar surgical approach and "bring the organism to the surface".

Six weeks after presentation, the patient was brought to the operating room, and under local anesthesia, a disposable Katena Barron-Hessburg suction trephine was used. The trephine was centered to include the worm, and trephination done to approximately 300 μm . The parasite was removed intact and passed directly to the parasitologist. The specimen, however, was lost during processing.

Prednisolone and homatropine were gradually tapered. Four months after surgery, there were no signs of active inflammation. Visual acuity was 20/50, correctable to 20/20. The patient showed no signs of systemic or further ocular parasitic infection.

2.2. Case 2

In November 2005, a healthy 8-year-old Chamorro male presented on Saipan with a 3 week history of redness, itching, glare sensitivity and blurred vision OD. He had no past ocular or medical history. He had traveled one month prior to Houston, Texas, and 10 months prior to that to southern California.

His visual acuity was RE 20/30, correctable to 20/25 ($-0.50 + 1.00 \times 065$), LE 20/20. External exam, pupils and motility were normal. Slit lamp exam of the right eye showed diffuse +1 conjunctival injection. The cornea showed diffuse subepithelial opacities from the 11:00 limbus to 2:30, with surrounding cell (Supplemental Fig. 2). The corneal epithelium showed no staining or ulceration. Within the corneal stroma, at the 2:30 o'clock position mid-periphery, a 1 mm long motile worm was visible. It was translucent with tapered ends and seemed to have a visible cavity through the center along the longitudinal axis. This translucency was presumed to be an intestine. The worm contracted to light. The anterior chamber showed no cell or flare. The iris and lens were normal. The slit lamp exam of the left eye was unremarkable. Intraocular pressure was normal in both eyes. Dilated retinal exam was normal in both eyes, with no vitreous cell, and no signs of retinal or choroidal tracks.

Systemic evaluation by a pediatrician was normal, as were laboratory examination of stool for parasites, complete blood count including eosinophils, abdominal ultrasound, and serum chemistries.

The patient was referred for removal of the corneal worm. Exams during the ensuing 2 weeks showed that the worm remained in the same area of the cornea, though it rotated its orientation relative to the limbus, from 90° to the limbus to 60° to the limbus. The worm was extracted in December 2005 through a freehand lamellar dissection with a limbal incision into the cornea. The worm, however, was not removed intact, and there was no specimen available for pathological or parasitological examination.

Postoperatively, the patient did well, with visual acuity returning to 20/20 within one week. Slit lamp exam showed no visible worm remnants. The patient has been followed for 10 years postoperatively, and his vision has remained 20/20, with slight corneal scarring superiomedially. There have been no signs of recurrence of the ocular worm during this period.

2.3. Case 3

In December 2008, a generally healthy, 34-year-old Chamorro woman with a history of soft contact lens wear for myopia presented to an optometrist with blurred vision in her right eye for one week. Her travel history was negative except for a one week trip to San Diego, California, 3 weeks prior.

Her visual acuity with soft contact lenses was 20/30 OD, 20/20 OS. Slit lamp exam OD noted no injection, +2 diffuse superficial punctate keratitis with staining, and +1 diffuse central stromal haze. There were no anterior chamber cells or flare. The anterior segment of the left eye was normal. It was assumed that the signs were related to the contact lens, and the contact lenses were discontinued, with instructions to return in 3 days. No improvement was noted, and the patient was referred to an ophthalmologist.

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