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Subretinal gnathostomiasis: A successful focal laser photocoagulation for a living parasite

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A R T I C L E I N F O

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A B S T R A C T

Purpose: To report a case of subretinal gnathostomiasis presenting with progressive subretinal tracts of a living parasite and successfully treated with focal laser photocoagulation.
Method: Observational case report.
Patient: A 29-year-old Thai male complained of blurred vision and floaters in his left eye for two weeks. An ocular examination showed multiple, whitish, subretinal tracks at the superotemporal retina. After 5 days of oral albendazole, a moving parasite was confirmed by multimodal retinal imaging. An immunoblotting analysis was positive for *Gnathostoma* species.
Result: The patient was treated by laser photocoagulation with frequency-doubled Nd:YAG laser around and over the parasite. Oral albendozole was continued and naproxen was prescribed for four weeks. His vision improved to 20/20 and the inflammation subsided completely within three months. The patient has been followed for five years without local and systemic complications.
Conclusions: Focal laser photocoagulation without systemic steroids could be a successful treatment for active subretinal gnathostomiasis with a satisfactory safety profile in a long-term follow-up.

1. Introduction

Gnathostomiasis is one of the most common parasitic infections in South East Asia and Latin America. There are 13 *Gnathostoma* species, but only 6 can cause diseases in humans.[1] This foodborne zoonosis is caused by the consumption of uncooked freshwater animals, such as fish, frogs, and eels, which are harboring third-stage larvae, or by the drinking of contaminated freshwater containing copepods.[2] The larvae or copepods then use humans as a dead-end host. The accidental consumption of roundworm larvae results in the parasite invading the visceral organs and causing the condition named visceral larva migrans. The creeping roundworm then migrates throughout the infected human body, typically leading to the more common signs of cutaneous larva migrans, which is a progressive swelling and an inflammatory response over a specific part of the skin. The migration of the worm into the human neurological system could cause a devastating outcome.

Ocular gnathostomiasis is a serious manifestation that has been increasingly found in recent years.[1,3] The highest incidences of ocular diseases occur in Japan, Thailand, and India, in decreasing order of frequency.[3] The ophthalmologic sites for this infection are the eyelids, conjunctiva, anterior chamber, vitreous cavity and subretinal space. Only two cases of a subretinal location have been reported.[4,5] To the best of our knowledge, the present case is the first to demonstrate successful focal laser photocoagulation of a moving *Gnathostoma* nematode in the subretinal space with long-term follow-up.

2. Case report

A 29-year-old Thai man complained of blurred vision and floaters in his left eye for two weeks. He had previously been diagnosed with panuveitis and referred for a retinal specialist consultation. He had a history of regularly consuming raw meat and also reported a history compatible with cutaneous larva migrans which disappeared without treatment. The best-corrected visual acuity (BCVA) of his left eye was

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20/30, and an ocular examination showed medium-sized keratic precipitates, koeppe nodules, anterior chamber cells (2+) and flare (2+), and anterior vitreous cells (3+). Funduscopic examination of the left eye showed multiple subretinal granular appearance at the midperiphery of the retina with focal optic nerve swelling on the superior part of the disc.

No notable skin lesions were seen on the systemic examination. A hematological investigation showed no leukocytosis and no eosinophilia. A stool examination was negative for intestinal parasites. However, owing to the history of raw meat consumption, possible larva migrans and the fundus appearance suggestive of subretinal tracks on the superior and temporal retina (Fig. 1, Left), diffuse unilateral subacute neuroretinitis (DUSN) was suspected. Oral albendazole and topical steroid eye drops were then prescribed. On the follow-up visit five days later, we observed the progression of a white track crossing underneath the superotemporal retinal vein over a period of 20 minutes, which was proved to be an active parasite on fundus photography (Fig. 2) and spectral domain optical coherence tomography (SD-OCT; Fig. 3). Fundus fluorescein angiography (FFA) (Fig. 1, Right) revealed multiple linear tracks of the parasite at the superior and temporal retina more prominent than the color fundus photographs. Some fluorescein leakages at the disc and retinal periphery were identified.

Frequency-doubled Nd:YAG laser (wavelength 532 nm) was applied firstly around, and subsequently over, the parasite (Fig. 4). A static worm was confirmed without any more movements (Fig. 4B and C). Albendazole and naproxen were then continuously prescribed for four weeks. Immunoblotting analysis for the common parasitic infections including *Angiostrongylus*, *Echinococcus*, *Filariae*, *Strongyloides*, and *Gnathostoma* was positive for *Gnathostoma* species.

During the follow up after the laser photocoagulation, no significant intraocular inflammation was observed. The BCVA came back to 20/25 and the vitreous cell reduced to 1+ at 3 months. The retinal laser scar became fibrotic with visible white thread of parasitic debris in the center. The patient has been followed for five years after the treatment. The fibrotic laser scar remained stable without abnormal systemic neurological signs and symptoms.

3. Discussion

We described a unique case of early stage DUSN. A living serologically confirmed *Gnathostoma* nematode was identified in the subretinal space and successfully treated with focal laser photocoagulation.

Gnathostoma spp. is a rare species that causes cutaneous larva migrans. Other more common species include *Ancylostoma caninum* and *Ancylostoma braziliense*.[6] However, in Thailand, half of the patients who were infected with Gnathostoma spp. initially presented with ocular invasion by the worm.[7] Eosinophilia may be present in 50%–70% of the cases with systemic infection but normal eosinophil level does not exclude the infection, especially in cases with ocular involvement.[8] Serological diagnosis is the gold standard for identification of gnathostomiasis in endemic areas including Thailand. ELISA analysis has low sensitivity and specificity and could show a cross reactivity with other nematodes, while immunoblotting analysis for *Gnathostoma* has much higher sensitivity (91.6%) and specificity (87.8%).[9] Although a surgical removal of the worm for histopathologic studies could be the most accurate method for nematode identification, it would require an urgent vitrectomy with risks of postoperative complications.

Multimodal retinal imaging suggested that the parasite might enter the globe at the superior border of the optic disc causing focal papillitis and continued to the superotemporal quadrant of the fundus through the subretinal space, leaving multiple whitish tracks behind as the worm moved. No hemorrhages from the choroid were identified to suggest parasitic invasion from the choroid. The reveal of the living subretinal worm could be precipitated by the effect of the systemic albendazole prescribed five days prior. A similar effect was reported in a study of cutaneous gnathostomiasis, in which outward movement of the *Gnathostoma spinigerum* into the dermis occurred after oral albendazole.[10,11] Therefore, ophthalmologists should be aware of possible progressive movement of the worm into the neurological system after prescribing systemic antiparasitic drugs and a low threshold of neurologist consultation is required when there is abnormal neurological presentation.

The length of *Gnathostoma* spp. ranges from 2 mm. to 2 cm. varying on the species.[12] In our case, we measured the length of the nematode which was approximately 2.6 mm. However, the whitish coiling track of the worm could mislead us that the worm itself looked longer than usual. The real-time movement of the worm we observe could help delineate the actual length of the worm.

To date, there have been only a few reported cases of DUSN caused by subretinal gnathostomiasis; one was treated with oral albendazole,[4] and the other was surgically removed by pars plana vitrectomy.[5] Other nematodes associated with DUSN include *Strongyloides stercoralis, Toxocara canis, Baylisacaris procyonis, Ancylostoma caninum*, and *Brugia malayi*. The effective use of focal laser photocoagulation has been demonstrated in early stage of DUSN patients.[13,14] A single shot at the advancing end of the nematode such as filariasis[13] could prevent the further progression of the worm. We elaborated in our case that focal laser photocoagulation is an effective treatment for DUSN caused by subretinal gnathostomiasis with long-term safety. To the best of our knowledge, focal laser photocoagulation of the living subretinal

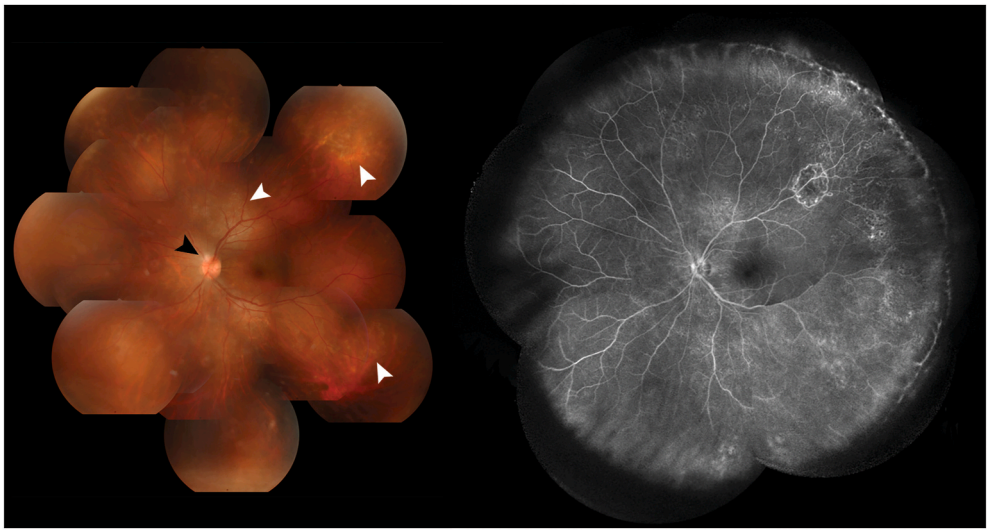

Fig. 1. Fundus photographs (Left) at the first visit shows multiple subretinal tracks (white arrowheads) at superior and temporal retina and focal papillitis (black arrowhead) at the superior part of the optic nerve head. A montage of fundus fluorescein angiography (Right) after the laser treatment shows more prominent abnormal hyperfluorescent tracks compared with the color fundus photograph with some leakages of fluorescein dye at the disc and retinal periphery. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

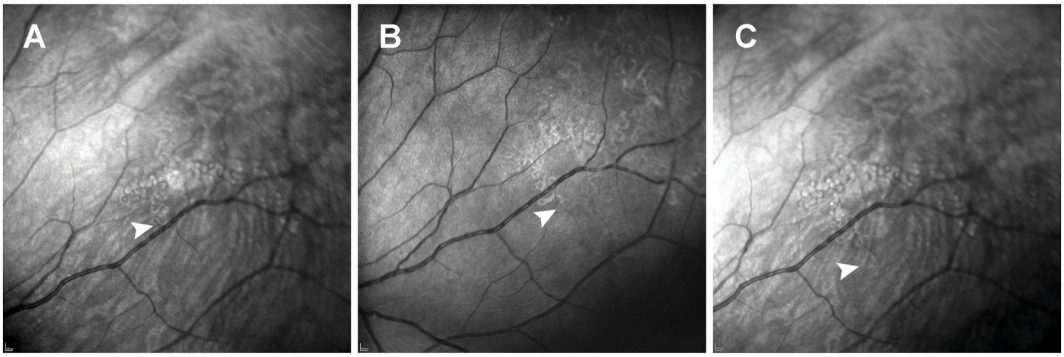

Fig. 2. The serial fundus photographs taken by infrared technique show the progression of the subretinal linear lesion of the living parasite (arrowhead). Figure (C) was taken 20 minutes after figure (A).

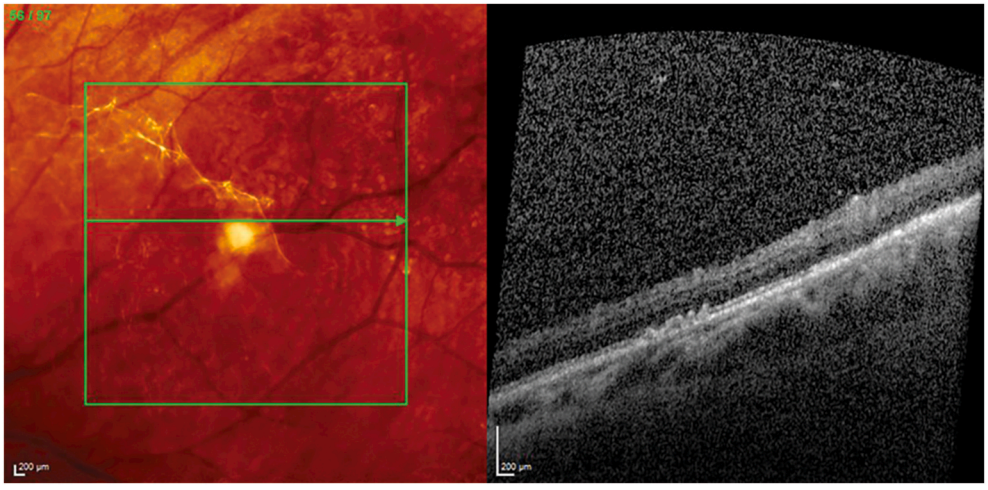

Fig. 3. Spectral domain optical coherence tomography before focal laser photocoagulation. shows multiple subretinal hyperreflective elevations corresponding with the living nematode.

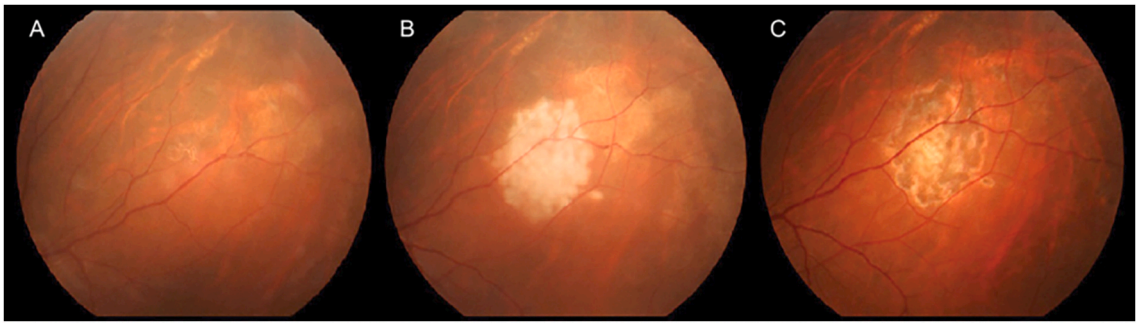

Fig. 4. The multiple color fundus photographs at different time points: (A) before the laser treatment, (B) immediately after the laser treatment and (C) 2-week post laser treatment with the static worm identified at the center of the laser scar. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

Gnathostoma nematode has never been described in the literature.

Systemic corticosteroids are generally prescribed to prevent intraocular inflammation after destruction of parasites.[13,14] However, we did not observe any aggravated inflammatory responses with the use of non-steroidal anti-inflammatory (NSAID) agent. Hence, oral steroid treatment might not be necessary for cases treated with focal laser photocoagulation and the side effects of systemic steroids could be avoided.

We suggested that focal laser photocoagulation to stop the migration of a living subretinal *Gnathostoma* spp. worm was a preferred treatment over creating a retinal hole and surgical removal by vitrectomy. The procedure was relatively simple, could be performed on an outpatient basis by general ophthalmologists, and complications of vitrectomy could be avoided.

4. Conclusions

A focal laser photocoagulation applied to the nematode without systemic steroids could be a successful treatment for a living subretinal gnathostomiasis without a long-term complication.

Summary statement

A 29-year-old male with panuveitis whose fundus showed multiple subretinal tracks in the affected eye. A moving subretinal parasite was identified after oral albendazole administration with a serologic result was positive for *Gnathostoma* species. A focal laser photocoagulation at the nematode was performed, oral albendazole continued and naproxen prescribed. The ocular inflammation subsided within three months. The patient has been followed for five years without local and systemic complications.

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Intellectual property

We confirm that we have given due consideration to the protection of intellectual property associated with this work and that there are no impediments to publication, including the timing of publication, with respect to intellectual property. In so doing we confirm that we have followed the regulations of our institutions concerning intellectual property.

Research ethics

We further confirm that any aspect of the work covered in this manuscript that has involved human patients has been conducted with the ethical approval of all relevant bodies and that such approvals are acknowledged within the manuscript.

Written consent to publish potentially identifying information, such as details or the case and photographs, was obtained from the patient(s) or their legal guardian(s).

Authorship

All listed authors meet the ICMJE criteria.
We attest that all authors contributed significantly to the creation of this manuscript, each having fulfilled criteria as established by the ICMJE.

We confirm that the manuscript has been read and approved by all named authors.

We confirm that the order of authors listed in the manuscript has been approved by all.

Declaration of competing interest

None of the authors have any financial/conflicting interests to disclose.

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