Ocular gnathostomiasis in Myanmar: two cases and literature survey

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Background: To report two cases of indigenous ocular gnathostomiasis in Myanmar with the update of the current status of this disease in Myanmar by literature survey.
Results: Two middle-aged women, 55-year-old from Pan Ta Naw and 40-year-old living in Yangon, Myanmar, presented individually to two different hospitals with several weeks history of blurred vision. In both cases, a live worm of about 4-5 mm was found moving in the anterior chamber of the eye in association with inflammatory cell infiltration. The worm in the anterior chamber of each patient was successfully removed by microscopic surgery. The visual acuity of the patients returned to normal after removal of the worm. Literature survey revealed that, including two cases reported here, the number of cumulative cases of ocular gnathostomiasis in Myanmar exceeds 20 cases, the most number recorded in the world.
Conclusions: Since ocular gnathostomiasis is an extremely rare form of gnathostomiasis, thousands of cutaneous and/or visceral cases have been undiagnosed in Myanmar. Nationwide epidemiological surveys for gnathostomiasis of any form should be performed.

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Introduction

Nematode parasites in the genus *Gnathostoma* are found in wild animals in various parts of the Americas, Eurasia, Oceania and Africa[1] and is known as the representative pathogen of fish-borne zoonotic helminthiases. In Asia[3], species namely *G. spinigerum*, *G. doloresi*, *G. hispidum* and *G. japonicum* are identified to infect humans, whereas *G. binucleatum* is a sole pathogen in the Americas. As for the human cases in Asia, Japan, the People's Republic of China and Thailand have long been known as the major endemic areas of human gnathostomiasis[2]. In addition, several Asian countries, such as Lao PDR[3], Vietnam[4] and India[5] have recently been proven as the gnathostomiasis endemic countries. Moreover, after an extensive local literature survey up to 2016[6], we have revealed that gnathostomiasis is endemic also in Myanmar.

Humans acquire gnathostomiasis mainly by ingesting uncooked or undercooked fish meat contaminated with the 3rd stage larvae (L3) of *Gnathostoma* species[1]. Although rare, infection after eating raw snake meat has been reported as an alternative route of infection[7]. After being ingested by humans, the third stage larvae (L3) of *Gnathostoma* preferentially migrate to subcutaneous connective tissues causing migratory erythema or serpiginous creeping eruption, and thus, gnathostomiasis is primarily known as a disease of the skin[1]. However, as a nature of zoonotic nematodiases, the larvae occasionally migrate into unexpected sites, such

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as the liver, lungs, brain[8] and eyes[5,9] to cause deleterious outcome[1].

In Myanmar, when an outbreak of gnathostomiasis had occurred among Korean residents in Yangon[10], all cases manifested cutaneous lesions. In contrast, in our recent extensive literature search on human gnathostomiasis in Myanmar up to 2016, the majority of indigenous gnathostomiasis cases (over 10 of about 15 estimated cases) were ocular cases[6]. Here we add two more cases of ocular gnathostomiasis we have recently experienced in Myanmar. In addition, 5 more indigenous ocular gnathostomiasis cases, which were reported in a poster in the local ophthalmology meeting, are added in the list and current status of ocular gnathostomiasis in Myanmar is discussed.

Case #1

A 55 year-old female from Pan Ta Naw Township, Ayeyarwadi Division, Myanmar, came to the out-patient ward of the Ophthalmology, North Okkalapa General/Teaching Hospital, Yangon, with the complaint of blurring vision and redness of her left eye for 2 weeks. The patient and her families sold freshwater and marine fish in the local market, where she often prepared and consumed undercooked/fermented fish (Burmese Name: Nga-Chin) with her family. Her visual acuity of the right eye was 6/36 (UA) and the left eye was hand motion. On slit lamp examination for the left eye, the conjunctiva was severely congested, cornea was hazy with massive cell infiltration. A live, actively moving worm of about 5 mm in length was seen in the anterior chamber of the left eye (Figure 1A). On the next day, the worm was removed under a surgical microscope and identified as a *Gnathostoma* L3 (Figure 1B), based on primarily the typical 4 rows of hooklets on the head bulb (Fig. 1B inset).

She was given an oral dose of 400 mg of albendazole for the precaution of the possible residual worms. After surgical extirpation of the worm, her visual acuity as well as ocular symptoms gradually improved and eventually normalized.

Case #2

One of the authors, Mya Mya Lwin, has received a short and thick nematode parasite of about 4-5 mm in length for the identification of the pathogen from a private hospital in Yangon, Myanmar. The worm was removed from the anterior chamber of an eye (affected side unknown) of a 40- year-old female patient who has visited a neuro-physician because of visual disturbance for several weeks. The patient was a housewife who often prepared and consumed freshwater fish dishes with catfish, snakehead fish and *Tilapia* species, etc., in particular, undercooked fermented fish (Nga-Chin). The worm was surgically removed from her eye (Fig. 2) which was about 4.0 x 0.5 mm (length x width), with a distinct head bulb equipped with 7-8 rows of hooklets (Fig. 2 inset).

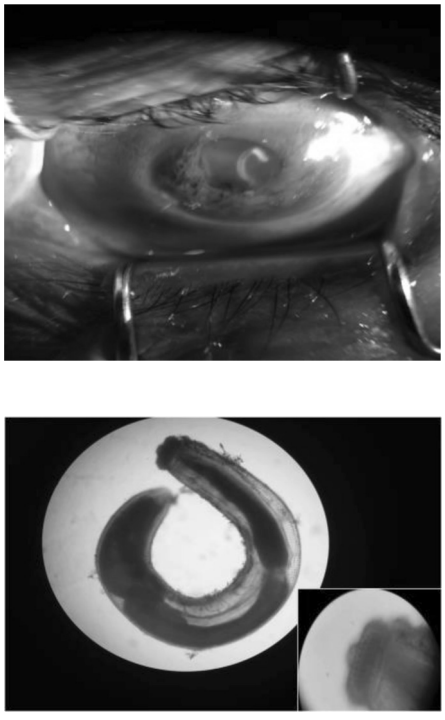

Figure 1: A worm floating in the anterior chamber of the left eye of the patient #1 (top) and the worm removed after surgery (bottom). Inset shows the close-up view of the head bulb showing 4 lines of hooklets.

The body is covered with fine cuticular spines and the inside contains reddish body fluid and dark-brown coloured intestine. Based on these features, the worm was identified as a young adult of *Gnathostoma spinigerum.* Visual acuity of the patient returned to normal after the surgery.

Figure 2: A surgically extirpated worm from the anterior chamber of the eye (side unknown) of the patient #2. Inset shows the close up view of the head bulb showing 8 lines of hooklets.

Discussion

While we have surveyed for gnathostomiasis cases in the proceedings and recent medical publications in Myanmar, we have found that Thi Thi Htoon et al. (2018) reported 12 ocular helminthiasis cases over a 9-year period (2009-2017) including 5 cases of ocular gnathostomiasis as a poster presentation at the 30th Myanmar Ophthalmological Conference 2018, held on the 15-16th October 2018, in Yangon, Myanmar. Thus, taking our two cases here and the 5 cases of Thi Thi Htoon (2018) together with the cases in our previous literature survey[6], a total of minimum 19, probably over 20, ocular gnathostomiasis cases so far were found in Myanmar (Table 1) since two classical cases[11,12] were reported in the 1960s.

As for the clinical manifestation of gnathostomiasis, cutaneous lesions, which can present as migratory oedema or serpiginous creeping eruptions, are the most common symptoms[1]. Although the frequency of ocular cases is far lower than that of cutaneous gnathostomiasis, high incidence of ocular gnathostomias (over 10 cases) has been reported in India, Japan, Mexico, and Thailand[5,9]. Now we may consider Myanmar as the country with the highest recorded incidence of ocular gnathostomiasis (Figure 3).

While Japan, Mexico and Thailand are known as highly endemic areas of gnathostomiasis, India and Myanmar are usually not listed in the high endemic countries. As is pointed out previously[5,9], it might be merely an ignorance of cutaneous and/or visceral cases. In fact, reports of cutaneous cases are recently emerging in India[13,14]. In Myanmar, collaboration of dermatologists is necessary if we are to elucidate an accurate situation of gnathostomiasis in general in Myanmar.

In the present study, the worm obtained from the patient #1 was morphologically a typical advanced third stage infective larva (AL3) of *G. spinigerum,* having 4 lines of hooklets on the head bulb and the whole body was covered with fine cuticular spines. In contrast, the worm recovered from the patient #2 has 8 lines of hooklets on the head bulb, a typical morphological feature of adult *Gnathostoma* worms. Generally, gnathostomiasis is known as a disease caused by the larval stage of *Gnathostoma* parasites. However, in Asia, *G. spinigerum* adult worms are occasionally found in the skin lesion of the human cases[15]. In the previous study, the worms recovered from the patients' eye are almost exclusively advanced 3rd stage infective larvae of *Gnathostoma*, our case suggests that *G. spinigerum* can develop into a young adult at least.

Although consuming raw fish and meat is not considered a traditional food habit in Myanmar[16], some Myanmar people lik our two present cases have a traditional custom of eating raw/undercooked fish. Previous study of Korean parasitologists showed high infection rate of freshwater fishes with *Gnathostoma* larvae sold in the markets in Myanmar[17,18]. Thus, at this occasion, one of the authors, Aung Phyo Wai, examined swamp eels sold in the market in Yangon and found high incidence of infection with significant seasonal variation (Table 2). Although this is a small scale survey, the results show that eating freshwater fish raw is a high risk of gnathostomiasis in this country.

Conclusion

We reported herewith two cases of ocular gnathostomiasis in Myanmar, together with 5 additional cases recently reported in the poster session of the local ophthalmology meeting. As shown in Table 1, over 20 cases of ocular gnathostomiasis have been accumulated in Myanmar. Therefore, not only ophthalmologists but also dermatologists, general physicians and home doctors in Myanmar should be aware of

the presence of this disease. Moreover, extensive epidemiological surveys not only of humans but also of intermediate fish/paratenic host animals is required to clarify the route of infection to humans and the maintenance of the parasite life cycle in this country to prevent the spreading of this disease.

Table 1: Indigenous ocular gnathostomiasis cases in Myanmar.

No.	Age (years)	Gender	Affected site	Year of infection	Reference
1	28	Male	Left eye	1958	Reported in ref. #6
2	48	Male	Left eye	1964	
3	N/A	Female	Left eye	1980?	
4-7	3-4 ocular cases during last 20 years			last 20 years	
8	ocular cases in National Health Laboratory			2005	
9-10	2 ocular cases (one worm each)			2007	
11	N/A	Male	Right eye	2001	
12	10	Male	Right eye	2013	
13	43	F	Anterior chamber	2010	Thi Thi Htoon (2018) Poster presentation
14	16	M	Anterior chamber	2012	
15	13	M	NA	2014	
16	41	F	Right eye	2016	
17	53	M	NA	2017	
18	55	F	Left eye (anterior chamber)	2018	
19	40	F	Side unknown, anterior chamber	2019	

N/A, not available.

Table 2: *Gnathostoma spinigerum* larvae in Asian swamp eels from local markets of Yangon, Myanmar

Year	Seasons	No. of eels Infected/examined	*Gnathostoma* L3 Total (per fish)	Highest No. of L3 per fish
2016	Rainy (Jun-July)	32/50 (64.0%)	185 (5.8)	69
	Cold (Nov-Dec)	27/65 (41.5%)	117 (4.3)	42
2017	Hot (Apr)	37/150 (24.7%)	85 (2.3)	8
2019	Cold (Jan – Feb)	61/150 (40.7%)	233 (3.8)	21
	Hot (Mar)	37/110 (33.6%)	76 (2.1)	14
	Rainy (Jun -Aug)	81/135 (60.0%)	452* (5.6)	52
Total		275/660 (41.7%)	1148 (4.2)	

*: both early and advanced 3rd larvae
Almost all *Gnathostoma* larvae were found in the liver of Asian swamp eels and only a few from the intestines of eels.

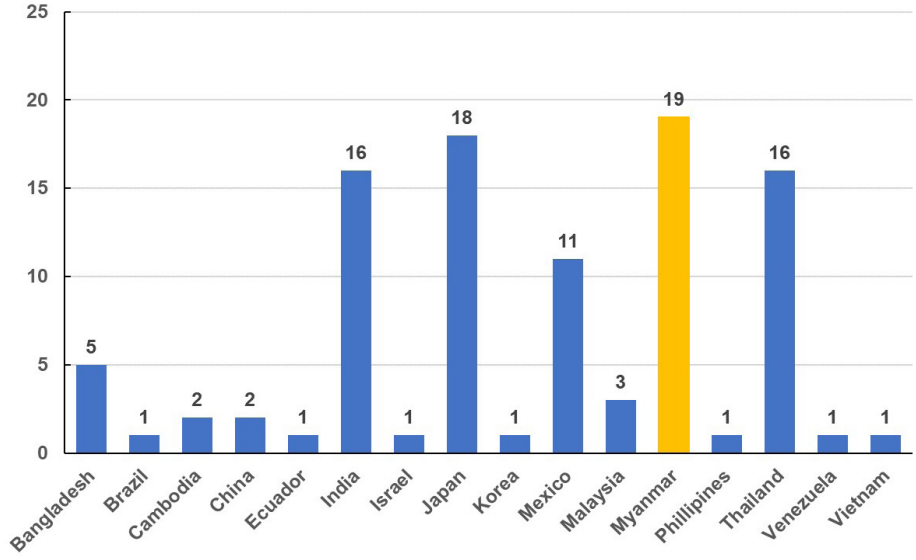

Figure 3: Cumulative distribution of ocular gnathostomiasis cases in each country. Modified from Fig. 1, Nawa et al. 2017 Am J Trop Med Hyg (Ref. #9) with permission.

References

1. Nawa Y, Maleewong W, Intapan PM, Díaz-Camacho SP. Chapter 21. *Gnathostoma.* In; Biology of Food-borne Diseases. (eds. Lihua Xiao, Una Ryan, Yaoyu Feng) CRC Press, Taylor & Francis (Boca Raton, London, NY). 2015; pp.405-426,
2. Miyazaki I. On the genus *Gnathostoma* and human gnathostomiasis, with special reference to Japan. Experimental Parasitology. 1960 Jun 1;9(3):338-70.
3. Vonghachack Y, Dekumyoy P, Yoonuan T, Sa-nguankiat S, Nuamtanong S, Thaenkham U, et al. Sero-epidemiological survey of gnathostomiasis in Lao PDR. *Parasitol Int* 2010;59:599-605.
4. Xuan LT, Hoa PTL, Dekumyoy P, Hoan NH, Khuong LH, Van TTH, et al., *Gnathostoma* infection in *South Vietnam. Southeast Asian J Trop Med Publ Health* 2004;35(suppl 1):97-9.
5. Nawa Y, Katchanov J, Yoshikawa M, Rojekittikhun B, Dekumyoy P, Kusolusuk T, et al. Ocular gnathostomiasis: A comprehensive review. *J Trop Med Parasitol* 2010;33:77-86.
6. Wai AP, Maw WW, Moe AC, Boonmars T and Nawa Y. Human gnathostomiasis in Myanmar: a review of local literature. *Southeast Asian J Trop Med Publ Health* 2018; 49(4):543-548.
7. Kurokawa M, Ogata K, Sagawa S, Miyaoka Y, Noda S, Nawa Y. Cutaneous and visceral larva migrans due to *Gnathostoma doloresi* infection via an unusual route. Archives of dermatology. 1998;134(5):638-9.
8. Katchanov J, Sawanyawisuth K, Chotmongkol V, Nawa Y. Neurognathostomiasis, a neglected parasitosis of the central nervous system. Em Infect Dis. 2011;17(7):1174-1180.
9. Nawa Y, Yoshikawa M, Sawanyawisuth K, Chotmongkol V, Figueiras SF, Benavides M, et al. Ocular gnathostomiasis—update of earlier survey. The American journal of tropical medicine and hygiene. 2017 Oct 11;97(4):1232-4.
10. Chai JY, Han ET, Shin EH, Park JH, Chu JP, Hirota M, *et al.* An outbreak of gnathostomiasis among Korean emigrants in Myanmar. *Am J Trop Med Hyg* 2003;69(1):67-73.
11. Gyi K. Intra-ocular gnathostomiasis. *British J Ophthalmol* 1960;44:42-45.
12. Khin T. Intra-ocular gnathostomiasis. *British J Ophthalmol* 1968;52(1):57-60.
13. Mukherjee A, Ahmed NH, Samantaray JC, Mirdha BR. A rare case of cutaneous larva migrans due to *Gnathostoma sp. Indian J Med Microbiol* 2012;30:356-8
14. Subhedar V, Nandedkar S, Jain M, Bajpai

T. (2014). Gnathostomiasis: a rare case of cutaneous creeping eruptions. *Ind J Dermatopathol Diag Dermatol* 2014; 1(1), 32-34.

15. Phetsouvanh R, Habe S, Newton P, Vongsouvaht M, Horii Y, Doanh PN, et al. Spontaneous emergence of a *Gnathostoma spinigerum* adult worm from the abdominal skin of a Laotian woman: a case report. Southeast Asian Journal of Tropical Medicine and Public Health. 2018;49(1): 1-5.
16. Khin Maung Win. Eating raw fish is not a Myanmar food habit. *Health Digest 2004;* 2004; 1(32,33): April 21st and 28th.
17. Jung BK, Lee JJ, Pyo KH, Kim HJ, Jeong HG, Yoon CH, *et al.* Detection of *Gnathostoma spinigerum* third-stage larvae in snakeheads purchased from a central part of Myanmar. *Korean J Parasitol* 2008;46(4):285-288.
18. Chai JY, Sohn WM, Na BK, Park JB, Jeoung HG, Hoang EH, *et al.* Larval *Gnathostoma spinigerum* detected in Asian swamp eels, Monopterus albus, purchased from a local market in Yangon, Myanmar. *Korean J Parasitol* 2015;53(5):619-625.