Cutaneous Gnathostomiasis: A Case Report from Thailand

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ABSTRACT

Gnathostomiasis is transmitted to humans via ingestion of third-stage larva of genus Gnathostoma. Thailand is considered an endemic area for the disease. We reported a 65-year-old Thai male patient with clinical presentation of a small number of nodular migratory swellings on the face and neck. The histopathology of skin biopsy revealed parasitic material surrounded by eosinophil infiltration and fibrosis in deep dermis. Therefore, we were able to confirm diagnosis of cutaneous gnathostomiasis. After treatment with ivermectin followed by albendazole, the lesions still persisted; thus, surgical excision of lesions may be the necessary curative treatment.

Keywords: Gnathostomiasis, migratory nodule, Thailand

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INTRODUCTION

Gnathostomiasis is a food-borne systemic parasitic infection. The disease is transmitted to humans via ingestion of third-stage larva of genus Gnathostoma, most notably G. spinigerum, which is commonly found in insufficiently cooked fish and other intermediate hosts. Gnathostomiasis-endemic regions and countries include Southeast Asia and Latin America, especially Thailand, Japan and Mexico.[1-2] From Thailand, we report a case of cutaneous gnathostomiasis with clinical presentation of nodular migratory swelling and parasitic material surrounded by eosinophil infiltration and fibrosis from skin biopsy specimens. The histopathology confirmed diagnosis of gnathostomiasis.

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CASE REPORT

A 65-year-old Thai male patient presented with a small number of painless migratory erythematous subcutaneous nodules with mild itching on the right infra-orbital region and on the right anterior neck for one week (Fig 1A). Underlying diseases included well-controlled type-2 diabetes mellitus and allergic rhinitis. A provisional diagnosis of angioedema was made on his first visit and he was prescribed prednisolone 20 mg/day, fexofenadine 180 mg/day, and loratadine 10 mg/day.

After two weeks, an erythematous nodule migrated from the right infra-orbital region to the right forehead, with persistence of lesion at the right anterior neck. The patient denied history of eating raw food. He reported no diarrhea, nausea, vomiting, or other systemic symptoms. Initial laboratory tests revealed normal leukocyte count (6,220 cells/uL) and elevated eosinophil count (13.5%, 840 cells/uL). Blood testing for Gnathostoma antibody by Western blot analysis

was positive. Stool exam by direct wet smear indicated no worm eggs or larvae. With history of nodular migratory lesions, high eosinophilia, and positive Gnathostoma antibody, a presumptive diagnosis of cutaneous gnathostomiasis was made. Accordingly, the patient was prescribed ivermectin 200 μg/kg for two consecutive days.

One week later, lesions persisted on the right side of his forehead and the right side of his anterior neck, suggesting the ineffectiveness of ivermectin in this case. As an alternative treatment, we prescribed albendazole 400 mg twice daily for 21 days.

Approximately one month after albendazole was started; the lesions on the right side of his forehead and the right side of his anterior neck were still evident, even though blood eosinophil count was decreased from 13.5% to 6.1% (absolute eosinophil count 353 cells/uL). An excisional skin biopsy from the neck showed existence of degenerated parasitic material embedded in granuloma, with eosinophil infiltration and fibrosis in deep dermis. Moreover, the cuticle with cuticular spines on the surface was noted (Fig 1B). The histopathology was compatible with gnathostomiasis, so cutaneous gnathostomiasis was definitively diagnosed. A second excisional biopsy from the right side of his forehead revealed subcutaneous fibrosis with eosinophil infiltration. Ultimately, the lesions were surgically removed.

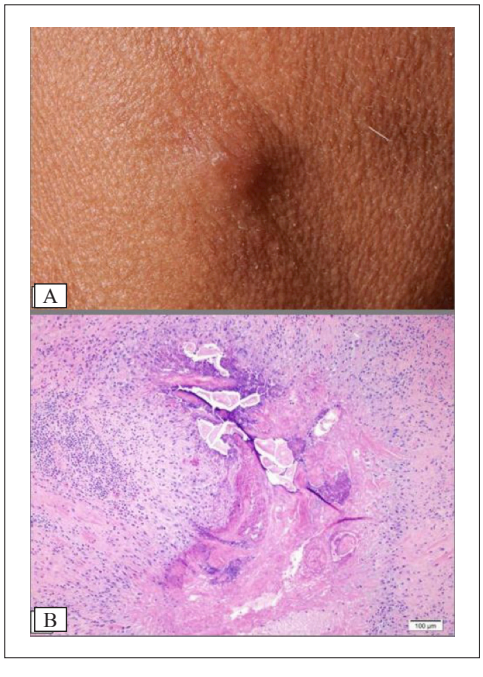

Fig 1. A: Subcutaneous erythematous nodule on right anterior neck; **B:** Section shows degenerated parasitic materials embedded in granuloma with eosinophil infiltration and fibrosis (hematoxylin and eosin; 100x)

DISCUSSION

Gnathostomiasis is a nematode infestation that causes an intermittent cutaneous or visceral larva migrans, resulting from an inflammatory reaction provoked by third-stage larval migration through human tissues. Duration from time of ingestion of parasite-infected raw fresh-water fish to the development of skin lesions varies from one to twenty weeks.[1]

Various studies have reported criteria that are highly suggestive of cutaneous gnathostomiasis diagnosis. Reported criteria include: (i) cutaneous manifestations attributable to gnathostomiasis, such as nodular migratory swelling or panniculitis, creeping eruption, and pseudofurunculosis; (ii) obvious exposure risk (e.g., history of consuming raw fish or other intermediate hosts and/or recent return from an endemic area); (iii) laboratory results with absolute eosinophilia count >500/mm^3, a positive finding of antibodies specific to Gnathostoma spp, by Western blot testing, and stool examination negative for other parasites.[3-5]

In our case, the erythematous subcutaneous nodule with swelling and mild itching predominately on the face led us to misdiagnose angioedema. After observing migration of a swelling nodule from the right infra-orbital region to the right side of his forehead and considering that Thailand is an endemic area, a presumptive diagnosis of cutaneous gnathostomiasis was indicated. We also looked for other factors consistent with a definitive diagnosis, including high absolute eosinophil count and positive for Gnathostoma-specific antibodies from blood testing. Additionally, the reduced absolute eosinophil count after 1 month of treatment using antihelminthic drugs in this patient also supported the diagnosis of parasitic infection.

The identification of Gnathostoma in a lesional skin biopsy specimen was required for certain diagnosis. Cross-sectional examination of intestinal canal for larva and presence of parasitic

material surrounded by a thin fibrous cuticle are the key points to identify Gnathostoma species[2,6], as shown in our case. A previous study reported the probability of detecting larva on a section is approximately 34%.[7] In most cases of cutaneous gnathostomiasis, biopsy specimens have failed to identify larva due to the migratory nature of the lesions.[3] Although it is possible that the histopathology (including eosinophilic panniculitis, eosinophilic cellulitis, and eosinophilic vasculitis) suggests tissue reaction, the result is insufficient to be considered a helpful diagnostic sign.[2]

Although it is generally difficult to do, we were able to successfully identify parasitic substance from skin biopsy in our case. One possible explanation for this success was the ivermectin and albendazole treatments given to the patient prior to the biopsy. Alvaro C, et al, reported that treatment with albendazole may stimulate development of papule or pseudo furuncle holding larva. In subsequent biopsy, the probability of identifying larva would be increased.[8]

Regarding treatment, cure rates from the use of ivermectin and albendazole are relatively high, but vary from several studies. One recent study reported cure rates of 93.9% and 94.1% in the patients receiving albendazole 400 mg twice daily for 21 days and 400 mg once daily for 21 days, respectively. Thus, the effectiveness of albendazole is not necessarily dose-specific.[5]

Nontasut P, *et al*, found that a single or double dose of ivermectin 0.2 mg/kg is expected to result in a 76%-95% cure rate.[9] Regarding the effectiveness of ivermectin versus albendazole, one study reported that treatment with albendazole 800 mg/day for 3 weeks resulted in a cure rate of 78.5%. In a relapse case that received albendazole from the same study, the disease was completely cured after the patient was prescribed ivermectin. In contrast, treatment with a double dose of ivermectin 0.2 mg/kg yielded a cure rate of 100%.[10] Thus, ivermectin and albendazole have similar levels of effectiveness, although albendazole has lower incidence of adverse effects than ivermectin.

In conclusion, this report profiles a case of cutaneous gnathomiasis in Thailand, based on clinical manifestation of migratory subcutaneous nodule, hypereosinophilia, positive serological finding for anti-Gnathostoma antibodies, and larva from histopathology. After unsuccessful initial treatment with ivermectin we prescribed albendazole for the second course of treatment. Surgical excision of lesions may also be the necessary curative treatment.

Author Disclosure Statement

All authors have no conflict of interest to declare.

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