

SPONTANEOUS EMERGENCE OF A *GNATHOSTOMA SPINIGERUM* ADULT WORM FROM THE ABDOMINAL SKIN OF A LAOTIAN WOMAN: A CASE REPORT

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Abstract. Gnathostomiasis caused by infection with the Spirurine nematode, *Gnathostoma* species, is a common fish-borne parasitic zoonosis in Asia. We present here the case of the spontaneous emergence of an adult *Gnathostoma spinigerum* worm from the abdominal skin of a Laotian woman. We review the literature on gnathostomiasis and discover that infective *G. spinigerum* larvae can grow into immature and mature worms in humans more commonly than expected.

Keywords: *Gnathostoma spinigerum*, adult worm, skin lesion, spontaneous emergence

INTRODUCTION

Gnathostomiasis is a disease caused by the nematode parasites belonging to the genus *Gnathostoma* (Nawa *et al*, 2015). It is a disease primarily of the skin and less frequently of the visceral organs (Nawa *et al*, 2015). The third stage larvae (L3) of *Gnathostoma* migrate through the tissues of the human body, preferentially to the skin, causing acute and chronic inflammation (Nawa *et al*, 2015). Human infection

occurs when people ingest the intermediate/paratenic hosts harboring *Gnathostoma* L3 (Nawa *et al*, 2015). Countries endemic for gnathostomiasis are found in Asia and Latin America, where people have a custom of eating raw or undercooked fish or amphibians (Nawa *et al*, 2015).

It is generally believed humans are not suitable definitive hosts for *Gnathostoma* species; L3 ingested by humans usually do not develop into adult worms (Nawa *et al*, 2015). Recovery of *Gnathostoma* adult worms from humans has been reported occasionally (Daengsvang, 1980; Miyazaki, 1991). We report here the case of the spontaneous emergence of an adult *Gnathostoma* worm from the abdominal

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Fig 1–Serpiginous eruption on the abdominal wall of the patient.



Fig 2–Low power view of the worm spontaneously emerged from the skin lesion.

skin of a Laotian woman. The parasite was identified morphologically and molecularly as a young adult *G. spinigerum* worm. We review the literature for cases where adult *Gnathostoma* worms are recovered from humans.

the anterior abdomen (Fig 1), otherwise was noted to have a normal examination.

Examination of the worm revealed it was 3 x 9 mm (Fig 2). Morphology under microscopy revealed the head bulb and the neck were buried in the body, with

CASE REPORT

A 32 year old Laotian female office worker living in Vientiane Province, Lao PDR presented to Mahosot Hospital, Vientiane, on 18th August 2007 complaining of a pruritic, painful serpiginous lesion on the skin of the anterior abdomen (Fig 1) for several weeks. Before admission, the patient ate “*Koi Plar*”, a local dish prepared with minced raw fish and spices marinated in lime juice. Although other family members also had this dish, none of them developed clinical symptoms. She has no history of any major medical problem. In the hospital, while waiting to be examined, a worm emerged through the skin of the anterior abdominal wall. On examination, the patient was noted to have a superficial serpiginous track on

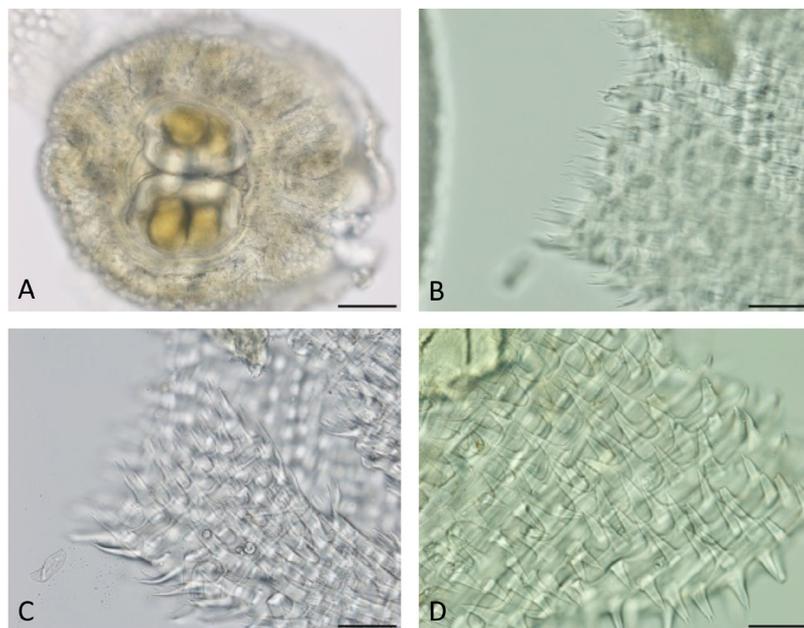


Fig 3—High power view showing characteristic features of a *Gnathostoma spinigerum* adult worm. A, head bulb having 8-9 rows of hooklets, scale bar=100 μ m. B-C, transitional morphological changes of the cuticular spines from trident (B), bifurcated (C) to single (D) spines on the anterior half of the worm body, scale bar=50 μ m.

5-6 lines of hooklets visible (Fig 3). The anterior half of the body was covered with cuticular spines and those on the anterior-most part had a trident shape, consistent with an adult *G. spinigerum* worm.

To identify the species, polymerase chain reaction (PCR) sequencing was carried out as described previously (Ando *et al*, 2006). DNA was extracted from a small piece of the tail of the worm, and the ribosomal internal transcribed spacer 2 (ITS2) sequence was amplified using a primer set for the LC1 (forward) and HC2 (reverse) sequences. A partial sequence of mitochondrial cytochrome oxidase 1 (Cox1) was also amplified using a primer set of the FH5 (forward) and MCO1B (reverse) sequences. The PCR conditions followed to those described previously

(Ando *et al*, 2006). The amplicons were purified using agarose gel electrophoresis and both directions were directly sequenced using the big dye termination method (Ando *et al*, 2006). The obtained sequences were analyzed to determine species by homology search using the BLAST (Basic Local Alignment Search Tool) of the GenBank server of the National Center for Biotechnology Information and by alignment using the ClustalX (Ando *et al*, 2006). Sequence identity of the Cox1 of the *Gnathostoma* worm

examined was a 99.7% (389/390) match with the sequence accession number AB180099 (*G. spinigerum* Cox1 gene) in the GenBank, and ITS2 was a 99.5% (601/604) match with sequence accession number AB181155 (*G. spinigerum* ITS2 sequence) in the GenBank, confirming the study worm was *G. spinigerum*. After the worm spontaneously emerged, the skin lesion gradually disappeared and no recurrent symptoms occurred. The patient did not receive any further medical treatments.

DISCUSSION

The spontaneous emergence of a *Gnathostoma* worm can occasionally be seen after anthelmintic treatment (Suntharasamai *et al*, 1992). Even without drug treatment, spontaneous emergence of

Gnathostoma worms have been reported from time to time. Kagen *et al* (1984) reported a case similar to ours: a 25 year old Laotian female immigrant to the US scratched a pruritic patch on the skin of her abdomen and removed a 1 cm long worm, identified as an immature female *G. spinigerum* worm. Samarasinghe *et al* (2011) described 2 cases from Sri Lanka: one where an adult worm emerged from the skin of the back, and another where it emerged from the skin of a finger. Subheddar *et al*, (2014) reported a case from India where an adult *G. spinigerum* worm emerged from the skin on the palm of the hand. Beside the skin, an adult *Gnathostoma* worm was reported twice in the urine of Laotian immigrants (Horohoe *et al*, 1984; Norcross *et al*, 1992).

As for the stage of the development of *Gnathostoma* worms at the time of diagnosis, we conducted a search on the literature to determine the maturation stages of *Gnathostoma* worms were. In 1934 from Thailand, Prommas and Daengsvang (1934) reported 9 cases of gnathostomiasis; all the worms isolated were adult male *G. spinigerum* worms. An immature male *G. spinigerum* worm was found in a surgical specimen from a male Thai patient presenting with acute ileus in Kuwait (Hira *et al*, 1989). A male *G. spinigerum* worm was recovered from the brain of a Thai woman in Korea suffering from meningoencephalitis (Lee *et al*, 1988). Jongthawin *et al* (2015) reported three cases of molecularly diagnosed gnathostomiasis in Thailand and adult *G. spinigerum* worms were identified in two cases. Miyazaki and Kikuchi (1954) reported the first case of an adult *G. spinigerum* worm from a woman in Japan, Ando (2003) reviewed a series of 103 gnathostomiasis cases diagnosed based on the morphology of surgically removed or spontaneously

appearing worms and found 57 of the 103 cases were infection with *G. spinigerum*, 20 (35%) either immature or mature adult worms. These suggest that *G. spinigerum* L3 larvae can grow into immature/mature worms in humans more frequently than we expected.

Lao PDR is a high risk country for gnathostomiasis: gnathostomiasis cases have been reported among Laotian immigrants in the US (Stowens and Simon, 1981; Kagen *et al*, 1984; Horohoe *et al*, 1984; Norcross *et al*, 1992), in Germany (Hennies *et al*, 2006) and France (Chabasse *et al*, 1988). A seroepidemiological survey of gnathostomiasis in Lao PDR revealed a prevalence rate of 47% in Vientiane, the Capital, and 39% in Champasak in southern Lao PDR (Vonghachak *et al*, 2010). These suggest gnathostomiasis is a public health problem in Lao PDR. Gnathostomiasis control and prevention programs need to be developed and implemented for Lao PDR.

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