

Case Report: Human Pentastomiasis Caused by *Armillifer moniliformis* in Malaysian Borneo

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Abstract. We report a case of visceral pentastomiasis caused by *Armillifer moniliformis* in a 70-year-old aboriginal farmer from rural Malaysian Borneo. The patient complained of upper abdominal pain, jaundice, and loss of weight. Radiological investigations and subsequent histopathological examination revealed an adenocarcinoma of the pancreas with an adjacent liver nodule containing a nymph of *A. moniliformis*. This report constitutes the first documented human pentastomid infection in the whole of Malaysia after nearly 40 years, and it is the third description from Malaysian Borneo. Cases of human and animal pentastomiasis in Malaysia are discussed.

INTRODUCTION

Pentastomiasis is a zoonotic parasitic disease with an increasing number of documented human infections caused by the larval stages (nymphs) of pentastomes (tongue worms).^{1,2} These vermiform organisms form a unique phylum and are related to branchiurans.^{1,3} Pentastomes possibly coevolved with their respective vertebrate hosts,¹ and adult parasites of most species live in the respiratory tract of snakes and other reptiles.² Most human infections are caused by tropical and subtropical snake pentastomid parasites of the genus *Armillifer*, which encompasses species with different geographical distribution. *A. armillatus* and *A. grandis* are found in Africa, whereas *A. agkistrodontis* has been reported in China and *A. moniliformis* in Southeast Asia.^{2,4,5} Human infection occurs after accidental ingestion of infective ova, which are shed into the environment by snake secretions and feces. Human pentastomiasis was reported among aborigines in West and East Malaysia in the 1960s (Table 1).^{6–10} The present report describes the third case of human visceral pentastomiasis in Sabah, Malaysian Borneo, caused by *A. moniliformis*, and it constitutes the first report of this disease after nearly 40 years in the whole of Malaysia.

CASE REPORT

A 70-year-old aboriginal farmer (Orang Asli) from Keningau, Sabah, East Malaysia (Figure 1), was admitted to the Queen Elizabeth Hospital, Kota Kinabalu, in October 2010 with a 1-month history of upper abdominal discomfort, weight loss, anorexia, jaundice, and dark urine. The patient lived in a rural area for his entire life. Imaging techniques showed a tumor of the head of the pancreas and a liver nodule with a diameter of 1–2 cm. No enlarged lymph nodes were seen. The patient underwent Whipple's procedure, and histopathological examination of the pancreas tumor revealed a moderately differentiated adenocarcinoma with lymphatic metastasis. The liver nodule was also excised, and microscopic examination showed a C-shaped vermiform parasite with a body length of approximately 11 mm. The

parasite's tegument was pseudosegmented and consisted of 26 annuli. The body tapered into a blunt-pointed cone with four rostral hooks (Figures 2 and 3). In accordance with published data,^{2,7,11} the parasite was diagnosed as an intact nymph of the Southeast Asian pentastome species *A. moniliformis*. The liver parenchyma showed obstructive changes within the portal tract, with chronic inflammatory cell infiltration and bile duct proliferation. A thick fibrous capsule associated with chitinous material was present on one free side of the liver tissue. There was intense chronic inflammatory cell infiltration directly adjacent to this fibrous capsule area.

The patient was discharged 15 days after surgery but did not return to the follow-up appointments.

DISCUSSION

Most human cases of pentastomiasis are asymptomatic and are incidental findings during surgery or autopsy.^{2,7,12} If symptoms occur, they depend on the localization of the nymphs and are caused by the death or migration of the larval parasites.² Most often, the liver, mesenteries, spleen, and lungs are affected,² and patients report fever, abdominal pain, vomiting, diarrhea, jaundice, and abdominal tenderness.⁴ Severe and possibly lethal cases, such as massive infection of the liver,¹³ mechanical ileus,¹⁴ and other dissemination,¹⁵ are very rare. In the case described here, the symptoms reported were most likely caused by the pancreatic malignancy, and the pentastome infection was an incidental finding. In a series of 30 consecutive autopsies performed on aborigines from five different states in West Malaysia, pentastome larvae were found in 33.3% of the cases, with a prevalence of 45.4% in adults.⁷ Only two cases of human pentastomiasis caused by *A. moniliformis* were reported in East Malaysia/Borneo.⁸ The present report is, thus, the third description of a human pentastome infection in Malaysian Borneo, and it is the first description in the whole of Malaysia after 37 years. Interestingly, all reports about pentastomiasis in all of Malaysia describe the infection in the Orang Asli aborigine population, except for one case; this one case was a European woman who was diagnosed with an *A. moniliformis* infection in Borneo in 1965.⁸ In the latter case, consumption of python meat 9 years before the onset of symptoms was revealed. The Indian python (*Python*

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TABLE 1
Reported cases of human pentastomiasis in Malaysia

Pentastome species	Organ	Geographic region	Diagnosis	Reference
<i>Armillifer</i> sp. (<i>A. moniliformis</i> assumed)	Neck and abdomen	East Malaysia (Sabah, Borneo; two cases, including one European patient)	Radiology (both cases)	Rail ⁸
<i>A. moniliformis</i>	Liver and lung	West Malaysia (Pahang state)	Autopsy	Prathap and others ⁹
<i>Armillifer</i> sp. (<i>A. moniliformis</i> assumed)	Chest and abdomen	West Malaysia (Selangor state)	Radiology	Burns-Cox and others ⁶
<i>A. moniliformis</i>	Liver, lung, mesenteric lymph nodes, and intestinal wall	West Malaysia (10 cases from five different states; most cases from Selangor)	Autopsy	Prathap and others ⁷
<i>Armillifer</i> sp.	Fallopian tube	West Malaysia (state not mentioned)	Histopathology	Ong ¹⁰

molurus) and the Asian reticulated python (*P. reticulatus*) are known final hosts of *A. moniliformis*.^{8,16} In a survey, adult parasites were recovered from two of six *P. reticulatus*,¹⁶ a snake also endemic to Malaysian Borneo. It is well-known that consumption of snake meat is a common practice in some parts of Southeast Asia,^{16–19} and among the aboriginal tribes in Malaysia, the Temiar, Semai, and Temuan are known habitual python eaters.⁷ Risk factors for infection include consumption of undercooked contaminated snake meat as well as contact with living snakes and their secretions (i.e., tropical snake farming, pet keeping, harvesting of their skins, or tribal totemism).^{1,2,20} All of the human *A. moniliformis* infections in West and East Malaysia were indirectly linked to eating snakes by either the patient's history or affiliation with a certain tribe (ethnicity), and thus, aboriginal people in rural areas are the population with the highest risk. In the case presented, the tribe of the patient was not determined, and no information about possible snake meat consumption was obtained. Another possible source of infection in Malaysian aborigines may be drinking of river water contaminated with snake secretions.⁷ In Malaysia, visceral pentastomiasis was also reported in wild and domestic animals as intermediate and final hosts (Table 2).^{6,16,18,19,21–27} In a large survey in 1981, the infection rate of different wild animals with nymphs of *A. moniliformis* was 1.7% in West Malaysia, with the highest individual number of parasites found in rodents and carnivores. Of the carnivora, 20.7% were infected.¹⁶

Of note, in some human cases, the diagnosis of visceral pentastomiasis was achieved by X-ray and not histology.^{6,8} Thus, in theory, an *Armillifer* species different from *A. moniliformis* could have caused the visceral pentastomiasis in these patients, because the radiological picture cannot discriminate between the parasites involved. However, it is generally accepted that *A. moniliformis* is the only *Armillifer* species found in Malaysia, which is underlined by the other reports about human^{7,9} and animal^{6,16,18,19,23–25} visceral pentastomiasis with definitive species diagnosis in Malaysia. In one report about rats, the pentastome species responsible was termed *Porocephalus armillatus*,²¹ which might have been confused with *A. (P.) moniliformis*.

In the present report, the parasite was diagnosed as a nymph of *A. moniliformis* based on morphology. Unfortunately, not enough tissue was available for polymerase chain reaction analysis and phylogenetic positioning of the parasite. Recent analysis revealed that the pentastome snake parasites from Africa (*A. armillatus*), China (*A. agkistrodontis*), and the Americas (*P. crotali*) clustered together in the minimal evolution model, indicating coevolution with their vertebrate final hosts.¹ *Armillifer* nymphs have a body length of 9–23 mm,² and *A. moniliformis* has been reported to have a minimal size of 11–12 mm in Malaysia.^{7,19} The species has about 30 rings,⁷ but



FIGURE 1. Map of Malaysia showing the patient's place of residence (5°20' N, 116°10' E) in Sabah, Malaysian Borneo.

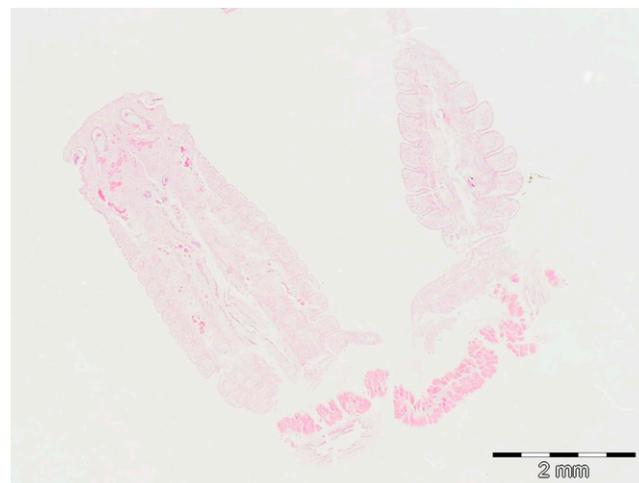


FIGURE 2. C-shaped nymph of *A. moniliformis* from the patient (hematoxylin and eosin stain) (Original magnification: 1.6×).

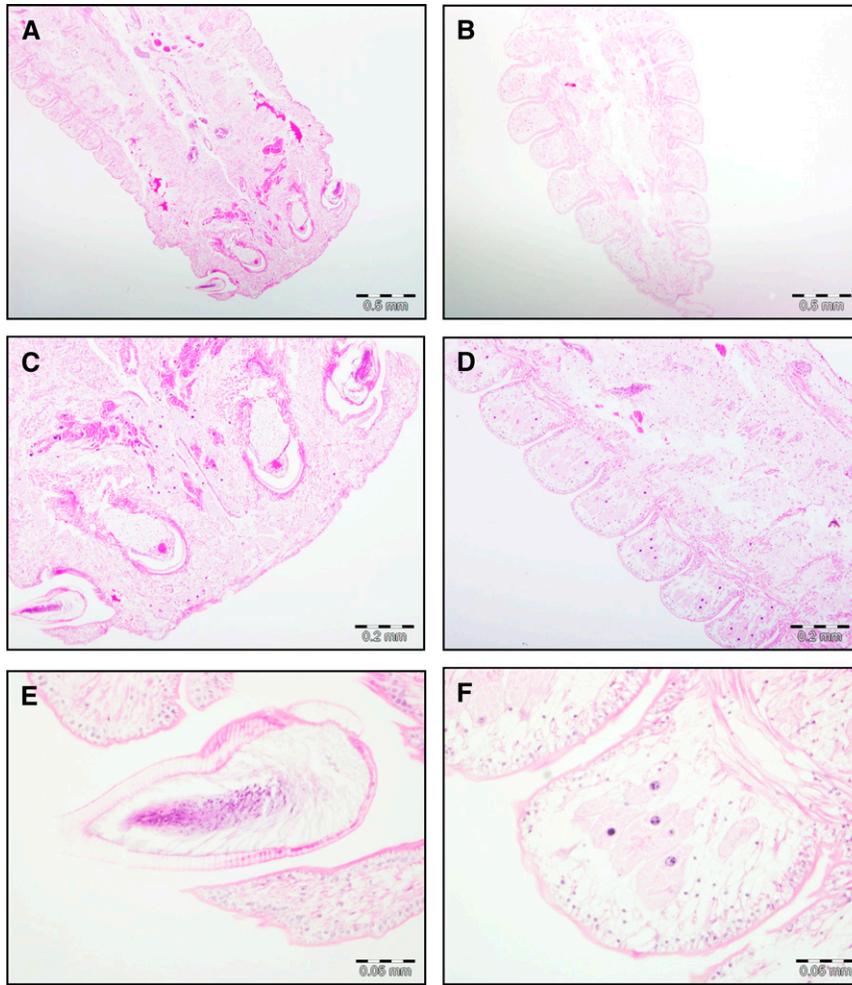


FIGURE 3. Sections of the nymph of *A. moniliformis*. (A) Rostral section of the parasite with four oral hooks. (B) Tapering posterior part of the parasite with annulations. (C) Close-up view of A. The mouth is surrounded by four hooks. (D) Annulated rings from the mid-body. (E) Close-up view of the oral hook. (F) Close-up view of a single annulated ring. All sections are stained with haematoxylin and eosin (Original magnification: A and B, 4×; C and D, 10×; E and F, 40×).

lower counts of 26 rings have also been reported.¹¹ In contrast, the geographically nearest neighbor, *A. agkistrodontis*, has only 7–9 spiral rings,⁴ whereas *A. armillatus* has 18–22 rings.^{6,7} To assess the full extent of human visceral pentastomiasis in

Malaysia, serological prevalence studies in risk populations should be performed; these studies were used in a recent investigation from The Gambia¹ and an earlier investigation from the Ivory Coast.²⁸

TABLE 2
Pentastomiasis among animals in Malaysia

Host	Pentastome species	Organ	Reference
Cynomolgus monkey (<i>Macaca irus/fascicularis</i>)	<i>A. moniliformis</i>	n/a	Burns-Cox and others ⁶
Giant swamp rat (<i>Rattus bowersi</i>)	<i>Porocephalus armillatus</i>	Lung, liver, spleen, and mesentery	Liat and Krishnasamy ²¹
House geckoes*	<i>Raillietiella hemidactyli</i>	n/a	Liat and Sen ²²
Wild animals (rodents, primates, carnivores, and reptiles*)	<i>A. moniliformis</i>	Abdominal cavity and lungs	Krishnasamy and others ¹⁶
Cat	<i>A. moniliformis</i>	Liver and spleen	Chooi and others ²³
Wild animals	<i>A. moniliformis</i>	n/a	Krishnasamy and others ¹⁸
Wild animals (house lizards,* snakes,* and others)	<i>A. moniliformis</i>	Lungs and mesenteries	Krishnasamy and others ¹⁹
Otter	<i>A. moniliformis</i>	Kidney, liver, spleen, and mesenteries	Cheah and others ²⁴
Lizard*	<i>Raillietiella</i> sp.	Lung	Jeffery and others ²⁶
Cockroaches	<i>Raillietiella</i> sp.	n/a	Jeffery and others ²⁷
Wild rats	<i>A. moniliformis</i>	n/a	Syed-Amez and Mohd Zain ²⁵

n/a = Information not available.
* Final hosts.

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