Case Report: *Angiostrongylus cantonesis* Myelitis in Thailand

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Abstract. A 67-year-old man presented with headache, middle back pain that radiated to both legs, and paresthesia in the right leg for 1 day. He had eaten raw shrimp 1 week previously. Over the next week after admission, he developed urinary retention and weakness in both legs. The numbness in his right leg expanded to below the umbilicus. Magnetic resonance imaging of the spinal cord showed myelopathy with minimal cord swelling at T9 to the conus medullaris and a hemorrhagic lesion from T10 to T11. A complete blood count on day 28 after the onset of symptoms revealed leukocytosis without eosinophilia and no white blood cells in his cerebrospinal fluid. Results of an immunochromatographic test kit were positive for *Angiostrongylus cantonesis* but negative for *Gnathostoma spinigerum*. After a 4-week course of albendazole combined with a tapering dose of dexamethasone, he achieved nearly complete recovery.

INTRODUCTION

Angiostrongylus cantonesis, also called rat lungworm, is a parasitic nematode and a leading cause of eosinophilic meningitis. Southeast Asia and tropical Pacific islands are considered endemic areas for this parasite, but there have been an increasing number of cases reported in South and Central America, the Caribbean, the United States, and Australia.^{1,2} In Thailand, approximately 100 *A. cantonesis* infections are reported annually, and the prevalence is particularly high in the northeast region.³ *A. cantonesis* completes its lifecycle in snails and *Rattus* rats, but many animals, such as freshwater shrimp, coconut crabs, frogs, and slugs, can be intermediate hosts. Ingestion of raw or undercooked animals or fresh vegetables contaminated with larvae from snails or slugs can cause infection in humans.⁴ Patients infected with *A. cantonesis* typically present with meningitis, which is accompanied by symptoms such as headache, photophobia, stiff neck, abnormal skin sensation, nausea, vomiting, fatigue, and body aches.⁵ *A. cantonesis* myelitis is rare and the diagnosis is often delayed because of under-recognition. We report a case of *A. cantonesis* myelitis encountered during October 2020 at Srinagarind Hospital in Khon Kaen, Thailand.

CASE PRESENTATION

A 67-year-old man with hypertension and hyperuricemia presented at a local hospital after experiencing headache, middle back pain that radiated to both legs, and paresthesia in the right leg for 1 day. He had eaten raw shrimp 1 week previously. After admission, he developed urinary retention and weakness of both legs that started in the right leg and then progressing to the left within 1 week. The area of numbness also expanded to the right side of the trunk below the umbilicus. He was bedridden and required intravenous morphine to alleviate the severe back pain. The pain resolved spontaneously within a few days, but the other neurological symptoms continued to worsen. Magnetic resonance imaging (MRI) of the thoracic and lumbar spine was performed during week 3 of admission and revealed myelopathy at T9 to the conus medullaris with spinal cord hemorrhage at T11–T12 and minimal spinal cord

swelling (Figures 1 and 2). The patient was subsequently referred for further investigation.

On the day of hospitalization at Srinagarind Hospital, his vital signs were as follows: blood pressure, 126/84 mmHg; heart rate, 108/min; body temperature, 38.0°C; and respiratory rate, 20/min. Neurological examination revealed muscle strength with grade 3/5 in the right lower limb, grade 4/5 in the left lower limb, and grade 5/5 in the upper limbs. Impaired pinprick sensations at the right side of trunk below the umbilical area and right lower limb were reported. Loose sphincter tone and diminished knee and ankle jerks were also detected bilaterally. Other physical examination findings were unremarkable.

Cerebrospinal fluid (CSF) examination on day 28 after the onset of symptoms showed a red blood cell count of 390 cell/mm³ and white blood cell count of 0 cell/mm³. His sugar and protein values were 82 mg/dL and 60 mg/dL, respectively. CSF Gram staining, bacterial culture, india ink staining, cryptococcal antigen, acid-fast bacilli staining, and polymerase chain reaction (PCR) assay for mycobacteria yielded negative results. A complete blood count (CBC) revealed leukocytosis without eosinophilia. Other blood chemistry findings were unremarkable. The results of the serum assay for *Angiostrongylus cantonesis* antibody that was performed using an immunochromatographic technique were positive, but the results of the test for *Gnathostoma spinigerum* were negative. The CSF was subjected to *A. cantonesis*-specific and *G. spinigerum*-specific PCR assays,^{6,7} but the results of both tests were negative.

The patient was treated with albendazole 800 mg/day for 4 weeks plus oral dexamethasone starting at 12 mg/day; then, it was tapered over the course of 4 weeks. At the 4-week follow-up examination, the patient's urinary retention had completely resolved and his muscle strength had significantly improved. Additionally, he had gone from being bedridden to being able to walk slowly with a walking aid.

DISCUSSION

We encountered a case of myelitis as an atypical presentation of *A. cantonesis*. Most cases of angiostrongyliasis present with eosinophilic meningitis lasting from 7 to 14 days. Patients usually have moderate to severe headache and are afebrile. Neck stiffness is detected in only 30% to 50% of patients; therefore, *A. cantonesis* infection cannot be excluded only if there is no sign of meningeal irritation. Although other forms

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FIGURE 1. Hyperintensity lesion at T9–T12 in the T1 sagittal view of the T-spine using magnetic resonance imaging (MRI).

of angiostrongyliasis are rare, ocular angiostrongyliasis has been occasionally reported.⁸ A thorough evaluation of the patient's history is an important part of the diagnosis because most infected patients have ingested freshwater raw snails or other intermediate hosts within 7 to 30 days before the onset of symptoms.⁵ Several other case reports of patients with *A. cantonesis* myelitis in the existing literature are listed along with their clinical presentations, diagnostic methods, treatment, and prognoses in Table 1.

FIGURE 2. Hyperintensity lesion at T10 level in the axial T1 view of the T-spine using magnetic resonance imaging (MRI) with gadolinium.

TABLE 1
Cases of *A. cantonesis* infection presenting with myelitic symptoms as a first manifestation reported in the literature from 1999 to 2020

Case	Year of report/country	Age, years/Sex	Risk factors	Presentation	Treatment	Outcome	Reference
1	1999/Australia	11 months/M	Travel to Fiji	Flaccid quadripareisis and high thoracic sensory level with dysesthesia	IV methylprednisolone	Death	14
2	2010/China	32/M	Consumption of inadequately cooked <i>P. canaliculata</i>	Headache and paresthesia of the upper limbs	Dexamethasone and albendazole	Complete recovery	15
3	2015/Japan	25/F	Vacation in Thailand and Laos	Numbness of the right flank	IV methylprednisolone	Recovery	16
4	2016/Israel	37/F	Probable consumption of contaminated vegetables	Leg weakness and urinary retention	IV methylprednisolone and then oral prednisolone	Partial recovery	17
5	2019/US	44/F	Consumption of various types of seafood	Paresthesia of the lower limbs, urinary retention, and midthoracic radicular pain		Complete recovery	18
6	2020/US	20/M	Consumption of raw giant African snail	Myalgia of the thighs, dysuria, and constipation	IV methylprednisolone and then oral prednisolone, albendazole, and IVIg	Partial recovery	12
7	2020/Martinique	13 months/F	Unknown	Hypotonia and quadriplegia	Prednisolone and albendazole	Psychomotor retardation	19

F = female; IV = intravenous; IVIg = intravenous immunoglobulin; M = male; *P. canaliculata* = *Pomacea canaliculata*; US = United States.

In Thailand, if patients with a history of raw food consumption present with self-limiting radicular pain followed by any myelitic signs at different anatomical levels, then gnathostomiasis is the provisional diagnosis.⁹ This case presentation reflects the migration of larva through the nerve root, thecal sac, and spinal cord.¹⁰ Although myelitis in neuro-gnathostomiasis is well-documented, *A. cantonensis* myelitis is rare. A previous case series of 12 patients with *A. cantonensis* infection reported no myelitis. Neither CBC results nor the presence of CSF eosinophilia can be used to distinguish between neuro-gnathostomiasis and neuro-angiostrongyliasis. A spinal MRI will show multiple linear, track-like, scattering punctate lesions with cord swelling with both diseases, but hemorrhagic lesions are more typically found with neuro-gnathostomiasis than with neuro-angiostrongyliasis.^{11,12} Molecular and serologic studies are important to confirm the diagnosis and distinguish between these diseases.

For this case, neither the CBC nor CSF examination revealed eosinophilia; however, MRI revealed minimal swelling of the spinal cord because its use was delayed. The patient had been admitted for supportive treatment and waited 3 weeks before MRI was performed and a diagnosis was made. The false-negative results of the CSF PCR assay for *A. cantonensis* may have been attributable to its timing. A diagnosis of neuro-angiostrongyliasis was confirmed by a positive antibody to *A. cantonensis* from an immunochromatographic test (ICT) kit developed by the Department of Parasitology of Khon Kaen University. The accuracy of this test is comparable to that of the standard method, and it is used as the referral method for measuring the accuracy of other test kits.^{6,13} The results of serologic testing for *G. spinigerum* were negative, thereby ruling out neuro-gnathostomiasis. A partial response to dexamethasone and albendazole was observed as expected. The minimal swelling of the spinal cord and absence of CSF pleocytosis indicated that the inflammation caused by *A. cantonensis* had nearly resolved on its own before treatment was initiated.

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