

Case Report

Angiostrongylosis meningomyelitis without blood eosinophilia

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Abstract

Angiostrongylus cantonensis and *Gnathostoma spinigerum* usually cause eosinophilic meningitis with associated peripheral blood eosinophilia. A 44-year-old man developed acute paraplegia with bowel and bladder dysfunction. Spinal magnetic resonance images showed a long T2W hyperintensity signal from the 1st to 8th spinal thoracic level. Cerebrospinal fluid analysis revealed eosinophilia and elevated cerebrospinal fluid protein, whereas differential leucocytes count in peripheral blood was unremarkable. Positive immunoblot tests for *A. cantonensis* antibody in serum and cerebrospinal fluid were reported. The patient had neither history of recent traveling to the high endemic areas of the parasite in Thailand, nor consumption the parasitic hosts. Immediate treatment with intravenous pulse methylprednisolone and oral albendazole resulted in complete recovery. Despite an unremarkable differential leucocytes count, absence a history of parasitic hosts consumption, and a less common presentation with meningomyelitis, *A. cantonensis* should be considered when cerebrospinal fluid eosinophilia presents.

Key words: myelitis; cerebrospinal fluid; angiostrongylosis.

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Introduction

Human gnathostomosis and angiostrongylosis are parasitic diseases endemic in Southeast Asia where people act as incidental hosts in the life cycle of both *Angiostrongylus cantonensis* and *Gnathostoma spinigerum*. Humans get infected through ingestion of infective larvae of both parasites in raw meat of the intermediate or paratenic hosts. Another route for *A. cantonensis* infection consists in the ingestion of fresh vegetables not properly washed or contaminated with the intermediate or paratenic hosts of *A. cantonensis*, i.e. snails, mollusks, crabs, fresh-water fishes, prawns, etc. (Figure 1).

The common presentations of human gnathostomosis and angiostrongylosis are eosinophilic meningitis and subarachnoid hemorrhage. Myelitis or meningomyelitis is scarcely reported. Moreover, most cases of the both parasitic infestations have peripheral blood eosinophilia associated to suggest the infestation. Normally, most of the eosinophilic meningitis cases relevant to both parasitic infestations are reported from the northeastern Thailand. Here, we report a case of acute meningomyelitis caused by *A. cantonensis*, without associated peripheral blood eosinophilia in the

southern Thailand. Also, the patient reported of never consumption of raw meat of the animals, which is known as the intermediate or paratenic hosts of the parasite at all.

Case report

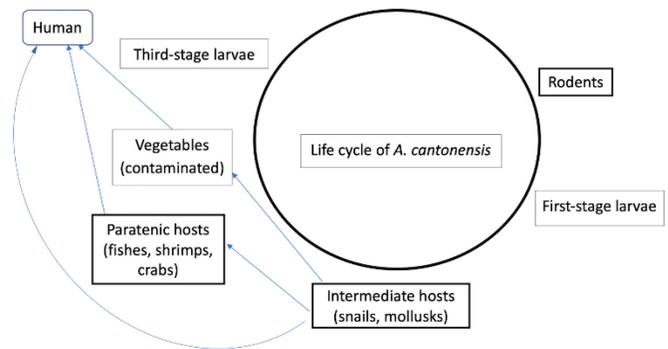
A previously healthy 44-year-old man had a febrile illness, which resolved spontaneously in a few days. Four days later, he presented to a local hospital with subacute paraparesis and bladder dysfunction. On the initial investigation, a peripheral blood cells count showed a hemoglobin level of 13.3 g%, total white blood cells count of 10,900 cells/mm³ with 60% polymorphonuclear (PMN) cells, 30% lymphocytes (Ly), 2% monocytes (Mo), and 2% eosinophils (Eo). An anti-HIV serology test was negative. Then, he was transferred to our hospital where the neurological examination revealed paraparesis of grade 3/5, brisk knee jerks, and urinary retention. There was no sensory deficit noted. A magnetic resonance images (MRI) showed only a long segment of T2W hyperintensity signal extending from thoracic spinal level (T) of T1 to T8 (Figures 2 and 3). To differentiate the causes of long extensive transverse myelitis in this case, CSF Venereal

Diseases Research Laboratory (VDRL) as well as Fluorescent Treponemal Antibody Absorption (FTA-ABS), polymerase chain reaction for human herpes virus types 1-6, antinuclear factor and NMO-IgG antibody were tested, which were unremarkable all. Subsequently, a lumbar puncture and a cerebrospinal fluid (CSF) analysis were done and showed colorless CSF and normal CSF pressure. With CSF leukocytosis of 310 cells/mm³, 48% Eo, 50% Ly, 2% PMN cells, and only 2 red blood cells/mm³ were reported. The CSF protein was 260.9 (14-45 mg %), CSF/serum glucose was 53/102 mg % (ratio = 0.52). The serology test by immunoblot technique for *A. cantonensis* antibody was requested, because of the high number of eosinophils in the CSF, and revealed positive results in both the serum and CSF samples. The patient was immediately treated with albendazole 800 mg/day orally combined with methylprednisolone 1 g/day intravenously for 5 consecutive days, followed by oral prednisolone 60 mg/d for another 5 days, and a course of tapered-off dosage in 15 days. The neurological disorders

Figure 2. Myelitis caused by *A. cantonensis*. Sagittal T2W magnetic resonance images of the thoraco-lumbar spine showed a long segment hyperintense spinal cord lesion, extending from T1 (the top arrow) to T8 spinal level with dominantly between T5 (bottom arrow) and T8 levels.



Figure 1. Life cycle of *A. cantonensis*.



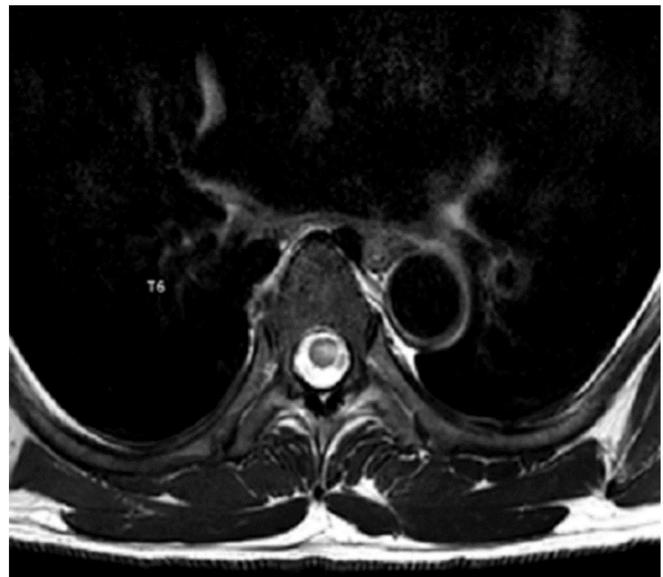
recovered completely 12 weeks after the treatment initiation.

The patient was a permanent resident of southern Thailand, where is a low endemic area of *A. cantonensis*. He had not travelled to any endemic areas of *A. cantonensis* recently prior to his illness, nor did he eat raw meat of the animals known as the hosts of *A. cantonensis* larvae. However, eating of various raw vegetables, particularly, morning glory which usually grows in water or muddy ground, served with a favorite “Thai papaya salad” was a possible route for the parasitic larvae to infest him.

Discussion

Eosinophilic meningitis and other neurological disorders with peripheral blood eosinophilia are mostly caused by parasitic infestations such as gnathostomosis, angiostrongylosis, toxocarosis, strongyloidosis. However, some non-parasitic infestation including

Figure 3. Myelitis caused by *A. cantonensis*. Axial T2W magnetic resonance images of the thoracic spine showed hyperintense spinal cord lesion at T6 spinal level.



coxsackie virus meningitis, neuro-Behcet's, and acute lymphoblastic leukemia could present with eosinophilic meningitis [1]. Among the causative parasites of eosinophilic meningitis mentioned, *G. spinigerum* and *A. cantonensis* are apparently endemic in Southeast Asia. While *Angiostrongylus costaricensis* is endemic in Central America, *Toxocara canis* and *Toxocara cati* are both found worldwide [2]. These parasites can cause a wide spectrum of nervous system disorders from asymptomatic cerebral and/or spinal MRI lesions to clinically obvious neurological disorders.

The different mechanisms of neural tissue injury between the two nematodes endemic in southeast Asia have been explained. While *G. spinigerum* usually invades neural parenchyma directly by hooks and spines on its head resulting in obvious neural tissue injury i.e., intracerebral or intraspinal hemorrhage, or subarachnoid hemorrhage, *A. cantonensis* typically induces immune-mediated inflammation of meninges or neural tissues i.e., meningitis, cranial neuritis or transverse myelitis. Nevertheless, isolated cerebritis or myelitis caused by *A. cantonensis* infestation is rarely reported. A case of gnathostomosis myelitis with merely sensory deficit in a European traveler who returned from a Southeast Asian country was reported [2]. A report of two cases of eosinophilic myelitis with positive IgG antibody for toxocarosis from Korea was published. Both cases presented with progressive stepwise sensory impairment without motor deficit. While slightly elevated blood eosinophilia was noted in one case, the CSF differential cell counts were unremarkable in both cases [3]. Another case of *T. canis* induced myelitis in which only right lower limb hypoesthesia presented, and concomitant eosinophilic pneumonitis was found [4]. Usually, peripheral blood or CSF eosinophilia, or both, is associated in cases of neurological disorders caused by the parasites. However, there is a variation of the presence of blood or CSF eosinophilia as the report cases above. Moreover, the common site of eosinophilic myelitis is the cervico-thoracic spinal cord level [2,5-6].

Because of the clinical syndromes of myelopathy, CSF eosinophilic leukocytosis, and high CSF protein but with few red blood cells, immunological meningomyelitis caused by *A. cantonensis* was considered in our case report, despite no gadolinium leptomeningeal enhancement (GLE) illustrated by the MRI. The visibility of abnormal GLE depends markedly on the degree of leptomeningeal inflammation, so, it can be variably visible on the gadolinium-enhanced MRI in cases of meningitis. Our diagnosis of meningomyelitis caused by *A. cantonensis*

was confirmed by a positive serology tests by immunoblot technique in both the serum and CSF. Like the previous case reports, the patient recovered completely with a course of high-dose prednisolone and oral albendazole [6-8]. We consider from our review that myelitis or other neurological disorders caused by the parasites mentioned earlier including *A. cantonensis* is an immunological reaction to the previous presentation of the parasites in the nervous tissue during their migration in human body. Thus, we think that the anti-inflammatory effect of corticosteroid plays a major role in neurological recovery in cases of angiostrongylosis with neurological involvement rather than antihelminth treatment.

Conclusions

Meningomyelitis is a rare neurological presentation in angiostrongylosis. CSF eosinophilia without obvious number of red blood cells is a significant clue for immunological meningomyelitis and the diagnosis of angiostrongylosis meningomyelitis. The absence of peripheral blood eosinophilia, no a history of eating raw meat of the animals known as the intermediate or paratenic hosts of *A. cantonensis*, and never traveling to nor living in a high endemic area of the parasite do not exclude the diagnosis of angiostrongylosis.

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Author contribution

Amornpojnimman and Sathirapanya contributed to the interpretation of data and wrote the manuscript. Sanghan, Ekpitakdamrong, Koonalintip and Leelawai contributed to the writing of the manuscript. All authors contributed to the final version of the manuscript.

References

1. Sick C, Hennerici MG (2014) Expect the unexpected: a case of isolated eosinophilic meningitis in Toxocarosis. *Case Rep Neurol* 6: 259-263.
2. Schmutzhard E (2007) Eosinophilic myelitis, a souvenir from South East Asia. *Pract Neurol* 7: 48-51.
3. Park CW, Choe WJ, Chun YI (2012) Eosinophilic myelitis in the cervical cord mimicking intramedullary cord tumor. *J Korean Neurosurg Soc* 52: 410-413.
4. Park KH, Kim YS, Kim SK, Choi NC, Kwon OY, Lim B, Park KJ (2016) *Toxocara canis*-associated myelitis with eosinophilic pneumonia. *Exp Neurobiol* 25: 139-142.
5. Oehler E, Ghawche F, Delattre A, Berberian A, Levy M, Valour F (2014) *Angiostrongylus cantonensis* eosinophilic

meningitis: a clinical study of 42 consecutive cases in French Polynesia. *Parasitol Int* 63: 544-549.

6. Diao Z, Jin E, Yin C (2010) *Angiostrongylus cantonensis*: lesions in brain and spinal cord. *Am J Trop Med Hyg* 82: 519.
7. Al Hammoud R, Nayas SL, Murphy JR, Heresi GP, Butler IJ, Perez N (2017) *Angiostrongylus cantonensis* meningitis and myelitis, Texas, USA. *Emerg Infect Dis* 23: 1037-1038.
8. Diao Z, Yin C, Jin E (2010) Images in clinical tropical medicine. Myelitis caused by infection of *Angiostrongylus cantonensis*. *Am J Trop Med Hyg* 83: 1176-1177.

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