Case Report

Spinal Gnathostomiasis: A Case Report with Magnetic Resonance Imaging and Electrophysiological Findings

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Background: *Spinal gnathostomiasis results in serious multiorgan impairments. Magnetic resonance imaging (MRI) and serology help in diagnosis, and assessing the severity and extent of the disease. However, the physiology of neural pathway could not be demonstrated. Electrodiagnosis may have a role in demonstrating the functions of the central and peripheral neural pathways and prognostic assessment of the disease.*
Material and Method: *An 18-year-old man presented with radicular pain and rapid progressive weakness of lower extremities, leading to paraplegia in three days. A clinical evaluation and laboratory tests, including serology, MRI, and electrodiagnosis, were performed.*
Results: *The investigation showed L1 paraplegia with urinary retention. The serial MRI of T-spine showed longitudinal T2 hypointense lesion along the lower thoracic spinal cord, representing track-like hemorrhage with spinal cord edema at the onset of symptoms, nodular enhancement of T11-T12 spinal cord, and enhancing and clumping of cauda equina nerve roots in the MRI at two months after the first MRI. The CSF and serum for Gnathostoma antibody were positive. Intravenous corticosteroid and oral albendazole were given. Three months after treatment, the symptoms improved in sensation but not in motor function. The electrodiagnosis was performed. There were very small amplitudes and no response in bilateral tibial and peroneal motor nerve conduction studies (NCS) respectively. The bilateral sural sensory NCS were normal. Neither tibial somato-sensory evoke potentials (SSEP) nor motor evoke potential (MEP) was recorded. No further improvement of patient's clinical status at eight months after onset was observed.*
Conclusion: *Electrophysiological findings demonstrated the function of spinal cord and peripheral nerves in the patient with spinal gnathostomiasis. There were correlations between MRI and electrophysiological findings that confirmed pathophysiology of the disease. Absence of SSEP and MEP response correlated with poor neurological outcomes in radiculomyelitis caused by Gnathostoma spp. infection.*

Keywords: *Spinal cord diseases, Gnathostoma, Magnetic resonance imaging, Electrodiagnosis*

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Gnathostomiasis is a foodborne parasitic infection from the third-stage larvae of *Gnathostoma* spp. Neurognathostomiasis results in serious illness and multiorgan impairments. Magnetic Resonance Imaging (MRI) and serology help in diagnosis, assessing severity, and extent of disease. However, the physiology of neural pathway could not be demonstrated. Electrodiagnosis may play a role in demonstrating central and peripheral neural pathway function and in making a prognosis. To our knowledge, there is no report of electrophysiological findings correlated with MRI in the patient with spinal gnathostomiasis. The authors conducted a physical evaluation; laboratory tests, including serology; MRI; and electrodiagnosis in a paraplegic man with spinal gnathostomiasis.

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Case Report

An 18-year-old man presented with radicular pain and rapid progressive weakness of the lower extremities, leading to paraplegia and loss of voiding ability in three days. He was referred to our hospital two months after onset. The MRI of thoracic spine at first admission in another hospital showed longitudinal T2 hypointense lesion along lower thoracic spinal cord, representing track-like hemorrhage with spinal cord edema up to T3 level and no definite enhancement (Fig. 1). Conservative treatment was given followed by discharge. The symptoms did not improve, the patient and his family came to our hospital, which is a tertiary

care hospital in north-eastern Thailand. Patient history was taken and an examination was performed. He was a high school student with a history of cutaneous itching of the left thigh before the onset of weakness. He denied history of eating raw food. Physical examination revealed flaccid tone with muscle power grade 0 of lower limb muscles. Pinprick, light-touch, and proprioception sensations were lost below L1 dermatome. The tone of anal sphincter was loose with negative bulbocavernosus reflex.

A full blood count showed normal values without eosinophilia. Cerebrospinal fluid (CSF) specimen showed no red blood cells, three white blood cells/mm^3, 112 mg/dl protein and 71.6 mg/dl glucose (65% compared with plasma glucose). CSF and serum for *Gnathostoma* and *Angiostrongylus* antibody were sent for analysis at the Faculty of Tropical Medicine, Mahidol University, Bangkok. Western blot assay that detected specific antibodies to a purified 24-kD antigen obtained from advanced third stage larvae of *Gnathostoma spinigerum* showed positive results in both CSF and serum. Results of serologic tests for *Angiostrongylus* were negative. Polymerase chain reaction (PCR) for *Mycobacterium tuberculosis* complex (MTB) and mycobacteria (NTB) were negative in CSF specimen. Stool for parasite showed *Blastocystis hominis* vacuolar form.

The second MRI of TL-spines (2 months after the first imaging) revealed decreased size of the track-like hemorrhage and spinal cord edema with small size of T11-T12 spinal cord. There was nodular enhancement of T11-T12 spinal cord, leptomeningeal enhancement along lower thoracic cord surface, and enhancing and clumping of cauda equina nerve roots (Fig. 2).

Treatment consisted of intravenous dexamethasone 24 mg/day for five days then pulse methylprednisolone 1 g/day for three days, oral albendazole 800 mg/day for 14 days, and metronidazole 1,200 mg/day for six days. The rehabilitation program to prevent complications was performed before discharged home.

Three months after treatment (five months after onset) sensation had improved, but the weakness of lower extremities remained. The electrophysiological studies, including motor and sensory nerve conduction study (NCS), somatosensory evoke potentials (SSEP), and motor evoke potentials (MEP) evaluation, were performed using Viking on EDX and Magstim Rapid 2 machine with double 70 mm air film coil and 0-3.5 Tesla at 100% output. The motor and sensory NCS of left median and ulnar nerves were normal. The motor

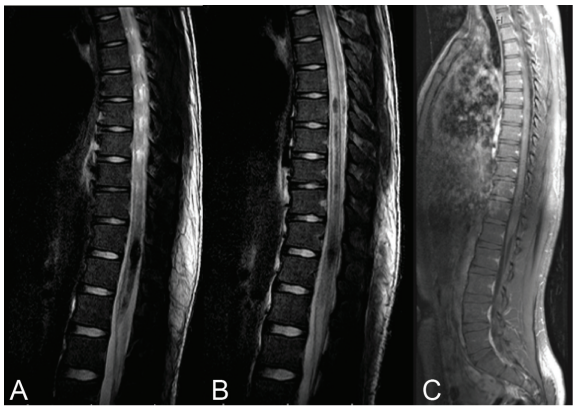

Fig. 1 MRI of thoracic spine. A, B) Sagittal T2 weighted images, showing abnormal longitudinal T2 hypointense lesion along lower thoracic spinal cord with spinal cord edema up to T3 level. C) Sagittal T1 weighted postgadolinium images, showing no definite enhancement.

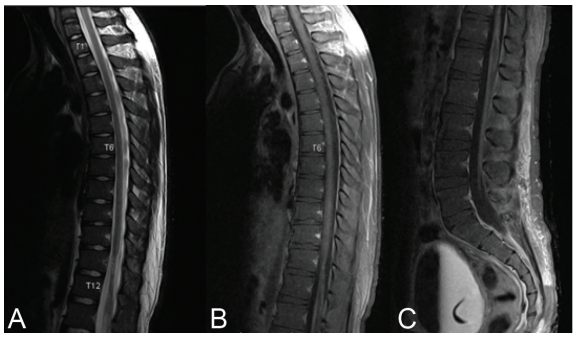

Fig. 2 MRI of thoracolumbar spines 2 months after first imaging. A) Sagittal T2 weighted imaging, showing decreased size of the longitudinal T2 hypointense lesion and spinal cord edema with small size of the T11-T12 spinal cord. B, C) Sagittal T1 weighted postgadolinium of thoracic and lumbar levels, showing nodular enhancement of the T11-T12 spinal cord, leptomeningeal enhancement along lower thoracic cord surface, and enhancing and clumping of cauda equina nerve roots.

NCS of bilateral peroneal and tibial nerves showed no response and very small amplitude respectively. Normal sensory NCS of bilateral sural nerves was recorded. The tibial SSEP showed normal response recorded from popliteal fossa area but no response obtained from spinal area and cortex. The MEP of upper extremities recorded at first dorsal interossei muscle was normal. However, the MEP of lower extremities recorded at quadriceps and iliopsoas muscles showed no response.

A second course of oral albendazole 800 mg/day for three weeks was administered. Follow-up MRI of TL-spine showed further decreased size of track-like hemorrhage with atrophic change of T11-T12 spinal cord and improvement of radiculomyelitis. Six months post-treatment (8 months after onset), no improvement of the patient's clinical picture was observed. The patient completed a comprehensive rehabilitation program for paraplegia to achieve maximum recovery. He could perform activities of daily living and use a wheelchair independently. He had a walking exercise program with knee-ankle-foot orthosis and gait aids.

Discussion

Gnathostomiasis is one of several common foodborne parasitic infections in Southeast Asia[1]. Humans are accidental hosts by ingestion of raw shellfish, freshwater fish, frog, and chicken. There are raw or undercooked food in some areas of Thailand. This patient had cutaneous symptoms that were localized skin pruritus of left thigh area without creeping eruption or migratory swelling, as in previous studies[2-4]. Larvae enter the central nervous system (CNS) by invading directly through intervertebral foramina along the spinal nerves and vessels[5] and damage the organs by direct mechanical injury, toxins, and immunological reaction[6]. No eosinophilia was demonstrated in this patient similar to the previous study that showed eosinophilia and systemic signs usually subside in the chronic stage[7]. Radicular pain is one of the outstanding features of neurognathostomiasis[6] that reflected the migration of nematode along the nerve root to the CNS. This patient had radicular pain three days before complete leg paralysis, which had the same duration with the previous study that showed the average of one to five days[6].

MRI was the first imaging for this patient to give the possible causes of paraplegia. Another role of MRI is assessing severity and extent of disease[8]. MRI of T-spines of this patient showed hemorrhagic track along thoracic cord, which is the hallmark sign of neurognathostomiasis[7,8]. The second MRI showed nodular enhancement of the T11-T12 spinal cord, leptomeningeal enhancement along lower thoracic cord surface, enhancing and clumping of cauda equina nerve roots, suggesting radiculomyelitis, which is the main manifestation of neurognathostomiasis[1,6]. Physical examination of L1 paraplegia with lower motor neurone signs in this patient correlated with these MRI findings. This patient did not have any symptoms of meningitis, meningoencephalitis, intracerbral, or subarachnoid hemorrhage.

CSF profile of this patient showed immune reaction inflammatory response, however there was no eosinophilic pleocytosis, which is a hallmark laboratory finding[6]. Immunodiagnosis used in this case was the 24-kD band on Western blot which has nearly 100% specificity for gnathostomiasis[9]. *Angiostrongylus cantonensis* infection is the main differential diagnosis in the case of neurognathostomiasis[7]. Immunodiagnosis and radiological appearance associated with neurological signs help to differentiate these two parasitic infections. The other causes of radiculitis such as *Mycobacterium* spp. were investigated with CSF PCR. The clinical presentation, MRI, and serology typically helped in the diagnosis of spinal gnathostomiasis. Isolation of the larvae from the lesions may be possible in cutaneous lesions but not practical for visceral involvement[7]. Living nematode was demonstrated intraoperatively in one study[10].

The treatments of gnathostomiasis were controversial. There is no randomized trial of anthelminthic therapy or corticosteroid. However, there were reports of albendazole treatment[1] that found that 56% fully recovered, 22% partially recovered, and 22% did not recover. In this case, the patient received two courses of albendazole treatment as in the previous study[11]. The MRI findings of radiculomyelitis and cord edema supported the use of steroid treatment in this patient. However, the patient's impairments did not improve. The improvement of radiculomyelitis, cord edema, and hemorrhagic track in follow-up MRI was demonstrated. Because of insufficient data about the previous treatment, the effectiveness of the treatment could not be concluded in this patient.

Electrodiagnosis including NCS, SSEP, and transcranial magnetic stimulation (TMS) were performed in this patient to evaluate the function of neural pathways. Abnormal amplitude of motor NCS but normal sensory NCS correlated with the findings of radiculitis in the MRI. SSEP evaluation helped in demonstrating dorsal column pathway and had a prognostic value in traumatic spinal cord injury[12]. Initial recordability of a tibial SSEP was associated with a favorable functional and neurological outcome, while initial absence of a tibial SSEP was associated with a poor outcome in 75% of the traumatic spinal cord injury patients[12]. In this patient, the absence of tibial SSEP recorded from spinal area and cortex suggested poor neurological outcome.

TMS was used in clinical diagnostic, prognostic tests, and treatment in various diseases[13,14]. The amplitude of MEP reflects integrity of the corticospinal tract, excitability of motor cortex, and the conduction along peripheral nerves to muscles[14]. There was absence of MEP recorded from lower extremities while normal MEP was recorded from upper extremities. Abnormal integrity of pyramidal tract below C-spinal cord, peripheral nerves, and muscles were considered. We could not evaluate the spinal conduction pathway in the case of abnormal peripheral conduction by electrodiagnosis. However, abnormalities in the electrophysiological findings might be the sequelae of radiculomyelitis and suggested poor neurological outcome.

Three months after the second course of albendazole or eight months after the onset, there was no further clinical improvement. The neurological and functional outcomes were correlated with the MRI and electrophysiological findings. Spinal cord atrophy in the MRI, absence, or abnormal amplitude in motor NCS, absence of SSEP, and MEP predicted poor outcome in this patient.

The limitations of this report were the details of previous treatment in the other hospital and late electrodiagnosis. Because lack of the treatment details, we could not evaluate the effectiveness of the treatment.

Conclusion

Electrophysiological findings demonstrated the function of spinal cord and peripheral nerves in the patient with spinal gnathostomiasis. There were correlations between MRI and electrophysiological findings that confirmed the pathophysiology of the infection. Absence of SSEP and MEP response correlated with poor neurological outcomes in radiculomyelitis caused by *Gnathostoma* spp. infection.

What is already known on this topic?

Radiculomyelitis is one of the manifestations of neurognathostomiasis. MRI and serology help in diagnosis, assessing severity, extent of disease, and management plan. The treatments of gnathostomiasis were controversial. There is no randomized trial of anthelminthic therapy or corticosteroid. However, there were reports of albendazole treatment with good result.

MRI demonstrated the pathology of spinal cord but not physiology. Electrodiagnosis may play a role in demonstrating central and peripheral neural pathway function and in making a prognosis.

What this study adds?

Electrodiagnosis demonstrated the function of spinal cord and peripheral nerves in the patient with spinal gnathostomiasis. Absence of SSEP and MEP response predicted poor neurological outcomes. There were correlations between MRI and electrophysiological findings that confirmed the pathophysiology of spinal gnathostomiasis.

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Potential conflicts of interest

None.

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โรคพยาธิตัวจี๊ดที่ไขสันหลัง: รายงานผู้ป่วยและผลการตรวจด้วยเอกซเรย์คลื่นแม่เหล็กไฟฟ้าและสรีรวิทยาไฟฟ้า

รัชวรรณ สุขเสถียร, สลินทิพย์ คุณาดิศร, อรกานต์ วงษ์ฟูเกียรติ, กัลยา อิงคสุทธิ

ภูมิหลัง: *โรคพยาธิตัวจี๊ดที่ไขสันหลังส่งผลให้เกิดความบกพร่องของร่างกายที่รุนแรง เอกซเรย์คลื่นแม่เหล็กไฟฟ้าและการตรวจทางซีรัมวิทยาช่วยในการวินิจฉัยและประเมินความรุนแรงของโรค แต่ไม่สามารถประเมินการทำงานของระบบประสาทที่แท้จริงของผู้ป่วยได้ การประเมินการทำงานของระบบประสาทส่วนกลางและส่วนปลายด้วยไฟฟ้าวินิจฉัยจึงสามารถช่วยในการให้การพยากรณ์โรค*

วัสดุและวิธีการ: *ผู้ป่วยชายอายุ 18 ปี มีอาการปวดและอ่อนแรงของขาทั้งสองข้างจนขยับไม่ได้ภายในเวลา 3 วัน ผู้ป่วยได้รับการวินิจฉัยและการตรวจทางห้องปฏิบัติการ เอกซเรย์คลื่นแม่เหล็กไฟฟ้า และไฟฟ้าวินิจฉัย*

ผลการศึกษา: *ผู้ป่วยมีอาการอัมพาตของขาจากรอยโรคที่ไขสันหลังระดับ L1 และปัสสาวะไม่ออก ผลการตรวจเอกซเรย์คลื่นแม่เหล็กไฟฟ้าครั้งแรกพบรอยโรค T2 hypotense ตามยาวของไขสันหลังระดับอก มีรอยเลือดออกและบวมในไขสันหลังสองเดือนหลังจากมีอาการ ผลของเอกซเรย์คลื่นแม่เหล็กไฟฟ้าพบความผิดปกติของเส้นประสาทบริเวณ cauda equina ผลการตรวจภูมิคุ้มกันต่อพยาธิตัวจี๊ดในเลือดและน้ำไขสันหลังให้ผลบวก ผู้ป่วยได้รับการรักษาด้วยยา corticosteroid และ albendazole สามเดือนหลังการรักษาอาการชาดีขึ้น แต่อาการกล้ามเนื้อขาอ่อนแรงยังคงเดิมจึงได้ตรวจไฟฟ้าวินิจฉัย ซึ่งพบการตอบสนองของเส้นประสาท tibial ขนาดเล็กและไม่มีการตอบสนองของเส้นประสาท peroneal โดยการกระตุ้นเส้นประสาทสั่งการ tibial และ peroneal การตรวจการรับความรู้สึกของเส้นประสาท sural ได้ผลปกติ ส่วนการตรวจ somato-sensory evoke potentials (SSEP) และ motor evoke potential (MEP) ไม่พบการตอบสนอง ผู้ป่วยมีอาการคงที่หลังจากเจ็บป่วยนาน 8 เดือน*

สรุป: *การตรวจไฟฟ้าวินิจฉัยช่วยในการประเมินการทำงานของไขสันหลังและเส้นประสาทส่วนปลายในผู้ป่วยโรคพยาธิตัวจี๊ดที่ไขสันหลัง พบความสัมพันธ์ระหว่างผลการเอกซเรย์คลื่นแม่เหล็กไฟฟ้าและการตรวจไฟฟ้าวินิจฉัยซึ่งยืนยันพยาธิสรีรวิทยาของผู้ป่วย การตรวจไม่พบการตอบสนองของ SSEP และ MEP มีความสัมพันธ์กับผลลัพธ์ที่ไม่ดีในผู้ป่วยโรคพยาธิตัวจี๊ดที่มี radiculomyelitis*