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Dual Cestode Infection in a Thai Patient (Spinal Sparganosis and Racemose Neurocysticercosis): A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ABCDEF 1 Verajit Chotmongkol
BCDE 2 Pewpan M. Intapan
BCDE 3 Kritsakorn Jingjit

1 Department of Medicine, Khon Kaen University, Khon Kaen, Thailand
2 Department of Parasitology and Research and Diagnostic Center for Emerging Infectious Diseases, Khon Kaen University, Khon Kaen, Thailand
3 Department of Surgery, Khon Kaen University, Khon Kaen, Thailand

Corresponding Author: Verajit Chotmongkol, e-mail: vercho@kku.ac.th
Conflict of interest: None declared

Patient: Male, 41
Final Diagnosis: Spinal sparganosis and racemose neurocysticercosis
Symptoms: Hip pain • numbness • weakness • blindness • hearing loss
Medication: —
Clinical Procedure: Laminectomy
Specialty: Infectious Diseases

Objective: Rare disease

Background: Spinal sparganosis and racemose (grape-like) neurocysticercosis are rare diseases. Co-infection with spinal sparganosis and racemose neurocysticercosis has never been reported in the English-language literature.

Case Report: A 41-year-old male presented after having experienced left hip pain with radiation to the leg, progressive weakness and numbness of the left leg, and difficulty urinating for 4 years, all of which was caused by spinal sparganosis. He had also experienced progressive vision and hearing loss on the right side for 3 years, which were caused by racemose neurocysticercosis. Magnetic resonance imaging (MRI) of the lumbosacral spine and the brain revealed intrinsic spinal cord lesion and racemose neurocysticercosis, respectively. Spinal sparganosis was diagnosed by histological examination and an immunochromatographic test. It was treated with a high-dose of praziquantel (75 mg/kg/day) combined with cimetidine; without clinical improvement. Treatment consisted of racemose neurocysticercosis with albendazole for 2 months. A follow-up MRI of the brain revealed a minimal reduction in the amount of racemose neurocysticercosis. Repeat treatment with albendazole was administered. However, further MRI examination of the brain revealed the condition to be unchanged. Vision and hearing loss had also not improved.

Conclusions: In this case report, we describe a middle-aged man who presented with spinal cord symptoms (hip pain, progressive weakness and numbness of the leg, and difficulty urinating) and cranial nerve palsies (vision and hearing loss) caused by spinal sparganosis and racemose neurocysticercosis, respectively. Neurological symptoms did not improve with anthelmintic drugs.

MeSH Keywords: Albendazole • Central Nervous System Parasitic Infections • Neurocysticercosis

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/910729

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Background

Sparganosis is an uncommon disease caused by the larvae of the *Spirometra* species of tapeworm. Cases of sparganosis are found mainly in China, Japan, Korea, and occasionally in Thailand. Most cases in Thailand have been found in the northeast, north, and central regions [1]. Humans are infected by drinking water contaminated with copepods carrying procercoids, ingesting the raw or uncooked meat of frogs, snakes, or birds containing plerocercoids, or using those meats as a poultice. Sparganosis frequently presents as a single subcutaneous nodule. Central nervous system (CNS) sparganosis is rare and usually presents with cerebral lesion. Intraspinal sparganosis can also occur but is extremely rare [2]. The diagnosis of sparganosis was confirmed though identification of the spargana by histopathological sectioning as well as by detection of specific antibodies in the serum using an enzyme-linked immunosorbent assay (ELISA) or immunochromatographic device [3]. Cysticercosis is a parasitic zoonosis infection caused by *Taenia solium* in its larval stage. Humans become infected either by eating fresh vegetables contaminated with feces containing *T. solium* eggs or by autoinfection when eggs are released from an adult tapeworm residing in the intestine and regurgitated into the stomach. In the intestine, the oncospheres hatch, penetrate the intestinal wall, and circulate to striated muscles, the brain, the eyes and various other types of tissue where the larvae become cysticerci. Neurocysticercosis (NCC) is the most common parasitic disease of the central nervous system. Patients present with varied neurological manifestations, depending on the location, size, and number of cysts. Racemose cysticercosis is a specific and extremely rare form of NCC that is characterized by excessive cysts without scoleces, which form grape-like clusters and present in either intraventricular or subarachnoid (cistern) form. The diagnosis of racemose NCC is typically based on neuroimaging, serology, and clinical data. We herein report a middle-aged man who presented with co-infection of both spinal sparganosis and racemose neurocysticercosis, which to our knowledge, has never been reported in the English-language literature.

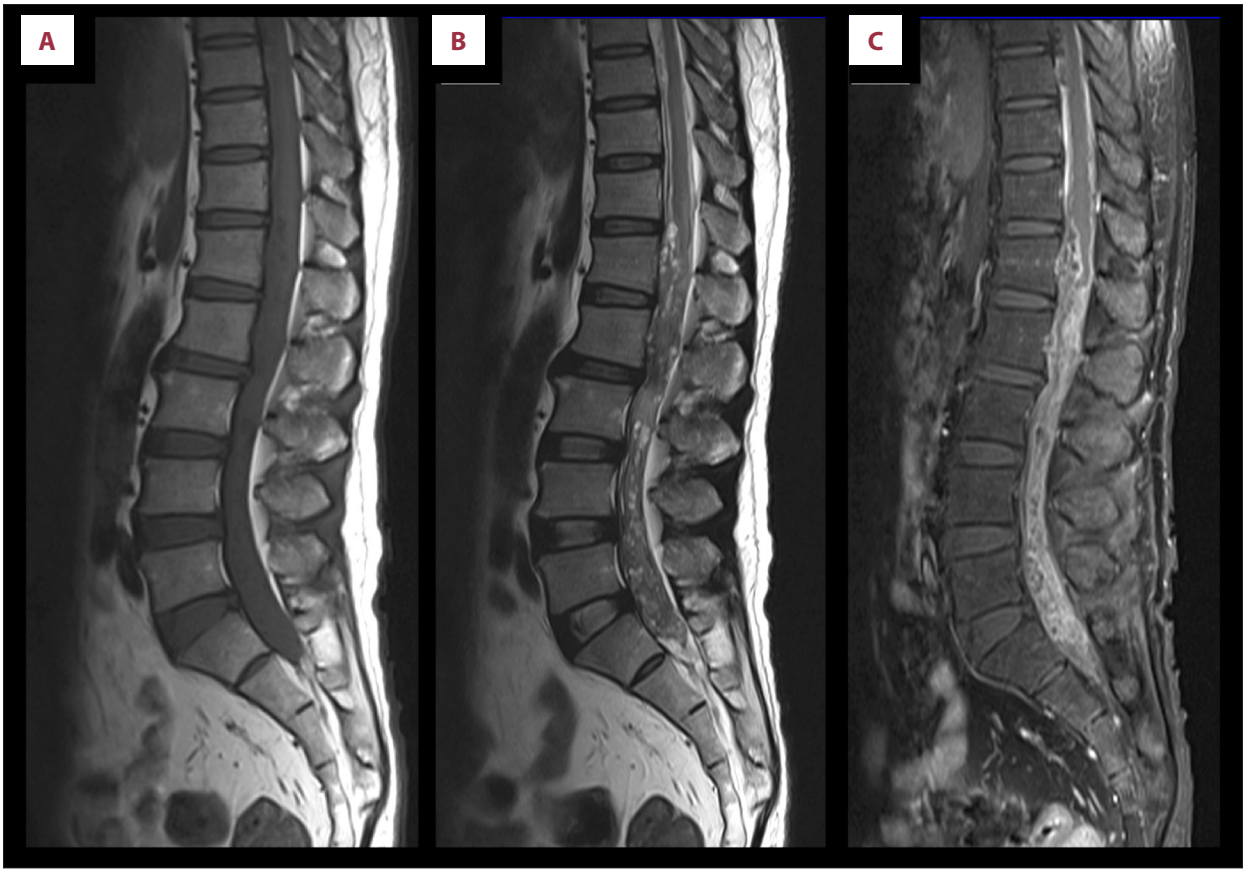

Figure 1. Lumbar sagittal magnetic resonance images: T1WI (**A**), T2WI (**B**), and gadolinium (Gd)-enhanced T1W with fat saturation (**C**) revealed intermediate signal intensity in T1WI, mixed hypo- hypersignal intensity in T2WI, and heterogeneous enhancement in T1FS with Gd at the thecal canal.

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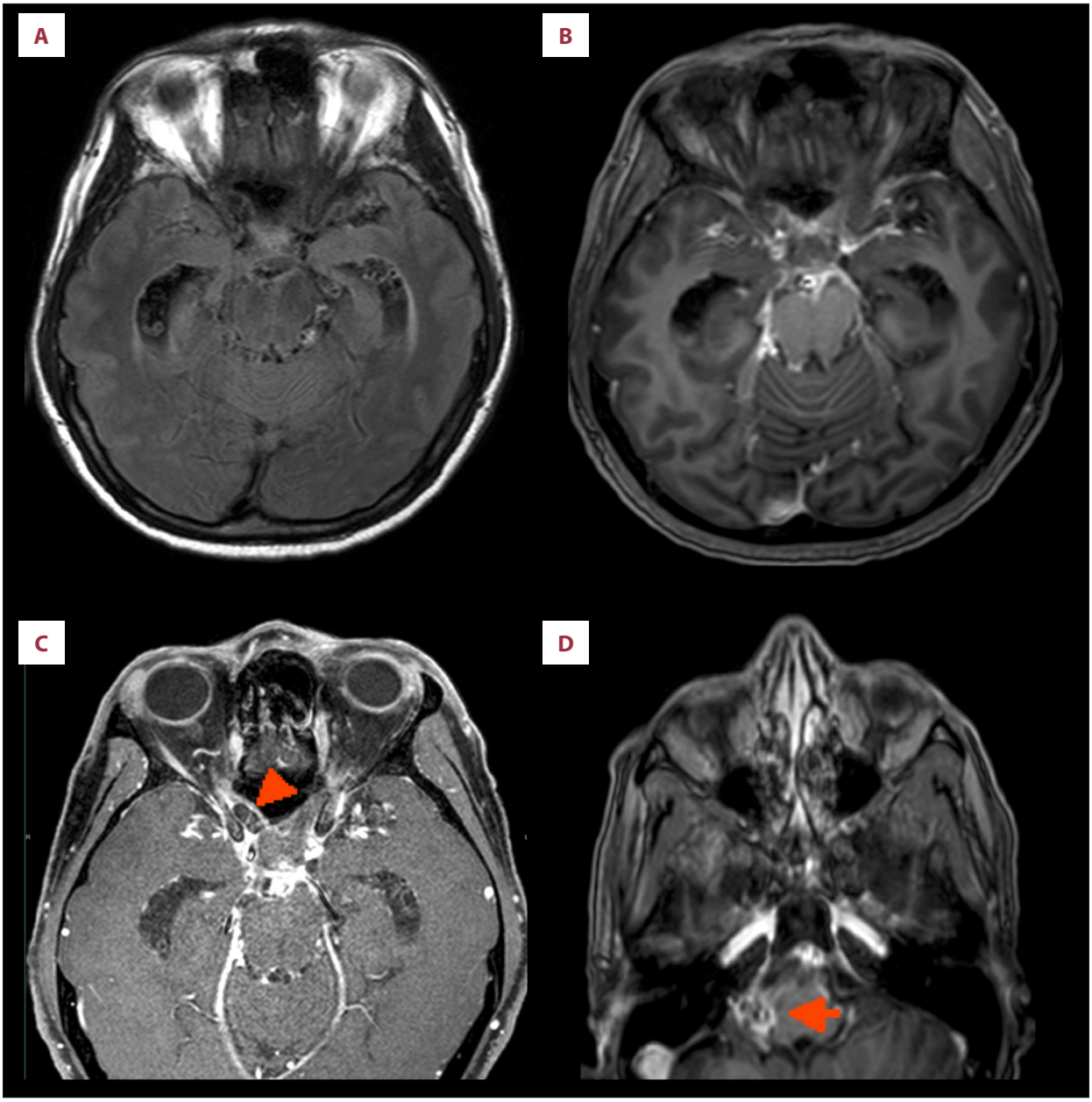

Figure 2. Magnetic resonance imaging of the brain: FLAIR (**A**) and gadolinium (Gd)-enhanced T1W with fat saturation (**B–D**) showed multiple small cystic-like lesions at the quadrigeminal, prepontine cisterns, and temporal horn of the lateral ventricles with heterogeneous enhancement (**A, B**), and rim-enhancing lesions in the right optic canal (**C**, arrow head), and right internal acoustic canal (**D**, arrow).

Case Report

A 41-year-old male living in a rural area of northeast Thailand came to the hospital in May 2016 after having experienced symptoms consisting of left hip pain with radiation to the leg, progressive weakness and numbness of the left leg, and difficulty urinating for 4 years. He had also experienced progressive vision and hearing loss on the right side for 3 years. Prior to experiencing these symptoms, he had been in good health. He had a history of occasionally consuming raw foods, but he denied having ever used a poultice. Neurological examination revealed mild muscle atrophy with motor power of grade IV/V at the proximal part and grade 0/V at the distal part, hyporeflexia, decreased pin-prick sensation, and absence of proprioception in the left leg. Reduced anal sphincter tone with absent anal reflex was also detected. An eye examination revealed blindness with optic atrophy of the right eye, and audiogram analysis showed profound sensorineural

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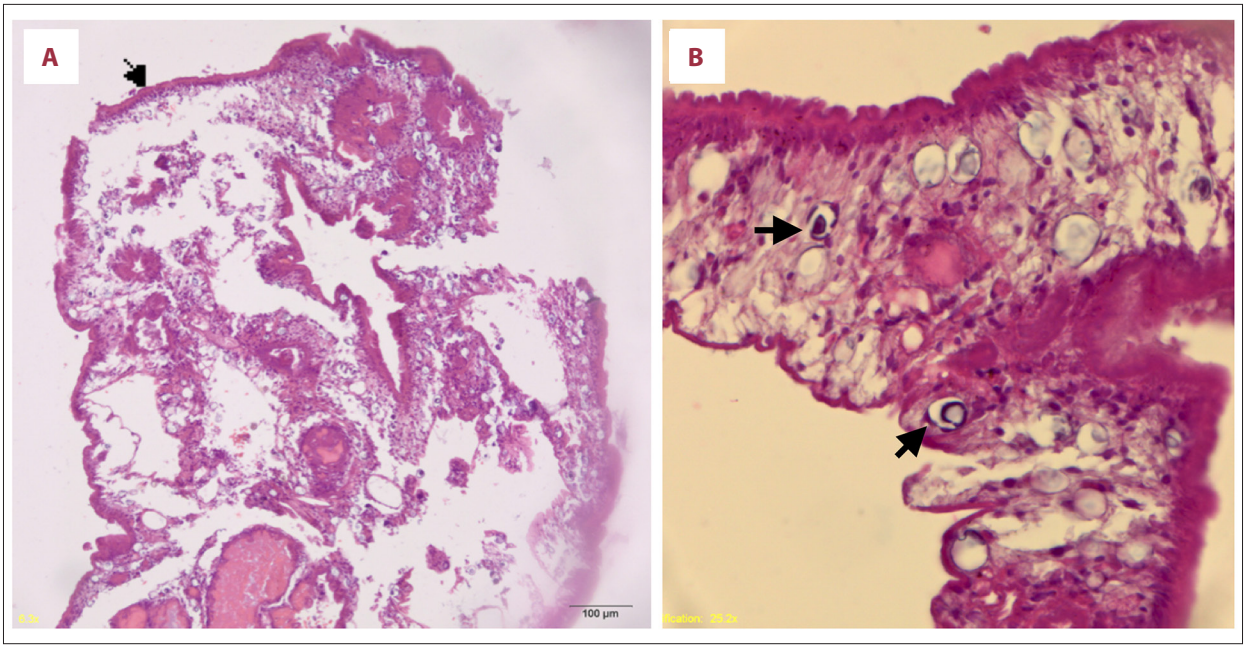

Figure 3. Hematoxylin and eosin-stained section: 10× (**A**) and 40× (**B**) revealed a piece of a parasite with a thick tegument (**A**, arrow), and calcareous corpuscles in a loose stroma (**B**, arrow).

Table 1. Clinical manifestations, laboratory results, treatment, and clinical outcomes of the patient.

Variables	Spinal sparganosis	Racemose neurocysticercosis
Symptoms	• Left hip pain with radiation to the leg, progressive weakness and numbness of left leg, and difficulty urinating for 4 years	• Progressive visual and hearing loss on the right side for 3 years
MRI abnormalities	• MRI of the lumbosacral spine revealed intermediated signal intensity in T1WI, mixed hypo- hypersignal intensity in T2WI, and heterogeneous enhancement at the thecal canal	• MRI of the brain showed multiple small cystic lesions at the quadrigeminal, prepontine cisterns, and lateral ventricles with heterogeneous enhancement, and rim-enhancing lesions in the right optic and internal acoustic canals
Serology	• An immunochromatographic test (ICT) for sparganosis antibody was positive.	• An ELISA test for cysticercus antibody was nonreactive
Treatment	• High-dose of praziquantel	• Long-term and repeat treatment with albendazole
Clinical outcome	• Not improved	• Not improved

hearing loss in the right ear. The results of MRI examinations of the spinal cord and brain are shown in Figures 1 and 2, respectively. The initial diagnosis; made based on the MRI findings; was spinal vascular malformation and racemose neurocysticercosis. Laminectomy was performed at levels L3 to L5 and inflamed infiltrative lesions with small cystic portions were observed. Tissue biopsy was only performed due to the presence of severely matted lesions. Histological examination revealed a piece of a parasite with a thick tegument and calcareous corpuscles in a loose stroma, consistent with sparganosis (Figure 3) [4]. An immunochromatographic test (ICT) for sparganosis antibody was positive, but an ELISA test to detect serum cysticercus total IgG antibody was nonreactive. The spinal sparganosis was treated with a high-dose of praziquantel (75 mg/kg/day) combined with cimetidine for 7 days. At a follow-up 9 months later, motor and sensory impairment had still not improved. The patient was given symptomatic treatment; which included analgesic drugs, physical therapy, and bladder training. Treatment consisted of racemose NCC with albendazole (15 mg/kg/day) for 2 months. A follow-up MRI of the brain revealed a minimal reduction in the amount of racemose NCC. Repeat treatment with albendazole was administered for 2 months. However, further MRIs of the brain revealed the condition to be unchanged. Vision and hearing

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loss had also not improved. Clinical manifestations, laboratory results, treatment, and clinical outcomes of the patient are summarized in Table 1.

Discussion

In cases of sparganosis that involve the nervous system, the infection is commonly found in the brain. Intraspinal sparganosis is extremely rare and is most commonly in the thoracic cord [2]. Kwon et al. reported on 1 case of spinal sparganosis and found 7 others in their literature review. These 8 cases included 6 male and 4 female patients ranging in age from 10 to 59 years. The mean duration of symptoms before diagnosis was 14.5 months (range, 3 days to 3 years) [5]. The findings of spinal cord MRI examination are nonspecific for spaganosis. Abnormal findings include nodular lesions, ill-defined; enhancing lesions, and heterogeneous intensity in T2WI with heterogeneous enhancement [2,5–7]. Conditions considered in the differential diagnosis of spinal sparganosis are granulomatous inflammations and tumors. Treatment of sparganosis consists of surgical removal of the parasite, as antiparasitic praziquantel is ineffective. In cases of spinal sparganosis, the prognosis has been found to range from fair to good [5]. However, the neurological symptoms in our patient did not improve due to the presence of severely matted lesions. Although a previous study reported treatment consisting of high-dose praziquantel therapy to be effective for cerebral sparganosis [8], the administration of high-dose praziquantel in our patient did not result in any improvement.

Neurocysticercosis frequently presents as focal lesions in the brain parenchyma (parenchymal form). Extraparenchymal NCC develops as the cysticerci reach the ventricles. Displacement of the cysts from the ventricles via cerebrospinal fluid lead to infection within the basal cisterns, sylvian fissures, and cortical/spinal subarachnoid spaces. The clinical manifestations of racemose NCC include chronic meningitis, hydrocephalus, dementia, cerebral infarction, mass effect, spinal arachnoiditis, and cranial neuropathy. The pathology of cranial neuropathy is arachnoiditis [9]. In our case, although an ELISA test to detect serum cysticercus total IgG antibody was nonreactive, racemose NCC was diagnosed based on brain MRI findings [10,11] and partial response to albendazole treatment. The ELISA test has been shown to yield a sensitivity of 64% [12]. The sensitivity of the test depends on the antigen type used for antibody detection in the serum sample. Blindness and profound sensorineural hearing loss in this patient were caused by cranial neuropathy shown in Figures 2C and 2D respectively. Treatment of racemose NCC is usually difficult and, as of yet, there have been no controlled treatment trials conducted in patients with this type of infection. Response to anthelminthic drugs varies and requires long-term treatment and repeated assessments [13]. The lack of improvement with regard to vision and hearing loss after treatment in this patient could be due to fibrous entrapment of the nerves [14].

Conclusions

Spinal sparganosis and racemose neurocysticercosis are rare diseases, and co-infection with spinal sparganosis and racemose NCC has never been reported. In our case, spinal sparganosis was diagnosed by histological examination and an immunochromatographic test. Racemose NCC was also diagnosed based on brain MRI findings and partial response to albendazole treatment. An ELISA test to detect serum cysticercus total IgG antibody was nonreactive due to the low sensitivity of the test. Neither spinal cord symptoms (hip pain, progressive weakness and numbness of the leg, and difficulty urinating) caused by spinal sparganosis nor cranial nerve palsies (blindness and sensorineural hearing loss) caused by racemose NCC, improved with the administration of anthelmintic drugs due to the presence of severely matted lesions of the spinal cord and fibrous entrapment of the cranial nerves.

Conflict of interest

None.

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