OBSERVATION

Sparganosis Presenting as a Conus Medullaris Lesion

Case Report and Literature Review of the Spinal Sparganosis

Jee-Hyun Kwon, MD; Jong S. Kim, MD

Background: Sparganosis is a rare parasitic infection affecting various organs, including the central nervous system. In rare cases, sparganosis may involve the spinal cord, usually at the thoracic area. We herein report, to our knowledge, the first case of sparganosis presenting as a conus medullaris lesion and review the literature of sparganosis involving the spinal cord.

Observation: A 42-year-old man presented with progressive perianal paresthesia and sphincter disturbances. Results of enzyme-linked immunosorbent assay of the cerebrospinal fluid and surgical biopsy were consistent with sparganum infection affecting the conus medullaris. We reviewed 7 other cases of spinal sparganosis.

Conclusions: Sparganosis may present as a conus medullaris lesion. This possibility should be considered when clinicians encounter patients with a conus medullaris lesion or cauda equina syndrome with uncertain diagnosis.

Arch Neurol. 2004;61:1126-1128

SPARGANOSIS IS A RARE PARASITIC infection caused by a migrating plerocercoid tapeworm larva of *Spirometra mansoni*. Subcutaneous tissue, skeletal muscle, visceral organs, and occasionally the central nervous system are involved.[1] In rare cases, a sparganum involves the spinal cord, usually at the thoracic area.[1-7] To our knowledge, sparganosis presenting as a conus medullaris lesion has not been reported. We herein describe such a patient and review the literature of sparganosis involving the spinal cord.

REPORT OF A CASE

Perianal paresthesia developed in a 42-year-old bus driver 2 years before the admission. The paresthesia was described as dull, heavy, and sometimes aching and was aggravated during defecation. The paresthesia gradually increased in severity, and 1 year later urinary difficulty, constipation, and erection failure developed. He had to perform intermittent catheterization by himself. He had a history of ingesting inadequately cooked frogs several years before the onset of perianal paresthesia. On examination, no skin nodule or organomegaly was found. His mental status and cranial nerve functions were all normal. He had paresthesia on both sides of the buttock and the right L5 and S1 dermatomes. There was hypesthesia in the right S3, S4, and S5 and left S4 and S5 dermatomes. His muscle power was normal in all extremities, and muscle atrophy was not observed. The Achilles tendon and bulbocavernosus reflexes were lost, and the anal tone was decreased. Babinski sign was absent on both sides.

Routine laboratory test results were within reference ranges, and no leukocytosis or eosinophilia was detected in the peripheral blood. Attempts to obtain a sample of cerebrospinal fluid (CSF) by means of spinal tapping failed, probably because of the severe adhesion caused by chronic inflammation. Magnetic resonance imaging of the spine showed nodular, masslike lesions in the conus medullaris and the adhesion of the cauda equina. Mild enhancement of the mass regions and the pial surface was seen (**Figure 1**). No lesions were found in the thoracic spinal cord, and

From the Department of Neurology, University of Ulsan College of Medicine, Asan Medical Center, Seoul, Korea.
The authors have no relevant financial interest in this article.

magnetic resonance image findings of the brain were normal. Results of an enzyme-linked immunosorbent assay were negative for cysticercosis or paragonimiasis, whereas the titer for sparganosis was significantly elevated in the serum (optical density, 0.864; cutoff value, 0.240) and CSF (optical density, 0.650; cutoff value, 0.250) samples obtained during the laminectomy. Results of acid-fast bacilli and India ink staining were negative, and CSF culture yielded no bacteria or tuberculosis.

On operation, the arachnoid membrane was thickened with yellowish-whitish discoloration due to severe inflammation and adhesion. Swollen, lobulated, yellowish granulation tissues were adhered to the cauda equina. These findings were consistent with chronic inflammation. The worm was not detected in the initial operation.

The patient received corticosteroid therapy, and his perianal paresthesia moderately improved. However, he persistently complained of a dull, uncomfortable sensation on both buttocks. To make a definitive diagnosis, and to remove the worm if possible, we performed a second operation 8 months later. This time, we were able to detect 1- to 2-mm milky white, soft, nodular masses surrounded by the adhesional nerve fibers beneath the dura mater and remove the worm. The worm had microscopic features characteristic of a sparganum (**Figure 2**). After the second operation, corticosteroid therapy was used again. His perianal paresthesia further improved. However, he still had a heavy sensation on both buttocks during defecation after 12 months of follow-up. Urination and defecation difficulties also persisted.

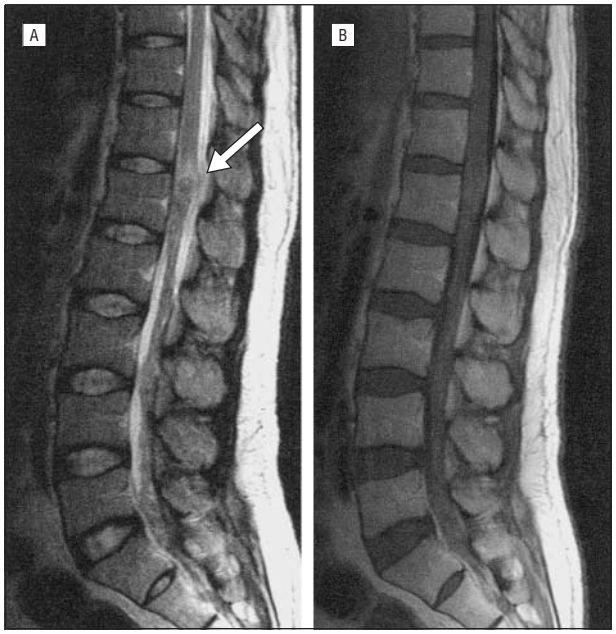

Figure 1. A, T2-weighted magnetic resonance image (MRI) shows nodular, masslike lesions in the conus medullaris (arrow) and a thickened thecal sac with adhesion of the cauda equina. B, T1-weighted MRI shows mild enhancement in the pial surface and the masslike lesion.

Figure 2. The histological specimen shows a worm with smooth outer surface of cuticle, widely distributed calcospherules (long arrows) and abundant muscle fibers (short arrow) (hematoxylin-eosin, original magnification ×200).

COMMENT

Our patient had paresthesia on the sacral areas and legs that was associated with sphincter disturbances. Magnetic resonance image and surgical findings confirmed the presence of chronic inflammation in the conus medullaris and the cauda equina. We initially made a diagnosis of sparganosis by means of enzyme-linked immunosorbent assay findings, which were finally confirmed by locating and removing a worm during the second operation. The assay using antisparganum antibody (IgG) has been shown to have a high sensitivity (85.7%-100%) and specificity (95.7%) in this condition.[8]

Although human sparganosis has been found worldwide, it is detected most frequently in east Asia. In this region, human infection develops by way of (1) drinking untreated water containing infected copepods (first intermediate host), (2) ingesting raw or inadequately cooked flesh of snakes or frogs infected with the sparganum (second intermediate host), and (3) applying the flesh of an infected intermediate host as a poultice to a wound.[9] The most likely pathogenesis of infection in our patient was the ingestion of inadequately cooked frogs.

In the literature review, we found 7 cases of spinal sparganosis. The details are summarized in the **Table**. Including our patient, there were 6 male and 2 female patients ranging in age from 10 to 59 years (mean age, 37.9 years). Presenting signs and symptoms included voiding difficulty (n=5), weakness of the limbs (n=6), sensory disturbances (n=6), and pain (n=4). Pleocytosis of the CSF was present in 2 patients, and elevated CSF protein levels were noted in 3 patients. The mean duration of symptoms before diagnosis was 14.5 months (range, 3 days to 3 years). The prognosis was good or fair.

In most of these patients, the lesions were detected in the thoracic cord. Our patient is unique in that the lesion was restricted to the conus medullaris/cauda equina. The route of the entry into the spinal cord remains unclear, but hematogenous spread seems likely. Although

Summary of Spinal Sparganosis Cases

Source	Age, y/ Sex	Mode of Infection	Initial Symptoms and Signs	Lesion Location	CSF Finding/ Eosinophilia, %*	Operative Findings/ Presence of Worm	Duration Before Diagnosis	Outcome
Lee and Sohn,[2] 1965	42/M	Raw snake	Burning sensation in abdomen/thorax, paraplegia, urinary incontinence	Extradural, T5	WBC, 0; protein, 45/4	Yellowish granulation tissue/yes	3 d	Fair
Park et al,[3] 1972	45/M	Raw frog and snake	Tingling sensation in arms, paraparesis, urinary incontinence	Extradural, C7-T2	WBC, 0; protein, 268/ no information	Yellowish granulation tissue/yes	5 mo	Good
Park et al,[4] 1983	40/M	Contaminated water	Low back pain after trauma	Intradural, below T10	WBC, 98; protein, 225/ no information	Caseous necrosis/no (diagnosis made by ELISA)	4 mo	Good
Lo et al,[5] 1987	43/F	Contaminated water and frog	Low back pain and paraparesis, sensory loss over the sacral region and left lateral aspect of leg	Spinal canal from the lower cervical region to the cauda equina	WBC, 1; protein 15/3	Adhesion of distal spinal cord and lumbosacral roots/yes	3 y	Fair
Fung et al,[6] 1989	22/M	Contaminated water	Lumbar pain, paraparesis, urinary incontinence, sensory loss below T8	Intradural, T8-T9	No information/4	Encapsulated solid mass/yes	3 y	Good
Cho et al,[1] 1992	59/M	Raw fish and cooked snake	Paraparesis, urinary incontinence	Intradural, T10-T11	No information	Milkish, nodular mass and granulation tissue/yes	3 mo	Good
Kudesia et al,[7] 1998	10/F	Unknown	Back pain, paraparesis, sensory loss (below T12 and in perianal area)	Intramedullary, T8-T10	No information/ mild	Irregularly lobulated mass, calcification/yes	8 mo	Good
Present report	42/M	Raw frog	Perianal paresthesia, sphincter disturbances	Intramedullary, conus medullaris, and cauda equina	WBC, 110; protein, 163/1.8	Nodular organisms surrounded by the adhesional nerve fibers and yellowish granulation tissue/yes	2 y	Fair

Abbreviations: ELISA, enzyme-linked immunosorbent assay; WBC, white blood cell count.
*WBCs are reported as 10^3 cells per microliter; protein, as grams per deciliter.

eosinophilia is often present in patients with parasitic diseases, it usually is not found in patients with spinal cord sparganosis, as in our patient. Our data illustrate that sparganosis should be suspected when clinicians encounter patients with conus medullaris or cauda equina syndrome with uncertain diagnosis.

Accepted for publication November 6, 2003.

Author contributions: *Study concept and design* (Dr Kim); *acquisition of data* (Dr Kwon); *analysis and interpretation of data* (Drs Kwon and Kim); *drafting of the manuscript* (Drs Kwon and Kim); *critical revision of the manuscript for important intellectual content* (Dr Kim); *administrative, technical, and material support* (Dr Kim).

Correspondence: Jong S. Kim, MD, Department of Neurology, Asan Medical Center, Song-pa PO Box 145, Seoul 138-600, South Korea (jongskim@amc.seoul.kr).

REFERENCES

1. Cho YD, Huh JD, Hwang YS, Kim HK. Sparganosis in the spinal canal with partial block: an uncommon infection. *Neuroradiology.* 1992;34:241-244.
2. Lee CW, Sohn K. A case of sparganum mansoni in spinal canal. *J Korean Surg Soc.* 1965;7:155-158.
3. Park SH, Rhee JD, Kang SK, Kim JK. A case report of *Sparganum mansoni* in the spinal canal. *J Korean Neurosurg Assoc.* 1972;1:204-207.
4. Park CK, Ha YS, Huh CW, Song JU. A case of sparganosis in the intradural space of the thoracolumbar spine. *J Korean Neurosurg Assoc.* 1983;2:739-743.
5. Lo YK, Chao D, Yan SH, et al. Spinal cord proliferative sparganosis in Taiwan: a case report. *Neurosurgery.* 1987;21:235-238.
6. Fung CF, Ng TH, Wong WT. Sparganosis of the spinal cord: case report. *J Neurosurg.* 1989;71:290-292.
7. Kudesia S, Indira DB, Sarala D, et al. Sparganosis of brain and spinal cord: unusual tapeworm infestation (report of two cases). *Clin Neurol Neurosurg.* 1998; 100:148-152.
8. Kim H, Kim SI, Cho SY. Serological diagnosis of human sparganosis by means of micro-ELISA. *Korean J Parasitol.* 1984;22:222-228.
9. Chang KH, Cho SY, Chi JG, et al. Cerebral sparganosis: CT characteristics. *Radiology.* 1987;165:505-510.