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.

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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
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 defeation after 12 months of follow-up. Urination and defeation difficulties also persisted.

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 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
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.

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

Abbreviations: ELISA, enzyme-linked immunosorbent assay; WBC, white blood cell count.

*WBCs are reported as 10³ cells per microliter; protein, as grams per deciliter.

REFERENCES