Sparganosis of the Spinal Canal: Rare Tapeworm Infection as a Cauda Equina Mass with Magnetic Resonance Imaging

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ABSTRACT

Intraspinal sparganosis is a very rare disease caused by infestation of migrating larva of Spirometra mansonoides. Only 7 cases of intraspinal sparganosis have been reported in the literature. We report a case in a 26-year-old woman presenting with left side sciatica and intermittent urinary incontinence for more than a year. Magnetic resonance imaging (MRI) revealed a large cauda equina mass with heterogeneous enhancement in the L4 to S2 spinal canal. An intradural mass matted with the nerve roots of the cauda equina was found during surgery. Pathological examination revealed chronic granulomatous inflammation with fragments of a helminth sparganum. We present this intraspinal sparganosis case with emphasis on its rarity, MRI features of this unique cauda equina inflammatory mass, and a literature review.

CASE REPORT

This 26-year-old woman developed skin rash and severe itching over the face, trunk and extremities for 6 months. In this period she was joining a study tour in the United States in 2007. She was treated as hay fever. One month later, she began to have lower back and left buttock pain with radiating pain and numbness in the left thigh, calf, and foot, which continued for about one and half years. Intermittent urinary incontinence was also noted for a year. Her left leg weakness and walking disturbance aggravated for 2 months before admission to our hospital. She had thalassemia but denied any other systemic diseases.

Her general physical examination was unremarkable. Neurological examination revealed hyposensation to heat, pin-trick and touch, numbness, and mild muscle weakness in the left lower extremity. The left Babinski reflex was absent. Routine laboratory study including complete blood count (normal eosinophil count 2.3%), blood biochemistry, glucose, eosinophil sedimentation rate, urinalysis, and chest radiograph showed no abnormalities. Urodynamic study revealed neurogenic bladder. Serological testing (enzyme-linked immunosorbent assay [ELISA]) for sparganum was not performed.
Lumbar spine MRI with and without gadolinium enhancement study revealed a large intradural mass lesion, about 10 cm in length, in the upper L4 to S2 spinal canal (Fig. 1), which extended along bilateral S1 and S2 nerve roots into the corresponding bilateral neuroforamina (Fig. 2). The cerebrospinal fluid (CSF)-filled subarachnoid space was completely obliterated in the L4 to S2 spinal canal. This intradural mass appeared with mild heterogeneous hypointense signal on T1W image and coarse heterogeneous hyperintense signal on T2W image, and with some small irregular extralow T1 and extralow T2 signal areas (Fig. 1). Some scattered tiny cystic lesions with bright T2...
signals (Fig. 1b, 3) were also seen in the cauda equina mass.
The enhancement of this mass was mild to moderate with
some heterogeneity (Fig. 1c, 1f). The adjacent dura was
only minimally enhanced, but without diffuse or nodular
thickening on MRI. The individual nerve roots of the cauda
equina could not be identified intradurally in the L4 to S2
spinal canal.

The patient underwent a lumbar laminectomy from L3
to S2 under the impression of cauda equina mass. The dura
was opened to expose the mass, which occupied the whole
spinal canal of L4 to S2 with complete obliteration of the
subarachnoid space. It was about 10 cm in length, elastic,
and gray-white (Fig. 4). The nerve roots of the cauda equina
were matted with the mass and could not be dissected away
from it. Only partial excision was performed. An artificial
dura graft was used for dural augmentation. After surgery
the left side sciatic pain subsided and left leg muscle power
improved. However, her neurogenic bladder persisted and
she received follow-up physical therapy for bladder training.

Pathological study revealed chronic granulomatous
necrotizing inflammation with fragments of helminth body
(Fig. 5a). Rudimentary bothrium at the anterior end of the
parasite and calcareous corpuscles were found in the para-
site, features of which are characteristics of the sparganum
(Fig. 5b).

A stool examination revealed no evidence of para-
sites or parasite ova after the operation. A single dose of
paraziquine (5 mg/kg) was given to this patient. Brain
MRI with and without enhancement study after surgery
showed no definite abnormality in the brain.

DISCUSSION

Manson discovered the first human sparganosis at
autopsy in Amoy, China, in 1882 [11]. Since then human
sparganosis have been reported worldwide, particularly
in China, Japan, Korea and Southeast Asia areas, and
less commonly in the United States and Europe [2]. The
life cycle of Spirometra has been well documented by
Muller and coworkers [2]. The adult tapeworm lives in the
gastrointestinal tract of the definite host, usually dogs, cats
and carnivores. Eggs are passed in the feces of the definite
host and develop into ciliated coracidia in water. The first
intermediate host, Cyclops, ingests the coracidia, which
develops into a procercoid (first stage larve). Frogs or snakes
drink water containing infected Cyclops with the procer-
coid, becoming the second intermediate host. In the second
intermediate hosts, the procercoid penetrates the intestinal
wall and migrates to various tissues, usually to muscle and

Figure 2. Axial a. and Coronal b. Gd-enhanced images
show extension of the inflammatory mass along the bilat-
eral S1 and S2 nerve roots into the corresponding bilateral
neuroforamina (arrows).
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subcutaneous tissue, and then grows into a plerocercoid (second stage larva), the sparganum. The sparganum in the infected second intermediate host is consumed by the definite host, thus continuing the life cycle. Humans can be infected accidentally as intermediate hosts by drinking water contaminated with infected copepods, such as Cyclops; by ingestion of raw or inadequately cooked snake or frog; or by applying the flesh or skin of an infected host as a poultice on a skin wound. Our patient lived in Taipei and she was not an aborigine. She denied having ingested raw flesh or having used a poultice. A review of her history no infection route could be traced in Taiwan. However, this patient had been on a study tour in San Diego, California, and Mexico in 2005 and had swum in a river in Mexico at that time. She participated in another study tour in California and New York in 2007. She started to have skin rash and severe itching during the stay of last study tour. However, the patient could not remember any probable contamination route she had experienced during the period of travel. Thus, the potential infectious route for this patient could not be ascertained.

When human acquires the infection, the larva penetrates the intestinal wall and migrates to various organs, with most of them lodging in the subcutaneous tissue and skeletal muscle of the body as slowly growing and migratory nodules. Intraspinal sparganosis is extremely rare. We believe the real number of cases might be underestimated. To our knowledge, only 7 cases of intraspinal sparganosis have been reported in literature. Five of these cases were intradural lesions with locations in the mid-thoracic, T11 to T12, T8 to T10, thoracolumbar, and cervical to cauda equina regions [6-10]. Two of the 7 cases were extradural lesions [12, 13]. In geographic distribution, four cases occurred in Korea, 1 in Taiwan, 1 in Hong Kong, and 1 in India.

In the previous 7 case reports of intraspinal sparganosis only that of Cho et al in 1992 included an MRI study [8]. In that case the brief description of the spine MRI revealed a diffuse intradural mass at T10 to T11 with mild enhancement. In our study, the lumbar MR study exhibited the following peculiar features: 1. the intradural inflammatory mass involving the cauda equina was large, up to 10 cm, and extended along the bilateral S1 and S2 nerve roots into the corresponding neuroforamina. The large size was probably related to the longevity of the worm, migratory movement and the effects of proteases secreted

**Figure 3.** MR myelography study shows obliteration of the cerebral spinal fluid (CSF) space below the upper L4. Multiple scattered, variably sized small cyst-like areas with high T2 signal are seen in the mass, probably due to trapped CSF spaces in the inflammatory mass.

**Figure 4.** The picture shows the dura opening after L-spine laminectomy. The dura mater is minimally thickened but does not severely adhere to the mass. The mass surface is rough and gray-white mixed with pink granulation tissue. The nerve roots of the cauda equina are matted in the granulomatous inflammatory mass with difficulty in dissecting them away from the mass.
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1. The abundant CSF-filled subarachnoid space was completely obliterated. Instead, we could see multiple scattered, variably sized small cysts with high T2 signal in the mass, which were rather unique findings in this MR image (Fig. 1b, 3). We supposed that these small cysts were likely due to small trapped CSF spaces in the inflammatory mass.  

2. The MRI showed some heterogeneous T1 and T2 signals within the cauda equina mass, including small irregular extralow T1 and extralow T2 signal areas. These heterogeneous signals probably resulted from this necrotizing granulomatous inflammation matted with the cauda equina nerve roots, trapped CSF cysts, fragments of helminth, some small chronic hemorhages and calcifications [4, 14-16]. Song et al described characteristic findings of a tunnel sign, approximately 4 cm in length and 0.8 cm in width, in cerebral sparganosis on MRI [14]. We could not see this tunnel sign on a lumbar MR, probably because of the small space in the spinal canal and absence of cord involvement.  

3. The mass had mild to moderate enhancement with some heterogeneity. This is different from a cauda equina tumor which is usually homogenous with more intense enhancement. 

The differential diagnosis should include other causes of intraspinal granulomatous inflammation and intradural tumors in the cauda equina area. The MRI findings exhibited some features of chronic granulomatous inflammation, but they were not specific for intraspinal sparganosis and may be indistinguishable from other causes of chronic granulomatous inflammation such as tuberculosis, fungus or even other parasites. The MRI findings in spinal tuberculosis meningitis or arachnoiditis include CSF loculi, obliteration of the spinal subarachnoid space, small nodules, and matting of the nerve roots in the lumbar region [17, 18]. Spinal tuberculosis meningitis is usually associated with nodular or linear dural thickening and enhancement. Thus, some of our MRI findings were similar, but not identical, to tuberculosis arachnoiditis and other causes of chronic granulomatous inflammation. Since our intraspinal sparganosis case was rather unique in resembling a cauda equina tumor, the differential diagnosis should also include the more frequently encountered filum terminale myxopapillary ependymoma and schwannoma [19]. Other possible differential diagnoses include meningioma, metastatic tumors, and rarely the paraganglioma, but these tumors are usually smaller in size. The myxopapillary ependymoma of the filum terminale or conus medullaris is a slow growing tumor that may become large and expand the lumbosacral canal and neural foramina. Ependymomas are usually enhanced strongly and homogenously on computed tomography or MRI, but may be heterogeneous in some cases when hemorrhage or necrosis is present. Usually the nerve roots of the cauda equina are displaced by an ependymoma and can be identified individually, rather than matting together. The schwannoma is generally a benign lesion but may become gigantic in the cauda equina area. Its MRI findings are similar to myxopapillary ependymoma. The rather homogeneous and intense enhancement in ependymoma, schwannoma, meningioma and paraganglioma is

Figure 5. a. Microscopic study shows chronic necrotizing granulomatous inflammation with the presence of a parasite body. (H&E, x100). b. A section of a parasite body shows cleft-like rudimentary bothrium at the anterior end of the worm (arrow), and parenchyma containing scattered bundles of longitudinal muscle fibers, branching excretory channels, mesenchymal fibers and calcareous corpuscles in a loose stroma. (H&E, x200)
characteristic and helpful in distinguishing these lesions from chronic granulomatous inflammation.

The common route of sparganosis infection in humans has been described, but the exact route of infection to the brain and spine is still not known. The larvae may migrate anywhere in the body, mostly at subcutaneous tissue and flesh, and it is believed that the foramina of skull base or spine might be the entry sites [14-16]. In our case the granulomatous inflammation extended along the bilateral S1 and S2 nerve roots into the corresponding neuroforamina. Some authors have also proposed a hematogenous route [9]. It is probable that a few tiny procercoid stage larva found in Cyclops which is consumed by human can accidentally enter the blood stream and migrate to different destinations.

The antiparasitic praziquantel is effective in cerebral cysticercosis and paragonimiasis, but its efficacy is not satisfactory in the treatment of CNS sparganosis. Surgical removal of the worm or granulomatous inflammation is the best treatment for intraspinal or CNS sparganosis [14-16]. Since the granulomatous inflammatory mass in our patient was severely matted with the intradural nerve roots of the cauda equina, a laminoplasty and only partial resection were performed. The left sciatic pain and left leg weakness improved after surgery. The neurogenic bladder persisted. She was followed up at our clinic and received physical therapy for bladder training.

The diagnosis of intraspinal sparganosis is rarely made before the surgery. It is difficult to make a definite diagnosis for intraspinal sparganosis based on clinical symptoms and neurological findings. Thus MRI imaging study plays an important role in this regard, as it identifies some peculiar features of intraspinal sparganosis. Combined with the clinical history and a positive ELISA serological test for sparganum, the MRI imaging findings with peculiar features of intraspinal sparganosis may establish a correct pre-operative diagnosis.

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