SNI

SURGICAL NEUROLOGY INTERNATIONAL

OPEN ACCESS

For entire Editorial Board visit : http://www.surgicalneurologyint.com

Editor: **James I. Ausman,** MD, PhD University of California, Los Angeles, CA, USA

SNI: Infection, a supplement to *Surgical Neurology International*

Case Report

A case of disseminated central nervous system sparganosis

Raywat Noiphithak, Gahn Doungprasert

Division of Neurosurgery, Department of Surgery, Thammasat University Hospital, Faculty of Medicine, Thammasat University, Pathumthani, Thailand

E-mail: *Raywat Noiphithak - raywat_n@yahoo.co.th; Gahn Doungprasert - Gahn.md@gmail.com
*Corresponding author

Received: 01 August 16 Accepted: 07 October 16 Published: 05 December 16

Abstract

Background: Sparganosis is a very rare parasitic infection in various organs caused by the larvae of tapeworms called spargana. The larva usually lodges in the central nervous system (CNS) and the orbit. However, lumbar spinal canal involvement, as noted in the present case, is extremely rare. We report a rare case of disseminated CNS sparganosis involving the brain and spinal canal and review the literature.

Case Description: A 54-year-old man presented with progressive low back pain and neurological deficit at the lumbosacral level for 2 months. Imaging indicated arachnoiditis and an abnormal lesion at the L4-5 vertebral level. The patient underwent laminectomy of the L4-5 with lesionectomy and lysis of adhesions between the nerve roots. Microscopic examination indicated sparganum infection. Further brain imaging revealed evidence of chronic inflammation in the left parieto-occipital area without evidence of live parasites. In addition, an ophthalmologist reported a nonactive lesion in the right conjunctiva. The patient recovered well after surgery, although he had residual back pain and bladder dysfunction probably due to severe adhesion of the lumbosacral nerve roots.

Conclusion: CNS sparganosis can cause various neurological symptoms similar to those of other CNS infections. A preoperative enzyme-linked immunosorbent assay is helpful for diagnosis, especially in endemic areas. Surgical removal of the worm remains the treatment of choice.

Key Words: CNS, diagnosis, disseminated, sparganosis, treatment

Access this article online

Website: www.surgicalneurologyint.com

DOI: 10.4103/2152-7806.195236

Quick Response Code:

INTRODUCTION

Sparganosis is a parasitic infection caused by the pleocercoid larvae of pseudophyllidean tapeworms, which are called spargana.[15] This infection is reported worldwide; however, it is most common in eastern Asia.[3] Sparganum infection has been documented in various organs, including the central nervous system (CNS) and the orbit;[15,17] CNS involvement is extremely rare. Therefore, preoperative diagnosis of CNS sparganosis is relatively difficult and is exclusively performed through histopathology. We report a rare case of disseminated CNS sparganosis involving the brain and the spinal cord and review the literature.

CASE REPORT

A 54-year-old Thai man presented with low back pain lasting for 2 months. The symptom was gradually progressive and was aggravated by movements. The

How to cite this article: Noiphithak R, Doungprasert G. A case of disseminated central nervous system sparganosis. Surg Neurol Int 2016;7:S958-61.
http://surgicalneurologyint.com/A-case-of-disseminated-central-nervous-system-sparganosis/

pain was referred to both legs and did not improve with medications. Later, the patient experienced weakness and numbness in his right leg and had walking difficulties. In addition, he had urinary retention which required urinary catheterization. The patient's symptoms worsened and limited his daily activities. Examination revealed generalized erythematous plaques on his head, trunk, back, and extremities. He also had several flaccid blisters. The patient had grade III motor weakness and paresthesia in his entire right leg; however, there was minimal motor weakness on the left side. Sacral sensation and sphincter tone were intact. Magnetic resonance imaging (MRI) of the lumbar spine showed a pattern of arachnoiditis at the L1-5 level. There were also multiple hypersignal intensity bead-like lesions on T2-weighted images in the spinal canal of the L4-5 vertebral level. The lesions were not enhanced with contrast [Figure 1]. Laminectomy in the L4-5 with lesionectomy was performed because we considered a spinal infection resulting in the recent neurological deficits. During surgery, we observed that the nerve roots were severely clumped together and had arachnoid adhesion. The lesions were found to be multiple tapeworms between the nerve roots and were completely removed with partial lysis of the adhesions under nerve root monitoring [Figure 2]. Microscopic examination demonstrated the presence of spargana with thick outer eosinophilic teguments and inner calcospherites [Figure 3]. Therefore, spinal sparganosis was diagnosed.

The patient was a monk on pilgrimage and had ingested uncooked frogs, snakes, and other amphibians for 10 years. The year prior to his visit he was diagnosed with bullous pemphigoid and was on high-dose prednisolone since then. Further investigation was done to rule out potentially disseminated sparganosis. In addition, the patient had a history of epilepsy, which was neither investigated nor treated. Although brain MRI indicated cerebral atrophy with surrounding hypersignal intensity in the left parieto-occipital area on T2-weighted images, which is compatible with old inflammation, there was no evidence of the live parasite in the brain [Figure 4]. An ocular examination by an ophthalmologist indicated the presence of an abnormal cystic, worm-like mass with calcification in the right conjunctiva. This was considered a nonactive lesion. However, we did not perform an enzyme-linked immunosorbent assay (ELISA) for sparganum or a cerebrospinal fluid (CSF) study. A stool examination revealed no evidence of parasite infection.

In the early postoperative period, the patient still had severe back pain; however, this improved during follow-up after a few months. Radicular pain, and weakness were also improved; however, neurological function of the urinary bladder was still deficit.

DISCUSSION

Sparganosis is a rare parasitic infection caused by larvae of the parasitic tapeworms of *Spirometra*.[1,3,5] Human sparganosis was first reported in China[6] and is most common in eastern Asia. The adult tapeworm lives in the gut of the definite host, which is usually dogs, cats, and carnivores. Eggs of the parasite are passed from the definite host into water and develop into coracidia, which are ingested by the first intermediate host, the Cyclops. The coracidia then develop into a procercoid stage in the Cyclops. Amphibians and reptiles, especially frogs and snakes, ingest water contaminated with infected Cyclops and become the second intermediate host. The procercoid in the second intermediate host migrates through the intestinal wall into other tissues and further develops into a plerocercoid, which is the sparganum. Finally, the sparganum is ingested by the definite host and continues its life cycle.[10] Humans can serve as an

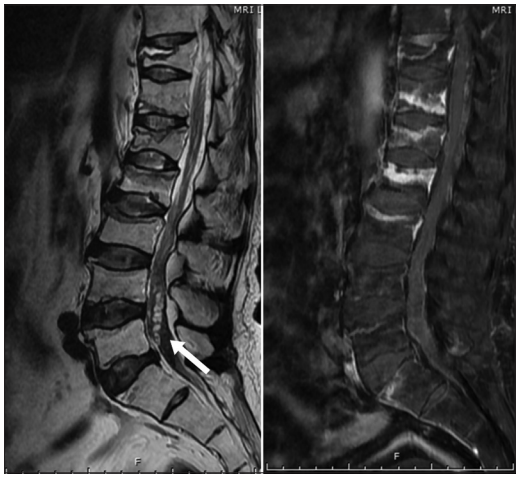

Figure 1: Left: MRI of lumbar spine showing a hypersignal intensity bead-like lesion on a T2-weighted image in the L4-5 vertebral level (white arrow). Right: the lesion was not enhanced on a contrast image

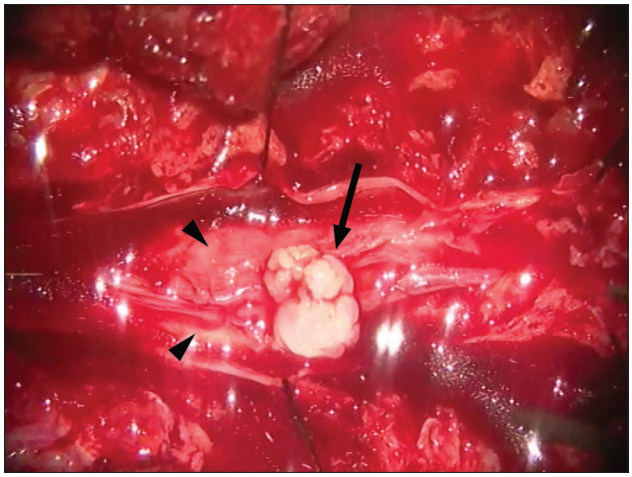

Figure 2: Intraoperative findings after laminectomy of the L4-5 demonstrated the presence of spargana (arrow) and clumping of the adjacent nerve roots (arrowhead)

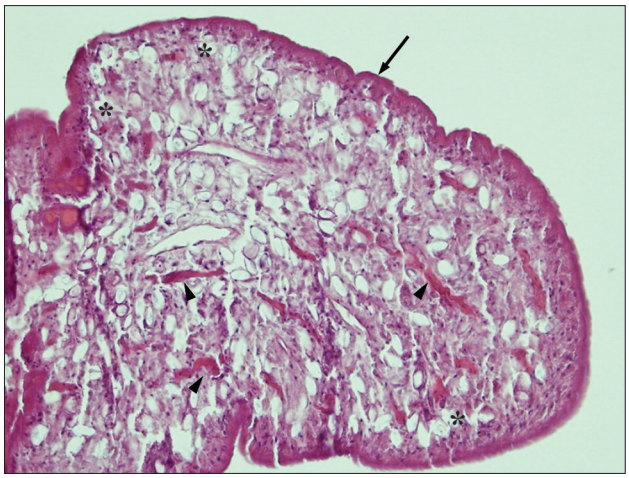

Figure 3: Microscopic findings of sparganum indicated thick eosinophilic tegument (black arrow), subtegumental calcospherites (asterisk), and longitudinal strips of muscle (arrowhead)

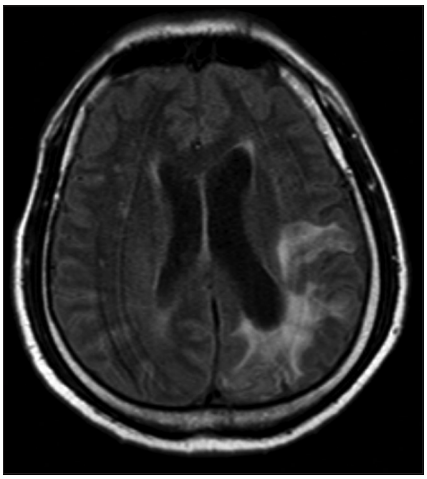

Figure 4: Brain MRI displayed evidence of an old lesion caused by larval migration. This lesion presented as cerebral atrophy with surrounding hypersignal intensity in the left parieto-occipital area on a fluid-atenuated inversion recovery image

intermediate host of the parasite and can be infected in three ways, namely by drinking water contaminated with copecidia, by eating amphibians or reptiles infected with sparganum, or by direct contact of wound with infected animals.[9,15,17]

Human sparganosis can occur in various organs. The most common sites of infection, as indicated in a publication from Thailand, are the eye, subcutaneous tissue, and the CNS.[17] A the study by Park *et al.* indicated that sparganosis in the CNS, particularly in the spinal canal, is relatively rare. Only nine cases have been reported in the literature.[12] The most common clinical presentation of cerebral sparganosis is a long history of seizure, as seen in our case.[15] However, our patient was diagnosed with sparganosis due to a spinal lesion instead of cerebral sparganosis owing to a history of epilepsy. This is because the cause of his epilepsy was not further investigated and he came to hospital with back pain, radicular pain, neurogenic bladder, and paraparesis. Nevertheless, our patient had a history of eating raw frogs, which contributed to the diagnosis.[11] Moreover, he was investigated for sparganosis at other sites because he had taken immunosuppressive drugs for a long time. Further, we found that he had ocular involvement, which could occur with sparganosis in patients with an immunocompromised status.[13]

Computed tomography and MRI may be helpful; however, these are nonspecific tools for detection of CNS sparganosis.[8,12,14,16] Although Song *et al.* have reported the tunnel sign as the most common finding in MRI, we did not observe this finding in our patient because his cerebral lesion may have been inactive and the lesions in the spinal canal were too small.[14] MRI of our patient indicated the presence of characteristics of arachnoiditis in the lumbar spinal canal and multiple regions of localized cerebral atrophy with white matter degeneration. This represented previous inflammation in these areas and was evidence that alerted us to possible parasitic infection. ELISA for anti-sparganum antibodies in the CSF or the serum is highly sensitive and specific for the diagnosis of sparganosis.[7] However, diagnosis is usually performed after surgical removal due to the rarity of the condition. Therefore, most patients are not diagnosed using this test.

Antiparasitic drugs for tapeworm, such as praziquantel, are ineffective for the treatment CNS sparganosis.[15,16] Surgical removal is the treatment of choice and provides favorable outcomes.[8,15,16] However, not all patients should undergo surgery because it seems to be beneficial only for cases with live worms or localized inflammatory granulation.[12] If patients are not good candidates for surgery, conservative treatment may be an acceptable option because of the spontaneous regression of granulation tissue after the death of the worm.[15] In these cases, ELISA can contribute to the diagnosis of sparganosis. Although it is difficult to determine whether the worm is alive on MRI,[2,4] highly enhanced and localized compressive lesions may indicate live worms. We considered surgical resection in this patient because there was a localized lesion in the spinal canal of the L4-5 vertebral level and the patient had symptoms compatible with the corresponding lesions in spite of the absence of enhancement on MRI. We found many worms loculating between the clumped nerve roots because of arachnoiditis, and attempted to remove all of the worms. We also lysed the adhesions. Even though back pain was not completely resolved because of the arachnoid adhesion between the nerve roots, the weakness and radicular pain were improved after the operation.

CONCLUSION

Although sparganosis is a very rare CNS infection, it can cause various neurological symptoms and should be considered as a differential diagnosis. In addition to history taking and imaging, preoperative ELISA can be helpful for the diagnosis of sparganosis, especially in endemic areas. Surgical removal of the worm is the treatment of choice and has good outcomes.

Acknowledgements

We are grateful to Waiwit Sanguanwongwarn, M.D. at Department of Orthopedic Surgery, Huahin Hospital, Prachuap Khiri Khan, Thailand, for support in clinical information of the patient, and Dussadee Sakonlaya, M.D. at Department of Pathology, Thammasat University Hospital, Pathumthani, Thailand, for support in the pathological analyses.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Andersen KI. Description of musculature differences in spargana of Spirometra (Cestoda; Pseudophyllidea) and tetrathyridia of Mesocestoides (Cestoda; Cyclophyllidea) and their value in identification. J Helminthol 1983;57:331-4.
2. Bao XY, Ding XH, Lu YC. Sparganosis presenting as radiculalgia at the conus medullaris. Clin Neurol Neurosurg 2008;110:843-6.
3. Beaver PC. Clinical Parasitology. 9 Sub edition., Lea & Febiger: Philadelphia, 1984.
4. Chang KH, Han MH. MRI of CNS parasitic diseases. J Magn Reson Imaging 1998;8:297-307.
5. Cho SY, Kang SY, Kong Y. Purification of antigenic protein of sparganum by immunoaffinity chromatography using a monoclonal antibody. Korean J Parasitol 1990;28:135.
6. Cobbold T. Description of Ligula mansoni, a new human cestode. Zoo J Linn Soc Lond 1883;17:78.
7. Kim H, Kim SI, Cho SY. Serological Diagnosis Of Human Sparganosis By Means Of Micro-ELISA. Kisaengchunghak Chapchi 1984;22:222-8.
8. Kudesia S, Indira DB, Sarala D, Vani S, Yasha TC, Jayakumar PN, *et al.* Sparganosis of brain and spinal cord: Unusual tapeworm infestation (report of two cases). Clin Neurol Neurosurg 1998;100:148-52.
9. Kwon J, Kim JS. Sparganosis presenting as a conus medullaris lesion: Case report and literature review of the spinal sparganosis. Arch Neurol 2004;61:1126-8.
10. Mueller JF, Hart EP, Walsh WP. Human Sparganosis in the United States. J Parasitol 1963;49:294-6.
11. Park HY, Lee SU, Kim SH, Lee PC, Huh S, Yang YS, *et al.* Epidemiological significance of sero-positive inhabitants against sparganum in Kangwon-do, Korea. Yonsei Med J 2001;42:371-4.
12. Park JH, Park YS, Kim JS, Roh SW. Sparganosis in the Lumbar Spine: Report of Two Cases and Review of the Literature. J Korean Neurosurg Soc 2011;49:241-4.
13. Roh SY, Lee JY, Park KW, Jung S-N. Sparganosis in a patient with diffuse large B cell lymphoma. J Cancer Res Ther 2013;9:712-4.
14. Song T, Wang WS, Zhou BR, Mai WW, Li ZZ, Guo HC, *et al.* CT and MR Characteristics of Cerebral Sparganosis. Am J Neuroradiol 2007;28:1700-5.
15. Sundaram C, Prasad V, Reddy JJ. Cerebral sparganosis. J Assoc Physicians India 2003;51:1107-10.
16. Tang TW, Huang JS, Huang SH, Su KE, Chang YL. Sparganosis of the Spinal Canal: Rare Tapeworm Infection as a Cauda Equina Mass with Magnetic Resonance Imaging. J Radiol Sci 2001;36:139-44.
17. Wiwanitkit V. A review of human sparganosis in Thailand. Int J Infect Dis 2005;9:312-6.