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## Cerebral sparganosis with intracerebral hemorrhage: A case report

**Article abstract**—We describe the clinical case and radiologic findings in a woman with cerebral sparganosis in which intracerebral hemorrhage was the presenting feature with hemiparesis and dysarthria. CT demonstrated high-density lesions in the right frontoparietal area, suggesting a hematoma. With conservative management, hemiparesis improved and follow-up CT revealed what looked like a resolving hematoma. Two weeks later, she complained once again of aggravated left hemiparesis and facial weakness. Diagnosis of sparganosis was made on the basis of brain MRI and ELISA. Stereotactic surgery was performed, and a live larva of sparganum was successfully removed.

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Sparganosis is caused by a migrating tapeworm larva of the genus *Spirometra*. Spargana invade subcutaneous tissue, muscle, eyeball, urogenital, and abdominal viscera, but the CNS is not a frequent site of invasion. Rare but increasing reports have described cerebral sparganosis from various parts of the world, and recent developments in serologic and radiologic studies make diagnosis of CNS infection by spargana much easier. However, cerebral sparganosis associated with intracerebral hemorrhage is very rare; so far, only one case has been reported in the literature<sup>1</sup> and another case with intraventricular hemorrhage rather than parenchymal hematoma has been reported.<sup>2</sup> Here we report another case of cerebral sparganosis with hemiparesis as the main manifestation of the disease. Apparently, this manifestation is attributable to the intracerebral hemorrhage caused by a live larva of sparganum of an unusual size.

**Case report.** A 61-year-old woman first visited the outpatient clinic of the neurology department of Hallym University Hospital because of left hemiparesis. Her history included eating half-cooked frog and intermittently drinking water from the ponds. She had no remarkable medical history up until 6 months ago when she suddenly developed left hemiparesis. CT of the brain was taken at the local hospital; no specific abnormal lesion was found. She was then treated for lacunar infarction with traditional medicine such as acupuncture and herbs, but her left hemiparesis improved only slightly over the following months. Neurologic examination revealed mild left hemi-

paresis and increased deep tendon reflexes (DTR). Diagnosis of an old lacune was presumed. Two months later, she came to the emergency room with more progressed left hemiparesis and newly developed left hemihypesthesia and dysarthria. Neurologic examination again revealed an intelligent woman of normal mentation with dysarthria. She had mild motor weakness in the left extremities, grade 4/5 in upper and lower extremity with increased DTR, and Babinski sign was noted on the left side. Primary sensory modalities of pain, temperature, touch, and vibration were decreased over the left extremities. Physical examination revealed no signs of abnormal palpable mass or lump over the entire body. Routine laboratory findings such as CBC (including eosinophil count), ESR, urinalysis, stool examination for parasites, ECG, and chest radiograph were normal. CT of the brain revealed a 2 × 1-cm high-density area in the right frontoparietal area with diffuse low-density area around it (figure 1A). Intracerebral hemorrhage was diagnosed and she was treated conservatively. One week later, her sensation became symmetrical, but the left hemiparesis and dysarthria were still persistent with only mild improvement. On the follow-up CT taken a week later, the high-density lesion in the right frontoparietal lobe shrank to 1 × 0.5 cm, but the surrounding low-density area was noticeably enlarged (figure 1B). We recommended MRI of the brain and cerebral angiography to exclude the possibility of an arteriovenous malformation or brain tumor. She refused to take the examinations and was discharged.

Two weeks later, she returned with more aggravated left hemiparesis. She had left facial palsy of central type and motor weakness with grade 0/5 in the left upper extremity

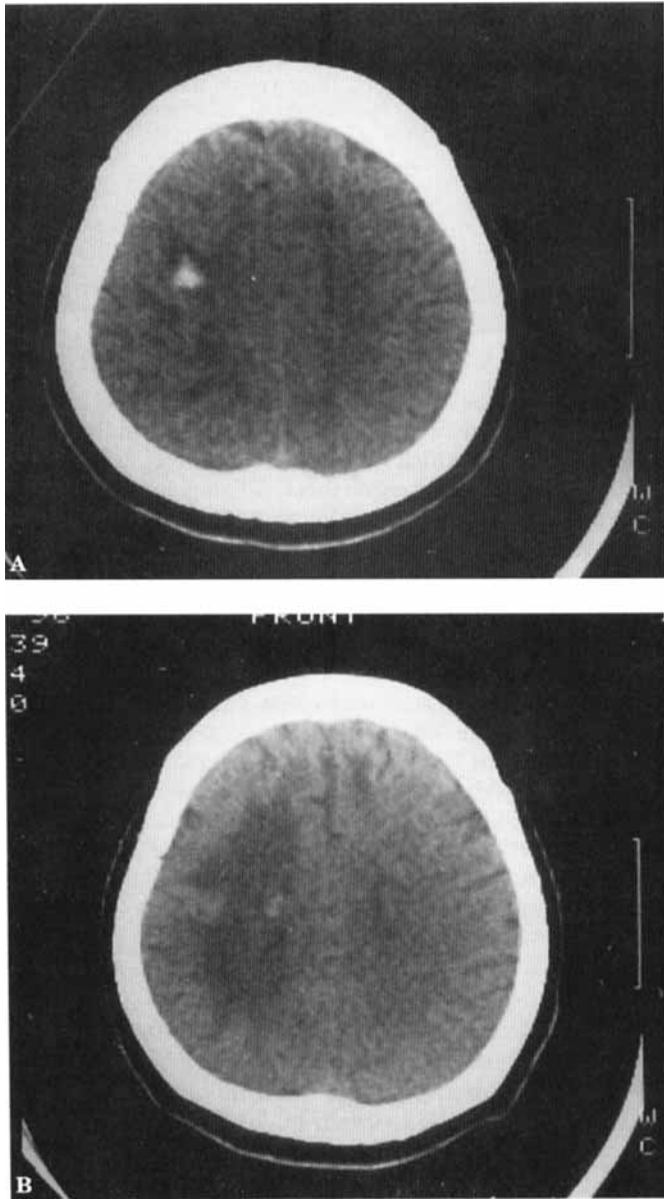


Figure 1. (A) Brain CT shows a high-density lesion in the right frontoparietal area with diffuse a low-density area around it that appears to be a small hematoma with surrounding edema. (B) Follow-up CT taken 1 week later shows a regressed high-density lesion, but surrounding diffuse low-density area is enlarged.

and grade 3/5 in the left lower extremity. MRI of the brain demonstrated multiple cystic low-signal lesions surrounded by the area of decreased signal change on T1-weighted images with rim enhancement (figure 2, A and B) and displayed as multiple high-signal lesions surrounded by an area of diffuse increased signal change on T2-weighted images (figure 2C). CSF examination revealed clear fluid with no cells, protein was 34 mg/mL, and glucose was 61 mg/mL (serum glucose, 60 mg/mL). Micro-ELISA test for sparganum was positive. At first it was difficult to diagnose the nature of this particular lesion, but the micro-ELISA test proved to be a useful tool for the evaluation of parasitic infestation. We performed the micro-ELISA test for sparganum twice, before and after the operation. The absorbance values of serum and CSF ELISA test for sparganum were 1.10 and 1.05 (positive

values > 0.22) before the operation. For cysticercus, they were 0.04 and 0.01 (positive values > 0.18). For paragonimus, they were 0.03 and 0.01, respectively (positive values > 0.25). Micro-ELISA test was an important clue in diagnosing this case. The absorbance value of the serum ELISA test for sparganum was decreased to 0.76 after the operation.

Three weeks later, she was transferred to the neurosurgical department, and stereotactic surgery was performed for cerebral sparganosis. During the surgery, the larva was cut in half, each measuring 20 and 30 cm. A living larva of 50 cm in total length with 2 to 3 mm in thickness was removed successfully from the lesion. Figure 3 shows part of the larva. She is now able to walk with assistance.

**Discussion.** Sparganosis is an infection caused by larvae plerocercoid of the genus *Spirometra*, a cestode tapeworm. In Asia, *Spirometra erinacei* is the indicated species.<sup>3</sup> The adult tapeworm can infect dogs, cats, and other carnivores, residing in their gastrointestinal tract. Eggs are passed in the feces of these primary hosts and then ingested first by intermediate hosts, called cyclops. Cyclops, in which eggs mature into proceroid larvae, are ingested by frogs, snakes, birds, and fishes. Proceroid larvae mature into plerocercoid in their second intermediate hosts, and the flesh of these animal is eaten by primary hosts, thus continuing life cycles. Human infestation is uncommon and seems to occur accidentally by ingestion of polluted water containing cyclops, by ingestion of raw or inadequately cooked flesh of snakes or frogs, and by applying the flesh of an infected intermediate host as a poultice to a wound. The exact transition pathway after infection has not been clearly understood, but the larvae is known to migrate from primary infection focus to a distant site, such as skin, skeletal muscle, orbital tissue, urogenital and abdominal viscera, and rarely to the CNS. In this patient, the route of infestation was thought to be through drinking water from ponds or eating poorly cooked flesh of frogs many years ago. No other signs or symptoms of infection of organs by sparganum could be detected other than the brain.

Since human infection was first discovered by Patrick Manson in 1882, more than 300 cases of human sparganosis have been reported worldwide, but it is more common in East Asia (China,<sup>2,4</sup> Japan, and Korea), and it is also reported in Southeast Asia (Malaysia, India, and the Philippines). Cases have been reported from the United States, especially in the Southeastern states.<sup>5</sup> Ordinarily, sparganosis involves subcutaneous soft tissue and superficial muscle fascia of the human body, and sparganosis of the brain is rare. Cerebral sparganosis was first reported by Takeuchi in 1918, and since then over 20 cases have been reported in the English literatures up to 1989. However, cerebral sparganosis has increasingly been detected in recent years (over 50 cases between 1990 and 1995), presumably due to advances in brain imaging and introduction of ELISA test. The cases of cerebral sparganosis too have been

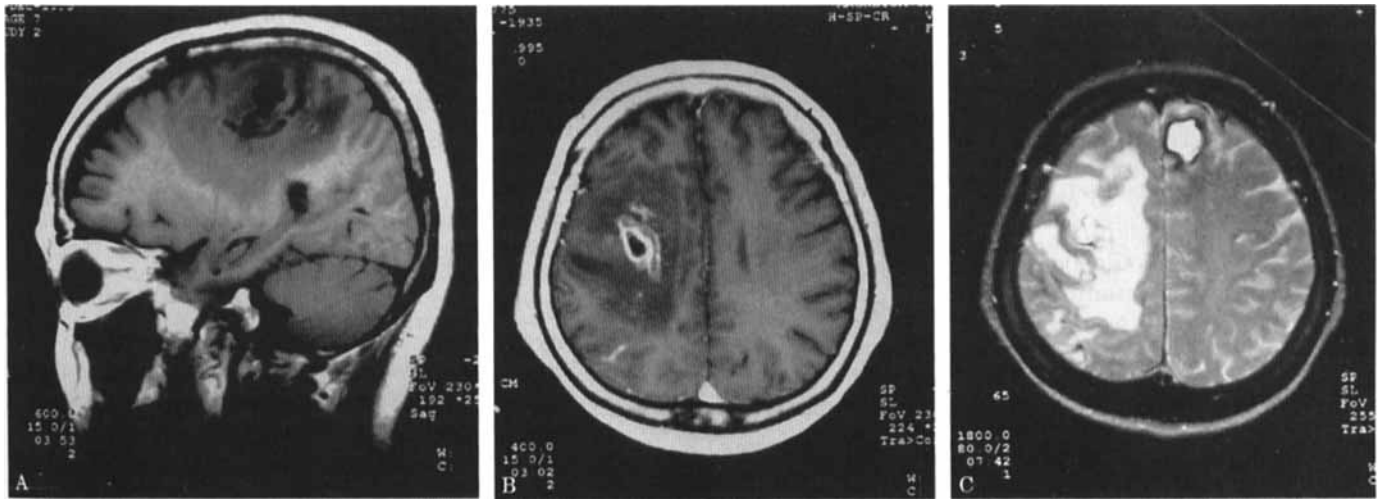


Figure 2. MRI of patient at presentation. (A) Sagittal T1-weighted image shows multiple cystic low-signal lesions with surrounding diffuse decreased signal change in the right frontoparietal area. (B) Enhanced axial T1-weighted image demonstrated rim enhancement of these low-signal lesions. (C) Axial T2-weighted image displays high-signal lesions with surrounding increased signal change in the same area.

more common in Asia than elsewhere,<sup>6,7</sup> perhaps because of the use of unsanitary drinking water and eating raw uncooked meat.<sup>4</sup>

The clinical manifestations of cerebral sparganosis are seizure, headache, and focal neurologic deficits such as hemiparesis, aphasia, dysarthria, and dizziness. Among these, seizure, headache, and hemiparesis are the most frequent symptoms and signs.<sup>6,7</sup> Our patient complained of recurrent weakness of the left extremities with waxing and waning course, sensory change, dysarthria, and left facial palsy without any seizures.

CTs of cerebral sparganosis usually demonstrate the presence of white matter hypoattenuation with dilatation of adjacent ventricle, irregular or nodular enhancing lesion, small punctate calcification, and a change in the location of the enhancing nodule on the follow-up CTs.<sup>6,7</sup> In this particular patient, the first CT could not be obtained. The CT performed a month before the first admission revealed a high-density area in the right frontoparietal region; the lesion regressed on repeat CT taken a week later. Brain MRI demonstrated multiple cystic areas of hypointensity on T1-weighted images with rim enhancement and areas of hyperintensity on T2-weighted images. Recently, Moon et al.<sup>7</sup> reported that areas of hyperintensity on T1-weighted images and areas of hypointensity areas on T2-weighted images were detected only in MRI. They believed that these changes in signal intensity represented petechial hemorrhage and may be due to capillary or venous injury by the migrating worm. Actually, some cases showed cortical petechial hemorrhage at the operation site.<sup>7</sup> We could not certify the cause and effect relationship between the intracerebral hemorrhage and the sparganum by the pathologic findings, but we agree that the idea of probable vascular injury was caused by the migrating larva.

Differential diagnosis should include other infec-

tious granulomas, neoplasms, chronic infarctions, and degenerative diseases. If an enhancing lesion shows an irregular, amorphous, beaded, or conglomerate nodular appearance together with extensive degeneration of the white matter with adjacent ventricular dilatation, differentiation from other granulomas or metastasis is not difficult. If the lesion has a mass effect, it may be misdiagnosed as a neoplasm or as other granulomas.<sup>6,7</sup>

ELISA test is a very sensitive test for the detection of sparganosis when the diagnosis is in doubt. Before the introduction of the ELISA test, human sparganosis had to be surgically diagnosed because its diagnosis depended almost entirely on the recovery of larvae from the lesion. In surgically proven cerebral sparganosis, both serum and CSF ELISA tests showed high sensitivity and concordance rate

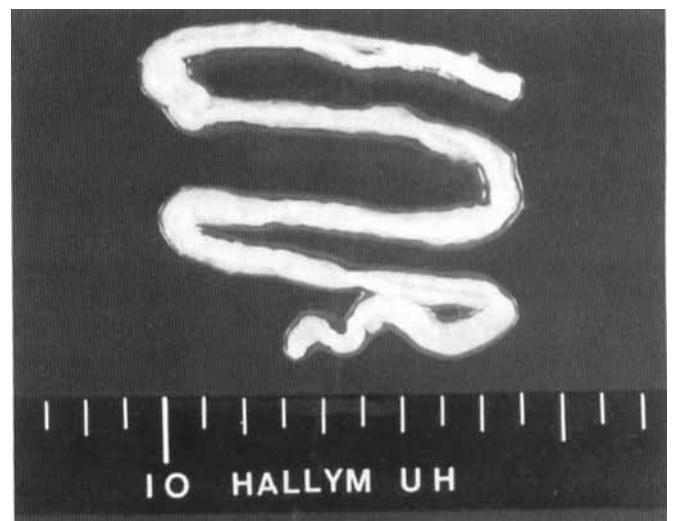


Figure 3. Part of live larva removed during the stereotactic operation with comparable scale under it. It was measured to be about 20 cm long.

in diagnosing cerebral sparganosis, and negative conversion was noted in patients with successful surgical removal.<sup>8</sup> However, it is difficult to say whether the decrement in the serum absorbance value was due to the treatment or to a natural course of human immune response to the spargnum. Further studies might be necessary to evaluate human immune response to the spargnum. The serologic diagnosis by means of ELISA could be a useful tool in the epidemiologic study of human sparganosis in a susceptible population, as well as in individual diagnosis. As for the management of cerebral sparganosis, surgical removal seems to be the most effective way of treatment.<sup>9</sup> It is a commonly held opinion that there is no known antihelminthic effective in sparganosis. Niclosamides are effective antihelminthic agents while the tapeworm is still within the intestine of the primary host. Once the tapeworm extends beyond the intestine, there is no evidence to support the therapeutic value of Praziquantel (Bayer Korea Pharm. Co., Korea) and Albendazole (Handok Pharm. Co., Korea), which have been effective agents for neurocysticercosis.<sup>10</sup>

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