Cerebral Sparganosis *Mansoni*

—Case Report—

Tatsunori OKAMURA, Mitsuo YAMAMOTO, Keiji OHTA,
Takashi MATSUOKA, and Tohru UOZUMI*

Department of Neurosurgery, Matsue Red Cross Hospital, Matsue;

*Department of Neurosurgery, Hiroshima University School
of Medicine, Hiroshima

Abstract

An 80-year-old male with paresis of the right upper extremity underwent surgery for a suspected metastatic brain tumor. A live *Sparganum mansoni* worm was removed from the parietal lobe through a left parietal craniotomy. Retrospective evaluation of magnetic resonance (MR) images revealed ipsilateral ventricular dilatation, despite the presence of a mass lesion resembling a metastatic brain tumor. This interesting MR imaging finding is an important point for differentiating between a brain tumor and cerebral sparganosis.

Key words: cerebral sparganosis, computed tomography, magnetic resonance imaging, parasite, *Sparganum mansoni*

Introduction

Human sparganosis caused by *Sparganum mansoni*, the plerocercoid larva of *Diphyllbothrium erinacei*, occurs worldwide but mainly in Asia, most commonly in Japan, Korea, and China.¹⁰⁾ Human cerebral sparganosis is an extremely rare parasitic zoonosis of the central nervous system. A live worm was removed from the brain in five of eight previously reported cases in Japan.^{2,7-9,11-13)} The symptomatological and neuroradiological findings are very similar to those of brain tumors, and the two conditions are difficult to discriminate preoperatively. Only one previous report described the magnetic resonance (MR) imaging features of cerebral sparganosis.²⁾

We report a recent case of cerebral sparganosis and describe the MR imaging and other interesting findings.

Case Report

An 80-year-old Japanese male had formerly worked

as a railway station attendant, and had never traveled overseas. He had sometimes drunk unboiled water and eaten wild herbs gathered at the water's edge in his childhood. However, he had no habit of eating raw meat, freshwater fish, frogs, or snakes. About 40 years previously, he had twice undergone removal of tumors from the umbilical region of the abdominal wall, but no details were available.

His present illness dated back to April 8, 1994, when he became aware of numbness and some associated difficulty in use of the right upper extremity. Computed tomography (CT) at a local hospital showed a hypodense lesion in the left parietal lobe. He was referred to the Department of Neurosurgery at Matsue Red Cross Hospital for further treatment of a possible brain tumor on April 11, 1994.

On admission, he had loss of appetite but without headache or nausea. He was a bright, intelligent man with normal mentation. Neurologically he was lucid with no abnormality except for slight right hemiparesis which was more severe in the upper extremity.

Precontrast CT revealed an extensive low-density

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Author's present address: T. Okamura, M.D., Department of Neurosurgery, Kitakyushu General Hospital, Kitakyushu, Fukuoka, Japan.

Fig. 1 Preoperative CT scans showing an extensive low-density area in the subcortical white matter of the left parietal lobe (*left*), and postcontrast enhancement of a nodule in this area (*right*).

area in the left parietal lobe without apparent distortion of the ventricles. Postcontrast CT revealed a homogeneously enhanced nodule in the low-density area (Fig. 1). T₁-weighted MR imaging showed a low-intensity lesion in the left parietal lobe that appeared as a high-intensity area on the T₂-weighted image. The nodule appeared as an isointense area on the T₁-weighted image, a hypointense area on the T₂-weighted image, and was enhanced by gadolinium-diethylenetriaminepenta-acetic acid (Gd-DTPA) (Fig. 2). MR imaging also revealed dilatation of the left lateral ventricle, which was obscure on CT scans (Fig. 3). Left carotid angiography showed normal findings. Results of routine hematological and biochemical investigations were normal except for a slight increase in the serum carcinoembryonic antigen and ferritin levels.

A left parietal craniotomy was performed under a diagnosis of metastatic brain tumor on April 17, 1994. The color tone and toughness of the brain tissue at the site of the lesion were indistinguishable from those of the normal brain. The exact site of the tumor was too difficult to localize, resulting in only an exploratory craniotomy.

On April 22, 1994, a second left parietal craniotomy was performed after the coordinates of the target, the nodular enhanced lesion, had been fixed by a CT-guided stereotactic system. The lesion coordinates corresponded to the region that had been examined at the first operation. As before, no abnormalities were found in the brain tissue. Manipulation of the aspirator in the subcortical layer at the bottom of the postcentral sulcus attached a milky-white, string-like body to the tip of

the aspirator (Fig. 4). Close observation disclosed slow peristaltic movement, suggesting that the body was a kind of worm. The worm was drawn out and removed carefully. Subsequently, the extracted worm, which was about 14 cm in total length and 1 mm in diameter, was identified as *Sparganum mansoni* (Fig. 5).

T₁-weighted MR imaging 20 days after the second operation revealed resolution of the low-intensity area and disappearance of the nodular enhanced lesion, and an elongated sinuous area of low intensity in the subcortex of the left parietal lobe, which was presumed to be the cavity resulting from extraction of the worm (Fig. 6). Postoperative hematological examination revealed normal findings with no increase in eosinophils or immunoglobulin E (IgE). Immunoserological examinations for parasitic infection using the Ouchterlony test and immunoelectrophoresis detected no evidence of a parasite. He made an uneventful recovery and was discharged with no neurological deficits on May 29, 1994.

Discussion

The final hosts of *Diphyllbothrium erinacei* are carnivores such as dogs and cats. The egg of the worm, excreted in the feces of the final host, hatches in fresh water and produces a coracidium. The coracidium is consumed by *Cyclops*, the first intermediate host, and then develops into a proceroid larva within the *Cyclops*. The second intermediate host, such as a frog, a snake, or a freshwater fish, ingests fresh water containing the *Cyclops*. The proceroid then develops into a plerocercoid larva, or sparganum, in the subcutaneous tissues or muscle of the new host. After the host containing the sparganum is consumed by the final host, the adult tapeworm is formed in the intestinal tract of the final host.¹⁰⁾

Humans may acquire this infection in three ways: 1) by drinking fresh water containing infected *Cyclops*, 2) by ingesting raw or inadequately cooked flesh of the infected second intermediate host, or 3) by applying the flesh of the infected host as a poultice to an open wound. A proceroid or plerocercoid larva in an infected human merely migrates to mainly soft subcutaneous tissues or muscle without growing into the adult tapeworm, since the human is only an accidental intermediate host. The present patient had no past history of ingesting the flesh of the intermediate hosts, but had twice undergone removal of a subcutaneous tumor in the abdominal wall about 40 years previously. Possibly he had already been infected with the sparganum at that time.

Fig. 2 Axial MR images revealing an extensive area of low intensity on the T_1 -weighted image (A: repetition time [TR] 400 msec, echo time [TE] 15 msec) and high intensity on the T_2 -weighted image (C: TR 3000 msec, TE 100 msec) in the left parietal lobe. The nodule appears as an isointense area on the T_1 -weighted image, as a hypointense area on the T_2 -weighted image, and is enhanced after Gd-DTPA administration (B). Coronal MR images revealing an area of low intensity in the sub-cortex of the left parietal lobe on a T_1 -weighted image (D) and nodular enhancement after Gd-DTPA administration (E).

Fig. 3 Axial (left) and coronal (right) T_1 -weighted MR images revealing the low-intensity lesion in the left parietal lobe associated with apparent adjacent ventricular dilatation.

Fig. 4 Intraoperative photograph showing the *Sparganum mansoni* worm (arrow).

Fig. 5 *Sparganum mansonii* worm extracted from the patient, measuring about 14 cm in length and 1 mm in diameter.

Fig. 6 Axial (left) and coronal (right) T₁-weighted MR images with Gd-DTPA 20 days after the second operation, revealing resolution of the low-intensity area with disappearance of the nodular enhanced lesion, and an elongated sinuous area of low intensity in the subcortex of the left parietal lobe, presumably the cavitory tract resulting from extraction of the worm (arrow).

Although the route of infection to the central nervous system is not yet known, the sparganum may migrate through the foramina of the skull base and vertebral column along the loose connective tissue around the vessels or nerves.^{3,11,13} A patient harboring a sparganum in the brain may suffer from various cerebral focal symptoms or convulsions due to formation of an inflammatory granuloma around the worm. The clinical symptoms are very similar to those of a brain tumor. Neuroradiological examinations such as CT or angiography cannot always distinguish between cerebral sparganosis and brain tumor.^{2,13} Almost all previous cases were initially

Table 1 CT findings in 43 cases of cerebral sparganosis and CT-pathological correlation

CT findings [Corresponding pathological features]	Case No./Total (%)
extensive low density in white matter [perifocal edema or fibrillary gliosis with loss of myelin]	42/43 (98)
ipsilateral ventricular dilatation [rapid regression of edema with chronicity of the disease]	32/39 (82)
irregular nodular enhanced lesion [dense collagenous wall surrounding worm (granulation tissue zone)]	37/43 (86)
small punctate calcifications [degenerated worm]	28/43 (65)
movement of enhanced nodule on serial CT scans [migration of the worm]	6/24 (25)

misdiagnosed as a metastatic brain tumor or glioma.

The CT findings of cerebral sparganosis were described in 42 previous cases.^{1-3,5,7,9,11,13} More than 80% of the 43 cases, including the present one, demonstrated a low-density area of variable extent in the cerebral white matter, associated with ipsilateral ventricular dilatation and irregular nodular enhancement within the low-density area (Table 1). Additional characteristic CT findings were small punctate calcifications and changes in the location of the enhanced nodule on serial CT scans.²⁻⁴ Chang *et al.*³ considered that these peculiar CT findings were due to the combined effects of long survival time, the active movement of the worm in the brain tissue, and the histotoxic effects of proteases secreted by the worm.⁶ Only one report described the MR imaging features of cerebral sparganosis.² In our case, MR imaging showed an extensive lesion resembling brain edema, which appeared as a low-intensity area on the T₁-weighted image and a high-intensity area on the T₂-weighted image. Surprisingly, closer observation revealed ipsilateral ventricular dilatation despite the mass lesion, which was not evident on the CT scans (Fig. 3). CT scans of metastatic brain tumors or gliomas often reveal a severe mass effect with perifocal edema, and such ipsilateral ventricular dilatation is an unlikely finding. We suggest that this feature might be an important point for differentiation between cerebral sparganosis and brain tumor. Rapid regression of edema associated with the chronicity of the disease and fibrillary gliosis with a loss of myelin may have caused the ipsilateral ventricular dilatation.³

Increased levels of eosinophils or IgE in the serum, or immunoserological examinations such as the

Ouchterlony test, are the basis for the diagnosis of human sparganosis, but these methods are seldom useful.^{2,7,13)} Recently, positivity for the sparganum-specific IgG antibody in serum or cerebrospinal fluid (CSF) using an enzyme-linked immunosorbent assay (ELISA) has been very useful for correct identification of cerebral sparganosis.²⁻⁴⁾

Cerebral sparganosis has often been misdiagnosed as brain tumor in cases where immunoserological examinations were not performed. The differential diagnosis of brain tumor should include the possibility of parasitic zoonosis. If parasitic infection is suspected from a history of ingesting raw flesh or drinking natural unboiled water, or from unusual CT and MR imaging findings, ELISA of both serum and CSF should be performed for a definitive diagnosis or a diagnosis by exclusion.

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Address reprint requests to: T. Okamura, M.D., Department of Neurosurgery, Kitakyushu General Hospital, 5-10-10 Yukawa, Kokuraminami-ku, Kitakyushu, Fukuoka 800-02, Japan.