CHRONIC CEREBRAL PARAGONIMIASIS COMBINED WITH ANEURYSMAL SUBARACHNOID HEMORRHAGE

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Abstract. A 67-year-old Korean woman attended our hospital complaining of a severe headache. A brain computed tomography scan showed conglomerated, high-density, calcified nodules in the left temporo-occipito-parietal area and high-density subarachnoid hemorrhage in the basal cisterns. Magnetic resonance imaging of the brain shows multiple conglomerated iso- or low-signal intensity round nodules with peripheral rim enhancement. She underwent craniotomies to clip the aneurysm and remove the calcified masses. *Paragonimus westermani* eggs were identified in the calcified necrotic lesions. Results of parasitic examinations on the sputum and an enzyme-linked immunosorbent assay for *P. westermani* were all negative. The patient presented with headache and dizziness that had occurred for more than 30 years. She had not eaten freshwater crayfish or crabs. However, she had sometimes prepared raw crabs for several decades. Overall, this case was diagnosed as chronic cerebral paragonimiasis, in which she may have been infected through the contamination of utensils during the preparation of the second intermediate hosts, combined with a cerebral hemorrhage.

INTRODUCTION

Paragonimiasis is an infection by the genus *Paragonimus*, the most common of which is *Paragonimus westermani*, the oriental lung fluke. *Paragonimus westermani* is a common human parasite in the Far East, and is particularly prevalent in Korea, China, Japan, and Taiwan. Paragonimiasis results from the ingestion of a raw or insufficiently cooked second intermediate hosts such as freshwater crayfish, crabs, or shrimp, where the encysted metacercarial stage occurs. The metacercariae excyst in the small intestine and penetrate the wall into the abdominal cavity, which then migrate through the viscera and diaphragm to the lungs. The lung is the principal habitat in the human host. The erratic migrations of the juvenile flukes result in ectopic paragonimiasis in various organs, frequently in the peritoneal and pelvic cavities, the diaphragm, the subcutaneous tissues, and the brain.1,2

The most serious erratic migration is a cerebral paragonimiasis, where the fluke enters the cranial cavity through the jugular or carotid foramen and usually invades the temporal and occipital lobes of the brain. Cerebral paragonimiasis is serious and is sometimes fatal, although symptoms of ectopic infections are solely dependent on the infected sites and the number of parasites therein.3−4 There are many factors known to cause intracranial hemorrhage including head trauma, hypertensive hemorrhage, coagulopathy, aneurysm, and arteriovenous malformation. Parasitic infections can also cause an intracranial hemorrhage, which have been reported for *Plasmodium*, *Toxoplasma*, sparganum, cysticercosis, and *Paragonimus*.5−10 We report a combined case of a chronic cerebral paragonimiasis and an intracranial hemorrhage.

CASE REPORT

A 67-year-old Korean woman was admitted to the Sun General Hospital in Daejeon, Republic of Korea on March 4, 2002 complaining of severe headache and drowsiness for the previous hour. A plain skull lateral view revealed radiopaque, conglomerated, calcified nodules in the temporo-occipito-parietal area (Figure 1A). Computed tomographic (CT) scans of the brain also showed conglomerated high-density calcified nodules with underlying focal encephalomalacia in the left temporo-occipito-parietal area and a subarachnoid hemorrhage in the basal cisterns (Figure 1B). A right lateral internal carotid cerebral angiogram showed a saccular aneurysm in the posterior communicating artery (Figure 1C). Magnetic resonance images (MRI) of the brain show conglomerated iso- or low-signal intensity round nodules with a peripheral rim enhancement in the left temporo-occipito-parietal area in both the T1- and T2-weighted axial images (Figure 1D–F). A chest radiograph showed mild right pleural thickening, but there was no active lesion. From these radiologic findings, the patient was diagnosed with multiple calcifications in the left temporo-occipito-parietal area due to a parasitic infection and a diffuse subarachnoid hemorrhage. She underwent an emergency craniotomy to decompress the intracranial pressure and to clip the neck of the aneurysm on March 5, 2002.

The headache and dizziness had occurred frequently for 30 years. She also had a history of hepatitis and pulmonary tuberculosis approximately 30 years ago. She had not eaten freshwater crayfish or crabs. However, she had sometimes prepared raw crabs to make the soybean sauce–soaked freshwater crabs or to cook for several decades. Prior to the emergency surgery, the laboratory results on the blood examination were as follows: red blood cell count = 3.6 × 10¹²/mm³, white blood cell count = 4.7 × 10⁹/mm³ (neutrophils = 46.9%, lymphocytes = 40.4%, monocytes = 6.1%, eosinophils = 4.6%, basophils = 1.9%), hemoglobin = 11.2 g/dL, platelet count = 3.0 × 10⁵/mm³. Results of an enzyme-linked immunosorbent assay (ELISA) for serum and cerebrospinal fluid and an IgG antibody test for *P. westermani*, *Clonorchis sinensis*, cysticercosis, and sparganum were all negative. No eggs, cysts, or larvae of the parasites were found in the stool and sputum.

The symptoms of headache and dizziness improved after the emergency surgery. The calcified masses in the left temporo-occipito-parietal area were resected on May 20, 2002 through the craniotomies. The resected masses were yellowish and looked like bunches of grapes (Figure 2A). Microscopically, the worm capsules were calcified and surrounded with granulation tissues. In addition, there were numerous eggs of *P. westermani* and Charcot-Leyden crystals within the
Cerebral paragonimiasis is diagnosed by radiologic findings, as well as by immunologic and parasitologic methods. The plain skull radiographs showed signs of an increased intracranial pressure or cerebral calcification. The CT and MRI findings for chronic cerebral paragonimiasis have been generally recorded as conglomerates of multiple ring-shaped shadows or enhancements so-called “grape cluster” or “soap bubble” forms in one hemisphere.\(^\text{3,4,14,15}\) The present case also showed typical radiologic findings of chronic paragonimiasis such as multiple conglomerated round calcified nodules in the left temporo-occipito-parietal area in the brain. An ELISA is recommended as a complementary tool for diagnosing cerebral paragonimiasis, and the serum and CSF antibody levels are generally positive in 48% and 31% in chronic cases, respectively.\(^\text{3}\) In the present case, the ELISA results for the diagnosis of paragonimiasis were negative, but there were many P. westermani eggs in the worm capsules resected from patient’s brain. A discrepancy between the ELISA and parasitologic results has also been reported.\(^\text{16}\)

These results suggest that the antibody titer against P. westermani, which blocks the antigenic stimulation of the worm capsules in the brain, returns to normal levels rather quickly because the antibody titers against Paragonimus species show seroconversion after approximately six months in most treated patients.\(^\text{1}\) It is interesting that numerous P. westermani eggs in the multiple nodules had been well preserved for a long time, but were not present the adult worm in the worm capsules. In addition, most of the eggs in the worm capsules retained a shell without a yolk. This indicates that the worm capsules in the brain had been established more than 20 years ago because adult worms die within 10–20 years, even without treatment.\(^\text{14}\) In addition, the worm capsules remain as multiple nodules with a low-density cavity containing the Paragonimus eggs in cases of cerebral paragonimiasis that persist for more than 20 years.\(^\text{3,14}\)

Pulmonary paragonimiasis is rarely fatal. However, most cases of cerebral disease are associated with chronic mortality due to epilepsy, dementia, and various neurologic sequelae. Moreover, 5% of the patients die as a result of hemorrhage.\(^\text{1}\) In the present case, the patient had headache and dizziness for approximately 30 years, which may be the clinical manifestations of the cerebral paragonimiasis. In addition, she had hepatitis and pulmonary tuberculosis 30 years ago. It is well-known that the clinical manifestations of pulmonary tuberculosis are similar to those of pulmonary paragonimiasis,\(^\text{1,2}\) and it was recently reported that active hepatic capsulitis can be caused by a P. westermani infection.\(^\text{17}\) It is possible that the symptoms of hepatitis and pulmonary tuberculosis 30 years ago were caused by the Paragonimus infection. The relation-
ship between a cerebral paragonimiasis and an intracranial hemorrhage has not been well described until now. In this case, a rupture of the aneurysm could have caused the diffuse subarachnoid hemorrhage. It is unclear if the Paragonimus infection in the brain contributed to the causes of the aneurysm or intracranial hemorrhage. However, there were reported two cases where a cerebral hemorrhage can be produced by a Paragonimus infection.\textsuperscript{9,10}

Based on the clinical, radiologic, and laboratory findings, the cerebral lesions of P. westermani in this patient may have been established at least 20 years ago because she had episodes of occasional headache and dizziness for 30 years, and pathologic findings of resected worm capsules have been observed in cases of chronic cerebral paragonimiasis more than 20 years after the infection. She may have been infected through the contamination of utensils during the preparation.
of the second intermediate hosts for several decades. This is the third case of a cerebral paragonimiasis combined with a cerebral hemorrhage. This case emphasizes that chronic cerebral paragonimiasis must be considered as a rare cause of a cerebral hemorrhage. Therefore, further study is needed to determine the relationship between the cerebral involvement of the Paragonimus species and a cerebral hemorrhage.

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