Cerebral Infarction and Cranial Venous Sinus Thrombosis Caused by Paragonimiasis

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Paragonimiasis is a zoonosis that is caused by *Paragonimus westermani*. Humans infected with the *Paragonimus* species have been reported worldwide and about 293.8 million people are at risk [1]. Patients infected with *Paragonimus westermani* may experience fever, cough, abdominal pain, or seizures. Most cases present with symptoms related to the lungs. This study reports a rare case of an individual infected with cerebral paragonimiasis and presented as cerebral infarction and cranial venous sinus thrombosis.

A 30-year-old man was admitted to our hospital because of sudden slow response and slurred speech for 3 days in July 2007. He also felt episodic numbness at right limbs. Neurologic examination showed alert, oriented, Wernicke aphasia. Right nasolabial fold was narrow and mouth deviated to left. Muscle strength of right upper limb was II/V, and other limbs were V/V. Bilateral Babinski sign was negative. Most laboratory tests were normal. Cranial MRI showed cerebral infarction of left temporal lobe and basal ganglia (Figure 1A–C). MRA showed occlusion of left cerebral middle artery (Figure 1D). Cerebrospinal fluid (CSF) analysis showed normal pressure, white cell counts, and total protein level. RPR and TPPA tests were negative. Patient was diagnosed with cerebral infarction and received aspirin 100 mg, qd. Several months later, the strength of right limbs returned to near normal.

In October 2010, this patient was admitted to our hospital again because of recurrent flexion of upper limbs, reflexion of lower limbs, and loss of consciousness for 3 days. After treatment with diazepam, spasms were controlled gradually except the frequency of spasm in the right upper limb became higher. Neurologic examination showed lethargy, increased muscle tone, and a positive Babinski sign on the right limbs. Most lab-

oratory tests were normal except D-dimer, which was elevated (1071 ug/L, rang: <200 ug/L). Cranial CEMRI suggested right frontal lobe lesion and gyrus enhancement, and left temporal lobe and basal ganglia cerebral infarction (Figure 2A–E). Cranial CTA showed significant stenosis of the left middle cerebral artery with a blockage after the M2 segment and significant stenosis of right middle cerebral artery (Figure 2F). Administration of anti-epilepsy drugs (Levetiracetam tablets 0.5, po, qd; Sodium valproate tablets 0.2, po, tid; Phenobarbital injection, im, q8 h) showed limited effect. Limb spasms became more frequent, and the patient went into a lasting coma. The round nodule in the frontal lobe hinted at the possibility of parasitic infection. The CSF analysis showed mild increase in protein (58.2 mg/dL) and positive paragonimiasis antibody reaction. Considering the clinical findings and the patient's history of eating raw freshwater crabs, cerebral paragonimiasis was diagnosed and 2 g of praziquantel was administered. Low molecular weight heparins calcium injection, warevan tablet, and carbamazepine were also administered because cranial CEMRV identified sinus thrombosis in the superior sagittal sinus, sinuses sigmoideus, and left transverse sinus (Figure 2G). The patient recovered quickly and could walk by himself 2 weeks later. The follow-up cranial MRV showed a decrease in thrombosis (Figure 2H), and cranial CEMRI showed reduction in the size of the right frontal lesion and no enhancement around the focus (Figure 2I). The 2-year follow-up showed no progression in neurological deficits.

A previous study investigated parasitic status of paragonimus metacercariae in freshwater crabs and crayfish, and 32.3% of the crayfish tested positive for paragonimus metacercariae [2].

Figure 1 The cranial MRI (July 11, 2007) showed cerebral infarction of left temporal lobe and basal ganglia (A) T1-weighted MRI; (B) T2-weighted MRI; (C) diffuse-weighted image. MRA (July 19, 2007) showed occlusion of left cerebral middle artery (D).

Human beings can be infected with Paragonimiasis by eating insufficiently cooked freshwater crabs or crayfish. The parasite can attack the lungs, brain, and sometimes other organs [1]. Cerebral paragonimiasis is an important form of extrapulmonary paragonimiasis with higher mortality, but is often misdiagnosed.

A definite diagnosis of cerebral paragonimiasis can be made when paragonimus eggs are found in brain lesions during pathologic examination, but such invasive examination is not suitable for all patients. The paragonimus-specific ELISA has demonstrated a high sensitivity (96%) and specificity (99%) to *Paragonimus westermani* [3]. Multiple, round, or oval-shaped calcifications on the computerized tomography CT scans were considered a defining characteristic of paragonimiasis infection, but cranial MRI is much more valuable for diagnosis [4–9]. MR images of cerebral paragonimiasis usually show conglomerates of multiple ring-shaped shadows or enhancements of so-called “grape cluster” or “soap bubble” forms in one hemisphere [6]. T1-weighted images of cerebral paragonimiasis nodules show a peripheral low density and central hypo- and isointensity, while T2-weighted images show widespread inflammatory changes in the surrounding tissue [4], and ring-like enhancement on the enhanced scanning [8]. In this case, the diagnosis of brain paragonimiasis was based on the following points: (1) the patient had a history of eating insufficiently cooked freshwater crabs; (2) the paragonimus-specific antibody reaction was positive; (3) cranial CEMRI identified obvious

edema and a round nodule in the lesion and the edema disappeared after treatment with praziquantel; (4) cranial CEMRI identified sinus thrombosis; (5) the symptoms disappeared quickly after praziquantel administration; and (6) D-dimer was found to be elevated.

It is reported that pulmonary symptoms typically present 1–27 months after infection. Symptoms of the central nervous system may present months after the onset of pulmonary symptoms. Headaches, seizures, or blurred visions may occur in patients with cerebral paragonimiasis. Lane et al.[7] reported a case of cerebral paragonimiasis where the patient suffered from diffuse headaches and blurred vision with for 3 months, and an enhanced lesion in the right occipital lobe. A Korean woman with headache and dizziness for 30 years was diagnosed with chronic cerebral paragonimiasis [5]. Another Korean man with seizures for 27 years was also diagnosed with cerebral paragonimiasis when eggs of *Paragonimus westermani* were found in the calcified necrotic lesions of the cerebrum [4]. Hemianopsia, convulsion, and gait disturbance were also seen in cerebral paragonimiasis [10]. Chen et al.[8] reported 14 cases with intracranial hemorrhage cerebral paragonimiasis in southwest China and revealed that intracranial hemorrhage cerebral paragonimiasis often represents as a small hemorrhage surrounded by disproportionately larger edema. The present case showed hemiplegia and seizure. Radiographic findings supported cerebral infarction and cranial venous sinus thrombosis.

Figure 2 The cranial MRI (October 5, 2010) showed right frontal lobe lesion and gyrus enhancement: (A) T1-weighted MRI; (B) T2-weighted MRI showed a round nodule (arrow); (C) diffuse-weighted image (DWI) showed not even high signal in the lesion; (D) apparent diffusion coefficient (ADC) showed high signal; and (E) CEMRI showed gyrus enhancement. Cranial CTA (Oct 11, 2010) showed left middle cerebral artery with blocking after the M2 segment, and significant stenosis of right middle cerebral artery (F). Cranial CEMRV (October 18, 2010) suggested sinus thrombosis in the superior sagittal sinus, sinuses sigmoideus, and left transverse sinus (G). Following up cranial CEMRV (Dec 2, 2010) showed that the thrombosis was decreased (H). Following up cranial CEMRI (Dec 3, 2010) showed reduction in the lesions and no enhancement around the focus (I).

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Conflict of Interest

The authors declare no conflict of interest.

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