**Angiographic findings in 2 children with cerebral paragonimiasis with hemorrhage**

**Case Reports**

**Case 1**

This 9-year-old previously healthy boy presented with sudden onset of headache, nausea, and vomiting during sleep. A CT scan (Fig. 1A) obtained the next day showed an acute intracerebral hemorrhage in the left temporal lobe, and the child was transferred to our hospital on the 3rd day. On admission, physical examination revealed no exact neurological deficit. Slight enhancement around the hypointense lesion was seen on postcontrast MRI (Fig. 1B), but a CT angiogram showed an abnormal vessel with irregular dilation in the area of the hemorrhage (Fig. 1C). Cerebral angiography revealed a beaded appearance of a branch of the left posterior cerebral artery, consistent with arteritis (Fig. 1D and E). The total white blood cell count was normal, while the absolute eosinophil count was $1.33 \times 10^9$ cells/L. A CT scan of the
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Chest showed cystic lesions (Fig. 1F). The child’s dietary history was taken again and disclosed that he had usually drunk untreated water from rivers. Enzyme-linked immunosorbent assay (ELISA) tests for multiple parasites were performed on his serum and were positive for *Paragonimus skrjabini*–specific IgG antibody and negative for others. The diagnosis of cerebral paragonimiasis was established, and the patient was treated with praziquantel at a dose of 75 mg/kg/day for 3 days followed 1 week later by a second course of praziquantel at the same dose. Follow-up brain MRI performed 1 month later revealed newly developed lesions located in the area of previous hemorrhage, and an additional course of praziquantel was administered. At 2 months after admission, a CT angiogram showed that the beaded appearance of the involved vessel was no longer present.

**Case 2**

This 5-year-old girl presented with a 1-month history of episodic cough and fever. She received antibiotic treatment from a nearby clinic, but did not appear to improve. Four days later, she developed sudden onset of headache, nausea, and vomiting. A CT scan performed in a nearby hospital revealed SAH, mainly in the left cerebral sulcus and, to a lesser extent, in the sylvian fissure (Fig. 2A). Cerebral angiography performed the next day (Fig. 2B) revealed a long segmental narrowing of the middle cerebral artery. The child was transferred to our hospital, and physical examination performed on admission revealed neck stiffness but no obvious focal deficits. Her total white blood cell count was $28.6 \times 10^9$ cells/L, with 75.2% eosinophilia. A CT scan of the chest demonstrated a subpleural mass and a pleural effusion in the left lung (Fig. 2C). These findings, together with an ELISA test that was positive for *P. skrjabini*–specific IgG antibody in serum, established the diagnosis of cerebral paragonimiasis. The control brain MRI examination obtained 1 week after SAH showed enhancement around the left cerebral sulcus and adjacent meninges, suggesting meningitis (Fig. 2D). The child was treated with 2 courses of praziquantel, as described above. At the 3-month follow-up, MRI showed that the enhanced lesion had diminished, while an MR angiogram showed normal intracranial vessels (Fig. 2E and F).

**Discussion**

Among *Paragonimus* species, *P. westermani* and *P. skrjabini* are the major pathogens for human paragonimiasis in China. The latter has been identified as the most significant agent of human paragonimiasis in our region (Three Gorges Reservoir region) and is also the main etiological agent of extrapulmonary paragonimiasis in humans.12 In human hosts, only a few *P. skrjabini* parasites reach the lungs and develop into adults; most remain in the juvenile stage, migrate into different organs, and cause extrapulmonary paragonimiasis, including the cerebral form.10,12
In the acute stage of cerebral paragonimiasis, the most common and characteristic CT or MRI findings are conglomerate, multiple, ring-shaped enhancements with surrounding edema of variable degree, resembling “grape clusters.” However, hemorrhage has frequently been found in association with cerebral paragonimiasis in young patients. When children with cerebral paragonimiasis present with intracranial hemorrhage, especially with subclinical pulmonary signs, diagnosis and treatment may be delayed. In the patient in Case 1, the intracerebral hemorrhage was also the initial finding. As a result of previous experience, cerebral paragonimiasis was included in the differential diagnosis of intracerebral hemorrhage and confirmed afterward despite subclinical pulmonary manifestations.

Although a few hemorrhagic events caused by cerebral paragonimiasis have been reported, vascular abnormalities have not been previously demonstrated on angiography. In both of our patients, involved vessels were located in the area of the hemorrhage. The hemorrhage, together with the new lesions seen nearby, improved after praziquantel treatment, and because of this the vascular changes and the hemorrhage were attributed to paragonimiasis infection. The mechanism of hemorrhage associated with cerebral paragonimiasis is unknown. It is possible that the migrating worm causes arteritis and capillary rupture, which might result in hemorrhage and infarction, although evidence of this is lacking. The angiographic findings in our patients included segmental stenosis and a beaded appearance of the arteries, which are characteristic of arteritis.

However, the angiographic appearance may also be mistaken for vasospasm as a result of SAH. In our 2 patients, angiographic vascular abnormalities were observed 3 days (Case 1) and 1 day (Case 2) after onset. In contrast, vasospasm following SAH usually develops 4–7 days after the hemorrhage. In the patient in Case 1, the angiogram revealed a fusiform expansion without obvious segmental narrowing of the vessel, which is not the typical appearance of SAH-induced vasospasm. In Case 2, the long segment of the severe vascular stenosis did not correlate well with the amount and location of subarachnoid blood, which was moderate and mainly located in the cerebral sulcus. Based on these observations, we considered that the vascular changes in our cases were more likely to be due to arteritis rather than vasospasm following SAH.

To the best of our knowledge, this is the first angiographic observation of cerebrovascular involvement associated with cerebral paragonimiasis. Hemorrhagic or ischemic stroke due to other cerebral parasitic diseases has also been reported. In neurocysticercosis, stroke is a well-known complication, with an incidence of 4%–12%, and the most common mechanisms have been found to be related to cerebral arteritis. Because of the limited information available, it is unknown whether the cerebrovascular
lar events mainly result from direct mechanical injury by the worms or are caused by indirect mechanisms such as inflammatory destruction.

Conclusions

The occurrence of cerebral paragonimiasis with hemorrhagic stroke is not rare, while cerebrovascular involvement has seldom been explored. The angiographic findings of cerebrovascular abnormalities in our patients demonstrated the existence of arteritis in cerebral paragonimiasis and suggested a possible relationship between cerebrovascular involvement and cerebral hemorrhage. Further studies of the frequency, mechanism, and prognosis of hemorrhagic cerebrovascular complications associated with cerebral paragonimiasis are clearly warranted.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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