Cerebral Paragonimiasis with Arteriovenous Malformation in a Child: A Case Report

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Abstract
Cerebral paragonimiasis with coexisting arteriovenous malformation is extremely rare. A 6-year-old girl was admitted to hospital due to confusion and frequent vomiting for 20 h. The physical examination was normal except for a slow light reflex on the left pupil. Blood tests showed a high white blood cell count and a normal eosinophil count. The head computed tomography (CT), advanced head CT scan and vascular reconstruction showed a linear fracture on the left harnpan, uneven thickness in the abnormal blood vessels of the left parietal region with a mixed local high-density area with clear edge. Arteriovenous malformation was diagnosed, and a vascular malformation resection was performed. Repeat blood tests showed an increased eosinophil count and the presence of Paragonimus-specific IgG antibodies. The patient was subsequently diagnosed with cerebral paragonimiasis and was given oral praziquantel as 75 mg/kg/day in three divided doses with an interval of four days. After two courses of treatment, the patient was discharged. Follow-up after six months showed negative Paragonimus-specific IgG antibodies titers. Vascular malformation resection combined with oral praziquantel administration provided a satisfactory therapeutic effect for cerebral paragonimiasis with coexisting arteriovenous malformation.

Keywords: Cerebral Paragonimiasis; Arteriovenous Malformation; Vascular Malformation Resection; Praziquantel

Introduction
Paragonimiasis is an orally acquired infectious disease and is widely distributed in China [1], the United States [2], South Korea [3], India, and other countries. Humans are infected by Paragonimus spp through eating raw or undercooked freshwater crabs or crayfish, rarely by eating raw boar meat. When infected crustaceans are consumed raw or undercooked by humans, the metacercarial cysts in the duodenum penetrate through the intestinal wall into the peritoneal cavity, then through the abdominal wall and diaphragm and develop into adults. Both of the adults and larvae of the lung fluke can invade cerebrum through the surrounding tissue of the carotid artery. The majority of lesions are located in the lung, and it is rare to have lesions in the brain only. The paragonimiasis IgG in the serum and the cerebrospinal fluid is positive. Arteriovenous malformation is an abnormal connection between arteries and veins, bypassing the capillary system. To the best of our knowledge, cerebral paragonimiasis combined with arteriovenous malformation has not yet been reported. Here, we report, for the first time, a case of cerebral paragonimiasis combined with arteriovenous malformation in a 6-year-old child and the experiences and lessons we learned after successful treatment.

Case Presentation
A 6-year-old girl was admitted to hospital due to confusion and vomiting for 20 h after a fall with head trauma, accompanied with frequent vomiting, convulsions and low-grade fever on the way to the hospital. The physical examination showed that the patient was pyretic, conscious but lethargic and had equally sized and shaped bilateral pupils, slow light reflex on the left and sharp light reflex on the right, massive swelling of the left scalp, no open wounds on the head or a subcutaneous hematoma, and normal limb muscle tension at a level of IV-V.

Emergency blood tests showed a high white blood cell count (15 × 10^9/L) and a normal eosinophils count (4%; absolute value 0.4 × 10^9/L); the head computed tomography (CT) scan showed a large area of high-density shadow on the left parietal lobe with low-density edema, midline shifting to the right, and a linear fracture on the left harnpan (Figure 1); the advanced head CT scan and vascular reconstruction showed a linear fracture on the left harnpan, uneven thickness in the abnormal blood vessels of the left parietal region with a mixed local high-density area of 2.7 cm × 3.1 cm × 2 cm, and a clear edge located at the lateral side of the curtain bleeding area (Figures 2-3). The patient was diagnosed with arteriovenous malformation. A vascular malformation resection was performed, the cortex was opened with a coagulation scalpel, and part of the hematoma was removed. The rest of the hematoma was separated from the brain tissue and removed completely. The necrotic softened brain tissue around the hematoma was also removed. A repeat head CT scan after surgery did not show any signs of cerebral hemorrhage (Figure 4). The suspected blood vessels were sent for pathological examination, which showed diffusive dots with inflammatory cell infiltration of predominantly eosinophils (Figure 5).

Figure 1: Flat head CT scan. A large area of high density is visible in the left temporoparietal lobe with the corresponding skull fracture.
Retrospective history analysis found that the patient lived in Chongqing, a paragonimiasis-endemic area, but did not have a clear history of eating raw crab or crayfish. Repeat complete blood count showed hemoglobin-110g/L, Neutrophils-85%, Lymphocytes-42%, Monocytes-2%, Eosinophils-10%, eosinophil count -12 × 10^9/L and Erythrocyte sedimentation rate- 50 mm at the end of 1st h (Westergren's method). Enzyme linked immunosorbent assay (ELISA) test for paragonimiasis were conducted, IgG in the serum and the cerebrospinal fluid were examined. Examination of stool sample and cerebrospinal fluid for Paragonimus eggs were negative. Chest X-ray showed no abnormalities. The patient was subsequently diagnosed with cerebral paragonimiasis and was given oral praziquantel at 75 mg/kg/d, three times per day for three days with an interval of four days, which was counted as one course of treatment. After two courses of treatment, the patient was discharged. After discharge, the patient continued to take one course of praziquantel treatment. Follow-up after six months showed that the patient was negative for paragonimiasis IgG antibodies.

**Discussion**

The majority of lesions in patients with paragonimiasis are located in the lung, and it is rare to have lesions only in the brain. While in this case, the patient presented only with cerebral hemorrhage. This is probably the main reason that the patient was not correctly diagnosed in the first place. The main complications of cerebral paragonimiasis consist of cerebral hemorrhage and subarachnoid hemorrhage [4], and occasionally venous sinus thrombosis [5] associated with arteriovenous malformation or aneurysm, which are easily missed. In this case, the arteriovenous malformation led to cerebral hemorrhage, and cerebral hemorrhage led to tumble, which caused a skull fracture; and after surgery, the pathological examination showed an increased eosinophils count. Together with the medical history of living in a paragonimiasis-endemic area, the presence of the paragonimiasis IgG antibodies in serum and in cerebrospinal fluid and the imaging manifestation, a diagnosis of cerebral paragonimiasis combined with arteriovenous

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**Figure 2:** Enhanced head CT scan. Abnormal enhancement of the vascular shadow is visible in the left temporoparietal lobe.

**Figure 3:** Head CT angiogram. A mass of abnormal blood vessels is visible in the left temporoparietal lobe.

**Figure 4:** Repeat head CT scan. No visible hemorrhage.

**Figure 5:** Pathology study. Infiltration of inflammatory cells was visibly dominated with eosinophils.
malformation was made. A comprehensive literature search did no similar case reported previously in the world.

Symptom onset in arteriovenous malformations often starts with acute cerebral hemorrhage caused by abnormal blood vessel rupture, and the onset of cerebral paragonimiasis also starts with acute cerebral hemorrhage caused by a different mechanism. After studying 135 young patients with arteriovenous malformation, Ellis et al. [6] found that vessel rupture in arteriovenous malformation is related to the size and location of the arteriovenous malformation and drainage by a single deep vein. In addition, Starke et al. [7] found that arteriovenous malformation consists of a mass of abnormal blood vessels, including supply arteries and drainage veins. Since there is no vascular bed between arteries and veins, bleeding occurs under an abnormal blood flow. For a simple arteriovenous malformation, pathological examination would show red blood cells and hemosiderin, etc. Clinical manifestations of cerebral paragonimiasis are mainly due to parasite migration, ovulation, and production of toxic substances. Intracranial hemorrhage is due to vasculitis and vasospasms [8]; therefore, if intracranial hemorrhage was caused by arteriovenous malformation, the pathological examination would show red blood cells and hemosiderin. Thus, in this case, the pathological examination showed diffuse clots with inflammatory cell infiltration of predominantly eosinophils, and then the main reason for intracranial hemorrhage was due to cerebral paragonimiasis rather than arteriovenous malformations.

Retrospective analysis of the diagnosis and treatment in this case indicates that cerebral paragonimiasis can be easily misdiagnosed [9] or missed. The case presented here had an acute onset and a short duration; in addition, the CT scan indicated skull fractures and intracranial hemorrhage, which are easily misdiagnosed as traumatic brain injury or even as arteriovenous malformations. The diagnosis of this case was based on the pathological exam, the presence of the paragonimiasis IgG antibodies, and the satisfactory effect of anti-paragonimiasis treatment. The diagnosis would have been incorrect if only the clinical symptoms were considered. Thus, we should be more vigilant and pay more attention to parasitic diseases. Patients with cerebral hemorrhage, subarachnoid hemorrhage, or brain trauma, who are from a Paragonimiasis-endemic area, should be taken to ask whether there is a history eating crayfish or crabs, or raw wild boar meat. Normal lung findings and a normal peripheral blood eosinophils count does not exclude paragonimiasis, especially cerebral paragonimiasis.

**Conclusion**

Vascular malformation resection combined with oral praziquantel administration provided a satisfactory therapeutic effect for cerebral paragonimiasis with coexisting arteriovenous malformation.

**References**


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