Case Report

Cerebral sparganosis: A case report

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Summary **Sparganosis is a rare parasitic infection in humans caused by a larval cestode of the genus Spirometra. Spargana invade muscle, subcutaneous tissue, the eye, urogenital, and abdominal viscera, but the central nervous system (CNS) is not a frequent site of invasion. Here, we describe an 18-year-old patient with cerebral sparganosis and review data from other cases reported in the medical literature.**

Keywords: Spirometra, cerebral sparganosis, albendazole

1. Introduction

Sparganosis is caused by a migrating tapeworm larva of the genus Spirometra. Spargana invade muscle, subcutaneous tissue, the eye, urogenital, and abdominal viscera, but the central nervous system (CNS) is not a frequent site of invasion. Rare but increasing reports have described cerebral sparganosis from many corners of the world (*1-3*), and recent developments in serologic and radiologic studies make diagnosis of CNS infection much easier than previously. We describe here a patient with sparganosis of the CNS and review data from other cases reported in the medical literature.

2. Case report

An 18-year-old female presented to the infectious department of Shanghai Public Health Clinical Center (SPHCC) with dizziness, headache, decreasing memory and reaction dullness, which had developed over the course of one month. Seizure occurred once during the course. On examination, her body temperature was 37.5°C, blood pressure was 120/80 mmHg and heart rate was 90/min. She had clear consciousness, but mental status was a little bit abnormal. Her reaction ability was dull and memory was decreased. Electrocardiogram (ECG) and X-ray checking for heart and lung was shown to normal signals. She had no nuchal rigidity with normal muscle strength and muscular tension. The signs of Kerning, Brudzinskin and Babinski were abnormal. Whole blood analysis revealed a leukocyte count of 6,500/mm^3, hemoglobin 11.2 g/dL and platelet count 210,000/mm^3. Data for her erythrocyte sedimentation rate, routine blood biochemistry and urine analysis were in the normal range. Anti-HIV antibodies, hepatitis B surface antigen, anti-hepatitis C virus antibodies, and rheumatoid factor were negative.

Lumbar puncture revealed high pressure in the cranial cavity, with a rapid dropping of cerebrospinal fluid (CSF). The leukocyte count and the level of glucose, protein and chloride were normal in CSF. There were no bacteria in Gram staining of the CSF sedimentation. However, the enzyme-linked immunosorbent assay for antisparganum antibody was strongly positive in blood and CSF confirmed by the Institute of Parasite of Chinese Center for Disease Control and Prevention. A cranial MRI was ordered. T1-weighted imaging (T1WI) showed that there was an irregular flake low signal focus in the white matter of left parietal lobe and its edge was vague (Figure 1A). T2-weighted imaging (T2WI) showed focus presented high signals (Figure 1B). Enhanced scans showed irregular enhancement focus and the range of enhancement was smaller than that of high signals observed in T2WI (Figure 1C). Diffusion weighted imaging showed tortuous beaded shape enhancement in the left corpus callosum and parietal lobe in sagittal plane (Figure 1D). Further inquiry into the medical history of the patient revealed she ate poorly cooked frog flesh five months ago.

The patient was finally diagnosed with cerebral

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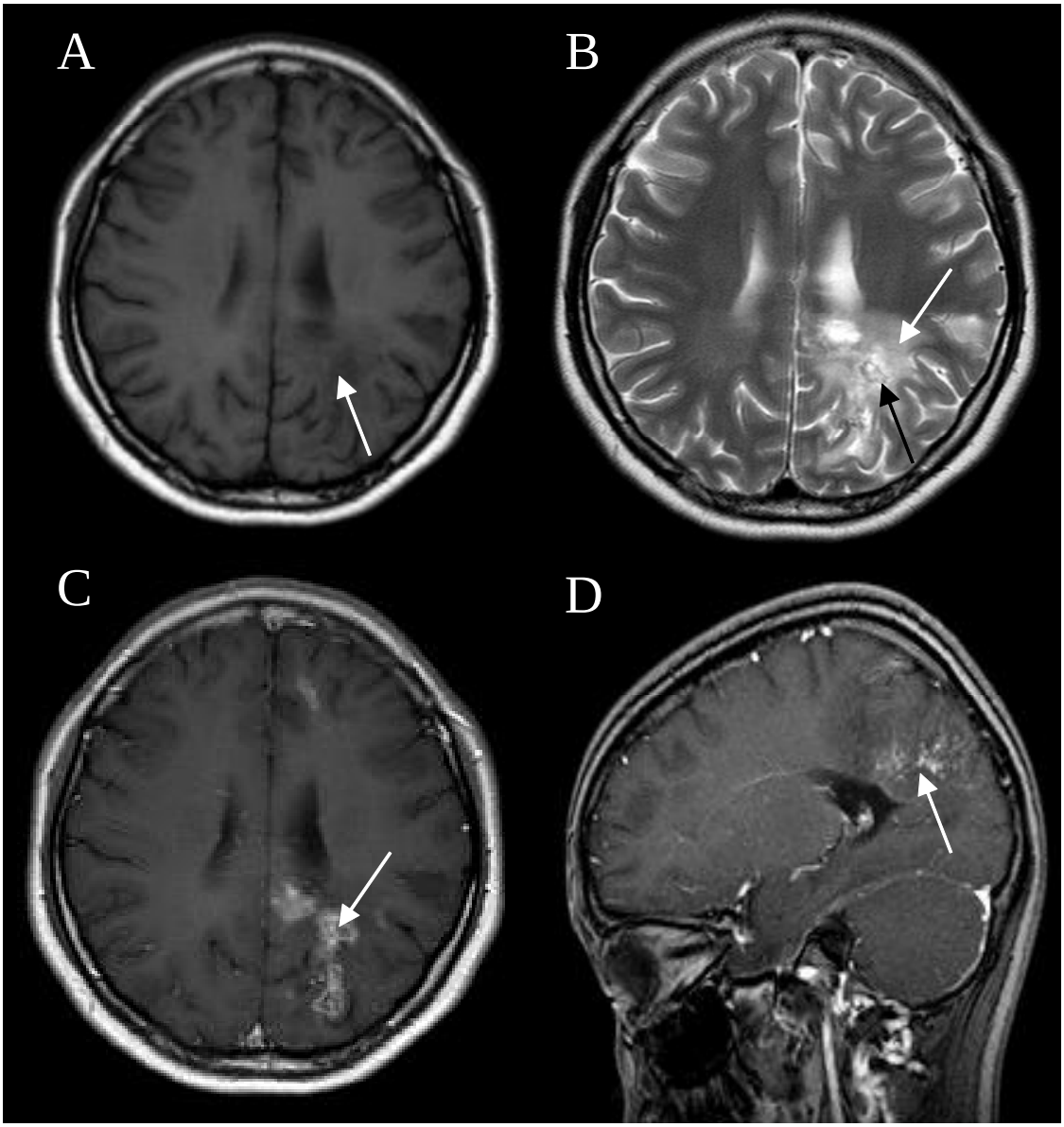

Figure 1. A cranial MRI taken during patient admission. (A) Irregular flake low signal focus in the white matter of left parietal lobe and its edge was vague (T1-weighted imaging, T1WI). **(B)** Focus presented high signals and the surrounding boundary was not clear (T2-weighted imaging, T2WI). **(C)** Enhanced scan showed irregular enhancement focus and the range of enhancement was smaller than that of abnormal high signals observated in T2WI. **(D)** Tortuous beaded shape enhancement in left corpus callosum and parietal lobe in sagittal plane (diffusion weighted imaging, DWI).

sparganosis. She rejected surgical operation and received abendazole (300 mg twice a day) pulsed sodium valproate therapy (200 mg twice a day). The course of treatment was 20 days. The patient was discharged 4 weeks later with no signs or symptoms. No neurological relapse was observed during a one-year follow-up.

3. Discussion

Sparganosis is a rare parasitic infection in humans caused by a larval cestode of the genus Spirometra. Human infection is uncommon and seems to occur accidentally by ingestion of polluted water containing cyclops in which tapeworm eggs mature into procercoid larvae, by ingestion of raw or inadequately cooked flesh of snakes or frogs, and by applying the flesh of an infected intermediate host as a poultice to a wound. The larvae can migrate from primary infection focus to a distant site, such as skin, skeletal muscle, orbital tissue, urogenital and abdominal viscera, and rarely to the CNS. The infective route in this case was probably through eating poorly cooked frog flesh five months ago. There were no other signs or symptoms resulting from sparganum detected other than the brain.

The clinical manifestations of cerebral sparganosis varied depending on the site of infection, including seizure, progressive headache, dizziness, consciousness disturbance and focal neurologic deficits such as hemiparesis, aphasia, and dysarthria. Among these,

seizure and headache were the most frequent symptoms and signs. Fever was rare. However, the clinical presentations of cerebral sparganosis were similar to some other slow progressive CNS diseases. Differential diagnosis such as other infectious granulomas, brain neoplastic lesions, and tuberculous meningitis should be done, especially when the lesions are not responding to treatment (*1*). The patient in this report complained of one month of dizziness, headache, decreasing memory and reaction dullness. Seizure occurred once during the course.

The final diagnosis depends on pathologic or immunologic examination results. ELISA test is a very sensitive test for the detection of sparganosis when the diagnosis is in doubt. In surgically proven cerebral sparganosis, both serum and CSF ELISA tests showed high sensitivity and a concordance rate in diagnosing cerebral sparganosis. The serologic diagnosis by means of ELISA could be a useful tool in the epidemiologic study of human sparganosis.

Computed tomographic (CT) scans and magnetic resonance imaging (MRI) can be very helpful in diagnosing cerebral sparganosis. Typical MRI patterns of cerebral sparganosis included abnormal enhancement such as a peripheral ring-type, tortuous beaded shape or a serpiginous tubular shape enhancement, and changes in location and shape of lesions in the follow-up MRI. Cong *et al*. (*4*) reported that the diagnostic accuracy rate for cerebral sparganosis at the first visit, after first imaging examination and after several follow-up MRI examinations were 0%, 11.8% and 28.6%, respectively. Song *et al*. (*5*) considered that the most characteristic finding was a tunnel sign on postcontrast MRI. The most common finding was bead-shaped enhancement. MRI is superior to CT in demonstrating the extent and number of lesions, except for punctate calcifications. Combined with clinical data and enzyme-linked immunosorbent assay, the preoperative diagnosis of cerebral sparganosis could be established on MR imaging.

The treatment of cerebral sparganosis included surgical operation and medicine. Surgical operations included stereotactic techniques and total removal of the lesion. Patients with no indications for operations can be treated with praziquantel or albendazole, but the therapeutic effect of surgical operation was better than medical treatment (*6*). Gao (*7*) thinks early detection and surgical removal of the parasite and the surrounding granuloma is the best option for its treatment. Our patient was treated with albendazole for 20 days and no neurological relapse was observed during a one-year follow-up. But we need to do further research on the long-term effects of medical treatment.

Cerebral sparganosis is extremely rare. However, the diagnosis of cerebral sparganosis should be considered if patients show seizure, progressive headache, dizziness and migrating granulomatous lesions in the area of epidemic sparganosis. We should inquire about medical history and take pathologic or immunologic examinations in order to exclude the disease.

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