Sparganosis of the brain: a case report and brief review

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ABSTRACT

Human sparganosis is a rare disease often affecting muscle, subcutaneous tissue and other locations, but sparganosis invading the brain is rather rare. Cerebral sparganosis has no specific symptoms which makes the diagnosis quite difficult and is usually neglected in the clinic. Here the authors reported a case of a 29-year-old female who was diagnosed with cerebral sparganosis and underwent surgery in their department and a brief review of the literature was conducted as well.

INTRODUCTION

Human sparganosis is a rare but increasing emerging food borne zoonosis caused by the plerocercoid larvae. The genus spirometra constitutes several species including S mansonoides, one of the most commonly reported human sparganosis infections in Asia[1]. The lifecycle of spirometra starts with adult worms living in the intestines of dogs and cats and eggs shed in faeces in the environment, while frogs and snakes usually serve as second intermediate hosts[2]. Humans are mainly infected through drinking untreated water containing spirometra larvae, consumption of undercooked frog or snake meat, or using raw flesh on open wounds as traditional poultices[3-7]. Sporadic human sparganosis has been documented worldwide, but the prevalence is higher in Asia countries, especially in China, Thailand, Japan and South Korea[3,4,4,8,9]. The most common target places in humans for spirometra are subcutaneous tissue or muscle[10], cerebral sparganosis is relatively rare but represents the most severe type[11], characterized by symptoms like seizures and the wandering sign in magnetic resonance imaging (MRI) scanning[12].
Management of cerebral sparganosis includes surgical removal of the worm as well as postoperative anti-parasite medication. However, cerebral sparganosis cases can be cured with medication alone. Cerebral sparganosis has a good prognosis after treatment. Here, we report a case of a cerebral *S. mansonoides* infection and provide a brief review of the literature.

**CASE REPORT**

**History and examination**

A 29-year-old female patient was admitted to the department of Neurosurgery due to intermittent right upper limb epilepsy and a left parietal lobe lesion. She experienced her first seizure attack half a year ago consisting of involuntary movements in the right upper limb. She did not take it seriously and did not take any anti-epileptic drugs (AED) medications. Two months before admission, seizures became more severe and she found a migratory nodule under her skin on her right thigh. She had lived in Fujian Province her whole life and enjoyed eating frogs in local restaurants. The people in Fujian Province have a habit of eating frogs, crabs, snake, chicken and pig meat, which are all host animals of the sparganosis. Computed tomography (CT) showed a left parietal lesion with large adjacent edema and the MRI showed an irregular hypointense lesion in T1 weighted imaging (T1WI) and hyperintense signal in T2 weighted imaging (T2WI). The lesion was homogenously enhanced in T1WI and with large perilesion edema. MR perfusion showed increased blood flow in the lesion area while spectroscopy showed an increased peak of choline (Cho) and decreased peak of N-acetyl-aspartate (NAA) [Figure 1]. A cerebral parasitic infection was suspected since the patient was from the endemic area with a previous history of eating undercooked frogs. Blood and cerebrospinal fluid (CSF) samples were sent to the China Institute of Parasite Research for detection, the antibody for *S. mansonoides* was positive in blood sample but negative in CSF sample. Another lumbar puncture was conducted and this time the antibody for *S. mansonoide* was positive in CSF sample. A craniotomy was performed to resect the lesion.

**Operation**

A right parietal under-navigation craniotomy was performed. A white long living parasite was seen and extracted. Postoperative pathology confirmed the diagnosis of *S. mansonoides*. The patient received anti-parasite medication after the surgery, postoperative MRI showed complete resection of the lesion. The patient had no seizures after 6-month follow-up [Figure 2].

![Figure 1: Preoperative radiological features of cerebral sparganosis. A: axial view of CT scanning showed irregular edema of the left parietal lobe; B: axial view of T1WI MRI showed a slight edema of the left parietal lobe; C: axial view of T2WI MRI; D: axial view of FLAIR MRI; E: sagittal view of enhanced T1WI MRI showing an irregular enhanced lesion of the left parietal lobe; F: axial view of enhanced T1WI MRS showed an increase of the Cho/NAA ratio; G: axial view of MR perfusion showed the lesion was hypometabolic; H: sagittal view of preoperative DTI. CT: computed tomography; MRI: magnetic resonance imaging; NAA: N-acetyl-aspartate; T1WI: T1 weighted imaging; DTI: diffusion tensor imaging](image-url)
Sparganosis of the brain

To investigate the epidemic and clinical characteristics of cerebral sparganosis. We searched Pubmed using the keywords “cerebral sparganosis” and “cerebral sparganosis mansonoide”. English literatures with at least abstract available were reviewed.

Epidemiology
Human sparganosis usually occurs when patients consume undercooked frog or snake meat, drink unboiled water contaminated by procercoidlarve. In some rural areas, people use fresh snake or frog meat directly on open wounds as a cure method. Some rural areas, people use fresh snake or frog meat directly on open wounds as a cure method. In China, the majority infected people had a history of consuming undercooked frog or snake meat. Besides histories of traveling or living in endemic areas, an active infection of other organs is also a useful clue for the diagnosis of cerebral sparganosis. In our case, the patient had a migratory nodule on the right upper limb, which makes us highly suspect a diagnosis of cerebral sparganosis.

Clinical characteristics
Cerebral sparganosis can cause various symptoms including headache, seizure, parenthesism, confusion, memory loss, etc. The symptoms are related to the location of the lesion[14], the most common symptoms were various types of seizures. In some cases, it can even present as an intracranial hemorrhage[9]. Fever is not a common symptom. Lesions are mainly located at frontal-partial lobes but invasion to cerebellums are also reported in very few cases[18]. Diagnosis of cerebral sparganosis is relatively hard since it has no specific manifestations. A history of traveling or living in endemic areas may indicate a possibility for the diagnosis. In China, the majority infected people had a history of consuming undercooked frog or snake meat. Besides histories of traveling or living in endemic areas, an active infection of other organs is also a useful clue for the diagnosis of cerebral sparganosis. In our case, the patient had a migratory nodule on the right upper limb, which makes us highly suspect a diagnosis of cerebral sparganosis.

Laboratory test and neuroimaging
Blood can show an increase of eosinophilia[13]. The disease can also be diagnosed with antigen specified IgG antibodies from blood and CSF samples, although cross-reactivity with other infestations or clonorchiasis limits specificity[15,16]. In our case, the first CSF sample was negative for immune-reactivity but positive for blood sample, however, the second blood and CSF sample were both positive.

CT scans usually show a mixed density lesion with peri-lesion edema, punctate calcification is shown in approximately 50% of patients[15,20]. The MRI of cerebral sparganosis is difficult to identify from low grade glioma or other tumors. Typically, ring-like or string-knot enhancement in T1WI images shows the sign of sparganosis movement in the brain parenchyma. Other characteristic features include a tunnel-shape configuration due to immigration of sparganosis through the brain parenchyma. Moreover, serial imaging may demonstrate the tunnel sign from a small nodular lesion and reflect the sparganosis activity in the brain[21,23].

MR spectroscopy of cerebral sparganosis (Bo and Xuejian[24] and Chiu et al.[25]) showed increased Cho and decreased NAA peaks in their cases, as in our case. Chiu et al.[25] also revealed peaks at 1.3 ppm and a peak between 14 and 1.8 ppm in his 46-year-old female patient, however, our case did not show a similar result. MR perfusion did not show higher cerebral perfusion in our case that was seen in the case of Chiu et al.[25].

Surgery
Although some studies report that using anti-sparganosis drugs can successfully cure the disease, the most efficient way to cure the disease still is...
surgical removal of the sparganosis from the infested site of the brain. Both stereotactic aspiration and an opening craniotomy were used by neurosurgeons. The goal of surgery is to completely remove the granuloma along with the larval. Deng et al.\textsuperscript{[26]} reported a series of 11 cases who underwent stereotactic aspiration surgeries, complete removal as achieved in 10 patients while incomplete removal in 1 patient. Due to the small wounds, stereotactic aspiration is recommended by most neurosurgeons. In the 26 cerebral sparganosis cases of Hong et al.\textsuperscript{[18]}, 16 of them were treated with craniotomy, 7 of them were treated with stereotactic aspiration and another 3 were treated with praziquantel only. None of them experienced a relapse of the disease. Yu et al.\textsuperscript{[13]} reported in 8 of 9 cases of cerebral sparganosis patients underwent a craniotomy due to lack of stereotactic equipment, with 1 patient dying due to unspecified reason. Both investigators reported that symptom duration more than one year indicated worse prognosis after surgery.\textsuperscript{[13,18]} In our case, we used a navigation guided craniotomy to resect the granuloma as well as the peri-lesion glia proliferation zone. The symptoms resolved immediately after surgery, the patient was administrated AED medication and 5 mg/day of praziquantel for 3 months. After 6-month follow-up, the patient was well without further seizures.

Cerebral sparganosis is a neglected food borne zoonosis and since it has no specific clinical characteristic, it is usually misdiagnosed until postoperative pathological finding. Patients who had a history of consuming undercooked meat or from endemic areas should be highly suspected. Immunosorbent assay for sparganosis antibody using blood and CSF samples as well as MRI images can provide evidence for preoperative diagnosis. Surgery should be performed and postoperative anti sparganosis drugs should be administered. Usually, cerebral sparganosis had a satisfying outcome based on surgery and drugs.

**DECLARATIONS**

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**Conflicts of interest**

There are no conflicts of interest.

**Patient consent**

Informed consent was obtained from the patient included in the study (KY-2012-019).

**Ethics approval**

All procedures performed in studies involving human participants were in accordance with the ethical standards of Huashan Hospital, Fudan University and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

**REFERENCES**


