Cerebral sparganosis is a rare but underestimated parasitic disease caused by infestation by sparganum. It is difficult to make a confirmed preoperative diagnosis of this disease given the absence of characteristic clinical manifestations. A detailed protocol for the diagnosis and treatment of cerebral sparganosis is still lacking in the literature. In this article the authors set out comprehensive procedures for the diagnosis and treatment of cerebral sparganosis, describing the use of a stereotactic aspiration technique complemented by microsurgery based on experience gained from multiple cases.

Methods. The disease history, clinical manifestations, imaging features, and therapeutic procedures for 11 patients with cerebral sparganosis were retrospectively analyzed. Stereotactic aspiration procedures were performed in all 11 patients and were complemented by microsurgeries in 3 patients. The learning and experience gained from these treatments were summarized, and a comprehensive protocol for the diagnosis and treatment of cerebral sparganosis was reviewed.

Results. Larvae of Spirometra mansoni were taken from all 11 patients: completely removed in 10 cases and partially removed in 1 case (discovered later). After surgery, clinical symptoms in all 11 patients were significantly improved. All epileptic symptoms were successfully cured, although in 1 case occasional seizures still occurred because of the incomplete removal of the larva. Muscle strength in the 4 patients who had hemiparesis prior to surgery recovered to normal. Symptoms in the 1 patient who had presented with partial body sensory disturbance resolved after surgery. There were no complications or deaths.

Conclusions. The authors concluded that an effective preoperative diagnosis of cerebral sparganosis can be made by detailed inquiry into the possible infection history and disease symptoms as well as careful scrutiny of characteristic radiological features and immunological testing results. In stereotactic operations performed to remove the larva, priority should be given to image-guided stereotactic aspiration given that it causes the smallest wounds. In cases in which stereotactic aspiration fails, stereotactic microsurgery should be performed to remove the larva. The surgeon must carefully avoid breaking the larva and leaving behind any larva residue during surgery.

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Key Words • cerebral sparganosis • Spirometra mansoni • stereotactic aspiration • microsurgery

Abbreviation used in this paper: ELISA = enzyme-linked immunosorbent assay.
monitoring of existing cases. In the present study, we retrospectively analyzed the clinical manifestations, imaging features, and therapeutic procedures for 11 patients with cerebral sparganosis and summarized the experience gained from stereotactic operations for the treatment of this disease.

Methods

General Information

Between October 2004 and January 2009, 11 patients (7 males and 4 females) suffering from cerebral sparganosis were treated in the Department of Neurosurgery at the No. 94 Hospital of People’s Liberation Army, Nanchang. Medical records, radiological images, and pathological specimens were retrospectively reviewed. The age of the patients at diagnosis ranged from 16 to 52 years, and the average age was 28.3 years. Disease history ranged from 2 weeks to 3 years. Among the 11 patients, 4 had eaten raw fish and frogs. All patients except 1 were from rural areas. Cerebrospinal fluid ELISA tests for the sparganum-specific antibody were performed in 5 patients. Nine patients with epileptic symptoms (seizures) were treated with antiepilepsy drugs. Stereotactic aspiration surgeries were performed to remove larvae in all 11 patients. In 3 cases in which the aspiration operations failed, stereotactic microsurgeries were performed. The follow-up period ranged from 6 to 49 months (average 26 months).

Stereotactic Operation

Prior to the operation, a stereotactic head frame was mounted, and enhanced CT scanning was performed. Targeting coordinates were calculated by selecting the highest density of the lesion as the origin. Regional anesthesia was then applied, and the incision site was selected based on the stereotactic targeting point. A 2- to 3-cm straight cut was made on the scalp, and a 10-mm-diameter hole was drilled in the skull. After carefully opening the dura mater, a lateral opening biopsy needle (Fig. 1A) was used to aspirate the larva from multiple directions. In 3 cases in which no larvae were obtained after repeated aspiration, microsurgeries were performed with stereotactic guidance. The scalp incision was extended to 4–5 cm, and a bone flap with a diameter of approximately 3 cm was removed using a milling cutter. After carefully opening the dura mater, the larva together with the lesion was cut from the brain tissue with the aid of a microscope.

Results

Clinical Data

All 11 cases of disease were single-site infestations. Infected sites included 5 parietal lobes, 3 frontal lobes, 2 occipital lobes, and 1 basal ganglion. Nine patients had seizures, 6 experienced headaches and dizziness, 4 had hemiparesis, and 1 presented with partial body sensory disturbance. None of the patients had a history of fever. Among the 5 cases in which CSF ELISA tests for the sparganum-specific antibody were performed, 2 were positive.

Radiological Findings

On plain CT scanning low-density lesions were observed in 7 patients. Punctate calcification, indicating degeneration of brain tissue, was demonstrated on presentation in 3 patients. Irregular high-density lesions accompanied by obvious surrounding edema, indicating granulomatous inflammation, was revealed in the other 4 patients. On enhanced CT scans 9 cases exhibited irregular enhancement in lesion density or punctate foci, whereas the other 2 cases showed no obvious enhancement. Plain MR imaging examinations in all 11 patients revealed irregular, patchy abnormal signals, with low signal for T1-weighted images and high signal for T2-weighted images. Boundaries were not clear. On enhanced MR imaging examinations, 2 cases demonstrated circular high signals and 3 showed twisted chain-like structures, indicative of plerocercoid larvae.

Treatment and Outcome

Stereotactic surgeries were performed to remove larvae from all 11 patients, which were completely removed in 10 patients and partially removed in 1. All larvae were determined to be spargana according to assays from the pathology departments at both our hospital and Nanchang University (Jiangxi Province). One larva example is shown in Fig. 1B. At the time of hospital discharge, all patients exhibited significant recovery from the preoperative clinical symptoms. The 9 patients with seizures continued to be treated with antiepilepsy drugs until 6 months after surgery. All epileptic symptoms were successfully cured, except in 1 case in which occasional seizures still occurred. Follow-up examination after 1 year in this case revealed that the lesion had transferred to the frontotemporal lobe from the parietal lobe, indicating that the larva was only partially removed during the initial operation. Muscle strength in 4 of the patients who had hemiparesis prior to the operation recovered to normal. Symptoms in the 1 patient who had presented with partial body sensory disturbance disappeared after surgery.

Follow-up radiological examinations in the 8 patients who were only treated with stereotactic aspiration surgery revealed deflated edema at previous lesion sites after 3 months (Fig. 2). After 6 months, both lesions and edema disappeared. For the 3 patients in whom larvae and lesions had been removed through microsurgeries after failed aspirations, radiological examinations after 3 months showed the disappearance of the lesions and surrounding edema, except for localized cerebral malacia (Fig. 3).

Discussion

Adult S. mansoni are mainly parasitized in feral cats and dogs; however, the larva form can survive in human tissues, causing sparganosis. Adult tapeworms in the intestines of cats and dogs produce eggs, which are passed out with feces and reach fresh water. Coracidia hatched from these eggs are then ingested by copepods (first intermediate host), developing into proceroid larvae that are ingested by frogs, snakes, birds, or mammals. In these...
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Fig. 1. A: A photo of the lateral opening biopsy needle set used for the stereotactic aspiration surgery. During the aspiration operation, the inner needle is pulled out so that the larva can be sucked into the outer needle jacket. B: Photograph of the plerocercoid larva taken out by stereotactic aspiration.

second intermediate hosts, procercoids mature into plerocercoid larvae. Humans can be considered dead-end hosts of spargana. The survival period of spargana in the human body ranges from 5 to 20 years. Once a human becomes infected, the plerocercoid larva can migrate to the brain, resulting in cerebral sparganosis. Cerebral sparganosis constitutes about 2.3% of the overall reported cases of human sparganosis. In China, more than 800 cases have been documented, and the patient age has ranged from several months to 62 years. The highest infestation rate is reported to occur between 10 and 30 years of age, and the male/female ratio is 2:1.

There are 2 main routes of human infestation by spargana: 1) penetration through skin and mucosa, and 2) ingestion of raw food or contaminated water through the mouth. Statistics show that more than half of infestations occur through a wound and skin/mucosa, and most infestations invade subcutaneous tissues and muscles. It is still not very clear how the sparganum enters the brain. Based on previous animal studies and observations of sparganosis in the human abdominal cavity, pleura, neck, and spinal cord, the following route can be proposed. After being ingested into the mouth and passing through the alimentary canal, the sparganum reaches the abdominal cavity. It then penetrates the diaphragm and mediastinum and reaches the neck, where it further migrates up through the perivascular space. After passing the foramen magnum, lacerate foramen, and jugular foramen, the sparganum enters the brain. Proteolytic enzymes in the scolex allow the sparganum to hydrolyze proteins and peptides, and thus facilitate the penetration of thick tissues and membranes.

Clinical manifestations of cerebral sparganosis depend on the infested site. The most commonly observed symptom is seizure, followed by hemiparesis, progressive headache, partial body sensory disturbance, and alteration in consciousness. Lesions are mainly located at the parietal lobe, frontal lobe, occipital lobe, cerebral ganglia, and basal ganglia. Patients typically have no history of fever. One characteristic helpful in the diagnosis of this disease is a possible alteration in the clinical manifestations of cerebral sparganosis with the migration of the larva. Radiological examinations typically reveal mechanical impairments and chronic inflammations, mainly due to the locomotion of the larva inside the brain tissue. Characteristic CT features include 1) a low-density lesion in the white matter with adjacent ventricular dilatation, that is, the so-called negative effect; 2) punctate calcification; and 3) an irregular or nodular enhanced lesion. Magnetic resonance imaging features manifest as 1) low signals for T1-weighted images and high signals for T2-weighted images; 2) a characteristic rosary or twisted chain-like lesion that is consistent with the larva shape on enhanced MR imaging; and 3) poor calcification boundaries. It is particularly helpful in diagnosing this disease when lesion migration is observed in sequential radiological examinations, indicating the existence of a live larva.

Since cerebral sparganosis is rare and lacks characteristic clinical manifestations, it is difficult to confirm the disease at the preoperative stage. We recommend the following as references for the diagnosis of this disease: 1) patients are young adults; 2) a history of eating raw frogs, snakes, or fish; 3) a prolonged course of disease and frequently changed symptoms; 4) radiological evidence.

Fig. 2. Preoperative and postoperative MR images obtained in a patient with cerebral sparganosis. The larva was removed through CT-guided stereotactic aspiration surgery. Enhanced MR images in the coronal (A) and axial (B) planes showing rosary enhancement at the right parietal lobe. Enhanced axial MR image (C) obtained 3 months after surgery, demonstrating no high signal except for local edema.
of the imaging features described above, especially lesion migration; 5) a positive ELISA test for the sparganum-specific antibody; and 6) a history of sparganum infestations of other organs.

As of today there is no specific medicine for the treatment of cerebral sparganosis, and most administered drugs play only supplementary roles. The most efficient way to cure this disease is to remove the sparganum from the infested site in the brain. Stereotactic surgery provides the most effective approach for this purpose. In the stereotactic operation, priority should be given to image-guided stereotactic aspiration, especially to remove lesions from deep brain and important functional regions, because this surgery creates the smallest wound and prevents broken larvae. Moreover, because of its intrinsic escaping mechanism, the larva is not adherent to surrounding tissue, which greatly enhances the success rate of aspiration surgery to cure this disease. We recommend devoting special attention to the following items during stereotactic aspiration surgery: 1) The most enhanced feature on the CT scan or MR image should be selected as the targeting point. 2) Aspiration should be performed in multiple directions radiating from the origin. 3) Carefully examine the integrity of the removed larva, paying particular attention to the scolex. If multiple strings were sucked out, it is likely that the larva is broken given that there has been no report of 2 spargana at 1 infestation site to date (the procercoid larva does not proliferate in its second intermediate host). In all 11 cases in this study, a lateral opening biopsy needle (Fig. 1A) was used to suck out the larvae by applying appropriate negative pressure. Avoid using biopsy forceps to directly remove the larva to prevent breaking of the string-like larva. Note that aspiration helps to induce localized negative pressure so that the larva can be sucked out even though there is no direct contact between the needle and the larva at the beginning. One should not give up in the event that the larva cannot be taken out after repeated aspirations but should undertake a craniotomy operation. After opening a small window (approximately 3 cm) in the skull, the larva can be removed by cutting out the lesion with the aid of a microscope. Again, carefully avoid breaking the larva body.

Conclusions

Effective preoperative diagnosis of cerebral sparganosis can be achieved by a detailed inquiry into any possible infection history and disease symptoms as well as careful scrutiny of characteristic radiological features and immunological testing results. Stereotactic surgery provides the most effective approach to cure this disease. During this operation, priority should be given to image-guided stereotactic aspiration, because it leaves the smallest wound, followed by a craniotomy in the event that aspiration fails. Follow-up examinations should be performed periodically to ensure that no larva residue remains in the brain after surgery.

Disclosure

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

Author contributions to the study and manuscript preparation include the following. Conception and design: Deng, Qian. Acquisition of data: Deng, Xiong. Analysis and interpretation of data: Deng. Drafting the article: Deng. Reviewed final version of the manuscript and approved it for submission: all authors.

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