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Case Report

Cerebral Sparganosis: A Case Report

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Abstract

Introduction: Cerebral sparganosis is a rare parasitic disease in Cuba.

Objective: To describe the treatment process of a patient with multiple brain injuries whose final diagnosis was sparganosis.

Presentation: A patient diagnosed with cerebral sparganosis performed by biopsy guided by stereotaxy is presented. He was consulted for difficult-to-control seizures and headache. Imaging studies showed brain lesions in the frontal and left thalamic areas. Histopathological study and immunohistochemical tests revealed a larva of Spirometra spp. He was given treatment with albendazole and praziquantel, after which he had clinical and imaging improvement.

Conclusion: Cerebral parasitic infection due to sparganosis is uncommon in our environment and stereotactic biopsy is an effective tool in the diagnosis and treatment of multiple brain lesions as a form of presentation of the disease.

***Keywords:** Sparganosis, biopsy, stereotaxy*

Introduction

Sparganosis is a zoonosis caused by the larva of the cestode Spirometra mansoni. It was first described by Patrick Manson in China in 1882, and the first case of human infection was reported by Charles Wardell Stiles in Florida in 1908.[1] Although this disease is reported around the world, it is more frequent in Southeast Asia (China, Japan and Korea). Humans are incidental hosts in the life cycle, while dogs, cats and other animals, which carry the parasite in the intestine, are the definitive hosts.[2] The incubation period is from 20 days to 3 years. Human infection occurs mostly by eating raw or poorly cooked meat of toads and snakes, chicken and fish (second intermediate host) or by ingesting water containing infected copepods of the genus Cyclops, and they grow in the muscles and subcutaneous tissue, eye, pleura, urinary tract and abdominal cavity but occasionally the brain can be affected (cerebral sparganosis), the latter being extremely rare. Neurologic manifestations of cerebral sparganosis include headache, focal motor deficit, and epileptic seizures. Diagnosis is made by history, clinical picture, laboratory tests, imaging and histopathology. Neuroimaging studies show one or several granulomatous lesions that capture contrast and that sometimes bleed or change their location due to the migration of the larva.3 There are few Cuban reports of cerebral infestation due to sparganosis.[4,5]

Clinical Case

A 47-year-old male patient from Villa Clara, an agricultural worker, with no history of eating raw or poorly cooked meat and with a history of post-traumatic epilepsy for which he had been treated with Carbamazepine (200mg) 2 tablets every 8 hours since he was 33 years regularly and without seizures for about 5 years. He was brought to the clinic due to reappearance of generalized tonic-clonic convulsive episodes with a postictal period of 5-6 minutes associated with holocranial headache of two months' duration and episodes of behavioral and anxiety disorders. Physical examination revealed disorientation in space and person; the neuropsychological evaluation revealed alterations in the behavioral sphere. Computed axial tomography (CAT) and magnetic resonance imaging (MRI) neuroimaging studies showed two annular lesions with perilesional edema and contrast uptake in the left frontal and parathalamic region, measuring 18x15x18mm and 14x15x15mm, respectively (Figure 1); he is referred to our service under diagnostic suspicion of brain metastasis. The extension studies performed did not show primary tumor lesion. Dexamethasone (4mg) 12mg daily was administered orally and brain biopsy was performed on the frontal lesion guided by stereotaxy using a Leksell frame and with the patient awake.

There were no complications during the procedure and a live and undamaged parasite was extracted (figure 2). He complied with treatment with Albendazole at a dose of 15 mg/kg/day divided into two subdoses for 6 weeks and Praziquantel 50mg/kg/day in three doses for 14 days with clinical and radiological improvement.

The sample was sent to the Pedro Kourí Institute of Tropical Medicine, where we were notified of the sparganosis infection.

Radiological CT studies were performed in the immediate postoperative period in order to rule out complications of the procedure, without the presence of hemorrhage and little air at the biopsy site. The MRI at three months after completing pharmacological treatment showed total remission of the lesions (figure 1).

He currently incorporated into his usual life with negativity in his imaging studies and control of his convulsive symptoms.

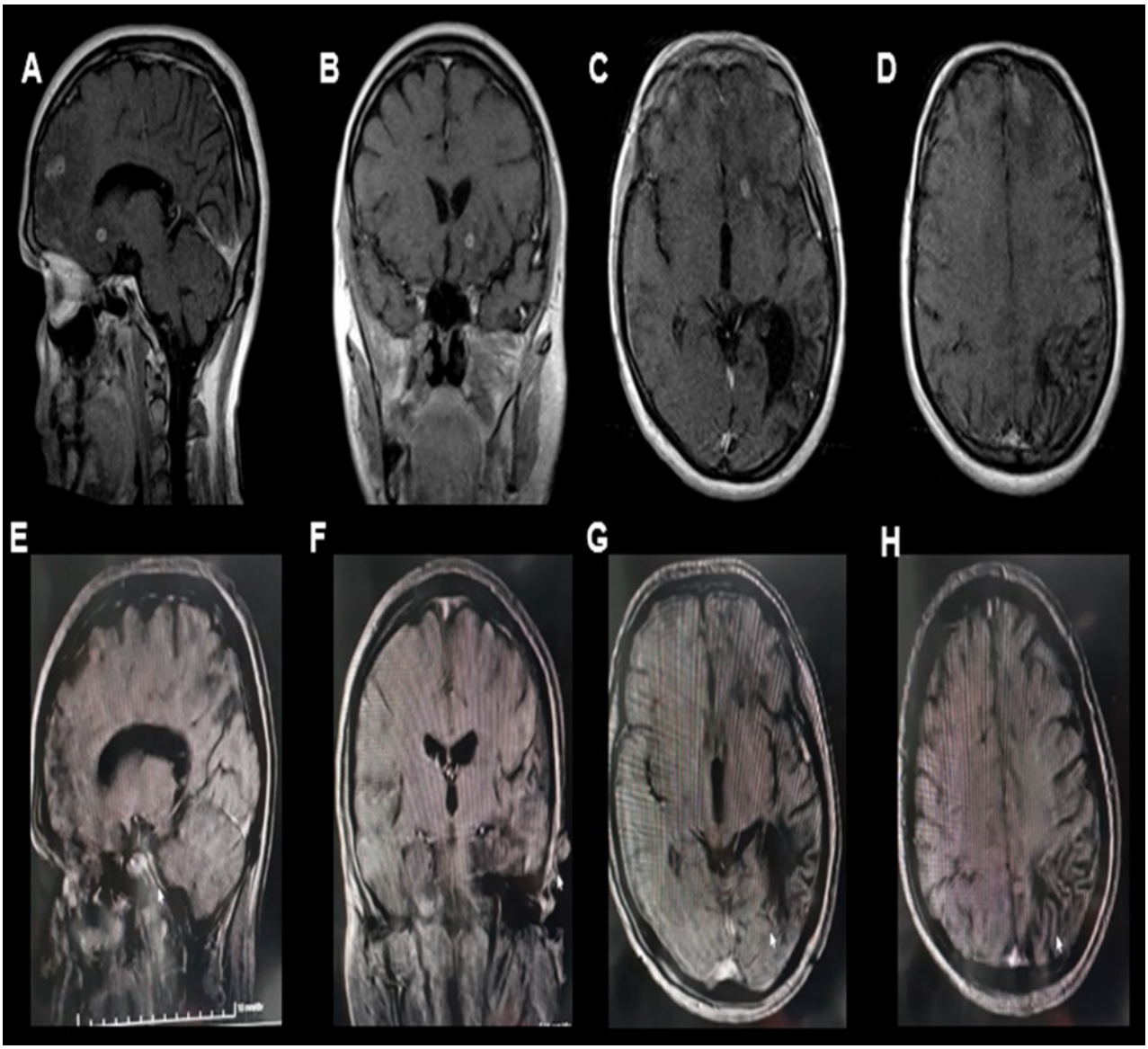

Figure 1: Magnetic resonance images, coronal and axial sagittal sections, respectively, preoperative (A, B, C, D), where two annular lesions with perilesional edema and contrast uptake are observed in the frontal and left parathalamic regions, 18x15x18mm and 14x15x15mm respectively; and postoperative 3 months later (E, F, G, H) where the absence of lesions is visualized.

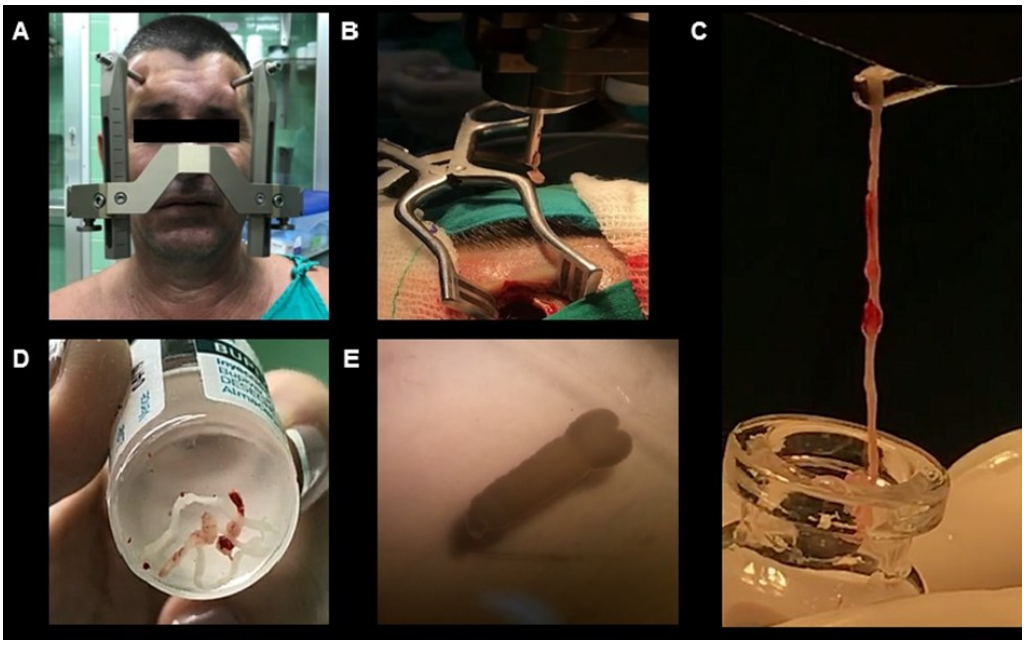

Figure 2. A Patient with the stereotaxic frame. B. Moment of removal of the parasite during surgery. C. Removal of the parasite from the Sedan cannula. D Sample sent to Pathological Anatomy and E. Parasite under microscopic vision fixed with formaldehyde

Discussion

Sparganosis is a zoonosis caused by the larva of the cestode of the genus Spirometra, family Diphyllobotridae, order Pseudophyllidea. This larva is also known by the generic name of espargano, from which it takes the name of the disease.[6,7]

Although there are several species of the genus Spirometra (Spirometra erinaceieuropaei (syn. S. erinacei or S. mansoni) and S. mansonoides), most infestations in humans are caused by the larvae of the adult tapeworms Spirometra mansoni and Spirometra erinacei. . S. erinaceieuropaei is most frequently found in Asia and Africa, while S. mansonoides is mostly reported in North America. In South America, the most common species is the mansoni.[8,9]

The adult parasite lives in the intestine of dogs, cats or other mammals. The first intermediate is clean water copepods while the second intermediate includes amphibians and reptiles such as frogs, toads and snakes. Humans are accidental hosts that become infected when they eat raw or poorly cooked meat with plerocercoid or when they use toad or snake meat as a poultice for the treatment of skin diseases or eye anti-inflammatory, or drink water contaminated with infected copepods. Once ingested, the larva penetrates the digestive tract and enters the peritoneal cavity, later invading muscle or subcutaneous tissue and other organs. The adult parasite is a white pseudosegmented helminthic 3 to 30 cm long. Plerocercoids are incapable of penetrating through intact skin, but they can penetrate through lesions in the skin, eyes, mucous and membranes.[6 -8]

The most common spargan infection is cutaneous, and extracutaneous infection can be located in muscles, eyes, tongue, breasts, lungs, pleural cavity, abdominal organs, urogenital tract, pericardium, mediastinum, neck, spinal canal, intraosseous and brain.[7 -10] The clinical presentation depends on the number and location of the larvae. In the early stages of human infection, it may be asymptomatic, but the migration and secretion of plerocercoids generally cause a localized inflammatory reaction and perilesional edema. The most common clinical manifestation of sparganosis is migratory subcutaneous nodules that appear and disappear over a period of time. The nodules are pruritic, with inflammatory signs, turn reddish and are accompanied by painful edema. Chills and fever can also accompany the infection, and eosinophilia is a common sign in patients.[7]

Cerebral localization is not frequent and represents 2.3% of all published cases. There is no confirmed theory that explains how the parasite reaches the brain, however Deng et al propose that once ingested it reaches the abdominal cavity, passes through the diaphragm and mediastinum and reaches the neck where it migrates through the perivascular spaces, after passing through the entrance holes of the cranial base, the spargan would enter the cranial cavity and brain.[11] The survival period of the spargan in man can extend up to 20 years.[12]

The most frequent clinical manifestations are seizures, hemiparesis, progressive headache and alteration of consciousness, although in cases located in the spinal canal they manifest as back and radicular pain, urinary incontinence and paraparesis.[8] The most frequent cerebral location is in the parietal, occipital and frontal lobes and basal ganglia.[13] Patients usually have eosinophilia and absence of fever when brain lesions are present. Imaging studies show areas of low density, the tunnel sign (corresponding to cavitated abscesses visible on biopsy), rosary or nodular lesions, and calcifications.

The presence of these last two characteristics usually generates the differential diagnosis with tuberculoma. Recognition of the migration of the lesion in the sequence of imaging tests shows one or several granulomatous lesions that enhance contrast and that sometimes bleed or change location and indicate the presence of a live larva.[13,14] In the case presented there was no presence of eosinophilia and neuroimaging studies showed multiple annular lesions that led to the suspicion of the presence of brain metastases. The clinical diagnosis of sparganosis is difficult because the symptoms are not pathognomonic and the definitive diagnosis is through biopsy of the subcutaneous nodule (in the case of cerebral or visceral sparganosis it is more difficult because it requires the removal of the larva), the Serological tests are the most used for diagnosis. ELISA (English acronym for Enzyme-Linked Immunosorbent Assay), using S. erinaceieuropaei sparganum antigens, is for the detection of specific anti-spargan antibodies, and has good sensitivity and specificity but is cross-reactive with patients with cysticercosis and paragonimiasis. ELISA using native or recombinant cysteine protease as antigen has higher sensitivity (100%) and specificity (97–98.22%) for serodiagnosis of human sparganosis compared with ELISA using sparagan ES antigens.[7] Specific recognition in blood or CSF(cerebrospinal fluid) was not performed before surgery because the presence of the parasite was not suspected due to its rarity in the environment. Surgical intervention is usually performed in the open or, in more experienced settings, through stereotactic surgery. The first allows a complete curative resection, although localization in depth is difficult when a small lesion does not have cortical representation. For its part, the second allows the lesion to be located in depth with certainty, but it must be performed by surgeons with experience in the procedure.

It also has the drawback that the lesion often has significant fibrosis that makes it difficult for the needle to penetrate. Pathological anatomy shows a chronic granulomatous inflammatory process with plasma cells and eosinophils, and marked collagenization. The cavitation areas present usually contain the cestode in the larval stage.[15] Diagnosis of cerebral sparganosis is frequently post-surgical and histopathological due to the low incidence and case reports in the area and the country, being difficult to confirm the disease in the preoperative stage, and in some circumstances the differential diagnosis with brain metastases is proposed , neurocysticercosis, abscesses and other multiple lesions before suspecting infection by asparagus, as in the case of the patient presented. Surgical treatment is the treatment of choice and the one with the best results,[15] however, in not all patients surgery seems to be beneficial, only in cases in which the parasite is alive or there is an inflammatory granulation, if the patient is not a candidate for surgery, conservative treatment may be acceptable due to spontaneous regression of granulation tissue after parasite death.[16] Surgical removal of the larva is the most effective therapy for subcutaneous, ocular, oral, and cerebral sparganosis. The body of the worm must be completely removed because the remaining scolex can cause recurrence of the disease. Praziquantel is the treatment of choice in case of multiple subcutaneous nodules and visceral sparganosis, the recommended full dose is 120-130 mg/kg divided into 2-3 doses for 2-3 days, however praziquantel does not have a complete effective therapy for cerebral sparganosis and its use is controversial, when associated with surgical removal it has better results.[17] In case of deep infections the use of albendazole is recommended.[10] In our opinion, resection, with selected and individualized criteria, was preferable to pharmacological treatment because it not only facilitates diagnosis, but also allows the disease to be cured in most cases.

This patient had two lesions, a brain biopsy guided by stereotaxy was planned and one of the parasites was removed, which allowed diagnosis and adequate treatment of the other, which was carried out with Albendazole 10-15mg/kg7day divided into two subdoses for 14 days. and Praziquantel 50 mg/kg/day divided into three subdoses for 14 days, associated with corticosteroids. Surgical removal of the thalamic lesion was not planned due to the high morbidity and sequelae associated with the procedure. Clinical and imaging follow-up was performed, observing the resolution of the condition.[16,17]

Knowledge of the clinical-epidemiological characteristics and distinctive radiological findings increase the detection and adequate treatment of sparganosis in the central nervous system.

Conclusions

Cerebral parasitic infection due to sparganosis is infrequent in our environment and can present as multiple lesions. Stereotactic biopsy is an effective tool in the diagnosis and therapy of this disease.

Conflicts of Interest

None

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