A Case of Acquired Immunodeficiency Syndrome With Cerebral Sparganosis and Review of the Literature.

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Case report

Keywords: HIV/AIDS, cerebral sparganosis, encephalopathy, praziquantel

Posted Date: April 20th, 2021

DOI: <https://doi.org/10.21203/rs.3.rs-434937/v1>

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Abstract

Background

AIDS existing with cerebral sparganosis is rare. Due to atypical symptoms of sparganosis, diagnosis and treatment are difficult and the misdiagnosis rate is very high. This paper analyzes the clinical data of the first case of AIDS existing with cerebral sparganosis in our hospital and reviews the relevant literature. It discusses the clinical manifestations, imaging features, diagnosis, and treatment of this disease and provides a reference for the treatment of AIDS existing with parasitic encephalopathy (cerebral sparganosis). Cerebral sparganosis has been reported worldwide especially in Asian countries. To date, this is the first reported case of sparganosis in HIV/AIDS patient.

Case presentation

A 34-year-old newly diagnosed HIV Chinese woman from Guanxi province presented at our hospital with a history of recurrent seizures for 5 years. The initial diagnosis was epilepsy but she was later diagnosed with cerebral sparganosis at our facility after thorough history, examination and investigations. Considering her immune status, she was treated with high dose praziquantel instead of surgery with significant improvement. She also received Zidovudine + Lamivudine + Nevirapine as the first-line antiretroviral treatment due to its high permeability across the blood-brain barrier.

Conclusion

Clinicians should pay attention to this rare entity in cases of HIV/AIDS-related encephalopathies which can be easily misdiagnosed as toxoplasmosis or cerebral tuberculosis. Thorough epidemiological history, clinical presentations, serologic and radiological tests are essential to reach the correct diagnosis and rule out other infections. Although the standard treatment for cerebral sparganosis is surgery, we opted for pharmacological treatment in consideration of immunological complications.

This case calls for more studies for standard treatment in HIV patients with cerebral sparganosis and requires clinicians to consider this rare entity when dealing with HIV related encephalopathies.

Background

Schizocephala mansoni is scientifically named *Diagonus mansoni*. It belongs to the class of tapeworms, schizocephae and diagonus. It is generally parasitic in animals such as cats and dogs. *Diegonella mansoni* is widely distributed, and it is reported to be endemic in 25 provinces in China, most of which are Guangdong, Henan, Fujian, Hunan and Guanxi⁽¹⁾. Humans can become the second intermediate host, the renewal host, and the final host of *Diegonella mansoni*. The larvae of *Diegonella mansoni* are called sparganosis mansoni, which parasitize the human body and cause sparganosis. Humans can be infected by the topical dressing of raw frog meat, swallowing raw or undercooked frog meat, snake meat, tadpoles, drinking contaminated water, using raw frog as poultices, and direct invasion of water

metacercariae. Sparganosis can cause ocular sparganosis, subcutaneous sparganosis, oral and maxillofacial sparganosis, cerebral sparganosis, and visceral sparganosis. Among them, cerebral spinal sparganosis is the most harmful. ⁽¹⁾

We report the case of cerebral sparganosis in HIV/AIDS patient with an emphasis on misdiagnosis, its treatment and briefly review the literature.

1. Li, Song, Li, Xie, Zhu, "Sparganosis", e154-e155.

Case Presentation

A 34-years old married Chinese woman, from Liuyang City in Hunan Province presented at our facility on April 2, 2018, with a 5 years history of seizures of the left limb. She reported having the first incidence of paroxysmal left limb twitching in 2013 which lasted for almost a minute and was fully conscious. Since then, she had suffered recurrent attacks with no predictable periodicity. The condition gradually worsened and involved both limbs of the left side as well as the left side of the face. She denied any history of loss of consciousness, frothing at the mouth or numbness. She was admitted to a local hospital in Guanxi Province with a diagnosis of epilepsy and received on oral Kaplan and Topatil with no further occurrence of convulsions until March 2018 when she was hospitalized in Guangdong due to convulsions again. The diagnosis was right frontotemporal lobe and parieto-occipital lobe lesions, with differential diagnoses of cerebral parasitic disease and secondary epilepsy. She was planned for surgery which was cancelled after preoperative tests showed she was HIV positive.

The patient was healthy in the past, born and raised in Guanxi, married at an age of 19 in Liuyang, Hunan and she has been working in Guangzhou for a long time. She reported drinking untreated/ unboiled water in Guanxi which is a common practice for the locals but denied eating raw/undercooked seafood such as snakes and frogs or using their flesh as a poultice. There was no history of genetic diseases or similar medical history in the family. She denied any history of sexual intercourse out of marriage. Physical examination was unremarkable.

Laboratory examinations done: complete blood count was normal, ALT 57.9U/L, AST 39.8U/L, globulin 34.4g/L, IgE 1360.8IU/ml, IgG 23.92g/L, CD4 + T cell count 229/uL, CD4/CD8 ratio 0.6, HIV RNA load 36 800 copies/ml.

Renal function, blood sugar, blood lipids, myocardial enzymes, electrolytes, procalcitonin, erythrocyte sedimentation rate, blood coagulation profile and thyroid function tests were normal. HBsAg, HBsAb, HBeAg, HBeAb were negative. TB test, hepatitis C antibody and syphilis antibody were negative.

Abdominal ultrasound revealed a slightly strong echo of liver parenchyma, a left kidney cyst and other findings were normal. A chest CT scan showed normal findings. Cerebrospinal fluid examination: intracranial pressure 200mmH₂O, protein 337.2mg/L, chlorine 131.1mmol/L, glucose 5.08mmol/L, there was no red blood cells or white blood cells. Gram staining, acid-fast staining, ink staining and culture

were all negative, cerebrospinal fluid and blood sparganosis IgG antibody was positive (antibody kit was purchased from Shenzhen Kangbaide Biotechnology Co. Ltd.)

Head MRI showed flaky and patchy long T1 and long T2 signals in the right frontotemporal and parietal occipital lobe, the adjacent sulcus was enlarged, the cerebral gyrus was shallow and a short strip of T1 signal running along the sulcus in the deep right temporal lobe. After contrast administration, the lesion showed irregular lines, nodules, and ring-like enhancements. The largest nodule was about 6mm in diameter. The right tentorium and part of the pia mater showed linear and nodular enhancement with the ventricular dilatation (Fig. 1). The diagnoses following blood tests and MRI scans were: Cerebral sparganosis, HIV/AIDS with differential diagnoses of secondary epilepsy and space-occupying lesion.

She was admitted for treatment and after a neurosurgery consultation, we agreed on pharmacological treatment as the patient was not suitable for surgery. She received four cycles of high dose praziquantel therapy with each cycle one month apart. Each course contained high dose praziquantel of 25mg/kg tds for 10 days; she developed a headache during the course of treatment which was relieved with IV mannitol 20% 250mls bid and dexamethasone 10mg daily. After the first course of anthelmintic treatment, we started her on antiretroviral therapy with Zidovudine + Lamivudine + Nevirapine regimen.

MRI scans after the anthelmintic treatment showed that the range of the right frontotemporal lobe-shaped shadow was slightly smaller than before the treatment (Fig. 2). There was no seizure attack during and after the course of the treatment. At present, the patient has improved and discharged, and she is being closely followed up and continues with antiretroviral treatment. The patient is currently being followed up every three months and the prognosis needs to be further observed.

Discussion And Conclusions

In this case of sparganosis in HIV/AIDS patient, we found out that the pathogenesis of sparganosis in this immunocompromised patient was the same as in other reported cases of immunocompetent patients.

In this case, the patient had lived in two endemic areas in China; Guanxi province- her home town and Guangdong province where she used to work. The most probable source of infection could be drinking infested water as per epidemiologic history. Sometimes the epidemiological history is atypical ^(2,3).

Cerebral sparganosis is a diagnosis of exclusion and is misdiagnosed in most cases. This case wasn't an exception as it was first diagnosed and treated as secondary epilepsy for 5 years. Wang et al found up to 11 years of misdiagnosis. In their study, all 24 cases were first misdiagnosed as glioma, brain abscess, brain tuberculosis and primary epilepsy ⁽³⁾. In a review of 6 European cases, the misdiagnosis rate was 100%. They were diagnosed as neurocysticercosis, cerebral abscess or dysembryoplastic neuroepithelial tumour ⁽⁴⁾. The clinical manifestations of sparganosis are atypical and vary depending on the site of infection. Cerebral sparganosis commonly involves the frontal lobe. It can also invade other parts such as the parietal lobe, temporal lobe, occipital lobe, basal ganglia or cerebellum. The main clinical symptoms

are seizures, limb weakness, headache, or physical disturbances ^(2,3). In severe cases, it can lead to intracranial hypertension, visual impairment, consciousness disorder, or sudden death.

In terms of auxiliary examination, the patient's peripheral blood white blood cell count and eosinophils count were not elevated but IgE was elevated. Apart from the raised opening pressure and glucose, there was no obvious abnormality in the routine examination of cerebrospinal fluid. The reported cases of cerebral sparganosis in non-HIV patients have shown some variability in the cerebral spinal fluid analysis with most cases presenting with raised white cells and proteins while others having normal values. Peripheral blood and cerebrospinal fluid sparganosis antibody test has high specificity and sensitivity and is an important auxiliary diagnostic method ⁽³⁾. Combined with head imaging, it can confirm the diagnosis and improve the diagnosis rate. Studies have reported immunologic tests to have cross-reactivity with other cestodes but when combined with a thorough history and radiological imaging especially MRIs, it plays a significant diagnostic role.

In particular, enhanced MRI of the head has significant advantages in the diagnosis of the disease, it shows the following features: multiple small nodules in the brain with low signal on T1WI and high signal on T2WI, which can be a small ring, bead-like, orbital or tubular enhancement. There is a large area of cerebral oedema around the lesion. The larva can migrate across the lobes of the brain and across the midline, and brain atrophy occurs in the primary lobe after the migration ^(2,5-6). This patient had multiple intracranial lesions and obvious oedema bands around the lesions, which showed linear, nodular, and ring-enhancing lesions after enhancement, which is consistent with the literature.

Surgical removal is the first choice for the treatment of cerebral sparganosis. Image-guided stereotactic aspiration is currently an ideal surgical option as it is highly effective and less traumatic ⁽⁷⁾.

For patients whose lesions are located in major functional areas, those who don't want invasive treatment or whose general conditions cannot tolerate surgery, medical treatment needs to be considered. Medical treatment with high dose praziquantel has shown to be effective in treating cerebral sparganosis in inoperable patients ^(2,8). In this case, we opted for pharmacological therapy in consideration of the compromised immunity in HIV/AIDS patients and the high risk of the operator's occupational exposure. At present, there is no unified medical treatment standard guide. The definitive treatment of cerebral sparganosis with medical therapy is still controversial. Some authors have declared it as effective as surgical treatment while others have declared it ineffective. A study involving 96 patients showed no significant difference between long term high dose praziquantel therapy and surgical treatment ⁽⁹⁾. Some patients often require multiple cycles of treatment, and there are still some patients, who cannot be cured with medical treatment ⁽⁸⁾.

This patient received four cycles of high dose praziquantel 25mg/kg tds for 10 days with and each cycle was a month apart therapy with significant improvement on clinical and radiological features. There was no seizure after the completion of the first cycle up to date, the patient is still being followed up every three months when she attends her routine HIV clinic.

The incidence of cerebral sparganosis is low and AIDS combined with cerebral sparganosis is even rarer. There are currently no reports of similar cases. We searched the CNKI, Wan Fang, Web of Science and Pub Med database with "HIV/AIDS + cerebral sparganosis" as the keywords but there were no related articles.

The clinical characteristics of this patient are similar to those of non-AIDS patients with cerebral sparganosis, which may be due to the high CD4+T cell count. Due to the immunosuppression of AIDS patients, parasitic infections in low immunity patients may have different characteristics. It is not excluded that patients with AIDS and cerebral sparganosis with low CD4+ T cell counts have more atypical intracranial lesions and their clinical manifestations may be more complicated and insidious, the discovery of more cases of cerebral sparganosis in HIV/AIDS patients may confirm this further. There is currently no consistent standard for the timing and choice of antiretroviral treatment for patients with HIV/AIDS existing with sparganosis, and there are no clinically available data on the merits of each regimen. In this case, we chose the regimen of Zidovudine + Lamivudine + Nevirapine as the first line mainly based on its high permeability of the blood-brain barrier.

Although cerebral sparganosis with HIV/AIDS is rare with no reported cases at present, it should be suspected especially in patients from endemic areas with possible epidemiologic history and radiological features.

When deciding the mode of treatment in an immunocompromised patient these factors need to be considered; the ability of the patient to withstand surgery, the surgeon's occupational risk and the post-operative recovery. Although surgical removal of the sparganum is the first choice of treatment, high dose praziquantel therapy has proved to be effective in inoperable cases. Clinical progress and follow up radiological images are of great use to ensure the death of the worms in the brain. Since there is currently no uniform standard for deworming and antiviral treatment guidelines, this case calls for immediate action on the matter.

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9. Hong, Xie, Wan, An, Xu, Zhang, "Efficacy", 10.

List Of Abbreviations

CNKI- China National Knowledge Infrastructure.

AIDS- Acquired Immunodeficiency Syndrome.

HIV- Human Immunodeficiency Virus.

ALT- Alanine Amino Transferase.

AST- Aspartate Amino Transferase.

Ig E- An immunoglobulin E.

IgG- Immunoglobulin G.

CD4- cluster of differentiation 4.

CD8- cluster of differentiation 8.

RNA- Ribonucleic acid.

HBsAg - Hepatitis B surface antigen.

HBsAb- Hepatitis B surface antibody.

HBeAg - Hepatitis B e antigen.

HBcAb - Hepatitis B core antibody.

TB- Tuberculosis

Bid- bis in die

Tds- ter die sumendum

Declarations

Ethics approval and consent to participate

The approval was sought from the Ethics Committee of The First Hospital of Changsha.

Consent for publication

Written informed consent for publication of the clinical details and/or clinical images was obtained from the patient. A copy of the consent form is available for review by the Editor of this journal.

Availability of data and materials

The datasets generated and/or analysed during the current study are not publicly available due to the hospital privacy policy but are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

Funding

This work was supported by the Prevention and treatment of major infectious diseases such as AIDS and viral hepatitis during the 13th Five-Year Plan project [2018ZX10302104-001-002]

Authors' contributions

ZC and UT conceptualized the case, prepared the original draft and was a major contributor in writing the manuscript. CL participated in writing and editing all the drafts. MW provided immediate care and treatment to the patient and obtained the patient consent. GZ supervision and grant acquisition. NW performed all laboratory investigations and their interpretations. YL Participated in writing, reviewing and editing the manuscript. All authors read and approved the final manuscript.

Acknowledgements

We thank the management of the First Hospital of Changsha as well as all health workers who were involved in the treatment and care of this patient.

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Figures

Figure 1

(A, B and C): MRI findings of the patient's head before treatment. legend; A: The flair sequence shows a patchy high signal (pointed by the arrow). B and C: The lesions are irregular lines, nodules, and circular enhancements (pointed by arrows). Figure keys: →- An arrow used to mark the affected areas on the brain. R- Right hand side A- Anterior FA- Fractional anisotropy WW- Window width WL- Window level

Figure 2

The head MRI images of the patient after treatment. legend; A1: The flair sequence showed a patchy high signal (pointed by the arrow), and the lesion shadow range was slightly smaller than before treatment. B1-C1: After the enhancement, the lesion showed irregular lines, nodules, and circular enhancement (pointed by the arrow), and the range of the lesion was slightly smaller than before treatment. Figure keys: →- An arrow used to mark the affected areas on the brain. R- Right hand side A- Anterior FA- Fractional anisotropy WW- Window width WL- Window level