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NEUROSCHISTOSOMIASIS IN YOUNG FILIPINO PATIENT PRESENTING WITH SEIZURE

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ABSTRACT

Neuroschistosomiasis, a rare manifestation of schistosomiasis affecting the central nervous system (CNS), can result in severe neurological complications if not promptly diagnosed and treated. The condition arises due to the development of granulomas around eggs that become lodged within the CNS. Here we present a case report involving a young Filipino individual who had recurrent, uncontrolled seizures, ultimately attributed to neuroschistosomiasis. On magnetic resonance imaging revealed that the patient had brain lesions consistent with neuroschistosomiasis. Concurrently, serologic tests revealed high titres of anti-schistosoma antibodies. Effective treatment encompassed the administration of corticosteroids and praziquantel, leading to a favourable clinical outcome. This case underscores the necessity of considering neuroschistosomiasis in seizure patients, particularly in regions where the condition is prevalent, and highlights the diagnostic and therapeutic complexities posed by the absence of a biopsy.

KEYWORDS

Neuroschistosomiasis, schistosomiasis, seizures, flukes, praziquantel

LEARNING POINTS

- *Diagnostic challenge and insight:* This case highlights the importance of considering a broad differential diagnosis in patients with atypical neurological presentations, particularly in those from regions where parasitic infections are endemic. The internist plays a crucial role in integrating clinical, serological, and imaging data to arrive at a correct diagnosis, in this case, neuroschistosomiasis.
- *Tailored treatment strategy:* The case illustrates the value of personalized patient care. Initiating empirical treatment based on a combination of clinical suspicion, high-risk patient demographics, and imaging findings, without invasive procedures, demonstrates the internist's role in managing complex cases where patient preferences must be balanced with medical necessity.

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INTRODUCTION

Schistosomiasis, a parasitic disease caused by blood flukes belonging to the Schistosoma species, currently affects over 200 million individuals worldwide[1]. While neuroschistosomiasis is considered rare globally, it is not uncommon in endemic regions such as sub-Saharan Africa and parts of Asia, including the Philippines. Cerebral symptoms are observed in around 2 to 4% of individuals who are infected, commonly affecting the parietal, temporal, and occipital lobes, as well as the cerebellum[2]. It is a serious condition that can present with seizures, focal neurological deficits, cognitive impairments, and other neurological manifestations. Additionally, spinal involvement might manifest as low back pain, lower limb radicular pain, muscle weakness, sensory loss, and urinary incontinence. A definitive diagnosis can only be established through histological analysis revealing schistosoma eggs and granulomas[3]. In managing neuroschistosomiasis, early detection and prompt intervention are of paramount importance. Given its prevalence in certain regions, clinicians should maintain a high index of suspicion, especially when patients from endemic areas present with unexplained neurological symptoms. The patient in this case study, a young Filipino male, had likely exposure to schistosomiasis during his time as a freshwater fisherman in rural rice paddies, which are known environments for the transmission of Schistosoma species. The exposure likely occurred 5 to 6 years prior to presentation, although the patient did not recall specific encounters with contaminated water.

CASE DESCRIPTION

A 27-year-old Filipino man, a non-smoker with no history of alcohol abuse, had a history of recurrent seizures since February 2022, which were initially managed using phenytoin. He had recently moved from a rural part of the Philippines, where he worked as a freshwater fisherman in rice paddies, both potential sources of schistosomiasis exposure. His most likely exposure occurred 5 to 6 years before presentation, though he had no clear recollection of contact with contaminated water. In September 2022, he presented to us after experiencing three episodes of generalized tonic-clonic seizures.

Upon examination, a head computed tomography (CT) scan revealed multiple calcified brain lesions, prompting suspicions of neurocysticercosis. However, further investigation via magnetic resonance imaging (MRI) revealed ill-defined cortical lesions situated in the right parieto-occipital lobes. These lesions were accompanied by adjacent edema and curvilinear enhancement. Several potential causes were considered, including fungal infections, tuberculosis, and vasculitis. Extensive diagnostic workups were conducted, encompassing autoimmune, tuberculosis, human immunodeficiency virus (HIV), toxoplasma, and echinococcus investigations, all of which yielded negative outcomes. Additionally, routine lab results were checked for eosinophilia, a potential indicator of parasitic infection, although results were within normal limits. Additional CT scans of the chest, abdomen, and pelvis yielded unremarkable results. Consultation with a neurosurgeon prompted contemplation of MRI spectroscopy and a brain biopsy; however, the patient opted against the latter procedure. MRI head spectroscopy indicated interval progression of lesions, leaning towards infection or vasculitis. The leading differential diagnosis was CNS tuberculosis (TB). As a result, empirical treatment was initiated, involving anti-TB medications, prednisone, and albendazole for possible neurocysticercosis. Levetiracetam was also administered to manage the seizures.

After 3 months, a brain MRI demonstrated an increase in lesion size, prompting a re-evaluation of the necessity for a brain biopsy. The patient was readmitted for a scheduled biopsy in February 2023. However, the preoperative MRI surprisingly revealed a reduction in the size of lesions, leading to the cancellation of the biopsy. Unfortunately, in June 2023, the patient was readmitted due to new breakthrough seizures, despite adhering to the previously prescribed medications. These seizures initially manifested as focal seizures in his left arm and eventually evolved into generalized tonic-clonic seizures. Comparing CT scans of the patient's head at admission to a scan done

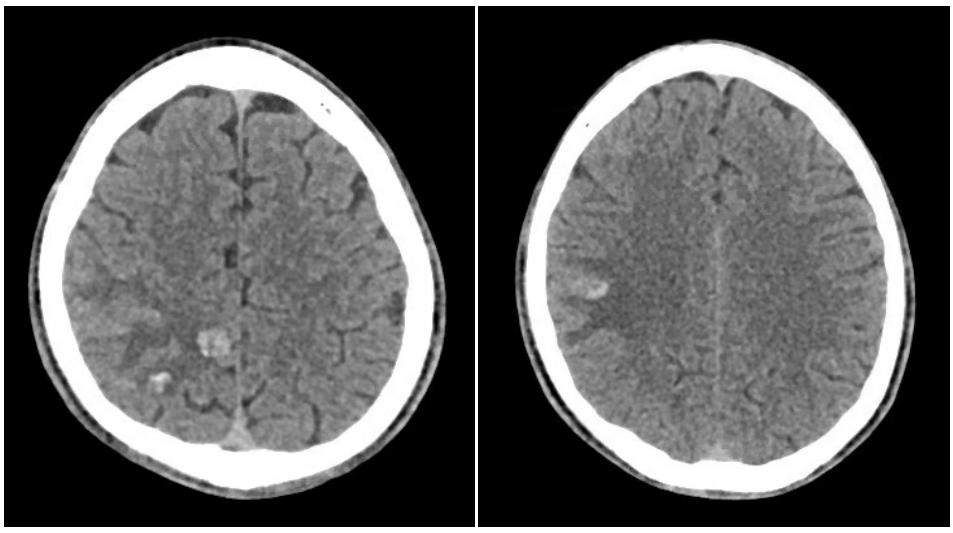

Figure 1. Computed tomography scan of the head (June 2023) showing multiple hyperdense lesions in the right temporoparietal and frontal regions.

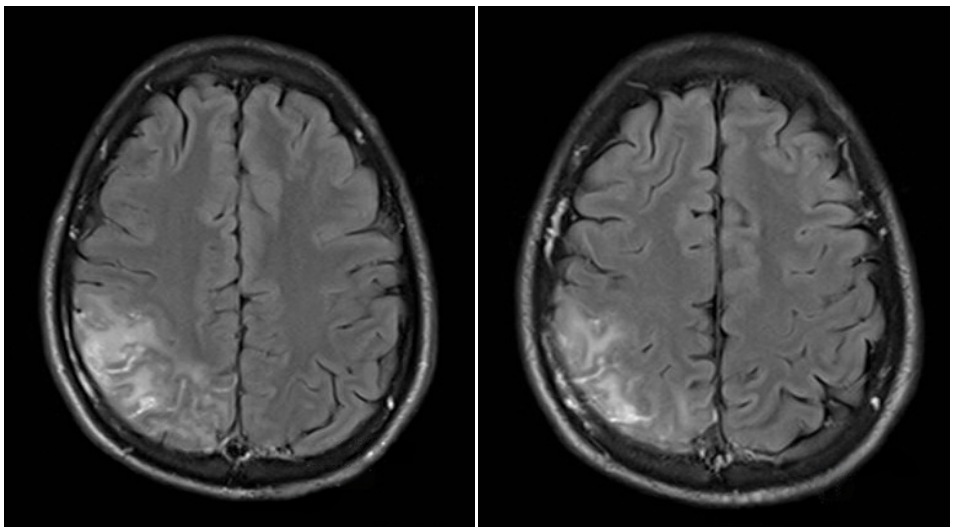

Figure 2. Magnetic resonance imaging of the brain (June 2023) showing right parietal essentially cortical gyriform pattern lesions with spotty nodular and curvilinear enhancement as well as adjacent focal leptomeningeal enhancement (pine tree pattern), with element of subcortical oedema.

in September 2022 showed that the number of lesions and perilesional edema had increased during the time between the two scans *(Fig. 1)*. However, CT alone played little role in the final diagnosis, which nearly always rests on other investigations. Meanwhile, MRI head findings strongly indicated a parasitic lesion, specifically suggesting schistosomiasis *(Fig. 2)*. Subsequently, a schistosoma titer was requested and returned notably high (1/160). Based on this information, the patient's presentation was attributed to neuroschistosomiasis, leading to the initiation of treatment with prednisone and praziquantel.

DISCUSSION

Schistosoma infection, also known as schistosomiasis or bilharzia, is a highly prevalent parasitic disease with global distribution, posing significant public health challenges, especially in tropical regions[4], such as the Philippine, the documented country of origin of our patient. Neuroschistosomiasis is a rare form of this disease that involves the CNS. It occurs when schistosomal eggs, instead of being excreted in the feces or urine, migrate to the CNS, and once there, the eggs release proteolytic enzymes that trigger eosinophilic inflammation and granulomatous reactions, ultimately leading to tissue replacement by fibrous scar tissue[5]. The vast majority of documented neuroschistosomiasis cases result from infections with *Schistosoma mansoni*, *Schistosoma haematobium*, or *Schistosoma japonicum* parasites[2]. The diagnosis of neuroschistosomiasis can be challenging. Despite being the gold standard diagnostic test, a tissue biopsy showing parasitic ova is invasive and carries a considerable surgical risk. So other diagnostic factors can also be additionally taken into consideration. The approach should rely on a comprehensive patient history that investigates symptoms, demographics, and potential sources of infection. Detecting parasite eggs in urine or stool is only achievable in 40-50% of cases[6]. Serological testing is a sensitive diagnostic method; however, due to its high false-positive rates, and the inability to distinguish old from acute infections, it is sometimes regarded as less reliable. Nevertheless, positive test results with high titers (>1:160), are considered meaningful and indicative of the presence of the infection[7,8]. A positive serological result detected in blood and/or cerebrospinal fluid could prompt a therapeutic trial for patients exhibiting consistent brain lesions[9].

Regarding neuroimaging, MRI is considered the best imaging modality, and several distinct post-contrast features have been documented. In our case, the brain MRI showed

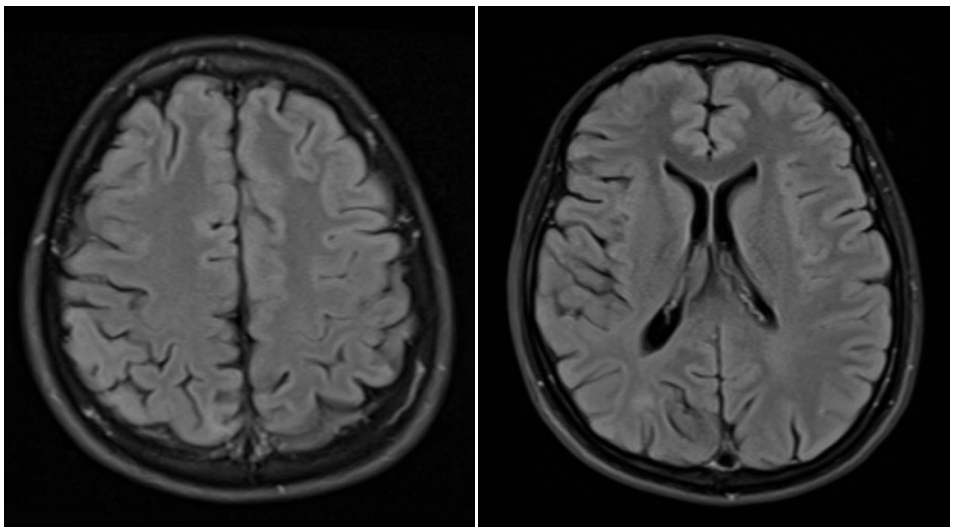

Figure 3. Magnetic resonance imaging of the brain done 6 months after anti schistosomal therapy given, showing nearly complete resolution of the previous lesions.

right cortico-parietal gyriform pattern lesions with spotty nodular and curvilinear enhancement, as well as adjacent focal leptomeningeal enhancement *(Fig. 2)*. These findings were suggestive of a parasitic infection, in particular schistosomiasis.

Our patient declined to undergo a brain lesion biopsy due to the invasiveness and substantial surgical risks associated with the procedure. So, we decided to start treatment for neuroschistosomiasis because the patient had recently lived in the Philippines, which is an area where the disease is common, had a positive serological result showing high titers, and had brain MRI findings that were suggestive. This course of action was chosen considering the potential significant neurological complications that can arise if the condition remains untreated.

Steroids (prednisone 1 to 2 mg/kg per day) and antiparasitic therapy (specifically praziquantel 40 mg/kg single dose) are the two main components of the treatment for neuroschistosomiasis. Praziquantel should be given a few days after the start of corticosteroid treatment since it can cause an inflammatory response that paradoxically worsens neurologic symptoms. The optimal duration of corticosteroid treatment remains uncertain; typically, therapy should extend over several months but may be gradually reduced based on individual considerations. Even in cases where the diagnosis of neuroschistosomiasis is suspected but not confirmed, corticosteroid therapy ought to be implemented. Antiepileptic drugs, such as levetiracetam, sodium valproate, or carbamazepine, are typically needed when patients present with seizures, aiming for reduced interaction with praziquantel. However, there are no established guidelines for the minimum duration of this treatment, and extended antiepileptic drug therapy for patients with cerebral neuroschistosomiasis is uncommonly warranted[10].

In this case, we initiated treatment by administering an initial dose of 50 mg of prednisone, followed by praziquantel 2 days later. The patient exhibited consistent improvement without experiencing new seizures or notable clinical alterations. Consequently, he was discharged with a treatment strategy involving high-dose steroids for a period of 2 weeks, followed by a gradual tapering regimen spanning 4 to 6 weeks. After 4 weeks, praziquantel administration was repeated before the tapering of steroid treatment. A follow-up appointment was scheduled with both neurology and infectious disease clinics 1 month and 6 months post-discharge, at which the patient's condition was asymptomatic, without headaches, seizures, or any focal neurological deficits. As the patient refused to go for brain biopsy initially, we decided to repeat MRI brain after 6 months of starting therapy, in order to monitor the MRI findings progression after anti schistosomal treatment was given, as this will further confirm our initial diagnosis. The MRI brain was done, and it showed interval significant resolution of the right parietal lesions, with no appreciable enhancement on post contrast images *(Fig. 3)*. The overall picture was suggestive of significant resolution of the disease pattern.

CONCLUSION

This case underscores the necessity of considering neuroschistosomiasis in seizure patients, particularly in regions where the condition is prevalent, and highlights the diagnostic and therapeutic complexities posed by the absence of a biopsy.

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