
ISSN 2622-7258

DOI: 10.31014/aior.1994.05.03.228

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Leptomeningeal Schistosomiasis: A Case Report of an Atypical Location of Neuroschistosomiasis Presenting as Adult-Onset Seizure

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Abstract
We report a case of a 19-year-old male, single, right-handed, student, Filipino currently living in Novaliches, Quezon City who consulted for the first time at our institution due to stiffening of extremities with a pertinent travel history from the Island of Samar. Evaluation and diagnostics showed a leptomeningeal enhancement and thereafter, a biopsy was made revealing deposition of schistosoma ova at the leptomeningeal area. Schistosomiasis (also known as Bilharzia or Blood Fluke Disease) is widely distributed in the Philippines affecting 24 provinces in Luzon, Visayas, and Mindanao, with 5 million people at risk and approximately 1 million affected in the year 2003. Cerebral schistosomiasis is a severe and neglected complication which occurs in less than 5% of infected individuals. Symptoms are non-specific such as headache, vomiting, confusional states, and focal seizures. Typical neuroimaging findings are expected at the spinal cord, cerebellum, and the subcortical area, however leptomeningeal involvement is rarely reported. We report this case to the medical community to give light on the different presentations of the said disease

Keywords: Schistosomiasis, Neuroschistosomiasis, Leptomeninges, Focal Seizure

1. Introduction
Schistosomiasis (also known as Bilharzia or Blood Fluke Disease) is widely distributed in the Philippines affecting 24 provinces in Luzon, Visayas, and Mindanao affecting 5 million people at risk, with approximately 1 million affected in the year 2003 (Olveda et al., 2014). Infection is transmitted by snails living in fresh water such as lakes, rivers, streams and ponds. Initial symptoms usually appear within days or weeks after cercarial penetration which migrates to the lungs then to the target organs via the vascular system. Symptoms include a skin rash, fever, headache, muscle ache, bloody diarrhea, cough, malaise, and abdominal pain in the early stages of the disease. If untreated, schistosomiasis can become a chronic illness as the flatworm eggs damage the lining or the main parenchyma of the target organ (Imai et al., 2011).
Figure 1: shows the distribution of Schistosomes in the Philippines

This type of parasitic infection is composed of several species with 5 of them causing significant disease entities. According to Olveda et al. (2014), there are three species are present in the Philippines namely S. japonicum, S. haematobium, and S. mansoni to which each species has a predilection to affect an organ system particularly the integumentary, urogenital, gastrointestinal, and hepatobiliary system. Although there are some cases in which the parasite may involve an “atypical location” so called ectopic schistosomiasis which affects the lung, heart, and in some cases the central nervous system.

Neuroschistosomiasis is a relatively uncommon disease with the risk of the infection depending on the level of immunity and the magnitude of the schistosomal invasion. About <5% of patients will develop neuroschistosomiasis where spinal and cerebellar complications are prevalent. This was further elaborated by Amaral and Andrade (2015) as S. japonicum infection has a predilection for cortex, subcortex, basal ganglia or internal capsule. The reason for this predilection is explained two different mechanisms. The first is by egg embolism and second is by worm migration through valveless veins as cited by the study of Wu et al. (2011) to which the study involved 11 patients from the Rennin Hospital, Wuhan University in China. Eggs from the portal system could embolize to the brain along the vertebral venous or Batsons’ plexus, via atrial/septal defects or patent foramen ovale, or via pulmonary venous shunts, as a result of hepatic and pulmonary hypertension. The size and shape of the eggs make it favorable in their migration to the brain especially in S. japonicum explaining it’s cerebral predilection. In contrast with S. mansoni and S. haematobium which have larger eggs and prominent spines. In relation to the second mechanism, adult flukes could enter the Batson plexus and then migrate upwards to the brain and deposit their eggs directly. Both theories explain the most common pathologies such as transverse myelitis and a cerebellar mass (George et al., 2009). In transverse myelitis, the most prevalent clinical manifestations as studied by Amaral and Andrade (2015) were weakness in the lower limbs (94%), back pain (84%), bladder dysfunction (75%) and impotence (80%). The most common site of injury was the thoracolumbar junction (65%) and 73% of parasitological stool examinations were negative.

Several neuroimaging techniques such as computed tomography scan (CT Scan) and magnetic resonance imaging (MRI) reveal certain patters in the diagnosis of neuroschistosomiasis. CT scans showed irregular low-dense and isodense lesions that were mixed in the focus area, as mixed boundary was not very clear. There are also reports of having multiple calcified areas at the subcortical area associated with edema and manifestations of the “glove sign” with obvious mass effect. Typical MRI findings showed a scattered gyrus-like, nodular, or irregular-shaped spots of strengthening (Wu et al., 2011). However due to the variation of clinical manifestations, a high index of suspicion is still warranted.
In our case, we describe a case of cerebral schistosomiasis presenting as a new onset seizure and a solitary mass-like lesion affecting the meninges of the brain.

2. Case Report

A case of a 19-year-old Filipino male, single, right-handed, unemployed, from Novaliches, Quezon City who sought consult for the first time at our institution on August 3, 2020 due to stiffening of extremities. He is a grade 9 student with a total of 8 formal years of schooling. The patient was born and raised in Catbalogan City, Samar Island for 13 years where they lived near rice-fields. He had no comorbidities with no history of trauma and surgery. He is a nonsmoker, non-alcoholic beverage drinker and denies illicit drug use. Review of the family history showed only hypertension on father side.

The patient was apparently well until the 2nd week of October 2019, when he started to experience headache, located at the right hemicranium, which was dull in character, graded 3-4/10 on PAS, non-radiating, occurring at no particular time of the day. After 1 month the patient was seen having seizures described as stiffening of the left extremities associated with head veering to the left, and loss of consciousness which lasted for 2-3 minutes which occurred for three times. The patient also had auditory and visual hallucinations prior to the seizure episode. He was initially brought to a nearby hospital which requested for neuroimaging and was initially given Levetiracetam 500mg tablet twice a day to which the patient was compliant. He was eventually sent home. In the interim the patient had recurrence of the seizures with the same semiology for about 2-3 times per month, from which he tolerated his condition. However, on the 1st week of August 2020 the patient had 6 episodes of seizure associated with vomiting hence consulted at our institution.

Upon consultation, the vital signs, general physical examination, and review of systems were unremarkable. Neurologic examination revealed that the patient was awake, oriented to time, place and person. He could follow commands. Cranial nerve examination noted no anosmia, 3mm equally briskly reactive to light pupillary response, no visual field cuts, primary gaze at the midline, full extraocular muscle movement, funduscopic exam was unremarkable, intact gross hearing, no facial asymmetry, uvula and tongue at midline. On manual motor testing, patient scored 5/5 on all extremities, with no sensory deficit and normoreflexive on all extremities. No Babinski was noted, no nuchal rigidity, no apraxia, no aphasia, no frontal release signs, no parietal signs.

Basic laboratory examinations (Appendix A) such as complete blood count, electrolytes and coagulation studies were normal. Other ancillaries such as Random blood sugar, Creatinine were normal. Chest Xray and 12-Lead ECG were normal (Appendix A).

Figure 1: The non-contrast CT scan on axial view showing finger-like hypodensities at the right temporal lobe with no midline shift nor hydrocephalus. The cranial vault was intact.
The cranial MRI done 6 months after the 1st imaging with gadolinium contrast shows areas of irregular hyperintensities likely corresponding to vasogenic edema on T2 segment (A – Axial, B – Coronal).

The cranial MRI with gadolinium contrast shows a serpentine-like enhancement at the right temporal lobe with the corresponding MRS ratio.

The patient was admitted and was initially managed as a case of adult-onset seizure probably secondary to an intracranial mass. The patient was started on Levetiracetam 500mg tab, 1 tablet twice a day for the control of seizures. Dexamethosone 4mg tablet, 1 tablet twice a day was also started for the vasogenic edema of the patient. Patient was admitted for 21 days with no neurologic nor behavioral changes. Patient was then subjected to excision of tumor and histopathologic studies were done which revealed a Cerebral Schistosomiasis at the subarachoid space and Virchow spaces overlying the right temporal lobe. He was given Praziquantel 60mg per kilogram as loading dose, a total of 2350 mg was loaded. Patient was observed for adverse drug reactions and was eventually discharged. Follow-up done after 1 month post-surgery with no recurrence of seizures and no focal neurologic deficits. Patient was able to go back to school.

3. Discussion

Focal epilepsy due to neuroschistosomiasis in the Philippines has been estimated to be from 2% to 5% among infected individuals. Diagnosing cerebral schistosomiasis can be difficult, since neurological symptoms and radiological findings are nonspecific. In some reported cases of neuroschistosomiasis, brain tumors, such as meningioma and glioma, had been suspected initially. Moreover, as in the present case, patients with neuroschistosomiasis may have no clinical evidence of systemic disease such as loose stools nor blood tinged urine. Stool/Urine examinations only reveal up to 40-50% of neuroschistosomiasis especially if the patient had the exposure a few years prior.

Our case demonstrates one of the neglected diseases in the Philippines. The patient had no history of ‘cercarial dermatitis’ nor the common prodromes of the said disease, possibly due poor health seeking behavior or perhaps
because it went unrecognized. Unremarkable examination and laboratory findings (on blood and stool exam) added to the diagnostic difficulty. With the benefit of histological diagnosis, the study suggests that the serpiginous enhancements seen on MRI represent focal zones of intense tissue reaction to eggs, leading to chronic pathology involving the vasculature. The treatment of cerebral schistosomiasis is highly effective and safe. Praziquantel eradicates the adult worms and concomitant corticosteroids reduce the granulomatous inflammation and are used for all schistosomal subtypes. However, the effect on chronic granulomatous disease and ova remains unknown. For this case, a high index of suspicion and a good clinical history revealing the pertinent travel history to endemic areas may help with the diagnosis and proper treatment for the patient.

4. Conclusion

This is a case of a 19-year-old male who presented with adult onset seizure with a history of travel to the province of Samar, with no known co-morbidities, no family history of seizure. Further laboratory work-up was inconclusive. Neuroimaging with MRS guidance revealed non-specific findings. We report this case as one of the neglected diseases in our country. Making the diagnosis is challenging, as manifestations may be protean and presentation with neurological complications can occur several years after the initial infection. Therefore, we recommend a high index of suspicion and an aggressive diagnostic approach in comparable cases.

Ethical Consideration
Patient form was secured before submission of manuscript

Financial Support and Sponsorship
None

Conflicts of Interest
There are no conflicts of interest.

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References
### Appendix A

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<td>ECG initial</td>
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Appendix B – BIOPSY SLIDES

Scanner View

MENINGES

CEREBRAL CORTEX

Scanner View
APPENDIX B – BIOPSY SLIDES

Low-power View

Low-power View
APPENDIX B – BOPSY SLIDES

High-power View

[Image of high-power view of tissue sample with labeled lateral spine]

High-power View

[Image of high-power view of tissue sample with labeled lateral spine]