Case Report

Neurocysticercosis in a 14-Year-Old Nigerian: A Case Report and Review of the Literature

Soyemi SS, Akinbo CO[1], Mgbehoma AI, Emiogun FE, Adegboyega OO[2], Obafunwa JO

Departments of Pathology and Forensic Medicine and [2]Radiology, [1]Division of Neurosurgery, Lagos State University Teaching Hospital, Lagos, Nigeria

Abstract

Neurocysticercosis (NCC), an infection of the brain by the larval form of *Taenia solium* tapeworm, has been described as the most common cause of acquired adult seizure and epilepsy in regions of the world where the worm infestation is endemic. It is acquired following ingestion of raw or partially cooked pork-containing cysts of *T. solium*. We report the case of a 14-year-old male patient, who presented with recurrent tonic–clonic partial seizures of 3 months duration, seizures, and progressive weakness of the left limb, while being fully conscious. He had a left facioparesis of the upper motor neuron type with plastic left hemiparesis. An impression of a right frontal cortical lesion was made. The laboratory investigations as well as the chest radiograph were reported as being essentially unremarkable. The magnetic resonance image of the brain revealed a circumscribed right parietal lesion which was circumscribed. A provisional diagnosis of intracranial tumor was then made. Surgery involved an *en bloc* resection of a cystic right parietal mass. The gelatinous fluid contained within was submitted for cytological examination. The latter revealed the presence of a cysticercus comprising the scolex, spiral canal, and the bladder wall. A diagnosis of NCC was made, and the patient was thereafter commenced on antihelminthics. The patient made an appreciable recovery thereafter. This report is to emphasize the continued existence of this disease, the need to improve diagnostic suspicion, and skill. This report further supports the proposal to declare NCC an international reportable disease.

Keywords: Diagnostic skills, neurocysticercosis, paresis, seizures, suspicion index

Received on: 23-02-20 **Review completed on:** 03-05-20 **Accepted on:** 25-05-20 **Published on:** 08-08-20

Introduction

Neurocysticercosis (NCC) is an infection of the brain by the larval form of *Taenia solium* tapeworm and has been described as the most common cause of acquired adult seizure and epilepsy, in parts of the world where this worm infestation is endemic.[1] It is acquired following ingestion of raw or partially cooked pork-containing cysts of *T. solium*. The gastric enzymes in the stomach break the eggs, liberating the enclosed larvae known as oncospheres. The latter penetrate the intestinal mucosa, enter the blood stream, and are then transported to all parts of the body. They are mostly deposited in the brain, muscle, and the subcutaneous tissue, where they eventually develop into cysts; serious manifestation of the disease usually follows lodgment of the cysts in the brain, spinal cord, or the eye.[2-4]

In the tissues, the oncospheres develop into metacestodes, and following the multiple stages of development, they become cysticerci. A membrane develops around each oncospheres with a vesicle-containing clear fluid and the parasite head/scolex. This first stage is called vesicular or viable stage. The cyst may remain in this stage for months or years depending on the immune response of the environment. The colloidal stage indicates transition, when the scolex deteriorates and the previously clear fluid turns turbid. In the granular-colloidal stage, the fluid becomes gradually more opaque, the parasite dies, and a dystrophic calcification ensues resulting in the formation of calcified nodules.[5,6]

Address for correspondence: Dr. Soyemi SS, Department of Pathology and Forensic Medicine, Lagos State University Teaching Hospital, 1-5 Oba Akinjobi Way, Ikeja, Lagos, Nigeria.
E-mail: sunday.soyemi@lasucom.edu.ng

Access this article online

Quick Response Code:

Website: www.atpjournal.org

DOI: 10.4103/atp.atp_12_20

How to cite this article: Soyemi SS, Akinbo CO, Mgbehoma AI, Emiogun FE, Adegboyega OO, Obafunwa JO. Neurocysticercosis in a 14-year-old Nigerian: A case report and review of the literature. Ann Trop Pathol 2020;11:72-5.

The clinical manifestations of NCC are nonspecific, and these include seizure, headache, and focal neurological deficits. These are essentially dependent on the locus of ultimate deposition in the brain. The use of high-resolution magnetic imaging (MRI) is helpful in identifying the cystic contents. Similarly, high-nuclear magnetic resonance (MR) spectroscopy, diffusion-weighted imaging, and MR perfusion imaging can all be of immense advantage. While their findings are not completely pathognomonic, it requires an experienced radiologist or imaging expert to recognize the peculiarities suggestive of NCC. Serologic test such as ELISA has very low sensitivity and specificity. A newer method known as enzyme-linked immunotransfer blot (EITB) has very good specificity and sensitivity, but it is expensive and not commercially available.[7-9] For these reasons, the diagnosis of NCC still poses a challenge to the clinician.

Case Report

We report the case of a 14-year-old right-handed male patient who presented with recurrent tonic–clonic partial seizures of 3 months duration. The seizures involved the left hemibody. The patient developed progressive weakness of the left limbs while being fully conscious, and there was left facioparesis of the upper motor neuron type with a plastic left hemiparesis. The findings in other systems were essentially normal. The patient had never lived in an environment where the pig is reared. He also claimed that he does not eat pork meat and lived with no domestic pet. A provisional diagnosis of right frontal cortical lesion was thereafter made.

The magnetic resonance image (MRI) of the brain revealed a right parietal lesion which was circumscribed [Figure 1]. It was hypointense in T1-weighted image (T1-WI) and fluid attenuated inversion recovery (FLAIR) sequences with moderate pressure effects. The lesion was hyperintense in the T2-WI and had perilesional hazy hyperintense signals in both FLAIR and T2WI sequences. The brain-lesion demarcation was poorly defined, and there was no contrast enhancement. There were no demonstrable lesions at the basal cisterns and the ventricles. The laboratory investigations as well as the chest radiograph were essentially unremarkable. An impression of an intracranial tumor was then made, and the neurosurgery team slated the patient for a surgical excision.

At surgery, a right parietal craniotomy was done, and an *en bloc* resection of a right parietal cystic mass was done. The cyst contained a gelatinous clear fluid, and this was submitted for cytological examination.

Cytological examination of the fluid revealed an essentially acellular smear. However, the cell block preparations from the fluid showed a cysticercus comprising the scolex, the spiral canal, and the bladder wall [Figures 2-4]. Higher magnification reveals the invagination of the larval form. Against this background, histopathological diagnosis of NCC was made. The patient was thereafter commenced on albendazole 300 mg twice daily for 30 days and subsequently made appreciable recovery.

The zeal to report this case is premised on three reasons: first – there is the possibility of underdiagnosis in endemic areas; second – extremely few cases appear in the literature in Africa; and third – the proposal to declare NCC an international reportable disease deserves some support.

Discussion

Despite the wide clinical arrays and heterogeneity of symptoms in NCC, all studies reported that seizures are the most common symptoms occurring in 60%–90% of patients.[10] In our case, tonic–clonic seizures and progressive body weakness were the presenting symptoms in this 14-year-old patient. Recent reports have shown that children are more likely to have NCC-related seizures than adults.[11]

Inflammation around the parasites is the main phenomenon triggering seizure in parenchymal lesions. This is commonly seen in degenerating cysts and calcified nodules.[12] In extra-parenchymal lesions, inflammation of the cerebrospinal fluid (CSF) space is usually severe with resultant arachnoiditis, vasculitis, ependymitis, and entrapment of cranial nerve which frequently requires ventriculoperitoneal shunt.[13] The

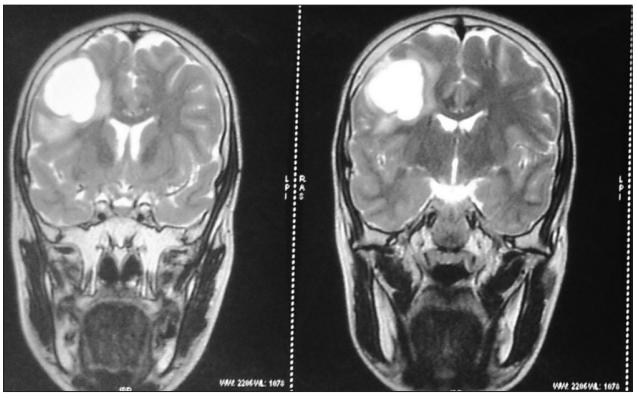

Figure 1: Magnetic resonance image (MRI) of the brain revealing a circumscribed right parietal lesion. The lesion was hypointense in T1 weighted image

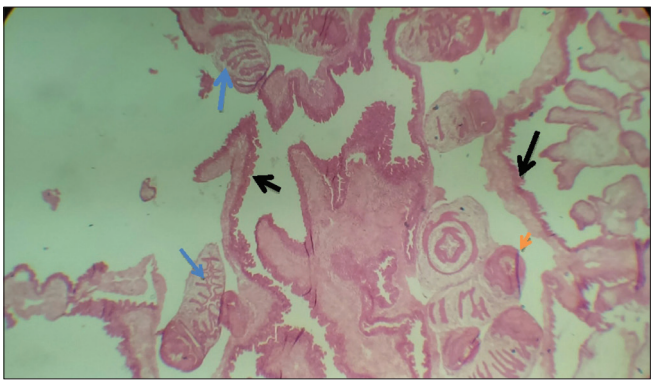

Figure 2: Scanning magnification image of a cell block showing cysticercus, comprising of the scolex (orange arrow), spiral canal (blue arrows), and bladder wall (black arrows) (H and E, ×40)

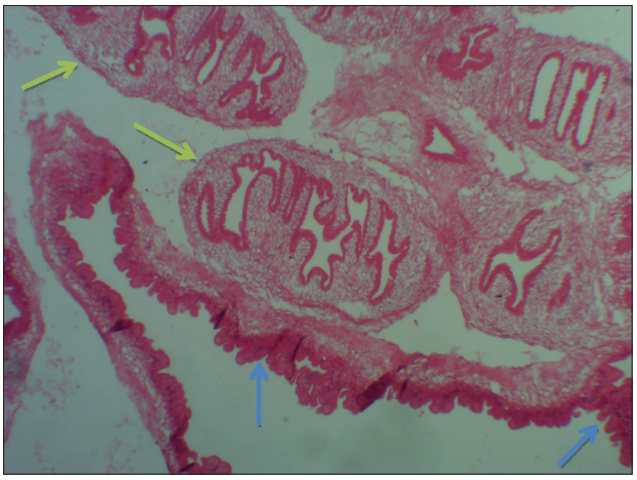

Figure 3: Photograph of the undulating bladder wall lined by a monolayer of rare nuclei (blue arrows), and larva forms, composed of duct-like invaginations (spiral canal) (yellow arrows) (H and E, ×100)

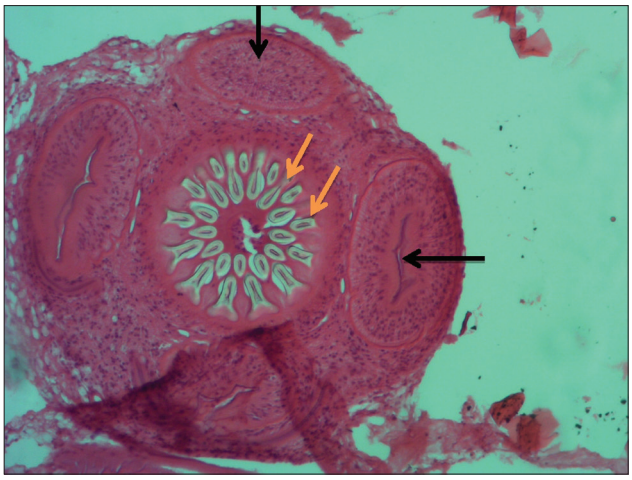

Figure 4: The scolex is seen consisting of a central rostellum, surrounded by two rows of 13 refractile hooklets (orange arrows) and 4 suckers (black arrows) (H and E, ×400)

inflammation in the CSF space is evident by pleocytosis, increased level of IgG subclasses, proteins, and interleukins 5, 6 and 19, as well as, eosinophils.[14] There was nothing to suggest cranial nerve entrapment or hydrocephalus in this patient. These negative findings further give credence to the fact that the lesion in this case was parenchymal.

Computerized tomography and MRI are all useful in establishing a diagnosis of NCC, identification of the developmental phase of the parasites, as well as, location of the lesion. It further allows the identification of any of the four morphological phases of disease, namely vesicular, colloidal, granular nodular, and the calcified nodule.[4] MR imaging is more sensitive than computed tomography in identifying the scolex and diagnosing parenchymal NCC.[11] Imaging studies are useful tools for making the diagnosis in experienced hands, particularly in the endemic regions. The diagnosis in the present case was, however, made from the cell block preparation of the cytology fluid sent to the anatomic pathology laboratory; the identification of the scolex comprising hooklets and the suckers confirmed the diagnosis [Figures 2-4]. Radiologists, therefore, need to have a high suspicion index and suspect the diagnosis when certain diagnostic features are observed. The extraparenchymal lesion (subarachnoid and ventricular) is, however, difficult to detect by imaging since attenuation and signal intensity of the cyst content is similar to that of the CSF; this is in addition to the poor enhancement after contrast administration.[7] In the present case, neither the scolex nor the cyst wall was recognized on MRI; what was recognized was the circumscribed right parietal lesion which was hypointense in T1WI.

Immunologic diagnosis of NCC requires serum, which is relatively easier to obtain than CSF. The EITB test performs better with serum than with CSF and is presently the best serological test for establishing the diagnosis of NCC. It has a sensitivity and specificity of 98% and 100%, respectively, in patients with more than one live cyst or subarachnoid disease.[15] Since there is the high prevalence of each condition in endemic countries, a causal as well as fortuitous relationship between the two pathologies might exist.[16] It is, however, worthy of note that EITB is expensive and not commercially available.[8] The commonly used ELISA test for the detection of antibodies is available but not recommended due to its low specificity and sensitivity.[9] Since there was no clinical suspicion of this lesion, ELISA test was not requested for in this case.

The treatment of NCC is usually complex and should be individualized, depending on the location and viability of the parasites. The intuitive approach is to kill the parasite using available antihelminthic drugs such as praziquantel or albendazole.[16] Mannitol is used to reduce the raised intracranial pressure; analgesics are prescribed for headache,[10] while steroids are used based on the assumption that they reduce edema and inflammation around parenchymal cysts.[3]

Cysticercosis is known to be endemic in many developing countries including Nigeria, where Ahidjo *et al*. reported a case of nuerocysticercosis with additional temporal bone changes in a-32 year-old Nigerian.[17] Similar cases had been reported in Central and South America, East, West and Central Africa, and Asia,[2,4,7,9,18,19] with recognized pig-to-pig transmission and a human-to-human transmission through the fecal-oral route to human. It is noteworthy that the authors were not able to find any literature report of cerebral cysticercosis from Nigeria. This said that it is pertinent to note that NCC is often encountered in developed countries as a result of migration.[20-23] It is for this reason that epidemiologists must remain vigilant to control the spread of this disease.

Conclusion

The present authors advocate that infestations by this parasite should be borne in mind when the clinicians are confronted with patients presenting as in the present instance. Specialists, particularly those in diagnostic medicine need to develop a high

suspicion index for diseases endemic in their region of practice. The zeal and willingness to report this case are further premised on the possibility of continued underdiagnoses, and the need to support the proposal to declare NCC an international reportable disease.[24] The latter proposal should help to prevent further spread to developed countries.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Commission on tropical diseases of the international league against epilepsy. Epilepsia 1994;35:89-93.
2. Gonzalez AE, Lopez-Urbina T, Tsang B, Gavidia C, Garcia HH, Silva ME, *et al*. Cysticercosis working group in Peru. Transmission dynamics of T solium and potential for pig-pig transmission. Parasitol Int 2006;55:131-5.
3. Nash ET, Pretell EJ, Lescano AG. Bustos JA, Gilman RH, Gonzalez AE. Perilesional brain edema and seizure activity in patients with calcified neurocysticercosis. Lancet 2008;7:1099-105.
4. Garcia HH, Del Brutto OH. Cysticercosis working group in Peru. Neurocysticercosis: Updated concepts about old disease. Lancet 2005;4:653-61.
5. Gripper LB, Welburn SC. Neurocysticercosis infection and disease – A review. Acta Tropica 2017;166:218-24.
6. Coral-Almeida M, Gabriël S, Abatih EN, Praet N, Benitez W, Dorny P. Taenia solium human cysticercosis: A systematic review of sero-epidemiological data from endemic zones around the world. PLoS Negl Trop Dis 2015;9:e0003919.
7. Del Brutto OH. Neurocysticercosis. A review. Sci World J 2012;2012:1-8. [doi: 10.1100/2012/159821].
8. Tsang VC, Brand JA, Boyer AE. An enzyme linked immunotransfer blot assay and glycoprotein antigens for diagnosing human Cysticercosis (*Taenia solium*). J Infect Dis 1989;159:50-9.
9. Mohan VR, Jayaraman T, Babu P, Dorny P, Vercruysse J, Rajshekhar V. Prevalence and risk factors for *Taenia solium* taeniasis in Kaniyambadi block, Tamil Nadu, South India. Indian J Public Health 2014;58:202-3.
10. Capio A. Neurocysticercosis: An update. Lancet Infect Dis 2002;2:751-62.
11. Kelvin EA, Carpio A, Bagiella E, Leslie D, Leon P, Andrews H, *et al.* The association of host age and gender with inflammation around neurocysticercosis cysts. Ann Trop Med Parasitol 2009;103:487-99.
12. Gupta RK, Awasthi R, Rathore RK, Verma A, Sahoo P, Paliwal VK, *et al.* Understanding epileptogenesis in calcified neurocysticercosis with perfusion MRI. Neurology 2012;78:618-25.
13. Fleury A, Carrillo-Mezo R, Flisser A, Sciutto E, Corona T. Subarachnoid basal neurocysticercosis: A focus on the most severe form of the disease. Expert Rev Anti Infect Ther 2011;9:123-33.
14. Adalid-Peralta L, Fleury A, Garcia-Ibarra TM, Hernández M, Parkhouse M, Crispín JC, *et al*. Human neurocysticercosis: *In vivo* expansion of the regulatory T cells and their requirements in the central nervous system. J Parasitol 2012;98:142-8.
15. Rajshekhar V. Neurocysticercosis: Diagnostic problems and current therapeutic strategies. Indian J Med Res 2016;144:319-26.
16. Carpio A, Fleury A, Hauser WA. Neurocysticercosis: Five new things. Neurol Clin Pract 2013;3:118-25.
17. Ahidjo A, Mustapha Z, Moghalu V, Pindiga UH, Ngadda HA, Mayun A, *et al*. Neurocysticercosis -Temporal bone changes on Computed Tomography- a case report. West AfricalJournal of Radiology 2009;16:35-9.
18. Onah DN, Chiejina SN. Taenia solium cysticercosis and human taeniasis in the Nsukka area of Enugu State, Nigeria. Ann Trop Med Parasitol 1995;89:399-407.
19. Weka RP, Ikeh EI, Kamani J. Seroprevalence of antibodies (IgG) to *Taenia solium* among pig rearers and associated risk factors in Jos metropolis, Nigeria. J Infect Dev Countries 2013;7:67-72.
20. McCormick GF. Cysticercosis: Review of 230 patients. Bull Clin Neurosci 1985;50:76-101.
21. Scharf D. Neurocysticercosis. Arch Neurol 1988;45:777-80.
22. White AC Jr. Neurocysticercosis: A major cause of neurological disease worldwide. Clin Infect Dis 1997;24:101-13.
23. Obafunwa JO, Hammerl E, Jaskierny D, Taylor L, Russell L, Reinhard K. Autologous bone flap resorption years after subtemporal craniotomy. Am J Forensic Med Pathol 2018;39:179-83.
24. Román G, Sotelo J, Del Brutto O, Flisser A, Dumas M, Wadia N, *et al.* A proposal to declare neurocysticercosis an international reportable disease. Bull World Health Organ 2000;78:399-406.