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Neurocysticercosis in a Nigerian woman -missed diagnosis of a neglected disease and surgical management

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ABSTRACT

Neurocysticercosis is a rarely diagnosed but significant clinical problem from infestation with taenia solium. It occurs as a result of ingestion of infested pork and the mode of transmission is usually feco-oral. It is commonly associated with non-specific neurologic manifestations expected of intracranial space-occupying lesions with its most common neurological presentation being seizures. However, its diagnosis without seizure is rarely reported. We report a recent experience in this regard and outcome of this disease which was inadvertently managed surgically. A good history and high index of suspicion especially in developing and low socioeconomic region is helpful for diagnosis in the presence of appropriate radiological findings. Prognosis is good when treatment is tailored to well selected cases. Treatment options include medical with or without surgical removal of encapsulated cyst. A high index of suspicion will be helpful in making appropriate diagnosis and proper management of this disease.

1. Introduction

Neurocysticercosis is a rare parasitic infestation of the brain parenchyma with the Taenia solium. T. solium is the specific tapeworm found in pig. Though few cases have been reported in the literature, it is generally considered a disease of the low socioeconomic status and endemic in developing countries. The typical route of infestation is feco-oral where the cyst of the tapeworm in the feces gets in contact with vegetables which is then consumed uncooked or partially cooked. The incidence of neurocysticercosis is largely unknown in different parts of the world.[1] It has been identified as a very common cause of seizure disorder in the developing world. As a result of the epidemic importance, there has been a proposal by WHO to make it a reportable disease worldwide.[2] Neurocysyticercosis in sub-Saharan Africa has been documented in the literature as a cause of secondary seizure but literature has been sparse on the surgical management.[1,3–5] There are however few literature on surgical management in patients presenting without seizure. This is perhaps the first to be reported as surgically managed in Nigeria (a West African country).

Clinical presentation depends on the site, size, number of intracranial lesions as well as the host immune response. Headache, seizures and other focal neurological deficits are common in patients with neurocysyticercosis. Diagnosis is mainly from these clinical presentations and appropriate neuroimaging. A high index of suspicion will aid early and appropriate diagnosis in endemic region.[3–6] Treatment involves the use of anthelmintic drugs, steroids with or without surgery depending on the presentation.[7–9]

However, diagnosis without seizure is rarely reported. We present this index case and document the incidence of missed diagnosis of this common but rarely diagnosed problem and subsequent surgical treatment.

2. Case presentation

A 35-year-old female staff nurse presented with 3 months history of progressively unabating headaches. There was no aggravating factor but the headache was temporarily relieved with use of non-steroidal anti-inflammatory drugs. There was no incidence of seizures. There was no

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significant past medical or family history. On examination, the only significant finding was left quadrantanopia. Physical examination showed no breast nor abdominal mass. On account of the chronic and worsening headache, an MRI scan of the brain was requested. The brain MRI showed a spherical lesion aborting the falx in the medial side of the left occipital lobe. The lesion was hypointense on T1 and hyperintense on T2 weighted imaging with peripheral enhancement on T1 with contrast (Fig. 1). There was significant perilesional edema and mass effect as evidenced by complete effacement of the posterior horn of the left lateral ventricle (Fig. 1). A working diagnosis of intracranial abscess with possible differentials of hydatid cyst, glial tumor or metastasis from an unknown primary origin was made. Because of increasing headache and large mass effect in addition to need for histologic diagnosis, she was counselled for excision biopsy. At surgery, left parieto occipital craniotomy was done, dural flap raised with the base along the sagittal sinus. Corticotomy of about a centimeter in the sulcus was done under operating microscope. This was dilated and deepened until the lesion was located. Careful dissection was done around the lesion with very minimal incursions into the normal brain tissues. There was intra-operative rupture of pus-like substance but it was prevented from spreading around the brain tissue. The whole lesion was removed en-bloc (Fig. 2) and the region was washed copiously with normal saline.

Histology confirmed cysticercosis with the use of Haematoxylin & Eosin staining technique which revealed multilayered cyst with invaginated scolices having numerous hooklets (Fig. 3A and B). The cystic cavity with fibrotic lesion and mixed pattern inflammatory cells-including plasma cells, lymphocytes, macrophages and eosinophils were observed. Masson's trichrome and Periodic Acid Schiff were the histochemical special staining techniques used to confirm fibrosis of the surrounding glia cells. Further examination with immunohistochemistry technique was also used to evaluate the larval stage of Taenia Solium and the surrounding neural tissues using anti-GFAP, anti-S-100 anti-CD20, anti-CD3, EMA and Vimentin. Immunohistochemical analyses revealed negativity with S100 protein, EMA (Epithelia Membrane Antigen) and Vimentin but positivity with GFAP (Glial Fibrillary Acid Protein), CD3 and CD20 expressing the level of activation of astrocytes and presence of both T and B cells respectively.

She was given oral anthelmintic for 3 months. Patient remained well all through a period of one year outpatient follow up. Her left quadrantanopia had completely resolved at follow-up review 3 months after surgery and she has remained well on follow-up.

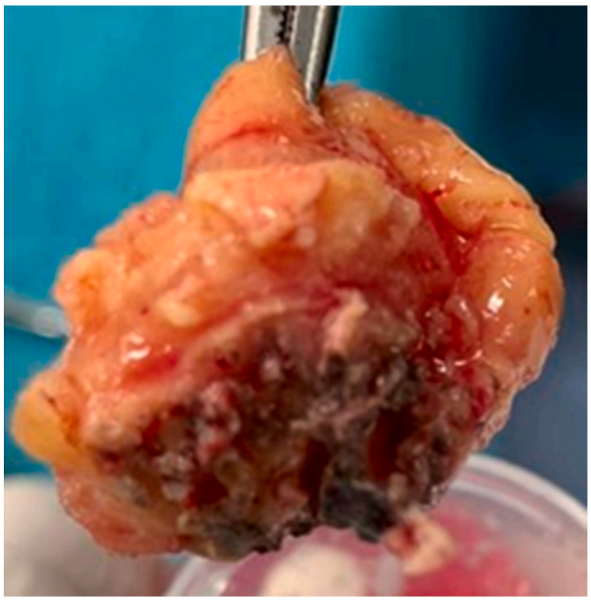

Fig. 2. Entire lesion removed en-bloc.

3. Discussion

Parasitic brain infection is a disease associated with developing nations and more commonly seen in rural and low socioeconomic regions. Typical brain infections include neurocysticercosis, echinococcus, schistosomiasis and others. Even though Nigeria is one of the endemic regions as published by the WHO, few cases of neurocysticercosis with seizures have been documented in the literature and even fewer cases of neurocysticercosis of surgical interest.[3–5,10] The life cycle of T. solium has been well documented in the literature. Typical endpoint after ingestion of the larvae of T. solium is the production of a space occupying lesion in form of a cyst in the brain with associated seizure and significant neurological deficits.

Neurocysticercosis in the West Africa subregion is most likely

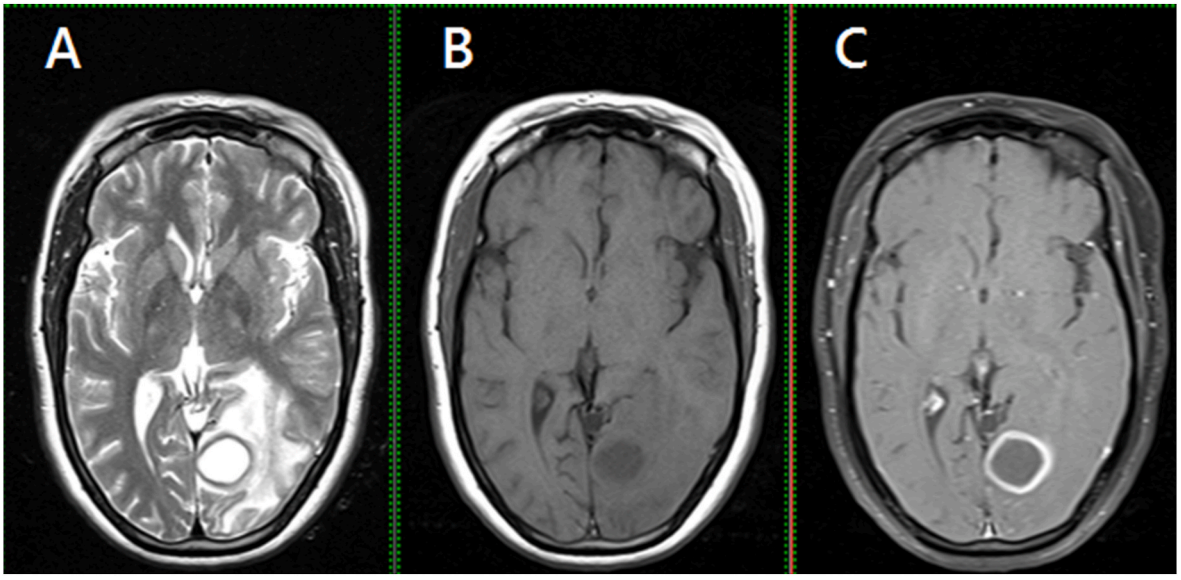

Fig. 1. A,B,C**:** The spherical lesion aborting on the falx along the medial side of the left occipital lobe. The lesion is hypointence on T1 *(1B)*, hyperintence on T2 *(1A)* with peripheral enhancement on T1 with contrast *(1C)*. Note the significant perilesional edema and mass effect as evidenced by complete effacement of the posterior horn of the left lateral ventricle *(1A)*.

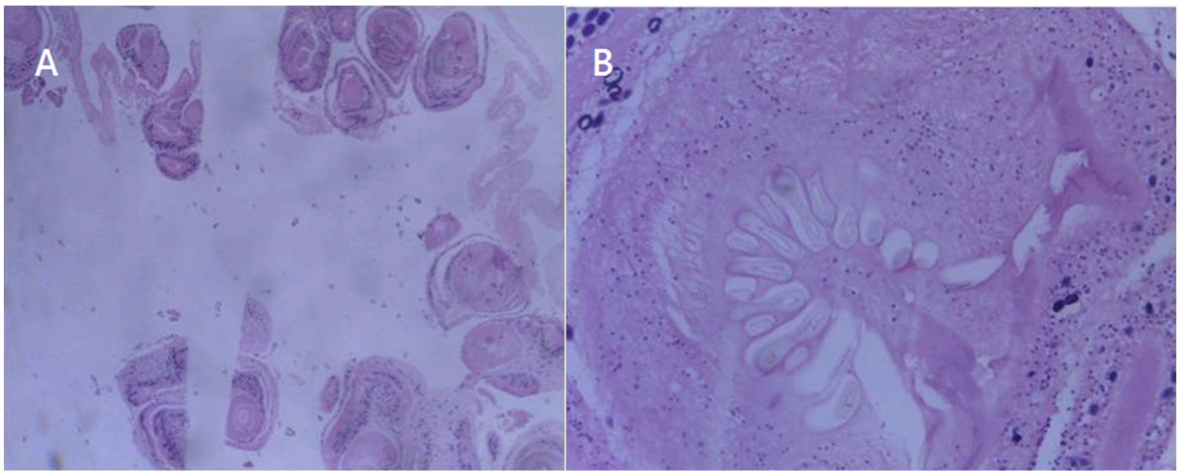

Fig. 3. A, B: *(using Haematoxylin & Eosin staining technique)* Cysticercosis seen as multilayered cyst with invaginated scolices having numerous hooklets.

underdiagnosed as a result of poverty, lack of the will to investigate with brain imaging or direct effect of poor healthcare.[1,6] The diagnosis was clearly missed in this case. There are two possible reasons for missing this important disease in an endemic region. First, the patient did not present with the common manifestation of seizures.[11,12] Second, as a result of rarity of neurocysticercosis, the brain MRI findings were not interpreted as possible intracranial neurocysticercosis but as an abscess to rule out metastasis.[13,14] Furthermore, the findings on MRI study depend on the particular stage of the neurocysticercosis at time of presentation which may be difficult for the pathologist to identify without appropriate experience. The stages include Vesicular, Colloidal vesicular, Granular nodular and Nodular calcified. One helpful radiologic characteristic from neuroimaging that should raise the index of suspicion for neurocysticercosis in the absence of any seizure is the typical subcortical cystic lesion(s) without any perilesional edema but with eccentric internal hypersdense foci on CT scan.[15] This radiologic finding are highly suggestive of cysts with scolices and is pathognomonic for Vesicular stage of parenchymal disease (classically referred to as "cyst with dot" sign).[15] Another finding that may be helpful is calcified cortical nodule, also seen in Vesicular stage of parenchymal disease.[15]

Just like other brain space-occupying lesions, presentation of a patient with neurocysticercosis is associated with impaired neurologic function of the part of the brain where the cyst is formed, though with increasing size of the cysts, many may present with features of raised intracranial pressure. The index patient only had headache and on clinical examination, quadrantanopia was elicited as a result of the cyst being in the occipital lobe. The typical presentation of seizure and raised intracranial pressure were however absent in the index case.[3,4,12,16] The lesion may grow bigger to the point of eliciting seizure activities in the brain, either from the direct pressure effect on the motor region or general irritation of the brain tissue. This underscores the importance of detailed clinical neurological examination in anyone with headache that is significant enough to seek medical help.

Detailed neurological examination is mandatory for all patients with chronic headache with neurological deficits as in the index case. Physical examination is equally important to rule out any mitotic lesions. Serology, molecular biology and neuroimaging are all necessary as well for diagnosis. Both MRI and CT scans are readily available in the developed countries but not so readily available within the regions that have higher incidence of parasitic infestations i.e low socioeconomic group.[6] Though these imaging modalities are available in some cities within the region, only few people are able to afford such investigations as healthcare in the region is mostly out of pocket as a result of poor coverage by the National Health Insurance Scheme. Consequently, fewer cases of neurocysticercosis are diagnosed and healthcare workers therefore have little familiarity with radiologic features of the disease. Molecular tests are equally expensive and not readily available in the region. In this index case, neurocysticercosis was not included as a differential diagnosis and so she was not treated as one.

Neurocysticercosis has been well managed medically in the literature. When presenting with seizures, appropriate anticonvulsant therapy is indicated. In cases where the diagnosis is made based on clinical history and neuro-radiology, anthelmintics have been noted to significantly help in symptom resolution.[7,9,17,18] There are however indications for surgical management especially when the diagnosis is made clinically as well as radiologically. Indication for surgical management include large lesions with mass effect in a symptomatic patient[8,9]. The index case however had a large lesion with perilesional oedema and mass effect which made her a surgical candidate regardless. Neurocysticercosis has been noted to mimic other pathologies in literature and therefore managed surgically, only to be diagnosed appropriately by histology post op.[19]Surgery for neurocysticercosis like any other intracranial lesions has to be carefully planned. The aim in neurocysticercosis excision is to achieve an en-block removal and to prevent rupture of the cyst content into the brain parenchyma. This is usually the case in a single lesion seen in an easily accessible brain region. Other factors considered in this case are the ease of localization and preservation of eloquent brain regions.[9] Neuronavigational equipment and intraoperative ultrasound scan are quite useful and helpful towards improving the ease of lesion localization. In cases with multiple cysts, a different approach would have been used. Treatment therefore has to be individualized.

Immunohistochemistry may be helpful in confirming histologic diagnosis of neurocysticercosis. The classical feature of hooklets within the scolices is classically indicative of the larvae of T. solium. Larval cysts evoke minimal surrounding inflammation. When cyst wall begins to leak C.cellulosae antigens, an intense inflammatory reaction develops in the adjusted brain. The immune response is both humoral and cell mediated.[10]

Further staining showed evidence of inflammatory changes within the brain tissues as seen in the presented case. Immunohistochemistry demonstrated the gliotic cells within the adjacent brain tissues. The brain tissue interestingly stained negative for Vimentin in this index case. Vimentin stain (an indicator of immature astrocytes) has, however been reported as positive contrary to what we saw.[10] Most of these studies were in pig models given the difficulties in analysing the developing immune response in infested human brains.[10,11] The discordance can, perhaps be as a result of the fact that in pigs the immune response can be examined earlier than in human patients, in whom sampling is inevitably made at a more chronic stage.[11] The variable inflammatory response seen in the index patient is not unusual as this has been reported in different experimental findings in the

literature.[10]

Prognosis A published series of 160 surgically treated cases of neurocysticercosis has shown that overall outcome is dependent on various factors[20] and that maleness as well as age younger than 40 years at the time of surgery and location of the cyst in basal cisterns all tend to be associated with the worst outcomes.[20] Also, prognosis tends to be worse in cases that require CSF diversion by shunting than in those who had only removal of the cyst.[20] In this index case, there was no indication for CSF diversion and we believe prognosis should be good when treatment is tailored to well selected cases.

4. Conclusion

Neurocysticercosis is a rarely diagnosed endemic health issue in Sub-Saharan Africa as a result of poverty and poor health care system. As a consequence of cost of proper evaluation, many cases are not managed appropriately. This case clearly shows that although the common presentation in the developing countries is seizure disorder, it could present with other features of space occupying lesion including headache and neurological deficits which are usually related to the involved location in the brain, instead of seizures. Neurocysticercosis diagnosis should hence be considered in people living in Sub Saharan Africa presenting with chronic headache, neurological deficits and suspicious neuroradiologic findings.

Informed consent

Obtained.

CRediT authorship contribution statement

Omotayo Abimbola Ojo: Supervision, Writing – review & editing. **Chiazor Udochukwu Onyia:** Conceptualization, Writing – original draft, Writing – review & editing. **Badirat Olukemi Lawal:** Writing – review & editing. **Nicholas Awodele Awolola:** Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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List of abbreviations

GFAP -: Glial Fibrillary Acid Protein
EMA -: Epithelia Membrane Antigen
MRI -: magnetic resonance imaging
WHO –: World Health Orgnisation
H and E –: Haematoxylin & Eosin
CT –: Computerized tomography
CSF –: Cerebrospinal fluid