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Case Report

Intraventricular adult *Taenia solium* causing hydrocephalus: A case report

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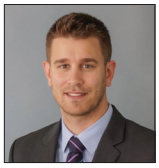

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ABSTRACT

Background: Neurocysticercosis (NCC) is the most common parasitic infection of the central nervous system worldwide and is caused by the larval form of the tapeworm *Taenia solium*. In general, *T. solium* larval form may be located in the neuraxis, resulting in pathology. Here, we report a rare case of female with a history of adult onset seizures presenting with adult form *T. solium* in the fourth ventricle, causing hydrocephalus.

Case Description: A 36-year-old female patient with a known history of adult onset seizures presented with a 1-year history of progressively worsening bilateral headaches with vertigo and intermittent nausea. A computerized tomography scan revealed ventriculomegaly and transependymal flow, with an obstruction at the level of the fourth ventricle. Outpatient magnetic resonance imaging demonstrated obstructive hydrocephalus secondary to a lobulated cystic mass within the fourth ventricle, demonstrating a gross appearance consistent with racemose NCC. The patient underwent endoscopic third ventriculostomy, and gross examination of the resected cyst revealed a mature *T. solium* larvae encased in a cystic membrane. Given that our patient was born and raised in Mexico but had not returned since the age of 8, NCC was an unexpected finding.

Conclusion: The present case highlights the importance of maintaining high suspicion for NCC in all patients presenting with seizures or hydrocephalus of unknown cause. Even in patients with a very remote history of residence in an endemic country, NCC can be an overlooked, underlying cause of both chronic neurologic symptoms, as well as acute, life-threatening neurologic emergencies.

Keywords: Hydrocephalus, Intraventricular, Neurocysticercosis, *Taenia solium*, Tapeworm

INTRODUCTION

Neurocysticercosis (NCC) is a parasitic disease endemic to most of the developing world and is a major source of admissions to neurological wards, as it is a significant cause of acquired seizures and epilepsy worldwide.[5] NCC is caused by the larval form (cysticerci) of *Taenia solium* and is one of the few rare helminth infections of the central nervous system (CNS). These cysticerci often develop in the ventricular system as free-floating cysts in ventricular cavities or are adherent to the choroid plexus.[15] Symptoms often arise when cysts obstruct hydrocephalus outflow in the ventricular system, resulting in obstructive hydrocephalus and increased intracranial pressure.[14] Here, we report a case of an adult suffering from *T. solium* brain infection in the United States.

CASE REPORT

History

Our patient is a 36-year-old female with a history of adult onset seizures. She was born and raised in Mexico and emigrated to the United States at the age of 8. Since arriving to the United States, she had never returned to Mexico. She had her first seizure about 10 years before presentation, at which point she was placed on anti-epileptic medications that were subsequently weaned off without any further seizure activity in the past several years. No prior imaging was available for our review, but the patient denied report of any intracranial lesions that could be identified as the culprit of the adult onset seizures.

Examination

She initially presented to an outside institution with progressively worsening bilateral headaches over the past year that were associated with vertigo and intermittent nausea. The headaches were worse at night and on waking up in the morning, and they improved throughout the day. A computerized tomography (CT) scan demonstrated ventriculomegaly and transependymal flow, with evidence of obstruction at the level of the fourth ventricle. She was referred to our clinic by our neurology colleagues, where she was found to have no neurologic deficits on examination. Outpatient magnetic resonance imaging (MRI) demonstrated obstructed hydrocephalus secondary to a lobulated cystic mass within the fourth ventricle, with a streak of internal linear enhancement most consistent with racemose NCC. A worm was easily identified within the cyst on the MRI [Figure 1].

Operation

Given the chronic nature of the lesion and her benign clinical examination, she was scheduled for elective surgery a few weeks later. She underwent right frontal burr hole for endoscopic third ventriculostomy and placement of external ventricular drain (EVD), then she was positioned prone and underwent suboccipital craniotomy for resection of the fourth ventricular cystic mass followed by duraplasty. The cyst was removed as a single *en bloc* specimen, although during the process of removal, a small portion of the cyst ruptured into the ventricle – this was quickly suctioned and extensively irrigated. Gross examination [Figure 2] and pathology [Figure 3] demonstrated a mature *T. solium* plerocercoid larvae measuring 1.2 × 0.5 × 0.2 cm encased in a tan, gelatinous, cystic membrane.

Postoperative course

Postoperatively, she had no neurologic deficits. Her EVD was quickly weaned with minimal drainage and low intracranial pressure, so it was removed on postoperative day 2. She underwent ophthalmologic examination, demonstrating no evidence of an active ocular infection or involvement of NCC, but concern for a chronic, inactive choroidal inflammatory process. Our infectious disease colleagues

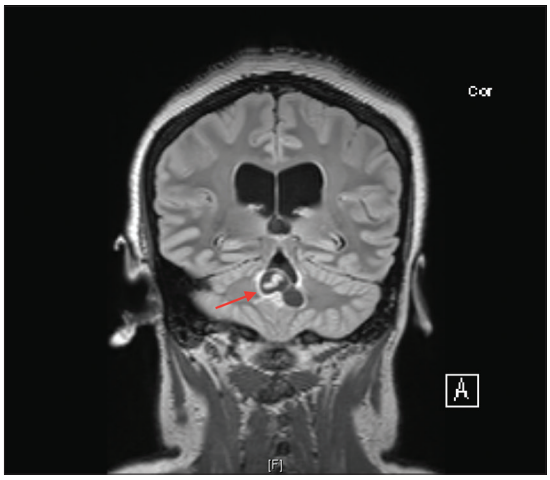

Figure 1: Preoperative magnetic resonance imaging with arrow pointing to intraventricular *Taenia solium* cyst.

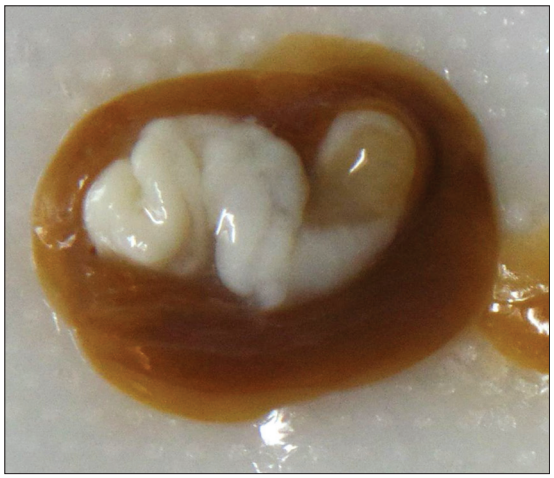

Figure 2: Gross mature *Taenia solium* plerocercoid larvae encased in a tan, gelatinous, cystic membrane.

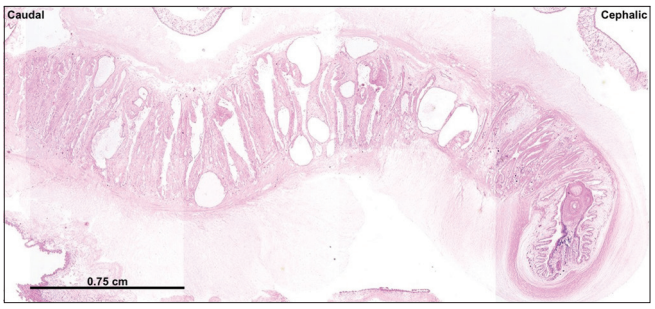

Figure 3: Histopathological examination of hematoxylin and eosin stained *Taenia solium* specimen.

were consulted and found no other risk factors for NCC aside from living in Mexico until the age of 8, and she was negative for tuberculosis infection. Given her longstanding history of seizures, but recent presentation with an active lesion, it was felt that she may have been auto-infecting. She underwent a 21-day course of albendazole with a 14-day dexamethasone taper for protection against aseptic meningitis. She was also prescribed a single dose of praziquantel at the end of her therapy given concerns for possible auto-infection from her gastrointestinal system. On discharge, her headaches, vertigo, and nausea had completely resolved.

DISCUSSION

NCC is the most common helminthic infection of the CNS worldwide and has recently become an emerging infectious disease in the United States, particularly in areas of high rates of immigration from endemic regions of Latin America.[10] Infection occurs primarily through ingestion of *T. solium* eggs, which hatch within the digestive system before releasing oncospheres that penetrate through the intestine, entering the circulation. These oncospheres passively reach the brain, forming larval cysts known as cysticerci. Extraparenchymal NCC occurs when the cysts reach the ventricles through the choroid plexus, where they may attach to the ependyma or pass freely.[19] Although intraventricular infection is rare, Figueroa *et al.* (2011) recently reported a case series consisting of 37 NCC patients in New Mexico and found such cysts in 30% of these individuals.[7] Amaral *et al.* (2003) reported a case series of 172 patients in Brazil with NCC and found greater than 54% of patients to have the intraventricular form of the disease.[2]

NCC has been widely reported in the literature; diagnostic criteria,[6] medical treatment,[1,10] and neurosurgical indications and approaches[12,17,18] have been well established by several groups. Racemose NCC is generally diagnosed by neuroimaging, laboratory evaluation, and epidemiologic findings. The Infectious Diseases Society of America (IDSA) and the American Society of Tropical Medicine and Hygiene (ASTMH) recommend MRI with 3D volumetric sequencing for the detection of intraventricular NCC,[20] which provides superior detection of the lesions as compared to CT, which has limited sensitivity for identifying subarachnoid and intraventricular cysts. Such lesions appear as hyperintense on T2-weighted images and provide clear visualization of potential hydrocephalus. These imaging and clinical evaluations in conjunction with serological testing – particularly in patients without the appropriate travel history – may provide additional data for a positive diagnosis. Common serologic findings include cerebrospinal fluid (CSF) lymphocytic pleocytosis in conjunction with increased protein levels dependent on the location and degree of infection.[11]

Intraventricular NCC presents more aggressively than the parenchymal form, often causing intracranial hypertension due to CSF obstruction and potential arachnoiditis.[17] Intraventricular cysts may be free floating and may cause obstruction of the fourth ventricle, Sylvian aqueduct, or the foramina of Monro, resulting in rapid deterioration as a result of acute hydrocephalus. In contrast, the cysts may become attached to the ependymal ventricular wall, resulting in ependymitis. In 2002, DeGiorgio and colleagues concluded that the risk of death in patients with NCC was higher in patients with large cyst burdens and those with intracranial hypertension.[4] Furthermore, the inflammatory process may contribute to vasculitis and further cerebrovascular disease.

The management and treatment of intraventricular NCC is distinct from the parenchymal form, which is the most widely published on. Indications for surgical management of intraventricular NCC include mass effect, CSF obstruction, and ventricular cysts. Acute hydrocephalus, as manifested in the case presented here, requires emergency ventriculostomy followed by cyst resection.[17] Today, the surgical modality of choice is neuroendoscopic cyst resection with ventriculostomy, with or without an EVD, or permanent shunt.[16] This has resulted in fewer complications and higher cure rates as compared to traditional open craniotomy and may obviate the need for anthelminthic agents.[13] In the case of an intraventricular cyst that is freely mobile, imaging directly before neuroendoscopic intervention is recommended to most accurately confirm location. However, a general contraindication for neuroendoscopic removal of such cysts is ependymal enhancement, which generally indicates dense, strong adhesion between the cyst and the ependymal wall. Therefore, attempted cyst removal in such cases has been associated with neurologic injury and intraventricular hemorrhage.[17]

In several cases, a ventriculostomy by itself may be enough to relieve hydrocephalus without a shunt; however, external drainage is often indicated in patients with extensive disease and damage. Furthermore, ventriculitis often complicates shunt placement in patients with NCC, resulting in shunt malfunction rates that are exceptionally high – reported to be over 30% – with a high rate of postoperative mortality.[9] In addition, shunts carry a risk of bacterial infection along with the potential for shunt failure by blockage.[20]

At present, there are no clinical trials or definitively established guidelines for the anthelminthic treatment of intraventricular NCC and remain poorly resolved; lesions may spontaneously resolve, and anthelminthic agents may lead to worsening of symptoms.[8] In general, it is often recommended that treatment be given under observation in the hospital coupled with corticosteroid administration to manage CNS inflammation. Many clinicians follow the 2013 American Academy of Neurology guidelines for the treatment

of parenchymal NCC, which recommends the use of albendazole and dexamethasone or prednisolone.[3] A longer dosing timeline of praziquantel is an acceptable alternative. The recent IDSA and ASTMH guidelines suggest using corticosteroids to decrease brain edema in the perioperative period. In addition, they recommend the use of antiparasitic drugs with corticosteroid therapy postshunt insertion to decrease the possibility of shunt failure in patients in whom surgical removal of intraventricular cysts is not possible. However, neither corticosteroids nor antiparastic drugs are recommended after successful removal of the intraventricular cysts

The present case highlights the importance of maintaining high suspicion for NCC in all patients presenting with seizures or hydrocephalus of unknown cause. Patients are most effectively evaluated for NCC by obtaining an MRI, serologic testing, and thorough neurologic examination. In addition, in the era of globalization and increased immigration and travel, it is critical to collect a thorough social history, including travel and residence history to any endemic country. This case illustrates that even in patients with a very remote history of residence in an endemic country, NCC can be an overlooked, underlying cause of both chronic neurologic symptoms, as well as acute, life-threatening neurologic emergencies.

CONCLUSION

Although rare in the United States, increased travel and immigration across endemic areas have increased the incidence of NCC in regions unfamiliar with the disease. Intraventricular cysticerci may be mistaken for other common intraventricular lesions, including colloid cysts. Here, we have presented an extremely rare case of an adult *T. Solium* within a cyst in the fourth ventricle, causing acute hydrocephalus.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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