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Case report

To intervene or not to intervene: A case of symptomatic neurocysticercosis complicated by ventriculitis

Syed H. Haq [a], Sidra R. Shah [a], Jannet Bux [a], Anh Si Le [a], Hafez Golzarian [a], Daniel Mueller [a], Joseph J. Sreenan [b], Sandeep M. Patel [c], Amanda Laird [d], William Cole [d,*]

[a] Department of Internal Medicine, BonSecour Mercy Health—St. Rita's Medical Center, Lima, OH, USA
[b] Department of Pathology Medicine, BonSecour Mercy Health—St. Rita's Medical Center, Lima, OH, USA
[c] Department of Interventional Cardiology, BonSecour Mercy Health—St. Rita's Medical Center, Lima, OH, USA
[d] Department of Critical Care Medicine, BonSecour Mercy Health—St. Rita's Medical Center, Lima, OH, USA

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ABSTRACT

Although well described in the current literature, Neurocysticercosis [NCC] remains an enigma when confronted by practitioners. This is in part due to the haphazard nature of the parasitic infection on the central nervous system [CNS]. These include single or multiple anatomic sites of infection, stage of parasitosis, and the resultant inflammatory response. As a result, NCC can present with a complex constellation of symptomatic presentations, making therapeutic regiments highly individualized. Despite intervention, other impediments may arise post-therapy due to the nature of the infection. We present a case of rapidly progressive symptomatic NCC that initially was successfully treated, however would eventually succumb to complications of ventriculitis.

Introduction

Neurocysticercosis is the most common parasitic disease of the central nervous system [1,2]. The causative agent is *Taenia solium* of NCC, with transmission originating from the consumption of fecally contaminated water or food containing *T. solium* eggs. The infection is more frequently endemic to third world nations, but outbreaks in the US are on the rise with most cases associated with high immigration centers, predominately in the southwestern US and with a NCC prevalence rate of 0.2–0.6 per 100,000 [3]. Treatment depends on a multitude of variables, often being highly individualized to the patient's symptomatology and prognosis. Furthermore, the nature of the infection predisposes individuals to determinantal complications despite initiation of appropriate therapy, or resolution of the cyst burden.

Case presentation

A 24-year-old Caucasian female with a past medical history of seizures presented for altered mental status. She was found minimally responsive and febrile. Her husband reported that she had flu-like symptoms and a headache prior to arrival. On exam, she was tracking with her eyes, moving all extremities but unable to converse. Upper motor neuron signs were present with clonus and a positive Babinski's sign. No nuchal rigidity was appreciated. A computed tomography [CT] head and computed tomography angiography [CTA] head and neck were insignificant. Lumbar puncture was concerning for bacterial meningitis with an opening pressure of 38 cm of water, with cerebral spinal fluid [CSF] analysis showing elevated neutrophil count and protein, with low glucose. A magnetic resonance imaging [MRI] showed innumerable ring-enhancing lesions, see Fig. 1. The patient was admitted to the intensive care unit, intubated for airway protection, and started on hypertonic saline and anti-epileptic therapy. Additionally, she was empirically started on Rocephin, Ampicillin and Vancomycin whiles cultures resulted.

During hospitalization, she continued to demonstrate intermittent seizure. An electroencephalogram demonstrated epileptogenic tendency and encephalopathy. CT head demonstrated findings of vasogenic edema, and the patient was subsequently started on steroids. CSF studies were negative for common bacterial, viral or HIV causes. Initial work up for septic emboli resulted in negative blood cultures and an

Abbreviations: NCC, Neurocysticercosis; CNS, Central Nervous System; CT, Computed Tomography; CTA, Computed Tomography Angiography; CSF, Cerebral Spinal Fluid; MRI, Magnetic Resonance Imaging; ICP, Intracranial Pressures.

* Correspondence to: Department of Critical Care, St. Rita's Medical Center (Mercy), 730 West Market Street, Lima, OH 45801, USA.
E-mail address: WCole@mercy.com (W. Cole).

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echocardiogram was significant only for a small patent foramen ovale. The patient underwent a right temporoparietal craniotomy with brain biopsy showing no larva, only reactive gliosis. Eventually, serology returned positive for neurocysticercosis. Stool sample was negative for ova or other parasites. Ophthalmologic examination was unremarkable. Therapy, as per current standard of care, was changed to Albendazole, Praziquantel and steroids. Dexamethasone 0.1 mg/kg/day was initiated one day prior and continued throughout the duration of antiparasitic therapy. One week later there was improvement in neurologic status with the patient following commands. She was extubated with follow up CT head showing improvement in cyst burden and cerebral edema. The patient completed her anti-parasitic therapy, a rapid steroid taper, and was discharged after 25 days of hospitalization.

The patient returned to the emergency department two weeks later for worsening mental status and tachypnea. On evaluation, she was febrile with leukocytosis and elevated proinflammatory markers. On physical exam, a horizontal nystagmus was appreciated. She soon became unresponsive, with a Glasgow coma scale of 5 and without gag reflex. She was subsequently intubated. CT head demonstrated a 5 mm left to right midline shift, a mass in the left hemisphere with adjacent

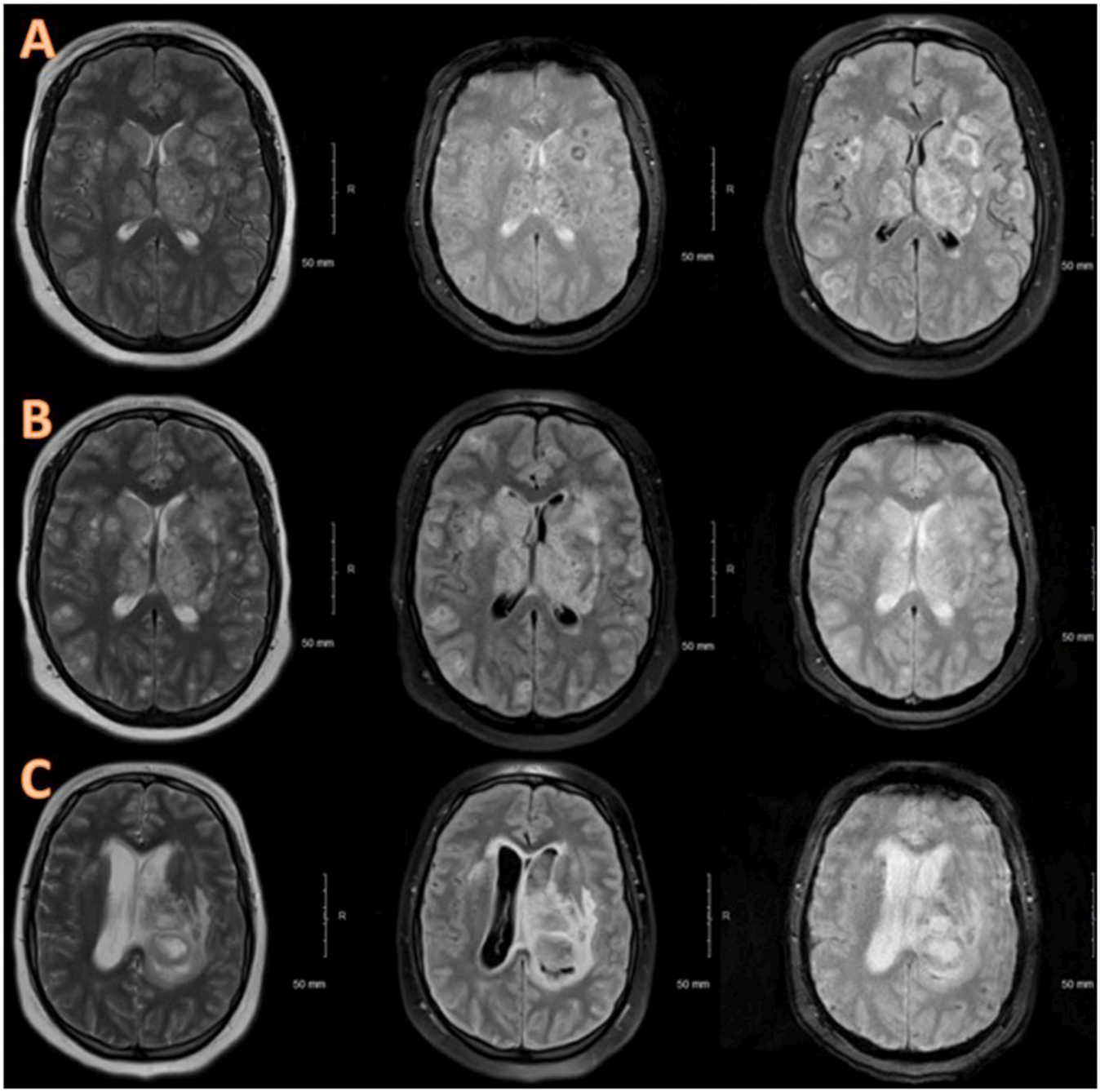

Fig. 1. (A) Day 1; Innumerable ring-enhancing central restriction diffusion lesions predominately involving the supratentorial brain with proclivity for the basal ganglia, thalami and gray-white matter junction, resulting in prominent disease burden and edema causing mass effect on the lateral ventricles. Additionally, midbrain lesion causing partial effacement of cerebral aqueduct. (B) Day 3; Redemonstration of lesions with milder involvement of the brainstem and bilateral cerebellum. Vasogenic edema appreciated. (C) Readmission Day 1; Interval development of hydrocephalus and ventriculitis. Left lateral ventricle periventricular/intraventricular lesions measure up to 3.1 cm with surrounding edema, consistent with NCC in colloidal vesicular phase. Left to right midline shift 0.4 cm. Demonstration of innumerable lesions now in nodular calcified stage. Right basal ganglia with findings indicative of pus.

mass-effect resulting in compression of left ventricle with surrounding edema concerning for an abscess, see Fig. 2. Hypertonic saline, steroids and antiparasitic therapy with albendazole and praziquantel were started. MRI brain demonstrated left basal ganglia and thalamic abscesses associated with ventriculitis, hydrocephalus, and persistent midline shift. Shortly after, unequal pupils were noted. CSF cultures were positive for Streptococcus agalactiae, and antiparasitic therapy was switched to Vancomycin and Meropenem. The patient underwent a right sided burr hole and ventriculostomy with external ventricular drain placement for imminent ventricular obstruction and cerebral decompression. Unfortunately, serial CT head scans demonstrated worsening progression of global edema, mass effect and midline shift, with signs of blurring of gray-white matter differentiation. Pupils remained fixed and dilated, with no appreciable brainstem reflexes. Brain death was declared after a positive apnea test.

Discussion

T. solium goes through numerous stages of infection once it infects the CNS. These include vesicular, colloidal, granular-nodular, and nodular-calcified. The organism can persist for 5–10 years with or without a symptomatic presentation in the vesicular phase. It does this by evading the immune system by disinhibiting the complement system, altering cellular response and degradation of attacking immunoglobulins [4]. Degeneration of the cysts occurs within the colloidal phase leading to more pronounced symptoms as the body's inflammatory system responds. This process can cause gliosis, fibrosis, edema, necrosis, ependymitis, and arachnoiditis [2]. This is followed by the granular nodular and nodular-calcified stage which are signified by progressive degradation and formation of a non-viable calcified nodule.

The site of infection can either be intraparenchymal or extraparenchymal. Intraparenchymal lesions are more common and are generally associated with seizures and headaches. Seizures are a result of immune reactions from degenerating NCC cysts. Other symptoms that may accompany intraparenchymal NCC are visual or cognitive changes and focal neurologic deficits. Individuals with a large cyst burden and an inflammatory response either by the host or provoked by antihelminthic therapy are predisposed to cysticercal encephalitis. Thus, although intraparenchymal NCC has a more favorable outcome compared to extraparenchymal lesions, the outcome is dependent on the number of cysts and degree of inflammatory response, which may lead to an epilepsy related death [4–7]. Extraparenchymal lesions have a diffuse presentation, affecting the ventricles, subarachnoid space, spine, and the eyes. These are typically associated with a larger parasite

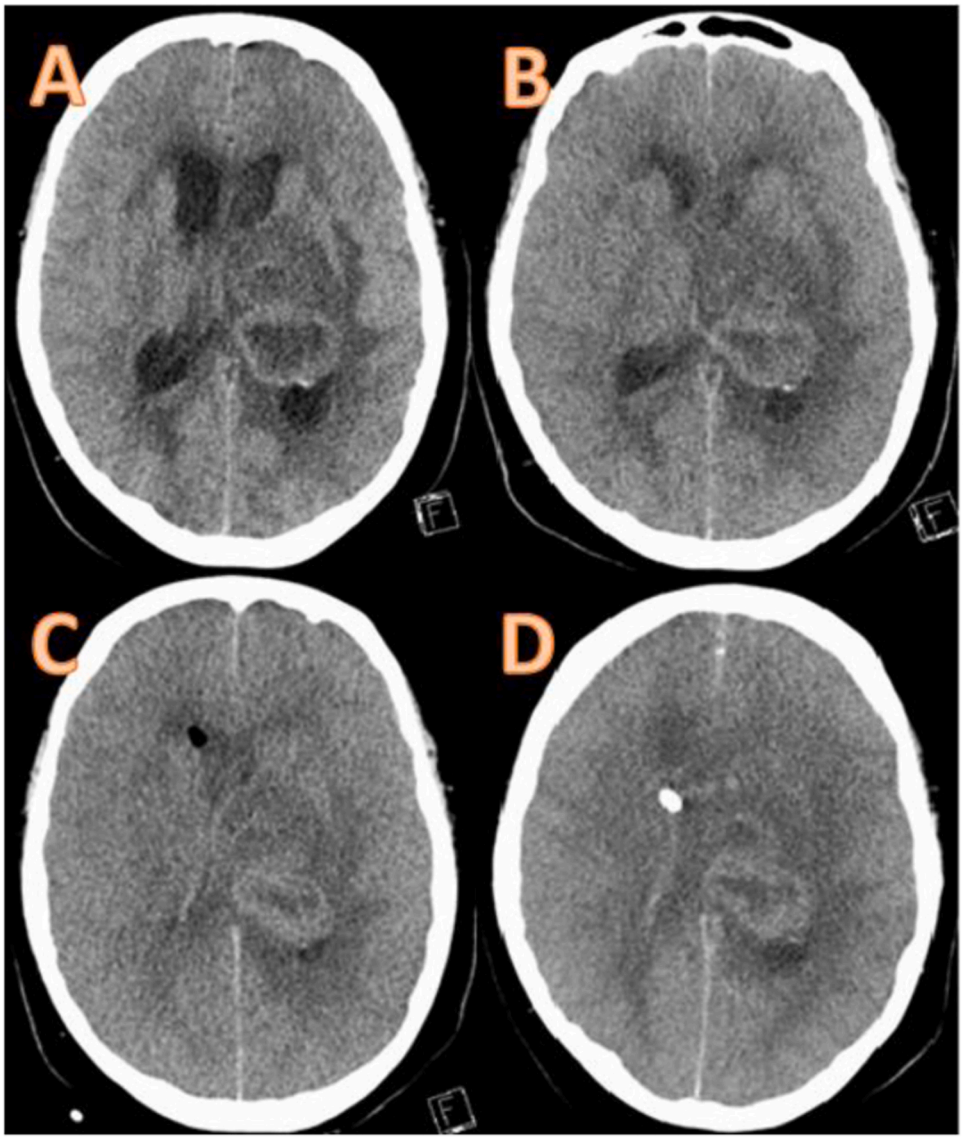

Fig. 2. (A) Readmission Day 1; 5 mm left to right midline shift. Left lateral ventricle is compressed. Extensive bilateral white matter edema in left frontal and parietal lobe, with 3.4 × 2.4 cm round mass. (B) Readmission Day 2; Worsening global edema and mass effect on basal cisterns and ventricles. Moderate hydrocephalus. Early blurring of gray-white matter differentiation. (C) Readmission Day 2; Interval placement of EVD. Ventricles are completely decompressed and flattened. Worsening diffuse edema with absence of cerebral edema. (D) Readmission Day 3; Diffuse brain swelling with effaced basilar cisterns and sulci. Slight rightward midline shift.

burden. Due to this process, these lesions are largely correlated with a worse prognosis, mortality most commonly a result of obstructive hydrocephalus, and the effects of elevated intracranial hypertension [8,9].

Treatment is centered around antiparasitic therapy, steroids, and symptomatic management. Furthermore, the effects are heavily reliant on degree, burden, and location of infection. Albendazole has been found to be most efficacious due to its optimal CSF penetration and less interactions with praziquantel, often used in conjuncture [10]. Steroids are utilized to prevent or decrease cerebral edema and intracranial pressures [ICP] [11]. Carbamazepine and Phenobarbital are optimal choices for control of seizures, such as those caused by intraparenchymal lesions [9]. Recent research has shown that methotrexate use with concomitant anti-TNF agents can play an anti-inflammatory role [8]. In severe cases, as seen with extraparenchymal NCC the picture may be complicated with hydrocephalus, intraventricular cysts or racemose NCC. In these circumstances, neurosurgical interventions may be warranted with ventriculoperitoneal shunt placement or cyst removal [1]. Additionally, prior to initiation of treatment an ophthalmic exam is necessary to exclude ocular cysticercosis, as inflammation induced by therapy can irreversibly threaten vision [5].

Treatment methodology remains a contentious area of debate within the literature. Cysticidal drugs, at times, have been attributed with greater risk than benefit. Mass cystic death within the CNS stimulates an extensive inflammatory response resulting in detrimental systemic effects, and therefore are contraindicated in situations of potential rise in ICP such as current or risk of hydrocephalus, subarachnoid, or encephalitic NCC [7]. Furthermore, in many cases parenchymal cysts naturally resolve or follow a benign and asymptomatic pathway [1,4]. However, several studies have demonstrated reduction in parenchymal lesion symptoms while on treatment, namely seizures and headaches [11]. Finally, neurocranial instrumentation increases the risk of infection, especially when on extended steroids.

Unique in our case is the fact that the patient was a Caucasian female with no history of travel or exposure to rural areas contaminated with *T. solium*. Reported by family, the patient would procure her own pork, grind and season to make chorizos. Despite the intraparenchymal NCC infection, she was symptomatic on presentation with seizures refractory to anti-epileptics, thus warranting treatment. Her initial CSF studies returned negative for Escherichia coli, Haemophilus influenzae, Listeria, Neisseria meningitides, Streptococcus agalactiae Pneumococcus, Tuberculosis, Cytomegalovirus, Epstein Barr Virus, Herpes simplex virus, Human immunodeficiency virus, Cryptococcus, and Syphilis. This prompted increasing suspicion for septic emboli, but was ruled out by negative blood cultures, echocardiogram, and resultantly a craniotomy with biopsy. As NCC was confirmed on serology, anti-helminthic therapy and steroids were initiated. Therapy exhibited success, with neurologic improvement over a prolonged hospitalization course. She completed 14 days of antiparasitic therapy with concurrent steroid use. However, she would return in a markedly worse status. Studies demonstrated significant midline shift and cerebral edema as result of ventriculitis and abscess formation. CSF culture was positive for Streptococcus agalactie. Her initial neurosurgical intervention, extended steroids utilization, and stay in healthcare setting may have predisposed her to acquire the infection. In any case, she rapidly deteriorated and inevitably succumbed to the infection despite aggressive measure to control the cerebral edema.

Conclusion

The symptomatology associated with NCC is contingent on numerous variables including cyst burden, size, location, and resultant inflammatory response. An individualistic approach must be applied when treating NCC. As treatment is accompanied by the potential to accelerate a poor clinical course due to exacerbation of symptoms or by causing inadvertent complication. Sequelae may include concurrent or resultant infectious, epileptic state or extended neurologic damage. Therefore, mortality and morbidity remain high with NCC. Further investigation is warranted to establish guidelines for post treatment monitoring and prophylaxis.

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Consent

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Disclosures

The authors have nothing to disclose.

CRediT authorship contribution statement

Syed H. Haq: Primary writer (original draft, review, editing, figures). **Sidra R. Shah**: Writing- (reviewing, case collection, and editing. **Jannet Bux:** Writer, reviewing, and resources. **Anh Si Le:** Writer and reviewer. **Hafez Golzarian:** Writer and reviewer. **Daniel Mueller:** Writing and case collection. **Joseph J. Sreenan:** Reviewer and figure. **Sandeep M. Patel:** Reviewer. **Amanda Laird:** Supervision and reviewer. **William Cole:** Supervision and editing.

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