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Case Report

Surgical nuances of giant neurocysticercosis according to intracranial location in the Southwest region of the Dominican Republic, presentation of cases, and literature review

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ABSTRACT

Background: Neurocysticercosis (NCC) is the most common infestation of the central nervous system, caused by the larval stage of the pig tapeworm *Taenia solium*. It is prevalent in regions with poor sanitation and underdevelopment, such as Latin America.

Case Description: We present four cases in which they harbored an intraventricular/intraparenchymal, frontal convexity, cerebellomedullary, and intraparenchymal NCC cyst of medium size, respectively. Three of them underwent complete removal of the cyst by craniotomy; the fourth had a shunt for obstructive hydrocephalus first, followed by excision of a suboccipital cyst 8 months later.

Conclusion: The intraventricular/intraparenchymal lesion was more complex to treat than its subarachnoid counterparts because the average brain should be transected and dissected away to achieve total removal. Waterjet dissection, arachnoid microdissection, and cyst drainage allowed minor brain damage than capsule coagulation and traction. Populated prospective studies are needed better to understand the surgical nuances of these rare entities.

Keywords: Cyst, Neurocysticercosis, Surgical technique, *Taenia solium*, Treatment

INTRODUCTION

Cysticercosis is a multi-system disease resulting from the seeding of the larval form of the pork tapeworm, *Taenia solium*, to various organs of the body. It is contracted through the fecal-oral route after ingesting viable eggs in foods contaminated with human feces.

T. solium is one of the seven neglected endemic zoonoses targeted by the WHO, and it is known to be endemic in broad areas of the world, including Latin America, Eastern Europe, sub-Saharan Africa, and parts of Asia, including the Indian subcontinent, Southeast Asia, and large regions of China.[1,2] The endemicity of *T. solium* is deeply rooted in poverty and involves domestic pig raising and poor sanitary conditions. The burden of neurologic disease associated with continued transmission falls mainly on impoverished rural populations.[3,8] However, neurocysticercosis (NCC) cases are diagnosed worldwide, even where transmission is not endemic.

Some extensive studies have shown increasing incidence in developed countries such as the United States, and it is very variable around the world. In Latin America, the incidence varies depending on the urban or rural region, from 121.7 to 138.4 cases/100,000 individuals per year. Sanitary conditions have a close relationship with NCC, and combating this disease is a priority for the World Health Organization (WHO).[4]

NCC may cause focal epilepsy from its lesions or lead to distant epileptogenesis through the development of hippocampal sclerosis and subsequent mesial temporal lobe epilepsy. Across all causes of acquired epilepsy (epilepsy not associated with genetic causative mechanisms or delivery), NCC is likely the most frequent causative mechanism in low-income countries.[5] Multiple reviews and meta-analyses[6] have consistently calculated that approximately 30% of all epilepsies in NCC-endemic regions are attributable to NCC. However, NCC may manifest as almost any neurological symptom depending on the cyst location and stage and the number of cysts,[3] frequently including chronic headache, intracranial hypertension, and cognitive impairment.

NCC has been reported as the most frequent helminthic infection of the central nervous system (CNS). Yet, there have been very few studies conducted in the Caribbean region to estimate the prevalence of NCC. This is primarily because NCC can only be diagnosed with certainty through neuroimaging or autopsy. Hence, the frequency of sequelae following infection with the larval stages of *T. solium* remains largely unknown.[7] Accurate data on the worldwide frequency of CNS infections with cysticercosis is lacking.

METHODS

The present article is a retrospective case series and literature review. We identified four cases of patients with giant neurocysticerci who underwent surgical treatment at our institution between 2015 and 2020 in the south region of the Dominican Republic [Table 1]. All patients were diagnosed using Del Brutto *et al*.[5] Diagnostic criteria for NCC with one major and two minor, we excluded patients who did not have complete clinical and radiological data available. Data collection was performed by reviewing electronic medical records and radiological images. We collected demographic data, clinical presentation, radiological features, surgical approach, intraoperative findings, and postoperative outcomes. Data were entered into a Microsoft Excel spreadsheet and analyzed using descriptive statistics. In addition to the case series, we performed a literature review to identify studies reporting the surgical management of giant neurocysticerci. We searched PubMed, Embase, and Cochrane Library databases using the following keywords: "neurocysticercosis," "giant neurocysticercus," "surgical management," "intracranial location," "case series," and "literature review." The search was limited to studies published in English from 2003 to 2020. We screened the titles and abstracts of identified studies for relevance and then reviewed the full text of potentially relevant studies. We extracted data on study design, patient characteristics, surgical approach, outcomes, and complications.

The results of the literature review were summarized narratively, with a focus on the surgical nuances of giant neurocysticerci according to intracranial location. This study was approved by the Institutional Review Board, and informed consent was obtained from all patients included in the case series.

CASE NO. 1

Clinical presentation and imaging

A 49-year-old male patient, obese, human immunodeficiency virus-positive (irregularly treated), presented to the emergency room (ER) with a 6-month history of altered mental status, left-sided hemiparesis (motor power grade 3/5), and left partial seizures. A computed tomography scan showed three cystic lesions, the biggest causing mass effect, in the right lateral ventricle extending to the ipsilateral basal ganglia. Contrast magnetic resonance imaging (MRI) showed a large (9.5 × 5.2 × 5.6 cm) enhancing cystic lesion with an enhancing nodule with substantial perilesional edema, compressing the ipsilateral ventricle and brain. The cyst was hypointense on the T1-weighted image and hyperintense on T2 and fluid-attenuated inversion recovery weighted image [Figure 1]. The signal intensity of the cyst was similar to cerebrospinal fluid (CSF) in all the sequences.

Techniques

A right 6 × 7 cm frontal craniotomy was performed, and a superior frontal gyrus (F1) approach was used, transecting its posterior portion. The first step was waterjet dissection between the cyst and the surrounding brain and ventricle, allowing adhesions separation. Then, we drained 20 mL of a yellowish serous fluid, reducing the cyst volume. Bipolar

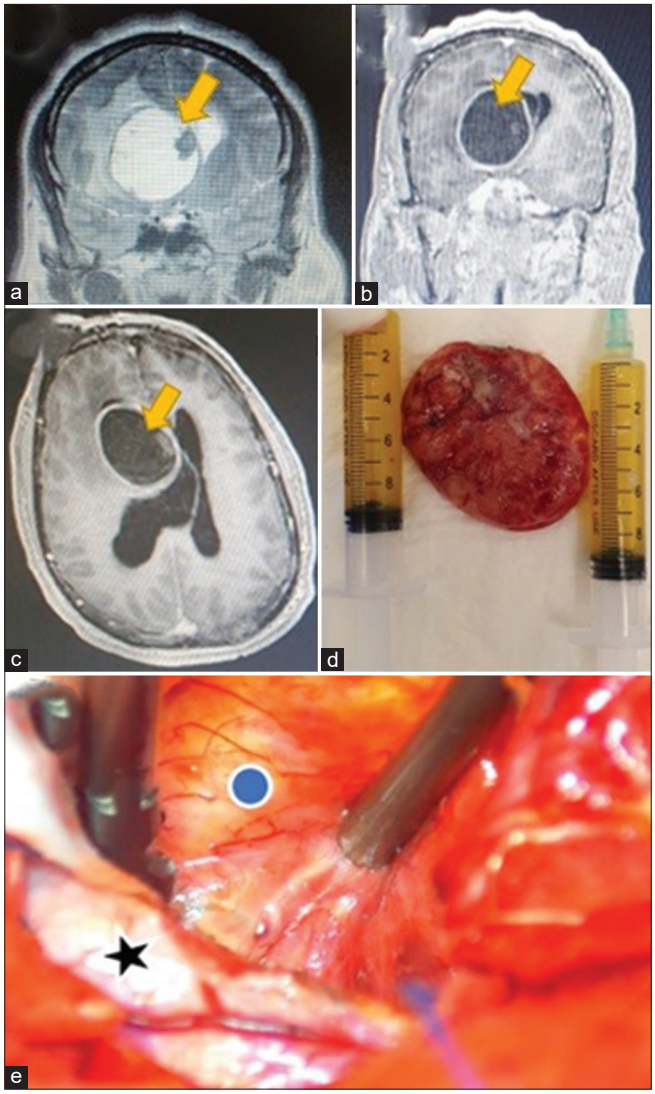

Figure 1: (a and b) Coronal view, T2- and T1-contrasted sequence magnetic resonance imaging (MRI) showing the intraventricular cyst, with scolex (yellow arrow). (c) Axial T1-contrasted MRI, the yellow arrow shows the giant cyst. (d) Excised cyst with intracapsular fluid drained (syringes). (e) Intraoperative exoscopic view of the cyst (blue dot) and the adjacent brain parenchyma (black star).

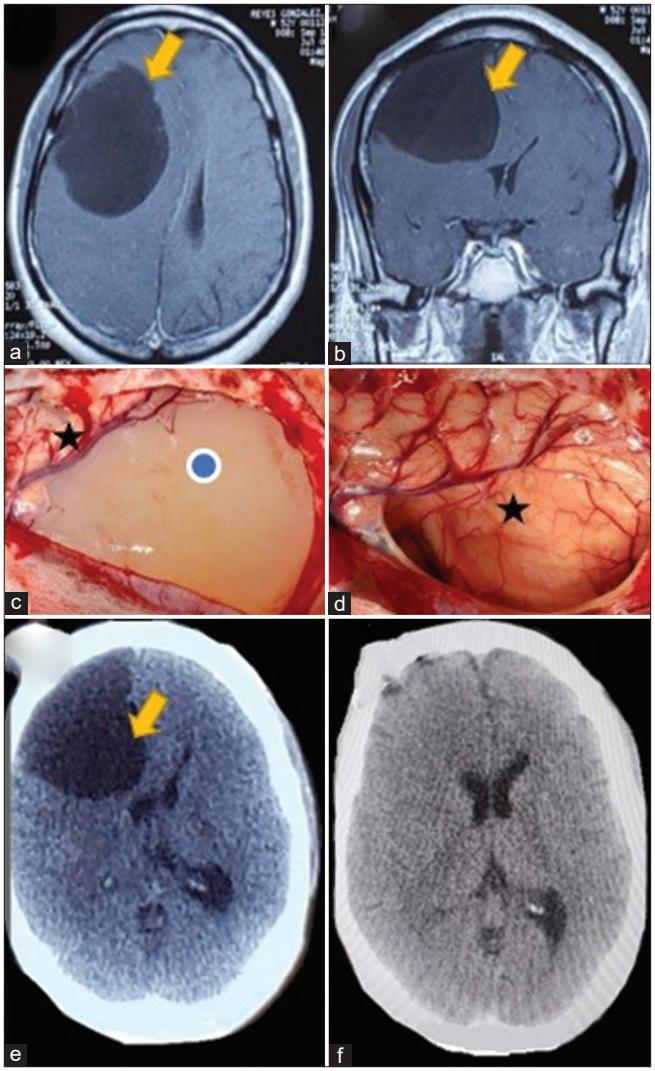

Figure 2: (a) Axial T1 contrast-weighted magnetic resonance imaging (MRI) with mass effect cyst (yellow arrow) and (b) coronal view, T1-contrasted sequence MRI showing the giant cyst (yellow arrow). (c) Intraoperative view of the cyst (blue dot) and adjacent brain (black star). (d) Intraoperative view of compressed brain after complete cyst removal (black star). (e) Preoperative axial view of a head computed tomography (CT) scan showing the compressing cyst (yellow arrow). (f) Postoperative head CT scan demonstrating the complete cyst removal.

coagulation shrank the capsule, and gentle traction of the lesion, protecting the edematous brain with patties, allowed the complete cyst excision. Histopathology confirmed the diagnosis of cysticercus cyst.

CASE NO. 2

Clinical presentation and imaging

A 44-year-old male patient, treated for 2 years by a psychiatrist for a supposed bipolarity, was seen in the outpatient clinic presenting personality changes, headaches, and generalized seizures, the latter 2 months before presentation. Computed tomography (CT) scan showed a prominent (12 × 8 × 8 cm) irregular subarachnoid hypodense cystic lesion above the right frontal lobe with a critical mass effect. Contrast MRI demonstrated a nonenhancing capsule and nodule without perilesional edema [Figure 2].

Techniques

A right 10 × 7 cm frontal craniotomy was done. The cyst was immediately beneath the dura, and it had a thin capsule filled with white fluid. We tried sharp dissection first,

between the lesion and brain, to develop a cleavage plane, but a spontaneous rupture occurred. Hence, we performed complete drainage followed by traction of the thin capsule.

CASE NO. 3

Clinical presentation and imaging

A 52-year-old male patient presented to the ER with a 2-week history of severe headaches. On examination, he was drowsy with moderate dizziness. No other neurological sign or symptom was found. CT scan showed acute hydrocephalus secondary to a cystic lesion in the cerebellomedullary cistern. He was shunted first, with immediate relief of the symptoms. He was discharged home to complete antiparasitic drugs treatment. Eight months later, the patient complained of nausea and vomiting; a contrast MRI demonstrated the nonenhancing cystic lesion without scolex, extending to the fourth ventricle [Figure 3].

CASE NO. 4

A 22-year-old female patient arrived at the emergency room presenting with generalized seizures, which had been preceded by a week of headaches. On examination using a head CT scan, an intracerebral cyst of middle size (2.3 × 2.1 × 3 cm) was identified in the right frontal lobe [Figure 4]. The cyst exhibited a calcified nodule and was accompanied by brain edema in the surrounding area. Considering these findings, the patient was promptly taken to the operating room for further evaluation and treatment, with a probable diagnosis of an oligodendroglioma tumor.

Techniques

We performed a right frontal craniotomy, posterior middle frontal gyrus corticectomy, capsule coagulation, and complete removal using gentle traction. The lesion was solid, yellowish, and avascular.

RESULTS

Table 2 describes four cases of NCC with intracranial cyst location, ranging in age from 22 to 52 and including both males and females. All four cases presented with a variety of symptoms, including seizures, headaches, altered mental status, and vomiting/dizziness. The cyst locations varied, with three cases involving the right side of the brain (in the lateral ventricle/ipsilateral basal ganglia, subarachnoid frontal lobe, and intracerebral frontal lobe) and one case in the posterior craniocervical subarachnoid space [Table 2]. Hydrocephalus was present in two of the cases, with one requiring shunting. The duration of symptoms varied from 1 week to 2 years, with one case presenting with hydrocephalus for 2 weeks before surgery. The surgical techniques used included waterjet, cyst drainage, capsule coagulation, traction, and sharp dissection. There were no intraoperative complications in three of the cases, while one case experienced a cyst rupture.

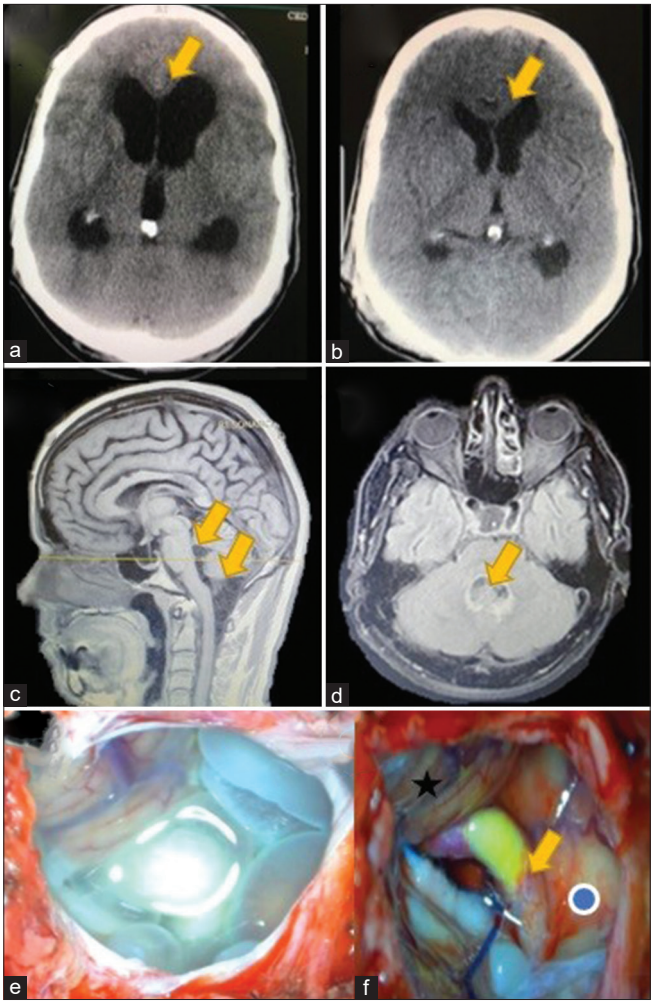

Figure 3: (a) Preoperative axial view of a head computed tomography (CT) scan demonstrating an acute hydrocephalus (yellow arrow) secondary to neurocysticercosis (NCC) and (b) postoperative CT scan showing resolution (yellow arrow) after cerebrospinal fluid shunt. (c) Sagittal view of a T1-weighted magnetic resonance imaging (MRI) showing a racemose NCC in the cisterna magna, extending to the fourth ventricle (yellow arrows). (d) Axial fluid-attenuated inversion recovery MRI demonstrating an intraventricular cyst with surrounding ependymitis (e) intraoperative view of racemose NCC inside cisterna magna, before excision (f) intraoperative view after cyst resection, medulla oblongata (blue dot), left posteroinferior cerebellar artery (yellow arrow) with an atheromatous plaque and right cerebellar tonsil (black star).

The Table 3 compares 17 different articles on various aspects of NCC. One notable observation is that the articles cover a wide range of topics related to the disease, including

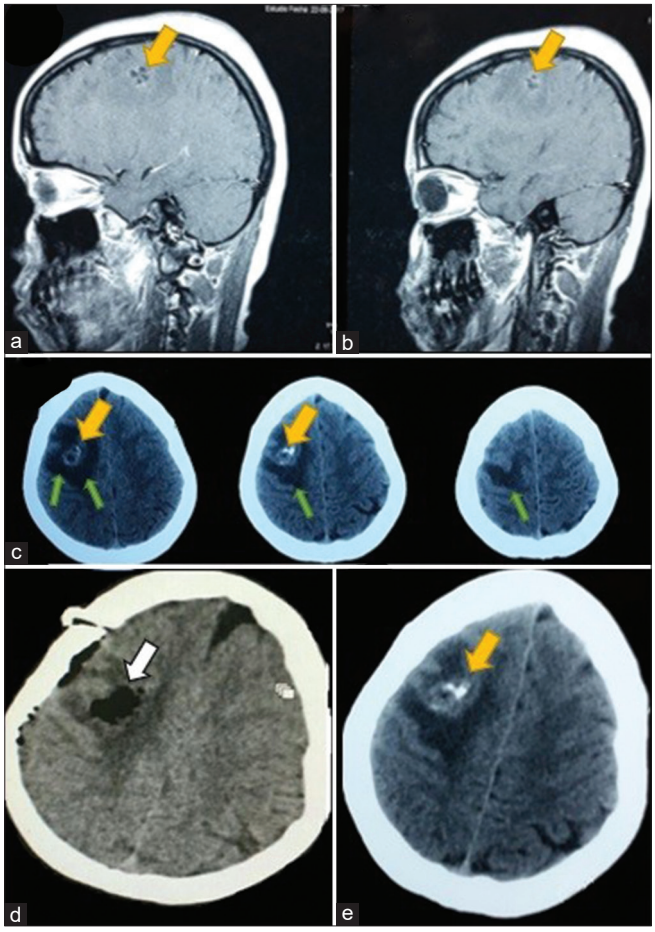

Figure 4: (a and b) Sagittal view of a T1-contrasted sequence magnetic resonance imaging showing a middle size neurocysticercosis cyst (yellow arrow). (c) Axial view of a head CT scan showing the cyst (yellow arrow) and surrounding brain edema (green arrows). (d) Postoperative CT scan demonstrating the complete resection of the cyst and (e) preoperative axial view of a head CT scan showing the compressing cyst (yellow arrow).

epidemiology, diagnosis, treatment, and management, among others. One of the most common themes throughout the articles is the prevalence of NCC in different regions [Table 3]. For example, Mafojane *et al*. (2003) provide an overview of the current status of the disease in Eastern and Southern Africa, while Cantey *et al*. (2014) focus on the prevalence of the disease in the United States.[3,11]

DISCUSSION

The mainstay treatment of NCC involves symptomatic therapy using cysticidal drugs (praziquantel and albendazole) as first-line drugs.[9] When seizures are part of the symptoms, antiepileptic drugs should also be administered. Steroid therapy may be required in some cases to control edema associated with the lesions. Surgery is indicated in the giant cyst and spinal NCC cases due to severe and progressive symptoms or acute neurological deterioration.[10]

Table 1: Del Brutto *et al*. diagnostic criteria for human neurocysticercosis.

Diagnostic criteria
Absolute criteria
• Histologic demonstration of the parasite from biopsy of a brain or spinal cord lesion
• Evidence of cystic lesions showing the scolex on neuroimaging studies
• Direct visualization of subretinal parasites by funduscopic examination
Major criteria
• Evidence of lesions highly suggestive of neurocysticercosis neuroimaging studies
• Positive serum immunoblot for the detection of anticysticercal antibodies
• Resolution of intracranial cystic lesions after therapy with albendazole or praziquantel
• Spontaneous resolution of small single-enhancing lesions
Minor criteria
• Evidence of lesions suggestive of neurocysticercosis on neuroimaging studies
• Presence of clinical manifestations suggestive of neurocysticercosis
• Positive cerebrospinal fluid enzyme-linked immunosorbent assay for detection of anticysticercal antibodies or cysticercal antigens
• Evidence of cysticercosis outside the central nervous system
Epidemiologic criteria
• Individuals coming from or living In an area where cysticercosis is endemic
• History of frequent travel to disease-endemic areas
• Evidence of a household contact with Taenia solium infection
Degrees of diagnostic certainty
Define diagnosis
• Presence of one absolute criterion
• Presence of two major plus one minor or one epidemiologic criteria
Probable diagnosis
• Presence of one major plus two minor criteria
• Presence of one major plus one minor and one epidemiologic criteria
• Presence of three minor plus one epidemiologic criteria

For practical purposes, these lesions can be classified according to the affected intracranial compartment (supratentorial, infratentorial, and spinal) or, more commonly: intraparenchymal/extraparenchymal (subarachnoid and intraventricular).[11] Sometimes, these parasites can mimic a brain tumor, as in one of our treated cases. Such lesions are more often associated with epileptic manifestations due to brain edema and cortical irritation. Small cystic lesions respond better to medical treatment and have a better prognosis if they are located into the brain

Table 2: Patients and surgical characteristics.

S. No.	Age and gender	Symptoms	Cyst location	Hydrocephalus	Duration of symptoms	Surgical technique	Intraoperative complication
1.	49/M	Left-sided hemiparesis/seizures/ altered mental status	Right lateral ventricle/ ipsilateral basal ganglia	Yes/No shunted	Six months	- Waterjet - Cyst drainage - Capsule coagulation - Traction	No
2.	44/M	Generalized seizures/headache/ altered mental status	Right Subarachnoid frontal lobe	No	Two years	- Drainage - Traction	Intraoperative cyst rupture
3.	52/M	- Severe headaches/drowsiness - Intermittent vomiting and dizziness	Posterior craniocervical subarachnoid	Yes/Shunted	- 2 weeks (hydrocephalus) - 4 months	- Sharp dissection - Water jet - Cyst drainage - Traction	No
4.	22/F	Headaches/Generalized seizures	Right intracerebral frontal lobe	No	One week	- Capsule coagulation - Traction	No

M: Male, F: Female

parenchyma than same found in giant cases; the latter trends to be resistant due to poor absorption by the cyst wall, causing mass effects besides the cysticerci death, as well as in the extraparenchymal form (presented in our cervicomedullary lesion patient).[12]

Giant cyst variants (over 5 cm in its larger diameter)[14] are rare. However, they cause mass effect symptoms or CSF flow obstruction, depending on the location. In those cases, surgery is preferred by cyst drainage alone or cyst removal if the pathology only causes brain compression.[14] In the setting of hydrocephalus, CSF diversion is recommended before the attempt of excision due to the inflammatory cascade surrounding the cyst, promoting arachnoiditis, and increased concentrations of proteins and cells that prolong hydrocephalus besides cyst removal. This manifestation depends on the host immune response, stage of the vesicular parasite, hydropic degeneration, and calcification[13,15] Lobato *et al*. found a 30% of hydrocephalus occurrence in their case report of 11 patients. Interestingly, those cases were nongiant cysts in its vast majority.[16]

According to Sotelo and Marin, the mortality rate for hydrocephalus secondary to cysticercosis arachnoiditis was 50% (41 patients) in a period of 2 years after shunting.[17] Due to purely mass effect obstruction of CSF, hydrocephalus seems somewhat less fatal than arachnoidal inflammation hydrocephalus. Nowadays, 30 years after those results, the mortality rate is lower with new therapeutic drugs and combinations. One of our two hydrocephalus patients died 12 days after surgery by septicemia. He had multiple complications due to nontreated HIV, high body index, and various NCC locations. He was extubated and alert, with mild deterioration of prior left-sided hemiparesis; 4 days after the procedure, he suddenly presented icteric skin, high hepatic markers, urea, and creatinine levels. Colli *et al*.[19] explained that patient status also should be considered. Severely deteriorated patients could benefit from cyst puncture and drainage through a burr hole and close follow-up. This author studied 160 surgically treated patients harboring all sizes of cystic NCC and found that most of them needed more than one procedure over time, with a complication rate of 30% and a mortality rate of 21.2%.

Microsurgical techniques are used for these pathologies, although it is not clear which of them are best suited.[18] We emphasize the role of preoperative imaging for preplanning, making a clear distinction between ventricular cyst extending into the brain parenchyma, complex racemose lesions extending from the cervicomedullary cistern into the fourth ventricle and cerebral aqueduct are of paramount help. Depending on how big the cyst is and the degree of vital nervous structures compressed, fluid drainage should be the first step; this reduces the main problem (mass effect) and allows surgeons to lower adjacent brain damage. This

Table 3: Different articles on various aspects of neurocysticercosis.

Reference	Region	Epidemiology	Diagnosis	Treatment	Complications
Mafojane *et al*. (2003)	Eastern and Southern Africa	High prevalence	Imaging techniques	Antiparasitic drugs	None reported
Winkler and Richter (2015)	Low- and middle-income countries	High burden	Improved diagnostic tools	Integrated management approach	Stigma and fear of transmission
Garcia *et al*. (2021)	Global	Eradicable cause of epilepsy	Improved diagnosis and surveillance	Integrated strategies and research	Stigma and misinformation about disease
Cantey *et al*. (2014)	United States	Low incidence	Imaging and serology	Antiparasitic drugs and symptomatic care	Few reported, mainly related to drug adverse effects
Garcia *et al*. (2020)	Global	High burden	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	None reported
Ndimubanzi *et al*. (2010)	Global	High burden	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	None reported
Ndimubanzi *et al*. (2010)	Global	High burden	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	None reported
Del Brutto *et al*. (1996)	Global	Low incidence	Clinical and imaging criteria	Antiparasitic drugs and symptomatic care	None reported
Li *et al*. (2019)	China	Uncommon presentation	Imaging techniques	Antiparasitic drugs and symptomatic care	None reported
Rajshekhar (2016)	Global	Improved diagnosis	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	None reported
Mahale *et al*. (2015)	Global	Various presentations	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	None reported
Umredkar *et al*. (2009)	India	Uncommon presentation	Imaging techniques and histopathology	Surgical removal of cysts and drugs	None reported
Paiva *et al*. (2017)	Brazil	Surgical treatment	Imaging techniques and histopathology	Surgical removal of cysts and drugs	Surgical complications
Fleury *et al*. (2011)	Global	Severe form of the disease	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	Neurological deficits and other complications
Rangel-Castilla *et al*. (2009)	Global	Various presentations	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	Surgical complications and adverse effects of drugs
Nash (2003)	Global	Improved management	Imaging techniques and serology	Antiparasitic drugs and symptomatic care	Hydrocephalus and other complications
Lobato *et al*. (1992)	Global	Various presentations	Imaging techniques and clinical signs	Shunting and antiparasitic drugs	Shunt-related complications

technique was performed in two cases due to the risk of jeopardizing the internal capsule and medulla oblongata.

After drainage, easy handling using graspers to apply some traction helps develop a cleavage plane that can be continued using waterjet dissection (instilling warm saline between the cyst wall and the brain) with a small feeding tube or a small catheter attached to a syringe. Sometimes waterjet dissection can remove the whole cyst if there are no adhesions between

the cyst wall and surrounding structures, similar to an Echinococcus hydatid cyst (Dowling-Orlando technique.[20] Capsule coagulation also helps shrink the cyst and makes the walls easier to hold, especially if the lesion has thick walls (a thin-walled cyst tends to open if coagulated or tractioned).

Another practical microsurgical maneuver is a sharp dissection (transection of arachnoid webs and fibrous adhesions) when the cyst is in the subarachnoid space, especially in basal cisterns, that allows cyst traction without brain, nerve, or vascular (torn veins) damage.[22] Care should be taken when using sharp dissection near a thin capsule cyst; it could tear the walls and cause fluid spreading, although this does not constitute further problems.[21]

The results of the studies we reviewed suggest that surgical management can be an effective treatment option for giant neurocysticerci, with good or excellent outcomes achieved in a majority of cases. However, the success rates varied depending on the location of the cyst and the surgical approach used. In terms of intracranial location, the studies we reviewed included patients with giant neurocysticerci in various locations, including the intraventricular region, brainstem, basal ganglia, and supratentorial region. Overall, the studies suggested that surgical management can be successful regardless of the cyst location, as long as the surgical approach is tailored to the specific location. In terms of surgical approach, the studies we reviewed included craniotomy with cystectomy or aspiration, endoscopic surgery, and stereotactic aspiration. The choice of surgical approach depended on various factors, including cyst size and location, as well as surgeon's preference and expertise. The studies suggested that both craniotomy and endoscopic surgery can be effective for removing giant neurocysticerci, but each approach has its own advantages and disadvantages. The studies also reported on complications associated with surgical management of giant neurocysticerci, including hemorrhage, infection, hydrocephalus, and neurological deficits. The incidence of complications varied across studies, but they were generally more common in patients who underwent craniotomy with cystectomy or aspiration compared to endoscopic surgery or stereotactic aspiration.

CONCLUSION

Surgical management is an important treatment option for giant neurocysticerci, especially in cases where medical management has failed or is contraindicated. The success rates and complication rates of different surgical approaches vary depending on the cyst location and surgeon expertise, but overall, surgical management can achieve good or excellent outcomes in a majority of cases. It is important for surgeons to carefully evaluate the patient's clinical and radiographic features and to tailor the surgical approach to the specific cyst location and characteristics. In addition, close postoperative monitoring is necessary to detect and promptly treat any complications.

There is no formal data about NCC in the Dominican Republic so far in the literature. Giant NCC variant is a rare presentation, comprising 0.4–5% of all cases, and constitutes a real endemic problem in our country. Populated prospective studies with larger sample sizes are needed to better understand the clinical and surgical nuances of these rare entities and noncystic type lesions.

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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