

Endoscopic endonasal approach for isolated subarachnoid neurocysticercosis in basal cisterns and its complications: illustrative case

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BACKGROUND Neurocysticercosis (NCC) is an infectious parasitic disease in which humans are the middle host in the life cycle of *Taenia solium*. It is currently considered to be a neglected tropical disease. According to their location, cysts can cause epilepsy, hydrocephalus, arachnoiditis, and intracranial hypertension. The subarachnoid is the rarest and most morbid form among all forms of NCC presentation.

OBSERVATIONS The authors report an odd case of subarachnoid NCC (SUBNCC). It was treated with expanded endoscopic endonasal surgery. Unfortunately, the patient developed a high-output nasal leak and meningitis after cyst removal, which demanded additional surgical procedures. Nonetheless, the patient showed a good clinical outcome after surgical interventions.

LESSONS Endoscopic endonasal approach of SUBNCC can be safe and curative. Nevertheless, the presented report shows that severe complications can arise from the procedure. The authors hypothesized that placing an early cerebrospinal shunt would reduce the patient's morbidity before difficulties arose.

<https://thejns.org/doi/abs/10.3171/CASE2229>

KEYWORDS neurocysticercosis; subarachnoid neurocysticercosis; sellar racemose neurocysticercosis; endoscopic endonasal approach

Neurocysticercosis (NCC) is a common parasitic disease caused by pork tapeworm cysts of *Taenia solium* in the central nervous system (CNS).¹ This disease is a significant health problem in Latin America, Africa, and some parts of Asia.² NCC is a neglected tropical disease. According to World Health Organization estimates, ~50 million people worldwide harbor neurocysticercosis, which causes ~50,000 deaths annually.³ It has three primary forms (parenchymal, ventricular, and subarachnoid neurocysticercosis [SUBNCC]), and clinical presentation varies accordingly: seizures, hydrocephalus, and intracranial hypertension, respectively. The SUBNCC usually shows cysts over the cerebral convexity in the sylvian fissure and basal cisterns.²⁻⁷ Basal cistern SUBNCC is the rarest and most morbid form. These cysts can grow, causing a mass effect in the adjacent cerebral areas. Indeed, they can cause endocrine abnormalities and visual deficits. These symptoms have similarities to those caused by pituitary tumors.^{1,4,7}

Treatment of SUBNCC is challenging because cysticide drugs generally achieve low concentrations in basal cisterns and intraventricular space and thus have insufficient efficacy. Hence, a surgical procedure is required in most cases.⁸

Herein, we discuss the case of a patient who presented with minimal symptoms of SUBNCC diagnosed using the expanded endoscopic endonasal approach and showed postoperative complications.

Illustrative Case

A 26-year-old man presented with progressive frontal headaches and dizziness episodes over the previous 2 years. On physical examination, no abnormality was noted. An expansive cystic lesion without contrast enhancement was shown on T1-weighted magnetic resonance imaging (MRI) at the retrochiasmatic space. Its dimensions were 3.4 × 2.9 × 3.2 cm, reaching the left prepontine

ABBREVIATIONS CNS = central nervous system; CSF = cerebrospinal fluid; ELD = external lumbar drain; MRI = magnetic resonance imaging; NCC = neurocysticercosis; SUBNCC = subarachnoid NCC.

INCLUDE WHEN CITING Published April 11, 2022; DOI: 10.3171/CASE2229.

SUBMITTED January 17, 2022. **ACCEPTED** February 18, 2022.

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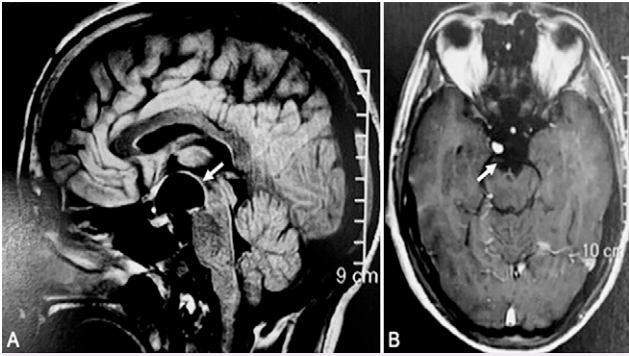


FIG. 1. A: Sagittal fluid-attenuated inversion recovery MRI shows a cystic lesion in the retrochiasmatic space. This lesion occupies the perimesencephalic cistern and displaces the floor of the third ventricle superiorly (arrow). It measures 3.4 × 2.9 × 3.2 cm. **B:** Axial T1-weighted MRI demonstrates a peripheral nodule with contrast enhancement (arrow).

cistern. The lesion displaced the third ventricle, optic nerves, and chiasma superiorly and the brain peduncles laterally. The fast imaging employing steady-state acquisition MRI showed a composite lesion of a hyperintense cyst with thin walls and a hypointense nodule (Fig. 1).

The primary imaging hypotheses were arachnoid cyst, craniopharyngioma, and SUBNCC. Additionally, we performed MRI of the entire spine, a neuroophthalmological examination, and hormonal evaluation. These evaluations did not show any alteration. Thus, a biopsy was performed using an expanded endoscopic endonasal approach.

After opening the dura mater of the suprasellar space during the proposed endoscopic approach, we proceeded with the arachnoid membrane dissection. We visualized the pituitary stalk in the middle of the operative field, the left optic chiasm on the ceiling, and a sessile lesion situated in the retrochiasmatic space (Fig. 2A and B). The capsule was a soft and slick and had a brownish-yellow appearance with no adherence to adjacent structures or tissue invasion. We proceeded with an excisional biopsy because gradual traction allowed it to be completely resected from the retrochiasmatic space (Fig. 2C). It is crucial not to provide excessive traction and cause cyst rupture. We observed an intense cerebrospinal fluid (CSF) flow through the surgical field. The closure was performed in a multilayer fashion using a dural substitute, a well-coaptated wide nasoseptal flap, Surgicel, and fibrin glue (Fig. 2D and E). No leak was observed with Valsalva maneuver after closure.

The histopathological analysis showed a lesion highly suggestive of neurocysticercosis in its racemose form. In this type, the scolex is not usually identified. However, it had the following characteristics: cystic cavity with a typical multilobulated grape-like cluster;

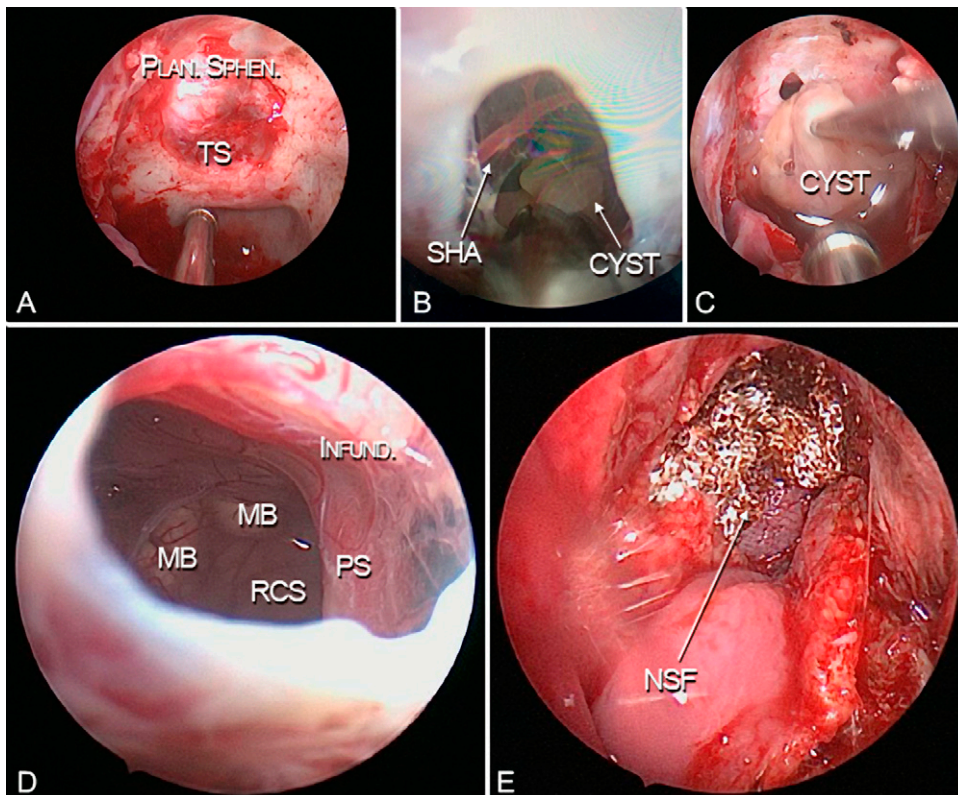


FIG. 2. A: The dural exposure achieved in the transplanum transtuberular approach. Plan. Sphen. = planum sphenoidale; TS = tuberculum sella. **B:** The dura mater is opened, and a sessile lesion is localized in the retrochiasmatic space. SHA = superior hypophyseal artery. **C:** The entire cystic lesion is removed. **D:** The mammillary bodies are visualized after complete lesion resection. Infund. = infundibulum; MB = mammillary bodies; PS = pituitary stalk; RCS = retrochiasmatic space. **E:** The skull base reconstruction is completed placing the nasoseptal flap in position. NSF = nasal septal flap.

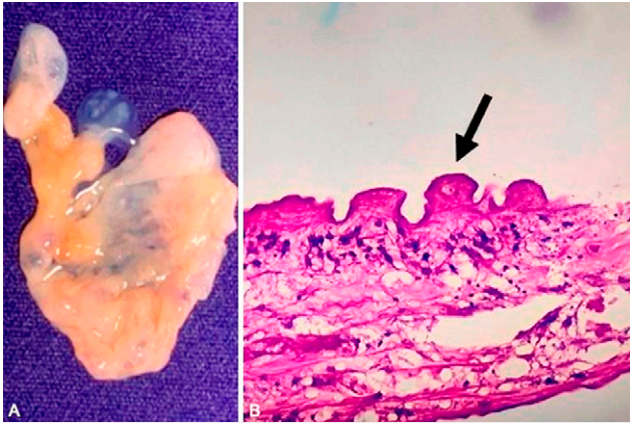


FIG. 3. A: Macroscopic characteristic of a cystic lesion with capsule soft and brownish-yellow in appearance. **B:** Double-layered eosinophilic membrane H&E histopathology image (original magnification $\times 40$) with a curved, wavy surface and numerous microvilli (black arrow).

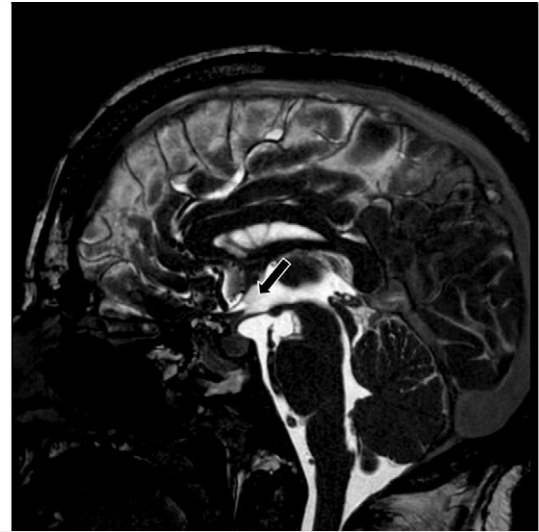


FIG. 4. Postoperative sagittal fast imaging employing steady-state acquisition MRI shows complete lesion resection (arrow).

numerous vesicles; double-layered eosinophilic membrane with a curved, wavy surface; and innumerable microvilli, which allowed the correct identification of the parasite (Fig. 3). Therefore, the patient's medical treatment was started by prescribing dexamethasone (12 mg/day) and albendazole (30 mg/kg/day) for 15 days.

After 5 days, the patient presented with severe headaches and rhinorrhea, showing a high-output CSF nasal leak. He was promptly treated with new endoscopic access and closure of the defect in three layers: fat, fascia lata graft, and nasoseptal flap. We also installed a CSF external lumbar drain (ELD) for 72 hours. He was discharged from the hospital on the fourth postoperative day.

After 5 days, the patient returned to the emergency department with a high-output nasal leak of CSF and signs of meningitis. Broad-spectrum antibiotics were prescribed for hospital-acquired meningitis for 15 days (cefepime 2 g three times a day and vancomycin 2 g twice a day). A new CSF ELD was placed, and during the lumbar puncture, the senior surgeon noted a high CSF pressure of >40 cm H_2O .

An endoscopic redo correction revealed necrosis of the nasoseptal flap's distal end secondary to multiple approaches. For reconstruction, we opted for a middle turbinate graft and triple-layer closure. After a week of antibiotic regimen and normalization of CSF parameters, we converted the ELD into a definitive lumbar peritoneal shunt. The postoperative brain MRI revealed complete lesion resection (Fig. 4). In addition, there was complete regression of symptoms.

Discussion

Observations

NCC is an infectious parasitic disease in which humans are the middle host in the life cycle of *Taenia solium*. The eggs are ingested and grow in the CNS. Although this disease is considered potentially eradicable, it is still endemic in many developing countries.^{1-3,5,6,9} Most imported cases come from Latin America, Spain (47.5%), France (16.7%), and Italy (8.3%). In Latin America, the average prevalence of NCC is 10%, varying among countries: Brazil 3.0%–5.6%; Mexico 1.3%–10%; Bolivia 22%; and Venezuela 4%–36.5%.³

NCC is one of the most common parasitic diseases of the CNS, occurring in brain tissue, spinal cord, subarachnoid space, and ventricular system. The racemose form is expected in the two last

locations, in which the scolex is not visible inside the cyst. On the other hand, sellar, suprasellar, or retrochiasmatic region presentation, as in our patient's case, is exceedingly rare and has seldom been published.^{4,7-10}

Most patients with intrasellar NCC present with a mass effect on adjacent cerebral areas. Therefore, ophthalmic and endocrine symptoms are the most common alterations. These are also featured in other sellar lesions, such as arachnoid cysts, pituitary adenomas, Rathke's cleft cysts, and craniopharyngiomas, hindering clinical diagnosis. Our patient reported only unspecific complaints such as worsening headaches and occasional dizziness.^{4,7-9,11,12}

Imaging can aid the diagnosis.¹ However, on top of a challenging clinical diagnosis, neuroimaging can present nonspecific findings for NCC, especially in its subarachnoid sellar form. These lesions can mimic arachnoid cysts or tumors usual to the location, often more common than the parasitic disease itself.^{4,7,8} Imaging findings of NCC are T1-weighted MRI hypointense and T2-weighted MRI hyperintense cyst and a minimal contrast enhancement of the walls and nodule (the scolex). Other sellar lesions, such as craniopharyngiomas, pituitary adenomas and arachnoid cysts, reveal different aspects on imaging: the first two present with vivid contrast enhancement and the last one presents without enhancement. When the scolex is absent, as in the racemose form presented by our patient, the examination interpretation can be even more difficult. In our case, a biopsy was performed to confirm the diagnosis.^{7,8,11-13}

Diagnosis of NCC is criteria-based. There are four criteria groups: absolute, major, minor, and epidemiologic.^{9,10} Our patient had a feature from the first group: microscopy finding of a multilobate cyst with fibrous walls and inflammatory nature and an abundance of vesicles in a grape-like distribution, without a visible scolex.⁸

The treatment recommended for racemose NCC is albendazole (15–30 mg/kg/day) for 15 to 30 days. We can add corticosteroids before and during treatment with antiparasitic drugs. Cysticidal action in SUBNCC has a significant drawback: an inadequate drug concentration is achieved in those spaces. Thus, resection is needed to aid treatment, as was performed in our patient.^{4,10,14}

The endonasal endoscopic approach is a safe and effective surgical option for sellar and suprasellar SUBNCC.^{1,4,7} We chose this approach because it is minimally invasive, resulting in acceptable surgical morbidity. However, the closure can be difficult, generating complications because patients with SUBNCC present a significant basal intracranial hypertension state. Therefore, in cases of high diagnostic suspicion of NCC, placing an early cerebrospinal shunt should be considered to reduce a patient's morbidity before complications arise. Unfortunately, our patient developed a high-output nasal leak and meningitis, requiring multiple surgeries. Meanwhile, we believe in the necessity for further studies to prove the shunt's efficacy simultaneously to the endonasal endoscopic approach.

Lines et al. recently brought another perspective to this procedure in a case of SUBNCC.¹⁵ The endoscopic endonasal approach was also conducted to evacuate multiple large tapeworm cysts in the basal and sylvian cisterns without postoperative complications. The cysts were removed with light suction or gentle forceps traction. The multilayer gasket seal technique for reconstruction was performed in three planes, with a dura mater replacement, bone pouch, nasoseptal flap, and fibrin sealant. Furthermore, the patient had previously received a ventriculoperitoneal shunt due to hydrocephalus, relieving intracranial hypertension.¹⁵

In our patient, the lumbar peritoneal shunt placement allowed complete hypertension pressure relief, thus supporting the nasal leak's resolution after a multilayered endonasal endoscopic reconstruction. Afterward, the patient's condition significantly improved in the 3 months of follow-up. Finally, we presented a patient with basal cistern NCC, which showed a good clinical outcome after multiple surgical interventions.

Lessons

This case is the second report of basal cistern SUBNCC treated with an endoscopic endonasal approach. Although this treatment can be curative, the presented case shows the importance of observing for complications. A high-output CSF fistula, for instance, may result in a worse prognosis due to the infectious risk. Therefore, even while more studies are needed to prove our hypothesis, we believe that placing an early cerebrospinal shunt may reduce a patient's morbidity before complications arise.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Goulart, Vieira Netto, Leão, Alencar. Acquisition of data: Cavalcante, Goulart, Vieira Netto, Marques, Leão, Alencar. Analysis and interpretation of data: Goulart, Vieira Netto, Marques, Leão, Alencar. Drafting the article: Goulart, Vieira Netto, Marques, Alencar. Critically revising the article: Cavalcante, Goulart, Marques, Alencar. Reviewed submitted version of manuscript: Goulart, Vieira Netto, Dias, Moraes, Marques, Leão, Alencar. Approved the final version of the manuscript on behalf of all authors: Cavalcante. Statistical analysis: Alencar. Administrative/technical/material support: Goulart, Dias, Moraes, Marques, Alencar. Study supervision: Cavalcante, Goulart, Vieira Netto, Alencar, Godoy.

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