CEREBROSPINAL FLUID LEAKAGE AND BONE EROSION CAUSED BY CYSTS IN BASAL CYSTERN NEUROCYSTICERCOSIS TREATED BY ENDOSCOPY

Fístula de líquido cefalorraquídeo y erosión ósea por quistes en neurocisticercosis de cisterna basal tratada por endoscopía

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ABSTRAC

Introduction: The extra-parenchymal neurocysticercosis and the racemose form are very predisposed to complications. Subarachnoid sellar cysts are rare, are associated with intracranial hypertension and disorders visual fields.
Clinical case: A 63-year-old male patient with racemose neurocysticercosis, hydrocephalus and cerebrospinal fluid fistula. He underwent endonasal endoscopy, removal of cysts from the sphenoid sinus, sellar, suprasellar, and prepontine regions, and fistula closure. He also presented erosion in the temporal bone and dural fistula, which were closed through microsurgery and endoscopy. The patient had a favorable initial evolution, with spastic quadriparesis, which improved with rehabilitation. Subsequently he presented episodes of ventriculoperitoneal shunt system dysfunction.
Conclusion: Neuroendoscopy is a diagnostic and therapeutic method of various forms of neurocysticercosis. Extra-parenchymal neurocysticercosis can produce bone and dural erosion, so must be considered in the differential diagnosis of cerebrospinal fluid.

Keywords Neurocysticercosis, Fistula, Neuroendoscopy, Sphenoid sinus, Cysts *(source: MeSH NLM)*

RESUMEN

Introducción: La neurocisticercosis extraparenquimal y la forma racemosa, tienen gran predisposición a complicaciones. Los quistes subaracnoideos en la región selar son raros, se asocian a hipertensión endocraneal y trastornos de campos visuales.
Caso clínico: Paciente varón de 63 años con neurocisticercosis racemosa, hidrocefalia y fístula de líquido cefalorraquídeo. Fue sometido a endoscopía endonasal, evacuación de quistes del seno esfenoidal, región selar, supraselar, prepontina, y cierre de fístula. Además, presentó erosión en el peñasco izquierdo y fístula dural, que se cerraron con ayuda de microcirugía y endoscopía. El paciente tuvo evolución inicial favorable, con cuadriparesia espástica, que mejoró con rehabilitación. Posteriormente presentó episodios de disfunción de sistema de derivación ventrículo-peritoneal.
Conclusión: La neuroendoscopía constituye un método diagnóstico y terapéutico de diversas formas de neurocisticercosis. La Neurocisticercosis extraparenquimal es capaz de producir erosión ósea y dural, por lo que se debe tener en cuenta en el diagnóstico diferencial de fístula de líquido cefalorraquídeo.

Palabras clave: Neurocisticercosis, Fístula, Neuroendoscopía, Seno esfenoidal, Quistes *(fuente: DeCS Bireme)*

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Neurocysticercosis is the most frequent parasitic disease of the human central nervous system, endemic in Latin America, India, regions of Asia and Africa. In the United States and Europe, it occurs with some frequency, mostly in immigrants. It is caused by the larvae of the cestode Taenia solium. In Latin America, cerebral cysticercosis with cystic forms is common, while in Asia, subcutaneous cysticercosis is common.

In general, neurocysticercosis presents as intraparenchymal forms, associated with convulsive symptoms, or extra-parenchymal forms, including subarachnoid or intraventricular cysts. The subarachnoid cysts can reach large size and form lobulations (appearance of bunch of grapes), lose the scolex, are usually located at the base of the brain or in the Sylvian fissure, and more rarely in the convexity. The subarachnoid cysts of the base lead to hydrocephalus, while those of the Sylvian or convexity fissures cause a mass effect (headache, neurological focality, seizures and deterioration of consciousness).

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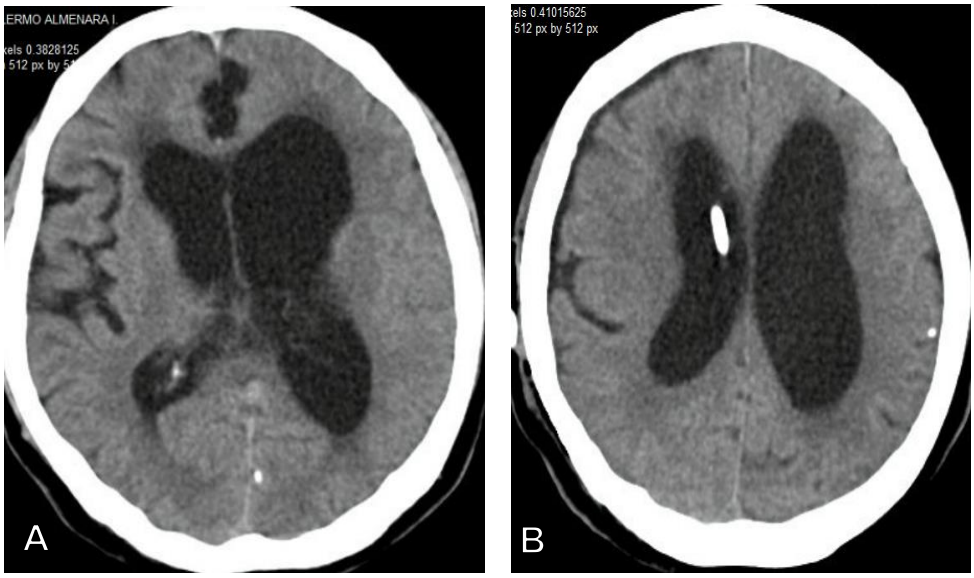

Fig 1. (A) Initial tomographic image March 2015 showing ventricular dilatation and transependymal edema, **(B)** Post operative and presence of ventriculoperitoneal shunt in right ventricle and moderate hydrocephalus

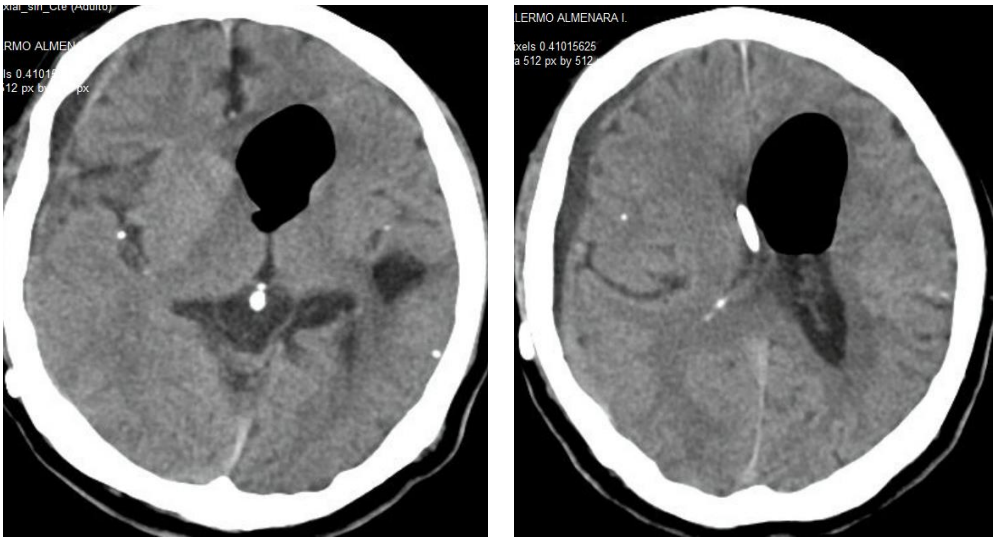

Fig. 2. Tomography without contrast in axial view showing pneumocephalus in the frontal horn

We present the case of a patient diagnosed with neurocysticercosis, who presents with hydrocephalus associated with subarachnoid cysts and cerebrospinal fluid fistula at the base of the skull.

CLINICAL CASE

History and examination: A 63-year-old male patient with a history of neurocysticercosis since 2010 (positive Western blot for cysticercosis - 6 bands), and hydrocephalus of 1 year of evolution associated with gait instability, nausea and vomiting, progressive right hemiparesis. He entered the emergency room at the Guillermo Almenara Irigoyen National Hospital (HNGAI) in March 2015 due to exacerbation of symptoms. He underwent a brain CT without contrast that identifies multiple cerebral calcifications and ventricular dilatation (Figure 1), so he was placed right ventriculoperitoneal shunt system (VPS), discharged with good evolution.

After two months of discharge, she presented postural instability associated with mild head trauma, with subsequent disorientation and progressive drowsiness. He attended emergencies of the HNGAI, and cerebral CT showed pneumocephalus (left frontal horn), empty sella image, which invaded part of the sphenoid sinus with erosion in the anterior wall (Figure 2), which was already visualized in images previous (Figure 3).

Treatment: At 5 months after VPS, the patient underwent a transnasal endoscopic approach (Figure 4), evidencing fluid leakage through the bone defect of the anterior wall of the sphenoid sinus, proceeding to sphenoidectomy with extraction of multiple cysticercus cysts from sphenoid sinus, Sella turcica and suprasellar and prepontin cisterns.

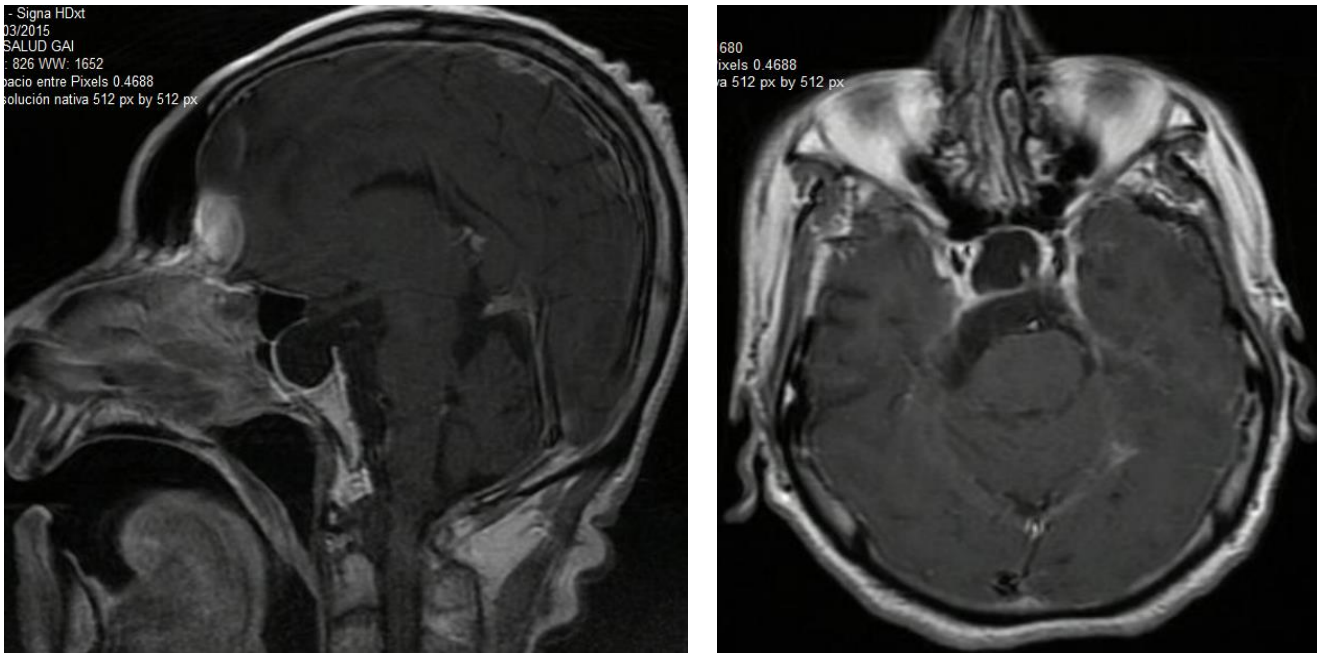

Fig 3. Magnetic resonance imaging (MRI) with gadolinium in sagittal and axial sections, prior to peritoneal ventricular bypass surgery, showing enlarged sella turcica apparently filled with cerebrospinal fluid covering almost the entire sphenoid sinus.

Hermetic closure of the fistula was performed, placing a small amount of fat wrapped in oxidized cellulose (Surgicel) inside the sella turcica, another similarly within the sphenoid sinus and finally the defect was covered with a mucosal graft obtained from the middle turbinate. (Figure 5)

The anatomopathological study of the sample confirmed neurocysticercosis. Subsequently, the patient presented with ventriculitis, receiving broad spectrum antibiotic treatment, the ventriculoperitoneal shunt (VPS) system was removed, and an external ventricular drain (EVD) was placed one month after the endoscopic approach. The persistent pneumocephalus forced us to study images in detail, identifying probable left anterior fistula of the left rock (Figure 6A), which was confirmed after surgery performed 2 months after the transnasal endoscopic approach, using pterional and subtemporal craniotomy, using a microscope. and endoscope. Bone erosion was found in the anterior superior aspect of the petrous temporal bone and dural fistula (Figure 6B), which were hermetically sealed with bone wax, superimposed layers of hemocollagen (Gelfoam), fat and suture of the dura with silk 4/0.

Evolution: The patient evolved favorably, being awake, with EG 13 points (O4, V3, M6), persisted with moderate spastic quadriparesis, which improved with rehabilitation

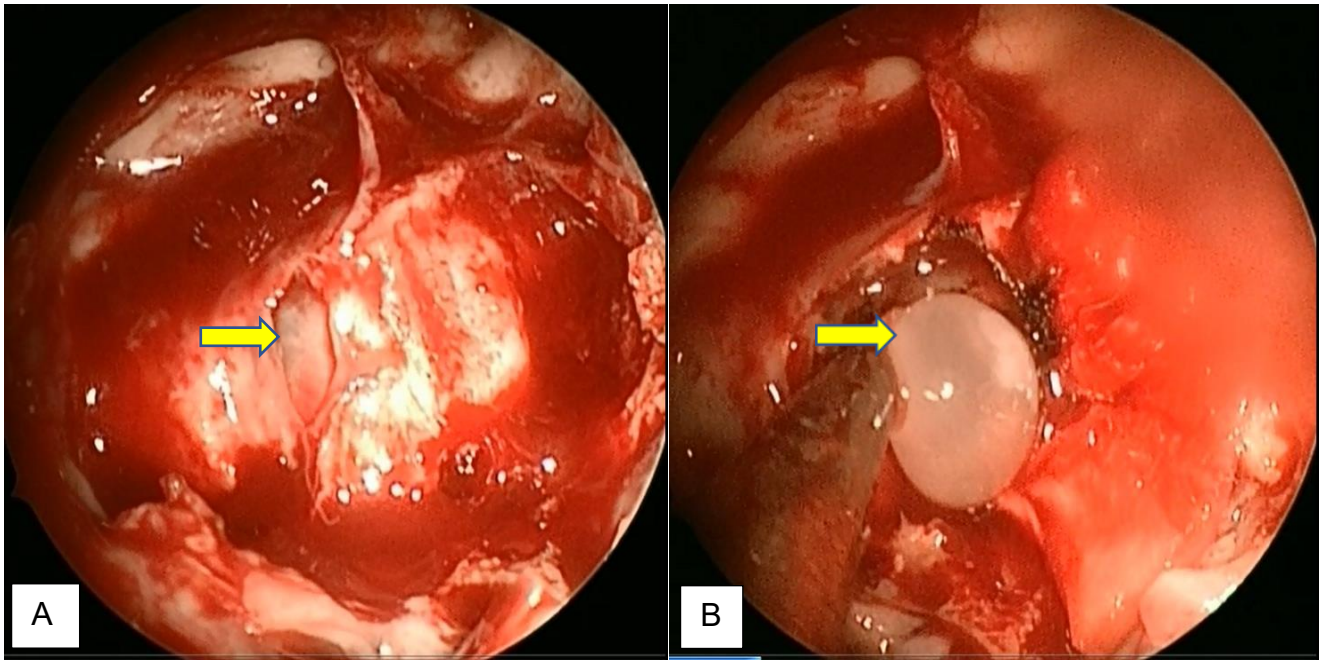

Fig 4. (A) Transnasal endoscopic approach showing evidence of bone defect in the anterior wall of the sphenoid sinus. **(B)** Output of cysticercus cysts through it.

over time. Posteriorly, the patient presented multiple episodes of valvular dysfunction, with multiple admissions and repetitive replacement surgeries, with a progressive deterioration of his health status.

DISCUSSION

Depending on the place where the cysts of the parasite are housed, neurocysticercosis is classified as medullary, parenchymal and extraparenchymal (which includes the subarachnoid and intraventricular forms). The latter is rare, and the combined presentation is exceptional[1]. The extraparenchymal form usually presents an aggressive behavior, given its great predisposition to produce complications[2]. The subarachnoid cysts of neurocysticercosis can form lobulations (cluster aspect), lose the scolex and are usually located at the base of the brain, Silvio's fissure, and more rarely in the convexity.

The neurocysticercosis racemosa differs from the parenchymal form. It originates from the aberrant proliferation of cestode larvae, is clinically more aggressive, and is reported in 15-54% of patients[13]. It usually presents as a meningeal, intraventricular or subarachnoid (cisternal) form. It produces meningitis and adhesions that result in hydrocephalus. It can trigger vasculitis and entrapment of cranial nerves in the inflammatory exudate. In the intraventricular and subarachnoid form, the oncospheres reach the ventricles through the choroidal plexuses. The death of the larvae causes ependymitis, which contributes to the production of hydrocephalus[13,15]. It is considered a

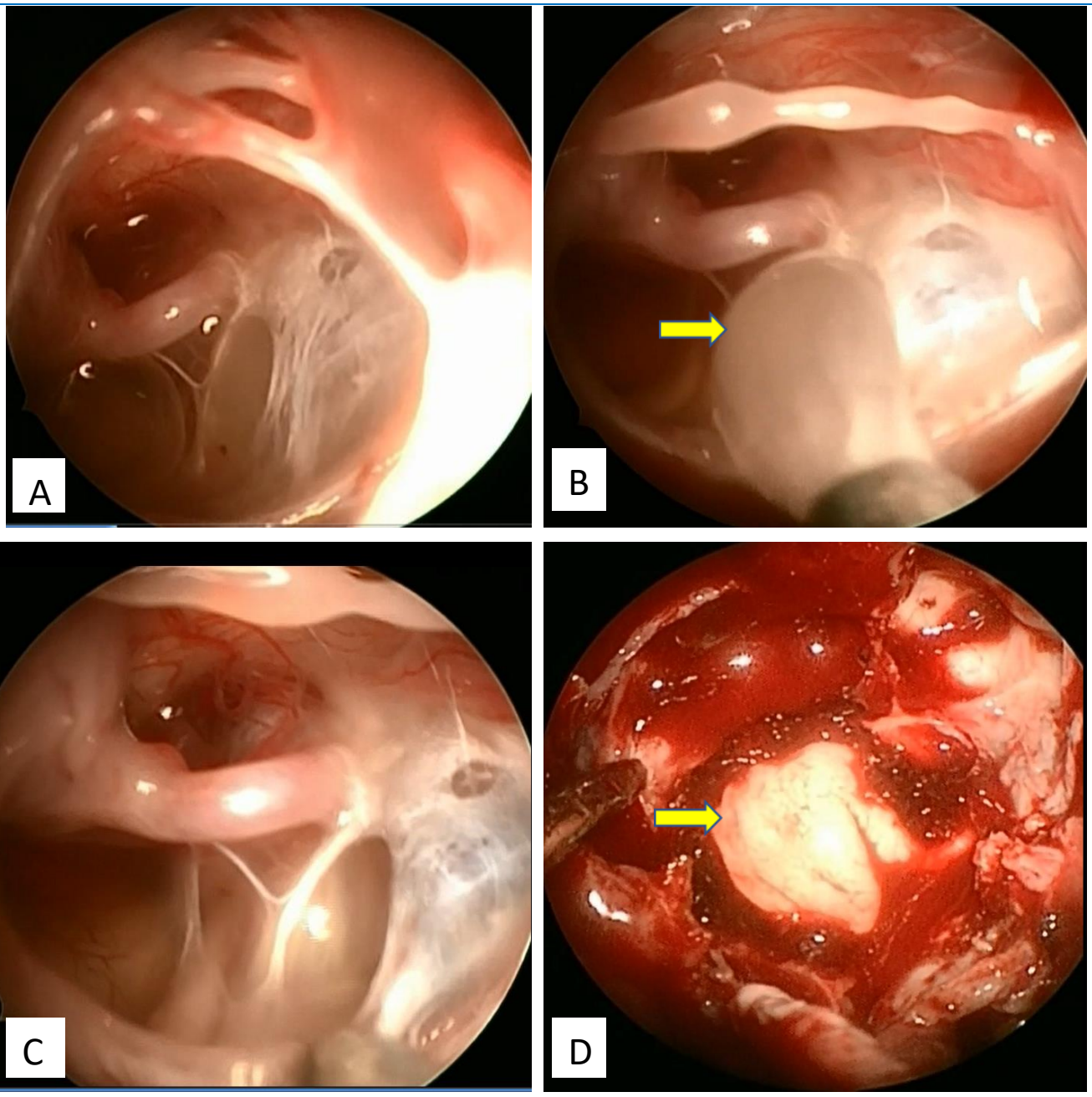

Fig.5: (**A, B, C**) Endoscopic view of cysticercus cysts extraction from the sella turcica, in addition the thickened arachnoid can be seen due to arachnoiditis caused by parasitic infection. (**D**) Bone defect covered with middle turbinate mucosal flap.

malignant form of infection if it is associated with hydrocephalus secondary to cysticercus meningitis, reaching a mortality rate of 50%.

In our case, the intense cisternal inflammatory reaction is evidenced by endonasal endoscopy, as a consequence of the racemic neurocysticercosis that the patient suffers (Figures 4 and 5). This would be related to the development of bone and dura mater erosion, with the subsequent cerebrospinal fluid fistula suffered by the patient, which, according to our literature review, has not been identified in another case report of patients with neurocysticercosis.

The cysts of subarachnoid cysticercus can reach a large size when they are located in a wide fissure such as Silvio's, and it is called giant racemose disease when the size of the vesicles is greater than 50 mm, as in the case presented by Kumar et al[3]. The subarachnoid cysts of the base lead to hydrocephalus, while those of the Silvio or convexity sulci cause mass effect[4]. Extraparenchymal neurocysticercosis is associated with a local inflammatory response with a high concentration of proteins and cells in the cerebrospinal fluid, which contributes to the development of hydrocephalus and the high rates of dysfunction of the ventriculo-peritoneal shunt systems, which exceed 50% at 5 years[5,12].

Neurocysticercosis, in particular the subarachnoid form, is also associated with other complications. It can cause cerebrovascular disease, including infarction, transient ischemic attack and intraparenchymal hemorrhage. The most common mechanism associated is the development of arteritis[6,7]. In rare cases, Neurocisticercosis racemosa can cause dementia. However, 66-87% of patients with neurocysticercosis have reported cognitive disturbances, and dementia or severe cognitive deterioration has been reported in 12-15% of patients[8], which would include the patient in our case report, who presented important commitment of their higher mental functions.

Cases of subarachnoid cysts in the sellar region are rare, and are frequently associated with intracranial hypertension secondary to hydrocephalus, visual field disorders such as hemianopsia or amaurosis, and endocrinological compromise (amenorrhea, weight gain) [9,10]. Arriada et al[11] reported the case of a 25-year-old patient with neurocysticercosis who debuted with the aforementioned condition, in whom the brain tomography with focus in the sellar region showed the presence of a cystic lesion with partial erosion of the sella turcica and the sphenoid sinus. , associated with displacement of the pituitary gland and optic chiasm. In this case, the surgical treatment was also performed transsphenoidal, achieving partial neurological improvement.

In our case report, we present a patient with an established diagnosis of neurocysticercosis, which develops hydrocephalus, which was initially treated by ventriculoperitoneal shunt. However, in the course of time despite being controlled intracranial hypertension, develops cerebrospinal fluid fistula, pneumocephalus and meningocele in the sphenoid sinus. The possibility of post-traumatic fistula associated with the aforementioned findings was initially raised, however, when performing the transnasal neuroendoscopy, the presence of a cysticercus cyst in the sphenoid sinus with erosion of the wall was evidenced, which would be the cause of the fistula. of the patient. In the same way, the presence of multiple cysts in the sellar, suprasellar and prepontine region was demonstrated. Finally, the diagnoses of hydrocephalus, neurocysticercosis and cerebrospinal fluid fistula were confirmed through the sella turcica, sphenoid sinus and through the petrous portion of the temporal bone, by direct vision with the endoscope.

We must consider this form of presentation as a differential diagnosis in a patient with subarachnoid extraparenchymal neurocysticercosis who presents with hydrocephalus and cerebrospinal fluid fistula. With the help of the

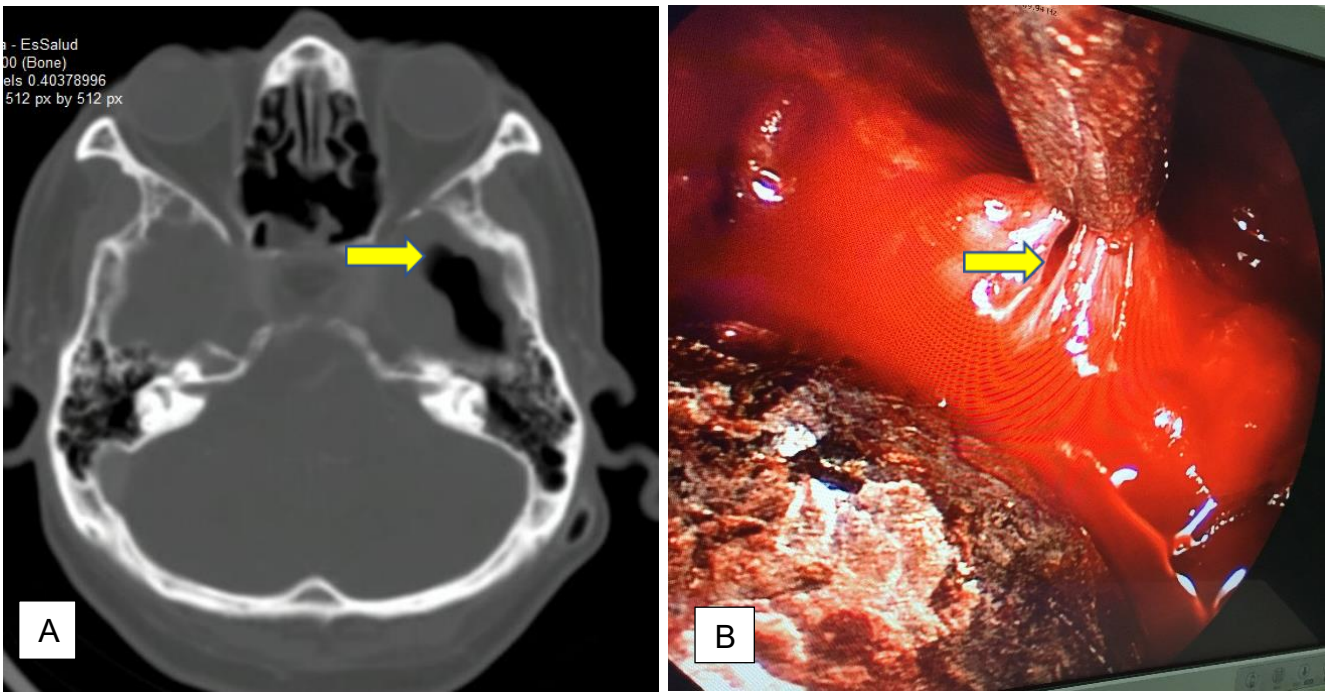

Fig. 6. (A) Brain CT in bone window (axial view) showing evidence of bone defect in the anterior wall of the sphenoid sinus. **(B)** Transnasal Endoscopic Approach where an output of cysticercus cysts through bone defect is showed.

transsphenoidal neuroendoscopy, the definitive diagnosis can be reached, and the removal of the cysts can be achieved as part of the treatment.

In the various reports of patients with the extraparenchymal form of the disease, neuroendoscopy has proved to be very useful, since it allows visualization and direct extraction of the cysts. Neuroendoscopy allows the management of post-hemorrhagic and post-infectious secondary hydrocephalus up to 60%, [16] although studies are lacking in post-infection due to neurocysticercosis, bearing in mind that antiparasitic drugs do not completely defeat the extraparenchymal type.

There is scarce literature on subarachnoid cysts that erode the sphenoid sinus, and after an exhaustive review of the literature, we have not found reports of patients with fistula associated with subarachnoid extraparenchymal neurocysticercosis as in our case, which also presented fistula and erosion in two bone points (Sella turcica floor and anterosuperior wall of the left petrous portion of temporal bone), so this case becomes of interest in the field of neurosurgery.

CONCLUSION

The endoscope is a fundamental diagnostic and therapeutic method of various forms of presentation of neurocysticercosis, thanks to its versatility of approach via ventricular, endonasal and parenchymal. The extraparenchymal neurocysticercosis is able to produce dura mater fistula and bone erosion, so in our environment we must take it into account in the differential diagnosis of cerebrospinal fluid fistula.

REFERENCES

1. Amaral L, Maschietto M, Maschietto R, Cury R, Ferreira NF, Mendonca R, Lima SS. Ununsual manifestations of neurocysticercosis in MR imaging: Analysis of 172 cases. ***Arq Neuropsiquiatr. 2003;61*:** 533-41.
2. Sanchez-Larsen A, Monteagudo M, Lozano-Setien E, et al. Neurocisticercosis racemosa subaracnoidea gigante y ventricular: a propósito de un caso. ***Rev Argent Microbiol. 2015*;47**(3):201-205
3. Kumar S, Thakur S, Jhobta A, Sood RG. Giant racemose neurocysticercosis with mass effect: Unusual presentation. ***Ann Indian Acad Neurol. 2013*;16**: 398-9.
4. Bickerstaff E, Cloake P, Hughes B, Smith W. The racemose form of cerebral cysticercosis. ***Brain. 1952;75***(1):116
5. Cardenas G, Jung H, Rios C, Fleury A, Soto-Hernandez JL: Severe cysticercal meningitis: clinical and imaging characteristics. ***Am J Trop Med Hyg 2010,82***(1):121-125.
6. Barinagarrementeria F, Del Brutto OH. Lacunar syndrome due to neurocysticercosis. ***Arch Neurol 1989;46***: 415-417.
7. Soto-Hernandez JL, Gomez-Llata Andrade S, Rojas-Echeverri LA, Texeira F, Romero V. Subarachnoid hemorrhage secondary to a ruptured inflammatory aneurysm: a possible manifestation of neurocysticercosis: case report. ***Neurosurgery 1996;38***: 197-199; discussion199-200.
8. Ciampi de Andrade D, Rodrigues CL, Abraham R, Castro LH, Livramento JA, Machado LR, et al. Cognitive impairment and dementia in neurocysticercosis: a cross-sectional controlled study. Neurology 2010;74: 1288-1295.
9. Del Brutto OH, Guevara J, Sotelo J. Intrasellar cysticercosis. ***J Neurosurg 1988;69***: 50 –60
10. Rafael H, Gómez S. Intrasellar cysticercosis. J ***Neurosurg1985; 63***: 975–97
11. Arriada N, Celis M, Higuera J, Corona T. Imaging Features of Sellar Cysticercosis: Case Report. ***Am J Neuroradiol 24***:1386–1389, 2003
12. Suastegui R, Soto-Hernández J, Sotelo J. Effects of prednisone on ventriculoperitoneal shunt function in hydrocephalus secondary to cysticercosis: a preliminary study. ***J Neurosurg 1996; 84***: 629-33.
13. Rohan R, Anish-Mehta, Srinivasa-Rangasetty. Extraparenchymal (Racemose) Neurocysticercosis and Its Multitude Manifestations: A Comprehensive Review. J Clin Neurol 2015;11 (3):203-211. Bangalore, Karnataka, India. http://dx.doi.org/10.3988/jcn.2015.11.3.203.
14. Takayanagui OM, Odashima NS. Clinical aspects of neurocysticercosis. ***Parasitol Int 2006;55*** Suppl: S111-S115.
15. Lobato R, Lamas E, Portillo J, Roger R, Esparza J, Rivas J, et al. Hydrocephalus in cerebral cysticercosis. Pathogenic and therapeutic considerations. **J *Neurosurg 1981;55***: 786-793.
16. Spyros Sgouros, Neuroendoscopy, ***Curren status and future trend based in the 5th World Congress of Neuroendoscopy in Athens, Greece. 2009***. Page 49, 4.5.

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: Flores JD, Malca J. *Drafting the article:* Flores JD. Critically revising the article: Flores JD. *Reviewed submitted version of manuscript:* Flores JD. *Approved the final version of the manuscript on behalf of all authors:* Flores JD.

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