Cyst-ventricular shunt by neuroendoscopic approach. First case of its kind in Guyana

Derivación cisto-ventricular por abordaje endoscópico. Primer caso de su tipo en Guyana. Presentación de caso

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Resumen

Los quistes aracnoideos son colecciones extracerebrales benignas formadas por líquido cefalorraquídeo rodeado de una membrana aracnoidea que histológicamente es indistinguible de la aracnoides sana. Su etiología la hipótesis más aceptada es la embrionaria según la cual se producen por una disgenesia en el desarrollo embriológico del espacio subaracnoideo. En este caso clínico presentamos a una paciente de 24 años de origen venezolano, actualmente residente en Guyana, quien presenta convulsiones tónico clónicas generalizadas y luego de estudios imagenológicos se evidenció un quiste aracnoideo gigante en la región temporal derecha, por lo que realizamos un abordaje endoscópico con el objetivo de crear una comunicación del quiste al sistema ventricular. Para realizar el procedimiento quirúrgico, que consistió en realizar una derivación cistoventricular endoscópica, se utilizó un endoscopio rígido Karls Storz con óptica de 30 grados, durante la cirugía tuvimos varias dudas retadoras que fueron resueltas a pesar de lo inusual del procedimiento, en cuanto a la selección del sitio de abordaje y a pesar de todos los inconvenientes anatómicos y la planeación acordada contrario a lo que usualmente se hace en la mayoría de los casos, los resultados fueron satisfactorios. La evolución postquirúrgica de nuestro paciente fue realmente satisfactoria, no se observaron complicaciones inmediatas ni tardías, tampoco se observaron secuelas relacionadas con la cirugía.

Palabras clave: Quistes aracnoideos, derivación quiste-ventricular, abordaje endoscópico.

Abstract

Arachnoid cysts are benign extracerebral collections formed by cerebrospinal fluid surrounded by an arachnoid membrane that is histologically indistinguishable from the healthy arachnoid. Its etiology the most widely accepted hypothesis is the embryonic one according to which they are produced by a dysgenesis in the embryological development of the subarachnoid space. In this clinical case we present a 24-year-old patient of Venezuelan origin, currently living in Guyana, who presents generalized tonic clonic seizures and after imaging studies showed a giant arachnoid cyst in the right temporal region, therefore we performed an endoscopic approach with the aim of creating a communication from the cyst to the ventricular system. To perform the surgical procedure, which consisted of performing an endoscopic cyst-ventricular shunt, a rigid Karls Storz endoscope with 30- degree optics was used, during the surgery we had several challenging questions that were resolved despite the unusual nature of the procedure, in terms of the selection of the approach site and despite all the anatomical inconveniences and the agreed planning contrary to what is usually done in most cases, the results were satisfactory. The post-surgical evolution of our patient was really satisfactory, no immediate or late complications were observed, no sequelae related to surgery were observed.

Keywords: Arachnoid cysts, cyst-ventricular shunt, endoscopic approach.

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Introduction

Endoscopic techniques have been used in neurosurgery since the beginning of the 20th century[1-5]. In 1910, urologist Even Victor.

L 'Espinasse performed endoscopic excision of the choroid plexuses in two patients with hydrocephalus and then in 1922 Walter Dandy replicated it in a single patient, obtaining a poor result. Due to the high rate of morbidity and mortality, this technique fell into disuse until 1970, when interest in it re-emerged thanks to new technological advances in optics and lighting systems[1-3,6-8].

In paraventricular cystic lesions and intraventricular pathology, neuroendoscopy is a minimally invasive, safe method with a favorable cost-benefit ratio and a low rate of complications compared to the classic microsurgical approach. Currently, the indications for this technique have expanded considerably; there are multiple articles in the literature on its use in different entities, such as hydrocephalus, intracranial cysts, intraventricular tumors, hypothalamic hamartomas, and skull base tumors[2,3,5,9,10].

The first description of cerebral arachnoid cysts was given by Richard Bright in the second volume of his Reports of Medical Cases, where he described them in 1831 as serous cysts in connection with the arachnoid[13]. Arachnoid cysts were subsequently given names such as circumscribed serous meningitis[14], pseudotumours of the brain[15] or chronic arachnoiditis[16]. Arachnoid cysts are benign extracerebral collections formed by cerebrospinal fluid surrounded by an arachnoid membrane that is histologically indistinguishable from the healthy arachnoid. Although they have been described in all ages, they are characteristic of childhood, when they represent up to 1% of all space-occupying intracranial lesions[11,12].

Regarding the etiology of arachnoid cysts, the most widely accepted hypothesis is the embryonic one[12,17,18] according to which they are produced by a dysgenesis in the embryological development of the subarachnoid space. Around day 35 of gestation, a loose membrane of mesenchyme appears between the neural tube, the somites and the notochord, which is the primitive meninge that will later differentiate into the pia mater, arachnoid and dura mater. On day 45 of embryonic development, the primitive meninge condenses, cavitates and forms a common space called the subarachnoid- subdural space.

The subarachnoid space develops in the fourth month of intrauterine life when the choroid membrane of the fourth ventricle is perforated, resulting in the release of cerebrospinal fluid from the primitive ventricular system with the formation of the cisterna magna, from which the subarachnoid space is created by a "water dissection" of the cerebrospinal fluid. At this stage of embryonic development, the arachnoid is not fully differentiated and, therefore, during the "dissection" a false pathway can be created between both layers of the arachnoid, which if enlarged will produce an arachnoid cyst. This explains the development of cysts in the posterior fossa, middle fossa and convexity, although not in other locations, such as suprasellar and intraventricular cysts.

Other hypotheses indicate that they are produced by the secretion of substances into the subarachnoid space from the venous system[19]. The theory proposed by Robinson[20] indicates that arachnoid cysts are secondary to focal encephalic hypoplasia in which the cysts occupy the vacated space. Exceptionally, hereditary cases have been published in an isolated form or related to other systemic malformations, such as polycystic kidney[21,22] trisomy of chromosome[12], neurofibromatosis[23] or glutaric aciduria type I[24]. Various theories have been formulated to explain cyst growth, the most accepted of which, because there are cases in which it has been possible to demonstrate in vivo, is the formation of valvular mechanisms that allow the entry without exit of liquid from the subarachnoid space ("communicating arachnoid cysts")[25]. The primary cyst has a congenital origin, either due to an alteration of the arachnoid membranes during the 15th week of the embryonic period or due to a possible agenesis of the temporal lobe, as mentioned above. The secondary cyst occurs through brain trauma, infections and/or intracranial hemorrhages.

According to the classification of Galassi et al.[26], these cysts are in the middle cranial fossa (MCF). They can be divided as follows: Grade I: they are small cysts limited to the anterior middle cranial fossa. Grade II: moderate size, triangular or quadrilateral in shape, occupying the anterior and central portion; extending to the Sylvian fissure. Grade III: large, round or oval in shape, occupying almost the entire MCF and extending to the hemisphere through numerous areas.

These cysts are usually asymptomatic and are found incidentally on neuroimaging. Other manifestations of arachnoid cysts include headache, seizures, signs of intracranial hypertension, and neurological deficit[27]. These cysts may also rupture, producing subdural hematomas and subdural hygromas [28]. Other symptoms may include neuropsychiatric symptoms. There are few studies and there is still no consensus; however, they may manifest with depressive and anxious symptoms and schizophrenia-type symptoms (e.g. paranoid ideas and hallucinations) and irritative or deficit neurological symptoms. In this clinical case we present a 24-year- old patient of Venezuelan origin, currently living in Guyana, with a first epileptogenic outbreak more or less five years ago, characterized by generalized tonic clonic seizures and with no previous personal history of head trauma or infectious diseases of the Central Nervous System, nor does he have a family history of related health, where when performing neuroimaging studies to rule out organicity we found a right parietotemporal arachnoid cyst Galassi type III with signs of compression of the right temporal lobe in the middle cranial fossa, as well as the ipsilateral ventricular system and displacement of midline structures (Figure 1).

Given the data in the international scientific literature on giant arachnoid cysts and the associations of neuropsychiatric symptoms, among which epilepsy stands out as the main symptom in a large number of cases. Considering that so far in Guyana this could be one of the first cases of its kind treated surgically by endoscopic means published internationally, in addition to new surgical experiences during the intervention, we decided to carry out this case publication with the aim of expanding scientific knowledge on the one hand and making known the scientific-technological advances that our country presents in the field of health on the other hand.

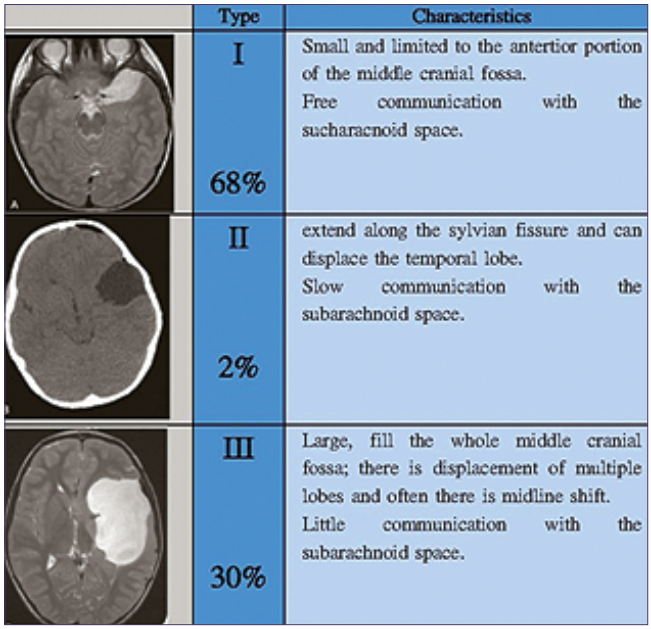

Figure 1. Classification of galassi for arachnoid cysts in the middle cranial fossa.

Clinical case presentation

This is a 24-year-old male, with a clinical picture of 5 years of evolution, according to the relatives, debuting with generalized tonic-clonic convulsive seizures, not previously observed by them in the patient during his life, accompanied by severe right hemicranial stabbing headache that increased in the morning hours, in all occasions it was presented without auras, and associated with gastric symptoms: vomiting and nausea.

Approximately a year ago, he came to our department for which he required admission on several occasions where the corresponding imaging studies were performed, where a Right Temporal Arachnoid Cyst Galassi Grade III was diagnosed, receiving conservative treatment and improvement of his initial clinical picture.

With no known personal medical history or family psychiatric history, he had adequate neurodevelopment and complete university education, and no history of toxic habits was recorded. Due to the constant recurrences of the seizures explained above, and the decrease in the inter seizure period during the evolution of her disease, we decided to discuss the case in our department with the aim of providing her with the best therapeutic option, where we decided after an exhaustive discussion that endoscopic cystoventricular shunt would be a magnificent definitive treatment option; for which we carried out a thorough surgical planning supported by the MRI and CT studies performed.

He was admitted to the Georgetown Public Hospital Corporation, Guyana, under the care of our Neurosurgery Service . No alterations in vital signs and tests performed in the emergency room prior to admission.

In-room laboratories: No abnormalities in complete blood count, renal function test, thyroid function test, lipid profile, fasting glucose, complete electrolytes and negative HIV serology.

Images: Plain brain CT with report: Galassi type III arachnoid cyst in the right middle cranial fossa with mass effects on the ipsilateral hemisphere and displacement of midline structures (Figure 2). Contrast-enhanced brain MRI reported: Galassi type III arachnoid cyst with displacement of the right temporal lobe into the medial cranial fossa, and midline structures to the left (Figure 3).

Electrophysiologicalstudies: Electroencephalogram not performed.

Cognitive screening: There are failures in attention and concentration.

Clinical evolution

a) Upon admission to the Emergency Center with mental examination:

- *Appearanceandbehavior:* patient on a stretcher restricted at four points, with apparent age consistent with chronological status, wearing hospital clothing, with unexpressive appearance, poor cooperation, suspicious, not shown to be aggressive, although with a perplexed look towards the ceiling.
- *State of consciousness:* awake, orientation: cooperative, although communication is difficult at times because she only speaks Spanish.
- *Memory:* no alterations.
- *Psychomotor activity:* without psychomotor agitation.
- *Language:* oral, good diction, sometimes low tone of voice and choppy words.
- *Thought:* consistent with reality, without delusional ideas.
- *Humorand affection:* he appears quiet and thoughtful
- *Critical judgment:* preserved, linked to reality.
- *Diseaseawareness:* present

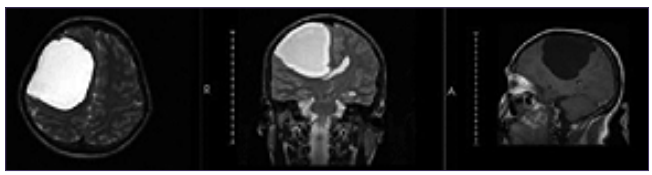

Figure 3. Contrast-enhanced brain MRI: Galassi type III arachnoid cyst with displacement of the right temporal lobe into the medial cranial fossa, and midline structures to the left.

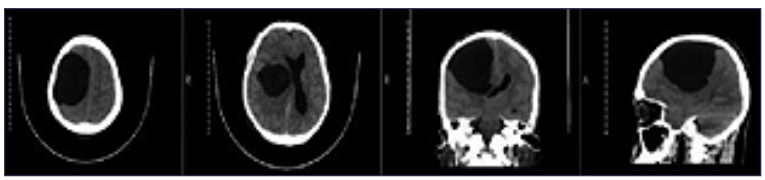

Figure 2. Plain brain CT: Galassi type III arachnoid cyst in the right middle cranial fossa with mass effects on the ipsilateral hemisphere and displacement of midline structures.

- *Neurological examination:* Glasgow scale: 15 pts., no motor defects, no sensory defects, intact cranial nerves, ROT present, isochoric and reactive pupils.

b) Surgical procedure in the operating room

To perform the surgical procedure, which consisted of performing an endoscopic cystoventricular shunt, a rigid Karls Storz endoscope with 30-degree optics was used, coupled to a double-entry working channel and connected to a continuous irrigation system with Lactated Ringer's solution.

The equipment used for the procedure included, among other essential instruments, a 3 French (Fr) Fogarty probe. The images were recorded digitally using a video capture card. The images were edited with Adobe Photoshop CS6 software.

The surgical procedure was performed with the patient under general anesthesia. Surgical planning was based on preoperative magnetic resonance imaging and computed tomography. Our intervention strategy was controversial, since it was not established in relation to the location of the cyst and its proximity to the cortical surface, as stated in the literature in this type of cases, but rather our approach route and direction was planned taking into consideration the eloquent areas, as well as vascular structures. That is why, contrary to the rules, we decided to penetrate the cyst at the most equidistant point with respect to the cerebral cortex and the arachnoid cyst, to avoid irreversible organic lesions and fatal angulation errors at the site of communication with the nearest avascular ventricular cavity. Our objective was to make a trajectory as direct and with the least possible angulation in order to reduce the margin of error, because in the most superficial site of the cyst, in addition to the intracystic path being longer, the angulation to enter the ventricular system was more prominent and we ran the risk of injuring the internal capsule and other anatomical structures such as the head of the caudate nucleus.

The patient was placed in the supine position with the head positioned in a ring-type headrest (San Antonio), rotated to the left side and elevated 30 degrees from the operating table, in order to reduce blood loss and postoperative pneumocephalus. The head was oriented according to the location of the planned trepanation in the paraventricular cysts, and was lateralized contralaterally to the cyst. The endoscopy tower was positioned in front of the surgeon handling the endoscope, so that he could have a view of it at all times. The height of the table was adjusted to allow the surgeon handling the endoscope to be in a flexed elbow position, thus achieving a comfortable and stable posture for handling it. The incision was marked, and two incisions were marked (Figure 4) an arcuate incision with the base oriented to respect the epicranial vasculature, towards the base; and another smaller one of 3 cm that coincided with the anterior end of the major incision. This surgical planning was based on two objectives: the first surgical objective was to confirm the initial diagnosis of an Arachnoid Cyst, after a meticulous observation of the cystic cavity, and in case of finding signs that did not confirm it at that moment we could change the surgical planning to a conventional surgery, second objective. Antisepsis and infiltration of the wound with 2% lidocaine with epinephrine (7 mg/kg as maximum dose) was performed. Trepanation was performed with a No. 16 drill, and the dura mater was opened linearly with a No. 11 scalpel, with an extension of 6-10 mm.

The cerebral cortex was then coagulated and then the puncture trocar was introduced according to the direction and depth pre-established. Once the trocar was removed, the working channel was introduced to the previously calculated depth, where the endoscope was subsequently placed.

The next step was to perform a thorough intracystic endoscopic navigation to confirm the diagnosis as mentioned above. The diagnosis was recognized from the finding of an opaque gray membrane that is usually vascularized, not corresponding to normal anatomy. Then, the most suitable site for fenestration was selected according to the pre-established angulation. The ostomy was performed with the help of a 3Fr Fogarty catheter, always taking care not to present blood vessels capable of producing bleeding.

Once the diagnosis was made, the corresponding anatomical characteristics were observed to let us know that we were in the ventricular system. After this procedure, the flow of cerebrospinal fluid and the rocking movement of the walls where the stoma was made were observed. Then, the area was reviewed under endoscopic vision to avoid leaving any active bleeding, placing a small fragment of Gealfom at the site of the corticotomy performed to prevent the leakage of cerebrospinal fluid and performing a layered closure with the subsequent placement of a sterile compressive bandage.

The days in hospital were 8 days, 3 of them corresponded to the preoperative preparation of the patient, where oral and intravenous medication was required, the latter in the immediate postoperative period, such as anticonvulsants and broad-spectrum antibiotic therapy (phenytoin 100 mg every 8 hours and augmnetin 1.2 g every 12 hours and metronidazole 500 mg every 8 hours for 5 days and then orally completing 10 days to prevent infections of the Central Nervous System, in addition to analgesics and gastric protectors accompanied by neurostabilizers.

c) Post-surgical clinical evolution

The post-surgical evolution of our patient was really satisfactory, no immediate or late complications were observed,

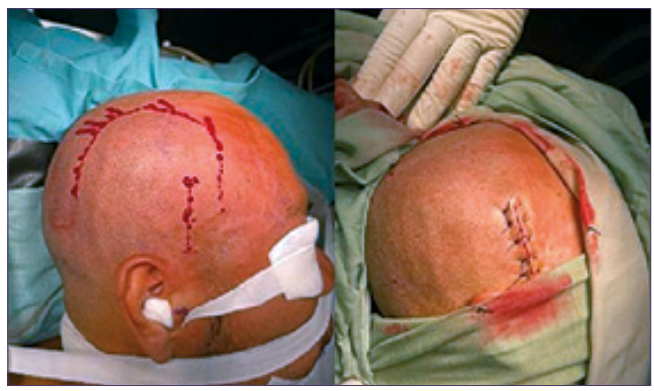

Figure 4. Two incisions were marked both based on two objectives: the initial diagnosis of an Arachnoid Cyst and in case of finding signs that did not confirm it at that moment we could change the surgical planning to a conventional surgery.

no sequelae related to surgery were observed, so he was discharged from hospital on the fifth day after surgery, once he had received parenteral antibiotic therapy, and then continued treatment and care at home, with follow-up at home.

During the evolution of the outpatient consultation, only one event of generalized tonic-clonic seizure was observed, which resolved with medication. After questioning the patient, it was found that it was an external withdrawal of the medication. Therefore, after 6 months of postoperative surgery, it was found that the patient had a permanent complete improvement of his symptoms. On the other hand, from the point of view of the follow-up and radiological evolution, with respect to the initial injury, we can consider that the patient presented a significant improvement because 3 months after surgery, a considerable reduction in the size of the arachnoid cyst was observed in his first evolutionary CT scan, as well as the mass effect observed in previous preoperative studies (Figure 5).

Discussion

The presence of an arachnoid cyst does not always constitute a surgical indication, although in our particular case, we had obvious reasons to consider that the symptoms presented constituted an evident disability to lead a normal life. Documentation of symptoms is the main indication for surgery[29]. Current surgical methods for the management of arachnoid cysts are: cystoperitoneal diversion, microsurgical fenestration and endoscopic fenestration. The reduction of symptoms between these three methods is similar, independent of the location 30. Endoscopic fenestration, despite the higher rate of immediate complications, allows a smaller craniotomy and visualization of the cyst in all directions; therefore, it should be used as a first option[31].

In our case, we had a controversy regarding the selection of the topographic site to perform the approach, since we had to carry out our planning against what is stated in the literature, due to the fact that we did not have Neuronavigation and the high risk of injuring eloquent areas of the brain.

Regarding the location of the approach, for those arachnoid cysts with a paraventricular location, the trephine hole is located on the most superficial portion of the skull and the cystoventricular communication is performed[32,33,34,35-37].

Our selected site to perform the fenestration of the cyst and to communicate with the Ventricular System was done according to the angulations previously performed, always taking into account avoiding very vascularized areas and using a 3Fr Fogarty catheter, which is a diameter sufficient to avoid the closure of the stoma. In our case, we did not use an electrocoagulator nor forceps or scissors in order to perform a dissection as blunt as possible, so we performed it only using the Fogarty catheter. The site and size of the ostomy to fenestrate the cyst is also a topic of debate35,38,39,40,41.

Regarding the site, in cysts it is suggested to make the stoma in the avascular region of the same[35,38,42]. On the other hand, Gangemi defines the site for the endoscopic cystoventricular ostomy where the ventricular wall generates a prominence within the cyst [14]. Regarding the size, the great majority of the authors perform it with Fogerty 3Fr without the need to place a catheter to prevent its occlusion[38,39,40,41,42,43]; others suggest coagulating whenever the edges of the stoma are allowed, extracting remnants with scissors, forceps or performing multiple fenestrations to decrease the risk of closure[35,41,44,45].

From a clinical point of view, the patient was classified as " improved " compared to the preoperative clinical status. Regarding imaging studies, the variables that indicated improvement were considered to be a reduction in the size of the ventricles or cysts or stable ventricles with disappearance of periependymal edema, an increase in the subarachnoid space in the cerebral convexity and centralization of the midline. For the evaluation of postoperative results, the 5-grade classification of Ross et al., was used, which considers preoperative objectives (including endoscopic planning, for example, fenestration of the cyst or reduction of the shunt systems to the minimum possible) and clinical and imaging changes (Table 1)[42].

In our case, in general, the symptoms were predominantly represented by neurological symptoms, although always associated with psychological disorders, although mild. However, our greatest effort was to improve the neurological symptoms, which were the most pointed out by the patient in this case.

We compare our clinical case with those of Da Silva J.A. et al., Baquero G.A. et al., Kahn A.H. et al., and Tsai T.Y. et al.[30,46,47,48], where there were some similar symptoms in addition to behavioral changes of an aggressive, suspicious and irritable type. Some patients had previous diagnoses or hereditary family history by psychiatry, and incidentally these types of cysts were found in them, where they were impressed that the persistent psychiatric symptoms despite pharmacological treatment were possibly secondary or exacerbated by organicity. These cases were evaluated by neurosurgery, where two of them underwent conservative management and the rest underwent surgical intervention with subsequent improvement. And precisely our case where the surgical treatment carried out caused almost complete improvement of his symptoms. Neuropsychological function in patients with arachnoid cysts showed no influence on verbal, performance or IQ scale, how-

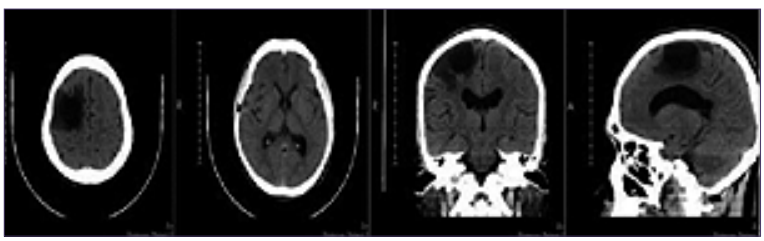

Figure 5. Evolutionary plain CT scan 3 months after surgery, presented a significant improvement without displacement of midline structures.

Table 1. Ross et al. Degree result definition, clinical status AND/OR MRI
Permanent complete improvement
Complete improvement transient
Partial improvement
Doubtful
Failed
All objectives persist at the time of assessment (minimum 6 months follow-up). Improved or unchanged AND At least one criterion is met
All objectives persist at the time of assessment (minimum 6 months follow-up), then one fails. Improved or no change AND At least one criterion is met
Not all pre-surgical objectives (if more than one) remain for 6 months post-surgical. Improvement or no change AND At least one criterion is met
Improved or unchanged AND No criteria are met, but ventricular size remains stable or cyst size
Worsening OR No criteria are met, and ventricular or cyst size increases

ever they did find that the subjects studied had 35.7% greater inattention and 39.7% greater impulsivity; and these values were not affected by surgical treatment[49]. Another study evaluated neurocognitive development in pediatric patients with Galassi II and III arachnoid cysts, through standardized tests, where no disorders in the neurodevelopment of the patients could be identified, however in the adaptive behavior test a tendency towards a poor outcome of their general function was found[50].

Other manifestations of arachnoid cysts are headache, seizures, signs of intracranial hypertension and neurological deficits[27] and in some cases surgical management may be required[51]. These cysts also rupture, producing subdural hematomas and subdural hygromas[28].

The main diagnostic method is magnetic resonance imaging; in simple phase, contrast and functional. Magnetic resonance imaging can demonstrate structural abnormalities, CSF flow patterns and brain organization[52]. Advances in the various diagnostic modalities have identified functional disorders of the brain related to the cyst 52 such as positron emission tomography, with which a hypometabolic pattern can be identified in the area of the arachnoid cyst[53]; electroencephalography shows increased irritability in the regions close to the cyst 52, plus a reduction in the amplitude of the acoustic evoked potentials in the frontotemporal area ipsilateral to the cyst[52], aspects that were not evaluated in our case, because these studies were not available due to situations beyond the control of our service. SPECT may detect alterations in perfusion in the regions adjacent to the cyst, along with impairment of regional cerebral flow, which correlate with clinical symptoms[54]. The existence of a decrease in the thickness of the cortex, as well as of the white and gray matter, is another aspect commonly found in this type of entity; this finding is proportional to the size of the cyst[55].

Conclusions

Despite the neurosurgical evidence that in the past, and in many places today, open surgery of these intracranial arachnoid cysts was a fundamental treatment pillar, we can conclude that today, and especially in developed countries, endoscopic surgery has become more important due to the reduction in surgical time, as well as the reduction in hospital time and the total recovery of the patient. The shortest transcerebral approach or transsurgical intracerebral route is not always the safest for planning and carrying out endoscopic transcerebral surgery, especially when the path, even if less elongated, presents a risk of injuring eloquent areas. Therefore, a more equidistant but well-planned intracerebral route that avoids injuring eloquent areas is preferable, in order to reduce disabling post-surgical neurological sequelae and achieve optimal post-surgical results, which allow us to reincorporate our patients to a full life without disability.

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