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Case Report

Sternberg's canal as a cause of encephalocele within the lateral recess of the sphenoid sinus: A report of two cases

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Abstract

Background: Intrasphenoidal encephaloceles are extremely rare findings. Sternberg's canal is a lateral craniopharyngeal canal resulting from incomplete fusion of the greater wings of the sphenoid bone with the basisphenoid. It acts as a weak spot of the skull base, which may lead to develop a temporal lobe encephalocele protruding into the lateral recess of the sphenoid sinus (SS).

Case Description: We present two cases of intrasphenoidal encephalocele due to persistence of the lateral craniopharyngeal canal. The first case presented with cerebrospinal fluid (CSF) rhinorrhea and the second one was referred to the neurosurgical department with CSF rhinorrhea and meningitis. Radiological investigations consisted of computed tomography (CT) scan, CT cisternography and magnetic resonance images in both cases. These imaging studies identified a herniated temporal lobe through a bony defect which communicates the middle cranial fossa with the lateral recess of the SS. Both patients underwent a transcranial repair of the encephalocele because of the previous failure of the endoscopic surgery. There was no complication related to the surgical procedure and no recurrence of CSF leakage occurred 2 and 3 years after surgery, respectively.

Conclusion: Encephalocele within the lateral recess of the SS is a rare entity which must be suspected in patients who present with spontaneous CSF rhinorrhea. Congenital intrasphenoidal encephaloceles, which are located medial to the foramen rotundum, seem to be due to persistence of the Sternberg's canal. Transcranial approach is a good option when a transnasal approach had failed previously.

Key Words: Cerebrospinal fluid rhinorrhea, intrasphenoidal encephalocele, lateral craniopharyngeal canal, lateral recess, sphenoid sinus, Sternberg's canal

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INTRODUCTION

Cranial encephaloceles, herniation of intracranial meninges and brain tissue through a defect in the cranium or skull base, are rare conditions with an incidence of approximately 1 in 35,000 people, and are more common in the anterior cranial fossa than in the middle one.[1,7,10,17,23] Basal encephaloceles represent up to 10% of them.[17] Intrasphenoidal encephaloceles are extremely rare findings. The medial or perisellar type

arises from the superior or posterior wall of the sphenoid sinus (SS). The lateral localization, which results from temporal lobe herniation through a middle fossa defect into the lateral recess of the SS, is even more rarer than the medial localization, and it is probably the least common type of basal encephaloceles.[4,9,10,16,19]

The etiology of SS bony defects may be acquired or congenital. Acquired cause may be traumatic or post-surgical and is the most frequent origin of these defects. Congenital abnormalities of the SS are likely to occur at the fusion plane of the ossified cartilaginous precursors of the sphenoid bone during development. Sternberg's canal is a lateral craniopharyngeal canal resulting from incomplete fusion of the greater wings of the sphenoid bone with the basisphenoid. It acts as a weak spot of the skull base, which may lead to develop a temporal lobe encephalocele protruding into the lateral recess of the SS.[1,4,6,7,13,19,20,23]

We present two cases of intrasphenoidal encephalocele due to persistence of the lateral craniopharyngeal canal.

CASE REPORTS

Case 1

A 73-year-old female with a 2-month history of permanent clear rhinorrhea, which had been treated previously as a rhinitis by a neumonologist, was referred to the ear–nose–throat (ENT) department. Physical examination revealed right-sided rhinorrhea. There was no history of head trauma or cranial surgery. The nasal discharge was interpreted as cerebrospinal fluid (CSF) rhinorrhea. A computed tomography (CT) scan revealed a defect in the middle cranial fossa communicated to the lateral recess of the right SS and a soft tissue mass within the sinus. Magnetic resonance (MR) imaging showed a brain tissue herniation through the bony defect [Figure 1c]. CT cisternography, following administration of intrathecal fluorescein, identified an intrasphenoidal encephalocele on the right side of the sinus [Figure 1a and b]. It appeared to be due to persistence of the Sternberg's canal. The patient underwent transnasal endoscopic surgery by an otorhinolaryngologist. The bony defect in the right SS was not identified during the procedure and the sinus was filled with abdominal fat. There was a CSF leak recurrence 1 month later. Thus, the patient underwent a second and a third transnasal endoscopic surgical repair by the same physician. She continued to have CSF rhinorrhea and was referred to the neurosurgical department. Because of the previous failure of the endoscopic surgery, the patient underwent a transcranial repair. A fronto-temporal approach was used, and under the operating microscope magnification, intradural dissection was performed until the temporal lobe encephalocele was identified [Figure 1d]. Once the lesion was correctly exposed, the protruded temporal tissue was reduced in volume by bipolar cauterization and removed. After that, the SS was filled with absorbable hemostatic gelatin sponge. The skull base defect was closed with a multilayer plasty: double layer of dural graft, bone fragments obtained from the craniotomy edge with rongeurs and fibrin glue. There was no complication related to the surgical procedure. No recurrence of CSF leakage occurred 2 years after surgery.

Case 2

A 46-year-old female was referred to the neurosurgical department with CSF rhinorrhea for 3 months and recently developed fever, nausea and headache. Meningitis was diagnosed by lumbar puncture and CSF cultures. Neuroradiological diagnosis was made in the same way as in the previous case. Radiological investigations identified a herniated temporal lobe through a bony defect which communicates the middle cranial fossa with the left lateral recess of the SS [Figure 2]. This bony defect may

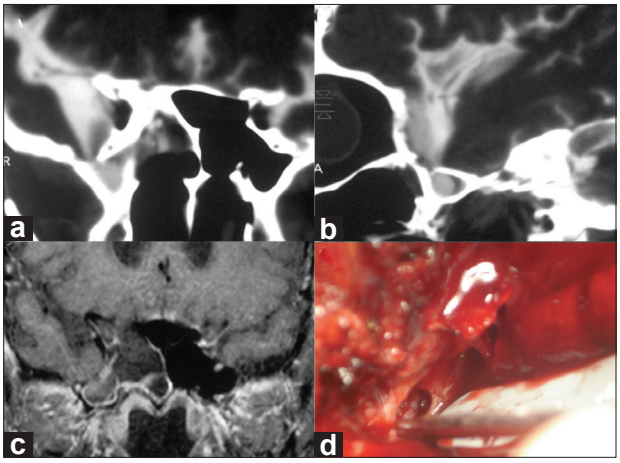

Figure 1: (a and b) Sagittal and coronal CT cisternography; (c) coronal MR image; (d) operative field during dissection of the skull base defect

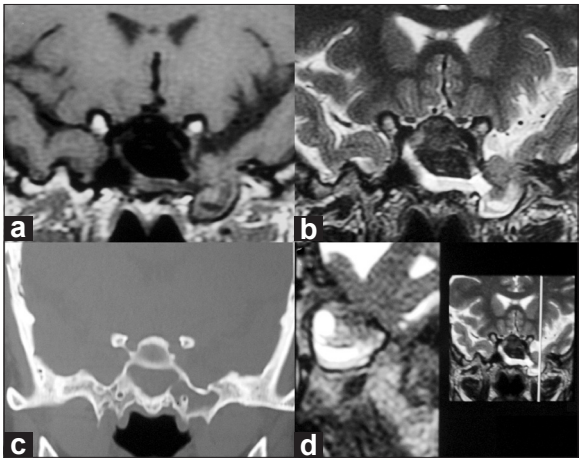

Figure 2: (a, b and d) Coronal MR images showing the temporal lobe encephalocele; (c) CT scan showing the lateral recess of the sphenoid sinus

be related to persistence of the lateral craniopharyngeal canal. Meningitis was treated with antibiotics. Then, the patient underwent microsurgical transsphenoidal surgery. The origin of the CSF leak and the site of the skull base defect was not identified during the procedure and the SS was obliterated with abdominal fat, absorbable hemostatic gelatin sponge and fibrin glue. There was a CSF leak recurrence 2 months after the procedure. Thus, she underwent a transcranial repair of the defect. A fronto-temporal approach was performed followed by extradural dissection of the temporal dura until the encephalocele was identified. The skull base defect was closed with a multilayer plasty as in the previous case. Immediate postoperative recovery was uneventful and no recurrence of CSF leakage occurred 3 years after transcranial surgery.

DISCUSSION

The SS is widely variable in size, shape, and degree of pneumatization. It reaches its full size during adolescence.[12,18,22] In the anatomical description of the SS type, its lateral wall does not extend laterally beyond the line connecting the medial edges of the anterior opening of the vidian canal and the extracranial end of the foramen rotundum.[22] The lateral recess of the SS is an extensive lateral pneumatization of the SS into the pterygoid process, the great wing of the sphenoid bone or both, and it is also known as lateral type of sinus [Figure 2c].[5,10,12,18,22] Rarely, the SS may extend up to the foramen ovale.[7] Those extensions of the SS are related to the middle cranial fossa, lateral to the cavernous sinus.[2] Lewin *et al*. found that 41 of 72 patients had lateral recess formation on at least one side and Barañano *et al*. found it in 35.3% of 1000 CT scans.[3,12] The size of the lateral recesses of the SS is highly variable and it can make the differentiation of normal asymmetry from pathologic expansion difficult.[5,12] The lateral type of SS creates a weak site in the skull base, in the junction of the middle cranial fossa and the lateral wall of the SS, which is relevant in the physiopathology of anteromedial temporal lobe encephaloceles and spontaneous CSF fistula.[7,20]

The sphenoid bone develops from the ossification of several independent cartilaginous precursors: presphenoid and postsphenoid/basisphenoid centers (body of the sphenoid bone), orbitosphenoids (lesser wings), and alisphenoids (greater wings). Union of those ossified components results in formation of the sphenoid bone.[15,19] If the posterior portion of the bony fusion of the greater wings with the bone's body is incomplete, it creates a lateral craniopharyngeal canal, which was described by Sternberg in 1888. In the presence of a lateral recess of the SS, the Sternberg's canal can communicate with the SS after its pneumatization, acting as a possible site of origin of congenital encephaloceles. Fusion planes offer resistance to pneumatization, so sphenoidal defects at fusion planes are more likely to be congenital than acquired.[1,3,4,19,21,23] Sternberg's canal has been reported in up to 4% of adults, but Barañano *et al*. found only one case in 1000 CT scans. They found that the development of arachnoid pits in the lateral recess of the SS, probably from underlying intracranial hypertension, was the major cause of acquired spontaneous CSF leaks and lateral intrasphenoidal encephaloceles in that study. Those arachnoid pits were present in 23.4% of patients and were located lateral to the foramen rotundum.[3] Unlike those reported arachnoid pits, the Sternberg's canal is a congenital defect at a fusion plane, which finishes its development after the SS pneumatization and its lateral recess formation.

Obesity is thought to be a risk factor for spontaneous CSF leak and encephaloceles, but it is only supported for anecdotal reports. The theoretic physiopathology of its association is that increased weight increases intraabdominal and intrathoracic pressure which could lead to the development of benign intracranial hypertension.[4,10] Our first patient was obese and the second one had a normal body mass index.

Intrasphenoidal encephaloceles that have an intact dura and no CSF leak are usually diagnosed during imaging studies for other problems.[10] CSF rhinorrhea is the most common clinical manifestation of temporal encephaloceles through Sternberg's canal and other previously occult malformations of the skull base. The CSF drainage is generally intermittent and not voluminous and may be ignored by the patient for a long time until complicated by meningitis. Recurrent meningitis may also occur. Others signs and symptoms of this entity are chronic headache, seizures and vertigo. Meningitis and pneumocephalus are common complications of CSF leaks of the SS, although tension pneumocephalus is an unusual presentation.[4,6,7,11,16,21] Patients with encephaloceles within the lateral recess of the SS classically present with CSF rhinorrhea during adulthood, enhancing the importance of pneumatization of the SS in the pathogenesis.[23] Regarding neuroradiological investigations, CT scan is a noninvasive imaging technique which gives good bone detail and identifies the site of the skull base defect. CT cisternography consists of injecting intrathecal water-soluble contrast medium before the CT scan and then visualizing it at the level of the dural and skull base defect [Figure 1a and b]. Intermittent or inactive CSF leaks are usually associated with a high incidence of false-negative results and MR imaging may be a better choice in those patients.[6,7,8,20,23] MR images give better information about the soft tissues like the encephalocele itself [Figures 1c, 2a, b and d].[6] Radiological images may also show partial or complete opacity of the SS.[14] Then,

the diagnosis is confirmed at the time of surgery.[21]

Persistent CSF leak is potentially lethal because it may lead to meningitis or brain abscess. Thus, repair of intrasphenoidal encephaloceles has two main objectives: prevention of CSF leak and to avoid central nervous system infection.[1,6,10,13]

Surgical treatment should be tailored to each patient. Transcranial approaches have been used for repair of encephaloceles within the lateral recess of the SS. Fronto-temporal craniotomy provides excellent access for exploration of middle cranial fossa floor. The dissection and repair of the encephalocele may be performed intradurally, extradurally or by a combination of both. Some authors believe that CSF leaks involving the lateral recess of a widely pneumatized SS require a transcranial approach for direct visualization and treatment of the defect. On the other hand, endoscopic transnasal approaches are less invasive and do not require a large external incision and temporal lobe retraction, minimizing brain manipulation.[1,4,6,9,10,16,21,23] The endoscopic transpterygoid approach reaches the lateral recess of the SS through the posterior wall of the maxillary sinus and the pterygopalatine fossa and it is a better technique than the traditional transnasal and transethmoid approaches to repair skull base defects in that location, allowing the surgeon to reach the most lateral portions of the SS and close the defect with a multilayer plasty. Microscopic transseptal and endoscopic paraseptal direct transsphenoidal approaches may not provide direct visualization of the lateral region of the SS, allowing the surgeon to treat the defect only by obliteration of the SS, without removing the underlying cause. Also, a complete removal of the mucosa of the lateral recess of the SS with these latter approaches may be impossible, leading to sinus obliteration failure.[2,4,10,20,23] Transcranial approach is a good option when a transnasal approach had failed previously, but probably, if we had performed a transpterygoid approach, we would have identified the skull base defect successfully and it would not have been necessary to use a transcranial approach.[1,2,11] On the other hand, the endoscopic transpterygoid approach is not actually a minimally invasive technique and it is a partially sterile approach because it reaches the lateral recess of the SS through the dirty nasal and paranasal sinuses cavities. In spite of needing more brain manipulation, transcranial approach is still safe to treat this challenging disease if it is performed by a skilful neurosurgeon with all current technology available. Transcranial surgery prevents central nervous system infection better than endonasal approaches because it allows the neurosurgeon to reach the defect through a more sterile operative field. There was no complication after transcranial surgery in our two patients. Thus, we think that transcranial approach is still a good option even as the first tier treatment instead of only a second tier technique.

CONCLUSION

Encephalocele within the lateral recess of the SS is a rare entity which must be suspected in patients who present with spontaneous CSF rhinorrhea. As it was stated in this article, congenital intrasphenoidal encephaloceles, which are located medial to the foramen rotundum, seem to be due to persistence of the Sternberg's canal. Although the endoscopic transpterygoid approach is probably the best technique to treat these lesions, transcranial approaches are an optimal option when a transnasal approach had failed previously.

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