ORIGINAL ARTICLE

Endoscopic Transpterygoid Approach and Skull Base Repair After Sphenoid Meningoencephalocele Resection. Our Experience

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KEYWORDS
Skull base; Sternberg’s canal; Endoscopic sinus surgery; Cerebrospinal fluid leak; Meningoencephalocele; Sphenoid lateral recess; Transpterygoid

Abstract
Introduction and objectives: Cerebrospinal fluid leaks associated to meningoencephaloceles of the sphenoid lateral recess are rare entities. A congenital bony defect at this level results in the persistence of Sternberg’s canal, or a lateral craniopharyngeal canal, which is supposed to be the origin of these lesions. Our objective was to show that the endoscopic transpterygoid approach is an effective technique for their treatment.
Methods: We present a series of 5 cases of meningoencephaloceles of the sphenoid lateral recess treated with endoscopic sinus surgery (4 women and one man; mean age = 59, range 37–72 years). Cerebrospinal fluid rhinorrhoea was present in all of them and they all underwent a transpterygoid approach with reconstruction of the skull base. We describe the surgical technique and review the literature.
Results: No complications were observed during surgery or the postoperative period. After a mean follow-up of 81 months, only one recurrence was seen.
Conclusions: The transpterygoid approach has proven to be effective for the treatment of meningoencephaloceles of the sphenoid lateral recess. Providing wide access to identify the defect, followed by meningoencephalocele ablation, is the key for successful surgery.

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Abordaje endoscópico transpterigoideo y reparación de base de cráneo tras resección de meningoencefalocele esfenoidal. Nuestra experiencia

Resumen

Introducción y objetivos: Las fistulas de liquido cefalorraquideo asociadas a meningoencefaloceles del receso lateral de esfenoides son entidades muy infrecuentes. Un defecto óseo congenito a este nivel da lugar a la persistencia del canal de Sternberg o canal craneofaríngeo lateral, supuesto origen de estas lesiones. Nuestro objetivo es exponer que la cirugía endoscópica nasosinusal con abordaje transpterigoideo es una técnica efectiva para su tratamiento.

Métodos: Presentamos cinco casos de meningoencefaloceles de receso lateral de esfenoides tratados con cirugía endoscópica (4 mujeres y un hombre, edad media = 37-72 años). Todos presentaban rinolítorreorra y en todos se realizó un abordaje transpterigoideo con reconstrucción de la base de cráneo. Realizamos una descripción de la técnica quirúrgica y una revisión de la literatura.

Resultados: Ninguno tuvo complicaciones intra o postoperatorias y solo se registró una recidiva, con una media de seguimiento de 81 meses.

Conclusiones: El abordaje transpterigoideo es efectivo para el tratamiento de meningoencefaloceles de receso lateral de esfenoides. Realizar un amplio acceso para identificar el defecto, seguido de la ablación del meningoencefalocele es la clave para una cirugía exitosa.

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Introduction and Objectives

Intranasal meningoencephalocele (MEC) is a protrusion of intracranial content, including meninges and brain tissue, through a defect in the base of the skull towards the nostrils or sinuses. It is a very rare lesion, with an approximate incidence of 1 per 35,000 persons, and are more common in the anterior skull base. Sphenoid sinus (SS) MEC are a very rare entity and can be arise spontaneously or have an acquired cause (traumatic or postoperative).

Depending on their location, they can be divided into medial or parasellar (through the superior or posterior wall of the SS) and lateral (towards the lateral sphenoid recess (LSR)); these last are the most infrequent.

Spontaneous lateral SS MEC originate from a congenital defect of the skull base, the result of an incomplete or premature fusion of the bone components involved in the complex ossification process of the sphenoids. This gives rise to a canal lacking bone covering, covered only by connective tissue, which is called the lateral craniofarygeal canal or Sternberg’s canal.

The persistence of Sternberg’s canal is sometimes associated with an extensive pneumatization of the sphenoid sinus (a condition that provokes a thinning of the LSR roof) and elevated intracranial pressure; the presence of this canal can cause the appearance of spontaneous lateral MEC linked to a cerebrospinal fluid fistula.

This type of lesions requires surgical treatment to repair the defect and prevent potential intracranial complications. The location of the defect in the skull base determines the type of endoscopic approach required and, in the case of the lateral sphenoid sinus recess, the transpterygoid (TP) approach has been shown to be the most appropriate pathway.

We present our experience in 5 cases of MEC treated using endoscopic TP approach. Likewise, the studies published are analysed and compared with our results.

Methods

We present a series of 5 diagnosed cases of LSR MEC and detail their characteristics in Table 1. All of them showed unilateral watery rhinorrhea as the presenting symptom and its analysis was positive for Beta-2 transferrin or Beta-trace protein. There was a history of prior head injury in 2 cases and 1 case presented meningitis 1 month after the rhinorrhea began.

All patients received preoperative a computed tomography (CT), a study that revealed a bone defect in one or the other lateral wall of the SS (Figures 1-3). In all cases sinonasal endoscopic surgery using TP approach and reconstruction of the skull base were performed (Figure 4A and B).

Results and Surgical Technique

The same surgical technique was performed for all 5 MEC cases: sinonasal endoscopic surgery using TP approach to access the LSR.

In the premedication phase, before anesthesia induction (some 30 minutes before the beginning of the surgery), the patient received an intrathecal injection of 0.5–1 ml of 5% fluorescein.

The nostril was first decongested with surgical patties and vasoconstrictor. The operation began with removal of the middle turbinate, from which the mucoperiosteal flap to be used in closing would be obtained. Next, a wide half meatotomy, complete ethmoidectomy and sphenoidotomy
Table 1  Characteristics of Our Series of Meningoencephalocele in Lateral Sphenoid Sinus Recess.

<table>
<thead>
<tr>
<th>Patient</th>
<th>Sex</th>
<th>Age (years)</th>
<th>Clinical presentation</th>
<th>Side</th>
<th>Approach</th>
<th>Flap (material)</th>
<th>Relapse</th>
<th>Follow-up (months)</th>
</tr>
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<tr>
<td>1</td>
<td>M</td>
<td>68</td>
<td>Unilateral watery rhinorrhea</td>
<td>L</td>
<td>TP</td>
<td>FL + MTMP</td>
<td>No</td>
<td>18</td>
</tr>
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<td>2</td>
<td>F</td>
<td>72</td>
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<td>L</td>
<td>TP</td>
<td>FL + MTMP</td>
<td>No</td>
<td>75</td>
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<td>54</td>
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<td>R</td>
<td>TP</td>
<td>FL + MTMP</td>
<td>No</td>
<td>82</td>
</tr>
<tr>
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<td>F</td>
<td>37</td>
<td>Unilateral watery rhinorrhea</td>
<td>L</td>
<td>TP</td>
<td>FL + MTMP</td>
<td>No</td>
<td>96</td>
</tr>
<tr>
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<td>F</td>
<td>63</td>
<td>Unilateral watery rhinorrhea</td>
<td>R</td>
<td>TP</td>
<td>FL + MTMP</td>
<td>Yes</td>
<td>132</td>
</tr>
</tbody>
</table>

F: female; FL: fascia lata; L: left; M: male; MTMP: middle turbinate mucoperiosteal; R: right; TP: transpterygoid.

Figure 1  Patient 1: coronal CT slice showing MEC in left lateral sphenoid recess (arrow). SED: right sphenoid sinus; SEI: left sphenoid sinus.

were performed. The sphenoidotomy was widened laterally with a Kerrison punch and drills. The mucosa of the posterior wall of the maxillary sinus and the periosteum of the ascending apophysis of the palatine bone were elevated; the sphenopatine artery and its branches were located and cauterised; the posterior wall of the maxillary sinus was widened using a Kerrison punch and drilling, with medial to lateral dissection until the fascia surrounding the fatty tissue of the pterygopalatine fossa was exposed. At this level, dissection of fatty tissue and neurovascular structures had to be precise, and bipolar electrocoagulation was used to control haemorrhaging. The medial plate of the pterygoid apophysis was carefully drilled to avoid any lesions in the pterygoid canal nerve or vidian nerve and in the maxillary branch of the trigeminal nerve (V2).

All of this achieved adequate access to the LSR and better control of the lesion, with the possibility of using optics of different angles, as well as sufficient space to manipulate and reconstruct the defect. Sinus mucosa was lifted and the bone adjacent to the defect drilled until the MEC was seen. The herniated tissue (normally non-functioning neural tissue) was reduced in size by coagulating with bipolar tweezers (ablation to the level of the skull base).

Millimetric-sized defects had to be enlarged slightly to be able to carry out the reconstruction. Fascia lata (TSF, Barcelona, Spain) was placed in underlay position (between the skull base bone and the dura mater in the epidural space), covered using a mucoperiosteal middle turbinate flap and stabilised with fibrin glue. In Case 5 the area corresponding to the infratemporal space was obliterated with abdominal fat, leaving the sinus vented. We placed

Figure 2  Patient 2: coronal CT slice showing MEC in left lateral sphenoid recess (asterisk). SED: right sphenoid sinus; SEI: left sphenoid sinus.

Figure 3  Patient 3: coronal CT slice showing MEC of the right lateral sphenoid sinus recess (asterisk), maxillary branch of the trigeminal nerve (V2), vidian nerve (V) and optic nerve (No). SED: right sphenoid sinus; SEI: left sphenoid sinus.
anterior packing covered by a glove finger and impregnated with antibiotic ointment as a means of fixation for 24 hours.

We did not use a Foley probe or lumbar drainage.

We ordered intravenous ceftriaxone as prophylactic antibioticotherapy due to its penetration in the central nervous system for 5–7 days. For patients allergic to cephalosporins, levofloxacin and trimethoprim/sulfamethoxazole constitute other appropriate alternatives.

We recommended a fibre-rich diet (sometimes laxative), immobilisation for 3 days with the thorax elevated, prophylactic subcutaneous heparin, sneezing with an open mouth and avoiding blowing the nose.

There were no intraoperative or immediate postoperative complications in any cases. In the follow-up (mean 81 months) only 1 relapse (reappearance of the fistula) was recorded, in a patient that presented a very large MEC (Figure 5). The relapse presented with bacterial meningitis 30 months after surgery and was solved with revision surgery and a new reconstruction of the skull base.

Discussion

The MEC of the LSR are very rare lesions. Our experience, as is that reported in the majority of published work, has shown that the TP approach is effective for reconstructing the skull base after its reduction. In our review of the literature, we found 87 cases of LSR MEC treated with endoscopic surgery published since 2000.

The MEC is considered to originate in Sternberg’s canal or lateral craniopharyngeal canal, and is thought to be the result of an incomplete fusion of the bone components of the sphenoid bone during the embryonic period. \cite{6,8,11,14,15,2} However, Barañano et al.\cite{9} suggest that cerebrospinal fluid fistulas are probably acquired. Based on an analysis of 1000 CT scans of sphenoid bones, they consider that the majority of the fistulas are found laterally to the spot where the ossification centres of the sphenoid fuse and laterally to V2. They believe that the development of recesses in the arachnoids in the context of intracranial hypertension cause the majority of LSR MEC.

The majority of the cases published in the literature, as well as those of our series, presented watery rhinorrhoea as a consequence of cerebrospinal fluid fistula and, in fact, this was the only clinical symptom reported in many cases. Other signs and symptoms have also been described in the presentation, isolated or in association with each other, such as headache, intracranial hypertension, diplopia or meningitis. Some patients \((n = 13)\) had a history of bacterial meningitis or had it at diagnosis and we found a case with subdural haematoma. \cite{13} We coincide with other authors in that the main objective of the treatment of these lesions is closing the fistula by repairing the defect in the skull base, so as to decrease the risk of intracranial infections. \cite{1,6,17}

To treat cases of MEC successfully, a good preoperative study that makes it possible to determine the location is
required. CT images are generally enough to locate the bone defect in the skull base, while magnetic resonance can provide information on soft tissues to evaluate the encephalic herniation.\textsuperscript{1} Isotope cisternography is not normally used, but it can be useful to evaluating cerebrospinal fluid pathways.\textsuperscript{1} If watery rhinorrhea is present, the suspicion of fistula should be confirmed with Beta-2 transferrin or Beta-trace protein tests.\textsuperscript{4,6,15,16} In a review of 39 studies published on the usefulness of several tests such as the Beta-trace protein or Beta-2 transferrin, Bachmann-Harildstad\textsuperscript{19} demonstrated that they are helpful in diagnosing the presence of cerebrospinal fluid. The Beta-trace protein test is highly specific and sensitive, it is quicker (20 minutes versus 120 minutes that the Beta-2 transferrin takes) and more inexpensive. Clinical suspicion with imaging tests and confirmation of cerebrospinal fluid were sufficient to orient the diagnosis in all of the studies published.

In comparison with external middle fossa approaches, endoscopic techniques are currently considered to represent the best path of access for treating sphenoid MEC, because they are less invasive and highly effective.\textsuperscript{20} Lesions in the sphenoid middle line can be repaired successfully with the traditional endoscopic pathways, transnasal-transsphenoidal or transethmoid-sphenoidal. However, these pathways are generally insufficient for cases located in the LSR due to limitations in access and the TP approach has been shown to be the most appropriate.\textsuperscript{1,3,4,6,8,10,14,16,21,23} Bolger was the first to gain access to the most lateral section of the SS through the posterior wall of the maxillary sinus and the pterygopalatine fossa, permitting the use of endoscopes of various angles to expose the defect appropriately and manage the tissue needed to eliminate the lesion and repair the skull base.\textsuperscript{24} This approach entails an increase in potential intraoperative neurovascular complications, due to the presence of the maxillary artery and its branches, the maxillary nerve (V2) and the vidian nerve. Nevertheless, these complications can be avoided by careful dissections, separating the content of the pterygopalatine fossa laterally.\textsuperscript{12,25} Hypoesthesia (associated with pain or not) in the area innervated by V2, 6,6,12,13 as well as dry eye through probable vidian nerve damage\textsuperscript{23,26} can be sequelae of the intervention.

Using intrathecal fluorescein (5%) as a diagnostic method or intraoperatively to locate the defect and check fistula closure during the operation has been shown to be of great help.\textsuperscript{3,6,17,24}

An extensive variety of materials have been described for repairing cerebrospinal fluid fistulas, including synthetic materials, autologous fascia, bone, fat and cartilage. In addition, given that elevated intracranial pressure contribute to the appearance of fistulas, a “robust” reconstruction has greater possibilities of resistance.\textsuperscript{15} This has been demonstrated by the low number of complications described in the series and cases that were handled in this way. Table 2 details the techniques and variability of materials used by various authors, in comparison with our series.

In our experience, placing fascia lata in an “underlay” position (epidural-intracranial between the skull base bone and the meninges) covered by a middle turbinate mucoperiosteal free flap has been effective. Tomazic et al.,\textsuperscript{8} however, applied the “overlay” technique (covering the defect in the nostril, above the margins of exposed bone)
in all their patients because they considered it less invasive, given that it infringes the sensitivity of the underlying temporal bone less.

Eradicating the SS with abdominal fat has frequently been used to seal the defect. However, unless it is associated with resection of the adjacent mucosa and repair of the skull base through multi-layer reconstruction, this technique can lead to persistence of cerebrospinal fluid fistula, appearance of arachnoid cysts and formation of delayed sinus mucocles.\(^6,11,12,25\) Abdominal fat has sometimes been used directly to seal the defect (in the intradural space, in the sense of a "‘bath-plug technique") before placing the fascia or mucosa flap.\(^1,6,15,17\)

Only 4 postoperative intracranial complications (3 meningitis and 1 cerebral abscess) have been published and all of them were resolved with endovenous antibiotics, plus additional lumbar puncture in 1 case.\(^6,12,25\) In 1 of the meningitis cases and in that of the cerebral abscess, the endoscopic approach was transethmoid-sphenoid,\(^6\) with the lesions located in the LSR. Persistence of cerebrospinal fluid fistula was described in 6 cases treated using approaches other than the TP (3 transphenoidal and 3 transethmoid-sphenoid).\(^2,6,12,20\) All 6 cases were repaired with complete resolution of the fistula (3 cases with eradication of the sphenoid sinus\(^6,16\) and 3 with revision endoscopic surgery\(^6,12\)).

Lumbar drainage was used in only 2 series from the moment of surgery until 2–4 days postoperative, in spite of the fact that there was no known elevated intracranial pressure.\(^6,11\) We decided not to put lumbar drainage in any of our cases, given that (coinciding with Castelnuevo et al.) we believe that the success of the reconstruction depends on correct closure of the defect in the skull base and that lowering intracranial pressure is not necessary in this sense because the persistence of certain pressure at the level of the fistula is useful for attachment of the "underlay" flap to the bone defect, without leaving any dead space.

Conclusions

Our experience, as well as that of most of the studies published, demonstrates that the transpterygoid approach is effective in treating meningoencephaloceles of the LSR. This approach achieves a wide space for manipulation and for optimal visualisation for repairing the defect in the skull base. The keys to success in the surgery are identifying the defect, removing the MEC and reconstructing in various layers.

Conflict of Interests

The authors have no conflicts of interest to declare.

References