Endoscopic treatment of traumatic basal encephaloceles: a report of 8 cases

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Object. Basal encephaloceles are rare entities that can present as congenital diseases; however, traumatic lesions due to head injuries or iatrogenic causes have been described in the literature. In this study the authors aimed to define placement techniques for free grafts in repairing traumatic basal encephaloceles and to describe the long-term effectiveness of endoscopic treatment.

Methods. Between September 1997 and December 2006, 8 patients with traumatic encephaloceles underwent endoscopic surgery. A free graft following an underlay (2 cribriform plate and 4 ethmoid fovea defects) or obliteration (2 sphenoid defects) procedure was used as the repair material.

Results. All traumatic basal encephaloceles with the associated skull base defects and cerebrospinal fluid (CSF) leakage were successfully treated via the endoscopic approach. There were no major complications or recurrence of meningitis or leakage of CSF encountered after an average follow-up of 77 months.

Conclusions. Long-term follow-up results demonstrated that endoscopic surgery was suitable for the treatment of traumatic basal encephaloceles. The underlay procedure is more appropriate than the overlay procedure in repairing large defects of the anterior skull base. Meticulous manipulations of the endoscope following precise autograft placement are mandatory for the successful repair of traumatic basal encephaloceles. (DOI: 10.3171/JNS/2008/108/4/0729)

KEY WORDS • basal encephalocele • meningitis • overlay • traumatic lesion • underlay procedure

B asal encephaloceles are rare entities produced by herniation of the cranial contents at the defective areas of the base of the skull.28 The subtypes of encephaloceles, namely intranasal and sphenoharyngeal encephaloceles, occur at bone defects located at the sphenoid bone and cribriform plate. They can appear in an oral pulsatile mass, and at other times they can cause nasal obstructions.8 Suwanwela and Suwanwela35 proposed an origin-based classification for encephaloceles, including basal encephaloceles: congenital,29 spontaneous,32 or traumatic. A traumatic lesion can be iatrogenically caused5,14,20,22,25,38 or the result of head injury.20,25

Endoscopic surgery provides a direct view of the skull base and is associated with lower morbidity rates compared with the transcranial approach in the treatment of CSF leakage.16,24 As instruments progressively improve, various surgeons have recommended endoscopic surgery for the treatment of more challenging entities, such as basal encephaloceles.5,20,22,23,38 There have been numerous reports on various surgical repair techniques for encephaloceles; however, a paradigm approach has not been elucidated in the literature.

Eight cases of basal encephalocele with a traumatic origin were treated at our institute (a tertiary medical center) within a 10-year period. In this report we describe the surgical repair technique applied via the endoscopic approach with a free tissue graft in an underlay or overlay fashion, and we assess its effectiveness through long-term follow-up of the repair.

Clinical Materials and Methods

Patient Population and Evaluation

Between September 1997 and December 2006, 8 patients with traumatic basal encephaloceles underwent endoscopic surgery. These patients included 2 men and 6 women with a mean age of 39 years (range 26–63 years). Five basal encephaloceles were attributed to head injuries and the other 3 were iatrogenic and represent Cases 3, 6, and 7 in Table 1. The 3 iatrogenic lesions were caused by a previous FESS or related endoscopic sinus procedures performed at another institution. All 5 patients with head injuries were referred from the neurosurgical department within our institution. Each patient had incessant watery rhinorrhea and a nasal polypoid mass on endoscopic exam-
The patients in 7 (88%) of the 8 cases presented with at least 1 episode of meningitis or brain abscess prior to our surgical procedures. Glucose test confirmed that the escaped liquid was CSF in these patients. The encephaloceles and accompanying skull base defects were recognized initially on radiographic studies, which included fine-cut contrast-enhanced CT scanning, magnetic resonance imaging, and contrast-enhanced CT cisternography. Four patients had an encephalocele in the ethmoid foovea, 2 in the cribriform plate, and 2 in the sphenoid sinus. There were no multiple or bilateral lesions. In the head injury group, the patients in Cases 1, 4, 5, and 8 were surgically treated within 3 months of trauma, whereas the patient in Case 2 underwent surgery 10 months after trauma. Surgical timing is listed in Table 1.

Operative Technique

The dural pouch in each patient was freed from surrounding tissue and mucosa, with its pedicle and base exposed. A large dural pouch that was pedunculated and drooped into the nasal cavity was found in 2 patients (Cases 3 and 7) and was tied up at the neck of the pouch. The drooping sack of the pouch was then excised using bipolar cautery, and the stalk and vascular structures were replaced intracranially. In each of the other 6 patients the dural pouch that protruded or herniated through the skull base without a drooping sack was simply reduced through bipolar cautery and then pushed intracranially. The skull bone around the defect was then carefully separated from the dura mater to prevent intractable intracranial bleeding or fracture of the attenuated skull base bone.

In accordance with the location of the encephalocele, a free graft was placed in an underlay fashion to repair the lesions at the anterior skull base (2 cribriform plate and 4 ethmoid foovea defects), whereas an obliteration procedure followed by an overlaying fat graft was used to repair the sphenoid sinus defects.

A free composite graft harvested from the inferior turbinate in the opposite side, which contained mucosa, submucosa, and concha bone, was used to perform repairs (underlay procedure) in the 6 patients with skull defects in the anterior skull base (Fig. 1). We trimmed the graft to fit in with the outline of the skull defect and shaped the submucosa of the graft slightly larger than the concha bone and the mucosal layer of the graft relatively broader than the submucosa. The epithelial side of the graft was positioned to face the nasal side, whereas the trimmed submucosa and concha bone of the graft were placed to face the cranial side (Figs. 2 and 3). After reducing the encephalocele and separating the dural pouch from the surrounding mucosa in 2 patients with sphenoid sinus encephaloceles, the mucosa was removed from the entire sphenoid sinus bone to avoid mucocele formation and to ensure that the graft would be viable. An overlaid fat tissue graft was used to seal off the sphenoid sinus skull defects by using an obliteration procedure (Figs. 4 and 5). A fluorescein test was not used in these patients during the operation because, during endoscopic control, CSF overflow is relatively plentiful, which made the skull base dehiscence sufficiently clear to visualize.

Results

Operative Findings

In addition to their identification on preoperative radiographic studies, all encephaloceles with skull defects were revealed on endoscopy during surgery. All skull defects associated with encephaloceles were 1 cm in diameter, making the overflow of CSF from the skull defect more plentiful than CSF leakage without encephaloceles. We used a single free graft as the repair material in each patient. An overlaid fat graft alone was used in the treatment of 2 sphenoid sinus lesions. Except for the underlaid free tissue graft, no other grafts beneath the skull base were used among patients whose skull defects were located in the cribriform plate or ethmoid foovea.

Infection Control in Patients With a History of Meningitis

Seven patients (88%) had suffered meningitis before

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs), Sex</th>
<th>Cause of Encephalocele</th>
<th>Location of Skull Defect</th>
<th>Meningitis/Brain Abscess</th>
<th>Time of Repair (mos since trauma)</th>
<th>FU (mos)</th>
</tr>
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<tbody>
<tr>
<td>1</td>
<td>40, M</td>
<td>head injury</td>
<td>fovea (ant ethmoid)</td>
<td>1×/never</td>
<td>3</td>
<td>115</td>
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<td>2</td>
<td>35, F</td>
<td>head injury</td>
<td>cribriform plate</td>
<td>2×/never</td>
<td>10</td>
<td>103</td>
</tr>
<tr>
<td>3</td>
<td>33, F</td>
<td>iatrogenic</td>
<td>cribriform plate</td>
<td>2×/never</td>
<td>14</td>
<td>98</td>
</tr>
<tr>
<td>4</td>
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<td>fovea (ant ethmoid)</td>
<td>1×/never</td>
<td>2</td>
<td>71</td>
</tr>
<tr>
<td>5</td>
<td>63, F</td>
<td>head injury</td>
<td>sphenoid</td>
<td>1×/1×</td>
<td>2</td>
<td>70</td>
</tr>
<tr>
<td>6</td>
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<td>fovea (pst ethmoid)</td>
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<td>6</td>
<td>64</td>
</tr>
<tr>
<td>7</td>
<td>35, F</td>
<td>iatrogenic</td>
<td>fovea (ant ethmoid)</td>
<td>2×/never</td>
<td>8</td>
<td>62</td>
</tr>
<tr>
<td>8</td>
<td>26, F</td>
<td>head injury</td>
<td>sphenoid</td>
<td>never/never</td>
<td>2</td>
<td>36</td>
</tr>
</tbody>
</table>

* ant = anterior; FU = follow-up; pst = posterior.
they presented to our clinic; 1 of these patients also had an episode of brain abscess. At a mean follow-up period of 77 months after surgical treatment, there were no central nervous system infections in any of these 7 patients.

Surgical Complications and Follow-Up

Successful endoscopic repairs were accomplished on the first attempt in all patients. No major postoperative complications or recurrences were noted at an average follow-up period of 77 months (range 36–115 months).

Discussion

Disease Origin and Anatomical Location

Basal encephaloceles represent rare entities that result from herniation of the meninges and brain parenchyma through a skull base defect into the sinonasal region. These lesions could be congenital in origin, representing a primary anomaly of the neural tube and its skeletal cover. Traumatic basal encephaloceles are categorized as lesions due to head injury or iatrogenic conditions following sinusonal surgery. In a literature review, cases of iatrogenic origin were noted to be less common than those caused by head injury (Table 2). A basal encephalocele is often complicated by CSF rhinorrhea, recurrent meningitis, headaches, and on occasion subdural hematoma (a sequela of loss of suspension from CSF). The communication with the nasal cavity in cases of basal encephalocele carries a significantly higher risk of CSF leakage and neurosurgical complications than that in other types of encephaloceles. Anterior basal encephaloceles usually involve herniation of the frontal lobe tissue through an anterior cranial fossa defect into the ethmoid sinus or nasal cavity. They can also result from temporal lobe herniation through a middle fossa defect into the sphenoid sinus. The ethmoid and sphenoid sinuses are the most common sites for encephalocele development.

Endoscopy: From Novel to Popular Treatment Method

With the rapid advancements in its instrumentation, the endoscopic approach greatly enhances visualization of the entire sinonasal roof and has become the mainstream approach in the treatment of skull base and sinonasal diseases, including the repair of CSF rhinorrhea. In 1981 Wiggand became the pioneer of endoscopy in the treatment of CSF rhinorrhea caused by FESS. Mattox and Kennedy further defined the technique and indications for the endoscopic management of CSF leakage and cephaloceles. Since then an increasing number of surgeons have used the endoscope to treat encephaloceles with accompanying CSF leakage. The successful outcomes of the endoscopic approach have opened surgeons to an alternative treatment of this skull base lesion without the risk of trancranial morbidity.
Note, however, that overlooking an encephalocele on endoscopy can lead to a failure of repair and fatal complications such as delayed meningitis. Meticulous endoscopic controls during the operation are essential to identifying skull lesions. Detailed, methodical history gathering and thorough evaluations of radiographic studies are also imperative for the diagnosis of basal encephaloceles.

**Encephalocele Incidence and Previous Treatment Experiences**

To our knowledge, encephaloceles are rare and reportedly occur in 1 in 35,000 live births. Stankiewicz reported no instances of traumatic encephaloceles in a review of complications in 300 cases of endoscopic ethmoidectomy. A low incidence of major complications with no instance of iatrogenic basal encephaloceles was cited by Lawson, who had reviewed published articles relating to 34 studies with > 20,000 procedures of intranasal sinus surgery. In 1990 Mattox and Kennedy reported 2 cases of traumatic encephaloceles, caused by a head injury in 1 patient and a transantral ethmoidectomy in another. Hudgins and colleagues presented a case of iatrogenic nasal encephalocele induced by an injury to the floor of the anterior cranial fossa following an FESS procedure of sphenoethmoidectomy and maxillary antrostomy. Wolansky and associates described cases of traumatic encephaloceles caused by head injuries.
Endoscopic treatment of traumatic basal encephalocele

Fig. 5. Case 5. A and B: Computed tomography cisternograms showing a sphenoid sinus encephalocele with CSF leakage (arrows, collections of contrast extracranially). C: Postoperative endoscopic image demonstrating mucosal epithelialization of an overlaid graft (arrows).

scribed a case of iatrogenic encephalocele in a patient who had undergone an intranasal ethmoidectomy. Boseley and Tami described 5 cases of encephaloceles in 4 patients; 2 lesions in 1 patient and 1 lesion in another patient had been caused by an endoscopic sinus procedure. Lanza and colleagues had the largest number of endoscopically repaired encephaloceles in a 4-year study, which included 11 patients with 12 encephaloceles out of 36 skull base defects. Marshall and associates identified 6 patients, each with 1 basal encephalocele, in a 6-year review of patients who had undergone endoscopic repair for CSF rhinorrhea. Note that in the latter 2 studies, the authors did not define the encephaloceles as congenital or traumatic lesions. The present study included 8 traumatic lesions, 3 of which were associated with previous FESS procedures.

Distinctive Operative Technique

In our patients, the dural pouch of the encephalocele was either excised (Cases 3 and 7) or reduced (all other cases) by bipolar cautery. Our experience revealed that a large or droopy sack of the encephalocele can be associated with difficulties in reducing the expanded pouch or replacing it into the intracranial position, leading to poor identification of the skull defect or difficulty in separating the dura mater from the skull bone. In addition, the pushing of an oversized dural pouch back into the cranium can act as a focus for infections and increase the risk of sepsis. Therefore, excision or amputation of the pouch is necessary because the neural tissue within the encephalocele is redundant, necrotic, and nonfunctioning. However, the loop of vessels should be preserved for their distal blood supply. The neck of the drooping dural sack in 2 of the patients in the present study (Cases 3 and 7) was tied up before cautery excision was performed, which made the endoscopic excision easier, and the reconstruction of the skull bone defect became safer and swifter. Hence, postsurgical morbidity may be avoided.

Indications for Underlay, Overlay, Turinate Graft, and Obliteration in Different Locations

Because dura mater does not regenerate, an adequate barrier separating the sinonasal region from the cranium is necessary for the reconstruction of the skull bone defect and the prevention of meningitis after amputation or excision of the encephalocele. Our results demonstrated that an underlaid composite graft offered a promising closure and efficaciously prevented ascending infection from the nose to the cranium. Based on the literature, both overlay and underlay procedures appear to offer similar success rates in the management of CSF rhinorrhea. However, an over-

<table>
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<tr>
<th>Authors &amp; Year</th>
<th>Age (yrs), Sex</th>
<th>Location of Skull Defect</th>
<th>Previous Meningitis</th>
<th>Prior Surgery (mos ago)</th>
<th>Treatment</th>
<th>FU (mos)</th>
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<tr>
<td>Mattox &amp; Kennedy, 1990</td>
<td>50, F</td>
<td>fovea (pst ethmoid)</td>
<td>1×</td>
<td>transanal ethmoidectomy (24)</td>
<td>ESS†</td>
<td>7</td>
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<td>Hudgins et al., 1992</td>
<td>43, F</td>
<td>ant cranial fossa</td>
<td>1×</td>
<td>FESS (8)</td>
<td>(refused surgery)</td>
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<tr>
<td>Wolansky et al., 1998</td>
<td>35, F</td>
<td>fovea &amp; cribiform plate</td>
<td>NA</td>
<td>intranasal ethmoidectomy (3)</td>
<td>craniotomy</td>
<td>NA</td>
</tr>
<tr>
<td>Boseley &amp; Tami, 2004</td>
<td>58, M</td>
<td>fovea (pt ethmoid)</td>
<td>1×</td>
<td>FESS (NA)</td>
<td>ESS‡</td>
<td>12</td>
</tr>
<tr>
<td>Lee et al., 2004</td>
<td>33, F</td>
<td>cribiform plate</td>
<td>2×</td>
<td>FESS (24)</td>
<td>endoscopic repair</td>
<td>6</td>
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<tr>
<td>present study</td>
<td>37, M</td>
<td>fovea (pt ethmoid)</td>
<td>1×</td>
<td>FESS (6)</td>
<td>ESS§</td>
<td>64</td>
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<tr>
<td>present study</td>
<td>35, F</td>
<td>fovea (ant ethmoid)</td>
<td>2×</td>
<td>FESS (8)</td>
<td>ESS</td>
<td>62</td>
</tr>
</tbody>
</table>

* ESS = endoscopic sinus surgery; NA = data not available.
† Endoscopic repair with middle turbinate bone and a pedicled septal mucosal graft.
‡ Endoscopic repair with acellular dermis and hydroxyapatite cement.
§ Endoscopic repair with an underlaid composite graft.
laid graft is inherently easier to displace than an underlay graft, especially on a steep or bumpy surface of the skull base bone. When the skull defect is > 1.0 cm in diameter, a sturdy graft is required to act as a stable barrier between the cranium and nasal cavity to uphold the herniated sac of the encephalocele. Based on this factor and according to our experience, a turbinate composite graft placed between the dura and skull bone (underlay procedure) is stable enough and less likely to slip away, and therefore is more favorable than placing the graft beneath the skull bone (overlay procedure). These matters are particularly important in the management of the anterior skull defect including the cribriform plate and anterior ethmoid fovea. Although the separation of the dura from the skull bone is dangerous in the region of the cribriform plate, cases in the present study involved encephaloceles with a large defect in the anterior skull base (> 1.0 cm in diameter), which made an underlay procedure more conceivable than in cases without encephaloceles.

Compared with the inferior turbinate, the middle turbinate—with its thinner layer of mucosa, flimsier concha bone, and sometimes hypoplastic anatomy—was deemed insufficient for repair of the skull defect. A composite graft of the inferior turbinate, as a repair material, contains thick mucosa, submucosa, and a broad concha bone and provides both a multiple-layer closure effect and enough volume to harvest. We stress the need for inferior turbinate composite grafts for the repair of encephaloceles and suggest trimming the graft to fit the outline of the skull defect before placement of the underlay graft, and shaping the submucosa and mucosa of the graft into a broader piece than the trimmed concha bone to prevent insufficient graft size and achieve an adequate seal. Among the patients with a basal encephalocele in the anterior cranial fossa (ethmoid fovea and cribriform plate) we did not use any additional grafts beneath the skull base, such as a pedicled septal mucosal graft or an overlaid temporalis fascia, apart from an underlaid free graft. Overall, a fitting composite graft in an underlay fashion provides a satisfactory closure effect for the reconstruction of the basal encephalocele in the anterior skull base.

Skull defects within the sphenoid sinus are difficult to manage and repair with neurosurgical or endoscopic approaches because of the angular surface or hypoplasia of the sphenoid sinus antrum. Placement of a graft in the epidural space within the sphenoid sinus is therefore time-consuming and appears to be unsafe. Concerns about inadvertent damage to the optic nerve or carotid artery might impose restrictions on graft placement during endoscopic sphenoid sinus surgery. Therefore, after detaching the overlying mucosa of the encephalocele and reducing the pouch intracrani ally, placement of an overlaid fat graft in a customized tissue bulk following an obliteration procedure of the sphenoid sinus, when supported by proper reinforced packing, seems to be a more practicable and efficient method than an underlaid grafting procedure for the management of the sphenoid sinus encephalocele. Furthermore, it is necessary to stress removal of the mucosa from the entire sphenoid sinus bone to ensure that the graft is viable and to avoid a mucocele.

Long-term observation is required for the evaluation of the seal-off effect of the obliteration procedure in sphenoid sinus lesions. In this study, the cases of 2 patients with a sphenoid encephalocele remained uneventful after follow-up periods of 70 and 36 months.

**Lumbar Drainage and Neurosurgical Backup in Special Cases**

Cerebrospinal fluid rhinorrhea should be treated aggressively, because of the potential risks of pneumocephalus, ascending meningitis, and fatal brain abscess. Most cases of CSF rhinorrhea can be initially managed by the insertion of a lumbar drain. Diverting CSF through a lumbar drain can prevent the formation of a fistula and thus reduce the risk of infection. Surgical repair was contemplated only if lumbar drainage failed to resolve the leak or the leak was a result of an intractable skull defect caused by encephaloceles. We observed that most skull defects in these patients were larger than those associated with CSF leakage without an encephalocele, making CSF overflow from the defect far more plentiful and surgical repair a necessity. Moreover, because the strong gush of the CSF will potentially push the graft out, a lumbar drain can decrease the intracranial pressure as well as minimize CSF pressure fluctuation. In our experience with promising results, lumbar draining was performed in 18 (46.2%) of 39 patients with CSF rhinorrhea at the time of surgical repair, including the defects repaired using an underlay procedure (8 patients) and those in the sphenoid sinus (7 patients). Although the lumbar drain is not indispensable in patients with CSF rhinorrhea, it was applied in patients in the present study to facilitate the graft adhesion rate and prevent graft displacement in the management of basal encephaloceles. Endoscopic treatment for a basal encephalocele is not without limitations. A skull defect > 1.5 cm in diameter and involving the area of the planum sphenoidale and tuberculum sellae may represent a limitation of the endoscopic procedure. Based on previous reports, the shortest distance (15 mm) between both sides of the carotid prominence was located just below the tuberculum sellae in 72% of cases. The risk of injury to major vessels or the pituitary-hypothalamus axis is high and has fatal consequences. Appropriate neurosurgical support is therefore required in such situations. A larger-scale study is warranted to evaluate possible complications of underlaid placement of a free graft at the anterior skull base, such as damage to the olfactory bulb with resultant anosmia.

**Treatment Timing: Before and During Meningitis Development**

Seven (88%) of 8 patients in this study had incurred bacterial meningitis before presenting to our institute, and 1 had suffered an episode of brain abscess, which can develop from an ascending infection from the sinonasal tract. Because the incidence of meningitis has been reported to be as high as 33.9%, we emphasized that the repair procedure obliterated the source of the CNS infection effectively, especially during an active infection. The effective and uneventful result perhaps can be attributed to our use of an allograft instead of foreign material.

**Conclusions**

As endoscopy becomes more popular, the deleterious complications of endoscopic surgery, such as CSF rhi-
norhea, seem unavoidable. With the improvement of endoscopic techniques and instrumentation, an increasing number of surgeons use the endoscope to manage CSF rhinorhea while avoiding intracranial morbidities. However, meticulous manipulation of the endoscope following a precise grafting procedure is mandatory for the successful repair of a more intractable skull defect in the basal encephalocele. In treating this challenging lesion, we stress the need for underlay placement of a turbinate composite graft with a skull defect can be successfully repaired by placing a free tissue graft via endoscopy. Experience in free graft placement techniques is of the utmost importance to the successful treatment of traumatic basal encephaloceles.

References


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