Operative Techniques in Otolaryngology 30 (2019) 30–36

Operative Techniques in

Otolaryngology

Pediatric skull base surgery: Encephaloceles and cerebrospinal fluid leaks

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KEYWORDS
Pediatric;
Cranial base;
Endonasal;
Endoscopic;
Encephalocele;
Cerebrospinal Fluid Leak

In recent years, endoscopic endonasal approaches to the pediatric skull base have become an increasingly popular method for treatment of a variety of malignant and benign lesions. These approaches have largely evolved from experience in adult patients and can be an excellent alternative to open procedures in carefully selected patients. Pediatric encephaloceles can be addressed via open craniotomy, transpalatal, and endoscopic endonasal methods. The surgical approach should be dictated by the anatomy of the patient as well as the size and location of the encephalocele in question. Pediatric cerebrospinal fluid leaks can occur due to iatrogenic causes during skull base resections or sinus surgery, as well as from traumatic injury. Multiple options for repair of cerebrospinal fluid leaks exist, but endoscopic nasoseptal flaps are among the most commonly utilized reconstruction options.

Introduction

An endoscopic endonasal approach (EEA) to the skull base has grown in popularity in recent years as an alternative to an open procedure.[1-4] Endoscopy offers exceptional visualization, no external incisions, and often more rapid recovery than open procedures.[5,6] As technology and experience with endoscopic techniques have progressed, Otolaryngologists and Neurosurgeons have increasingly employed endoscopy to approach lesions of the skull base.[7,8] EEAs have been shown to be safe and efficacious alternatives to open approaches in the hands of experienced surgeons.[9]

Initially utilized in adult patients, surgeons more recently have begun to utilize EEAs in pediatric patients. Initial concern that pediatric EEA may interfere with cran-

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http://doi.org/10.1016/j.otot.2019.01.012
1043-1810/

iofacial growth has not turned out to be an issue in several published series. Additionally, the relatively small nasal passages and underdeveloped sinuses typical of the pediatric population have not prevented safe EEA surgery even in very young patients. Multiple centers have reported applicability, safety, and efficacy of the endoscopic approach in pediatric cohorts.[5,10-12]

As pediatric EEA experience has grown, surgeons have increasingly used it to address a variety of complex skull base lesions. This manuscript focuses on the current state of pediatric cerebrospinal fluid (CSF) leak and encephalocele management. Focus is given to alternatives to open surgery. This includes review of the current state of pediatric EEA surgery in managing these conditions. Additionally, the transpalatal approach is reviewed as it can be a useful approach, particularly in appropriately selected neonates.

Encephalocele

Defects in the skull base, either congenital or acquired, can lead to herniation of cranial contents. Traditionally, meningocele designates herniation of meninges and CSF, while meningoencephalocele refers to herniation of meninges, CSF, and brain tissue.[13] Pediatric encephaloceles are most commonly congenital, but can be the result of surgical interventions or traumatic injury.[14,15] Congenital encephaloceles may be noted incidentally when imaging is obtained for other reasons or due to symptoms of intracranial complication (meningitis, central nervous system infection).[16] Additionally, encephalocele can herniate in the nasal cavity or nasopharynx and present with varying degrees of nasal obstruction, snoring, or obstructive sleep apnea.[17]

Evaluation of an encephalocele requires nasal endoscopy, full neurologic examination, imaging often with both computed tomography (CT) and magnetic resonance imaging (MRI). Encephaloceles that involve the sellar region can present with endocrinopathy, necessitating a full endocrinology work up.[18-20] A small number of traumatic encephaloceles are self-resolving, but the majority of congenital or postsurgical encephaloceles require operative repair.[21] The primary goals of surgery are to reduce the encephalocele, reconstruct the skull base defect, and preserve function of involved central nervous system structures. Consequently, this should also relieve associated symptoms such as nasal obstruction.[22]

The majority of pediatric encephaloceles are congenital, which can be further subdivided into frontoethmoid (60%), basal (30%), or both (10%). Basal defects are described with respect to the location of the anatomic defect. Spheno-orbital, transethmoidal, sphenoethmoidal, and transsphenoidal basal encephaloceles have been described.[22] The size, location, and contents of the encephalocele, as well as the age and comorbidities of the patient, help guide the surgeon in choosing an appropriate operative approach.

There are at least 3 approaches to pediatric encephalocele: open, transpalatal, and endoscopic transnasal (Table 1). All 3 approaches have a role in modern skull base surgery, and careful patient selection is critical. Historically, pediatric encephaloceles were addressed using an open craniotomy approach.[11] Very large encephaloceles with facial deformity or intracranial complications such as rupture of the encephalocele are relative indications for open craniotomy approach.[14,23] Various open craniotomy approaches are well described for management of pediatric encephalocele.[23-25]

The transpalatal approach is less commonly used than either open or endonasal approaches as evidenced by the infrequent number of times it is reported in the literature.[26] The rare use of the transpalatal approach could be in part due to relative surgeon unfamiliarity with the technique. Further, the transpalatal approach is most useful only when the encephalocele and defect are located in the nasopharynx (the skull base location that is situated behind the palate). When appropriate conditions arise, however, the transpalatal approach can provide excellent exposure with little morbidity related to the palate division and repair.[26] The senior author (JCR) finds the transpalatal approach most useful for patients with both of the following conditions: (1) neonatal age whereby small nasal passages may limit an EEA approach, and (2) the encephalocele sack and skull base defect are located in the nasopharynx. In such

Table 1 Comparison of surgical approaches used in repair of pediatric encephalocele

Approach	Advantages	Disadvantages
Open Craniotomy	Able to address all locations of skull base	Requires external incisions and traditional craniotomy, often longer recovery time and increased postsurgical morbidity, can be difficult to reduce hernia sac contents from above
Transpalatal	Excellent visualization of many nasopharyngeal lesions even in very young children, able to reduce hernia sac contents from below	Requires palatal incision, poor visualization of sellar and anterior skull base lesions
Endonasal Endoscopic	Excellent endoscopic visualization of most skull base locations, able to reduce hernia sac contents from below	Limited visualization for more lateral skull base locations, Potentially more difficult in younger children, bloody surgical field can challenge visualization, endonasal skull base reconstruction can be challenging

a situation, division of the soft palate, and sometimes the posterior portion of the hard palate, provides direct visualization of the nasopharynx, encephalocele sac, and skull base defect.

Case example

A 10-month-old male infant with history of term twin birth following an uncomplicated pregnancy presented to Otolaryngology clinic with complaints of progressive noisy breathing and poor weight gain. Parents had noted severe snoring and poor sleep and polysomnogram showed severe obstructive sleep apnea. On examination, he was found to have subtle dysmorphic features but no identifiable genetic syndrome. Flexible nasal endoscopy in clinic showed midline nasal mass. MRI imaging showed a midline transsphenoidal encephalocele with brain tissue herniated into the nasopharynx associated with dysgenesis of the corpus callosum and dilation of the third ventricle (Figure 1). After appropriate preoperative workup including endocrine and ophthalmology evaluation, he was taken for transpalatal approach and repair of the encephalocele with a free calvarial bone graft (Figures 2 and 3). Follow-up imaging 4 months after surgery showed stable skull base reconstruction and no recurrence of encephalocele (Figure 4). See the supplemental resources for a video highlighting the transpalatal approach (Video 1).

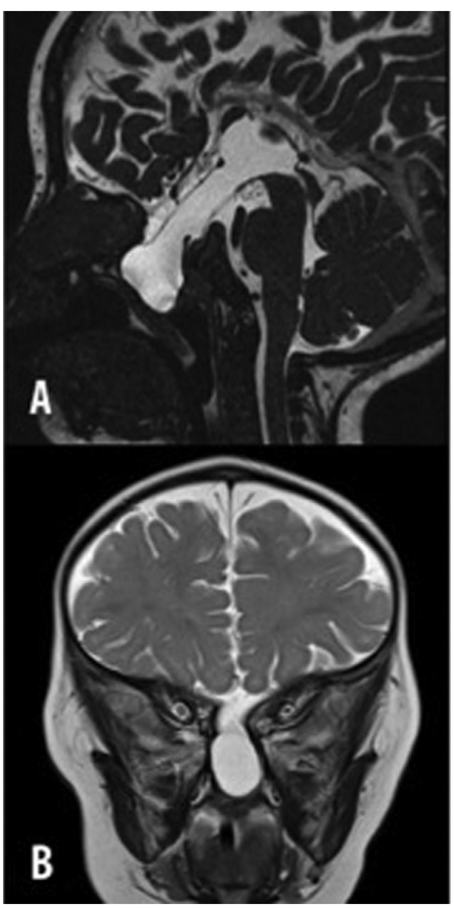

Figure 1 Coronal and sagittal views of T2 MRI showing large midline transsphenoidal encephalocele with herniation into the nasopharynx. MRI, magnetic resonance imaging.

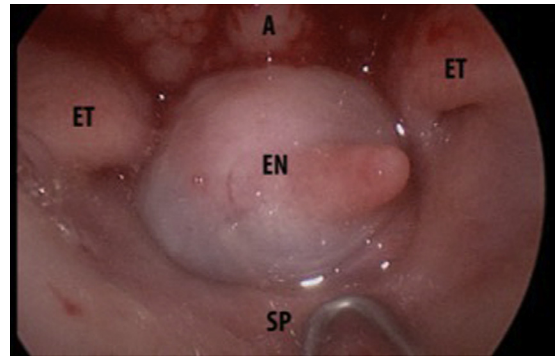

Figure 2 Intraoperative view via 120° transoral endoscope (A, adenoid; ET, eustachian tube; EN, encephalocele; SP, soft palate).

As surgeons have gained experience with EEAs in pediatric patients, endonasal approaches are more frequently being used to treat encephaloceles in appropriately selected patients. EEAs to address an encephalocele ultimately have the same goals as open or transpalatal approach as previously discussed. Advantages to using endoscopes include avoiding external incisions and often excellent visualization. A bloody field can present challenges with endoscopic visualization, but this can be adequately managed with experienced endoscopic surgeons and modern equipment. There have historically been several considerations unique to pediatric EEA (1) that skull base intervention will negatively affect skull base growth and development and (2) that the small nasal passages in young children will prohibit effective endoscopic instrumentation.

The skull develops from 3 components, membranous neurocranium (forming the flat bones of the skull), cartilaginous neurocranium or chondrocranium (forming most of the skull base), and viscerocranium (forming the bones of the facial skeleton).[27] The anterior skull base acts as a scaffold for develop of the facial skeleton, and, as a result, endoscopic intervention has the potential to negatively impact craniofacial growth and development. However, in case series to date, there has been no impairment of craniofacial development reported to date.[5,10-12] Long-term follow-up will be required for patients who have undergone endoscopic resection of skull base lesions to determine if there are any delayed effects on skull growth that have not been represented in the current data.

At the advent of the endonasal approach, there was also concern that in very young children the nasal passage may not be large enough to accommodate an endoscope and instrumentation necessary for safe resection of skull base lesions. While it is true that the age of the patient and size of the sinonasal cavities are inversely correlated, the EEA approach has been successfully used in children even under 2 years old.[28-30] Authors have noted that the procedure is somewhat more cumbersome in extremely young patients, but has been made possible by using smaller endoscopes

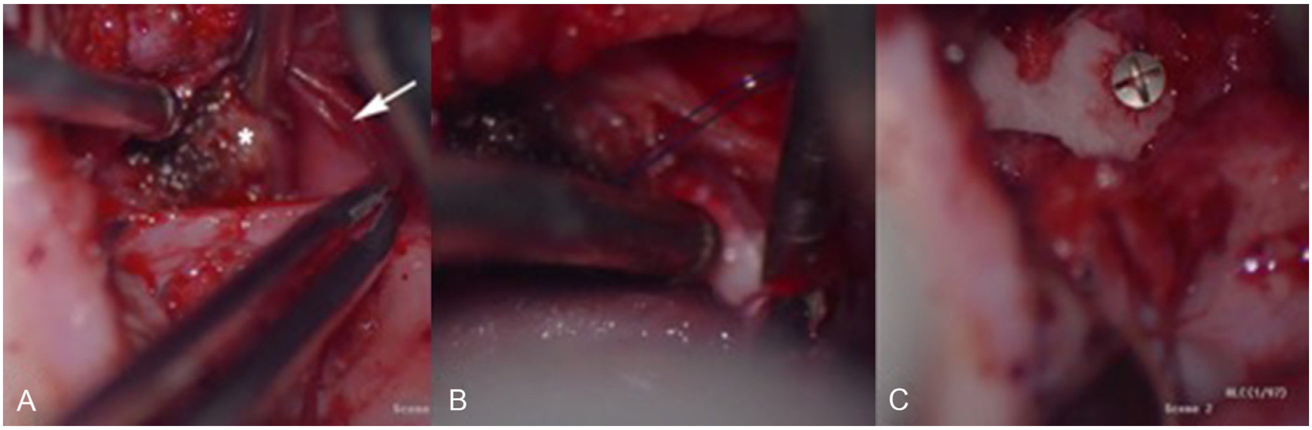

Figure 3 (A) Incision of the nasopharyngeal mucosa (white arrow) and separation of the dural sac (white asterisk) from the mucosa. (B) Purse string suture for dural plication and further reduction of the dural sac. (C) Free calvarial bone graft affixed to the clivus with a screw. Following this, the surgery is completed with closure of the nasopharyngeal mucosa over the bone graft, replacement of the hard palate bone flap, and standard multilayer repair of the soft palate.

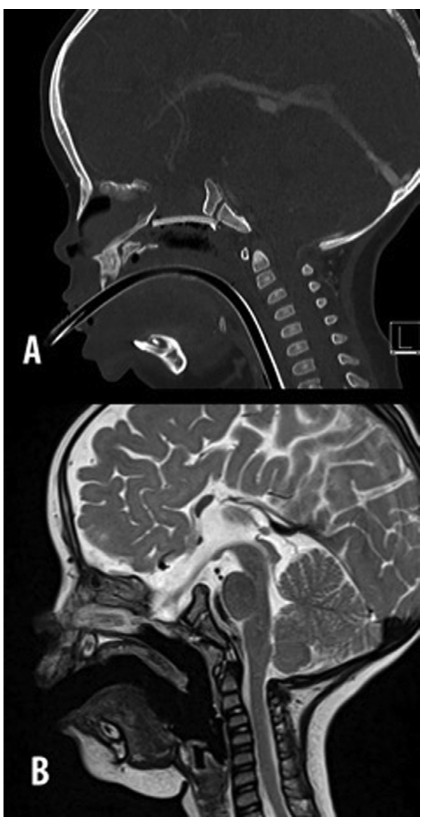

Figure 4 (A) Postoperative CT scan showing bone flap in place. (B) T2 MRI obtained 4 months postoperatively, showing stable reduction of encephalocele. Patient is now 2 years postoperation with stable vision and endocrine function. CT, computed tomography; MRI, magnetic resonance imaging.

and adopting the use of microsurgical instruments rather than traditional endoscopic sinus surgery instruments.[21]

In addition, the adoption of intraoperative CT- and MRI-based navigation has been helpful in young children with undeveloped paranasal sinuses. EEA uses the nasal passage and paranasal sinuses to access the skull base. If a sinus has not formed, the operation often it requires more extensive drilling with fewer surgical landmarks. Intraoperative navigation allows the surgeon to confirm his or her anatomical position with good reliability, although it is no substitute for careful dissection and identification of intranasal landmarks.

Finally, the importance of a team approach to skull base surgery cannot be overstated. Surgery of the skull base requires specialized knowledge of the 3-dimensional relationships between the lesion and critical structures including the carotid arteries, cranial nerves, orbit, and brain. Research has shown that effective collaboration is improved with increasing case volume and consistent roles within the Otolaryngology and Neurosurgery teams.[31] Furthermore, teams that work together on an ongoing basis are more effective at dealing with intraoperative emergencies such as internal carotid injury in animal models.[32] It is also crucial to collaborate with Neurosurgeons facile at open craniotomy in case the encephalocele cannot be addressed endoscopically or if there are intracranial complications from the encephalocele or during surgical resection.

CSF leak

Pediatric CSF leak repair borrows many of the same repair techniques that have been popularized in adults. CSF leaks may be categorized into iatrogenic versus noniatrogenic and traumatic versus spontaneous causes. Noniatrogenic, spontaneous CSF leaks are associated with increased intracranial pressure in about 70% of cases, with benign intracranial hypertension being the most common cause of elevated intracranial pressure.[33] Traumatic, noniatrogenic CSF leaks are most commonly due to accidental injury and occur in 2%-4% of head injuries. Most resolve with conservative measures including bed rest, head elevation, lumbar drainage, and diuretics.[34,35] If persistent for more than 1-2 weeks, repair is recommended to prevent meningitis.[33] Iatrogenic CSF leaks occur during skull base resections as well as sinus dissection for chronic rhinosinusitis.[33,36]

Table 2 Sinonasal and scalp regional flaps useful in skull base reconstruction. Associated arterial pedicles are noted on the right

Sinonasal	
Nasoseptal flap	Posterior septal arteries
Inferior turbinate flap	Branches of the sphenopalatine artery
Middle turbinate flap	Branches of the sphenopalatine artery
Scalp	
Pericranial/galeocranial flap	Supraorbital and supratrochlear arteries
Temporoparietal flap	Superficial temporal artery
Temporalis muscle flap	Deep temporal arteries

Repair of CSF leak involves dissection of native sinuses, identification and exposure of skull base defect, denuding of surrounding mucosa, and then watertight closure with a flap or graft. Available reconstruction options fall into several categories: nasal, scalp, free grafts, and free flaps (Table 2).

The "work horse" reconstruction flap option for pediatric skull base reconstruction is the nasoseptal flap. Initially described in 2006, it is a pedicled mucoperichondrial and mucoperiosteal flap based on the posterior septal branches of the sphenopalatine artery.[37] See supplemental resources for a video demonstrating raising of a nasoseptal flap (Video 2). Advantages include ease of harvest and no need for separate operative field or incision outside the nose. CSF leak rates after endoscopic reconstruction are comparable to open techniques in the adult literature.[38] There are less data in pediatric patients but current data suggest that endoscopic and open techniques are comparable in regard to CSF leak.[5,39] The length of flap that can be harvested is dependent on nasal growth. A 2009 study using craniofacial CT scans and NSF flap modeling suggested that the width of NSF is likely to be sufficient at all ages but length may be insufficient under 6-7 years old for transsellar defects.[39] While it remains important to consider whether NSF size will be adequate in children, more recent case series have shown adequate length and successful CSF leak control in children as young as 2-4 years old following transsellar surgery.[5,11]

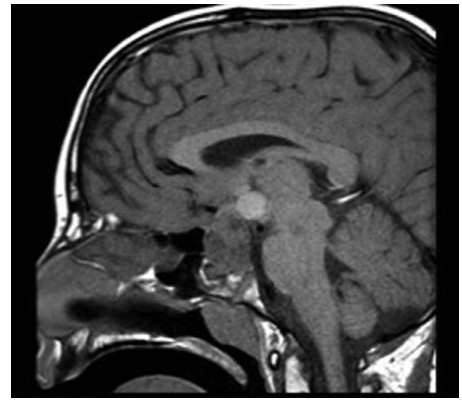

Figure 5 Preoperative MRI T1 postcontrast image showing a sagittal section demonstrating a large, heterogeneous and partially cystic mass, centered around the sella. This appearance is characteristic of craniopharyngioma. MRI, magnetic resonance imaging.

Case example

A 12-year-old male presented to Otolaryngology clinic for evaluation of sellar lesion extending into the suprasellar space found during work up for growth delay and growth hormone deficiency. MRI and CT preoperatively were consistent with probable craniopharyngioma (Figure 5). After formal visual field testing was found to be intact and appropriate counseling of the parents and patient, he was taken for endoscopic endonasal resection. Resection resulted in an expected intraoperative CSF leak. Reconstruction of the skull base and control of the CSF leak were performed with a multilayer repair augmented with a NSF (Figure 6). The NSF provided complete coverage and the intraoperative CSF leak was successfully controlled (Figure 7). See supplemental materials for a video demon-

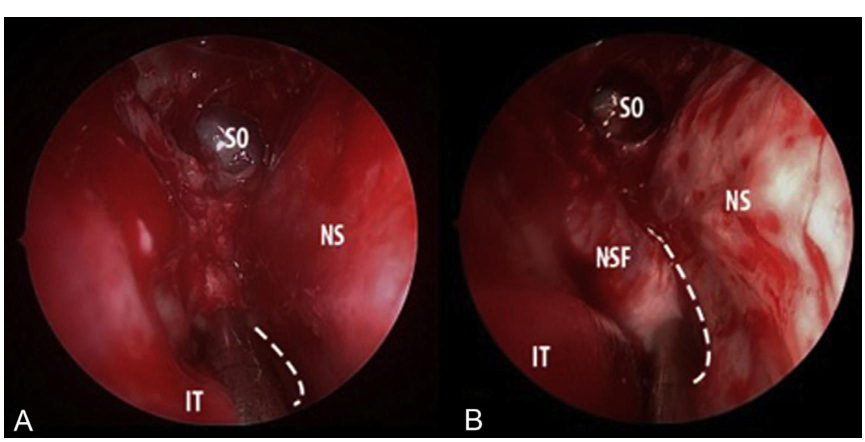

Figure 6 (A) Endoscopic view of the right nasal cavity prior to NSF elevation. The sphenoid ostium has been surgically opened. Dashed line represents the right choana and posterior aspect of the septum (IT, inferior turbinate; NS, nasal septum; SO, sphenoid ostium). (B) Endoscopic view of the right nasal cavity after elevation of the NSF. The exposed bone and cartilage of the septum can be seen. The NSF is stored in the nasopharynx to protect it while the sellar lesion is resected. NSF, nasoseptal flap.

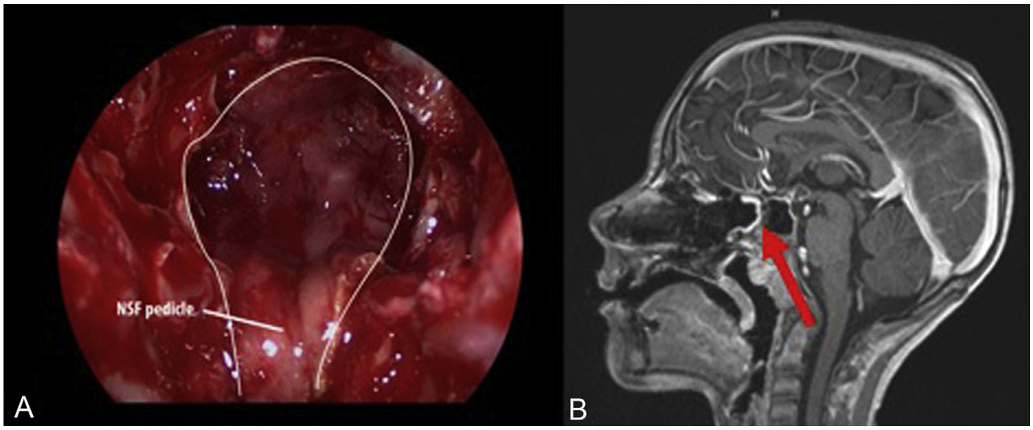

Figure 7 (A) Endoscopic view of the NSF following inset. The sphenoid face has been open widely and the NSF flap is outlined in white. The NSF sits flush against the resection border creating a watertight seal. The pedicle of the NSF arising from the septal branch of the right sphenopalatine artery is also illustrated crossing the sphenoid rostrum. (B) T1 postcontrast MRI showing good position and adequate coverage of the NSF (arrow). CT, computed tomography; NSF, nasoseptal flap.

strating raising a NSF (Video 2) and reconstruction of the skull base using the NSF (Video 3).

In patients who have a skull base lesion not amenable to NSF or in patients who have had a previous NSF that failed, surgeons have turned to a variety of other flaps and grafts. One such option is the temporoparietal fascial flap which is raised based off the superficial temporal artery and can be tunneled through the pterygopalatine fossa to be placed on the skull base.[40] The pericranial flap based off of supraorbital and supratrochlear arteries can be used to address lesions of the anterior cranial skull base or frontal sinus. Free flap reconstruction of the skull base remains difficult given the narrow working corridor of the nose and relative paucity of vessels for microvascular anastomosis.

Conclusion

The field of pediatric endoscopic endonasal surgery has progressed rapidly in the last several years as surgeons become more comfortable visualizing and resecting increasingly challenging lesions. Techniques currently used have largely been adopted from the adult skull base experience. Pediatric encephaloceles can be addressed via the open craniotomy approach, transpalatal, or EEA. The surgical approach should be tailored to the unique skull base lesion being addressed, although endoscopic endonasal approaches are now often being used for lesions that traditionally would require open craniotomy. Likewise, CSF leak repairs via the endoscopic view have become more widely adopted, with the nasoseptal flap being one of the most common reconstructive methods employed.

Disclosure

The authors reported no proprietary or commercial interest in any product mentioned or concept discussed in this article.

Supplementary material

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.otot.2019.01.012.

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