Repair of Giant Anterior Skull Base Encephalocele Containing Intralesional Eloquent Brain: Technical Note

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BACKGROUND: Encephaloceles are herniations of intracranial neural tissue and meninges through defects in the skull. Basal encephaloceles are rare anterior skull base defects incident in 1 in 35,000 live births. Sphenoethmoidal encephaloceles are even more uncommon, with an incidence of 1 in 700,000 live births. Anterior skull base encephaloceles may be life-threatening in infants, presenting as airway obstruction and respiratory compromise. They can also present with cerebrospinal fluid (CSF) rhinorrhea, purulent nasal drainage, or meningitis.

OBJECTIVE: To report a novel technique for repairing a giant sphenoethmoidal encephalocele containing eloquent neural tissue.

METHODS: A 16-mo-old girl presented with progressive airway obstruction from a giant sphenoethmoidal encephalocele that filled her oral cavity. She had multiple congenital anomalies including agenesis of the corpus callosum and cleft lip and palate. Computed tomography showed complete absence of the bony anterior cranial base, and magnetic resonance imaging demonstrated the presence of the pituitary gland and hypothalamus in the hernia sac.

RESULTS: We repaired the encephalocele using a combined microsurgical and endoscopic multidisciplinary approach working through transcranial, transnasal, and transpalatal corridors. The procedure was completed in a single stage, during which the midline cleft lip was also repaired. The child made an excellent neurological and aesthetic recovery with preservation of pituitary and hypothalamic function, without evidence of CSF fistula.

CONCLUSION: The authors describe a novel multidisciplinary technique for treating a giant sphenoethmoidal encephalocele containing eloquent brain. The cleft lip was also repaired at the same time. The ability to work through multiple corridors can enhance the safety and efficacy of an often-treacherous operative endeavor.

KEY WORDS: Encephalocele, Meningoencephalocele, Sincipital, Skull base defect, Transcranial, Transnasal, Transpalatal

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Congenital encephaloceles are protrusions of the brain and its meninges through a defect in the developing skull. They are categorized by location and are differentiated from meningoceles by the neural or glial elements contained within the sac.[1,2] Himly and Serres[3] were the first to propose this concept of herniation through bony defects in the early 19th century. Basal encephaloceles are the rarest variety and occur in one of 35 000 live births. They are categorized with other anterior (sincipital) encephaloceles, differentiating them from those of the cranial vault, such as occipital encephaloceles.[4,5] The further classification of basal herniations include transethmoidal, transsphenoidal, sphenoethmoidal, sphenomaxillary, and sphenoorbital subtypes. Although some anterior encephaloceles protrude between the frontal, nasal, and ethmoid bones as visible, skin-covered protrusions on the face, others can present as insidiously expanding lesions that are typically discovered in the naso-oropharynx.[6,7] Despite their concealment, they may become life-threatening in obligate nasal-breathing infants, presenting with airway obstruction, snoring, cerebrospinal fluid (CSF) rhinorrhea,

ABBREVIATION: EXIT, Ex Utero Intrapartum Treatment

recurrent meningitis, or purulent nasal discharge. Associated neurologic conditions may include hypothalamic-pituitary dysfunction, optic nerve anomalies, and agenesis of the corpus callosum.[8,9] Associated craniofacial malformations may include cleft lip, cleft palate, and hypertelorism, which may further impact treatment strategies.[1]

Several surgical approaches have been described for the management of these complex anterior skull base encephaloceles, including transnasal, transpalatal, and transcranial approaches. The transcranial approach was first described by Walter Dandy in 1929[1] and has been the mainstay of treatment for complex giant encephaloceles since. Substantial advances in anatomical modeling, hemostasis in young infants, and the management of postoperative meningitis, and infection have increased the safety of this approach.[3,10] Recent reports have introduced minimally invasive endoscopic transnasal and transpalatal approaches as effective ways to manage frontobasal encephaloceles. In such cases, the lesions were often associated with small bony defects and did not contain eloquent brain tissue in the encephalocele sac. These newer approaches are powerful but may provide insufficient exposure for giant encephaloceles with eloquent tissue.

Here, we describe a novel 3-discipline, 3-corridor approach for the repair of a large skull base encephalocele containing eloquent brain, rarely reported in the literature. We utilized a combined transcranial, transnasal, and intraoral transpalatal technique, and the procedure was carried out in a single sitting by neurosurgery, otolaryngology, and plastic and reconstructive surgery. Although a subset of these disciplines and corridors have been utilized in the past, the use of all for this large repair, their timing and coordination throughout the procedure, as well as the harvesting of a bone graft from the same cranial incision makes this approach novel. Neurological function was preserved, and there was no evidence of a CSF leak. The patient's cleft lip was repaired with a nice aesthetic result.

METHODS

Clinical Presentation

This 16-mo-old girl was referred for evaluation of airway obstruction, copious oral secretions, and swallowing difficulties secondary to an expanding, giant sphenoethmoidal encephalocele containing eloquent brain. She also had a cleft lip and palate.

She was born via cesarean section at 36 weeks' gestation with an Ex Utero Intrapartum Treatment (EXIT) procedure for placement of a tracheostomy due to airway obstruction from the encephalocele diagnosed prenatally. The child was given up for adoption by her birth mother, who had Asperger syndrome and thromboangiitis obliterans (Buerger disease). She underwent placement of a right occipital ventriculoperitoneal shunt for hydrocephalus in early infancy, followed by a percutaneous endoscopic gastrostomy for feeding. She was adopted at age 10 mo and was followed for the enlarging encephalocele that occupied most of her oropharynx and hypopharynx and was felt to be inoperable by the referring hospital.

The patient was slightly developmentally delayed but was alert and interactive and began sitting at age 11 mo. She had hypertelorism and a cleft lip and palate through which the encephalocele was clearly visible (Figure 1). Echocardiogram demonstrated a right-sided aortic arch and an aberrant left subclavian artery diverticulum with mass effect on the esophagus and the left mainstem bronchus.

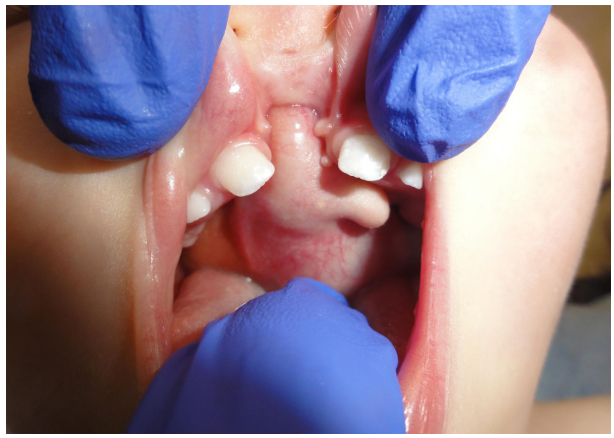

FIGURE 1. *Intraoperative image demonstrating the encephalocele as visualized through the oral cavity. Note the multiple daughter sacs on the lesion.*

Magnetic resonance imaging (MRI) showed the large sphenoethmoidal encephalocele filling the oropharynx and extending inferiorly to the level of the 2nd cervical vertebra. The sella turcica was grossly underdeveloped, with components of the pituitary gland and hypothalamus plastered to the posterior wall of the encephalocele sac. Portions of the inferior frontal lobes and the proximal anterior cerebral arteries had herniated through the skull base defect. Agenesis of the corpus callosum was noted with associated colpocephaly (Figure 2). Computed tomography (CT) showed that the bony floor of the anterior cranial fossa was essentially absent, with an enormous defect spanning from the orbits to the underdeveloped clivus (Figure 3).

Institutional Review Board permission was granted, and written consent was obtained from the patient's mother for the presentation of this case and for the publication of the patient's image.

Surgical Management

A multidisciplinary approach was selected and rehearsed preoperatively by teams from neurosurgery, otolaryngology, and plastic and reconstructive surgery. Endocrinologic work-up was unremarkable.

The patient was positioned supine with the brow up and the head supported on a horseshoe rest (Figure 4). She was registered to the frameless electromagnetic guidance system (Axiem, Medtronic, Dublin, Ireland). Intravenous clindamycin and hydrocortisone were administered. The entire face and scalp were prepared and draped. Neurosurgery began with a bicoronal scalp incision and mobilization of a large vascularized pericranial graft based on the supraorbital arteries. A bifrontal craniotomy was elevated, and the dura was separated from the anterior skull base under microsurgical guidance. The brain was full in spite of intravenous mannitol and hyperventilation. Therefore, the orbital bar was removed bilaterally creating better access to the anterior cranial base. We identified a widened area at the foramen cecum, where the anterior portion of the encephalocele was identified coursing inferiorly.

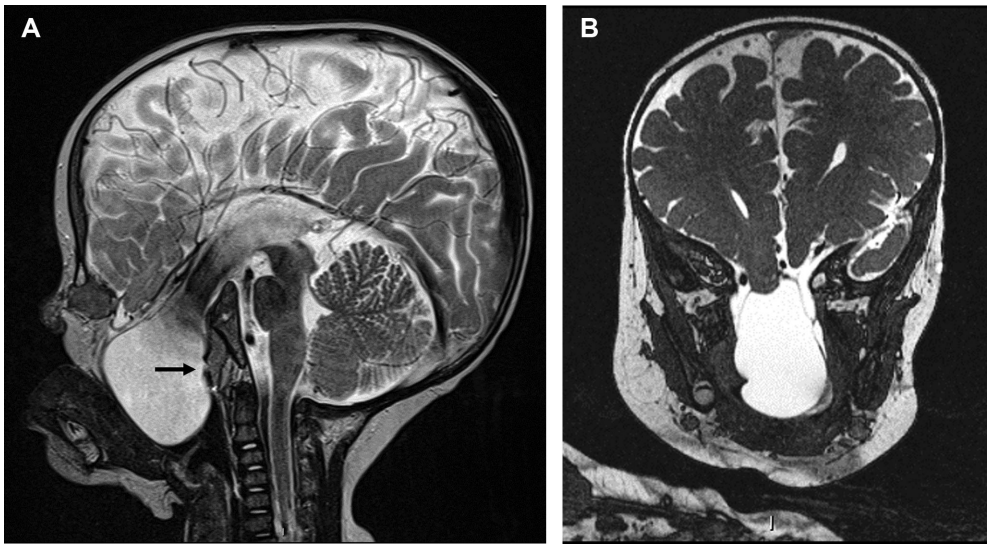

FIGURE 2. *Sagittal* **A** *and coronal* **B** *T2-weighted MRI demonstrating the large anterior skull base defect with the protruding encephalocele into the oral cavity. The giant sphenoethmoidal encephalocele is noted to be extending through the anterior cranial base and filling the oropharynx. Dysmorphic pituitary and hypothalamic tissue are in the sac posteriorly (arrow), along with portions of the frontal lobes and proximal anterior cerebral arteries. There is agenesis of the corpus callosum.*

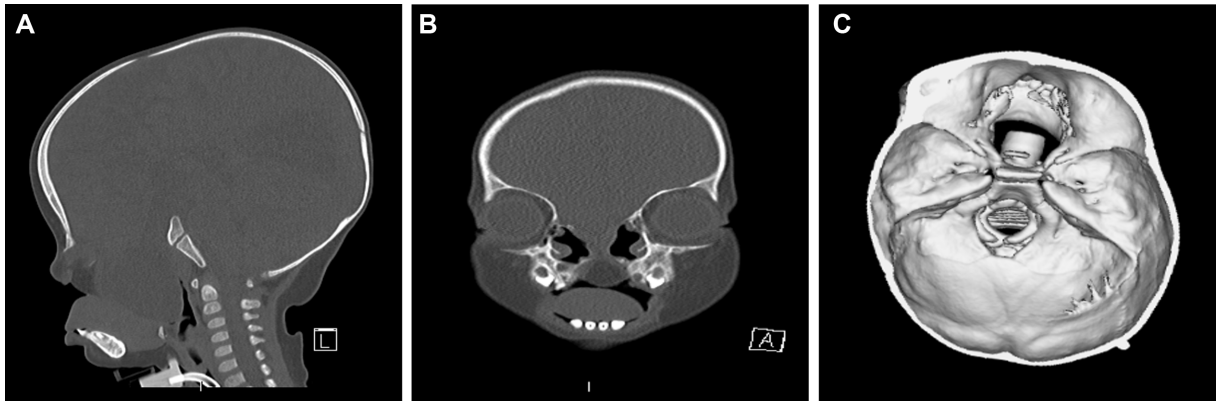

FIGURE 3. *Sagittal* **A**, *coronal* **B**, *and 3-dimensional reconstructed axial CT demonstrating the extent of the large skull base defect.* **C**, *The tracheostomy tube is seen from above through the defect in the skull base.*

Next, the plastic surgery team worked in the oral cavity. Exposure was maximized with a Dingman retractor. They performed a tedious dissection, separating the encephalocele sac from the oral mucosa. The otolaryngology team worked transnasally using 0- and 30-degree endoscopes. A midline nasal split was performed to facilitate the dissection through the open cleft lip and nose. Otolaryngology and plastic surgery widened exposure of the extracranial portion of the encephalocele, working endoscopically through the nasal and oral cavities in an anterior to posterior direction.

At this point, the 3 surgical teams began working simultaneously to come around the neck of the large encephalocele, with neurosurgery working microsurgically from above and otolaryngology and plastic surgery working endoscopically from below. The dissection was carried out posteriorly to within 1 cm of the posterior lip of the skull base defect adjacent to the clivus. Because the encephalocele sac was too large to mobilize further, we chose to decompress it in a controlled fashion. We punctured the anterior sac through the oral cavity with a needle and removed CSF, taking care not to injure the viable pituitary and hypothalamus. The puncture site was then closed with 4-0 NUROLON sutures (J&J Medical Devices). The posterior sac was separated from adenoid tissue working endoscopically from below.

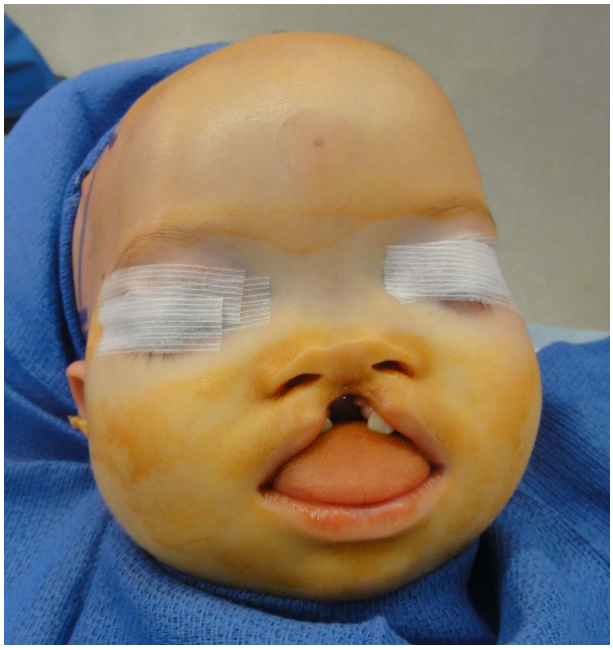

FIGURE 4. *Intraoperative image demonstrating the patient's cleft lip and palate as well as hypertelorism.*

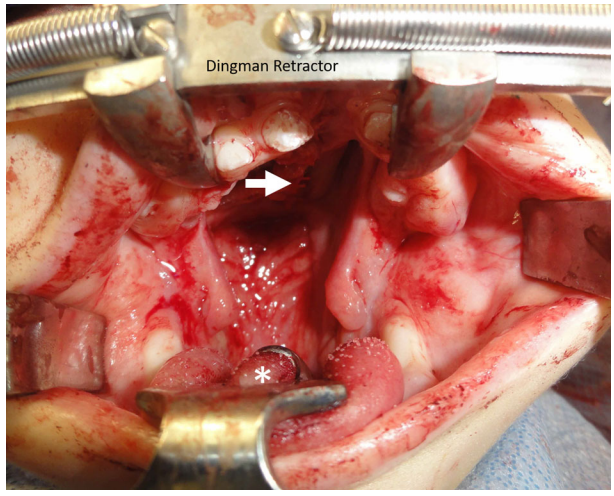

FIGURE 6. *Intraoperative transoral image demonstrating the palatal repair from below utilizing a Dingman retractor to optimize visualization of the oral cavity and the suture line along the mucosa (white arrow). The tongue is denoted by the white asterisk, and the teeth are in view superiorly in the image underneath the Dingman retractor.*

The deflated sac was then pulled up into the intracranial compartment (Figure 5). The vascularized pericranial graft was then mobilized and brought inferiorly into the nasopharyngeal cavity where it was tacked down from below to the periosteum, and the surrounding tissue of the clivus with 4-0 NUROLON sutures (J&J Medical Devices). This provided complete coverage of the defect. Biological glue (Evicel Fibrin Sealant®, Ethicon, Bridgewater, New Jersey) was then applied intracranially to cover the vascularized pericranial graft and exclude the intracranial compartment from the nasopharyngeal and oral cavities below.

Next, a left parietal bone graft was elevated and remodeled to fit the contours of the large defect in the skull base and wedged in place supported by bone over the orbits bilaterally. Bone shavings from the inner table of the graft along with demineralized bone matrix were used to fill in the defect at the cranial donor site. The nasal mucosa was reconstructed in the mouth with interrupted 4-0 VICRYL sutures (J&J Medical Devices) (Figure 6). The bifrontal craniotomy and orbital bar were reconstructed with absorbable plates and screws (SonicWeld, KLS Martin, Jacksonville, Florida). The scalp was closed in standard layers,

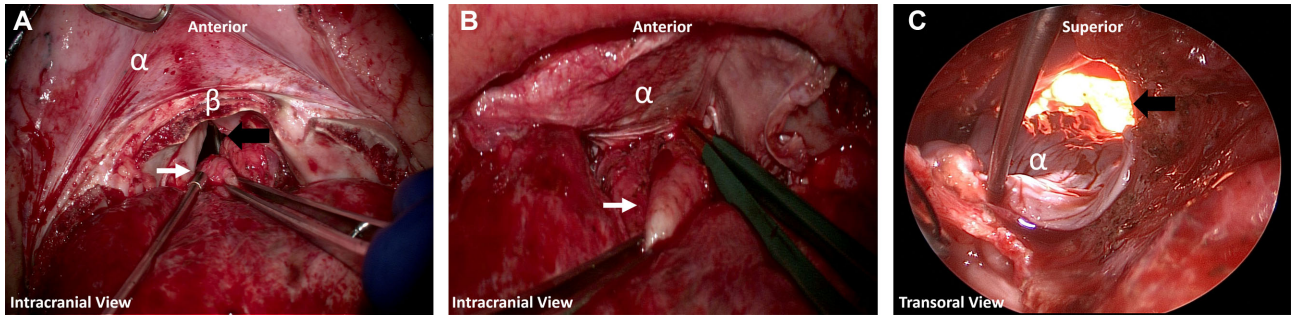

FIGURE 5. *Intraoperative views from the transcranial and transoral approaches.* **A**, *Demonstrated is an intracranial view of the meticulous reduction of the encephalocele (white arrow) into the intracranial cavity through the skull base defect (black arrow) after release from the oral and nasal mucosa below. The alpha symbol denotes the pericranial flap and the beta symbol the cut bone edge from the craniotomy.* **B**, *Demonstrated is the lowering of the pericranial graft (alpha symbol) into the defect in order to exclude the encephalocele (white arrow) from the oral cavity.* **C**, *An endoscopic transoral view from within the oropharynx looking up into the intracranial cavity demonstrating the skull base defect (black arrow), and the pericranial graft (alpha symbol) prior to its suturing to the posterior dura in the first step of defect closure.*

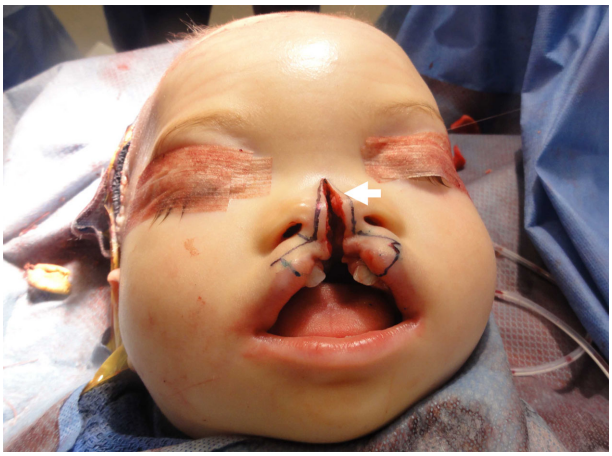

FIGURE 7. *Intraoperative plan for the repair of the patient's nasal deformity and cleft lip and palate utilizing a modified inferior triangle technique. The white arrow denotes the incision line created at the start of the procedure.*

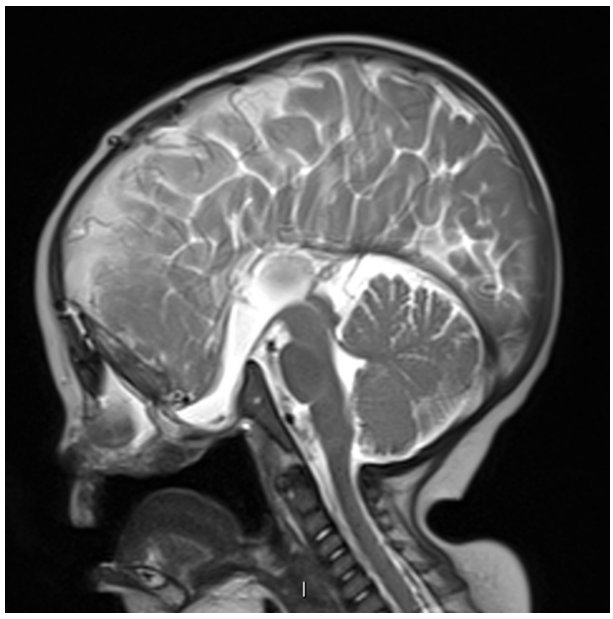

FIGURE 8. *Postoperative sagittal T2-weighted MRI demonstrating the repair of the encephalocele and its return into the cranial cavity overlying our operative repair of the skull base.*

and a drain was left in place. The nasal deformity and midline cleft lip were repaired utilizing a modified inferior triangle technique (Figure 7). The palatal defect was large and the lateral palatal shelves so small that it was decided that the palatal repair may require microvascular free tissue transfer in the future.

The patient awakened from anesthesia hemodynamically stable, moving all extremities. The estimated blood loss was 100 ml, and the operative time was 8 h. Although no specific considerations were given to regards to anesthetic usage, a senior anesthesia team experienced with complicated cases such as these was used. No tranexamic acid or aminocaproic acid was administered.

RESULTS

Postoperative Course

There were minor fluctuations in the patient's serum sodium immediately postoperatively that normalized within 48 h. There was no evidence of diabetes insipidus and no evidence of a CSF fistula. She had episodic fevers on postoperative day 3 and was started on broad spectrum antibiotics with sputum cultures growing *Klebsiella pneumoniae*. She developed midface swelling on postoperative day 5 after the postoperative drain had been removed the day prior and needle aspiration of the scalp recovered purulent material. She returned to the odds ratio (OR) for oral exploration to ensure the safety and sterility of our construct. Although this was an unfortunate complication, it was the consequence of the surgical field being located near the oral and nasal cavities, and occurred on day 5 after the drain in the same area was removed. The return to the OR was swift in order to protect the integrity of the construct and minimize the possibility for further infection. The mucosal closure was intact, but 5 ml of purulent fluid was evacuated from the anterior oronasal cavity in the vicinity of the glabella. A red rubber catheter was left in place temporarily. Wound cultures grew multiple organisms and she received broad spectrum antibiotics. She underwent further transoral irrigation and 2 wk of antibiotics with resolution of the infection. She was discharged home in good condition. Postoperative MRI showed significant reduction in the size of the encephalocele and removal of the obstructing mass in the oropharynx (Figure 8). By 6 mo after surgery, she had made a dramatic improvement and was happy, alert, and interactive, making developmental gains, pulling to a stand, and taking steps with assistance. Her incisions healed with an excellent cosmetic result (Figure 9).

DISCUSSION

Key Results

Here, we describe a novel 3-discipline, 3-corridor technique to repair a giant anterior skull base sphenoethmoidal encephalocele that contained eloquent brain. The repair permitted safe mobilization of the encephalocele and its eloquent neural contents. Utilization of a vascularized pericranial flap and a parietal calvarial autograft from the same incision led to a durable reconstruction of the anterior skull base without a CSF leak. The patient's cleft lip was repaired in the same sitting with a nice aesthetic result. She continues to improve neurologically without any endocrinopathies.

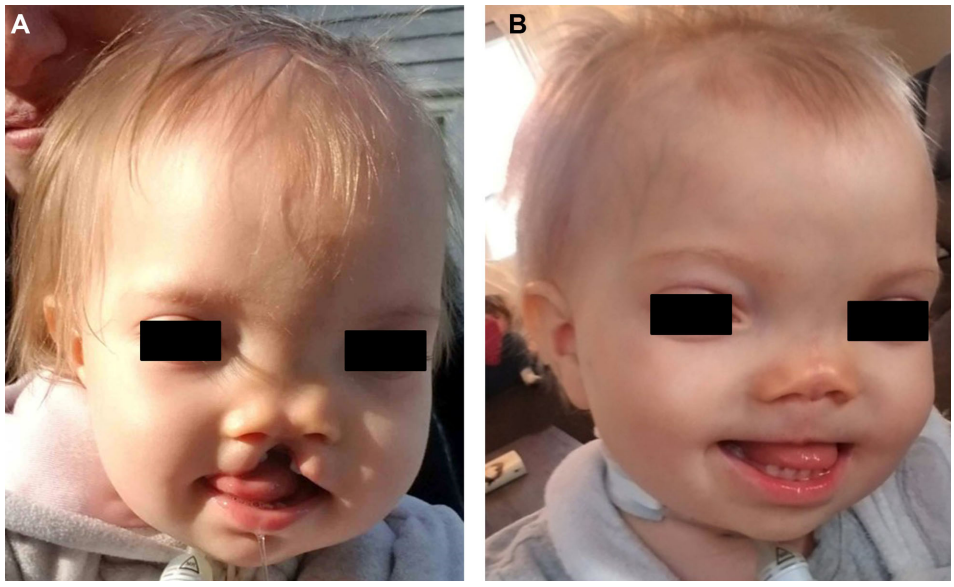

FIGURE 9. *Gross appearance of the patient before* **A** *and 3 mo after* **B** *operative repair.*

Clinical Diagnosis

On clinical examination, encephaloceles of the anterior cranial base may be confused with other lesions of the naso- and oropharynx including dermoids, lipomas, fibromas, teratomas, hemangiomas, cysts, polyps, and nasal gliomas. Radiographic examination with MRI will usually confirm the diagnosis. The Furstenberg test has been described as a way to help differentiate encephaloceles from these other lesions by visualizing a glistening blue or pink mass that pulsates with the heartrate, swells with compression of the jugular veins and is cross-fluctuant with pressure on the anterior fontanelle.[1]

Operative Approach

There is no one-size-fits all technique for the surgical correction of basal encephaloceles. Transnasal and transpalatal routes with endoscopic guidance may be ideal for repair of some lesions without eloquent neural tissue that are associated with small bony defects. Drawbacks of this technique are the small size of the working space and the possible need for an additional incision to source a bone graft to avoid utilizing artificial materials in infants.[11-15]

Transcranial approaches remain the mainstay of treatment for giant anterior skull base encephaloceles.[15,16] In the past, poor posterior visualization utilizing this approach sometimes led to incomplete repairs and the development of meningitis.[15,17] Combining the transcranial approach with endoscopic transnasal and/or transoral approaches can enhance the safety and efficacy of the procedure and offer a substantial benefit for circumferential visualization of the defect.[15] Others have described cases in which a subsequent delayed second surgical procedure was required for cleft lip repair, when a single combined approach would have sufficed.[18,19] Although postoperative infections are a significant concern in these cases, there were no special antibiotic considerations made preoperatively in this case; however, because of the requirement for swift washout of a focal postoperative collection, a broader and longer course of antibiotics may be considered for these case in the future.

Limitations and risks are reported with transcranial approaches, as well as for endoscopic repairs, and include seizures, meningitis, sinusitis, and intracranial abscess formation.[20,21] Thus, all approaches pose risks, and each must be carefully selected to maximize the intended repair. A greater risk of infection is present with incomplete repair of the encephalocele when there is poor visualization of the entire lesion.[19]

Surgical Pearls

Based on the senior author's experience with this and other large, complex encephaloceles of the cranial base, we offer several pearls for the successful management of these defects. (1) When a multidisciplinary approach is chosen, detailed preoperative planning and rehearsal should be performed with careful review of the imaging to identify eloquent structures in the sac that must be preserved and to anticipate difficulties in reconstructing the cranial base. (2) The order of surgical exposures should be determined preoperatively: which team starts, who goes next, and when the teams will work simultaneously. (3) For young children, in whom lumbar spinal drainage to relax the brain may be challenging, removal of the orbital bandeau can facilitate exposure of the cranial base. (4) Deflating the sac can help with its mobilization. Eloquent neural tissue within the sac should not

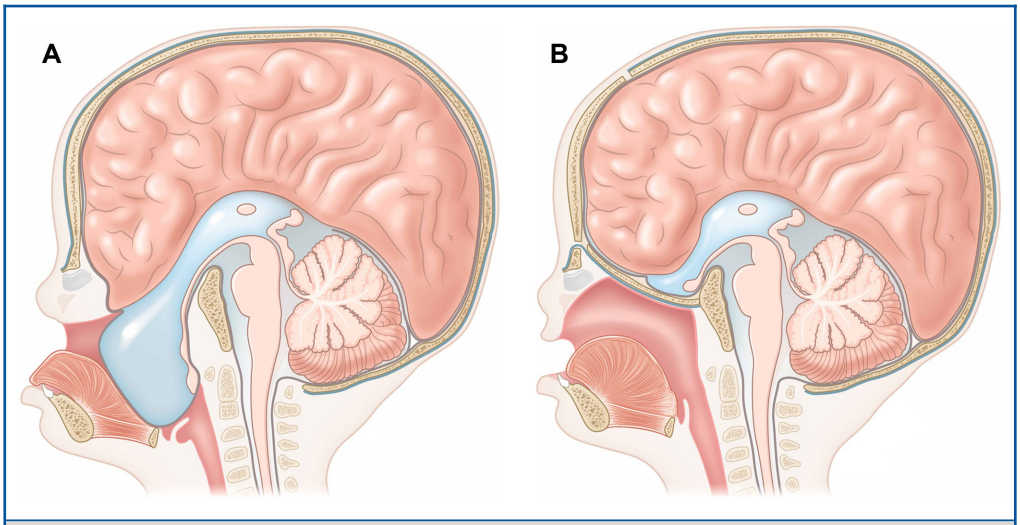

FIGURE 10. *Artist's depiction of the repair of the herniated encephalocele.* **A**, *Depicted is the preoperative defect.* **B**, *Depicted is the postoperative repair. Note the pericranial graft tucked underneath the harvested parietal bone slinging up the encephalocele sac. The repaired mucosa is noted in the oral cavity, and midline tissue has been reinstated to close the cleft defect.*

be sacrificed.[16] (5) Endoscope-assisted microsurgery can aid in visualizing the geometry of the encephalocele. (6) Coagulation of the hernia sac should be avoided when it contains eloquent brain, as this can lead to neurological and endocrinological abnormalities.[22-24] In a report of 6 transsphenoidally resected encephaloceles, all 6 patients developed endocrinopathies as a result of aggressive resection of herniated sacs.[25] Basal encephaloceles that do not contain eloquent neural tissue can be managed endoscopically with coagulation of the sac to shrink it. (7) A vascularized pericranial graft can help prevent a CSF leak.[26] (8) Whenever possible, autologous calvarial grafts should be utilized for reconstructing the skull base in young children, to avoid the use of titanium mesh and other foreign bodies. The use of rib grafts in infants and toddlers has been reported[27], but cranial bone harvested from the same incision seems superior. (9) Facial deformities (eg, cleft lip and palate), when present, can be repaired during the same surgical sitting. (10) The swift return to the OR with concern for infection in the operative bed, given the proximity to the oral and nasal cavities, can help prevent further complications and protect the construct.

CONCLUSION

We report the successful management of a giant sphenoethmoidal encephalocele containing eloquent brain using a combined intracranial, transnasal, and transoral approach (Figure 10). Meticulous attention must be paid to preserve eloquent intralesional tissue, and deflating the sac can help with its management. The use of a vascularized pericranial graft and autologous bone can provide durable reconstruction of the cranial base and help avoid a CSF fistula. Cleft lip and palate, when present, can be repaired in the same sitting if surrounding tissue is adequate for closure. A multidisciplinary team approach that crosses the boundaries of the skull base can facilitate the repair of complex anterior basal encephaloceles.

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Disclosures

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

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