J Neurosurg (3 Suppl Pediatrics) 107:266–271, 2007

Transpalatal endoscopic endonasal resection of a giant epignathus skull base teratoma in a newborn

Case report

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✓Teratomas are neoplasms composed of tissues from all three germ layers with varying degrees of differentiation. They are most commonly found in the sacrococcygeal and gonadal regions and rarely occur in the head and neck region. A teratoma is termed "epignathus" when it arises from the skull base or hard palate and is located in the oral cavity. The authors describe a case of a giant epignathus teratoma originating in the skull base of a neonate, extending bilaterally via two pedicles throughout the hard palate and protruding through the oral cavity. The tumor was completely resected using a transpalatal endoscopic endonasal approach. The excised tumor proved to be an immature teratoma with well-differentiated yolk sac elements. At the 1-year follow-up the patient showed no evidence of tumor recurrence and the child remains neurologically intact.

This report demonstrates the use of a transpalatal endonasal corridor in a preterm infant. This approach provided an ample corridor into the ventral skull base without the need for external excisions and/or disruption of osseous elements. *(DOI: 10.3171/PED-07/09/266)*

Key Words • endoscopy • epignathus teratoma • pediatric neurosurgery • skull base • transpalatal endonasal approach

Teratomas are tumors that derive from all three germ cell types and can be mature or immature. Teratomas can occur in any location in the body, although they usually occur in the midline and are congenital.[3,7,21] Teratoma tumors are generally rare, with an incidence of one in 4000 live births; are found predominantly in females; and are associated with an 18% risk of other congenital malformations.[3,5] Only 10% of all teratomas occur in the head and neck region and only 2% are oropharyngeal.[5,30]

The term "epignathus" is used for teratomas located in the oral cavity or pharyngeal region when they arise from craniofacial bones, mainly from the palatine, sphenoid, or ethmoid bones.[10] Epignathus presentation of teratomas is very uncommon and this tumor presentation is associated with a high mortality rate during the neonatal period.[2,10,11,25] The incidence rate of epignathus teratomas has been estimated to be between one in 35,000 to one in 800,000 live births.[5,31]

Epignathus teratomas can be so large that they can protrude from the oral cavity or extend along the eustachian tube into the middle ear. By filling up the mouth and nasal cavity, these tumors naturally cause a substantial obstruction of the upper airways. Because the presence of an epignathus teratoma is a life threatening condition at the time of delivery, a prenatal diagnosis is essential to coordinate the treatment and appropriate management of these tumors.[2,10,11,14,28] The patient's airway must be secured by performing either an endotracheal intubation or tracheostomy. Definitive treatment consists of completely resecting the tumor.[10,14,31] To date, the standard technique used for resecting these lesions has consisted of traditional craniofacial approaches with microscopic assistance.[5,10,14,23,25,28] In the last decade, there has been a progressive introduction of the endoscope to surgically access the ventral skull base. Recently, various skull base diseases have been managed with the assistance of endoscopes, including small teratomas.[20,22,32] This is the first

Abbreviations used in this paper: AFP = alpha-fetoprotein; CT = computed tomography; MR = magnetic resonance; NICU = neonatal intensive care unit.

report in the literature, however, of using an endoscopic endonasal approach to resect a giant epignathus teratoma arising from the skull base.

Case Report

History and Examination. This 33-week preterm female infant with a large skull base teratoma was born to a 36-year-old healthy woman. The mother had two other children from past pregnancies who were both delivered full-term without complications. The mother's first antenatal visit occurred at the 14th week of pregnancy. Her first ultrasonography examination was performed at approximately 21 weeks of pregnancy, and was found to be unremarkable. At 27 weeks of pregnancy, however, a follow-up ultrasonographic image showed a fetal neck mass. On subsequent ultrasonography, it was noted that the fetal neck mass was increasing in size, and that moderate to severe polyhydramnios had developed.

At 28 weeks of pregnancy the mother was briefly admitted to the hospital for preterm labor symptoms, treated using indomethacin and betamethasone to induce lung maturity, and was sent home. As part of her evaluation at this time, an MR image of her pelvis and lower abdomen was obtained to more accurately evaluate the infant's neck mass, which showed a complex cystic-solid mass in the skull base extending back to the oropharyngeal region (Fig. 1). The woman had a normal fetal echocardiogram and normal levels of AFP.

Delivery Planning and Course. The mother was readmitted at 32 weeks of pregnancy and observed very closely with serial abdominal ultrasonography examinations. At this time, it was noted that the polyhydramnios as well as the size of the fetal neck mass was increasing. A multidisciplinary approach was used to address the patient's problems. The mother was regularly examined by physicians and staff in the Neonatology, Pediatric Surgery, Maternal Fetal Medicine, Otolaryngology, and Neurosurgery departments. Because the fetal neck mass was increasing in size, a decision was made that the infant would be electively delivered at 33 weeks of gestation.

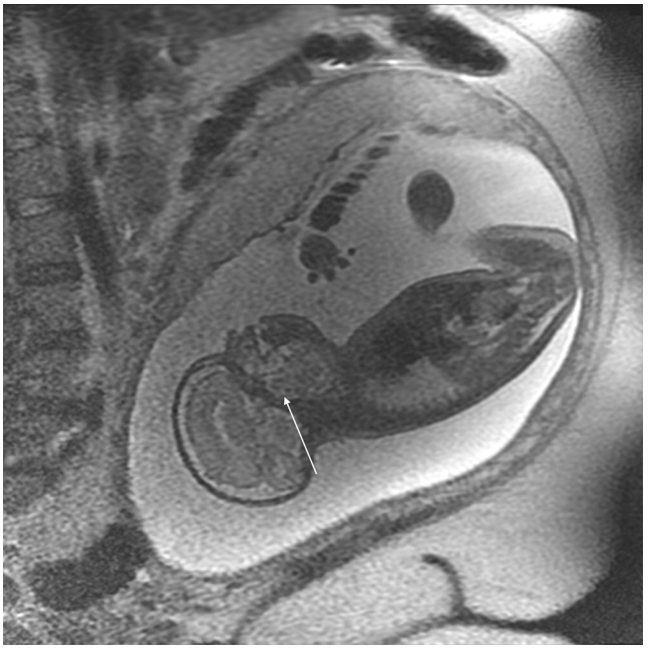

FIG. 1. Pelvic MR image obtained in a 36-year-old pregnant woman, showing a fetus with a skull base teratoma in an initial stage in which the tumor had not yet protruded through the mouth. The *arrow* points to the skull base origin of the disease.

It was the belief of the multidisciplinary team that blockage of the airway would become a significant problem because the epignathus teratoma had caused complete obstruction of the oropharynx. The need for an immediate tracheostomy procedure was anticipated and scheduled to be performed immediately after the head and neck of the baby had emerged from the mother and while the child was still attached to the placenta. At the time, there was a concern that the tracheostomy would not be feasible if the location of the neck mass precluded direct access to the trachea; therefore, cannulization using extracorporeal membrane oxygenation was planned.

The mother was admitted prior to the anticipated delivery date because of a premature rupture of membranes that initiated labor. A cesarean section was performed 1 day before it was planned. In an organized manner, the delivery was attended by physicians and staff from the Pediatric Surgery, Maternal Fetal Medicine, and Pediatric Otolaryngology and Craniofacial departments. When the baby's head and neck emerged, it was noted that the neck area was accessible and not obstructed by the teratoma, and therefore a tracheostomy could be performed prior to separating the baby from the placenta. The mass was noted to be located completely intraorally with a substantial exophytic component obliterating the entire oral cavity and extending well beyond the mandible, with the tongue protruding below it. At the time of the tracheostomy procedure, the infant was noted to be bradycardic with a heart rate of approximately 60 beats per minute. A neonatal cardiac emergency was called. Chest compressions were started and intramuscular epinephrine was administered. The infant received three administrations of epinephrine and a dose of atropine during this resuscitation. The resuscitation continued while the tracheostomy was being attempted, which took 8 to 9 minutes. The child tolerated resuscitation well, with excellent improvement in opening of the airway and in perfusion. The infant was immediately transported to the NICU with a 2.5 mm tracheostomy tube sutured in place.

Surgical Planning. We felt it was appropriate to allow the child to stabilize after delivery prior to proceeding with definitive tumor resection. Therefore, the child remained in the NICU undergoing fluid optimization for pending surgery. The neonate's preoperative serum level of AFP was 94,035 ng/mL, within the normal range ($<$ 170,000 ng/mL). Preoperative CT scans with 3D reconstruction (Fig. 2) showed that the teratoma originated along the entire ventral skull base from the level of the cribriform plate through the planum sphenoidale and along the ventral clivus. The tumor then extended along a defect in the hard palate into the oropharynx, creating a large exophytic component that filled the oropharynx. Secondary effects of the tumor were noted within the oropharynx and nasopharynx as well as a dramatic expansion of the mandible and remodeling of the entire oropharyngeal cavity. The tumor was a heterogeneous mass formed by fat and calcified tissues extending through both sides of

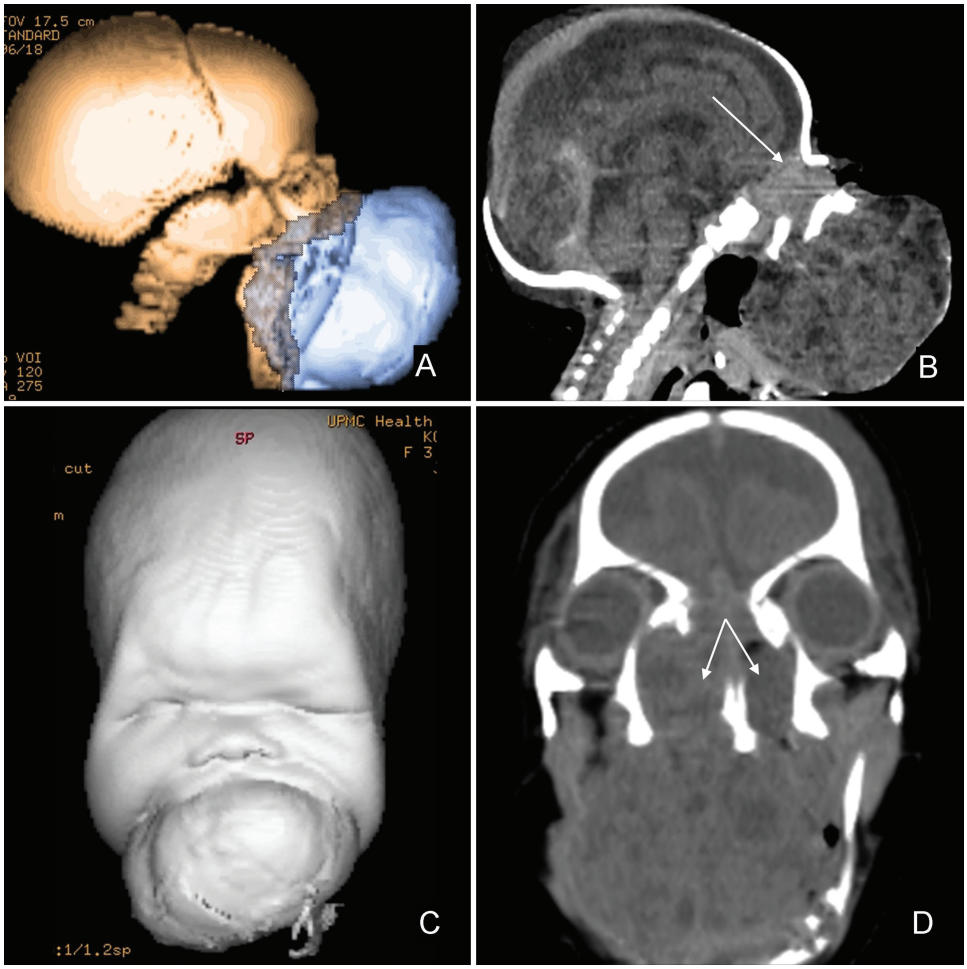

FIG. 2. Preoperative CT scans showing an epignathus teratoma measuring 6.8 × 6.5 × 6.2 cm in a neonate. A: Three-dimensional reconstructed side view showing the tumor (in *blue*) extending through the mouth. The horizontal and vertical rami of the mandible are elongated, and the mouth is opened to a greater than 90° angle. B: The tumor origin is visible in relation to the entire ventral skull base *(arrow)*. C: Three-dimensional reconstructed view of the grossly expanded nasal cavity causing severe hypertelorism secondary to the expansion of the nose. The interorbital distance was 30 mm. D: Axial view of the heterogeneous mass formed by fat and calcified tissues extending through both sides of the palate into the oral cavity *(arrows)* and out of the mouth without any intracranial extension.

the palate into the oral cavity and out of the mouth without any intracranial extension. The nasal cavity was grossly expanded, causing severe hypertelorism secondary to the expansion of the nose. The interorbital distance was 30 mm. The horizontal and vertical rami of the mandible were elongated, and the mouth was opened to a greater than 90° angle. The inferior orbital rims and lateral walls were rotated upward.

Subsequent MR imaging complemented the CT findings by identifying multiple foci of variable increased T-2 signal intensity, which showed low signal intensity on the fluid-attenuated inversion-recovery images, suggesting that much of the interior of the tumor was cystic. Magnetic resonance angiography revealed one large artery centrally located within the mass that appeared to arise from the left nasopharynx (possibly an ethmoidal artery). A fairly large external carotid branch (presumably a facial artery) was noted on the surface of the lesion on the left side (Fig. 3).

Tumor Resection. At 3-weeks postpartum (36 gestational weeks), the child was brought to the operating room. After induction of general endotracheal anesthesia through the prior tracheostomy location, the patient was prepared using sterile techniques, draped, and placed supine. With the assistance of surgical loupe magnification, the surgery was initiated by circumferential dissection of the exophytic component of the tumor transorally. The dissection was then performed using a 45° endoscope to gain access to the pedicle, while allowing for bipolar electrocautery coagulation prior to transection to minimize blood loss. This technique allowed the pedicle to be divided after adequate circumferential bipolar electrocoagulation at the level of the hard palate defect.

Once the exophytic component was exenterated, two excellent corridors were created through the bilateral defects in the hard palate, offering direct access to the residual teratoma. The endonasal dissection was initiated with the aid of a 0° endoscope, using a four-hand, two-surgeon technique to resect the origin of the teratoma from the ventral skull base through the hard palate defects in an extracapsular fashion. This technique consisted of one surgeon holding and manip-

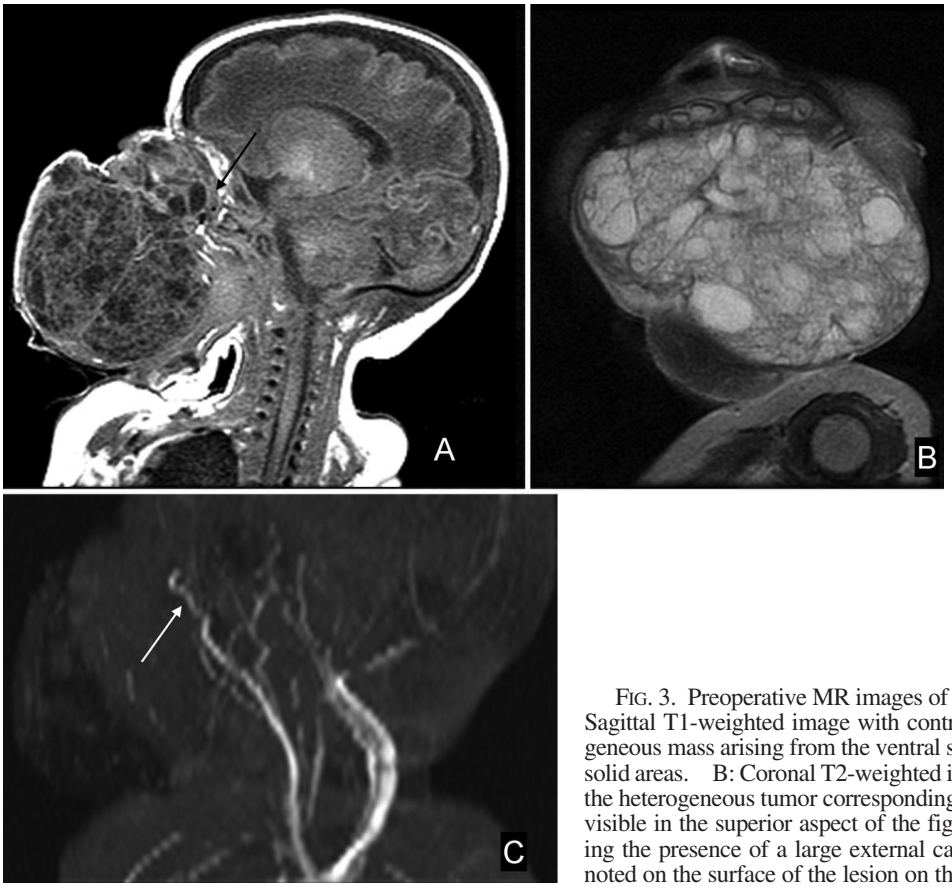

FIG. 3. Preoperative MR images of an epignathus teratoma in a neonate. A: Sagittal T1-weighted image with contrast enhancement showing a very heterogeneous mass arising from the ventral skull base *(arrow)* consisting of cystic and solid areas. B: Coronal T2-weighted image showing high signal intensity inside the heterogeneous tumor corresponding to cystic areas. The nose of the patient is visible in the superior aspect of the figure. C: An MR angiogram demonstrating the presence of a large external carotid branch (presumably a facial artery) noted on the surface of the lesion on the left side *(arrow)*.

ulating the endoscope while using another instrument in the other hand (to perform suctioning or traction using forceps, for example), while the second surgeon operated one instrument in each hand, usually suctioning with the left hand and using variable instruments with the right (such as dissectors, bipolar coagulators, and scissors).

The large prominent vessel passing through the tumor was isolated and coagulated. The tumor origin was adherent to the region of the ethmoidal cells anterosuperiorly along the anterior skull base and the cribriform plate. The tumor had extended caudally down to the region of the skull base that would have eventually formed the sphenoid sinus in the sphenoid bone. Obviously in this premature infant, none of the paranasal sinuses had been adequately formed, making identification of key anatomical landmarks very difficult. The extracapsular dissection progressed caudally until the level of what would have been the sphenoid bone. Once this dissection was performed, the anterior part of the teratoma on the right side, followed by the left side, was brought into the oral cavity. This maneuver left only the exophytic origin of both pedicles attached to the posterior midline at the level of the floor of the sphenoid sinus. Both pedicles were meticulously resected from the skull base using coagulation and drilling of the bone along the attachment to the skull base.

The lateral margins of the tumor were adherent to the carotid canals below the region that would have differentiated into the pterygoid plates, and these margins were carefully resected away. To gain access to this space, the vomer and rostrum of the sphenoid had to be drilled to allow for endoscopic dissection. After this drilling was performed, the area was examined with a 45° endoscope, revealing what appeared to be a gross-total resection. Hemostasis was achieved using bipolar electrocautery, Avitene pledgets, and Surgicel, an extremely tedious process due to the bloody nature of the tumor and the size of the patient. The dura mater was exposed but not transgressed in various locations; therefore, no special dura reconstruction was undertaken because there was no cerebrospinal fluid leakage during the procedure. The entire endonasal dissection of the tumor from the skull base and subsequent hemostasis was fully and exclusively performed under endoscopic visualization.

Mandible Correction. Following the resection, the maximal mandible opening was 60 mm and the fixed anterior open bite was 45 mm. The deformity caused by the chronic intraoral mass had affected the temporomandibular joint region as well. A preoperative CT scan demonstrated that the gonial angle measured approximately 180°. Immediately after the resection, two layers of ultrathin DuoDERM (ConvaTec) were placed on the chin and the mental region (anterior part of the mandible). An Ace bandage was then placed in a Barton-type dressing (figure-eight bandage providing support below and anterior to the lower jaw) to resuspend the mandible and begin molding it in an attempt to close the anterior open bite. The patient tolerated the procedure well; estimated blood loss was between 200 and 220 ml.

Pathological Findings and Postoperative Course. A com-

plete resection of the lesion was achieved as demonstrated on postoperative images (Fig. 4). Pathology tests proved that the lesion was a nasopharyngeal immature teratoma with well-differentiated yolk sac elements. The infant required a 1-month hospitalization during ventilator support in the NICU while the mandibular remodeling and upper airways reformed. During this period, she experienced episodes of tracheitis and pneumonia that were treated medically. At the 1-year follow-up, the child's mandibular remodeling has been completed, and from a craniofacial perspective she has recovered well. The patient has intact cranial nerves and no evidence of recurrence of the tumor (Fig. 5).

Discussion

The presence of a mass with mixed echogenicity and multiloculated cystic or solid regions on obstetric ultrasonographic images indicates a teratoma, and its location on the mouth indicates the epignathus variety.[2,24,26] An epignathus teratoma can also cause polyhydramnios due to impaired fetal swallowing that frequently leads to an inability to identify the fetal stomach.[6,9] An elevation of AFP levels can also be detected and assist in the diagnosis and follow-up of the patient.[27] In the present case, however, the levels of AFP were within the normal limits, thereby neither facilitating in the diagnosis nor helping in the monitoring of postoperative AFP levels.

A prenatal diagnosis is probably the most important step in the management of these life-threatening lesions. It is essential to allow for a multidisciplinary preparation, particularly for the moment of the delivery.[14,24,26]

These tumors have various challenging characteristics. Even when the diagnosis is accurate before delivery, there remains a high rate of mortality with these tumors.[5,10,14,28,31] A significant source of morbidity and death involving epignathus teratomas is related to maintaining a neonate's airway and providing adequate ventilation. The placenta is responsible for oxygenating the newborn's blood, and therefore after its disconnection the infant depends on his or her lungs exclusively to breathe and maintain oxygenation. An epignathus teratoma functions as a grotesque obstructive foreign body, and an infant delivery followed by inadequate protection of the airway invariably leads to death.[14,25] A tracheostomy procedure is necessary in the majority of cases to keep the infant alive. For cases such as the one described here in which the epignathus teratoma protrudes through the mouth, the possibility of securing the airway by orotracheal intubation is not feasible. Alternatives such as extracorporeal membrane oxygenation must be available at the time of the delivery to avoid the disaster of a tracheostomy failure.[19]

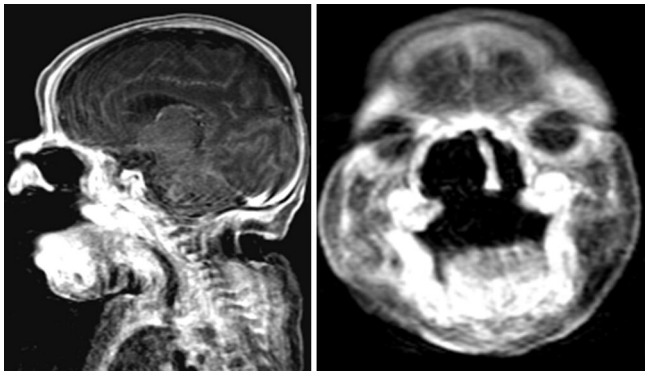

FIG. 4. Postoperative sagittal *(left)* and coronal *(right)* MR images with contrast enhancement demonstrating gross-total resection of the epignathus teratoma.

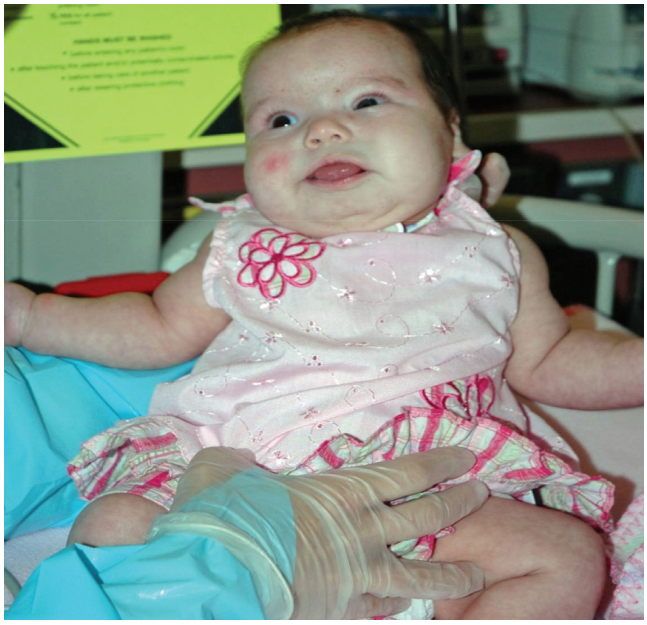

FIG. 5. Photograph of the patient at the 8-month follow-up visit demonstrating craniofacial recovery. (Photograph published with permission of the parent.)

Once the neonate is stable, the subsequent challenge is how to manage the tumor itself. The treatment of choice is resection of the tumor, and gross-total resection should always be attempted.[10,12,14,31] If residual disease is left, the recurrence rate approaches 100%, and there are no options for adjuvant therapy (such as radiation) in this setting.[5,20] Traditionally, access to the attachment of epignathus teratomas at the level of the ventral skull base has been gained through open craniofacial approaches with the aid of surgical loupes or microscopes.[12,29] Extensive incisions on the face and/or neck are conventionally performed to achieve total resection of teratomas of the skull base.[1,4] The use of endoscopes to assist with resections performed with microscopes has been described as improving visualization to secure a better and more precise dissection of skull base teratomas.[12] A purely endoscopic approach, however, obviates the need for a microscope completely and therefore does not require the use of any transfacial approaches to create a corridor to the skull base for visualization, as required when using a microscope. Instead, once the exophytic component of the tumor is removed, the corridor created within the hard palate by the tumor itself is utilized to gain access to the skull base attachments.[8,15–18] There are very few reports of teratomas resected using a purely endoscopic approach in the literature.[20,22,32] These reports identify the use of a purely endoscopic approach in relatively small teratomas. In the case of a giant lesion with significant adherence through the entire ventral skull base in a newborn infant, no one has previously been able to demonstrate the use of an endoscopic approach for complete access. In this case, there were obvious advantages of avoiding the craniofacial approach and not

disturbing the osseous framework to allow for subsequent growth and development,[13,29] and achieving a complete resection after a 12-month follow-up period is satisfying.

Conclusions

In this report, we demonstrated the feasibility of accessing the ventral skull base even in a newborn. A gestational age of 36 weeks represents the youngest age that a transpalatal endonasal approach has been used for access to the ventral skull base. A purely endoscopic transpalatal endonasal approach, such as the resection of the epignathus teratoma described in this report, is an excellent alternative to conventional transfacial/transcranial approaches for the treatment of neonatal skull base lesions. This new approach has the advantage of allowing better visualization for adequate resection of skull base lesions in the neonate while preserving craniofacial growth harmony with extremely low morbidity. Optimistically, this approach may prove to be a method that can diminish the incidence of recurrence of skull base teratomas. Yet because these are very rare lesions, a long period of time will be necessary before this question will be adequately answered.

Disclosure

Drs. Kassam, Snyderman, and Carrau are paid consultants for the Karl Storz, KLS Martin, and Stryker Corporations.

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Manuscript submitted February 20, 2007.
Accepted May 29, 2007.

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