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Approaching the Sella through the Nonpneumatized Sphenoid in Pediatric Patients

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Abstract

Objective The purpose of this study is to specifically assess pediatric patients with nonpneumatized sphenoid sinuses who have undergone transsphenoidal resections of skull base tumors and assess the complications and outcomes.

Methods Data was collected by a retrospective chart review done on children ages 7 and under who underwent endoscopic tumor resection and had a partially or completely nonpneumatized sphenoid sinus on preoperative computed tomography imaging. Surgical data collected included surgical corridor, gross total versus subtotal resection, repair method, use of septal flap, intraoperative and postoperative cerebrospinal fluid leak, and estimated blood loss.

Results Six patients were identified that fit our inclusion criteria who underwent surgery between November 2015 and April 2018 (3 males, 3 females; average age = 4.28 years). Tumor pathologies include three craniopharyngiomas, Rathke cleft cyst, meningocele, and neuroblastoma. All cases involved varying percentages of sphenoid sinus pneumatization. All cases except one craniopharyngioma and the neuroblastoma required removal of cancellous bone to access pathology. Degree of sphenoid pneumatization tended to be greater with age and resulted in less intraoperative bleeding. The two youngest patients with completely nonpneumatized sphenoid sinuses lost 61.73 and 17.52% of their total blood volume intraoperatively. Procedures were able to be adequately performed and pathology completely addressed with minimal postoperative complications and no postoperative CSF leaks.

Conclusions Intraoperative challenges including hemorrhage are likely in nonpneumatized patients, and thus surgeons should be prepared with adequate vascular access and blood products. Hemorrhage and total blood volume loss are increased in the younger patients with no sphenoid pneumatization. Despite the additional challenges, a nonpneumatized sphenoid sinus is not a contraindication for an endoscopic resection in the pediatric population.

Keywords
- transsphendoidal resection
- nonpneumatized sphenoid sinus
- skull base tumor
- endoscopic tumor resection

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Introduction

Endoscopic endonasal approach (EEA) is a surgical technique used for the resection of skull base tumors through the paranasal cavity and is performed by otolaryngologists and neurosurgeons. Before EEAs, skull base tumors were either removed through an open craniotomy or with the use of a microscope. An endoscopic approach via natural orifices allows for less tissue disruption, no external incisions, and less brain manipulation, ultimately leading to shorter hospital admissions and quicker recoveries.[1–4]

EEAs have been well established in the adult population with a small yet growing literature base on this approach in pediatrics.[2,5,6] Our group recently published a study reporting that EEAs are safe and efficacious in the very young pediatric population under 6 years old.[7] Small nasal cavities and sinuses in young children and thus smaller surgical corridors pose challenges to the EEA surgeon.

An additional, unique challenge in approaching the sella in young children is going through a nonpneumatized sphenoid. In most cases, the sphenoid does not become pneumatized until about age 7, which makes access to the skull base through a nonpneumatized sinus challenging because of bleeding and loss of visual landmarks.[6,8] To date, the only publication that illuminates these intraoperative challenges in transsphenoidal approaches through a nonpneumatized sphenoid was a report on a small, adult case series.[6] They reported that total tumor resection was achieved and postoperative complications were limited.

Brockmeyer et al suggested that skull base procedures like EEA can be safely performed in children but that surgeons must consider whether age-dependent anatomical structures are developed.[5] Some of the current literature even suggest that that EEA should be avoided, if possible, in patients with nonpneumatized sinuses so as to not disrupt normal sinus development.[9,10] However, these recommendations are theoretical, and to our knowledge no empirical evidence exists yet to verify whether EEA is more beneficial or harmful for very young patients with nonpneumatized sinuses.

This study aims to assess outcomes and complications in children without complete sphenoid pneumatization who have undergone an EEA for tumor resection. Approaching the sella through a nonpneumatized sphenoid has unique challenges.

Methods

Rady Children's Hospital Institutional Review Board approval was obtained to access patient charts for this study. A retrospective chart review was done on children aged 7 and under who received tumor resection by a sellar approach via EEA and showed lack of complete sella pneumatization on preoperative computed tomography (CT) imaging. Eight patients aged 7 and under were identified who underwent EEA via transellar approach between November 2015 and April 2018. Two of these patients had chordomas and were excluded from analysis because the surgical approach was much more extensive and outside of the scope of this study. Six patients met the inclusion criteria. None of the patients included had reported personal or family history of bleeding problems or relevant medication history that would suggest coagulation abnormalities. Patient age, gender, indication for surgery, tumor pathology, and percent pneumatization were evaluated for all patients by the same otolaryngologist from our institution. Surgical data including surgical corridor, gross total versus subtotal resection, repair method, use of septal flap, intraoperative cerebrospinal fluid (CSF) leak, and estimated blood loss (EBL) were recorded. Postoperative CSF leak, immediate or residual diabetes insipidus (DI), endocrine sequelae, optic outcomes, and other postoperative complications were recorded.

An otolaryngologist used a pre-operative, mid-sagittal computed tomography scan of each patient to estimate percent pneumatization in the region between the anterior sphenoid wall and posterior sella (►**Fig. 1**).

Total blood volume was determined by using an *Estimated Blood Volume Calculator*.[11]

Results

Six patients between November 2015 and April 2018 met the inclusion criteria. Patients were between 0.4 and 7 years of age (average = 4.28 years).

Sphenoid Characteristics

►**Table 1** includes sphenoid characteristics including percent of pneumatization preoperation. Six patients with nonpneumatized sphenoids as visualized on preoperative CT imaging underwent EEA for resection of a skull base tumor. The two youngest patients, aged 5 months and 13 months, had a completely nonpneumatized sphenoid. The patients aged 4 and 6 years had 30 and 45% sphenoid pneumatization, respectively. The two patients aged 7 had 55% sphenoid pneumatization and 70% sphenoid pneumatization.

Tumor Characteristics

Tumor pathologies include three craniopharyngiomas: Rathke cleft cyst, meningocele, and neuroblastoma. The first

Table 1 Demographics, sphenoid, and tumor characteristics

Patient	Age (years)	Weight (kg)	Percent of pneumatization	Indication for resection	Pathology	Pathology site
1	7	26.9	70%	Encephalitis, seizures	Craniopharyngioma	Sella
2	4	15.4	30%	Hydrocephalus	Craniopharyngioma	Sella/suprasella
3	1.3	10.7	0%	Optic nerve compression	Rathke cleft cyst	Sella
4	0.4	4.05	0%	Recurrent meningitis	Meningocele	Sphenoid

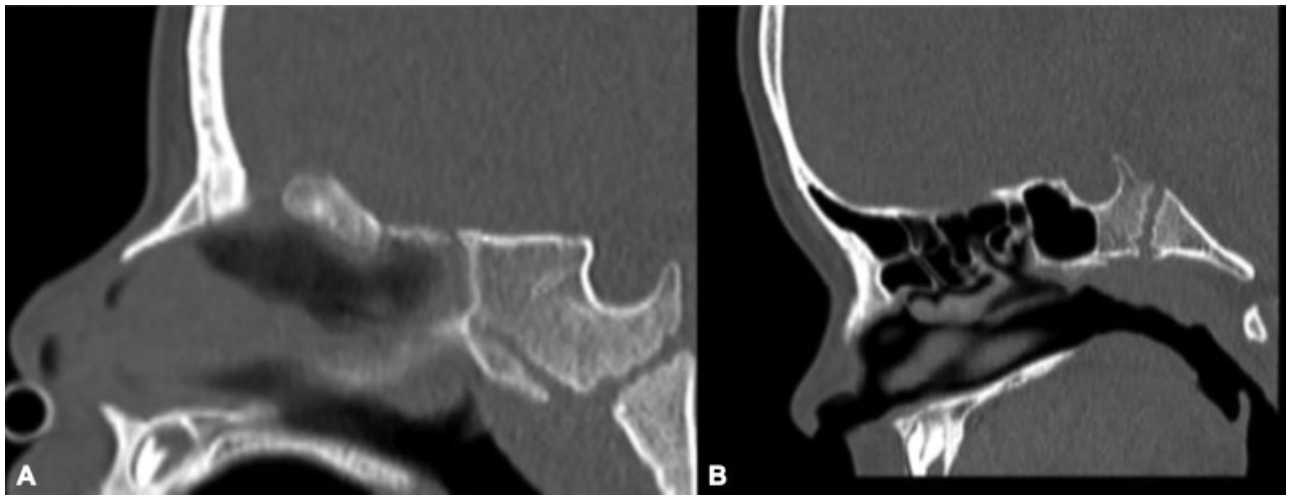

Fig. 1 Preoperative sagittal computed tomography images. (**A**) The completely nonpneumatized sphenoid sinus of a 13-month-old patient with a Rathke cleft cyst. (**B**) A partially pneumatized sphenoid sinus, ~70%, in a 7-year-old patient with a sellar craniopharyngioma.

craniopharyngioma presented with hydrocephalus and measured 8 × 9.5 × 15.5 mm on magnetic resonance imaging (MRI). The second craniopharyngioma presented with meningoencephalitis and seizures, and measured 36 × 30 × 20 mm on MRI. The third craniopharyngioma presented with headache and seizures and measured 14 × 11 × 19 mm. The Rathke cleft cyst measured 10 × 11 × 5 mm and presented with optic nerve compression. This resulted in visual deficits including color asymmetry, anisometropia, and left greater than right astigmatism. The meningocele presented with recurrent meningitis, and measured 7 mm thick. The neuroblastoma measured 47 × 25 × 33 mm.

Surgical Approach

All six cases underwent a transsphenoidal approach for resection. Preoperative embolization would not have been helpful in any of these cases because it was considered possible or safe. All patients except one of the craniopharyngiomas (Patient 6) and the neuroblastoma (Patient 5) required additional curetting and drilling through cancellous bone to access the pathology (►**Fig. 2**). A binaural approach was utilized in all cases. The inferior third of the middle turbinate was trimmed to provide access to the sphenoid. In the two cases of a completely nonpneumatized sphenoid, cortical bone was encountered at the location of the anterior sphenoid wall. This cortical bone was opened widely using a Kerrison bone punch. Once this was removed, the sphenoid was noted to be full of soft cancellous bone. This was removed with a high-speed drill and curette. In most cases, the drill was not needed, as the bone could be removed easily with gentle curettage. The curette was noted to be safer as it could provide tactical feedback when palpating the cortical bone of the skull base, laminae

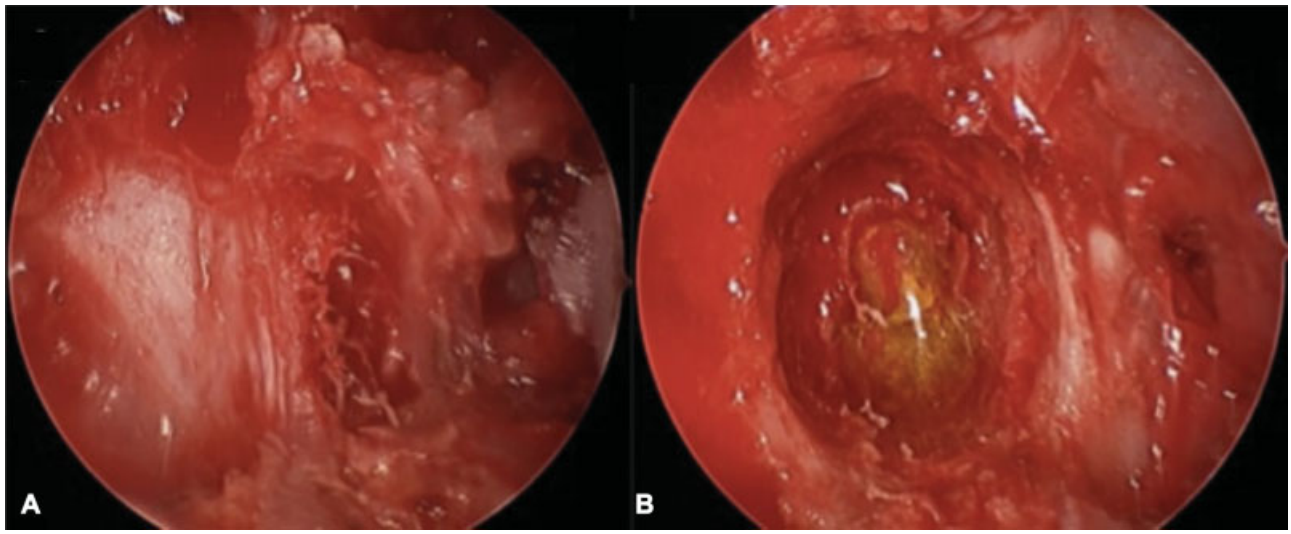

Fig. 2 Intraoperative endoscope images. (**A**) The anterior sphenoid wall with cancellous bone in the center at the site of the future sphenoid ostia, and the cortical bone of the anterior sphenoid wall to the left. (**B**) The brownish Rathke cleft cyst seen at region of sella turcica identifying posterior extent of sphenoid removal.

papyracea, and posterior sphenoid wall including the sella turcica. The cancellous bone of the non-pneumatized sinus was very vascular and hemorrhaged during removal, sometimes causing issues with visualization. Bleeding had to be controlled using Surgiflo and compression with pledgets, and in some cases bone wax. The cancellous bone was completely removed until surgical landmarks of the planum sphenoidale, lamina papyracea, and posterior sphenoid wall were identified. In each case, image guidance was used to verify the locations of the internal carotids, opticocarotid recess, and sella turcica, at which point the sella was safely entered.

All cases were repaired with an acellular dermal matrix (AlloDerm, LifeCell Corp, Woodlands, Texas) in an underlay fashion, and in two of the cases a fat graft was placed into the sellar defect. In patient 2, a nasoseptal flap was used for repair because of a high flow intraoperative CSF leak. No lumbar drains were used. Complete gross total resection was obtained in the craniopharyngioma cases. The Rathke cleft cyst was fenestrated with all debris removed. The meningocele was resected and the skull base was repaired.

Intraoperative Complications

►**Table 2** includes intraoperative and postoperative patient complication data. All patients had significant EBL intraoperatively, and three of the cases required an intraoperative blood transfusion (EBL range = 50–200 mL). The craniopharyngioma cases, patients 1, 2, and 3, lost 3.49% and 4.06% and 4.29% of total blood volume, respectively. The Rathke cleft cyst lost 17.52% of total blood volume, the meningocele lost 61.73% of total blood volume, and the neuroblastoma lost 1.5% of total blood volume. Three patients (50%) required an intraoperative blood transfusion. The Rathke cleft cyst required 150 mL of packed red blood cell (pRBC) and the meningocele required 160 mL of pRBC transfused intraoperatively. The neuroblastoma required 300 mL of pRBC. All patients were infused with Plasma Lyte A intraoperatively, which is an isotonic volume expander used in cases with substantial blood loss.

Postoperative Complications and Outcomes

Despite all patients having significant intraoperative bleeding, none had postoperative bleeding complications or a postoperative CSF leak. Patient 3 had preoperative opthalmic changes that persisted in the postopertaive period. These included color asymmetry, anisometropia, and L > R astigmatism. Patient 4 experienced low-grade fever, congestion, and diarrhea within 1-week post-operation which quickly resolved and was thought to be viral in origin. Patient 4 experienced left eye swelling 1-week post-operation but did not have any visual disturbances. Patient 2 had a large craniopharyngioma with extension into the 4th ventricle, and suffered from thyroid stimulating hormone and antidiuretic hormone deficiencies, while the other three patients did not have any postoperative pituitary dysfunction. Three patients had transient postoperative DI, and one had residual DI. Patients followed up with their primary surgeon from 0.33 to 2.33 years. The patient with the neuroblastoma had worsened metastatic disease in multiple sites that did not respond to chemotherapy and ultimately expired 7 months after EEA surgery.

Table 2 Intraoperative and postoperative patient complication

Patient	1	2	3	4
Pathology	Craniopharyngioma	Craniopharyngioma	Rathke cleft cyst	Meningocele
Follow-up (years)	0.33	1.5	0.75	
Plasma Lyte A (mL)	600 mL	800 mL	500 mL	50 mL
Intraoperative blood transfusion	No	No	150 mL pRBC	160 cc pRBC
Estimated blood loss (mL)	75	50	150	200
Percentage of total blood loss	3.49%	4.06%	17.52%	61.73%
Other intraoperative complication	No	No	No	Hypoglycemic but resolved with fluids
Postoperative CSF leak	No	No	No	No
Intraoperative CSF leak	No	Yes, high flow	No	No
Residual DI	No	Yes	No	No
Immediate DI	Yes	Yes	Yes	No
Endocrine sequelae	No	TSH, ADH deficiencies	No	No
Optic outcomes	No vision disturbances	No vision disturbances	Persistent: Color asymmetry, anisometropia, astigmatism (left > right)	1 week postoperative L eye swelling

Abbreviations: ADH, antidiuretic hormone; CSF, cerebrospinal fluid; DI, diabetes insipidus; TSH, thyroid-stimulating hormone; pRBC, packed red blood cell.

Discussion

Endoscopic resections have become more popular in the last decade, as the improved technology has made this type of approach feasible and safe.[2,4] Most of these surgeries are done in the adult population and less often in the pediatric population. Because of the novelty and recent adoption of this type of procedure, there is still a lot unknown about its effect on the developing pediatric anatomy.

Variation in pediatric anatomical development poses a significant challenge to surgeons performing EEA.[8,10] The sphenoid is estimated to become pneumatized from age 3 to age 10.[5,12] Thus, very young pediatric patients without sphenoid sinus pneumatization offer additional challenges to the EEA surgeon, as important visual landmarks may be hidden behind the layer of cancellous bone.[8] The surgeon must also take the additional step to dissect through cancellous bone to expose and penetrate the posterior sphenoid wall.[7]

Many studies have described the complex anatomy of the sphenoid and skull base and its role in EEA in detail.[10,13–15] However, few have offered empirical evidence to clarify whether sphenoid nonpneumatization should be an exclusion criterion for EEA candidates. Vaezi et al suggested that sinus pneumatization affects surgical window, which in turn may put passing neurovascular structures at risk during skull base surgeries.[4] In a study done by Kikuchi et al, sphenoid pneumatization was wider in male than female patients, suggesting that sphenoid pneumatization may be gender dependent.[16] Song et al investigated the challenges involved in EEA of nonpneumatized adults undergoing EEA, citing difficulty visualizing anatomical landmarks and a small surgical corridor.[6] No study, however, has yet to investigate actual clinical outcomes and intra- and postoperative risks in nonpneumatized pediatric patients undergoing EEA.

Our study looked at six young EEA patients that had absent or incomplete sphenoid pneumatization. In congruence with the literature, sphenoid pneumatization in our patients increased with age (see ►**Table 1**). Among our patients, all had successful surgeries and were able to adequately address the pathology. No patients had a postoperative CSF leak. None of our patients had personal or family history or medication history that would suggest coagulation problems, yet all patients had intraoperative blood loss (range = 75–200 mL) and three required intraoperative blood transfusions. The reason for bleeding was not because of any disorder, but because of the nature of going through the cancellous bone in the nonpneumatized sphenoid. Only two patients presented with postoperative sequelae: the Rathke cleft cyst presented with ophthalmic complications and one of the craniopharyngioma cases presented with pituitary dysfunction. In the case of the RCC, the ophthalmic outcomes were not a postoperative complication; it was present prior to surgery from the cyst pushing against the optic chiasm, and was still present postoperatively. In all patients, the tumor was completely resected despite anatomical challenges and nonpneumatization of the sphenoid sinus, and few intra- and postoperative complications were present. While it is thus reasonable to infer that EEA is a generally safe and effective procedure, nonpneumatized patients offer unique intraoperative challenges such as bleeding that the EEA surgeon must be prepared for.

This study is limited due to its sample size of only six patients, thus preventing us from forming any definitive conclusions on efficacy and outcomes of EEA for different tumor types in nonpneumatized patients. Future studies should also investigate long-term follow-up data to assess future complications that may not be captured in the 1 to 2 year follow-up period. Assessing outcomes in a longer follow-up period and comparing EEA patients to controls would provide more information on whether EEA procedure affects long term sinus and facial growth.

Conclusions

Despite the anatomical challenges and the developmental concerns mentioned in the literature, this case series demonstrates that nonpneumatized pediatric EEA cases may still benefit from transellar approaches for skull base tumor resection. The safest surgical approach includes careful dissection through the nonpneumatized sphenoid and use of intraoperative image guidance to expose the important landmarks. However, this series also demonstrates that intraoperative challenges including hemorrhage are likely in nonpneumatized patients, particularly in the younger patients with no pneumatization. Because of the difficulty in controlling hemorrhage, surgeons should be ready with adequate vascular access and blood products. Despite the additional challenges that surgeons must be aware of and prepared for, a nonpneumatized sphenoid sinus is not a contraindication for an EEA in the pediatric population as the surgery can be performed safely and effectively.

Note
This manuscript has been accepted for podium presentation at the 2018 North American Skull Base Society Conference in Orlando.

Financial Disclosures
None of the authors has a financial interest in any of the products, devices, or drugs mentioned in this manuscript.

Conflict of Interest
None declared.

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