Endoscopic nonembolized resection of an extensive sinonasal cavernous hemangioma: A case report and literature review

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ABSTRACT

Sinonasal hemangiomas, although rare, must be considered in the evaluation of intranasal masses with profuse epistaxis. Although the availability of literature discussing cavernous hemangiomas in this location is limited, there have been no case reports of exclusively soft tissue sinonasal cavernous hemangiomas extending to the anterior skull base (ASB) that were resected purely endoscopically. Here, we describe the successful endoscopic resection of an extensive right sinonasal cavernous hemangioma extending to but not invading the ASB. Although highly vascular, in select cases, these tumors can be successfully resected endoscopically without embolization by experienced endoscopic sinus and skull base surgeons.

(Allergy Rhinol 4:e179–e183, 2013; doi: 10.2500/ar.2013.4.0068)

Hemangiomas are benign vascular tumors that most commonly arise early in life. More than 75% are evident at birth, but most involute spontaneously before adulthood.[1] Hemangiomas are the most common benign tumors of the head and neck, but typically are located in areas outside of the sinonasal cavity, including the lateral skull base, parotid gland, tongue, larynx, or cutaneous surfaces.[2–4] Hemangiomas of the nose and paranasal sinuses are exceedingly rare, and, in particular, cavernous hemangiomas are among the least commonly found. Indeed, to the best of our knowledge, <40 cases of sinonasal cavernous hemangiomas have been reported in adulthood since 1959.[5–17] Typically, cavernous hemangiomas are associated with the lateral wall of the nasal cavity or with the inferior turbinate.[5,15,18] To date, there are no publications of a soft tissue sinonasal cavernous hemangioma that extends up to the anterior skull base (ASB).

Instances of purely endoscopic resection of small sinonasal cavernous hemangiomas have been previously described.[5–7,10,18,19] However, none have involved or extended to the ASB or were resected without preoperative embolization. Here, we describe the management of a 66-year-old man with a large sinonasal cavernous hemangioma extending to the ASB managed purely endoscopically without preoperative embolization. The protocol for this study was reviewed and approved by the Institutional Review Board of Rutgers New Jersey Medical School, Newark, NJ.

CASE REPORT

A 66-year-old man was referred to our tertiary care center with profuse epistaxis and a large right-sided sinonasal mass. Head and neck examination revealed a severely deviated nasal septum ipsilaterally. Nasal endoscopy (after removal of nasal packing) showed marked epistaxis. The patient was repacked uneventfully with Merocel (Medtronic Xomed, Jacksonville, FL) nasal packing. Computed tomography (CT) scan showed a 3.7 × 2.3-cm soft tissue mass deforming the medial wall of the right maxillary sinus (Fig. 1). Magnetic resonance imaging (MRI) revealed extension of the lesion to the ASB with signs of increased vascularity with significant contrast enhancement (Fig. 2). CT and MRI studies also revealed postobstructive right frontal and maxillary sinusitis (Figs. 1 and 2).

Endoscopic endonasal resection of the lesion was performed without complications after performing a septoplasty for adequate access (Fig. 2 *E*). Histopathological analysis was consistent with cavernous hemangioma (Fig. 3). The patient's postoperative course was unremarkable. Postoperative imaging at 3 months revealed no residual disease (Fig. 1, *C* and *D*). At 9 months follow-up, the patient had no evidence of recurrence on nasal endoscopy (Fig. 2 *F*). He remained disease free at 2 years.
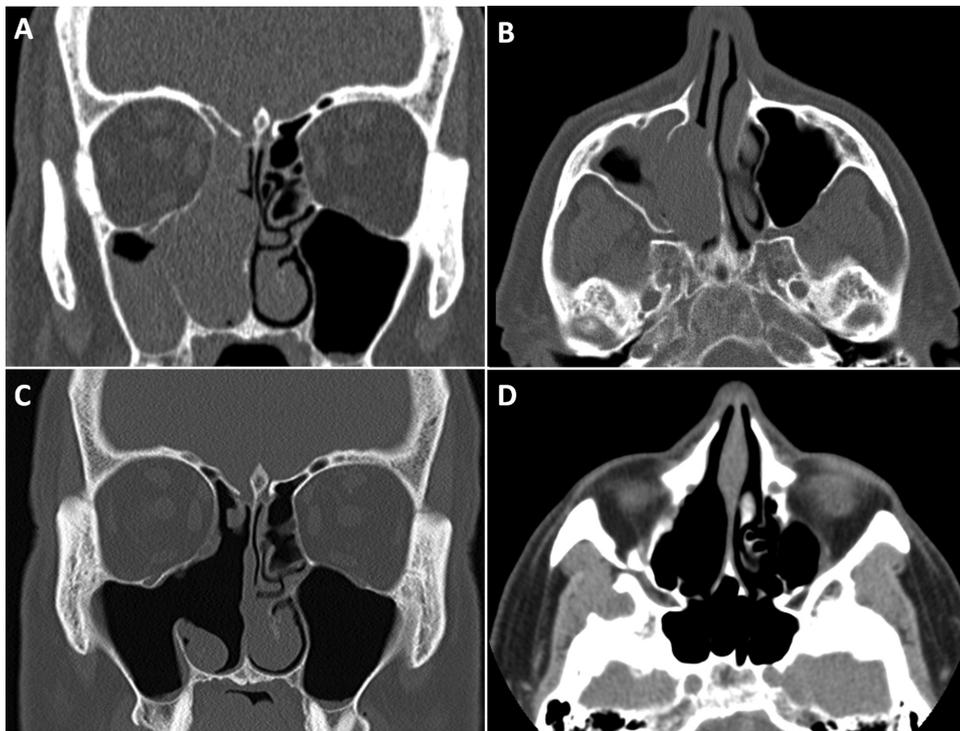
Figure 1. Preoperative (A) coronal and (B) axial computed tomography (CT) scans depicting a right sinonasal mass with extension up to the anterior skull base (ASB); postoperative (C) coronal and (D) axial CT scans at 3 months showing complete tumor resection without residual disease.

DISCUSSION

Hemangiomas are benign vascular tumors most commonly found in skin and mucosa. They are composed of a proliferation of blood vessels lined by endothelial cells.[2,3] Hemangiomas are frequently confused with vascular malformations but are differentiated based on cell behavior, history, and histology.[20,21] Typically, hemangiomas are small or absent at birth, but undergo a rapid proliferative phase characterized by increased mitotic activity during infancy. Commonly, they will involute and leave signs of fibrosis in the process.[21] However, they may appear stable for some time and proliferate with age.[1] In contrast, vascular malformations are present at birth, grow without regression, and exhibit a normal rate of cell turnover without a proliferative phase.[21] Hemangiomas have a stronger predilection for female subjects, with a 5–6:1 female-to-male ratio, whereas vascular malformations have no predilection for gender or race.[1,7,21]

The exact origin of hemangiomas remains a topic of controversy. Some have proposed that embryonic unipotent cells are involved, and others suggest that blood vessels elongate by some unknown mechanism.[2] It has also been cited that hemangiomas are benign neoplasms, similar to hamartomas.[15] Ultimately, however, hemangiomas represent endothelial proliferations.[21]

Hemangioma classification corresponds to the vessels contained within: capillary, cavernous, or mixed.[1,3] Capillary hemangiomas are the most common and are more frequently seen among children, typically in the skin or oral mucosa. They usually will involute completely within 6–12 years without intervention; therefore, treatment is not recommended.[1,2] Cavernous hemangiomas are even rarer congenital malformations, which can manifest in adulthood as deeper lesions that usually do not regress.[7,15,21]

The most common localized benign vascular tumor in the head and neck, hemangiomas are very rarely found in the sinonasal soft tissue.[15,22] In the largest study to date, reviewing biopsy materials of 51 cases of vascular growths of the nose between 1948 and 1958, only two cavernous hemangiomas were identified.[15] Since then, only sporadic cases have been reported in adults, totaling <40 sinonasal cavernous hemangiomas.[6–14,17,23–25] Within the sinonasal region, the most commonly reported sites include the nasal septum, nasal vestibule, inferior nasal turbinates, and maxillary sinus.[15,18] Capillary hemangiomas typically present at the nasal septum, whereas cavernous hemangiomas more often occur at the lateral nasal wall.[5,8,15]

Hemangiomas of the sinonasal cavity most commonly present as epistaxis, nasal obstruction, and a sinonasal mass.[18] A cavernous hemangioma is more dilated than a capillary hemangioma and is likely to develop to a massive size, as seen in our patient. Usually, it is a soft, easily compressible mass with a reddish–purple appearance, as was seen in this current case. However, if it extends deeper into subcutaneous tissue this discoloration may no longer be evident.[2]
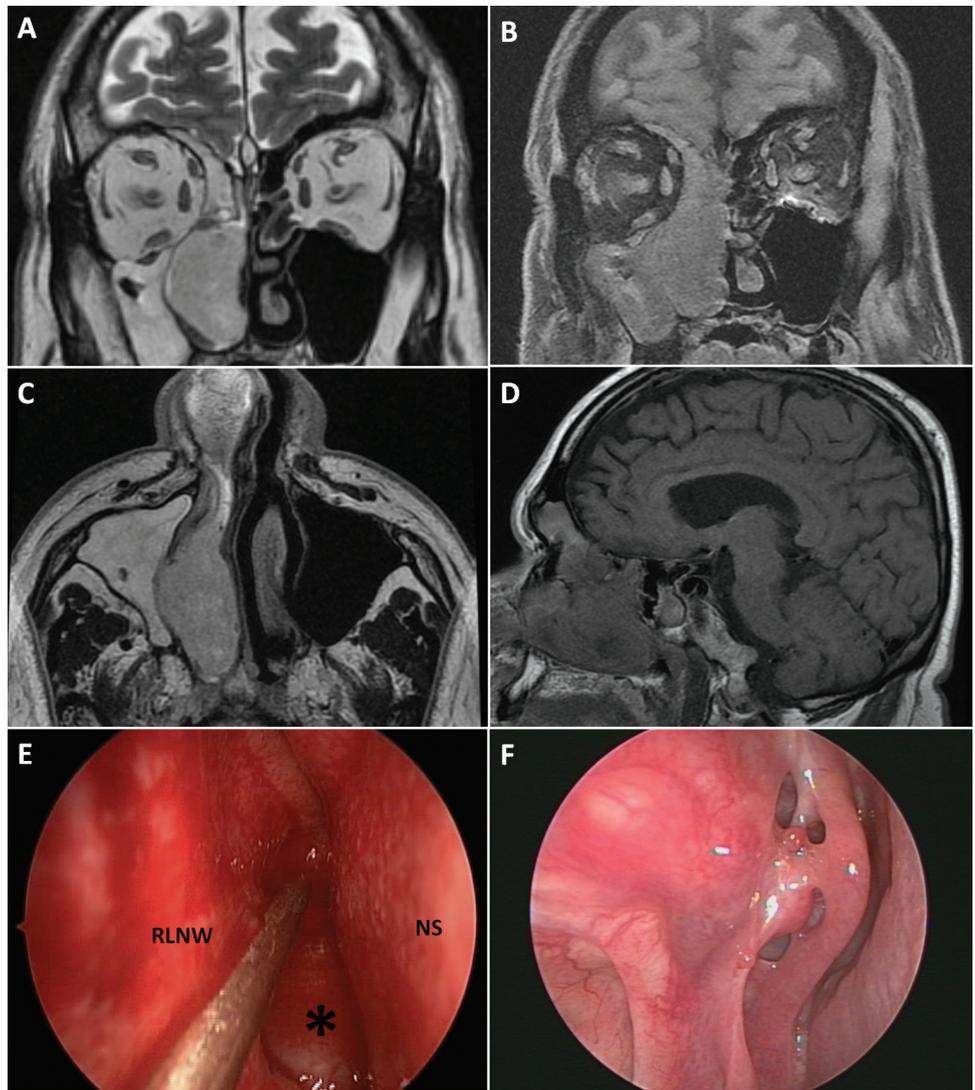
Figure 2. Preoperative T2-weighted (A) coronal and (C) axial and gadolinium (B) enhanced coronal and (D) sagittal magnetic resonance images (MRIs) show a large right sinonasal mass with significant enhancement. (E) Intraoperative depiction of the tumor after an endoscopic septoplasty. (F) Postoperative 30° rigid nasal endoscope at 9 months follow-up indicating no evidence of disease.

Because the differential diagnosis for recurrent epistaxis includes malignancy and with it, the potential for metastasis, it is important to correctly determine the nature of any nasal mass. The differential for benign causes includes inverted papilloma, arteriovenous fistula, lymphangioma, glomangioma, angiofibroma,[26–28] and inflammatory tumor, such as granuloma pyogenicum or granuloma gravidarum. Some possible malignant neoplasms that might present similar to our patient's include angiosarcoma, hemangiopericytoma,[29,30] and paraganglioma.[1]

Fortunately, hemangiomas can be identified based on characteristic histological and radiological findings. In the proliferative phase, multiple laminas of basement membrane underlying the endothelium along with an active rough endoplasmic reticulum can be visualized. After involution, fatty deposit islands among fibrous tissue may be evident. If the hemangioma underwent incomplete regression, foci of endothelial proliferation may be seen concomitantly with areas of fibrofatty infiltration.[21]

Cavernous hemangiomas are well visualized by MRI and appear as extensively dilated vessels as well as discrete areas of hyperintensity.[25] Diagnosis of nasal mucosal hemangioma should be suspected in patients presenting with profuse epistaxis with a contrast enhancing nasal mass with or without bone remodeling on imaging.[31] Malignant growths are more likely to infiltrate adjacent tissues and show bone erosion and destruction. However, there have been exceptional cases of sinonasal cavernous hemangiomas that have eroded bone.[11]

Treatment in this rare tumor has yet to be clearly elucidated. In the past, the treatment of choice for this highly vascular tumor was complete excision using an open approach in the form of craniofacial resection, with preoperative embolization.[16,32] Because removal of vascular tumors can be challenging, if an enhancing mass appears on CT and MRI concurrently with a history of recurrent epistaxis, arteriography should be considered.[31] Preoperative embolization can minimize intraoperative blood loss.[32] Furthermore, the decreased
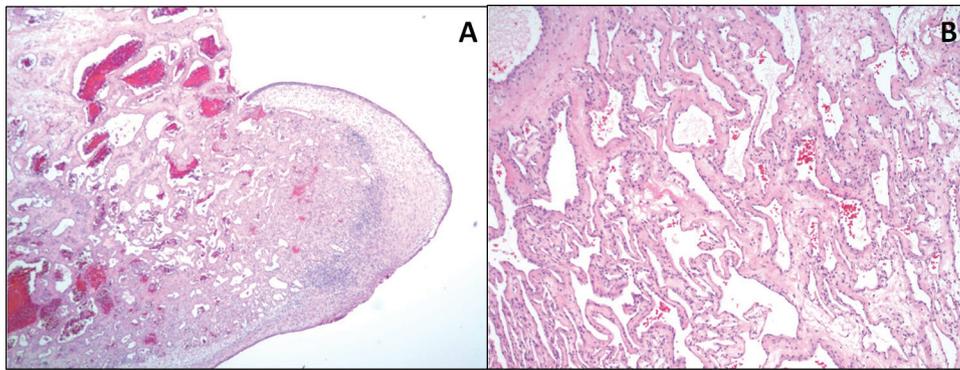
Figure 3. *Histopathological analysis of specimen using hematoxylin and eosin (H&E) stain at (A) low power and (B) high power showing dilated vessels.*

blood supply to the tumor would facilitate its removal. Nonetheless, embolization has been associated with significant morbidity (*e.g.*, neurovascular and integumentary system injury) and mortality and may not be necessary in all cases.

With the growing experience and evidence of positive outcomes associated with endoscopic surgery, its use as the sole treatment modality for both benign and malignant tumors of the sinonasal and ASB is rising.[10,33–36] Discussions of the endoscopically assisted craniofacial resection to remove a sinonasal growth involving the ASB began in the 1990s.[37,38] By 2002, Iwata *et al.*[8] concluded the safety and efficacy of using transnasal endoscopic surgery in epithelioid hemangiomas. The first case in the literature of successful endoscopic removal of a sinonasal cavernous hemangioma was in 2003.[24]

Two cases of successful endoscopic removal of sinonasal cavernous hemangiomas have been described. In 2008, Bakhos *et al.*[6] made the recommendation to perform selective embolization before endonasal endoscopic resection of a cavernous hemangioma in the nasal cavity. Later in the same year, Caylakli *et al.*[7] performed endoscopic resection of a cystic mass found exclusively in the middle turbinate. Although these cases were successful in resection of tumors within the nasal cavity, tumors that involve the ASB have been associated with increased morbidity and mortality.[39,40] Issues that increase the risk of surgery in this area include cerebrospinal leakage, infectious complications, and neurovascular injury.[39,41] Although evidence of the safety of the technique is rapidly increasing along with its popularity, use of the endoscope in this area in the past had been cautious to avoid these complications.

In our patient, the endoscopic endonasal technique was used successfully to resect this massive sinonasal cavernous hemangioma without preoperative embolization and limited intraoperative blood loss. This technique proved to be a direct, fast approach that provided the necessary exposure needed for adequate visualization and complete removal of the tumor. This technique was chosen because this tumor did not show signs of intracranial involvement on imaging and did not expand laterally. Additionally, given the lack of pterygopalatine fossa expansion, we felt that if this tumor was an angiofibroma (although unlikely in this 66-year-old man), adequate resection could still be safely achieved without embolization. In our institution, small-to-medium size vascular lesions such as nasopharyngeal angiofibromas without significant lateral expansion are typically resected without embolization, whereas larger vascular lesions are removed using a multiangle approach after embolization.[42] Although we achieved our desired goal in this patient, familiarity with endoscopic technique and ability to obtain hemostasis are paramount in nonembolized resection of sinonasal vascular lesions.

CONCLUSIONS

Sinonasal cavernous hemangiomas are extremely rare tumors, and their optimal management has yet to be defined. Although preoperative embolization has been advocated for these highly vascular lesions, in select cases with sufficient endoscopic experience, endonasal resection may be considered without embolization.

REFERENCES

1. Duvvuri U, Carrau RL, and Kassam AB. Vascular tumors of the head and neck. In Head and Neck Surgery—Otolaryngology. Bailey BJ, Johnson JT, Newlandds SD (Eds). Philadelphia, PA: Lippincott Williams & Wilkins, 1811–1825, 2006.
2. Das Gupta TK, and Brasfield RD. Blood and lymph vessel tumors. CA Cancer J Clin 22:176–183, 1972.
3. Shpitzer T, Noyek AM, Witterick I, et al. Noncutaneous cavernous hemangiomas of the head and neck. Am J Otolaryngol 18:367–374, 1997.
4. Eivazi B, Ardelean M, Baumler W, et al. Update on hemangiomas and vascular malformations of the head and neck. Eur Arch Otorhinolaryngol 266:187–197, 2009.
5. Archontaki M, Stamou AK, Hajiioannou JK, et al. Cavernous haemangioma of the left nasal cavity. Acta Otorhinolaryngol Ital 28:309–311, 2008.
6. Bakhos D, Lescanne E, Legeais M, et al. Cavernous hemangioma of the nasal cavity. Ann Otolaryngol Chir Cervicofac 125: 94–97, 2008.

7. Caylakli F, Caĝici AC, Hurcan C, et al. Cavernous hemangioma of the middle turbinate: a case report. Ear Nose Throat J 87:391–393, 2008.

8. Iwata N, Hattori K, Nakagawa T, and Tsujimura T. Hemangioma of the nasal cavity: A clinicopathologic study. Auris Nasus Larynx 29:335–339, 2002.

9. Jammal H, Barakat F, and Hadi U. Maxillary sinus cavernous hemangioma: A rare entity. Acta Otolaryngol 124:331–333, 2004.

10. Jungheim M, and Chilla R. The monthly interesting case—Case no. 64. Cavernous hemangioma. Laryngorhinootologie 83:665–668, 2004.

11. Kim HJ, Kim JH, and Hwang EG. Bone erosion caused by sinonasal cavernous hemangioma: CT findings in two patients. AJNR Am J Neuroradiol 16:1176–1178, 1995.

12. Kitzmann AS, Moore EJ, Salomão DR, and Woog JJ. Cavernous hemangioma involving the lacrimal outflow system. Ophthal Plast Reconstr Surg 23:488–490, 2007.

13. Palacios E, and Daroca PJ Jr. Nasal cavernous hemangioma. Ear Nose Throat J 86:326–328, 2007.

14. Vargas MC, and Castillo M. Sinonasal cavernous haemangioma: A case report. Dentomaxillofac Radiol 41:340–341, 2012.

15. Osborn DA. Haemangiomas of the nose. J Laryngol Otol 73:174–179, 1959.

16. Loh LE, Ho KH, Thoo A, and Kwan C. Hemangioma of the ethmoidal sinuses. Singapore Med J 35:211–214, 1994.

17. Sarkar S, Roychoudhury A, and Roychaudhuri BK. Intractable anemia: A case of bleeding nasal cavernous hemangioma. Indian J Otolaryngol Head Neck Surg 63:23–24, 2011.

18. Song CE, Cho JH, Kim SY, et al. Endoscopic resection of haemangiomas in the sinonasal cavity. J Laryngol Otol 123:868–872, 2009.

19. Pasquini E, Sciarretta V, Frank G, et al. Endoscopic treatment of benign tumors of the nose and paranasal sinuses. Otolaryngol Head Neck Surg 131:180–186, 2004.

20. Donnelly LF, Adams DM, and Bisset GS III. Vascular malformations and hemangiomas: A practical approach in a multidisciplinary clinic. AJR Am J Roentgenol 174:597–608, 2000.

21. Mulliken JB, and Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. Plast Reconstr Surg 69:412–422, 1982.

22. Sheppard LM, and Mickelson SA. Hemangiomas of the nasal septum and paranasal sinuses. Henry Ford Hosp Med J 38:25–27, 1990.

23. Apfelberg DB. Argon and YAG laser photocoagulation and excision of hemangiomas and vascular malformations of the nose. West J Med 163:122–127, 1995.

24. Kelley TF. Endoscopic management of an intranasal hemangioma: A case report and literature review. Otolaryngol Head Neck Surg 128:595–597, 2003.

25. Itoh K, Nishimura K, Togashi K, et al. MR imaging of cavernous hemangioma of the face and neck. J Comput Assist Tomogr 10:831–835, 1986.

26. Ruiz JW, Saint-Victor S, Tessema B, et al. Coblation assisted endoscopic juvenile nasopharyngeal angiofibroma resection. Int J Pediatr Otorhinolaryngol 76:439–442, 2012.

27. Boghani Z, Husain Q, Kanumuri VV, et al. Juvenile nasopharyngeal angiofibroma: A Systematic review and comparison of endoscopic, endoscopic-assisted, and open resection in 1047 cases. Laryngoscope 123:859–869, 2013.

28. Vazquez A, Shukla PA, Choudhry OJ, et al. Geometric alopecia after preoperative angioembolization of juvenile nasopharyngeal angiofibroma. Allergy Rhinol (Providence) 4:e21–e24, 2013.

29. Tessema B, Eloy JA, Folbe AJ, et al. Endoscopic management of sinonasal hemangiopericytoma. Otolaryngol Head Neck Surg 146:483–486, 2012.

30. Dahodwala MQ, Husain Q, Kanumuri VV, et al. Management of sinonasal hemangiopericytomas: A systematic review. Int Forum Allergy Rhinol 3:581–587, 2013.

31. Dillon WP, Som PM, and Rosenau W. Hemangioma of the nasal vault: MR and CT features. Radiology 180:761–765, 1991.

32. Biller HF, Krespi YP, and Som PM. Combined therapy for vascular lesions of the head and neck with intra-arterial embolization and surgical excision. Otolaryngol Head Neck Surg 90:37–47, 1982.

33. Eloy JA, Vivero RJ, Hoang K, et al. Comparison of transnasal endoscopic and open craniofacial resection for malignant tumors of the anterior skull base. Laryngoscope 119:834–840, 2009.

34. Higgins TS, Thorp B, Rawlings BA, and Han JK. Outcome results of endoscopic vs craniofacial resection of sinonasal malignancies: A systematic review and pooled-data analysis. Int Forum Allergy Rhinol 1:255–261, 2011.

35. Ong YK, Solares CA, Carrau RL, and Snyderman CH. New developments in transnasal endoscopic surgery for malignancies of the sinonasal tract and adjacent skull base. Curr Opin Otolaryngol Head Neck Surg 18:107–113, 2010.

36. Wood JW, Eloy JA, Vivero RJ, et al. Efficacy of transnasal endoscopic resection for malignant anterior skull-base tumors. Int Forum Allergy Rhinol 2:487–495, 2012.

37. Thaler ER, Kotapka M, Lanza DC, and Kennedy DW. Endoscopically assisted anterior cranial skull base resection of sinonasal tumors. Am J Rhinol 13:303–310, 1999.

38. Yuen AP, Fung CF, and Hung KN. Endoscopic cranionasal resection of anterior skull base tumor. Am J Otolaryngol 18:431–433, 1997.

39. Batra PS, Citardi MJ, Worley S, et al. Resection of anterior skull base tumors: Comparison of combined traditional and endoscopic techniques. Am J Rhinol 19:521–528, 2005.

40. de Almeida JR, Witterick IJ, Gullane PJ, et al. Physical morbidity by surgical approach and tumor location in skull base surgery. Head Neck 35:493–499, 2013.

41. Ransom ER, and Chiu AG. Prevention and management of complications in intracranial endoscopic skull base surgery. Otolaryngol Clin North Am 43:875–895, 2010.

42. Eloy JA, Murray KP, Friedel ME, et al. Graduated endoscopic multiangle approach for access to the infratemporal fossa: a cadaveric study with clinical correlates. Otolaryngol Head Neck Surg 147:369–378, 2012. □