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.

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 imagining 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.
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. Hemangiomas are frequently confused with vascular malformations but are differentiated based on cell behavior, history, and histology. 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. However, they may appear stable for some time and proliferate with age. In contrast, vascular malformations are present at birth, grow without regression, and exhibit a normal rate of cell turnover without a proliferative phase. 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.

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. It has also been cited that hemangiomas are benign neoplasms, similar to hamartomas. Ultimately, however, hemangiomas represent endothelial proliferations.

Hemangioma classification corresponds to the vessels contained within: capillary, cavernous, or mixed. 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. Cavernous hemangiomas are even rarer congenital malformations, which can manifest in adulthood as deeper lesions that usually do not regress.

The most common localized benign vascular tumor in the head and neck, hemangiomas are very rarely found in the sinonasal soft tissue. 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. Since then, only sporadic cases have been reported in adults, totaling <40 sinonasal cavernous hemangiomas. Within the sinonasal region, the most commonly reported sites include the nasal septum, nasal vestibule, inferior nasal turbinates, and maxillary sinus. Capillary hemangiomas typically present at the nasal septum, whereas cavernous hemangiomas more often occur at the lateral nasal wall.

Hemangiomas of the sinonasal cavity most commonly present as epistaxis, nasal obstruction, and a sinonasal mass. 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 red-purple appearance, as was seen in this current case. However, if it extends deeper into subcutaneous tissue this discoloration may no longer be evident.
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, 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, and paraganglioma.

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.

Cavernous hemangiomas are well visualized by MRI and appear as extensively dilated vessels as well as discrete areas of hyperintensity. 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. 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.

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. 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. Preoperative embolization can minimize intraoperative blood loss. Furthermore, the decreased
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, Caylalaki 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.

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