

# Removal of an Intraosseous Venous Malformation of the Maxilla via a Transoral-transconjunctival-Transcaruncular Approach

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**Summary:** Intraosseous vascular anomalies represent less than 1% of all bony tumors. When they involve the maxillofacial skeleton, these masses may cause substantial morbidity and aesthetic concern. Herein, we highlight a case of a maxillary intraosseous venous malformation manifesting as functional lacrimal duct obstruction with epiphora, sinus pressure, and increasing medial canthal/maxillary fullness over 2 years in a young, healthy woman. Surgical excision of the lesion was accomplished without a skin incision by coupling a transoral, midface degloving technique with a transconjunctival–transcaruncular approach. This minimally invasive technique led to complete resolution of symptoms and a satisfactory cosmetic outcome, without a cutaneous scar. (*Plast Reconstr Surg Glob Open* 2023; 11:e5155; doi: 10.1097/GOX.0000000000005155; Published online 3 August 2023.)

Intraosseous venous malformations may affect the bones of the maxillofacial skeleton and present as gradually enlarging, painless masses.<sup>1,2</sup> When indicated, the preferred management is en bloc resection with bony defect reconstruction, requiring awareness of facial contour, symmetry, and vasculature.<sup>3,4</sup> Removal of small, benign, medial maxillary bone lesions via a minimally invasive, skin-sparing strategy may allow for a successful functional and cosmetic outcome. Herein, we describe removal of an intraosseous maxillary venous malformation using a combined transoral and transconjunctival–transcaruncular approach, a technique previously described in orbital fracture repair.

## CASE PRESENTATION

A 26-year-old woman with a history of migraines presented with increased tearing and sinus pressure for 2 years and development of a painless mass below her left

medial canthus over 8 months. She denied vision changes and constitutional symptoms. Of note, magnetic resonance imaging (MRI) completed a decade before for her migraines showed no evidence of facial lesions.

The examination was significant for a firm, smooth mass inferior to the left medial canthus at the base of the nose with mobile overlying skin and soft tissues. There was an increase in tear lake volume, delay in fluorescein dye disappearance, and patent nasolacrimal irrigation, confirming functional nasolacrimal obstruction. The remainder of the ophthalmic, head, and neck examination was unremarkable; there were no abnormal vessels on nasal endoscopy. A computed tomography (CT) scan of the paranasal sinuses demonstrated a well-demarcated left anterior maxillary osseous lesion, consistent with an intraosseous venous malformation, and compression of the left nasolacrimal duct (Fig. 1). MRI of the paranasal sinuses revealed clear sinuses and no soft tissue infiltration from the lesion; there were no feeder vessels or signs suggestive of a high flow lesion. Surgical excision of the mass via a combined transoral and transconjunctival approach, to avoid a skin incision, performed by an otolaryngology-oculoplastics team was proposed.

Intraoperatively, nasolacrimal duct probing and silicone lacrimal intubation was performed to protect the lacrimal system. The nasojuugal fold was pre-marked in case the need to convert to an open approach arose. Exposure of the mass was pursued via a transgingival incision and a

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Received for publication May 31, 2022; accepted June 15, 2023.

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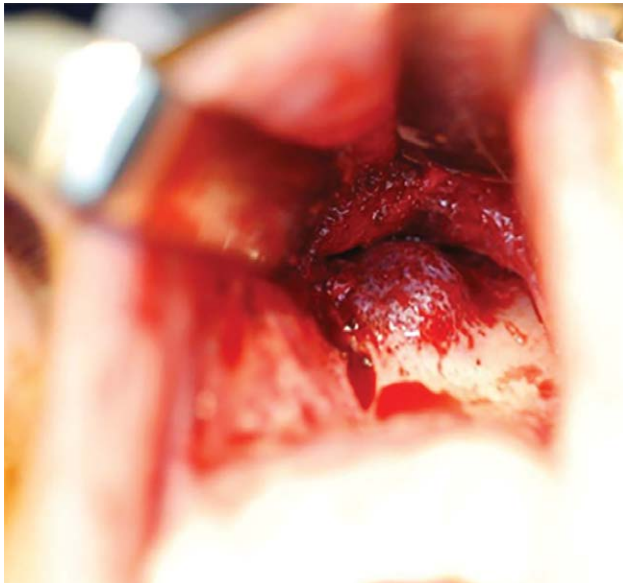
DOI: 10.1097/GOX.0000000000005155

Disclosure statements are at the end of this article, following the correspondence information.

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**Fig. 1.** CT scan image of the paranasal sinuses without contrast showing a well-demarcated, left anterior maxillary osseous lesion and compression of the left nasolacrimal duct. Note the typical starburst pattern of the intraosseous hemangioma.



**Fig. 2.** Visualization of the intraosseous hemangioma via a transoral, subperiosteal degloving approach.

degloving technique in the subperiosteal plane (Fig. 2). To provide additional superior exposure, a transconjunctival incision with extension into a transcaruncular incision was performed. A Colorado needle tip on monopolar cautery was used to incise through the conjunctiva and lower eyelid retractors just inferior to the inferior tarsal border, extending from the lateral-most aspect of the eyelid to 1 mm lateral to the punctum. Dissection was carried

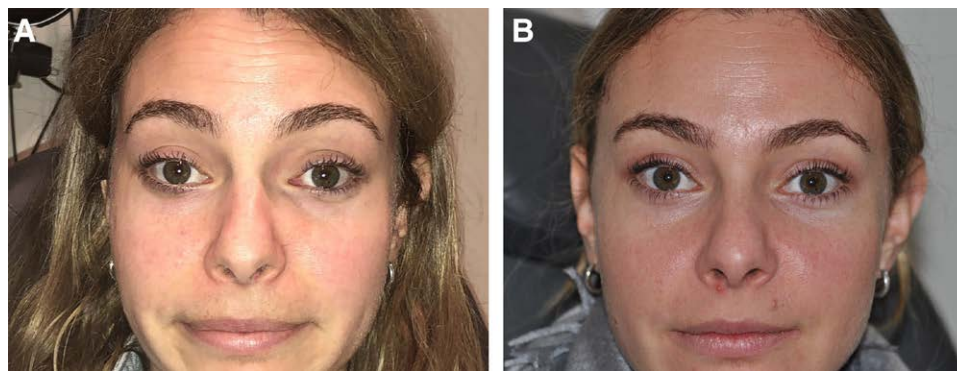
out in the preseptal plane down to the inferior orbital rim. The periosteum was incised and elevated away from the inferior orbital rim and onto the maxillary face, meeting the transoral dissection plane. The periorbita was elevated away from the medial orbital floor in the area of the mass, including elevation of the inferior oblique muscle origin. To provide additional exposure, a transcaruncular incision was made and extended inferolaterally to connect with the transconjunctival incision. Inspection of the exposed lesion, a well-circumscribed, violaceous mass, was consistent with a low-flow, intraosseous venous malformation, which was then removed en bloc using an osteotome. The defect along the inferomedial orbital rim and base of nose was plated with a titanium mesh sheet, which was fixated with screws to the inferior orbital rim via the transconjunctival incision and along the anterior maxillary face via the transgingival incision. [See figure, **Supplemental Digital Content 1**, which shows inferomedial orbital rim defect following lesion removal, visualized via transconjunctival-transcaruncular incision. A, Titanium mesh, molded and plated over the maxillary/orbital rim defect, visualized via the transoral incision (B). <http://links.lww.com/PRSGO/C701>.] Forced ductions confirmed no restriction of globe movement. Histopathologic examination confirmed diagnosis of an intraosseous venous malformation.

Postoperatively, the patient endorsed significant improvements in tearing and a satisfaction with her cosmetic result. Four weeks postoperatively, she demonstrated resolved nasolacrimal duct obstruction, preserved facial nerve function, and minor maxillary nerve hypesthesia. At 6 weeks, her lacrimal tubes were removed. At 5 months, preoperative maxillary fullness (Fig. 3A) was resolved, with an excellent cosmetic and functional outcome (Fig. 3B). At four years, there have been no signs of recurrence.

## DISCUSSION

Intraosseous venous malformations are rare, representing less than 1% of all bony tumors.<sup>1</sup> Although they typically involve the vertebral column and skull, they may also involve the maxillofacial skeleton, impacting the mandibular, maxillary, and nasal bones, with decreasing frequency.<sup>3,5</sup> These lesions predominantly present in women around the fourth decade of life as gradually enlarging, painless masses, posing an aesthetic concern; additional signs and symptoms vary with the location of the mass.<sup>2</sup> A differential diagnosis is included in Table 1.

CT remains the preferred imaging modality for intraosseous venous malformations and usually demonstrates a well-circumscribed, expansile lesion with a “honeycomb,” “sunburst,” or “soap bubble” appearance, owing to malformed venous channels within bone trabeculae.<sup>1</sup> On T2-weighted MRI, the lesion is typically hyperintense, reflecting slow-flowing blood or subacute thrombus; on T1-weighted imaging, some intraosseous venous malformations are hyperintense from thrombus or fat.<sup>1,4,5</sup> Management may be indicated with mass effect, repetitive bleeding, and cosmetic disfigurement. The treatment of choice is en bloc resection, with a sufficient margin of healthy bone to reduce bleeding risks. Reconstruction of



**Fig. 3.** Before and after facial photographs. A, Preoperative examination revealing a firm, smooth mass inferior to the level of the left medial canthus at the base of the nose with mobile overlying skin and soft tissues. B, At 5 months postoperative inspection, demonstrating resolution of maxillary fullness, with absence of cutaneous scarring.

**Table 1. Differential Diagnosis of Intraosseous Cavernous Venous Malformation of the Maxilla**

**Arterio-venous malformation**

Aneurysmal bone cyst\*

Fibrous dysplasia\*

Osteoma\*

Ossifying fibroma\*

Giant cell granuloma\*

Histiocytosis X\*

Ameloblastoma

Osteosarcoma

Hemangiopericytoma

\*These lesions may be amenable to combined transoral and transconjunctival-transcaruncular approach depending on size and relationship to adjacent structures.

the bony defect may be achieved via autogenous grafting or alloplastic materials.<sup>3,4</sup>

In our case, surgical excision of the vascular malformation was accomplished without a skin incision by coupling a transoral, midface degloving technique with a transconjunctival-transcaruncular approach to the inferomedial orbital rim, allowing for maximal exposure of the lesion. Combined transoral-transconjunctival exposure of the midface has been described in the treatment of zygomaticomaxillary complex fractures.<sup>6,7</sup> Combined transconjunctival-transcaruncular incision has been used as an approach to plating large fractures of the orbit involving the medial orbital wall, orbital strut, and orbital floor, safely protecting the lacrimal system.<sup>8</sup> With this technique, a single subperiosteal operating plane incorporating the medial orbit wall and floor can be achieved by disinserting the inferior oblique muscle at its origin at the posterior lacrimal crest.<sup>9,10</sup> Tagging the origin of the inferior oblique muscle with a suture allows it to be easily re-attached at the completion of surgery; failure to reattach the inferior oblique may be associated with postoperative muscle under-action.<sup>11,12</sup>

Potential surgical complications include lower eyelid retraction, infraorbital hypesthesia, diplopia secondary to inferior oblique muscle disruption, epiphora

secondary to lacrimal system damage, and orbital hematoma. We pre-marked the nasojugal fold to allow for a well-placed cutaneous incision, should there have been need for conversion to an open approach. An incision in this location is cosmetically acceptable and avoids damage to the facial nerve and infraorbital nerve; the angular artery in this region may be encountered and cauterized without significant consequence. In addition, the internal maxillary artery can be accessed via nasal endoscopy and cauterized in the event of severe bleeding from the maxilla.

Although technically challenging, this minimally invasive technique can be used to provide access to small (generally <3 cm), benign, medial maxillary bone lesions while avoiding a cutaneous scar.<sup>13,14</sup> Careful case selection is necessary to ensure that the mass can be accessed and removed while avoiding critical structures, including the lacrimal apparatus and infraorbital nerve; the approach may not be appropriate for high-flow vascular or malignant lesions.

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**DISCLOSURES**

*The authors have no financial interest to declare in relation to the content of this article. This study was supported, in part, by the UCSF Vision Shared Resource Core Grant (NIH/NEI P30 EY002162) and by an unrestricted departmental support grant from Research to Prevent Blindness to UCSF Department of Ophthalmology.*

**PATIENT CONSENT**

*The patient provided written consent for the use of her image.*

**HELSINKI DECLARATION**

*This report is in compliance with the Declaration of Helsinki and the Health Insurance Portability and Accountability Act.*

## REFERENCES

1. Strauss SB, Steinklein JM, Phillips CD, et al. Intraosseous venous malformations of the head and neck. *Am J Neuroradiol*. 2022;43:1090–1098.
2. Ramchandani PL, Sabesan T, Mellor TK. Intraosseous vascular anomaly (haemangioma) of the zygoma. *Br J Oral Maxillofac Surg*. 2004;42:583–586.
3. Koybasi S, Saydam L, Kutluay L. Intraosseous hemangioma of the zygoma. *Am J Otolaryngol*. 2003;24:194–197.
4. Powers DB, Fisher E, Erdmann D. Zygomatic intraosseous hemangioma: case report and literature review. *Craniomaxillofac Trauma Reconstr*. 2017;10:1–10.
5. Moore SL, Chun JK, Mitre SA, et al. Intraosseous hemangioma of the zygoma: CT and MR findings. *Am J Neuroradiol*. 2001;22:1383–1385.
6. Cortese A, D'Alessio G, Brongo S, et al. Management of zygomatic fractures in young patients: technical modifications for aesthetic and functional results. *JCraniofac Surg*. 2016;27:2073–2077.
7. Baumann A, Ewers R. Midfacial degloving: an alternative approach for traumatic corrections in the midface. *Int J Oral Maxillofac Surg*. 2001;30:272–277.
8. Imaizumi A, Ishida K, Nishizeki O. An extended transcaruncular approach for naso-orbito-ethmoid and Le Fort II fracture repair. *J Craniomaxillofac Surg*. 2016;44:1922–1928.
9. Cho RI, Davies BW. Combined orbital floor and medial wall fractures involving the inferomedial strut: repair technique and case series using preshaped porous polyethylene/titanium implants. *Craniomaxillofac Trauma Reconstr*. 2013;6:161–170.
10. Ishida K. Evolution of the surgical approach to the orbitozygomatic fracture: from a subciliary to a transconjunctival and to a novel extended transconjunctival approach without skin incisions. *J Plast Reconstr Aesthet Surg*. 2016;69:497–505.
11. Ahn JH, Jung JH, Choi HY. Ocular motility after repair of combined medial and inferior orbital wall fractures with extended conjunctival incision with inferior oblique reattachment. *JCraniofac Surg*. 2016;27:1312–1315.
12. Tiedemann LM, Lefebvre DR, Wan MJ, et al. Iatrogenic inferior oblique palsy: intentional disinsertion during transcaruncular approach to orbital fracture repair. *J AAPOS*. 2014;18:511–514.
13. Muscat K, Cobb R, Vassiliou L, et al. Scarless total maxillectomy: midfacial degloving with extended transconjunctival retrocaruncular approach. *Br J Oral Maxillofac Surg*. 2017;55:857–858.
14. Dikarev A, Porhanov V, Kochergina E, et al. Minimally invasive approach in surgical treatment of tumors of maxilla. *Head Neck Cancer Res*. 2016;1.