

A NOVEL MINIMALLY INVASIVE APPROACH TO AND RECONSTRUCTION OF A SOLITARY FIBROUS TUMOR OF THE NASOLACRIMAL DUCT

REBECCA PAQUIN, MD, DMD, CAMILO REYES GELVES, MD, LAUREN REID, MS, EDMOND RITTER, MD, J. KENNETH BYRD, MD, FACS

Abstract

Background: Nasolacrimal tumors present a diagnostic challenge given their rarity and the similarity of presenting symptoms to those of benign conditions. Minimal discussion is present in the literature regard reconstruction following resection of nasolacrimal tumors.

Methods: In this report, we present the case of a 60-year-old female found to have a unilateral solitary fibrous tumor (SFT) of the proximal nasolacrimal duct. En bloc resection was performed utilizing a completely internal, endoscopic Denker maxillectomy with split rib graft reconstruction.

Results: Tumor free margins were achieved with minimal cosmetic impact.

Conclusions: While en bloc resection is a common treatment for nasolacrimal tumors, this is the first recorded case using a minimally invasive technique without external incisions. We achieved an excellent cosmetic result, and the patient was satisfied with the outcome.

Introduction

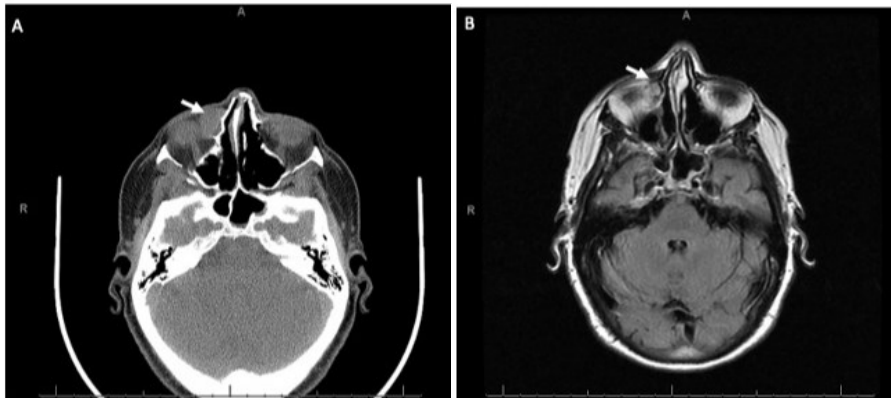
The solitary fibrous tumor (SFT) was first described by Klemperer and Coleman in 1931 as a mesenchymal neoplasm of the pleura.¹ Although initially thought to arise only from the pleura, SFTs are now known to arise from a diverse range of anatomic sites.²⁻⁵ According to the most recent literature, approximately 50-70% of all SFTs are localized outside of the thorax.⁶ There have now been numerous reports of SFTs occurring in the head and neck. Sites reported in the head and neck include the meninges, thyroid, larynx, nasal cavity, paranasal sinuses, orbit, nasopharynx, oral cavity, parapharyngeal space, neck, scalp, and facial soft tissues.⁵ Orbital involvement was first reported in 1994.⁷ SFTs are generally considered to exhibit benign behavior, but some case series have found rates of metastasis as high as 34%.⁶

There is also a paucity of literature regarding ablative and reconstructive surgery of the nasolacrimal system. While nasolacrimal tumors are rare, the spectrum of lacrimal sac tumors carries a 55% malignancy rate.⁸ Further, the diagnosis of nasolacrimal tumors tends to be delayed due to rarity of tumors of this site, slow onset of symptoms, and presumed diagnosis of dacryostenosis or dacryocystitis.⁹⁻¹³ An ablative approach previously used for nasolacrimal tumors was the dacryocystectomy, but this tended to leave positive margins and carried a mortality rate as high as 50%.^{10,14,15} Valenzuela et al., in 2006, were the first in the literature to suggest en bloc excision as a radical treatment for nasolacrimal tumors.⁹ Albiad et al. modified the reported technique by performing a combined sinus-orbit approach with endoscopic guidance. The authors were also the first to describe reconstruction of the bone defect left from en bloc resection, for which they used titanium mesh.¹⁵ There have been no reports to date in which external incisions were able to be avoided. In this article, we describe a case in which we performed a minimally invasive en bloc resection of a nasolacrimal SFT with an entirely internal approach and followed with immediate reconstruction with a split rib graft.

Case Description

The patient is a 60-year-old female with a history of chronic rhinosinusitis with nasal polyposis who had undergone functional endoscopic sinus surgery in the past and was known to our rhinology team. She presented to ophthalmology with the complaint of three months of gradual enlargement of the right medial canthal region as well as epiphora (figure 6A). She denied pain or discharge.

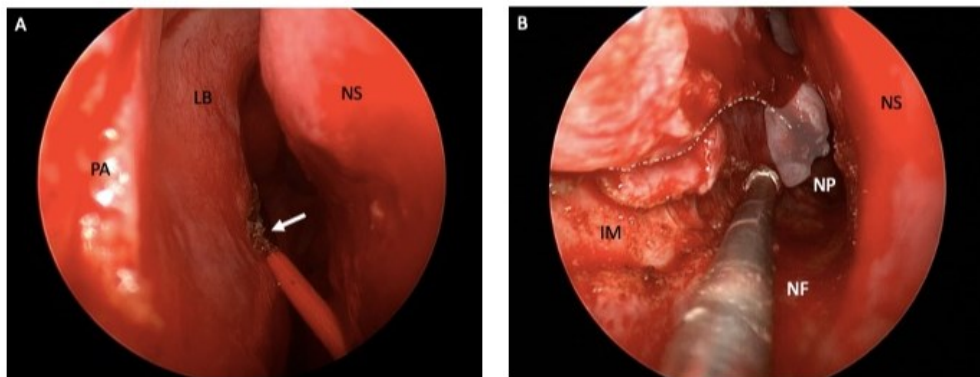
She was noted on exam to have mild swelling and erythema overlying the nasolacrimal region without fluctuance or discharge. The initial differential diagnosis included chronic dacryocystitis versus dacryocystocele. As the patient reported a distant history of bilateral orbital floor fractures, a computed tomography (CT) scan of the orbits and paranasal sinuses was obtained. This demonstrated a soft tissue mass at the level of the right lacrimal sac measuring 19 x 14 x 16 millimeters with uniform enhancement. There was no cystic component noted, which was incongruous with a dacryocystocele.



Figures 1A and 1B

Widening of the bony walls of the proximal nasolacrimal duct was noted, consistent with chronic pathology (figure 1A). On review of a previous CT scan of the sinuses from four years prior, the lesion was again noted, at that time measuring 9 millimeters in largest dimension. Biopsy of the lacrimal sac was performed, which demonstrated SFT. The oculoplastic surgery team then performed a right dacryocystectomy and dacryocystorhinostomy with placement of Crawford tubes.

The Crawford tubes were removed five months postoperatively. A magnetic resonance imaging (MRI) scan of the face, neck, and orbits performed at this time demonstrated asymmetric soft tissue enhancement and enlargement along the right medial orbit and nasolacrimal duct (figure 1B).

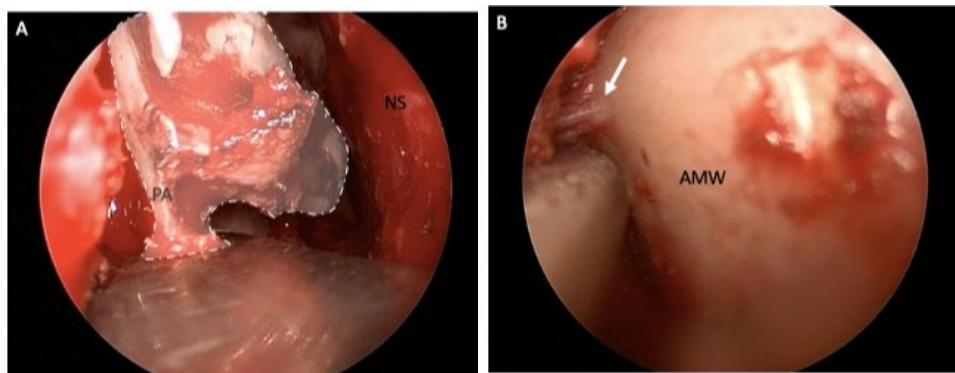


Figures 2A and 2B

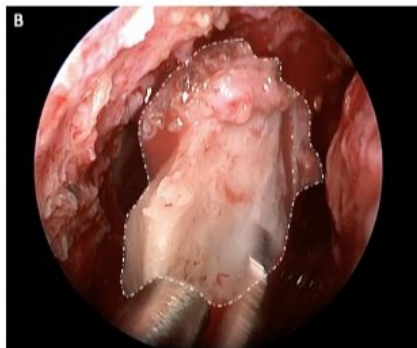
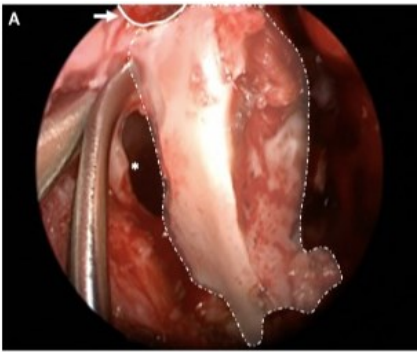
After discussion of the case by our multidisciplinary head and neck tumor board, it was decided that the patient should undergo an endoscopic Denker maxillectomy with en bloc resection of the nasolacrimal apparatus and immediate reconstruction, to be performed as a joint case by otolaryngology-head and neck surgery and plastic surgery.

After discussion of the case by

Using image guidance and endoscopic instrumentation, an incision was made along the lateral nasal wall from the piriform aperture to the previous maxillary antrostomy (figure 2A). The incision was then extended superiorly to the level of the orbital floor. The mucosa was elevated in a subperiosteal plane anterolaterally to the orbital foramen. Using a surgical handpiece with a diamond bur, an osteotomy was performed within the medial maxillary sinus wall along the vertical incision (figure 2B). This was carried laterally, with protection of the infraorbital nerve, using a curved osteotome (figure 3A-B).



Figures 3A and 3B



Figures 4A and 4B

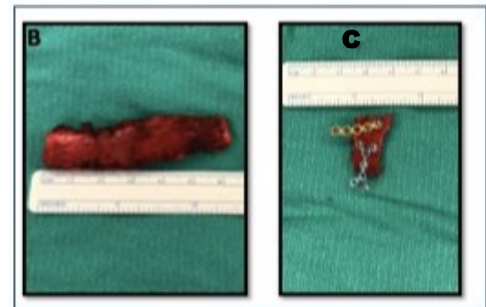
Then, with appropriate protection of the orbital contents, the diamond bur was carried superiorly from there to the orbital rim, then posteriorly to the level of the previous maxillary antrostomy, and finally medially to the initial bur cut, removing the specimen en bloc (figure 4A-B). Anterior, posterior, superior, and inferior soft tissue margins were then sent for frozen section analysis. These were all noted to be negative for tumor. Next, the left fifth rib was harvested. This was split and contoured appropriately on the back table using an oscillating saw and osteotome. A maxillary labial incision was performed and dissected superiorly in the subperiosteal plane to fully expose the bone defect. The tailored rib graft was fixated with two low-profile plates. Cancellous bone harvested from the remaining rib was packed around the edges of the bone graft (figure 5 A-C). The buccal sulcus incision was closed with chromic suture. Doyle splints were placed in both nares and secured anteriorly with a nylon suture. The patient had an uncomplicated postoperative course. The intranasal splints were removed on postoperative day 11, and the patient resumed nasal saline irrigations and fluticasone nasal spray that day. The patient's only complaint was hypesthesia of the right infraorbital nerve distribution, but sensation has progressively returned with time. An MRI of the face, neck, and orbits obtained six months postoperatively

demonstrated no abnormal enhancement. The patient is quite satisfied with her appearance (figure 6B).

Discussion

Tumors of the nasolacrimal system are rare.¹⁶ Nasolacrimal neoplasms typically present gradually with epiphora, dacrocystitis, and a palpable mass.¹¹⁻¹³ A high index of suspicion is necessary when these signs are present, as diagnosis of neoplasms of the nasolacrimal system tend to be delayed due to a misdiagnosis of dacrocystitis or dacrostenosis.⁷⁻¹¹ Krishna et al., in their review of lacrimal sac tumors, found a minimum 55% malignancy rate (range 55-100%) for primary lacrimal sac tumors in addition to a high rate of local invasion and recurrence.¹²

Management of nasolacrimal tumors depends on tumor type, malignancy, size, extension, and the patient's general health.¹² Due to the lack of randomized controlled trials regarding SFTs, there is no established, universally accepted treatment strategy. The general recommendation is that this tumor be treated similar



Figures 5A, 5B, and 5C

to other soft tissue sarcomas.¹⁷ Gold et al. found that positive surgical margins after resection of SFT predicted both worse local recurrence-free survival and worse metastasis-free survival.¹⁸ In one of the largest clinicopathological studies regarding SFT, Demicco et al. found 5- and 10-year rates of metastasis of 74% and 55%, respectively.¹⁹ En bloc resection of the nasolacrimal apparatus and its enveloping bony structures has gained support among orbital and head and neck surgeons in recent years as an approach to nasolacrimal tumors that is able to achieve good local control.^{8,9,15,16,19} In this case, we were able to perform a minimally invasive en bloc resection of a nasolacrimal tumor without any skin incision and achieve tumor-free margins.



Figures 6A and 6B

There is little published in the literature regarding reconstruction following resection of nasolacrimal tumors. Albiad et al. described simultaneous reconstruction of the bony defect with contoured titanium mesh through a combined sinus-orbit approach, with the finding that they were able to provide fixation for the medial canthal tendon, provide globe support, and prevent midface collapse.¹⁵ There has otherwise been no significant mention of reconstruction following resection of nasolacrimal apparatus. We found that reconstruction with a split rib graft allowed for excellent cosmesis following resection.

Based on findings in the literature and in this case, there are a few concluding remarks to highlight. First, a high index of suspicion should be had when patients present with epiphora, dacryocystitis, and a palpable nasolacrimal mass, as these are commonly misinterpreted signs of potentially malignant nasolacrimal neoplasms. Second, it is possible to provide en bloc resection with clear margins without disfiguring scars by using an endoscopic-assisted internal approach to nasolacrimal tumors. Third, a split rib graft, as demonstrated, above can provide excellent cosmesis following en bloc resection of nasolacrimal tumors.

References

1. Klemperer P, Coleman BR. Primary neoplasms of the pleura. A report of five cases. *Am J Ind Med.* 1992;22(1):1-31.
2. Hanau CA, Miettinen M. Solitary fibrous tumor: histological and immunohistochemical spectrum of benign and malignant variants presenting at different sites. *Hum Pathol.* 1995;26(4):440-449.
3. Suh YJ, Park JH, Jeon JH, Bilegsaikhan SE. Extrapleural solitary fibrous tumor of the thyroid gland: A case report and review of literature. *World J Clin Cases.* 2020;8(4):782-789.
4. Smith SC, Gooding WE, Elkins M, et al. Solitary Fibrous Tumors of the Head and Neck: A Multi-Institutional Clinicopathologic Study. *Am J Surg Pathol.* 2017;41(12):1642-1656.
5. Ayad T, Ghannoum J. Solitary fibrous tumor with pseudo-lipoblasts involving the sublingual gland: report of a case and review of the literature. *Eur Arch Otorhinolaryngol.* 2007;264(1):93-98.
6. van Houdt WJ, Westerveld CM, Vrijenhoek JE, et al. Prognosis of solitary fibrous tumors: a multicenter study. *Ann Surg Oncol.* 2013;20(13):4090-4095.
7. Westra WH, Gerald WL, Rosai J. Solitary fibrous tumor. Consistent CD34 immunoreactivity and occurrence in the orbit. *Am J Surg Pathol.* 1994;18(10):992-998.
8. Gervasio KA, Mellen PL, Boparai RS, et al. Combined Endoscopic and External Resection of an Inverted Papilloma of the Lacrimal Sac: A Case Report and Review of the Literature. *Allergy Rhinol (Providence).* 2019;10:2152656719876254.
9. Valenzuela AA, Selva D, McNab AA, Simon GB, Sullivan TJ. En bloc excision in malignant tumors of the lacrimal drainage apparatus. *Ophthalmic Plast Reconstr Surg.* 2006;22(5):356-360.
10. Hornblass A, Jakobiec FA, Bosniak S, Flanagan J. The diagnosis and management of epithelial tumors of the lacrimal sac. *Ophthalmology.* 1980;87(6):476-490.
11. Valenzuela AA, McNab AA, Selva D, O'Donnell BA, Whitehead KJ, Sullivan TJ. Clinical features and management of tumors affecting the lacrimal drainage apparatus. *Ophthalmic Plast Reconstr Surg.* 2006;22(2):96-101.
12. Krishna Y CS. Lacrimal Sac Tumors - A Review. *Asia-Pacific Journal of Ophthalmology.* 2017;6(2):173-178.
13. Song X, Wang J, Wang S, Wang W, Wang S, Zhu W. Clinical analysis of 90 cases of malignant lacrimal sac tumor. *Graefes Arch Clin Exp Ophthalmol.* 2018;256(7):1333-1338.
14. Ni C, D'Amico DJ, Fan CQ, Kuo PK. Tumors of the lacrimal sac: a clinicopathological analysis of 82 cases. *Int Ophthalmol Clin.* 1982;22(1):121-140.
15. Albiad CR, Weed DT, Walker TJ, et al. En bloc resection of lacrimal sac tumors and simultaneous orbital reconstruction: surgical and functional outcomes. *Ophthalmic Plast Reconstr Surg.* 2014;30(6):459-467.
16. Curragh DS, James C, Selva D. Swinging inferior turbinate approach to the nasolacrimal duct. *Orbit.* 2020;39(2):112-117.
17. Davanzo B, Emerson RE, Lisy M, Koniaris LG, Kays JK. Solitary fibrous tumor. *Transl Gastroenterol Hepatol.* 2018;3:94.
18. Gold JS, Antonescu CR, Hajdu C, et al. Clinicopathologic correlates of solitary fibrous tumors. *Cancer.* 2002;94(4):1057-1068.