Warthin's Tumor of the Nasolacrimal Duct

Neda Ahmadi, MD[1]; Lauren Z. Gavaris, MD[2]; Nazaneen Grant, MD[1, 2]
[1]Georgetown University Hospital, [2]Washington Hospital Center

Georgetown University Hospital
MedStar Health

ABSTRACT

Objectives:
The objectives of this study are to review the anatomy of the nasolacrimal apparatus and to present a rare tumor of the nasolacrimal duct.

Methods:
Retrospective case report and review of literature

Results:
A 67-year-old woman presented with right nasal obstruction. Examination revealed a right sided nasal cavity mass. Imaging revealed a mass originating from the right nasolacrimal duct with extension into the right orbit. Histopathologic examination of the tumor identified the tumor as Warthin's tumor.

Discussion/Conclusion:
Primary tumors of the nasolacrimal apparatus are rare. Most originate from the lacrimal sac and more than half of these tumors are malignant. Primary epithelial tumors are the most common type of tumors encountered. These tumors should be considered in the differential diagnosis of a patient presenting with nasal obstruction, epiphora, or a nasal cavity mass. Given advances in endoscopic techniques, it is important for otolaryngologists to be familiar with the different pathologies that involve the nasolacrimal apparatus and their clinical presentation. In this article, we report a rare type of nasolacrimal duct tumor, and a present a literature review of primary tumors of the nasolacrimal apparatus.

CONTACT

Neda Ahmadi, MD
Georgetown University Hospital
Email: NXA113@gunet.georgetown.edu

INTRODUCTION

Tumors of the nasolacrimal system are rare and mostly arise from the lacrimal sac.[1] More than 400 primary lacrimal sac tumors have been reported.[2] The frequency of lacrimal sac tumors is between 3.3 to 6.1%.[3] Primary tumors of the nasolacrimal duct (NLD) are even more rare. The most common clinical presentations of these tumors include epiphora, pain, medial canthal swelling, blood-tinged tears and epistaxis.[1]

CASE PRESENTATION

A 67-YEAR-OLD WOMAN PRESENTED with a history of right nasal obstruction and pain over the nasal bridge. She denied epistaxis and ocular symptoms such as pain, diplopia and epiphora. Her past medical history was significant for right dacryocystorhinostomy (DCR) fifteen years ago. She denied history of smoking.

Physical examination was significant for fullness of the right medial canthal area, and tenderness of the right nasal sidewall. Nasal endoscopy revealed a large erythematous polypoid lesion protruding from the anterior aspect of the right middle meatus. There was no bleeding noted. Extra-ocular muscle movements were within normal limits and there was no proptosis or globe displacement. Computed tomography demonstrated a 9 × 12 × 14-mm lesion originating from the right nasolacrimal duct (NLD) and entering into the nasal cavity medially through the previous DCR and entering the orbit superolaterally (**Figure 1A**). The orbital component was extraconal and measured 15 × 13 × 17-mm (**Figure 1B**). The biopsy of the intra-nasal portion of the lesion in the clinic demonstrated inflammatory cells.

The patient underwent excision of the lesion and dacryocystorhinostomy using a combined endoscopic and external approach. The orbital portion was removed by ophthalmology using a frontoethmoidal "Lynch" incision (**Figure 2**). Hematoxylin-eosin staining revealed a benign tumor composed of bilayered oncocytic epithelium with fluid containing microcystic spaces and small foci of lymphocytic aggregates (**Figures 3**) consistent with Warthin's tumor.

DISCUSSION

An appreciation of the nasolacrimal anatomy is important in understanding the pathologies involved. The nasolacrimal apparatus starts with the 0.3mm medial canthal punctal openings that lead to the superior and inferior vertical canaliculi (each measuring 2mm in length). These canaliculi make 90 degree turns leading to the 8mm superior and inferior horizontal canaliculi, which form the common canaliculus prior to entering the lacrimal sac. The common canaliculus and the lacrimal sac are located between the anterior and posterior limbs of the medial canthal ligament. The lacrimal sac measures 12 – 15mm in length and lies within the lacrimal fossa. The anterior crest of the lacrimal fossa is formed by the frontal process of the maxilla and the posterior crest is formed by the lacrimal bone.[4]

Intranasally, a significant (10mm) portion of the lacrimal sac is located above the insertion of the anterior end of the middle turbinate.[5] The sac leads to the NLD (18-24mm in length), which is located approximately 4mm anterior to the maxillary sinus ostium.[6] The NLD opens into the inferior meatus and a mucosal valve (Hasner) prevents retrograde reflux of nasal contents into the NLD. This opening is located approximately 25mm from the anterior nasal spine. It is also 13.7mm superior to the nasal floor and 14.3mm posterior to the anterior attachment of inferior turbinate.[6]

Lesions of the nasolacrimal system are classified into epithelial and non-epithelial tumors. Primary epithelial tumors are the most common accounting for 70% of all reported cases, followed by mesenchymal tumors (12%), lymphoid lesions (11%), malignant melanoma (4%) and others (1%).[2,7] Benign epithelial tumors include squamous and transitional cell papillomas, oncocytomas and benign mixed tumors. The mean age of patients with benign epithelial tumors is 52 years. Excision of the lesion and DCR are considered adequate when managing these benign tumors.

Malignant epithelial tumors include squamous cell carcinoma, papillomas with foci of carcinoma, transitional cell carcinoma, adenocarcinoma, mucoepidermoid carcinoma, adenoid cystic carcinoma and poorly differentiated carcinoma. Squamous cell carcinoma is the most common. Lateral rhinotomy with extensive surgical excision of the nasolacrimal system including the canaliculi, sac and NLD is the recommended surgical management. The recurrence rates are estimated to be 12.5%.[2] Pre- or post-operative radiation therapy along with lymph node dissection is indicated in larger widespread tumors.[2]

Primary non-epithelial tumors include mesenchymal lesions (fibrous histiocytoma, hemangiopericytoma, histiocytoma, angiosacoma), lymphoproliferative lesions (lymphoma), malignant melanoma and others such as neurofibroma or granulocystic sarcoma. Fibrous histiocytoma is the most common non-epithelial malignant tumor, followed by lymphoma and malignant melanoma.[2]

The pathological finding in our patient is unusual. Warthin's tumor (papillary cystadenoma lymphomatosum) is a common benign tumor of the parotid gland, accounting for 14% of all benign and malignant epithelial neoplasms of the gland.[8] A correlation between Warthin's tumor and cigarette smoking is hypothesized. However, our patient was not a smoker. It has been reported that 8% of Warthin's tumors occur in extraparotid locations, with cervical lymph nodes being the most common location.[8] The embryogenesis and histogenesis of extra-parotid Warthin's tumors (EPWT) is controversial. The pathophysiology that currently explains the presence of Warthin's tumor in periparotid and cervical regions is explained by the later encapsulation of the parotid gland.[9] However, despite this, EPWTs occurring in unusual sites such as the larynx, and the hard palate have been reported.[9] According, to Snyderman, *et al*.[10] EPWTs of distant sites lack the characteristic lymphoid component and are best classified as oncocytic papillary cystadenomas. Histological evaluation of our case demonstrates the presence of bilayered epithelium lining the cystic spaces and the surrounding lymphoid stroma; thus, confirming the diagnosis of EPWT.

To our knowledge this is the first reported case of Warthin's tumor involving the nasolacrimal system. Given the benign nature of these tumors, the management includes excision and DCR. At 1 year postoperatively, our patient has no evidence of disease and her presenting symptoms of nasal obstruction and pain have resolved.

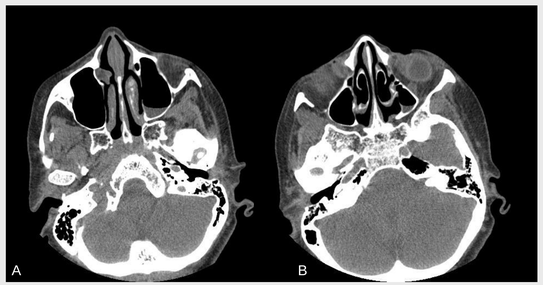

Figure 1. Axial non-contrast CT demonstrating the lesion in the right NLD (A) extending into the orbit (B).

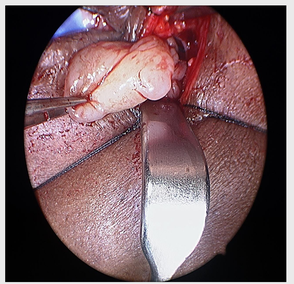

Figure 2. The orbital portion of the tumor being excised through a Lynch incision.

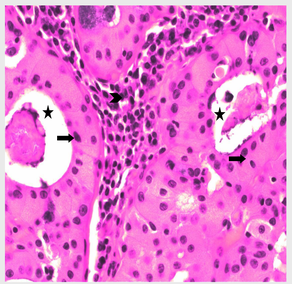

Figure 3. This section demonstrates the bilayered epithelium (arrows) lining the cystic spaces (stars), and the surrounding lymphoid stroma (arrowhead) (H&E 400x).

CONCLUSIONS

Although tumors of the nasolacrimal apparatus are rare, otolaryngologists will likely encounter patients who present with epiphora or unilateral pain over the nasal bridge. Therefore, familiarity with the anatomy of the nasolacrimal apparatus and the tumors arising from this structure are essential.

REFERENCES

1. Sakaida H, Kobayashi M, Yuta A, et al. Squamous cell carcinoma of the nasolacrimal duct. *Eur Arch Otorhinolaryngol*. 2009 Mar;266(3):455-8.
2. Heindl LM, Jünemann AG, Kruse FE, et al. Tumors of the Lacrimal Drainage System. *Orbit*. 2010 Oct;29(5):298-306.
3. Marthin JK, Lindegaard J, Prause JU, et al. *Acta Ophthalmol Scand*. 2005 Feb;83(1):94-9.
4. Chastain JB, Sindwani R. Anatomy of the orbit, lacrimal apparatus, and lateral nasal wall. *Otolaryngol Clin North Am*. 2006 Oct;39(5):855-64, v-vi.
5. Wormald PJ, Kew J, Hasselt AV, et al. Intranasal anatomy of the nasolacrimal sac in endoscopic dacryocystorhinostomy. *Otolaryngol Head Neck Surg*. 2000 Sep;123(3):307-10.
6. Tatlisumak E, Aslan A, Comert A, et al. Surgical anatomy of the nasolacrimal duct on the lateral nasal wall as revealed by serial dissections. *Anat Sci Int*. 2010 Mar;85(1):8-12.
7. Stefanyszyn MA, Hidayat AA, Pe'er JJ, et al. Lacrimal Sac Tumors. *Ophthal Plast Reconstr Surg*. 1994 Sep;10(3):169-84.
8. Patterson JW, Wright ED, Camden S. Extraparotid Warthin's tumor. *J Am Acad Dermatol*. 1999 Mar;40(3):468-70.
9. Astor FC, Hanft KL, Rooney P, et al. Extraparotid Warthin's tumor: clinical manifestations, challenges, and controversies. *Otolaryngol Head Neck Surg*. 1996 Jun;114(6):732-5.
10. Snyderman C, Johnson JT, Barnes EL. Extraparotid Warthin's tumor. *Otolaryngol Head Neck Surg*. 1986 Feb;94(2):169-75.