ONCOCYTIC CARCINOMA OF THE NASOLACRIMAL DUCT TREATED BY TRANSNASAL ENDOSCOPIC RESECTION

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Abstract: *Background.* Malignancies of the nasolacrimal apparatus are rare lesions that usually originate from the lacrimal sac. To the best of our knowledge, this is the first case of a primary oncocytic carcinoma of the nasolacrimal duct reported in the literature.

Methods. A 43-year-old woman presented with a 3-month history of right epiphora. At nasal endoscopy, an exophytic lesion occupying the right inferior meatus was visible. Imaging studies showed an expansile lesion involving the nasolacrimal duct and the inferior meatus, but not invading the orbit or adjacent soft tissues. A biopsy was suggestive for an oncocytic lesion.

Results. A radical resection was obtained through an endoscopic extended medial maxillectomy with a definitive diagnosis of oncocytic carcinoma. Forty-two months after surgery there was no evidence of disease.

Conclusions. In selected malignant lesions of the nasolacrimal duct not involving the adjacent soft tissues or lacrimal sac, an exclusive endoscopic approach can be considered. © 2011 Wiley Periodicals, Inc. *Head Neck* **00:** 000–000, 2011

Keywords: nasolacrimal duct; malignant tumors; oncocytic carcinoma; endoscopic resection; transnasal resection

Malignancies of the nasolacrimal drainage apparatus are rare. Most originate from the lacrimal sac, with only a few reports of primary nasolacrimal duct malignancies.[1–5] A wide range of different histologies have been observed, including lymphoma, Kaposi sarcoma, adenocarcinoma, squamous cell carcinoma, mucosal melanoma, and metastases from distant sites. Oncocytic carcinoma has been reported in only 8 patients, where the lesion developed from the lacrimal sac.[6–11] To the best of our knowledge, this is the first reported case of oncocytic carcinoma of the nasolacrimal duct treated by a pure endoscopic approach.

CASE REPORT

A 43-year-old woman presented with a 3-month history of right epiphora. Nasal endoscopy revealed a gray-reddish, partially necrotic lesion filling the anterior third of the right inferior meatus (see Figure 1). CT and MRI showed an expansile lesion occupying the right inferior meatus and nasolacrimal duct. Apparently, the lesion originated from the nasolacrimal duct without involving the middle turbinate. The medial wall of the maxillary sinus was deformed but not infiltrated by the tumor, which was also in contact with the orbital wall. The lacrimal sac did not appear to be involved by the lesion (see Figure 2). A transnasal biopsy performed under local anesthesia was indicative of an oncocytic lesion, but left open the differential diagnosis between benign oncocytoma and low-grade adenocarcinoma. Excision via an extended endoscopic medial maxillectomy, also known as endonasal Denker operation,[12] was planned. Frozen sections obtained at the level of the lacrimal sac and the residual mucosa of the inferior meatus were negative. The procedure was completed by resecting the medial wall of the lacrimal sac, and creating a dacryocystorhinostomy; the residual lacrimal pathway was stented. The postoperative course was uneventful. Histologic examination of the neoplasm showed a combination of papillary-cystic, solid, and cribriform growth patterns. Cells were polyhedral in shape, characterized by abundant, finely granular, eosinophilic cytoplasm, and showed diffuse immunoreactivity for mitochondria. Nuclei were characterized by a moderate pleomorphism with mitoses. There was no evidence of infiltration of either the medial wall of the lacrimal sac or the bony wall of the nasolacrimal duct. A definitive diagnosis of oncocytic carcinoma was rendered. No adjuvant treatment was administered. The patient has been followed by endoscopic and MRI controls every 6 months. When last seen, 42 months postoperatively, she was free of disease (see Figure 3).

DISCUSSION

Whenever faced with a patient complaining of persistent epiphora, especially if unilateral, the clinician should consider the possibility that the symptom

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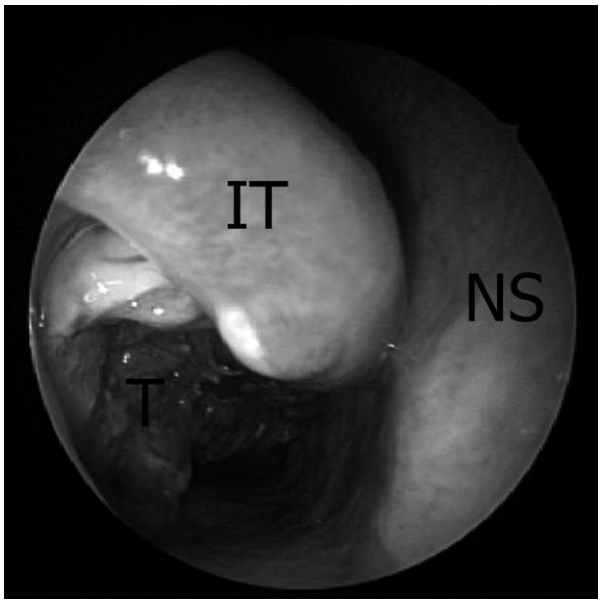

FIGURE 1. Preoperative endoscopic view of the tumor (T). The figure shows a mass growing into the inferior meatus. IT, inferior turbinate; NS, nasal septum.

might be related to a tumor originating within the lacrimal pathway or from an adjacent site such as the sinonasal tract, with secondary obstruction by compression or infiltration of the nasolacrimal duct. Currently, benefiting from increased collaboration, the ophthalmologist and otolaryngologist may obtain complete assessment of the lacrimal system and the adjacent nasal cavity with intracanalicular and nasal endoscopy. In the present case, the patient was first referred to an otolaryngologist and a mass in the inferior meatus impairing the drainage of the lacrimal system was easily recognized by rigid endoscopy.

Even though the biopsy did not assess the benign or malignant nature of the lesion, a proliferation of cells with an abundant acidophilic cytoplasm rich in mitochondria, called oncocytic cells, was observed. Oncocytic cells, first described by Hamperl[13] in 1931, are transformed cells characterized by a large size and abundant acidophilic cytoplasm rich in mitochondria.[14] It is generally thought that this transformation is related to the aging process. Lesions composed predominantly of these cells can be subdivided into oncocytic hyperplasia, benign oncocytoma, or oncocytic carcinoma, depending on the histologic features.[15] In the sinonasal tract, most oncocytic lesions

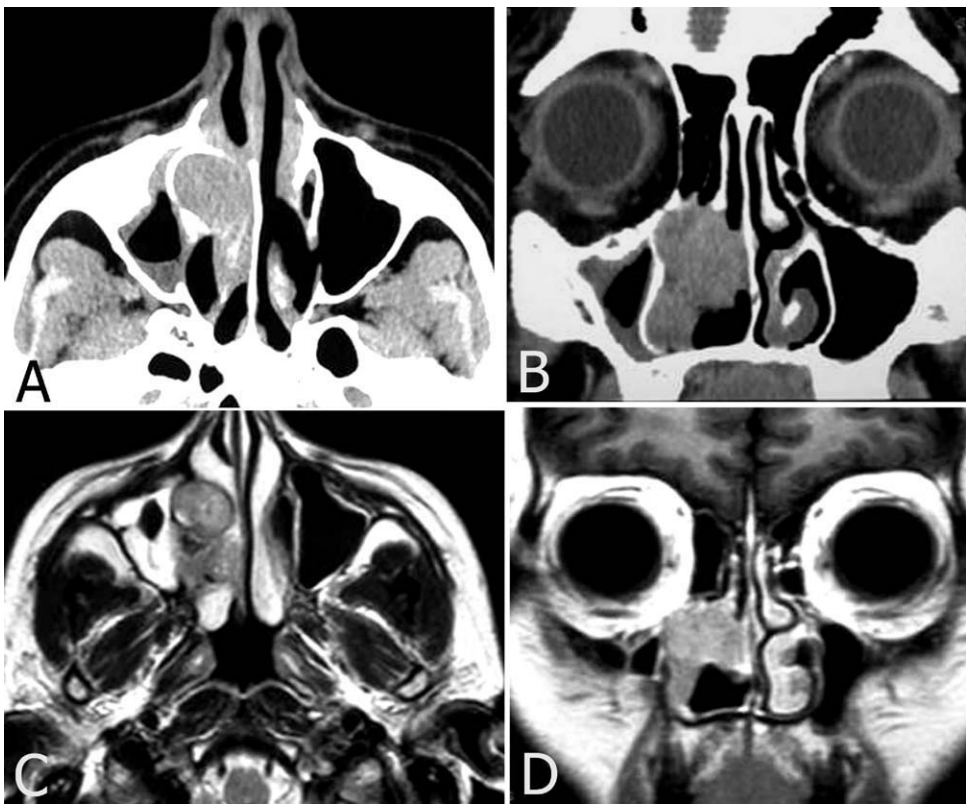

FIGURE 2. Plain CT on axial (**A**) and coronal (**B**) planes; MRI SE T2 on axial plane (**C**); MRI SE T1 after gadolinium administration on coronal plane (**D**). On the right side, the inferior meatus is partially occupied by a lesion arising from the most caudal part of the nasolacrimal duct. The duct itself is grossly enlarged; its bony walls are remodeled. Inferior growth along the lateral wall of the nasal cavity is observed, although the maxillary sinus is not involved.

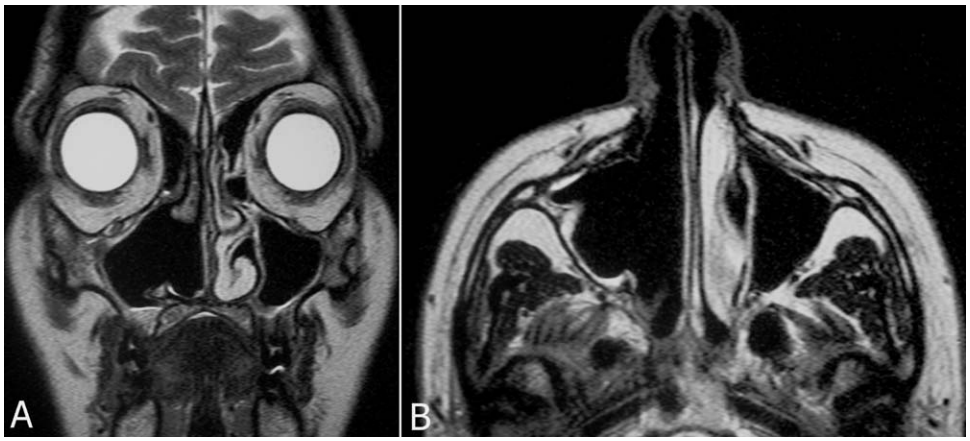

FIGURE 3. MRI SE T2 on coronal (**A**) and axial (**B**) planes. Follow-up MRI performed 42 months after surgery shows no sign of recurrence.

arise from the nasal septum,[16] whereas their origin from the nasolacrimal apparatus is very rare. Pe'er et al[11] reviewed 118 primary lacrimal sac tumors and found that benign and malignant oncocytic tumors accounted for 6% of all cases. Only 3 of these (2.5% of the total) were oncocytic carcinomas. After reviewing the literature, we identified 8 cases of oncocytic carcinoma, all of which originated from the lacrimal sac.[6–11] To the best of our knowledge, this is the first reported case of primary nasolacrimal duct oncocytic carcinoma. Even benign oncocytoma more frequently affects the lacrimal sac, and only 2 cases stemming from the nasolacrimal duct have been reported in the literature.[10,17]

The treatment of choice for malignant tumors of the nasolacrimal pathway is wide surgical excision with free margins.[7,10,11,18] An open approach, modulated on the extent of the disease, is classically used. Adjuvant chemotherapy or radiotherapy can also be added, depending on the histology and status of resection margins.[8,19,20] The use of an endoscopic technique, although in combination with an external approach, has been addressed in only 2 reports.[20,21] This is the first case of a primary nasolacrimal malignancy excised via an exclusive transnasal endoscopic approach. CT and MRI findings suggested that the lesion was confined to the nasolacrimal duct, which was considerably increased in size but still surrounded by a thin layer of bone, with no involvement of adjacent soft tissues (see Figure 2). The absence of any critical extent to the sac and the medial wall of the orbit as well as the presence of a limited nasal involvement of the inferior meatus allowed us to plan an endoscopic resection via a Sturmann–Canfield operation (or endonasal Denker operation). This approach, popularized by Brors and Draf,[12] is performed under endoscopic control and enables complete exposure of the nasolacrimal duct and sac without resorting to external access. This is achieved by making an incision anterior to the inferior turbinate, exposing the piriform crest, dissecting the soft tissues of the anterior wall of the maxillary sinus until the infraorbital nerve is visualized, and drilling the anteromedial corner of the maxillary sinus.

Not unexpectedly, endoscopic resection of tumors involving the nasolacrimal apparatus has not been reported to date. In addition to the rarity of the disease, the anatomic configuration of the nasolacrimal apparatus makes endoscopic management of tumors rarely feasible. In fact, the majority of tumors of the nasolacrimal apparatus involve the lacrimal sac; the proximity of the lesion to the orbit, skin, and subcutaneous tissues, together with frequent extension outside of the limits of the sinonasal cavity, represents a clear indication for an open approach. In contrast, in the present case the tumor originated within the nasolacrimal duct and did not extend to the sac. Another element that made the endoscopic resection feasible was the fact that, as suggested by imaging findings, the tumor was intraoperatively found to simply expand the nasolacrimal duct without infiltrating the surrounding bone. Although involvement of the nasolacrimal duct by tumors originating from the sinonasal tract is not a rare finding, only 3 cases of malignancies originating from and confined to the nasolacrimal duct have been reported in the literature.[5,22] In all cases, in view of the histologic diagnosis of malignant melanoma, extensive resection using an external approach was performed.

Oncocytic carcinoma is characterized by a relatively high rate of recurrence and a slow pattern of growth. These factors call for periodic endoscopic and imaging examinations, which in our patient were performed every 6 months, similar to the surveillance strategy adopted at our institution for malignancies of the sinonasal tract.

CONCLUSION

Oncocytic carcinoma of the lacrimal drainage system is a rare lesion. In all cases reported so far the tumor

originated from the lacrimal sac. We present the first case of oncocytic carcinoma limited to the nasolacrimal duct, which was radically removed with an exclusive endoscopic approach. In carefully selected lesions that do not extend beyond the limits of the nasolacrimal duct, a pure transnasal endoscopic approach may be considered as a valid option to more extensive transfacial techniques.

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