Sinonasal (Schneiderian) papilloma of the lacrimal sac

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ABSTRACT

Sinonasal papillomas, also known as Schneiderian papillomas, are benign but infiltrative epithelial neoplasms that arise from ectodermally derived pseudostratified ciliated (or Schneiderian) epithelium that lines the sinonasal tract and nasal cavity. We describe a rare case of a sinonasal papilloma confined to the lacrimal duct and sac in a 45-year-old man who presented with 10 months of right-sided epiphora and swelling of his medial right lower eyelid. He was found to have a 10 × 10 mm area of distension near the right lacrimal sac and underwent an external dacryocystorhinostomy. A computed tomography scan revealed an expansile mass in the right nasolacrimal duct area. Right endoscopic sinus surgery and endoscopic dacryocystorhinostomy were performed. This case illustrates the importance of including sinonasal papilloma, a benign but invasive neoplasm, in the differential diagnosis of nasolacrimal duct obstruction.

KEYWORDS Inverted papilloma; lacrimal sac; nasolacrimal duct; Schneiderian papilloma; sinonasal papilloma

The ectodermally derived ciliated respiratory mucosa that lines the sinonasal tract, called the Schneiderian membrane, gives rise to three morphologically distinct papillomas. These are referred to individually as exophytic, inverted, and oncocytic papillomas or collectively as sinonasal papillomas.¹ Sinonasal papillomas are uncommon, representing only 0.4% to 4.7% of all sinonasal tumors.² Inverted and oncocytic types are characterized by a potentially invasive nature and relatively high recurrence rates.^{3,4} Recurrence rates quoted in the literature range from 2% to 80%.⁵ This rate of recurrence depends mostly on the extent of disease, completeness of resection, tumor location, and biological variability of the tumor.⁶

CASE PRESENTATION

A 45-year-old man presented with right-sided epiphora and swelling of his right medial lower eyelid area beginning in August 2013. He was initially evaluated by oculoplastic surgery and underwent an attempted external dacryocystorhinostomy (DCR) in February 2014. He underwent dilation of his proximal lacrimal system and was subsequently found to have abnormal tissue within the lacrimal sac. A biopsy of this tissue revealed sinonasal (Schneiderian) papilloma with inverted features and focal associated verrucoid squamous epithelial hyperplasia. A computed tomography scan revealed

an expansile mass in the right nasolacrimal duct area as well as bilateral mild pan-sinus disease and a left septal deviation (*Figure 1*).

He was referred to otolaryngology and underwent a right endoscopic DCR, as well as a right frontal sinusotomy, total ethmoidectomy, maxillary antrostomy, and sphenoid sinusotomy for chronic rhinosinusitis in March 2014. The patient was found to have polyps in his nasal cavity, along with a papilloma fungating through the previous external DCR into the middle meatus. The lacrimal sac and tissue surrounding the DCR site were completely removed via a combined open and endoscopic approach (*Figure 2*). Pathology revealed areas of sinonasal papilloma, mostly exophytic, and isolated to the lacrimal sac, with acute inflammation and fibrosis without any evidence of dysplasia or malignancy. The patient did not have postoperative complications and currently has no evidence of recurrence. His last follow-up appointment was in July 2014.

DISCUSSION

Almost 300 distinct lacrimal sac tumors have been reported, most being primary epithelial tumors (73%), of which 75% are malignant.⁷ Most lacrimal sac tumors present insidiously with symptoms of associated chronic dacryostenosis or dacryocystitis, but up to 43% are found inadvertently

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Figure 1. Preoperative computed tomography scan of the right lacrimal sac sinonasal papilloma (white arrow).

Figure 2. (a) Right middle meatus after external dacryocystorhinostomy with verrucous lesion (white arrow). The middle turbinate is seen on the right, and a Crawford tube is seen exiting the nasolacrimal duct. (b) Excision of the lacrimal sac (white arrow) through the right middle meatus endoscopic dacryocystorhinostomy. (c) Right lacrimal sac fully excised. (d) Four-month postoperative healing of the right nasal cavity.

at the time of DCR, as occurred in this case.⁸ Primary sinonasal papillomas of the lacrimal sac are exceedingly rare, with only a handful of case reports in the literature.^{7,9–14} Histologically, they can be grouped into three subtypes, each of which shares the distinct characteristics of epithelial thickening, scattered mucous cells, and intraepithelial neutrophils with microabscesses.¹ Specific histological features unique to the three subtypes include downward growth of surface epithelium with an intact basement membrane (inverted), branching fronds with a fibrovascular core (exophytic), and

inverted and exophytic growth with pseudostratified columnar epithelial cells (oncocyctic).¹ Interestingly, this papilloma had evidence of both exophytic and inverted features on separate biopsies. It has been suggested that ectopic migration of the Schneiderian membrane during embryogenesis could account for these aberrant papillomas in sites contiguous with the sinonasal tract.¹⁵ An association with human papillomavirus has been established for the exophytic subtype (low-risk human papillomavirus types 6 and 11), with an unclear association for inverted and no association with

oncocytic.^{1,16} The rate of malignant transformation is morphology specific, with inverted and oncocytic having the highest rates (5%–15% and 4%–17%, respectively), and the exophytic having a rate close to 0.¹

Although rare, sinonasal papillomas can arise de novo from the lacrimal sac and other areas discrete from the sinonasal tract. Due to potentially high recurrence rates (up to 80%), close follow-up with these patients is necessary.⁵

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