

A Case of Recurrent Schneiderian Papilloma of the Lacrimal Sac Invading the Nasal Cavity

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A 44-year-old man presented with a history of chronic epiphora, discharge from the right eye, and a palpable mass in the medial canthal area. Irrigation of the lacrimal system revealed bloody discharge. Orbital magnetic resonance imaging (MRI) showed a well-defined heterogeneous enhanced mass filling the lacrimal sac and upper nasolacrimal duct (NLD). A wide excision and surgical biopsy were performed. Histopathology showed the tumor to be an exophytic Schneiderian papilloma with moderate to severe dysplasia. Three months later, the mass was found to be invading the nasal cavity through the NLD. Endoscopic histopathological evaluation confirmed that it was identical to the originally identified papilloma.

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Key Words: Lacrimal sac, Recurrent exophytic papilloma, Schneiderian papilloma

The Schneiderian papilloma originates from the mucosal epithelial lining of the nasal cavities, the paranasal sinus, the nasolacrimal duct (NLD) and the lacrimal sac.¹ The majority of cases develop in the lateral nasal wall or paranasal sinus.² A lacrimal sac is a rare location for the Schneiderian papilloma.³ We report the case of a recurrent Schneiderian papilloma of the lacrimal sac that was removed surgically, but eventually invaded the nasal cavity through the NLD.

Case Report

Clinical History

A 44-year-old male patient visited the outpatient department with complaints of a chronic epiphora, and discharge from the right eye. A palpable mass had been noted in the right medial canthal area 10 days prior to the visit.

On ophthalmological examination, the visual acuity was 1.0 in both eyes without correction and there was no relative afferent pupillary defect. A firm and tender mass was palpated above the right medial canthal ligament area. Bloodstained discharge was noted after right nasolacrimal duct irrigation. Examination of the other eye and nasal cavity was normal.

Dacryocystography showed evidence of a soft tissue mass

as an uneven mottled density of the contrast media in the right lacrimal sac (Fig. 1). Magnetic resonance imaging (MRI) of the orbit revealed a well-defined heterogeneous enhancing mass lesion, approximately 18×9 mm, filling the lacrimal sac and extending to the upper NLD. The mass extended anteriorly to the right facial soft tissues with no any evidence of bony structure destruction (Fig. 2).

Under local anesthesia, excision of a mass was performed by dacryocystectomy using a standard dacryocystorhinostomy skin incision. When the lacrimal sac was removed, the mass was found within lacrimal sac, extending into the NLD. Adhesions were adjacent to the sac and obstruction of the NLD occurred at the junction with the bony canal due to tumor extension to the excision margins. We removed as much of the tumor as possible, but could not excise a portion that was located in the bony canal duct. Surgical biopsy demonstrated an exophytic Schneiderian papilloma with focal moderate to severe dysplasia on the histopathology.

Three months after the operation, the orbital MRI showed a recurrent mass in the right lacrimal sac invading the nasal

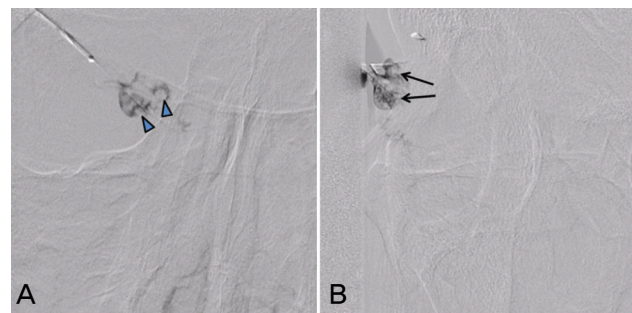


Fig. 1. Dacryocystography shows an uneven, mottled density of contrast media, evidence of a soft tissue mass (A: blue arrow, B: black arrow) at the right lacrimal sac.

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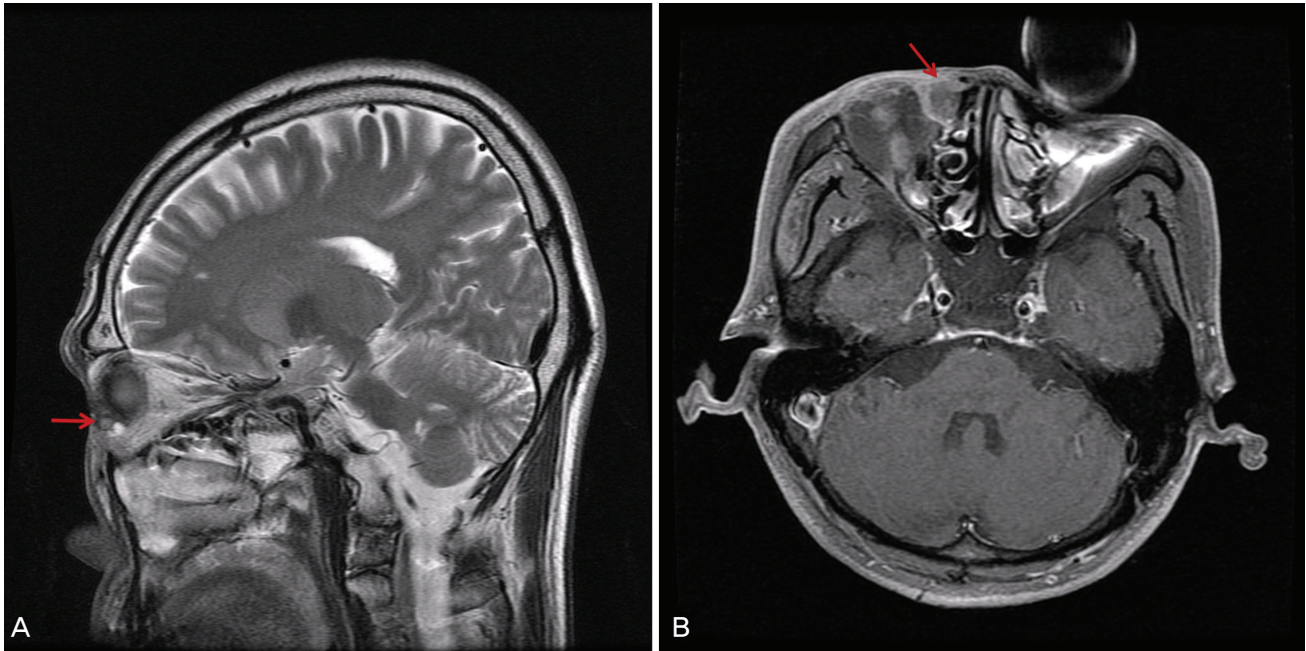


Fig. 2. Orbital MRI before surgery. (A) A well-defined mass lesion (arrow) at the right lacrimal sac (T2WI, sagittal view, fat saturation enhancement). (B) A heterogeneous enhancement in the lesion that extends anteriorly adjacent to the right facial soft tissues without evidence of bony structure destruction (T1WI, axial view, Gadolinium enhancement).

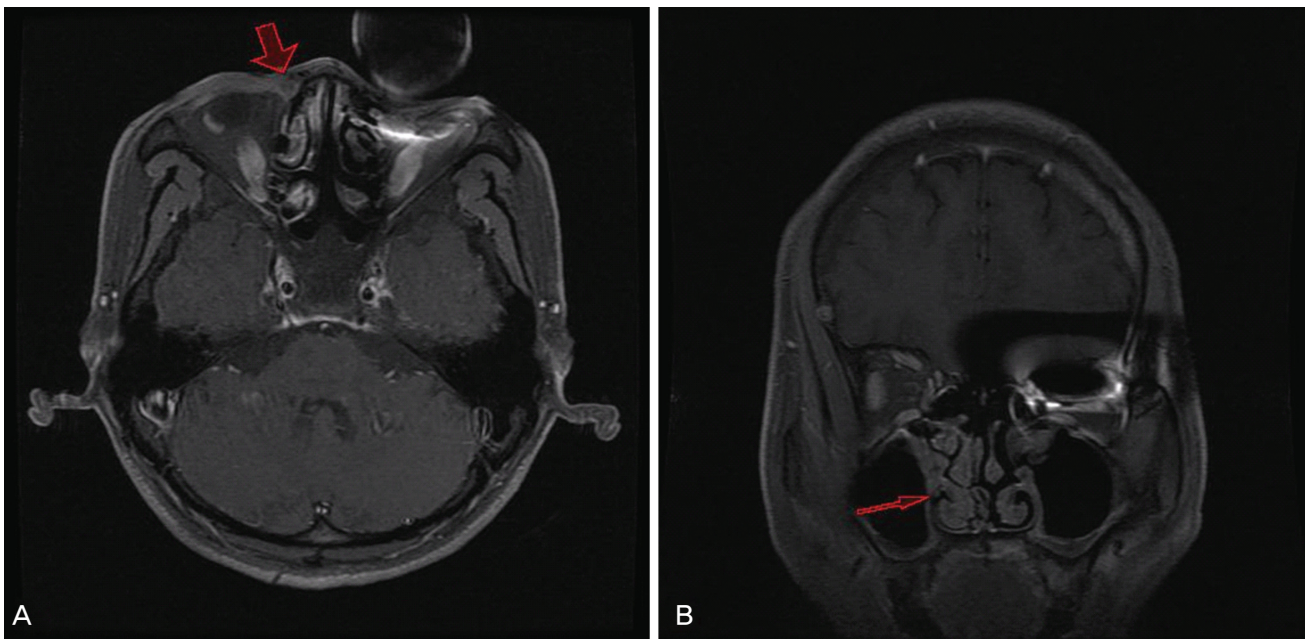


Fig. 3. Orbital MRI, 3 months after surgery. (A) A small enhancing recurrent lesion (thick arrow) is seen in the right nasolacrimal sac (T1WI, axial view, Gadolinium enhancement). (B) A recurrent mass (thin arrow) invading the nasal cavity via the inferior meatus (T1WI, coronal view, Gadolinium enhancement).

cavity via the inferior meatus (Fig. 3). After referral to the Department of Otorhinolaryngology, the patient underwent endoscopic biopsy of the nasal cavity lateral wall through a nasal approach. An identical tumor was confirmed, and endoscopic medial maxillectomy and dacryocystorhinostomy has been planned for its removal.

Histopathology findings

Histological examination revealed a fungiform mass with projecting finger-like proliferation of the epithelium. The tumor revealed an exophytic growth pattern with branching fibrovascular stalks covered by an epithelial cell layer (Fig. 4). According to the cytological features, the lesion was a transi-

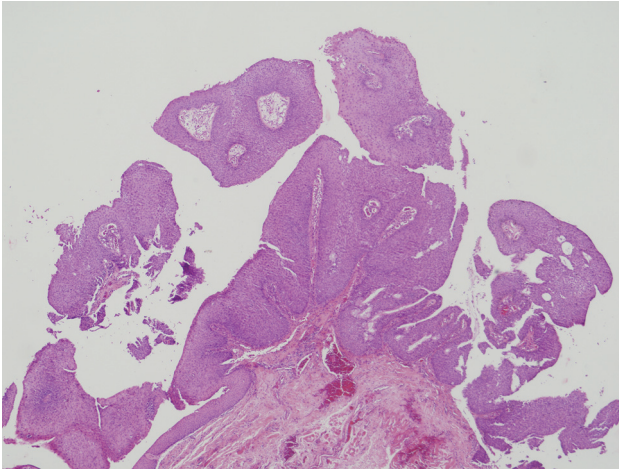


Fig. 4. Histological examination reveals a fungiform mass with projecting finger-like proliferation of the epithelium. The tumor has an exophytic growth pattern with branching fibrovascular stalks covered by an epithelial layer (Hematoxylin-eosin stain, $\times 40$).

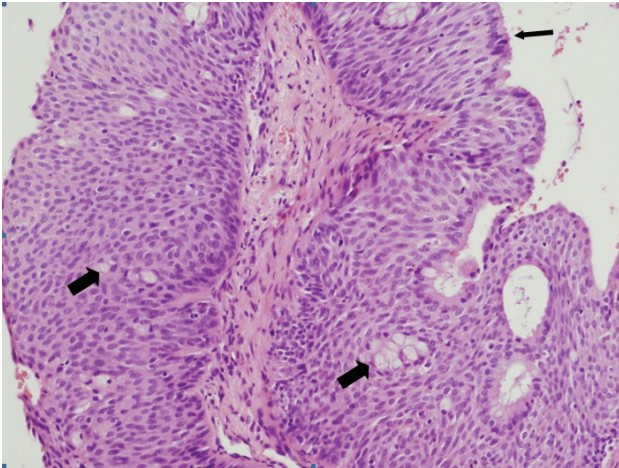


Fig. 5. Transitional-type epithelial lining containing goblet cells (thick arrow). Occasionally, a single layer of columnar ciliated respiratory epithelial cells (thin arrow) is seen to line the surface of the hyperplastic transitional cells (Hematoxylin-eosin stain, $\times 200$).

tional cell papilloma. The epithelial lining was a transitional type and contained goblet cells. Occasionally, a single layer of columnar ciliated respiratory epithelial cells lined the surface of the hyperplastic transitional cells (Fig. 5). The papilloma had a partial severe dysplasia (Fig. 6). Pathologic examination confirmed the diagnosis of an exophytic Schneiderian papilloma with moderate to severe dysplasia.

Discussion

The nasolacrimal drainage system shares its epithelial origin with that of the sinonasal tract. The mucosal epithelium that lines the nasal cavity, paranasal sinuses, and lacrimal apparatus is unique in its embryogenesis, in that it is ectodermal

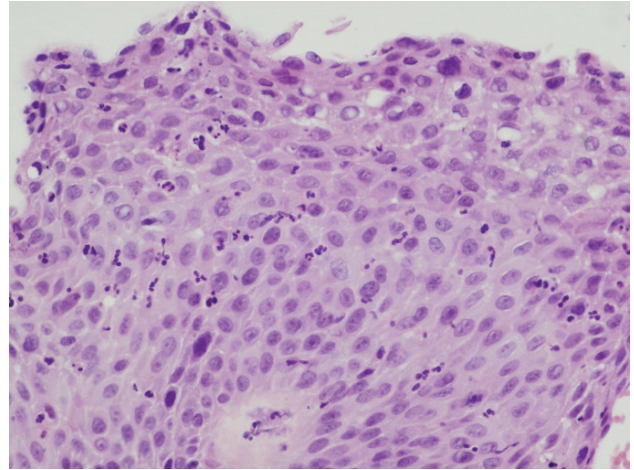


Fig. 6. A partial severe dysplasia in the papilloma (Hematoxylin-eosin stain $\times 600$).

in origin.^{1,2} A common synonym for this ectodermally derived mucosa and papilloma is the “Schneiderian mucosa” or “Schneiderian papilloma”.⁴

The Schneiderian papilloma is an uncommon neoplasm. The lateral nasal wall is the most common site of origin.² The lacrimal system is one of the most unusual primary locations,³ having been reported in Korea in only two cases reported.^{5,6} This case is a recurrent Schneiderian papilloma of the lacrimal sac that eventually invaded the nasal cavity through the nasolacrimal duct after mass excision of the lacrimal sac tumor. There is no prior report of recurrence and invasion of the nasolacrimal duct by an exophytic papilloma originating in the lacrimal sac.

Valenzuela et al.⁷ reported the clinical presentation of lacrimal drainage apparatus tumors in 37 patients. Symptoms included epiphora, a palpable mass, dacryocystitis, discharge, a visible lesion, bloody discharge or tears, epistaxis, and non-axial globe displacement. This patient had chronic epiphora and a small palpable mass at the nasal canthal area. Irrigation of the lacrimal system revealed bloody discharge. Bloodstained discharge from the puncta should always be regarded as a significant finding whether spontaneous, secondary to pressure on the sac, or after irrigation.⁸ When found, surgical intervention is usually recommended, because it is suggestive of a lacrimal sac tumor.

Radiographic examination of all masses that develop at the medial canthus is essential. Dacryocystography allows the identification of space-occupying tumors in the lacrimal sac. A characteristic dacryocystogram, as described by Veirs,⁹ shows a distended sac shadow, uneven or mottled density of the contrast media, and patency with residual media present 30 minutes after injection. Orbit and sinus CT scan, and MRI provide the most useful information about the extent of tumors and their relationship with surrounding bony structures and soft tissues.¹⁰ In this case, dacryocystography showed uneven contrast media at the right lacrimal sac, and evidence of a soft tissue mass. MRI of the orbit revealed a hetero-

geneous enhancing mass, approximately 18×9 mm, in the right lacrimal sac and upper nasolacrimal duct. The mass lesion extended anteriorly to the facial soft tissue, but no bony destruction was observed.

Histological examination of excised tissue is essential to confirm diagnosis. As presented by Ryan and Font,³ lacrimal papillomas can be subdivided by growth pattern into exophytic, inverted, or mixed in architecture. Another classification, reported by Ashton et al.¹¹ defines transitional cell papillomas, intermediate types, and transitional cell carcinomas. In this case, the Schneiderian papilloma had an exophytic growth pattern. The lesion was a transitional cell papilloma, with stratified columnar epithelium and scattered goblet cells, with partial severe dysplasia.

The possibility of malignant change must always be considered. In the cases studied by Ryan and Font,³ 7 out of the 12 inverted or mixed-type papillomas presented either as carcinomatous lesions or progressed to invasive carcinoma; none of six exophytic papillomas developed into carcinomas, however. This patient had an exophytic Schneiderian papilloma with moderate to severe dysplasia, which may precede the development of a malignancy. An exophytic type papilloma does not rule out malignant potential.

Recurrence is a major clinical problem in the treatment of Schneiderian papillomas. Hyams,¹² reported on papillomas of the sinonasal tract, and found a recurrence rate of 40-60% for all papillomas, regardless of type. All recurrences occurred at essentially the same anatomical site as the previous surgically removed tumor. We concluded that this case was a recurrent Schneiderian papilloma caused by incomplete removal of the lacrimal sac papilloma.

The therapeutic approach to this type of tumor has to be planned carefully. Sham et al.¹³ recommended that guidelines for the surgical treatment of Schneiderian papillomas include the following: 1) the papilloma should not be managed as a completely benign lesion; 2) the best opportunity for successful control of the papilloma is the first surgical procedure and; 3)

the more open the approach, the better the accessibility, the more complete the resection, and the lower the chance of recurrence.

In conclusion, a Schneiderian papilloma of the lacrimal sac is a locally aggressive benign epithelial tumor with malignant potential. Incomplete tumor excision causes higher recurrence rates with spreading into the other nasolacrimal system. Therefore, a Schneiderian papilloma of the lacrimal sac must be accurately diagnosed by histopathological examination, and completely removed by surgical excision.

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