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Case Report

Inverted Papilloma as a Rare Differential Diagnosis of a Medial Canthal Swelling

Deivanraj Palanimuthu[1]*, Abdul Razak Ahmad[2], Mohamad Azizul Fitri Khalid[3], Hasmah Hashim[4] and Ahmad Izani Mohd Safian[1]

[1]Department of Otorhinolaryngology-Head and Neck Surgery, Hospital Putrajaya, Jalan P9, Presint 7, 62250 Putrajaya, Wilayah Persekutuan Putrajaya, Malaysia

[2]Department of Otorhinolaryngology-Head and Neck Surgery, Hospital Melaka, Jalan Mufti Haji Khalil, 75400 Melaka, Malaysia

[3]Department of Otorhinolaryngology-Head and Neck Surgery, Hospital Sultanah Bahiyah, Km 6, Jalan Langgar, 05460 Alor Setar, Kedah, Malaysia

[4]Department of Pathology, Hospital Melaka, Jalan Mufti Haji Khalil, 75400 Melaka, Malaysia

***Corresponding Author:** Deivanraj Palanimuthu, Department of Otorhinolaryngology-Head and Neck Surgery, Hospital Putrajaya, Jalan P9, Presint 7, 62250 Putrajaya, Wilayah Persekutuan Putrajaya, Malaysia.

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Abstract

Inverted papilloma (IP) originating primarily in the nasolacrimal duct (NLD) is considered very rare. Most of IP cases arising from the ethmoid region, lateral wall of nasal fossa and maxillary sinus. It is an aggressive neoplasm characterized by high recurrence rates and prone towards malignant transformation, albeit it's a benign neoplasm histologically. IP of NLD may present as non-progressive medial canthal swelling, masquerading as more common conditions such as dacryocystitis, or other primary acquired NLD obstruction. Here, we report a case of a 70-year-old gentleman, presented with a swelling in his right medial canthal region for a 3-month duration, associated with tender on palpation and unilateral epiphora. Nasoendoscopy showed a granulation tissue arising from the right Hasner valve and biopsy revealed IP features. Radiological imaging revealed an enhancing soft tissue lesion arising from the right NLD associated with a mild expansion of the duct but no bony erosion seen. He underwent an open right medial maxillectomy with right dacryocystectomy via lateral rhinotomy approach, and the patient made an uneventful recovery with normal visual acuity and intact eye movements.

Keywords: Inverted Papilloma; Nasolacrimal Duct; Medial Canthal Swelling; Open Right Medial Maxillectomy; Case Report

Introduction

Inverted papilloma (IP) is a benign neoplasm yet behave almost similar to what malignant neoplasm is. It can be locally invasive and have a tendency to recur following surgical removal. A small yet crucial proportion of cases may undergo malignant transformation [1]. These tumours are commonly originating from the nasal cavity and paranasal sinuses. Hence, it is extremely rare to find an IP originating primarily from the lacrimal system, though more infrequently, the nasolacrimal duct (NLD) involvement may be secondarily due to the extension from the paranasal sinuses to the orbit [2]. To date, there are fewer than 20 cases reported in the literature [3].

Case Presentation

A 70-year-old gentleman, with underlying hypertension, presented with a 3-month history of swelling over the right medial canthal region associated with unilateral epiphora and tender on palpation of the swelling. Initially, he was treated as dacryocystitis by the ophthalmology team with 2 weeks course of antibiotic. He denied any rhinosinusitis symptoms or visual disturbance. There were no loss of appetite, loss of weight, night sweats, prolonged cough or any familial case of malignancy. In view of the persisted symptoms, he was referred to an otorhinolaryngologist for nasal endoscopy to rule out nasal pathology causing the NLD obstruction.

On examination, there was a right medial canthal swelling measuring 2 x 2 cm, firm in consistency, slightly erythematous and mild tender (Figure 1). Visual acuity and ocular movements were normal. There was an increased tear lake with a delayed dye disappearance test and a hard stop on syringing suggestive of a postsaccal obstruction. Nasoendoscopy revealed a small granulation tissue over the right Hasner valve with bulging over the lateral wall of the nasal cavity, anterior to the middle turbinate with normal nasal mucosa which most likely caused the obstruction. Otherwise, no hypertrophy of inferior turbinates, no mucopus discharge seen, no deviated nasal septum and osteomeatal complexes were pattern. Intraoral, ear and neck examinations were unremarkable.

Figure 1: Clinical photograph showing a firm, medial canthal mass measuring 2 x 2 cm, with slightly erythematous overlying skin (black arrow).

Biopsy was taken over the right Hasner valve and revealed features of inverted papilloma characterised by an inverted growth pattern of epithelium with ramifications into the underlying stroma, which was most likely originated primarily from NLD. Contrast-enhanced computed tomography (CECT) scan of paranasal sinus demonstrated an enhancing soft tissue lesion measuring 1.9 x 2.0 x 2.6 cm seen at the medial aspect of the right orbit within the extraconal space appeared to arise from the right NLD (Figure 2). There was a mild expansion of the NLD as well. However, no significant bony erosion seen or any suspicious lesion except for the hyperdense bony lesion of the right anterior ethmoid sinus wall measuring 0.7 x 0.7cm in keeping with benign osteoma. There were no other polypoidal lesion or features of sinusitis seen within the paranasal sinuses.

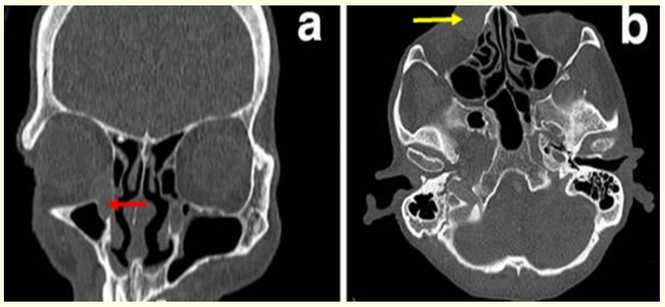

Figure 2: The CECT paranasal sinus in coronal (2a) and axial (2b) views. 2a) The red arrow showing homogenous mass at medial aspect of the right orbit appeared arising from the right NLD measuring 1.9 x 2.0 x 2.6cm. 2b) The yellow arrow showing homogenous mass at medial aspect of the right orbit displacing the right orbit laterally and posteriorly just anterior to anterior ethmoid sinus wall.

The patient underwent open right medial maxillectomy with right dacryocystorhinotomy (DCR) via lateral rhinotomy approach. Intra-operatively, right lacrimal sac with 2 x 2 cm mass were excised and removed together with right lacrimal bone (dacryocystectomy), right inferior turbinate and right medial maxillectomy. A Crawford tube were passed through the remaining superior and inferior canaliculi to replace and stent the lacrimal system.

All 4 samples were sent for histopathological examination. Two out of 4; right lacrimal sac and right medial maxillectomy revealed features of inverted papilloma, characterised by partially capsulated papillomatous tumour composed of inward growth of mildly dysplastic squamous epithelium with delicate fibrovascular stroma. There was a loss of polarity and few mitoses seen. The underlying stroma was mildly edematous with focal areas of haemorrhage. Ki-67 showed an increased proliferative index involving the lower third of the epithelial thickness. No stroma of bone invasion seen and no evidence of malignancy noted. All surgical margins were free from the tumour (>10 mm). The other 2 samples; right lacrimal bone and right inferior turbinate revealed no tumour tissue seen. Hence, the diagnosis of IP with mild dysplasia originating from the right nasolacrimal system was confirmed (Figure 3).

The patient was reviewed regularly post-operatively. The wound was well-healed with no orbital complication detected. To date, he was found to be well and no evidence of recurrence seen after almost one year post-operation. Throughout follow-up, his eye vision was intact and nasoendoscopy revealed no significant finding.

Discussion

Inverted papilloma (IP) or formerly known as Schneiderian papilloma was first described by Ward and Billroth in 1854. It was then changed the name to IP in 1938, by Ringertz based on the inverted epithelium appearance from histopathological finding [4]. The lesion is usually unilateral, with a male to female ratio of 3:1 and affected in the sixth to seven decades of life, consistent with this patient who is a 70-year-old gentleman.

Despite being benign in origin, IP raised such a great concern among clinician and surgeons for few distinctive reasons. One of the most distinctive features is, the IP may undergo malignant transformation into squamous cell carcinoma at the rate of 5-21% [5]. It is also known for its aggressiveness and invasiveness as it may cause an adjacent bony destruction without malignant transformation.

The most common sites of IP are the lateral nasal wall near the middle meatus, ethmoid sinuses as well as maxillary sinuses. There are less than 20 cases reported in the literature by far, similar to this case whereby the IP is originating primarily in NLD instead. Furthermore, an extension of IP from paranasal sinuses or sinonasal cavity to the orbit via NLD is equally uncommon.

Although the actual pathogenesis of IP originating from NLD is still unclear, however, there is literature suggested that an ectopic migration of the Schneiderian membrane during embryonic life is responsible for the uncommon site of IP originates [6]. Patient with IP in the NLD usually presented with epiphora and medial canthal mass at the initial stage at which clinicians may confuse with dacryocystitis or other types of infection involving the lacrimal system. Similarly, in our case, the patient was initially treated with antibiotics for acute dacryocystitis by the ophthalmology team. However, upon completion of the antibiotic, reassessment had been made and showed a negative sign of remission, hence the case was referred to the otorhinolaryngologist for further management.

In the initial stages, IP of NLD may present in a similar manner with other primary acquired NLD obstruction or chronic dacryocystitis. If left untreated, IP in the NLD may invade the orbit and leads to blindness or oculomotor paralysis. Thus, when a patient presented with medial canthal swelling with unilateral epiphora, excluding an obstructing neoplastic mass is extremely crucial. Nasoendoscopy and CECT scan may reveal neoplasm of unsuspected benign symptoms of such medial canthal swelling with epiphora.

Nasoendoscopy and CECT scan remain the important measures to evaluate the lesion and to exclude other differential diagnoses of a persistent NLD obstruction. Besides, it also helps for pre-treatment staging of IP and to determine surgical strategies for both the patients and the surgeons.

As the IP has the ability to locally destroyed the adjacent bony structures, it is not recommended to delay the crucial investigations and surgical treatment to the patient with suspected IP. Thus, this patient has been offered for open medial maxillectomy and DCR via lateral rhinotomy approach, albeit the procedure is considered traditional compared to the recent endoscopic approach.

There were few literatures had mentioned regarding combined endoscopic-assisted with purely endoscopic approach to have a similar recurrence rate of IP post-operatively if not better with the more invasive external approach [7], however, we had opted the latter to be more suitable with the case. In view of patient age and the high potential of malignant transformation, open medial maxillectomy with DCR via lateral rhinotomy were our best option for tumour control. Sauter., *et al*. [8] recommend en bloc resection of any potential sites of tumour extension, such as the lateral nasal wall, ethmoid labyrinth and medial part of the maxilla, to remain as gold standard as they were before.

In this case, we had removed 4 important structures (right lacrimal sac, right lacrimal bone, right NLD and medial portion of maxillary wall) which were affected with the tumour invasion or as potential sites for recurrence or tumour extension. In comparison with usual medial maxillectomy, in which the lacrimal system is not fully resected, we had done a combination of open medial maxillectomy with DCR via lateral rhinotomy for the purpose mentioned. Although this open approach comes with several complications mostly being massive bleeding from important adjacent structures, an experienced surgeon with a great team should encounter these predicted complications, as there are still tumours that may lend themselves to the traditional external medial maxillectomy [9]. IP of NLD is being one of the best examples to address via this method, besides other malignant tumours involving the lateral nasal wall, medial wall of the maxillary sinus and the adjacent ethmoid cells.

Adjuvant radiotherapy may be considered in the case of early recurrence, incomplete tumour removal with positive margins and tumour with associated malignancy [10]. However, due to the com-

pleteness of tumour resections and negative margins (>10 mm) in this case, the patient was regularly follow-up for recurrence surveillance and to date, no sign of recurrence noted in nearly one year duration post-operatively.

Conclusion

Although primary IP of NLD is extremely rare, one must not underestimate a simple case of dacryocystitis who is not responded well to the antibiotics. Nasoendoscopy and CECT scan are both crucial to exclude such invasive tumour of NLD. We recommend a complete en bloc resection of the lacrimal system together with the adjacent potential structure of tumour extension with clear margins to avoid the need for adjuvant therapy.

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