

Case Report

A Rare Otological Presentation of Squamous Papilloma

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ABSTRACT

Introduction

Squamous papillomas (SPs) are benign epithelial neoplastic lesions commonly found in skin, oral mucosa, upper aerodigestive tract and genitals. Involvement of external auditory canal (EAC) by SP is extremely rare and whatever scarce literature is available is in the form of case reports and small case series. The exact incidence of this entity is still unknown. Consequently, its etiology, histopathological features, differential diagnoses and optimal management merit a better understanding by the Otolaryngologists.

Case Report

We present a case of SP of EAC in a 15 year old girl hailing from north-eastern India who presented to us with an acutely painful mass in the EAC. Contrast Enhanced Computed Tomography remained the gold standard and imaging study of choice for evaluation. However, the diagnosis was clinched only by histopathology. We also discuss our strategy of management along with a brief review of literature.

Conclusion

Squamous papilloma has rarely been reported to affect the EAC. Human papilloma virus remains the most consistent etiological agent. Advanced imaging like contrast enhanced high resolution computed tomogram remains gold standard to evaluate any mass seen in EAC. Complete excision with cold steel technique or coblation with good post-operative care and follow up is the standard of care.

<u>Keywords</u>

Squamous Papilloma; External Auditory Canal; Human Papilloma Virus; Koilocytosis

quamous papillomas (SPs) are benign neoplasms of epithelial origin affecting skin, oral mucosa, upper aerodigestive tract and genital organs. However, SPs of the external auditory canal (EAC) are rare in occurrence. It is sporadic in nature with equal distribution amongst all age groups withoutany gender predilection.¹ Its exact incidence affecting the ear is still unknown owing to its rarity.

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It is attributed to low risk human papilloma virus (HPV) usually type 6 or 11. The portal of entry is unestablished but most acceptable hypotheses propose use of unsterilized instruments for traditional shaving inside ear (especially in south China population) or vertical transmission during parturition as the initial causative insult.²

There are various cutaneous lesions which may mimic SPs in gross appearance like verruca vulgaris (common warts), canal cholesteatoma, carcinoma in situ and invasive squamous cell carcinoma.² Hence its diagnosis mandates histopathological examination which is characteristic. The features which are pathognomonic of HPV infection are koilocytosis with cytoplasmic clearing and micronuclear hyperchromatism.²

Here we present a rare case of SP involving the EAC skin. The case report aims to highlight the clinical presentation, histopathological features and management from the perspective of an otologist.

Case Report

Fifteen years old girl presented at a peripheral hospital in north-eastern India with complaint of pain in left ear associated with blocked sensation of the ear for last 5 days. She denied otorrhoea, hearing loss, tinnitus and vertigo. There was no history of previous ear surgery or ear trauma. Otoscopy revealed a solitary, pinkish, fleshy, minimally tender mass filling the entire EAC which extended into the concha, completely obscuring the EAC lumen (Figure 1).



Fig. 1. A solitary pinkish fleshy mass completely filling the left external auditory canal

On probing it appeared to arise from posterior EAC wall with non-visualization of the tympanic membrane. Contralateral ear was normal. There were no other similar cutaneous/orogenital lesions found in the patient.

Patient was initially managed conservatively with medicated aural packing and ototopic medication. However, upon no clinical improvement, imaging in the form of Contrast Enhanced High Resolution Computed Tomogram (CE-HRCT) of Temporal bone with 0.5 mm sections was requisitioned. It revealed a fairly circumscribed soft tissue attenuated mass measuring 1.1×0.6 centimeter in inner $1/3^{rd}$ of left EAC with no significant enhancement (Figure 2).

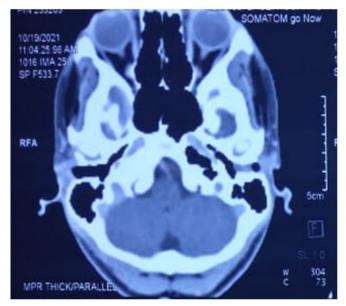


Fig. 2. CE-HRCT Temporal bone with 0.5 mm sections revealed a fairly circumscribed soft tissue attenuated mass measuring 1.1 x 0.6 centimeter from inner 1/3rd of left external ear canal with no significant enhancement.

Patient was counselled for excision biopsy of the lesion and consent for same was obtained. The lesion was excised in toto under local anesthesia using an operating microscope and sent for histopathological examination (HPE). The post-operative period was uneventful. HPE revealed stratified squamous epithelium with features of acanthosis, hyperkeratosis, marked koilocytic cells with dark nuclei, clear cytoplasm and no cellular atypia (Figures 3a and 3b). The patient was kepton regular follow up till 1 year. The surgical sitehealed well without any recurrence or stenosis of the EAC.

Photomicrographs of Haematoxylin and Eosin stain, 40 x (Figure 3a) &100 x (Figure 3b) magnification showing stratified squamous epithelium with features of acanthosis, hyperkeratosis, marked koilocytic cells with dark nuclei and clear cytoplasm and no cellular atypia.

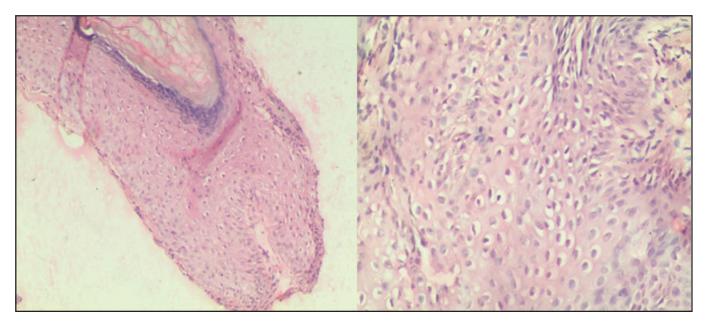


Fig. 3a. & b. Photomicrographs of Haematoxylin and Eosin stain, 40 X (Fig 3a) &100 X (Fig 3b) magnification showing stratified squamous epithelium with features of acanthosis, hyperkeratosis, marked koilocytic cells with dark nuclei and clear cytoplasm and no cellular atypia.

Discussion

Papillomata are benign epithelial tumors commonly caused by HPV which affect the epidermis and mucous membranes in the head and neck region. Although SPs are relatively common benign lesion of the aero-digestive tract, benign papillomatosis affecting the EAC is an atypical presentation with under recognized incidence.² SPs are rare in children younger than 5 years of age though it has been reported in a 3 year old toddler.³ In the present case report the patient was a teenage girl with no history of trauma to ear. Possibly the patient acquired the disease by vertical transmission as suggested in literature.

SPs of the EAC are similar to papillomas in other cutaneous sites and can become inflamed with mechanical irritation like scratching and subsequently undergo ulceration.⁴ Grossly SP of EAC vary greatly with both flat plaques and raised lesions of different sizes coexisting in a single patient and may arise from any site of either bony or cartilaginous EAC.⁵ Unlike the existing literature, in the present case report the papilloma was pinkish, fleshy and globular. In its natural course, SPs of EAC may remain solitary, grow at a slow pace and might cause mechanical obstruction to hearing pathway leading to conductive deafness. Rarely pressure necrosis of underlying bone or involvement of tympanic membrane has also been reported.⁶ In the present reported case, patient had presented with otalgia and was detected to have a mass in the EAC on otoscopy with conductive hearing loss.

The possibility of spontaneous resolution of SPs of EAC unlike cutaneous warts is still unclear but its malignant transformation to squamous cell carcinoma has been mentioned in literature and its association with HPV has also been established in a high proportion of cases. Still further genotyping of such a benign lesion to gauge its malignant potential is a matter of debate.⁷ In present reported case no genotyping was done because of non availability of genetic test in a peripheral hospital in north east India.

Several cutaneous lesions can mimic SPs in appearance like verruca vulgaris (common warts), carcinoma in situ and invasive squamous cell carcinoma. However, the definitive diagnosis of SP is clinched by

histopathological examination which shows characteristic multiple finger-like projections of benign squamous epithelium surrounding fibrovascular cores with hyperkeratosis and parakeratosis.² Investigations to

hyperkeratosis and parakeratosis.² Investigations to evaluate a growth in EAC should include radiological imaging in form of CE-HRCT Temporal Bone and histopathological evaluation. In the present case report the histopathological findings were consistent as reported in the existing literature.

Surgical removal of the lesion by either by cold steel or coblation stays to be the most efficacious method in the treatment of SPs of EAC. Many other methods described like cryosurgery, electrodessication with/ without curettage and carbon dioxide laser (5 watts at continuous pulse) are also fruitful. Topical treatment with agents like salicylic acid and cryotherapy has been shown to be effective in obtaining clearance of cutaneous warts. However, studies using Dinitrochlorobenzene, 5 Flurouracil, intralesional bleomycin, intralesional interferon and photodynamic therapy did not give any certain evidence of effectiveness for SPs of EAC and therefore these are not recommended as primary management modality.8 In the present case report the lesion was excised in toto under microscope avoiding circumferential incision with cauterization of base. No topical medicationswere used in the post-operative period.

The major post-surgical complication can be scarring and consequent EAC stenosis which can be prevented by avoiding circumferential incisions and ensuring meticulous post-operative care to promote uncomplicated healing and if required stenting of the EAC with silastic tube. The reported case had no post op stenosis of EAC upto 1 year after surgical excision of lesion.

Recurrence is rare after complete excision. However, recurrences can be easily managed with revision surgery being an easily accessible part of the ear. Treatment of recurrent SPs of EAC, middle ear and mastoid cavity with radiotherapy has shown promising outcomes but there is also risk of malignant transformation, hearing loss, EAC stenosis and vestibular, trigeminal and facial neuropathies.⁹ We suggest that since most SPs of EAC arise from a single locus in the EAC without satellite lesions, focal resection with cold steel technique and

regular post-operative follow up should be sufficient for treatment including recurrences, although rare. The patient did not have any recurrence upto 1 year post primary excision of the lesion.

Conclusion

Squamous papilloma is a common benign epithelial neoplasm occuring in the head and neck commonly seen in oral cavity and larynx. However, rarely it has been reported to affect the EAC. Human papilloma virus remains the most consistent etiological agent. Advanced imaging like contrast enhanced high resolution computed tomogram remains gold standard to evaluate any mass seen in the external auditory canal. Complete excision with cold steel technique or coblation with good postoperative care and follow up is the standard of care.

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