

The Challenging Diagnosis of Verrucous Carcinoma of the Auricle and Its Reconstruction: A Case Report

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Abstract

Background: Verrucous Carcinoma (VC) is a low-grade squamous cell carcinoma. Histopathology of VC lacks signs of epithelial dysplasia. Delayed diagnosis may lead to more radical and aesthetically unfavorable results.

Methods: In this report, we present are rare case of auricular VC, which was repeatedly misdiagnosed.

Results: Due to the histological structure of the auricle, only deep biopsies containing both the perichondrial layer and the auricular cartilage allow accurate diagnosis of VC. In tumors with verrucous morphology or recurrent benign lesions of the auricle, VC must be considered.

Conclusion: Biopsies containing the perichondrial layer as well as the auricular cartilage are mandatory to diagnose VC.

Keywords: Verrucous carcinoma; Squamous cell carcinoma; Auricle; Head and neck; Histopathology

Introduction

Six percent of all skin malignancies occur in the auricular region. Of these, Squamous Cell Carcinoma (SCC) accounts for 60% to 70%, followed by basal cell carcinoma and malignant melanoma accounting for 30% to 40% and 2% to 6%, respectively [1].

The highly-differentiated Verrucous Carcinoma (VC) is an exophytically growing, rare histological subtype of low-grade SCC first described by Ackermann in 1948 [2]. Most common localizations are the oral cavity and larynx [3]. Non-mucosal occurrence, such as in the auricular region, is extremely rare [4]. VC presents with a slow tumor growth as well as a low tendency of regional and distant metastatic spread. At the same time, the ability for local infiltration and the rates of local recurrence are high [5].

Established risk factors for the development of mucosal VC are tobacco use, alcohol consumption and poor oral hygiene [6]. Extensive sun exposure is another well studied risk factor of SCC of the skin [7]. VC predominantly affects elderly white men [8].

Histopathological features comprise hyperkeratosis, acanthosis and papillomatosis. Characteristically, tumor cells lack invasion of the basement membrane, but rather show deep pushing borders. Typical signs of epithelial dysplasia are minimal and rare [3,9]. Therefore, superficial biopsies may lead to benign differential diagnosis such as seborrheic keratosis, verruca vulgaris, papilloma or keratoacanthoma.

Surgical resection in primary and recurrent VC is recommended as first-line treatment. Radiation-based therapies are controversial, as there is evidence of malignant tumor transformation into higher grade SCC with subsequent worse outcome [8,10]. The five-year relative survival of mucosal VC reaches approximately 80% [8].

We herein present a rare case of a repeatedly misdiagnosed VC of the auricle, caused by too superficial excisions. In this case, we focus on the specific histopathological and clinical features of the auricular region that are important to properly diagnose VC and discuss reconstruction techniques in case of aesthetic alterations.

Case Presentation

A 79-year-old male presented to the outpatient clinic of the department of Otorhinolaryngology

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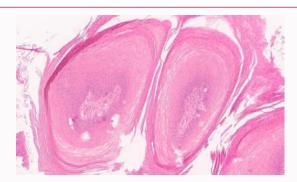


Figure 1: Hematoxylin-eosin stain of the superficial biopsy from auricular mass. Proliferative well differentiated epithelium. Epidermis with hyperkeratosis, papillomatosis, hypergranulosis and acanthosis. No signs of dysplasia or aplasia. Basement membrane intact. Pathological assessment: Compatible with seborrheic keratosis of hyperkeratotic type.



Figure 2: Macroscopic picture of the left auricle. Conchal cavum six months post reconstruction with full-thickness skin graft. Inferior border of skin graft shows an exophytically growing mass.

of the Technical University Munich with a 5-months history of a growing mass of the left auricle in March 2018. The patient reported of a previous excision of this mass 6 months ago at an otolaryngologic practice without histologic analysis of the excised tissue. At presentation he denied any otalgia, bleeding or history of head and neck skin malignancies. Past medical history revealed a left-sided mastoidectomy due to cholesteatoma seven decades ago. The patient had no history of tobacco and alcohol consumption or extensive sun exposure. On physical examination the patient's left conchal cavum showed an exophytic verrucous mass of approximately 2.5 cm \times 2 cm \times 0.5 cm, not adherent to the cartilage layer. The external auditory canal was not affected. Further palpation and sonography of the parotideal and cervical lymph nodes were negative for suspicious nodes. The facial nerve was intact.

Results of the superficial biopsy, taken from the auricular mass showed squamous papilloma without dysplasia. Circular radiofrequency-assisted excision of the mass was performed under local anesthesia. The deep surgical excision margin comprised only parts of the perichondrial layer as the tumor was not attached to the cartilage. Postoperative histopathological results revealed seborrheic keratosis of hyperkeratotic type without signs of malignancy (Figure 1).

Thus, the defect was reconstructed using a full-thickness skin graft from the lateral region of the ipsilateral neck. Six months after the reconstruction, a recurrent exophytic mass of the conchal cavum was detected, located in the inferior border of the skin graft (Figure 2).

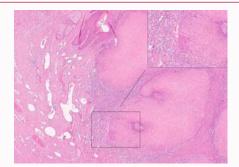


Figure 3: Hematoxylin-eosin stain of the resected tissue. Epidermis with hyperkeratosis, papillomatosis, hypergranulosis and acanthosis. Minimal signs of atypia. Rete ridges extending deep into dermis in pushing manner. Inlet showing epidermal invasion front with interruption of basement membrane and distinct stromal inflammation reaction. Pathological assessment: Overall, low-grade squamous cell carcinoma of verrucous type.



Figure 4: Macroscopic picture of the reconstructed left auricle. Conchal cavum and caudal conchal cymba six weeks post second reconstruction with full-thickness skin graft. Scar tissue after resection of cranial anthelical cartilage.

Re-excision was performed. Histopathology of the re-excised tumor repeatedly revealed hyperkeratotic seborrheic keratosis without dysplasia. At the next follow-up, three months later, recurrence of the mass was evident once again. Another biopsy (1 cm diameter) did not reveal malignant criteria either. Against the background of multiple recurrences of the mass, the patient was recommended a more radical resection of the tumor including the cartilage layer despite the risk of overtreatment with subsequent cosmetic alteration of the auricle. After receiving the patient's informed consent, deep radiofrequency-assisted en-bloc-resection of the mass with the underlying conchal cartilage was performed under local anesthesia. Synthetic wound dressing was used for temporary closure. Eventually, the histopathological findings showed low-grade squamous cell carcinoma of verrucous type (Figure 3).

Sonographic re-staging of the parotideal and cervical lymph nodes was clear. Re-resection was necessary to enlarge the perifocal surgical margins to 4 mm to 5 mm. Upon this, the lateral part of anthelical cartilage could be preserved. On final histopathological examination, all surgical margins were tumor free with minimal margins of at least 4 mm. Reconstructive options, especially in terms of full-thickness skin grafts vs. local skin flaps, were discussed with the patient. According to the patient's preference, a full-thickness skin graft from the lateral region of the contralateral neck was used for reconstruction under local anesthesia. The 5-weeks postoperative follow-up showed uneventful healing without evidence of graft necrosis or unpleasant scarring. Moreover, the aesthetic result was satisfying, showing a stable auricular cartilaginous rim despite

subtotal resection of the anthelix and antitragus (Figure 4).

Discussion

We herein present the case of a 79-years-old patient with a verrucous carcinoma of the auricle that was repeatedly misdiagnosed as a benign lesion although multiple biopsies and excisions were serially performed. While the histological structure of most skin regions consists of an epidermal, a dermal and a subcutaneous layer, the anterior auricle is lacking a subcutis with a soft tissue basal lamina [11]. Thus, the dermis of the anterior auricle is tightly adherent to the perichondrial layer, whereas the posterior auricle contains a thin subcutaneous layer [12]. These special features of the auricle's histology fundamentally complicate histopathological assessment. Further, the elastic cartilage is crucial for the auricle's flexibility and its aesthetic appearance. Therefore, surgery of the cartilaginous auricle needs to be performed particularly cautiously.

Nevertheless, sufficient biopsies of skin lesions are crucial for a detailed histopathological assessment to avoid delayed diagnosis due to false-negative histopathological results. Delayed diagnosis typically results in destruction of important anatomical structures and compromises the oncologic and aesthetic outcome. Choosing the appropriate biopsy technique is particularly challenging in presence of auricular VC as it lacks clear histopathological signs of dysplasia in superficial layers and can only be assessed if the invasion border can be detected at or within the underlying cartilage layer.

In the present case, several specimens (biopsies and excisions) including only the perichondrial layer but lacking the cartilage itself revealed repetitive false-negative benign entities such as papilloma and hyperkeratotic seborrheic keratosis. In clinical routine, excisions of auricular cartilage are typically avoided due to the following facts: on the one hand superficial biopsies are usually sufficient to diagnose basal cell carcinoma and poorly-differentiated squamous cell carcinoma and on the other hand the excision of auricular cartilage with wound healing by secondary intention can lead to aesthetic deformities. This can be caused by loss of tissue thickness and scarring with subsequent deformity of the auricle's outline. Additionally, VC of the auricle is extremely rare.

Hence, we are facing two different scenarios that should be avoided. First, a benign auricular skin lesion may be excised including the underlying cartilage with an unsatisfactory aesthetic result representing overtreatment. Secondly, an auricular malignant skin lesion may be excised too superficially to avoid an unfavorable cosmetic result, leading to a wrong histopathological diagnosis.

We recommend that in case of a clinically suspicious history of recurrent or rapidly growing masses of the auricle in combination with a benign histopathology after biopsy, at least an incisional wedge biopsy containing underlying cartilage should be ensured, despite the potential risk of aesthetic alterations.

In respect to the aesthetic subunits of the auricle as well as principles of reconstructive principles the following aspects should be kept in mind: Depending on the aesthetic subunit of the auricle, the size of the excised cartilage can vary in conjunction with the reconstructive options of the auricular defect. Within the conchal cavum and the auricular scapha, loss of tissue thickness will rather be invisible. Skin defects may either get closed by undermining the wound edges or by reconstruction with full-thickness skin grafts. Therefore, more extensive en-bloc resection of cartilage can be performed to increase the likelihood of a correct histopathological

result. In aesthetically more demanding regions such as the anthelix, fossa triangularis or antitragus, skin defects including the cartilage may require more advanced surgical techniques such as locoregional flaps or skin grafts combined with cartilage transplantation [13-15]. Thus, an incisional wedge biopsy should be preferred representing a reasonable compromise between good reconstructive options and histopathological assessability. More extensive cartilaginous defects of the anthelix affecting the stability of the auricle typically need more comprehensive reconstruction. Retroauricular flaps such as the pulltrough island flap can be used in these situations. Depending on the size of the defect, these can lead to decreased auricular stability.

In the present case, the mass was originally located in the conchal cavum. Consequently, an en-bloc excision of the underlying conchal cartilage was possible to finally diagnose VC. The presence of remaining tumor cells in the surgical margin led to resection of medial parts of the antitragus as well as medial parts of the anthelical rim. History of ipsilateral mastoidectomy with including retroauricular tissue loss limited the opportunity for retroauricular local flap reconstruction. Moreover, the patient preferred a simple reconstruction with a full-thickness skin graft again. The chosen reconstructive option resulted in sufficient stability of the auricle with only a slightly changed auricular contour.

In case of recurrent exophytic tumor growth of the auricle, VC must be considered. Due to the fact that VC lacks typical signs of epithelial dysplasia, histopathological diagnosis of VC is challenging and often requires excision of the auricular cartilage. Depending on the size and auricular region of excised cartilage, various surgical options are available to achieve an aesthetically satisfying reconstruction.

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