Verrucous Carcinoma of the Lips: Diagnostic Challenges and Surgical Management (Review of Literature and Case Report)

Case Report

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ABSTRACT

IIntroduction: Verrucous carcinoma, often known as Ackerman's tumor, is a rare variant of squamous cell carcinoma characterized by slow growth and minimal metastatic potential despite its local aggressiveness. Diagnosis hinges upon clinical, macroscopic, and microscopic criteria, complicated by its benign histological appearance.

Observation:A 70-year-old woman with a history of prolonged sun exposure presented with leukokeratoses involving the vermilion borders of both lips and the left labial commissure, accompanied by infiltrative lesions showing verrucous surfaces over a 3-year period. Initial histological examinations suggested papilloma altered by inflammation. Dermatological consultation and polarized dermoscopy revealed hairpin vessel-like structures, bright white features, and yellowish crusts. Subsequent guided biopsies confirmed extensive verrucous carcinoma upon histopathological evaluation.

Surgical excision encompassed the entire tumor involving the left half of both lips and the left labial commissure, followed by reconstruction using the "lip-shave" technique. Immediate postoperative findings demonstrated successful commissuroplasty. Complete excision with clear margins was achieved, and no recurrence or metastasis occurred during the 3-year follow-up.

Discussion: Verrucous carcinoma, originally described by Ackerman, represents a distinct entity from conventional squamous cell carcinoma, with a favorable prognosis and minimal metastatic potential. Clinical diagnosis is challenging, often resembling benign papillomatous lesions histologically. Treatment typically involves surgical excision, with varying outcomes based on tumor extent**.**

Conclusion: Buccal verrucous carcinoma poses diagnostic challenges, with divergent views on its classification relative to squamous cell carcinoma. Close collaboration with dermatologists is crucial for accurate diagnosis and management. Surgical excision remains the primary treatment modality, although its effectiveness diminishes in extensive cases.

Key Words: verrucous carcinoma, squamous cell carcinoma, buccal, surgical excision

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INTRODUCTION

Ackerman's tumor is typically considered a low-grade variant of squamous cell carcinoma, characterized by slow growth and a low incidence of metastasis despite local invasiveness and destructive tendencies. Diagnosis relies on clinical, macroscopic, and microscopic features of the lesion; its histologically benign appearance complicates diagnosis.

It is a slowly growing and locally aggressive tumor, with surgical excision with clear margins being the treatment of choice.

We present a case of a patient with challenging histological diagnosis, managed primarily surgically, with a favorable long-term outcome and no recurrence reported.

OBSERVATION

This concerns a 70-year-old female with a history of prolonged sun exposure. Physical examination reveals leukokeratoses involving the vermilion border of the upper and lower lips, as well as the left commissure, with multiple infiltrative lesions showing verrucous surfaces evolving over 3 years. Cervical lymph nodes were non-palpable. General examination and preoperative assessments were unremarkable. Initial histological examination of multiple biopsies suggested a papilloma altered by inflammation. Due to inconclusive histological results, dermatological consultation was sought, revealing via polarized dermoscopy findings resembling hairpin vessels, bright white structures, and yellowish crusts (Fig.1).

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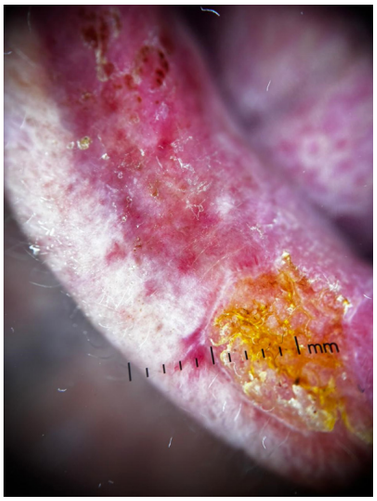

Figure 1: Polarized dermoscopy showing hairpin vessels, bright white structures, and yellowish crusts.

These findings prompted guided dermoscopy-directed biopsies, leading to the subsequent discovery upon histopathological examination of an extensive verrucous carcinoma covering the entire surface of the aforementioned lesions.Treatment consisted of surgical excision of the entire tumor involving the vermilion of the left half of both lips and the left commissure (Fig.2), with lip reconstruction using the "lip-shave" technique.

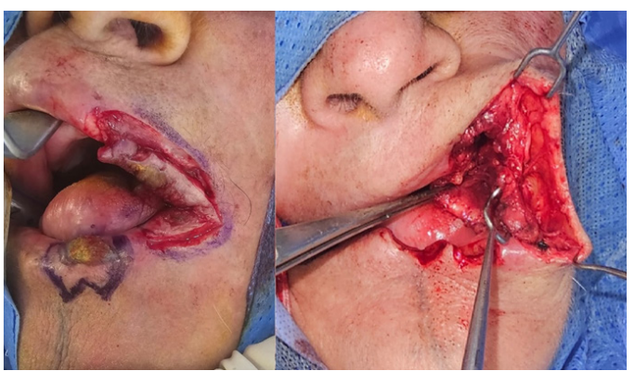

Figure 2: Image showing surgical excision of the entire tumor involving the vermilion of the left half of both lips and the left labial commissure.

This involved separating the mucosa from the muscular plane to the vestibule, gently pulling it over the excised vermilion area, and suturing it at the skin-mucosa junction. Additionally, commissuroplasty was performed (Fig.3). Excision was complete with clear margins. There has been no recurrence or metastasis noted in the 3 years following surgical treatment.

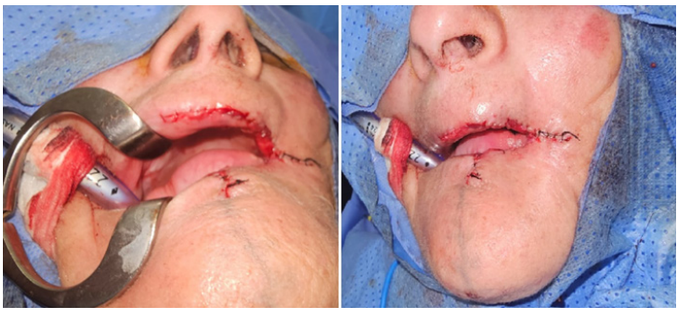

Figure 3: Immediate postoperative image demonstrating reconstruction of the left lips and the left labial commissure.

DISCUSSION

The verrucous carcinoma, formerly known as oral florid papillomatosis or Ackermann's tumor, is a rare, well-differentiated, non-metastatic variant of squamous cell carcinoma. It was first described by Ackermann in 1948. Its incidence is low compared to squamous cell carcinoma, which is the predominant cancer of the oral cavity and head and neck region. Verrucous carcinoma represents only 2% of oral cavity cancers. It exhibits slow growth and does not metastasize locally or distantly. Verrucous carcinoma of the buccal mucosa predominantly affects the attached gingiva and buccal mucosa [1]. The pathogenesis of buccal verrucous carcinoma remains debated [2]. Some studies, including those involving lips, suggest a potential role of UV exposure as a pathogenic factor in verrucous carcinomas [3], a principal risk factor described in our patient.

Clinically, it presents as an exophytic, keratinized lesion with a verrucous or papillomatous appearance, palpable and flexible to touch [4]. Diagnosis relies on anatomo-clinical correlation. Histologically, there is epithelial proliferation with a keratotic surface displaying verrucous features, along with sharp or blunt epithelial invaginations, either exophytic or endophytic, filled with keratin that pushes but does not invade the underlying connective tissue, giving a papillomatous appearance [5]. There are few cellular atypias, no vascular or neural invasion, and a base infiltrate of lymphoplasmacytic cells [6].

Two categories of verrucous carcinoma can be distinguished: those respecting the basal membrane and hybrid forms containing foci of squamous cell carcinoma within them. Chorionic infiltration may be considered a poor prognostic factor, warranting more aggressive management akin to squamous cell carcinoma. Histological diagnosis can be misleading with superficial biopsies. The lesion's regularity may mimic other benign papillomatous lesions such as verrucous hyperplasia, common wart, papillomatosis, or Schneiderian papilloma. Therefore, clinical correlation is essential, and multiple levels of cross-sections may be necessary in cases of doubt [7].

Treatment of buccal verrucous carcinoma is less radical than squamous cell carcinoma due to its lack of metastasis, but consensus on management is lacking. Common therapeutic options include surgical excision with or without cervical lymph node dissection, surgery combined with external beam radiotherapy, and radiotherapy alone (external or brachytherapy).

In a multivariate analysis, Alonso et al. [8] suggest that surgical management is a prognostic factor associated with improved overall survival. Surgical excision is the most widely described first-line treatment in the literature, involving clear margin excision without cervical lymph node dissection, although its efficacy diminishes in extensive cases [2].
Medina et al. [9] conclude from their cohort study that surgery is effective in achieving initial tumor control in 82% of cases, improving to 94% after recurrence. Koch et al. [10] report that surgical intervention alone is predominantly chosen for treating oral verrucous carcinomas, with rates as high as 85.5% for tongue lesions, 83.3% for lip lesions, and 82.6% for gingival mucosa.For extensive lesions, other therapeutic modalities may be considered, such as external beam radiotherapy, which remains an acceptable alternative in cases where surgery is contraindicated [11], despite historical controversy.The role of adjuvant therapies is not clearly defined in terms of indications and modalities but appears to be reserved for advanced stages.

CONCLUSION:

Buccal verrucous carcinoma can be diagnostically challenging. According to some authors, it may represent a minimally invasive variant of well-differentiated, low-grade squamous cell carcinoma, with specific clinical characteristics. Others view it as a distinct entity rather than a variant of squamous cell carcinoma, with a favorable prognosis.In the discussed clinical case, the superficiality of initial biopsy samples may have led to falsely negative results, highlighting the importance of close collaboration with dermatologists. Surgery remains the treatment of choice, although its efficacy is limited in extensive cases where complete excision may be technically challenging.

CONFLICTS OF INTEREST

The authors declare that there are no conflicts of interest.

Ethical approval

The authors declare that ethical approval was not required

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REFERENCES:

1. Zhu LK, Ding Y, Liu W. A clinicopathological study on verrucous hyperplasia and verrucous carcinoma of the oral mucosa. J Oral Pathol Med 2012;41:131-5.

2.Wu CF, Chen C, Shen Y, Huang I, Chen C. Effective eradication of oral verrucous carcinoma with continuous intra-arterial infusion chemotherapy. Head Neck 2008;30:611-7.

3.Zhu, L.-K., Y.-W. Ding, W. Liu, Y.-M. Zhou, L.-J. Shi, et Z.-T. Zhou. « A clinicopathological study on verrucous hyperplasia and verrucous carcinoma of the oral mucosa ». Journal of oral pathology & medicine 41, no 2 (2012): 131 35. https://doi.org/10.1111/j.1600- 0714.2011.01078.x.

4. Batsakis JG, Hybels R, Crissman JD, Rice DH. The pathology of head and neck tumors: verrucous carcinoma, Part 15. Head Neck Surg 1982;5:29-38.

5. Batsakis JG, Suarez P, El-Naggar AK. Proliferative verrucous leukoplakia and its related lesions. Oral Oncol 1999;35:354-9.

6. Santoro A, Pannone G, Contaldo M. A troubling diagnosis of verrucous squamous cell carcinoma (“the bad kind” of keratosis) and the need of clinical and pathological correlations: a review of the literature with a case report. J Skin Cancer 2011;2011: 370605.

7. Badoual, C., H. Péré, J. Cros, et H. Roussel. « Carcinome épidermoïde des voies aérodigestives supérieures : quoi de neuf en 2009 ». Annales de pathologie 29, no 4 (2009): 265 73. https://doi.org/10.1016/j.annpat.2009.07.004.

8. Alonso, J. E., E. C. Kuan, A. Arshi, et M. A. St John. « A population-based analysis of verrucous carcinoma of the oral cavity ». The laryngoscope 128, no 2 (2018): 393 97. https://doi.org/10.1002/lary.26745.

9. Medina, J. E., W. Dichtel, et M. A. Luna. « Verrucous-squamous carcinomas of the oral cavity. A clinicopathologic study of 104 cases ». Archives of otolaryngology 110, no 7 (1984): 437 40. https://doi.org/10.1001/archotol.1984.00800330019003.

10. Koch, B. B., D. K. Trask, H. T. Hoffman, L. H. Karnell, R. A. Robinson, W. Zhen, H. R. Menck, et al. « National survey of head and neck verrucous carcinoma : patterns of presentation, care, and outcome ». Cancer 92, no 1 (2001): 110 20. https://doi.org/10.1002/1097-0142(20010701)92:13.0.CO;2-K.

11. Kang CJ, Chang JTC, Chen TM, Chen IH, Liao CT. Surgical treatment of oral verrucous carcinoma. Chang Gung Med J 2003;26:807-12..