

## Oral Carcinoma Cuniculatum: A Case Report

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### ABSTRACT

We report a case of oral carcinoma cuniculatum, an exophytic variant of oral squamous cell carcinoma that has bland cytomorphic features, and a peculiar and characteristic growth pattern. Despite the lack of cytologic atypia, the tumor exhibited locally aggressive and infiltrative behavior with bone and cutaneous involvement. Pertinent benign and malignant mimics, and helpful differentiating features are also discussed.

*Key words:* oral squamous cell carcinoma, oral squamous cell carcinoma variant, oral carcinoma, mouth neoplasms

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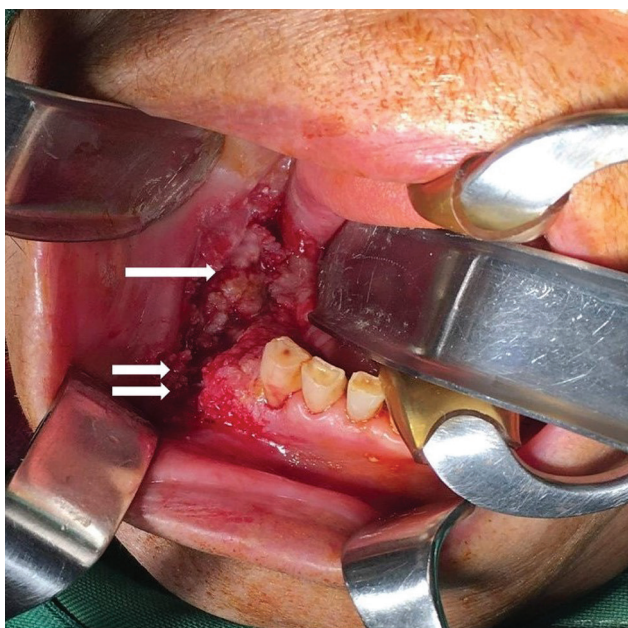
### CASE

A 75-year-old female consulted for a warty mass at the lower right premolar-molar area of 6 months' duration. Examination showed a sessile papillomatous gingival mass on the right posterior mandible with intra-osseous extension that produced a defect on the mandibular ridge (Figure 1). There was also an extra-oral fluctuant and draining communication to the overlying skin (Figure 2). An excision of the lesion was performed. Histological sections from the mass showed an exophytic superficial component composed of blunt papillomatous squamous proliferations with surface orthokeratosis (Figure 3). There was also an underlying invasive component composed of burrowing and branching channels that were lined by the same neoplastic squamous epithelium, and that contained keratinaceous debris within these channels (Figure 4). The tumor cells displayed minimal cytological atypia (Figure 4, inset). Based on these features, we signed the case out as Oral Carcinoma Cuniculatum (OCC).

### DISCUSSION

OCC is a rather unusual morphological variant of Oral Cavity Squamous Cell Carcinoma (OSCC) included in the current World Health Organization (WHO) Classification of Head and Neck Tumors and College of American Pathologists (CAP) Cancer Checklist for Lip and Oral Cavity.<sup>1,2</sup> It is chiefly characterized by an exophytic papillary surface with a blunt, "cobble-stone" appearance, a deeply invasive underlying component composed of anastomosing keratin-filled channels that are likened to "rabbit burrows," and minimal cytological atypia of the neoplastic squamous cell lining.<sup>3,4</sup> Because of the exophytic surface with minimal histological atypia, cogent differential diagnoses include innocuous benign lesions such as Squamous Papilloma, and another variant of OSCC characterized by a bland cytology - Verrucous Carcinoma (VC).<sup>1</sup> Squamous papillomas are benign and have limited growth potential. These are entirely superficial lesions that should be devoid of an invasive component. Both

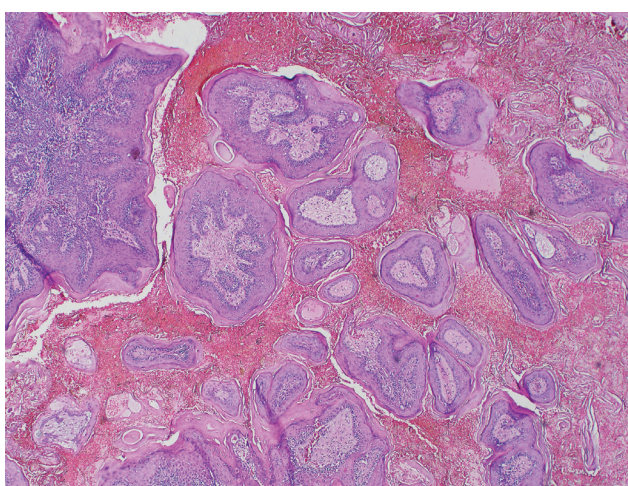




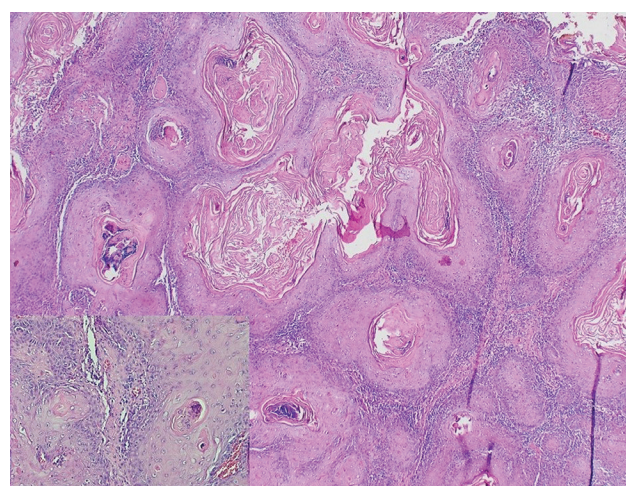
**Figure 1.** Papillomatous gingival mass on the right posterior mandible (arrow) with bone invasion (double arrow).



**Figure 2.** Fluctuant extra-oral extension to the overlying skin.



**Figure 3.** Blunt, papillomatous exophytic surface with orthokeratosis (H&E, X100).



**Figure 4.** Underlying invasive burrowing channels containing keratinaceous debris (H&E, X100); Neoplastic stratified squamous epithelial lining with minimal cytological atypia (Inset, H&E, X400).

OCC and VC are exophytic lesions that have a bland cytology, an invasive component, and remarkable keratin production.<sup>1,3,4</sup> However, the exophytic component of VC has a warty, pointed, “church spire-like” surface with excessive surface orthokeratin, in contrast to the blunt, “cobble-stone” surface of OCC. The endophytic invasive component of VC has a rounded, cohesive, and pushing front that is often limited to the lamina propria without infiltrating detached islands of tumor cells, while that of OCC is composed of keratin-filled, deeply burrowing, and anastomosing channels that frequently invade bone.<sup>1,3-5</sup> VC also only has rare mitoses confined to the basal layer, and without any abnormal forms.<sup>5</sup> Particularly difficult however when addressing these differential diagnoses, especially with VC, is if one is faced with limited tissues which is often the case with oral biopsies. Limited,

fragmented, or too superficial biopsies may prevent adequate evaluation of the architectural characteristics of the endophytic component. This uncertainty may have to be relayed to the clinician if a confident distinction cannot be made. Papillary Squamous Cell Carcinoma (PSCC) may enter among the considerations because of the exophytic nature of the lesion. However, PSCC displays severely dysplastic epithelial cells or cells devoid of maturation lining the papillary fronds, quite unlike the bland epithelial cells of OCC.<sup>4,6</sup> Also, PSCC usually has a filiform rather than a bulbous surface and absent to little keratinization that is largely limited to the surface.<sup>6</sup>

As with conventional OSCC, OCC affects the adult population and is associated with smoking.<sup>1,4</sup> HPV association is infrequently reported. The most commonly

reported site of involvement of OCC is the mandibular gingiva, as with our patient. Prognosis is suggested to be worse than that of VC but better than that of conventional OSCC.<sup>3</sup> Although metastasis is rarely reported, OCC is locally aggressive and infiltrative.<sup>3,4</sup> Our patient manifested with bone invasion and draining cutaneous communication. Two months after surgery, the patient had clinical findings indicative of a recurrence despite clear surgical margins.

## CONCLUSION

Because of the unusual growth pattern and bland cytology, the unacquainted pathologist may find it disconcerting to render a diagnosis of malignancy in OCC, especially if faced with limited tissues. Clinical and ancillary parameters, familiarity with the entity, and histologic clues of locally aggressive behavior, will certainly aid in the correct diagnosis.

## ETHICAL CONSIDERATION

Patient consent was obtained before submission of the manuscript.

## STATEMENT OF AUTHORSHIP

All authors certified fulfillment of ICMJE authorship criteria.

## AUTHOR DISCLOSURE

The authors declared no conflict of interest.

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## REFERENCES

1. Sloan P, Gale N, Hunter K, et al. Malignant surface epithelial tumours: squamous cell carcinoma. In: El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ, eds. WHO classification of tumours of the head and neck. 4th ed. Lyon: IARC Press; 2017.
2. Raja RS, Weinreb I, Bullock MJ, et al. College of American Pathologists Protocol for the examination of specimens from patients with cancers of the lip and oral cavity Version LipOralCavity 4.0.0.1; 2017. Available from: <https://cap.objects.frb.io/protocols/cp-headandneck-lip-oralcavity-17protocol-4001.pdf>. Accessed May 9, 2019.
3. Datar UV, Kale A, Mane D. Oral carcinoma cuniculatum: a new entity in the clinicopathological spectrum of oral squamous cell carcinoma. *J Clin Diagn Res.* 2017;11(1): ZD37-9. PMID: 28274074. PMCID: PMC5324519. <https://doi.org/10.7860/JCDR/2017/23437.9226>.
4. Farag AF, Abou-Alnour DA, Abu-Taleb NS. Oral carcinoma cuniculatum, an unacquainted variant of oral squamous cell carcinoma: a systematic review. *Imaging Sci Dent.* 2018;48(4):233-44. PMID: 30607347. PMCID: PMC6305781. <https://doi.org/10.5624/isd.2018.48.4.233>.
5. Zidar N, Cardesa A, Gillison M, Hekkiwekk T, Hille J, Nadal A. Verrucous carcinoma. In: El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ eds. WHO classification of head and neck tumors. 4th ed. Lyon: IARC Press; 2017.
6. El-Mofty SK, Cardesa A, Helliwell T, et al. Papillary squamous cell carcinoma. In: El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ, eds. WHO classification of head and neck tumours. 4th ed. Lyon: IARC Press; 2017.

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