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CLINICAL REPORT



Extensive carcinoma cuniculatum of the maxilla: A rare variant of oral carcinoma.

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ABSTRACT

Carcinoma cuniculatum (CC), is a rare well-differentiated, low-grade variant of squamous cell carcinoma. However, diagnosis of oral CC has remained very difficult, because many pathologists and clinicians remain unacquainted with oral CC, because there are very few reported cases of this disease in the oral cavity. To our knowledge, no more than 60 head and neck cases have been reported since Flieger and Owinski first described a case involving the mandible in 1977, because there are very few published cases of this disease in the oral cavity, the aim of this report is to provide a detailed clinical and histopathologic description of carcinoma cuniculatum of the maxilla, provide a brief review of the literature, and highlight the difficulties in arriving at the correct diagnosis.

KEY WORDS:

Oral cancer; Cuniculatum; Oral diagnosis.

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INTRODUCTION

Carcinoma cuniculatum (CC), is a rare well-differentiated, low-grade variant of squamous cell carcinoma; initially described by Aird et al. in 1954⁽¹⁾. Initially thought to be limited to cutaneous sites, most commonly the plantar surface of the feet, this neoplasm has since been described in several noncutaneous sites, including the oral mucosa^(2,3). To our knowledge, no more than 60 head and neck cases have been reported since Flieger and Owinski first described a case involving the mandible in 1977⁽⁴⁾, because there are very few published cases of this disease in the oral cavity, the aim of this report is to provide a detailed clinical and histopathologic description of carcinoma cuniculatum of the maxilla, provide a brief review of the literature, and highlight the difficulties in arriving at the correct diagnosis.

CASE REPORT

A 85-year-old male was referred to the author's institution for evaluation of persistent pain and swelling of the anterior maxillary region, 5 months of evolution without receiving prior treatment. The patient's past medical and dental histories were noncontributory and he denied tobacco and alcohol use. On extraoral examination, a slight swelling of the upper lip was noted. The intraoral examination (Fig. 1) revealed a exophytic nodular soft lesion, uneven surface, with the same color as the adjacent mucosa, and focal white patches as well small red dots, in the anterior region of the maxilla. Computed tomography showed a osteolytic lesion with poorly defined margins and irregular trabeculation in the anterior maxilla. Incisional biopsy specimen was obtained with the patient under local anesthesia. The biopsy report was indicative of CC, showing the sections: an orthokeratinized papillomatous epithelial surface, with mild nuclear pleomorphism and hyperchromatism, infiltrating the subyacent fibrous connective tissue stroma, forming cyst like sinuses and tracks, filled with keratin debris. (Fig. 2) The diagnosis was confirmed with the excisional biopsy. (Fig. 3)

DISCUSSION

CC is an extremely rare form of oral cancer. To the 2020, just 50 cases



Figure 1. Exophytic nodular soft lesion, uneven surface, with the same color as the adjacent mucosa and focal white patches, located in the anterior region of the maxilla.

of oral CC have been reported in English-language literature. Possible etiologic factors include trauma, chronic inflammation, radiation, etc; and it is reported a 3:1 male-to-female ratio^(5,6). In a retrospective analysis conducted by Sun et al⁽⁷⁾, CC had an incidence rate of 2.7% of all oral squamous cell carcinomas at their medical facility. According to these authors, the most frequent intraoral sites of appearance being the jaw and tongue. In our case, the site of appearance of the CC was the anterior maxillary region, making the case report even more exceptional, even Fonseca et al.⁽⁸⁾ for 2013 reported in their review only 4 cases of oral CC in the anterior maxillary region.

In 2005, the World Health Organization (WHO) included oral CC as a new variant of squamous cell carcinoma. According the WHO classification (2017), the features of oral CC are described as follows: well differentiated, usually on mucoperiosteum, locally destructive deep

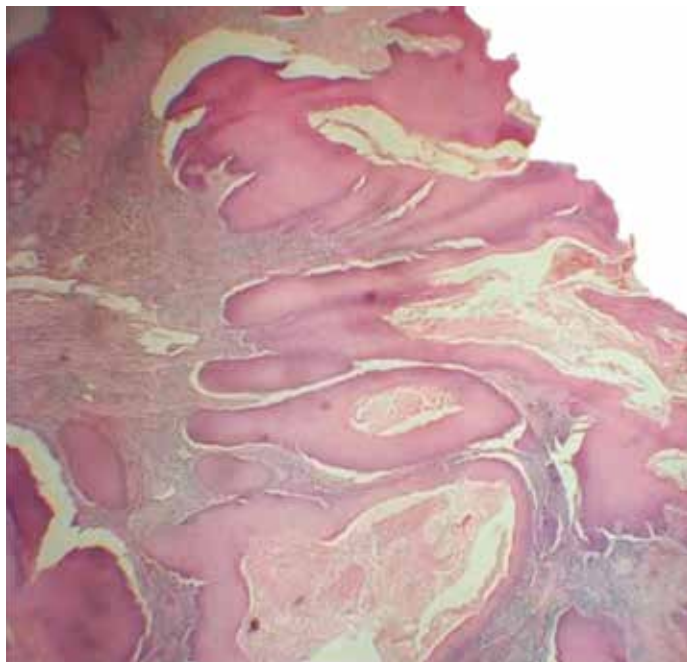


Figure 2. Incisional biopsy H&E 20X: orthokeratinized papillomatous epithelial surface, with mild nuclear pleomorphism and hyperchromatism, infiltrating the subacute fibrous connective tissue stroma, forming cyst like sinuses and tracks, filled with keratin debris.

burrowing pattern, metastasis rare, recurs locally but rarely if ever metastasizes. However, diagnosis of this entity has remained very difficult, because many pathologists and clinicians remain unacquainted with oral CC⁽⁹⁾.

Classic histology of CC is comprised of complex and branching networks of crypts resembling rabbit burrows, from which the term "cuniculatum" is derived. These crypts are characteristically lined by well-differentiated, stratified squamous epithelial cells with minimal cytological atypia⁽¹⁾. These characteristics being found in the biopsies performed in our case. However, because of numerous clinical and histologic similarities between this variant and other pathologies widely described, the true incidence of oral CC may have been underestimated and underdiagnosed in the general population⁽⁸⁾. The primary treatment option is total surgical resection, with high rates of tumor control demonstrated in the literature⁽¹⁻⁴⁾.

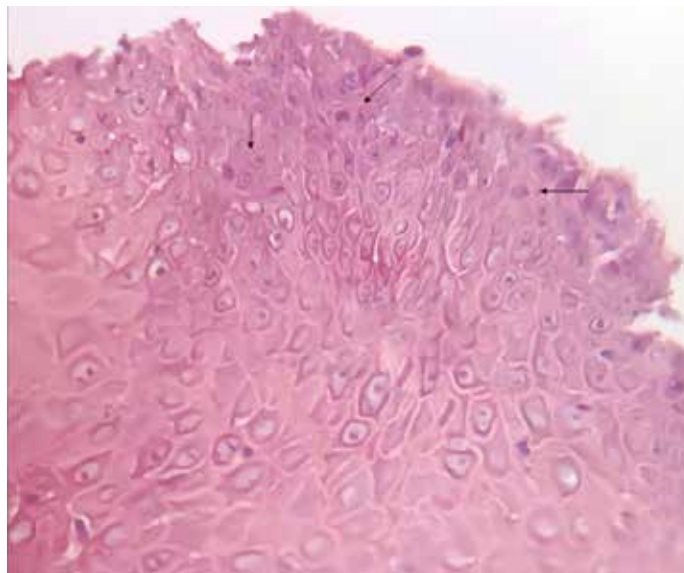


Figure 3. Excisional biopsy H&E 40X: Some cells (arrows) with cytologic changes.

CONCLUSION

This investigation underlines the difficulty of diagnosing CC, without clinicopathologic correlation and proper sampling, carcinoma cuniculatum may be underdiagnosed and subsequently undertreated. Histological evaluation is critical for an accurate diagnosis. The defining and distinguishing microscopic attributes of CC are complex. Complete surgical resection remains the mainstay of treatment and provides excellent rates of local tumor control.

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CONFLICTS OF INTERESTS

None.

ETHICAL APPROVALS

This study was approved by the Ethics Committee School of Dentistry, Universidad del Zulia

References

- Shay S, Choy W, Christensen R, John M. Extensive carcinoma cuniculatum of the mandible. *Am J Otolaryngol*. 2015;36(3):446-50. doi: 10.1016/j.amjoto.2015.01.002.
- Barreto J, Velazquez E, Ayala E, et al. Carcinoma cuniculatum: a distinctive variant of penile squamous cell carcinoma: report of 7 cases. *Am J Surg Pathol*. 2007;31:71-5. doi: 10.1097/01.pas.0000213401.72569.22.
- Puxeddu R, Cocco D, Parodo G, et al. Carcinoma cuniculatum of the larynx: a rare clinicopathological entity. *J Laryngol Otol*. 2008;122:1118-23. doi: 10.1017/S0022215107000163.
- Flieger S, Owinski T. Epithelioma cuniculatum rzadka postać nowotworu jamy ustnej i szczek [Epithelioma cuniculatum an unusual form of mouth and jaw neoplasm]. *Czas Stomatol*. 1977;30(5):395-401. Polish. PMID: 266444.
- Shapiro M, Wong B, O'Brien M, Salama A. Mandibular destruction secondary to invasion by carcinoma cuniculatum. 2015. *J Oral Maxillofac Surg*. 73:2343-51. doi: 10.1016/j.joms.2015.05.034.
- Neves-Silva R, Trivellato A, Sverzut C, Nelem C, Yamamoto L, Molena K, et al. Gingival carcinoma cuniculatum: Rare variant of squamous cell carcinoma often misdiagnosed as an inflammatory process. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2020;129(1):e90. doi.org/10.1016/j.oooo.2019.06.374
- Sun Y, Kuyama K, Burkhardt A, et al. Clinicopathological evaluation of carcinoma cuniculatum: A variant of oral squamous cell carcinoma. *J Oral Pathol Med*. 2012;41(4):303-8. doi: 10.1111/j.1600-0714.2011.01116.x.
- Fonseca F, Pontes H, Pontes F, Carvalho P, Sena-Filho M, Jorge J, et al. Oral carcinoma cuniculatum: two cases illustrative of a diagnostic challenge. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2013;116(4):457-63. doi: 10.1016/j.oooo.2013.06.006.
- Takashi I, Miura K, Kakehashi H, Shiraishi T, Egashira K, Asahina I. Carcinoma cuniculatum of the lower gingiva masked with leukoplakia: A case of difficult diagnosis. *J Oral Maxillofac Pathol*. 2021. 33:281-5. doi.org/10.1016/j.ajoms.2020.11.006