Case Report

Carcinoma Cuniculatum of the Oral Cavity – A Rare Entity

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Abstract:

Carcinoma cuniculatum is a rare lesion occurring in the oral cavity. Owing to its protracted growth and benign appearance it becomes a challenge for the clinician to diagnose. Therefore it is paramount importance that the diagnosis of carcinoma cuniculatum should be based on clinical feature and its histological architecture.

Keywords: Carcinoma cuniculatum, squamous cell carcinoma, verrucous carcinoma.

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Introduction

Carcinoma cuniculatum (CC) a variant of squamous cell carcinoma which exhibits verrucous or papillary growth with ulceration and is an uncommon lesion to occur in the oral cavity. CC is widely known to occur in other parts of the body such as esophagus, penis, and soles of the foot. CC was first described by Aird in 1954.1 The term cuniculatum derives from a Latin word 'cuniculus' meaning rabbit, because of a 'rabbit burrow' appearance of the complex branching keratinfilled crypt. The diagnosis of CC is a challenging task for a clinician because of its protracted growth, benign appearance and the rarity of its occurrence. Here we report a case of CC occurring on buccal mucosa.

Case Report

A sixty three year old female patient reported to the department of oral and maxillofacial surgery with a reddish white ulceroproliferative cauliflower like growth measuring 2cm x3 cm with a history of eight months duration. An excsional biopsy was done under local anesthesia and was sent for histopathological examination. Gross morphology of the lesional tissue showed the presence of irregular surface (Figure 1) and was firm in consistency. Routine hematoxylin and eosin staining of the lesion was done.



Figure 1: Showing gross photograph of excised tissue specimen.

On histopathological examination the lesion classical exophytic and endophytic growth pattern (Figure with undulating papillomatosis. The down growths of the tumor were club shaped and had a very distinct line of invasion into deeper tissue with an intact basement membrane and a single basal layer (Figure 2 and 3). The verrucous surface revealed cleft like spaces covered by parakeratinised cells extending deep into the lesion. The keratinocytes appeared to stain lightly with eosin and possessed a small nucleus. Deeper tissue invariably revealed numerous inflammatory cells, mainly lymphocytes beneath the tumor. (Figure 4)

seen in the mean age of 55 years and most common site being upper jaw.

Etiology and pathogenesis of CC are still remains unknown. A human papilloma virus (HPV) association has been implicated, but cannot be verified in every case.³ Alcohol and tobacco were possible etiologic factors in some reported cases,³ other causative factors described in literatures are traumatic event, chronic inflammation, radiation or arsenic ingestion.4

Due to rarity of the lesion the epidemiologic data are less to be found in literature.

Histologically the diagnosis of CC is usually difficult principally because of cellular features of a benign lesion.

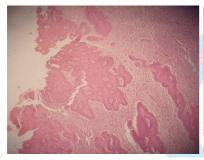
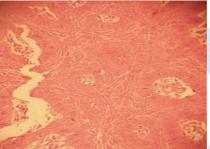
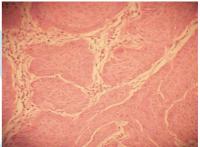


Figure 2: Photomicrograph **Figure 3:** Photomicrograph (10x) (4x) showing endophytic and exophytic proliferation.



showing downgrowth of tumor exhibiting club shaped invasion into deeper connective tissue.



4: **Figure** Under magnification

Collagen appeared to be compressed at the lower margin of the tumor. Based on these features final diagnosis of Carcinoma cuniculatum wad given.

Discussion

Carcinoma cuniculatum is a rare slow growing lesion which is a variant of oral squamous carcinoma. Clinically it is known to occur as a proliferative or verrucous growth with ulceration. It has frequently described in other parts of the body but very few cases have been reported in the literature when it is concerned with oral mucosa. The oral lesion is more common in males with a male: female of 14:5

superficial biopsy often reveals hyperkeratosis, acanthosis, benign squamous papilloma and mild dysplasia. Final diagnosis should not be based on just cytological features alone but on the clinical macroscopic microscopic, architectural features. Differential diagnosis of CC includes squamous papilloma and verrucous CC keratin-filled crypts carcinoma, typical, and in VC, the keratinization pattern is vertical or 'church-spire-like'. The pattern of hyperplasia in CC is a formation of canaliculi, and in VC, a bulbous expansion. comparison to papilloma with no inflammation and papillary squamous cell carcinoma with a moderate chronic, nongranulomatous inflammation, VC and CC show heavy inflammation.⁵

Conclusion

Carcinoma cuniculatum is a rare lesion to occur in the oral mucosa and can be a challenge in the diagnosis owing to its clinical and biological behavior. Thereby it is imperative that the diagnosis should be done based on its clinical features and histological architecture to avoid misdiagnosis.

References

1.Aird I, Johanson HD, Lennox B, Stansfeld AG. Epithelioma cuniculatum: a variety of scuamous carcinoma peculiar to the foot. Br J Surg.1954;42(173):245-50.

- 2.Bouquot JE. Oral verrucous carcinoma. Incidence in two US populations. Oral Surg Oral Med Oral Pathol Oral Radiol Endod.1998;86(3):318-24.
- 3.Raguse JD, Meneking H, Scholamnn HJ, Bier J. Manifestation of carcinoma cuniculatum in the mandible. OralOncol EXTRA 2006;42:173-175
- 4.Schwartz RA. Verrucous carcinoma of the skin and mucosa.J Am Acad Dermatol, 1995;32 (1):1–2.
- 5.De Petris G, Lewin M, Shoji T. Carcinoma cuniculatum of the esophagus. Ann Diagn Pathol.2005 Jun;9(3):134-8.

