



Case report

Peripheral ameloblastoma: use of cytokeratin 19 and Ber-EP4 to distinguish it from basal cell carcinoma

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Summary Rare tumor of the oral cavity presenting as a gingival swelling without bone involvement was present. The lesion, resected for mastication troubles, histologically presented a basaloid cell proliferation predominantly organized in nests formed by a peripheral palisading of cells with apically disposed nuclei. Immunohistochemical stainings for the cytokeratin 19 and Ber-EP4 showed a marked and diffuse positivity for the former, while the latter was negative. These results support the diagnosis of peripheral ameloblastoma and allow us to distinguish it from the basal cell carcinoma, which shares some histological features with this unusual tumor of the oral cavity.

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Introduction

Peripheral ameloblastoma is a rare tumour of the oral soft tissue presenting the same pathologic features of the osseous counterpart.¹ Because of the location and its histopathologic features it is possible to mistake it with basal cell carcinoma.² These two tumours present different biological behaviour so the differential diagnosis is important.³ This could make necessary the use of cytokeratin 19 and Ber-EP4, the first positive in ameloblastic cells⁴ and the second positive in the neoplastic basal cells.⁵ We describe a case of

peripheral ameloblastoma and discuss the differential diagnosis with basal cell carcinoma and the usefulness of the immunohistochemical staining with cytokeratin 19 and Ber-EP4.

Case report

Female, 69 years old, presented for 1 year on the gingiva a firm submucosal nodular growth. The lesion, 1.5 cm in the maximum diameter, brought about trouble for the mastication. For this reason she went to the surgeon to excise the lesion. A radiographic examination excluded involvement of the bone. The clinical diagnosis was "epulis". The

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surgical specimen was submitted for the histological examination.

Macroscopically the specimen was a whitish, firm, superficially ulcerated nodular lesion. The sections obtained from the surgical specimen showed microscopically a proliferation of basalioid cells arranged in cords, islands and nests originating from the overlying gingival epithelium. The neoplasm presented a peripheral palisading pattern. The architecture was highly suggestive of basal cell carcinoma, but some of the proliferative nodules showed a central area of polyhedral cells, sometimes splinded sometimes with squamous metaplasia, reminiscent of the immature enamel organ. The cells rimming the islands were cubic or cylindrical and had elongated, vesicular nuclei apically polarized toward the central reticular areas. Other areas showed small nests of immature cells surrounded by abundant fibrous stroma. Immunohistochemical procedures showed diffuse positivity of the neoplastic cells for CK19, in some areas more marked in the palisading cells. Also positive for the CK19 were some nests of epithelioid cells scattered into the gingival epithelium. Ber-EP4 was negative except some rare areas frankly basalioid where the neoplastic cells were slightly positive. EMA was focally positive in areas with squamoid metaplasia, but otherwise it was negative. The diagnosis of peripheral ameloblastoma was made.

Discussion

Peripheral ameloblastoma is an unusual variant of odontogenic tumor. ¹ Extraosseous location is the peculiar feature of this type of tumor, otherwise similar to the classical ameloblastoma. It is rare and its incidence is about 1% of all ameloblastomas. It is an indolent tumour, often presenting as a nodular growth of the gingiva, and it is characterized by a benign behaviour, cured by local excision. ³ Histologically the major differential diagnosis is the basal cell carcinoma of the skin with which it shares some microscopical features, so sometimes the differential diagnosis can be difficult. ² Like the basal cell carcinoma of the skin, the peripheral ameloblastoma is characterized by a proliferation of basal cells often arranged in nests included in a fibrous stroma with areas of contact with the epidermis. The arrangement of the rim cells of the nests are the same, that is typically palisading, but in contrast with the basal cell carcinoma, whose cells have the nuclei located on the

basis of the cytoplasm, the rim cells of the peripheral ameloblastoma have the nuclei located on the upper part of the cytoplasm. Furthermore the immunohistochemical analysis outlines the differences between these two types of tumors.

The basal cell carcinoma shows diffuse and marked positivity for Ber-EP4 ⁵ and negativity for cytokeratin 19; ⁶ on the contrary, the peripheral ameloblastoma is always negative for Ber-EP4, except very focal areas in which a true basalioid differentiation is present, and diffusely and markedly positive for the cytokeratin 19. ⁴ Very peculiar is the presence in the gingival epithelium of some clusters of cytokeratin 19 positive cells otherwise negative. It could be that these cells represent epithelial rests of the dental lamina in the gingival epithelium.

Therefore the immunohistochemical stainings are the clue for the differential diagnosis between peripheral ameloblastoma and basal cell carcinoma overall in the cases in which peculiar histological features are not present or when an incisional biopsy is made. Moreover it is important to differentiate these two neoplasms because having different biological behaviour they need different treatment. In fact while a simple local excision is often resolutive for peripheral ameloblastoma without risk of recurrence, the basal cell carcinoma needs a larger excision to exclude the risk of local recurrence and/or regional metastases ³. For this reason we suggest the use of cytokeratin 19 and Ber-EP4 when a tumor of the soft part of the oral cavity with histological features of basal cell carcinoma and/or peripheral ameloblastoma occurs.

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