

Case Report

Peripheral ameloblastoma

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Abstract : A case of peripheral ameloblastoma in a 57-years-old woman is presented, along with a discussion of the clinical and histological characteristics of the lesion. After clinical and radiographic examinations, and with a differential diagnosis of pyogenic granuloma, an excisional biopsy was performed and the material collected was sent for histological examination. On the basis of the histopathological diagnosis, a second operation was performed with a wide safety margin, including bone tissue, which did not show any involvement with the odontogenic neoplasm.

Keywords: ameloblastoma; peripheral ameloblastoma; gingival lesions.

Introduction

Peripheral ameloblastoma is a rare lesion usually occurring on the gingiva with histological characteristics similar to those of centrally located ameloblastoma, although it appears exclusively in the gingiva or mucosa covering the tooth-bearing areas of the jaws^[1,2]. While intraosseous ameloblastoma is locally aggressive with invasion and destruction of the bone, peripheral ameloblastoma generally lacks this persistent invasiveness^[3].

One case of peripheral ameloblastoma, with a previous differential diagnosis of pyogenic granuloma, is presented along with a discussion of the clinical and histological characteristics of the lesion.

Case Report

A female caucasian patient, aged 57 years, came to the Department of Diagnosis and Oral Surgery of the School of Dentistry of Sao Jose dos Campos - UNESP, complaining of a bleeding lesion in the region of the lower left second molar, which had increased in volume with time (nearly 1 year). Clinical examination showed an

exophytic, hyperemic lesion, bleeding at the touch but otherwise causing no symptoms, in the retromolar and vestibular marginal gingiva. A CT scan showed no bone alteration (Fig.1). With a differential diagnosis of pyogenic granuloma, an excisional biopsy was made with a small margin, and the material collected was sent for histological examination. As the findings indicated peripheral ameloblastoma, an expansion of the biopsy area was suggested, including removal of the alveolar bone, to examine for a possible central ameloblastoma in the buccal cavity. After the second operation, the material sent for examination revealed no histological alteration. A clinical and radiographic follow-up of the patient has revealed no sign of recurrence.

Histological examination of the excised specimen revealed mucosal fragments with an odontogenic epithelial neoplasm, showing sheet-like proliferation of cubic cells, with round or oval nuclei containing loose chromatin and showing various stages of mitosis. Such cellular masses were sometimes continuous with the basement membrane of the mucosal covering epithelium, showing squamous metaplasia in some areas. In plexiform regions, anastomotic chains generally consisted of a double layer of columnar cells (Fig.2). There were also peripheral columnar cells with polarized nuclei located next to the basement membrane, along with central cells which sometimes resembled cells of the enamel organ stellate reticulum, and sometimes were squamous (Fig.3). Merging of the last two patterns (plexiform and follicular) could also be observed in the surface epithelium, and in the peripheral region of the neoplasm there was continuity with the surface epithelium (Fig. 4). The latter areas showed ulceration covered by a fibrous-purulent exudate, showing severe spongioblastosis and exocytosis. Some neoplastic epithelial cells were detected in the lateral and deep parts of the fragments examined. The fibrous connective stroma of the neoplasm showed areas of mononuclear inflammatory infiltration.

Discussion

Peripheral ameloblastoma, although relatively rare in the buccal cavity, has been given a number of different names, including extra-osseous ameloblastoma, soft tissue ameloblastoma, ameloblastoma of mucosal origin,

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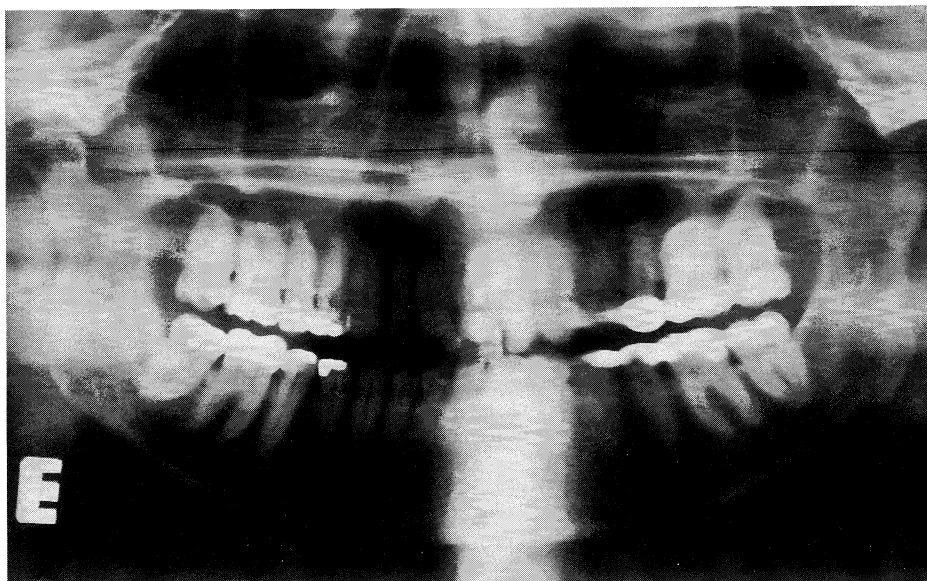


Fig. 1 : Panoramic X-ray of the jaw showing absence of bone involvement.

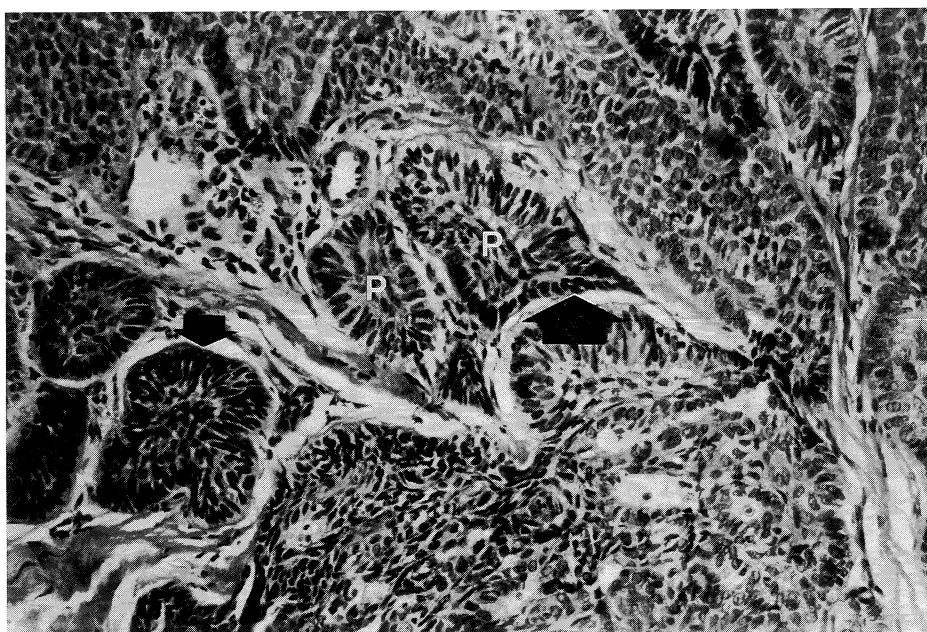


Fig. 2 : Region with plexiform structure (P), showing anastomotic chains consisting of a double layer of columnar cells (arrows). Hematoxylin and eosin staining. X210

ameloblastoma of the gingiva and odontogenic gingival epithelial hamartoma^[2], and may occur at any age except in young adults^[3]. The neoplasm shows as apparent predilection for the dentulous jaw^[4].

Peripheral ameloblastoma, although presenting histological characteristics similar to intraosseous ameloblastoma, attacks only soft tissue, mostly in the area of the teeth involved. Tumors such as basal cell adenoma, adenoid-cystic carcinoma of the minor salivary glands and the adamantoid variation of squamous cell carcinoma present histological characteristics quite similar to peripheral ameloblastoma^[2,4].

The equivocal clinical characteristics of peripheral ameloblastoma may lead to a wrong differential diagnosis, resulting in incorrect planning or insufficient surgery without total resection with an adequate safety margin or removal of the underlying bone tissue for appropriate histopathological analysis, as in the present case. The clinical similarity with lesions such as peripheral reactive hyperplasia, ossifying fibroma, peripheral odontogenic fibroma, or inflammatory hyperplastic lesions, such as peripheral giant cell granuloma or pyogenic granuloma may also contribute to erroneous diagnosis and in some cases, only histopathological findings provide the final correct diagnosis^[2,3]. Another important diagnostic modality for peripheral ameloblastoma is radiographic examination, as the lesion does not invade the bone tissue, although there have been some reported cases of minimal bone erosion, invasion or reabsorption, without severe bone destruction. This characteristic differentiates peripheral ameloblastoma from intraosseous ameloblastoma, which causes marked bone tissue lysis.



Fig. 3 : Mass of cubic neoplastic cells with a basaloid aspect (arrows), continuing from the surface epithelium and neoplastic areas with columnar peripheral cells (CC). Hematoxylin and eosin staining. X157

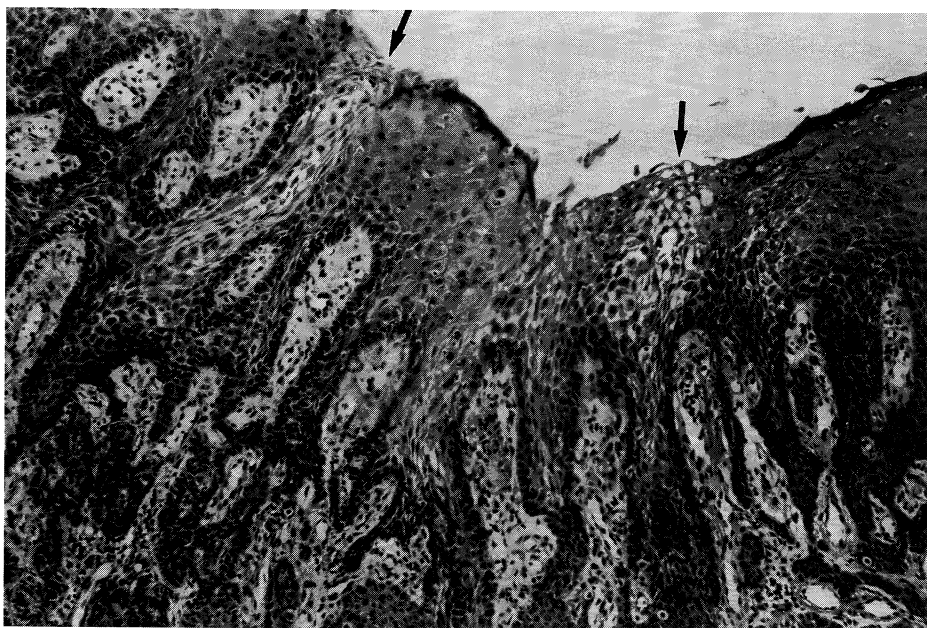


Fig. 4 : Peripheral region of the neoplasm showing its continuity with the surface epithelium (arrows). Hematoxylin and eosin staining. X100

In all cases, treatment should be surgical, with a safety margin and curettage, and also, depending on the radiographic findings, the extent of lesion resection should be increased to include the alveolar bone and extraction of the involved teeth. Peripheral ameloblastoma does not tend to be recurrent and, although in some cases malignancy may be found, this is not common^[5,6].

When studying peripheral ameloblastoma, the biggest consideration is its etiology, which is largely controversial and for which several hypotheses have been put forward. Possible origins include epithelial remnants of the dental lamina and derivatives of the enamel organ or the basal cell layer of the oral mucosa^[7,8,9]. As Woo *et al.*^[10] have stated, it is difficult to explain the origin of a peripheral ameloblastoma that arises in the buccal mucosa on the basis of dental lamina rest proliferation, since such rests would rarely occur in such a location. This may explain the extremely rare incidence of peripheral ameloblastoma in the buccal mucosa. Another two histogenic origins have been proposed: tumors that show complete separation from the overlying surface epithelium, probably arising from odontogenic epithelial remnants^[11], and tumors showing direct extension from the basal cell layer of the overlying epithelium^[12]. Due to understand the etiological complexity of this lesion, further detailed studies should be carried out.

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