

A Recurrence of Intra-Osseous Ameloblastoma in Soft Tissue

Recurrencia de Ameloblastoma Óseo en los Tejidos Blandos

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ABSTRACT: Ameloblastomas are usually found in the hard tissues of the jaws, but have also been reported in the adjacent soft tissues. The extensive resection including bone as well as adjacent soft tissues is critical because the recurrences seem to stem from the soft tissues. Here we report the unusual occurrence of an extraosseous ameloblastoma in the tissues of the cheek.

KEY WORDS: ameloblastoma, odontogenic tumors, recurrence.

INTRODUCTION

The most common odontogenic neoplasm that affect the gnathic osseous is the ameloblastoma, still it accounts less than 1% of all tumors of the jaws (Rapidis *et al.*, 2004). Its origin from odontogenic epithelium and may arise from enamel organ, remnants of dental lamina, the lining of an odontogenic cyst (dentigerous) or possible from the basal epithelial cells of the oral mucosa (Small & Waldron, 1955). It often presents as a slow-growing, painless swelling causing expansion of the cortical bone, perforation of the lingual or buccal plates, and infiltration of soft tissue (Ghandhi *et al.*, 2006).

The peripheral ameloblastoma is rare, its location is in soft tissue and the behavior is less aggressive. It corresponds from 1-5% of all ameloblastomas (El-Mofty *et al.*, 1991). Ameloblastomas have a biologic behavior which is characterized by local invasion, rare capacity to metastasize and great capacity of recurrence (Campbell *et al.*, 2003). The rates of recurrence depend on the type of surgery and range from 15-25% after radical surgery to 75-90% after conservative surgical management (Nakamura *et al.*, 2002).

Most recurrences occur at the site of the primary tumor. However, lesions are also known to recur in bone grafts, and rarely, recurrence involves adjacent soft tissues (Ferretti *et al.*, 2000).

This report presents a case of a recurrence of ameloblastoma intra-osseous in soft tissue.

CASE REPORT

A male Caucasian patient, 53 years-old was attended at Hospital Universitário Oswaldo Cruz in Recife, Pernambuco, Brazil with a complain of tissue augmentation in the oral cavity. The medical history revealed 24 years ago, patient was submitted an enucleation and curettage in mandible angle with a histopathological diagnostic of ameloblastoma. Another surgical procedure for relapse in right body, ramus and angle with great dimension has been performed 19 years ago. Partial resection was realized with safety margin and the proximal and distal stumps were fixed

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with Kirschner thread. The patient returned to the hospital 13 years ago because he was victim from physical aggression in mentum region with intra-oral exposure of Kirschner thread that was removed under general anesthesia. Currently, the patient returned and during intra-oral exam was noticed an asymptomatic lesion in right buccal mucous with a smooth surface, firm rubbery consistency, normal roseate coloration and sessile (Fig. 1). It can be noticed the osseous defect in right alveolar region of mandible (Fig. 2). The x-ray exam showed the area in right mandible that suffered resection (Fig. 3). The diameter of the tumour was about 25mm. The management was

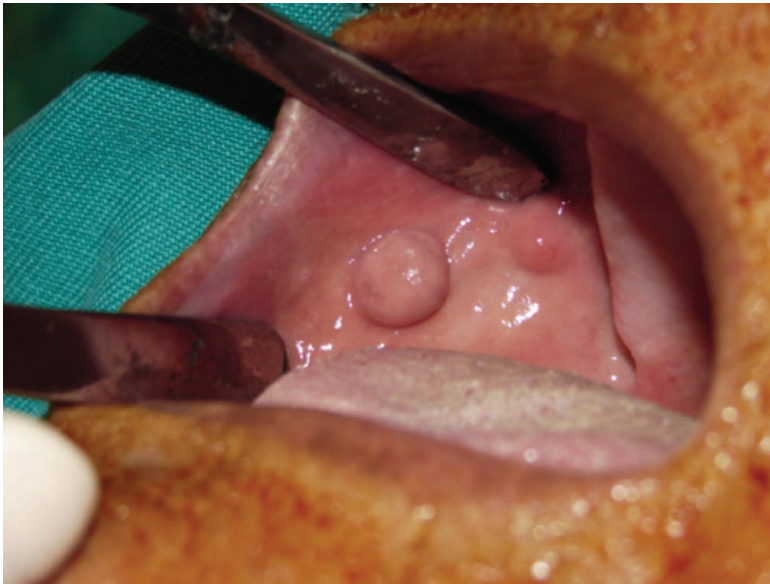


Fig. 1. An asymptomatic lesion in right buccal mucous.



Fig. 2. Osseous defect in right alveolar region of mandible (patient with upper dental prosthesis).

excisional biopsy and sent to the Pathological Laboratory. The histopathological analysis presented ameloblastoma (Fig. 4 and 5). Therefore, it was a recurrence from ameloblastoma intra-osseous in the soft tissue. The wound healed well and there has been no sign of recurrence after 2 years.

DISCUSSION

Recurrence of ameloblastoma is thought to be related predominantly to inadequate surgical removal of the primary tumor (Rapidis *et al.*). Although, the recurrence may be further attributable to these reasons: firstly, the presence of small islands of neoplastic tissue in the cancellous bone at the margins of the specimen or the implantation of tumor cells during enucleation. Secondly, the consequence of soft tissue recurrence (Gold, 1991). The overlying mucosa should be included in the resection if the tumor invades the alveolus and perforates through the alveolar bone. Thirdly, tumor seeding. This should be considered as the most important causative factor in the recurrence of ameloblastoma in bone grafts (Zachariades, 1988).

The literature has a few reports of recurrence of ameloblastoma intra-osseous in soft tissue (Olasoji *et al.*, 2004). This report is corroborated by this work that illustrated the necessity of taking into consideration the adjacent soft tissue in cases of tumor with cortical bone perforation during surgical treatment. This is especially important in an environment where patients are not likely to attend follow-up review for a long period. It also demonstrates the ability of intra-osseous ameloblastoma to grow in soft tissues. It was concluded that extensive resection including bone as well as adjacent soft tissues is critical because the recurrences seem to stem from the soft tissues, especially from the adjacent periosteum.



Fig. 3. The x-ray exam showing the area in right mandible that suffered resection.

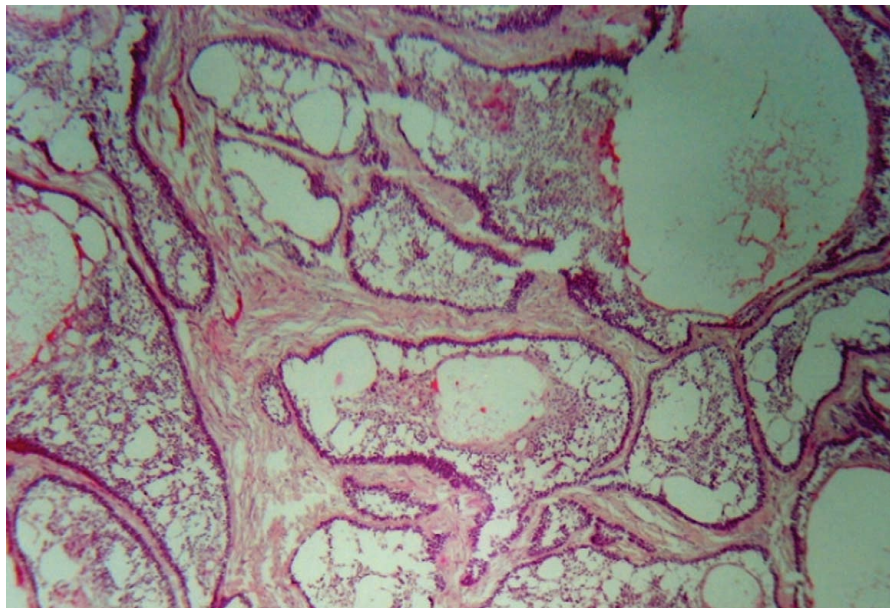


Fig. 4. Follicle type ameloblastoma with odontogenic epithelium islands in a stroma of fibrous connective tissue. HE. 40x.

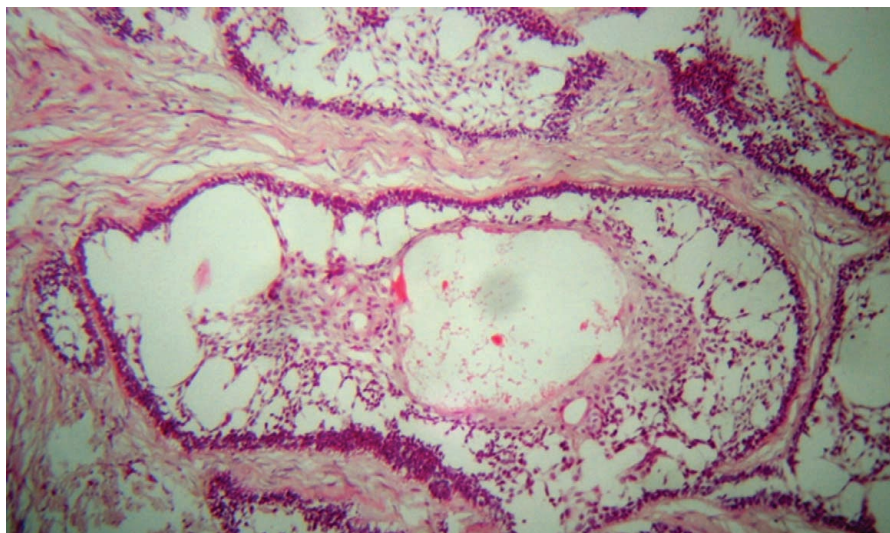


Fig. 5. Epithelium odontogenic island presenting peripheral cells with inverted polarized nucleus and middle cells similar to stellate reticulum. HE. 100x.

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RESUMEN: Ameloblastomas se encuentra normalmente en el tejido óseo de las mandíbulas, sino que también han sido reportados en los tejidos blandos adyacentes. Incluida la amplia resección ósea, así como los tejidos blandos adyacentes es fundamental porque el recidivas parecen provenir de los tejidos blandos. Aquí el informe de un inusual extraosseous ameloblastoma en los tejidos de la mejilla.

PALABRAS CLAVE: ameloblastoma, tumor odontogénico, recurrencia.

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