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Peripheral Ameloblastoma, Rare and Often Misdiagnosed Entity. Case Report and Review of Literature

Ameloblastoma Periférico, Una Entidad Rara y Frecuentemente Mal Diagnosticada. Reporte de Caso y Revisión de la Literatura

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ABSTRACT: Peripheral ameloblastoma (PA) is a rare benign odontogenic tumor that accounts for only 2 to 10% of all ameloblastomas. It originates in the soft tissues, typically in the gingiva, and may mimic common reactive lesions, making diagnosis challenging. This paper presents a rare case of peripheral ameloblastoma and reviews the related literature, highlighting diagnostic challenges, histopathological features, and current treatment approaches. A 43-year-old male patient presented with a firm, painless, non-bleeding 3¥3 cm gingival lesion adjacent to the upper right third molar. Radiographic evaluation revealed no bone involvement. A clinical diagnosis of pyogenic granuloma was made, and complete excision was performed along with the extraction of the impacted tooth. Histopathological analysis showed ameloblastomatous epithelium and immunohistochemical positivity for CK19, confirming the diagnosis of peripheral ameloblastoma. Findings were consistent with ameloblastic islands beneath intact squamous epithelium. No bone invasion or malignant transformation was observed. At the two-month follow-up, no recurrence was detected. Peripheral ameloblastoma occurs more frequently in men in their fifth decade and typically affects the mandibular gingiva. Its differential diagnosis includes reactive lesions such as pyogenic granuloma and basal cell carcinoma. Immunohistochemistry, particularly CK19 positivity and Ber-EP4 negativity, is key for differentiation. Although conservative surgical excision is usually effective, cases of recurrence and malignant transformation have been reported, making long-term clinical follow-up essential.

KEY WORDS: Peripheral ameloblastoma; Odontogenic tumor; Differential diagnosis.

INTRODUCTION

Ameloblastoma is a rare odontogenic benign tumor that represents 1 % of all head and neck tumors (Ghai, 2022). First reported by Cusack in 1827, ameloblastoma is classified by the World Health Organization (2022) in two distinct forms: central (intraosseous), the most prevalent form (80 %), and peripheral (extraosseous), representing only 20 % of all ameloblastoma cases (Vered & Wright, 2022).

The extraosseous variant of ameloblastoma, also known as peripheral ameloblastoma, typically occurs in the soft tissues of the tooth-bearing area. However, though in a few cases the extra gingival location has also been reported (Shi, *et al*., 2021). This paper aims to present a rare case of peripheral ameloblastoma and describe from a literature review his diagnostic features highlighting the challenges in identifying this tumor that can be mistaken for more common reactive lesions on the gingiva.

CASE REPORT

A 43-year-old male patient presented in the department of oral surgery with the complaint of an asymptomatic gingival growth in the region of the right maxillary wisdom tooth, which had grown over the last year according to the patient. The patient's medical history was unremarkable, with no reported systemic diseases, drug use, cigarette, or alcohol consumption.

The intraoral examination showed a 3¥3 cm gingival outgrowth at the site of the right maxillary

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wisdom tooth. The lesion displayed a non-bleeding, and pebbly surface with a sessile base. on palpation it was firm and painless (Fig. 1). No extraoral swelling or lymphadenopathy was noted in the extraoral examination.

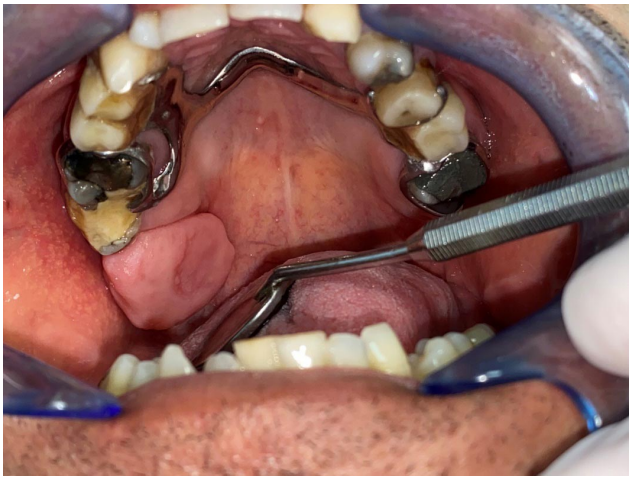

Fig. 1. Intraoral view showing an exophytic growth in the region of maxillary wisdom tooth.

The radiographic examination showed the impacted upper wisdom right tooth with no evidence of bone involvement (Fig. 2).

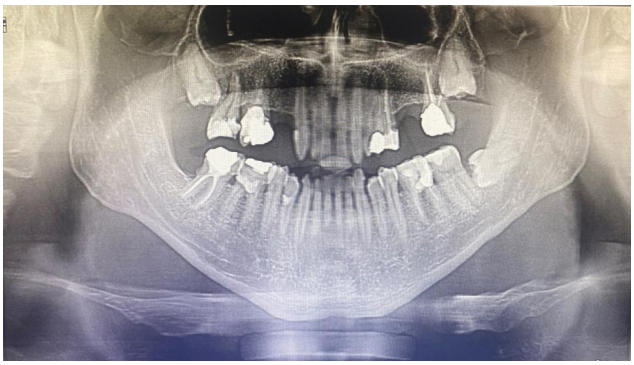

Fig. 2. Radiographic presentation without bone involvement and impacted 18.

A presumed diagnosis of pyogenic granuloma was established based on clinical and radiological findings. A complete excision was performed under local anesthesia, with the extraction of the impacted maxillary wisdom tooth situated submucosally with no bone in the supra-coronal region, then the specimen was submitted to histopathologic examination (Fig. 3).

Microscopically, the lesional tissue showed a hyperplastic stratified squamous epithelium with numerous papillary projections and multiple areas that showed ameloblastomatous change. Immuno histochemical analysis showed positivity of the superficial epithelium for the odontogenic marker cytokeratin 19 (CK 19), confirming its odontogenic transformation.

Based on the histopathological presentation, the lesion was diagnosed as peripheral ameloblastoma.

At the control examination, two months after excision, the alveolar site of 8 was completely healed and no recurrence was noted. Additionally, the patient was informed about the importance of regular controls annually for early diagnosis of possible recurrences.

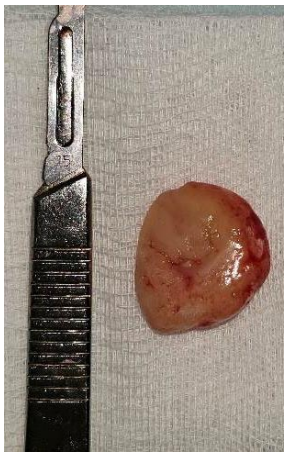

Fig. 3. Post-operative view of operative piece.

DISCUSSION

Peripheral ameloblastoma, ameloblastoma of the soft tissues, gum ameloblastoma, and ameloblastoma of mucosal origin are terms to designate the extraosseous form of ameloblastoma that was first reported in the literature by Kuru in 1910 (Vezhavendhan *et al*., 2019; Ide *et al*., 2020).

Peripheral ameloblastomas are more common in male patients, with a maximum incidence between the fifth and sixth decades of life. The most common location is the premolar region with the retromolar aspect of the mandible. Other less common regions include the posterior tuberosity in the maxilla and the floor of the mouth. The lesion was in line with what was described in previous literature and was identified in a man in his 40s (Vanoven *et al*., 2008; Singh *et al*., 2022).

The actual histogenesis of PA is still controversial. It may originate from the “Serre’s pearls” (the extraosseous cellular residuals of the dental lamina) or cells in the basal cell layer of the surface

epithelium. The origin from stratified squamous epithelium or pluripotent cells of minor salivary glands has also been proposed (Ide *et al*., 2009).

The lesion usually presents as a slow-growing exophytic mass from the soft tissues, most frequently encountered in the edentulous area. It can also manifest at the base of the tongue or on the oral floor. It is generally painless, firm, granular, or pebby-like surfaced growth, sessile or pediculated, with a normal mucosa color as we noted in our case, or it can be dark red (Luciani *et al*., 2023).

Peripheral ameloblastomas don't penetrate bone structure, and lesions don't affect the cortical bone. In our case, the observations were in line with this information, as there was no cortical bone alteration in the radiological assessment. No change or invasion of cortical bone had been noticed in our case during the radiological examination. Furthermore, during the surgery, there was no bone in the supra-coronal region, with the tooth situated submucosally. A few cases have shown some bone involvement, which was referred to as cupping or saucerization. This process refers to a depression made from the pressure of the tumor on bone; however, saucerization is usually mild, and no neoplastic invasion or bone infiltration is seen (Wettan *et al*., 2001; Borrello *et al*., 2016).

Histologically, PA reveals ameloblastic growth under an intact layer of overlying squamous epithelium with islands of tissue that show a central part rich of stelliform cells. The most common histological form presents follicular or plexiform cells. The epithelium is always well outlined but not capsulated with fibrous tissue; therefore, very often, the tumor epithelium is in close contact with the surrounding mucosa. In contrast to intraosseous ameloblastoma, mitosis is rare. However, stromal tissue tends to contain lymphocytic inflammatory infiltration (Philipsen *et al*., 2001; Ide & Kusama, 2004).

Clinically, this lesion can be misinterpreted as other reactive lesions of the oral mucosa, which are much more common (such as pyogenic granuloma, peripheral ossifying fibroma, and peripheral giant cell granuloma), as well as basal cell carcinoma (BCC) (Manor *et al*., 2004).

(BBC) and (PA) in some cases present histological similarities, sometimes requires the use of immunohistochemistry to differentiate the two lesions. Both lesions show a proliferation of basal cells organized in a nest surrounded by stromal fibrous tissue. (PA) can be recognized by the presence of nuclei in the upper portion of the cells, whereas in (BCC) these nuclei are localized in the lower part of the cell's cytoplasm. In the immuno-histochemical analysis, (PA) is positive for cytokeratin 19 and negative for Ber-EP4, on the contrary of (BCC) (Tajima *et al*., 2001; Chhina & Rathore, 2015; Upadhyaya *et al*., 2018).

The treatment of this lesion consists of a conservative excision of the tumor, generally without removing bone or teeth. In our observation, the extraction of tooth 18 was performed due to its non-functional status, facilitating the complete elimination of the pathological tissue (Borrello *et al*., 2016; Anpalagan *et al*., 2021).

It was reported that peripheral ameloblastoma, unlike the central form, has a good prognosis with low recurrence potential; however, cases of recurrence and malignant transformation have been reported in the literature, which requires regular and long-term monitoring (Baden *et al*., 1993; Tajima *et al*., 2001).

CONCLUSION

In conclusion, peripheral ameloblastoma is a variant of ameloblastoma that shares its histologic features but is found to be less aggressive and don't usually invade the underlying bone. Nerveless, due to his rarity, the lesion may be mistaken for more common reactive lesions on the gingiva, and the diagnosis is usually made based on their histologic appearance and immunohistochemical results.

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RESUMEN: El ameloblastoma periférico (AP) es un tumor odontogénico benigno poco frecuente, que representa solo entre el 2 y el 10 % de todos los ameloblastomas. Se origina en los tejidos blandos, típicamente en la encía, y puede imitar lesiones reactivas comunes, lo que dificulta su diagnóstico. Este trabajo presenta un caso poco común de ameloblastoma periférico y revisa la literatura relacionada, destacando las dificultades diagnósticas, las características histopatológicas y los enfoques terapéuticos actuales. Se trata de un paciente masculino de 43 años que consultó por una lesión gingival firme, indolora, no sangrante de 3×3 cm, adyacente al tercer molar superior derecho. La radiografía no mostró compromiso óseo. Se diagnosticó clínicamente como granuloma piogénico y se realizó su escisión completa junto con la extracción del diente incluido. El análisis

histopatológico reveló epitelio con cambios ameloblastomatosos y positividad inmunohistoquímica para CK19, confirmando el diagnóstico de ameloblastoma periférico. Los hallazgos fueron consistentes con islotes ameloblásticos bajo un epitelio escamoso intacto. No se observó invasión ósea ni transformación maligna. A los dos meses de seguimiento, no se evidenció recurrencia. El ameloblastoma periférico afecta más frecuentemente a hombres hacia la quinta década de vida y compromete principalmente la encía mandibular. Su diagnóstico diferencial incluye lesiones reactivas como el granuloma piogénico y el carcinoma basocelular. La inmunohistoquímica, especialmente la positividad para CK19 y negatividad para Ber-EP4, resulta clave para su diferenciación. Aunque el tratamiento conservador suele ser eficaz, se han reportado casos de recurrencia y transformación maligna, lo que hace necesario un seguimiento clínico a largo plazo.

PALABRAS CLAVE: Ameloblastoma periférico; Tumor odontogénico; Diagnóstico diferencial.

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