J Clin Periodontol 2005; 32: 12–15, doi: 10.1111/j.1600-051X.2004.00627.x

Journal of Clinical Periodontology

Peripheral ameloblastoma of the gingiva: the importance of diagnosis

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López-Jornet P, Bermejo-Fenoll A: Peripheral ameloblastoma of the gingiva: The importance of diagnosis. J Clin Periodontol 2005; 32: 12–15. doi: 10.1111/j.1600-051X.2004.00627.x. © Blackwell Munksgaard, 2004.

Abstract

Background: Peripheral ameloblastoma is an extremely rare epithelial odontogenic tumor, limited to the soft tissues of the gums or oral mucosa. Although the lesion is benign, it may be locally aggressive.

Methods: The present study describes the case of a 31-year-old male presenting a firm, symptomless tumor mass of irregular appearance and measuring approximately 12 mm in diameter, located in the distal zone of 4.7.

Results: An excision biopsy was performed. The lesion was covered with hyperplastic squamous epithelium, with islets of epithelial cells located at subepithelial level. The cells in the peripheral zone adopted a palisade distribution, and presented the appearance of a lax reticulum at central level. A fibroblastic stroma was observed between the islets. The diagnosis was peripheral ameloblastoma.

Conclusions: Although the origin of the lesion remains unclear, it is able to recur and undergo malignant transformation. Consequently, peripheral ameloblastoma should not be viewed as a harmless mass.

Key words: ameloblastoma; diagnosis; odontogenic tumor; peripheral ameloblastoma

Accepted for publication 29 March 2004

Peripheral ameloblastoma is a very rare odontogenic tumor, representing approximately 1–5% of all ameloblastomas (El-Mofty et al. 1991, Gardner 1996). The lesion is exclusively located in the gingiva or oral mucosa (Guralnick et al. 1983, Buchner & Sciuba 1987).

The tumor was first described by Kuru (1911). In a review by Mintz et al. (1999) covering the period between 1959 and 1989, only 33 cases were documented, while Redman et al. (1994) found 55 cases in the Anglo–Saxon literature. Posteriorly, Zhu et al. (1995) identified 42 cases in the Japanese literature, corresponding to the period between 1967 and 1992. Reichart et al. (1995), in a review of 3677 cases of ameloblastoma, only 2% were found to possess peripheral ameloblastomas. Likewise, Gurol and Burkes (Gardner 1996) reviewed 205 ameloblastomas in the period from 1956 to 1994, and found only eight cases to correspond to peripheral ameloblastoma. Posteriorly, in a 2002 review conducted by Philipsen et al. (2001), peripheral ameloblastoma was seen to account for 2–10% of all ameloblastomas.

One of the main uncertainties regarding the origin of this tumor, although it is believed to derive directly from the lining epithelium or from remains of the dental lamina located in non-bony soft tissues (Moskow & Baden 1982, Kaneko & Ueno 1986, Horowitz et al. 1987, Gurol & Burkes 1995, Ide et al. 2002). Histologically, peripheral ameloblastoma is similar to intrabony ameloblastoma, although the former is less aggressive (Buchner & Sciuba 1987).

The maximum incidence is observed between the fifth and sixth decades of life, with an average patient age at initial appearance of the lesion of 50 years. A slight male predominance has been reported (Gardner 1996). In most cases (65%), the lesion is located in the mandible, particularly on the lingual aspect of the premolar zone (representing 32.6% of all locations) (Philipsen et al. 2001). In the upper maxilla the most frequent location corresponds to the anterior zone for Japanese patients, while in Caucasians it corresponds to the region of the tuberosity (Shiba et al. 1983, Orsini et al. 2000). Our study presents and analyses a case of peripheral ameloblastoma located in the gingiva.

Clinical case

A 31-yearold Caucasian male presented with a painless gingival tumor, identified on occasion of a routine exploration. The patient presented a history of duodenal ulcer and irritable colon, with no smoking, alcohol abuse or regular utilization of mouthrinses. The clinical exploration revealed a tumor located in the distal gingiva of 4.7, and measuring approximately 12 mm in diameter. The

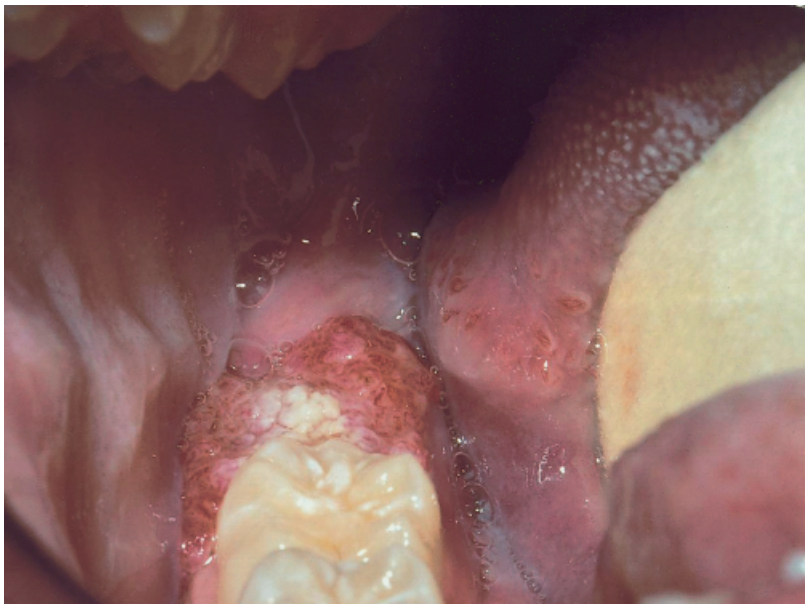

Fig. 1. Clinical image of the lesion, showing a raised tumor mass in the distal zone of 4.7.

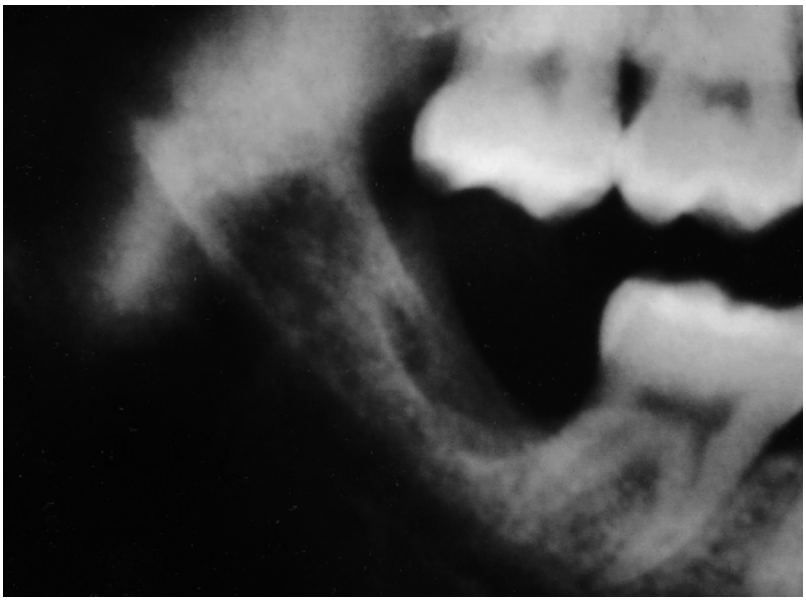

Fig. 2. X-ray image showing retromolar trigone erosion.

Table 1. Possible causes of gingival tumors, excluding pharmacological factors

Malformations	Irritation or traumatism	Infectious processes	Neoplasms
hamartoma	pyogenic granuloma	gingival abscesses	connective tissue
torus	peripheral giant cell granuloma	papilloma	lymphomas
angioma	fibroma		metastasis

growth was slightly erythematous, of firm consistency and presented an irregular surface, without signs of ulceration or fistulas (Fig. 1). All the teeth on that side were vital, and the patient referred no tenderness to percussion. The blood test parameters were all normal. The X-ray study revealed slight cortical bone erosion, with no root reabsorption or tooth displacement (Fig. 2). An excision biopsy was performed under local anesthesia, eliminating the tumor lesion and performing curettage of the bony region. The area was sutured and the patient was given instructions on oral hygiene, advising the local application of ice. Anti-inflammatory medication was prescribed (ibuprofen, 600 mg/8 h); no other pharmacological treatment proved necessary. The tumor specimen was immersed in 10% formalin solution for the histopathological study (Fig. 3A–C), which showed the lesion to be covered with hyperplastic squamous epithelium, with islets of epithelial cells located at subepithelial level. The cells in the peripheral zone adopted a palisade distribution, and presented the appearance of a lax reticulum at central level. A fibroblastic stroma was observed between the islets. The diagnosis was peripheral ameloblastoma. More than 2 years later, there have been no recurrences or changes in the zone.

Discussion

Ameloblastoma is a benign but locally aggressive epithelial odontogenic tumor. Three clinical variants have been described for therapeutic purposes: solid/multicystic (86%), single cyst (13%) and peripheral or extraosseous (1%) (Gardner 1996). Peripheral ameloblastoma, also known as extraosseous ameloblastoma or soft tissue ameloblastoma, is a rare odontogenic tumor that should be differentiated from gingival tumors. The lesions are most often located in the region of the lower jaw, and are generally single – although Hernandez et al. (1992) described the case of a 54-year-old male simultaneously presenting two separate peripheral ameloblastomas: one in the gingival of the lower left incisor region, and the other in the lower right incisor zone.

Peripheral ameloblastoma and basal cell carcinoma are presently considered to be the same entity (Buchner & Sciuba 1987). The lesion is often pediculate or sessile, measures 1–2 cm in diameter, and grows slowly. While asymptomatic in the early stages, it posteriorly may produce slight symptoms in some cases (Nauta et al. 1992, Almenar et al. 1999). In our patient the lesion was asymptomatic and was identified on occasion of a routine examination. The tumor has been described as pinkish in color, or erythematous (as in our case) (Gurol & Burkes 1995). The surface may be granular or irregular, and tends to ulcerate.

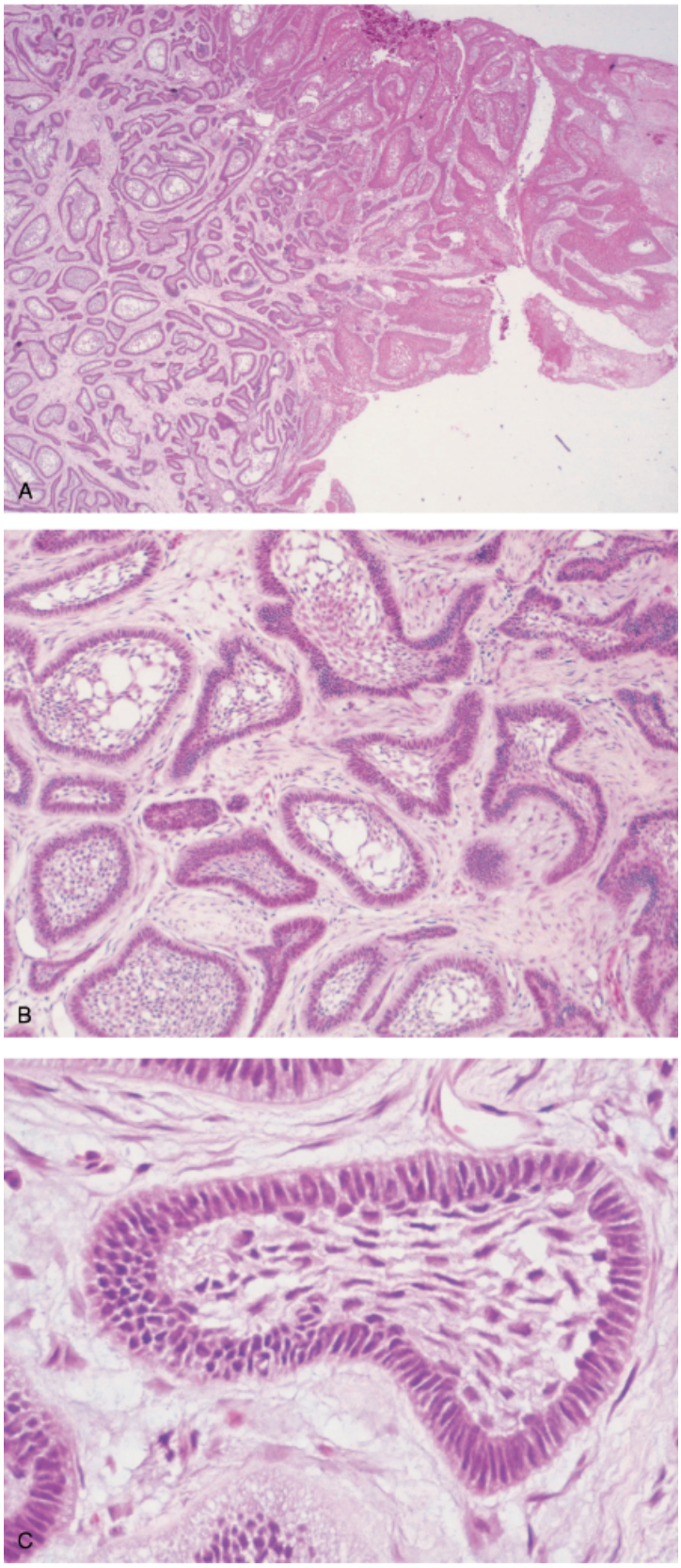

Fig. 3. (A–C). Histological image of the hematoxylin–eosin-stained lesion, covered with hyperplastic squamous epithelium and presenting islets of epithelial cells located at subepithelial level. The cells in the peripheral zone adopt a palisade distribution, and present the appearance of a lax reticulum at central level. A fibroblastic stroma is observed between the islets.

The radiological study may reveal a slight radiotransparency in alveolar bone, resulting from pressure exerted by the tumor growth, as in our patient. If the lesion is located in the interdental papillary region, it may cause separation of the teeth. Reichart et al. (1995) found only five of 73 peripheral ameloblastomas to present radiological evidence of superficial bone reabsorption or erosion.

The differential diagnosis is established with other gingival neoplasms such as papilloma (Kaneko & Ueno 1986), fibroma, telangiectatic granuloma, or peripheral giant cell granuloma (Table 1). In some cases it may be diagnosed preoperatively as squamous cell carcinoma (Buchner & Sciuba 1987). The distinction between peripheral ameloblastoma and odontogenic hamartoma or peripheral odontogenic fibroma may prove difficult. In this context, peripheral hamartoma consists of a collection of inactive odontogenic remains, while odontogenic fibroma is predominantly fibrous. The criteria for diagnosing peripheral ameloblastoma comprise the presence of a tumor located external to bone, with histopathological characteristics corresponding to islets and filaments of odontogenic epithelium, generally similar to the follicular pattern of common intrabony ameloblastoma. The epithelial cell islets exhibit the acanthotic variant of this pattern, with central keratin-forming areas or the cystic pattern. In some lesions the epithelial filaments tend to be continuous with the superficial epithelium or with the epithelium located in the lesion. The epithelial filaments and islets are typically surrounded by fibrous tissue (Shiba et al. 1983, Buchner & Sciuba 1987, Hernandez et al. 1992, Nauta et al. 1992, Almenar et al. 1999, Orsini et al. 2000).

Recurrences are not infrequent; consequently, wide surgical resection is advised. Cases of malignant transformation have been published (Yamamoto et al. 1990, Baden et al. 1993, Philipsen et al. 2001, Tajima et al. 2001, Wettan et al. 2001). Tajima et al. (2001), found those cases of peripheral ameloblastoma presenting as an exophytic mass measuring over 1.5 cm in diameter, with cortical erosion to require wide surgical resection and long-term patient follow-up. Philipsen et al. (2001), in a review of 160 cases, found malignant transformation in six patients. Baden et al. (1993) reported malignant degenerative changes in peripheral ameloblastoma,

and considered the latter to present the same transformation potential as intrabony ameloblastoma. Consequently, peripheral ameloblastoma should not be viewed as a harmless mass.

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