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Case report

Peripheral ameloblastoma: case report and our experience 20 years

Fabio Luciani[1], *Edoardo Papa*[2], *Andrea Leggeri*[2], *Roberta Condò*[2], *Filippo Giovannetti*[3], *Leonardo Calabrese*[2]

[1]Catholic University "Our Lady of Good Counsel", Tirana, Albania; [2]University of Roma "Tor Vergata", U.O. Oral-Maxillofacial Surgery, Post Graduate Course in "Oral Surgery, Roma, Italy; [3]University of L'Aquila, U.O.S.D. Maxillofacial Surgery P.O., L'Aquila, Italy

Abstract. The purpose of this study was to determine the clinical and histological features and treatment of peripheral ameloblastoma (P.A.). Peripheral ameloblastoma is a rare benign odontogenic tumor that concerns soft tissue and have a typical extraosseous localization. Aim of this work is to show its clinical and histological characters, in order to define some useful information for differential diagnosis with other oral neoformations, comparing literature with our data, collected in twenty years of clinical activity of Oral and Maxillofacial Surgery Unit of Policlinico Tor Vergata in Rome. Prognosis of peripheral ameloblastoma is certainly favourable, with a restitutio ad integrum close to 100%. In the period between October 2002 and November 2021, we reported 8 diagnoses of peripheral ameloblastoma. Medium age of the group with diagnosis of P.A. was 71,4 years with a standard deviation: 3,65. P.A.'s incidence in our sample of patients was 0,26%. Peripheral ameloblastoma is a benign odontogenic tumor that requires a careful diagnosis, a complete surgical eradication and adequate follow up, because malignant evolution is rare but possible. (www.actabiomedica.it)

Key words: Odontogenic Tumors; Peripheral Ameloblastoma; Oral Biopsies;

Introduction

Ameloblastoma is the second most common odontogenic tumor, known to be slow-growing, persistent, and locally aggressive (1). This tumor is benign but locally aggressive and is often discovered clinically as a painless swelling or incidentally on a routine panoramic radiograph. It can result in variable symptoms secondary to displacement or destruction of surrounding tissue ranging from mobile teeth, swelling, pain, numbness, and pathologic fractures in advanced stages (2).

The World Health Organization classifies ameloblastoma into 3 subcategories based on clinical presentation, radiographs, histopathology, treatment considerations, and prognosis (3, 4):

1. Intraosseous multicystic or solid type;
2. Intraosseous unicystic type;
3. Peripheral or extraosseous type (P.A.).

Multicystic type represents 86% of all ameloblastomas, unicystic type 13% and the peripheral form 1% (5). Gardner et al. suggested that P.A. have %1.3-10 ratio among all ameloblastomas (8). The diagnosis is based on both the clinical and histologic characteristics. Caution is required during their surgical excision to avoid recurrence.

Peripheral ameloblastoma is a rare and benign extraosseous ameloblastomas which effects soft tissues (8). It is also known as Extraosseous Ameloblastoma, Ameloblastoma of the soft tissues, Gum Ameloblastoma, Ameloblastoma of mucosal origin (6).

Typical site is the mandible in 65% whereas maxilla is interested in 35% (5). In the maxilla the tumour is often found on the tuber, in the mandible on the lingual area near premolar teeth. Sex prevalence is 2:1 for man, and the mean age is 53 y for men and 51 y for females (4,5,6). The etiology of this tumour is still not clear. Some Authors have pointed out in some cases a DNA hybridization in presence of Papillomavirus 16 and 18.

Histologically P.A. presents a squamous epithelial layer within a dense connective stromal tissue. During its growth tumour acquires an arrangement in islands of tissue that show a central part rich of stelliform cells. The most common histological form presents follicular or plexiform cells. Epithelium is always well outlined but it's not recognizable a capsule of fibrous tissue therefore very often the tumour epithelium is in close contact with the surrounding mucosa. Mitosis are rare like also in intraosseous ameloblastoma. Very frequent instead is the presence of a lymphocytic inflammatory infiltrate within stromal tissue (4,7). The lesion is usually single, sessile or stalked, with a smooth surface and a diameter between 1-2 cm. P.A. can have a colour like normal mucosa or it can be dark-red. In non-edentulous patients can be found close to interdental papilla (4). The lesion is usually asymptomatic and shows a slow and exophytic growth (4,7).

Radiographically it appears to be external to cortical bone. In some cases, it's possible to point out the cupping or saucerization effect, because the tumour due to its pressure effects a concavity on the osseous surface (4,6,7).

P.A. have a non-invasive behaviour, but it has however a recurrence rate of 16-19%.

Case report

On July 2019 M.F., male, 64 years old comes to our observation, in the Oral and Maxillofacial Surgery Unit of Policlinico Tor Vergata in Rome. He presented a neoformation in the last 15/18 months in the premaxilla area that didn't allow to him the common feeding (Figure 1).

At medical history the patient reported a slight hypertension in treatment with β-blockers in a good hemodynamic offset. He denied other medical and surgical pathology and to have allergy to common drugs used in dentistry. He reported he has been a heavy smoker and to have given up smoking for 28 years. At the oral inspection he presented a bulky exophytic neoformation, strongly edematous, not aching, not ulcerated, with a hard-elastic consistency that impeded normal mastication

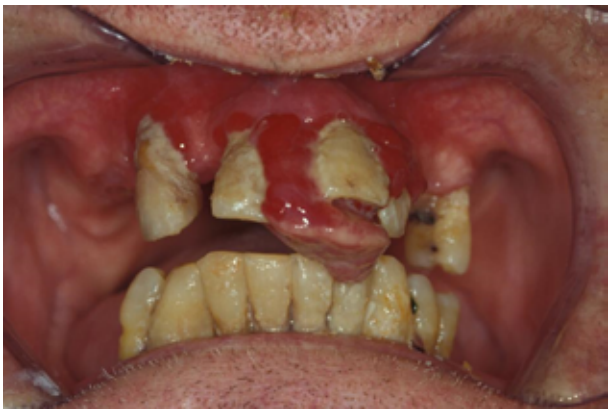

Figure 1. Preoperative view of Peripheral Ameloblastoma.

Teeth 1.1, 2.1 and 2.2 appeared involved by the neoformation and presented a third degree of mobility. He was affected by a severe chronic periodontitis with high levels of plaque and bleeding index. It was prescribed an orthopantomography, periodontal therapy associated with antiseptic therapy with chlorhexidine 0,2%. Afterwards the radiographic evaluation it was decided to remove the neoformation and the impaired teeth. Patient's informed consent has been obtained.

Detachment of mucoperiosteum flap showed a close adherence of the neoformation with teeth 1.1, 2.1 and 2.2 and residual part of alveolar bone so they were removed in one-piece (Figure 2).

Bone contour was regulated with a bone-cutting tungsten carbide cutter, gum plastic was performed and after a careful haemostasis, a single points of silicone silk suture was made. The suture was removed at twelfth day. Check healing at 30 days (Figure 3).

The histologic appearance of Peripheral Ameloblastoma, observed by light microscopy after appropriate treatment and colouring based on haematoxylin and eosin, showing proliferation of cellular elements of numerous small polygonal, with hyperchromatic

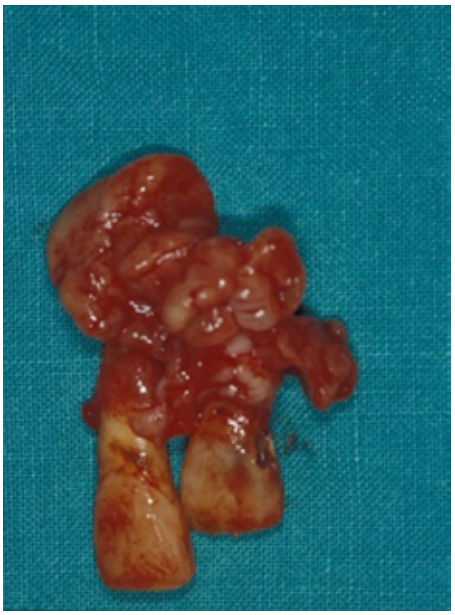

Figure 2. Neoformation, teeth 1.1, 2.1 and 2.2 and residual part of alveolar bone once removed in one-piece.

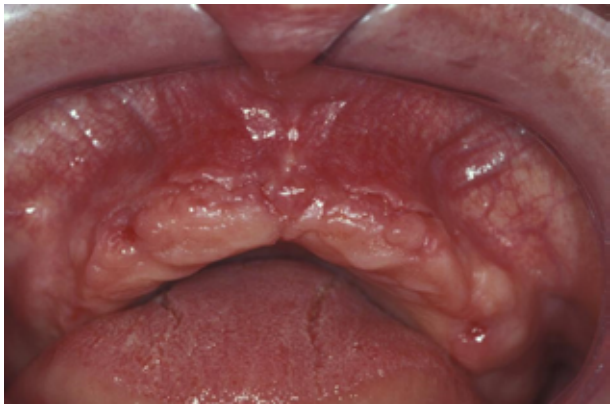

Figure 3. Intraoral view after healing at 30 days.

nucleus identified as the pre-ameloblasts devoid of secretory activity, with reverse nuclear polarity. Mitotic divisions are not observed, with tumor basaloid largest within the cytoplasm (Figure 4).

This clinical case report gave us the opportunity to retrospectively evaluate P.A.'s incidence in twenty years of clinical activity in the Oral and Maxillofacial surgery Unit of our Hospital. Between 2002 and 2021, among 3048 cases of oral lesions diagnosed in the Department of Oral-MaxilloFacial Surgery, University

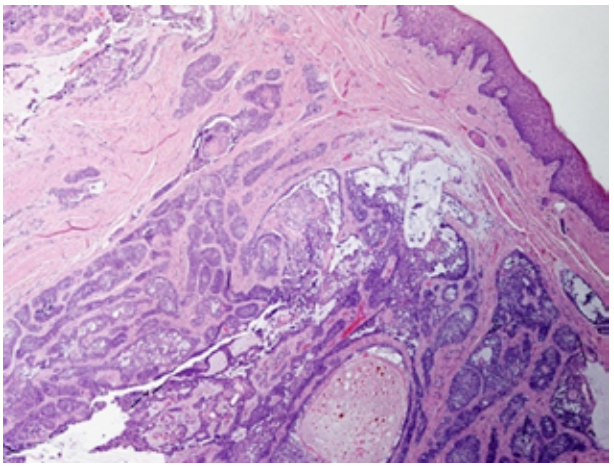

Figure 4. Histologic view of Peripheral Ameloblastoma.

of Tor Vergata Roma; 52% were male (1585 subjects) and 48% were female (1463 subjects). Medium age was 55 years with a standard deviation (S.D. 18, older in female, 57 years, than in male, 53 years (p=0.001).

In the period between October 2002 and November 2021 we reported 8 diagnosis of P.A., 3 female (medium age 71,5 years, S.D. 2,12) and 5male (medium age 71,6 years, S.D. 4,43). Medium age of the group with diagnosis of P.A. was 71,5years with a S.D.: 3,65. Peripheral ameloblastoma's incidence in our sample of patients was 0,26%. Our data are in agree with the data in the international literature, they show that P.A. was a neoformation of rare clinical finding. The histopathological diagnosis was "peripheral Ameloblastoma".

Univariate analysis was performed to evaluate the prevalence of pathologies. The Chi-squared test was used for comparisons between groups and the t-test between means. A P-value of <0.05 (two tailed) was considered statistically significant.

Discussion

Differential diagnosis is with giant cell peripheral granuloma, peripheral odontogenic fibroma, ossifying peripheral fibroma, pyogenic granuloma, fibroma, papilloma, epulis and basal cell carcinoma (B.C.C.) (4,6,7,14,17,18).

P.A. and B.C.C. present some histological common aspects. Both the lesions show a proliferation of basal cells organized in nest surrounded by stromal fibrous tissue.

P.A. can be recognized by the presence of nuclei in the upper portion of the cells whereas in B.C.C. these nuclei are localized in the lower part of the cells cytoplasm (4). Another difference can be emphasized by the immunohistochemical analysis: P.A. is positive for Keratin 19 and negative for Ber-EP4 on the contrary of B.C.C. (4).

Immunohistochemically, P.A. differs from IAs for a lower positivity to Ki-64. Ki-64 low values are sign of a lower malignancy of PA compared to IAs and this suggests a good prognosis (11).

P.A. is a rare and benign odontogenic tumour with extraosseous site. First case has been reported in literature by Kuru in 1910 (12).

From epidemiological point of view Garner et all. (13) have remarked the most interested decade is that between 60 y and 70 y with a rate of 2:1 for men.

In a literature review is reported that P.A. can be inclined to invade alveolar bone (14). Mintz and Manor have pointed out the most affected area is anterior region of mandible (15,16). P.A. appears typically like a small neoformation of pink gum. Diagnosis is made by biopsy and the support of radiographic examinations (6). The treatment consists in complete surgical excision of tumour. Ampalagan et. Al suggests a primary surgical excision of peripheral ameloblastoma and any recurrent lesions, involving the lesion in its entirety, including a cuff of normal tissue, generally without the removal of teeth and afterwards a long-term follow up of at least 10 years of both primary and recurrent lesions (21). Some other authors however stress the importance of annual follow-up for at least 8 years (17). This, as we said before, due to some cases reported in literature that showed a malignant evolution (19,20).

In conclusion it's possible to consider P.A. a benign odontogenic tumour with a good prognosis that requires however a careful diagnosis, a surgical removal approach and follow-up to observe rare malignant evolution (19,20).

Conflict of Interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

Informed Consent: Written informed consent was obtained from the patient concerned.

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Correspondence:
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Andrea Leggeri
Viale Trieste 16
00019, Tivoli (RM), Italy
Phone: +393388445970
E-mail: dr.andrealeggeri@gmail.com
Orcid number: 0000-0003-0927-4254