

Case Report

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Peripheral Ameloblastoma of the Mandible: A Rare Case Report

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Abstract

The purpose of this study is to present the authors experience in the management of a rare clinical condition. A 31-year-old female was referred to Maxillo-facial Surgery Department, reporting the presence of a growing mandibular lesion. The radiological examination confirmed the presence of osteolytic unicystic lesion that was excised and after a clinical pathological exam, diagnosis of ameloblastoma was established. Ameloblastoma represents 1% of all oral tumors and 11% of odontogenic tumors and it has been reported to be more prevalent in Asian or African-Caribbean individuals. Most of patients first present symptoms between the ages of 30 and 40 years; the most common presentation is a painless and gradually growing swelling. Peripheral Ameloblastoma (PA) represents a rare subtype, comprising only 1% to 5% of all ameloblastomas, and it features more benign behavior than other types, characterized by minimal bone involvement. Differential diagnosis should include reactive swelling such as peripheral giant cell granuloma, peripheral odontogenic fibroma and others. Treatment for ameloblastoma could include a traditional approach using extensive resection but also more conservative techniques. Due to its rarity, there is no strong consensus relating to the surgical margins. In this case a revision surgery wasn't necessary and regular controls were performed as planned. Since literature described it, the possibility of recurrence should be considered mandatory.

Keywords: Peripheral ameloblastoma; Rare disease; Head and neck; Extraosseus; Gingival lesion.

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Introduction

Ameloblastoma represents 1% of all oral tumors and 11% of odontogenic tumors. They are located much more frequently on mandibular bone, rather than maxillary bone, more often in the posterior region instead of the anterior one, except for the African black race which is localized more frequently at the symphysis. It's extremely rare to find these tumors in extra-mandibular locations, because of the association between their etiology and dental structures. However, extra bony localizations at the alveolar mucosa known as peripheral or extraosseous ameloblastomas are described. Based on the aspects found in pathological anatomy, it is possible to recognize the following four histological subtypes: conventional (also called solid or multicystic), unicystic, desmoplastic, and peripheral (extraosseous). Current evidences support the contention that peripheral ameloblastoma is the most common epithelial odontogenic tumor of the gingival/alveolar mucosa, but it represents 4.5% of ameloblastomas. It is characterized by a histological picture that can be superimposed on follicular ameloblastoma, that is an unicystic subtype. The surface of the peripheral ameloblastoma may be ulcerated, usually does not infiltrate the bone and does not present radio logically with features of radio transparency. It is limited to the gum or alveolar mucosa; it infiltrates the surrounding tissues, mostly the gum connective tissue, but does not involve the underlying bone. Peripheral ameloblastoma originates from residues of the dental plate, the so-called glands of Serres, pluripotent cells located in the basal cell layer of the mucosal epithelium and in the minor salivary glands and often a continuity with the basal cells of the gingival epithelial plate. Almost all oral pathologists and dentists are experienced in diagnosing peripheral/extraosseous ameloblastoma but unfortunately, several authors chose to use the terminology "peripheral alveolar type or typical epulis" for ordinary intraosseous ameloblastomas that breached the alveolar bone, grew in the gingiva and exhibited the same clinical appearance as epulis. The most common clinical presentation is a painless and gradually growing swelling, not involving mandibular bone. Peripheral ameloblastoma usually shows several histologic characteristics of an intraosseous infiltrating ameloblastoma, but the disease with histologically low-grade malignant features is extremely rare. Extraosseous ameloblastoma was first reported in the literature by Kuru in 1911 [1], and nowadays a case report by Stanley and Krogh published in 1959 is considered to be the first well-established case of peripheral ameloblastoma [2]. Even more frequently extraosseous ameloblastoma is an incidental finding during a routine dental examination; as such, to make the correct diagnosis at its first presentation is always challenging for medical doctors. More specific radiological imaging modalities, such as a CT scan and MRI, should be included in the diagnostic process as they can most of the time demarcate the lesions better than a traditional bi dimensional orthopantomography performed by the dentist. This is because in most cases, the lesions are located near the bone and within the normal tissue margins. Bone involvement of the peripheral ameloblastoma is usually represented by cupping or saucerization that refers to a depression made from the pressure of the tumor on the bone. However, patients affected by peripheral ameloblastoma with bone involvement are a rare finding and it is usually mild with no neoplastic invasion or marrow infiltration. Thanks to the dense fibrous tissue of the gingiva and periosteum and the cortical plate of the alveolar pro-

cess, a well defined physical barrier could be represented to the bone infiltration of the peripheral ameloblastoma. The biological features of peripheral ameloblastoma are similar with that of a hamartoma or persistent hyperplasia rather than that of a neoplasm. Moreover, the clinical characteristic of peripheral ameloblastoma, for example the course of disease, lesion growth and patient symptoms, are not specific for peripheral ameloblastoma and this makes it difficult to distinguish between this kind of neoformation and other type lesions growing on the gingival mucosa. Neof ormations that should be considered with similar manifestation could be epulis, fibroma, squamous cell carcinoma, and lymphoma and all of these are lesions that should be investigated when thinking about differential diagnosis. For intraoral lesions, ultrasonic examination is rarely performed and the tumor could be incorrectly diagnosed as epulis or periapical fistula. This makes this way of studying this lesion not reliable in helping for peripheral ameloblastoma diagnosis. Instead, the diagnosis of PA should be considered if: the mass grows slowly without pain and trismus; no cauliflower-like changes in the superficial mucosa could be observed; less mucosal lesion than submucosal mass is represented; CT scan imaging or MRI shows clear demarcation between bone and medial pterygoid muscle, uniform density and less enhanced images. In these cases, fine needle aspiration or incisional biopsy is strongly recommended to prevent unnecessary surgical intervention such as overtreatment. When approaching small lesions, conservative supra-periosteal surgical excision with an adequate margin free from disease is recommended even in case diagnosis is not confirmed. While when large lesions are approached, incisional biopsies should be performed since differential diagnosis include malignant tumor before proceeding with surgical procedures. Partial bone should be resected if cup-like or saucerized bone involvement is detected during surgery. Anyway, after surgical treatment, continuous follow up is necessary due to the possibility of late recurrence or malignant changes, even if they are rarely described.

Materials and methods

A 31-year-old female was referred to Maxillo-facial Surgery Department, University of Siena, in July 2020 from her dentist. The patient reported the presence of a mandibular growth, that she first noticed one month earlier. She presented no pain or bleeding in that area. Anamnestic information about the patient revealed that patient has no systemic disease and no drug use; she also reported no use of cigarette and alcohol. Thanks to an extraoral examination a swelling in the left emimandible region was detected. No lymphadenopathy was noticed. Through the intraoral examination the presence of a swelling, that spanned from the left second premolar to the left third molar region was observed; sizes were around 3 x 1,5 cm and overlying mucosa was flushed. No intraoral fistula was noticed and there was no pain at the palpation of the lesion. A dental CT scan was performed by the patient. The radiological examination confirmed the presence of an osteolytic unicystic lesion ranged from left second premolar to left third molar in mandibular bone; no cortical bone invasion was observed (Figures 1-3). According to clinical and radiological evidences, the authors decided to proceed with lesion removal.

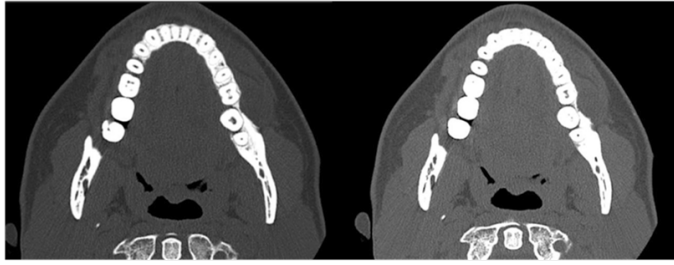


Figure 1: Image showing retrograde pyelography. Ureter appeared to be dilated with? vujo.

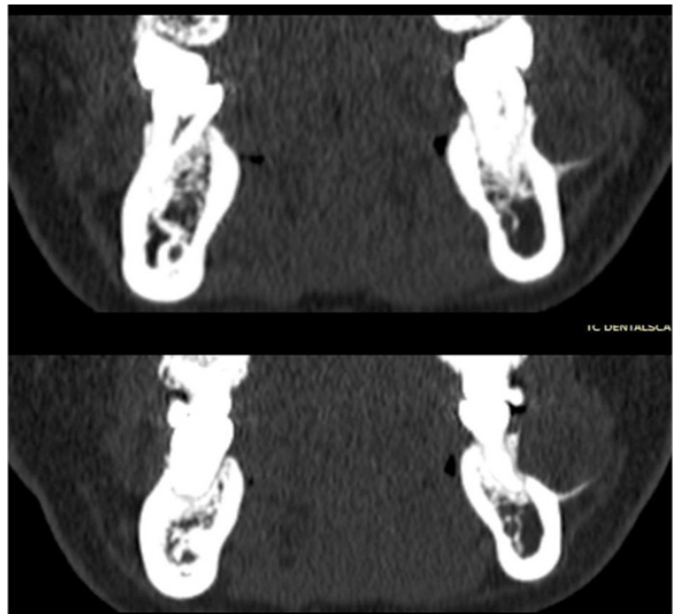


Figure 2: Preoperative CT scan: coronal view of the unicyclic lesion.

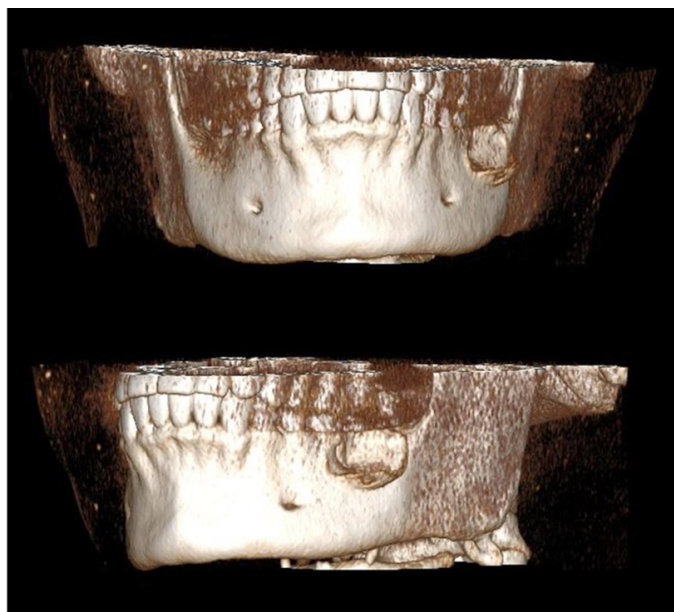


Figure 3: Preoperative CT scan 3-dimensional reconstruction.

Results

In September 2020 a surgical procedure was performed. Using transoral approach, after local anesthesia infiltration, an interpapillary incision was performed between first premolar and retromolar trigone of left hemimandible. The unicyclic lesion was identified, adherent to cortical bone and excised (Figures 4,5). Moreover, the lesion was formalin-fixed and prepared for being submitted to a definitive histological examination. No major or minor intraoperative complications had occurred and the operative time was around 1 hour and half. The patient hospitalization lasted for about 1 night. After clinical, histological and radiological examinations lesion was described as a “*peripheral unicyclic ameloblastoma with intraluminal growth*”. At the follow-up examination scheduled two weeks later there was no recurrence and patient had no complaint. At the follow-up performed one month and two months after the excision there’s was still no evidence of recurrence. Further surgical approach (a radical resection) was deemed unnecessary and wound healing was good (Figure 6). Additionally, patient was informed about the importance of regular follow-up that were made during one year after excision for early diagnosis of possible recurrences. After two months the lesion area was clinically unchanged. A second CT performed 10 months later did not show the superficial bone resorption, confirming the tumor was not infiltrating the bone. Patient consent form was obtained by the patient for participation in this case report.

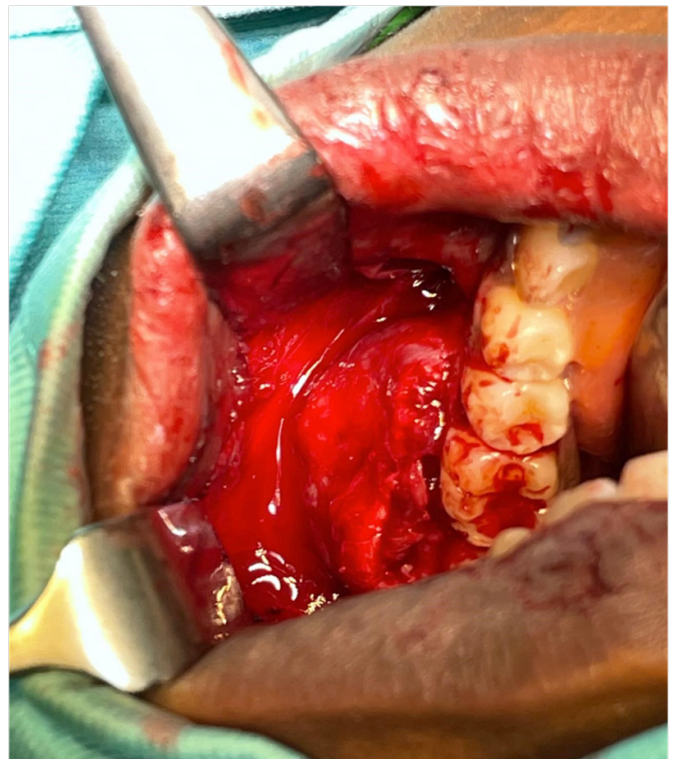


Figure 4: Intraoperative view of the unicyclic lesion.



Figure 5: Excised unicystic lesion.



Figure 6: View of postoperative healing of surgical site.

Discussion

Ameloblastoma represents 1% of all oral tumors and 11% of odontogenic tumors [3]. Current evidence supports the contention that PA is the most common epithelial odontogenic tumor of the gingiva/ alveolar mucosa [4]. Most ameloblastomas are characterized by benign behavior and slow growth, with features of local aggression due to their infiltrative ability that can result in significant clinical conditions that may require an aggressive surgical approach. Ameloblastomas are considered locally aggressive tumors that through bone can also invade surrounding soft tissues if not treated in time, however, ameloblastoma remains a benign tumor, therefore lymph node or distant metastases are rare and it define the behavior of the neoplasm as malignant, as happens in less than 1% of cases. Most of patients usually present symptoms for the first time between the ages of 30 and 40 years; according to literature, our case patient was in fact 31 years old. Despite African people may often present it at an earlier appearance; this tumor has been reported to be more prevalent in Asian or African-Caribbean individuals which was another characteristic of our patient. New neof ormations can be classified depending by histological features, and, according to the most recent World Health Organization (WHO) classification system [5], it is possible to distinguish four types of ameloblastoma: conventional (also called solid or multicystic), unicystic, desmoplastic, and peripheral (extraosseous). Peripheral ameloblastoma (PA) represents a

rare subtype, comprising only 1% to 5% of all ameloblastomas, and it features more benign behavior than other types, characterized by minimal bone involvement. This makes diagnosis of the peripheral subtype an important finding because treatment may consequently be much more conservative. Moreover, peripheral ameloblastomas are generally common at one location. Only Hernandez *et al.* reported one case that was placed at two different locations at the same time [6]. These benign tumors usually occur primarily in mandibular premolar area, followed by lower anterior and maxillary tuber areas. Mean age of appearance is 52.1, but it can be seen at ages ranging from 9 to 92. In contrast to other ameloblastomas subtypes, they are more common in men with a male/female ratio of 1.9:1 [7]. The most common presentation is a painless and gradually growing swelling. The deep margin does not tend to invade bone extensively, nevertheless radiographically it may result as a scalloped lesion. On the other hand, sometimes peripheral ameloblastoma could invade bone structures, create nerve damage that could involve inferior alveolar nerve. However, innovative microsurgical repairing techniques exist today to fix the issue [8,9]. Peripheral ameloblastoma differential diagnosis should include reactive swelling such as peripheral giant cell granuloma, peripheral odontogenic fibroma, peripheral ossifying fibroma, papilloma, pyogenic granuloma, epulis, and fibroma [10]. Basal cell carcinoma of the gingiva is also considered as an analogous neof ormation to peripheral ameloblastoma. However, the way these diseases manifest is similar. Radiological and histological investigations are necessary to make differential diagnosis through specific characteristics. Malignant transformation of the PA is exceedingly rare [11].

Consequently, 3-dimensional imaging such as Computed Tomography (CT) or Magnetic Resonance Imaging (MRI) helps to demarcate the lesions accurately. Despite this characteristic appearance, certain diagnosis requires histological examination to exclude other peripheral odontogenic tumors. In this case patient's characteristic and radiological findings were suggestive of peripheral ameloblastoma so the authors decided to proceed with surgery without histological diagnosis. Rationale for the management has been questioned. In many cases, the traditional approach using extensive resection is avoided in favor of the more conservative techniques. However, due to its rarity, there is no strong consensus relating to the surgical margins and, although less aggressive than other types of ameloblastoma, excision using a local conservative approach [12] or more extensive aggressive treatment [13] has been suggested.

Conclusion

The purpose of this case report is to present our surgical experience approaching a rare pathology. In the reported case PA appeared as a swelling range from left second premolar and left third molar region and the patient didn't have symptoms. According to literature, after the tumor was locally excised and during follow-up, no bone's invasion was observed, confirming the lesion was completely extra osseus. Further surgical approaches were judged as unnecessary over treatments. Although recurrence rate of peripheral ameloblastomas are low, long-term follow-ups are suggested [14]. It was reported that a benign peripheral ameloblastoma was recurred as an ameloblastic carcinoma [15]. Additionally, a metastatic peripheral ameloblastoma and a recurrence of a peripheral ameloblastoma which shows dysplasia

was reported too [16,17]. Due to such information, long term and regular controls should be mandatory.

Declarations

Conflicts of interest: The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

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