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Case Report

Mandibular unicystic ameloblastoma revealed by florid epulis of the gum: Case report

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ABSTRACT

Ameloblastomas are quite frequent odontogenic tumors, they can be intraosseous or peripheral to the gum, it's rare to find an intraosseous ameloblastoma with gingival extension. Here, we report the case of a 40 years old woman who presented with an exophytic gingival lesion at the gum level of the 46 tooth that was extracted at a quack 01 years before her consultation due to pain in this area. Clinical and radiological examination revealed the presence of two tumors in the same area.

The patient benefited from an excision of her gingival tumor and an enucleation of her intraosseous tumor. Histopathological and immunohistochemical examinations revealed that the respective tumors were a spindle cell epulis and an intraluminal unicystic ameloblastoma of different origins, leaving the question and search for the relationship between these two tumors. The postoperative course was uneventful during the 12-month follow-up period after surgical treatment.

1. Introduction

Ameloblastoma is a rare benign odontogenic tumor with a slow evolution. It represents 1% of maxillary tumors and 11% of odontogenic tumors [1]. It is a benign tumor with local invasive and evolutionary potential, usually occurring between the third and fifth decade. Men are more affected than women. The mandibular location is more frequent than the maxillary [2,3]. It is a tumor originating from epithelial and/or ectomesenchymal elements that participate in tooth formation [4].

Peripheral ameloblastomas originate from remnants of the dental lamina of superperiosteal location and will develop outside the bone, in the soft tissue. In contrast to intraosseous ameloblastomas, which rarely extend into the overlying mucosa [5].

Epulis is an inflammatory pseudotumor of the gingiva with no degenerative potential and a recurrent tendency. It is a gingival growth resulting from chronic local irritation or endocrine variations [6].

The presence of two tumors in the same location makes the diagnosis difficult, raising the question of whether these lesions are peripheral ameloblastomas that invade the underlying bone or intraosseous ameloblastomas that extend to the overlying gingiva, or are unrelated tumors. The final diagnosis requires a histopathological and immunohistochemical study of the surgical specimens.

We report a case of a woman with an intraosseous ameloblastoma discovered incidentally following examination of an epilus present in the same anatomical location, this work has been reported in line with the SCARE 2020 criteria [24].

2. Case report

40-year-old woman, without any particular pathological history, referred by a dental doctor to our department for florid epulis of the right mandibular gum. The patient reported that the gingival lesion appeared 2 months after the extraction of the 46 tooth due to pain and mobility of this tooth. Exobuccal examination revealed a right jugal swelling opposite the mandibular horizontal branch, hard, fixed to the bone plane, 08 cm in length, with no disturbance of facial sensitivity. The endobuccal examination reveals a pedunculated gingival tumor mass, originating from the gingiva of the 46 tooth extracted from a quack, 04 cm long, ulcerated, not painful, bleeding at contact at its base, there is also a hard vestibular swelling, fixed to the bone, going from 44 to 47 tooth, with bleeding mucosa opposite, mobility of 45 tooth [Fig. 1].

A cone beam was performed showing radiolucency extending in the right horizontal branch from tooth 42 to tooth 47 with disappearance of

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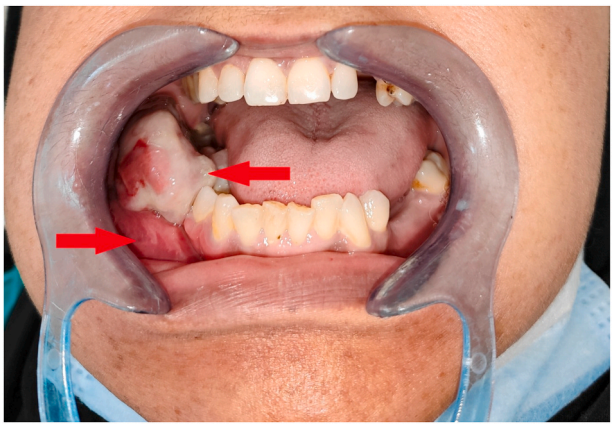

Fig. 1. Preoperative image showing the epulis and the swelling of the vestibule below.

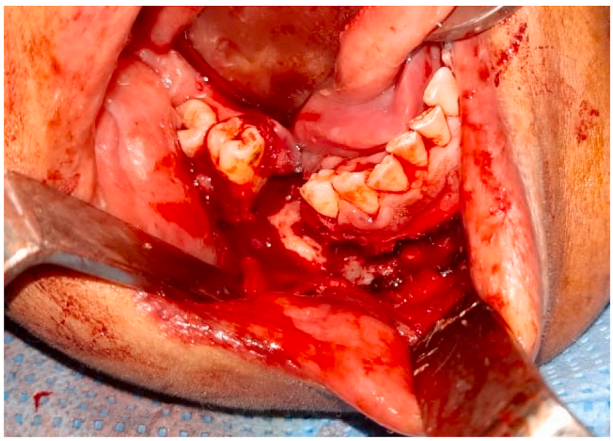

Fig. 3. Intraoperative image showing the cystic cavity after enucleation and removal of the epulis.

the lingual bone cortex from 47 to 45 and disappearance of the vestibular bone cortex from 47 to 43 with the alveolar nerve being pushed back from 47, the lesion respects the basilar margin **[Fig. 2]**.

Under general anesthesia, we performed an exeresis of the two tumors with extraction of the 45 mobile tooth. The enucleated tumor was a solid, friable mass, invading the alveolar nerve that had to be sacrificed and leaking a yellowish fluid during its removal **[Figs. 3 and 4]**.

Anatomopathological examination revealed a florid epulis with a positive and diffuse AML marker for the gingival tumor, as well as an intraluminal unicystic ameloblastoma for the bone tumor.

The patient had no complications after her surgery during the 12-month follow-up period.

3. Discussion

Ameloblastoma is considered a rare disease, accounting for 1% of all head and neck tumors, and is more common in Africa, China and India [7,8]. Ameloblastoma occurs more frequently in men than in women in the age range of 30–60 years with an average age of 36 years [8,9]. It is an odontogenic tumor derived from the epithelium involved in the formation of the tooth, the enamel organ.

Almost 80% of ameloblastomas are located in the mandible, usually in the posterior part of the horizontal branch, while only 20% are found in the maxilla [10]. Generally, ameloblastoma is discovered by chance following an X-ray taken for another reason, as in our patient's case [11].

Clinically, the symptoms are nonspecific, usually manifesting as a prominent jugal swelling at the vestibular level rarely accompanied by pain. Different types of ameloblastoma are described in the literature, we have: multi-cystic/infiltrating is the most frequent type of ameloblastoma, unikystic diagnosed in our patient, peripheral/extraosseous ameloblastoma and desmoplastic type [7].

The unicystic form represents 10–15% of ameloblastomas, it occurs during the second or third decade and appears on radiography as a clear image with regular walls, most often at the level of the mandibular angle associated with an included tooth, it is admitted that this type is formed following a neoplastic modification of the wall of an odontogenic cyst, it can take on significant dimensions before its discovery because it remains asymptomatic for a long time [12,17,18]. The cyst is composed of an epithelium of 5–15 layers with luminal and stellate cells in the center with an acanthomatous appearance. There are three forms of unicystic ameloblastoma: an intraluminal form, a luminal form and an intramural or plexiform form [13,14].

Unicystic ameloblastoma is characterized by an inflammatory transformation that can alter the typical appearance and lead to the wrong diagnosis of a radicular inflammatory cyst, which leads us to systematically look for an ameloblastic component in the anatomopathological study. Immunohistochemistry can be of great help, as we observe an inconstant positivity of anti-CD56 and anti-calretinin antibody [15,16]. This type of tumor is considered less aggressive than other ameloblastomas and can be treated like odontogenic cysts by simple enucleation.

In the literature, recurrence after conservative treatment is reported between 10 and 25%, but this report does not specify the histological subtypes, for this reason, several authors advocate resection, which may

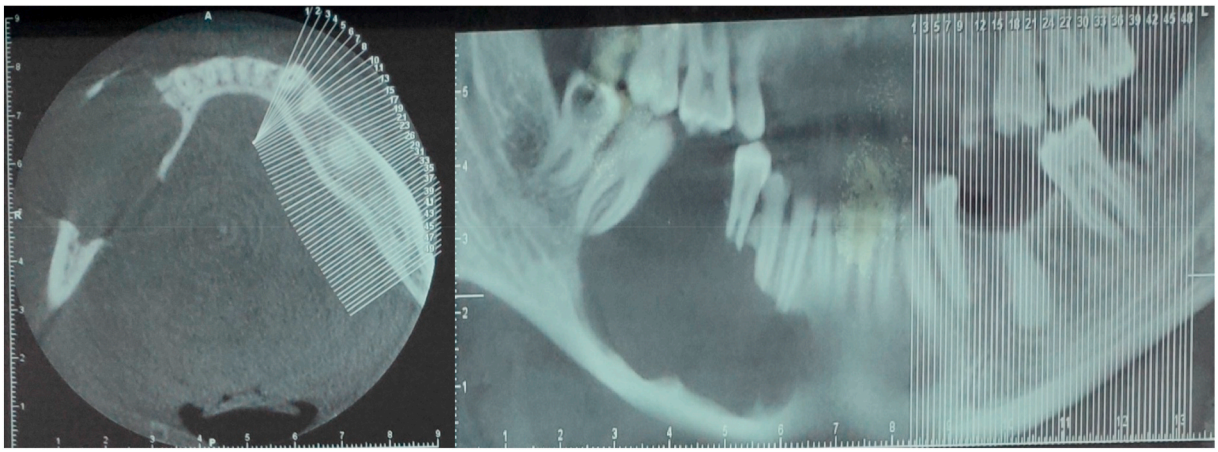

Fig. 2. Cone beam radiography showing the intraosseous cystic image at the level of the mandibular horizontal branch.

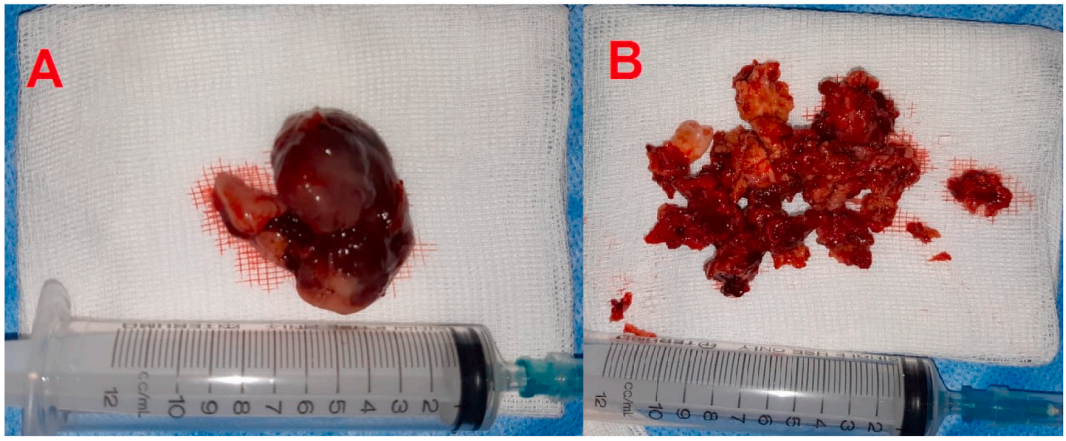

Fig. 4. Image showing the operative specimens: A. the epulis. B. Cystic content.

be unnecessary [20,21]. Li et al., in his study, found that recurrence has a relationship with histological subtypes, those that invade the fibrous wall have a rate of 35.7%, but the others have a rate of only 6.7% [19].

In our case, we wondered if there is a relationship between the occurrence of unicystic ameloblastoma and florid epulis since they originate in the same location (opposite to the 46 extracted) and are of synchronous occurrence.

The epulis is a benign tumor of the gum, it appears more in women, most often at the mandibular level following a chronic local irritation or hormonal perturbations, clinically it appears as a red fleshy mass very vascularized, bleeding on contact, circumscribed sessile or pediculated, several histopathological forms are found: peripheral ossifying fibroma, fibroma/fibrosis, with giant cells, pyogenic granuloma, with hyperplastic squamous epithelium, with granulation tissue and peripheral odontogenic fibromas. Treatment consists of surgical excision [22,23].

No anatomopathological or immunohistochemical relationship was found between the two tumors, which means that they are of different origin. It is assumed that the occurrence of the ameloblastoma originating from the tooth triggered pain, which led the patient to extract it at a quack without prior radiological examination, the ameloblastoma caused irritation of the gum which was a factor favoring the appearance of the epulis, moreover that it is a woman in a period of genital activity.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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None.

Author contribution

Amine kaouani: Corresponding author writing the paper. Ouassime kerdoud: writing the paper. Rachid Aloua: writing the paper. Faiçal Slimani: Correction of the paper.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Guarantor

Amine Kaouani.

Declaration of competing interest

The authors declare no conflict of interest.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102422.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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