

Up-to date review and case report

A histologic variant of ameloblastoma: the acanthomatous type

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Abstract – Introduction: Ameloblastoma is the most common aggressive benign odontogenic tumor of the jaws. This kind of tumor arises from dental embryonic remnants. **Clinical case:** We report a case of acanthomatous ameloblastoma of the mandible occurring in a 79-year-old male patient. Through this case, we describe clinical and radiological findings in ameloblastoma, discuss different variants of this tumor, and emphasize the histological diagnosis. **Discussion:** Among the various types of aggressive benign odontogenic tumors, acanthomatous ameloblastoma represents a subtype of the solid multicystic ameloblastoma with specific microscopic features. Its course is controversial. **Conclusion:** Although the diagnosis of acanthomatous ameloblastoma is based on histologic features, dentists and oral surgeons should consider this possibility in patients with an aggressive tumoral process.

Mots clés :
améloblastome
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Résumé – Une variante histologique d'améloblastome : le type acanthomateux. Introduction : L'améloblastome est la plus commune des tumeurs agressives odontogènes bénignes des maxillaires. Elle provient des vestiges embryonnaires de l'organe dentaire. **Observation :** Nous rapportons un cas d'améloblastome acanthomateux de la mandibule chez un patient âgé de 79 ans. A travers cette observation clinique, nous décrivons les caractéristiques cliniques et radiologiques de l'améloblastome en rappelant les différents types histologiques particulièrement le type acanthomateux. **Discussion :** L'améloblastome acanthomateux est une variante de l'améloblastome solide multikystique qui possède des caractéristiques microscopiques spécifiques. Son évolution reste controversée. **Conclusion :** Même si son diagnostic est histologique, le chirurgien-dentiste doit savoir évoquer une telle hypothèse face à un processus tumoral agressif.

Introduction

Ameloblastomas account for about 1% of all tumors of the jaw and mainly develop during the third to fifth decades of life [1]. Ameloblastomas exhibit distinct microscopic characteristics and variable histological patterns. Histopathologically, six patterns are discernible: follicular, plexiform, acanthomatous, granular cell, basal cell, and desmoplastic types [2]. The reported case illustrates hallmarks of acanthomatous ameloblastoma.

Clinical case

A 79-year-old man consulted for dysphagia in our Oral, Maxillofacial and Esthetic Surgery Unit. Intraoral examination found a hard and painless swelling extending from the lower right third molar to the ramus. The mucosa over the swelling appeared normal. No tooth displacement was noted but dental health was poor. The extraoral examination did not show any facial asymmetry. The patient looked in good health and did not have any significant medical history. He was unaware of the swelling noticed during intraoral examination.

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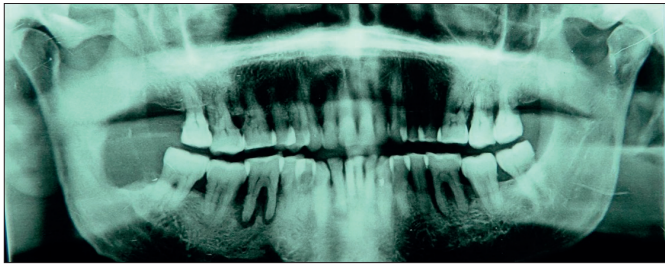


Fig. 1. Initial OPG: Unilocular radiolucency involving the anterior portion of the right ramus with thinned cortical plates.

Fig. 1. Panoramique initial montrant une image radioclaire au niveau de la partie antérieure du ramus droit avec des corticales osseuse amincies.

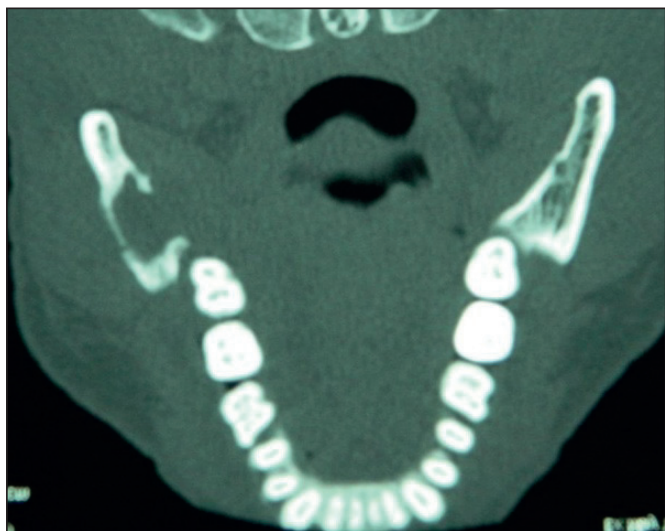


Fig. 2. CT scan showed a well-defined lytic lesion of the right ramus with a cortical break on the internal side.

Fig. 2. Le scanner montrait une lésion ostéolytique bien circonscrite au niveau du ramus droit ainsi qu'une effraction de la corticale interne en regard.

No neck nodes were palpable or hypoesthesia recorded. Orthopantomogram (OPG) showed a well-demarcated radiolucent area extending from the cervical part of the third right lower molar to the mid height of the ramus. There was no root resorption (Figure 1).

Computed tomography (CT) scan showed a well-defined lytic extended unilocular lesion involving the anterior portion of the right ramus. Cortical thinning with areas of cortical break was noted (Figure 2).

A benign odontogenic tumor was suspected, such as a keratocyst or a unicystic ameloblastoma. Surgical excision was decided under general anesthesia. The lesion was solid, encapsulated and totally removed with an intra-oral approach. The remaining cavity was then irrigated to remove any residual

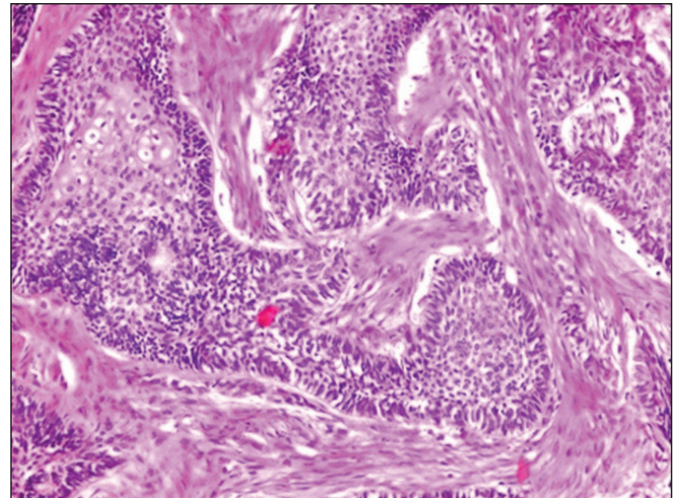


Fig. 3. Hematoxylin Eosin ×10: solid epithelial cell nests with peripheral palisading ameloblastic cells and central squamous cells.

Fig. 3. Hematoxyline Eosine ×10. Des nids de cellules épithéliales bordés de cellules améloblastiques en palissade et des cellules squameuses au centre.

fragment and debris. Histologic examination of the excised sample revealed solid epithelial cell nests with peripheral palisading ameloblastic cells and central squamous metaplasia consistent with diagnosis of acanthomatous ameloblastoma (Figure 3). No immunoeexpression of protein markers (P16, Ki67) was recorded.

No recurrence was observed 18 months after surgery.

Discussion

Ameloblastomas are rare benign odontogenic tumors arising from dental tissue, specifically the ectodermal cells responsible for the formation of tooth enamel. This tumor was described for the first time by Broca in 1868 as adamantinoma, then coined with the name "ameloblastoma" by Churchill in 1934 [3]. On the basis of previous reports, acanthomatous ameloblastoma occurs in older patients rather than younger ones, while in developing countries ameloblastoma occurs in younger patients [1, 4]. According to the current World Health Organization (WHO) classification of odontogenic tumors, ameloblastomas are divided into four types: solid/multicystic, extraosseous/peripheral, desmoplastic and unicystic. Histologically, the most common types are plexiform and follicular and four other uncommon variants include acanthomatous, granular cell, desmoplastic and basal cell types. The most prevalent histological subtype is the follicular variant (64.9%), followed by the plexiform (13%) and the acanthomatous variants (3.9%) [5]. Clinical findings are highly variable, such as a slow-growing mass, loose or displaced teeth, and rarely paresthesia

and pain. However, many lesions are discovered incidentally on radiographic examinations in asymptomatic patients. The most common radiographic presentation of ameloblastoma is an extended, radiolucent, multiloculated cystic lesion, with a characteristic “soap bubble-like” appearance. Other findings also include unilocular radiolucency, tooth displacement and root resorption. CT scan shows thinning of the bone, cortical destruction and extensive local infiltration [6]. In the reported case, the radiographic images revealed a well-demarcated unilocular radiolucent area with cortical destruction and no dental signs.

Both follicular and plexiform ameloblastoma show basal cells arranged in a peripheral palisading pattern, with the cells in the central portion mimicking stellate reticulum. Acanthomatous variants have central portions composed of squamous cell differentiation with keratin formation [7]. Squamous metaplasia as in acanthomatous ameloblastoma may be attributed to chronic irritation due to calculus and oral sepsis as seen in our patient [3].

Some authors believe that the acanthomatous variant, if untreated, can develop into an invading and metastasizing squamous cell carcinoma. Furthermore, there is controversy about the biological behavior of the acanthomatous ameloblastoma. Some researchers believe that this type is locally aggressive and frequently invades the alveolar bone or recurs after marginal surgical excision. Others suggest that there is no difference between the various subtypes of ameloblastoma and no special extensive local infiltration or bone destruction or risk of recurrence [8, 9]. Some aspects, as regards both the pathogenesis and the invasive growth of ameloblastoma remain unclear. Thus, the study of molecular mechanisms of cell proliferation can be helpful to predict aggressiveness of ameloblastoma and especially P16 (cyclin-dependent kinase inhibitor) which is a tumoral suppressor protein encoded by the *CDKN2A* gene. A high score in the immunoeexpression of

the P16 protein may indicate lower aggressiveness and a lower rate of recurrence [10].

Conflicts of interests: none

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