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Case Report

Cone-beam computed tomography imaging of benign odontogenic lesions in the maxilla: A report of 2 cases ☆,☆☆

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ABSTRACT

Odontogenic tumors are slow-growing and generally nonaggressive; however, aggressive characteristics appear to exist in particular tumors. The author reports two cases of benign odontogenic lesions at the anterior maxilla. A 44-year-old woman came to an oral-maxillofacial surgeon with a complaint of asymptomatic swelling of the left anterior region of the maxilla 15 years ago, which started gently but gradually increased over time. The patient experienced a physical injury 15 years ago, resulting in the loss of the upper left front teeth. In the other case, a 50-year-old woman complained for 5 years about a little swelling in her left anterior maxilla that became larger over time without causing any discomfort or pain. The Cone-beam computed tomography (CBCT) exam revealed a partially multilocular radiopaque mixed radiolucent lesion in the anterior maxilla, with margins that are both well-defined and ill-defined. Both display features of infiltrative, expansive, and moderately aggressive growths, leading to the erosion and perforation of the cortical plates in the buccal and palatal regions. Based on the biopsy results, both samples showed similar findings, specifically a benign odontogenic lesion, likely desmoplastic ameloblastoma, and squamous odontogenic tumor without any atypic cells or malignancy. This study aims to further our understanding of the clinical and radiological features of these patients and emphasize the importance of investigating specific lesions as potential diagnoses.

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Introduction

The oral and maxillofacial region presents a diverse range of pathological disorders as a result of the complicated structure of the tissues. The size of the tooth-forming tissues can range from tiny enlargements to massive enlargements, with the nature of the abnormalities varying from benign to malignant. Odontogenic tumors exhibit a slow growth rate and are often nonaggressive, although some specific types of tumors may display aggressive characteristics. In order to provide accurate treatment, it is crucial for the clinician to have in-depth knowledge of the pathological and radiological presentation [1].

According to the World Health Organization's (WHO) 2017 classification, benign odontogenic tumors that form only from odontogenic epithelium include ameloblastoma, calcifying epithelial odontogenic tumor, squamous odontogenic tumor, and adenomatoid odontogenic tumor. In the 2005 WHO classification, ameloblastomas were categorized into 3 types: solid/multicystic, extraosseous/peripheral, desmoplastic, and unicystic. The current classification has been simplified to include only 3 types: ameloblastoma, unicystic ameloblastoma, and extraosseous/peripheral ameloblastoma. The fifth edition of the World Health Organization (WHO) Classification of Head and Neck Tumors (2022) introduced adenoid ameloblastoma as a newly identified benign epithelial odontogenic tumor [2]. The term "solid/multicystic" for conventional ameloblastoma was removed as it lacks biological significance and may cause confusion with unicystic ameloblastoma. Desmoplastic ameloblastoma is now considered a histologic subtype rather than a separate clinicopathologic entity. Despite its distinct clinical and radiographic features, it behaves similarly to conventional ameloblastoma. The conventional form of ameloblastoma is distinguished by its multilocular and expanding behavior, as well as its infiltrating histological margin. The desmoplastic variant of ameloblastoma is uncommon and has unique characteristics in terms of its histology, imaging, and clinical presentation. This occurrence stands out due to its distinct appearance, possible violence, and high likelihood of misinterpretation [3].

Maxillary ameloblastomas pose a greater risk compared to mandibular lesions. Treating tumors in the posterior maxillary region is challenging due to their tendency to expand beyond the borders of the thin maxillary cortical bone, making containment difficult. Identifying something at an early stage is likewise highly challenging. In order to effectively treat maxillary lesions, a drastic approach involving maxillectomy (either partial, whole, or subtotal) is necessary, depending on the extent of the tumor [4].

Cone-beam computed tomography (CBCT) is a preferred diagnostic imaging technique for obtaining 3-dimensional (3D) images of the oral and maxillofacial region [5]. CBCT scans provide a clearer view of the lesion's location and the interaction between teeth and the floor of the nasal and maxillary sinus compared to periapical and panoramic radiographs. CBCT provides clinicians with the ability to monitor changes in growth, accurately perceive borders in depth, which may be challenging to distinguish otherwise, and evaluate the relative proximity of neighboring critical structures [6]. Both imaging and biopsy are necessary to definitively establish the diagnosis [7].

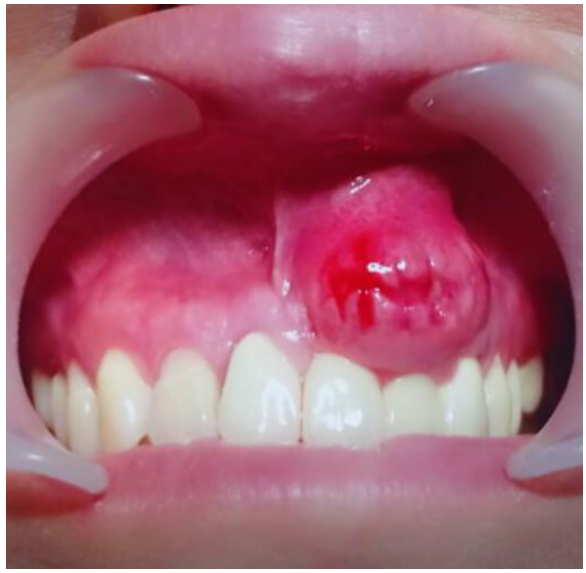

Fig. 1 – Case 1, An intraoral examination reveals the presence of a lesion on the anterior left maxilla.

Case report

This case report was prepared and written in accordance with the CARE reporting guidelines for case reports in order to support accuracy and transparency in the dissemination of case reports and the reporting of data from patient interactions [8]. Informed consent was obtained from the patient to be included in this study.

Case 1

A 44-year-old woman came to an oral-maxillofacial surgeon with a complaint of asymptomatic swelling of the left anterior region of the maxilla for 15 years ago (Fig. 1), which started gently but gradually increased over time. The patient experienced a physical injury 15 years ago, which led to the loss of the upper left anterior teeth. She underwent a dental procedure to restore the upper left front teeth region. She had a bridge restoration in the upper left front teeth region.

A Cone-beam CT scan was performed and reconstructed on the maxilla with a 9 × 14 cm field of view (FOV). The images were assessed, and sectional images of the region to be evaluated were selected, with the following results: On the panoramic view (Fig. 2), there is evidence of missing/agenesis of teeth 18, 21, 22, and 28. Tooth 12 shows radiopaque filling material in the crown reaching the pulp with a lesion indicating widening of the periodontal membrane space in the apical third, suggesting apical periodontitis. Tooth 11 appears post-root canal treatment. There is a radiopaque unit crown and bridge in the region of 11-21-22-23. Additionally, there is a radiolucent lesion with internal radiopaque septa, with partially

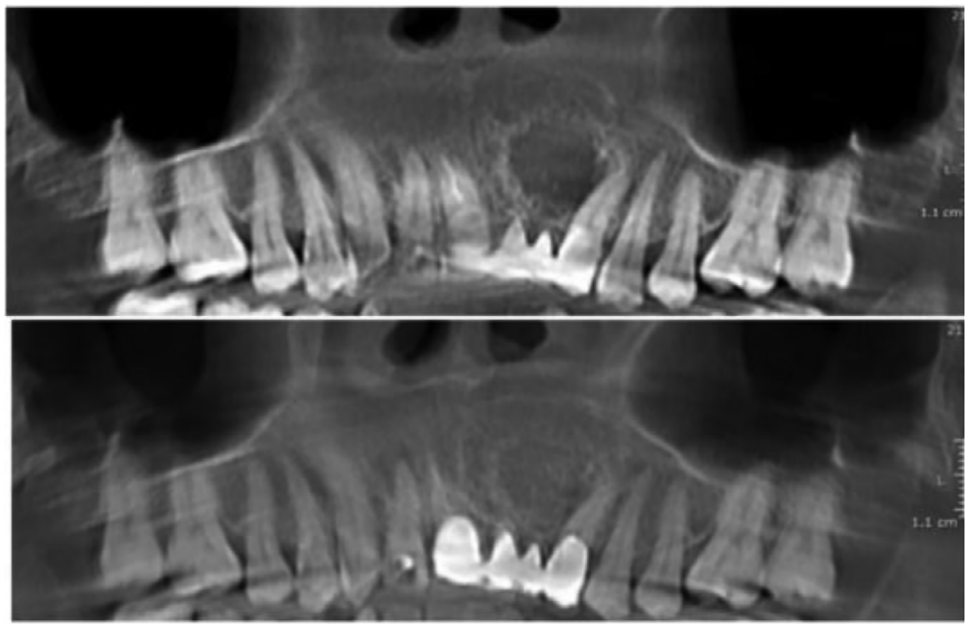

Fig. 2 – Case 1: from a panorama view, a radiolucent lesion with internal radiopaque septa has partially well-defined and partially ill-defined margins and rough and destructed cortical bone edges.

well-defined and partially ill-defined borders (with rough and destructed cortical bone edges), tending to be ovoid in shape, located in the anterior left maxilla in the region of 21-23.

From coronal view (Fig. 3A), a mixed radiolucent-radiopaque lesion is observed in the anterior maxilla, located in the edentulous ridge region of 21-22 and mesial to tooth 23. The lesion is semi-ovoid/circular in shape with dimensions of approximately 21 × 17 × 18 mm. Its borders are partially well-defined and partially ill-defined, with rough and partially destructed bone edges. Internally, the lesion is radiolucent and semi-multilocular, with septation observed on some sides. Density analysis shows values ranging from 129.8 to 252.9 gray values, equivalent to the density of fluid or soft tissue. The labial cortical plate appears expanded and partially discontinuous (destructed), whereas the palatal cortical plate remains intact, indicating the lesion tends to expand labially/buccally (Fig. 3B). The lesion is in contact with the apical region of tooth 23, causing distal displacement without resorption, and is also in contact with the nasopalatine canal without discontinuity of its walls (Fig. 3C). The lesion is located inferior to the nasal cavity and the maxillary sinus, with no involvement observed. Three-dimension (3D) lesion segmentation (Fig. 3D) was performed to simulate the dimension and location of the lesion.

The radiographic findings suggest a differential diagnosis primarily involving a benign aggressive odontogenic tumor, with desmoplastic ameloblastoma as a key consideration. This type of ameloblastoma is known for its aggressive nature and can present with features such as ill-defined borders, cortical expansion, and mixed radiolucent-radiopaque areas on imaging. Another possible diagnosis is a benign fibro-osseous lesion, which includes conditions like fibrous dysplasia and ossifying fibroma. Fibrous dysplasia typically exhibits a ground-glass radiopacity and well-defined borders, while ossifying fibroma often shows a mixed radiolucent-radiopaque appearance with more defined margins. The presence of cortical perforation, expansion, and poorly defined borders in the radiographic image raises the possibility of an inflammatory lesion or malignancy, as these features can be indicative of more aggressive processes. Conversely, the likelihood of a cystic lesion is considered minimal due to its generally well-defined borders and less aggressive behavior compared to the lesions currently under consideration.

The biopsy results indicate the presence of a benign maxillary tumor. Two tissue samples, measuring 5×5×4 mm and 10×5×4 mm, respectively, were analyzed. Both samples exhibited a firm, white appearance and were processed separately. Microscopic examination (Fig. 4) revealed extremely dense fibrous connective tissue containing bundles of small to medium-sized spindle-polygonal epithelial cells. Notably, no atypical or malignant cells were observed in the samples. Based on these findings, the diagnosis is consistent with desmoplastic ameloblastoma, a type of benign but aggressive odontogenic tumor known for its dense fibrous stroma and the presence of spindle-shaped epithelial cells.

Case 2

A 50-year-old woman presented with a gradually enlarging swelling in her left anterior maxilla that had been developing over a period of 5 years. Despite the progressive increase in size, she reported no associated discomfort or pain. Additionally, she did not have any complaints regarding her teeth. Swelling was oval, firm, nontender, and attached to the underlying structures. No surface ulcerations were noticed (Fig. 5).

A Cone-beam CT scan with a field of view (FOV) of 9×14 cm of the upper jaw reveals a lesion in the anterior maxillary region, particularly between teeth 21, 22, and 23. The panoramic

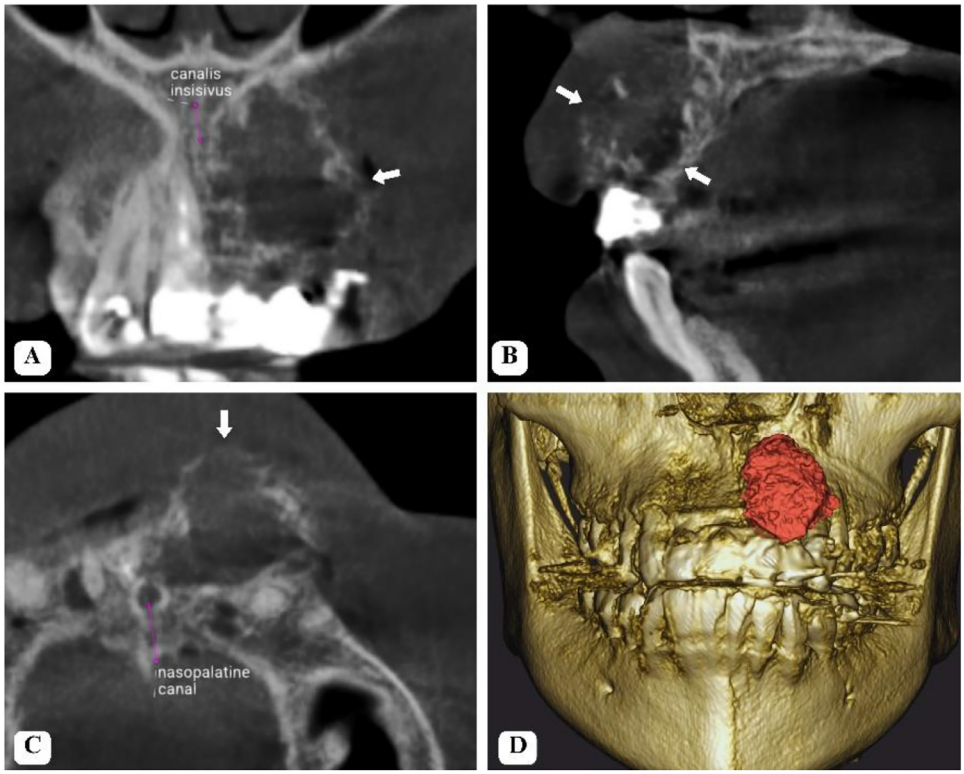

Fig. 3 – Case 1, Multiplanar reformation (MPR) CBCT images. (A) coronal view, (B) sagittal view, (C) axial view, (D) 3D view with lesion segmentation.

view (Fig. 6) shows displacement of tooth 22 and a diffuse radiolucent area around the alveolar region of tooth 22, along with gangrene of the roots of teeth 28 and 46. Other structures appear normal.

Specifically, the lesion is characterized as mixed radiolucent-radiopaque with a semi-ovoid or slightly irregular circular shape (Fig. 7A), measuring approximately 14 × 18 × 16 mm. The lesion's borders are partially well-defined and ill-defined, with rough bone margins, cortical thinning resembling eggshell thinning, and partial destruction. Internally, the lesion is radiolucent with scattered irregular radiopaque spots, and the density measures approximately 41.7-243.0 gray values, equivalent to soft tissue density.

Expansion is observed in the labial and palatal cortical plates (Fig. 7B), with some discontinuity. The lesion extends towards the labial/buccal direction, contacting the apices of teeth 21, 22, and 23, resulting in displacement of tooth 22 mesially and tooth 23 distally without resorption. The lesion also intersects with the nasopalatine canal without showing wall discontinuity (Fig. 7C). It is located inferiorly to the nasal cavity and maxillary sinus, with no apparent involvement of these areas.

In conclusion, the findings indicate a mixed radiolucent-radiopaque lesion in the anterior maxilla, specifically in the edentulous ridge area from 21-22 to the mesial of tooth 23. The lesion is infiltrative, expansive, and somewhat aggressive, with labial/buccal expansion, destruction, and perforation of the labial/buccal cortical plates and rough bone margins. Differential diagnoses include benign odontogenic tumors such as desmoplastic ameloblastoma, adenomatoid odontogenic tumor, squamous odontogenic tumor, or giant cell granuloma. Additionally, the possibility of an odontogenic cyst with inflammatory features is considered, given the cyst-like appearance and cortical expansion and perforation.

The macroscopic examination of the histopathological analysis revealed 2 biopsy tissue samples measuring 7 × 5 × 3 mm and 7 × 4 × 4 mm, respectively. The samples were solid and white and were processed in separate cassettes. Microscopic analysis (Fig. 8) showed that the tissue sections consisted of dense fibrous connective tissue with fragments of bone and benign epithelial foci, including areas with squamous epithelium. The overall conclusion of the biopsy is a benign tumor of the maxilla, specifically a squamous odontogenic tumor, with desmoplastic ameloblastoma considered as a differential diagnosis.

Discussion

Ameloblastoma is the common odontogenic tumor that originates from the odontogenic epithelium. In the 4th edition of the WHO classification, terms like follicular, plexiform, basaloid, granular, or desmoplastic have been eliminated since they lack clinical significance in describing pathological classifications [9]. Conventional ameloblastomas exhibit the distinct features of being multilocular and having a tendency to

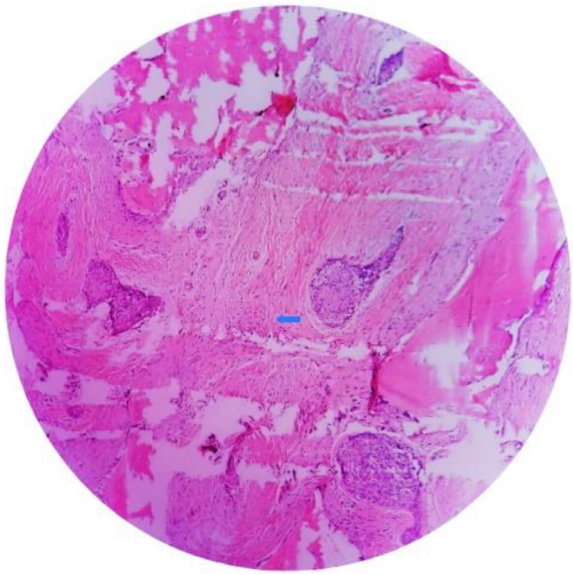

Fig. 4 – Histological specimen of case 1 presenting with dense fibrous connective tissue with small to medium sized polygonal spindle nucleus epithelial cells (blue arrow) were observed (HE Hematoxylin and eosin staining, original magnification ×100).

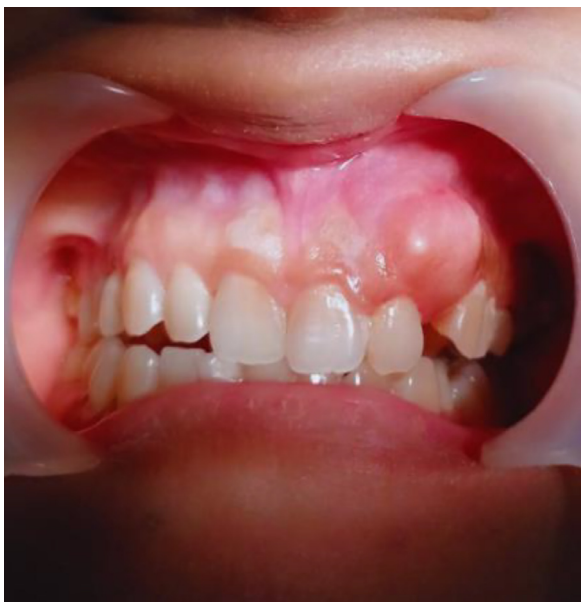

Fig. 5 – Case 2, An intraoral examination reveals swelling on the gingiva anterior left maxilla.

grow. The solid/multicystic or intraosseous ameloblastoma is characterized by its local invasiveness, slow growth, and high tendency to recur if not adequately excised due to its invasion of the marrow spaces. It has no tendency to metastasize, no gender preference, and occurs equally in both sexes [10].

The condition is typically identified in individuals aged 30 to 60 and primarily affects the posterior mandibular region in over 80% of instances. It might manifest as swelling of the jaws that varies in size. It occasionally is associated with an impacted tooth and causes expansion of the bonny cortex with the possibility of resorption of roots of the involved teeth. The predominant histological patterns observed are the plexiform and follicular types. The remaining types are desmoplastic, acanthomatous, granular, and basal [11].

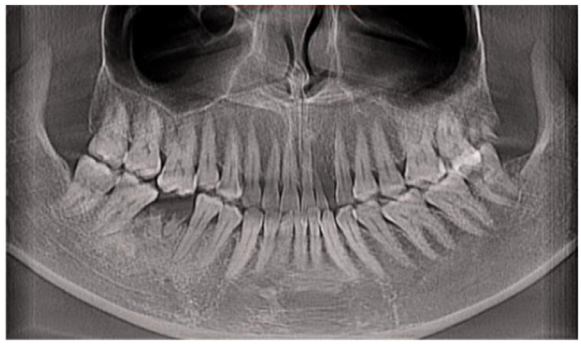

Fig. 6 – Case 2: From a panorama view, it shows displacement of tooth 22 and a diffuse radiolucent area around the alveolar region of tooth 22.

The radiographic characteristics of desmoplastic ameloblastoma (DA) also vary from those of common ameloblastoma. A wide range of appearances, from radiolucency to radiolucency with flecks of opacity to mixed radiolucent/radio-opaque, have been reported. Typically, conventional ameloblastoma appears as a clearly defined radiolucent area that can be either unilocular or multilocular. Nonetheless, the borders of DA are typically indistinct [12].

Adenomatoid odontogenic tumor (AOT) was previously known as ameloblastic adenomatoid tumor or adenoameloblastoma due to its histological feature of ameloblastoma-like clusters of epithelial cells in a developed connective tissue stroma. Irregular keratinization can be observed as ghost cells in conjunction with various levels of dysplastic dentin. There are 3 variants of AOT: follicular, extra-follicular, and peripheral. The peripheral type originates from the gingival tissues and is exceptionally uncommon. The follicular type is frequently linked to an impacted tooth and is present in 75% of instances. On the other hand, the extrafollicular type occurs between the roots of adjacent teeth and is not associated with an unerupted tooth [13].

Squamous Odontogenic Tumor (SOT) is a type of tumor that is identified by the presence of squamous epithelial islands surrounded by mature connective tissue stroma. The SOT is sometimes incorrectly identified as squamous cell carcinoma, keratocanthoma, ameloblastoma, or verrucous carcinoma. On occasion, this tumor mimics ameloblastoma, yet it is less aggressive than ameloblastoma. The condition is characterized by the abnormal growth of epithelial cells originating from the cell rest of Malessez. The potential differential diagnoses for SOT include acanthomatous and desmoplastic ameloblastoma types, as well as well-differentiated squamous cell carcinoma. Its clinical features range from painless swelling to mildly painful gingival swelling, which equally affects the maxilla anterior region and posterior mandible region [14]. Both men and women are equally affected, and the tumor does not exhibit a preference for either jawbone. The

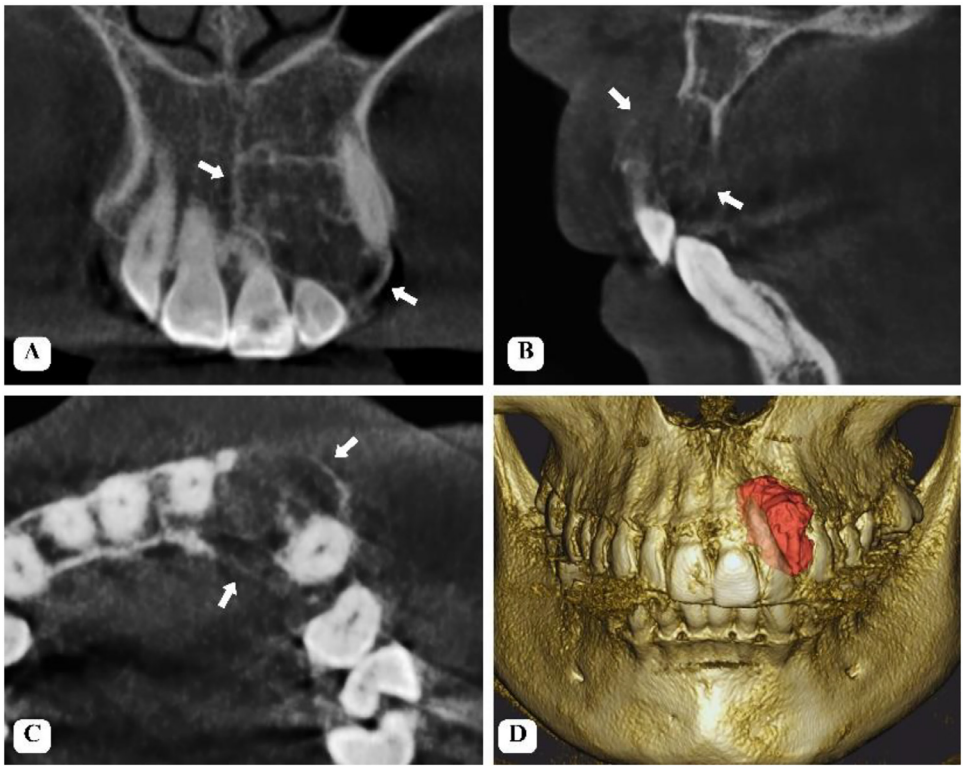

Fig. 7 – Case 2, Multiplanar Reformation (MPR) CBCT images. (A) coronal view, (B) sagittal view, (C) axial view, (D) 3D view with lesion segmentation.

predominant sites are the front upper jaw and back lower jaw. The majority of SOTs are managed with conservative approaches such as enucleation or curettage. However, aggressive or recurring tumors necessitate radical resection to ensure complete removal and reduce the risk of further recurrence [15].

Considering the suspicion of radiodiagnosis for case 1, desmoplastic ameloblastoma emerges as a likely diagnosis for several reasons. The expansive nature of the lesion combined with the presence of ill-defined areas suggests that it could potentially represent an ameloblastoma. However, given the mixed radiodensity of the lesion, it aligns with one of the possible variants of ameloblastoma—specifically, desmoplastic ameloblastoma—or alternatively, a benign fibro-osseous lesion. First, desmoplastic ameloblastoma is known for its slow growth pattern, which aligns with the clinical presentation of case 1, where the lesion has gradually enlarged over a longer duration. Second, while desmoplastic ameloblastomas can exhibit aggressive behavior, they often maintain well-defined borders, particularly in their early stages. The presence of well-defined borders in case 1 supports this diagnosis, as it indicates a relatively contained lesion. Additionally, desmoplastic ameloblastomas typically cause less extensive bone destruction compared to other more aggressive odontogenic tumors. The absence of significant bone destruction and the gradual, localized expansion observed in case 1 are consistent with the behavior of a benign desmoplastic ameloblastoma. Lastly, the dense, hard consistency of the lesion and its asymptomatic nature are characteristic of desmoplastic ameloblastomas, which often present as firm and asymptomatic until they reach considerable size. Therefore, the well-defined borders, slow and localized growth, and firm consistency in case 1 strongly support the suspicion of desmoplastic ameloblastoma, reinforcing its diagnosis.

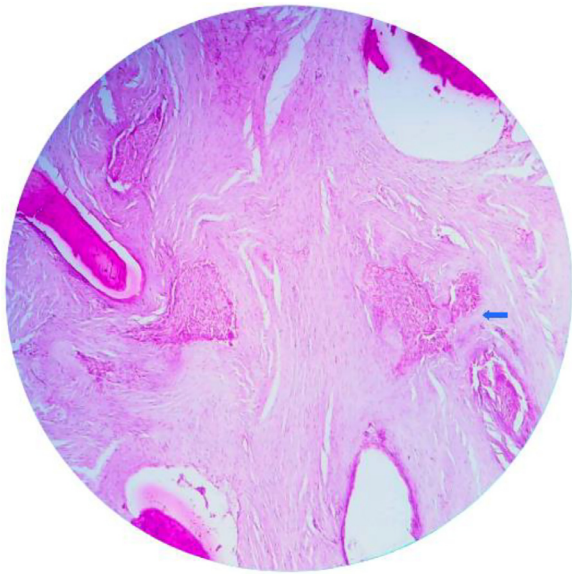

Fig. 8 – Histological specimen of case 2 presenting with irregularly shaped epithelial island, surrounded by fibrous connective tissue with fragments of bone and benign epithelial foci (blue arrow) were observed (HE Hematoxylin and eosin staining, original magnification x100).

Table 1 – Comparison table of lesion characteristics based on literature and case examination results [16–19].

Odontogenic tumor	Age/gender/localization/clinical feature	Common radiographic features	Essential diagnostic criteria
Ameloblastoma (desmoplastic type)	Fourth-fifth decades/distinct male predilection/slight predilection for the mandible compared to the maxilla/painless swelling	Mixed radiopaque-radiolucent lesion resembling benign fibro-osseous lesions, poorly marginated, honeycombing	Small nests and strands of "compressed" odontogenic epithelium supported by pronounced collagenized stroma. Interspersed epithelial cells arranged in compressed islands or strands.
Adenomatoid odontogenic tumor	Second-third decades/female/anterior maxilla, pericoronal/painless swelling	Well-defined mixed lesion	Site in alveolar processes of jaws. Epithelial nodular structure/rosettes of spindled to columnar epithelial cells. Duct-like structures with minimal stroma.
Squamous odontogenic tumor	Third decade/no gender predilection/anterior maxilla, posterior mandible/painless swelling	Well-defined mixed lesion	Site in tooth-bearing areas of the jaw, close to lateral surface of root. Mature fibrous connective tissue. Uniform squamous differentiation without significant keratinization. No peripheral palisading and stellate reticulum.
Case 1	44 years old/female/anterior maxilla/painless swelling	Semi-ovoid/circular in shape with partially well-defined and partially ill-defined, with rough and partially destructed bone edges	Dense fibrous connective tissue with small to medium sized polygonal spindle nucleus epithelial cells, no atypical cells or malignancies were found.
Case 2	50 years old/female/anterior maxilla/painless swelling	Mixed radiolucent-radiopaque with a semi-ovoid or slightly irregular circular shape with rough bone margins, cortical thinning resembling egg-shell thinning, and partial destruction	Fibrous connective tissue with bone fragments and benign epithelial foci together with an epithelial impression, no atypical cells or malignancies were found.

The biopsy and histopathological analysis (HPA) of the specimen revealed dense fibrous connective tissue interspersed with small to medium-sized polygonal spindle nucleus epithelial cells, with no atypical or malignant cells identified. A final diagnosis of desmoplastic ameloblastoma was made.

In contrast, case 2′s radiodiagnosis encompasses several possibilities, including adenomatoid odontogenic tumor, squamous odontogenic tumor, or desmoplastic ameloblastoma. The lesion exhibits a mixed radiodensity with smaller radiopaque spots compared to case 1 and lacks distinct septation, suggesting a closer resemblance to adenomatoid or squamous odontogenic tumors. However, the potential for desmoplastic ameloblastoma cannot be excluded. The HPA results for case 2 indicated the presence of bone and epithelial fragments, and although the findings included epithelial foci, the overall characteristics of the lesion direct the diagnosis more towards a squamous odontogenic tumor. However, it is still important to investigate the possibility of desmoplastic ameloblastoma because it shares similar characteristics with other conditions. The squamous odontogenic tumor consists of a peripheral cell layer composed of flat to cuboidal cells [20]. Desmoplastic ameloblastomas often exhibit elongated and compressed islands and strands, rather than spherical and broad-based structures seen in other types of ameloblastomas. Desmoplastic ameloblastoma often exhibits patterns of squamous cells in the squamoid areas, which are not seen in SOT [21].

Utilizing 3D Cone-beam CT (CBCT) provides a more detailed view of the lesion's borders, direction of expansion, and its relationship with surrounding tissues compared to traditional 2D imaging [13]. This advanced imaging technique is invaluable for narrowing down the differential diagnosis of odontogenic tumors, offering insights that facilitate a more accurate assessment. A panoramic examination might initially lead to a preliminary diagnosis of ameloblastoma due to its prevalence among odontogenic lesions and the commonality of its clinical presentation, which includes infiltrative, expansive, and sometimes aggressive characteristics with ill-defined lesion borders. To achieve a definitive diagnosis, histopathological analysis (HPA) is essential. Key histological features include compressed epithelium and stroma collagen for desmoplastic ameloblastoma, duct-like structures for adenomatoid odontogenic tumors, and squamous epithelium for squamous odontogenic tumors. Some conditions have similar histological features, requiring additional evaluation or supplementary tests to differentiate them [22]. Frequently, there is ambiguity regarding the diagnosis and the appropriate treatment plan to be implemented. Immunohistochemistry can be a valuable resource in determining the precise diagnosis, directing the treatment plan, and predicting the probable prognosis of these lesions [23].

After analyzing the differences and further characteristics as shown in Table 1, it is crucial to approach each case with a detailed and careful classification based on specific characteristics observed in the imaging and histopathological findings. By thoroughly assessing these features, oral radiologists can refine the differential diagnosis and provide more accurate and focused guidance to the referring clinicians. Although the treatment approach, often involving surgical resection, may ultimately be similar regardless of the precise diagnosis, a meticulous evaluation ensures greater confidence and precision in diagnosing the lesion. This careful differentiation helps in narrowing down the possible diagnoses, thereby enhancing the overall diagnostic accuracy and reducing potential confusion for the referring physician.

Conclusion

Evaluating oral radiographs of lesions affecting the hard tissues of the oral cavity is crucial for accurate diagnosis. Both cases presented involve rare pathologies. Many cases like this should come to the forefront of medical literature so that the biological behavior of hybrid lesions can be thoroughly described. A thorough and careful review of radiographic interpretation is essential, as it aids in the precise classification of these lesions based on their visual characteristics, thereby narrowing down the differential diagnoses. Although various potential diagnoses may be considered, the chosen treatment approach is typically surgical resection. This careful approach not only improves diagnostic accuracy but also guides appropriate treatment planning.

Patient consent

Complete written informed consent was obtained from the patient for the publication of this study and accompanying images.

Data availability

The manuscript has been read and approved by all the authors, that the requirements for authorship as stated earlier in this document have been met, and that each author believes that the manuscript represents honest work.

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