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Maxillary Unicystic Ameloblastoma: A Case Report

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Abstract:

Background:

Ameloblastoma is a benign epithelial odontogenic tumor. It is often aggressive and destructive, with the capacity to attain great size, erode bone and invade adjacent structures. Unicystic ameloblastoma is a rare odontogenic lesion, with clinical, radiographic and gross features of jaw cysts. The lesion histologically shows typical ameloblastomatous epithelium lining part of the cyst cavity with or without and/or mural tumor growth. Unicystic ameloblastoma usually presents in posterior mandibular ramus region, while it is rare and atypical in posterior maxillary region.

Case presentation:

We report a case of 60 year old male patient with a medical history of diabetes and hypertension who was referred by his dentist for further evaluation due to delayed healing of the extraction socket of tooth (27), removed two years earlier. Clinical examination revealed halitosis and a non-healed socket with epithelialized walls and no bone exposure with suspicion of an oroantral communication. The Valsalva maneuver was negative. Initial panoramic radiography revealed an irregularly healed tooth (27) socket with indistinct margins and a heterogeneous left maxillary sinus floor. Computed tomography showed a well-defined hypodense lesion lacking a sclerotic border, displacing the left maxillary sinus and nearly obliterating its lumen. The patient was treated by surgical enucleation of the lesion and extraction of first molar tooth which was present inside the lesion. The histopathological examination of the lesion revealed confirmed finding for unicystic ameloblastoma mural form. No recurrence was observed in 1 year follow-up.

Conclusion:

Maxillary region is considered a rare and atypical location for unicystic ameloblastoma. We emphasize the importance of differential diagnosis of an odontogenic lesion with common clinical and radiological features that will impact the treatment planning and follow up. As oral health providers we should be aware that the unilocular radiolucencies may be unicystic ameloblastoma.

Keywords: Ameloblastoma, Unicystic ameloblastoma, Maxillary, Enucleation

Introduction:

Ameloblastoma is a benign odontogenic tumour with an aggressive growth tendency and a high risk for malignant transformation and metastasis. The pooled incidence rate of ameloblastoma is 0.92 per million person-years.[1] They account for 1% of all oral tumours.[2] The World Health Organization classifies ameloblastomas into four clinical categories as solid multicystic, unicystic, desmoplastic, and extraosseous/peripheral.[3] Unicystic ameloblastoma (UA) is a distinct kind of ameloblastoma characterized by slow growth and is relatively locally aggressive.[4] UA is a rare variant of ameloblastoma frequently encountered in second or third decade. This report highlights the importance of radiological and pathological examination of maxillary Unicystic Ameloblastoma.

Case Report:

A 60-year-old male patient with a medical history of diabetes and hypertension was referred by his dentist for further evaluation due to delayed healing of the extraction socket of tooth (27), removed two years earlier. Clinical examination revealed halitosis and a non-healed socket with epithelialized walls and no bone exposure (Fig.1). Probing did not reach the base of the socket, raising suspicion of an oroantral communication. The Valsalva maneuver was negative.

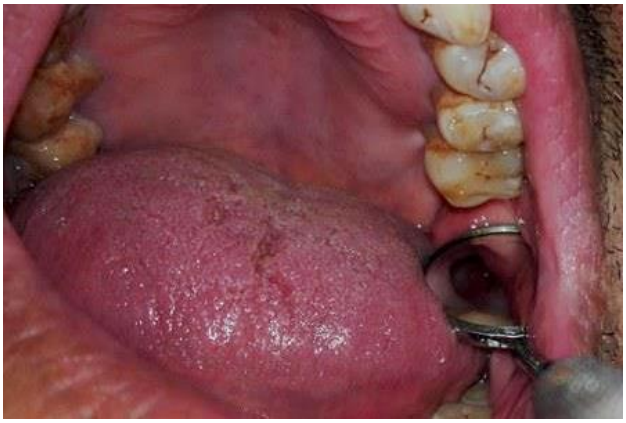


Figure1: Intraoral photograph showing the non-healed of the extraction socket of tooth(27)

Initial panoramic radiography showed an irregularly healed socket of tooth (27), with indistinct margins and an absent left maxillary sinus floor that appeared heterogeneous (Fig.2).

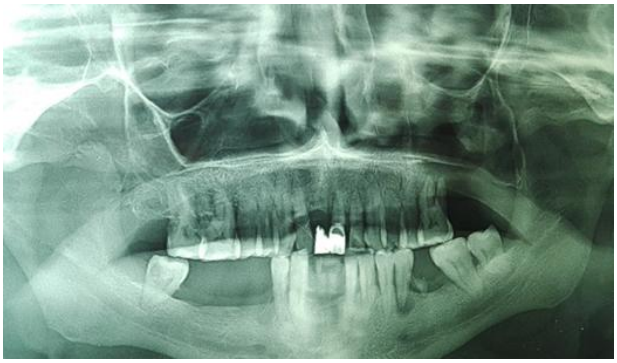


Figure 2: Panoramic radiography showing non-healing of the tooth (27) extraction site and heterogeneity of the left maxillary sinus.

Subsequent computed tomography revealed a well-defined hypodense lesion with no peripheral sclerotic border, in the region of the tooth (27) socket. This lesion displaced the left maxillary sinus superiorly, nearly obliterating its entire lumen(Fig 3.A). Additionally, there was evidence of alveolar bone loss affecting the support of tooth (26) (Fig 3.B). The presence of an air density within the lesion confirmed an oroantral communication (Fig3.C,D).

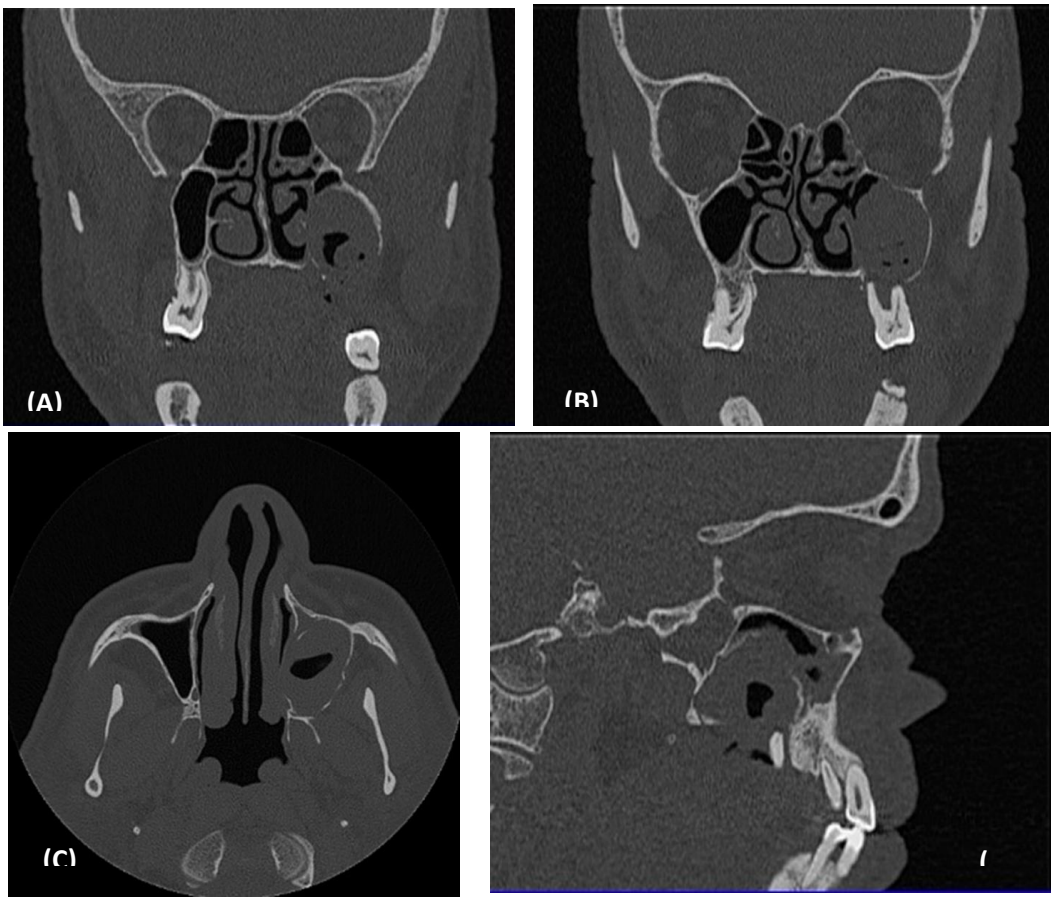


Figure 3: Computed tomography examination showing: in Figure A, a coronal slice with bone window revealing a hypodense lesion in the left maxilla, displacing the ipsilateral sinus floor superiorly; in Figure B, alveolar bone loss affecting the support of tooth (26). Figure C displays an axial slice through the maxillary sinuses, and Figure D, a sagittal slice, highlighting air density indicative of an oroantral communication.

All the routine pre-operative evaluation was carried out for the patient, and he was prepared for surgery. The surgical site was prepared using betadine and draping was performed. Under aseptic conditions local anesthesia was injected and infiltration at the local site was also performed. A triangular mucoperiosteal flap was raised in relation to the cystic lesion and then the involved tooth was extracted. A bony window was created followed by the removal of the cystic lining.The cavity was inspected for any remnants of the lining and for bony chips. Finally, the cavity was irrigated with saline. Once hemostasis was achieved primary closure was performed (Fig.4). Post-operative instructions were given.

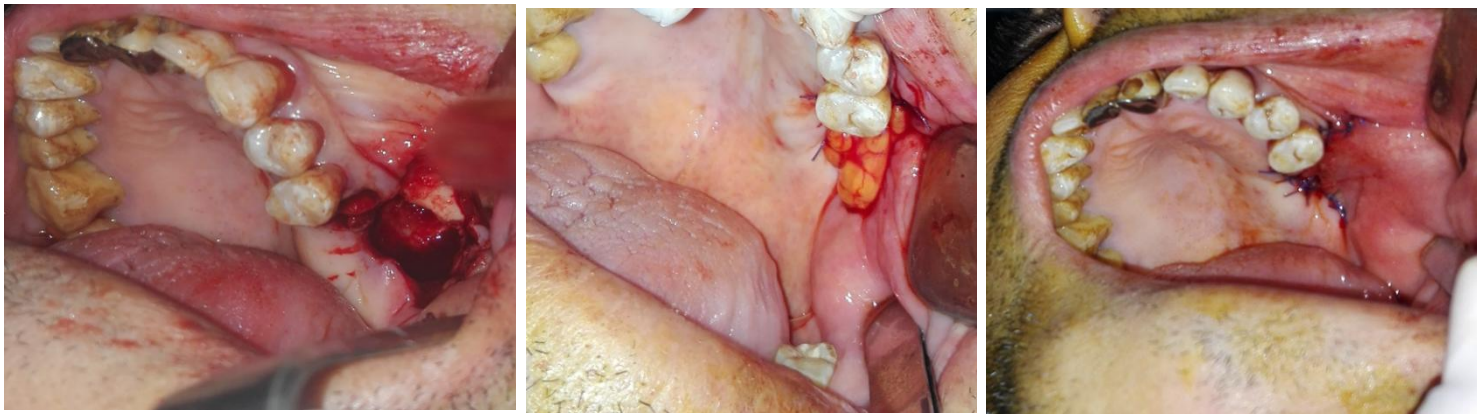
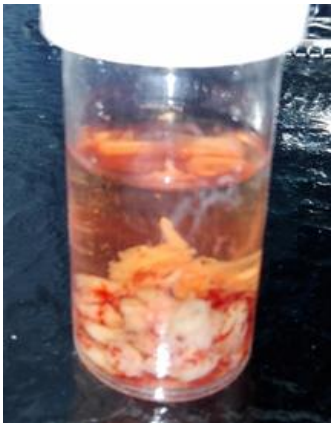


Figure 4: Intraoral perioperative photographs illustrating the surgical steps of cyst enucleation.

Post-operatively, the specimen was sent to the pathologist to confirm the type of unicystic ameloblastoma (Fig.5).

Regular clinical and radiological follow-up was conducted, revealing proper healing of the surgical site and no radiological evidence of recurrence.



HISTOLOGIE :
Les fragments adressés ont été inclus en totalité
Ils ont intéressé une prolifération tumorale agencée en massifs et en amas composés par une double population cellulaire : périphérique cylindrique disposée de façon palissadique, et centrale losangique peu cohésive, rappelant les cellules du corps muqueux de Malpighi, réalisant par places des images d'enroulement cellulaire
Les noyaux sont pâles, finement chromatiques, sans mitoses

Figure 5: Showing the specimen and histopathological features of the anatomical pathology results.

Discussion:

UA is a rare type of ameloblastoma, accounts for about 6 % of all ameloblastomas. It affects mandible more often than maxilla and in about 50 % of the cases occur in the second decade of life [5].It is presented more commonly in the mandible than in the maxilla in the ratio of 13:1. T he tumor is observed in mandibular-ramus region, while posterior region of maxilla is considered to be rare and atypical [6]. T he lesion is usually found in association with the crowns of mandibular third molar teeth, but can be seen also in interradicular, periapical and edentulous regions as well [7]. In our case it is associated with the non-healing extraction site of tooth 27. It is presented as a painless swelling, facial asymmetry, tooth impaction, tooth displacement, mobility, or tooth resorption. On radiographic imaging the unilocular lesion with well defined sclerotic borders is seen [8]. The differential diagnosis of UA should include keratocystic odontogenic tumor, residual cyst, central fibroma, central giant cell granuloma and dysplastic fibrosis[9] .Ackermann et al. (1988) [8]and Robinson and Martinez (1977)[10] argued that as the epithelium of odontogenic cysts and ameloblastomas have a common ancestry, a transition from a nonneoplastic cyst to a neoplastic one could be possible, even though it occurs infrequently. Radiographically there are 2 main patterns: Unilocular and multilocular [11,12]. Based on histological examination, to diagnose a lesion as unicystic ameloblastoma, the minimum criteria is the demonstration of presence of a single cystic sac lined by odontogenic ameloblastomatous epithelium which is seen only in focal areas [13]. T here are different classifications of unicystic ameloblastoma. Based on the clinicopathologic study of 57 cases of unicystic ameloblastoma, Ackerman's classification into three histologic groups is as follows: I. Luminal UA (tumor confined to the luminal surface of the cyst); II. Intraluminal/plexiform UA (nodular proliferation into lumen without infiltration of tumor cells into connective tissue wall); and III. Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium) [8]. T here is another grouping by Philipsen and Reichart [14] which describes the forms of UA as follows: Subgroup 1. Luminal UA; Subgroup 1.2. Luminal and intraluminal; Subgroup 1.2.3. Luminal, intraluminal and intramural; and Subgroup 1.3. Luminal and intramural. UA is considered to be a less aggressive form of ameloblastomas that can be successfully removed by

simple enucleation or other less aggressive surgery [15]. T he use of Carnoy's solution to decrease the risk of recurrence after conservative surgical treatment of UA's was initially suggested by Stoelinga and Bronkhorst in 1988 [16]. Also it is advocated that vigorous curettage of the bone should be avoided because it may implant foci of ameloblastoma more deeply in bone [17]. The recurrence rate for UA's after conservative surgical treatment (curettage or enucleation) is generally reported 10–20 % [18] and on average, <25 % [19]. This is considerably less than 50–90 % recurrence rates which are noted after the conventional curettage of solid or multicystic ameloblastomas [18,20]. Lau and Samman [21] reported recurrence rates of 3.6 % for resection, 30.5 % for enucleation alone, 16 % for enucleation followed by Carnoy's solution application, and 18 % by marsupialisation followed by enucleation, where the lesion is reduced in size.

Conclusion:

Every unilocular radiolucency of the jaw should be closely monitored and examined since UA shares significant clinical and radiographic similarities with odontogenic cysts and tumors. Neither the incisional biopsy may be able to reflect the true nature of the lesion nor the aspirational cytology. Long-term follow-up is mandatory because of the recurrence risk of unicystic ameloblastoma, which may occur after a long time.

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