**Dentomaxillofacial Radiology (2004) 33,** 60–62

http://dmfr.birjournals.org

CASE REPORT

# Maxillary unicystic ameloblastoma

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The authors present the case of a 17-year-old White male patient complaining of enlargement in the gingival region and the fundus of the left maxillary anterior vestibular sulcus. The clinicopathological diagnosis was plexiform unicystic ameloblastoma. With this report, the authors illustrate the importance and complexity of a differential diagnosis of lesions with a cystic aspect in the anterior region of the maxilla, among them inflammatory radicular cysts, odontogenic keratocysts, adenomatoid odontogenic and unicystic ameloblastoma.
*Dentomaxillofacial Radiology* (2004) **33,** 60–62. doi: 10.1259/dmfr/32974609

**Keywords:** ameloblastoma; unicystic; plexiform; cysts; maxilla; keratocyst

## Case report

A 17-year-old White male patient came to the Oral Medicine Service of the Dental School of Araraquara – UNESP complaining of enlargement in the gingival region and the fundus of the left anterior vestibular sulcus, with discrete painful symptoms upon palpation and of hard consistency similar to bone tissue. The patient did not report any systemic health problems.

Clinical examination revealed vestibular swelling in the left anterior region above the maxillary canine root, of hard consistency and similar in colour to the oral mucosa, as well as discrete bulging of the palate between the lateral canine and incisor. In addition, erythematous tissue with a granulomatous aspect was observed in the vestibular marginal gingival region of the canine (Figure 1).

The patient reported previous surgery performed 2 years before for removal of a lesion in the same region. Radiography performed at that time had revealed a circumscribed radiolucency surrounded by a radiopaque halo, which extended from the distal side of the left maxillary lateral incisor and overlapped the root of the left maxillary canine, with evident thickening of the periodontal ligament throughout the canine root (Figure 2).

The present panoramic and periapical radiographs showed a unilocular radiolucency with clear contours, which extended from the distal side of the root of the left maxillary lateral incisor, including the root of the left maxillary canine, to the nasal fossa floor and the distal side of the root of the left maxillary second premolar. A circular radiopaque 2 cm diameter halo surrounding the radiolucency was observed. There was no radiographic evidence of bone cortex close to the cervical third of the left maxillary canine. The lamina dura of this tooth was found to be interrupted in the apical third (Figure 3).

Pulp vitality tests of the left maxillary central and lateral incisors, canine and premolars were positive.

Aspiration of the lesion, performed by puncture of the gingival fold owing to the apparent lack of bone cortex in this area, yielded a brownish-orange fluid. Microscopic analysis did not show the presence of cholesterol crystals, typical of periapical cysts, but was suggestive of a diagnosis of an odontogenic keratocyst owing to the presence of keratin lamellae.

After endodontic therapy was performed on the left maxillary canine, the patient was treated surgically with enucleation of the lesion and curettage of the surgical site. No root resorption of the left maxillary canine was observed during surgery.

Histological analysis of the surgical specimen revealed a plexiform unicystic ameloblastoma, according to the World Health Organization classification.[1] The ameloblastoma was completely surrounded by a dense fibrous capsule and lined with ameloblastic epithelium, with proliferation of star-shaped cell chains inside the tumour, causing the plexiform pattern (Figure 4). The capsule of the lesion was free of intramural infiltration by ameloblastic tissue islands.

Healing of the area was satisfactory and the patient is being currently followed-up periodically at the Oral Medicine Service in view of the need of early identification of possible recurrences.

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Received 6 May 2003; accepted 4 January 2004

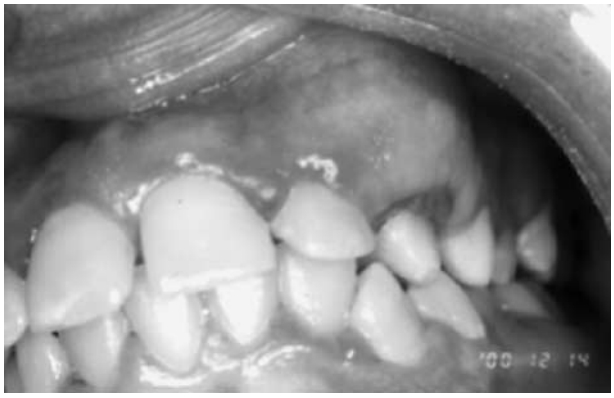

**Figure 1** Vestibular enlargement and granulomatous tissue in the marginal gingiva of the maxillary left canine

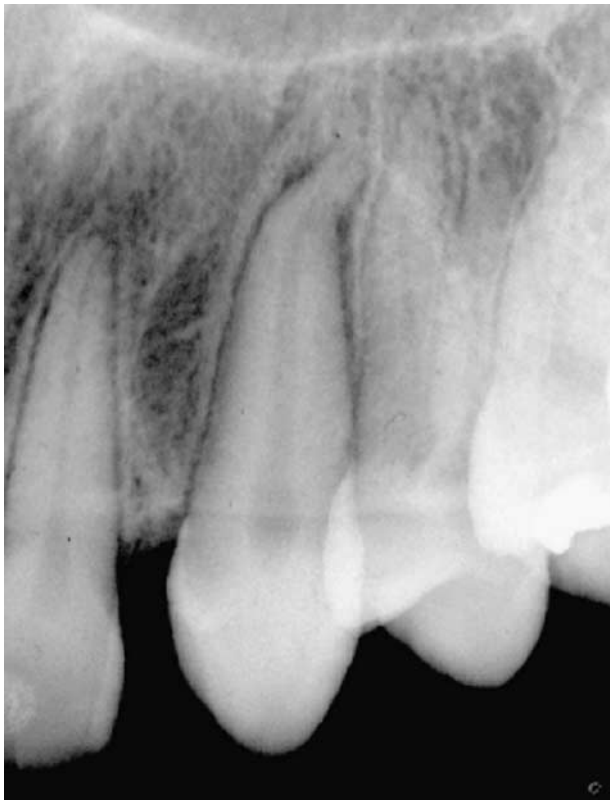

**Figure 2** Periapical radiograph taken at the time of the first surgery (2 years previously)

## Discussion

The present report describes the case of a 17-year-old patient who presented with swelling in the vestibular region of the maxilla, histologically diagnosed as plexiform unicystic ameloblastoma. The location of this lesion in the anterior region of the maxilla is considered to be rare and atypical, since this lesion predominantly occurs in the mandible, with the molar region and the ascending ramus being the most affected areas.[2–4] The ratio of mandibular to maxillary unicystic ameloblastoma has been reported to be 13:1.[3]

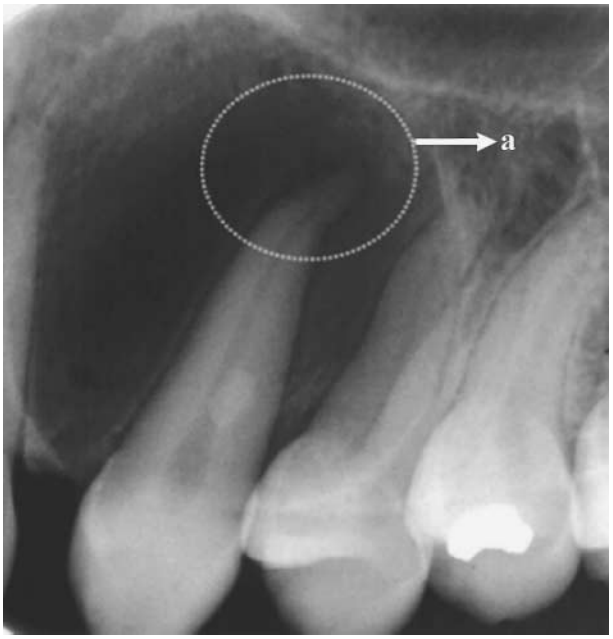


**Figure 3** Present periapical radiograph showing extensive radiolucency involving the root of the canine. The lamina dura of this tooth is interrupted in the apical third (a)

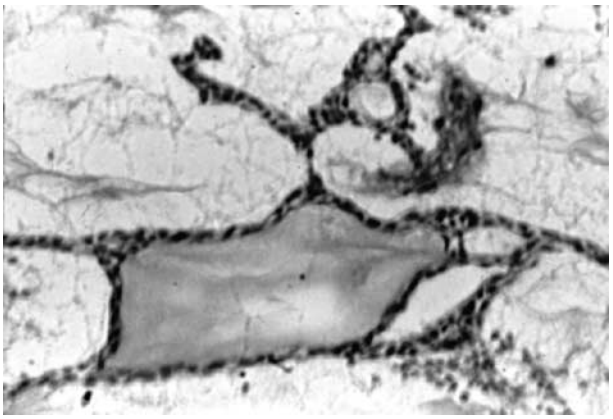

**Figure 4** Star-shaped cells with a plexiform arrangement containing hyaline material

Clinical examination revealed enlargement in the vestibular region of the left maxillary canine, and periapical radiography was required to verify bone involvement. Radiography showed a radiolucency involving the canine root, extending to the periapex and to the lateral margins, with a cystic appearance.

In young individuals, ameloblastoma is usually associated with impacted teeth;[5] however, in the present case no impacted teeth were observed and the patient did not report any possible previous impaction.

At first the radiolucency seemed to resemble an inflammatory periapical cyst, but detailed analysis of the image revealed an interrupted lamina dura on the root apex. In addition, the dental crown was intact and no broad restorations or carious lesions, which could justify the

pathogenesis of a periapical cyst, were observed. The tooth also showed positive pulp vitality.

The pulp vitality test was important for the differential diagnosis between a periapical cyst and an odontogenic keratocyst. Diagnosis of the latter still could not be excluded, although this lesion is also rare in the anterior region of the maxilla.[6] Based on this suspicion and on the supposed absence or thinning of bone cortex in the marginal gingival region, puncture of the gingival sulcus was performed. Keratocysts usually produce large amounts of keratin, which can be identified microscopically inside the cyst, indicating the nature of the lesion. Analysis of the punctured fluid suggested a keratocyst owing to the presence of keratin lamellae, supporting the probable diagnosis.

Clinical examination showed the presence of erythematous tissue with a granulomatous appearance located in the marginal gingival region of the left maxillary canine. This did not appear to be classical gingival inflammation since the patient presented satisfactory oral hygiene not compatible with the occurrence of periodontal inflammation. The erythematous and granulomatous tissue of the gingiva was clinically interpreted as being an exteriorization of a part of the lesion, supposedly a capsule, and this clinical hypothesis was supported by the absence of bone cortex in this area.

Enucleation was planned based on the suspected diagnosis of an odontogenic keratocyst. The radiographic image of an encapsulated unicystic lesion, which developed without causing resorption of the root apex, was confirmed during surgery. The lesion was completely removed and only clinically healthy bone tissue remained. In contrast to the most probable diagnosis, *i.e.*, odontogenic keratocyst, microscopic analysis of the surgical specimen revealed plexiform unicystic ameloblastoma.

Fortunately, the surgical conduct was compatible with the biological nature of unicystic ameloblastoma, which does not present an aggressive clinical behaviour. In addition, analysis of the surgical specimen revealed the absence of ameloblastic cell chains infiltrating the fibrous capsule, indicating a good prognosis and low recurrence potential.

The patient reported that 2 years previously he had undergone surgery for removal of a lesion in the same bulging region, a fact suggesting that the present lesion was a recurrence, which is compatible with the biological behaviour of ameloblastoma and keratocysts.[7,8] According to the patient, the surgical material had not been submitted to microscopic analysis at that time. Systematic and periodic follow-up is essential based on the high recurrence rate of ameloblastomas, including the unicystic type.

The periapical radiograph of the patient taken 2 years before, together with the present clinical and radiographic aspects, led us to exclude the diagnosis of a radicular cyst or periapical granuloma. The previous radiograph from the time of first surgery showed a small circumscribed lesion adjacent to the middle third of the canine root, which may have been previously diagnosed as a lateral periapical cyst.

In conclusion, the differential diagnosis of lesions of the anterior region of the maxilla, mainly periapical cysts and keratocysts, is difficult and extremely important for correct management. On the other hand, in the present study, only microscopic examination of the surgical specimen allowed the establishment of the final diagnosis of unicystic ameloblastoma, illustrating the complexity of the diagnostic process of bone pathologies, especially when the lesions present non-classical aspects and atypical locations.

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