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Unicystic Ameloblastoma: A Case Report and 4 Years Follow Up

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Abstract

Ameloblastoma is an invasive tumor which originates from remnants of the dental lamina and odontogenic epithelium and represents 1 % of all maxillary tumors.

A case of unicystic ameloblastoma and a conservative therapeutic attitude are reported with a four-year follow-up.

The importance of biopsy before surgical excision is underlined.

Keywords: *Unicystic, ameloblastoma, impacted, molar, mandible.*

Introduction

Ameloblastoma, odontogenic tumors of epithelial origin without induction of mesenchyme that represent 1 % of oral tumors, represent a very particular clinical and radiological entity **[**1, 2**]**. The World Health Organization (WHO) classified ameloblastoma in four types: multicystic, peripheral, desmoplastic and unicystic ameloblastomas **[**1, 2**]**.

The topography of these tumors is variable. They are rarely observed at the level of the maxilla and are found at the level of the mandibular symphysis, but their most frequent situation is at the level of the mandibular angle and can invade the ascending ramus or even the coronoid process **[**3, 4**]**.

Radiologically, ameloblastoma are described as a radiolucent, multilocular image leading to expansions, 90% of cases present a multilocular aspect and 10% are unilocular. They can be associated with impacted teeth and/or cause root resorption or tooth displacement **[**5, 6**]**.

The differential diagnosis arises with dentigerous cysts **[**7**]**. This is why it is advisable to carry out the histopathological examination of all suspected cases of dentigerous cysts, especially to detect unicystic ameloblastoma, which form from their epithelial lining, without radiological signs indicating this significant variation **[**4, 8**]**.

Unicystic ameloblastoma, a very rare variety, constitutes 5% of ameloblastoma **[**9**]**.

Various studies have shown that 15 to 30% of all ameloblastoma form from the wall of a dentigerous cyst **[**6-8**]**.

On the other hand, 85% of unicystic ameloblastoma are associated with observed dentigerous cysts, all in patients who have not exceeded the third decade [6, 7, 9].

Three histological types are recognizable according to the degree of ameloblastomatous epithelial extension, namely luminal, intraluminal, and mural types [4, 5].

Compared to solid and multi-cystic ameloblastoma, unicystic ameloblastoma are considered to be less aggressive and respond more favorably to conservative management [8, 10].

In this article we present a case of mural ameloblastoma with a conservative therapeutic attitude and a clinical and radiological follow-up of 4 years.

Clinical Case

A 16-year-old female consulted us for left an intra-oral swelling in the retro-molar region with absence of her second mandibular molar. (Figure 1).

Palpation reveals liquid contents of the swelling. Depressibility test indicates the presence of a thin osteo-periosteal shell opposite to the tumefaction. Endo-buccal examination reveals a fluctuating collection filling the bottom of the vestibule facing the mandibular premolars and molars. The wisdom tooth and the second molar were missing from the dental arch. The first molar as well as the second premolar were mobile and sensitive to percussion. No sign of right inferior alveolar nerve paresthesia.

A panoramic radiograph shows expansive unilocular radiolucency's with a well-demarcated margin extending from the right mandibular second premolar to the right ramus posteriorly. the right mandibular condyle is spared. The lesion caused the expansion of both vertical and horizontal branches with thinning of the basilar rim. From a dental point of view, there is no root resorption of the first mandibular molar, while the left mandibular second molar is included in the lesion and pushed anteriorly down to the root of the first molar and the third molar is also included in the lesion and pushed back towards the right mandibular angle. (Figure 2). The preliminary diagnosis was a dentigerous cyst, keratocyst or a type of ameloblastoma.

In order to decide the extension of the surgical act and the histological nature of the lesion; an incisional biopsy is decided under local analgesia.

The histological examination confirms the diagnosis of a mural ameloblastoma

Following the histological results, it was decided to perform a conservative excision surgery.

An intermaxillary arch retainer is placed preoperatively to prevent a possible fracture of the left mandibular angle and under local analgesia the tumor was excised and both molars removed (Figure. 3a, 3b and 3c).

A paresthesia of the right inferior alveolar nerve is observed postoperatively. She was a passenger.

Histopathological examination reveals the presence of a cystic wall on which are grafted colonies of ameloblasts outside the cystic cavity and highlights a polarization of the basal cells with a displacement of the nucleoli, hyperchromatism and vacuolation of the cytoplasm, which confirms the diagnosis of a cystic ameloblastoma. (Figure 4).

One week later a clinical healing was observed and the control X-ray shows bone healing well underway. (Figure. 5a and 5b).

One month later, the clinical exam and the panoramic X-ray reveals a good healing process and the intermaxillary arch was removed. (Figure. 6a and 6b).

One year later the panoramic X-ray reveal a complete bone healing. (Figure 7).

Four years later the bone apposition is complete and the radiographic examination shows no recurrency. (Figure 8)

Unicystic Ameloblastoma

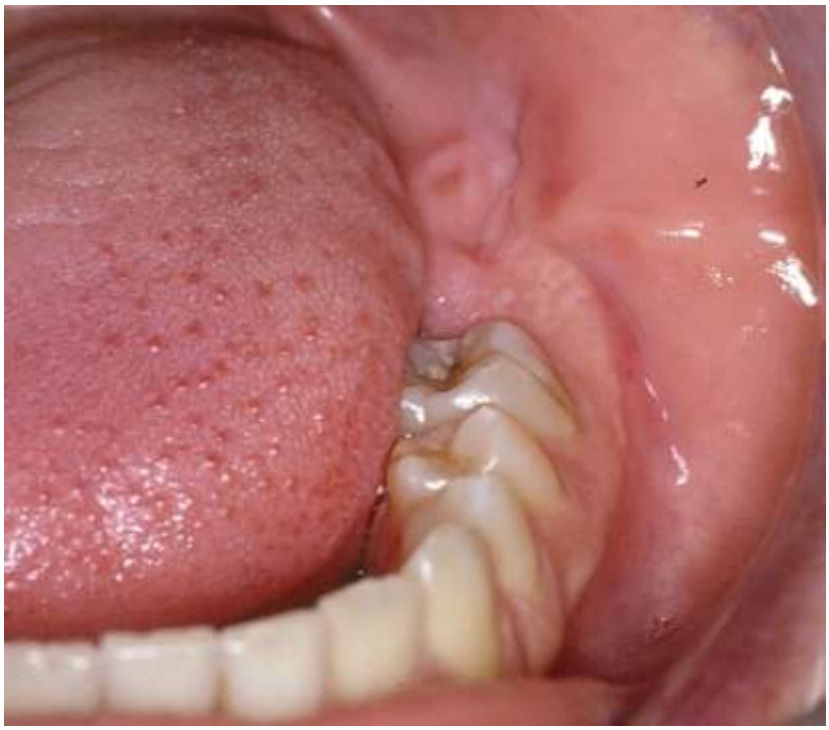


Figure 1: Clinical view.

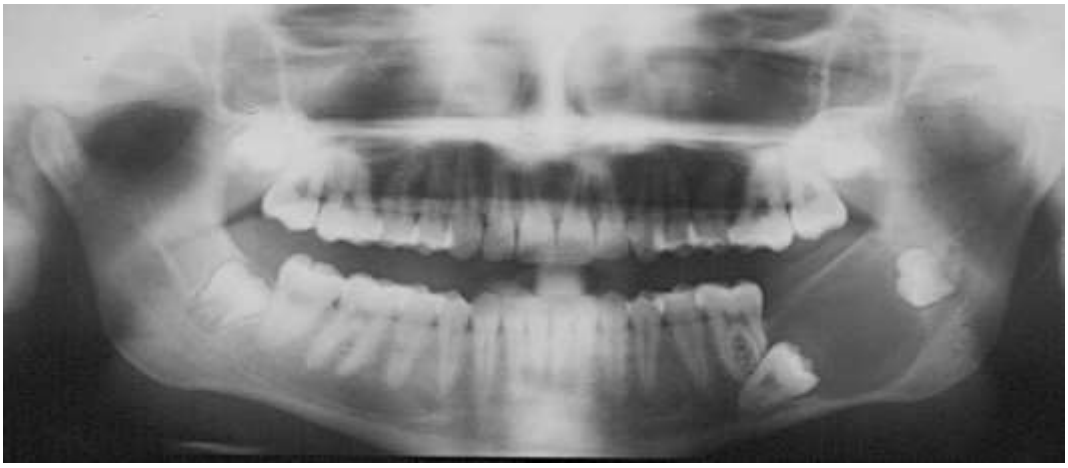


Figure 2: Preoperative panoramic X-ray showing the extension of the lesion and the associated impacted molars.

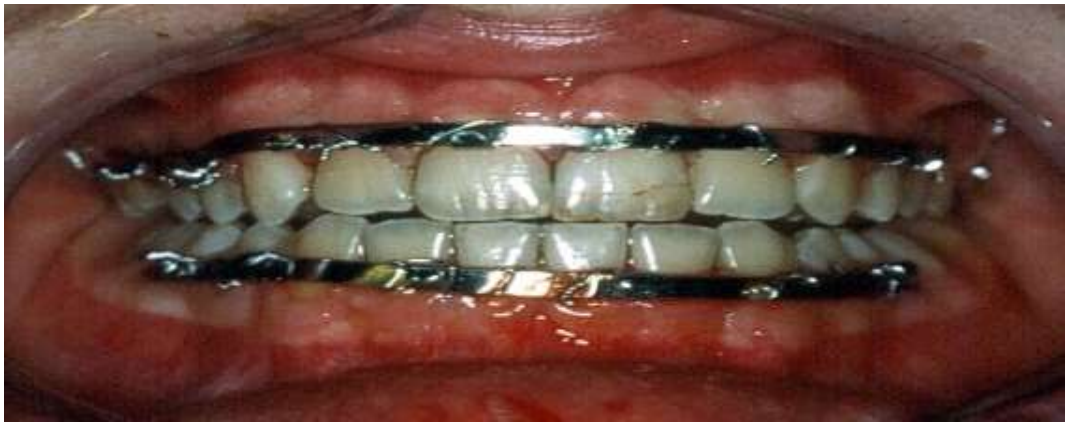


Figure 3: a- the intermaxillary arch

Figure 03 b- per-operative view of the lesion with the position of the third molar.

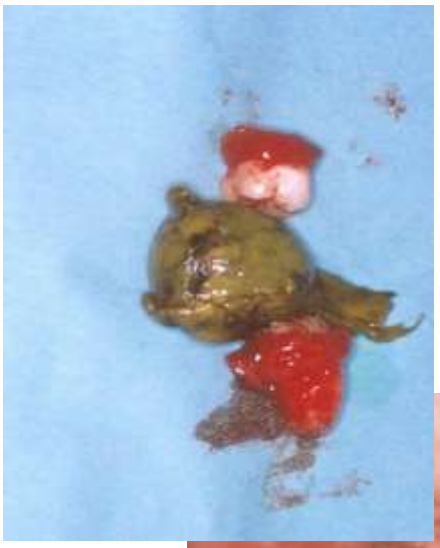
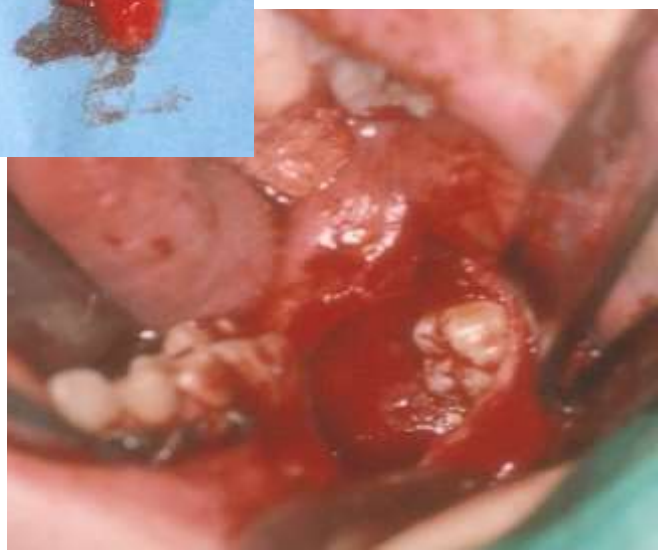


Figure 03 c- lesion specimen with the two molars.



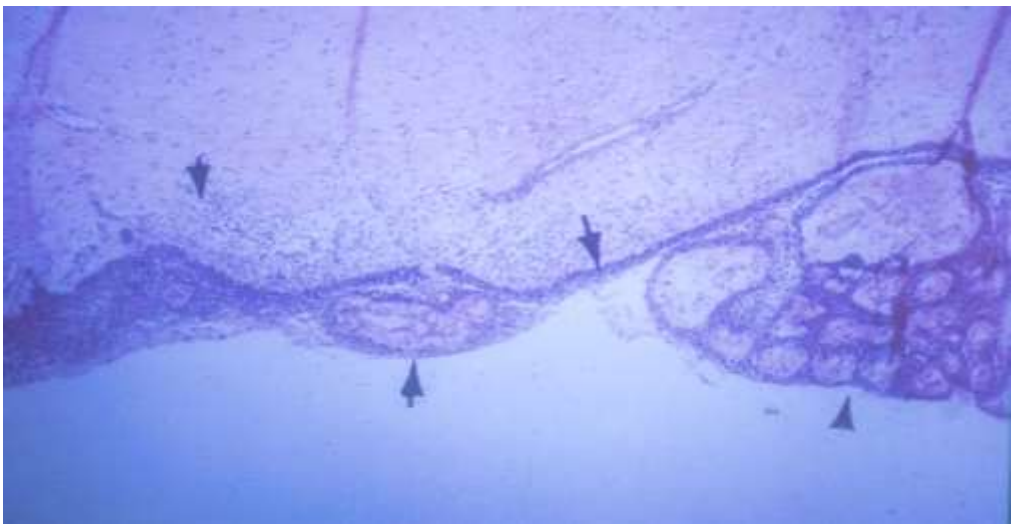


Figure 4: a- histological section at low magnification (x 10 HE): the cyst cavity is lined with an adamantine epithelium with, on the periphery, a palisade layer bordered by a layer of ameloblasts.

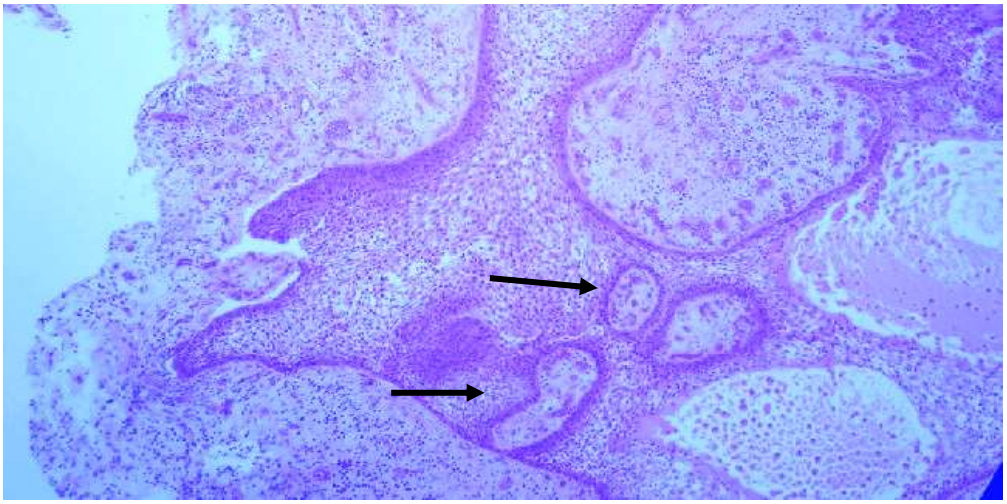


Figure 04 b- histological section at high magnification (x 40 HE): note the presence of ameloblastic follicles within the stroma.

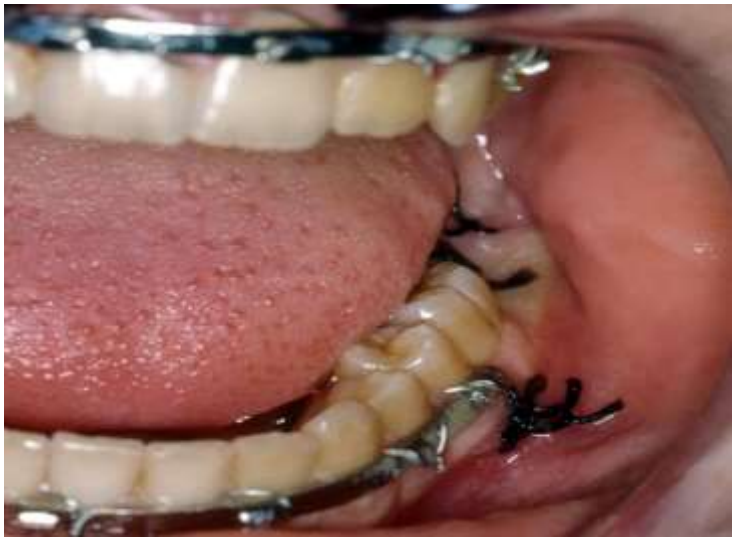


Figure 5a - clinical healing before taking out the stiches

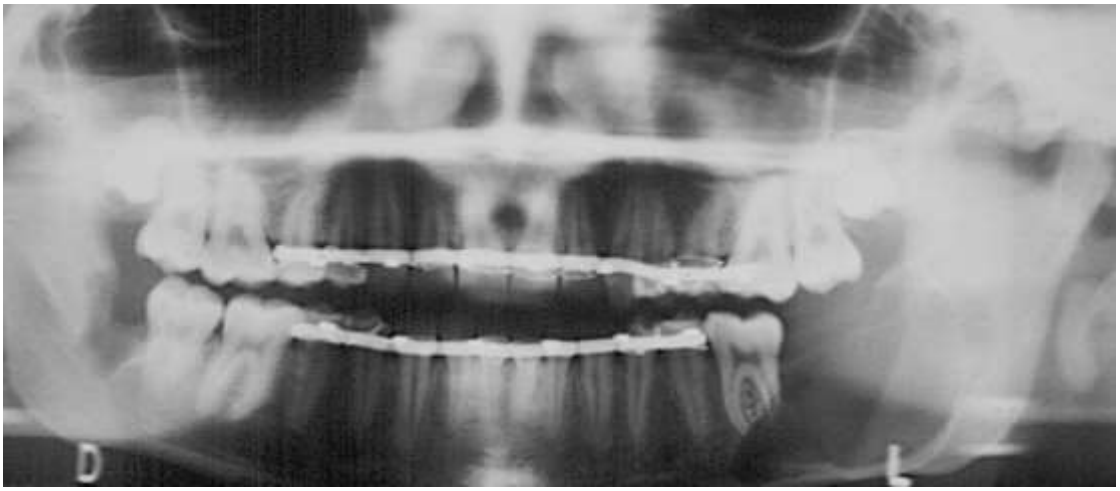


Figure 05 b- panoramic radiograph at one week.

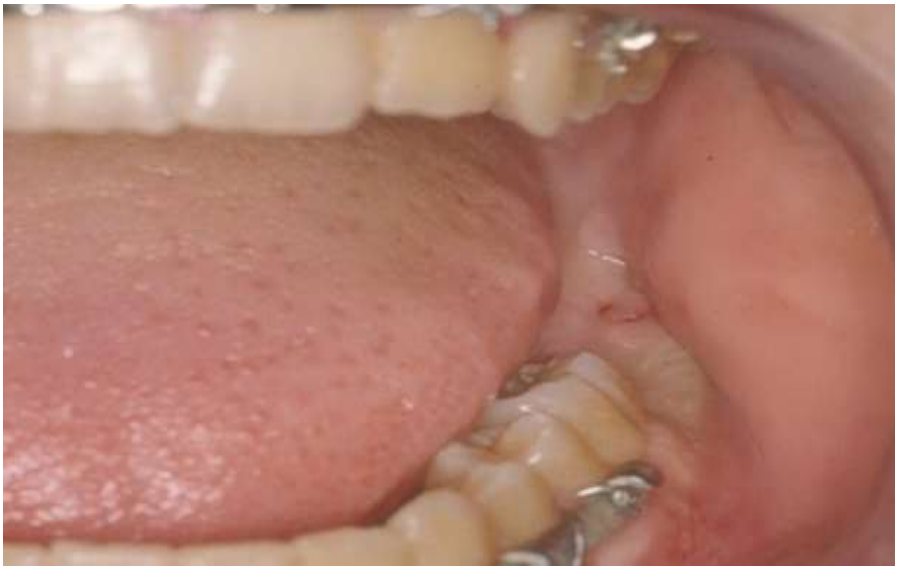


Figure 6: a- clinical healing

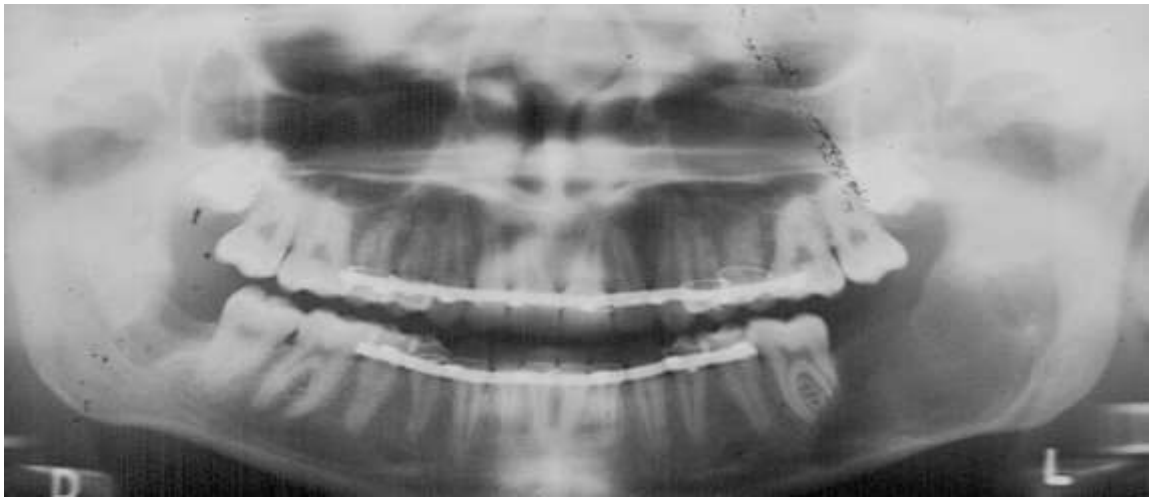


Figure 06 b- postoperative panoramic radiograph at one month.

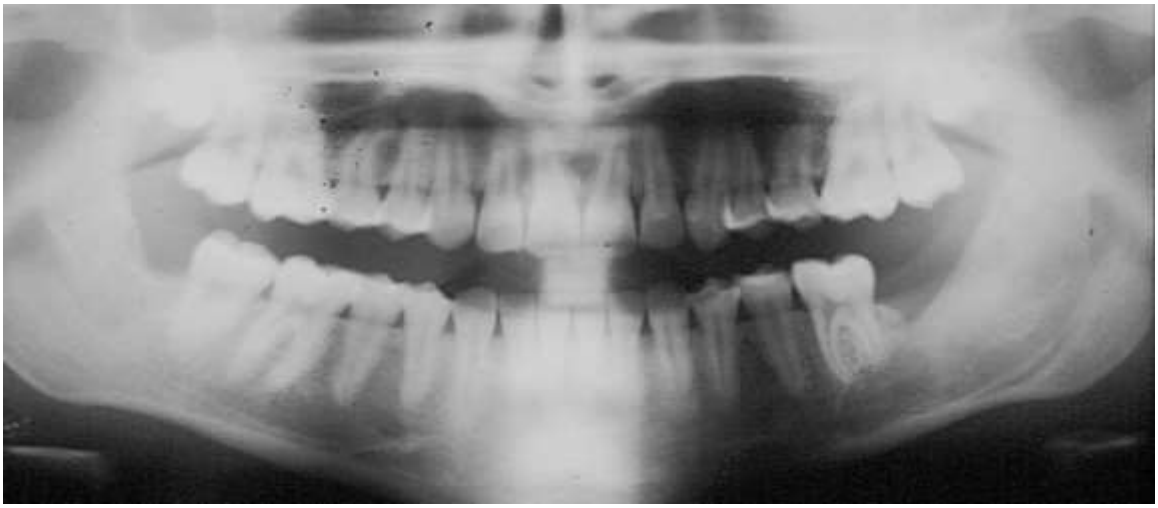


Figure 7 X-ray at 12 months.

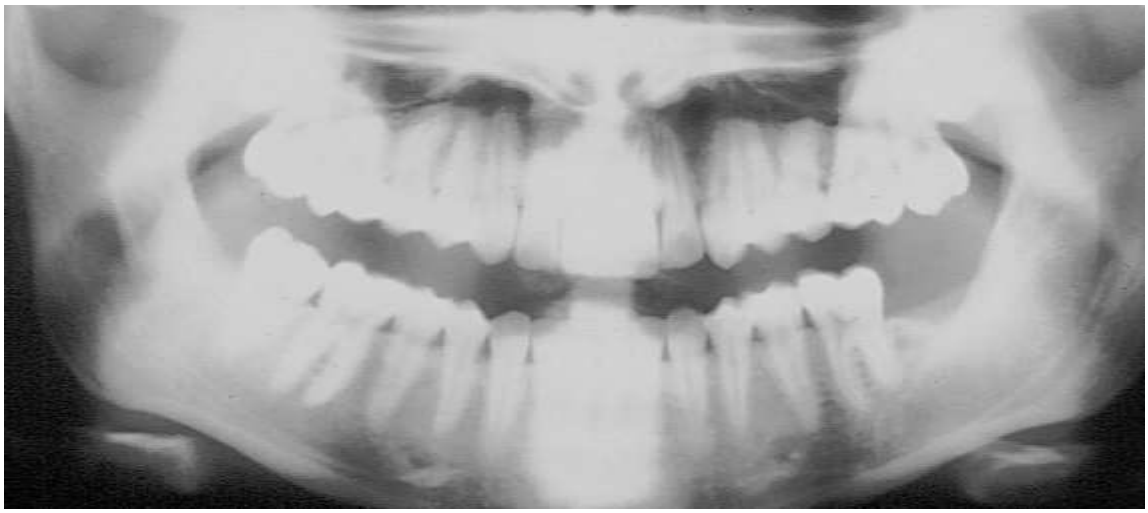


Figure 8: X-ray at 4 years, note complete healing.

Discussion

Ameloblastoma are subdivided into 2 biological/microscopic subtypes:

solid or multilocular ameloblastoma and unicystic ameloblastoma **[**3,4**]**.

There is a very significant rationale for such a subdivision as treatment and prognosis differ **[**10**]**. Solid ameloblastoma is more aggressive and requires more radical treatment than cystic ameloblastoma and shows a high rate of recurrence (50-90%) if treated by curettage **[**8, 10**]**. Unicystic ameloblastoma, on the other hand, is an ameloblastoma that has a wide cystic space in which there can be wall growth **[**5, 9**]**.

It could represent a cystic ameloblastoma which is unilocular or, an odontogenic cyst in which there have been ameloblastic transformations at the level of the epithelial wall **[**7, 9**]**. A histological variant of this type is cystic plexiform ameloblastoma in which the cyst wall shows a network of ameloblastic epithelium **[**11**]**.

Unicystic ameloblastoma are rare and constitute about 10% of all ameloblastoma: which usually are observed in young population with an average of 22 years **[**9**]**. They are characterized by slow growth and being moderately aggressive, with the highest localization in the posterior part of the mandible **[**12, 13**]**.

Radiographically, the lesions usually show expansive unilocular radiolucency's with a well-delimited margin. Around 50–80% of cases are allied with an impacted or unerupted tooth **[**9,12**]**.

Thus, the clinical and radiographic appearances of unicystic ameloblastoma are in some cases indistinguishable from those of dentigerous cysts **[**9, 13**]**.

The categorization of unicystic ameloblastoma cannot be made only on the radiological appearance, histological detecting of ameloblastic cells infiltration of the cystic wall or extramural proliferation should be performed **[**9, 12, 13**]**.

Unicystic ameloblastoma may show a variable, plexiform or follicular epithelial wall, frequently with typically ameloblastic cells in the basal layer in several places. The wall of the cystic area becomes flattened and although a few ameloblastic cells may be seen, it may resemble that of a non-neoplastic cyst **[**9,13**]**.

Conclusion

The incisional biopsy substantiated to be essential in the choice of our therapeutic attitude. It limited the extent of our surgical excision; which spared our patient a hemi mandibulectomy. The long term follow-up is essential to control bone healing.

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