

Long-term follow-up of a case of multicystic ameloblastoma treated with curettage: case report

Acompanhamento a longo prazo de um caso de ameloblastoma multi-cístico tratado com curetagem: relato de caso

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ABSTRACT

The conventional or multicystic solid ameloblastoma is the most common type with the majority of cases affecting the posterior region of the mandible and occurring between the third and fourth decades of life. The present study aims to report a case of multicystic ameloblastoma in which treatment by means of curettage was chosen and no evidence of recurrence was noted over 7 years of follow-up. Although the literature indicates

aggressive surgical therapy as the best form of treatment for multicystic ameloblastoma, factors such as the clinical, radiographic and histopathological aspects of the lesion, patient age, postoperative morbidity, possibilities for aesthetic and functional changes must be evaluated together during treatment planning. In some cases, as in the report presented, conservative therapies such as aggressive curettage of the lesion can be effective in the long term, with no recurrence of the lesion and providing quality of life to the patient.

Keywords: case report, ameloblastoma, treatment, curettage.

RESUMO

O ameloblastoma sólido convencional ou multicístico é o tipo mais comum com a maioria dos casos acometendo a região posterior da mandíbula e ocorrendo entre a terceira e quarta décadas de vida. O presente estudo tem como objetivo relatar um caso de ameloblastoma multicístico em que optou-se pelo tratamento por curetagem e não se observou evidência de recorrência ao longo de 7 anos de seguimento. Embora a literatura indique a terapia cirúrgica agressiva como a melhor forma de tratamento para o ameloblastoma multicístico, fatores como os aspectos clínicos, radiográficos e histopatológicos da lesão, idade do paciente, morbidade pós-operatória, possibilidades de alterações estéticas e funcionais devem ser avaliados conjuntamente durante o planejamento do tratamento. Em alguns casos, como no relato apresentado, terapias conservadoras como curetagem agressiva da lesão podem ser eficazes a longo prazo, sem recidiva da lesão e proporcionando qualidade de vida do paciente.

Palavras-chave: Relato de caso, Ameloblastoma, Tratamento, Curettage.

1 INTRODUCTION

Ameloblastoma is a benign epithelial neoplasm of odontogenic origin. It can originate from the epithelium of the enamel organ, remnants of the dental lamina, epithelium of dentigerous cysts or from the basal cells of the epithelium of the oral mucosa.^{1,2} Clinically, it can be classified as conventional or multicystic, unicystic or peripheral solid. The conventional or multicystic solid type is the most common type with the majority of cases affecting the posterior region of the mandible and occurring between the third and fourth decades of life. In Blacks, more cases are seen in the third decade, while Caucasians have a maximum incidence during the fourth decade.³

Ameloblastoma usually presents as an intraoral swelling of lateral and painless expansion or as an incidental finding on routine radiographic images. Radiographically, multicystic ameloblastoma appears as a well-delimited multilocular radiolucent image with the appearance of “soap bubbles”.^{3,4} There are six histopathological subtypes of ameloblastoma, follicular, plexiform, acantomatous, granular cell, basal and desmoplastic types.⁵ Although histologically benign 4.5% of all cases have malignant potential and metastasize most commonly to the lungs.⁶

The therapeutic management of these neoplasms remains a challenge due to their locally aggressive behavior and the high rate of recurrence after treatment.⁷ Previously, conservative strategies such as curettage, enucleation or marsupialization have been reported. However, they were associated with higher rates of recurrence such as 75 and 90%.^{8,9,10,11} The ideal treatment aims to achieve complete excision of the lesion with safety margins and the lowest possible morbidity, which can be more easily achieved through more radical surgical techniques such as marginal segmental or block resection with safety margins and defect reconstruction bone.^{12,13} However, in many cases, aggressive management brings with it the challenge of postoperative rehabilitation.¹⁴

Despite the high rate of recurrence of this neoplasm in the face of conservative therapies, the present study aims to report a case of multicystic ameloblastoma in which treatment by means of curettage was chosen and no evidence of recurrence was noted over 7 years of follow-up.

2 CASE PRESENTATION

A 17-year-old female patient attended a private clinic complaining of a volumetric increase in the posterior and left regions of the mandible, with mild pain symptoms. On extra-oral clinical examination, there was no facial asymmetry or lymphadenomegaly. However intra-oral clinical examination revealed volumetric increase the triangle retomolar and bottom left mandibular vestibule Sessi, similar staining adjacent mucosa and consistency hardening palpation (Figure 1).

Figure 1: Intra-oral aspect of ameloblastoma



Source: Own authorship.

Panoramic radiography was requested as a complementary examination as soon as we observed an extensive lesion that extended over the region of the body, angle, branch and left mandibular coronoid process, root resorption of teeth 36 and 37 and association of tooth 38 with the lesion (Figure 2).

Figure 2: Radiographic aspect of the lesion.



Source: Own authorship.

Thus, a computed tomography scan was performed to better assess the lesion, where the real proportions of the lesion were observed, such as the vestibulo-lingual extension and the actual involvement of the dental units involved (Figure 3).

Figure 3: Tomographic aspect of the lesion.

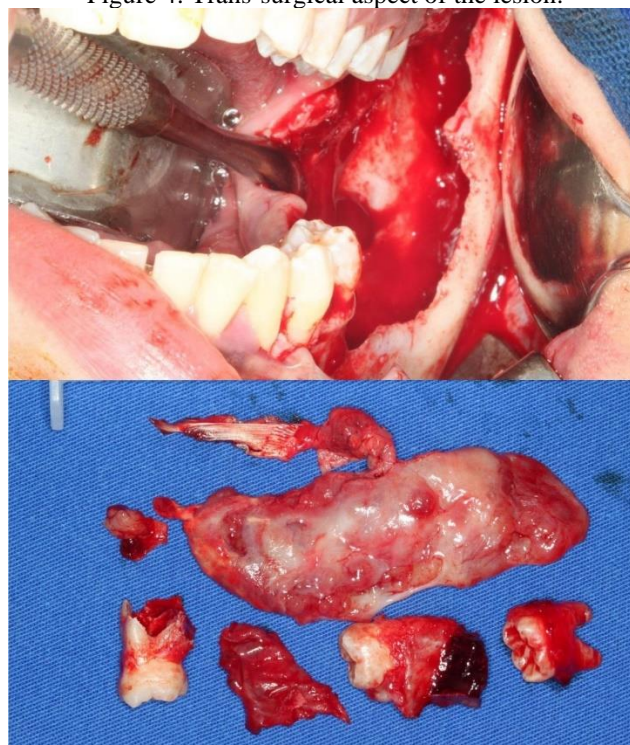


Source: Own authorship.

As hypotheses for diagnosis, the following possibilities were raised: dentigerous cyst, keratocyst or ameloblastoma. An incisional biopsy under local anesthesia was performed and, after histopathological evaluation, the diagnostic hypothesis of ameloblastoma was confirmed associating the clinical and radiographic findings and the histological result, the final diagnosis was multicystic ameloblastoma.

For follow-up treatment, the patient underwent pre-anesthetic evaluation, where no contraindications for surgical approach of the tumor under general anesthesia were identified. After nasotracheal intubation, we performed intraoral infiltration with bupivacaine with 1:100,000 noradrenaline for hemostasis of the surgical field during the procedure. Then, we performed intraoral access with full mucoperiosteal detachment. Aggressive curettage under the lesion affected large mandibular extension. We performed extraction of teeth 36, 37 and 38, abundant irrigation with saline and primary closure of the surgical bed with resorbable suture threads.

Figure 4: Trans-surgical aspect of the lesion.

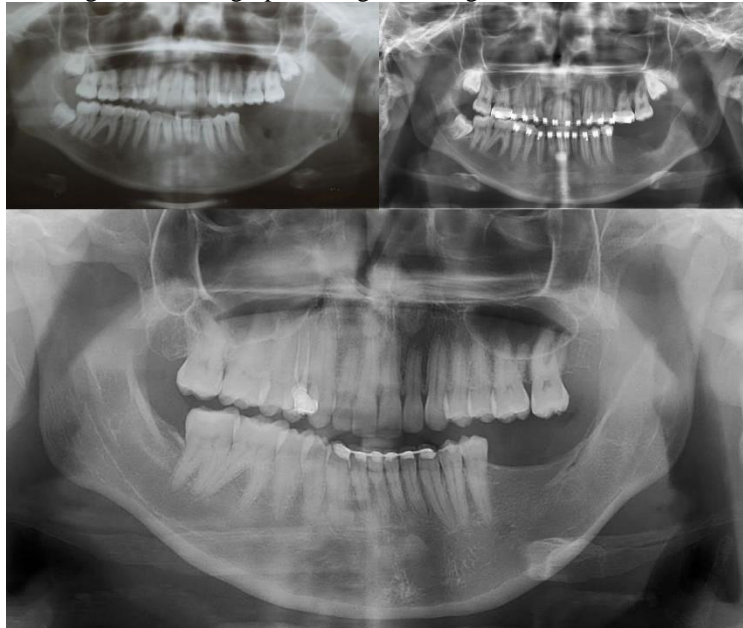


Source: Own authorship.

In the immediate postoperative period, the patient was prescribed amoxicillin 500 mg three times a day for 7 days, dexamethasone 4 mg twice a day for 3 days and sodium dipyrone 1 g four times a day for 5 days and was restricted to an exclusively liquid/past diet for 60 days.

The patient was followed up weekly during the first month after surgery and, later, the returns became more spaced with the performance of panoramic control radiographs. In the radiographic images of postoperative control, at 30 and 90 days, there was satisfactory bone repair and in the radiographic control at 7 years postoperatively, there was no radiographic sign of tumor recurrence **Fig. 5** Radiographic image showing bone neoformation

Figure 5: Radiographic image showing bone neoformation.



Source: Own authorship.

3 DISCUSSION

Although authors claim that the multicystic variant of ameloblastoma occurs more frequently during the third and fourth decades of life, in the present case the patient was diagnosed at 17 years of age [3]. At the expense of her age, the likelihood of aesthetic and functional defects in the face of invasive treatment and the possibility of long-term follow-up, we opted for aggressive lesion curettage. According to Curi et al., 1997 [15], these variables must be taken into account when determining the type of therapy, aggressive or conservative, to be instituted. However, conservative management is associated with higher rates of recurrence and curettage may bring a greater risk of aggressive relapse of the lesion with malignant transformation of ameloblastoma and the possibility of local, regional and distant metastases [11, 16]. However, we did not observe anything that recurrence of the lesion over 7 years of clinical and radiographic follow-up.

In order to make treatment more effective, conservative therapies such as enucleation and curettage were performed in conjunction with cryotherapy and application

of Carnoy's solution [15, 17]. However, the combination of the respective interventions still leads to higher recurrence rates than do invasive treatments and can lead to complications such as pathological fracture and wound dehiscence. These aspects are mainly relevant in view of fenestrations of the bone cortex and extension of the lesion in soft tissues [18, 19].

Due to its slow growth, ameloblastoma recurrences are usually seen many years after the initial intervention. Therefore, authors recommend that the clinical and radiographic follow-up be extended for at least ten years or, preferably, for the entire life of the patient [20]. We agree with the authors and we are already in the 7th years of postoperative follow-up of the patient in the case presented, without any clinical signs that indicate recurrence of the lesion. However, we also recognize and agree with Faqi et al., 2019 [21], who stated, according to the results of their systematic review with meta-analysis, that there are still no comparative studies in the literature between conservative and surgical treatments with high levels of scientific evidence involving randomized clinical trials. This can be correlated to problems with ethical issues, heterogeneity of the therapies involved, sample and reduced standardization of results related to quality of life and time of recurrence.

As much as the literature shows benefits in terms of less recurrence of the lesion in the face of radical surgical treatments such as bone resection with safety margins, other aspects must be taken into account during the treatment decision. In younger patients, such as children or adolescents, there may be greater aesthetic repercussions, changes in maxillofacial growth and development and psychological problems [22, 23]. Therefore, we evaluated all these factors together with the clinical, radiographic and histopathological aspects of the lesion. We chose to maintain a conservative approach with aggressive intra-lesional curettage and, to date, we have not had recurrence or problems with the factors already mentioned, which were assessed during treatment planning.

4 CONCLUSION

Although the literature indicates aggressive surgical therapy as the best form of treatment for multicystic ameloblastoma, factors such as the clinical, radiographic and histopathological aspects of the lesion, patient age, postoperative morbidity, possibilities for aesthetic and functional changes must be evaluated together during treatment planning. In some cases, as in the report presented, conservative therapies such as aggressive curettage of the lesion can be effective in the long term, with no recurrence of the lesion and providing quality of life to the patient.

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