

Conservative treatment of large unicystic ameloblastoma in a young patient: a 7-year follow-up

Tratamento conservador de grande ameloblastoma unicístico em paciente jovem: um acompanhamento de 7 anos

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ABSTRACT

The unicystic ameloblastoma is an odontogenic tumor that is frequently found in young patients and usually affects the posterior region of the mandible. This paper aimed to describe a case report and 7-year follow-up of a young patient. The 12-year-old patient presented with increased volume in the lower third of the right hemiface with a duration of 6 months. Radiographic examination showed a unilocular radiolucent lesion extending along the body, angle and right mandibular ramus. An incisional biopsy was performed and a histological diagnosis of luminal unicystic ameloblastoma was obtained, opting for a conservative treatment with decompression and subsequent complete enucleation of the lesion. The patient has been under follow-up for 7 years, with a good evolution of the condition and without clinical and radiographic signs of recurrence of the lesion. Thus, it is ratified that timely conservative intervention and conservative surgical treatment should be the first choice for ameloblastoma management in young patients.

Keywords: oral surgery, ameloblastoma, conservative treatment.

RESUMO

O ameloblastoma unicístico é um tumor odontogênico que é frequentemente encontrado em pacientes jovens e geralmente afeta a região posterior da mandíbula. Este artigo teve como objetivo descrever um relato de caso e acompanhamento de 7 anos de um paciente jovem. O paciente de 12 anos apresentou aumento de volume no terço inferior da hemiface direita com duração de 6 meses. O exame radiográfico mostrou uma lesão radiolúcida unilocular que se estende ao longo do corpo, ângulo e ramo mandibular direito. Foi realizada biópsia incisional e obtido diagnóstico histológico de ameloblastoma unicístico luminal, optando por tratamento conservador com descompressão e subsequente enucleação completa da lesão. O paciente está sob acompanhamento há 7 anos, com boa evolução da condição e sem sinais clínicos e radiográficos de recorrência da lesão. Assim, ratifica-se que a intervenção conservadora oportuna e o tratamento cirúrgico conservador devem ser a primeira escolha para o manejo do ameloblastoma em pacientes jovens.

Palavras-chave: cirurgia oral, ameloblastoma, tratamento conservador.

1 INTRODUCTION

Ameloblastoma is a rare, slow-growing benign neoplasia with a locally aggressive behavior. Despite its low incidence, it was reported to be the second most common type of odontogenic tumor after the odontoma, accounting for 11.7% of all cases¹. Solid/multicystic ameloblastoma, the most common form of ameloblastoma, usually has a higher destructive and infiltrative potential and a higher propensity to relapse. In contrast, unicystic ameloblastoma occurs mainly in younger patients, commonly appearing as a single cystic cavity with a similar behavior to odontogenic cysts².

The literature describes it as a slow growing tumor that rarely leads to metastasis, but may spread and cause destruction of the cortical bone, as well as invading the surrounding soft tissue, causing pain, facial asymmetry, dysphonia and dysgeusia, malocclusion, root resorption, pathological migration and paresthesia³.

Unicystic ameloblastoma was first described in 1977. It refers to a cystic lesion that presents clinical and radiographic characteristics that are similar to odontogenic cysts, but histological evaluation shows ameloblastic epithelium and its microscopic variants⁴.

Despite its clinicopathological subtypes, ameloblastoma is generally characterized by an aggressive potential for local invasion and a high tendency to recur, therefore, aggressive treatment through surgical resection is commonly recommended⁵. However, surgical resection of mandibular ameloblastoma can cause complications and functional, physical and psychological sequelae, especially in young patients.

Diagnosis is usually achieved with panoramic radiography and cone beam computed tomography, and this finding may be occasional for asymptomatic patients or in the early stages of the disease. It usually presents radiographically as a unilocular lesion, with a microscopic cystic nature, and responds best to conservative therapy⁶. Nevertheless, the definitive diagnosis is only obtained after a biopsy.

Such lesions can be classified into three histological variants: luminal, intraluminal and mural. The prognosis and treatment plan are determined according to the histological subtype⁵. Numerous treatment modalities can be implemented, ranging from enucleation, marsupialization, and resection of the affected area; however, more conservative measures tend to be adopted in young patients⁷.

Conservative treatment, such as decompression and enucleation, can preserve bone integrity and allow the mandible to continue growing. Nevertheless, such approach is associated with a high level of postoperative recurrence, in the range of 60-80%, mainly due to failure in removing remnant islands of tumor that infiltrate the bone walls¹. In answer to this challenge,

adjunctive therapies such as peripheral osteotomy, chemical cauterization and use of liquid hydrogen are added to the conservative approach, in order to mitigate the chances of lesion recurrence. Therefore, the present article aimed to describe the 7-year follow-up of a clinical case of unicystic ameloblastoma, in which a conservative treatment was proposed for a young patient.

2 CASE REPORT

A female patient, Brazilian, fair-skinned, non-alcoholic, non-smoker, presented initially to Oral and Maxillofacial Surgery Service at the age of 12 years, with no systemic comorbidities, with a main complaint of increased facial volume, which had been observed for 6 months.

The extraoral clinical examination revealed an expansile lesion, with increased volume in the lower third of the right hemiface. Intraoral clinical examination revealed increased volume in the region of the body and angle of the right mandible and the presence of tooth 46 with a temporary seal. Panoramic radiography showed a unilocular radiolucent lesion extending over the body, angle and right mandibular ramus, causing the displacement of elements 47 towards the basilar and 48 towards the mandibular ramus (Figure 1). Tomographic examination showed that the lesion caused expansion of the buccal and lingual cortical plates with areas of fenestration.

Figure 1. Initial Radiographic Image.

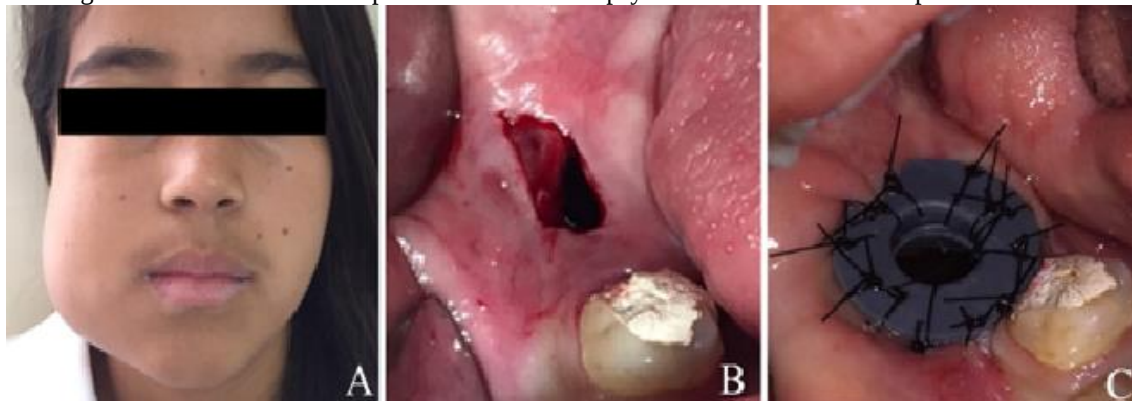


Source: Personal files, 2016.

Initially, the aspiration puncture procedure was performed to evaluate the aspect of the intralesional material and to exclude the possibility of a vascular lesion. Subsequently, an

incisional biopsy was performed and the specimen was sent for histological evaluation. In the opening left by the removal of the specimen for incisional biopsy, a flexible rubber device (adapted dropper) was installed to perform decompression and subsequent reduction of the lesion volume and consequently mitigate the possibility of a mutilating surgery in a young patient (Figure 2).

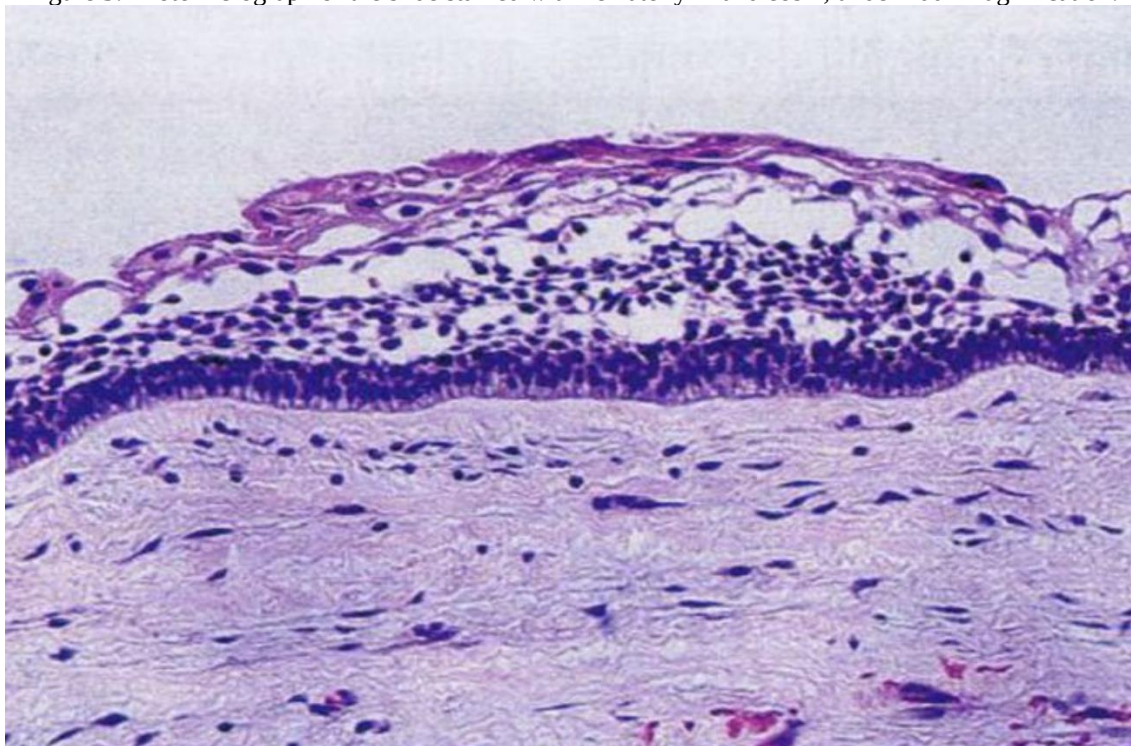
Figure 2. A: Initial clinical aspect. B: incisional biopsy. C: Installation of decompression device.



Source: Personal files, 2016.

Given the clinical and radiographic characteristics, two diagnostic hypotheses were proposed: unicystic ameloblastoma or dentigerous cyst. The initial histological diagnosis was unicystic ameloblastoma. The decompression technique was continued with daily irrigations with sterile 0.9% sodium chloride saline solution interspersed with 0.12% chlorhexidine digluconate irrigation to clean the pathologic cavity and for clinical and radiographic follow-up. After 6 months of decompression evolution, a significant decrease in the diameter of the lesion was observed and radiographic evidence of bone neoformation at the periphery of the lesion was found. Given the favorable evolution, it was decided to completely excise the lesion under general anesthesia, using the surgical technique of enucleation associated with peripheral osteotomy, in order to reduce the chances of recurrence of the lesion, along with the application of liquid nitrogen. The pathological specimen removed by surgical enucleation was sent for histopathological evaluation to ratify the initial diagnosis, with the final result of the biopsy being Unicystic Ameloblastoma (Figure 3).

Figure 3. Photomicrograph of the slide stained with hematoxylin and eosin, under 200x magnification.



Source: Personal files, 2016.

The patient has been under follow-up for 7 years, with a good evolution of the condition and without clinical or radiographic characteristics of recurrence of the lesion (Figure 4).

Figure 4. Radiographic follow-up after 7 years.



Source: Personal files, 2023.

3 DISCUSSION

The optimal approach to ameloblastoma should aim to minimize recurrences, restore function and aesthetics and present minimal physical, functional, and psychological sequelae^{8,9}. Kalaiselvan et. al.⁷ mention that surgical planning should take into consideration parameters such as the presence of comorbidities and the patient's age, the dimensions and location of the tumor and available techniques for reconstruction.

Unicystic ameloblastomas usually present as slow-growing, persistent, locally invasive lesions that can result in bone deformities⁷. These expansive lesions present well-defined radiolucent areas, which are surrounded by a sclerotic border, preferentially in the mandible posterior region⁵. In this case report, it is possible to observe a unilocular radiolucent alteration that extended over the body, angle and right mandibular ramus, causing the displacement of teeth 47 and 48. Therefore, this clinical presentation corroborates the previously found literature.

Among the age range of incidence of these lesions, most cases are diagnosed up to the second decade of life¹⁰. Painful conditions are mainly associated with older patients and not directly correlated to tumoral growth¹¹. Thus, as in this report, a young patient (12 years old), asymptomatic, exhibiting root resorption, corroborates previously published findings in which the average age of these lesions varies around 13 years and is predominantly found in the mandible⁴.

Unicystic ameloblastomas exhibit three histological variants, where the luminal subtype of the tumor is confined to the luminal surface of the cyst and the cystic wall is totally or partially lined by ameloblastic epithelium; the intraluminal variant presents ameloblastoma nodules projecting into the cystic lumen; and finally, the third variant called mural, in which the cystic wall is infiltrated by ameloblastoma¹¹. The characteristics described after the analysis of the surgical specimen in the present case are compatible with the luminal aspect.

However, there is a philosophical current^{12,13} that disseminates that ameloblastomas should be treated radically to avoid recurrence. Nevertheless, there is a dilemma whenever the treatment plan involves children, because mandibular lesions at this age are usually benign, and thus, conservative surgeries are the treatment of choice, since at this age there will be continued bone growth and presence of unerupted teeth³.

Jitesh and Madhulaxmi¹⁰ believe that radical surgery involves large resections such as marginal or segmental mandibulectomy with the need for 1-to-1.5-centimeter margins, since ameloblastoma cells are usually found 8 mm away from the radiographic and clinical borders

of the tumor. Nevertheless, the low recurrence rate, around 0-10%, makes this technique a viable option to avoid new approaches and interventions².

Among the reported procedures, it is worth mentioning enucleation, marsupialization with subsequent enucleation, enucleation and chemical cauterization (Carnoy's solution), intralesional application of liquid nitrogen, decompression followed by enucleation and peripheral osteotomy; the cases in which en bloc resection is chosen are rare¹. These findings are in agreement with the conduct adopted in the present clinical case, where decompression was performed through a flexible device installed at the time of the incisional biopsy, followed by enucleation associated with peripheral osteotomy and application of liquid nitrogen to mitigate the chance of recurrence. The patient has now been under follow-up for seven years, with no evidence of relapse.

The efficacy of a given treatment modality is determined by means of the recurrence rate after treatment. The recurrence rate is lower with radical treatment when compared to more conservative treatment modalities. Lau and Samman¹⁴ studied through a systematic literature review the recurrence rate after several treatment modalities and observed that the recurrence rate was 30.5% after enucleation alone, 18% for marsupialization, 16% for enucleation followed by application of Carnoy's solution and 3.6% for resection.

According to Jitesh and Madhulaxmi¹⁰ although a low recurrence rate after resection is reported, radical treatment should be avoided in children for the following reasons: (1) continued facial growth in children (2) possibility of causing aesthetic disfigurement (3) and masticatory and functional problems, which may be psychologically deleterious to the child's development. Thus, a conservative treatment approach performs a salutary role in pediatric and adolescent patients, since it is associated with satisfactory success rates when well indicated and resolute, presenting bone neoformation and restoration of bone architecture and topography, as described in the present case.

4 CONCLUSION

Thus, it is ratified that timely conservative intervention and conservative surgical treatment associated with local adjuvant therapies such as the use of liquid nitrogen, peripheral osteotomy, and extraction of teeth closely related to the lesion should be the first choice for treating unicystic ameloblastoma in young patients. This approach provides good results and reduces the chance of complications and aesthetic, psychological, social, and functional sequelae related to large resections.

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