EFFECTIVE DECOMPRESSION AS A CONSERVATIVE TREATMENT FOR A LARGE UNICYSTIC AMELOBLASTOMA OF THE MANDIBLE: CASE REPORT

LAIZ MOREIRA DE **PAULA**[1*], LUCAS TEIXEIRA **BRITO**[2], LEONARDO VITOR **MAGRI**[1], FLAVIA ANDREZZA GOMES **ALVES**[3]

1. Dentist, Resident of Oral and Maxillofacial Surgery, State Emergency Hospital of the Northwest Region of Goiânia Governador Otávio Lage de Siqueira (HUGOL), Goiania, Goias, Brazil; **2.** Dentist, Professor and Oral and Maxillofacial Surgeon, Goiania, Goias, Brazil. **3.** Dentist, Professor and Oral and Maxillofacial Surgeon, State Emergency Hospital of the Northwest Region of Goiânia Governador Otávio Lage de Siqueira (HUGOL), Goiania, Goias, Brazil.

* Street T-33, Nº 125, Bueno district. Goiania, Goias, Brazil. ZIP CODE: 74215-140. laizmoreirap@gmail.com

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ABSTRACT

Ameloblastoma is a benign odontogenic tumor that can be highly invasive and aggressive. It often occurs in the jaw of young patients. Surgical decompression followed by enucleation is commonly used as a treatment for this injury. We report the clinical case of a 19-year-old male patient who was diagnosed with a large intraluminal unicystic ameloblastoma. The proposed treatment was surgical decompression for 12 months, followed by enucleation and peripheral osteotomy. The objective of the report is to present the benefits of conservative treatment and the possibility of its implementation in cases of unicystic ameloblastomas, in order to conserve soft, bone and dental tissues, eliminate facial disfigurement, preserve masticatory capacity and reduce psychosocial consequences for the patient.

KEYWORDS: Ameloblastoma; Surgical decompression; Conservative Treatment; Jaw Neoplasms; Mandibular Osteotomy.

1. INTRODUCTION

Ameloblastoma is a benign odontogenic tumor, derived from the epithelium, which can be highly invasive and aggressive[1]. The most common site is the mandible, affecting about 87% of reported cases[2], and its slow and asymptomatic growth can cause severe facial asymmetries. It is a tumor with high recurrence rates after surgical treatment and that can rarely turn into a malignant lesion if not treated[3].

The new classification of odontogenic tumors, updated in 2017, reclassified ameloblastoma into: "conventional ameloblastoma", "unicystic ameloblastoma", "peripheral/ extraosseous ameloblastoma" and "metastatic ameloblastoma", based on their clinics characteristics. The subdivisions 'solid/ multicystic ameloblastoma' and 'desmoplastic ameloblastoma' is not used in the classification actually[4]. Unicystic ameloblastoma is found in 26.2% of reported cases[2] and has a lower recurrence rate after conservative treatment. Microscopically, three histopathological variants can be identified: luminal, intraluminal and mural.

The treatment options commonly used for large cystic lesions are: enucleation, decompression or marsupilization followed by curettage or enucleation, radical resection or mandibulectomy. Conservative treatment is related to a high recurrence rate, while aggressive treatment can cause severe deformities and require challenging reconstructions[5]. Surgical decompression followed by enucleation is considered an excellent alternative for conservative treatment, when properly indicated and performed[6].

The objective of this report is to present the benefits that conservative treatment can provide to the patient, when performed correctly and with collaboration and commitment, through the case report of a young patient who was diagnosed with a large intramural unicystic ameloblastoma in the mandible.

2. CASE REPORT

A 19-year-old male patient, leucoderma, was admitted to an emergency hospital complaining of painful symptoms with a 3-week evolution, associated with a hardened volume increase in the mandibular region on the right side and lower lip numbness. He was in good general condition and denied known comorbidities. The intraoral examination revealed a deep effacement of the mandibular vestibule in the region between teeth 44 to 48, associated with expansion of the buccal and lingual bones and mild mobility in teeth 46 and 47.

A computed tomography (CT) image was requested at the hospital to better elucidate the case, and an aspiration puncture was performed. The CT images concluded a hypodense, multilocular, well-delimited area, located from the alveolar crest to the basilar cortical bone, extending from tooth 44 to near the angle of the mandible, measuring about 55x33x29mm, causing expansion and thinning of corticals, in addition to root resorptions in teeth 46 and 47 (Figure 1). The aspiration puncture resulted in a yellow citrine liquid.

Initial hypotheses suggested the presence of a cystic lesion. An arch bar was installed in the lower dental arch to provide additional stabilization for the mandible and for the affected teeth. We performed an incision at the

mandibular vestibule to collect a fragment of the lesion capsule for biopsy and installation of a decompression device, made from a Foley probe cut and sutured to the mucosa oral with 2-0 Nylon suture thread (Figure 2). The patient was instructed to irrigate the cystic cavity daily.

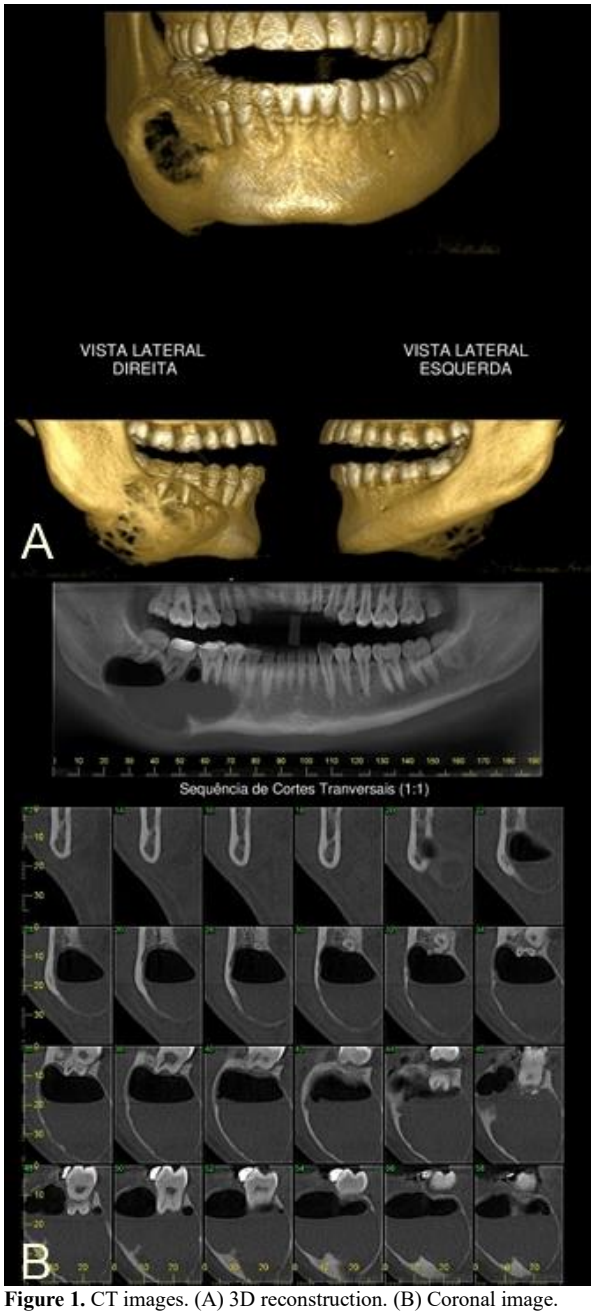

Figure 1. CT images. (A) 3D reconstruction. (B) Coronal image.

Cytologcal examination of the yellow citrus fluid concluded inflammatory cells. The collected fragment was processed in hematoxylin and eosin and the histopathological exam concluded the presence of an intraluminal unicystic ameloblastoma.

The patient was instructed to maintain the irrigation of the cystic cavity and the monthly follow-up. The CT image after 02 months of decompression revealed that the lesion was regressing and there was bone growth in its margins. Therefore, the treatment initially proposed was being satisfactory for the present case. The device was then kept in position for a period of 12 months and a new CT image revealed that there was a sufficient reduction of the lesion to avoid damage to the involved vital structures and mandibular fracture (measurement 22x10x18mm) (Figure 3).

Figure 2. Clinical image of arch bar and decompression device installed.

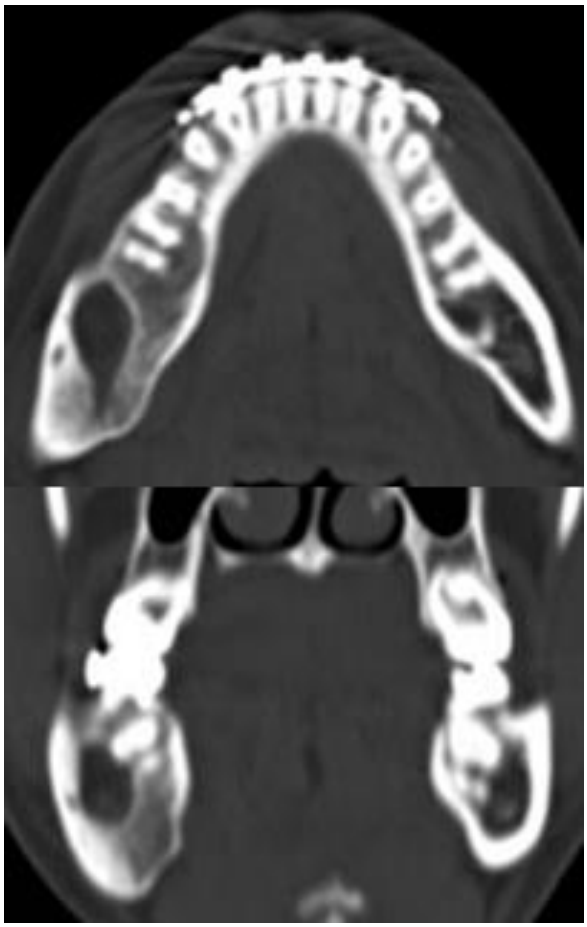

Figure 3: Axial and Coronal CT images after 12 months of decompression.

A new surgical intervention was performed, which consisted of removing the arch bar from the lower dental arch and removing the decompression device. The remaining lesion was enucleated from the mandible, and curettage and peripheral osteotomy of approximately

1mm from the lesion margin were performed (Figure 4).

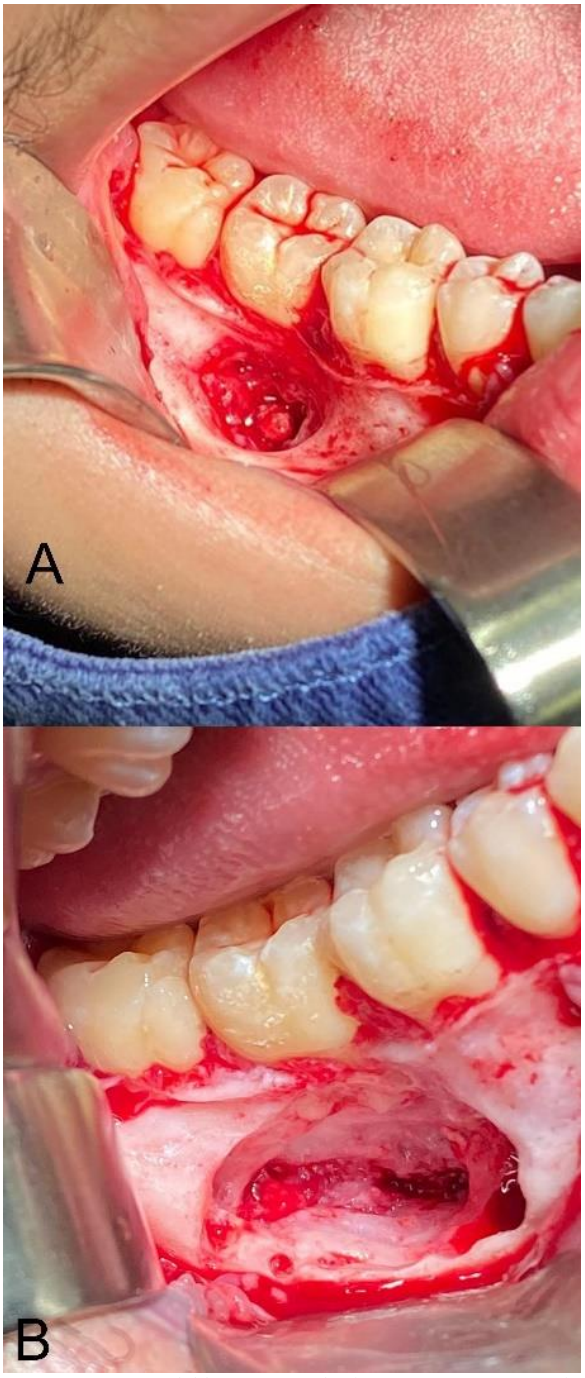

Figure 4. (A) Ameloblastoma enucleation. (B) Appearance after peripheral osteotomy.

After abundant irrigation with saline solution, the surgical access was sutured with a 4-0 Vicryl suture thread. Biopsy of the remaining lesional tissue maintained the diagnosis of unicystic ameloblastoma. After 12 months of follow-up, a CT showed bone formation in the entire bone cavity (Figure 5). The apexes of the roots of the affected teeth were expelled from the bone cavity and were covered with newly formed bone, and the inferior alveolar nerve canal was moved to the normal anatomical position. The patient evolved with no facial asymmetry, no motor or sensory deficits, normal mandibular bone contour, complete bone remodeling, maintenance of pulp vitality in teeth 44, 45, 46 and 47 and so far, no signs of ameloblastoma recurrence.

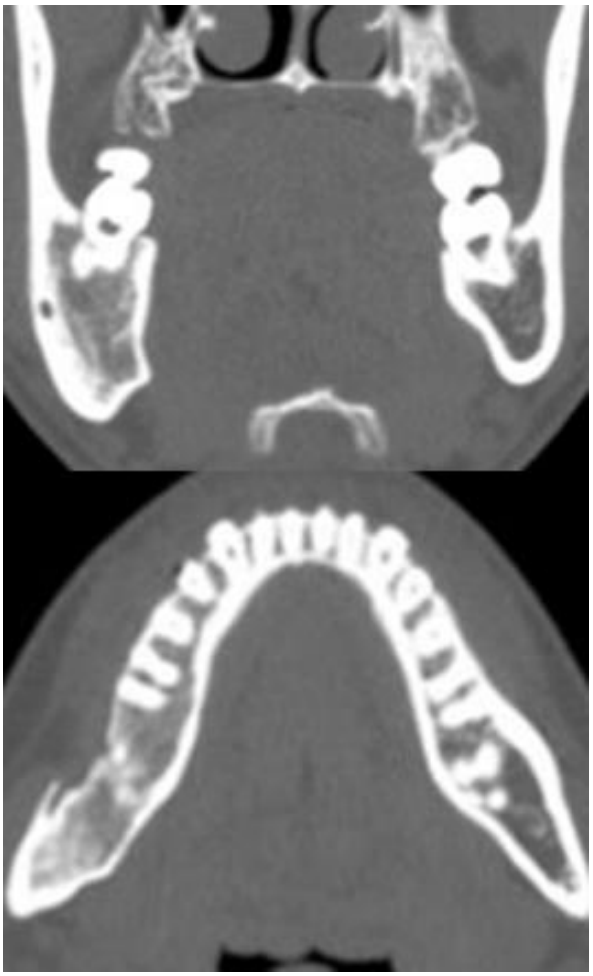

Figure 5: Coronal and axial CT images after 12 months of definitive treatment.

3. DISCUSSION

The different cystic lesions of the mandible, such as odontogenic keratocyst, dentigerous cyst and unicystic ameloblastoma, have similar radiographic and clinical presentations, which make them indistinguishable during the initial examination. Although ameloblastoma is characterized by a larger size and expansion than the other compared lesions, when found in the mandible, it is essential to perform complementary examinations such as aspiration puncture and incisional biopsy before making any treatment decision. Computed tomography is the gold standard imaging exam to visualize the buccolingual expansion, bone and root resorptions, calculate the volume of the hypodense area and verify the relationship with noble structures of large cysts[8].

Unicystic ameloblastoma is the cystic variant of ameloblastoma. It is represented by a localized, encapsulated lesion filled with citrine yellow liquid and can be uni or multilobulated. The tumor can grow into the lumen or into fibrous connective tissue. Among the histological types, the mural component has a greater number of reports of recurrence after surgical treatment,

therefore, it should be conducted as a conventional ameloblastoma[9,10]. As it is commonly confused with other odontogenic and inflammatory cysts, unicystic ameloblastomas are usually treated by enucleation, when presented in smaller size. The biopsy of these lesions is important to correctly diagnose the lesion found, and to guarantee the necessary follow-up[11].

There is a predilection for the location in the mandible, especially in the body and ramus regions. When located in the maxilla, the tumor has a aggressive clinical presentation, due to the spongy characteristic of the bone tissue. By invading the trabecular spaces, ameloblastoma can extend its limits and invade the central nervous system, if not treated in the initial phase. Its increased invasiveness and makes it difficult to treat secondary injuries. Therefore, unlike mandibular ameloblastomas, aggressive treatment must be rigidly performed when diagnosed in the maxilla[12,13].

Even today, it is quite controversial and there is no agreement on the best form to treat ameloblastoma. Surgeons have divergences in the proposed treatment modes, so the discussion between the choices of conservative or radical treatment is still quite present in the literature[14]. A study by Hendra in 2019 concluded that conventional ameblastoma is usually treated radically and unicystic ameloblastoma is treated conservatively. The recurrence rate after conservative treatment was 41% for the solid variant and 21% for the unicystic variant. Therefore, after evaluating the patient's factors, clinical, tomographic and histological aspects of the lesion, it may be possible to indicate conservative treatment for unicystic ameloblastoma[15,16].

Within the conservative treatment options, decompression is performed through the installation of a device that communicates the cystic cavity with the oral cavity, allowing constant irrigation and washing of the interior of the lesion. The device sutured in the oral mucosa prevents this communication from closing during treatment and facilitates irrigation by the patient. This type of treatment allows for the reduction of intracystic pressure, maintenance of pulp vitality and the eruption of the teeth involved, preservation of anatomical structures and avoiding pathological fractures and the need for major surgical procedures and reconstructions. The time required for a satisfactory reduction of ameloblastoma is not yet described in the literature, but it ranges from 2 to 80 months[6].

The patient in the clinical case presented above is young and was diagnosed with an extensive lesion, which included the inferior alveolar nerve canal, the lower edge of the mandible and posterior teeth, so the decision to perform conservative treatment was intended to avoid grotesque tissue losses, need for major reconstructions and changes in their psycho-emotional state. The proposed treatment was surgical decompression followed by enucleation and peripheral osteotomy with a 1mm margin. The benefits of this choice were: procedures performed under local anesthesia, no hospitalization required, less morbidity due to the absence of a donor tissue, preservation of involved teeth, reestablishment of the sensory innervation of the inferior alveolar nerve and absence of aesthetic deformities.

The disadvantages of conservative techniques are related to the risk of the patient being submitted to a second approach, in cases of recurrence and the need for collaboration and patient commitment throughout the follow-up[17]. The irrigation and maintenance of the device in position are essential for a successful treatment and these factors are the responsibility of the surgeon and the patient. In cases where there is no satisfactory interrelationship between both parties, surgical decompression must be aborted and a aggressive treatment instituted.

Radical treatment of maxillofacial pathologies assumes that tumor cells infiltrate the bone beyond the radiographic contour, requiring 1-1.5 segmental bone resections beyond the apparent tumor margins. As a result, extensive and considerable losses are generated for the patient, who will need complex reconstructive and rehabilitative processes[18]. The reconstruction must be performed immediately after the resection, ideally, to reduce functional and aesthetic defects[19]. Additionally, even if the removal of a considerable and sufficient area is performed, there is still a risk of recurrence of the lesion. The treatment of a recurrent lesion in a resected site and in the process of reconstruction is even more challenging[20].

Although radical treatment provides a shorter treatment time and reduces the chances of recurrence, it is suggested by several authors that a conservative approach be used as the initial treatment, waiting for physiological bone regeneration and avoiding aggressive surgeries when dealing with large and aggressive lesions in the jaws[21]. Young patients are the most elected for conservative treatments, which implies a follow-up period of approximately 5 to 20 years[22]. In the case presented, the period of 12 months was sufficient to reduce more than 90% of the volume of the initial lesion.

4. CONCLUSION

Conservative treatment of ameloblastoma in young patients allows for the conservation of soft tissue, bone and teeth, eliminates facial disfigurement, preserves masticatory capacity and reduces psychosocial consequences. As it is a variant with a lower recurrence rate, it is still valid to consider the conservative approach in cases of unicystic ameloblastoma. It is important to make it clear to the patient that the follow-up must remain rigid for several years and that in the presence of recurrence of the lesion, radical surgical treatment will be necessary.

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