Recurrence of multicystic ameloblastoma: case report

Recorrência de ameloblastoma multicístico: relato de caso

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Introduction

Ameloblastoma is an odontogenic tumor that grows slowly, but is locally invasive and infiltrating1-3. It has a destructive potential and the combination of proliferative lesions to malignant transformation have been documented4,5. Its etiology is not completely understood, but is believed to be originated from the remaining dental lamina, reduced enamel epithelium, epithelial cell rests of Malassez, or from the basal cells of the epithelial surface layer5,6. There is no prevalence for gender or race, and it affects predominantly the mandible (80-85% of cases)7.

The solid or multicystic ameloblastoma is a subtype that shows a greater tendency to infiltrate the surrounding tissues. It shows a higher recurrence rate, and requires quick and precise approach for treatment implementation1-8. Thus, once the diagnosis is established, the treatment plan is drawn from the clinical and radiographic findings, and may be aided by CT and MRI1-9.

By presenting unique biological behavior, there is controversy about the best way to treat ameloblastomas. Several therapeutic modalities have been proposed, including curettage, marsupialization, cryosurgery, electrocautery, sclerotherapy, and radiotherapy. However, the conventional treatment for solid ameloblastomas, in most cases, is radical.
surgical excision with 1 to 2 cm of safety margin beyond the limits of the radiographic lesion\textsuperscript{12,10}.

Radical techniques bring with them serious inconvenience to the patient, including masticatory dysfunction, change of mandibular movements, and facial mutilation and deformity\textsuperscript{11}. Treatment should include rehabilitation, restoring functional, anatomical, and esthetic abilities, as well as assisting the reintegration of the patient to society\textsuperscript{12}.

This study aims to report the case of multicystic ameloblastoma treated with marginal resection of the mandible, and to discuss relevant issues regarding diagnosis and treatment of this condition.

### Case report

A 34-year-old patient was referred to the Stomatology service at the Bauru School of Dentistry of the University of São Paulo (USP), Brazil, for assessment of multilocular radiolucent lesion involving the areas of the body, angle, and the right mandibular ramus. Clinically, the lesion was covered with mucosa of regular color and there was no facial asymmetry (Figure 1).

![Initial panoramic radiograph](source: all figures are prepared by the authors.)

Figure 1 – Initial panoramic radiograph

Marsupialization was performed to collect material for biopsy and diagnosis was pulse granuloma. After 4 months, the lesion receded (Figure 2).

![Panoramic radiograph after marsupialization](source: all figures are prepared by the authors.)

Figure 2 – Panoramic radiograph after marsupialization
In the clinical and radiographic postoperative control after one year, multilocular radiolucent area in the mandibular angle was observed (Figure 3).

New biopsy was performed and the histopathological examination revealed an ameloblastoma. The patient underwent a surgical procedure via three-dimensional margin resection including larger cortical and medullary portion of the mandibular body and ascending ramus, preserving the base of the mandible and the posterior region of the ascending ramus. A 2.4 mm reconstruction plate system was fixed, overlapping the lower residual cortical bone for maintenance of the surgical site (Figure 4).
Postoperative controls were conducted every three months, and 20 months after the surgery the patient returned for postoperative control without signs and symptoms of recurrence (Figure 5).

Figure 5 – Postoperative radiograph (1 year)

Discussion

The WHO classifies ameloblastoma as a benign epithelial odontogenic tumor without ectomesenchyme with locally invasive behavior and a high recurrence rate. It represents 11 to 18% of all odontogenic tumors primarily affecting the posterior mandible\textsuperscript{6-13}. Generally asymptomatic and slow growing, it can perforate the cortical bone, resorb teeth, and cause facial asymmetry\textsuperscript{1}.

Odontogenic cysts and tumors are a group of lesions arising from the tooth-producing apparatus or its remnants. They may originate from odontogenic epithelium and/or ectomesenchyme with varying degrees of inductive tissue interaction\textsuperscript{14-16}. Odontogenic cysts and tumors are rare, and the lack of familiarity with these lesions and their variable appearance may lead to difficulties in diagnosis, with occasional serious confusion with other lesions. Thus, the clinician should perform a careful biopsy, with sufficient amount of tissue, avoiding misinterpretation of the histopathological examination due to lack of material, as probably occurred in the first biopsy\textsuperscript{12,17,18}.

Ameloblastomas are classified into unicystic, multicystic, peripheral, and malignant subtypes, whereas conventional ones are solid or multicystic (86% of cases), unicystic (13% of cases), and peripheral or extra-osseous (about 1% of cases) (6). Such distinction is important, as it aids treatment selection.

Conventional ameloblastoma tends to infiltrate the bone trabeculae of cancellous bone in the periphery of the lesion, prior to bone resorption being visible radiographically. Therefore, the true tumor margin often extends beyond the apparent clinical or radiographic margin\textsuperscript{2}.

The best form of treatment has been controversial for several years. Radiotherapy is not indicated because the lesion is radioresistant. There is also an indication in the literature of electrocautery, cryosurgery, and injection of sclerosing agents as alternative treatments\textsuperscript{9,16}.

A high potential for recurrence (60 to 80%) after simple treatment with enucleation or curettage is frequently observed, justifying the choice of professionals for more aggressive treatments\textsuperscript{10}. An adequate alternative for the treatment of small lesions of multicystic ameloblastoma is marginal resection, which is widely used; however, due to the recurrence rate of approximately 15%, it is recommended that the safety margin should be at least 1 cm beyond the radiographic limits\textsuperscript{6}.

Evaluating several studies in order to compare the recurrence rate of treatments with complete and marginal resection, no significant difference was observed between the approaches\textsuperscript{16,17}.

The advantages of less extensive surgical treatments such as marginal resection of the mandible are significant, improving patient prognosis. Facial deformity, masticatory dysfunction, abnormal jaw movement, and the need for further reconstructive surgeries are consequences often associated with treatments of complete resection, and may often be minimized with the use of marginal resection\textsuperscript{10,14}. However, cases of extensive lesions affecting great part of the maxillary cortical bone or mandibular lesions by anatomic considerations suggest a more
aggressive approach with segmental resection (without maintaining bone continuity) including periosteum and overlying soft tissues\textsuperscript{15,16,17}.

The interpretation of clinical and imaging data of patients with ameloblastoma is essential for early diagnosis and effective treatment of the lesion. The therapeutic decision must aim to eliminate the pathology, considering the morbidity of treatment method and the quality of life in the rehabilitation of patients\textsuperscript{15,18}.

The long-term postoperative follow-up with imaging is essential because, although more than 50\% of recurrences happen within the first five postoperative years, the lesion has the potential to develop late recurrences\textsuperscript{2,9,18}. Due to its slow growth, the reappearance of the lesion may occur after many years or even decades from the first surgery, and when this happens, the lesions tend to show a higher potential for invasion and bone destruction than the original ones\textsuperscript{15}.

Conclusions

The treatment by marginal resection with a safety margin was an efficient alternative in this reported case of multicystic ameloblastoma in the mandible, avoiding mutilation by a more radical treatment. Although the conservative approach proved itself effective so far in the case described, the long-term follow-up is an essential part of treatment considering their local aggressiveness and high recurrence rate.

Resumo

Introdução: o ameloblastoma é classificado pela Organização Mundial de Saúde (OMS) como um tumor odontogênico epitelial benigno sem ectomesenquima odontogênico com comportamento localmente invasivo e alta taxa de recorrência. Representa 1\% dos tumores orais, afetando principalmente a região posterior da mandíbula. Geralmente assintomático, de crescimento lento, pode perturbar a cortical óssea, reabsorver e movimentar dentes e causar assimetria facial. Esse tipo de tumor pode ser classificado, de acordo com os aspectos clínicos e radiográficos, em ameloblastoma sólido ou multicístico, unicístico e periódico. Objetivo: relatar um caso de ameloblastoma multicístico tratado com ressecção marginal da mandíbula. Relato de caso: paciente de 34 anos encaminhado para avaliação de lesão radiolucente multiloculada envolvendo as regiões de corpo, ângulo e ramo mandibular direito, revelando ao exame histopatológico o diagnóstico de ameloblastoma. O paciente foi submetido à cirurgia resseciva e à fixação com placa reforçadora. O acompanhamento com consultas periódicas é uma parte essencial do tratamento, considerando a agressividade local e a alta taxa de recorrência desse tipo de lesão.


References


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