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Enucleation of large magnitude meroblastic fibroma: Case report

Manoel Olavo Valentim Fernandes de Castilho[1], Karen Tamires Viau[2], Tulio Avelar Ferreira[3], Ana Luísa Bacile Katsui[4], Emilly Anne Teixeira Pereira[5], Lucas Fernando da Silva Moraes[6].

ABSTRACT

Although ameloblastic fibroma is a benign neoplasm, it has considerable rates of recurrence and malignant transformation into an ameloblastic fibrosarcoma. In this context, we seek to provide important information about the clinical presentation, diagnosis, and treatment of this rare and challenging tumor. To this end, we present a case report of a 24-year-old female patient with asymptomatic volume increase in the left mandibular region together with a panoramic X-ray showing a multilocular radiolucent area involving the ramus region, angle and mandibular body associated with element 47 included. Initial clinical management was complementary exams accompanied by incisional biopsy under LA with anatomopathological results of ameloblastic fibroma, the treatment of choice was enucleation + peripheral osteotomy of the lesion under GA. She is currently under a one-year follow-up with no evidence of recurrence and healthy bone neoformation at the site. Ameloblastic fibroma is a rare benign odontogenic lesion with a high recurrence rate and potential for malignant transformation. The treatment of choice is enucleation and curettage, and resection with a safety margin may be necessary in cases of larger lesions and recurrences. Long-term follow-up is essential for monitoring possible relapses. Therefore, treatment should be individualized, taking into account factors such as age, location, recurrences, and postoperative morbidity.

Keywords: Mandible, Pathology, Oral surgery, Neoplasms.

INTRODUCTION

Ameloblastic fibroma is a rare, mixed benign odontogenic neoplasm composed of connective and epithelial tissue, originating from the enamel organ. (MELO, 2015 and SLOOTWEG 2006) Although it is a benign tumor, its high recurrence rate and potential for malignant transformation into an ameloblastic fibrosarcoma make it important to correctly identify and treat it. The mean age of presentation is 10 to 20 years, with a slight predominance in males. The literature indicates that the most common region of occurrence is the mandible, followed by the maxilla, with preference for the posterior region. Diagnosis is made through clinical, radiographic, histopathological, and immunohistochemical examinations (CHRCANOVIC, 2017). The most common treatment is enucleation and curettage. Although surgery is successful in many cases, recurrence is still a significant problem (Chen, 2007 and Slootweg, 2006).

[1] Mandaqui Hospital Complex - SP
[2] Guairacá University Center – PR
[3] University of Franca - SP
[4] City University of São Paulo – SP
[5] University Center of the United Metropolitan Colleges – SP
[6] University Center of the Associated Colleges of Education – SP

OBJECTIVE

In the previous context, we present a case report of a 24-year-old woman who underwent excision of ameloblastic fibroma through enucleation and curettage treatment. Thus, we seek to provide important information about the clinical presentation, diagnosis, and treatment of this rare and challenging tumor.

CASE REPORT

A 24-year-old female patient sought care at the Oral and Maxillofacial Surgery and Traumatology Service of the Conjunto Hospitalar do Mandaqui de São Paulo, presenting with a slight increase in volume, asymptomatic in the right mandible (Figure 1A) together with a radiographic finding identified in a panoramic X-ray examination after a consultation with a private dentist approximately 30 days ago. She denied difficulty chewing, paresthesia or any local discomfort. The patient was systemically well, reporting no comorbidity, addiction or use of medications.

Clinical examination revealed a slight facial asymmetry due to increased volume in the region of the right mandibular angle, with the absence of associated lymphadenomegaly or other local phlogistic signs. Intraoral, the patient was absent from element 47, had a slight bulging of the buccal and lingual alveolar ridge region with normal mucous membranes in normal appearance and color, absence of purulent output or active intraoral bleeding, mild mobility of element 46, and good oral hygiene (Figure 1B). Palpation showed apparent expansion of the bone corticals of hardened consistency involving the angle region and the right mandibular branch.

Radiographically, an extensive multilocular radiolucent lesion in a "soap bubble" pattern was observed, affecting the ramus region, angle and mandibular body, with bone fenestrations and tooth 47 associated with the lesion, located in the mandibular body below the apex of element 46, which presented significant bone resorption around its distal root (Figure 2).

Computed tomography revealed a hypodense, multilocular image with well-defined borders, involving the entire coronoid process, branch, angle, and mandibular body up to close to the apex of element 45, with bulging of the buccal and lingual corticals, with small areas of bone fenestration measuring approximately 88x51x42 mm (Figure 3).

First, an incisional biopsy was performed under local anesthesia in order to obtain an accurate diagnosis and determine the treatment plan. The specimen submitted to anatomopathological examination showed the presence of an epithelial component formed by anastomodating cords or trabeculae, mesenchymal components formed by stellar or triangular cells. The epithelial cells are similar to ameloblasts, as well as to the stellate reticulum of the enamel organ, and the histopathological findings are suggestive of ameloblastic fibroma (Figure 4).

The case was discussed as a team with the patient, and conservative surgical treatment was chosen, involving total enucleation of the associated lesion and peripheral osteotomy due to the patient's early age and the large size of the lesion. This therapeutic option was exhaustively discussed with the patient, exposing clearly and precisely the possibilities of recurrence, as well as the risks and benefits of a more conservative treatment.

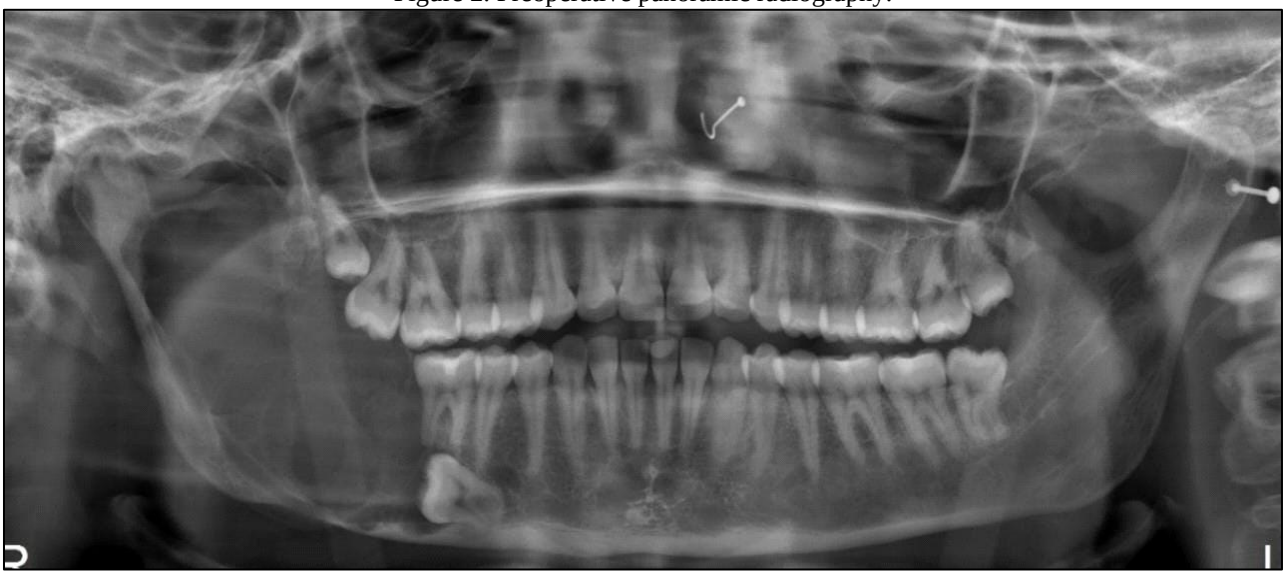
The surgery was performed under general anesthesia with intraoral access through an extended incision from the retromolar region to tooth 41. After total exposure of the lesion (Figure 5A), extraction of element 46 was performed, which showed significant periradicular bone resorption and distal root resorption; extraction of element 47 included and associated with the lesion and total enucleation of the lesion along its entire length (Figure 5B), with peripheral osteotomy (Figure 5C and D). The osteotomy was performed with a spherical diamond drill throughout the residual bone cortical, with care to preserve the integrity of the inferior alveolar nerve, followed by a careful review of the region of bone fenestrations in order to remove possible lesional remnants in soft tissue, followed by the suture through planes in a judicious manner. The removed lesion was about 9 cm long, with a rubber-like consistency (Figure 5E).

In postoperative the patient had mild edema compatible with the surgical procedure, and the patient was discharged from the hospital the day after surgery and was instructed to maintain a liquid diet for about 28 days. Currently, the patient is under 1-year postoperative follow-up, with no signs of lesion recurrence or postoperative complications. It has a good appearance of healing and bone remodeling, as well as preservation of the dental condition and the alveolar ridge.

Figure 1. (A) Extraoral aspect with mild facial asymmetry (B) Intraoral clinical aspect of the lesion.

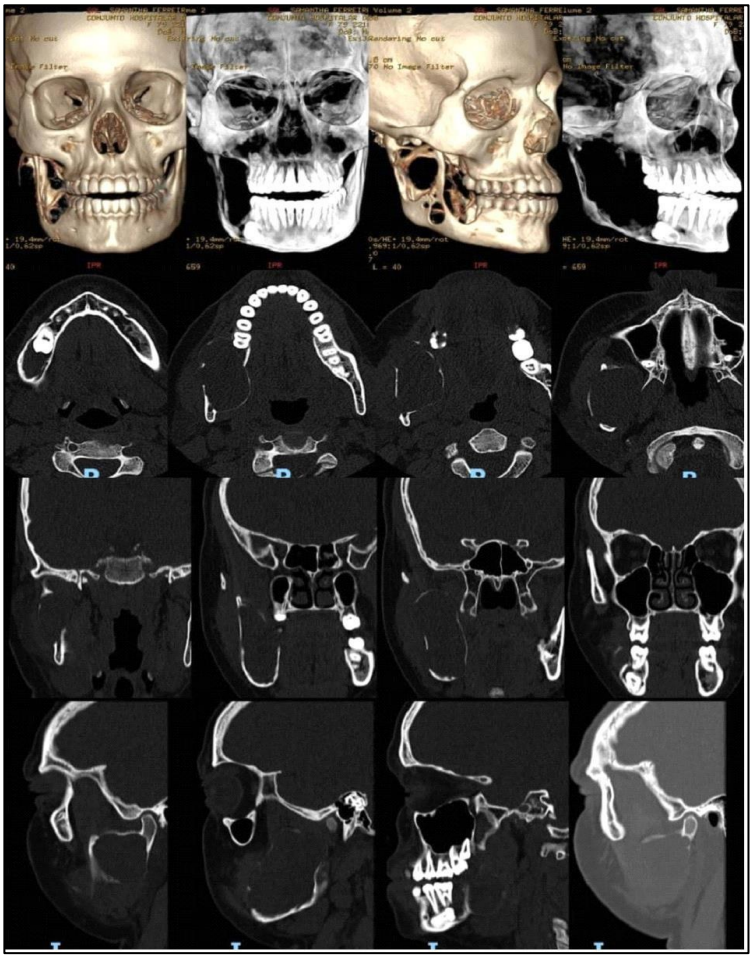
Source: Division of Oral and Maxillofacial Surgery and Traumatology, Conjunto Hospitalar do Mandaqui.

Figure 2. Preoperative panoramic radiography.



Source: Division of Oral and Maxillofacial Surgery and Traumatology, Conjunto Hospitalar do Mandaqui.

Figure 3. Extensive multilocular radiolucent lesion in a "soap bubble" pattern affecting the ramus region, angle, and mandibular body associated with tooth 47.



Source: Division of Oral and Maxillofacial Surgery and Traumatology, Conjunto Hospitalar do Mandaqui.

Figure 4. Result of anatomopathological examination.

Macroscopia:

Bloco e lâminas (2 HE) para consulta.

Microscopia:

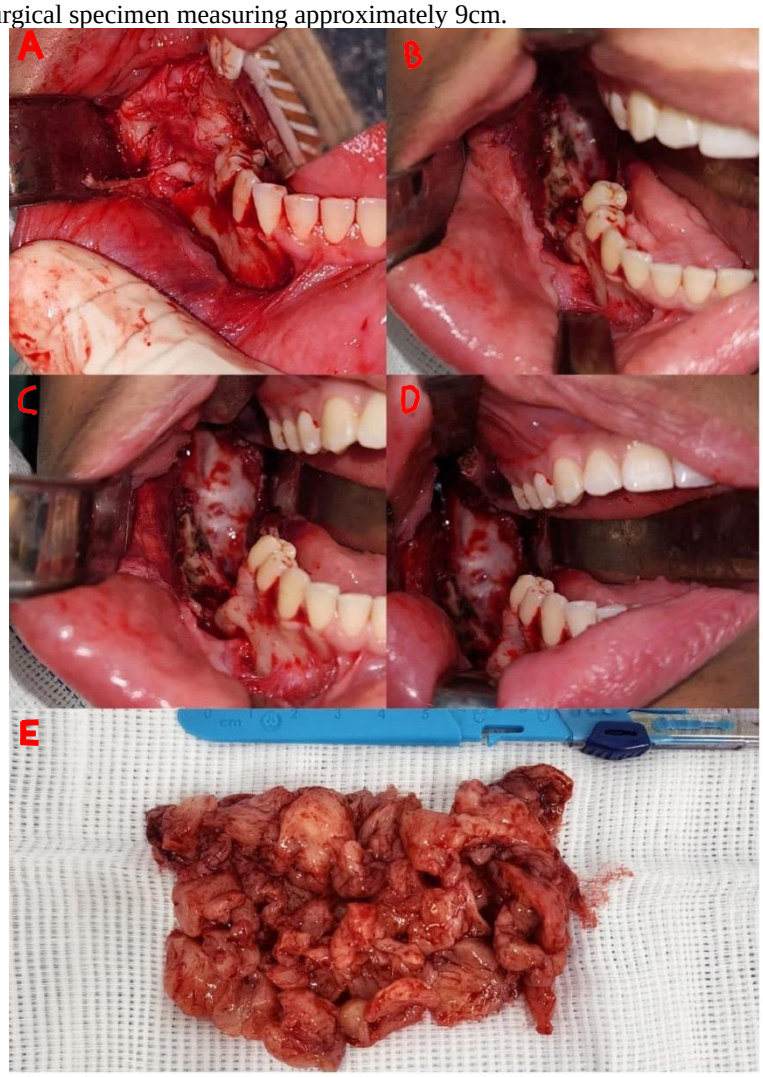
Os cortes mostram neoplasia odontogênica mista caracterizada pela presença de componente epitelial formado por cordões ou trabéculas anastomosantes, e componente mesenquimal formado por células estrelárias ou triangulares. As células epiteliais são colunares semelhantes a ameloblastos e geralmente dispõem-se em cordões de duas fileiras. Por vezes há também a formação de ilhotas epiteliais com as células centrais semelhantes a retículo estrelado do órgão do esmalte. Em raras ocasiões há metaplasia escamosa nas células centrais. O componente mesenquimal é composto de células triangulares ou estrelárias em estroma mixomatoso. Há áreas de poucas células e outras mais celulares.

Diagnóstico:

Mandíbula: Fibroma ameloblástico

Source: Division of Oral and Maxillofacial Surgery and Traumatology, Conjunto Hospitalar do Mandaqui.

Figure 5. (A) Intraoral access and total exposure of the lesion. (B) Immediate clinical appearance after excision of the lesion and extraction of associated included element and element 46. (C) Clinical appearance after curettage. (D) Clinical appearance after peripheral osteotomy (E) Surgical specimen measuring approximately 9cm.

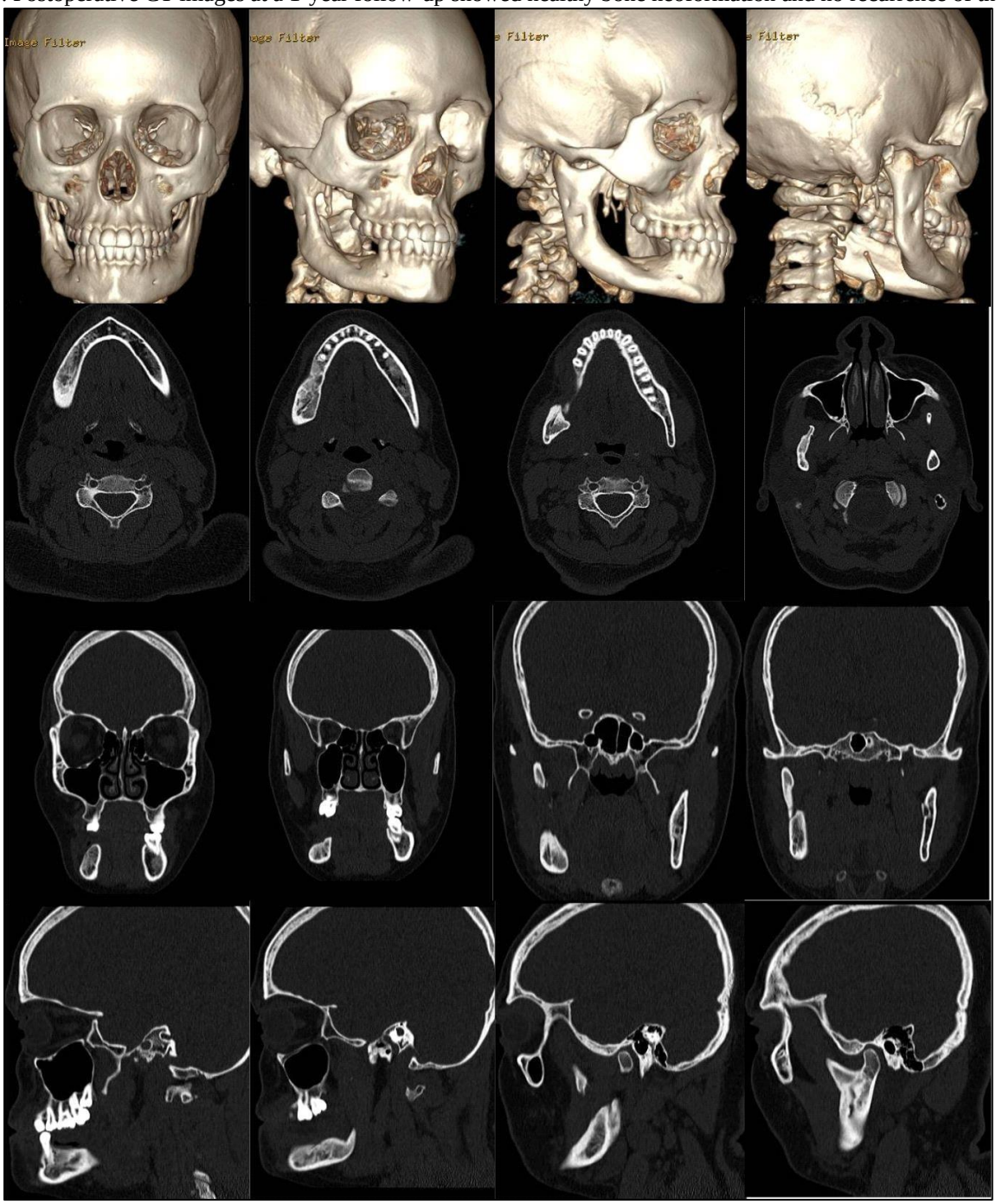


Source: Division of Oral and Maxillofacial Surgery and Traumatology, Conjunto Hospitalar do Mandaqui.

Figure 6. (A) Extra oral appearance after 1 year, with no evidence of facial asymmetries. (B) Intraoral appearance after 1 year, maintaining all mandibular circumference, healthy mucous membranes, stable and maintained occlusion.

Source: Division of Oral and Maxillofacial Surgery and Traumatology, Conjunto Hospitalar do Mandaqui.

Figure 5. Postoperative CT images at a 1-year follow-up showed healthy bone neoformation and no recurrence of the lesion.



Source: Division of Oral and Maxillofacial Surgery and Traumatology, Conjunto Hospitalar do Mandaqui.

DISCUSSION

Ameloblastic fibroma is a rare benign odontogenic neoplasm that accounts for about 1.5% to 4.5% of odontogenic tumors. Its etiology is unknown, but it may be related to genetic mutations, traumatic injuries, and dental abnormalities. Despite recent progress in understanding the pathogenesis and management of AF, there are still gaps in knowledge about the disease, such as its origin and underlying pathology (MELO, 2015; TOZOGLU, 2016).

Although it is a benign lesion, AF exhibits a potentially aggressive biological behavior, with reported cases of local invasion and recurrence. The AF recurrence rate can reach 18% depending on the extent of the lesion, invasion or not of soft tissues after bone fenestration, and the surgical technique used. In addition, ameloblastic fibroma has a potential for malignant transformation into an ameloblastic fibrosarcoma with recurrence rates close to 45%, making its diagnosis and treatment important (KULKARNI, 2013).

Clinically, it presents as an asymptomatic, well-defined, slow-growing lesion, it is usually discovered in routine examinations, as in the case described, but in cases of greater extension, dental mobility, paresthesia and root resorption may occur, also observed in the case described. It is a true mixed odontogenic tumor, composed of connective tissue and epithelials, originating from the enamel organ, with or without the formation of hard dental tissue (CHEN, 2007; TOZOGLU 2016).

AF presents radiographically as a unilocular or multilocular radiolucent lesion when it reaches greater proportions, occurring predominantly in the posterior region of the mandible, often associated with an included tooth, with a predilection for young patients between the first and second decade of life and a slight predilection for males (MELO, 2015; CHRCANOVIC, 2017; CHRCANOVIC, 2017). Cortical expansion is common, being observed in about 80% of cases (CHRCANOVIC, 2017). In the case reported, all characteristics converge to the diagnosis, the only difference being that the patient is female.

It is considered the most common mixed odontogenic tumor in young patients, with the differential diagnosis being odontogenic myxoma, solid ameloblastoma, dentiger cyst, keratocyst, and odontoma (MELO, 2015), hypotheses that were considered prior to the initial incisional biopsy. Therefore, the anatomopathological examination is essential for the diagnosis of the lesion and treatment plan. There is no consensus for the treatment of AF, the most indicated is enucleation and curettage, a technique that was adopted for this case; However, marginal or segmental resection can be chosen depending on each case, taking into account the patient's size, stage, age, and recurrences, since its malignant variant has been more associated with patients with larger lesions, with large invasion of soft tissues, recurrences, and depending on the patient's age. Therefore, treatment should be individualized for each case. The literature suggests that for larger lesions and/or older patients, the treatment of choice is resection with a safety margin (CHRCANOVIC, 2015).

In the case presented, despite the size of the lesion and soft tissue involvement, the treatment was widely debated between the team and the patient, who, due to their age, good oral hygiene conditions, and difficulty in finding segmental resection associated with a microvascularized graft (which would be necessary for the local case, if this technique was chosen) and the patient's correct capacity/collaboration, she agreed to perform a more conservative treatment for the case, generating less postoperative morbidity for the patient.

Postoperative follow-up is essential, since recurrence can occur late, even years after surgery. Most cases of relapse occur within the first two years after treatment, however. Therefore, regular monitoring with imaging and clinical tests is recommended to detect recurrence (MELO, 2015; CHRCANOVIC, 2017). In this case, the clinical follow-up was done weekly until completing approximately 42 days postoperatively, being changed to monthly until completing 6 months, and a new return after another 6 months, thus completing 01 year of follow-up.

Currently, the patient is under a one-year follow-up, with no signs of recurrence, with a good and healthy local bone neoformation, absence of paresthesia and good bone remodeling, reducing the asymmetry caused by the cortical expansion of the lesion. She is properly instructed that she should follow an annual radiographic follow-up for the case.

It is important to note that, despite the potential for malignant transformation, ameloblastic fibrosarcoma is a rare variant of ameloblastic fibroma. Malignant transformation occurs in less than 1% of cases, and is characterized by the presence of high-grade areas of cellular atypia, invasion of adjacent tissues, and metastasis to regional lymph nodes (MELO, 2015; CHRCANOVIC, 2017; KULKARNI, 2013).

In summary, ameloblastic fibroma is a rare benign odontogenic lesion that has a high recurrence rate and potential for malignant transformation. The treatment of choice is enucleation and curettage, and resection with a safety margin may be necessary in cases of larger lesions and recurrences. In the case presented here, the patient opted for a more conservative surgical treatment and was followed up for one year without recurrences and with normal bone formation to date. Long-term follow-up is essential for monitoring possible relapses. The successful treatment of ameloblastic fibroma depends on an accurate diagnosis, careful planning, and appropriate surgical approach, followed by regular postoperative follow-up. With these measures, we can provide patients with a better quality of life and reduce the risks of complications and relapses.

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