



ISSN: 2230-9926

Available online at <http://www.journalijdr.com>

# IJDR

International Journal of Development Research

Vol. 10, Issue, 05, pp. 36182-36184, May, 2020

<https://doi.org/10.37118/ijdr.18958.05.2020>



RESEARCH ARTICLE

OPEN ACCESS

## CONSERVATIVE APPROACH TOUNICYSTIC AMELOBLASTOMA IN A PEDIATRIC PATIENT - CASE REPORT AND CLINICAL CHALLENGES

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### ARTICLE INFO

#### Article History:

Received 20<sup>th</sup> February, 2020  
Received in revised form  
18<sup>th</sup> March, 2020  
Accepted 11<sup>th</sup> April, 2020  
Published online 30<sup>th</sup> May, 2020

#### Key Words:

Ameloblastoma, Odontogenic Tumors,  
Marsupialization, Curettage,  
Conservative Treatment.

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### ABSTRACT

Ameloblastomas are benign odontogenic neoplasms characterized by the proliferation of epithelial cells that have characteristics similar to those of the enamel organ. They are tumors that affect the gnathic bones that show faster growth and greater potential for recurrences than other odontogenic tumors. Two basic forms of distinct behavior are found: conventional (solid / multicystic) and unicystic. Unicystic ameloblastomas are more rare and less prone to recurrence than conventional ameloblastomas. This last characteristic allows the efficient use of more conservative surgical therapeutic approaches. This article aims to report the case of a patient with unicystic ameloblastoma in the jaw treated by marsupialization and later curettage, under general anesthesia, and with follow-up for three years without recurrence of the disease.

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Citation: *Izabella Sol, Cristóvão Marcondes de Castro Rodrigues, Marcella Sol, Jéssica Brenda Rodrigues de Medeiros, Cláudia Jordão Silva4; Marcelo Caetano Parreira Silva.* "Conservative approach tounicystic ameloblastoma in a pediatric patient - case report and clinical challenges", *International Journal of Development Research*, 10, (05), 36182-36184.

## INTRODUCTION

Ameloblastomas are benign intraosseous lesions that affect the maxillary bones, interfering with function and aesthetics (Bisinelli et al., 2007), and can be classified as solid or multicystic, unicystic, peripheral or extrasosseous and desmoplastic (Antonoglou, 2015). Ameloblastoma is classified as the second most common odontogenic tumor, with no predilection for sex and ethnicity. Characterized by a slow and asymptomatic growth pattern, they cause bone cortical expansion, which can lead to tooth movement and root

resorption, being more observed in the posterior region of the mandible (branch and mandibular angle) (Bisinelli et al., 2007; Hendra et al., 2019). They are locally invasive and have a high recurrence rate if not removed properly (Hendra et al., 2019). Its treatment and prognosis depend on multiple factors, such as its form of clinical presentation, affected area, time of evolution and type of tumor (Bisinelli et al., 2007; Hendra et al., 2019). The treatment of choice for multicystic tumors generally requires a more aggressive approach due to its higher recurrence rate<sup>2</sup>, while the unicystic and peripheral forms, due to the lower rate of recurrence, respond well to less invasive

conservative treatment (Bisinelli *et al.*, 2007). The management of tumors in pediatric patients remains controversial in the literature, as it affects the growth dynamics of the craniofacial skeleton, dentition and soft tissues (Bansal, 2015). The conservative treatment of unicystic ameloblastoma in a 13-year-old patient with a follow-up time of 36 months is reported below.

## CASE REPORT

Male patient, thirteen years old, melanoderm, attended the Buccomaxillofacial Surgery and Traumatology outpatient clinic at the Federal University of Uberlândia in september 2016, complaining of a volumetric increase in the oral region of the right mandible for six months, without previous allergies and comorbidities. The clinical examination showed facial asymmetry in the mandibular region without condylar or mental deviation, asymptomatic, with aesthetic complaint. The intraoral examination showed a rigid painless swelling in the region of the mandible, involving the region of the lower right second premolar to the right mandibular branch, with expansion of both cortices. There were no signs of inflammation and lymph node enlargement. Panoramic radiography showed a well-delimited unilocular radiolucent image involving the right mandibular branch up to the distal face of element 45, with an investigation germ of element 48 and element 47 included inside the lesion (Figure-1).



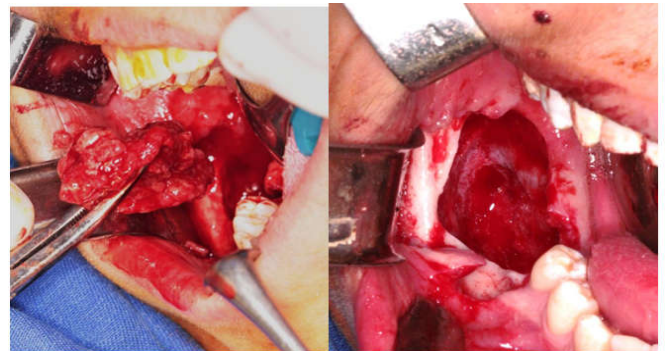
**Figure 1. Panoramic radiography showing unilocular radiolucent lesion in the body region, angle and right mandibular branch up to the distal face of the right lower second premolar, involving the dental germ of the right third and second lower molars inside the lesion, with cortical expansion**



**Figure 2. Panoramic radiograph after 8 months of marsupialization, we observed a significant decrease in the lesion**

The diagnostic hypotheses were odontogenic keratocyst, dentigerous cyst and unicystic ameloblastoma. Incisional biopsy and intraoral marsupialization was performed under local anesthesia in september/2016. The aspiration puncture prior to biopsy showed slightly mucous citrus liquid inside the

lesion. The result of the histopathological examination proved to be compatible with unicystic ameloblastoma. The patient was instructed on local hygiene and periodic controls were performed until visualization of the significant volumetric reduction of the lesion (Figure-2). The enucleation of the lesion associated with peripheral osteotomy was then planned after 08 months of marsupialization (Figure-3). Under general anesthesia and nasotracheal intubation, the total excision of the pathology was performed, with the sample collected for histopathological analysis. His report showed unicystic ameloblastoma with luminal and mural proliferation. During the postoperative period, the patient evolved with small suture dehiscence, without pain complaints and without inflammatory signs. Periodic returns were kept for control. Currently, the patient is in a 36-month term. The panoramic control radiograph shows satisfactory bone neoformation and the absence of clinical and radiographic signs of recurrence in an enucleated location (Figure-4). The patient awaits older age for rehabilitation with osseointegrated implants.



**Figure 3. Trans-operative aspect of enucleation of unicystic ameloblastoma (a) and surgical store after peripheral osteotomy (b)**



**Figure 4. 36-month postoperative panoramic radiograph showing new bone formation with no evidence of recurrence**

## DISCUSSION

Unicystic ameloblastoma is more observed in patients aged 1-2 decades, characterized by slow, asymptomatic and locally aggressive growth, with the posterior region of the mandible being the main site of involvement (Bisinelli *et al.*, 2007). Radiographically the edges of the lesion are well defined, with no radiographic evidence of infiltration in adjacent bone<sup>3</sup>. The cortical bone is frequently expanded and with areas of perforation but without clinical and histological signs of neoplastic tissue in adjacent soft tissues (Isacsson, 1986). All of these common clinical and radiographic characteristics of unicystic ameloblastoma are present in our case report. Histopathological variants, classified according to the degree of extension of the ameloblastic epithelium into luminal

(tumor confined to the cyst lining epithelium associated with a fibrous cystic lining membrane), intraluminal (projections of the neoplastic epithelium in the lumen of the cystic cavity) and mural (follicular or plexiform ameloblastoma infiltrate in the fibrous wall) are related to the biological behavior of the tumor (Bisinelli *et al.*, 2007; Garcia *et al.*, 2016). Despite being classified as a benign tumor, due to its characteristics of local invasion and recurrence rate, the management of patients with unicystic ameloblastoma must be carefully decided. Much has been discussed about the most appropriate treatment for pediatric and growing-age patients, and this topic is still controversial. The biological behavior of unicystic ameloblastoma tends to be less invasive when compared to the solid variant (Antonoglou *et al.*, 2015), with conservative surgical treatment being recommended by some authors (Bisinelli *et al.*, 2007; Antonoglou *et al.*, 2015; Bansal, 2015; Nakamura, 2009). Thus, the continued growth of the facial bones is allowed, the facial tissues remain preserved and the aesthetic and functional sequelae are limited (Laborde, 2017). Several surgical treatments are reported in the literature, including more aggressive modalities such as marginal or segmental resection (Laborde, 2017) to more conservative treatments such as enucleation (Garcia, 2016), marsupialization<sup>7</sup>, application of Carnoy's solution (Haq, 2016), or a combination of these techniques (Nakamura *et al.*, 2002; Laborde *et al.*, 2017; Haq, 2016). In view of the large number of treatment options, the choice of the modality that will best suit the case must take into account some factors such as the histological type, clinical and radiographic characteristics, location of the lesion and its relationship with adjacent structures, age, impact psychological treatment of the patient and cooperativeness (Bisinelli *et al.*, 2007; Garcia, 2016), thus aiming at the most effective and least morbid choice for each patient.

In the present study, the pathological anatomy of the lesion showed the presence of two histological types, luminal and mural, associated with more aggressive behavior of the lesion. However, during the analysis of the patient's age (thirteen years) and the good response of the lesion to conservative treatment, where a significant decrease in its total size was observed within six months of marsupialization monitoring, associated with the patient's good cooperation during the period, we opted for the choice of conservative treatment aiming at lower morbidity of the case, without great aesthetic and functional losses. The marsupialization technique followed by enucleation associated with peripheral osteotomy of the lesion in a pediatric patient with no recurrence in 36 months of follow-up reported in this study was also observed in similar cases of patients of the same age group reported by Bisinelli (2007) and Garcia (2016), who demonstrated the absence of recurrence of the lesion in a follow-up period of 10 years and 12 months, respectively. The goal of marsupialization is to reduce the size of the lesion so that less extensive surgery can be performed later, as corroborated by this report. The internal decompression promoted by marsupialization promotes bone remodeling<sup>10</sup>, where it is possible to see in the accompanying radiographic exams the reduction of the lesion concomitant to peripheral osteogenesis. However, this conservative modality is more related to recurrences, with rates from 36% to 18%<sup>3,8</sup> being generally observed 15 months after the initial treatment<sup>1</sup>. Factors such as treatment performed on the initial injury and quality of the surgical technique, extent and location of involvement, influence the prognosis of the case and are directly related to the rate of recurrence (Bisinelli *et al.*, 2007;

Nakamura, 2002). Thus, postoperative monitoring is essential for early diagnosis of this pathology, as well as an operator with experience to perform the technique leading to immediate and late postoperative surgical success.

### Final considerations

The available evidence suggests that conservative treatment has a favorable outcome for the treatment of unicystic lesions in pediatric patients when possible. As observed in this case report, marsupialization followed by enucleation had a positive effect on the patient's aesthetics and masticatory function, in addition to not showing recurrence in a 36-month follow-up period. Prospective, multicenter and controlled studies, associated with long-term follow-up periods, are necessary to define the effectiveness of the treatments proposed in the literature.

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