Research, Society and Development, v. 9, n. 7, e960975194, 2020
(CC BY 4.0) | ISSN 2525-3409 | DOI: http://dx.doi.org/10.33448/rsd-v9i7.5194

Ameloblastoma de células granulares em maxila: atualização das aparências clínica, radiográfica e histológica incomuns

Granular cell ameloblastoma in maxilla: update of unusual clinical, radiographic and histological appearance

Ameloblastoma de células granulares en maxila: actualización de las apariencias clínica, radiográfica e histológica inusuales

Recebido: 02/06/2020 | Revisado: 04/06/2020 | Aceito: 06/06/2020 | Publicado: 16/06/2020

Bruno Coelho Mendes
ORCID: https://orcid.org/0000-0001-7896-8909
Sao Paulo State University, Brazil
E-mail: brunocoelho.mnds@gmail.com
João Paulo Bonardi
ORCID: https://orcid.org/0000-0002-1687-1340
Sao Paulo State University, Brazil
E-mail: joao_bonardi@hotmail.com
Leonardo de Freitas Silva
ORCID: https://orcid.org/0000-0002-6345-1269
E-mail: leonardofreitas86@gmail.com
Sao Paulo State University, Brazil
Marcelo Macedo Crivelini
ORCID: https://orcid.org/0000-0002-0174-439X
Sao Paulo State University, Brazil
E-mail: mmcrivel@foa.unesp.br
Ana Paula Farnezi Bassi
ORCID: https://orcid.org/0000-0002-0031-4953
Sao Paulo State University, Brazil
E-mail: apfbassi@foa.unesp.br

Resumo

Objetivo: descrever o tratamento de sucesso de um ameloblastoma de células granulares em região posterior de maxila, de 5mm de tamanho, descoberto em exame de rotina durante a avaliação de uma comunicação buco-sinusal. **Pacientes e métodos**: o relato de caso é apresentado baseado em uma revisão simples da literatura dos últimos 10 anos com os

descritores (Ameloblastoma; Maxilla; Surgery; Diagnosis Oral; Neoplasms) no Pubmed. O tratamento composto pela fistulectomia e fechamento da comunicação buco-sinusal com corpo adiposo da bochecha foi associado a enucleação e curetagem da lesão adjacente sob anestesia local. **Resultados:** evolução satisfatória foi observada em 1 ano de pós-operatório, com bom aspecto cicatricial, sem sinais de complicações buco-sinusais ou recidiva da lesão. **Conclusão**: para lesões menores, propostas de tratamento mais conservadoras como a enucleação representam uma alternativa viável e, além disso, a utilização de técnicas adjuvantes como a curetagem pode oferecer benefícios quando o diagnóstico diferencial inclui lesões mais agressivas ou com maior taxa de recidiva.

Palavras-chave: Ameloblastoma; Maxila; Cirurgia; Diagnóstico bucal; Neoplasma

Abstract

Objective: to describe the successful treatment of a 5mm-sized granular cell ameloblastoma in the posterior region of the maxilla, discovered in a routine examination during the evaluation of an oroantral communication. **Patients and methods**: the case repot is presented based on a simple literature review of the last 10 years with the descriptors (Ameloblastoma; Maxilla; Surgery; Oral Diagnosis; Neoplasms) in Pubmed. Treatment consisting of fistulectomy and closure of the oroantral communication with the buccal fat pad and vestibular flap was associated with enucleation and curettage of the adjacent lesion under local anesthesia. **Results:** a satisfactory evolution was observed in the 1 year postoperative, with a good healing aspect and no signs of oroantral complications or recurrence of the lesion. **Conclusion:** for minor lesions, more conservative treatment proposals such as enucleation represent a viable alternative and, in addition, the use of adjuvant techniques such as curettage can offer benefits when the differential diagnosis includes more aggressive lesions or with a higher rate of recurrence.

Keywords: Ameloblastoma; Maxilla; Surgery; Diagnosis Oral; Neoplasms

Resumen

Objetivo: describir el tratamiento exitoso de un ameloblastoma de células granulares en la región maxilar posterior, de 5mm de tamaño, descubierto en un examen de rutina durante la evaluación de la comunicación oroantral. **Pacientes y métodos**: el informe del caso se presenta en base a una revisión simple de la literatura de los últimos 10 años con los descriptores (Ameloblastoma; Maxilar; Cirugía; Diagnóstico oral; Neoplasias) en Pubmed. El tratamiento consistente en la fistulectomía y el cierre de la comunicación oroantral con el

cuerpo adiposo de la mejilla se asoció a la enucleación de la lesión adyacente con legrado de la cavidad bajo anestesia local. **Resultados**: se observó una evolución satisfactoria 1 año después de la operación, con un buen aspecto de cicatriz, sin signos de complicaciones oroantral o recurrencia de la lesión. **Conclusión:** para lesiones menores, las propuestas de tratamiento más conservadoras, como la enucleación, representan una alternativa viable y, además, el uso de técnicas adyuvantes como el legrado puede ofrecer beneficios cuando el diagnóstico diferencial incluye lesiones más agresivas o con una mayor tasa de recurrencia.

Palabras clave: Ameloblastoma; Maxilar; Cirurgía; Diagnóstico bucal; Neoplasias

1. Introduction

Ameloblastoma is an uncommon but clinically significant odontogenic neoplasm, because despite slow growth and benignity in most cases, it is locally invasive and aggressive (Siar, Lau & Ng, 2012). Clinical and histomorphologic patterns are variable and identified in three types: conventional, unicystic ameloblastoma and the peripheral (Wright & Vered, 2017). In general, the age group that can be affected varies from 07 to 85 years, with a higher incidence in the third decade of life and development more frequently in the mandible (Siar et al., 2012; Zwahlen & Grätz, 2002).

Granular cell ameloblastoma is a rare histological variant of the conventional one. Characterized by presenting cells with abundant cytoplasm in eosinophilic granules, similar to a starred reticulum, it presents the highest rates of relapse. His pathogenesis has been attributed to the dysfunctional state of neoplastic cells, either by degenerative changes or even aging in long-term lesions (Chae, Smoll, Hunter-Smith & Rozen, 2015; Gunawardhana, Jayasooriya, & Tilakaratne, 2014; Siar et al., 2012; Zwahlen & Grätz, 2002).

The diagnosis is obtained through clinical and imaging characteristics and confirmed by histopathological exam of the specimen collected in biopsy. The treatment modalities may vary from enucleation / curettage, segmental resection, enucleation associated with peripheral osteotomy, curettage associated with cryotherapy (Curi, Dib, & Pinto, 1997). Patient follow-up should be rigorous in order to identify possible relapses, which may require more aggressive treatment (Ooi, Feng, Tan, & Ong, 2014).

The aim of this article is to describe the successful treatment of a 5mm-sized granular cell ameloblastoma in the posterior region of the maxilla, discovered in a routine examination during the evaluation of an oroantral communication.

2. Patients and Methods

This case repot is presented based on a simple literature review with the descriptors (Ameloblastoma; Maxilla; Surgery; Oral Diagnosis; Neoplasms) in Pubmed, similar to (Dave, Arora, Shetty & Saluja, 2015).

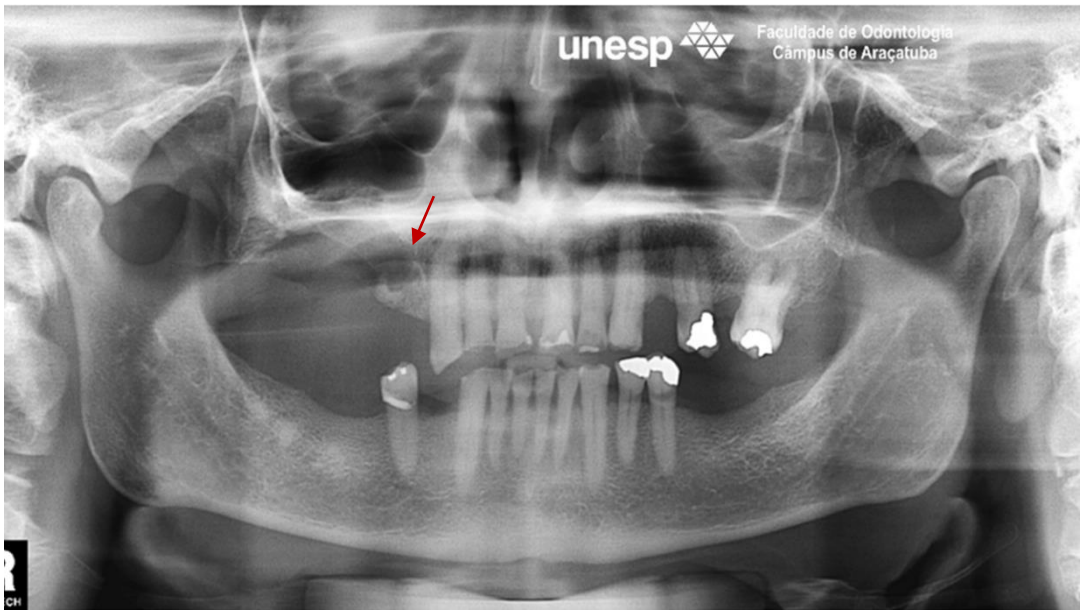
Case Report

A 56-year-old male patient, referred to Aracatuba School of Dentistry, complaining of oroantral communication occurred after 15 tooth extraction, which had undergone three unsuccessful closing attempts. At oroscopy, oroantral fistula at the region of the 15 tooth were noted (Figure 01). He was on the sixth day of antibiotic therapy with amoxicillin/clavulanic acid 875mg/125mg and and maxillary sinus irrigation with 60ml of saline three times a day. The panoramic radiography showed a 5mm unilocular circumscribed radiolucent image with defined borders and regular contours adjacent to the fistula (Figure 02). The diagnostic hypotheses established for the lesion were respectivelly residual cyst, keratocyst and ameloblastoma. After signing informed consent, the patient was treated through fistulectomy, closure of the communication with the pedicled buccal fat pad and vestibular flap, in addition to the enucleation and curettage of the associated lesion (Figure 03). The specimen was submitted to histopathological examination whose diagnosis revealed granular cell ameloblastoma (Figure 04). Patient was followed up clinically and radiographically and the satisfactory local repair, after 01 year-postoperative, shows the efficacy of the communication closure technique and enucleation associated with curettage (Figure 05).

Figure 01: Clinical aspect of oroantral communication at region of the 15 tooth.

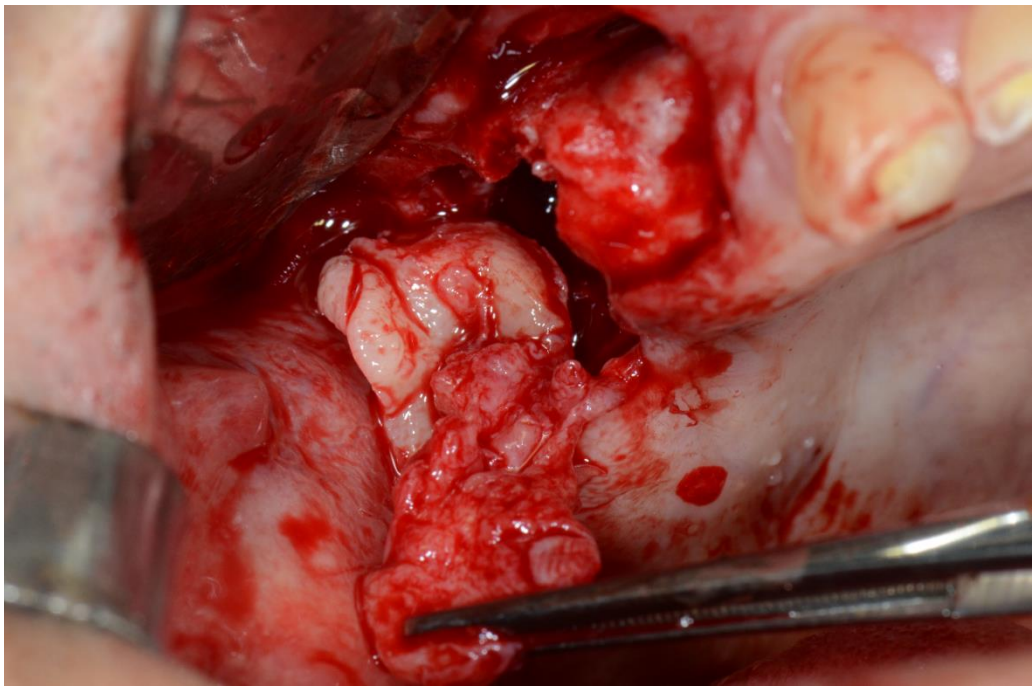
Source: authors

Figure 02: Panoramic radiography shows a unilocular circumscribed radiolucent image with defined borders and regular contours adjacent to the fistula, with aproximatelly 5mm.



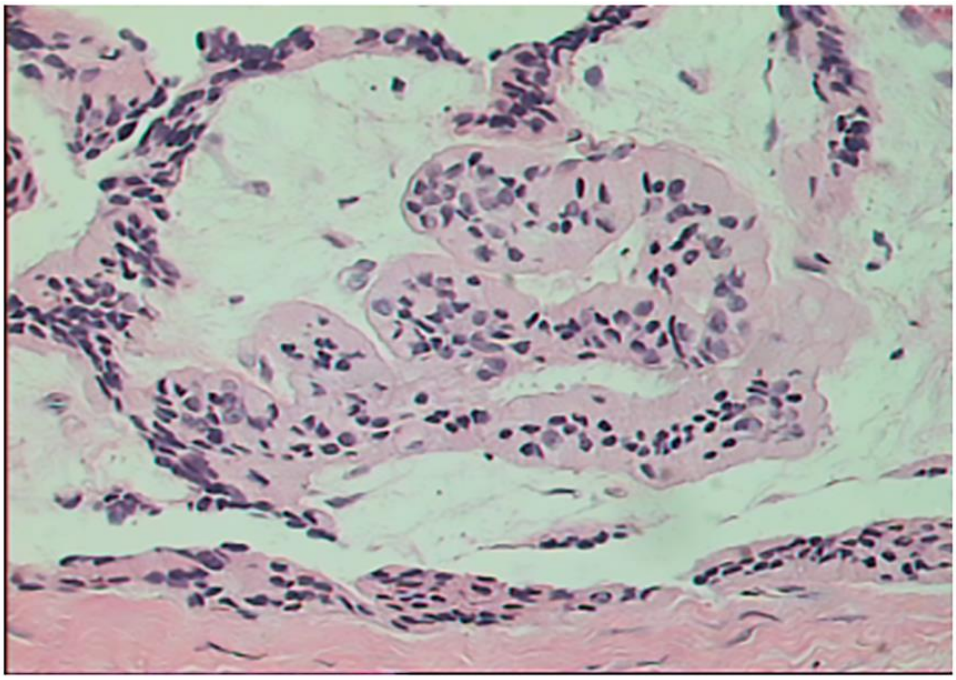
Source: authors

Figure 03: Lesion clinical aspect during its enucleation and closure of the oroantral communication.



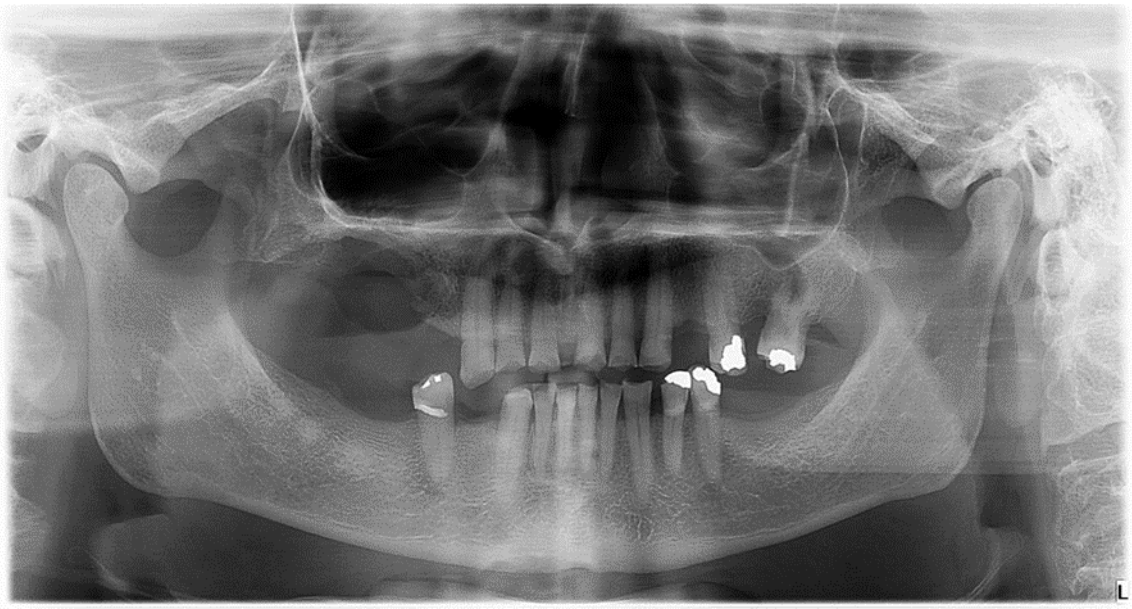
Source: authors

Figure 04: Microscopic features of ameloblastoma showing epithelial strands of hyperchromatic cells with abundant eosinophilic granular cytoplasm. HEx400



Source: authors

Figure 05: One-year postoperative panoramic radiography showing no signs of recurrence at the surgical region.



Source: authors

3. Discussion

Although it occurs more often in the mandible, 91.5% of cases, the occurrence of ameloblastoma in the maxilla should be considered, even without of the most common clinical features such as painless volumetric increase of slow growth, tooth dislocation and paresthesia, associated with classic multilocular radiographic appearance, similar to soap bubbles or honeycombs with dental involvement. Furthermore, early diagnosis can avoid more invasive treatments such as large resections with margins of 1-1.5 cm, which can lead to large bone defects and consequently require further reconstructions. (Bansal et al., 2015; Chae et al.,2015; Curi et al., 1997; Dave et al., 2015; Gunawardhana et al.,2014; Ooi et al., 2014; Siar et al., 2012; Wright & Vered, 2017; Yamunadevi et al., 2014; Zwahlen & Grätz, 2002).

In this case, the lesion detected in a routine examination, asymptomatic, could be completely enucleated, although presenting features that did not individualize it of the oroantral communication. Another important factor was radiographic presentation. Although granular cell ameloblastoma is a rare histological variation of the conventional, which in turn presents a multicystic radiographic pattern, this case presented it as unilocular radiolucency, which is not the pattern frequently observed for this variant of ameloblastoma. Thus, if the differential diagnosis was not composed of an aggressive pathological process, treatment could be carried out through simple enucleation and the possibility of recurrence would be greater. Therefore, it is important that conventional ameloblastomas be included in the differential diagnosis of unilocular lesions (Bansal et al., 2015).

Although the treatment performed was conservative, under hypothesis of residual cyst, it is necessary to point out the importance of curettage performed after enucleation considering the differential diagnosis, even in a remote possibility. Furthermore, the referral of the specimen for histopathological examination was indispensable (Bansal et al., 2015; Chae et al.,2015; Curi et al., 1997; Dave et al., 2015; Gunawardhana et al.,2014; Ooi et al., 2014; Siar et al., 2012; Wright & Vered, 2017; Yamunadevi et al., 2014 Zwahlen & Grätz, 2002). In this scope, it is notable that posterior curettage offers therapeutic advantage compared to isolated enucleation, to cover more aggressive lesion.

More conservative treatments such as enucleation or curettage have higher rates of recurrence, as high as 83.3% when compared to wide-margin resection, and more than 50% of recurrences occur within the first 5 postoperative years. In addition, evidence in the literature indicates greater aggression of the lesion when development occurs in the maxilla, with possible expansions to paranasal sinuses and compression of noble structures. (Dave et al., 2015; Pogrel & Montes, 2009; Siar et al., 2012; Yamunadevi et al., 2014; Zwahlen & Grätz, 2002). Thus, the location, size, diagnosis, treatment performed and possibilities of recurrence, suggest a periodic follow-up of the patient so that any alteration is still detected in the initial phase (Zwahlen & Grätz, 2002).

4. Conclusion

In conclusion, for minor lesions, more conservative treatment proposals such as enucleation represent a viable alternative and, in addition, the use of adjuvant techniques such as curettage can offer benefits when the differential diagnosis includes more aggressive lesions or with a higher rate of recurrence.

References

Bansal, S., Desai, R. S., Shirsat, P., Prasad, P., Karjodkar, F., & Andrade, N. (2015). The occurrence and pattern of ameloblastoma in children and adolescents: An Indian institutional study of 41 years and review of the literature. *International journal of oral and maxillofacial surgery*, 44(6), 725–731. https://doi.org/10.1016/j.ijom.2015.01.002

Chae, M. P., Smoll, N. R., Hunter-Smith, D. J., & Rozen, W. M. (2015). Establishing the natural history and growth rate of ameloblastoma with implications for management: systematic review and meta-analysis. *PloS one*, 10(2), e0117241. https://doi.org/10.1371/journal.pone.0117241

Curi, M. M., Dib, L. L., & Pinto, D. S. (1997). Management of solid ameloblastoma of the jaws with liquid nitrogen spray cryosurgery. *Oral surgery, oral medicine, oral pathology, oral radiology, and endodontics*, 84(4), 339–344. https://doi.org/10.1016/s1079-2104(97)90028-7

Dave, A., Arora, M., Shetty, V. P., & Saluja, P. (2015). Granular cells in ameloblastoma: An enigma in diagnosis. *Indian journal of dentistry*, 6(4), 211–214. https://doi.org/10.4103/0975-962X.165048

Gunawardhana, K. S., Jayasooriya, P. R., & Tilakaratne, W. M. (2014). Diagnostic dilemma of unicystic ameloblastoma: novel parameters to differentiate unicystic ameloblastoma from common odontogenic cysts. *Journal of investigative and clinical dentistry*, 5(3), 220–225. https://doi.org/10.1111/jicd.12071

Ooi, A., Feng, J., Tan, H. K., & Ong, Y. S. (2014). Primary treatment of mandibular ameloblastoma with segmental resection and free fibula reconstruction: achieving satisfactory outcomes with low implant-prosthetic rehabilitation uptake. *Journal of plastic, reconstructive & aesthetic surgery*: JPRAS, 67(4), 498–505. https://doi.org/10.1016/j.bjps.2014.01.005

Pogrel, M. A., & Montes, D. M. (2009). Is there a role for enucleation in the management of ameloblastoma? *International journal of oral and maxillofacial surgery*, *38*(8), 807–812. https://doi.org/10.1016/j.ijom.2009.02.018

Siar, C. H., Lau, S. H., & Ng, K. H. (2012). Ameloblastoma of the jaws: a retrospective analysis of 340 cases in a Malaysian population. *Journal of oral and maxillofacial surgery:* official journal of the American Association of Oral and Maxillofacial Surgeons, *70*(3), 608–615. https://doi.org/10.1016/j.joms.2011.02.039

Wright, J. M., & Vered, M. (2017). Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and Maxillofacial Bone Tumors. *Head and neck pathology*, 11(1), 68–77. https://doi.org/10.1007/s12105-017-0794-1

Yamunadevi, A., Madhushankari, G. S., Selvamani, M., Basandi, P. S., Yoithapprabhunath, T. R., & Ganapathy, N. (2014). Granularity in granular cell ameloblastoma. *Journal of pharmacy & bioallied sciences*, 6(Suppl 1), S16–S20. https://doi.org/10.4103/0975-7406.137253

Zwahlen, R. A., & Grätz, K. W. (2002). Maxillary ameloblastomas: a review of the literature and of a 15-year database. *Journal of cranio-maxillo-facial surgery*: official publication of the European Association for Cranio-Maxillo-Facial Surgery, 30(5), 273–279. https://doi.org/10.1016/s1010-5182(02)90317-3

Porcentagem de contribuição de cada autor no manuscrito

Bruno Coelho Mendes – 20%

João Paulo Bonardi – 20%

Leonardo de Freitas Silva – 20%

Marcelo Macedo Crivelini – 20%

Ana Paula Farnezi Bassi – 20%