Figura 4: A) Fotografía frontal previa a la enucleación y curetaje de la lesión. **B)** Ortopantomografía previa a la enucleación y curetaje de la lesión. **C)** Fotografía frontal un año después de la enucleación y curetaje de la lesión. **D)** Ortopantomografía un año después de la enucleación y curetaje de la lesión.

A) *Frontal picture prior to enucleation and curettage of the lesion.* ***B)*** *Orthopantomography prior to enucleation and curettage of the lesion.* ***C)*** *Frontal picture one year after enucleation and curettage of the lesion.* ***D)*** *Orthopantomography one year after enucleation and curettage of the lesion.*

ca. Con este caso se demuestra que la descompresión más enucleación y curetaje son una opción de tratamiento importante para este tipo de lesiones aun cuando sean de gran tamaño, siendo fundamental el nivel sociocultural, compromiso y apego del paciente a sus controles clínicos e imagenológicos.

El control que se presenta de la paciente a un año después de haber realizado la enucleación y curetaje de la lesión no sólo permite observar que no hay hallazgos imagenológicos que indiquen una recidiva, sino que además se observa la regeneración ósea del sitio de donde se realizó la enucleación, aun así, el paciente continuará bajo control estricto clínico e imagenológico durante un periodo de 10 años.

Clinical case

Unicystic ameloblastoma treated by decompression and enucleation. Case report and literature review

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ABSTRACT

Background: Ameloblastoma is a benign tumor that originates from odontogenic epithelium. It develops in the region of the jaws but in 80% of cases it is located in the posterior region of the mandible. It has an intraosseous and progressive growth, causing expansion of cortical bone and up to 50% recurrence during the first five years if it is not treated properly. A conservative treatment may prevent esthetic and functional sequelae entailed by more aggressive treatment options for this type of lesions. **Objective:** We emphasize the importance of a strict clinical and imaging follow up to give the patient the possibility of undergoing conservative treatments for this type of odontogenic tumors. **Material and methods:** A 22-year-old female patient presented with a mural unicystic ameloblastoma in the region of the right mandibular body and angle. She was initially treated by decompression plus extraction of the associated tooth. Two years later, enucleation and curettage of the lesion was performed. Clinical and imaging follow up of the patient was done two months after surgery. **Results:** The conservative treatment by marsupialization with subsequent enucleation and curettage resulted in the complete neoformation of the condylar neck, coronoid process, ramus, angle, and mandibular body. **Conclusions:** Unicystic ameloblastomas can be treated by means of an initial decompression that helps to preserve and regenerate the affected anatomical regions to subsequently perform the enucleation of the lesion and thus prevent the esthetic and functional sequelae of conventional treatment.

Keywords: Unicystic, mural, enucleation, curettage, lesion, odontogenic tumor.

BACKGROUND

The unicystic ameloblastoma (UA) was first described by Robinson and Martinez in 1977 as a tumor derived from the remains of the dental lamina. The UA causes cortical bone expansion due to its slow growth and is usually asymptomatic. By imaging, it appears as a unilocular radiolucent lesion, with well-defined sclerotic borders, and is associated with retained third molar teeth in 80% of cases.[1-3] According to the imaging characteristics, there are two types of UA, namely a dentigerous and a nondentigerous variant, which depends on whether it is related or not to a retained tooth.

Robinson and Martinez proposed that the UA originated from a dentigerous cyst. This theory was refuted by Li et al. by comparing the expression of PCNA (Proliferating Cell Nuclear Antigen) in the epithelium of both the UA and the dentigerous cyst. Li et al. demonstrated that the PCNA antigen was expressed only in the epithelium of the dentigerous cyst. Even in the epithelium portions of the UA that most resembled those of the dentigerous cyst the PCNA antigen was not expressed. The origin of the UA from a dentigerous cyst was thus discarded.[4-7]

In 2017, the World Health Organization included ameloblastoma in the group of Benign epithelial odontogenic tumors and subclassified it into 4 types: ameloblastoma, unicystic ameloblastoma (UA), peripheral ameloblastoma, and metastasizing ameloblastoma.[8] UA occurs in the posterior region of the mandible in a 7:1 ratio relative to the maxilla. It accounts for 5% to 22% of all cases of ameloblastoma and most of these tumors are diagnosed in the first and second decade of life with a peak incidence at 16 years of age, even though they can also occur in the fourth and fifth decade of life.

The dentigerous variant of UA is more common in men in their teens and twenties, whereas the nondentigerous variant develops mostly in women in their thirties.[9]

The epithelium of the tumor must have the following characteristics to be able to make a positive diagnosis (described by Vickers and Gorlin in 1970 for ameloblastoma): basal cells in palisade, reverse nuclear polarization, cytoplasmic vacuolization, and cellular hyperchromatism.[10]

On the other hand, Akerman et al. in 1992 studied the type and zone of cell proliferation of the UA epithelium in relation to the capsule and the lumen of the tumor and classified it as follows:

1. Luminal. The tumor lining is ameloblastic epithelium.
2. Intraluminal. Projections of ameloblastic tissue in the lumen without evidence of growth within the connective tissue.
3. Intramural. It shows islands of ameloblastic growth within the connective tissue.[10,11]

Treatment of UA is controversial as it ranges from simple enucleation, enucleation and curettage, decompression, to block resection with safety margin to avoid relapse and adjuvant combination to decrease the percentage of relapse.

With regard to decompression, it was first described in 1671 by Lean Scultet as a means of changing the internal pressure of cystic lesions. Decompression nullifies the constant stimulus that causes peripheral bone reabsorption and thus favors the decrease in the size of the lesion with the consequent bone apposition.[12-15]

The objective of this report is to emphasize the importance of providing a conservative treatment to the patient with this type of dental tumors to prevent the functional and esthetic sequelae entailed by the excision of the tumor through a conventional treatment.

CLINICAL CASE

A female patient aged 22 years presented with progressive and asymptomatic swelling of seven months of evolution in right facial middle and lower thirds. Orthopantomography and computerized axial tomography studies showed an irregularly shaped radiolucent lesion, measuring 10 × 8 × 4 cm, with well-delimited sclerotic borders in mandibular ramus. There was also perforation of the right vestibular cortical bone, angle and mandibular body with a third molar tooth shifted to the lower edge of the mandibular body *(Figure 1)*.

The patient underwent an incisional biopsy and related tooth extraction. The histopathological examination revealed a UA of mural type with plexiform pattern. Due to the size of the lesion, age of the patient, and possibility of adherence to appointments, a conservative treatment was chosen, which consisted of placing a decompression tube under local anesthesia on the anterior edge of the right mandible ramus to perform decompression of the lesion. Clinical and imaging follow-up was performed for two years in which the complete remodeling of right-side neck of the condyle, ramus, angle, and anterior part of the mandibular body was achieved. It only remained a lesion in the retromolar area of approximately 4 × 3 × 5 cm, so it was decided to perform enucleation and curettage *(Figures 2 and 3)*. The tissue obtained was sent to histopathological study resulting again in

diagnosis of UA of mural type with plexiform pattern. Clinical and imaging follow-up were done one year after enucleation. It is planned to maintain long-term clinical follow up *(Figure 4)*.

DISCUSSION

Since UA was first described in 1977 several treatments have been proposed in the literature. The size of the tumor in our patient made us choose the procedure of surgical resection with a safety free margin of 15 mm to obtain a percentage of recurrence of 3%.[14]

A systematic review showed that treatment by enucleation and curettage has a percentage of relapse of 30%. When the treatment is combined with decompression, relapse decreases to 16%, and if treatment consists of enucleation and curettage plus application of Carnoy's solution or liquid nitrogen the percentage of relapse drops to 10%.[15] Morgan et al. found that peripheral osteotomy (with or without Carnoy's solution application) significantly decreases recurrence rates;[16] in contrast, Tolstunov observed that performing peripheral osteotomy causes the epithelium of the lesion to be dispersed, thus increasing the possibility of recurrence.[17]

Tonietto et al. support the use of liquid nitrogen as adjuvant treatment. Liquid nitrogen preserves the inorganic properties of the bone compared with Carnoy's solution, which destroys the bone osteogenic and osteoconductive properties. In addition, Carnoy's solution is carcinogenic due to the amount of chloroform it contains.[18] Nakamura et al. observed that decompression is most effective in large lesions, although they do not define clearly when decompression is indicated. Yet, they reported a series of 50 cases in which the lesions resolved significantly with this type of treatment and surgical removal was performed to prevent a possible transformation of the residual epithelium.[19] Despite what is described in the literature and based on the experience of our service in the conservative treatment of this type of dental tumors, we decided to perform a treatment of decompression plus enucleation and curettage to avoid esthetic and functional sequelae in the patient presented in our clinical case.

CONCLUSIONS

There is controversy over the best UA treatment to achieve the lowest percentage of relapse. With the experience of our institution we decided to provide the patient with a conservative treatment option to maintain both function and esthetics. According to the literature, the size of the lesion makes it necessary to perform a resection with free margins. However, because of the good adherence and commitment of the patient to her appointments of follow up and post-surgical care we were able to perform a significant decompression of the lesion in the first instance to carry out in a second surgical time the enucleation and curettage of the residual epithelium. This allowed not only preserving anatomical structures, but also regenerating lost anatomical structures. Besides, it was possible to avoid surgical resection, which entails functional, esthetic, and psychosocial sequelae. This case shows that decompression plus enucleation and curettage are an adequate treatment option even for larger lesions. In addition, the sociocultural level, commitment and attachment of the patient to clinical and imaging follow up is fundamental.

Imaging follow up one year after the enucleation and curettage of the lesion showed bone regeneration and no sign of recurrence. The patient will continue under strict clinical and imaging follow up for a period of 10 years.

REFERENCIAS / REFERENCES

1. Seintou A, Martinelli-Kläy CP, Lombardi T. Unicystic ameloblastoma in children: systematic review of clinicopathological features and treatment outcomes. *Int J Oral Maxilofac Surg*. 2014; 43: 405-412.
2. Ming-Hsuan H, Meng-Ling C, Jyh-Kwei C. Unicystic ameloblasatoma. *J Dent Sci*. 2012; 20: 1-5.
3. Heikinheimo K, Kurppa KJ, Elenius K. Novel targets for the treatment of ameloblastoma. *J Dent Res*. 2015; 94 (2): 237-240.
4. Argandoña JP, Espinoza JY. Ameloblastoma uniquístico, bases del tratamiento conservador. Presentación de caso clínico y actualización de la bibliografía. *Rev Esp Cirug Oral y Maxilofac*. 2011; 33 (2): 88-92.
5. Flores JAE, Romero JF, Millan AFO. Ameloblastoma uniquístico. Reporte de un caso. *Rev Mex Cir Bucal Maxilofac*. 2009; 5 (3): 95-98.
6. Kurppa KJ, Catón J, Morgan PR et al. High frequency of BRAF V600E mutations in ameloblastoma. *J Pathol*. 2014; 232 (5): 488-491.
7. Tie-Jun L, Yun-Tag W, Shi-Feng Y, Guang-Yan Y. Unicystic ameloblastoma: a clinicopathology study of 33 chinese patients. *Am J Surg Pathol*. 2000; 24: 1385-1392.
8. Hirschhorn AI, Vered M, Buchner A, Greenberg G, Yahalom R. Unicystic ameloblastoma in an infant: a managment dillema. *J Craniomaxillofac Surg*. 2013; 30: 1-5.
9. Pogrel MA, Montes DM. Is there a role for enucleation in the management of ameloblastoma? *In J Oral Maxillofac Surg*. 2009; 38: 807-812.
10. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma:a clinicopathological study of 57 cases. *J Oral Pathol*. 1988; 17: 541-546.
11. Samman N. Recurrence related to treatment modalities of ameloblastoma: a systemic review. *Int J Oral Maxillofac Surg*. 2006; 35: 681-690.
12. Lee PK, Samman N. Unicystic ameloblastoma- use Carnoy's solution after enucleation. *Int J Oral Maxillofac Surg*. 2004; 33: 263-267.

13. Abdularahmman EA, Khalid AA. Management of large pediatric ameloblastoma: Conservative approach with 4 years follow up. *Oral and Maxillofacial Surgery Cases*. 2019; 100093.
14. Pogrel MA. Treatment of Keratocysts. The case for descompression and marsupialization. *J Oral Maxillofac Surg*. 2005; 63: 1667-1673.
15. Castro-Nuñez J. Decompression of odontogenic cystic lesions: past, present, and future. *J Oral Maxillofac Surg*. 2016; 74: 104. e1-104.e9.
16. Morgan TA, Burton CC, Qian F. A retrospective review of treatment of odontogenic keratocyst. *J Oral Maxillofac Surg*. 2005; 63 (5): 635-639.
17. Tolstunov L, Treasure T. Surgical treatment algorithm for odontogenic keratocyst: combined treatment of odontogenic keratocyst and mandibular defect with marsupialization, enucleation, iliac crest bone graft, and dental implants. *J Oral Maxillofac Surg*. 2008; 66 (5): 1025-1036.
18. Tonietto L, Borges HO, Martins CA, Silva DN, Sant'Ana Filho M. Enucleation and liquid nitrogen cryotherapy in the treatment of keratocystic odontogenic tumors: a case series. *J Oral Maxillofac Surg*. 2011; 69 (6): 112-117.
19. Nakamura N, Mitsuyasu T, Taketomi T. Higuachi Y, Ohishi M. Marsupialization for odontogenic keratocysts: long-term follow-up analysis of the effects and changes in growth characteristics. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2002; 94 (5): 543-553.

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