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Unicystic Ameloblastoma in a Pediatric Patient: Case Report

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ABSTRACT

This article describes the clinical, histological, radiological characteristics, and treatment of unicystic ameloblastoma by presenting a clinical case of an 11-year-old patient, over a period of 2 years of clinical control and follow-up, in which histopathological diagnosis and radiographic control play a crucial role in diagnosis and the choice of an appropriate treatment plan. In this case, the patient underwent marsupialization, and after 4 weeks, enucleation in the mandibular symphysis and parasymphysis, along with odontectomy under general anesthesia. A treatment success rate of 100% without recurrence was achieved, and postoperative follow-up every 6 months was recommended.

Objective: To describe the characteristics and management of Unicystic Ameloblastoma, as well as the occurrence of the lesion in a pediatric patient.

KEYWORDS: Unicystic Ameloblastoma, odontogenic tumor, marsupialization, enucleation.

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INTRODUCTION

Ameloblastoma is a benign odontogenic tumor with aggressive behavior and a high risk of recurrence.[1,2] Its origin is from remnants of the dental lamina, enamel organ, Malassez epithelial remnants, and cyst epithelium.[3] This lesion is classified as Unicystic and Solid or Multicystic, Peripheral, and Malignant.[2] Unicystic Ameloblastoma (UA) is characterized by being a single, well-defined lesion, the cystic cavity is lined by typical ameloblastic cells and may or may not show luminal proliferation.[1,4,5] UA occurs in 50% of cases in the second decade of life and commonly localizes in the mandible.[5,6]

The purpose of this article is to present a rare case of UA with atypical localization in the mandibular symphysis and parasymphysis of a child associated with impacted teeth.

Clinical Characteristics

UA often presents clinically as a painless, slow-growing swelling, causing expansion of the cortical bone, extension of lingual and/or buccal plates, and penetration of soft tissues.[7] This lesion occurs mostly in young individuals and responds better to conservative treatment. [4,6,8,10] Approximately 50% of cases are diagnosed in the second decade of life, with a slight male predilection, and are most frequently located in the mandibular third molar area, ascending ramus, followed by the mandibular body and symphysis.[3,6]

Histological Characteristics

The typical morphology of ameloblastomas is odontogenic epithelial islands resembling the stellate reticulum, peripherally palisaded by columnar cells with inverted nuclear polarity.[11] UA is histologically classified into luminal type when the single cyst is lined by ameloblastic tissue, intraluminal type when ameloblastic tissue projects into the cyst lumen usually in a plexiform pattern, and mural type in which the epithelium with ameloblastic proliferation invades the fibrous wall of the cyst.[3,8] There are also epithelial variants, the most common being follicular, resembling enamel organ epithelium within a fibrous stroma; peripheral cells are columnar to cuboidal (similar to ameloblasts), with hyperchromatic nuclei arranged in a reverse-palisading pattern.[3] Other types include acanthomatous, granular, and basaloid. [3]

Radiological Characteristics

Radiographically, UA has unilocular, radiolucent characteristics and shows internal uniformity, resembling a Dentigerous Cyst or an Odontogenic Keratocyst.[6,8]

Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) show homogeneous internal density and signal intensity, with well-defined margins and thinning of the cortical bone. [8]

Treatment

When treating a patient with any type of ameloblastoma, surgery is the treatment of choice, however, the decision between conservative or radical approaches remains controversial.[2] Management depends on the clinical type of ameloblastoma, whether multicystic or unicystic.[12] When referring to conservative surgery, we mean enucleation or marsupialization, combined or not with support techniques such as curettage. Carnoy's solution is used to decrease the risk of recurrence after conservative surgical treatment, and liquid nitrogen as an adjunct method that preserves the inorganic properties of the bone.[5,12,13]

Siriwardena recommends that in pediatric patients, conservative treatment should be attempted to avoid facial deformity and minimize dysfunction in the context of unerupted teeth and continuous facial growth.[14] Furthermore, conservative treatments, while preserving bone integrity, appear to be associated with a high recurrence rate.[12] In the case of opting for radical surgery, the procedure refers to mandibulectomy or segmental resection of the lesion.[12] When there is a solid or multicystic ameloblastoma, the treatment of choice is radical surgery with safe margins of 1 cm and resection of adjacent soft tissue, followed by reconstruction.[2]

Some authors have suggested combined conservative surgery and long-term follow-up as a possible option, especially in cases with good patient compliance and low risk of affecting adjacent structures.[2] After the described treatments, it is necessary to mention that the evaluation of ameloblastoma treatment is a complex problem, as ideally, it should not be so destructive due to the benign nature of this lesion, but it should be extensive enough to avoid recurrences. Recurrence rates for UA vary, with reported values ranging from 5% to 25% after treatment.[6,8]

PRESENTATION OF THE CASE

An 11-year-old male presented with his guardian to the maxillofacial surgery clinic, reporting that he was taken to a health center for dental caries in the lower right quadrant. Upon clinical examination, a swelling in the area was evident, and purulent content evacuation was performed without systemic complications. The patient was referred to the maxillofacial surgeon.

On physical examination, he presented with increased volume in the right mandibular body, presence of deciduous teeth, and crepitus on palpation. Orthopantomography and cone-beam computed tomography (CBCT) were performed, identifying an intraosseous hypodense lesion in the mandibular body extending from the midline to the area of tooth 4.6, approximately 2.5cm x 5.2cm x 3.8cm in size, tooth 4.2 was impacted, 4.3 and 4.5 teeth were in root formation and horizontally mesioangular, near the basal border, in close contact with the inferior alveolar nerve, and the 4.4 tooth was distoangular intraosseous; 4.6 tooth showed root resorption (Figure 1-2).

Figure 1: Initial panoramic X-ray.

Figura 2: Tomografía corte axial.

Marsupialization was performed (Figure 3 A and B) and after 4 weeks, enucleation of the mandibular symphysis and parasymphysis along with odontectomy under general anesthesia was carried out (Figure 3 C); histological analysis reported stratified non-keratinized squamous epithelium about 3 to 4 layers thick, composed of cuboidal and hyperchromatic cells at the basal layer, with cytoplasmic vacuolization and intercellular spacing. Irregular masses projecting into the lumen were constituted by cuboidal or columnar cells with polarized nuclei similar to ameloblasts or pre-ameloblasts, surrounding a central portion of elongated cells resembling the stellate reticulum, mimicking the dental follicle stage during organogenesis. The lesion capsule showed connective tissue with abundant collagen and blood vessels filled with erythrocytes without epithelial rests. The final diagnosis was luminal type Unicystic Ameloblastoma. (Figure 4 A and B)

(A)

(B)

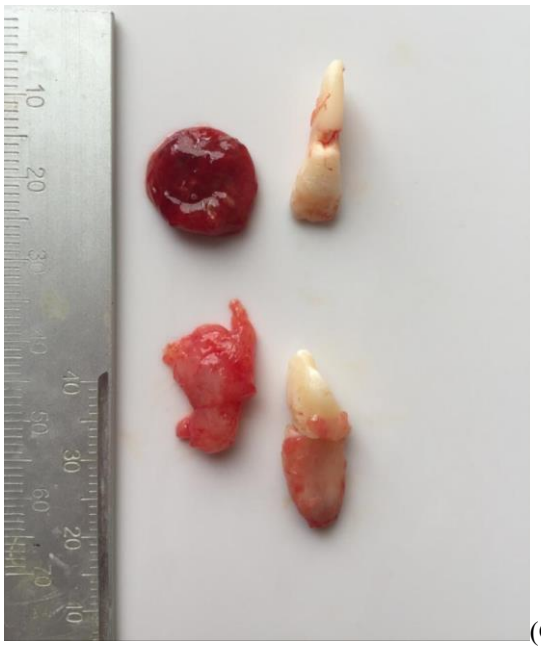
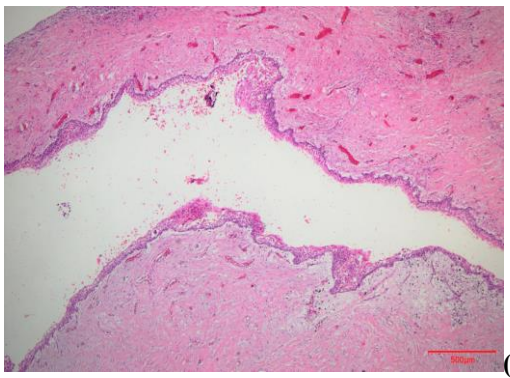
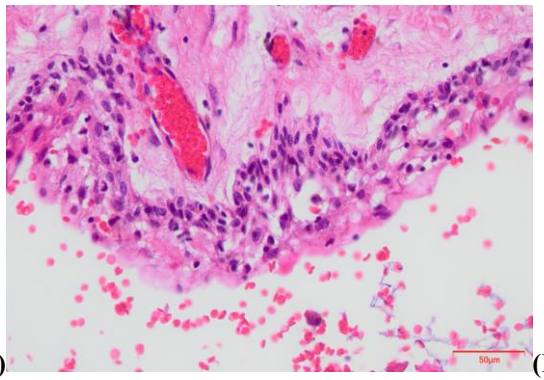

(C)

Figure 3: A) Marsupialization B) Macroscopy Lesion C) Enucleation.


(A)

(B)

"Figure 4: A) Histology staining H&E 100x B) Histology staining H&E 400x."

Subsequently, clinical and radiographic follow-ups were performed, recommending semi-annual controls (Figure 5-6-7).

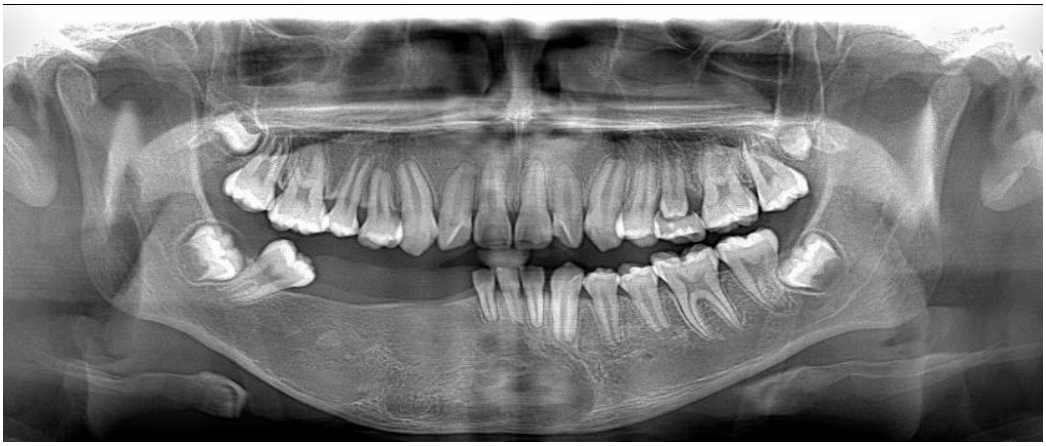


Figure 5: One-year follow-up X-ray.

Figure 6: Two-year follow-up X-ray.

Figure 7: Follow-up after two years.

DISCUSSION

To obtain a definitive diagnosis in this clinical case, we relied on the results of complementary tests of great importance such as lesion biopsy and CBCT tomography. With these tests, an accurate result was obtained that this patient had a unicystic ameloblastoma. It should be noted that there are various differential diagnoses that can be confused with this lesion, including odontogenic keratocyst, nasosinusal papilloma, ameloblastic fibroma, squamous cell carcinoma, adenoid cystic carcinoma, and basal cell carcinoma.[11,15]

According to the criteria of the World Health Organization (WHO), there is a slight predilection for males. According to Hendra et al. 2019, in a worldwide study with 6,435 AU patients, 3,427 (53.2%) cases were male and 3,008 (46.7%) were female, and ultimately, this study indicates that sex prevalence was not statistically significant.[3,16]

Other authors found no significant difference in AU predilection in relation to a specific gender.[6,11,14,16,17]

AU is the second most common of ameloblastomas worldwide, accounting for 26.2%; in North America, it represents 7.4%, and in South America, 11% of all types of ameloblastomas.[7]

This case demonstrates the importance of making an early diagnosis by identifying clinical and radiographic alterations in the patient's anatomy, taking into account their age because it is important to provide appropriate treatment so that their growth and development, both functional and aesthetic, are not affected.

Risimati, in his case report, conducted follow-ups every 3 to 6 months over a period of 3 years and presented complete decompression and bone regeneration in the defect, with no signs of recurrence; furthermore, it was indicated that long-term follow-up should include both clinical and radiographic examinations for at least 10 years, with yearly reviews.[18] Similarly, Neagu and colleagues suggest annual follow-up for 10 years due to the potential for recurrence.[12] CBCT can be performed 6 months after the final curettage to detect any recurrence and to evaluate bone density.[2]

CONCLUSION

Ameloblastoma is a benign, aggressive odontogenic tumor with a high likelihood of recurrence. Within its classification, this article presents a case of a Unicystic Ameloblastoma, characterized by being a single, well-defined lesion located in

the mandibular symphysis and parasymphysis of a pediatric patient associated with impacted teeth.

Conservative treatment of UA in young patients is indicated for the preservation of function and aesthetics. Histopathological diagnosis, CBCT computed tomography, and differential diagnosis significantly influence the choice of appropriate treatment to prevent recurrence in the future.

REFERENCES

I. Zheng CY, Cao R, Hong WS, Sheng MC, Hu YJ. Marsupialisation for the treatment of unicystic ameloblastoma of the mandible: a long-term follow up of 116 cases. British Journal of Oral and Maxillofacial Surgery [Internet]. Septiembre de 2019 [consultado el 7 de noviembre de 2022];57(7):655-62. Disponible en: https://doi.org/10.1016/j.bjoms.2019.06.002

II. Nowair IM, Eid MK. A modified surgical approach for the treatment of mandibular unicystic ameloblastoma in young patients. Journal of Cranio-Maxillofacial Surgery [Internet]. Febrero de 2020 [consultado el 7 de noviembre de 2022];48(2):148-55. Disponible en: https://doi.org/10.1016/j.jcms.2019.12.013

III. WHO Classification of head and neck tumours. [lugar desconocido]: International Agency for Research on Cancer (IARC); 2017.

IV. Ricci M, Mangano F, Tonelli P, Barone A, Galletti C, Covani U. An unusual case of unicystic intramural ameloblastoma and review of the literature. Contemporary Clinical Dentistry [Internet]. 2012 [consultado el 7 de noviembre de 2022];3(6):233. Disponible en: https://doi.org/10.4103/0976-237x.101104

V. Agani Z, Hamiti-Krasniqi V, Recica J, Loxha MP, Kurshumliu F, Rexhepi A. Maxillary unicystic ameloblastoma: a case report. BMC Research Notes [Internet]. 18 de octubre de 2016 [consultado el 7 de noviembre de 2022];9(1). Disponible en: https://doi.org/10.1186/s13104-016-2260-7

VI. Seintou A, Martinelli-Kläy CP, Lombardi T. Unicystic ameloblastoma in children: systematic review of clinicopathological features and treatment outcomes. International Journal of Oral and Maxillofacial Surgery [Internet]. Abril de 2014 [consultado el 7 de noviembre de 2022];43(4):405-12. Disponible en: https://doi.org/10.1016/j.ijom.2014.01.003

VII. Hendra FN, Natsir Kalla DS, Van Cann EM, de Vet HC, Helder MN, Forouzanfar T. Radical vs conservative treatment of intraosseous ameloblastoma: Systematic review and meta-analysis. Oral Diseases [Internet]. Enero de 2019 [consultado el 7 de noviembre de 2022];25(7):1683-96. Disponible en: https://doi.org/10.1111/odi.13014

VIII. Wakoh M, Matsuzaka K, Imoto K, Goto T, Kamio T, Shibahara T. Luminal, Intramural Unicystic Ameloblastoma with Marked Fluid-Fluid Level: Validity of CT and MRI Findings. The Bulletin of Tokyo Dental College [Internet]. 2020 [consultado el 7 de noviembre de 2022];61(1):61-9. Disponible en: https://doi.org/10.2209/tdcpublication.2018-0064

IX. Takahashi S, Idaira Y, Sato T, Asada Y, Nakagawa Y. Unicystic Ameloblastoma in a Child Treated with a Combination of Conservative Surgery and Orthodontic Treatment: A Case Report. Journal of Clinical Pediatric Dentistry [Internet]. 1 de enero de 2019 [consultado el 7 de noviembre de 2022];43(2):121-5. Disponible en: https://doi.org/10.17796/1053-4625-43.2.9

X. Isolan C, Moreira A, Edges A, Post L, Aitken-Saavedra J. Successful conservative treatment of a mandibular unicystic ameloblastoma: 13-year follow-up. Journal of Clinical and Experimental Dentistry [Internet]. 2018 [consultado el 7 de noviembre de 2022]:0. Disponible en: https://doi.org/10.4317/jced.54897

XI. EVANGELOU Z, ZARACHI A, DUMOLLARD JM, PEOC'H M, KOMNOS I, KASTANIOUDAKIS I, KARPATHIOU G. Maxillary Ameloblastoma: A Review With Clinical, Histological and Prognostic Data of a Rare Tumor. In Vivo [Internet]. 2020 [consultado el 7 de noviembre de 2022];34(5):2249-58. Disponible en: https://doi.org/10.21873/invivo.12035

XII. Neagu D, Escuder-de la Torre O, Vazquez-Mahia I, Carral-Roura N, Rubin-Roger G, Penedo-Vazquez Á, Luaces-Rey R, Cedrun-Cembranos J. Surgical management of ameloblastoma. Review of literature. Journal of Clinical and Experimental Dentistry [Internet]. 2019 [consultado el 7 de noviembre de 2022]:0. Disponible en: https://doi.org/10.4317/jced.55452

XIII. Liceaga Escalera CJ, Montoya Pérez LA, Vélez Cruz M, Jiménez de la Puente G. Ameloblastoma uniquístico tratado mediante descompresión y enucleación. Reporte de un caso y revisión de la literatura. Revista Odontológica Mexicana [Internet]. 13 de noviembre de 2020 [consultado el 7 de noviembre de 2022];24(1). Disponible en: https://doi.org/10.22201/fo.1870199xp.2020.24.1.77550

XIV. Siriwardena BS, Tennakoon TM, Hunter KD, Tilakaratne WM. Unicystic ameloblastoma: Analysis of 370 cases in a single center in Sri Lanka. Journal of Oral Pathology & Medicine [Internet]. 20 de junio de 2018 [consultado el 7 de noviembre de 2022];47(7):706-9. Disponible en:

https://doi.org/10.1111/jop.12740

XV. Anandani C, Metgud R, Singh K. Calretinin as a Diagnostic Adjunct for Ameloblastoma. Pathology Research International [Internet]. 15 de abril de 2014 [consultado el 7 de noviembre de 2022];2014:1-7. Disponible en: https://doi.org/10.1155/2014/308240

XVI. Hendra FN, Van Cann EM, Helder MN, Ruslin M, de Visscher JG, Forouzanfar T, Vet HC. Global incidence and profile of ameloblastoma: A systematic review and meta-analysis. Oral Diseases [Internet]. 25 de enero de 2019 [consultado el 7 de noviembre de 2022];26(1):12-21. Disponible en: https://doi.org/10.1111/odi.13031

XVII. Al Sineedi F, Aruveetil YA, Kavarodi AM, Harbi SO. Bilocular unicystic ameloblastoma of the mandible in a 9 yr old child – A diagnostic and management dilemma. The Saudi Dental Journal [Internet]. Julio de 2018 [consultado el 7 de noviembre de 2022];30(3):250-5. Disponible en: https://doi.org/10.1016/j.sdentj.2018.03.001

XVIII. Rikhotso RE, Premviyasa V. Conservative Treatment of Ameloblastoma in a Pediatric Patient: A Case Report. Journal of Oral and Maxillofacial Surgery [Internet]. Agosto de 2019 [consultado el 7 de noviembre de 2022];77(8):1643-9. Disponible en: https://doi.org/10.1016/j.joms.2019.02.025

XIX. Reichart PA, Philipsen HP, Sciubba JJ. The new classification of Head and Neck Tumours (WHO) — any changes? Oral Oncology [Internet]. Septiembre de 2006 [consultado el 8 de noviembre de 2022];42(8):757-8. Disponible en: https://doi.org/10.1016/j.oraloncology.2005.10.011