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Clinical Case

Conservative Surgery to Approach Odontogenic Keratocyst in Young Patient: Case Report

Cirurgia Conservadora no Manejo de Ceratocisto Odontogênico em Paciente Jovem: Relato de Caso

Cirugía Conservadora en el Manejo de Queratoquiste Odontogénico en Paciente Joven: Reporte de Caso

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Abstract

The odontogenic keratocyst consists of a benign lesion of the oral cavity that involves peculiarities regarding its growth, treatment, and prognosis. The main objective in the treatment of this pathology is to guarantee small morbidity to the patient and to reduce recurrences, which are characteristic of this type of injury. In this case report, we demonstrate the effectiveness of less invasive treatments for smaller-sized lesions that responded well to cystic decompression. A 33-year-old female, diagnosed with odontogenic keratocyst in the region of the angle and left mandibular ramus, was surgically treated by five months of marsupialization and subsequent enucleation with a 30-month follow-up without relapsing. The diagnosis of this pathology is performed through radiographic and histopathological exams associated with clinical findings. Due to the slow asymptomatic growth, the identification of the keratocyst is often made lately, and a radical treatment can promote functional and esthetic deficiencies. Even with favorable results of conservative treatment, long-term follow-up should be performed to detect possible relapses.

Descriptors: Decompression; Odontogenic Cysts; Recurrence; Conservative Treatment.

Resumo

O ceratocisto odontogênico consiste em uma lesão benigna da cavidade oral que envolve peculiaridades quanto ao seu crescimento, tratamento e prognóstico. O principal objetivo no tratamento dessa patologia é garantir pequena morbidade ao paciente e reduzir as recorrências, muito comuns nesse tipo de lesão. Neste relato de caso demonstramos a eficácia de tratamentos menos invasivo para lesões de tamanho menor e que respondem à descompressão cística. Paciente feminina de 33 anos, com diagnóstico de ceratocisto odontogênico na região de ângulo e ramo mandibular esquerdo, foi tratada cirurgicamente por cinco meses com marsupialização e posterior enucleação, com acompanhamento pós-operatório de 30 meses sem recidiva. O diagnóstico desta patologia é realizado através de exame radiográfico e histopatológico associados aos achados clínicos. Devido ao crescimento lento e assintomático, a identificação do ceratocisto costuma ser feita tardiamente, e um tratamento radical pode promover deficiências funcionais e estéticas. Mesmo com resultados favoráveis ao tratamento conservador, um acompanhamento em longo prazo deve ser realizado para detectar possíveis recidivas.

Descritores: Descompressão; Cistos Odontogênicos; Recidiva; Tratamento Conservador.

Resumen

El queratoquiste odontogénico consiste en una lesión benigna de la cavidad oral que presenta particularidades en cuanto a su crecimiento, tratamiento y pronóstico. El objetivo principal en el tratamiento de esta patología es asegurar una baja morbilidad para el paciente y reducir las recidivas, muy frecuentes en este tipo de lesiones. En este informe de caso demostramos la eficacia de tratamientos menos invasivos para lesiones más pequeñas que responden a la descompresión quística. Paciente femenina de 33 años con diagnóstico de queratoquiste odontogénico en región de ángulo y rama mandibular izquierda, fue tratada quirúrgicamente durante cinco meses con marsupialización y posterior enucleación, con seguimiento postoperatorio de 30 meses sin recurrencia. El diagnóstico de esta patología se realiza mediante examen radiográfico e histopatológico asociado a hallazgos clínicos. Debido al crecimiento lento y asintomático, los queratoquistes suelen identificarse tardíamente y el tratamiento radical puede provocar deficiencias funcionales y estéticas. Incluso con resultados favorables del tratamiento conservador, se debe realizar un seguimiento a largo plazo para detectar posibles recidivas.

Descriptores: Descompresión; Quistes Odontogénicos; Recurrencia; Tratamiento Conservador.

INTRODUCTION

The odontogenic keratocyst is a benign lesion of the oral cavity, which deserves special attention due to its histopathological and clinical behavior specificities. Such injury originates from remnants of the dental lamina and may present locally aggressive behavior. Its growth typically occurs in the anteroposterior direction, resulting in lesions with low cortical expansion, but can cause dental dislocations depending on their dimensions. It is worth mentioning that the growth of the odontogenic keratocyst is caused by unknown factors, which may be related to the epithelium itself or enzyme activity in the cyst wall[1].

Radiographically is characterized by a single, radiolucent and ill-defined lesion, which may be related to the crown of an unerupted tooth, commonly diagnosed in routine exams due to the asymptomatic growth in most patients[2]. This cyst represents 10% to 20% of all types of odontogenic cysts and is the third most common cystic type in the mandible, with greater involvement in the

regions of the posterior body and mandibular ramus, with a slight predilection for males. The definitive diagnosis occurs after the histopathological analysis of material collected in the biopsy, and the treatment of choice depends on its clinical presentation (size, location, proximity to noble structures) [2].

The present article aims to report on a singular case of a slow and asymptomatic odontogenic keratocyst in the posterior region of the mandible, whose diagnosis was initially masked by a local chronic infection. This lesion received surgical treatment for decompression with subsequent enucleation and is still under clinical follow-up without recurrences.

CASE REPORT

A 33-year-old female patient was referred to clinical evaluation due to severe pain and intraoral suppuration in her left jaw in April of 2018. During anamnesis, the patient reported a lower third molars extraction for approximately 9 years and a radiolucent lesion was seen associated with the crown of the unerupted left lower third molar (Figure 1). No allergies or comorbidities were present.

Figure 1: Panoramic radiograph before left lower third molar extraction. A radiolucent lesion associated with the crown of the unerupted molar is seen.

On physical examination, the patient presented moderate trismus, facial asymmetry, and left vestibular fundus erasure. In a panoramic radiographic examination, a well-defined radiolucent lesion was observed in the region of the angle and left mandibular ramus, where the clinical hypotheses were dentigerous cyst and odontogenic keratocyst (Figure 2). Due to the acute infection, Amoxicillin 500mg was prescribed every 8 hours for seven days and the patient returned for reevaluation in one week, with an incisional biopsy scheduled for this second moment.

After seven days, without signs of infection, an incisional biopsy was performed under local anesthesia and marsupialization of the lesion was made with 4-0 nylon thread. The primary anatomopathological result was a cystic capsule showing chronic foreign body granulomatous inflammation.

After five months of follow-up, a reduction in the size of the lesion was observed (Figure 3).

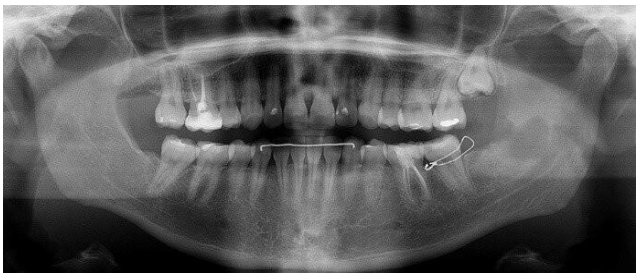

Figure 2: Initial radiographic image made in 2018 after clinic examination with visualization of a well-circumscribed radiolucent lesion in the angle and left mandibular ramus region.

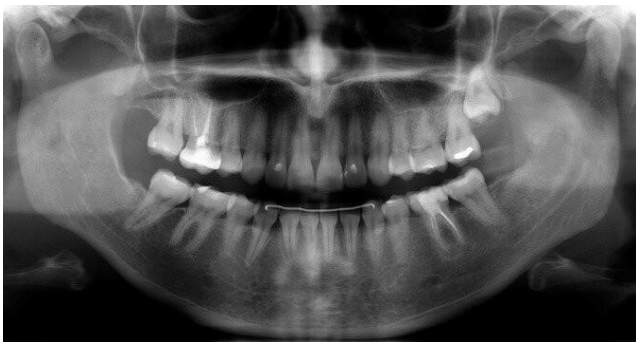

Figure 3: Panoramic radiograph after 05 months of decompression of the lesion, showing a reduction in the total size of the lesion. At this moment, total enucleation was planned.

As a definitive treatment, we opted for total enucleation of the lesion, under general anesthesia, through intraoral access. The enucleated material of dimensions 13x7x4mm was sent for histopathological analysis, with a definitive result of an odontogenic keratocyst. Monthly returns were performed, with no signs of infection and postoperative paresthesia being observed. After 30 months of follow-up of total enucleation, an appropriate bone neoformation could be observed radiographically, with no signs of lesion recurrence (Figure 4).

Figure 4: Radiographic control of 30 months of follow-up of odontogenic keratocyst enucleation showing satisfactory bone neoformation, with complete remission of the lesion and no signs of recurrence.

DISCUSSION

Odontogenic keratocysts are benign lesions of the oral cavity with peculiar characteristics that deserve attention. The diagnosis is usually made from routine panoramic radiographs used to investigate other causes, such as an impacted tooth[2]. It is worth noting that the patient of this report

had a history of radiolucent lesion associated with the unerupted lower third molar before its extraction, which was not initially treated and there was growth progression over the years, being discovered in a routine examination for investigation of pain in the lower left quadrant. In addition, the maintenance of the remnants of the cystic capsule, due to its incomplete removal at the time of extraction, provided a means of continuous communication between the cyst and the oral cavity, causing a chronic and prolonged infection, which made the initial diagnosis of the correct pathology difficult by modifying the microenvironment and characteristics of the cystic capsule, with a definitive diagnosis being reached only after total enucleation of the lesion[1].

Anand et al.[3] highlighted the need for differential clinical and radiographic diagnosis of the dental follicles associated with unerupted molars. The absence of symptoms is not necessarily associated with the absence of an existing pathology, where it is not possible to determine radiographically which dental follicle with higher thickness will progress to detectable clinical lesions[3]. This case reinforces the need for careful diagnosis during the evaluation of the included teeth, for the detection of alterations and early management, and, in the presence of thicker follicles, they must be removed during extraction and submitted to histopathological analysis.

Different modalities of treatment of odontogenic keratocyst were observed in the literature, ranging from conservative (enucleation, cryotherapy, marsupialization) to radical approaches (surgical resection)[4]. Each type of treatment has advantages and disadvantages, in addition to different rates of recurrence, so it is necessary to analyze the specificities of each case in the planning and establishment of the therapeutic conduct.

The main discussion among studies on this topic is related to what type of approach should be chosen to guarantee the reduction of the high rates of recurrence involving this injury, which can reach 62%[5]. In a study by Al-Moraissi et al.[1,5], several forms of treatment were analyzed, suggesting enucleation after the cyst decompression. The authors also claim that radical resection is the optional treatment with the lowest recurrence rate, but to ensure a lower rate of morbidity and esthetic-functional deficits, resections should be reserved for multiple and recurrent lesions and the possibility of syndromic cases, which is not the case of the patient in this report. Therefore, the decompression followed by enucleation guarantees the preservation of noble structures and anatomy for lower lesions. After the individual analysis of the case, the therapeutic approach is determined based on a combination of factors that guarantee a better prognosis and less morbidity to the patient[6,7]. Corroborating with the literature, we demonstrated a reduction in the total size promoted by marsupialization, which allowed enucleation through intraoral access, preventing the formation of unsightly scars in the patient and preservation of the lower alveolar nerve, without the evolution of postoperative paresthesia.

According to Maurette et al.[8], the decompression time varies from 1 to 14 months, which can be a disadvantage in the choice of this type of approach, in addition to being a factor of abandonment or lack of cooperation on the part of the patient. In the present case, the patient was well-educated and collaborative, with regular attendance at clinical follow-ups, committed to the hygiene of the marsupialization site during the time of decompression and showed significant results in just 5 months. This treatment modality has advantages because it is less traumatic to the patient, reduces the need for hospital care and the spent on medication[6-8]. In addition, it is worth mentioning that, as it is a conservative treatment modality, it allows the dismissal of small or extensive reconstructions after the removal of the lesion, prolonging the total treatment time, since the surrounding bone structure can recover naturally and gradually.

Still, many authors are aware of the need for clinical and radiographic monitoring of patients for a certain period after the surgical treatment, since there are high rates of recurrence, detected in the first 5-15 years after treatment[9]. According to Kshirsagar et al.[9], follow-up should be done once a year for the first 5 years and, after that period, if there are no recurrences, it should be performed once every 2 years. Vallejo-Rosero et al.[11] confirm that continuity of follow-up is mandatory since relapses can occur after years of surgical treatment. In the study presented by Borrás-Ferreres et al.[10], there was no evidence of recurrence after 2 years of follow-up in cases treated conservatively, which is compatible with our findings. No relapses were observed in 30 months of follow-up.

CONCLUSION

The combination of correct diagnosis, an adequate therapeutic approach, and the patient's cooperation with team care are the main variables involved in the success of a clinical case. In this report, the positive results evidence a less traumatic technique that promoted the removal of the lesion without causing higher morbidity to a cooperative patient and a period of significant clinical follow-up without the presence of recurrences.

REFERENCES

1. Neville, W.B., Damm, D.D., Allen, C.M., Bouquot, J.E. Patologia oral & maxilofacial, Rio de Janeiro, Guanabara Koogan, 3ªed, 2009.

2. El-Naggar A.K., Chan J.K.C., Grandis J.R., Takata T., Slootweg P.J. (Eds): WHO Classification of Head and Neck Tumours (4th edition). IARC: Lyon 2017.
3. Anand S, Kashyap B, Kumar GR, Shruthi BS, Supriya AN. Pericoronal radiolucencies with significant pathology: clinico-histopathologic evaluation. Biomed J. 2015;38(2):148-52.
4. Tabrizi R, Hosseini Kordkheili MR, Jafarian M, Aghdashi F. Decompression or Marsupialization; Which Conservative Treatment is Associated with Low Recurrence Rate in Keratocystic Odontogenic Tumors: A Systematic Review. J Dent Shiraz Univ Med Sci. 2019; 20(3):145-151.
5. Al-Moraissi EA, Dahan AA, Alwadeai MS, Oginni FO, Al-Jamali JM, Alkhutari AS, Al-Tairi NH, Almaweri AA, Al-Sanabani JS. What surgical treatment has the lowest recurrence rate following the management of keratocystic odontogenic tumor?: A large systematic review and meta-analysis. J Craniomaxillofac Surg. 2017;45(1):131-144.
6. Yildirim G, Ataoglu H, Kalayci A, Ozkan BT, Kucuk K, Esen A. Conservative Treatment Protocol for Keratocystic Odontogenic Tumour: a Follow-up Study of 3 Cases. J Oral Maxillofac Res. 2010;1(3):e7.
7. de Castro MS, Caixeta CA, de Carli ML, Ribeiro Júnior NV, Miyazawa M, Pereira AAC, Sperandio FF, Hanemann JAC. Conservative surgical treatments for nonsyndromic odontogenic keratocysts: a systematic review and meta-analysis. Clin Oral Investig. 2018;22(5):2089-2101.
8. Maurette PE, Jorge J, de Moraes M. Conservative treatment protocol of odontogenic keratocyst: a preliminary study. J Oral Maxillofac Surg. 2006;64(3):379-83.
9. Kshirsagar RA, Bhende RC, Raut PH, Mahajan V, Tapadiya VJ, Singh V. Odontogenic Keratocyst: Developing a Protocol for Surgical Intervention. Ann Maxillofac Surg. 2019;9(1):152-157.
10. Borrás-Ferreres J, Sánchez-Torres A, Alberdi-Navarro J, Aguirre-Urizar JM, Mosqueda-Taylor A, Gay-Escoda C. Therapeutic management of the odontogenic keratocyst. An energetic approach with a conservative perspective and review of the current therapeutic options. J Clin Exp Dent. 2020;12(8):e794-9.
11. Vallejo-Rosero KA, Camolesi GV, de Sá PLD, Bernaola-Paredes WE. Conservative management of odontogenic keratocyst with long-term 5-year follow-up: Case report and literature review. Int J Surg Case Rep. 2020;66:8-15.

CONFLICT OF INTERESTS

The authors declare no conflict of interest.

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