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CASE REPORT

Decompression followed by enucleation in the treatment of orthokeratinized odontogenic cyst: case report

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Abstract:

The orthokeratinized odontogenic cyst, currently classified as a separate entity from the odontogenic keratocyst, is a rare developmental cyst that affects the gnatic bones. It predominantly affects young male patients. Its clinical behavior is benign with a predominance of involvement by the posterior region of the mandible. Treatment involves enucleation, with or without prior decompression. The present case reports a 17-year-old male patient with bilateral lesion manifestation, the left osteolytic process being of large proportions. The patient was subjected to decompression prior to enucleation, and this conservative treatment is essential for successful treatment. The patient was followed up for 1 year with no signs of recurrence.

Keywords: Decompression; Bone cysts; Pathology.

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INTRODUCTION

The orthokeratinized odontogenic cyst (COO) was first described by Schultz in 1927[1,2], however it was in 1981 that Wright identified COO as a variant of the odontogenic keratocyst[2]. Later, WHO defined the orthokeratinized odontogenic cyst as an entity separate from the odontogenic keratocyst[1]. Although originally classified as a variant of the keratocyst, it is currently placed in a different category due to its own characteristics[2,3] such as low aggressiveness, low relapse rate and no association with Basal Cell Nevoid Carcinoma Syndrome[4–6]. COO represents 7% to 17% of all keratinizing cysts in the gnathic bones[7].

The COO can appear under the influence of the dental papilla or just the oral epithelium. On occasions when COO is associated with an impacted tooth, it is stated that due to the pluripotentiality of the odontogenic cyst epithelium, the reduced enamel epithelium after completing its dental formation function, has the ability to keratinize under appropriate stimuli[1]. Often seen in the 3rd and 4th decades of life and predominantly in men, it shows a predilection for the posterior region of the mandible and usually presents as a single lesion; multiple occurrences are rare[4,8]. COO's are often asymptomatic, discovered as an incidental finding, however, swelling and pain can be reported[1,9].

Radiographically, they are well-defined unilocular radiolucencies, in most cases associated with an impacted tooth. Few cases are multilocular and their diameter varies from 20 to 70 mm with an average size of 48 mm[2,9]. Its growth causes buccolingual expansion, a characteristic not frequently found in odontogenic keratocyst, which shows anteroposterior growth pattern[9]. Histopathologically, the epithelial lining shows four to eight cell layers of thick, uniform ortho-keratinized stratified squamous epithelium with a prominent granular cell layer. Basal cells are flat to cuboidal, without the characteristics of palisade or polarization, which are commonly seen in keratocyst, as well as the absence of parakeratin, also found in keratocyst[1,2,8].

Treatments range from decompression, marsupialization and enucleation or a combination of these therapies. Decompression as an initial procedure is a common conservative approach that requires an opening in the cystic wall keeping the patent with its own device such as a tube or stent. The objective is to decrease intraluminal pressure, allowing new centripetal bone growth in the cyst walls[10,11]. The main advantage of decompression is the preservation of important anatomical structures, such as teeth and the lower alveolar nerve and the prevention of a possible consequent deformity and / or mandibular fracture[12].

CASE REPORT

Male patient, 17 years old, went to the outpatient clinic of the Buccomaxillofacial Surgery and Traumatology service at the Municipal Hospital Dr. Mario Gatti, showing wide bulging in the buccal and lingual regions in the region of the left mandibular body, elements 35 and 36 showed degree 1 mobility, patient denied paresthesia, pain or any other symptom. The right side was normal. The panoramic radiographic examination showed a wide osteolytic, radiolucent, unilocular lesion, involving the left mandibular body, covering elements 32 to 38 that were included, and it was also possible to notice root spacing, without reabsorption. On the right side, it was possible to observe unilocular, radiolucent, osteolytic lesion, in a region distal to element 47 (Figure 1). The patient was submitted to an aspiration puncture on the left side, obtaining straw-colored liquid content as material. Two biopsies under local anesthesia were performed. The excisional biopsy was performed on the right side and the incisional biopsy was performed on the left side along with the placement of two rigid drains for decompression. A number 12 urethral tube was used as a drain and it was fixed with steel wire number 1 and nylon 4.0 wire. (Figure 2).

Computed tomography was performed after surgery to accurately assess the position of the drains, as well as the total extent of the lesion (Figure 3), the examination evidenced the large bucolingual expansion caused by the cyst. Both injuries were referred to the Pathology Center of Faculdade São Leopoldo Mandic, resulting in an Orthokeratinized Odontogenic

Figure 1. Initial panoramic radiograph showing bilateral unilocular radiolucent lesions in the mandibular body. It is possible to observe root clearance of premolars and molars.

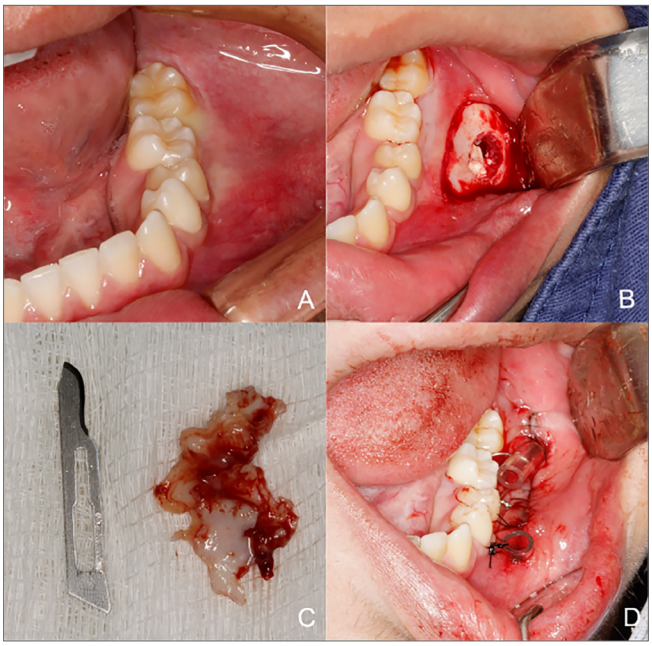

Figure 2. A) Bulging in the buccal and lingual regions of the left mandibular body B) Excisional biopsy. C) Fragment of the lesion. D) Apposed rigid drains.

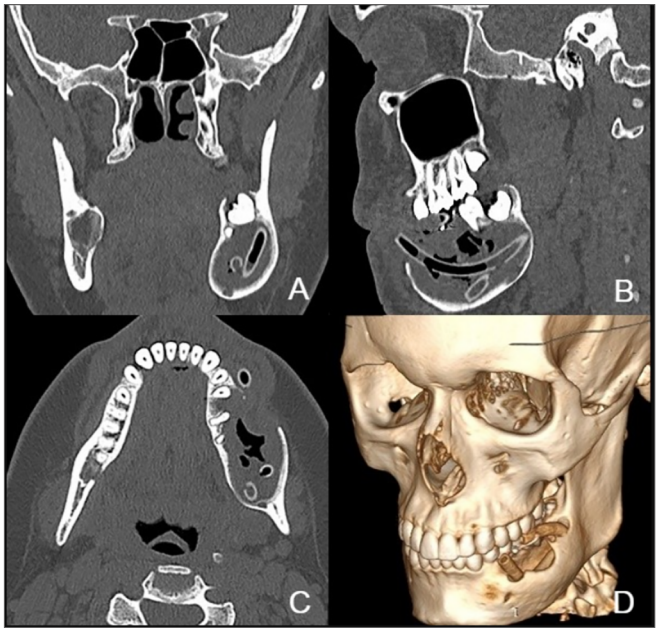

Figure 3. Computed tomography in the coronal (A), sagittal (B), axial (C) and 3D reconstruction (D) sections performed after the placement of the drains. It is possible to perceive the bucolingual expansion of the region of the left mandibular body, as well as the resorption of part of the vestibular cortex.

Cyst. Histopathological analysis revealed cystic capsules covered by stratified squamous epithelium, orthokeratinized and with few layers, presenting a flat interface with the connective tissue capsule and absence of basal cell palisades. The biopsy on the left side also showed fragments of keratin lamellae, hemorrhagic exudate and mixed inflammatory infiltrate (Figure 4). The patient was instructed to perform irrigation four times a day with 0.9% SF, as well as to maintain strict oral hygiene.

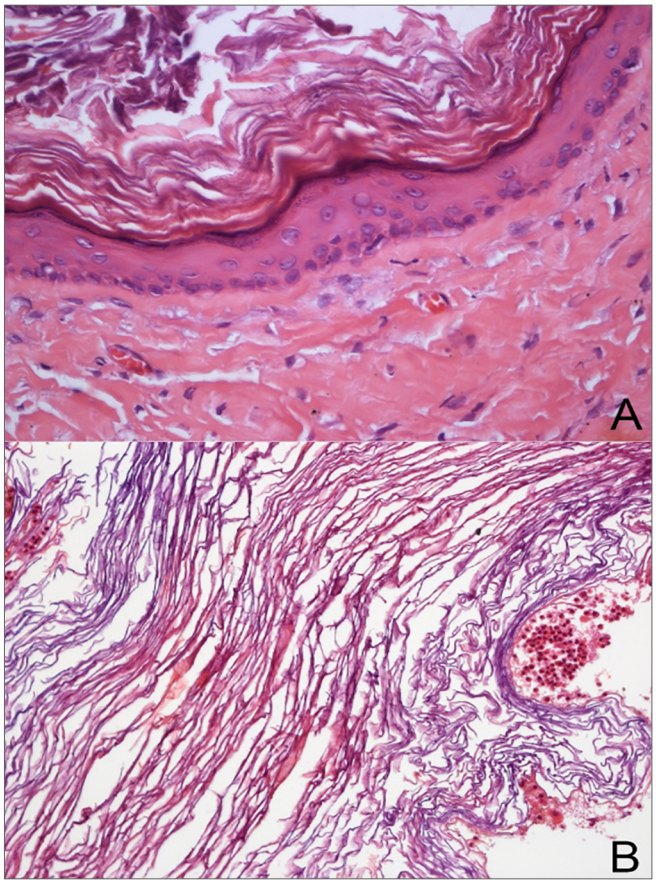

Figure 4. A) Cystic capsule covered by stratified orthokeratinized squamous epithelium B) Fragments of keratin lamellae, hemorrhagic exudate and mixed inflammatory infiltrate.

The total decompression time was 12 months, being monitored periodically through panoramic radiography every 4 months and the drains being repositioned when necessary. After verifying the maximum possible decompression limit, the patient underwent surgery under general anesthesia for enucleation, peripheral ostectomy of the lesion and extraction of elements 18, 28 and 38 (Figure 5). The patient is being followed up and after 1 year it is possible to observe new bone formation in the region (Figures 6 and 7), the patient is without sensorineural disorders and the teeth involved in the lesion are vital, without mobility and without signs of recurrence of the lesion.

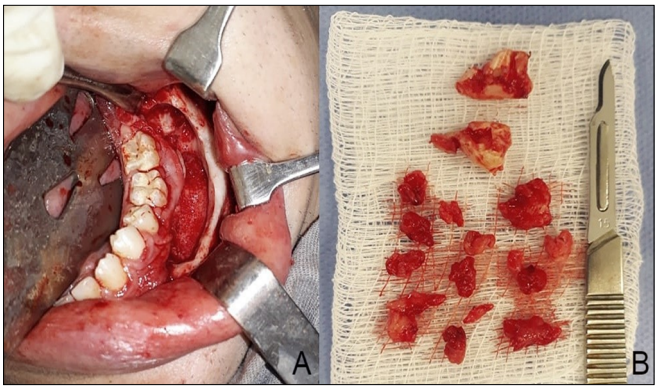

Figure 5 . A) Enucleation and peripheral ostectomy of the lesion. B) Fragments of the cystic capsule and element 38 removed.

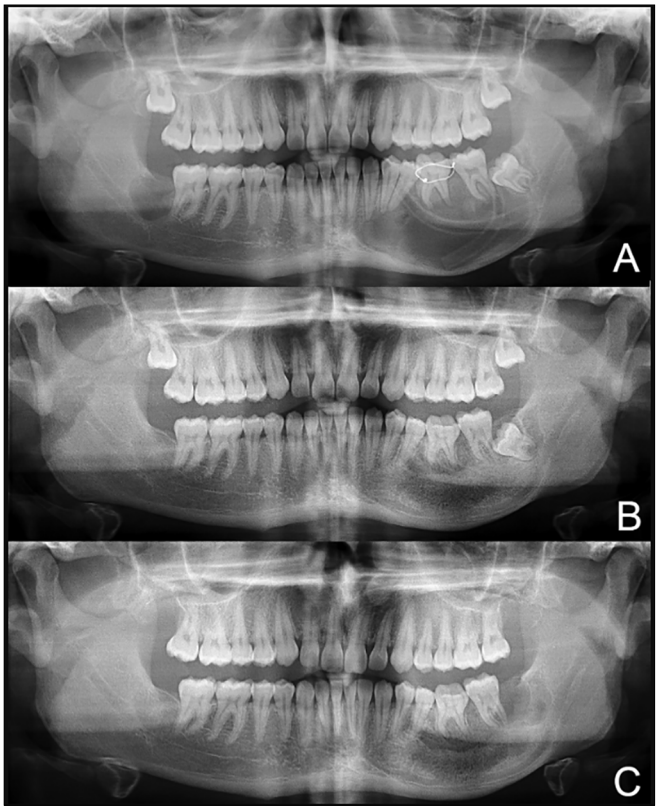

Figure 6. Sequence of panoramic panoramic radiographs. A) Radiography showing the two rigid drains apposed. B) X-ray after 8 months showing bone neoformation. C) Immediate postoperative radiograph of enucleation surgery and extraction of included teeth.

Figure 7. anoramic radiograph after one year of follow-up, with new bone formation and no signs of recurrence. It is possible to observe displacement of the mandibular canal to the basal region.

DISCUSSION

The literature is corroborated in the reported case regarding the location of the lesion and the male gender being the most affected. The posterior region of the mandible is the most affected region, which can be explained by the cellular debris of the dental lamina, which is more active in this area at the age when many patients develop their cysts[7]. The maxilla can also be affected, but less frequently[5,9].

The clinical differential diagnosis involves numerous lesions, but the differential histopathological diagnosis must include the dentigerous cyst, root cyst and the intraosseous epidermoid cyst, however, the absence of skin attachments in the COO is the differentiating feature[4]. The frequency with which COO's occur with unerupted teeth has led some authors to speculate that the orthokeratinized cyst really represents a dentigerous cyst with orthokeratinization, resulting from the reduction of the enamel epithelium[8]. On rare occasions, COO may be associated with a non-vital tooth, offering an erroneous diagnosis of root cyst[6,13]. The peripheral variant of the orthokeratinized cyst is rare, but there are reports in the literature. This variant has a benign and non-aggressive clinical course and is located within the soft gingival tissues[14].

The bilateral and/or multiple form of the lesion is an unusual phenomenon. The most common bilateral lesion is the keratocyst[1] followed by the dentigerous cyst[7]. In the present case, the lesion was bilateral as well as a case reported in the literature[1]. Multiple cases of keratocyst generally occur associated with the Basal Cell Nevoid Carcinoma Syndrome, which does not occur with orthokeratinized cysts[2]. However, the patient reported in the study was investigated about the syndrome, obtaining a negative result, as he did not meet the diagnostic criteria.

Parakeratinized and orthokeratinized mandibular cysts shared a common diagnosis as a keratocyst, however, several studies carried out over the years have shown that these two entities are distinct from each other[1]. COO's do not have satellite cysts like keratocysts, which may contribute to their low recurrence rate. The COO recurrence rate varies between 2.2%[4] and 4%[7]. The reported case was followed up for 1 year without signs of recurrence of the lesion, which confirms the literature in which patients were followed up for 62 months without recurrence[2] and longer follow-up up to 12 years without signs of recurrence of the lesion[15].

Immunohistochemical findings in the COO confirm its non-aggressive behavior when compared to

the keratocyst[1], which occurs in the oral cavity about eight times more common than that[6,7]. Despite its benign nature, the orthokeratinized cyst can displace the neighboring teeth and also the mandibular canal[7], as observed in the case described in which there was root removal and the mandibular canal moved to the basal region.

At the base of the growth and expansion of cysts, there is an increase in osmotic pressure within the cystic lumen, where the increasing hydrostatic pressure improves osteoclastic resorption and contributes to its expansion[16], due to this, the decompression prior to enucleation brings numerous advantages how to save tissues, minimize the likelihood of damage to adjacent structures and avoid the cost of hospitalization[10]. It is possible to notice the decrease in the lesion in the case presented, verified by the absence of tooth mobility and maintenance of the inferior alveolar vasculonervous bundle. The reduction of the lesion through this technique allowed the second enucleation surgery to be performed when the cortical-basal bone had a thickness of at least 3 mm, ensuring sufficient mechanical strength to operate safely[16].

The disadvantages of decompression include the duration of treatment, discomfort, dependence on patient compliance and generally the need for 2 surgical procedures[10,16]. Enucleation following decompression was necessary in 54.5% of patients undergoing decompression and was used more frequently when the cyst diameter exceeded 40 millimeters[10]. Complications are reported more frequently when enucleation is performed as a single procedure for extensive cysts[16], therefore, for the treatment of extensive cysts, prior decompression is a smart choice for preserving vital structures.

CONCLUSION

COO is considered a lesion apart from the odontogenic keratocyst due to its own characteristics, which is reflected in the recent WHO classification that included COO as a distinct cyst. COO has a benign course with few reports of recurrence, but it can reach large proportions in the gnathic bones. Routine panoramic radiographs are of great importance for the diagnosis, since most cases are asymptomatic and can reach large proportions, early diagnosis improves the prognosis. Decompression is an easy procedure that can decrease the morbidity of large cysts and make enucleation a safer procedure for the patient, avoiding damage to blood vessels, nerves and teeth, as presented in the report described.

REFERENCES

1. Pinpalkar RD, Berpande SR, Bhavthankar JD, Mandale MS. Bilateral orthokeratinized odontogenic cyst: a rare case report and review. J Oral Maxillofac Pathol. 2014 May/Aug;18(2):262-66.
2. Uddin N, Zubair M, Abdul-Ghafar J, Khan ZU, Ahmad Z. Orthokeratinized odontogenic cyst (OOC): clinicopathological and radiological features of a series of 10 cases. Diagn Pathol. 2019 Apr;14(1):28.
3. Neville D, Damm DD, Allen C, Chi A. Patologia oral e maxilofacial. 3ª ed. Philadelphia: Saunders; 2009.
4. Kamat M, Kanitkar S, Datar U, Byakodi S. Orthokeratinized odontogenic cyst with calcification: a rare case report of a distinct entity. J Oral Maxillofac Pathol. 2018 Jan;22(Suppl 1):S20-3.
5. Chien CC, Wu YH, Kuo YS, Chiang CP. Orthokeratinized odontogenic cyst in the right maxila. J Formos Med Assoc. 2019 Sep;118(9):1375-7.
6. Rajalakshmi R, Sreeja C, Vijayalakshmi D, Leelarani V. Orthokeratinised odontogenic cyst mimicking periapical cyst. BMJ Case Rep. 2013;2013:bcr2013200883. DOI: https://doi.org/10.1136/bcr-2013-200883
7. Sarvaiya B, Vadera H, Sharma V, Bhad K, Patel Z, Thakkar M. Orthokeratinized odontogenic cyst of the mandible: a rare case report with a systematic review. J Int Soc Prevent Communit Dent. 2014;4:71-6.
8. Crane H, Forno P, Kyriakidou E, Speight PM, Hunter KD. Multiple orthokeratinized odontogenic cysts: a report of two cases and review of the literature. Head Neck Pathol. 2020 Jun;14(2):381-5.
9. Mahdavi N, Taghavi N. Orthokeratinized odontogenic cyst of the maxilla: report of a case and review of the literature. Turk Patoloji Derg. 2017;33(1):81-5.
10. Marin S, Kirnbauer B, Rugani P, Mellacher A, Payer M, Jakse N. The effectiveness of decompression as initial treatment for jaw cysts: a 10-year retrospective study. Med Oral Patol Oral Cir Bucal. 2019 Jan;24(1):e47-52.
11. Oh JS, You JS, Kim SG. Clinical and histomorphometric evaluation of decompression followed by enucleation in the treatment of odontogenic keratocyst. J Dent Sci. 2018 Dec;13(4):329-33.
12. Tabrizi R, Kordkheili MRH, 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). 2019 Sep;20(3):145-51.
13. Servato JPS, Cardoso SV, Silva MCP, Cordeiro MS, Faria PR, Loyola AM. Orthokeratinized odontogenic cysts presenting as a periapical lesion: report of a case and literature review. J Endod. 2014 Mar;40(3):455-8.
14. Boffano P, Gallesio C. Peculiar case of orthokeratinised odontogenic cyst: a peripheral counterpart of the intraosseous entity?. Br J Oral Maxillofac Surg. 2012 Jul;50(5):e75-7.

15. Li TJ, Kitano M, Chen XM, Itoh T, Kawashima K, Sugihara K, et al. Orthokeratinized odontogenic cyst: a clinicopathological and immunicytochemical study of 15 cases. Histopathology. 1998 Mar;32(3):242-51.

16. Bonavolontà P, Orabona GDA, Friscia M, Sani L, Abbate V, Iaconetta G, et al. Surgical management of large odontogenic cysts of the mandible. J Craniofac Surg. 2019 Oct;30(7):e658-61.