Case report

CLINICAL MANAGEMENT OF ODONTOGENIC KERATOCYST: CASE REPORT AND INTEGRATIVE LITERATURE REVIEW

ABSTRACT

Introduction: The odontogenic keratocyst is a common odontogenic cyst which that affects mainly the posterior region of the mandible.

Aims: The aim of this study was to discuss the clinical management of odontogenic keratocyst, based on a case report and integrative a literature review.

Presentation of case: A 34-year-old female visited the Oral Surgery Division with the complaint of discomfort with alignment of her teeth. Radiographic examination showed a well-defined multi-focus radiolucent area in the left posterior region of the mandible with involvement of cortical lingual bone. The incisional biopsy was performed and the diagnosis of Odontogenic Keratocyst was confirmed. Conservative treatment with decompression management was accomplished. The follow-up of 11 months reveled a considerable reduction in lesion size, bone neoformation and mandibular cortical bone consolidation.

Conclusion: The conservative or radical management of odontogenic keratocyst depends on several factors such as location and size of the lesion, involvement of nerves and vessels and area of bone destruction. The choice of treatment must be discussed and defined for each case, aiming at the patient's well-being and the resultativity of the case.

Keywords: Odontogenic keratocyst. Decompression. Odontogenic cyst. Treatment.

1. INTRODUCTION

The odontogenic keratocyst (OKC) is a cystic pathological condition that originates from the dental lamina, representing approximately 11,7% of cystic odontogenic lesions [1].

The OKC can affect the maxilla and mandible [2], however it have highest prevalence in posterior region of the mandible [3].

The World Health Organization has classified the OKC as a neoplasm, due to the aggressive clinical course, the high potential for recurrence, mutations in the PTCH1 tumor suppressor gene, satellite cysts and the association with Gorlin-Goltz syndrome [4]. Due these characteristics the WHO has considered the OKC as keratocyst odontogenic tumor, however, in 2017, WHO published a new classification of head and neck tumors. In this context, the keratocyst odontogenic tumor was moved back to cyst category called odontogenic keratocyst because there was not enough evidence to characterize the lesion as neoplastic [5].

Most keratocysts are diagnosed in routine imaging exams in the dentistry officer. In radiograph exam, the OKC shows as radiolucent image single or multi-focus can be associated or not for impacted teeth [5]. It's often asymptomatic, but when they reach large proportions, it can cause discomfort and facial asymmetry [6, 7].

Typically, histologic findings of OKC show a para-keratinized stratified squamous epithelium, usually 6–8-cell-layer thick, which demarcates a cystic lumen filled with desquamated keratin. There is a well-defined, often palisaded, basal layer of columnar or cuboidal cells, whereas the parakeratotic cells in the upper layers often show a corrugated surface [8].

Various treatment modalities have been reported for OKC. Enucleation, enucleation with cryotherapy, marsupialization, decompression, and resection are example of therapies applied to OKC [9]. The application of Carnoy's solution also was described such as complementary therapy after the removal of cist. Carnoy's solution promotes cauterization and superficial bone necrosis with destroys the epithelial remnants of the dental lamina of the bone margins [10].

Considering the characteristics and clinical heterogeneity of OKC, the aim of this study was to discuss the clinical management of the OKC based on a case report and an integrative literature review.

2. PRESENTATION OF CASE

This study was realized in the School of Dentistry of Faculdades Unidas do Norte Minas, MG, Brazil, and approved by the Research Ethics Committee under protocol no. 2.844.090.

A 34-year-old female visited the Oral Surgery Division with the complaint of discomfort with alignment of her teeth. Medical history was not contributory. On extraoral examination was identified facial symmetry and no dermal lesions. The intraoral examination revealed satisfactory oral health, absence of carious lesions or periodontitis. Dental restorations were observed. Satisfactory mucous membranes and dental crowding in the lower arch were seen. Computed tomographic examination showed a well-defined multi-focus hypodense area in the left posterior region of the mandible with involvement of cortical lingual bone (Figure 1).

After thorough examination of the case the patient was diagnosed with a mandibular cystic lesion. The diagnostic hypotheses of odontogenic keratocyst and multicystic ameloblastoma were suggested.

Aiming to verify the content of the lesion the aspiration puncture was performed. A provisional diagnosis was made as OKC due to presence of whitish liquid and a thick appearance on aspiration, compatible with keratin (Figure 2A). Due to high size of the lesion, mandibular fragility, and risk of inferior alveolar nerve injuries, was performed incisional biopsy and additional decompression of the lesion with a silicone ring (Figure 2B).

The sample biopsied were included in paraffin and stained with hematoxylin and eosin (H&E). Histopathological examination revealed a fragment of cystic capsule of odontogenic origin, partially coated by a few layers of epithelium parakeratinized. The basal layer of the epithelium consisted of columnar cells with palisade morphology and hyperchromatic nuclei (Figure 3). The most superficial layer of the epithelium was observed the presence of keratin. The capsule was composed of loose connective tissue, blood vessels and moderate mononuclear inflammatory infiltrate. These findings leaded to the definitive diagnosis of odontogenic keratocyst.

Computer tomography with 3D reconstruction was performed 11 months after the decompression surgery procedure to evaluate the reduction in lesion size. The findings of computer tomography revealed that that there was a considerable reduction in lesion size, bone neoformation and mandibular cortical bone consolidation (Figure 4).

3. DISCUSSION

Odontogenic keratocyst is a recognized pathological condition worldwide for its aggressive behavior and propensity to recurrence. This context the management of OKC treatment is of fundamental importance for the outcome resolutive pathology.

The origin of odontogenic keratocyst is based on the association between remaining cell remains of the dental lamina with genetic changes in genes PTCH1, increased expression of the PCNA and Ki 67 genes, and the functional dysregulation of tumor suppressor genes [4, 11, 12]. Additionally, the elevated expression of the anti-apoptotic gene B2 (BCL2) cell lymphoma in OKC in comparison with other odontogenic lesions [11]. This set of changes provides proliferation on epithelial remains and formation of the cystic cavity.

The etiological and pathogenesis aspects of OKC reveal that recurrences of lesion may be related to remnants of epithelial remains of the primary lesion during enucleation, or the presence of satellite cysts and microcysts in the tissues underlying [13]. Unfortunately, the cystic coating of most of the OKC is quite and friable, which makes the total enucleation of the lesion extremely difficult, especially in large and multilocular lesions [14, 15]. The use of Carnoy's solution (absolute alcohol, chloroform, acetic acid, and ferric chloride) in the surgical site, after removal of the lesion, may assist in the destruction of epithelial remnants and consequently reduce recurrence rates [14].

Decompression or marsupialization are considered a less invasive technique in the management of OKC that aimed at decreasing the size of extensive lesions prior to enucleation or completely solve the lesions [16]. The main of these approaches are the preservation of important anatomical structures such as inferior alveolar nerve and the prevention of a consequent possible deformity [17]. Marsupialization and decompression have been defined as different surgical techniques. The marsupialization technique, a part of the cyst is removed followed by suturing the lesion's exposed boundaries to the adjacent mucosa exposing the cyst to the oral cavity. In decompression, a drain is placed in the lesion, which maintains communication with the cystic cavity [17].

The decompression or marsupialization leads to decreases the intracystic pressure and may result in bone neoformation in addition to thickening of the cystic epithelial lining. This allows for better surgical management in the total removal of the lesion and consequently reducing the potential for recurrence [13].

In the integrative review about OKC in the last 10 years published in the PubMed database, of the 27 cases analyzed, the majority presented unilocular lesions (n=23), and in 19 studies the lesions were not associated with unerupted teeth. For treatment options, enucleation followed by curettage was the most frequent (n=22), decompression or marsupialization was utilized in the minority of cases (n=5), and the use of Carnoy's solution was observed in 5 cases [6, 10, 18-27]. The data of the articles can be found in Table 1.

The treatment modalities for OKC have differing recurrence rates and include enucleation [28], enucleation with cryotherapy [29], enucleation with application of Carnoy's solution [30], marsupialization [16], decompression [31] and resection [32]. Interestingly, the

minority of cases analyzed used the marsupialization o decompression techniques. The literature highlights the benefits of use of the marsupialization o decompression maneuver in extensive lesions, as well application of Carnoy 's solution in the sugery area after cyst removal [13, 14]. Such procedures when well indicated and well performed can reduce the potential for recurrence of the lesion.

In the present case, and to minimize the risk of injuries of noble structures such as inferior alveolar nerve and reduce the risk of pathologic fracture of the mandible, the decompression was performed as the primary treatment. The decompression aimed at gradually reducing the lesion for subsequent enucleation of the cyst in a less traumatic way [33]. This technique can promote epithelial differentiation and modify expression of cytokeratin 10 expression from positive to negative, making the cystic epithelium more similar to the epithelium of the oral cavity and improve the sugerycal management of cystectomy step [24, 34].

The literature points out that despite the size of the lesion, there is a preference to opt for surgical treatments such enucleation or resection with or without the application of Carnoy's solution or cryotherapy in hospital environment under general anesthesia [23]. Meanwhile, on the other hand, it is known that there are regions where access to dental service of high complexity is restricted placing decompression or marsupialization as a viable therapeutic for the management of OKC.

The percentage of recurrence of odontogenic keratocyst can reach 62,5%, demonstrating aggressive growth pattern despite being benign [7]. In this context, more experienced professionals with highly complex resources opt for more radical treatments such as cystectomy with therapy adjuvant such as application of Carnoy's solution. The application of Carnoy's solution can destroy the remaining cystic epithelial cells and generate a superficial bone necrosis reducing the possibility of recurrence [6, 10, 20].

Given the above, it is possible to state the clinical management of odontogenic keratocyst depends on several factors, among them: the location and size of the lesion, the experience of the professional, and the means available for treatment modality. The option for more radical or conservative techniques should be debated and well defined for each clinical case, always aiming for the patient's well-being and reluctivity of the case.

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ETHICAL APPROVAL

This study was realized in the School of Dentistry of Faculdades Unidas do Norte Minas, Brazil, and approved by the Research Ethics Committee under protocol no. 2.844.090.

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LEGEND OF FIGURES

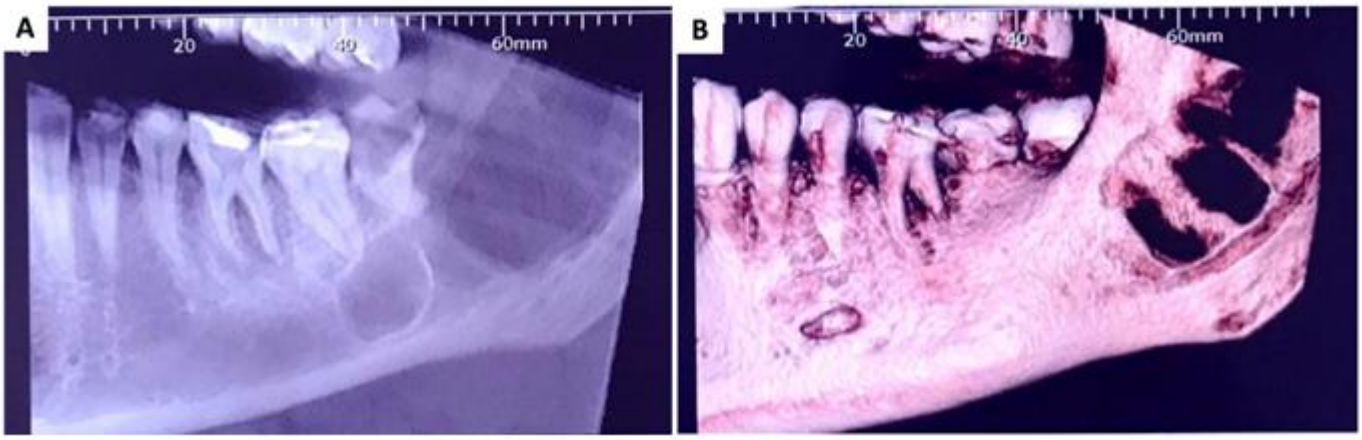

Figure 1: A) Computed tomographic examination showed a well-defined multi-focus hypodense area in the left posterior region of the mandible with involvement of cortical lingual bone. B) 3D reconstruction of the left posterior region of mandible.

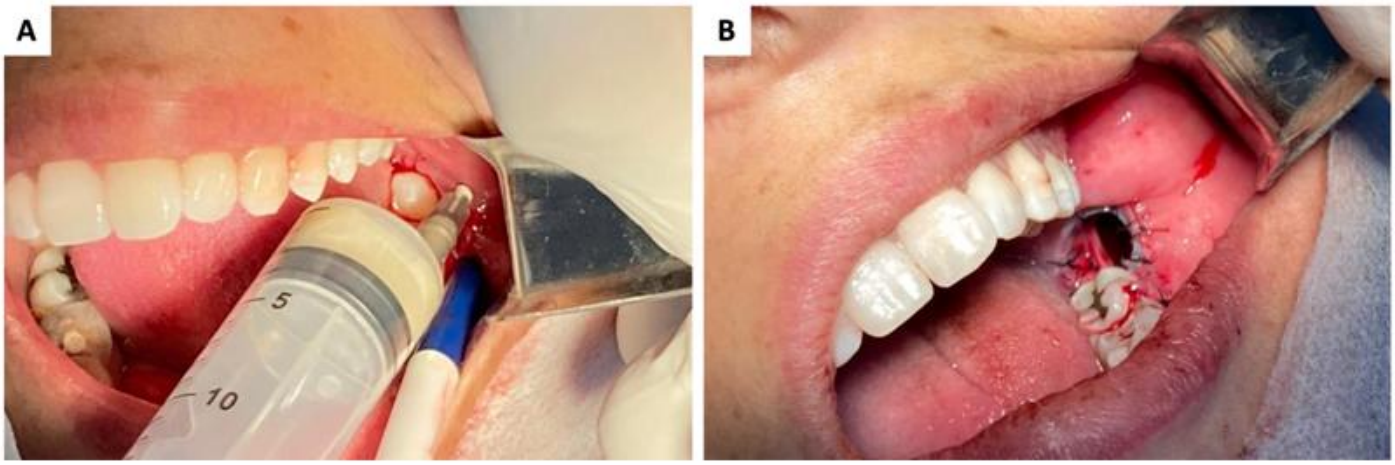

Figure 2: A) Aspiration puncture showed the presence of whitish liquid. B) Decompression of the lesion with a silicone ring.

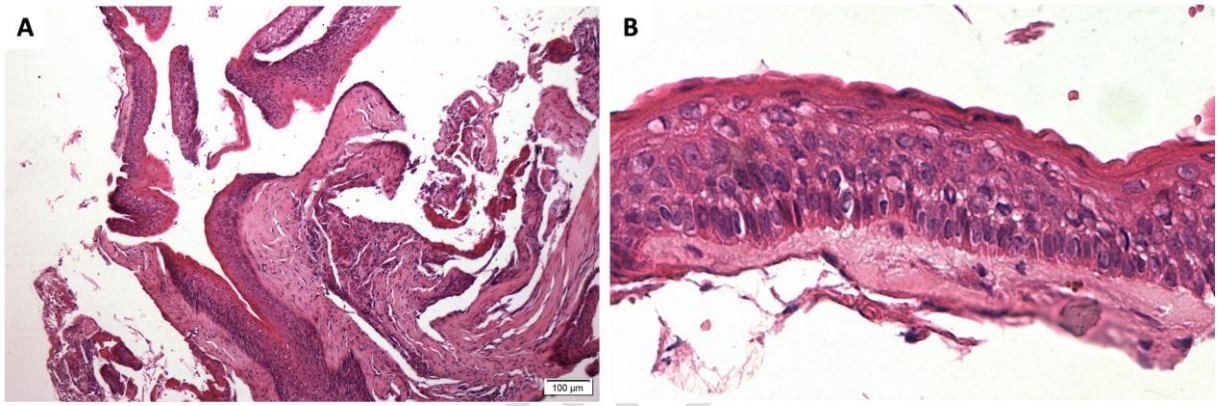

Figure 3: Histopathological examination stained with hematoxylin and eosin. A) 10x and B) 40x.

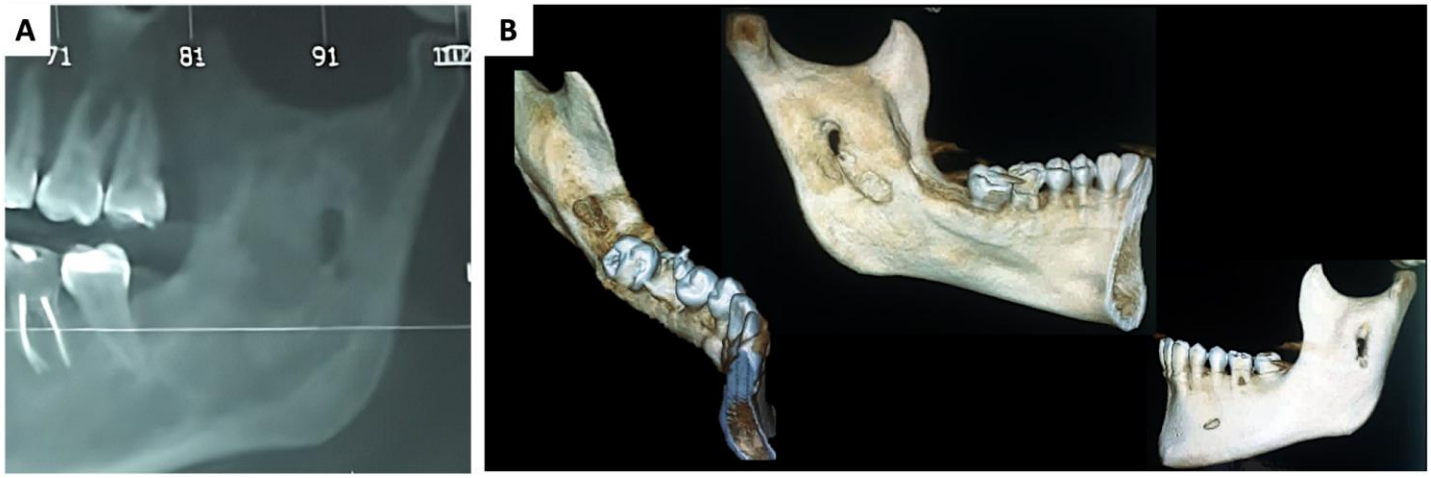

Figure 4: Computer tomography examination after decompression procedure showed a considerable reduction in lesion size. A) Left posterior region of the mandible. B) 3D reconstruction of mandible.

Table 1: Data from the articles included in the study.

Autor/Year	Gender	Age	Location	Included tooth	Aspect	Size	Marsupialization or Decompression	Enucleation	Carnoy's solution
Matijevic, S., et al. (2012)	Male	52	Mandibular body and branch	Yes	Single	4.0 cm	Yes	Yes	Yes
Priya, K., et al. (2014)	Male	18	Left side maxilla	No	Single	1,0 cm	No	Yes	No
	Female	20	Right side maxilla	No	Single	5 cm	No	Yes	No
	Female	16	Left side maxilla	No	Single	6 cm	No	Yes	No
	Male	20	Left maxilla	No	Single	5 cm	No	Yes	No
	Male	16	Right side maxilla	No	Single	1.5 cm	No	Yes	No
Gupta, R. K., et al. (2016)	Male	22	Body and mandibular branch on the left side.	Yes	Single	> 7 cm	Yes	Yes	No
Arshad F. 2016	Male	20	Mandible/region near the apexes of teeth 13 a nd 23	Yes	Multi-focus	2,0 cm	No	Yes	No
Gopalkrishna, A. A., et al. (2018)	Female	82	Mandibular body on the left side	Yes	Single	2.5 cm	No	Yes	No
Shetty, P., et al. (2018)	Female	22	Posterior maxilla on the left side	Yes	Single	> 7 cm	No	Yes	No
Rodrigues-Fernandes, C. I., et al. (2018)	Male	14	Anterior mandible	No	Single	< 7 cm	No	Yes	No
Khan, A. A., et al. (2019)	Female	35	Bilateral body and mandibular branch	No	Single	> 7 cm	Yes	Yes	No
Polak, K., et al. (2019)	Not reported	53	Anterior region of mandible	No	Single	> 7 cm	Yes	Yes	No
Radia, S., et al. (2019).	Male	10	Left side maxilla.	Yes	Single	> 7 cm	No	Yes	Yes
Sheethal, H. S., et al. (2019).	Female	15	Posterior maxilla the left side invading the sin us	Yes	Single	> 7 cm	No	Yes	No
Telagi, N. and B. M. Ahmed (2019).	Male	40	Mandibular body on the right side	No	Multi-focus	6 cm	No	Yes	No
Vijayarangan, S. (2019)	Female	19	Left side mandibular angle	No	Single	> 7 cm	Yes	Yes	Yes
	Male	39	Mandibular body	No	Multi-focus	> 7 cm	No	Yes	No
	Female	35	Element maxilla 14 to 24.	No	Single	> 7 cm	No	Yes	No
	Male	29	Mandibular body	No	Single	> 7 cm	No	Yes	No
	Female	35	Posterior mandible	Yes	Single	> 7 cm	No	Yes	Yes

Firth, N., et al. (2020)	Female	57	Between teeth 42 and 43	No	Single	1.5 cm	No	Yes	No
Goto, M., et al. (2020)	Male	21	Maxillary sinus and left side sphenoid bone	No	Single	> 7 cm	No	Yes	No
Vallejo-Rosero, K. A., et al. (2020)	Female	67	Anterior mandible	No	Single	> 7 cm	Yes	Yes	Yes
Prashanth, B. R., et al. (2020)	Male	29	Anterior region of mandible	No	Single	> 7 cm	No	Yes	No
Roman, C. R., et al. (2022)	Male	19	Posterior region of the left side jaw	No	Single	4 cm	No	Yes	No
Lohokare, A. U., et al. (2022)	Male	34	Right mandibular body	No	Multi-focus	3 cm	No	Yes	No