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Research Article

Managing Odontogenic Keratocysts: An Enygma? Considerations to Choose the Correct Approach

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Abstract

Purpose: The aim of this study is to propose a protocol for the appropriate management and treatment of odontogenic keratocysts.

Materials and Methods: A longitudinal descriptive study on patients with odontogenic keratocysts surgically treated in the Oral and Maxillofacial Surgery Unit of the University Hospital of Maracaibo, Venezuela during the period between January 2015 and January 2020. Using a management algorithm, the treatment modality for each patient was described based on clinical, radiographic characteristics and biological behavior of the lesion.

Results: 10 patients were included in the study. The average age was 44.7 ± 21 years and 60% of the patients were female. All lesions were located in the mandible, 2 of the lesions had radiographic evidence of cortical perforation. The average radiographic diameter of the lesions was 6.3 ± 2.2 cm. Regarding the surgical variables, 7 patients received decompression + residual cystectomy, 1 patient enucleation and curettage and 2 patients received marginal or en-bloc resection.

Conclusion: It is necessary to unify different variables, like biological behavior and surgical and radiographic characteristics, since they are widely scattered in the literature and thus being able to protocolize the management of these lesions to obtain better results and decrease the recurrence rate.

Keywords: Odontogenic Keratocyst; Decompression; Enucleation; Resection; Recurrence

Introduction

The Odontogenic Keratocyst (OKC) is a benign lesion recently considered as an odontogenic cyst in the present World Health Organization (WHO) classification of Head and Neck tumors of 2017, mainly based on its clinical and histopathological features [1]. They have an embryonic origin from the cellular remnants of the dental lamina. This cyst is aggressive, with a slow growth pattern typically in the anteroposterior direction, covering the medullary spaces of the bone and with a high recurrence rate [2,3]. These lesions could be diagnosed at any age stage; however, 60% of cases occurs in individuals between 10 and 40 years of age [4].

To confirm the diagnosis of OKC, it is necessary to perform a complete intra and extraoral clinical evaluation, thorough radiographic analysis and pathological examination to establish an accurate final diagnosis [4]. Radiographically, the lesion's appearance is of a unilocular or multilocular radiolucency, most often surrounded by smooth or scalloped margins with sclerotic borders and defined edges. Larger lesions are often multilocular [5,6].

Different treatment modalities have been described, ranging from conservative techniques to radical surgeries. Throughout the literature, different judgments (either conservative or radical) are applied for the right treatment of OKCs, but they are based in a specific characteristic in an isolated way without unifying criteria to choose the best treatment depending on all the variables in which this pathology is displayed.

Purpose of the Study

The purpose of this study is to propose a protocol for the appropriate management and treatment of OKCs with unified judgment according to its characteristics based on our experience.

Materials and Methods

Study design and sample

A longitudinal descriptive study on patients with odontogenic keratocysts surgically treated in the Oral and Maxillofacial Surgery Unit of the University Hospital of Maracaibo, Venezuela during the period between January 2015 and January 2020. OKCs were evaluated through clinical and imagenological examinations, such as panoramic x-rays and CT scans.

Inclusion criteria were OKCs diagnosed in our unit. Patients diagnosed with Gorlin-Goltz syndrome, lesions that had been previously treated at other centers, patients with irregular follow-ups and patients with absent or incomplete medical data were excluded from the study.

This study was approved by the Institutional Review Board of our institution.

Study variables

The heterogeneous variable groups were categorized as demographic, radiographic and surgical. The demographic variables included age and gender. The radiographic variables included location of the lesion, association with impacted teeth, evidence of cortical perforation, locularity (uni or multilocular), soft tissue affectation and size of the lesion. Surgical variables included 3 treatment modalities: 1. Enucleation and curettage. 2. Decompression and subsequent residual cystectomy. 3. En-bloc/marginal resection.

The radiographic size of the lesion was obtained using the longest diameter of the lesion in any direction through preoperative panoramic x-rays. Likewise, the presence of recurrence, time of follow-up and presence of complications were registered after the surgery.

Treatment protocol

The treatment modality was chosen based on the clinical and radiographic characteristics of the lesion, as well as its biological behavior (Figure 1).

Figure 1: Keratocyst located on the condyle.

OKCs with less than 3 cm in diameter that did not involve the inferior alveolar nerve were treated with enucleation and curettage. Larger lesions were treated using decompression and residual cystectomy or marginal/en-bloc resection.

The treatment modality based on decompression with residual cystectomy was applied on collaborative patients with OKCs greater than 3 cm in diameter involving vital anatomical structures

who were able to obtain periodic radiographic controls during the decompression time. At the time of the incisional biopsy, a fenestrated polyethylene tube was placed inside the lesion and fixated using non absorbable suture. Instructions were given to the patient in order to irrigate through the drain twice a day using saline water and periodic radiographic controls were obtained to assess the size of the lesion. A monthly follow-up regime was established, including clinical and radiographic evaluation and the drain tube was adjusted accordingly in order to allow for bone formation inside the cystic cavity.

The definitive secondary surgery consisting of enucleation and curettage was performed when there was enough bone to prevent damage to the adjacent vital structures, when the radiolucency decreased by approximately 2 cm in diameter, when the OKC was not in contact with the inferior alveolar nerve or when the drain tube was ejected. If an adjacent tooth was still involved or interfered with the correct removal of the cyst, it was removed. At the same time as the enucleation and curettage of the lesion, the overlying mucosa was excised to remove the daughter and satellite cysts between the mucosa and the OKC.

En-bloc/marginal resection was chosen for OKCs greater than 3cm in diameter that met the following criteria:

- Cortical perforation and/or soft tissue affectation
- Recurrence after conservative treatment
- Aggressive behavior
- Non collaborative patients
- Malignant transformation within the cyst
- Lesions located in sites where conservative treatment was difficult (e.g. mandibular condyle) (Figure 1).

For all treatment modalities, a follow-up schedule was applied using panoramic x-rays at the first, third and sixth month during the first year. Then, every 6 months during the second year and finally, one panoramic x-ray annually for the next 5 years on asymptomatic patients.

Statistical analysis

For the interpretation of the results, the data were recorded retrospectively on standardized collection forms. A database was built with Microsoft Excel for statistical analysis. Descriptive statistics were calculated for all study variables.

Results

During the period of the study, 11 patients with OKCs were evaluated and treated in our institution; however, 1 patient was excluded due to inconsistent attendance to follow-up appointments. The final sample was comprised of 10 patients without any previous treatment. The average age was 44.7 ± 21 years, ranging from 11 to 72 years. 60% of the patients were female. All lesions (100%) were located in the mandible, 6 lesions (60%) were associated with impacted teeth, 2 lesions (20%) presented radiographic evidence of cortical perforation and 2 lesions (20%) presented soft tissue affectation. The average radiographic diameter was 6.3 ± 2.2 cm and 6 lesions (60%) were multilocular.

Regarding the surgical variables, 7 patients (70%) received conservative treatment (decompression + residual cystectomy), 1 patient enucleation and curettage (10%) and 2 patients (20%) received marginal or en-bloc resection as a first treatment.

The average follow-up period was 48.9 ± 14.5 months. Only 1 patient (10%) presented recurrence of the lesion after 8 months, which was treated with decompression with subsequent residual cystectomy. Regarding complications after the definitive surgery, 1 patient (10%) had a mandibular fracture, 2 patients (20%) presented inferior alveolar nerve transient paresthesia and 1 patient (10%) presented a granuloma, associated to the extraoral approach as shown in supplemental table 1.

Descriptive Statistics	
Sample Size (n)	
Patients	10
OKCs	10
Demographic Variables	
Age	44.7 ± 21 years
Gender Male Female	 4 (40%) 6 (60%)
Surgical Variables	
Enucleation and Curettage	1 (10%)
Decompression + Residual Cystectomy	7 (70%)
Marginal/En-bloc Resection	2 (20%)
Radiographic Variables	

Location of the lesion Mandible	10 (100%)
Association with impacted teeth Yes No	6 (60%) 4 (40%)
Greater Diameter	6.3 ± 2.2 cm.
Evidence of Cortical Perforation Yes No	 2 (20%) 8 (80%)
Locularity Multilocular Unilocular	 6 (60%) 4 (40%)
Other Variables	
Recurrences	1 (11.1%)
Time of recurrence	8 months
Follow-up average time	48.9 ± 14.5 months
Complications Mandibular fracture Granuloma IAN Paresthesia	 1 (10%) 1 (10%) 2 (20%)

Table 1: Study variables distribution.

Discussion

The OKC is an odontogenic cystic lesion that demonstrates the behavioral characteristics of a benign neoplasm and has a propensity to recur after surgical treatment. Different surgical strategies and protocols have been proposed to facilitate complete removal of the lesion and minimize recurrent or residual disease. A greater understanding of the clinical behavior of the OKC has prompted more meticulous surgery with or without peripheral ostectomy, tissue fixation methods, soft tissue excision, radical treatment when necessary and more meticulous follow- up. The optimum treatment modality has yet to be realized [7,8].

In the present study, we performed a retrospective analysis of a cohort of OKC cases, using a standardized protocol. OKCs were found to affect patients of a wide age range, with an average patient age of 41.8 years, similar to other studies. On the other hand, the literature reports that it is found mostly in men and in our study, women were affected in 55.6% of cases. The mandibular molar region was the most affected site, agreeing with the literature, but the locularity differs: the multilocular appearance was more common than the unilocular appearance in our study [5,9]. Regarding the size of the lesion, we selected the largest diameter of the lesion in any dimension estimated by panoramic radiographs taken before treatment, similar to the studies of Anavi., *et al*. [10] and Madras and Lapointe [11] where they measured the maximum diameters of the lesions in 2 dimensions on panoramic radiographs as well as the standard lesion area index (SLAI) using the maximal vertical length (in cm) multiplied by the maximal horizontal length (in cm). They found this method to be both practical and useful, and it is probably applicable for use in daily practice to plan stages before surgery and for follow-up.

Although experimental molecular pathology studies have brought some insights into the biological profile of the OKC, the issue of appropriate treatment for the lesion remains a subject of controversy. Different treatment modalities generally classified as conservative or aggressive are reported. “Conservative” treatment usually includes enucleation and/or marsupialization, and “aggressive” treatment includes enucleation accompanied by adjunctive therapies or resection [4,5].

Different adjunctive treatment methods like Carnoy’s solution and 5-Fluorouracil have been proposed to eradicate the recurrence rate of OKCs [12,13]. Sharif., *et al.* [14] compared enucleation of OKCs alone versus enucleation and adjunctive treatment with Carnoy’s solution and found benefit in the use of adjunctive treatment. However, even after using Carnoy’s solution, microcysts and epithelial islands were always seen in the overlying attached mucosa of OKCs and so recurrence took place. In our study, after the enucleation and curettage of the lesion, the overlying mucosa was excised to remove the daughter and satellite cysts between the mucosa and the OKC; this may eliminate newly developing cysts from epithelial islands or microcysts, which are found in approximately 50% of the cases [5,9,15].

Variables such as location and size of the lesion, evidence of cortical perforation, soft tissue involvement and history of previous treatment are significantly important to determine the treatment type [6,8,16,17]. Kolokythas., *et al.* [4] used a treatment modality based on clinical or radiographic evidence of cortical perforation and subsequent soft tissue involvement and history of previous recurrence of the same lesion. Cases associated with any of these findings were considered to have a more aggressive behavior and were treated with resection and/or enucleation with peripheral ostectomy. For the rest of the cases, when the patient demonstrated willingness to comply with frequent follow-ups and to participate in treatment, a more conservative approach was taken, with decompression through a silastic drainage tube.

In our study, these variables were taken into account; however, we included others such as biological behavior of the lesion, locularity, radiographic size of the lesion and associated impacted teeth. From the aforehead mentioned, impacted or associated teeth were found in our study in approximately 60% of OKCs and injuries with involved teeth were more likely to recur. Farias Cunha., *et al.* [9] and Varinauskas., *et al.* [18] agree in their studies that the insinuation of the OKC into the root of a tooth may increase the risk of recurrence. Therefore, they suggest the extraction of the affected tooth in cases of root involvement by the lesion [9,18]. As in our point of view, preservation of the tooth in such cases may compromise proper enucleation of the cyst and therefore it must be extracted.

The literature supports that larger lesions were more likely to undergo decompression with residual cystectomy or enucleation with peripheral ostectomy, whereas smaller lesions were more likely to undergo enucleation without peripheral ostectomy [4,15,19,20]. Sigua-Rodriguez., *et al.* [5], implemented a treatment modality using a 1-step protocol (OKCs smaller than 3 cm in diameter that did not involve the inferior alveolar nerve were treated by enucleation and curettage with peripheral ostectomy) or a 2-step protocol (larger lesions were treated using decompression and then, after lesion reduction, enucleation with curettage and peripheral ostectomy). Because of their 27-year experience in the conservative treatment of OKCs, the authors believe that this type of treatment does not need for partial or total resections [5].

This differs from our study, because there are situations where total or partial resections could be justified, such as cystic lesions with multiple recurrences, large multilocular cysts with severely thinned out bone or multiple perforations, cases of malignancy transformation within the cysts, patients with poor compliance to follow-up appointments or lesions that have already recurred after conservative treatment to the next anatomic boundary [15-17,21].

Regarding the surgical treatment for the OKC, it is known that there are multiple postoperative complications that can occur. Infection, recurrence, paresthesia, and pathologic fractures occur more frequently. Lee., *et al*. [22] design a study of 249 patients who underwent surgical removal of intrabony cysts. Outcome variables were postoperative complications, infection, and recurrence. There were 59 postoperative infections in their study, differing from ours, in which there was no postoperative infections. They also reported 14.5% of paresthesia after treatment and 1.5% of the cases had fractures after cyst enucleation. These findings are similar to our study, where these complications were evidenced.

OKCs have a considerable rate of recurrence, which varies significantly according to the type of management applied. Studies published over the last three decades have reported recurrence rates after initial treatment with various procedures that ranged from 2.5% to 62.5% [23,24]. Chrcanovic., *et al*. [24] in their analysis study of 6427 cases, suggested that the highest probabilities of recurrence of OKCs were presented by marsupialization (28.7%) and enucleation alone (22.5%), and the lowest probabilities of recurrence were presented by enucleation plus the use of adjuvant therapies (5.3%), decompression followed by enucleation (11,3%) and marginal/segmental resection (2.2%). However, in the present study, the only patient who presented a recurrence was in fact treated with decompression and subsequent enucleation.

The literature does not present any consensus on a uniform treatment plan for OKCs and does not provide adequate evidence for determining which modality is most effective in lowering morbidity or preventing recurrence. However, in our study, despite presenting a limited sample, the recurrence of the OKCs was evident only in one patient with a mean follow-up of more than 2 years. Therefore, it is suggested, according to our results, to consider the different unified variables hereby described (scattered in the literature until now) for decision-making at the time of treating the OKC as shown in supplemental algorithm 1.

Conclusion

A myriad of surgical strategies have been proposed to facilitate complete removal of odontogenic keratocysts and minimize recur-

rent or residual disease. It is necessary to unify different variables, like biological behavior and surgical and radiographic characteristics, since they are widely scattered in the literature and thus being able to protocolize the management of these lesions to obtain better results and decrease the recurrence rate.

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