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Case Report

Dental Science and Oral Maxillofacial Issues

Odontogenic Keratocyst: A Diagnostic and Surgical Challenge

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Received Date: October 08, 2024; Accepted Date: October 25, 2024; Published Date: November 11, 2024

Citation: Gauri Bargoti, Vidya Iyer, Amit Gupta (2024). Odontogenic Keratocyst: A Diagnostic and Surgical Challenge, J Dental Science and Oral Maxillofacial Issues.1(2):6, **DOI**: DSOMI-24-CA-006.

Abstract

The odontogenic keratocyst (OKC) is an epithelial developmental cyst. Several theories of the development of the OKC have been presented. According to one theory, OKCs develop from the remains of the dental lamina. These lesions have posed a great difficulty for the surgeons and pathologists. The surgeons since the beginning have been experimenting with OKC treatment modalities to find a way of treating it without any recurrences. This case series comprises of four such cases of OKC, unique in their character, making their diagnosis and treatment plans very different from each other.

Key words: odontogenic keratocyst; cornoy's solution; cryotherapy; decompression; enucleation.

Introduction:

The odontogenic keratocyst (OKC) is an epithelial developmental cyst. Several theories of the development of the OKC have been presented. According to one theory, OKCs develop from the remains of the dental lamina [1]. According to other theories, OKCs emanate from the basal cell layer of the oral mucosal epithelium or from the stellate reticulum of the enamel organ [2].

The aggressive nature, relatively high recurrence, and range of treatment options of the Odontogenic Keratocyst (OKC) are well documented. Since being first identified by Mikulicz in 1876, OKC has undergone conceptual and terminological alterations. The World Health Organization Classification (WHO) gave it the term "keratocystic odontogenic tumour" in 2005 after observing biological and genetic anomalies consistent with neoplastic growth. Later in 2017, it underwent another renaming and was given the odontogenic cysts classification once more.

Once a diagnosis of OKC is made, surgical intervention is required, given the expansile and locally aggressive nature of this type of cyst, as they can cause cortical expansion, displacement and resorption of teeth. They have also been shown to cause sensory loss due to compression [3].

These lesions have posed a great difficulty for the surgeons and pathologists. The surgeons since the beginning have been experimenting with OKC treatment modalities to find a way of treating it without any recurrences. On the other hand, eminent pathologists have been struggling to determine the true nature of OKC so that a definite line of action can be devised [4] .

This case series comprises four such cases of OKC, unique in their character, making their diagnosis and treatment plans very different from each other.

Case Series

- The first case was of a 38 years old female who reported to the Department of Oral and Maxillofacial Surgery, ITS-CDSR, Muradnagar with a swelling in upper left back region of the face. She got her CBCT done from a hospital where the Doctor gave a diagnosis of residual cyst. There was a history of extraction from the same region done around 8 years ago. When the patient reported to our Department reluctant to undergo biopsy or any other treatment under Local Anaesthesia, hence, direct enucleation was planned under General Anaesthesia considering the lesion to be a residual cyst. During the procedure, an incision was given in the maxillary vestibule from the second premolar till the tuberosity region and a full thickness mucoperiosteal flap was raised followed by extraction of teeth 26 and 27 and enucleation. A cheesy white discharge was encountered from the cavity pointing towards OKC. Complete enucleation was done till sound bone was visible followed by wound closure and the samples were sent for histopathological study.
- The second patient was a 16 years old male who reported to ITS-CDSR, Muradnagar for his Orthodontic treatment. During radiographic examination, there was an accidental

finding of a cystic lesion around impacted right upper canine extending from 13 to 16. Under general anaesthesia an incision was given in the maxillary vestibule from 12 to 17 region and a full thickness mucoperiosteal flap was raised followed by enucleation. Primary closure of the cavity was not done and an opening was left. This was followed by cryotherapy with liquid nitrogen.

- The third case involved a 46 years old male patient who reported to our department with a complaint of pain in lower right back tooth for the past 3 months along with pus discharge which stopped with medication. Radiographically, a cystic lesion was seen extending from 43 to 47 region. Again, under general anaesthesia an incision was given from 42 to 48 region and a mucoperiosteal flap was raised. Enucleation was done followed by treatment with Carnoy's solution.
- The fourth case was a 16-year-old female who reported to our department with pain in left back tooth for 1 year. After radiographic assessment, an incisional biopsy was done. When the cavity was opened, grey particles like mercury were recovered from it. The patient informed us that the particles were placed by the person previously treating her. The biopsy report indicated that it was an Infective OKC. Enucleation under general anesthesia was done, followed by iodoform dressing.

Discussion:

There is an ongoing debate regarding the biological behavior and classification of the OKC since the first edition of the World Health Organization (WHO) atlas on the histological typing of odontogenic tumors and jaw cysts [5]. In 1971, the WHO classified this lesion as an odontogenic cyst [6] and maintained it in 1992 [7], whereas in 2005, it was classified as a Keratocystic Odontogenic Tumor (KCOT) [8] , but reclassified into OKC again in 2017 (El-Naggar et al., 2017)[9] . OKC's grow in the medullary space of the mandible or maxilla with no or little expansion of the cortical bone. OKCs of the maxilla are particularly rare with less than 1% of cases reported in the literature [10].

The first case presented in our series outlines the management and findings of a solitary OKC in the maxilla which was treated by enucleation alone.

Our second case was treated with cryotherapy using liquid nitrogen. Liquid nitrogen has a unique ability to devitalize bone in situ while leaving the inorganic framework untouched, cryotherapy has been used for a number of locally aggressive jaw lesions, including odontogenic keratocyst, ameloblastoma, and ossifying fibroma [11]. In a study by Schmidt et al, Twenty-three of the 26 patients (88.5%) had no evidence of clinical or radiographic recurrence after treatment by enucleation and cryosurgery [12].

The third case in our series was treated by enucleation followed by chemical cauterization with Carnoy's solution. Given the concern regarding recurrence, Carnoy's solution had been proposed as an adjuvant treatment post-marsupialization. The solution itself is made up of chloroform, absolute ethanol, glacial acetic acid and ferric chloride. A systematic review of Carnoy's solution used in treating OKC's categorized the evidence as grade C [13].

The majority of patients are of a younger generation and the preferred course of treatment would be a conservative enucleation as opposed to an aggressive resection that mutilates patients. The role of the anti-aging gene Sirtuin 1 may be relevant to the treatment of OKC since Sirtuin 1 is a critical regulator of p53 that controls OKC lesion formation. The role of Sirtuin 1 activation versus inhibition may be important to the treatment of OKC in these patients and be relevant to surgeons and pathologists [14, 15, 16].

In Browne's series, 3 different treatment methods were evaluated: marsupialization, enucleation and primary closure, and enucleation and packing open. There was no correlation between treatment method and the rate of recurrence. The overall rate of recurrence was almost 25% [17]

Conclusion

In order to detect OKCs at an early stage, protocols should be in place that call for dentists to do a diagnostic OPG for patients on their initial visit, followed by imaging being done once every three years for a given person. Treatment methods include performing a biopsy while completely aspirating the fluid inside to allow the epithelium to thicken. Given that the majority of these patients are of a younger generation, the preferred course of treatment would be a conservative enucleation as opposed to an aggressive resection that mutilates patients. None of these recommendations change the reality that periodic radiographic examinations must be accompanied with long-term follow-ups.

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