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International Journal of Head and Neck Pathology

Case Report

Diagnostic Dilemma of Odontogenic keratocyst Mimicking a Dentigerous Cyst: A Case Report

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How to cite: Georgia Benitha., Diagnostic Dilemma of Odontogenic keratocyst mimicking a Dentigerous Cyst: A Case Report. Int J Head Neck Pathol 2023; 6(2):6-11.

DOI: https://doi.org/10.56501/intjheadneckpathol.v6i2.978

Received:08/08/2023 *Accepted:19/08/2023* *Web Published:25/08/2023*

Abstract

Odontogenic keratocysts (OKCs) are aggressive cysts with high recurrence rates and potential for infiltration. This case report presents a rare follicular OKC mimicking a dentigerous cyst in a 63-year-old male. Clinical suspicion and thorough histopathological examination were crucial for differentiating the two, leading to appropriate surgical management and ultimately preventing recurrence.

Keywords: Follicular OKC, Dentigerous cyst, Recurrence, aggressive cyst

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INTRODUCTION

The odontogenic keratocyst (OKC) is a locally aggressive developmental odontogenic cyst with a high relapse rate. [1][2][3][4] According to the WHO's updated classification of head and neck tumors, published in 2005, the term "KCOT" is appropriate because it reflects the tumor's neoplastic nature, and it is defined as "a benign uni-or multicystic intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and the potential for aggressive, infiltrative behavior."

The mandible is the most often presenting location. [5] It is a cystic jaw lesion that is locally aggressive and has a high rate of growth and recurrence. [6] There is a reported 0% to 100% recurrence rate. [7] It affects more men than women and has a bimodal age distribution, with the first peak appearing in the second and third decades and the second peak appearing in the fifth decade or beyond.

Because of its pathognomonic microscopic traits, potential for aggression, and close association with nevoid basal cell carcinoma syndrome (NBCS) or Gorlin-Goltz syndrome, this cyst stands out among odontogenic cysts. OKCs are made up of a cystic region lined by a uniform parakeratinized squamous epithelium with five to ten cell layers and a distinctive basal layer. The cystic area contains desquamated keratin.

Generally, the interface with the surrounding connective tissue is flat, albeit small satellite cysts and basal layer budding are possible [3]. Compared to other odontogenic cysts, this cyst has increased mitotic activity [4].

One variant of OKC is the follicular keratocyst, which is characterized as a cyst that surrounds the crown of an unerupted tooth and is linked to the tooth's neck while exhibiting the characteristic histology of an OKC. A preexisting keratocyst may give rise to a follicular keratocyst. When viewed using radiographic imaging, it resembles a dentigerous cyst. 22–44% of all cases of OKC that have been documented are related to it.

Differentiating a dentigerous cyst from an extrafollicular OKC is crucial since the former is less aggressive than the latter. This case report highlights a rare case of odontogenic keratocyst associated with an impacted mandibular molar tooth and its surgical management.

Case report:

A 65-year-old male patient came to the Department of oral and Maxillofacial Surgery with a complaint of pain and swelling in relation to the right lower jaw region for two months. The pain was dull, aching persistent and the swelling gradually increased to the present size.

On general examination, the patient was well-oriented with time and surroundings. On extraoral examination, there is evidence of soft to firm swelling with mild tenderness and no increase in temperature. No pus discharge or pulsation was felt. Intraoral examination revealed swelling on the right buccal vestibule and tenderness on percussion noted in relation to 47. Routine laboratory investigations were normal. On radiographic examination, orthopantomography revealed diffuse radiolucency in the periapical region of 47 showing well-defined unilocular radiolucency with well-defined corticated borders seen associated with horizontally impacted 48. The radiolucency was noted extending from the crown of 48 involving the root of 47 regions mesiodistally and superoinferiorly extending from the alveolar ridge of 48 region up to a few millimeters above the inferior border of the mandible. There is no evidence of displacement of the tooth and no resorption of the root. (Figure1)

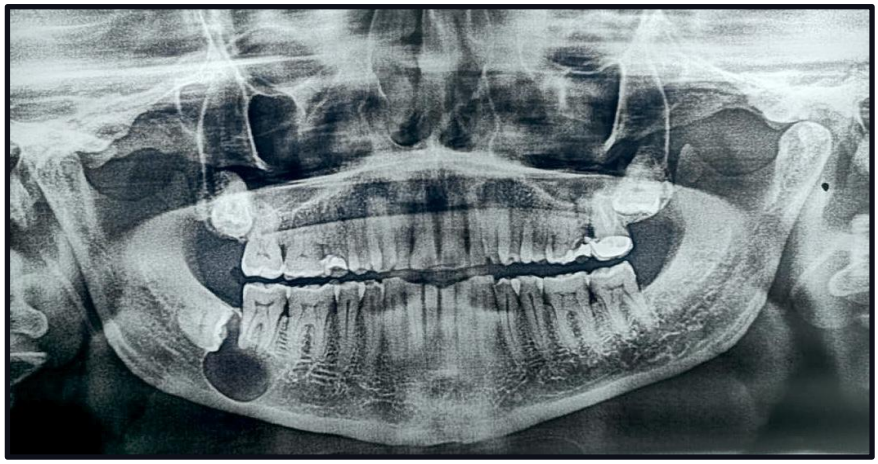

FIGURE 1: Indicates the unilocular well-defined corticated radiolucency noted with horizontally impacted 48 extending from the coronal surface of 48 up to the root of 47 regions.

Based on history, clinical and radiographic examination, a differential diagnosis of dentigerous cyst, OKC, and Unicystic ameloblastoma was suggested. After getting an informed consent signature from the patient surgery was performed to remove the lesion. Under general anesthesia, surgical enucleation of the cystic area was done along with the extraction of the impacted tooth. Excised specimens were submitted to the Department of Oral and Maxillofacial Pathology for providing a correct diagnosis. (Figure 2)

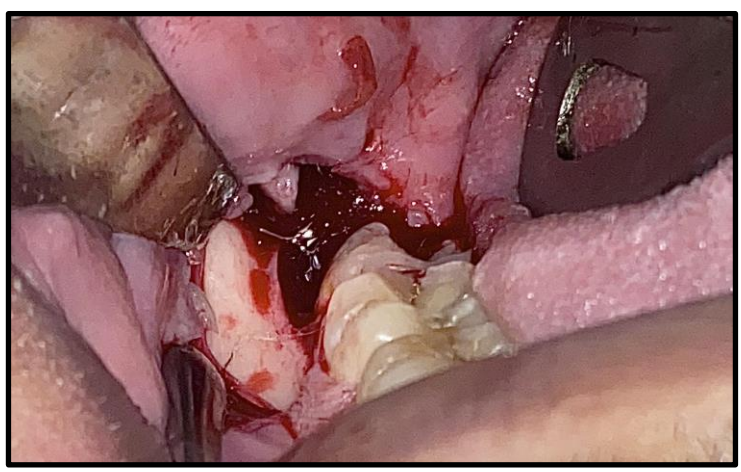

FIGURE 2: Enucleation and complete removal of the cyst along with the impacted 48

After grossing, processing, and sectioning the specimen was viewed under a microscope. The stained H and E section showed a cystic lining showing few areas of the keratinised stratified squamous epithelium of variable thickness, predominantly 5-6 cells layer thick and few areas of non keratinized stratified squamous epithelium of variable thickness predominantly 3-4 cells layer thick. The basal cells are cuboidal with hyperchromatic nuclei arranged in a palisading manner. Underlying dense connective tissue wall shows moderate chronic inflammatory cell infiltrate, vascularity, and areas of hemorrhage. Cholesterol clefts and peripheral resorbing are also seen. (Figure3)

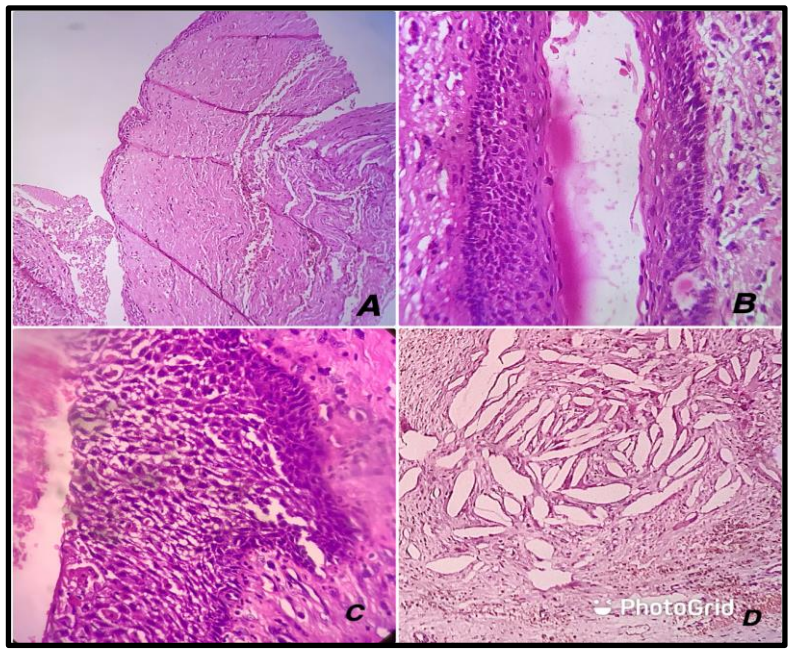

FIGURE 3: H and E Section
A) shows 3 to 4 cell layer thickness of non-keratinized stratified squamous epithelium with flat epithelium connective tissue interface(10X)
B) 5 to 6 cell layer parakeratinized stratified squamous epithelium with palisading hyperchromatic basal cell layer (40X)
C) Shows the infected hyperplastic non-keratinized epithelium of variable thickness (10X)
D) Cholesterol clefts are evident (40X)

Based on the clinical, radiological and histopathological features the diagnosis was given as was given as infected odontogenic cyst - odontogenic keratocyst? The patient was followed up and no recurrence was noted.

Discussion:

The odontogenic keratocyst (OKC) is considered to be one among the most aggressive odontogenic cysts, owing to its relatively high recurrence rate and local invasion [8][9][10][11]. Based on its clinical behavior, it has been categorized as a benign neoplasm in the 2005 WHO odontogenic tumors and cysts classification but reclassified as a cyst in the 2017 WHO odontogenic tumors and cysts classification.[12]

Pain, soft tissue swelling, and bone expansion with gross facial asymmetry are observed clinically. It is more common in females and the peak age of occurrence of OKC is in the second and third decades of life. In our case, the patient is in his sixth decade, which reflects its bimodal presentation. [3] There are mixed reports present in the literature for gender predilection.

The presence of well-defined radiolucency encircling the crown of an impacted tooth is normally diagnosed as a dentigerous cyst on clinical radiographic examination, it was a close differential. The attachment of the cystic lining to the neck of the tooth also contributed to the possibility of diagnosis as a ‘Dentigerous cyst’. The microscopic examination of the cyst fitted well in favor of OKC, thus, the possibility of considering this as a DC with a keratinized lining epithelium did not have credibility.

In 1982, Altini and Cohen coined the term "Follicular keratocyst" to describe a group of lesions in which the cystic lining was characteristic of an odontogenic keratocyst on histological examination but had completely encircled the crown of the tooth and was firmly attached to the neck on gross examination. When an enlarging keratocyst surrounds an unerupted tooth's follicle, the cystic lining may fuse with the reduced enamel epithelium.(5)The cystic epithelium immediately surrounding the tooth's neck is not keratinized in these situations, and the underlying capsule displays inflammatory alterations. In the present case, the findings were similar to the literature with the cyst surrounding an impacted tooth, the lining observed to be attached to the neck of a tooth, giving the appearance of a dentigerous cyst. The bulk of the cystic lining had 6-8 layers of parakeratinized stratified squamous epithelium along with a few areas showing 3-4 cell layers of non-keratinized stratified squamous epithelium resembling reduced enamel epithelium, giving credence to the earlier mentioned theory that the cyst surrounding an impacted tooth could have fused with the reduced enamel epithelium.

The diagnosis of follicular keratocyst was made based on the gross, radiographic, and histological examination. Multiple treatment options have been proposed, including marsupialization or decompression, enucleation with peripheral ostectomy, and a combination of these. In the current case, enucleation with peripheral ostectomy was carried out. The patient has been on regular follow-up for 1 year with no signs of recurrence.

Kim DK and colleagues found that the staining pattern and intensity for Ki-67 were the same for both the follicular and extrafollicular variants of OKC. As a result, follicular OKC is similar to extra-follicular OKC in terms of aggressiveness, and it should be treated with the appropriate therapeutic method to avoid recurrences. [8] As a result, it's critical to identify between a follicular keratocyst and a dentigerous cyst, as their clinical behaviour differs.

CONCLUSION

As a result, it was concluded that diagnosing keratinized odontogenic cysts is challenging since these cysts had the radiographic appearance of other non-keratinizing odontogenic cysts because of their unique placement around the crown of an unerupted tooth. A complete histological and radiological examination is performed, and advanced molecular analysis and correlation are also necessary.

Financial support and sponsorship:

Nil

Conflicts of interest

There are no conflicts of interest

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