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Case Report

Orthokeratinized Odontogenic Cyst of Maxilla: A Case Report

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Abstract

Background: A variety of Odontogenic cysts frequently present in the jaws. Cysts that develop from epithelial tissues involved in tooth formation are classified as "Odontogenic" and can be inflammatory or developmental in origin. The OKC is a very different form of developmental Odontogenic cyst because of peculiar clinical behavior, distinctive origin and development, specific histopathological features, unique tendency for recurrence and disputed treatment modalities. Orthokeratinized Odontogenic cyst is a relatively rare developmental Odontogenic cyst which was initially considered as Orthokeratinized type of OKC by WHO-1992, and later on OKC as KCOT in WHO-2005 classification, but now it is designated as different entity from OKC as OOC in WHO-2017 classification of head and neck region of Odontogenic tumors. Here we present a clinically diagnosed case of maxillary sinusitis which turned out to be Orthokeratinized Odontogenic cyst on histopathological examination.

Keywords: Odontogenic Cyst; Orthokeratinized Odontogenic Cyst; Odontogenic Keratocyst

Introduction

Odontogenic keratocysts (OKCs) are developmental odontogenic cysts of epithelial origin, first identified and described in 1876 and further characterized by Phillipsen in 1956 [1]. Pindborg and Hansen suggested the histologic criteria necessary to diagnose OKC in 1962 [2]. Phillipsen introduced the term OKC in 1956 to describe all odontogenic cysts showing keratinisation of lining epithelium [3]. It is derived from the remnants of the dental lamina which represent a primordial epithelium with the ability of keratinisation, proliferation and infiltration. OKCs are keratinized epithelium lined cysts of the jaws with well defined histologic criteria. Currently, significant difference between keratinized cystic lesions are recognized and Orthokeratinized Odontogenic cyst (OOC) is no longer part of the spectrum of Odontogenic Keratocyst (OKC) [4].

Orthokeratinized Odontogenic cyst (OOC) is a relatively rare developmental jaw cyst, considered as distinct entity from Odontogenic Keratocyst as it exhibits a less aggressive behavior and a

very low rate of recurrence arising from the cell rests of the dental lamina or from the basal cell layer of the oral mucosal epithelium mainly located in the posterior segment of the mandible with specific histological features and clinical behavior [5]. The OOC occurs predominantly in males between the third and fourth decades, with a mean age of 33.5 years. It is not until 1998 that Li., *et al*. suggested the term "Orthokeratinized Odontogenic cyst", which is the most accepted at the present time [6,7].

Here, we present histopathologically diagnosed case of OOC of maxillary sinus with distinct clinical, radiological and histopathological behavior thus, differentiating it from OKC.

Case Presentation

A 32 years old male patient reported to the department of Oral and Maxillofacial Surgery, ESIC Dental College and Hospital, Rohini, Delhi, with a complaint of large Oro-antral communication in the left posterior region of the jaw associated with pus discharge from left zygoma. On intra-oral examination, there was a hard tissue defect in the left first molar of the maxilla. There was existing epithelialized mucosa along opening of sinus and also the defect was extending upto sinus. On radiological examination, the computed tomography showed an expansive multilocular radiolucency and Oro-antral communication with respect to 27 and 28 region of maxilla. Clinically the case was diagnosed as chronic maxillary sinusitis with Oro-antral communication.

Investigations

A fine needle aspiration cytology (FNAC) was done with 26 bore needle, and a creamish-white liquid material was obtained. The cytology of the aspirated material revealed the presence of squamous cells and few inflammatory cells. All hematological investigations were within normal limits. Excisional biopsy was done in the department of Oral and Maxillofacial Surgery and multiple bits of soft tissues, reddish-brown in color and irregular in shape and surface texture were received and sent to the Department of Oral and Maxillofacial Pathology for histopathological examination.

Grossing

Gross examination showed multiple bits of soft tissues measuring approximately 3.0x3.0x2.0 cms, 1.5x1.2x0.5 cms and 1.5x1.0x0.4 cms respectively. The soft tissues were reddish-brown in color, irregular in shape and size and firm in consistency (Figure 1). The tissues were kept for routine processing and microscopic examination after Haematoxylin and Eosin staining.

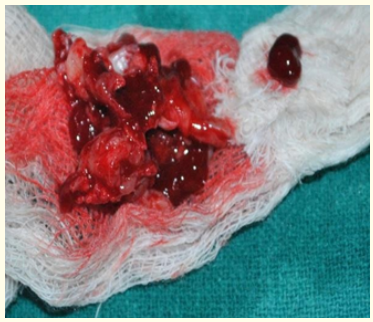

Figure 1: Grossing of specimen.

Histopathology

Histopathology showed a cystic lining of orthokeratinized stratified squamous epithelium of uniform thickness consisting of 6-8 cell layers showing basal cell polarization. Cystic lumen showed traces of keratin. Sinus lining of pseudostratified ciliated columnar epithelium with inflamed connective tissue containing glands were also seen (Figure 2a). The fibro-cellular connective tissue stroma revealed chronic inflammatory cells chiefly lymphocytes. Hemorrhagic areas were also evident at certain places (Figure 2b). These features were typical of orthokeratinized odontogenic cyst (OOC).

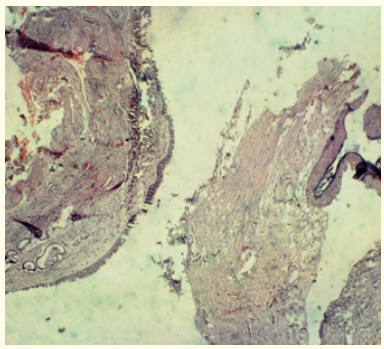

Figure 2a: H&E section shows antral lining of pseudostratified ciliated columnar epithelium with goblet cells and cyst lining seen in adjacent area - X4x.

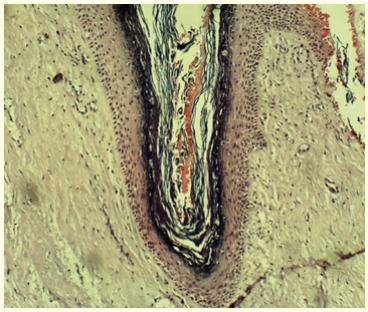

Figure 2b: H&E section shows orthokeratinized stratified squamous epithelium with fibro-cellular CT stroma and cystic lumen containing keratin -X 10x.

Discussion

OOC is an uncommon developmental Odontogenic cyst and constitutes about 5.2% to 16.8% of the cases that had been previously diagnosed as OKC in different case series [6]. OOC was described as distinct individual entity from other odontogenic cysts by Wright in 1981 and the term orthokeratinized odontogenic cyst was given by Li., *et al*. in 1998 which is the most widely accepted at present [6,8]. OOC occurs predominantly in young adults and shows 2:1 male:female ratio [9]. According to previous studies, OOC occurs mostly in the third and fourth decades of life [6,10]. Moreover, MacDonald., *et al*. stated that there is preponderance for occurrence in females in the second decade of life. The mandible was more commonly involved than the maxilla, the most common location being the mandibular molar and the ramus region [11]. The size can vary from <1 cm to >7 cm in diameter9.

The lesions of OOC present clinically and radiologically as hard, progressively growing, non- tender swelling, with a well defined radiolucency. OOCs are usually asymptomatic and a slow growing swelling is the most frequent presenting symptom and is occasionally accompanied by pain [7]. Large lesions can cause cortical expansion [12], but in our case, its clinical behavior was totally different and was misdiagnosed as chronic maxillary sinusitis on clinical and radiological evaluation.

Radiographically, the cyst appears as a well-circumscribed, unilocular or multilocular radiolucency that occasionally is associated with an unerupted tooth or with the root, without causing resorption [12]. Both the OOC and KCOT show similar findings clinically regarding age, sex, and site of occurrence but the OOC are generally solitary asymptomatic lesions whereas KCOT associated with Nevoid basal cell carcinoma syndrome exhibits multiple lesions [13].

Histopathologically, OOC have a cystic lining of orthokeratinized stratified squamous epithelium of uniform thickness and basal cell polarization with prominent granular cell layer in varying thickness. The basal cells are flat or low cuboidal that exhibits palisading nature with superficial layer of orthokeratin. Cystic lumen may also contain keratin. In our case, histopathological features were also in accordance with conventional histopathological characteristics of OOC which confirmed our final diagnosis of the presented case as OOC. This entity must be differentiated from the KCOT that shows a regular epithelium of 5 - 10 cell layers thick with basal cells lined with an elongated nucleus and the presence of characteristic superficial corrugated layer of parakeratin [14,15].

The differential diagnosis of the OOC on the basis of clinicoradiographic features includes KCOT, Dentigerous cyst or Paradental cyst and Ameloblastoma. In our case, the clinical and radiographic features were suggestive of chronic maxillary sinusitis which was erroneous from final diagnosis as OOC. The histopathology of the lesion confirmed the final diagnosis as Orthokeratinized Odontogenic cyst. For further confirmatory diagnosis, immunohistochemical studies can be done for the expression of Bcl-2, Ki-67, P-53 and TGF-α. The reduced expression of all these tumor markers can be observed in OOC. Molecular analysis can also be done for its pathogenesis and clinical behavior. The immunohistochemical studies and the molecular analysis will help in confirmation of the diagnosis, prognosis and treatment planning of the lesion.

Management

The best treatment course recommended for OOC is conservative surgical removal with complete enucleation of the cystic lesion to prevent its recurrence. Dong., *et al*. in their study found that enucleation with or without curettage, combination of enucleation followed by marsupialisation and peripheral osteotomy for large multilocular lesion of OOC showed no recurrence [6].

Conclusion

Orthokeratinized Odontogenic Cyst is relatively more common in posterior region of mandible, mostly body and ramus. It is considered as a distinct entity from other Odontogenic cysts because

of its peculiar clinical and less aggressive behavior, erroneous radiological and histopathological features and a very low recurrence rate. A definite diagnosis of OOC can be made by histopathology of lesion, and advanced technology includes immunohistochemistry and molecular analysis to explore its histopathogenesis, clinical behavior and best treatment approach.

The case being presented was that of a maxillary pathology which was clinically diagnosed as chronic maxillary sinusitis and the diagnosis was established only on microscopic examination of the curetted lesion. The less aggressive behavioral pattern of the lesion should be kept in mind and a more conservative treatment approach should be considered compared to the more commonly occurring Odontogenic Keratocyst which warrants a radical treatment modality.

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