Ortho keratinized Odontogenic Cyst of Mandible: A Rare Case Report

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Abstract

Objective: The Ortho keratinized Odontogenic Cyst (OOC) is a rare lesion originates from dental lamina and clinically, it may be mistaken for many other odontogenic cysts and Tumors. Microscopically, It should be distinguished from KCOT because of differences in biologic behavior and histologic features.

Case: An interesting case of OOC arising in the edentulous mandibular right first premolar region of a 55-year-old woman is reported. Under the initial clinical diagnosis of a residual cyst, the excisional biopsy was performed. Because of detection of an orthokeratinized epithelium lining, a definite diagnosis of OOC was made.

Conclusion: Microscopic examination is crucial for making the correct diagnosis of such lesions, therefore establishing patients' prognosis accurately.

Key words: Bone, Jaw, Keratocyst, Odontogenic Cyst, Orthokertinized Odontogenic Cyst.

Please cite this article as:

Sargolzaei S, Mashhadi Abbas F, Hesami L, Jabbari S, Zadsirjan S. Ortho keratinized Odontogenic Cyst of Mandible: A Rare Case Report. J Dent Sch 2015; 33(3): 233-237.

Received: 28.04.2015 Final Revision: 16.06.2015 Accepted: 20.07.2015

Introduction:

Ortho keratinized odontogenic cyst (OOC) is a developmental cyst relatively uncommon, arising from the cell rests of the dental lamina (1, 2).For the first time, Schultz described OOC in 1927 and considered it to be a type of formerly called Odontogenic Kerato Cyst (OKC)(3). Li *et al.* (1998) suggested the term "ortho keratinized Odontogenic cyst ", which is now the most accepted (1). The WHO's 2005 edition reclassified the parakeratotic type as a neoplasm and designated it as KCOT. The 2005 edition clearly excluded OOC from its description of a KCOT (4).

The occurrence of OOC in oral cavity is considered to be eight times less than KCOT (5).Due to its different histopathology, less aggressive clinical behavior and less recurrence pattern, it is important to distinguish this lesion from KCOT.

OOC usually is seen more commonly in males between third and fourth decades , with a mean age of 33.5 years (1-3).The mandible is affected twice as often as maxilla(5). The posterior portion of the lower jaw is often involved (5).

The most frequent symptom is swelling, with or without pain. The size can vary form less than 1cm to large lesions greater than 7cm in diameter (6).

Radiographically, the cyst appears as unilocular and occasional multilocular radiolucency with well-defined border. OOC is most often associated with an unerupted mandibular third molar tooth (6). The root resorption is not a feature of the COC but it can displaced adjacent teeth and inferior dental canal (1, 5)

Microscopically, OOC is characterized by the presence of uniform ortho keratinized stratified epithelium with a thick granular layer and non-

palisaded basal layer (1, 3)

Surgical nucleation with curettage is the treatment choice for COCS .The rate of recurrence has been reported between 0 and 2% which is in marked contrast with the 40% recurrence of KCOT (2, 7).

The object of this article is to describe a rare case of COC appeared a residual cyst and presents a pertinent review of this odontogenic lesion including its clinicopathologic behavior and histopathologic pattern.

Case:

A 55-year old woman was referred to the oral pathology department of Shahid Beheshti dental faculty .Beforehand, the lesion was detected incidentally during radiographic evaluation, in the edentulous region of tooth #28.Her dental history revealed that the tooth #28 had been extracted three months ago. The patient had no chief complaint. Clinical assessment showed no swelling, discoloration or any other abnormalities at the region. Her medical history was noncontributory. On palpation, both buccal and lingual structures surrounding the area were painless.

Orthopantogram exhibited a unilocular well - defined radiolucency with sclerotic border measuring about 15×10 mm between the region of teeth #27 and #29. (Figure.1)On the basis of the radiographic features in the edentulous area, a hypothesis of residual cyst was raised. Excisional biopsy was performed under local anesthesia .Gross examination of the excised Specimen revealed three pieces of cystic creamy-brown elastic tissue measuring 1×1×0.4 cm. In cut surface was cystic creamy with caseous material and lining thickness of 0.6 cm. After surgery, the material obtained was immediately submerged in 10% neutral buffered formalin and sent to the laboratory to analyze its histopathological features. At the oral pathology laboratory, the lesion was cut in to small pieces, processed according to standardized histotechemical methods, and then embedded in paraffin. Sections 5-µm-thick were obtained from the paraffin block and stained with hematoxylin-eosin.

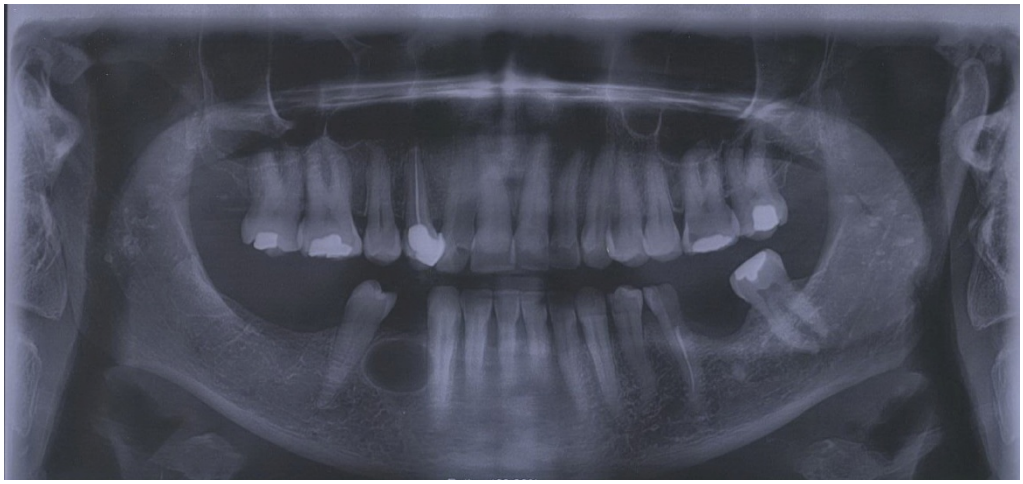

Figure 1- Orthopanthogram showing a unilocular well-defined radiolucency With sclerotic border between the region of teeth #27 and #29]

Microscopically, sections showed a cystic lesion lined by varying thickness ortho keratinized stratified squamous epithelium. A prominent flattened basal cell layer which is characteristic of the ortho keratinized odontogenic cyst was present in this case. Granular cell layer was noticeable in the superficial layer subjacent to the orthokeratin material. Detached epithelium

of cyst wall in some areas and a thick layer of orthokeratin material in cystic lumen were present (Fig 2, 3). The final diagnosis was OOC. The excisional biopsy performed beforehand, was considered as a treatment. Since careful follow-up is needed in order to detect any signs of recurrence, patient was asked to attend for six-month and one-year follow up appointment. The time for the scheduled appointment has not arrived yet.

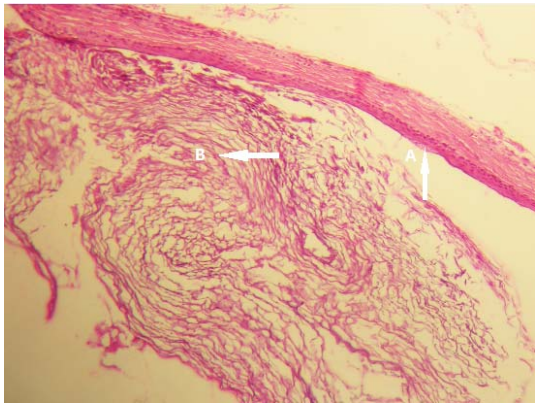

Figure 2- The lining of ortho keratinized odontogenic cyst shows stratified squamous epithelium (A) with surface orthokeratin material (B). Hematoxycilin eosin stain (×40)

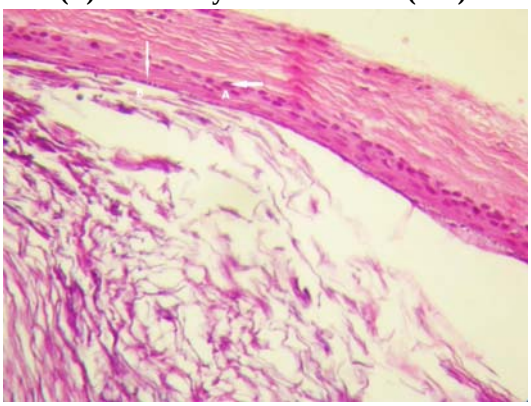

Figure 3- The stratified squamous epithelium with flattened basal cells (A) and granular layer (B). Hematoxycilin eosin stain (×400)

Discussion:

In the present case, we described a rare case of OOC developing in the edentulous area, with appearance of a residual cyst. In contrast to residual cysts that are primarily associated with inflammatory process; OOCs are known to have a developmental origin. They present 10% of all cases previously Diagnosed as odontogenic keratocyst (1, 2, 5).

OOCs occur predominantly in young adults and show a 2:1 male-to-female ratio (6). Curiously in this case, 55-year-old female was diagnosed with OOC. In agreement with previous studies, our reported lesion was located in the posterior mandible (8).

Swelling is the main clinical symptom and is accompanied on occasions with pain (1, 2, 5).The present case was discovered as incidental finding and it was completely asymptomatic.

Radiographically, OOC presents a unilocular radiolucency, but occasional multilocular. About two thirds of occs appear clinicaly and radiographically a dentigerous cyst and most often involve an unerupted mandibular third molar tooth (6).Our patient's Radiographic evaluation revealed the unilocular radiolucent lesion without unerupted tooth in an edentulous region starting from lower right canine extending to lower right second premolar and appear a residual cyst , other radiographic features were typical and with no signs of resorption or displacement.

The incidence in the region where extractions were performed before, the quite small dimension, the radiographic appearance with sclerotic margin and the absence of any kind of symptoms made it challenging to promptly identify the precise nature of the lesion. The provisional diagnosis was residual cyst and because of this, histologic examination was essential to conclude the diagnosis.

Histologically, the OOC shows a cystic cavity lined by a regular stratified squamous epithelium, usually uniform about 4- to 9-cell layers thick. The basal cell layer is less prominent and has a more fiattened or squamoid appearance. A prominent granular layer is found immediately below a flat and non-corrugated surface(9). In this case surface was corrugated

but composed of orthokeratin with prominent granular layer in some area. It is important to differentiate this entity from KCOT that presents a regular epithelium of 5-to 10-cell layers thick and the basal cells lined with a prominent palisaded nucleus. The presence of a superficial corrugated layer of parakeratin is characteristic in KCOT (10, 11).In this case, areas of superficial corrugated layer of orthokeratin was seen.

Other than typical microscopic differences, KCOT can be distinguished from OOC on several features like association with basal cell nervous syndrome, older age group, high recurrence rate, more antero-posterior extension without (5) .In relation to the immunochemical pattern , the level of expression of p53 in OOC is lower than on KCOT that propose a reduced proliferative potential. Recent immunocytochemical results proved fewer Ki-67-positive proliferating cells which are mostly limited to the basal call layer in the epithelial lining of OOC than KCOT .Hence, KCOT lining shows more prominent suprabasal activity than of OOC(2, 12) .The reactivity to cytokeratin 1,2 and 10 which would recommend a normal differentiation of the epidermis whereas the KCOT reacts to cytokeratin 4, 13, 17 and 19 , indicating that there are different entities.

This paper reports a case of OOC occurred at the mandibular right region, initially assumed to be a residual cyst. In residual cyst, keratinizing epithelium has been observed in some cases and it is considered as a metaplastic change and usually is a focal phenomenon. The presence of keratin in residual cyst is considerably related with the age of patients (11). As pointed out elsewhere, a differential diagnosis among them may be challenging particularly when there is a secondary infection and keratin is not noticeably evident on the lining surface(13).Therefore a complete examination of the lining and the attention of oral pathologist during the examination of the specimens is mandatory to confirm the diagnosis.

The differential Diagnosis of the OOC contains other radiolucent lesions of the jaws mostly odontogenic lesions such as dentigerous cyst or ameloblastoma. The OOC reveals similar radiographic characteristics with the ameloblastoma, such as its propensity to appear as a multilocular radiolucency. Unlike ameloblastoma, the OOC does not cause root resorption which is a common feature on ameloblastoma. In the differential diagnosis of all the radiolucent lesions involving impacted teeth like dentigerous cyst, OOC should always be considered. OOC has also been reported imitating as periapical lesions (14).Therefore, distinction from an indolent to a life-threatening disease developing in the periapical area, although infrequent, is potentially real.

Conclusion:

Through experience gained in the current case, we emphasize the importance of concise histhopathological evaluation of all excised lesions as well as cautious clinical diagnosis of an individual case, because a misdiagnosis may be made. Furthermore, it is important to remember that all keratinized cysts growing in the edentulous region should be warily assessed to rule out the occurrence of KCOT or even OOC, which in turn shows more aggressive behavior than residual cyst.

Conflict of Interest: “None Declared”

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