

Case Report

Follicular Keratocyst of Mandible- A Case Report

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Abstract:

Follicular keratocyst is a variant of odontogenic keratocyst (OKC). It has typical lining of OKC but surrounds the crown of impacted tooth and attached at the cemento-enamel junction, thus mimicking a dentigerous cyst on radiographical examination. Follicular keratocyst is similar to extra-follicular variant in behavior and more aggressive compared to dentigerous cyst. Thus it is important to distinguish this entity from dentigerous cyst. Review of literature shows very few cases of follicular keratocyst. Hereby we are reporting a case of follicular keratocyst which was diagnosed as dentigerous cyst based on clinical and radiographic findings and confirmed diagnosis of follicular keratocyst was made on histopathological examination.

Key words: Follicular keratocyst, Odontogenic keratocyst, Dentigerous cyst, Cemento-enamel junction.

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Introduction:

The Odontogenic keratocyst (OKC) was first described by Philipsen in 1956.^[1] They are aggressive in nature, have a high recurrence rate, greater tendency to invade adjacent tissues and often associated with unerupted teeth.^[2] It has been reclassified as “keratocystic odontogenic tumour (KCOT)” by the WHO and it is defined as “a benign uni- or multicystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and has the potential for aggressive, infiltrative behavior”.^[3]

Follicular keratocyst is considered as a variant of OKC where the cyst has typical histology of an OKC but surrounds the crown of an unerupted tooth and attached to the neck of the tooth. Follicular keratocyst may arise from the pre-existing keratocystic cavity following the eruption

of tooth, in a similar manner as the tooth erupts into the oral cavity. On radiographic picture they resemble dentigerous cyst. Follicular keratocysts account for 25 to 40 % of all the odontogenic keratocysts and are comparatively uncommon.^[4]

Follicular keratocyst is aggressive in nature and similar to extra-follicular OKC in its clinical behavior.^[4] Therefore it is important to distinguish this entity from dentigerous cyst. Hereby we are reporting a case of follicular keratocyst involving the impacted canines in the mandibular anterior region.

Case Report:

A 39 year old male patient was reported to the dental OPD complaining of pain in the mandibular anterior region since 3 months. The patient complained of paresthesia of lower lip bilaterally. The patient had no

significant medical and dental history. The extra oral and intra oral examination did not show obvious swelling.

Intra oral examination revealed missing permanent mandibular left and right canine. Therefore an orthopantomogram was taken to assess the condition of canines. Orthopantomogram revealed a well-defined radiolucency with corticated borders, extending from lower left second premolar to right second premolar region which was also enclosing impacted mandibular right and left canine (Figure 1).



Figure 1: Orthopantomogram, showing a well-defined radiolucency with corticated borders, enclosing impacted mandibular right and left canine.

A provisional diagnosis of KCOT/Dentigerous cyst was made. The lesion was surgically enucleated with chemical treatment (Carnoy's solution) along with the impacted lower canines and sent for histopathological examination.

The gross specimen was greyish white in color, measuring approximately 10x6cm in size. The specimen appeared as a cystic lesion with a cystic cavity surrounded by a well-defined capsule. The cystic lining was attached to neck of mandibular left canine in the region of cemento-enamel junction.

The histopathological examination stained with H & E showed cystic cavity lined by epithelium of variable thickness. The major portion of the lesion revealed keratinized stratified squamous epithelium of 6-8 layers thickness. Basal layer showed columnar cells with palisading arrangement of nuclei (Figure 2). The section taken from the tissue attached to the neck of the tooth

exhibited thin epithelial lining resembling reduced enamel epithelium which was 2 to 3 layers thick (Figure 3).

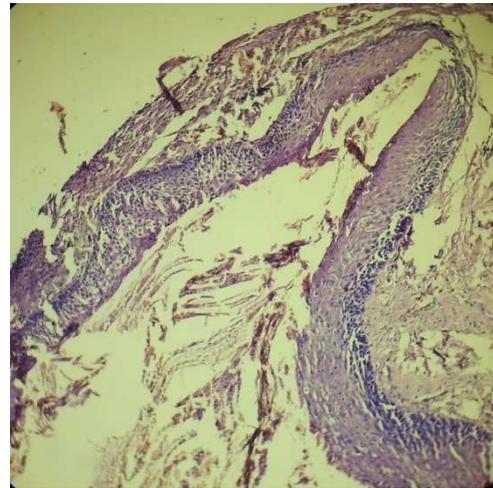


Figure 2: Cystic cavity lined by epithelium of 6-8 cell thickness and basal layer showing columnar cells with palisading arrangement of nuclei (H & E X10).

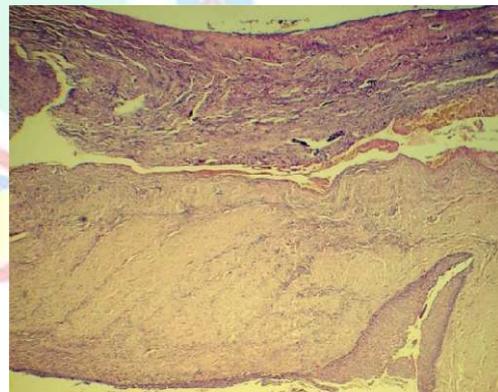


Figure 3: The section taken from the tissue attached to the neck of the tooth, exhibiting thin epithelial lining resembling reduced enamel epithelium of 2 to 3 layers thick(H & E X 4).

The connective tissue demonstrated few chronic inflammatory cells predominately lymphocytes and few plasma cells. The blood vessels lined by endothelial cells and cholesterol clefts were also observed.

Thus correlating the clinical, radiographic and histopathological features, a diagnosis of follicular keratocyst was made.

Discussion:

Follicular keratocyst is a relatively uncommon entity. Only few cases of follicular keratocyst have been reported so far in English literature. Incidence of follicular keratocyst is more in mandible compared to maxilla.^[5] The present case was reported in the mandibular anterior region which coincided with the previous studies. In 1969, Browne suggested that when an enlarging keratocyst surrounds the follicle of an unerupted tooth, there is possibility of fusion of cystic lining with the reduced enamel epithelium. In such cases the cystic epithelium immediately around the neck of the tooth is not keratinized and shows inflammatory changes in the underlying capsule. Later in 1982, Altini and Cohen introduced the term "follicular keratocyst" for group of lesions where the cystic lining was characteristically of odontogenic keratocyst on histological examination but which on gross examination had completely surrounded the crown of the tooth and had been firmly attached to the neck. Accordingly, they suggested that follicular keratocysts are extrafollicular in origin and may develop following eruption of a tooth into a pre-existing keratocystic cavity.^[6] In the present case, the cyst was enclosing impacted teeth and lining was attached to the neck of one of the teeth giving an impression of a dentigerous cyst. On histopathological examination, the majority of the cystic lining showed 6-8 layers thickness and the basal layer of cells with palisading arrangement of nuclei. The cystic lining attached to the neck of tooth resembled reduced enamel epithelium giving the impression of dentigerous cyst. Thus correlating with the gross, radiographic and histopathological examination, the diagnosis of follicular keratocyst was made.

Various treatment modalities have been suggested that includes marsupialization or decompression, enucleation with peripheral ostectomy, enucleation with Carnoy's solution, physical treatment with cryotherapy using liquid nitrogen and

resection.^[7] In our case the mode of treatment done was enucleation with peripheral ostectomy. Patient has been on regular follow up since 1 year with no signs of recurrence.

Kim DK et al in 2003 conducted an immunohistochemical study that revealed the staining pattern and intensity for Ki-67 was same for both the follicular and extra-follicular variant of OKC. Thus follicular OKC is similar to the extra-follicular type in aggressiveness and should be treated with the correct therapeutic approach in order to prevent recurrences.^[8] Therefore it is important to distinguish follicular keratocyst from dentigerous cyst as the clinical behavior of these two lesions are different.

Conclusion:

Follicular keratocysts are rare entities and should be distinguished from dentigerous cyst. Although clinical and radiographic findings are contributory, the final diagnosis should be based on histopathological examination. Upto date only few cases of follicular keratocyst have been reported. Analysis of additional cases of follicular keratocyst with long term follow up may add clarity to our current understanding of this rare entity.

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