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Case report

Keratocystic Odontogenic Tumor of Buccal Mucosa: A Rare Case Report and Histochemical Comparative Study

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Keratocystic odontogenic tumor (KCOT) is a benign intraosseous neoplasm of the jaw occurring most commonly in mandibular ramus molar area with male predilection. Peripheral KCOTs are very uncommon. Here we report a case of keratocyst in buccal mucosa in a 55 years old female patient, the diagnosis of which was based on subjective histological evaluation and further confirmed by immunohistochemical stain and compared its immunohistochemical features with intraosseous KCOT. Intraosseous KCOT and buccal mucosa keratocyst were stained with hematoxylin and eosin, picrosirous red, Ck 17, Bcl 2 and Ki 67, and compared. Keratocyst of buccal mucosa showed positive expression for CK17 and Ki 67, and negative expression of Bcl2. On polarizing microscope, it showed yellowish red birefringence of collagen fibers in connective tissue wall. Keratocyst in the buccal mucosa is rare. Though it mimics intraosseous counterpart histopathologically, the nature of connective tissue and low proliferation index makes it less destructive. Therefore, it requires only conservative excision with follow up.

Keywords: Keratocystic odontogenic tumor, buccal mucosa, immunohistochemistry, picrosirous red stain

Keratocystic odontogenic tumor (KCOT) is one of the most frequent benign, multicystic odontogenic tumors, comprising about 11% of all jaw cysts (1). KCOT may be uni- or multicystic, with characteristic parakeratinized squamous epithelium lining, which has a potential for aggressive and infiltrative growth (2). KCOTs have bimodal age allocation with incidence in 3rd and 5th decades and slight male predilection. They occur usually in the mandibular posterior body and ramus region (3). They almost always occur within the bone, however small number of peripheral KCOT cases has been reported in gingiva (15 cases) (4) and buccal mucosa (5 cases) (5). KCOT has also been reported in maxillary antrum, temporomandibular joint, skin and temporal region (5-6). Among their clinical features, local destruction potential, as well as tendency for multiplicity specially when associated with nevoid basal cell carcinoma, can be noted (7). KCOTs originate from remnants of dental lamina and from basal cells of the oral epithelium (8). The aim of the present study was to describe a case of keratocyst of buccal mucosa and compare the immunohistochemical features of this lesion with sporadic intraosseous KCOT.

Case report

A 55 years old female patient presented with a painful swelling in the left cheek region present since 3 months. The swelling was progressive and was associated with pain since 15 days. On clinical examination, a solitary firm submucosal nodule, tender on palpation of size 1x1cm was located on left buccal mucosa in relation to 25 and 26 region

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Figure 1. Solitary submucosal mass on left buccal mucosa in relation to 25 and 26 region.

(Figure 1). Since it was a small lesion, excisional biopsy was advised. On surgical intervention, a thick walled mass was observed, which was sent for histopathological examination. Clinical differential diagnosis of fibroma, neurofibroma and lipoma were given.

On histopathology, cystic lining and fibrous connective tissue were observed. The cystic epithelial lining was parakeratinized stratified squamous epithelium with 4-5 cell thickness. The basal cells were low columnar with hyperchromatic nuclei showing palisading arrangement with corrugations at surface epithelium (Figure 2a). The surrounding connective tissue showed dense bundles of collagen fibers arranged irregularly, inflammatory infiltrate with numerous eosinophils and budding blood capillaries with proliferating endothelial cells (Figure 2b). Adipose tissue, bundles of nerves were also seen along with muscle degeneration (Figure 2c). Based on these features it was diagnosed as keratocyst of buccal mucosa. Considering the rarity of the lesion and to extricate tissue extension of central lesion, orthopantamogram was taken but no apparent pathology was seen confirming our diagnosis. In an attempt to elucidate the nature of the lesion and to discern any differences with that of intraosseous KCOT, picrosirous red staining, as well as Ck17, Ki67 and Bcl2 immunohistochemical staining were further performed. Moreover, another case of KCOT of maxillary sinus, and 2 cases of intraosseous KCOTs were retrieved from archives and served as control cases. 5 microns serial sections were cut from paraffin blocks and stained with routine hematoxylin and eosin and were used for re-evaluation of the histological diagnosis. The ethical clearance for the present study was approved by institutional ethical committee.

While studying the nature of connective tissue by picrosirous red, keratocyst of buccal mucosa and maxillary KCOT showed yellowish red birefringence of fibers, whereas intraosseous KCOT revealed greenish yellow color (Table 1 and Figure 3).

Three micrometer sections of 1 keratocyst of buccal mucosa, 2 intraosseous KCOT and 1 maxillary sinus KCOT were deparaffinized and rehydrated in serial alcohol solutions. Antigen retrieval was performed in a microwave oven in TRIS-EDTA buffer (pH 9). Tissue sections were quenched of endogenous peroxidase followed by washing step with TRIS buffer saline with pH 7.4. Incubations with primary antibodies: Ki67, Ck17 and Bcl2 were performed. Reactions were developed with post primary block for 30 min at 37

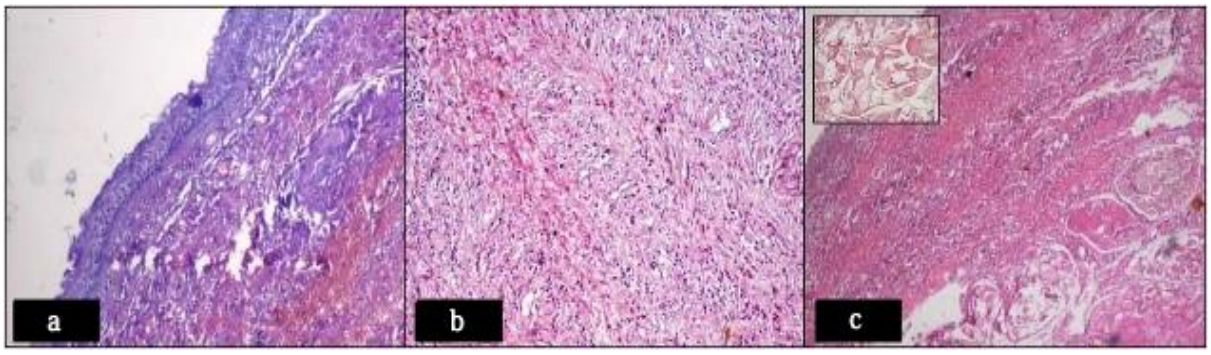


Figure 2. Hematoxylin and eosin staining of KCOTs. Characteristic lining of KCOT (a); dense connective tissue with proliferating endothelial cells and numerous eosinophils (b); deeper connective tissue showing muscle degeneration (inset) (c).

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Table 1. Immunohistochemical pattern of expression in keratocyst of buccal mucosa, sporadic KCOT and KCOT of maxillary sinuus

References	Buccal mucosa keratocyst	Intraosseous KCOT	Maxillary sinus KCOT
13	Ck17(+) suprabasal layer	Ck17(+) suprabasal layer	-
5	Ck17(+) in all layers	Ck17(+) in all layers	-
	Bcl2(+) basal layer	Bcl2(+) basal layer	
Present report	Ck17(+) basal and suprabasal layer	Ck17(+) basal and suprabasal layer	Ck17(+) basal and suprabasal layer
	Bcl2(-)	Bcl2 (+) in basal layer	Bcl2 (+) in basal layer
	Ki67(+) sporadically in basal layer	Ki67(+) basal, suprabasal and spinous cell layers	Ki67(+) basal and suprabasal layer in focal areas
Picrosirous red (present case)	Yellowish red	Greenish yellow	Yellowish red

KCOT: keratocystic odontogenic tumor; (+): positive staining; (-): negative staining.

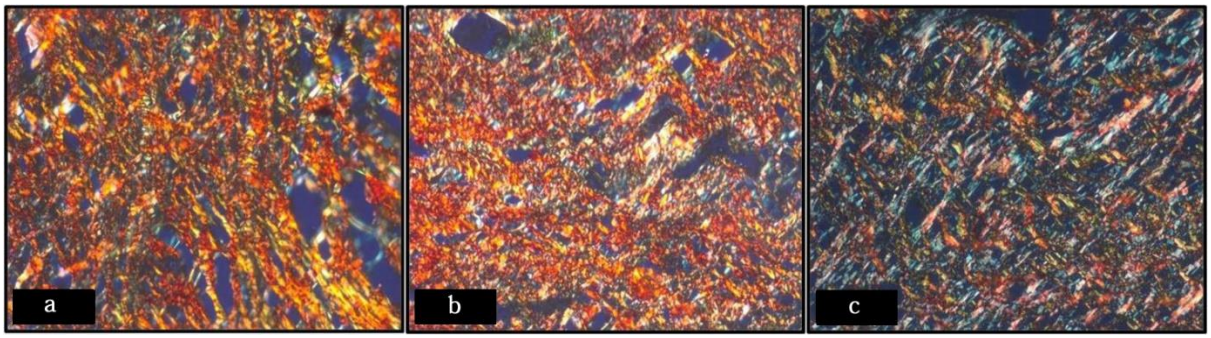


Figure 3. Picrosirous red staining. KCOT of buccal mucosa (a) and maxillary sinus (b) showpredominantly yellowish red birefringence and intraosseous; KCOT (c) showing greenish yellow birefringence under polarizing microscope.

°C and were visualized using DAB chromogen (diaminobenzidine). The sections were counterstained with Harris' hematoxylin. The epithelium lining of KCOT was histologically divided into basal and suprabasal layers. Ck17 was positive in basal and suprabasal layers of both cases and controls, which confirms the odontogenic origin of cysts (Figure 4). Ki67 was strongly positive in

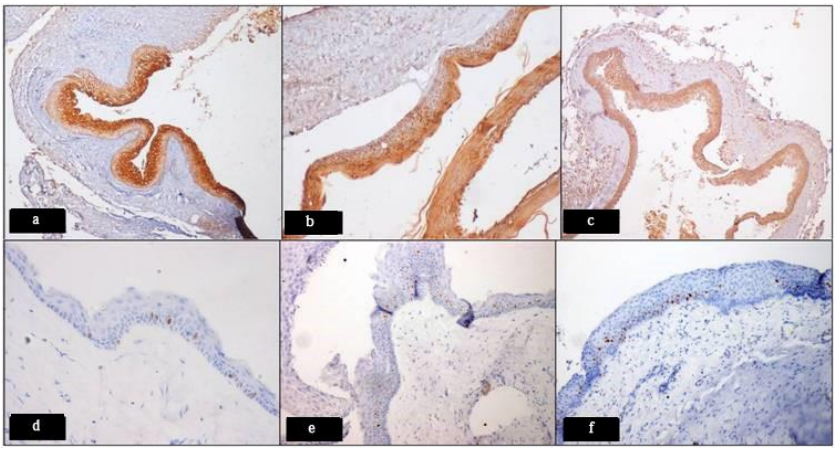


Figure 4. Immunohistochemical staining of KCOTs with Ck17 and Ki67. Positivity of Ck17 in lining epithelium of KCOT of buccal mucosa (a), maxillary sinus (b) and intraosseous KCOT (c); Ki67 staining in lining epithelium of KCOT of buccal mucosa (d), maxillary sinus (e) and intraosseous KCOT (f).

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basal, suprabasal and spinous cell layers in intraosseous KCOT, weakly positive in basal layer in keratocyst of buccal mucosa, whereas in maxillary sinus KCOT, basal and suprabasal layer showed positivity in focal areas (Figure 4). Bcl2 was expressed strongly in basal layer of intraosseous KCOT, weakly in KCOT of maxillary sinus and negative in keratocyst of buccal mucosa (Figure 5). Therefore, the histopathological features of the present case showed typical features of a keratocystic odontogenic tumor.

PTCH function results in increased transcription of genes responsible for cell proliferation and finally results in tumor formation. Once mutation has occurred in *PTCH*, KCOT becomes target for additional genetic alterations, facilitating tumor progression. Besides genetic factors, dysregulation of cell cycle and proliferation may be important for KCOT pathogenesis (9-10).

In the present case, in an attempt to provide greater clarity regarding histogenesis, we performed immunoreactions for Ck17. Expression of Ck17 was

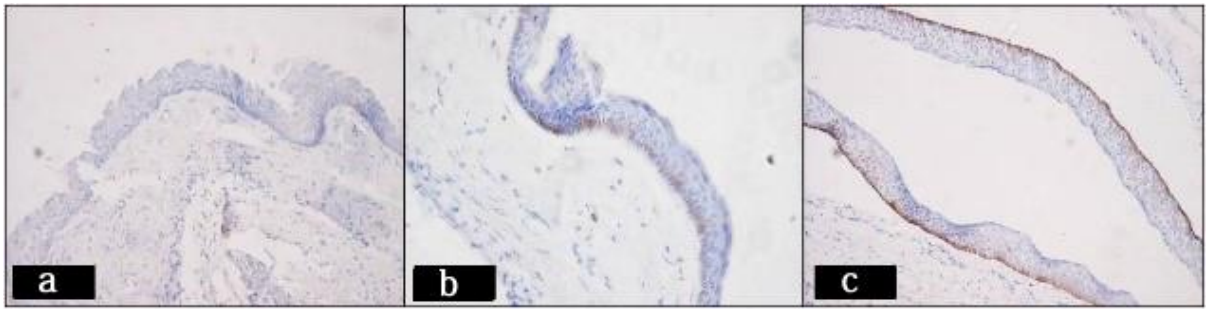


Figure 5. Bcl2 immunohistochemical staining. Lining epithelium of KCOT of buccal mucosa (a), maxillary sinus (b), and intraosseous KCOT (c).

Discussion

Keratocyst of buccal mucosa is an extremely rare entity with only few cases reported in literature. KCOTs commonly occur in the posterior mandibular region, peripheral KCOTs are rare, and in addition KCOTs in buccal mucosa are extremely rare (4). The mechanism of KCOT development in the buccal mucosa is not well understood. Various studies have explained the probable mechanisms, such as dislocation and persistence of the dental lamina in this region during odontogenesis, integration of vestibular lamina into upper molar areas distal to parotid duct during embryogenesis, and epithelial cell proliferation from epidermal tissue and its appendages (4-5). Regardless of the source of epithelial cells, the etiology of KCOT is strongly related to mutation in *PTCH* (patched-1) gene, which is a tumor suppressor that encodes for a transmembrane protein. PTCH together with SMO (smoothened) forms a receptor for sonic hedgehog ligands and suppresses SMO mediated transcription of cellular proliferation genes. Therefore, lack of

observed in basal and suprabasal layers of cystic epithelium, thus confirming odontogenic origin of lesion.

Restricted weak expression of Ki67 in keratocyst of buccal mucosa suggests its less proliferative potential when compared to intraosseous KCOT.

In intraosseous KCOT, Bcl2 expression was strongly positive in basal layer of cystic lining, confirming abnormal control of cell cycle. The Bcl2 overexpression increases the survival of epithelial cells, which may have led to peculiar growth pattern of KCOT. However, Bcl2 expression was negative in buccal mucosa keratocyst.

Collagen which forms the principal component of connective tissue was also evaluated to note if there was any discrepancy in the nature of collagen fibers in intraosseous KCOT, buccal mucosa keratocyst, and maxillary sinus KCOT. Intraosseous KCOT showed greenish yellow birefringence which was indicative of young and immature fibers. Keratocyst of buccal mucosa and KCOT of

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maxillary sinus showed yellowish red birefringence, which was accredited to the presence of mature and dense packed collagen fibers. As the maturation of collagen proceeds, there is an amend in proteogylcan content of fibers causing dehydration of fibers, thereby resulting in increased diameter and intensity of birefringence. Hence a change in polarizing color of the fibers from greenish yellow to yellowish red occurs (11).

Moreover, inflammation influences the polarization color and packing of fibers in connective tissue wall of keratocysts. In buccal mucosa keratocyst, moderate inflammation was seen, whereas in control KCOTs mild inflammation was observed. The presence of inflammation results in well packed and thickened fibers of collagen giving yellowish red birefringence (11).

The diagnosis of KCOT is based on characteristic histological features. But when it occurs in buccal mucosa, definite diagnosis of KCOT is difficult in the absence of evidence of the origin. The main differential diagnosis includes epidermal cyst and steatocystoma simplex. Epidermal cyst resembles KCOT histopathologically, but differs in site of occurrence. Epidermal cyst occurs in relation to line of closure of the embryonic fusion plane. A different pattern of cytokeratin expression is noted in both lesions. Epidermal cyst is positive for Ck10 and negative for Ck17, whereas KCOT is negative for Ck10 and positive for Ck17 (12). Steatocystoma simplex presents with undulating walls of stratified squamous cells with eosinophilic cuticle and have sebaceous glands in their walls (5).

Due to very few cases in literature, it is difficult to envisage the behavior and recurrence of keratocyst of buccal mucosa. To add to the literature, keratocysts of buccal mucosa are considered to be less aggressive due to low proliferative activity than their intraosseous counterparts, so simple surgical resection and excision of immediate adherent soft tissue would be satisfactory.

In summary, concerning KCOTs within the soft tissues of the face, few case reports have been documented. Peripheral KCOTs, especially those in buccal mucosa, are rare. The present case showed typical histopathological features of KCOT. However, picrosiorus red staining revealed matured connective tissue and immunohistochemical markers exposed low proliferative activity, thereby attributing to low aggressiveness of peripheral KCOTs when compared to intraosseous KCOT.

As only few cases have been reported to date, future genetic studies would add up knowledge to the literature.

Conflict of interest

The authors declared no conflict of interest.

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