Peripheral Odontogenic Keratocyst of Buccal Mucosa

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ABSTRACT

The odontogenic keratocyst is a developmental cystic lesion of jaw bones. There is an on-going debate about the pathogenesis of this entity. Odontogenic keratocysts are usually seen intraosseously in jaws with a predilection in the mandibular molar ramus regions. Extra-skeletal variants are reported rarely in the gingiva. Extra-skeletal variants occurring peripherally in other soft tissue components of the oral cavity are extremely rare. Even though histogenesis is ambiguous, such presentation may be related to the tumour-like behaviour of odontogenic keratocysts. Here, we present a case of peripheral odontogenic keratocyst in a 62-year male who presented with complaint of a painless lump on the right buccal mucosa. The diagnosis was made on biopsy of the lesion. The lesion was excised completely.

Key Words: *Buccal mucosa, Pathology, Keratocyst, Odontogenic.*

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INTRODUCTION

Odontogenic keratocysts (OKCs) constitute 4%–12% of all cysts arising from odontogenic structures. The origin of OKC is considered to be from the remnants of the dental lamina, the primitive embryonal ectodermal ridge that forms the enamel organ. Unlike other cysts of the jaw bones, OKC exhibits atypical clinical features; notably, locally invasive behaviour, minimal cortical bone expansion, high recurrence rate (> 60 %), and coexistence with the nevoid basal cell carcinoma syndrome (NBCCS).[1]

As many researchers reported chromosomal abnormalities associated with OKC, the World Health Organization (WHO) reclassified it from a benign cyst to a neoplasm in 2005 under the nomenclature of keratocystic odontogenic tumour. This decision was reversed in 2017 as the authors and participants of the Consensus and Editorial Panel of the WHO suggested the evidence was not sufficient at the time to consider it as a neoplasm.[2] However, cases of solid OKCs reported by many authors are against the cystic nature of the lesion.[3,4]

OKCs are usually seen intraosseously in the posterior region of the mandible but can occur anywhere in the jaw bones. Extra-skeletal variants are reported rarely in the gingiva.[1]

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Very rarely, they are found in other soft tissue components of the oral cavity.[5] Importantly, the peripheral variants occurring in gingiva and other intraoral soft tissue sites may be related to the neoplasm-like behaviour of the entity. This article aims to present a rare case of peripheral OKC (POKC) on buccal mucosa with a review of literature related to non-gingival POKC.

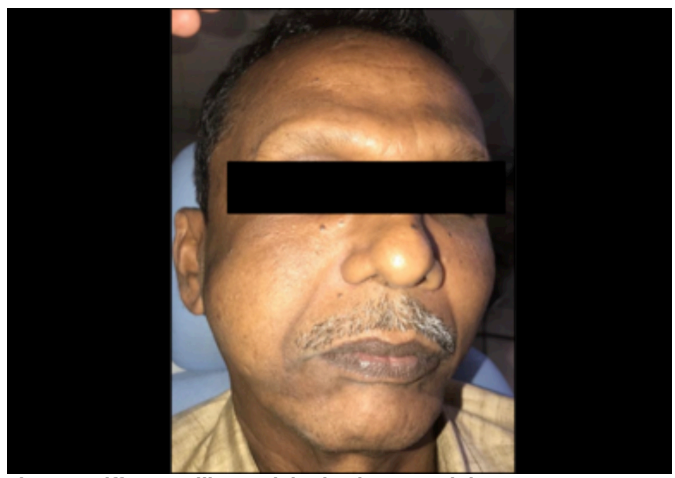

Figure 1: Diffuse swelling on right cheek. External view.

CASE REPORT

A 62-year male reported to the Department of Oral Medicine and Radiology, with the complaint of a painless lump in the right cheek for the previous 4 years (Figure 1). The swelling was small initially which gradually enlarged to the present size. It was not associated with any other symptoms and did not interfere with speech or mastication. His medical, family and dental histories were not relevant. The patient was a cigarette smoker for the last 40 years, well-built and nourished. His vital signs were within limits.

On examination, a diffuse swelling measuring approximately

4×3 cm was present on the right cheek. The swelling was centred over the right cheek without obliterating the nasolabial fold. Overlying skin was of normal colour and the surface of the swelling appeared smooth with no visible pulsations. On palpation, the swelling was non-tender, firm in consistency and not fixed. Overlying skin was pinchable and no pulsations were felt.

Intraorally, the swelling was evident on the right buccal mucosa with no secondary changes of sinus opening or ulceration (Figure 2). The swelling was non-tender, non-pulsatile, firm in consistency and mobile. There was no obliteration of the upper and lower buccal vestibule. His periodontal status was poor and salivary flow was satisfactory.

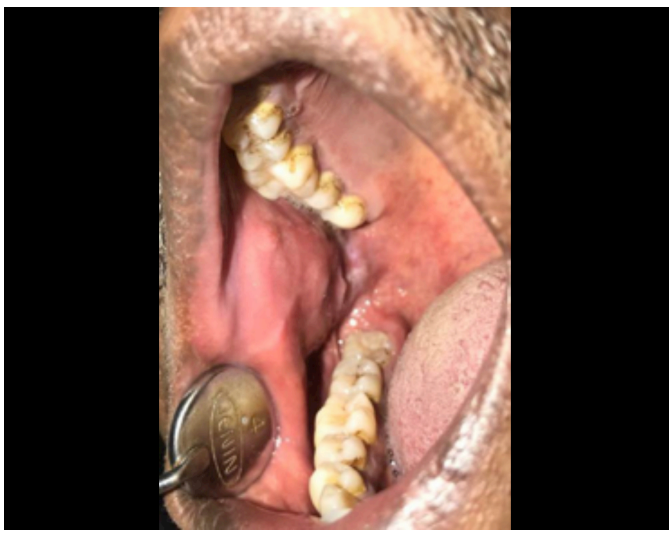

Figure 2: Intraoral examination shows a swelling on the right buccal mucosa.

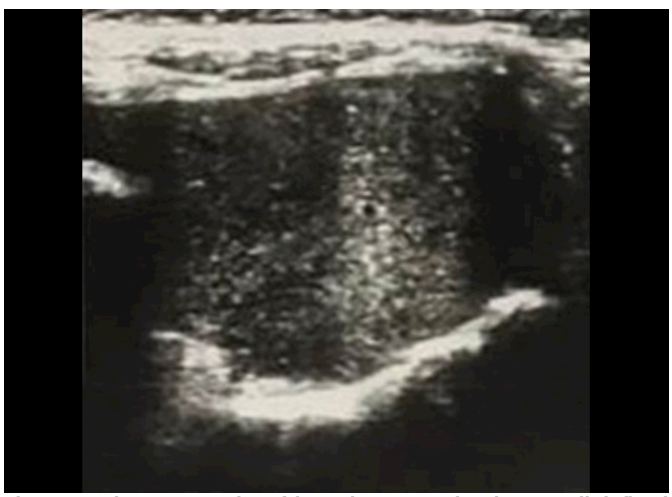

Figure 3: Ultrasonography of buccal mucosa showing a well-defined hypoechoic lesion with homogenous low-level internal echoes.

The differential diagnosis considered included benign salivary gland tumour and mesenchymal neoplasms. On orthopantomograph (OPG), there were no significant bone changes related to this lesion. Ultrasound scan showed a well-defined hypoechoic lesion measuring 4.3×2.7×3.4 cm with homogenous low-level internal echoes. (Figure 3). Colour Doppler study ruled out vascular lesion. Parotid gland and Stenson duct were normal.

Fine needle aspiration cytology of the swelling showed numerous desquamated epithelial cells and mixed inflammatory cells. An incisional biopsy was performed. The histology showed a cystic lesion with lumen and capsule. The lining displayed parakeratinisation with surface corrugation, 4-6 cell layer thick stratified squamous epithelium and basal cell palisading. The capsule showed bundles of collagen fibres. No skin appendages were seen in the cyst wall (Figure 4 a-c). These findings were correlated with clinical presentation and the final diagnosis of POKC of right buccal mucosa was reached. A few weeks later, surgery was carried out under general anaesthesia with complete removal of the cyst and the patient was put under periodic follow-up.

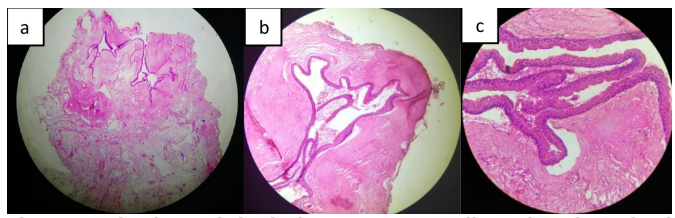

Figure 4: Histology of the lesion. A. Hematoxylin and eosin stained sections showing cystic lesion with lumen and capsule. b. The capsule with bundles of collagen fibers and corrugated lining. c. Parakeratinised lining with surface corrugation, 4-6 cell layer thick stratified squamous epithelium and basal cell palisading.

DISCUSSION

POKC is a rare variant of OKC's intraosseous counterpart but occurs in the soft tissues without significant bone involvement. Very rarely, they are found in non-gingival soft tissues of the oral cavity.

The histogenesis of POKC occurring in the buccal region is ambiguous. The presence of the displaced remnants of dental lamina in buccal mucosa and their persistence during embryogenesis is essential for such ectopic pathology.[6] During the embryological stage, the developing deciduous dentition and the oral vestibule maintain close proximity. Additionally, this stage is characterised by repeated reciprocal migration of the embryonic structures involved in the formation of the prospective oral vestibule and buccal mucosa with the odontogenic epithelium around the upper molar areas.[7,8] These observations support the ectopic histogenesis of a keratocyst with the features of OKC in the buccal tissue.

However, there is a controversy whether non-gingival soft tissue OKC truly represents a peripheral counterpart of the intraosseous OKC. Authors have related odontogenic or epidermal origin for the keratocysts in the buccal mucosa, making the histogenesis ambiguous.[9] Epidermal structures like sebaceous glands are not uncommon occurrences on the buccal mucosa in the form of Fordyce spots. The epithelial lining of OKC, shows a striking resemblance with the epithelial lining of the sebaceous gland duct.[5] POKC may be distinguished from skin lesions such as trichilemmal cysts and steatocystoma by its hair growth pattern (Catagen phase) and the presence of pilosebaceous units, respectively.[10] However, non-syndromic cutaneous keratocysts are histologically indistinguishable from POKCs.[10,11]

Ida *etal*. proposed a nonspecific term, "mucosal keratocyst," for the cystic lesions of buccal location that are difficult to distinguish from OKC and have ambiguous histogenesis.[12] Major flaw in the sebaceous ductal origin theory of buccal keratocyst is the case reports of other odontogenic tumours reported in the buccal mucosa, for example, ameloblastomas.[12]

Epidermoid cyst exhibits similar histological features as POKC. However, the preferred site for epidermoid cyst is along the line of closure of the embryonic fusion plane unlike POKC.[9] Histologically, the lining epithelium of POKC shows parakeratinisation with a corrugated surface and characteristic palisading of cuboidal basal cells as observed in this case. Additionally, a different immunohistochemical pattern of cytokeratin expression is also helpful for differential diagnosis. CK10, which is a specific marker for cornified structures like skin, will be negative in POKC. On the contrary, CK17, which is positive in POKC, will be negative in epidermoid cysts.[12]

Even though the histogenesis of POKC in the buccal mucosa is ambiguous, an odontogenic origin is suspected based on the typical histological pattern of the lesion.

PATIENT'S CONSENT:
Informed consent was obtained from the patient.

COMPETING INTEREST:
The authors declared no competing interest.

AUTHORS' CONTRIBUTION:
VK: Concept, data collection and interpretation, literature search, and manuscript writing.
PM: Concept, data collection, and literature search.
PC: Concept.
All the authors have approved the final version of the manuscript to be published.

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