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Case Report

Dentistry Section

Epidermoid Cyst of the Buccal Mucosa: A Case Report and Review of Literature

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ABSTRACT

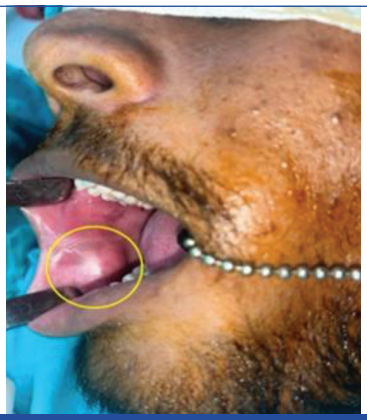
Epidermoid Cysts (ECs) are rare, benign cystic lesions that result from surface epithelium entrapment or, more frequently, from abnormal infundibular epithelium repair following a flare-up of follicular inflammation. They occur anywhere on the body, but are more commonly found on the neck, scalp and torso. In the head and neck region, ECs constitute 7% out of which only 1.6% are reported in the oral cavity. The most common sites for oral ECs include the floor of the mouth, lips, palate, tongue and jaws. Here, we present a case of EC of the buccal mucosa in a 21-year-old male. The patient reported with a swelling on the right buccal mucosa. After clinical examination, it was diagnosed as a cystic lesion. The lesion was excised under general anaesthesia. Histopathological examination showed the presence of keratinised stratified squamous epithelium and a cystic lumen filled with keratin. There was no evidence of dermal appendages. Hence, a final diagnosis of EC of the buccal mucosa was made. The ECs are common in the skin (scalp, back, trunk) but rare in the oral cavity. The novelty of the case lies in the extremely rare intraoral occurrence of an EC in the buccal mucosa, a site seldom reported in the literature. Thereby, highlighting the importance of considering this entity in the differential diagnosis of buccal swelling like mucocoele, lipoma, fibroma and minor salivary gland adenoma.

Keywords: Cyst, Epidermal inclusion cyst, Epidermoid cyst, Keratin, Oral cavity

CASE REPORT

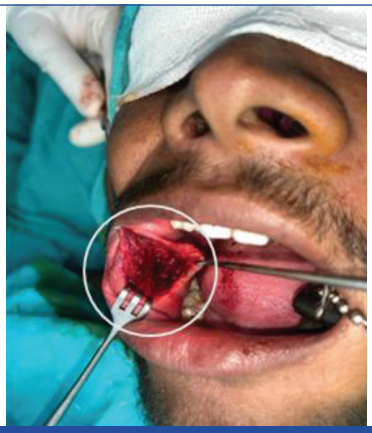
A male patient, aged 21 years, reported to the institute with an intraoral swelling of the buccal mucosa on the right side for six months. The patient did not have any associated habits. The swelling was pea-sized initially and had progressively increased to the current size of 2×2 cm. There was no associated pain or trauma. No pus discharge or bleeding was observed. He had no underlying medical conditions or any drug allergies.

On examination, a smooth, nodular, sessile swelling was seen over the right buccal mucosa of approximately 2×2 cm, which was pinkish and round, with well-defined borders and a smooth surface [Table/Fig-1]. The local temperature was not raised on palpation, and the consistency was soft to firm. There was no tenderness or induration present.

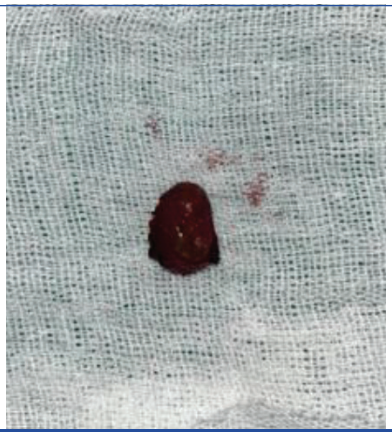


[Table/Fig-1]: Intraoral photograph of patient showing lesion over right buccal mucosa (yellow circle).

The lesion was nodular, soft to firm and fluctuant without any pus discharge. Based on these findings, a clinical diagnosis of a lipoma on the right side of the buccal mucosa was made. Fibroma, epidermoid cyst, dermoid cyst, sebaceous cyst, and mucocoele were considered for differential diagnosis. After haematological investigations, excision of the lesion was done in toto under general anaesthesia [Table/Fig-2], and the sample was sent for microscopic assessment [Table/Fig-3].

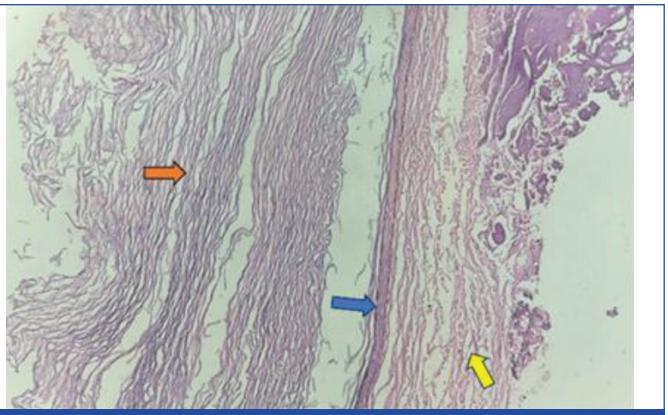


[Table/Fig-2]: Photograph showing complete excision of cystic lesion (enucleation) under general anaesthesia (white circle).



[Table/Fig-3]: Gross examination: Single, oval, reddish in colour, soft to firm in consistency, excised tissue specimen received.

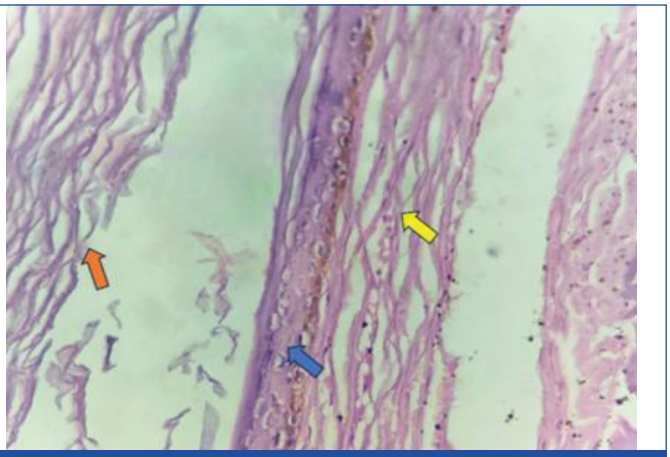
The haematoxylin and eosin slides showed the presence of a cystic cavity lined by 2 to 3-layered keratinising stratified-squamous-epithelium with abundant keratin within the cystic lumen [Table/Fig-4]. The cystic wall was comprised of collagen fibers, fibroblasts and inflammatory cell infiltrate. There was no evidence of adnexal structures or skin appendages.



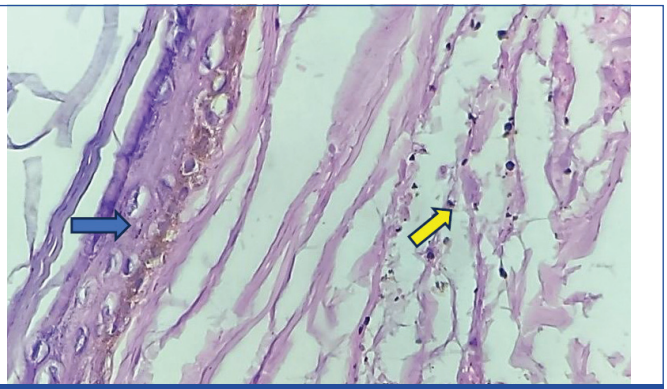
[Table/Fig-4]: Haematoxylin and eosin-stained section showing cystic lining (blue arrow), surrounding connective tissue wall (yellow arrow) and abundant keratin within the cystic lumen (orange arrow) at 10x.

Under high magnification [Table/Fig-5,6], all features of low magnification- 10x were confirmed.

These histopathological findings were suggestive of an EC. The surgery had a favourable outcome, and on a six-month follow-up, the patient presented with no signs of infection/recurrence.



[Table/Fig-5]: Haematoxylin and eosin-stained section showing keratinised stratified squamous epithelium (blue arrow), surrounding connective tissue wall (yellow arrow) and abundant keratin within the cystic lumen (orange arrow) at 20x.



[Table/Fig-6]: Haematoxylin and eosin-stained section showing keratinised stratified squamous epithelium (blue arrow), surrounding connective tissue wall (yellow arrow) at 40x.

DISCUSSION

The ECs, also called inclusion cysts, are non-cancerous cystic lesions originating from the germinal epithelium. They are formed either because of the entrapment of the surface-epithelium or due to the abnormal or unusual healing of the infundibular epithelium, which mainly occurs when the hair follicle undergoes inflammation [1]. The common sites of EC include the facial region, neck, trunk and back [2,3]. The head-neck region comprises 6-7% of ECs and the oral cavity accounts for 1.6% [4]. ECs are seen at different sites in the oral cavity, including the floor of the mouth, lips, palate, jaws and tongue [1].

The ECs occur in individuals from 15 to 35 years of age and also show a male predominance. They arise from the hyperplasia of the infundibular epithelium as a reaction to inflammation of the hair follicle [5]. They are usually asymptomatic and painless unless secondarily infected [6]. Clinicians still have significant challenges when diagnosing ECs because of their non-specific clinical presentation, which can mimic other diseases or conditions. An anatomo-pathological examination is required for the definitive diagnosis of the lesion [6].

ECs mainly develop in those regions of the body where embryonic elements merge. The most frequently affected area is the floor of the mouth. In contrast, the buccal mucosa is an uncommon location [7].

Roser initially reported EC in the oral cavity in 1859. ECs are infrequent, noncancerous cystic lesions resulting from surface epithelium being trapped in or, more frequently, from abnormal infundibular epithelium healing, usually due to follicle inflammation [8].

The ECs can arise in any part of the body and account for <0.01% of all oral cavity cysts. Their incidence ranges between 1.6-6.9% in the oro-facial region and 1.6% in the mouth [9].

ECs are categorised into congenital or acquired types based on the pathophysiology. The former is believed to arise from the congenital inclusion of ectodermal tissue while embryonic development occurs. The acquired type, originally called “implantation cysts”, is thought to result from the accidental or surgical implantation of the epithelium into deeper tissues of the mesenchyme. The history of previous surgery and trauma are the two main contributing factors in developing acquired types of ECs [10].

Clinically, ECs manifest as firm, fluctuating, painless, slow-growing masses, often ranging from 1 to 4 cm. Even if they are congenital, they are usually discovered in the 2nd and 3rd decades. They do not interfere with functionality and are considered asymptomatic unless their size causes disturbance. Some uncommon symptoms like headache, obstructive-sialadenitis, and facial deformity have also been documented in the neck and head [11].

The mouth floor comprises 25% of ECs in the oral cavity. Other reported sites include lips, jaws, palate, tongue and pterygopalatine fossa [1]. The buccal mucosa is an uncommon site of occurrence. Here, we have described a case of EC in a male, aged 21 years, who complained of swelling for six months on the right buccal mucosa.

A few cases of ECs of buccal mucosa present in the literature are mentioned [Table/Fig-7] [6,7,12-16].

Author(s)	Year	Gender	Age of the patient in years	Onset	Buccal mucosa (left/ right)	Size of lesion in cm
Ozan F et al., [12]	2007	Female	38	Six months	Left-side	2×3×4
Kini YK et al., [13]	2013	Male	25	Two years	Left-side	1.5×1.5×1.5
Costa FWG et al., [7]	2015	Male	29	Four years	Right side	3.5
Srivastava A et al., [14]	2015	Male	35	Three months	Bilateral	1.5×1.5
Chakraborty PS et al., [15]	2018	Male	82	One year	Left-side	4.5×2
Rohde RL et al., [16]	2019	Female	2	Two years	Left-side	4.5×3.5×1.5
Dammak N et al., [6]	2021	Male	56	Five years	Left-side	3.4×3.1×2.1
Present case	2024	Male	21	Six months	Right-side	2×2

[Table/Fig-7]: Review of cases of Epidermoid Cyst (EC) occurring on the buccal mucosa [6,7,12-16].

The ECs commonly develop in young adults, mainly in the second and third decades. Men tend to be twice as commonly affected as women, with a ratio of 3:1 [17,18].

Although ECs are mostly benign, they may progress into malignancy over the years if left untreated [19]. A few malignant transformations evolving from ECs have been described in the literature, including Basal-Cell-Carcinoma (BCC), Bowen disease, and Squamous-Cell-Carcinoma (SCC) [18]. Dini M et al., in 2001, reported a case of BCC, which developed in the wall of an EC [20]. They can also be seen in Gardner's syndrome in isolated or multifocal form [21].

Soft-tissue swellings, including dermoid cyst, lipoma, neurofibroma, myxoma, irritational fibroma, and neurilemmoma, should be considered in the differential diagnosis of ECs as they present a similar clinical presentation to ECs [1].

Histopathological assessment is the gold standard for differentiating ECs in these cases. On histology, ECs show a cystic cavity lined by keratinising stratified-squamous epithelium with abundant keratin in the lumen. They are called "pearly tumours" because of the cyst's smooth, shiny, waxy keratin content. In contrast to the dermoid cyst, ECs lack the presence of dermal appendages like sebaceous glands, hair follicles and sweat glands. Histopathologic/microscopic assessment unquestionably is the primary aid of diagnosis in dermoid and ECs [22].

Large cysts may cause pain and infection; hence, needle aspiration and fenestration are usually not advised [23]. ECs have a low recurrence rate of <3%; however, it can be prevented by enucleation or complete cyst removal [24]. In this case, excision of the cyst was done in toto under general anaesthesia.

This case highlights the incidence of EC on buccal mucosa, which is an unusual site of occurrence. For proper treatment, a thorough examination, including the site of occurrence along with the symptoms, is of utmost importance. ECs mimic other lesions like dermoid cysts and sebaceous cysts. Histopathology remains the gold standard in the diagnosis of such lesions.

CONCLUSION(S)

The ECs are less frequently seen in the oral cavity. These cysts have a malignant potential, so a close follow-up is required. Prompt diagnosis and eradication of ECs have critical importance. ECs should also be included in the differential diagnosis of benign soft-tissue tumours. Histopathological evaluation is the gold standard for diagnosing ECs. Enucleation or complete excision of the cyst remains the standard mode of treatment.

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