



Epidermoid Cyst in an Unusual Location: A Case Report

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ABSTRACT

Epidermoid cysts account for approximately 80% of follicular skin cysts. They are considered a benign condition that can occur anywhere in the body; in the microscopic examination, this cyst reveals a cavity lined by stratified squamous epithelium resembling an epidermis, a lumen filled with degenerating orthokeratin. It is sporadic in the oral cavity, representing only 1.6% of all cysts. Its appearance in the buccal mucosa has been poorly reported. The purpose of this article is to describe a case of a 22-year-old male with an epidermoid cyst in this unusual location. This entity should not be ruled out of lesions that may occur in the oral cavity, even when its incidence is extremely low.

1. Introduction

Epidermoid cysts (EC) account for approximately 80% of follicular skin cysts and are considered benign conditions that can occur anywhere in the body.^[1] EC can arise at any age, but it is more frequent adulthood; it is more predominantly found in males. The face, neck, periauricular area, and upper trunk are more commonly involved. EC are fluid-filled protrusions originating from the follicular infundibulum and lying just under the surface of the skin. Lesions usually occur spontaneously; however, implantation of the epithelium resulting from trauma is considered an etiologic factor.^[2] Rarely, such epidermal inclusion can develop in the mouth.^[3] It may result in a complicated, correct diagnosis of these lesions in the oral cavity due to its unusual presentation. To achieve a proper diagnosis and develop a correct surgical strategy, specialized imaging examinations such as ultrasonography, computed tomography, magnetic resonance imaging, and histopathological examination, should be carried out.^[1] In a microscopic examination, this cyst reveals a cavity lined by stratified squamous epithelium resembling an epidermis, with the lumen filled with degenerating orthokeratin.^[3] The purpose of this article is to describe a rare case of an epidermoid cyst in the right buccal mucosa of a 22-year-old male as well as a review of the literature.

2. Case presentation

A 22-year-old male patient attended our service, presenting a painless tumefaction in the right facial region with an undefined time of evolution. His medical record was unremarkable. A nodular lesion in the proper oral mucosa, firm, well-circumscribed, and painless on palpation was evidenced on clinical

examination. An isodense, encapsulated, well-circumscribed unilocular lesion was observed in the right lateral buccal mucosa on the CT scan. Among the differential diagnoses, lipoma and a salivary gland tumor were managed, so it was decided to perform excisional biopsy intraorally under local anesthesia. The cystic lesion was enucleated using a vertical incision at the right buccal mucosa surface, obtaining a 1.5 x 1.4cm sample of brownish coloration with an irregular surface and texture (Figure. 1).



Figure 1. Sample of brownish colouration with an irregular surface and texture.

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The histopathological study (hematoxylin-eosin stain 400X) described a cystic cavity lined by orthokeratinized stratified squamous epithelium and a lumen partially filled orthokeratin. A fibrous connective tissue wall was also observed (Figure. 2).

A final diagnosis of EC was determined. After two years of follow-up, the patient has not shown any clinical or imaging signs of recurrence.

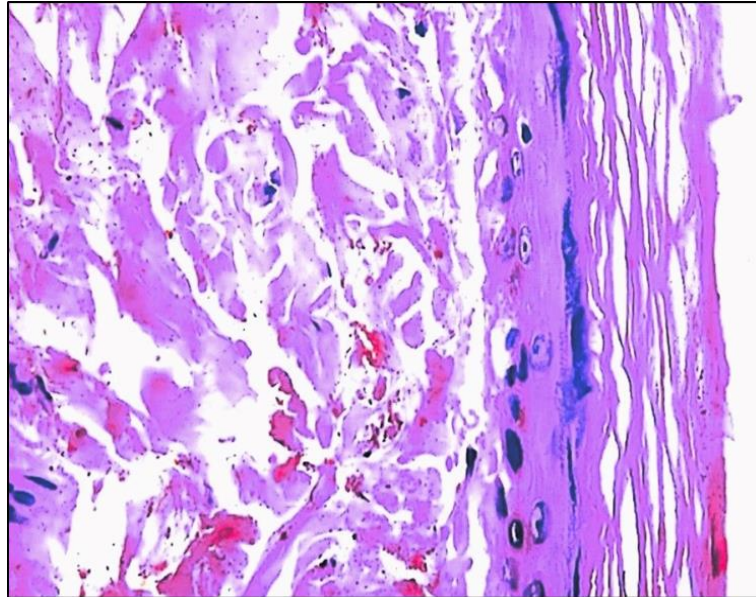


Figure 2. Photomicrograph of the histological cut (hematoxylin-eosin stain 400X). The cystic cavity lined by orthokeratinized stratified squamous epithelium and lumen partially filled by orthokeratine is observed and the fibrous connective tissue wall.

3. Discussion

EC are fluid-filled protrusions originating from the follicular infundibulum.^[2] Lesions usually occur spontaneously; however, implantation of the epithelium resulting from trauma is considered an etiologic factor, and rarely, such epidermal inclusions can develop in the mouth.^[3]

The cyst wall is lined with stratified squamous epithelium. Therefore, the peeling of keratin layers will accumulate inside the cyst. A noticeable characteristic of this pathology is that the cyst can communicate with the skin surface through a keratin-filled orifice, so-called punctum. EC may also occur due to obstruction of the follicular orifice, as seen in patients with acne vulgaris.^[2]

EC can occur in a wide age range, from 10 to 45 years of age, but most

typically arise in the third and fourth decades of life.^[4, 5] EC can be found anywhere in the body, but are most commonly seen on the face, neck, chest, upper back, scrotum, and genitals.^[6] Janarthanam and Mahadevan^[7] showed that 7% of these cysts occur in the head and neck; the oral cavity representing only 1.6%. Different studies agree that when this cyst appears in the oral cavity, the most frequent location is the floor of the mouth 1, 4, 8, 9. Other numerous areas are the lingual lateral border, lateral pharyngeal wall, the soft palate, and mandible.^[4] The most frequent intraoral locations of this entity reported in the literature at the time of this review are shown in Table 1. EC is predominantly found in males, as in our case.

Table 1. Review of the literature about the most frequent locations of EC in the maxillofacial region.

| AUTHOR | YEAR | TITLE | INVESTIGATION TYPE | LOCATION | TREATMENT |
|---------------------------------------|------|---|--------------------|-----------------|-------------------|
| Yosuke Sakamoto et al. ^[4] | 2014 | A case of an epidermoid cyst arising in the maxillary sinus. | Case report | Maxillary sinus | Surgical excision |
| Janarthanam et al. ^[7] | 2012 | Epidermoid cyst of the submandibular region | Case report | Floor of mouth | Surgical excision |
| Tanaka et al. ^[8] | 2011 | Large epidermoid cyst in lateral floor of mouth and submandibular region. | Case report | Floor of mouth | Surgical excision |
| Zeeshan et al. ^[9] | 2016 | Sublingual epidermoid cyst mimicking as plunging ranula – A case report. | Case report | Floor of mouth | Surgical excision |

| | | | | | |
|------------------------------------|------|---|-----------------------------------|----------------|-------------------|
| Koca et al. ^[10] | 2007 | Epidermoid cyst on the floor of the mouth: report of a case. | Case report | Floor of mouth | Surgical excision |
| Turetschek et al. ^[11] | 1995 | Case report: epidermoid cyst of the mouth floor: diagnostic imaging by sonography, computed tomography, and magnetic resonance imaging. | Case report | Floor of mouth | Surgical excision |
| Jham et al. ^[12] | 2007 | Epidermoid Cyst of the Floor of the Mouth: A Case Report. | Case report | Floor of mouth | Surgical excision |
| Da Silveira et al. ^[13] | 2020 | Intraoral Epidermoid Cyst With Extensive Elastofibromatous Changes: An Extremely Rare Finding. | Case report | Floor of mouth | Surgical excision |
| Papanayotou et al. ^[14] | 1977 | Epidermoid implantation cyst of the lower lip: report of a case. | Case report | Lip | Surgical excision |
| Phukan et al. ^[15] | 2014 | Cytodiagnosis of epidermoid cyst of the upper lip; A common lesion in an uncommon site. | Case report | Lip | Surgical excision |
| Kuroyanagi et al. ^[16] | 1973 | Epidermoid cyst of the upper lip: report of a case. | Case report | Lip | Surgical excision |
| Maranhão et al. ^[17] | 2011 | Atypical appearance of epidermoid cyst in tongue's ventral surface. | Case report | Tongue | Surgical excision |
| Gurgel et al. ^[18] | 2015 | Epidermoid Cyst Arising in the Buccal Mucosa: Case Report and Literature Review. | Case Report and Literature Review | Buccal Mucosa | Surgical excision |
| Ozan et al. ^[19] | 2007 | Epidermoid Cyst of the Buccal Mucosa: A Case Report. | Case Report | Buccal Mucosa | Surgical excision |
| Costa et al. ^[20] | 2020 | Epidermoid Cyst Of The Buccal Region Of The Face | Case Report | Buccal Mucosa | Surgical excision |

Even though the intraoral location at the mouth level has been the most frequently documented, EC in the buccal mucosa has been poorly reported. In fact, in 2014, Gurgel et al.^[18] conducted a review regarding EC located in this area, which yielded only seven cases described in the world literature. Ozan et al.^[19] reported a case of a 38-year-old female patient with a diagnosis of EC and only mentioned two other cases of this pathology in the left buccal mucosa. In their case, the lesion's excision was done under local anesthesia, and the patient was followed up for 18 months, without showing signs of recurrence. Their approach is very similar to the one used in the present study, where no reproduction was observed after two years. CT scans play an essential role in diagnosing epidermoid cysts and determining the surgical strategy. CT usually demonstrates a well-encapsulated mass of various densities representing a mixture of fat and keratin.^[21] On ultrasound, the lesions appear round or oval, well-circumscribed, and avascular.^[2] In our case,

the tomographic study revealed an isodense, encapsulated, well-circumscribed, and unilocular lesion in the right lateral buccal mucosa. Through this imaging method, the authors established the location, size, and differential diagnoses (lipoma and salivary gland tumor) of the existing lesion and decided to practice an excisional biopsy intraorally under local anesthesia. The biopsy needs to be done to confirm the diagnosis. Histologically, an EC is lined by an epithelial cell wall. This epithelium is stratified squamous epithelium resembling the epidermis and includes a granular layer and keratin lamellae in the lumen. In contrast, "true" dermoid cysts contain skin adnexa such as hair follicles, sebaceous, and sweat glands.^[22]

There is scarce information about the immune profile of oral EC. It is reported to possess negative immunoreactivity to apoptosis-related molecules (ssDNA, cleaved Lamin A, gamma-H2AX, and cleaved caspase-3). CD138

is expressed in the squamous epithelium (mainly in the basal and spinous layers) but not in the keratinizing components.^[23] Terada^[24] and Gurgel et al.^[18] observed positive marking for the CK5/6 and CK34BE14 in the EC. Histological and immunohistochemical staining patterns differentiate epidermoid cysts from other types of cystic lesions, although the usual diagnosis for EC is based on histopathological findings. In the present case, the diagnosis was clarified with a histopathological study; Hematoxylin-eosin staining revealed the classic features of EC (cystic cavity lined by orthokeratinized stratified squamous epithelium and a lumen partially filled by orthokeratin). Even though immunohistochemistry was not necessary in the present case, it is essential to remark that this study helps clarify histopathological diagnoses for the management head and neck tumors, mainly when dealing with sarcomas and lymphomas, to establish an adequate classification and treatment.^[25] Future research on this type of cyst should be reported to contribute to recording the immunohistochemical profile of oral EC. Small uncomplicated cysts usually do not need treatment. Removal may be accomplished by simple complete surgical excision of the cyst with the cyst wall intact. A local anesthetic with epinephrine is preferred to minimize bleeding. The drug should be injected around the cyst, with avoidance of direct injection into the cyst.^[26] The complete excision should be delayed if an active infection is present as the dissection planes will be difficult. An alternative surgical approach can also be made with a punch biopsy and expulsion of the intact cyst through the small defect. If the entire cyst wall is not removed, the cyst may recur. Cysts are more challenging to remove once they have ruptured.^[27] CO₂ or erbium-YAG-laser^[2] may treat some small cysts.^[2] To preserve aesthetics, this entity's approach, if possible, must be carried out intraorally^[20], as in this case. However, occasions where the extraoral method should be practiced due to the location and size of the lesion. The low frequency of the EC in the oral cavity (and even more unusual in the buccal mucosa) exposes why this entity is rarely included among the differential clinical diagnosis of lesions in the oral cavity. We presented a very particular case of EC on the right buccal mucosa, where an excisional biopsy was performed due to the benign nature of the differential clinical diagnoses and the behavior that the lesion presented. This entity should not be ruled out of lesions that may occur in the oral cavity, even when its incidence is extremely low. During the literature review developed for our research, no similar case reported in Latin America was found.

4. Conclusion

Due to the scarcely reported cases in the literature, thinking about an epidermoid cyst in the oral cavity is unusual. Even less frequent at the level of the buccal mucosa. However, imaging studies, clinical history, and histopathology may confirm this entity's presence, and thus, as shown in our study, it should not be ruled out of the maxillofacial diagnostic repertoire.

Conflict of Interest

The authors declared that there is no conflict of interest.

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