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Epidermoid Cyst of the Buccal Mucosa: A Case Report

Fatih Ozan, DDS; Hidayet Burak Polat, DDS;
Sinan Ay, DDS, PhD; Fahrettin Goze, MD

Abstract

Aim: The purpose of this article is to describe a rare case of an epidermoid cyst in the buccal cheek region and a review of the literature.

Background: Dermoid and epidermoid cysts are developmental pathologies occurring in the head and neck region with an incidence ranging from 1.6% to 6.9%, and both cysts reported in less than 0.01% of all oral cavity cysts.

Report: A rare case of an epidermoid cyst originating from the buccal mucosa in a 38-year-old woman with a complaint of swelling and facial asymmetry in the left cheek just distal to the commissure for six months is presented in this report.

Keywords: Cyst, epidermoid cyst, buccal mucosa

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Introduction

Dermoid cysts can be found anywhere in the body, particularly in areas where embryonic elements fuse together. Most cases have been reported in the ovaries, the testicles, as well as the hands and feet. Dermoid and epidermoid cysts in the mouth are uncommon and account for less than 0.01% of all oral cysts.[1-2] The great majority arise in the floor of the mouth, but there are rare and usually individual case reports of examples in other sites.

Epidermoid, dermoid, and teratoid cysts are nonodontogenic cystic lesions. They are rare lesions derived from germinal epithelium. While a dermoid cyst has an epidermal lining with skin adnexa such as hair follicles and sebaceous glands, the epidermoid cyst contains no such adnexa. Teratoid cysts have been rarely described in the floor of the mouth. These cysts contain respiratory, gastrointestinal, and connective tissues such as bundles of striated muscle and distinct areas of fat.[1-4]

The purpose of this article is to describe a rare case of an epidermoid cyst in the buccal cheek region of a 38-year-old woman and a review of the literature.

Case Report

A 38-year-old woman had a conspicuous swelling of the left cheek, just behind the commissure (Figure 1).

The patient reported the swelling had been present for about six months. She had been prescribed an antibiotic by a general dental practitioner but stated the antibiotic had not helped. She had no history of surgery and/or trauma related to the lesion location.

Intraoral examination revealed a 2 x 3 x 4 cm swelling extending from the left commissure to the first molar region. The swelling was not tender to palpation and formed a doughy mass freely mobile between the buccal mucosa and the buccinator muscle. The overlying mucosa was normal in color and texture with no apparent abnormality of the facial skin in the area of the swelling. Submental or submandibular lymph nodes were not palpable.

Under local anesthesia, the cyst was enucleated using a horizontal incision in the buccal mucosa (Figure 2).

Figure 1. Extraoral appearance of the patient.

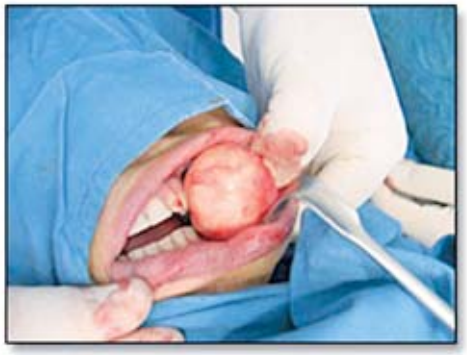

Figure 2. Intraoperativeappearance of the cyst.

The wound was closed with 3.0 silk sutures. The postoperative period was uneventful and healing was good.

Histopathological examination of the surgical specimen revealed a keratinized squamous epithelial lining with the inner surface lined with keratin lamellas and the outer surface lined with gingival connective tissue components (Figure 3, HE X10). In a detailed examination keratin lamellas, epithelium, epithelial lining, and fibrovascular gingival tissue were observed (Figure 4, HE, X25).

The patient has shown no clinical or radiographic evidence of recurrence during 18 months of follow-up evaluation.

Discussion

Dermoid cysts are classified into the following three categories[3-6]

1. Epidermoid cysts - the cystic cavity is lined with epithelium without skin appendages;
2. Dermoid cysts - the cystic cavity includes skin appendages such as hair, hair follicles, sebaceous, and sweat glands;

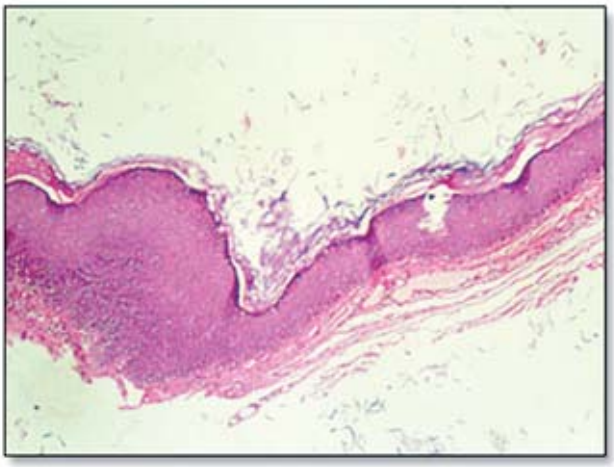

Figure 3. Microscopic appearance ofcyst (HE X10).

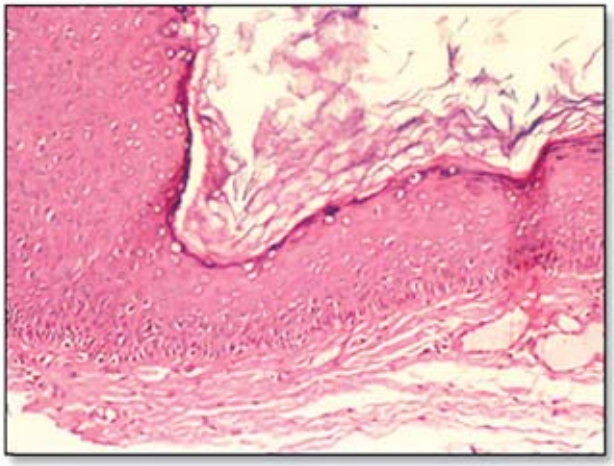

Figure 4. Increased magnification of an area of Figure 3 (HE X25).

3. Teratoid cysts - in addition to skin appendages the cystic cavity elements of the mesoderm can be found such as bone and muscle as well as gastrointestinal and respiratory tissue.

Dermoid cysts of the floor of the mouth are classified topographically as sublingual and submental in relation to the mylohyoid muscle.

Dermoid and epidermoid cysts are uncommon in the mouth. The incidence in the head and neck has been reported to range from 1.6% to 6.9%.[1,2,4,5,7] Most reported cases have involved the floor of the mouth (sublingual dermoids), usually in the midline.[2-8] Rare cases have been reported in the tongue,[1] lips,[1] uvula,[9] temporomandibular joint dermal graft,[10] intradiploic,[11] intracranial,[12] and intraosseously within the mandible and maxilla;[1] only two case reports were presented[1,13] associated with an epidermoid cyst of the buccal mucosa.

The etiology of these cysts is essentially unknown. The most popular theory is epithelium being sequestered in lines of fusion of embryonic processes[1,2,8] as a result of epithelial tissue being traumatically implanted in utero[1] or to the traumatic implantation of a piece of epidermis.[6,14-16] Implantation cysts were first recognized by Werhner[17] in 1855, originally referred to by Sutton[18] in 1895, and believed to originate through implantation of epithelium by either surgical or accidental trauma into deeper mesenchymal tissues. Since trauma is said to always precipitate in the formation of the implantation-type epidermoid cyst, King[19] preferred the term 'post traumatic cyst.' Similar observations of cyst formation have been made in humans following the inclusion of epithelium in surgical wound sites.[1,14,16]

However, this traditional view of processes fusing with the breakdown of the intervening epithelium has been challenged for this theory does not explain the presence of adnexa in dermoid cysts or the absence of cysts of this type in zones of known fusion such as the palate. Shafer and co-authors[20] postulate cysts of this type in the floor of the mouth arise from totipotent blastomeres trapped during closure of the mandibular and hyoid branchial arches. However, none of the traditional explanations account for the present case.

Although the epidermoid cyst rarely discloses malignancy, isolated cases of premalignant and malignant conditions (Bowen's disease, Paget's disease, and squamous cell carcinoma) have been found in their walls. Dini et al.[21] described a patient with basal cell carcinoma arising in the wall of an epidermoid cyst. Ikeda et al.[21] presented a case report stating basal cell carcinoma originated from an epidermoid cyst in which they found nests of basal cell carcinoma connected with the epidermoid cyst and partially replacing the cyst wall. Lopez-Rios et al.[22] described a patient with squamous cell carcinoma arising in

the wall of an otherwise conventional epidermoid cyst. An incorrect diagnosis could result in inappropriate therapy. If the lesion is completely excised, the treatment is definitive.

In making a differential diagnosis the clinician should first entertain a broad variety of conditions. Conditions resembling this clinical presentation are swelling of the face as odontogenic infection, buccal space infection, masseteric space infection, and dermoid cysts. These conditions can be classified as developmental, neoplastic, and infectious processes. Infectious processes, as odontogenic infection, buccal space infection, and masseteric space infection, are unlikely in this case because the lesion/mass has attained considerable size without constitutional symptoms such as malaise or fever and there was no response to antibiotic treatment. A salivary obstruction with a mucocele or ranula formation is a possibility. A bluish-gray hue is characteristic of a ranula, but the lesion in the present case had an overlying mucosa which was normal in color.

A primary malignant process is very doubtful owing to the lesion's size, normal overlying mucosa, cystic homogeneity, and lack of nodal involvement. Benign neoplastic processes in this region may include lipoid, salivary, and vascular lesions. A lipoma would tend to be yellowish and nodular. A vascular lesion such as a hemangioma or lymphangioma is also unlikely for these are usually lobulated and have an irregular mucosal surface. One would expect an obvious reddish-purple color (hemangioma) or almost clear translucent color (lymphangioma) with lesions of this magnitude.

Most salivary gland tumors probably cause an obstructive phenomena when they reach this size. If this were a cystic hygroma, one would expect it to be present at birth (50%) or develop by age two (90%). Cystic hygromas are more common in the posterior triangle of the neck and are soft, fluctuant masses. The most frequent oral site is in the anterior two thirds of the tongue.

A lesion to consider in the developmental category is the oral lymphoepithelial cyst. This cyst is an uncommon lesion developing within oral lymphoid tissue. It can occur anywhere within or adjacent to Waldeyer's ring. The oral lymphoepithelial cyst presents as a small (small than 1 cm), smooth, whitish-yellow, firm, painless mass. It often contains cheesy keratinaceous material in the lumen. Because of the size, localization, and hue, the lesion presented this diagnosis was unlikely.

Summary

The differential diagnosis for an epidermoid cyst should include developmental, neoplastic, and infectious lesions. The developmental lesion was the most compatible category for this patient who presented with a large, slowly enlarging, smooth, doughy, painless lesion.

The diagnosis between an epidermoid and dermoid cyst was initially considered and later confirmed through histopathological examination of the surgical fragment. The cyst's features characterized by stratified squamous epithelium with laminas of keratinization on the surface and lumen of the cyst cavity determine the histological classification. The lack of epithelial adnexa excludes the diagnosis of dermoid cyst.

The proposed treatment of epidermoid cysts is surgical removal.[3,8,23] The cyst removal procedure was simple and effective, and its success was confirmed by the lack of postsurgical alterations and no recurrence of the lesion.

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About the Authors

Fatih Ozan, DDS

Dr. Ozan is a Resident Assistant in the Department of Oral and Maxillofacial Surgery of the Faculty of Dentistry at Cumhuriyet University in Sivas, Turkey.

e-mail: fozan@cumhuriyet.edu.tr

Hidayet Burak Polat, DDS

Dr. Polat is a Resident Assistant in the Department of Oral and Maxillofacial Surgery of the Faculty of Dentistry at Cumhuriyet University in Sivas, Turkey.

Sinan Ay, DDS, PhD

Dr. Ay is an Assistant Professor in the Department of Oral and Maxillofacial Surgery of the Faculty of Dentistry at Cumhuriyet University in Sivas, Turkey.

Fahrettin Goze, MD

Dr. Goze is a Professor in the Department of Pathology of the Faculty of Medicine at Cumhuriyet University in Sivas, Turkey.