Case Report

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EPIDERMOID CYST OF THE BUCCAL MUCOSA: A CASE REPORT

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ABSTRACT

Dermoid and epidermoid cysts are developmental pathologies occurring in the head and neck region with an incidence ranging from 1.6% to 6.9%. Both cysts reported are less than 0.01% of all oral cavity cysts. The article describes a rare case of an epidermoid cyst of the left buccal mucosa in a 38-year-old man and review of the literature.

KEYWORDS: Cyst; Dermoid Cyst; Epidermoid cyst; Buccal mucosa

INTRODUCTION

Epidermoid cysts are non-odontogenic inclusion cyst lined by ectoderm. These are rare lesions derived from germinal epithelium and are encountered throughout the body, in areas where embryonic elements fuse together.[1] Most cases have been reported in ovaries and the testicles, with 7% occurring in the oro-facial area and 1.6% in the oral cavity, representing 0.01% of all oral cavity cysts.[1-4] The floor of the mouth is one of the most commonly affected area, however these cysts can also be found in the tongue, lips, buccal mucosa and jaws.[1-5] Epidermoid cysts are indolent in nature, slow to progress and remains asymptomatic unless secondarily infected.[5,6] We present a rare case of an epidermoid cyst in the nasolabal area of buccal region of a 38-year-old man and review of the literature.

CASE REPORT

A 38-year-old man reported with the complaint of swelling of the left cheek area, just lateral to nasolabial fold which was present for about 2 years. The swelling was painless. He had no history of surgery and/or trauma related to the lesion. He was prescribed an antibiotic by a general dental practitioner which did not reduce the size of swelling. Extraoral examination showed a 2 x 3 x 4 cm swelling extending from the left commissure to midbuccal region anteroposteriorly and up to ala of nose laterally in nasolabial fold superoinferiorly. Overlying skin was apparently normal. On palpation swelling was non tender and freely mobile. Submandibular & submental lymph nodes were not palpable (Fig. 1). Intraoral examination revealed a 2 x 3 x 4 cm swelling extending from the left commissure to the lateral incisor region. The swelling was non tender and was freely mobile between the buccal mucosa and buccinators muscle. The overlying mucosa was normal in color and texture. A provisional diagnosis of benign soft tissue lesion was made. Under local anesthesia, an incision was given in the buccal mucosa and blunt dissection was carried out and mass was exposed which was well encapsulated, was excised. During dissection angular artery was encountered which was cut and ligated (Fig. 2). The wound was closed in layers with 3.0 vicryl sutures. The postoperative period was uneventful and healing was good (Fig. 3). On gross examination the mass was oval in shape, pinkish white in color and doughy in consistency measuring 2 x 3 x 4 cm (Fig. 4). The specimen was submitted for histopathology. Cystic lumen was filled with thick creamy, cheesy like material. On microscopic 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 covered by fibrous capsule composed of epithelium, without any skin adnexa (Fig. 5). This was consistent to the diagnosis of epidermoid cyst.[1,2,6] The patient was followed up for 2 years after surgery there was no sign of any recurrence.

DISCUSSION

Dermoid cysts are nonodontogenic cystic lesions. Roser, in 1859 first described epidermoid cyst. These are rare benign conditions in the oro-facial region derived from abnormally situated

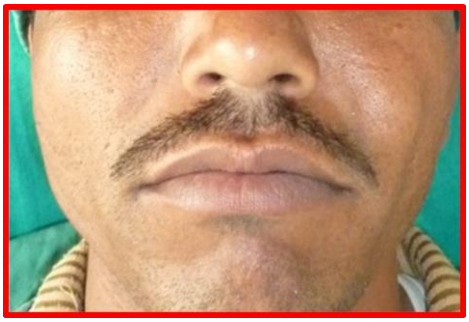

Fig. 1: Extraoral View Showing Swelling in Left Nasolabial Region

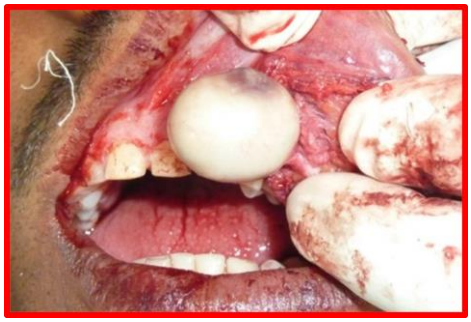

Fig. 2: Intraoperative Appearance of the Mass

Fig. 3: Showing Left Buccal Region After Excision of Mass

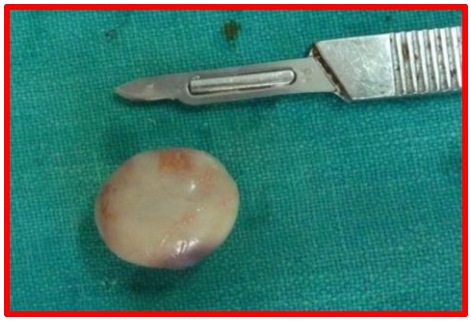

Fig. 4: Excised Mass

ectodermal tissue. Depending on the pathogenesis, Epidermoid cyst can be divided into: 1) Congenital; 2) Acquired

Congenital cysts are dysembryogenic lesions that arise from ectodermal elements entrapped during midline fusion of the first and second branchial arches between the third and fourth week of the intrauterine life. Alternatively, they may also arise from tuberculum impar. Acquired cyst is derived from traumatic or iatrogenic inclusion of epithelial cells or from occlusion of sebaceous gland duct. It was first recognized by Werhner in 1855 and originally referred to as "Implantation cyst" by Sutton in 1895. There are two theories for epidermoid cyst formation: Firstly, Epidermoid cyst may occur when two epidermal surfaces fuse together during early intrauterine life and an ectodermal implant is retained deep to the surface. Secondly, due to traumatic entrapment of surface epithelium in the connective tissue; later these cells may differentiate to form cyst.[1,3,6,7] In 1955, Meyer classified Dermoid cysts into the following three categories.[1,6]

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;
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 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%. Most reported cases have involved the floor of the mouth, usually in the midline. Rare cases have been reported in the tongue, lips, uvula, temporomandibular joint dermal graft, intracranial, and intraosseously within the mandible and maxilla. So far very few case reports of an epidermoid cyst of the buccal mucosa have been reported in the literature.[8] Epidermoid cysts are generally diagnosed in young adults in the second and third decades of life. It is twice as common in men as in women with a male to female ratio of 3:1.[1,4,6] 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, benign tumors and dermoid cysts. In the case reported odontogenic infection is ruled out as it is of long duration i.e. 2 years. The mass has attained considerable size without constitutional symptoms such as malaise, fever and pain. As there was no response to antibiotic treatment.

A salivary obstruction with a mucocele formation is a possibility. A bluish-gray hue is characteristic of a mucocele, but the lesion in the present case had an overlying mucosa which was normal in color. 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 reddishpurple color & pulsation (hemangioma) or almost clear translucent color (lymphangioma) with lesions of this magnitude.[8]

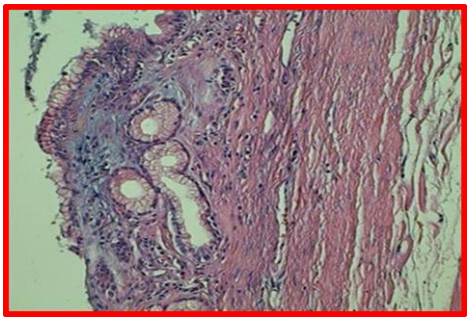

Fig. 5: Photomicrograph showing ortho-keratinised stratified squamous epithelium with flat epithelial-connective tissue interface lining the cystic cavity (H&E)

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 (smaller 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.[8] In most cases, dermoid cysts are treated by enucleation. Surgical access depends on the location and size of the lesion. Marsupialization has also been proposed as a treatment alternative in cases of giant cysts. When intraoral access is complicated, a combined intraoral and extraoral approach should be used.[4,7,9] 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.[6] Dini *et al.*, described a patient with basal cell carcinoma arising in the wall of an epidermoid cyst. Ikeda et al. presented a case report of 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.*, 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.[1,8] In our case, excision was carried out without any major complications by employing intraoral access under local anesthesia. This approach is supported by Akao and colleagues who state that intraoral access must be attempted first, even if dealing with a large cyst. The intraoral approach leads to good cosmetic and functional results.[6,7] Surgical excision is normally achieved without major complications and prognosis is very good.

CONCLUSION

Epidermoid cyst of the buccal region is quite rare and need to be differentiated from several other conditions like odontogenic infection, benign tumors and dermoid cysts of the area. To obtain the correct diagnosis of cystic swellings, fine needle aspiration cytology should always be performed. Surgical excision is the treatment of choice and may be performed under local anesthesia through intraoral access, with no recurrence expected.

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