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An Unusual Incidental Finding of Multiple Micro-Epidermoid Cyst Present Bilaterally In Buccal Mucosa of Patient with Oral Sub-Mucous Fibrosis.

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ABSTRACT

Epidermoid cyst are benign cyst which results from entrapped epidermal elements without adenexal appendages. It is a rare cyst in oro-facial region which represents less than 0.01% of all oral cavity cyst . It is usually asymptomatic but can present itself acutely if secondarily infected or grows over time to attain large sizes and cause significant anatomical disfigurement and systemic complications as well. Complete enucleation of the cyst is the choice of treatment .Here we report an unusual case of incidental finding of multiple micro-epidermal cyst of bilateral buccal mucosa in a 35 yrs-old male patient with Grade 2 oral sub-mucous fibrosis.

KEYWORDS

Introduction:
Dermoid cyst are developmental lesions found anywhere in body, particularly because of inclusion of tissues from variant sources (ectoderm, mesoderm, endoderm) along the embryonic lines of fusions.

Acquired variants occur due to traumatic or iatrogenic inclusion of epithelium or obstruction of sebaceous gland duct, hence also known as implantation keratinizing epidermoid cysts [1 and 4.]

Within the oral cavity, its a rare entity which accounts for less than 0.01% of all the cyst [2,3,4 and 7]. A large majority (one – fifth) of which is found in the floor of the mouth [2.5,6 and 7 .]

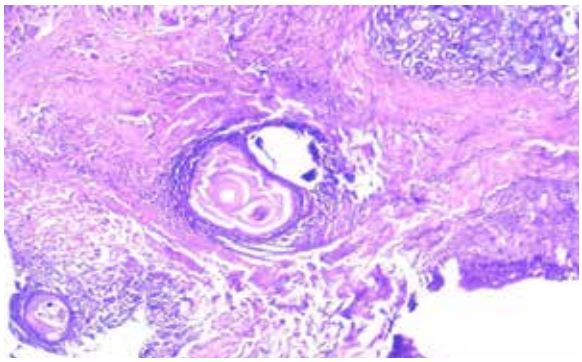
Meyer[7] in 1955, described the three histological types of dermoid cyst i.e. true demoid cyst, epidermoid cyst and teratoid cyst Dermoid cysts are lined by keratinized epithelium and contain skin adenexa, epidermoid cyst on the other hand are only lined by simple squamous epithelium. Teratoid cyst contain dermoid material plus tissues of other embryonal sources e.g. ciliary respiratory epithelium, gastrointestinal tissues, muscles e.t.c.

We present in this case report an exceptionally atypical existence of multiple micro-epidermoid cyst in buccal musosa of a patient with oral submucous fibrosis.

Case report.
A 35-year-old male reported to our out –patient dept. with the chief complaint of reduced ,mouth opening and burning sensation on consumption of spicy foods for more than a year. Personal history revealed habit of chewing pan-masala (areca nuts), 7-8 pouches per day for the last 15 yrs. Past history revealed that he had undergone conservative management for the same elsewhere but with no apparent relief .

On detailed intra-oral examination ,rigid fibrous bands were palpated bilaterally over the buccal mucosa. Mucosa over the upper-lower lips, corner of the mouth ,soft-palate, retromolar area and along the tonsillar fauces were significantly blanched . Inter-incisal distance was reduced to 5mm.(stage 4)[16] .Mobility of the tongue was normal.

Other dental and extra-oral examinations were insignificant.



Two samples each containing small piece of buccal mucosa measuring approximately about 0.8 x0.5x0.5 cm was incised from each side and sent for histopathological examination followed by Bitateral fibrous band resection and reconstruction with buccal fat pad under local anesthesia (Fig.1 and 2). Intraoperative mouthopening of 33 mm was achieved with the help of Hiester's mouth gag.

Histopathological examination of both the samples showed fibro- collagenous tissue stroma with area of collagen deposition and hyalinization and multiple foci of entrapped squamous cell epithelium filled with keratinous material in the lumen.(Fig. 3). Foci of intact lobules of minor salivary glands were also seen. The finding was consistent with Submucous fibrosis with multiple epidermal inclusion cyst.

The patient is under observation for the last 3 months following the surgery without any obvious clinical presentation till date.

Discussion:

The term epidermoid cyst was given by Roser in 1859. Most commonly they appear between the age group of 15-35 yrs and can be categorised depending on the pathogenesis as congenital or acquired[4].

The etiology of both the dermoid and the epidermoid cyst is the failure of the surface ectoderm to detach from the underlying connective tissues , resulting in the sequestration and the implantation of the surface ectoderm. Congenital variety are derived from the entrapment of ectodermic elements in the midline of embroyenic line of fusion [5,8] .Acquired cyst can arise from traumatic or iatrogenic implantation of surface epithelium [1 and 4]. However, for this to happen a cycle of events like, history of trauma, proliferativeand diffentiative property of the implanted surface epithelium along with mild degree of inflammatory process are required to occur simultaneously [1 AND 4]. The pathogenesis of oral submucous fibrosis which is described as microtauma to the oral mucosa produced by the friction of coarse fibres of areca nut which in turn facilitates diffusion of the alkaloids into the subepithelial connective tissue resulting in juxtaepithelial inflammatory cell infiltrate [17]. In our case, the trauma caused due to coarse fibres of areca nuts to the oral mucosa must have lead to buccal epithelial inclusion at numerous micro sites along with simultaneous chronic slow insidious inflammatory process leading to multiple micro -epidermoid cysts formation at numerous sites within the buccal mucosa.

The incidence of the epidermal cyst in oro-facial region has been reported as 7% and 1.6% in oral cavity ,representing 0.01% of all cysts of oral cavity[2,4,5 and 9]. Most of the intraoral cases are reported in the midline and floor of the mouth [3,5,6 and 7] . Cases are seen with respect to tongue, buccal mucosa, maxilla , mandible and uvula but are rare [4,8,10 and 11]. Common syndromes associated with eidermoid cyst are Gardner's Syndrome, Basal cell nevus syndrome[12]

Several other lesions occurring in buccal mucosa including, lipoma, lymphangioma, and mucocele can be taken into consideration for differential diagnosis. However , our finding was co-incidental and apart from the reduce mouth opening and rigid submucous fibrous bands no other clinical presentation of any cystic lesion were evident before and during the surgery. Histopathology report later established the diagnosis which revealed the presence of multiple micro-epidermoid cyst along with submucous fibrosis.

Although ,acute symptomatic manifestation are possible if secondarily infected ,they usually remain aymptomatic [13] .Rarely an epidermal cyst undergoes malignant transformation. However, few cases has been reported [15.] Basal cell carcinoma arising from the walls of epidermoid cyst has been reported by **Ikeda** [14] .

Conclusion:

Epidermoid cyst of oral cavity is s a rare entity. Moreover, the anatomical variation as well as the histopathology variation in our case prove to be highly significant. Establishment of proper diagnosis need to be done after taking into consideration all other congenital, developmental, neoplastic ,traumatic or infectious lesions. Therefore ,we emphasize the importance of a close histologic examination of lesions that are seemingly benign in order to avoid incorrect diagnosis.

References:

1. Ettinger RL, Manderson RD. Implantation keratinizing epidermoid cyst . Oral Surg. 1973;26:225-230. Doi:10.1016/0030-4220(73)90242-9.
2. Rajayogeshwaran V , Eveson JW . Epidermoid cyst of the buccal mucosa . Oral Surg Oral Med Oral Pathol. 1989;67:184. doi: 10.1016/0030-4220(89)90326-5.
3. Worley CM, Laskin DM. Coincidental sublinguala and sub mental epidermoid cyst. J Oral and Maxillofac Surg;1993;51:789-790. doi: 10.1016/S0278-2391(10)80424-0.
4. Kini YK, Kharkar VR, Rudagi BM, Kalburge JV. An unusual occurance of epidermoid cyst in the buccal mucosa : A case report with review of literature. J Maxxillofac Surg . 2013:12:90-3
5. Longo F , Maremonti P, Mangone GM, De Maria G , Califano L. Midline (dermoid)cyst of the floor of the mouth : Report of 16 cases and review of surgical techniques. Plas Reconstr Surg. 2003;112:1560-5.
6. King RC , Smith BR, Burk JL. Dermoid cyst in the floor of the mouth . Revie of reports and case reports. Oral Surg Oral Med Oral Pathol. 1994;78:567-76.[PubMed
7. Meyer I . Dermoid cysts(dermoids)of the floor of the mouth . Oral Surg Oral Med Oral Pathol.1955;27:1149-64.[PubMed : 13266349]
8. Pryor SG, Lewis JE , Weaver A, OrvidaS LJ. Pediatric dermoid cyst of the head and neck . otolaryngol Head and Neck Surg. 2005;132:938-42.[PubMed: 15944568]
9. Ozan F, Polat HB, Ay S , Goze F. Epidermoid cyst of buccal mucosa : A case report . J Contemp, Dent Pracc 2007;8(3):1-6.
10. Flom GS, Donovan TJ, Landgraf JR. Condenital dermoid cyst od=f anterior tongue. Otolaryngol Head Neck Surg. 1998;101:8. [PubMed]
11. Noffke CE. Implantation-type epidermoid cyst of the mandible. Dentomaxillofac Radiol. 1999;28:385-5.
12. Shafer WG, Hine MK, Levy BM. A textbook of oral pathology . 4. Philadelphia: WB Saunders; 1983.
13. Cortezzi W , De Albuquerque EB. Secondarily infected epidermoid cyst in the floor of the mouth causing a life threatening situation: reporty of a case . J Oral Maxillofac Surg. 1994;52:762-764 . doi: 10.1016/0278-2391(94)90496-0
14. Ikeda I, Ono T. Basal cell carcinoma originating from an epidermoid cyst. J Dermatol. 1990;17:643-646.
15. Devine JC , Jones DC. Carcinomatous transformation of a sublingual dermoid cyst-a case report . Int J Oral Maxillofac Surg. 200;29:126-127 . doi: 10.1016/S0901-5027(00)80009-0.
16. Haider SM, Merchant AT, Rahbar MH: Clinical and functional staging of osmf. British journal of oral and maxillofacial surgery. 38,12-15.
17. Chiang CP, Hsieh RP, Chen TH, Chang YF, Liu BY(2002). High incidence of autoantibodies in taiwanese patient with oral submucous fibrosis. J Oral Pathol Med 31:402-9.