

Epidermoid cyst: Report of two cases

Surekha R Puranik, Rudrayya S Puranik¹, Satya Prakash, M Bimba²

Departments of Oral Medicine and Radiology and ¹Oral and Maxillofacial Pathology, PMNM Dental College and Hospital, Bagalkot, ²Department of Oral Medicine and Radiology, Subbaiah Institute of Dental Sciences, Shimoga, Karnataka, India

Abstract

Epidermoid cysts (ECs) are uncommon, benign cystic lesions derived from the entrapment of surface epithelium or more often from the aberrant healing of infundibular epithelium during an episode of follicular inflammation. ECs occur anywhere on the body, particularly along embryonic fusion lines, most commonly on the face, scalp, neck, chest and upper back. Head and neck ECs constitute only about 7%, whereas only 1.6% of ECs are reported in the oral cavity. They comprise <0.01% of all the oral cysts. Floor of the mouth, tongue, lips, palate, jaws, etc., are some of the reported sites of ECs in the oral cavity. Microscopically, ECs are lined with plain stratified squamous epithelium filled with laminated layers of keratin. Here, we report two rare cases of ECs, one occurring in the gingival aspect and other in the lower third of face. The cases are reported due to rarity of ECs in the head and neck region.

Key Words: Epidermoid cyst, gingiva, keratin

Address for correspondence:

Dr. Surekha R Puranik, Department of Oral Medicine and Radiology, PMNM Dental College and Hospital, BVVS Campus, Bagalkot - 587 101, Karnataka, India.
E-mail: puraniksurekha19@rediffmail.com

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INTRODUCTION

Epidermoid cysts (ECs) are uncommon, benign cystic lesions derived from the entrapment of surface epithelium or more often from the aberrant healing of infundibular epithelium during an episode of follicular inflammation. ECs occur anywhere on the body, particularly along embryonic fusion lines, most commonly on the face, scalp, neck, chest and upper back. Head and neck ECs constitute only about 7%, whereas only 1.6% of ECs are reported in the oral cavity. They comprise <0.01% of all the oral cysts. Floor of the mouth, tongue, lips, palate, jaws, etc., are some of the reported sites of ECs in the oral cavity. However, gingival ECs are infrequently reported. In a case series of ECs by Ravindranath *et al.*, out of 13 cases, only 3 occurred in the gingiva (2 in the left posterior and 1 in the right posterior mandibular gingiva). To the best

of our knowledge, this is the fourth case report of EC in the region of gingiva in English literature. Clinically, they present as slow-growing, painless, fluctuant masses often detected during the second or third decade of life. Rarely, they cause functional interference; however, size and location of the cyst are being considered as prime concern for seeking the expert opinion. Microscopically, ECs are lined with plain stratified squamous epithelium filled with laminated layers of keratin.

CASE REPORTS

Case report 1

A 28-year-old male patient reported to the clinic with a swelling in the posterior lingual aspect of gingiva of 34, 35 and 36 regions for 1 year. It was asymptomatic which gradually increased to the present size. Medical/dental history

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was not relevant. The patient denied any previous history of trauma or prior surgery at the same site. He expressed growing concern over interference in the tongue movement and inability to occlude properly on occasions. On examination, intraorally, a solitary, well-defined, ovoid-shaped swelling measuring approximately 3.5 cm × 2.5 cm was noted on the lingual gingival aspect. It was firmly attached to the underlying tissue with stalk. It was nontender, firm in consistency, covered with fibrous pale yellowish tint mucosa. There was no bleeding on contact and secondary changes of mucosa. Mild degree of stains and calculi were noted. Physical examination was noncontributory [Figure 1].

Occlusal radiograph revealed roughly oblong-shaped, soft tissue density shadow on the lingual aspect of left floor of the mouth corresponding to 34, 35 and 36 with no underlining bony changes. Two small radiopaque spicules were seen emanating from cortical bone [Figure 2].

The lesion was completely excised surgically. At the time of excision, two small bony projections appeared underlining the lesion, which were removed with a rongeur. The postoperative course in healing was free of complication. Follow-up of 1 year showed no signs of recurrence.

Macroscopically, the specimen was yellowish white soft tissue mass measuring 3 cm × 2 cm, cut surface of which showed pale white cheesy material [Figures 3 and 4].

Microscopically, hematoxylin and eosin-stained sections showed a large prominent circular microcystic space lined by flat squamous epithelium surrounding a lumen filled with abundant compressed keratin arranged in lamellar pattern with no skin appendages. The overall histopathological features were suggestive of “epidermoid cyst” [Figure 5].

Case report 2

A 68-year-old male patient reported to the Department of Oral Medicine and Radiology for missing teeth and wanted new teeth set. He gave a history of asymptomatic swelling on the right side of lower one-third of the face for 5 years, which gradually increased to the present size of a pear. The patient was concerned about his esthetics. There was no history of trauma or relevant medical and dental history. There was a history of tobacco chewing habit for 30 years. A solitary swelling was seen near the right angle of mouth measuring about 3.5 cm × 2.5 cm in dimension, with intact, stretched overlying skin of normal color [Figure 6]. On palpation, it was elastic rubbery, nontender and movable. Intraorally, the patient was edentulous with favorable upper and lower ridges. Orthopantomograph revealed completely edentulous, resorbed maxillary and mandibular arches.

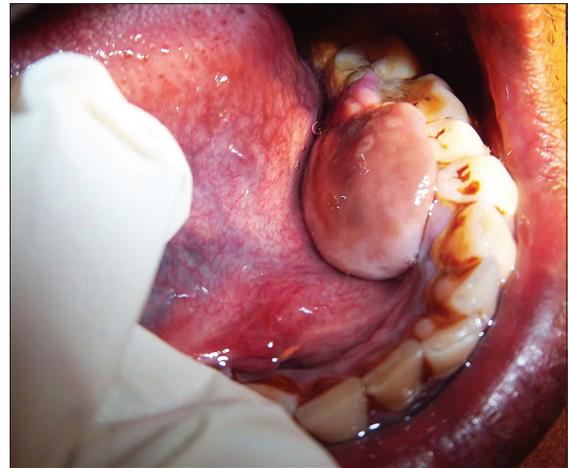


Figure 1: Clinical photograph shows swelling in posterior lingual aspect of gingiva of 34, 35 and 36 region

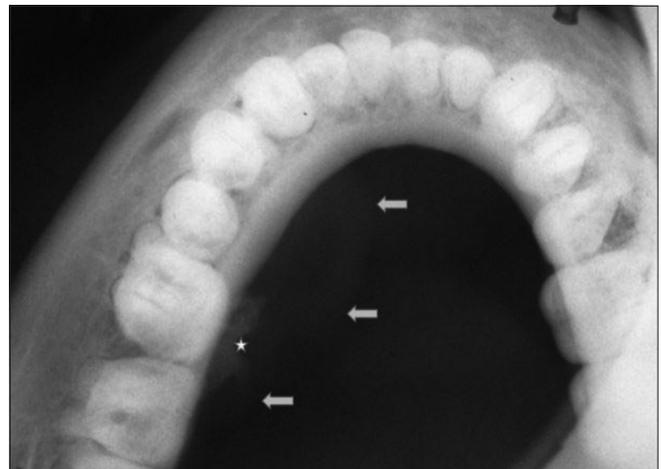


Figure 2: Occlusal radiograph shows roughly oblong-shaped, soft tissue density shadow with two spicules



Figure 3: Macroscopically, specimen shows yellowish white soft tissue mass measuring 3 cm × 2 cm

The lesion was surgically excised. On surgical excision, yellowish cheesy material extruded from the swelling [Figure 7]. The

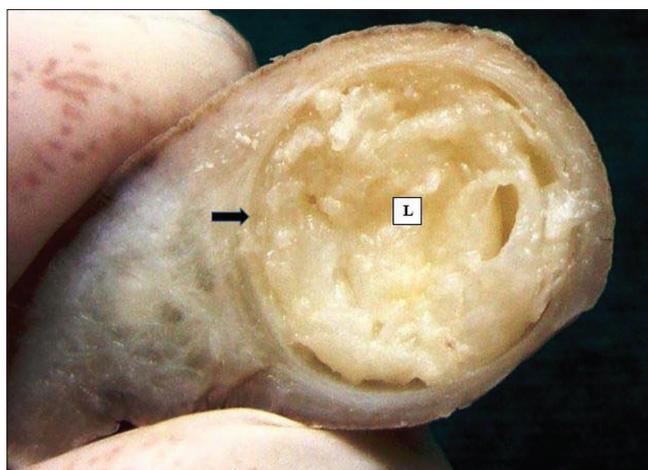


Figure 4: Excised specimen shows pale white cheesy material on cut surface

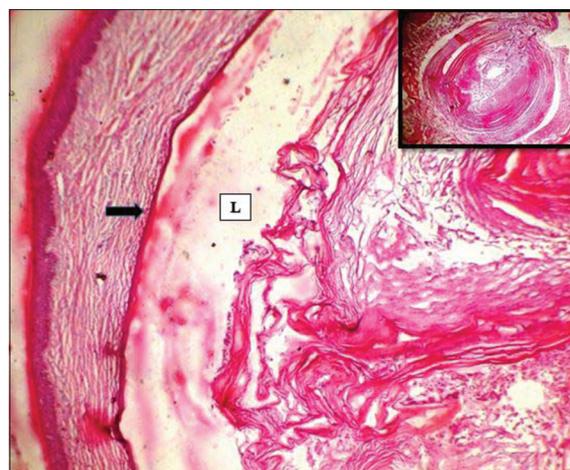


Figure 5: Histopathology shows microcystic space lined by flat squamous epithelium surrounding a lumen filled with abundant compressed keratin arranged in lamellar pattern with no skin appendages (H&E stain, $\times 40$). [Inset: Photomicrograph showing the lamellar pattern of keratin (H&E stain, $\times 40$)



Figure 6: Clinical photograph shows swelling near the right angle of mouth measuring about 3.5 cm \times 2.5 cm in dimension, with intact, stretched overlying skin of normal color



Figure 7: Excised specimen shows yellowish cheesy material extruding from the swelling on cut section

postoperative course and healing was free of complication. Follow-up of 2 years showed no signs of recurrence.

Microscopically, hematoxylin and eosin-stained sections showed a cystic cavity lined by stratified squamous orthokeratinized layers, lumen filled with fragments of keratin with no skin appendages. The overall histopathological features were suggestive of “epidermoid cyst” [Figure 8].

DISCUSSION

Roser in 1859 first described EC in the floor of mouth. ECs are uncommon, benign cystic lesions derived from the entrapment of surface epithelium or more often from the aberrant healing of infundibular epithelium during an episode of follicular inflammation.^[1]

New and Erich (1937) reported 24 (1.6% cases of ECs occurring in the floor of mouth out of 1495 cases of dermoid cysts.

In 1955, Meyer described three variants of teratoid cysts; (1) EC always lined by stratified squamous epithelium without dermal appendages within the underlying connective tissue. (2) Dermoid cyst, in addition to typical squamous epithelium, contains dermal appendages such as hair, hair follicles, sebaceous and sweat glands. (3) Teratoid cyst wall is lined with squamous epithelium and consists of tissues from all three germ layers such as respiratory, gastrointestinal and nervous system. The lumen of all three types of cyst displays greasy, cheese-like, white-gray content formed by shed keratin and sebaceous material.^[2,3]

Depending on pathogenesis, ECs are divided into congenital and acquired type. The former is thought to develop from congenital inclusion of ectodermal tissue during embryological development. The acquired type, originally referred to as “implantation cyst,” is believed to originate through

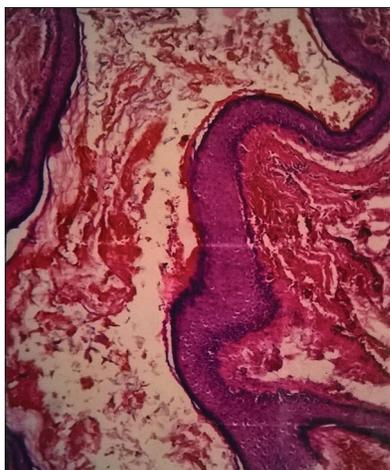


Figure 8: Histopathology shows cystic cavity lined by stratified squamous orthokeratinized epithelium, filled with keratin with no skin appendages (H&E stain, $\times 40$)

implantation of epithelium by either surgical or accidental trauma into deeper mesenchymal tissues. Trauma and previous surgical history have been reported as major factors in etiology of acquired type of ECs. Our cases of ECs appeared to be congenital in origin owing to lack of trauma or prior surgical history.^[4,5]

Clinically, ECs present as slow-growing, painless, firm and/or fluctuant masses often detected during the second or third decade of life even if they are congenital in origin. A characteristic doughy consistency is noted, especially with cysts of the floor of the mouth. They are considered to be asymptomatic with no functional interference unless the size causes annoyance. Dysphonia, dysphagia, dysarthria, dyspnea, double chin, interference in tongue movement and mastication, speech, etc., have been reported with ECs in different locations and with increasing size. Unusual manifestations such as headache, obstructive sialadenitis and facial asymmetry have also been reported with ECs of other sites in the head and neck. Our patient with gingival EC sought dental opinion due to interference in the tongue movement and occlusion, and in case report 2, except esthetics, there were no other manifestations.^[4-8]

In oral cavity, 25% of ECs are primarily found in floor of the mouth. Tongue, lips, jaws, palate, pterygopalatine fossa, etc., are some of the reported sites. However, gingival ECs are infrequently reported. In a case series of ECs by Ravindranath *et al.*, out of 13 cases, only 3 occurred in the gingiva (2 in the left posterior and 1 in the right posterior mandibular gingiva). To the best of our knowledge, this is the fourth case report of EC in the region of gingiva in English literature. ECs grow anywhere in the body with few cases reported in the temporal region, submental region, submandibular region, calvarium, orbit, maxillary sinus, intracranially in the posterior and

middle fossae. The age range between 10 and 72 years has been reported for ECs, with most becoming apparent during 15–40 years. Hypersecretion of fat influenced by hormonal stimulus during puberty may stimulate growth of cyst which would explain greater incidence in young adults of 16–40 years of age. Tuz *et al.* presented a case of sublingual dermoid cyst that enlarged rapidly during pregnancy, causing deglutition and mild respiratory problems. Sex predilection is reported to be contradictory with few papers reporting male predominance. Ravindranath *et al.* reported average of 37.4 years (20–70) with 11 out of 13 cases in males and 2 in females in their case series concurrent with both the present cases, occurred in male patients with ages being 28 and 68 years in case 1 and case 2, respectively.^[1,2,8,9]

Fine needle aspiration cytology (FNAC) is safe, cost-effective complimentary tool for preoperative diagnosis of ECs. However, it is emphasized that it is not possible to determine specific histologic subtypes through FNAC.^[2,3]

Imaging plays an important role to delineate cystic masses from solid lesions and to assess the luminal content. In cases of ECs, located in surgically inaccessible areas such as floor of the mouth, uvula, temporomandibular joint, jaws, temporal region, intracranial, orbit, magnetic resonance imaging (MRI) and computed tomography (CT) allows more precise localization, anatomic extension and topographic relation. This helps in preoperative diagnosis and enables the surgeon to choose the most appropriate surgical approach. CT reveals expansile, hypointense areas with smooth sclerotic margins. MRI findings include smooth masses with iso or slightly high signal intensity relative to adjacent muscles on T1-weighted and very high signal intensity on T2-weighted images, with superior soft tissue imaging than CT. It also shows keratin debris in the cyst. On ultrasound, most ECs appear as hypoechoic masses containing variable echogenic foci and display posterior sound enhancement with no color Doppler signals.^[10-12]

A variety of plaque and nonplaque irritant-associated gingival swellings are encountered in clinical practice. The characteristic presentation, demographic data, gingival-periodontal evaluation, radiographic changes and appropriate investigations should be considered for final diagnosis. The differential diagnosis for localized gingival masses comprises pyogenic granuloma, peripheral ossifying fibroma, peripheral giant cell granuloma, irritational fibroma, etc. Certain rare benign mesenchymal lesions such as neurofibroma and oral focal mucinosis have been reported on gingival aspect. Peripheral odontogenic tumors are rarely included in the differential diagnosis of gingival swellings because of paucity of occurrence. Peripheral ameloblastoma, calcifying epithelial odontogenic tumor, odontogenic fibroma, adenomatoid odontogenic tumor are some of such tumors

presenting as sessile or pedunculated, nontender, firm, reddish- or normal-colored focal growths of varying sizes in the mandibular gingiva. Definitive histology remains the gold standard in these cases and helps in differentiating from ECs. A preoperative clinical diagnosis may be impossible because of rarity of ECs in the gingiva. Extraoral ECs as in the case 2 mandate the exclusion of certain mesenchymal tumors such as lipoma, fibroma and myxoma. Proper clinical evaluation is required in such cases.^[13-15]

Histologically, ECs reveal cystic wall lined by keratinized stratified squamous epithelium filled with keratin. ECs are described as “pearly tumor” due to shiny, smooth, waxy keratinous content of the cyst. Unlike dermoid cyst, they exhibit no adnexal structures such as hair follicle, sebaceous gland and sweat gland. Microscopic examination undoubtedly remains the primary means of diagnosing ECs as evidenced in both the cases.^[3-5]

Various intra- and extra-oral approaches have been discussed for surgical treatment of ECs depending on sites. We excised the lesion with a simple scalpel. The use of laser for excisional biopsy in the intraoral approach has been advocated for achieving the goals such as lessening postoperative discomfort and improved surgical access than scalpel by Romeo *et al.* for the small EC of 1 cm on the lateral border of tongue. High cutting ability, bloodless operative field, relative ease and rapidity of use are some of the advantages of lasers. KTP, diode, erbium, neodymium-doped yttrium aluminum garnet lasers have been tested for intraoral ECs.^[3,6]

Recurrence rate of ECs is reported to be very less with good prognosis. Although malignant transformation has been considered to be extremely rare for head and neck ECs, a few cases have reported carcinomatous changes in ovarian, cutaneous, intracranial ECs. There are no data regarding such change in gingival ECs.^[8,11,15]

CONCLUSION

Although EC of the head and neck region is quite a rare entity, particularly in the gingival aspect, it should be included in the differential diagnosis of benign gingival swellings. Thorough clinical evaluation and imaging may sometimes be necessary to

rule out ECs in the head and neck region. Complete surgical excision is needed for successful outcome. Definitive histologic examination is required for final diagnosis. We report two cases of EC of left mandibular gingiva intraorally and near right corner of mouth extraorally, successfully managed with surgical excision.

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Conflicts of interest

There are no conflicts of interest.

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