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An Unusual Presentation of an Epidermoid Cyst in the Floor of the Mouth – A Case Report

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Abstract:
Epidermoid cysts are uncommon, benign cystic lesions occur anywhere in the body, particularly along embryonic fusion lines, most commonly on the face, scalp, neck, chest, and upper back. Head and neck epidermoid cysts constitute only about 7%, whereas only 1.6% of epidermoid cysts are reported in the oral cavity. They comprise <0.01% of all the oral cysts. Clinically, the lesion presents as a slow-growing asymptomatic mass, usually located in the midline that may occasionally cause elevation of the tongue, interference of speech, and the appearance of a double chin. Histopathologic examination will reveal a cyst cavity lined by stratified squamous epithelium with a well-developed granular layer resembling the epidermis. Lumen will be filled with abundant degenerating ortho-keratin. Underlying connective tissue will be densely collagenous with endothelium lined vascular spaces filled with RBCs. Microscopic examination undoubtedly remains the primary means of diagnosing Epidermoid cysts. In this report, we describe the case of a 56-year-old female patient who presented with a massive sublingual epidermoid cyst. The lesion was treated satisfactorily by surgical excision and the diagnosis was corroborated by histopathological examination. We discuss the clinical steps required to achieve a final diagnosis, the differential diagnosis, useful imaging techniques, and treatment of oral epidermoid cysts.

Key Words*: Oral epidermoid cyst; Benign tumor of the oral cavity; Floor of mouth swelling.*

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I. Introduction

Epidermoid cysts are rare, benign cystic lesions that occur anywhere in the body, particularly along the embryonic lines of fusion, with common occurrences on the face, scalp, neck, chest, and upper back. Head and neck epidermoid cysts constitute only about 7%, whereas only 1.6% are reported as intra-oral epidermoid cysts. They comprise less than 0.01% of all the oral cavitycysts[1]. There are two main theories on the etiology of dermoid and epidermoid cysts that appear on the midline floor of the mouth According to first, Dermoid cysts of the floor of the mouth are dis-embryogenetic lesions result from entrapment of ectodermal tissue during the midline fusion between the first and second branchial arches that occur in the third and fourth embryonic weeks. The second theory suggests that midline dermoid and epidermoid cysts may be a variant of the thyroglossal duct cyst with ectodermal elements predominating[2]. Acquired forms derived from iatrogenic or traumatic inclusion of epithelium and skin appendages have also been reported.

According to the anatomic relationship between the cyst and the muscles of the floor of the mouth, dermoid cysts can be sublingual, submental, or lateral. The sublingual cyst is in the midline under the tongue, above the genioglossal muscles. As the cyst grows, swelling of the floor of the mouth appears, and the tongue is raised and pushed backward. The submental cyst is in the midline of the submental region, between the geniohyoid and mylohyoid muscles. Cyst enlargement leads to a typical "double-chin" appearance. The lateral cyst is located under the mandibular body; it enlarges downward and toward the hyoid bone, and it may displace the

submandibular gland and the tongue on the opposite side. Lateral cysts are rare, and some authors do not recognize them as a separate entity but consider them to be median cysts that have expanded laterally during their enlargement[3].

Epidermoid cysts are a histological variant of dermoid cysts. Dermoid have three histological subtypes, namely, true dermoid cysts, epidermoid cysts, and teratoid cysts[4]. These cysts are lined by stratified squamous epithelium. The cysts are filled with muddy material composed of desquamated cells. Epidermoid cysts may sometimes show the presence of secretory or ciliated epithelium which is why mucous has been found in some cases reported in the literature.

These cysts occur most often in patients in their second or third decade of life. Clinically, the lesion presents as a slow-growing asymptomatic mass, usually located in the midline that may occasionally cause elevation of the tongue, interference with speech, and the appearance of the double chin. Because they are almost always asymptomatic, dermoid cysts are usually diagnosed only after they have attained a considerable size. Recommended treatment is surgical excision via transoral or extraoral access, which mainly depends on the size and location of the lesion.

In this report, we describe the case of a56-year-old lady who presented with a massive swelling in the floor of the mouth which was diagnosed as an oral epidermoid cyst. We discuss the clinical steps required to achieve a final diagnosis, the differential diagnosis, useful imaging techniques, and treatment of dermoid cysts.

II. Case Report

A 56-year-old female patient presented to the Department of Oral and Maxillofacial Surgery, Govt Dental College, Kozhikode, Kerala, India, with a chief complaint of swelling below the tongue that had appeared 6 months back. She neglected the swelling as it was asymptomatic, but her son noticed changes in speech and asked her to consult a doctor. The swelling has not enlarged in size ever since she noticed it. There was no difficulty in swallowing or breathing. There was no history of any pus or fluid discharge.

On general examination, the patient was moderately built and nourished. Vitals were stable, but BP was found to be very high and a medical consultation was sought. There was no evidence of lymphadenopathy. On extra-oral examination, swelling of size 4x4 cm resembling sub-mental fatwas noticed. The overlying skin was normal in color, texture, and temperature. The mass was not fixed to the skin or underlying structures. It was soft, compressible on palpation, and non-tender (figure 1).Intraoral examination revealed a single well-defined ovoid swelling of size 4x3x5 cm in the floor of the mouth towards the right side, raising the tongue onto the palate, which appears normal in color, smooth-surfaced and non-ulcerated. Overlying mucosa over the swelling was normal (figure 2). On palpation, the swelling was non-tender, soft, cystic in consistency, compressible with a positive Paget's test. Also, it was non-trans-illuminant. The slip sign was negative. No additional mucosal lesions were noted. Tongue movements were normal. No dysphagia or dyspnea, but dysarthria was present.

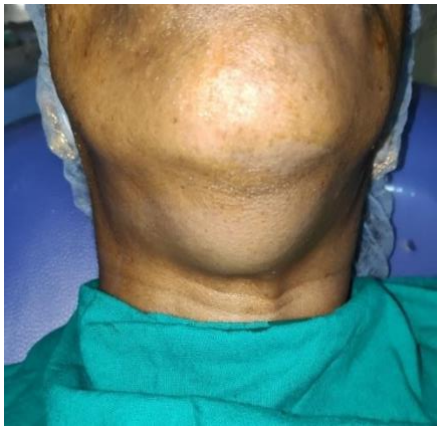

Figure 1: Extra-oral view

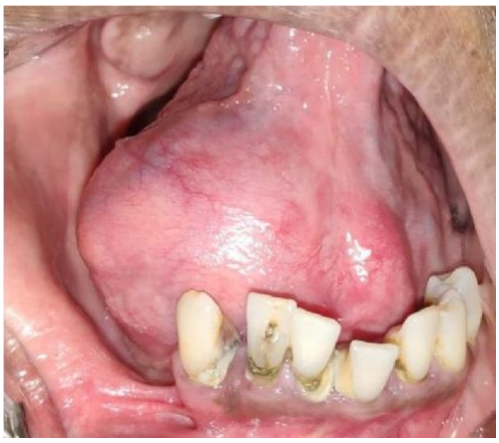

Figure 2: Intra-oralview

Keeping in mind the period of evolution (6 months) and the absence of pain, the possibility of infection was ruled out. So with a tentative diagnosis of ranula, aspiratory punction was carried out which yielded dirty white thick fluid with dull whitish solid flakes resembling keratin that obstructed the wide bore needle lumen (18 G), which hinted towards the differential diagnosis of sebaceous cyst, lipoma, dermoid cyst, and branchial cleft cyst. Microscopic examination of the aspirate identified degenerated epithelial cells in an eosinophilic background.

She underwent ultrasonography of the neck which showed features of cystic swelling of average transverse dimension 2 cm in the sublingual space extending to the submandibular compartment.

Magnetic Resonance Imaging of the neck revealed a well-defined altered signal intensity lesion measuring 4.3x3.1x5.6 cm involving sublingual space. The lesion is noted displacing tongue superiorly, mylohyoid muscle inferiorly, and splaying bilateral genioglossus muscle laterally suggestive of congenital cystic lesion, likely sublingual dermoid cyst. Routine blood and urine investigations were in the normal range. No abnormality was detected on the chest radiograph.

An incision biopsy was planned to confirm the diagnosis. Under local anaesthesia linear mucosal incision is placed over the most prominent part. Blunt mucosal dissection was done, and the lesion was identified. Part of the lesion was incised in a wedge-shaped manner and taken out. Thick pultaceous material popped out through the defect. Hemostasis was achieved and the site closed with 2-0 silk suture. Specimen sent for histopathologic examination in fixative medium formalin and the diagnosis was oral epidermoid cyst with foreign body giant cell reaction.

The patient was operated on under general anaesthesia with nasotracheal intubation. One traction suture was passed through the tip of the tongue, and the tongue was pulled forward and upward. An intraoral paramedian incision without injuring the submandibular duct was placed. The cyst lining was identified after blunt mucosal dissection using mosquito forceps (figure 3). Most of the dissection was performed through blind finger manipulation. The cyst is then completely freed and removed. Layered suturing was performed using 3-0 vicryl suture material to reduce dead space. Final closure was done with 3-0 silk sutures (figure 4).

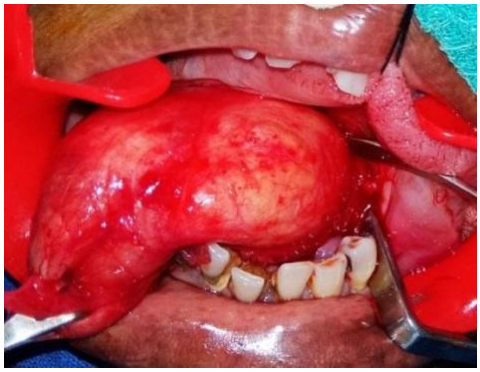

Figure 3: Intra-operative view

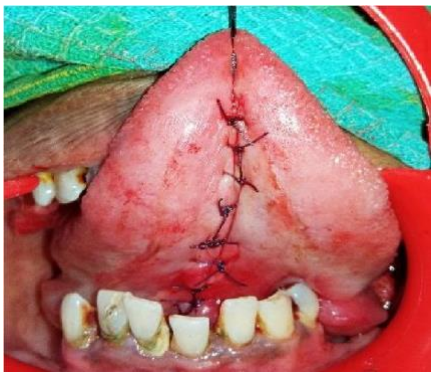

Figure 4: Final Closure

Macroscopically, the specimen consisted of an oval mass of tissue (6.3x3.5x5.5 Cm3 in size), whose surface appeared smooth (figure 5). Specimen sent for histopathologic examination in fixative medium formalin. Cut surface showed yellowish cheesy white material (figure 6).

On histopathologic examination, the tissue showed a cyst cavity lined by stratified squamous epithelium with a well-developed granular layer resembling the epidermis. Lumen was filled with abundant degenerating ortho-keratin. Underlying connective tissue was densely collagenous with endothelium lined vascular spaces filled with RBCs (figure 7). Microscopic examination undoubtedly remains the primary means of diagnosing epidermoid cysts.

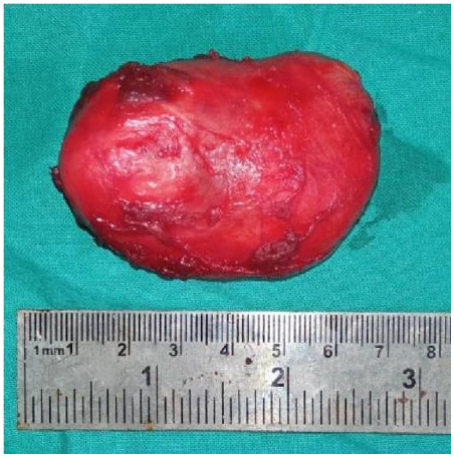

Figure 5: Excised specimen

Figure 6: Cut surface (Gross)

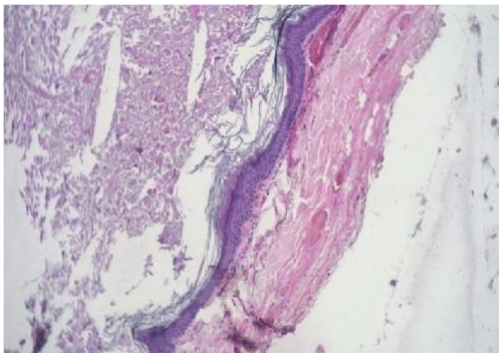

Figure 7: Histopathology showing cyst cavity lined by stratified squamous epithelium with a well-developed granular layer resembling epidermis

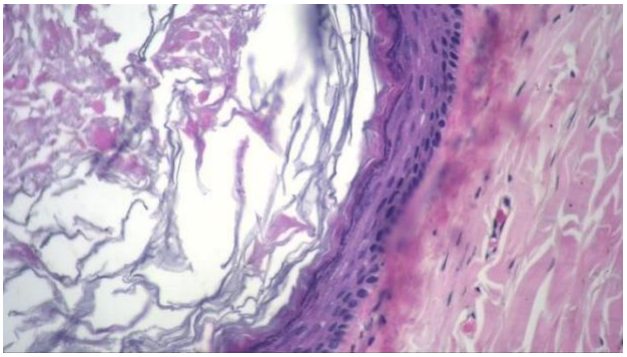

Figure 8: Lumen filled with degenerating ortho keratin

III. Discussion

Dermoid cysts are chronic, slow-growing, and non-tender soft tissue cysts. In 1955 Meyer updated and classified the dermoid cysts into three groups, namely true dermoid, epidermoid, and teratoid. True dermoid show skin adnexa in the surrounding wall namely sebaceous and sweat glands and hair and hair follicles. If the cyst does not contain dermal appendages, the variety is called an epidermoid cyst. Teratoid cyst contains elements derived from all three germ layers: ectoderm, mesoderm, and endoderm, forming bone, muscle, gut, or respiratory mucosa.

These cysts are rare in the head and neck region. They comprise less than 0.01 % of cysts of the oral cavity. In a review of 1,459 cases of dermoid cysts, 7 % were found in the head and neck region and only1.6 % in the oral cavity[5]. Another review by the same author reported the percentage of intraoral dermoid to be 6.5 %. Amongst intraoral sites, floor of mouth is most common. Most patients with epidermoid cyst are in the range between 10 and 35 years of age[6]. Longo and others found that there is a male preponderance in the ratio 3:1, with a mean age of 28 years. Other authors claim that no gender predilection exists. Dermoid cysts mostly arise in embryonic fusion lines, so they commonly present as midline swelling. However, their site of origin may not always be in midline. If they start growing laterally and breach the mylohyoid curtain partially or completely, they may present as lateral neck swelling. Occasionally, there might not be any breach, rather stretching/ expansion of mylohyoid by the pathology, which may present as lateral neck swelling. Most of the cysts in the floor of the mouth occur in the midline, and a lateral dermoid cyst is rarely observed. Epidermoid cysts of the neck have been classified into three categories[7].

- Supra mylohyoid (intraoral)
  - Supra geniohyoid
  - Infra geniohyoid
  - Sublingual
- Infra mylohyoid (cervical)
  - Submental
  - Submandibular
- Peri and trans mylohyoid (intraoral and cervical)
  - Submental trans mylohyoid
  - Lateral peri mylohyoid
  - Trans mylohyoid

When dealing with swellings in the sublingual region, 4 main groups of lesions should be considered: infections, tumours, mucous extravasation phenomena, and anatomic abnormalities arising during embryonic development[8].

In the present case, the patient was a 56-year-old female. The sublingual swelling suggests that the lesion is above the mylohyoid muscle, which is the most common location but the cyst was lateral and there was a midline neck swelling in contrast to a lateral neck swelling thus we concluded it as submental fat. She reported speech difficulties, which is a common symptom. However, the patient could not determine precisely when the swelling had initially developed. We believe it is unlikely that the lesion achieved the present size in only 6 months; most likely, the cyst had gone unnoticed until it grew large enough to cause speech impairment. The patient did not experience any problem except speech difficulty. Being from a rural area and low socioeconomic status, the patient neglected it until functional impairment occurred. The hypothesis of infection was discarded due to the period of evolution and the absence of pain. A malignant tumour was ruled out given the lesion's clinical nature and appearance and the absence of lymphadenopathy.

We were then left with 2 main diagnostic possibilities: a plunging ranula and an anatomic abnormality. Because the clinical picture was compatible with ranula and because ranulas are far more common than dermoid cysts, this was our first impression. Later, the nature of the aspirate tapered the diagnosis to a developmental cystic lesion.

The differential diagnosis of sublingual swellings is more exacting. Imaging techniques may be used for preoperative diagnosis and surgical planning. Fine-needle aspiration is a safe, cost-effective, and reliable tool for preoperative diagnosis of dermoid cysts. Magnetic resonance imaging (MRI) and computed tomography (CT) allow a more precise localization of the lesion and enable the surgeon to choose the most appropriate approach[9]. Some authors prefer MRI over CT as a diagnostic tool for dermoid cysts, as it is superior in terms of soft-tissue resolution and, thus, better able to depict the internal structure of a mass lesion. In our case, we did FNAC, USG, and, MRI. It should be emphasized that it is not possible to determine the specific histologic subtype through fine-needle aspiration, MRI, or USG. Thus, we did an incision biopsy to confirm the diagnosis.

Histologically, Epidermoid Cysts shows a cystic wall lined by keratinized stratified squamous epithelium filled with abundant keratin. Epidermoid Cysts are described as "pearly tumour" due to the shiny, smooth, waxy keratinous content of the cyst[15]. 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 Epidermoid Cysts.

Complete surgical removal is required for the treatment of epidermoid cysts. For operation, there are two approaches, intra-oral approach, and extra-oral (cervical incision) approach. The approach tends to be decided based on the size and the location of the dermoid cyst on the floor of the mouth. In our case, we did this through an intraoral approach as the swelling was above the mylohyoid muscle. In some cases, a combined approach is selected. In the intra-oral approach, an incision is performed usually through the ventral surface of the tongue. While marking this incision, the surgeon should give care to avoid both submandibular ducts[3]. In most cases in which the intra-oral approach is employed, operations are performed under the general anaesthesia delivered by nasotracheal intubation. To avoid the post-operative airway compromise, pre-operative tracheostomy and overnight intubation may be performed. When the cyst is too large to be removed, the cyst may be decompressed for safety removal[10]. Dermoid cyst lining is generally very thick, and the dermoid cyst is relatively easy to be removed completely. Therefore, an intra-oral approach should be attempted at first, as it leads to very good cosmetic and functional results, even if the cyst was a large or inferior type. When the removal of the cyst was difficult using only the intra-oral approach, the extra-oral approach should be combined[9].

IV. Conclusion

Epidermoid cysts are rarely seen intraorally. When present, they might go unnoticed and assume large dimensions to impinge on the upper aerodigestive tract. Aspiration in a hospital setting and contrast-enhanced MRI are useful tools in diagnosis. Excision is the treatment of choice. The intraoral approach is the first preference, as it leads to very good cosmetic and functional results, but massive cysts may require an extraoral approach or combination of both approaches. Recurrence is generally uncommon.

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