

# Sub lingual Epidermoid Cyst in an Elderly Individual - Globus in the Oral Cavity - A Case Report and Literature Review

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**Abstract:** *Sublingual Epidermoid cyst is a rare lesion seen over floor of the mouth with a reported incidence of 1.6%. It can vary in size; symptoms can range from a complete asymptomatic presentation to a massive swelling causing airway obstruction (rare cases). Proper Clinical Examination and judicious advice of investigations such as FNAC, CT, MRI and Ultrasound can lead to diagnosis. Surgical Excision remains the main stay of treatment with very rare incidence of recurrence. In this article we reported a case of 6 x 6 cms sublingual epidermoid cyst which displaced the tongue to a greater extent and got managed by surgical excision via intraoral approach.*

**Keywords:** Epidermoid cyst, Sub lingual, Surgical Excision, intraoral approach

## 1. Introduction

Epidermoid cyst is a rare benign cystic malformation of epidermal origin. Head and neck epidermoid cysts have an incidence of 6.9 % of which 1.6 % cysts can be seen intraorally and floor of the mouth constitutes 0.01 % of all epidermoid cysts indicating the rarity of this cyst in this region. Teszler et al in 2007 gave a comprehensive classification for cysts over floor of the mouth in which they grouped these cysts into three main categories and each category involving median and lateral variants.1

Supra mylohyoid cysts (intraoral or sublingual).  
Inframyoeloid cysts (cervical).  
Peri and trans mylohyoid cysts (dual intraoral and cervical).

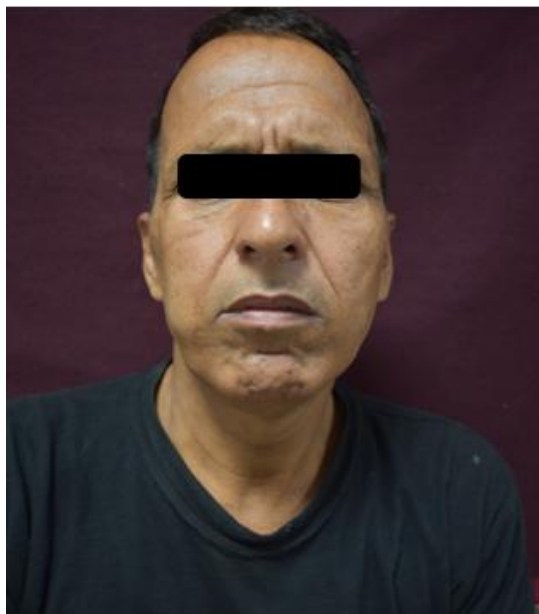
Here we present a case report on intraoral epidermoid cyst over floor of the mouth which was managed with surgical excision via intraoral approach.

## 2. Case Report

A 52-year-old Male reported to our department outpatient unit with a chief complaint of painless swelling measuring 6 X 5 cms in dimension below the tongue (Fig –1) and associated slurred speech for the last 9 months.



**Figure 1:** Clinical photograph showing intraoral appearance of the lesion Patient gave no history of any trauma, infection or previous surgery. No marked lymphadenopathy present. Extra oral examination revealed a mild swelling over sub mental area which is non-tender upon palpation (Fig –2).

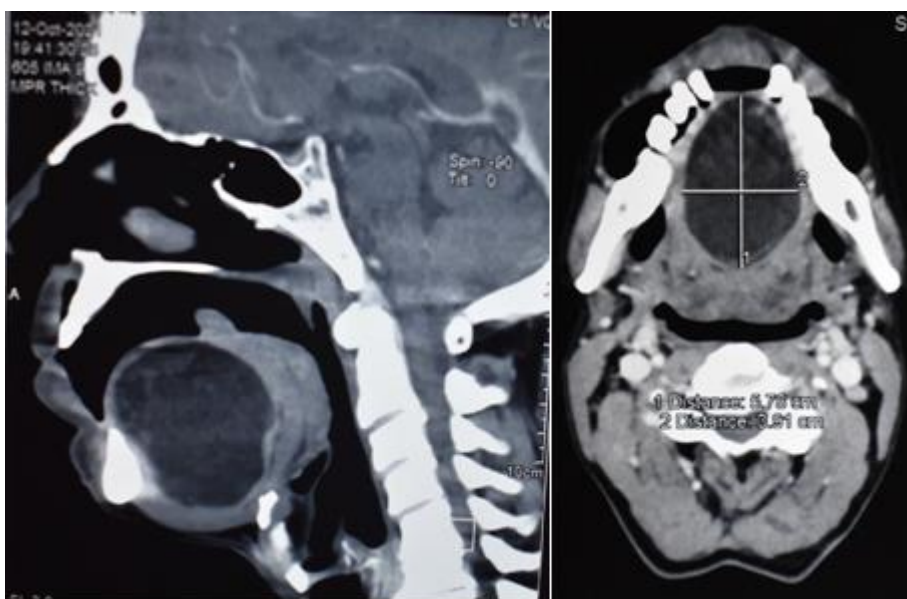


**Figure 2:** Clinical photograph showing extraoral appearance of the patient

A smooth surfaced swelling seen over the floor of the mouth displacing the tongue superiorly till the level of hard palate, covered with normal mucosa with no signs of any inflammation or infection. Upon palpation, swelling was nontender, non-compressible and had dough like consistency.

Clinical findings led us to provisionally diagnose it as either ranula (most common location) or dermoid cyst (midline swelling).

Ranula was ruled out by Sialographic examination. Ultrasonography and Contrast Enhanced Computed Tomography (CECT) revealed a well-defined cystic lesion measuring 6x4 cms filled with homogenous material. (Fig -3)



**Figure 3:** Contrast enhanced computed tomographic image (CECT) of the lesion

Upon examination, as the patient general health status presented no contraindication for surgery, he got operated under general anesthesia by surgical excision of the swelling by a horizontal incision on the mucosa over swelling. Using Alternate blunt dissection and cauterization, in toto delivery of the cyst was done via the intraoral supramylohyoid approach. (Fig -4). Surgical specimen (Fig -5) was sent for histopathological study. Figure -4: In toto delivery of cyst via intraoral approach

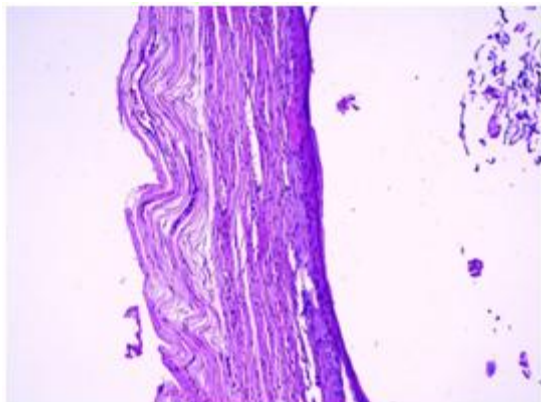


**Figure 5:** Surgical specimen

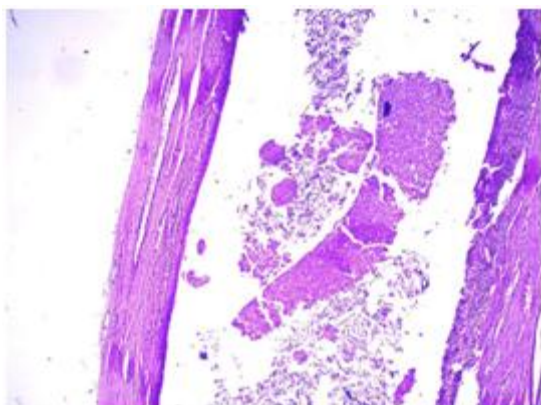
Histopathological analysis of surgical specimen revealed the cyst wall made of Fibrocollagenous tissue with occasional giant cells. cystic cavity is occupied with white cheesy material composed of abundant keratin flakes suggestive of an epidermoid cyst. (Fig-6A-6C)

Healing was uneventful and 6 months follow up showed no

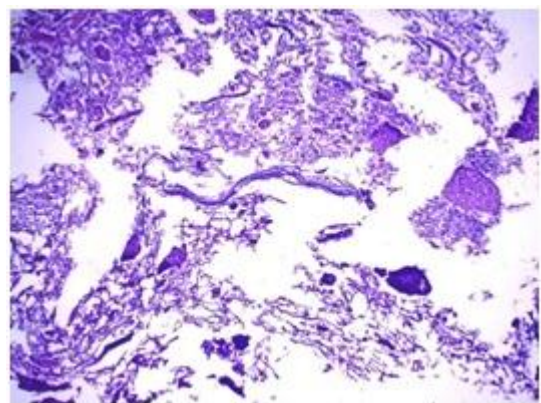
signs of any recurrence. Figure –6: Photomicrographs showing microscopic sections of the surgical specimen



**Figure 6 (A):** H&E (10×)-section of cyst lined by thinned out stratified squamous epithelium and filled with keratin flakes and inflammation.



**Figure 6 (B):** H&E (4×)-section of cyst wall lined by stratified squamous epithelium and filled with keratin flakes, inflammatory cells and occasional multinucleated giant cells.



**Figure 6 (C):** H&E (10×)-section show sheets of inflammatory cells with keratin flakes and multinucleated giant cells

### 3. Discussion

Epidermoid cysts constitute a rare variant of cystic malformations of head and neck region. Origin of which can be of two types.<sup>2</sup>

- 1) Developmental-Due to inclusion of epithelial remnants between midline structures, most commonly thought to

be the fusion area of 1st and 2nd branchial arches or from the remnants of tuberculum impar.

- 2) Acquired-post traumatic or iatrogenic implantation of epithelial cells into deeper tissues. Dermoid cysts are histologically classified into 3 subtypes by Meyers as 1) Epidermoid cyst-lined by stratified squamous epithelium with no skin adnexa.
- 3) Dermoid cyst/compound cyst-lined with stratified squamous epithelium and having skin adnexa like hair follicles.
- 4) Teratoma/complex cyst-epithelium lined cavity containing structures raising from all 3 germ layers.

Epidermoid cysts can measure from few millimeters to few centimeters with no sex predilection but few authors proposed a slight male predilection.<sup>3</sup> It can present from the time of the birth (congenital variant) but most cases will become apparent during 2nd-3rd decade of life<sup>4</sup> and there was a case reported even in an 84 old year-old female individual.

Sublingual epidermoid cyst presents clinically as a painless (unless infected) smooth surfaced firm swelling covered with normal oral mucosa. Swelling will displace the tongue giving a double tongue appearance intraorally in case of supramylohyoid variant and double chin appearance extra orally, in case of inframylohyoid variant.<sup>5</sup>

Symptoms can range from asymptomatic swelling to a massive swelling occupying the floor of mouth leading to dysphagia and in severe cases causing airway obstruction which has very rare incidence reported in literature.<sup>16</sup>

Clinical presentation gives the picture of a ranula which is common in this location.

FNAC of epidermoid cyst yields thick whitish material which under microscope shows keratin flakes and anucleate squames. The presence of squamous cells on FNAC rules out the possibility of a ranula.<sup>7</sup>

Computed Tomography and Magnetic Resonance Imaging are widely accepted modalities for imaging epidermoid cyst but MRI having obvious advantages over CT in terms of detailed soft tissue imaging and proper delineation of the extent of cyst.<sup>8</sup> Ultrasound examination will provide details of the cystic contents.

Best time to operate a sublingual epidermoid cyst is when it is first noted. Surgical treatment of choice is Enucleation of the cyst in toto. literature review shows decompression followed by marsupialization, performed in case of large sublingual cyst compromising the airway.<sup>9, 10</sup> Intraoral surgical approach is indicated for supramylohyoid variants and extra oral approach is indicated for inframylohyoid variants, if no contraindication exists.

Recurrence is very rare when it is excised completely.

Malignant transformation is also rare but there are cases reported in past where in a squamous cell carcinoma developed from an epidermoid cyst.<sup>11</sup>

#### 4. Review of Literature

Literature search for case reports of intraoral epidermoid cyst over floor of the mouth was done using keywords like

sublingual epidermoid cyst, floor of the mouth swellings in PubMed, some scientific websites and 31 articles were selected. Key findings from few of those articles were summarized in Table –1.

**Table 1:** Key points from previously published case reports on sublingual epidermoid cyst

S. No.	Author	Age	Sex	Key points
1	Katz and Passy.3	Mean age below 35 years	Equal sex predilection	3 variants sublingual group Geniohyoid group Lateral variants
2	Potts et al.8	22 years	Female	Clear delineation and nature of luminal fluid can be better defined using MRI than CT.
3	C. Macdonald worley.4	2-3rd decade of life	No sex predilection	Reported the unusual case of multiple sublingual and submental epidermoid cysts
4	A. Di Francesco.9	-	-	If the cyst is too large to be totally delivered through the midline vertical incision, it can be aspirated to reduce its volume and allow the dissection to continue to completion.
5	Vyomesh Bhatt.11	64 years	Female	Presented the case of squamous cell carcinoma that arose in the lining of an epidermoid cyst that developed within the sublingual gland
6	Lyngdoh T. S.10	24 years	Male	Introduced technique of initial decompression followed by partial excision and marsupialization in case of large sublingual cyst.
7	Mainak Dutta.5	23 years	Female	Stated the terms like “double chin” and “second tongue” appearances of sublingual epidermoid cysts
8	Chintan-C. Nishar.6	60 years	Male	Reported a case of tracheal collapse immediate post-surgery needing tracheostomy to maintain the airway.

#### 5. Conclusion

Case presented in this article is showing maximum of intraoral appearance and mild swelling extra orally giving double chin and double tongue appearance. Proper preoperative planning using ultrasound and CT examination lead to the proper diagnosis and revealed the supramylohyoid presence of the swelling which got surgically enucleated via intraoral approach. Even though it is having rare incidence, proper diagnosis leads to correct management with almost zero chances of recurrence.

#### 6. Disclosures

All authors report no disclosures associated with this case report Abstract word count: 105

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