

## Epidermoid Cyst of the Floor of the Mouth

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### Introduction

Epidermoid cyst in the floor of the mouth is an uncommon developmental lesion. The pathogenesis is the entrapment of the ectodermal tissue along embryonic fusion lines. The clinical presentation is a progressive swelling in the floor of the mouth and neck. If large enough, the cyst can potentially compromise the airway, speech, and swallowing. The main treatment is surgical excision by either intraoral or extraoral approach depend on the size and location of the cyst.

We present the case of a large sublingual epidermoid cyst with extension to the basal region of the tongue and compressing the oropharyngeal lumen excised by an intraoral approach. The patient was managed at our academic institution. [1]

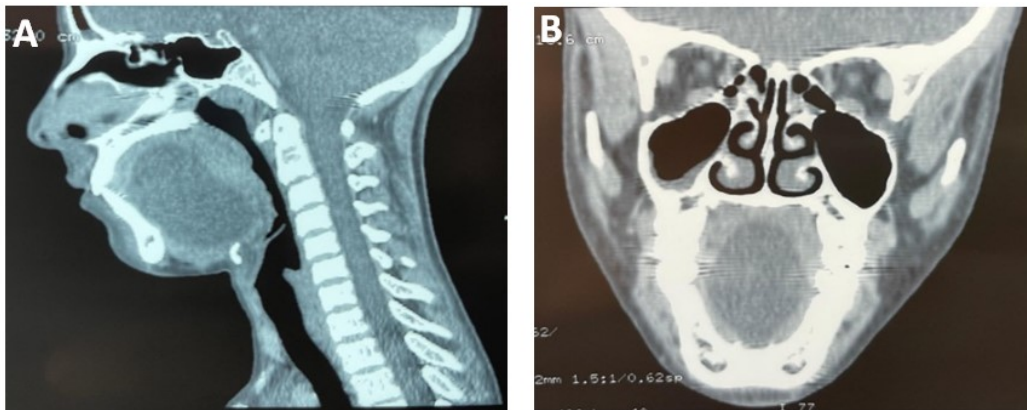
### Case Presentation

B. F, 15 years old, was admitted to our department with a swelling of the floor of the mouth, which had been present since birth and had progressively increased in size. Endobuccal examination revealed a regular swelling on the anterior floor of the mouth, 5 cm long in its long axis, soft and covered with normal-looking mucosa. This swelling pushes the tongue backwards, causing slurred speech and difficulty chewing. On clinical examination, a well-defined, rounded, soft mass measuring 5 cm in diameter and mobile in both planes is found in the submental region (Figure 1). Bipalpal palpation revealed a single mass with endobuccal and cervical expression. The rest of the cervicofacial examination was normal. Cervical ultrasound revealed a homogeneous, well-limited echogenic mass occupying the entire floor of the mouth; CT examination of the floor of the mouth showed a voluminous cystic lesion of the floor of the mouth measuring 52x30x57, without a septum, extending to the inferobasal region of the tongue and compressing the oropharyngeal lumen (Figure 2)

These clinical and radiological findings point to the diagnosis of a benign cystic lesion of the floor of the mouth, of unknown nature. Under general anaesthesia and nasotracheal intubation under nasofibroscopy, the cystic lesion of the floor of the mouth was completely removed in a single operation (Figure 3 and 4). Post-operative management was straightforward, with antibiotic treatment and parenteral nutrition for 48 hours (figure 5). Pathological examination concluded that the lesion was a epidermoid cyst of the floor of the mouth, with no histological signs of malignancy (Figure 6).



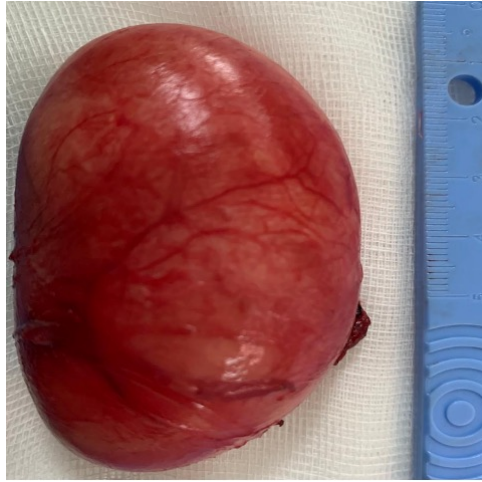
**Figure 1:** Endobuccal view



**Figure 2:** CT of sagittal view (A) and coronal view (B) demonstrates the left sublingual epidemoid cyst extended to the basal region of the tongue and compressing the oropharyngeal lumen measuring 55x30x55 mm.



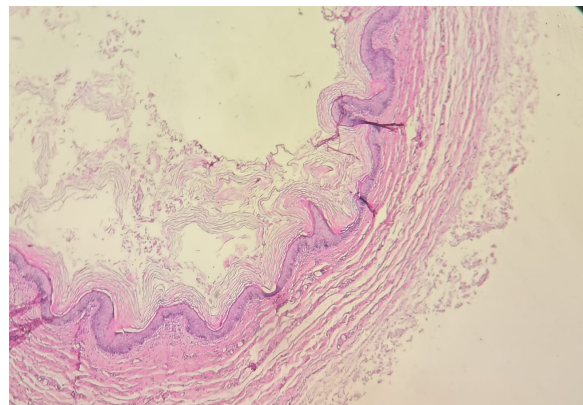
**Figure 3:** Intraoral Excision Sublingual Epidermoid Cyst



**Figure 4:** Surgical Specimen



**Figure 5:** Photo after 3 Days Of Surgery



**Figure 6:** Epidermoid Cyst: Cystic Formation Bordered By Squamous Epithelium and Containing Keratin (Hesx40)-457200278574500

## Discussion

Epidermoid and dermoid cysts constitute 1.6 to 6.9% of all cysts in the head and neck area. The incidence in the floor of the mouth of the oral cavity is rare and represented less than 0.01% of all cysts of the oral cavity. Sublingual, submaxillary and submandibular spaces are common localization in the floor of the mouth [2]. The floor of the mouth dermoid cyst is the second most common for the head and neck region after the lateral eyebrow. Two pathogenesis theories of the dermoid cyst have been described. The first theory is acquired implantation in which there is a traumatic inclusion of epithelial cells within the underlying tissue or from occlusion of the sebaceous gland. But the lesions only present following the clear history of trauma or surgery. The second theory, which is more common, is the congenital inclusion cysts that are formed by the epithelial entrapment during midline fusion of the first and second pharyngeal arches during the third and fourth weeks of gestation. Meyer I et al. firstly classified floor of mouth dermoid cyst into three variants based on histopathology which was epidermoid, dermoid and teratoid variants [3]. In 2013, Gordon et al. proposed to use the terminology “congenital germline fusion cyst” instead of “dermoid cyst” to avoid confusion with the teratoma entity, which is the true solid neoplasm. They also recommended adding the specific Meyer’s variant behind this terminology to specify three the histopathologic subtypes which are 1) Epidermoid cyst: epithelial-lined cyst wall with no adnexal structure. 2) True dermoid cyst: epithelial-lined cyst with skin appendages (sebaceous and sweat gland and hair follicle) in cyst wall or compound cyst. 3) Teratoid cyst: epithelium lining with the derivative of all three germ layers (bone, muscle, gastrointestinal tissue) or complex cyst [4]. Of the three variants, epidermoid is the most common, followed by dermoid and teratoid, respectively. There is a bimodal age distribution that can first present during infancy and later during puberty when hormone change leads to increased sebum production, but they can occur at any age.

Anatomically, the floor of mouth dermoid cyst can be divided into three regions as sublingual, submental and lateral according to the anatomic relationship between cyst and floor of mouth muscle. The majority of the cysts (78 %) are in the midline, 16 % involve more than one space and only 6% are located in the submandibular space [5]. The signs and symptoms depend on the location of the cyst. Above the mylohyoid muscle, the clinical presentation is swelling at the floor of the mouth or posterior tongue displacement which can later cause swallowing, speaking and even airway problems. Beneath the mylohyoid muscle, the patient will present with a submental or submandibular mass depending on the laterality of the cyst. In our case presentation, although the location of the cyst is at lateral sublingual space above the mylohyoid muscle, it displaced the submandibular gland posteroinferior more than the above sublingual gland. This made the patient presented with submandibular swelling rather than the floor of mouth swelling. The important differential diagnosis includes plunging ranula, vascular or lymphatic malformation, chronic sialadenitis, and salivary gland neoplasm.

MRI imaging allows us to define the location of the cyst and its relationship to geniohyoid and mylohyoid muscle. This information is important for selecting the optimal surgical approach. Dermoid cysts are isointense or hypointense to muscle on T1-weighted image and hyperintense or heterogeneous on T2-weighted image.

Fine needle aspiration cytology is useful in differentiating the cyst as it did in our case. However, aspiration of the high viscosity content cyst may be difficult and inadequate for interpretation.

A definite pathologic examination is essential for confirming the diagnosis. Surgical excision is the treatment of choice. An aspiration to decompress the cyst before endotracheal intubation may be useful for a very large cyst obstructing the airway. The surgical approach can be an intraoral or extraoral approach depending on the size and location of the cyst. The intraoral approach is recommended for a small to a medium sublingual cyst which is less than 6 cm in size and above the mylohyoid muscle, whereas an extraoral approach is appropriate for large cysts over 6 cm located or transgress below the mylohyoid muscle [5–7]. Although the cyst in our case was large (6.5 cm) and extended behind mylohyoid to involve the submandibular and parapharyngeal space, we chose the intraoral approach to avoid cosmetic problems. Most literature reports the cases of midline sublingual dermoid cysts transorally excised by dissection through the midline raphe of the tongue and floor of the mouth [8–11]. Gulati U et al. reported

the combined intraoral and extraoral approach to remove large lateral sublingual epidermoid cyst [12]. They did the submandibular approach and divided the mylohyoid muscle to expose the inferior part of the cyst and additional approach from the floor of mouth to dissect the superomedial aspect. In our present case, the cyst was in lateral sublingual space with posterior extension to parapharyngeal space. We proposed to remove the sublingual gland for two reasons. Firstly, the posterior portion of the sublingual gland obscures the view of posterior part of Wharton's duct, lingual nerve, and medial pterygoid muscle which are important structures needed to be preserved. After sublingual gland removal, there was more space to dissect the cyst from the surrounding muscle without disruption of the capsule. Secondly, the mucosal incision at lateral floor of mouth disrupts the duct opening of sublingual gland, if the gland is left, the mucocele or ranula will occur in the future. We report the first case of a large lateral sublingual epidermoid cyst with parapharyngeal extension removed by an intraoral approach. By excision of the sublingual gland first, an intraoral approach is possible for large lateral sublingual dermoid cysts that transgress mylohyoid muscle. The intraoral approach provided the patient with excellent cosmetic and functional outcomes. Recurrence is rare after complete excision but a 5% rate of malignant transformation of the dermoid and teratoid variant has been reported [5-7].

## Conclusion

Swelling on the floor of mouth has been a challenging site for diagnosis and management. A case of epidermoid cyst was diagnosed and treated effectively with imaging techniques. Surgical excision is the treatment of choice with intraoral approach in cysts lying above mylohyoid muscle and external approach in those lying below it. Differential diagnosis includes congenital lesions, infections, tumors and mucous extravasation cysts. Recurrence after complete surgical excision and malignant transformation are rare.

## References

1. RA Agha, MR Borrelli, R Farwana, K. Koshy, A Fowler, DP Orgill (2018) For the SCARE Group, The SCARE 2018 statement: updating consensus surgical case report (SCARE) guidelines, *Int. J. Surg*, 60: 132-6.
2. De Ponte FS, Brunelli A, Marchetti E, Bottini DJ (2002) Sublingual epidermoid cyst. *J Craniofac Surg*, 13: 308-10.
3. I. Meyer (1955) Dermoid cysts (dermoid) of the floor of the mouth, *Oral Surg. Oral Med. Oral Pathol*, 8: 1149-64.
4. PE Gordon, WC Faquin, E Lahey, LB Kaban (2013) Floor-of-mouth dermoid cysts: report 3 variants and a suggested change in terminology, *J. Oral Maxillofac. Surg*, 71: 1034-41.
5. CB Tezler, IA El-Naaj, O Emordi, M Luntz (2007) Dermoid cysts of the lateral floor of mouth : a comprehensive anatomic-surgical classification of cyst of oral floor, *J. Oral Maxillofac. Surg*, 65: 327-32.
6. N Ohta, T Watanabe, T Ito, T Kubota, Y Suzuki, A Ishida, et al. (2012) A case of sublingual dermoid cyst: extending the limits of the oral approach, *Case Rep. Otolaryngology*, 2012: 634949.
7. F Longo, P Maremonti, GM Mangone, G De Maria, L Califano (2003) Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques, *Plast. Reconstr. Surg*, 112: 1560-5.
8. S Aydin, MG Demir, N Demir, S Sahin, SS Kayipmaz (2016) A giant plunging sublingual dermoid cyst excised by intraoral approach, *J. Maxillofac. Oral Surg*, 15: 277-80.
9. RK Sahoo, PK Sahoo, D Mohapatra, S Subudhi (2015) Two concurrent large epidermoid cysts in sublingual and submental region resembling plunging ranula: report of a rare case, *Ann. Maxillofac. Surg*, 7: 155-8.
10. M Baliga, N Shenoy, D Poojary, R Mohan R (2015) Naik, Epidermoid cyst of the floor of the mouth, *J. Maxillofac. Surg*, 5: 79-83.
11. JO Voss, S Buehling, N Thieme, C Doll, K Hauptmann, M Heiland, et al. (2018) Sublingual cysts of different entities in an infant - a case report and literature review, *Int. J. Pediatr. Otorhinolaryngol*. 113: 260-5.
12. U Gulati, S Mohanty, J Augustine, SR Gupta (2015) Potentially fatal supramylohyoid sublingual epidermoid cyst, *J. Maxillofac. Oral Surg*, 14: 355-9.