

## Case Report

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# Sublingual dermoid: an uncommon presentation and review of literature

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## ABSTRACT

Dermoid cysts are generally seen in the areas of embryonic fusion, in the midline, either following sequestration of ectodermal tissue, or due to failure of separation of the ectoderm from the mesoderm during third to fifth weeks of gestation. Patient information: An eighteen year old female patient presented to department of otorhinolaryngology with a painless swelling in the floor of mouth on right side. Physical examination: the swelling was cystic in consistency, bluish in color, translucent, globular shaped, nontender with a right soft submental swelling. Diagnostic assessment: both ultrasound and computed tomography of the swelling was done which reported as ranula. Interventions; under aseptic precautions and general anesthesia, elective excision of the cyst was done by intraoral approach and specimen was sent for histopathological examination. Postoperative period was uneventful. Follow up and outcome: on follow up the patient was stable and the histopathological report revealed dermoid cyst. A lateral to midline presentation of sublingual dermoid cyst in the floor of mouth with recent occurrence of symptoms and no history of any trauma is rare, hence should be considered as one of the differential diagnosis for floor of the mouth cystic swellings. Ranula and dermoid cysts have similar clinical presentation with similar ultrasound, computed tomography and magnetic resonance imaging findings. The only distinguishable investigation is histopathological examination and all the specimens have to be sent for histopathological examination irrespective of the preoperative investigation reports.

**Keywords:** Dermoid cyst, Sublingual, Ranula, Cystic swelling

## INTRODUCTION

The aetiology of dermoid cyst is not clear but can be associated with trapped cells as a result of the inclusion error resulting in the development into the ectoderm, mesoderm and endodermal tissues.<sup>1</sup> These conditions may produce hair, muscle, bone, cartilage, teeth, and mucous membranes. Historical trauma, infection, and spontaneous autonomous new growth are closely related to these lesions.<sup>1</sup>

These benign lesions are encountered throughout the body and 7% in the head and neck region, 1.6% in oral

cavity, and represent less than 0.01% of all oral cavity cysts.<sup>2</sup> Etiologically, dermoid cysts may be congenital or acquired. The congenital type found in the cervicofacial region is derived from entrapment of epithelial cells during midline fusion in embryonic development. Acquired forms develop from traumatic or iatrogenic implantation of epithelial cells into surrounding tissues.<sup>3</sup> Even though they are generally diagnosed in the second and third decades of life, they can present at any age with equal frequency of occurrence to both genders.<sup>4</sup> Depending on the size of the lesion, it can displace the tongue and cause dysphagia, dysphonia, and dyspnea.<sup>5</sup>

In current article we present a rare case report of a case reported as ranula in all the investigations and diagnosed as dermoid cyst on histopathology.

## CASE REPORT

### Patient information

An eighteen year old female patient presented to the department of otorhinolaryngology, Vijayanagar institute of medical sciences, Ballari during September 2019 with complaints of swelling in the floor of the mouth since the past 2 months, with no other known comorbidities. The swelling was insidious in onset, gradually progressive. The swelling was not associated with pain or excessive salivation, fever, difficulty in opening mouth or difficulty in swallowing or breathing. No history of any dental complaints, trauma or any increase in the size of swelling or pain with the oral feeds. No history of any surgeries or dental procedures in the past. No history of similar complaints in the family.

### Physical examination

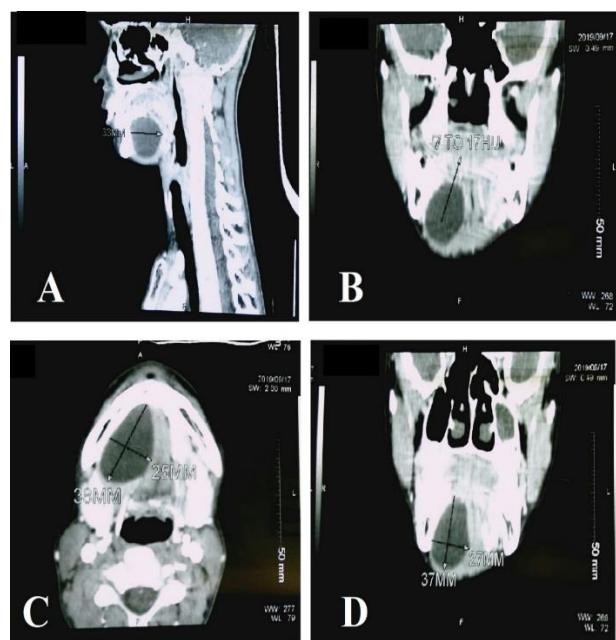
A young female patient who was moderately built and nourished, conscious, cooperative, well oriented presented with stable vitals. On examination of the oral cavity, a solitary bluish swelling was in the floor of the mouth, on the right side of the midline, which was extending from the frenulum anteriorly to posteriorly upto the level of the third molar tooth along the gingivolingual sulcus. The swelling was 3x4 cm in size, oval in shape, regular borders and had smooth surface (Figure 1A). On palpation there was no local rise in temperature, was nontender, cystic in consistency, fluctuation was present. The swelling was translucent in nature. On bidigital palpation, ballotment was absent. The swelling over the floor of the mouth elevated the tongue, hence restricted the right lateral movement of the tongue. Oropharyngeal and indirect laryngoscopic examination was normal. On examination of neck a diffuse solitary swelling of approximately 3x3 cm was present in the right submental region, which was nontender with no local rise of temperature, soft in consistency, smooth surface and diffuses margins. The skin over the swelling was normal and pinchable (Figure 1B). No enlarged lymph nodes. Ear, nose and paranasal sinus examination was normal.



**Figure 1: (A) Clinical photograph showing right sided floor of the mouth cystic swelling and (B) right submental swelling (black arrow).**

### Diagnostic assessment

Patient was subjected to routine blood tests which were found to be in normal range. An USG neck suggested a well-defined cystic lesion in the floor of the mouth. CT neck was done which suggested well defined cystic lesion in the floor of the mouth of size measuring 37x25x33 mm), of 7 to 17 HU, suggestive of ranula (Figure 2A-2D). Considering the history, examination findings and investigation reports, patient was diagnosed to have ranula over the floor of mouth on the right side and was posted for elective surgical excision/marsupialization of the cyst under general anesthesia.



**Figure 2: (A)-(D) CECT scan of neck in sagittal, coronal and axial planes showing a well-defined cystic lesion in the floor of the mouth.**

### Intervention

After taking informed and written consent from patient and patient's parents, she was posted for excision of the ranula via intraoral approach under general anesthesia (Figure 3A). A curvilinear mucosal incision was made on the cyst; cyst wall was dissected protecting the submandibular gland, duct, lingual nerve and lingual artery. It was observed that the cyst was present above the mylohyoid muscle, pushing the muscle downwards which presented as the neck swelling. The cyst was excised intoto and sent for histopathological examination (HPE) (Figure 3B). Thorough saline wash was given and was inspected for any remnants. Hemostasis achieved and the wound was closed with 3-0 vicryl absorbable sutures.

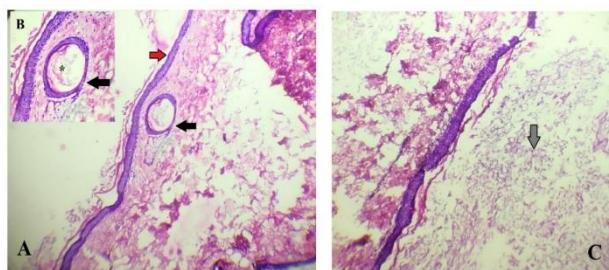
Postoperative period was uneventful and was discharged on second postoperative day with oral antibiotics and anti-inflammatory medications and reviewed regularly.



**Figure 3: (A) Intra-operative picture of the cyst excision and (B) post-operative dissected specimen of the cyst.**

#### Follow up and outcome

Patient was called for follow up after 3 days of discharge, wound healing well with no signs of infection. The histopathology report (HPE) of the cyst revealed the cyst wall was composed of stratified squamous epithelium enclosing abundant keratin debris and lamellar keratin and resting on a fibrocollagenous and fibroadipose tissue stroma containing pilosebaceous glands features suggestive of dermoid cyst (Figure 4A-4C).



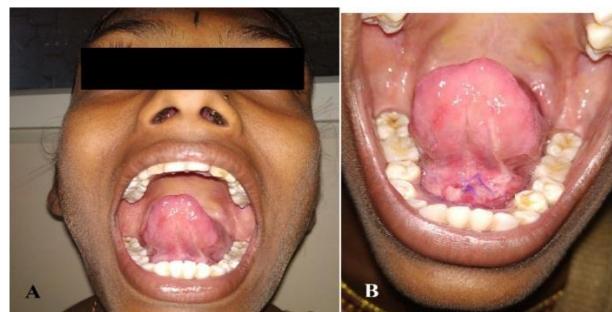
**Figure 4: (A) Histopathological report of the specimen showing the cyst wall composing of stratified squamous epithelium (red arrow) enclosing abundant keratin debris (grey arrow), (B) picture in the inset, high power 100X view showing pilosebaceous glands (black arrow) and (C) and lamellar keratin (\*) and resting on a fibrocollagenous and fibroadipose tissue stroma containing pilosebaceous glands (black arrow), features suggestive of dermoid cyst, abundant keratin debris (grey arrow).**

Patients attenders were explained about the histopathological report and were reassured about the benign nature of the cyst, however to do a regular follow up with us to look for signs of recurrence. Patient was later followed up at regular intervals and no signs of recurrence or inflammation were observed at the surgical site (Figure 5A-5B).

#### DISCUSSION

Dermoid cysts are generally seen in the areas of embryonic fusion, in the midline, either following sequestration of ectodermal tissue, or due to failure of separation of the ectoderm from the mesoderm during

third to fifth weeks of gestation.<sup>6-8</sup> However, their most common location in the head and neck region is around the eyes, followed by dorsum of the nose and anterior aspect of neck.<sup>9</sup> When found in oral cavity, dermoid cysts are classified as nonodontogenic lesions and about 7% occur in head and neck region, among them 23% are located at floor of mouth and can be found either lateral to tongue or in the midline.<sup>10,11</sup> While dermoid cysts can be seen at virtually any age, the highest incidence occurs in patients between 15 and 35 years of age, with no gender predilection.<sup>12</sup>



**Figure 5: (A) Postoperative day 10 picture showing the normal of the position of tongue and (B) intraoral picture showing healed scar.**

The international literatures suggested three theories with regard to the origin of cysts in the floor of the mouth. According to the first and most prevalent theory, these cysts originate from embryonic cells of the first and second branchial arch entrapped in the mesenchyme of the area during the third to fourth week of embryonic life. With regard to the second theory, it explains the pathogenic mechanism of the acquired form. The acquired cysts may be due to the implantation of epithelial cells subsequent to accidental or surgical injury (traumatic causes, iatrogenic antecedents, or an occlusion of a sebaceous gland duct). Lastly, the third theory maintains that these cysts are considered a variation of the cyst of the thyroglossal pore.<sup>13-17</sup> They are classified histologically as epidermoid cyst (lined by simple squamous epithelium without any adnexal structures), true dermoid cyst (epithelium with pilosebaceous apparatus and apocrine or eccrine skin appendages), and teratoid (contains ectodermal, mesodermal and endodermal derivatives). The most common of these cysts in head and neck are epidermoid, followed by true dermoid and then teratoid variants.<sup>18</sup> In our patient, the histology was consistent with true dermoid cyst.

Clinically, these lesions present as slow-growing and painless swellings, soft, well encapsulated, and without lymphadenopathy.<sup>19</sup> Depending on the anatomic location of the cyst and the muscles of the floor of the mouth, dermoid cysts may be defined as sublingual or submental.<sup>20</sup> A sublingual cyst is located above the geniohyoid muscle and swelling with displacement of the tongue may occur. In cases that develop below the geniohyoid, a submental swelling with a double chin

appearance may occur.<sup>21</sup> In current case, the swelling present in floor of the mouth, above mylohyoid and geniohyoid muscle, pushed the tongue upwards which restricted right lateral movement of tongue and produced a right submental swelling. These lesions may become secondarily infected and in rare cases may undergo a malignant change.<sup>21</sup> In current case, there was no signs of inflammation.

In present case report, the cyst presented with lateral presentation. They are considered to occur in the midline, though there are reports of dermoids occurring in the lateral aspect of the floor of the mouth.<sup>22,23</sup> The differential diagnosis for floor of the mouth cystic lesions are ranula, cystic hygroma, thyroglossal duct cyst, brachial cleft cyst, infectious cysts, lymphatic malformation, tumors, hemangioma, salivary lesions, and Ludwig's angina only in cases of inflammatory complications.<sup>24</sup>

An ultrasound scan is commonly used as the first choice to investigate the lesion.<sup>25</sup> Dermoid cysts are unilocular, well circumscribed, anechoic or hypoechoic on ultrasound scan. On CT scan they are homo or heterogenous with or without subtle rim enhancement. On MRI dermoid cysts are hyperintense on T2 and variable intensity on T1. Dermoid cysts have a sack of marbles appearance due to the presence of fat globules.<sup>26</sup> Ranulas are anechoic, lobulated cystic lesion with no doppler flow on USG, homogenous unilocular on CT, T2 hyperintense and T1 variable on MRI.<sup>26</sup> Due to the similarities in the USG, CT, MRI findings in both ranula and dermoid cysts, they can be confused and hence a mandatory HPE has to be done. In our case report all the investigations including ultrasound and CT scan pointed towards ranula, but the HPE report suggested dermoid cyst which gives us the conclusion that all the postoperative specimens must be sent for histopathology irrespective of the pre-operative investigation reports.

The dermoid cyst may cause life threatening situations if left untreated.<sup>27</sup> The treatment of choice is surgical enucleation via an intraoral or extraoral approach, which is facilitated by the presence of a capsule.<sup>27</sup> The intraoral approach is used in large, deeply seated, non-infected lesions and led to good cosmetic and functional results.<sup>27</sup> The extraoral approach is used for very large dermoid involving the floor of the mouth and submental space and in severe infection that compromises the patient's airway.<sup>27</sup> In our case an intraoral approach was sufficient as the cyst was non-infected and was present above the mylohyoid muscle.

## CONCLUSION

A lateral to midline presentation of sublingual dermoid cyst in the floor of mouth with recent occurrence of symptoms and no history of any trauma is rare. Ranula and dermoid cysts have similar clinical presentation with similar USG, CT and MRI findings. The only

distinguishable investigation is histopathological examination. Hence, we suggest that lateral to midline presentation of dermoid cyst can occur rarely and should be considered as one of the differential diagnosis for floor of the mouth cystic swellings and all the specimens have to be sent for histopathological examination irrespective of the preoperative investigation reports.

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