

# Sublingual dermoid cyst: case report and literature review

## Précis

Sublingual dermoid cysts are rare, benign cysts containing dermal adnexal structures, which present in the floor of the mouth. This case highlights the clinical findings and management of a sublingual dermoid cyst.

## Abstract

This case report reviews the clinical features and surgical management of a large sublingual dermoid cyst. A 15-year-old male patient presented with a large midline floor of the mouth swelling and associated displacement of the tongue. Speech changes were noted on presentation. A magnetic resonance imaging (MRI) scan revealed a 7x4x3cm mass in the submental and sublingual spaces. Fine needle aspiration suggested a sublingual dermoid cyst. The cyst was excised via an intraoral approach. Sublingual dermoid cysts are rare entities, and their management has been poorly described. This case report illustrates the benefit of an intraoral surgical approach.

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## Introduction

Sublingual dermoid cysts are rare entities that occur in the floor of the mouth, representing less than 0.01% of all oral cavity cysts.<sup>1-5</sup> The cyst can develop superior to the geniohyoid/mylohyoid muscles, resulting in an upward displacement of the tongue towards the palate and oropharynx.<sup>1,2</sup> The resulting functional implications include potential difficulty with eating (dysphagia), speech (dystonia), respiration (dyspnoea) and cosmesis. They may also develop inferior to the geniohyoid/mylohyoid muscles, giving rise to a 'double chin' appearance.<sup>1</sup>

Sublingual dermoid cysts can vary in size. They are slow growing and are usually diagnosed in the first to third decade.<sup>1,2</sup> There is no specific gender predisposition; however, the literature has noted a greater male predominance in some case series.<sup>1</sup> They are usually asymptomatic and indolent.<sup>3</sup>

Sublingual dermoid cysts originate in the midline of the floor of the mouth, but may extend laterally and inferiorly, and can attain a large size before clinical presentation.<sup>6,7</sup> A sudden increase in size has been reported during the onset of puberty due to an increased production of sebum from the sebaceous glands.<sup>8</sup> Alternatively, sudden expansion may be due to secondary infections with drainage intraorally or cervically.<sup>6,9,10</sup>

Sublingual dermoid cysts are squamous epithelial-lined cysts containing dermal adnexal structures within the cyst cavity. These cysts differ

histologically from epidermoid cysts, which lack skin appendages, and the latter may represent the simplest form on the teratoma spectrum. These are not to be confused with epidermoid cyst of the skin, a non-teratomatous cyst arising from the hair follicle.<sup>4</sup> The true incidence of sublingual dermoid cysts is not precisely known due to the inclusion of epidermoid cysts and teratomas in many publications.<sup>9,11,12</sup> True sublingual dermoid cysts are rare entities, and their management is poorly described.

The literature concludes that definitive management of sublingual dermoid cysts is complete surgical excision.<sup>1-8</sup> Multiple surgical approaches have been advocated depending on the location of the cyst. Those located superior to the geniohyoid can be removed intraorally,<sup>13-16</sup> while sublingual dermoid cysts occurring inferior to the geniohyoid may require an extraoral approach.<sup>14-16</sup>

The clinical presentation, appropriate investigations, and surgical management of a large sublingual dermoid cyst via an intraoral approach are described here.

## Case

A 15-year-old male presented with a sublingual swelling, located in the anterior floor of the mouth. The swelling had been increasing in size over the previous months. He complained of swallowing difficulties, voice change and restrictive tongue movements. Examination revealed a firm, non-tender



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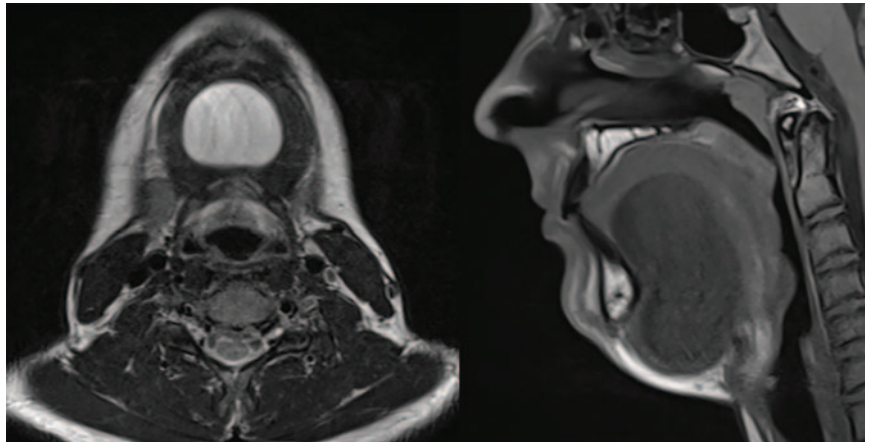
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**FIGURE 1:** Intraoral view of the midline sublingual swelling in the floor of the mouth. The swelling had a dough-like consistency and was mobile with no evidence of mucosal changes.



**FIGURE 2:** Axial (left) and sagittal (right) magnetic resonance imaging (MRI) scan of a 7x4x3cm mass in the floor of the mouth. The mass is homogenous and had a high signal on T2 and a low signal on T1 MRI. The superior margin abuts the genioglossus, and extends to the base of the tongue posteriorly, between the geniohyoid muscles, to the level of the hyoid inferiorly.

sublingual expansion located in the midline of the floor of the mouth (**Figure 1**). The swelling was mobile, had a dough-like consistency on bimanual palpation, and was well demarcated. No overlying mucosal or skin changes were noted. There was displacement of Wharton's ducts laterally and caudally; however, salivary flow was satisfactory.

### Cytology

Aspiration of the mass revealed cloudy, straw-coloured intraluminal debris. Cytological examination of this fluid demonstrated occasional benign squamous epithelial cells together with proteinaceous material. These features were suggestive of a sublingual dermoid cyst and the authors elected to excise the sublingual mass.<sup>3,4</sup>

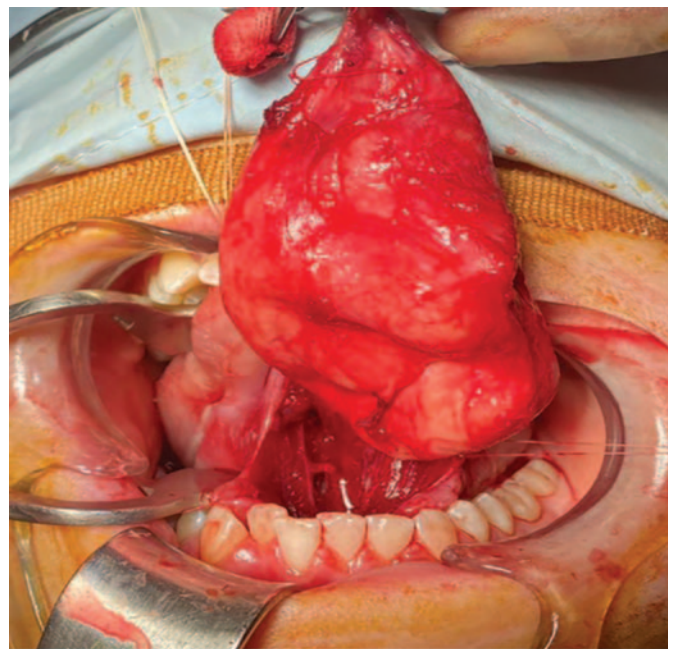
### Imaging

Radiographic imaging may include computerised tomography (CT), magnetic resonance imaging (MRI) and ultrasonography. Soft tissues are represented accurately on MRI imaging and this is helpful to visualise the extent of the sublingual dermoid cyst.<sup>10</sup> **Figure 2** shows an MRI scan of the sublingual dermoid cyst. This scan highlights vital adjacent structures such as the muscular margins, the dimensions of the sublingual dermoid cyst, and its relationship to the airway. The MRI influences the treatment planning process and the surgical access for evacuation of the cyst and its contents.<sup>17,18</sup>

### Surgical procedure

An intraoral excision was the approach of choice. Under general anaesthetic via a nasal intubation, traction sutures were secured through the tip of the tongue and to allow access to the ventral aspect of the tongue and floor of the mouth.

Initial cannulation of Wharton's duct with lacrimal probes facilitated identification and orientation of the ducts. The dissection was carried out using monopolar electrocautery immediately superficial to the mass. Care was taken not to interfere with the cyst lining and retraction of Wharton's ducts. Once the dissection plane between the cyst wall and mucosa was established, dissection along this avascular plane was completed. This involved dissecting over the superior aspect of the cyst and elevating the genioglossus muscle



**FIGURE 3:** Intraoperative view of the intact cyst after dissection. Areas of adhesion to the surrounding muscles were apparent between the geniohyoid and mylohyoid muscles during the excision.

fibres off the capsule. Laterally, the cyst travelled between the geniohyoid muscles, and lateral retraction of the geniohyoid muscles allowed access to the submental space. The inferior surface was relieved with gentle digital pressure from the submental region. The inferior cyst wall was dissected from the mylohyoid and the lesion delivered intact (**Figures 3, 4, and 5**). Meticulous haemostasis was achieved to avoid haematoma formation. Submental drains were inserted and secured following excision of the lesion.

### Histopathology

The specimen was a 7.4x4.2x3.5cm orthokeratinised squamous epithelial lined cyst with skin adnexal structures in the cyst wall consistent with a sublingual



FIGURE 4: Sublingual cavity post excision. Haemostasis was achieved perioperatively. Two submental drains were inserted to avoid haematoma in the large dead space.



FIGURE 5: A) Dermoid cyst excised B) Incision into the capsule with layers of keratinous sebum-like content.

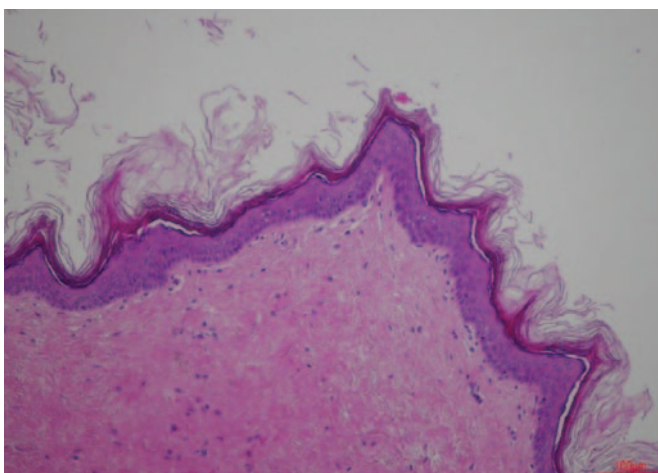


FIGURE 6: Photomicrograph of the dermoid cyst containing keratin lined by orthokeratinised squamous epithelium with skin adnexal structures in the cyst wall (haematoxylin-eosin stain).

dermoid cyst.<sup>10</sup> Haemorrhage, inflammatory infiltrate, and multinucleated cells were noted, in keeping with the previous aspiration.<sup>10</sup> Figure 5 shows the sublingual dermoid cyst lining intact (A) and excised (B). Cystic contents such as skin adnexal structures and keratin are evident in the grey/cream matter. (Figure 6)<sup>10</sup>

There were no postoperative complications. The drains were removed on the first postoperative day and the wound healed without complication. There were no changes in speech, neurosensory deficits, salivary pathology, or functional problems at six-month follow-up. No recurrence was detected.

### Discussion

Sublingual dermoid cysts are exceptionally rare with no significant gender predilection.<sup>13</sup> They present as a painless, indolent swelling that may cause functional and cosmetic deficits. Their pathogenesis is uncertain but theories include developmental malformations caused by a defect in the fusion of embryonic mesenchyme,<sup>7</sup> entrapment and proliferation of epithelial debris during midline fusion of the embryonic tongue during the third and fourth intrauterine weeks,<sup>19</sup> traumatic implantation of epithelium,<sup>20</sup> or displacement of normal epithelial bands, destined to differentiate from their usual

destination.<sup>20</sup>

Histologically, sublingual dermoid cysts are lined by orthokeratinised stratified squamous epithelium with a prominent granular cell layer. Abundant keratin is often found within the cyst lumen. Areas of respiratory epithelium can be seen. The cyst wall is composed of fibrous connective tissue that contains one or more skin appendages such as sebaceous glands, hair follicles or sweat glands. Anatomically, sublingual dermoid cysts are divided into three categories: median genioglossal (supramylohyoid), median geniohyoid (inframyllohyoid) and lateral. Our case anatomically represents a large median geniohyoid sublingual dermoid cyst. The range of size of sublingual dermoid cysts varies in the literature from 12mm to 12cm at the greatest dimension.<sup>21</sup> Dimensions of 6cm and more have been classified as large. Size can influence the surgical approach by means of an intraoral or a submental external approach.<sup>22</sup>

The differential diagnosis for a floor of the mouth swelling can include: neoplasms (lymphangioma, lipoma, haemangioma, angioma); infections (odontogenic, salivary gland); and, developmental processes (ranula, cystic hygroma, thyroglossal duct cyst and ectopic thyroid tissue).<sup>17,18,22</sup>

The diagnosis is predominantly made using imaging such as CT, MRI or ultrasonography demonstrating a cystic nature, size and anatomical location.<sup>10,17,22,23</sup> In a study to determine different methods of radiographic imaging, MRI was identified as the recommended method in demonstrating exact position, extension and demarcation.<sup>24</sup> Fine needle aspiration cytology and ultrasound with Doppler mode can also be utilised for the diagnosis.<sup>25,26</sup>

Aspiration of the cyst contents has been advocated by Di Francesco;<sup>27</sup> however, this approach is controversial as it poses the risk of spilling the luminal contents and increases the risk of infection.

The definitive treatment of sublingual dermoid cysts is surgical excision. Multiple techniques have been described, which are divided into intra- and extraoral techniques, depending on the size and location of the cyst.<sup>28</sup> For small to moderate-sized cysts, an intraoral approach is advocated, while an extraoral submental approach is advocated for large cysts or cysts below the geniohyoids.<sup>1,29,30,31</sup> This is due to the concern of inadequate access for removal and the risk of recurrence.<sup>27</sup> Secondly, salivary contamination and infection have been described as potential risk factors for intraoral excision.<sup>27</sup> The literature reports an intraoral approach predominance (59.3%), versus 30.5% for an extraoral approach in the review by Vélez Cruz *et al.*<sup>21</sup> A



combined approach was advocated in two of the 58 cases reported.<sup>21</sup> Intraorally, Brusati<sup>30</sup> proposed a midline glossotomy, while Di Francesco<sup>27</sup> described a modified version with partial evacuation of the cyst. The midline sagittal glossotomy provides good access, but has the drawback of increasing postoperative pain and oedema, and potential airway compromise. Additionally, it may result in a forked tongue or changes in articulation.<sup>11</sup> Extraorally, a transcuteaneous approach has been described for sublingual dermoid cysts between or inferior to the geniohyoid. This results in a submental scar and also decreases access during the superior dissection of the capsule.<sup>27</sup> A secondary intraoral incision can be used, but may result in an oro-cutaneous fistula.<sup>27</sup> Finally, a symphyseal mandibular osteotomy has been described for very large sublingual cysts in the dentate population.<sup>32</sup> This increased exposure comes at the expense of increased pain, oedema, additional risk of damage to the dentition, and periodontal defects.<sup>32</sup> General anaesthetic is recommended for all cases except for very small, superficial cases as it permits greater patient comfort and greater retraction, visualisation and access. Nasotracheal intubation is indicated to avoid any interference with the surgical field.<sup>33</sup> The procedure is performed with the patient in supine position.<sup>33</sup> The chosen surgical approach must provide appropriate access to the cyst to permit complete excision, as any cystic remnant will result in recurrence.<sup>33</sup> The authors disagree that only small to moderate cysts can be surgically excised using an intraoral approach.<sup>32</sup> As demonstrated in this case, large cysts can be excised using an intraoral approach with good cosmetic and functional results. Cannulation of the Wharton's ducts will help to identify and avoid inadvertent damage to them. Intraoral access to sublingual and even submental spaces can be achieved through an intraoral approach, thereby avoiding an osteotomy of the mandible or skin incision in the submental fold, which should be considered when the cyst is well defined.<sup>33</sup>

## Conclusion

Sublingual dermoid cysts are rare. Their management has been reported in case reports and case series, which have been based on surgical expertise and experience. This case highlights important diagnostic measures, surgical planning and intra-operative techniques to manage a large sublingual dermoid cyst.

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## CPD questions

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1. Dermoid cysts in the oral cavity predominantly occur in the:

- A: Buccal mucosa
- B: Hard palate
- C: Floor of mouth
- D: Dorsum of tongue

2. Anatomical demarcation of a floor of mouth dermoid cyst is based upon:

- A: Size
- B: Stage of development
- C: Suprahyoid muscular group
- D: Proximity to the hyoid bone

3. Dermoid cysts differ from epidermoid cysts due to the presence of:

- A: An epithelial lining
- B: Adnexal structures
- C: Keratin
- D: Laminated internal contents



CPD