

# Sublingual dermoid cyst: A case report and review of the literature

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## Abstract

**Background:** Dermoid cysts in Oral cavity & mouth are relatively uncommon developmental anomaly. They are thought to arise in the midline and along the lines of embryonic fusion of the facial processes containing ectodermal tissue.

*Case report:* A 21-year-old female presented with a 2 year history of a swelling in the floor of mouth. Clinical examination revealed a solitary midline swelling in floor of mouth measuring approx. 3\*3 cm non-tender, pinkish, non-fluctuant and doughy consistency with no secondary changes over mucosa displacing the tongue superiorly. The fast growing nature and size of the lesion raised suspicion of potential compromise to the airway. Surgical excision was therefore performed.

*Conclusion:* Differential diagnosis of cystic lesions in the floor of the mouth is of paramount importance, as the recommended surgical techniques vary depending on the anatomical site of the lesions. Treatment depends upon the size of the dermoid cyst and the site and its relation with mylohyoid muscle. Cyst could be excised by intraoral approach or extra-oral approach. The intraoral approach is generally preferred for those lesions that do not extend beyond the mylohyoid muscle boundaries; this leads to a satisfactory cosmetic and functional outcome.

## Introduction

Dermoid cysts in the floor of the mouth are relatively uncommon developmental anomaly. They are thought to arise as a result of a defective development along embryonic lines of fusion containing both ectodermal and endodermal elements. The lesions tend to be slow growing. Geographical incidence rates are variable, ranging from 0.02 to 1.8 per cent.<sup>1</sup> The reappears to be no sex predilection. The lesions are associated with a bimodal age distribution, with the greatest peak during the teenage years and a smaller peak during the first year of life, although cases have been reported in patients as young as 7 months and as old as 77 years.<sup>2-5</sup>

In this paper, we present an unusual case of a 21-year-old female diagnosed with a rapidly growing dermoid cyst in the mouth floor. A review of the literature was performed; the aetiology and management of these lesions are discussed.

## Casereport

A 21-year-old female came to E.N.T. OPD of RDGMC Ujjain with a 2 year history of rapidly growing swelling in the floor of mouth. The patient did not report any eating or swallowing difficulties but her speech was mildly affected. She had no dyspnea or pain. There was no history of any previous surgery or trauma to oral cavity of neck. Patient was otherwise healthy.

Clinical examination revealed solitary midline swelling in floor of mouth measuring approx. 3\*3 cm non-tender, pinkish, non-fluctuant and doughy consistency with no secondary changes over mucosa displacing the tongue superiorly

Magnetic resonance imaging (MRI) demonstrated a 3 cm well-defined, thin-walled cystic mass that lay within the sub-lingual space, with no extensions through the mylohyoid muscle (Figure 2). There was obstruction to the submandibular glands due to a pressure effect on the submandibular papillae. Minor displacement of the epiglottis was also evident.

The fast growing nature of the cyst and its size raised the suspicion of potential compromise to the airway. Surgical excision was therefore advised.

Under general anesthesia, the patient underwent surgical enucleation of the cyst via an intraoral approach. The cyst was dissected from surrounding tissues en bloc (Figure 3) and the wound was closed primarily.

The procedure was uncomplicated and the tongue went back to its original position. Histopathology confirmed the diagnosis of a dermoid cyst (Figure 4).

## Discussion

According to the literature it was found that only 6.94% of the dermoid cyst occurred in the head and neck of which 11% were located in the floor of mouth. Malignant tumor was ruled out in view of the lesions clinical aspect and the absence of lymphadenopathy. The two most plausible differential diagnosis of the condition were ranula or an embryonic abnormality. (Table 1) summarizes differential diagnosis of sublingual and cervical swelling. The pathogenesis of midline shift of mouth floor is not well established. Meyer divided cysts of the mouth floor into three categories based on histology: dermoid, epidermoid and teratoid.<sup>6</sup> In fact, dermoid cysts occur primarily in the testes and ovaries, and the most common location in the head and neck is the external third of the eyebrow.<sup>9</sup>



FIG.1

Pre-operative clinical presentation of the dome-shaped, symmetrical sublingual swelling.

The most recent classification in the literature was outlined by Gordon *et al.*, in 2013.<sup>10</sup> These authors modified Meyer's classification of the cysts,<sup>8</sup> using new terminology, referring to the cysts as congenital germline fusion cysts. Congenital germline fusion cysts have traditionally been referred to as dysontogenic cysts. The following classification is specific to those cysts within the floor of the mouth: (1) epidermoid – stratified squamous epithelium lining with no dermal appendages; (2) dermoid – stratified squamous epithelium lining with dermal appendages, present within associated underlying connective tissue, including sebaceous glands, hair follicles and sweat glands; and (3) teratoid – stratified squamous epithelium lining with elements of all three germ layers, ectoderm, mesoderm and endoderm, in the underlying tissue.

The cysts can be further classified according to their site of development during embryogenesis:<sup>11</sup> group I develop along the nasolacrimal groove, and give rise to periorbital cysts; group II develop within the prenasal space, giving rise to cysts over the dorsum of the nose; group III develop along the midline of the first and second pharyngeal arches, giving rise to cysts within the sublingual, submental and sub-mandibular regions; and group IV develop in the ventral dorsal midline, typically in the thyroidal, suprasternal or sub-occipital regions.

Three modes of pathogenesis have been described: acquired implantation, congenital teratoma and congenital inclusion.<sup>6,11</sup> However, as highlighted by Gordon *et al.*, a teratoma is a solid neoplasm and should be considered separate to a teratoid variant germline fusion cyst.<sup>10</sup> In view of this, there are only two true modes of pathogenesis: acquired implantation and congenital inclusion.<sup>9</sup>

Acquired implantation is the method by which a small sample of epithelium is separated and implanted within the underlying tissues, typically following surgery or trauma. This was simulated in a study by Baker and Mitchell by surgically implanting an autogenous sample of epithelium into the deeper connective tissue on the backs of rats.<sup>12</sup>



FIG.2

(a) Sagittal and (b) coronal magnetic resonance imaging scans of the 6 cm well-circumscribed cystic mass, which extends from the sublingual area at epiglottis level.

Congenital inclusion cysts are formed during embryogenesis, where disruption occurs during the fusion of the embryological components, causing in-folding of the epithelium.<sup>13</sup> The mechanism underlying group II (nasal) cysts is described in particular detail within the literature, with interest from the neuroectodermal involvement during development which results in the intracranial extension of some cysts. There have been many proposed theories; however, the most widely accepted theory is that initiated by Grunwald in 1910, and further interpreted by Pratt and Bradley.<sup>14-16</sup>

A number of symptoms can be experienced by patients, including dysphagia, dyspnoea, and difficulty masticating



FIG.3

(a) Intra oral midline incision from the tongue base to the mouth floor ,providing adequate access to the lesion. (b) Peri-operative view of the cystic mass.(c) View following removal; the specimen measure dapproximately 3×3cm.

And speaking.<sup>17,18</sup>Cysts occurring above the mylohyoid muscle tend to displace the tongue superiorly,and those that occur below may cause a double chin appearance extra-orally.<sup>19</sup>There have been reports in the literature of dermoidcysts dramatically increasing in size during pregnancy. It has been postulated that this may be caused by an increasein sebum production from the sebaceous glands present inthe cyst lining.<sup>20</sup>Mesoelleaet al. proposed an alternative theory, suggesting that the rapid growth is associated with increased plasma levels of oestrogens and progesterone,which act as growth factors on the cyst.<sup>19</sup>Very large dermoid cysts have been reported in the literature,but these are rare. When they do occur, there may be a risk tothe airway, and tracheotomy may even be required as part of management.<sup>20</sup>

Radiography is an important adjunct in the diagnosis and management of dermoid cysts. Ultrasound, computed tomography (CT) and MRI have all been reported in the litera-ture. Ultrasound may be helpful in differentiating between solid, vascular and cystic lesions.<sup>21</sup>It is generally accepted that CT and MRI imaging allow more precise localisation

in comparison to ultrasound.A high signal on T2-and a low signal on T1-weighted imaging typically indicate the presence of cystic structures.<sup>22</sup>

Aspiration cytology may be useful in differentiating between types of cysts.<sup>18</sup>However, the high-density fluid contents of dermoid cysts do not easily aspirate, limiting the usefulness of this technique.<sup>19</sup>Histological examination is essential to confirm the diagnosis.

Although there are no set rules, the most appropriate time to operate is when symptoms such as dysphagia, dysphonia and dyspnoea are present. Treatment for sublingual dermoid cysts is almost always surgical. The aspiration of cystic contents was previously attempted by Mesoelleaetal. In order to debulk a largest sublingual dermoid cyst;

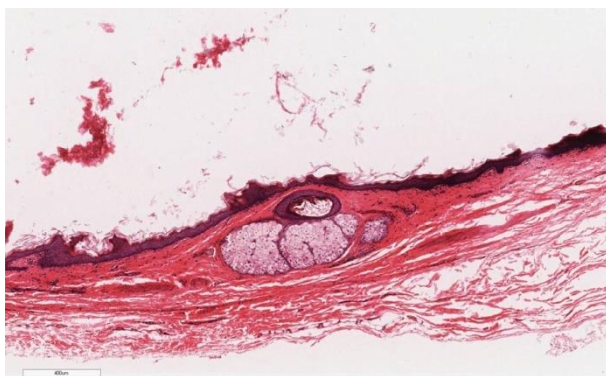


FIG.4

Histological image of the cyst,showing keratinizing squamous epithelium with a distinct granular layer,with a sebaceous gland in the cyst wall.(H&E;40×).

TABLE I  
DIFFERENTIAL DIAGNOSES OF MOUTH OR NECK SWELLINGS<sup>7</sup>

Category	Lesion
Developmental	Dermoid cyst
	Thyroglossal duct cyst
	Branchial cleft cyst
	Ectopic thyroid
Infection	Sublingual abscess
	Sublingual cellulitis
	Ludwig's angina
Salivary gland	Sublingual sialadenitis
	Mucocele
	Ranula
	Unilateral or bilateral Wharton's duct blockage
	Salivary gland tumour (benign or malignant)
Hamartomatous	Cystic hygroma
	Neoplasia
	Squamous cell carcinoma
	Other benign & malignant (lipoma, neurofibroma, haemangioma, lymphangioma)
Other	Epidermoid cyst
	Orally lymphoepithelial cyst
	Heterotopic gastro intestinal cyst
	Enteric duplication cyst

However, this proved unsuccessful because of the high-density fluid content.<sup>19</sup>

There is debate in the literature as to the suitability of intraoral and extraoral surgical approaches when removing dermoid cysts. Generally, the size and anatomical location of the cysts are the two most significant considerations.<sup>23</sup> It has been stated that an intraoral approach is more suitable for sublingual cysts of up to 6 cm in size, whereas an extra-oral approach may be advantageous for those cysts greater than 6 cm.<sup>18</sup> There are, however, case reports that detail the removal of very large dermoid cysts carried out by intra oral approach, which claim a cosmetic advantage. Boko *et al.* reported on the intraoral surgical excision of a dermoid cyst measuring 13 cm in length.<sup>20</sup> McGregor proposed a symphyseal mandibular osteotomy approach for larger lesions, citing restricted surgical access as a limiting factor for conventional intraoral and extraoral approaches.<sup>24</sup> With regards to anatomical location, Longo *et al.* reported that extraoral incision is mandatory if the cyst is located beneath the mylohyoid muscle.<sup>25</sup>

Prognosis following surgical removal is excellent, with a low incidence of relapse. Malignant changes have been recorded in dermoid cysts, by New and Erich, but not in the mouth floor.<sup>6</sup> However, a 5 per cent rate of malignant transformation of the teratoid type or oral dermoid cysts has been reported by other authors.<sup>26</sup>

- Dermoid cysts in the mouth floor are relatively uncommon developmental lesions
- They tend to be slow growing
- Differential diagnosis is of paramount importance, as recommended surgical techniques vary depending on lesion size and anatomical position
- Prompt diagnosis and treatment can prevent potential airway compromise

Differential diagnosis of cystic lesions in the mouth floor is of paramount importance, as the recommended surgical techniques vary depending on the size and anatomical position of the lesions. The intraoral approach is the preferred treatment modality for those lesions that do not extend below the mylohyoid muscle boundaries; this leads to a satisfactory cosmetic and functional outcome. Extraoral incision is mandatory only if the cyst lies below the mylohyoid muscle.

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