

## Case Report

# Giant epidermal inclusion cyst: an unusual case presentation

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### ABSTRACT

Epidermoids or epidermal inclusion cysts are developmental malformations rarely seen in the oral cavity. They are a histological variant of lesions broadly termed as dermoid cysts. The case discussed here stands out not only due to its unique clinical presentation which can put a clinician in a diagnostic dilemma but also because of its anatomical variation upon surgical exploration which can add to the conundrum.

**Keywords:** Epidermoid, Dermoid, Cyst

### INTRODUCTION

Epidermoid cysts are slow growing, rare growths and are generally due to location of ectodermal tissue at an unusual site. They are also known as follicular infundibular cysts, epidermal inclusion cysts or epidermal cysts and are benign developmental anomalies. Epidermoid cyst is often used interchangeably with the term dermoid cyst, however they have histological variation as discussed further. Mostly such cysts are seen in the testes and ovaries, while 7% of dermoid cysts are seen in the head and neck area.<sup>1</sup> Epidermoid cysts however represent only less than 0.01% of all intraoral cysts and is even less frequently seen than dermoid cysts of the head and neck.<sup>2,3</sup> Hence it is quite an unusual differential in cases of cystic lesions of the oral cavity.

Dermoid cysts may be congenital or acquired although the presentation of both is similar.<sup>4</sup> They may also be classified clinically depending on their location i.e superior or inferior with respect to mylohyoid muscle and central i.e median or lateral cysts in relation to midline of the neck.<sup>5</sup> A supramylohyoid cyst presents as a sublingual swelling while an inframylohyoid cyst manifests as submental swelling.<sup>5,6</sup> Dermoid cyst encompasses a broader term to include epidermoid cysts, dermoid cysts

and teratomas into which they are classified histologically.<sup>4</sup>

In this case report we describe the case of an 11 year old female child who presented with a sublingual mass alongwith description of course of management adopted for the patient. The case reported here is fascinating because of its unusual and complex anatomy and presentation which not only caused a diagnostic dilemma and but also was unique on surgical exploration.

### CASE REPORT

An 11 year old female patient presented to the outpatient unit of Otorhinolaryngology department at a tertiary care rural Medical College with a painless hard gradually progressive swelling in floor of mouth and extending gradually to involve submental region since last two years as shown in Figure 1. It was associated with discomfort during swallowing and difficulty in speech i.e. dysarthria which was more marked in the last six months.

On extraoral examination, there was a swelling measuring about 4.5×5.5 cms in size in median submental position. Overlying skin was intact, non-pulsatile and unpigmented. On palpation, the mass was cystic, firm,

fluctuating and nontender. Transillumination test was negative.

Intraoral examination indicated a cystic sessile dome shaped swelling occupying almost entire floor of mouth and partially lifting tongue. Overlying mucosa was intact but congested as seen in Figure 1. The swelling was bimanually palpable and transmission of movements between intraoral and submental swelling was felt. The swelling also moved with deglutition. There was no palpable lymphadenopathy on presentation.



**Figure 1: Sublingual swelling occupying whole of floor of mouth.**



**Figure 2: Typical double chin appearance of submental swelling.**

A differential of plunging ranula, dermoid cyst, lipoma, thyroglossal duct cyst, cystic hygroma, odontogenic cyst, tumour of floor of mouth or salivary gland tumour was considered.

Ultrasonography of the neck and local area was suggestive of a circumscribed and encapsulated swelling with distinct boundaries but was inconclusive. Routine haematological investigations were normal. Fine needle aspiration cytology yielded pultaceous material indicative of features consistent with dermoid cyst.

Patient was hospitalized and taken up for surgical excision under GA. An intraoral approach was initially

used taking care of Wharton's duct as presented in Figure 3. However on further exploration, the cyst was found to be of a gigantic size with a lobed and dumbbell shaped structure. It was passing through the mylohyoid muscle and adherent to adjacent structures, hence a transverse extraoral incision was also given, mylohyoid muscle was separated and the complete intact trilobed cyst was excised after careful dissection and separating it from muscles of floor of mouth and from the hyoid bone to which it was attached. The wound was stitched in three layers and the mass was sent for histopathological examination. Grossly the dumbbell appearing cyst measured about 7.5×6.5 cms in size with a trilobite structure and a thin wall as in Figure 4. Postoperative period was uneventful and the child was discharged after 5 days of indoor care. Follow-up at 8 months revealed no recurrence and the patient is healthy till date.



**Figure 3: Intraoperative picture showing mass via intraoral approach.**



**Figure 4: Postoperative specimen showing trilobed appearance.**

On histopathological examination, there was cheesy material within the sac and microscopic examination confirmed the diagnosis of an epidermal inclusion cyst with flattened stratified squamous epithelium and

acidophilic stratum corneum. No hair follicles sweat or sebaceous glands were identified within the epithelial lining.

## DISCUSSION

Dermoid cysts are benign squamous epithelial lined masses usually filled with pultaceous cheesy material. The commonest site of dermoid cysts are in the testes and ovaries. In the head and neck region, epidermoid cysts are unusual and represent less than 0.01% of all oral cavity cysts. In a study at Mayo clinic, New and Erich found that amongst 1459 cases only 1.6% cases were seen in the oral cavity.<sup>7, 8</sup> This is due to presence of wide number of other masses, particularly odontogenic and non-odontogenic cysts in the oral cavity. Dermoid cyst may be defined differently by dermatologists, otolaryngologist, gynaecologist, surgeon, neurosurgeon or pathologist because though they exist in each of these specialists's domain, they present with a wide & variable spectrum of symptoms. Hence to clarify, on the basis of histology, Meyer's classification of dermoid cysts is almost universally used to differentiate dermoid cysts into 3 variants i.e epidermoid, dermoid and teratoid cysts. Epidermoid cyst is the basic type which is lined by squamous epithelium, dermoid cysts also have adnexal tissue in the wall alongwith squamous epithelium while teratoid variety may have tissues arising from trigermental layers viz ectoderm, mesoderm and endoderm.<sup>9</sup> Epidermoid cysts are less commonly seen than dermoid cysts in the head and neck region. The term sebaceous cyst often used interchangeably with dermoid cyst is a misnomer because study of the content of these cysts with respect to lipid pattern and cytokeratins suggest their proximity to epidermis rather than to sebaceous origin.<sup>10</sup>

The exact aetiology of origin of epidermoid cysts is not known and various theories have been postulated. Most widely accepted amongst these suggests that such cysts are of congenital origin where malformation at third to fifth week of gestation during the stage of embryonic fusion results in sequestration of ectoderm into deeper tissues. The occurrence of epidermoid cysts along the lines of fusion supports this hypothesis. Others consider them to be of acquired aetiology like accidental or surgical trauma causing implantation of epidermis into deeper tissues.<sup>4,10,11</sup> Certain other theories like occlusion of the pilosebaceous or eccrine duct, or origin from remnant of thyroglossal duct and HPV infections have also been suggested as contributory factors.<sup>10</sup>

Most authors suggest no gender predilection. These oral cavity cysts are generally slow growing and indolent with late presentation. Dermoids are generally found in patients between the ages 15 to 35 years.<sup>4,11</sup> In certain congenital syndromes like Gardner's syndrome, presentation is seen early.<sup>10</sup>

Clinical presentation depends on site, size and duration of the swelling. Due to insidious progression presentation is

generally delayed even in cysts of congenital origin until they develop to a size causing significant complaints like speech alteration, dysphagia and cosmetic deformity.

It may present as intraoral or cervical swelling on basis of its superior or inferior relation to the mylohyoid muscle. However in unique cases as the one discussed, due to giant multilobed size and anatomical distinctness presentation is varied and cause diagnostic confusion. Other presenting features are dysphagia, dysarthria, dyspnoea due to displacement of the tongue. A typical double chin appearance or cosmetic deformity may be seen in inframylohyoid anomalies. A long standing dermoid cyst may lead to secondary infections and symptoms like pain, recurrent swelling with foul smelling discharge and excoriation.<sup>2,4,10</sup> Very rarely cases of malignant transformation especially from the teratoid variant have been reported. Chronicity of such cysts alongwith prolonged irritation have been said to be responsible for such dysplasia.<sup>12</sup>

Cervical swellings especially of the sublingual and submandibular regions are seen in a variety of lesions and may be included in differential diagnosis like congenital lesions, developmental lesions, inflammatory lesions, benign or malignant tumour of floor of mouth, jaw or salivary gland tumour.<sup>4,11,13</sup> In this patient, infectious lesions like abscesses of the sublingual and submandibular spaces and cellulitis were ruled out due to insidious onset and absence of signs of inflammation like fever, pain, erythema. A malignant lesion was also not likely due to the age of the patient and absence of lymphadenopathy after a significant history. The diagnosis of a congenital developmental or anatomic anomaly was more likely on basis of history and presentation of the patient. The likelihood of ranula, mucous extravasation phenomenon primary or secondary to lithiasis, dermoid cyst, thyroglossal duct cyst, cystic hygroma, branchial cleft cyst were also considered. In our patient, fine needle aspiration yielded cheesy material which was indicative of a dermoid cyst. CT scan was not done in this patient as she came from a poor socioeconomic strata and ultrasound provided us a rough estimation of the extent of the disease.

The treatment of choice for dermoid cyst is complete excision.<sup>4,5,10,13</sup> Generally patients with dermoid cysts present late when the swelling has reached a considerable size compromising oral functions and causing dysarthria, dysphagia or cosmetic deformity. Approaches used may be introral, extraoral or combined approach depending on location, size and attachments of cyst. An intraoral approach is generally used when the cyst is lying superior to mylohyoid muscle without any attachments. Extraoral approach is generally avoided unless the cyst is of gigantic size with attachments to surrounding structures. In our case we first attempted an intraoral approach, but upon separating mylohyoid it was found to be adherent to underlying structures. An extraoral incision was then given and upon exploration it was found to be adherent to

hyoid and its musculature from which it was separated. Hence a combined approach was used to remove the cyst in totality. Care was taken in order to avoid spillage as it may lead to irritation, secondary infection and adhesions impeding function of surrounding structures. Histopathological examination is the only manner to ensure the diagnosis and confirm the histology as even aspiration techniques cannot predict the subtype of a dermoid cyst. Acidophilic stratum corneum and basophilic dot like stippling of stratum granulosum on microscopic examination are the hallmark signs of an epidermoid cyst.

Prognosis is very good and recurrence of an epidermoid cyst is seldom seen if the cyst is removed completely with its wall and attachments.<sup>4, 13</sup> However incomplete removal might lead to residual deformity and an infected suture line hence stress should be emphasized not only upon complete removal of cyst but also upon complete removal of all attachments. Malignant transformation especially in the head and neck region has rarely been reported.<sup>12</sup>

To summarize, we presented a case report which presented with a mobile submental as well as intraoral swelling which is a presentation of various differential entities as discussed. Unconventional clinical features as in the current case report add to the clinician's dilemma. Epidermoid cyst is a rare diagnosis in such cases and having a detailed knowledge of its variable presentations is mandatory. An elaborate history, examination and investigations are helpful in confirming the diagnosis. Careful surgical exploration, total excision and histopathological confirmation are essential to avoid complications.

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