Journal of Craniomaxillofacial Research

Vol. 4, No. 1 Winter 2017

Sublingual epidermoid cyst: A case report

Jyotsna Galinde [1], Suraj Arjun Ahuja [2]*

1. Professor, Department of oral and Maxillofacial Surgery, MGM Dental College & Hospital, Mumbai, India.

2. Resident, Department of oral and Maxillofacial Surgery, MGM Dental College & Hospital, Mumbai, India.

ARTICLE INFO

Article Type:
Case Report

Received: 10 Oct 2016
Revised: 21 Nov 2016
Accepted: 29 Des 2016

**Corresponding author:*
Suraj Arjun Ahuja
Department of oral and Maxillofacial Surgery MGM Dental College & Hospital Mumbai, Maharashtra 410209, India.

Tel: +9819985991
Fax: +9819985991
Email: drahujasuraj@gmail.com

ABSTRACT

The occurrence of epidermoid and dermoid cysts in the oral cavity is extremely rare accounting 0.01% of all cysts. In our case report we hereby present a large epidermoid cyst in the floor of mouth imitating a plunging ranula. A 26-year-old female reported to us with a complaint of a swelling below the tongue since 2 year. On examination there was a swelling in the left side of the floor of the mouth, with anextra oral component in the submental area, the patient underwent surgical removal of the mass under local anaesthesia. Diagnosis of an epidermoid cyst was confirmed by the histopathological report.

Introduction

Dermoid cysts are benign lesions, primarily seen in the testes and ovaries, with 7% occurring in the head and neck area and 1.6% within the oral cavity [1]. They represent less than 0.01% of all oral cavity cysts [2]. Dermoid cysts usually occur in young adults in their 2nd and 3rd decades; however they may also occur in infants. There is no gender discrimination [3]. Histologically, there are three types of dermoid cysts; epidermoid, dermoid, and teratoidt the cysts can be defined as epidermoid when the lining presents only epithelium, dermoid cysts when skin adnexa are found, and teratoid cysts when other tissue such as muscle, cartilage, and bone are present [4]. It is suggested that these cysts are derived from epithelial remains from the closure process of the first and second branchial arches [5]. Particularly in the tongue region, these lesions may be formed by remains of the tuberculum impar, which, together with the lateral lingual prominences, form the body of the tongue and floor of the mouth [6].

Usually, the term of dermoid cyst is used for all of these types in clinical practice. Sublingual dermoid cyst cases usually present themselves as a painless swelling in the floor of mouth. In addition, these patients might also experience dysphonia, disarticulation, dysphagia, and dyspnoea symptoms [7]. Treatment of dermoid cysts is by surgical excision; by extraoral, intraoral or combined approaches. Some authors believe that the treatment consists of excision by intraoral approach in sublingual cysts and extraoral approach in submental cysts [8,9]. Recurrence is rare when the cyst is completely removed [10].

Case Report

A 26 year old female reported to our department, with the history of a persistent swelling below the tongue since 2 year and is now visible near the left side of neck since 4-5 months. It was a soft, painless swelling, which was initially small and increased in size over duration of 2 years. She had no difficulty in moving the tongue and there was no history of dysphagea or dyspnoea. On intraoral examination, there was a swelling in the left side of the floor of the mouth, pushing the tongue to the right side. Upon bimanual palpation it was soft in consistency, non-tender with indistinct edges. The mucosa over the swelling were intact and normal (Figure 1). On extra oral examination a diffuse extra oral swelling was present over left sub mental & sub mandibular area around 4x3 cm, ovoid in shape, over lying skin appears normal with no secondary changes , no movement with deglutition or with protrusion of tongue was noticed (Figure 2). On palpitation swelling was non tender had a dough like consistency with ill-defined borders, swelling was non reducible nor compressible. Transillumination was negative and there was no evidence of cervical lymphadenopathy given the clinical appearance of the swelling, provisional diagnosis of a plunging ranula or benign/malignant tumours of mucosa or salivary glands was made. A orthopantomography x-ray was taken to rule out and odontogenic infection & salivary gland pathology such as salivary calculus or stone. To confirm the diagnosis a CT scan was advised but could not be undertaken because of patient's financial limitations, a USG guided FNAC was done which revealed the presence of epithelial remnants, desquamated tissue and cellular debris suggestive of dermoid cyst.

Following discussion with the patient, the decision to operate under LA was made. A mucosal incision was made over the swelling. Blunt dissection through the pericapsular tissue was performed, taking care in the regions of the submandibular ducts and lingual nerves bilaterally. Dissection was done to expose the cyst which was lying superficial to the mylohyoid muscle (Figure 3). Cyst was removed per oral in toto (Figure 4A & 4B). After achieving haemostasis, the wound was sutured in layers and a corrugated rubber drain was placed in position. The specimen was sent for histopathological examination. The maximum thickness of the wall of the cyst was 2mm. The cyst contained a yellowish greyish, gritty, paste-like material (Figure 5). Histology revealed an epidermoid cyst with orthokeratinized stratified squamous epithelial cystic lining with prominent stratum granulosum (Figure 6).

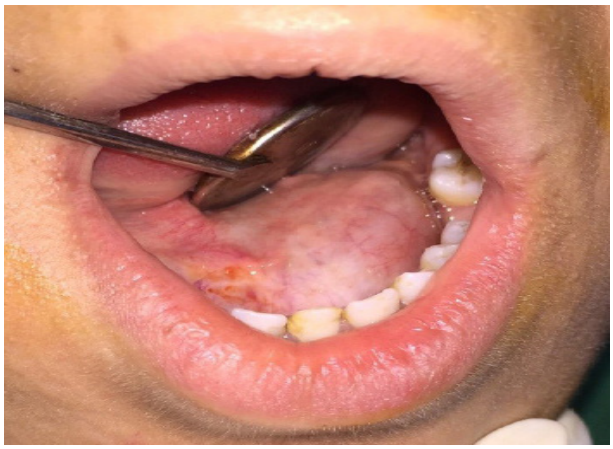

Figure 1: Swelling in the left side of the floor of the mouth, pushing the tongue to the right side. The mucosa over the swelling were intact and normal.

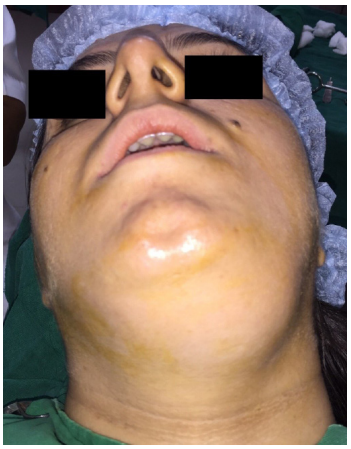

Figure 2: A diffuse, ovoid, non tender, dough like extra oral swelling present over left sub mental & sub mandibular area around 4x3 cm.

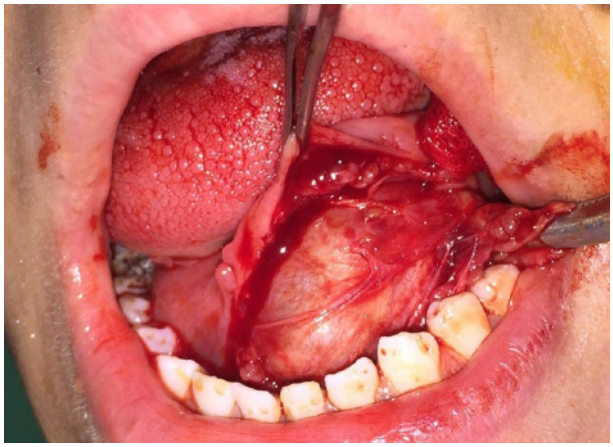

Figure 3: Cyst exposed intra orally which was lying superficial to the mylohyoid muscle.

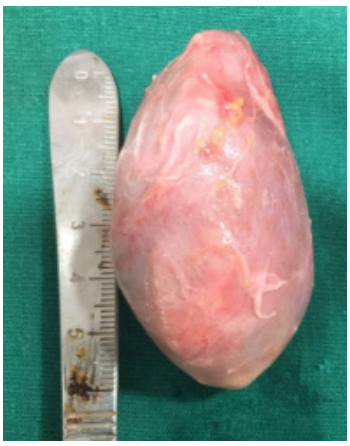

Figure : 4A.

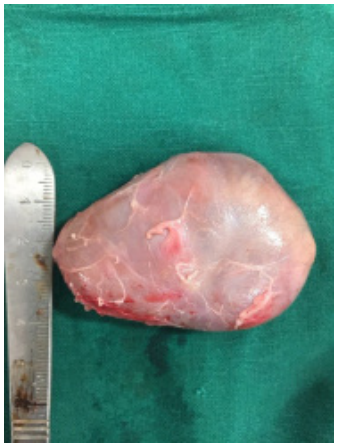

Figure : 4B.

Figure 4A **&** *4B:* Cyst was removed per oral in toto.

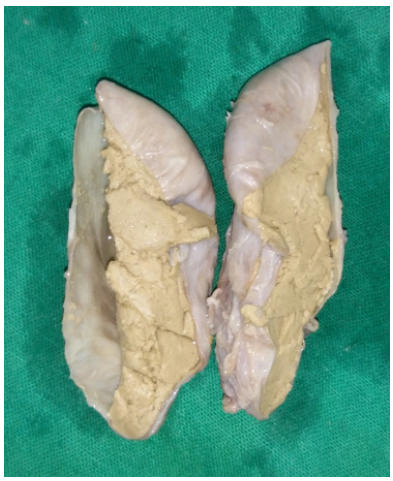

Figure 5: Cyst containing a yellowish, greyish, gritty, paste-like material.

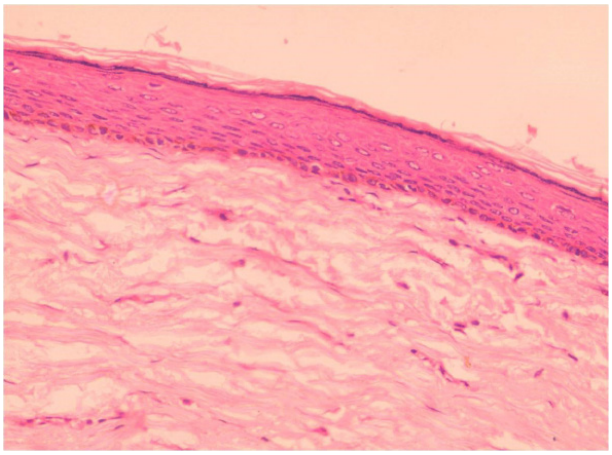

Figure 6: H & E stained slide shows orthokeratinized stratified squamous epithelial cystic lining with prominent stratum granulosum. The connective tissue capsule shows collagen fibres with absence of skin appendages and minimal chronic inflammatory cell infiltrate.

Discussion

The term 'dermoid cyst' causes some confusion, as it has historically been used differently by different specialties. Some authors use the term dermoid in lieu of teratoma. Others define a dermoid as a subcutaneous, congenital cyst. Meyer (1955) classified congenital cysts of the floor of the mouth as epidermoid, dermoid, or teratoid, according to their histological features. All three types of cyst contain a greasy, cheese like, white/grey/tan material. Dermoid and teratoid cysts may also contain fragments of hair, nails, or dental enamel in their lumen.

Congenital floor of mouth cysts are thought to arise from entrapment of ectodermal tissue in the midline during fusion of the first and second brachial arches, during the third and fourth embryonic weeks [11]. Acquired forms are likely to be a result of either iatrogenic or traumatic implantation of epidermal fragments into the underlying tissues. Other hypothesis include the possibility that floor of mouth cysts may represent a variation of the thyroglossal duct cyst [12].

The differential diagnosis of sublingual lesions includes ranula, lymphatic malformation, dermoid cyst, epidermoid cyst, and heterotopic gastrointestinal cyst (Table 1) [13]. For this reason, bimanual palpation and conventional radiography are not always sufficient in making differential diagnoses. In these cases, it is necessary to use ultrasonography, CT scan, or MR imaging together with cytology examination by fine-needle aspiration biopsy [14]. Floor of mouth cysts generally

present as slow growing, painless, doughy/fluctuant swellings. They are asymptomatic until large enough to cause dysphonia, dysphagia or dyspnoea. The tongue will be displaced poster-superiorly, and the patient may present with a double chin, particularly if the mass sits superficial to the mylohyoid. Treatment is by surgical enucleation. Surgical approach is determined by the cyst's relationship to the muscles of the floor of the mouth, particularly the myohyoid. The mylohyoid muscle separates the sublingual space from the submental and submandibular spaces. If the mass is in the sublingual spaces superficial to mylohyoid, an intraoral incision is preferred. Care must be taken to preserve the lingual nerve and the submandibular ducts. If the mass lies deep to mylohyoid, a submental/submandibular extraoral incision is required. The incision is made in a natural skin crease, but some degree of scarring is inevitable. There is a risk of damage to the marginal mandibular branch of the facial nerve. Care is taken to avoid rupture of the cyst, as the contents may be irritant, causing postoperative inflammation. Once removed, recurrence is unlikely.

Mainak Dutta et.al. 15 in 2013 analysed 28 cases of Head and neck epidermoid cyst occurring at various sites such as the submandibular region [5], pinna [5], sublingual region [1], periorbital [6], suprasternal [6], along the anterior border of sternocleidomastoid [1]and glabella [3] were involved, along with an iatrogenic implantation epidermoid cyst in a tracheostomy scar. Excision was the preferred treatment in 20 cases. There are sporadic case reports of malignant transformation arising in the lining of dermoid, epidermoid and teratoid cysts. One case of a squamous cell carcinoma in the lining of an epidermoid cyst in the sublingual gland has been reported [16].

Conclusion

Dermoid cysts in the floor of the mouth are quite rare and need to be differentially diagnosed from several other diseases and conditions of the area. For their diagnosis, their clinical picture is essential involving a detailed examination of their size and anatomical location. Furthermore, invaluable assistance is provided by the ultrasonic scan, computed tomography of the area. Their treatment calls for careful planning and execution of the surgical operation an intra or an extra oral approach can be opted for. The ultimate confirmation and definite diagnosis of the disease is always effected via histopathological examination.

Differential Diagnosis Of Dermoid Cyst Of The Floor	
Mucus extravasation phenomena	Mucocele
	Plunging (cervical) ranula
Embryologic anomalies	Thyroglossal duct cyst
	Branchial cleft cyst
	Dermoid cyst
	Cystic hygroma
Infection	Acute bacterial infection/cellulitis of oral floor
	Sialadentitis of sublingual or submandibular gland
	Viral lymphadenitis (EBV, CMV)
Granulomatous disease	Infectious
	Mycobacterial disease
	Cat-scratch disease
	Actinomycosis
	Toxoplasmosis
	Tularaemia
	Histoplasmosis
	Blastomycosis
	Noninfectious
	Sarcoidosis
	Wegener's granulomatosis
	Langerhans' cell histiocytosis
	Crohn's disease
Non-granulomatous inflammatory disease	Kawasaki's disease
Tumour	Benign tumours of salivary glands
	Lipoma
	Fibroma
	Haemangioma
	Lymphangioma
	Angioma
	Neurofibroma
	Malignant tumours of salivary glands*
	Lymphoma
	Rhabdomyosarcoma
	Neuroblastoma
	Metastatic neoplasm
Other	Normal fat in the submental or submandibular area
	HIV-related lymphadenopathy
	Pneumocystis lymphadenitis
	Persistent generalized lymphadenopathy
	Nocardiosis
	Non-Hodgkin's lymphoma
	Metastatic Kaposi's sarcoma

Table 1. Differential diagnosis of dermoid cyst of the floor of.

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Please cite this paper as:
Galinde J, Arjun Ahuja S; Sublingual epidermoid cyst: A case report. J Craniomaxillofac Res 2017; 4(1): 313-318