

Epidermal Inclusion Cyst of Tongue-A Case Report

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ABSTRACT

The epidermal inclusion cyst constitutes less than 0.01% of all the oral cysts in head and neck region. They are congenital lesions caused due to defective fusion of the embryonic lateral mesenchymal tissues. It has a variable tissue origin and can originate from ectoblastic, mesoblastic or endoblastic tissues. The incidence rate of epidermal cyst involving head and neck region is 1.6-7.0%. Clinically the epidermal inclusion cyst is characterized by dysphagia, dyspnoea and dysphonia. Here we are presenting a case of epidermal inclusion cyst in a 45 year old female involving left lateral border of tongue.

Keywords: Congenital abnormalities, Epidermoid cysts, Tongue, Dermoid cyst

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INTRODUCTION

Epidermal inclusion cyst also known as dermoid cysts are congenital lesions caused due to defective fusion of the embryonic lateral mesenchymal tissues. It has a variable tissue origin. They can originate from ectoblastic, mesoblastic or endoblastic tissues. A true dermoid cyst cavity is covered with keratinized epithelium with dermal appendices¹. The epidermal inclusion cyst rarely occur in the head and neck with an incidence ranging from 1.6 to 6.9%. They constitute less than 0.01% of all oral cysts^{2,3,4}. Roser was the first to designate dermoid cysts in the floor of the mouth as epidermoid tumours⁵. Intra-orally epidermal inclusion cyst most commonly involve the sublingual area however it can be found on lip, tongue and bone⁶. The isolated epidermoid cyst of the tongue without sublingual involvement is very rare. Till date only 14 cases have been reported in literature involving tongue however only in 4 cases lateral border of tongue is affected⁷. The term "dermoid cyst" characterizes a distinct entity. The word "dermoid" has been used to designate true dermoid cysts, epidermoid cysts, epidermal inclusion cyst and teratoid cysts^{8,9,10}. Following theories of etiopathogenesis of epidermal inclusion cyst have been proposed¹¹

1. Congenital inclusion of dermal and epidermal elements of germ layers in deeper tissues along the embryonic lines of fusion.

2. Acquired traumatic implantation of dermal and epidermal elements of surface epithelium which may proliferate and keratinize.
3. Growth from rest of totipotent cells displaced from the blastomere.

These cysts show variation in size and weight. The symptoms of dysphagia, dyspnoea and dysphonia may occur due to upward displacement of tongue by these sublingual swellings. Furthermore growth of dermoid cyst in an inferior direction may give rise to appearance of characteristic "double chin". These well encapsulated lesions typically feel "dough like" on palpation and consistency may range from a cheesy, sebaceous to liquefied substance^{5,10}. Fine needle aspiration cytology, ultrasound, CT and MR imaging provide essential information on the cyst location. Ultrasonographic findings comprise solid and cystic structures within a heterogeneous mass. On CT scans, epidermal inclusion cyst appear as moderately thin walled, unilocular masses filled with a homogeneous, hypoattenuating fluid substance with numerous hypoattenuating fat nodules giving the pathognomonic "sack-of-marbles" appearance. On MR imaging dermoid cysts give variable signal intensity on T1-weighted images and are usually hyperintense on T2-weighted images. Fine needle aspiration cytology has been advocated as an essential investigation. Treatment comprises total surgical excision. Recurrences are unusual after absolute surgical excision. Reports of malignant transformation of sublingual dermoid and epidermoid to squamous carcinoma and basal cell carcinoma are present. A 5% rate of malignant transformation of the teratoid variety of oral dermoid cysts has also been quoted in literature^{4,5,9}.

CASE REPORT

A 45 year old (Fig.1) female patient reported to us with presenting complaint of a swelling involving left side of tongue since 2 years (Fig.2). The swelling was

progressively increasing in size and causing difficulty in eating and chewing of food. There was no significant past medical and dental history. The patient had no drug allergy and no history of any habit. She was well nourished and well built and had normal intelligence. She was well oriented to surroundings and had normal gait and posture. Her vitals were normal. She had no history of any systemic disease. No abnormal findings were detected on extra-oral examination. On intra-oral examination a diffuse swelling of 3x3 cm noted involving left lateral border and dorsal surface of tongue (**Fig.2 & Fig.3**). The overlying mucosa over the swelling was normal having well studded with tongue papillae. The swelling was slightly elevated from the surface of tongue (**Fig.3**). All other structures were normal. On palpation a non-tender firm to hard diffuse swelling of 3x3 cm involving left lateral border and dorsal surface of tongue in anterior 1/3rd third region has been noted. The swelling was non-fluctuant, non-compressible and non-reducible. There was no associated pain and bleeding. The swelling was interfering with tongue movements causing difficulty in chewing food. There was no associated lymphadenopathy. The patient is advised for high resolution USG of tongue and MRI with contrast. The high resolution USG of tongue (**Fig.4**) shows a well-defined heterogeneous predominantly hyperechoic lesion noted in left lateral border of tongue measuring approx. 2.29x1.59 cm. The MRI shows a well defined lesion (1.5x1.0x1.2cm) with mild peripheral enhancement and display signal intensity alteration seen in anterior 2/3rd of tongue on left side (**Fig.5,6,7,8,9,10**). Few round oval lesions display signal intensity alteration suggestive of lymph nodes in level 1a and bilateral level 1b. On the basis of clinicoradiological features a provisional diagnosis of retention cyst have made. Fine Needle Aspiration Cytology was done and revealed the presence of yellowish thick material which was sent for cytological examination. Cytological examination revealed presence of desquamated epithelial cells and lots of keratin flecks (**Fig.11**). The patient is further advised for excisional biopsy of the lesion. The routine blood investigation is carried out which was found to be normal. The excisional biopsy is done under local anesthesia. The histology (**Fig.12**), shows connective tissue surrounded by stratified keratinizing epithelium with cystic degeneration and keratin fluid without any dermal component. On the basis of histopathology a final diagnosis of epidermoid inclusion cyst is reached. The patient is followed for 6 months but no recurrence has been reported.



Fig.1: Profile photograph of patient showing no extra-oral abnormality

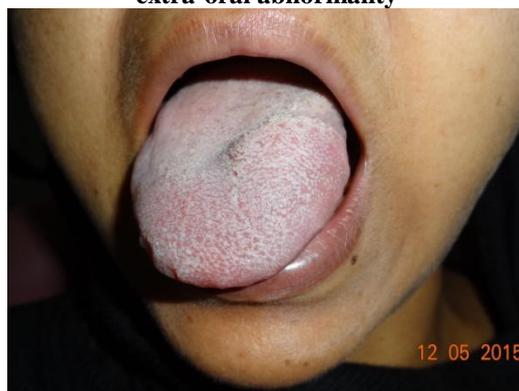


Fig.2: Intra-oral view of patient showing diffuse swelling of 3x3 cm noted involving left lateral border and dorsal surface of tongue



Fig.3: Intra-oral view showing overlying mucosa over the swelling was normal having well studded with tongue papillae. The swelling was slightly elevated from the surface of tongue

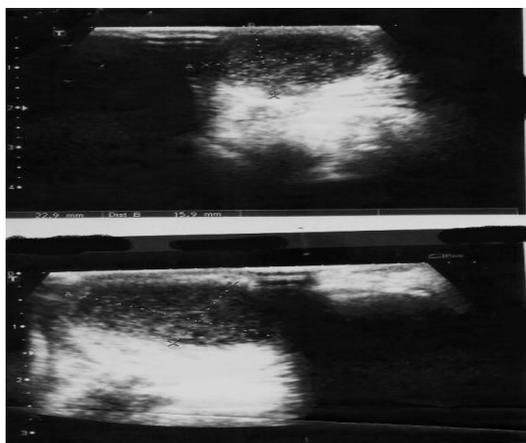


Fig.4: HR USG of tongue showing a well defined heterogeneous predominantly hyperechoic lesion noted in left lateral border of tongue measuring approx. 2.29x1.59 cm

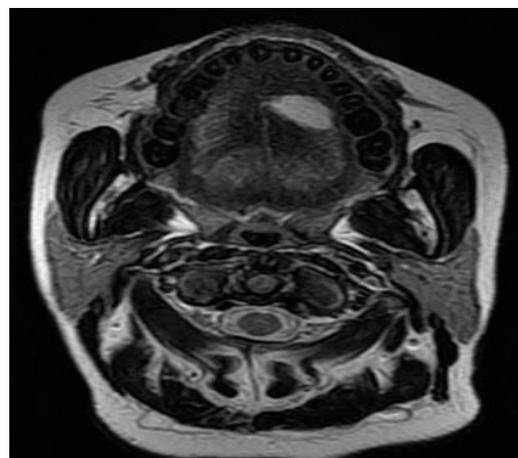


Fig.7: Axial T2 FSE image showing heterogeneous intra-lingual mass with high signal intensity



Fig.5: Axial T1 FSE showing heterogeneous intralingual mass with low signal intensity involving anterior 2/3 rd of tongue on left side

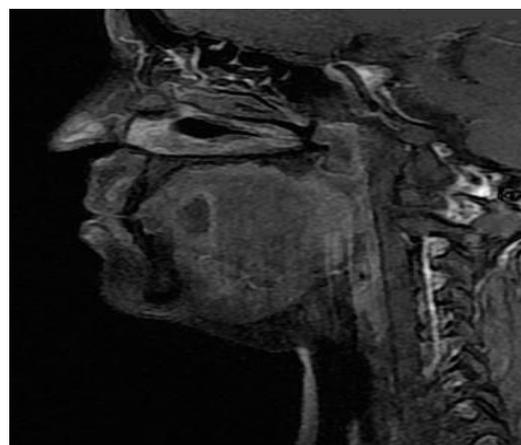


Fig.8: Saggital T1 with contrast image shows heterogeneous intralingual mass with low signal intensity involving anterior 2/3 rd of tongue on left side with mild peripheral enhancement



Fig.6: Axial T1 with contrast shows a well defined lesion (1.5x1.0x1.2cm) with mild peripheral enhancement with low signal intensity

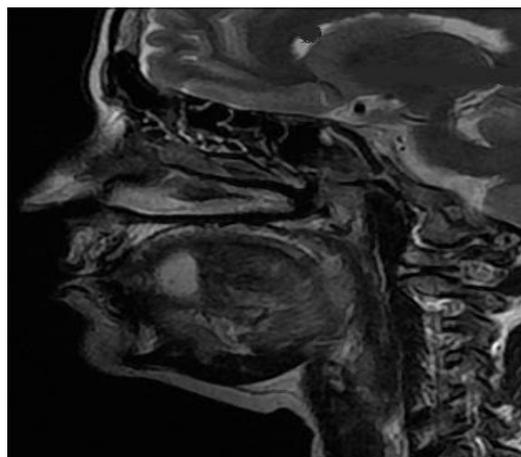


Fig.9: Saggital T2 FSE image showing heterogeneous intra-lingual mass with high signal intensity

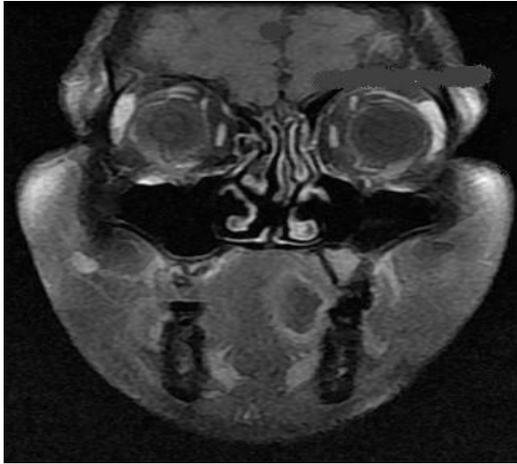


Fig.10: Coronal T1 weighted image intralingual mass with low signal intensity involving anterior 2/3 rd of tongue on left side of tongue



Fig.11: Fine Needle Aspiration Cytology (Pap stain) showing presence of desquamated epithelial cells and lots of keratin flecks

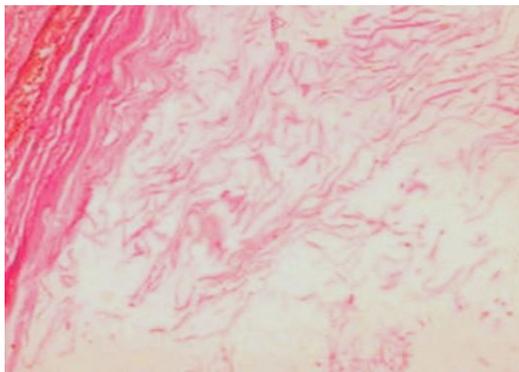


Fig.12: Photomicrograph showing cystic cavity lined by stratified squamous epithelium with presence of lots of keratin flecks supported by connective tissue stroma (H&E x100)

DISCUSSION

Epidermoid cyst is a congenital cyst which occurs due to entrapment of ectoderm at the time of fusion of neural tubes however they can be acquired type also which occurs due to secondary inclusion of epidermal elements into dermis post trauma or iatrogenetically¹². The epidermal cyst of tongue is formed by remains of the tuberculum impar¹³. The

incidence rate of epidermal cyst involving ovaries and testicles is reported to be 80%⁹ however in head and neck region it is 1.6-7.0%. The epidermal cyst constitutes less than 0.01% of all the oral cysts in head and neck region¹⁴. Epidermal cyst is classified histologically by Meyer in 1955 in three types as epidermoid, dermoid or teratoid¹². All three types of cyst contain a greasy, cheese like, white/gray/tan material¹³. Dermoid and teratoid cysts may also contain hair, nails or dental enamel in their lumen¹¹. The epidermal cyst is the slow growing in nature, varies in size from few mm to 10 cm having normal or yellow-reddish colour. In most cases epidermal cyst is painless swelling with soft consistency. The clinical diagnosis should always be supported with the histological examination¹⁴. The unoperated epidermoid cyst can achieve increase in size causing discomfort during mastication, swallowing, and speaking¹⁵. The differential diagnosis of epidermoid cyst includes ranula, mucocele, lymphangioma, lymphoepithelial cyst and thyroglossal duct cyst¹⁵. The choice of imaging in epidermal cyst of head and neck region is ultrasonography due to its reliability and economical feature however the computed tomography and magnetic resonance imaging allow precision localization of epidermoid cyst in relation to geniohyoid and mylohyoid muscles¹⁶. The epidermoid cyst of congenital and acquired type has no clinical or histological difference in spite of different pathogenesis mechanisms¹⁸. Histologically the epidermal cyst is characterized by stratified squamous epithelium with laminae of keratinization on the surface and lumen of the cyst cavity¹⁴. Treatment of choice is complete surgical resection or enucleation. The recurrence of epidermoid cyst is rare however the sporadic cases of malignant transformation have reported in epidermoid cyst. These malignant transformations arise in epithelium of dermoid, epidermoid and teratoid cysts¹⁸. Only one reported case of squamous cell carcinoma in the lining of an epidermoid cyst in the sublingual gland has been reported¹⁹.

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