Clinico-Radiological Analysis of Dermoid Cysts of Head and Neck Region-A Meta-Analysis

Akhilanand Chaurasia*, Vaibhav Mishra**

Abstract

Objective: The aim of present study is evaluation of site predilection and age related incidence of Dermoid cyst in head and neck region. *Materials and Methods*: The central search engines like Pubmed, Scopus, Medline, Science direct, Mendley and Google scholar is used to search all cases of dermoid cyst pulished till date in English language. Total 47 published studies have been retrieved from database meeting the search criteria. Out of 47 published studies, 20 studies have no radiological features. The remaining 27 cases have classical radiological presentations on Conventional radiographs (Occlusalradiographs, Panoramic radiographs), Ultrasound, CT and MRI. *Results*: Sublingual region was the most common site followed by tongue and floor of the mouth. Lip, soft palate and buccal mucosa were least frequently involved with only one case reported in each location. Dermoid cysts were clearly depicted on CT and MR imaging and on ultrasonography as well defined encapsulated cystic masses. Displacement of adjacent anatomical structures were evident in most of the radiological findings. *Conclusion*: Dermoid cyst of head and neck is uncommon lesion. Whenever, a well defined cystic lesion is appreciated in head and neck region, the Dermoid cyst should always be included in differential diagnosis.

Keywords: Dermoid Cyst; Epidermoid Cyst; Congenital Abnormalities; Hamartoma.

Introduction

Dermoid cysts are congenital lesions caused by a defect in the fusion of the embryonic lateral mesenchymatic mass and composed of tissues with different origins: ectoblastic, mesoblastic, or endoblastic. A true dermoid cyst cavity is covered with epithelium showing keratinisation and presenting identifiable dermal appendices [1]. These lesions rarely occur in the head and neck with an incidence ranging from 1.6 to 6.9% and represent less than 0.01% of all oral cavity cysts [2,3,4].

Roser was the first to designate dermoid cysts in the floor of the mouth as epidermoid tumours [5].

Author's Affiliation: *Assistant Professor **Resident, Department of Oral Medicine & Radiology, Faculty of Dental Sciences, King George's Medical Unniversity Lucknow.

Reprints Requests: Akhilanand Chaurasia, Assistant Professor, Department of Oral Medicine & Radiology, Faculty of Dental Sciences, King George's Medical Unniversity, Lucknow, Uttar Pradesh 226003

Email-chaurasiaakhilanand49@gmail.com

Received on 18.01.2017, Accepted on 27.01.2017

Even though the expression "dermoid cyst" characterizes a distinct entity, the word "dermoid" has been used to designate true dermoid cysts, epidermoid cysts, and teratoid cysts [6,7,8]. Based on the histopathological picture Meyer divided the floor of the mouth cysts into following types [4,6,9] Epidermoid cysts - where in the cystic cavity is lined with epithelium without skin appendages, Dermoid cysts here the epithelial lined cystic cavity encloses skin appendages such as hair, hair follicles, sebaceous, and sweat glands. Teratoid cysts - in this entity, the cystic cavity in addition to skin appendages also encloses mesodermal derivatives such as bone, muscle, gastrointestinal and respiratory tissue [7,8]. All these three cysts owing to their squamous epithelium lining may enclose cheesy keratinaceous material within their lumen. Hence the fundamental difference between the dermoid and the epidermoid is the presence of skin appendages within the wall of the former and the lack of the same in the latter. Epidermoid cysts may be categorized as congenital or acquired based on their origin although there is no disparity between the two either clinically or histologically [4,6,10]. Ambiguity about their exact pathogenesis exists and dysontogenetic, traumatic,

and thyroglossal anomaly theories have been postulated [4,6,8,9]. Most congenital dermoid and epidermoid cysts perhaps begin due to an embryologic accident during the early stages of development but hardly get perceived until their size causes annoyance [7]. The origin of epidermoid cysts is believed to be from entrapment of epithelial remnants during midline closure of the bilateral first and second branchial arches [5,10]. It has also been opined that ectodermal differentiation of multipotential cells, most probably pinched off at the point of anterior neuropore closure may give rise to these cysts. On the other hand, they may also crop up from the tuberculum impar of His [4,5]. Traumatic or iatrogenic inclusion of epithelial cells or the blockage of a sebaceous gland duct have been postulated as the pathogenesis of acquired cyst. However, some authors have also stated that midline cysts may represent a diverse form of thyroglossal duct cyst [8,10]. They may be found in any age group but show preponderance between 15-35 years of age with no gender predilection. Although floor of the mouth in the midline is most favored site, occasional occurrence involving the buccal mucosa, tongue, lips, uvula, temporomandibular joint dermal graft, intradiploic, intracranial, and intraosseous location within the mandible and maxilla also have been cited in literature [11,12]. These lesions show variation in size and weight from few millimeters to centimeters and a gram to several hundred gram respectively. Symptoms of dysphagia, dyspnoea and dysphonia may occur due to upward displacement of tongue by these sublingual swellings. Further more growth in a inferior direction may give rise to appearance of characteristic "double chin". These well encapsulated lesions typically feel "dough like" on palpation, although they may be fluctuant and cyst like based on consistency of the luminal contents, that may range from a cheesy, sebaceous to liquefied substance [5,8]. Fine needle aspiration cytology, ultrasound, CT and MR imaging provide essential information on the cyst location that allows optimal preoperative planning. Ultrasonographic findings comprise solid and cystic structures within a heterogeneous mass. On CT scans, the dermoids 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. Although not equivalent to CT and MRI, it is safe, economical and dependable technique and is therefore useful for analysis of sublingual lesions. Treatment comprises total surgical excision. Caution should be taken not to rupture the cyst, as cystic contents may act as irritants to fibrovascular tissues, causing postoperative inflammation. 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 [4,5,7]. Differential diagnosis of dermoid cyst includes cystic hygroma, neurofibroma, haemangioma, sublingual ranula, Ludwig's angina, Lymphangioma, lipomas, acute infection, neoplasm, infections.

Materials and Methods

The central search engines like Pubmed, Scopus, Medline, Mendley and Google scholar is used to search all cases of dermoid cyst pulished till date. The word dermoid, epidermoid, tumors of head and neck, hamartoma and hamartomatous malformations are used for boolian search. Total 47 pulished studies have been retrieved from database meeting the search criteria. Out of 47 published studies 27 studies have been selected meeting inclusion criteria for meta-analysis. The inclusion criteria includes radiographic features on any imaging modality including ultrasound, CT, MRI etc.

Results

The categorical variables are presented in number and percentage and continuous variables are presented as mean and SD. Quantitative variables are compared using Unpaired t-test between two groups. Chi square test is used to know the frequency. A p value of <.05 will be considered statistically significant. The data will be entered in ms excel sheet and analysis will be done using SPSS version 21.0.

The study population consists of total 47 study subjects. The sex ratio in the study population showed that male patient proportion was higher than female. 29 subjects (61.7%) were male and 18 (8.3%) were female in study population (Table 1). The study subjects age ranged from 2 months to 60 years with the mean age of 19.86±15.24 years. The study population is divided in 5 age groups. The majority of study population (34%) belongs to 5 to 18 years followed by 19 to 35 years (27.7%),<5 years(21.3%),36

to 50 years (12.8%),50 to 60 years (4.3%) (Table 2). The Unpaired t-test is used to evaluate the site of occurance of dermoid cyst in male and female population. It is found that sublingual area is most common site of occurance in males (34.5%) and females (33.3%). The site of occurrence in sublingual area is followed by tongue(17.0%), Floor of the mouth(10.6%), Neck (8.5%). Parotid, nasal, mastoid regions and mandible were less frequently involved with two cases reported for each location. Other less frequently reported locations were temporal region, lip, soft palate, buccal mucosa, sphenoid region and forehead with only one case reported for each location (Table 3).

The age groups are corelated with site of occurrence, it was found that Out of 16 cases reported at the sublingual site, 8 cases occurred in the age group 5-18 years and 6 cases occurred in the age group 19-35 years and reaming 2 were in 36 to 50 years. In 36-50 years no case was reported. For 5 cases of dermoid cysts occurring in the floor of the mouth, all age groups were affected except 36-50 years in which no case was reported. At tongue, a total of 8 cases were reported, out of which, 5 occurred in the age interval of 5-18 years and 3 occurred in cases less than 5 years of age. In the neck region, out of 4 reported cases, 2 occurred in the age interval of 19-35 years, 1 was reported in each of 36-50 and less than 5 years age group (Table 4).

Out of total forty-seven (47) cases included in metaanalysis, 20 cases had no reported radiological findings (Table 5). Among the reported cases with radiological findings, Ultrasound, CT and MRI were the most commonly used radiological methods for diagnosis of Dermoid cyst.

The most common presentation of Dermoid cyst on Ultrasound is a well-defined cystic lesion with multiple rounded lobulated echogenic structures with mixed internal echoes. No significant vascularity is reported. Contrary to the low detection of plain radiograph, the positive rate of CT in diagnosis of dermoid cyst is much higher. CT has lower space resolution but is more sensitive in revealing bone shape than radiograph which can display trabecular bone pattern clearly. In a case of dermoid cyst, it is more important to demonstrate the change of bone shape than the trabecular bone pattern changes. CT can also reveal the relationship between the lesion

and adjacent hard or soft tissues after multiplanar reconstruction. On CT, most common presentation of Dermoid cyst is a well defined radiolucent, encapsulated cyst-like lesion containing low attenuation rounded structures of fat density.

MRI provides good contrast between different tissues and more detailed information than CT images especially for the soft tissues. Many tissues such as fatty tissue and ¹H-rich tissue (corpora vitreum, cerebrospinal fluid et al) have their unique features on MR image. Fatty tissue is of high signal intensity on T1 and T2 image and without change on T1 post-contrasted image and ¹H-rich tissue is of high signal intensity on T2 weighted-image, low signal intensity on T1WI and T1WI after contrasting. Functional MR imaging including MR spectroscopy, dynamic contrast-enhanced MRI, diffusion-weighted MR imaging (MR-DWI) has progressed rapidly in recent years offering more molecular biological evidences for diagnosis.

MRI of a Dermoid cyst demonstrated a lesion with low signal intensity on the T1-weighted image and high signal intensity on the T2-weighted image. The formation appeared encapsulated. Lesion appeared as containing multiple uniformly rounded foci, creating a "sack-of-marbles" appearance. However MRI is inferior to CT in demonstrating early bone changes therefore CT scan may be considered in some cases when bone changes was suspected. In one case reported by Kandogan T et al [4] at the sublingual region, dermoid cyst appeared as a wellcircumscribed non-enhancing cystic mass with homogenous content. In two cases of dermoid cysts reported by Christos Makos et al [13] at floor of the mouth region Scintiscan with a Tc^{99m} was performed which demonstrated no evidence of the existence of ectopic thyroid tissue within the lesion. In one case of dermoid cyst occurring in midline neck region, a mandibular occlusal plain radiograph revealed no calcifications with normal buccal and lingual cortical plates and no indication of expansion or decortication. Dardo Menditti et al [14] reported a case of Dermoid cyst in right mandible between the roots of canine and 1st premolar, in which a panoramic radiograph revealed a radio-transparent, monolocular area with very large displacement between these teeth.

Table 1:

Sex	N	0/0
Male	29	61.7
Female	18	38.3
Total	47	100.0

Table 2:

Age intervals	Frequency	Percent	
Below 5 years	10	21.3	
5 to 18 years	16	34.0	
19 to 35 years	13	27.7	
36 to 50 years	6	12.8	
50 to 60 years	2	4.3	
Total	47	100.0	

Table 3:

	s	Sex		
Site of Occurence	Male	Female		
Sublingual	10	6	16	
G	34.5%	33.3%	34.0%	
Floor of the mouth	3	2	5	
	10.3%	11.1%	10.6%	
Tongue	5	3	8	
_	17.2%	16.7%	17.0%	
Neck	3	1	4	
	10.3%	5.6%	8.5%	
Temporal region	0	1	1	
	.0%	5.6%	2.1%	
Parotid	1	1	2	
	3.4%	5.6%	4.3%	
Nasal	2	0	2	
	6.9%	.0%	4.3%	
Mastoid region	2	0	2	
_	6.9%	0.0%	4.3%	
Mandible	0	2	2	
	0.0%	11.1%	4.3%	
Lip, Soft Palate, Sphenoid sinus, Buccal Mucosa,	3	2	5	
Forehead	10.3%	11.1%	10.6%	
Total	29	18	47	
	100.0%	100.0%	100.0%	

Table 4:

SITE	Below 5 years	5 to 18 Years	50 to 60 years	Total		
Sublingual	0	8	6	2	0	16
	.0%	50.0%	46.2%	33.3%	.0%	34.0%
Floor of the mouth	1	1	2	0	1	5
	10.0%	6.2%	15.4%	.0%	50.0%	10.6%
Tongue	3	5	0	0	0	8
O	30.0%	31.2%	.0%	.0%	.0%	17.0%
Neck	1	0	2	1	0	4
	10.0%	.0%	15.4%	16.7%	.0%	8.5%
Temporal region	1	0	0	0	0	1
1 0	10.0%	.0%	.0%	.0%	.0%	2.1%
Parotid	0	0	1	0	1	2
	.0%	.0%	7.7%	.0%	50.0%	4.3%
Nasal	1	1	0	0	0	2
	10.0%	6.2%	.0%	.0%	.0%	4.3%
Mastoid region	1	0	1	0	0	2
· ·	10.0%	.0%	7.7%	.0%	.0%	4.3%
Mandible	0	1	1	0	0	2
	.0%	6.2%	7.7%	.0%	.0%	4.3%
Lip, Soft Palate,	2	0	0	3	0	5
Sphenoid, Buccal	20.0%	.0%	.0%	50.0%	.0%	10.6%
Mucosa, Forehead						
Total	10	16	13	6	2	47
	100.0%	100.0%	100.0%	100.0%	100.0%	100.0%

Table 5:

S.N.	Author	Year	Age	Sex	Site	Radiological	Radiological finding
1	Sellappampatty Veerappapillai	Dec	11	M	Sublingual	method USG	well-defined cystic lesion in the floor of the mouth), Cyst-
	Dhanasekaran, Shivakumar Senniappan, Shankar Radhakrishnan	2014			Ü	CT	like lesion, with a thin wall in the sublingual region.
2	Dr. Souvagini Acharya, Nilamadhab Prusty	Nov 2014	33	F	Right Parotid	USG	Mixed internal echoes due to fat content and shows the presence of osseous structures within
						CT	Fat globules floating within the lesion along with fat and fluid levels).
3	Shakeel M, Keh Sm, Chapman A, Hussain A	Nov 2014	62	M	Right Parotid	MRI	A 3 x 3 cm mass in the parotid tail
4	Mirza S, Fadl S, Napaki S, Abualruz A	June 2014	43	M	Sublingual	USG CT	A relatively well circumscribed midline cystic structure measuring approximately 8.3 * 7.7 * 5 cm with multiple rounded lobulated echogenic structures within. No significant vascularity was noted. No associated enlarged cervical lymph nodes were seen), Showed a well-defined encapsulated cystic mass lesion in the floor of the mouth occupying the submental region containing low attenuation rounded structures of fat
5	Uma Garg, Ritika Batra, M.K. Garg	Nov.	10	M	Sublingual	USG	density. revealed cystic mass of the size of 7 · 6 cm.
6	Zhao D, Han Y, Chen Y, Qiu J	2013 2013	22	M	Subcutaneous	None	None
7	Diercks GR, Iannuzzi RA, Mccowen	July 2013	31	F	Tissue Of The Mastoid Region Lateral Neck	Tione	
	K, Sadow PM.	Mar.			Associated With The Thyroid Gland Inferiorly	None	None
8	Sellappampatty Veerappapillai Dhanasekaran, Shivakumar Senniappan,	Dec 2012	11	M	Sublingual	None	None
9	Shankar Radhakrishna N K Sahoo, A K Choudhary	Dec 2012	28	M	Midline Neck	СТ	Demonstrated a cyst like lesion originating just off the midline in the sublingual region and elongated backwards. Well encapsulated cystic lesion with compression of the mylohyoid muscle could be seen in the mid line
10	WG CDR, Priya Jeyaraj And Brig NK Sahoo	Nov 2012	37	M	Midline Neck Extending From Inferior Border Of Mandibular Symphysis To The Level Of Cricoid Cartilage	Mandibular Occlusal Radiograph CT	No calcifications. The buccal and lingual cortical plates were normal with no indication of expansion or decortication revealed a large radiolucent space occupying lesion in the sublingual space
11	Nobuo Ohta, Tomoowatanabe, Tsukasa Ito, Toshinori Kubota, Yusuke Suzuki, Akihiro Ishida, Seiji Kakehata, Andmasaru Aoyagi	Sep. 2012	21	F	Sublingual	Axial and sagittal magnetic resonance	showed that tumor had low signal intensity on the T1- weighted image and high signal intensity on the T2- weighted image.
12	Marino R, Pentenero M, Familiari U And Gandolfo S	2012	15	M	Sublingual	imaging Magnetic Resonance Imaging (MRI)	showed a large lesion on the floor of the mouth about 4.5 cm in diameter. Body of the tongue appeared to be occupied by a voluminous cyst. The formation appeared encapsulated and caused a lifting of the body of the tongue dislocating caudal structures of the mouth floor.
13	Herlin C, Béziat Jl, Koppe M, Nimeskern N, Gleizal A.	2011 Nov	47	M	Bilateral Upper Lip	None	None
14	Lin Hw, Silver Al, Cunnane Me, Sadow Pm, Kieff Da.	2011 Oct	60	M	Submandibular Region (Floor Of The Mouth)	MRI	Demonstrated a lesion containing multiple uniformly rounded foci, creating a "sack-of-marbles" appearance
15	Christos Makos, George Noussios, Marinos Peios, Spyridon Gougousis, Andpantelis Chouridis	July 2011	21	F	Floor Of The Mouth	USG CT	infrasonic formation with distinct boundaries semi-transparent cystic formation with distinct boundaries
			17	M	Floor Of The Mouth	SCINTISCAN with a Tc ^{99m} USG CT	showed no evidence of the existence of ectopic thyroid tissue. sizeable infrasonic formation with distinct boundaries a sizeable cystic formation with distinct boundaries,
						SCINTI SCAN with a Tc ^{99m}	showed no evidence of the existence of ectopic thyroid tissue.
16	Harsha Jain, Sanjay Singh, Amit Singh	Mar. 2011	17	F	Sublingual	CT	Showed a well defined lesion in the floor of the mouth
17	Nwojo R, Roy S, Chang CY	2011 Jan	9 Month	M	Facial Canal In The Mastoid Segment Causing Facial Paralysis	MRI and CT	revealed marked widening of the facial canal in the mastoid segment consistent with facial nerve schwannoma or hemangioma
18	Suga K, Muramatsu K, Uchiyama T, Takano N, Shibahara T.	2010	4 Week	M	Midline Region Of The Soft Palate Close To The Uvula	None	None
19	Sanjay Jadwani, Biswajit Misra, Shreenivas Kallianpur, Snehal Bansod	Apr 2010	22	M	Sublingual	None	None
20	Emel Cadalli Tatar, Ömer Tarik Selçuk, Güleser Saylam, Ali Özdek,	Dec 2009	18 2Y 6	M M	Nasal Midline Nasal Dorsum	CT CT	no intracranial extension no intracranial extension
21	Hakan Korkmaz <u>Dizon M, Ozturk A, Redett Rj, Izbudak I</u>	2009 Aug	Months 5	F	Frontosphenoidal Region With Sinus Tracts In The Frontal Bone Extending To The Dura	CT, MRI	well-circumscribed non-enhancing cystic mass extending from the sublingual area to the thyroid notch level. The content of the cyst was homogenous

22	Danda Manditti Luigi Laina Nicola	Tom	30	T.	Dt. Mandible P /M	OPG	Radio transport manulacular area on the right side of
22	Dardo Menditti, Luigi Laino, Nicola Ferrara And Alfonso Baldi	Jan. 2008	30	F	Rt. Mandible B/W Roots Of Canine And 1st Premolar	OFG	Radio-trasparent, monolocular area on the right side of the mandibule, located between the roots of a canine and the first premolar with a very large displacement of the roots of these teeth.
23	Tolga Kandogan, Murat Koç, Enver Varda, Elif Selek And Özlem Sezgin	Sep. 2007	11	M	Sublingual	MRI	showed an 40 × 35 mm well-circumscribed non-enhancing cystic mass extending from the sublingual area to the thyroid notch level. The content of the cyst was homogenous
24	Bruno C. Jham, Gabriela V. Duraes, Andre C. Jham, Cassio R. Santos	Aug 2007	25	M	Sublingual	None	None
25	Ozan F, Polat Hb, Ay S, Goze F	2007 Mar	38	F	Buccal Mucosa	None	None
26	Mark F. Burger, Paul Holland, Bradford Napier	Nov 2006	25	M	Sublingual	MRI	demonstrated a sharply demarcated, fluid-filled, sublingual cyst that measured approximately $7.1\times4.5\times2.9$ cm.
27	Paolo Scolozzi, Tommaso Lombardi, Bertrandjaques	Feb 2005	1	F	Frontotemporal Region	CT and MRI	Revealed a cystic lesion within the frontal bone with a stalk extending down to the dura mater of the right middle cranial fossa
28	Paul C. Edwards, Liz Lustrin And Elsa Valderrama	Nov Dec.	Case 1	F	Sublingual	MRI	Indete crantal 1000a
		2003	Case 2	F	Ventral Tongue Extending To The Floor Of The Mouth		demonstrated a large irregular hyperintense tongue mass surrounded by a hypointense rim on T2-weighted
			Case 3	M	Ventral Surface Of Tongue		imaging
			Case 4	M	Tongue With Associated Submental		
			Case 5	M	Swelling Ventral Surface Of Tongue		
29	Tuz M, Dogru H, Uygur K, Baykal B	2003 Sep	18	F	Sublingual	None	None
30	Maria Salete Nahás Pires Corrèa, Ricardo De Nardi Fonoff, Henrique Castilhos Ruschel, Symonne Pimentel Castro De Oliveira Lima Parizotto, Fernanda Nahás Pires Corrèa	Apr 2003	15 Month	M	Left Ventral Part Of Tongue	None	None
31	Uppal Hs, D'souza AR, De R, Irving RM	2002 Feb	17	F	Infratemporal Fossa	None	None
32	Bloom D, Carvalho D, Edmonds J, Magit A	2002 Jan	Neonate (1 Week)	M	Floor Of The Mouth To Midline Neck	USG MRI	revealed a fluid-filled sublingual mass A 2-cm, cystic, left-sided, sublingual mass that crossed the midline without extension inferior to the Mylohyoid muscle.
33	Lohaus M, Hansmann J, Witzel A, Flechtenmacher C, Mende U, Reisser C	1999 Aug	19	F	Floor Of The Mouth	USG CT, MRI	a 5 x 5 x 4 cm cystic tumor with the unusual aspect of multiple smaller spherical formations
34	Hong SW	1998 Apr	2.5	F	Rt. Temporal Region (Frontotemporal)	CT, MRI	
35	Yamaki T, Higuchi R, Sasaki K, Nozaki M	1996 Dec	41	M	Forehead	CT	Multiple subcutaneous lesions in the frontal region with no intracranial extension.
36	Worthington JP, Sinclair JH	1992 Jan	25	M	Sublingual	None	None
37	Vucković N, Vucković D	1991	1.5	M	Midline Neck	None	None
38	Reddy Vs, Radhakrishna K, Rao Pl	1991	Infant	M	Intralingual	None	None
39	J M Goldman, D J Barnes, D V Pohl	Jan 1990	38	F	Sublingual	CT	showed a cystic mass beneath the tongue and narrowing of the nasopharynx (at the level of the uvula) and oropharynx.
40	Al-Fallouji Ma, Butler MF.	1983 July	15	F	Behind Ramus Right Mandible	None	None
41	J. E. Musgrove	Nov. 1950	23	F	Sublingual	None	None

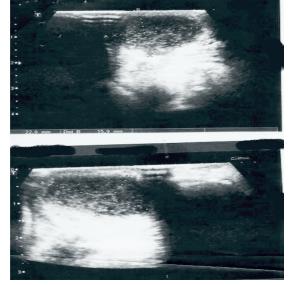


Fig. 1: HR USG of tongue showing a well defined heterogeneous predominantly hyperechoic lesion noted in left lateral border of tongue measuring approx. 2.29x1.59 cm

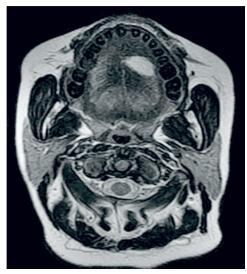


Fig. 2: MRI (Axial T2 FSE) image showing heterogenous intralingual mass with high signal intensity



Fig.3-MRI (Axial T1 FSE) showing heterogenous intralingual mass with low signal intensity involving anterior 2/3 rd of tongue on left side



Fig. 4: CT (Axial section) showing well defined hypodense, encapsulated cyst-like lesion containing low attenuation rounded structures of fat density

Discussion

Dermoid cysts are true developmental cysts, which arise from the entrapment of pluripotent ectodermal and mesodermal primordia in the embryonic lines of fusion, between the 3rd and 5th weeks of gestation. Hence, they are dysontogenic in origin, that is, they result from defective embryonic development. In the head and neck region, these ectodermal and mesodermal components get entrapped when the 1st and 2nd branchial arches of each side fuse in the midline. The entrapped tissues then undergo proliferation and cystic transformation. The dermoid cysts of the mouth are most frequently located on the median line of the mouth floor, most likely caused by the retention of the germinal epithelium during the growth of the mandible and hyoid branchial arches and are considered rare [2,3,8,15-19]. Although these lesions typically manifest during the second or third decade of life, however may present since birth with equal frequency of occurrence is both genders [20]. Clinically, the dermoid cysts usually present as a painless slow-growing mass at the sublingual, submental and submandibular region [8]. Dermoid cyst of the oral cavity is either sublingual or submental and starts in the midline, may extend laterally and downward and can attain a large size before presentation [1,8,21].

However, there can be sudden increase in the size of these lesions and it can be due to the onset of puberty when there is an increase in the secretion of sebum from the sebaceous glands 48 or may be due to secondary infection of the cyst contents either by blockage of salivary glands involved in the cyst or by implantation of oral microbials into the cyst through trauma causing pain, trismus, fever, dysphagia, odynophagia and cervical lymphadenopathy [8,22,23].

The word "Dermoid cyst", is a generic term which has been used to encompass and describe three different histological varieties. All three types may contain a cheesy keratinous material. Based on the histopathologic picture, Meyer divided the floor of the mouth cysts into following types [13,24,25]. Epidermoid cysts which are simple cysts lined with simple squamous epithelium, with a fibrous wall and no skin appendages or adnexals like hair follicles, sebaceous or sweat glands in their connective tissue wall. As these cysts develop from the upper part of the pilosebaceous unit they are incapable of producing sebum and do not contain any skin appendages. True Dermoid cysts also known as compound cysts, which are lined with keratinizing

stratified squamous epithelium with skin adnexals in the connective tissue wall. The lumen will contain keratin, sebum, varying amounts of fat and occasionally hair. Teratoid cysts also known as complex cysts which are lined with epithelium ranging from simple stratified, keratinized stratified squamous to stratified columnar respiratory type of epithelium and containing derivatives from all the three germ layers (ectoderm, mesoderm and endoderm) within their lumen. The cystic cavity in addition to skin appendages also encloses mesodermal derivatives such as bone, muscle, gastrointestinal and respiratory tissue. Theories of etiology of dermoid cysts includes congenital inclusion of dermal and epidermal elements of germ layers in deeper tissues along the embryonic lines of fusion, acquired traumatic implantation of dermal and epidermal elements of surface epithelium, which may proliferate and keratinize and growth from rest of totipotent cells displaced from the blastomere. Although floor of the mouth in the midline is most favoured site, occasional occurrence involving the buccal mucosa, tongue, lips, uvula, temporomandibular joint dermal graft, intradiploic, intracranial, and intraosseous location within the mandible and maxilla also have been cited in literature [9,11,18].

These lesions show variation in size and weight from few millimetres to centimetres and a gram to several hundred grams respectively [7,25]. Dermoid cysts present as a single, localized, slow growing, mobile, soft, firm or doughy, usually well circumscribed soft tissue mass. In the head and neck region, they are mostly seen in the midline and even when they are laterally positioned; their stalk can often be traced to the midline. They are usually seen in children, adolescents or young adults. They may present later in life as they are slow growing and become evident only after reaching an appreciable size. They may at times show a sudden increase in size during puberty or pregnancy, due to the hormonal changes (increased plasma levels of estrogen and progesterone), which stimulate the sebaceous glands that these cysts contain, with a resultant abundant sebum secretion and accumulation.

The sublingual dermoids raise the tongue and may cause difficulty in speech, swallowing and breathing, while the submental dermoids produce the characteristic "double chin" appearance. Symptoms of dysphagia, dyspnoea and dysphonia may occur due to upward displacement of tongue by these sublingual swellings [7,9]. Further more growth in a inferior direction may give rise to appearance of characteristic "double chin" [9,10,15]. These well encapsulated lesions typically feel "dough like" on

range from a cheesy, sebaceous to liquefied substance [4,7]. On an ultrasound, they typically appear as a well circumscribed, thin walled, unilocular mass with coalescence of fat globules into small, discrete, echogenic nodules within the fluid matrix, producing a characteristic "sac-of-marbles" appearance. On histopathologic examination, dermoid and epidermoid cysts are lined with stratified squamous epithelium with keratin and / or sebaceous material filled cavity.

In a true dermoid cyst, the connective tissue wall shows variable dermal appendages, including sebaceous glands, hair follicles and sweat glands. Treatment comprises total surgical excision [4,9,15,24]. Caution should be taken not to rupture the cyst, as cystic contents may act as irritants to fibrovascular tissues causing postoperative inflammation [25].

Recurrences are unusual after absolute surgical excision [15,25]. Reports of malignant transformation of sublingual dermoid and epidermoid to squamous carcinoma and basal cell carcinoma are present [12,26]. A 5% rate of malignant transformation of the teratoid variety of oral dermoid cysts has also been quoted in literature [11,12,26].

Conclusion

Epidermoid cyst of the oral cavity is an uncommon entity. Ample understanding and vigilance about this slow growing painless mass is essential not only because of the symptoms it produces but also due to its malignant potential.

References

- Vargas Fernandez JL, Lorenzo Rojas J, Aneiros Fernandez J, Sainz Quevedo M. Dermoid cyst of the floor of the mouth. Acta Otorrinolaringol Esp. 2007; 58:31–33.
- Ege G, Akman H, Senvar A, Kuzucu K. Case report: sublingual dermoid cyst. Tani Girisim Radyol 2003; 9:57–59.
- De Ponte FS, Brunelli A, Marchetti E, Bottini D. Sublingual epidermoid cyst. J Craniofac Surg 2002; 13:308–310.
- Kandogan T, Koc M, Vardar E, Selek E, Sezgin O. Sublingual epidermoid cyst: a case report. J Med Case Rep. 2207; 1:87.
- 5. Damle MV, Irani DK, Hiranandani NL: Epidermoid cyst of the floor of the mouth. case report. J Med case Reports. 2007; 1:87.

- Ozan F, Polat HB, Ay S, Goze F: Epidermoid cyst of the buccal mucosa: A case report. J Contemp, Dent Pract. 2007; 3:90-96.
- Pancholi A, Raniga S, Vohra PA, Vaidya V: Midline submental epidermoid cyst: A Rare Case. Internet J Otorhinolaryngol 2006.
- 8. Seah TE, Sufyan W, Singh B: Case report of a dermoid cyst of the floor of the mouth. Bombay Hos J. 2002.p.44.
- Koca H, Seckin T, Sipahi A, Kaznac A: Epidermoid cyst in the floor of the mouth: report of a case.Quintessence Int. 2007; 38:473-477.
- Hemaraju N, Nanda SK, Mediker SB: Sublingual dermoid cyst. Indian Journal of Otolaryngology and Head and Neck Surgery. 2004; 3:218-220.
- 11. Shear M, Speight P: Cysts of Oral and Maxillofacial Region. 4th edition. Blackwell Munksgaard; 2007: 181-183.
- 12. Rutherford SA, Leach PA, King AT: Skull Base: An Interdisciplinary Approach. 2006; 2:109-115.
- Christos Makos, George Noussios, Marinos Peios, Spyridon Gougousis, Andpantelis Chouridis. Dermoid Cysts of the Floor of the Mouth. Two Case Reports. Case Rep Med 2011.
- 14. Menditti D, Laino L, Ferrara N, Baldi A. Dermoid cyst of the mandibula: a case report. Cases J. 2008 Oct 22; 1(1):260.
- Black EE, Leathers RD, Youngblood D. Dermoid cyst of the floor of the mouth. Oral Surg Oral Med Oral Pathol. 1993; 75:556–558.
- Lozano Blanco I, Perez Climent F, Carbonell Sanchis R, Gimenez Vaillo F, Oliver Guasp C. Giant dermoid cyst of the floor of the mouth. An Otorrinolaringol Ibero Am. 1995; 22:601–608.

- 17. Terribile Wiel Marin V, Passi P, Miotti A, Bragaggia P. A case of giant dermoid cyst of the mouth floor. Stomatol Mediterr/ 1985; 5:195–199.
- Hunter TB, Paplanus SH, Chernin MM, Coulthard SW. Dermoid cyst of the floor of the mouth: CT appearance. AJR Am J Roentgenol. 1983; 141:1239– 1240.
- Akao I, Nobukiyo S, Kobayashi T, Kikuchi H, Koizuka I. A case of large dermoid cyst in the floor of the mouth. Auris Nasus Larynx. 2003; 30:S137– 139.
- Mancuso AA, Hanafee WN. Kirchner JA. Computed tomography and magnetic resonance imaging of the head and neck, 2nd edn. Williams & Wilkins, Baltim. 1985.
- 21. Ramsey RG. Neuroradiology with computed tomography. Saunders, Philadelphia. 1981.
- Shaari CM, Ho BT, Shah K, Biller HF. Lingual dermoid cyst. Otolaryngol Head Neck Surg. 1995; 112:476– 478.
- King RC, Smith BR, Burk JL. Dermoid cyst in the floor of the mouth. Review of the literature and case reports. Oral Surg Oral Med Oral Pathol. 1994; 78:567– 576.
- 24. Janjua TA, Goravalingappa R. Quiz case 1. Submandibular dermoid cyst. Arch Otolaryngol Head Neck Surg. 1999; 125:1270, 1272.
- Teszler CB, El-Naaj IA, Emodi O, Luntz M, Peled M. Dermoid cysts of the lateral floor of the mouth: a comprehensive anatomosurgical classification of cysts of the oral floor. J Oral Maxillofac Surg. 2007; 65:327–332.
- 26. Mustafa Tuz, Harun Dogru, Kemal Uygur. Rapidly growing sublingual dermoid cyst throughout pregnancy. Am J Otolaryngol. 2003; 24:334-337.