

Dermoids of the Maxillofacial Region: A Study on Clinical Presentation and Surgical Management

Divashree Sharma, Geeta Mishra Tripathi, Ambrish Mishra¹, Neelam Shakya²

Departments of Dentistry and ¹Community Medicine, Shyam Shah Medical College, Rewa, ²Department of Oral Surgery, Government College of Dentistry, Indore, Madhya Pradesh, India

Abstract

Introduction: Dermoid and epidermoid cysts commonly termed as “Dermoids” are known to be developmental benign swellings that possibly occur as a result of traumatic implantation of epithelium or entrapment of epithelial remnants during embryonic fusion. The occurrence is rare (about 7%) in the head-and-neck region and they represent <1.6% of all cysts of the oral cavity. **Patients and Methods:** A retrospective study was done on 12 histopathologically proven cases of epidermoid and dermoid cysts of the maxillofacial region that were treated in Shyam Shah Medical College, Rewa, Madhya Pradesh, India, from January 2013 to December 2019. The cases were evaluated for clinical presentation, demographic characteristics, and treatment performed. **Results:** Out of a total of 12 cases that were included in this study, 10 cases were diagnosed as epidermoid and 2 cases as true dermoid cysts. The mean age of occurrence was 31.66 ± 9.97 years. Male predominance was seen with a male-to-female ratio of 1.4:1. The floor of the mouth was the most commonly involved site in ten (83.34%) cases. Sublingual cysts were seen in five (41.67%) cases, combined sublingual-submandibular involvement in three (25%) cases, and midline floor of the mouth cyst in two (16.67%) cases. All the cases were treated by surgical enucleation, and no recurrence or malignant transformation was reported in any case at a minimum follow-up of 2 years. **Conclusion:** Based on the findings of this retrospective study, it is suggested that these lesions are very rare in the maxillofacial region and pose a diagnostic challenge. However, fine-needle aspiration cytology, ultrasound imaging, computerized tomography, and magnetic resonance imaging techniques can be relied upon for precise treatment planning. Surgical enucleation of these lesions is the preferred treatment modality.

Keywords: Buccal, dermoid cyst, epidermoid cyst, floor of the mouth, sublingual, submandibular, surgical enucleation

Submitted: 05-Mar-2022; **Revised:** 31-May-2022; **Accepted:** 03-Jun-2022; **Published:** 27-Aug-2022

INTRODUCTION

Epidermoid, dermoid, and teratoid cysts are closely related uncommon, benign, developmental cystic malformations and are commonly termed as “dermoids” or “dysontogenetic cysts.”^[1] Epidermoid cyst is described as “a simple cyst lined with stratified squamous epithelium containing cystic fluid or keratin and no other specialized structure in its lumen.”^[2] The characteristic feature of dermoid cyst is the presence of the skin appendages, whereas in the teratoid cyst, the lining varies from squamous to ciliated respiratory epithelium with derivatives from ectoderm, mesoderm, and/or endoderm.^[3] The most common location of these cysts is ovaries, and sacral region (about 80%) and only 1.6% to 6.9% of epidermoid and dermoid cysts have been reported in the head-and-neck area predominantly in orbit, calvarial diploic space, and intracranial region.^[4,5] The incidence of epidermoid

cyst in the oral cavity is extremely rare (1.6%).^[6] This was a retrospective study conducted on 12 histopathologically proven cases of dermoid cysts (ten cases of epidermoid and two cases of true dermoid cyst) of the maxillofacial region that were treated by surgical enucleation.

PATIENTS AND METHODS

A retrospective study based on the clinical records was conducted on 12 patients who were diagnosed with epidermoid or dermoid cysts of the maxillofacial region upon histopathologic examination

Address for correspondence: Dr. Divashree Sharma,
F-12/1, New Doctors Colony, Arjun Nagar, Rewa - 486 001,
Madhya Pradesh, India.
E-mail: divashri.19.sharma@gmail.com

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Sharma D, Tripathi GM, Mishra A, Shakya N. Dermoids of the maxillofacial region: A study on clinical presentation and surgical management. Indian J Dent Sci 2022;14:127-31.

of the enucleated specimen from January 2013 to December 2019 in the Dentistry Department, SS Medical College and associated Hospital, Rewa, Madhya Pradesh, India. Only the cases which underwent ultrasound and/or computerized tomography (CT) scan and/or magnetic resonance imaging (MRI) and fine-needle aspiration cytology (FNAC) report as a part of preoperative assessment were included in the study. The study focused on evaluating the anatomic location, clinical and demographic features, treatment performed, and treatment outcomes of these patients. The patients were operated either under general anesthesia with rhino-tracheal intubation or under local anesthesia. Surgical enucleation was performed in all cases (through intraoral approach in 11 cases and transcutaneous extraoral approach in 1 case), followed by histopathologic examination of the excised specimen [Figures 1-5]. The postoperative follow-up period was a minimum of 2 years in all patients. A follow-up period of maximum 7 years was reported in one patient. The location, size, histopathological diagnosis, and surgical treatment performed for all cases are summarized in Table 1.

RESULTS

A male-to-female ratio of 1.4:1 and a mean age of 31.66 ± 9.97 years were reported in this study. Epidermoid

cyst of the floor of the mouth was reported in nine (83.34%) patients (sublingual epidermoid cyst in four patients (33.34%), combined sublingual–submandibular epidermoid in three patients (25%), and midline floor of the mouth cyst in two (16.67%) patients), epidermoid cyst of left cheek in one (8.33%) patient, and sublingual dermoid cyst and dermoid cyst of upper labial mucosa in one patient each (8.33%). All the cases were treated by surgical enucleation [Table 1]. Postoperative complications such as infection, wound dehiscence, or nerve injury did not occur in any of patients. No recurrence or malignant transformation was reported in any of the cases at a minimum follow-up of 2 years.

DISCUSSION

Meyer proposed the term “dermoid” to denote all the three categories of the cysts, i.e., epidermoid, true dermoid, and teratoid cysts.^[3] When the cyst presents with a lining of only squamous epithelium with a fibrous wall, it is regarded as an epidermoid cyst, when keratinization with skin adnexa such as pilous follicles, and sudoriparous and sebaceous glands are



Figure 1: Extraoral photograph revealing a swelling below the chin and lower border of mandible on left side and intraoral photograph showing a swelling in the floor of the mouth leading to tongue elevation



Figure 2: Exposure of the cyst through intraoral elliptical incision made in the floor of the mouth



Figure 3: The enucleated specimen and the aspirated fluid from the lesion

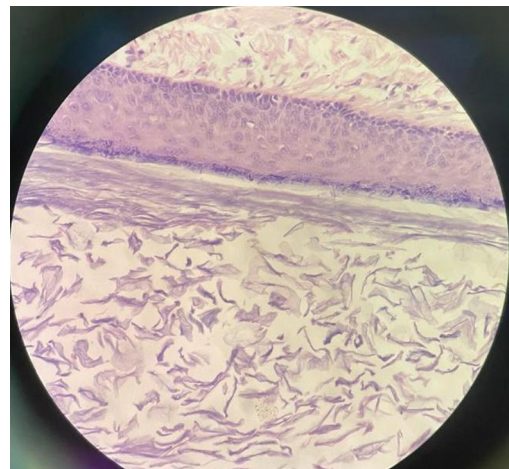


Figure 4: Histopathologic appearance of epidermoid cyst

Table 1: Distribution of patients according to the demographic data, clinical characteristics, histopathology, and treatment performed

Serial number	Location of the cyst	Age of the patient	Gender	Size of excised specimen (cm)	Histopathologic diagnosis	Treatment performed
1	Left sublingual region	32	Male	3.5 × 2.0	Epidermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth under LA
2	Left sublingual region and submandibular region	38	Female	6.2 × 5.2	Epidermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth under GA
3	Left sublingual and submandibular region	28	Female	5.5 × 4.8	Epidermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth under GA
4	Right sublingual and submandibular region	33	Male	5.0 × 4.5	Epidermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth under GA
5	Midline floor of the mouth	38	Male	3.5 × 3.0	Epidermoid cyst	Enucleation by intraoral approach through a median mucosal incision in the floor of the mouth under GA
6	Right sublingual region	19	Male	4.0 × 3.8	Epidermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth under GA
7	Right sublingual region	25	Female	3.9 × 2.7	Epidermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth under GA
8	Right sublingual region	29	Female	4.0 × 3.5	Epidermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth. under GA
9	Midline floor of the mouth	22	Male	3.5 × 2.4	Epidermoid cyst	Enucleation by intraoral approach through median mucosal incision in the floor of the mouth under LA
10	Well circumscribed swelling in left submandibular region	39	Male	5.6 × 5.1	Epidermoid cyst	Enucleation through extraoral submandibular approach under GA
11	Midline upper labial mucosa	55	Male	1.0 × 0.5	Dermoid cyst	Enucleation by intraoral approach through a circumferential incision in upper labial mucosa under LA
12	Right sublingual region	22	Female	2.0 × 1.5	Dermoid cyst	Enucleation by intraoral approach through an elliptical incision in the floor of the mouth under LA

GA: General anesthesia, LA: Local anesthesia

**Figure 5:** Exposure of a large extraoral well-circumscribed swelling through left submandibular incision which was diagnosed as epidermoid cyst on histopathological examination

found on the cyst wall it is regarded as a true dermoid cyst, and when the lining varies from simple squamous to a ciliate respiratory epithelium containing ectodermal, mesodermal, and/or endodermal derivatives (muscle, cartilage, or bone), the cyst is classified as a teratoid cyst.^[3,7] A thick, greasy-looking material is found on aspiration in all three variants.^[8] Epidermoid cysts may be classified as congenital or acquired. The most common theory of the occurrence of epidermoid and

dermoid cysts is that they usually appear in the midline floor of the mouth as a result of entrapped ectodermal tissue of the first and second branchial arches, which fuse during the 3rd and 4th weeks *in utero*. A second theory proposes that these midline cysts may be a variant of the thyroglossal duct cyst.^[9] Acquired epidermoid cysts are also thought to arise from a traumatic or surgical injury which causes the inclusion of ectoderm or occlusion of sebaceous gland duct.^[10] Certain drugs such as cyclosporine, imiquimod, and tacrolimus have also been implicated in the eruption of epidermoid cysts.^[11] However, none of our patients presented with the history of intake of any such drugs. Smoking has also been reported to be associated with the occurrence of facial epidermal inclusion cysts.^[11] The male patients who presented with facial epidermoid cyst on the left submandibular region and right sublingual epidermoid cyst were long-standing chronic smokers.

The occurrence of dermoid and epidermoid cysts in the floor of the mouth is rare and represents <0.01% of all cysts of the oral cavity.^[12] The other reported sites are buccal mucosa, lips, tongue gingiva, uvula, temporomandibular joint dermal graft, intracranial, intradiploic, and intraosseous within jaw bones.^[12-14] Dermoid cysts of the scalp or ear regions may present with intracranial extension. When such a cyst enlarges and ruptures, the contents pass into the cerebral ventricle or to subarachnoid space progressing to the development of encephalomeningitis and seizures.^[15]

The lesions exhibit slow, painless growth pattern and are commonly diagnosed in the second or third decade of life even if they are congenital.^[16] The lesion is known to occur predominantly in males.^[17,18] Our study was compliant with the findings in literature with a male preponderance and in terms of mean age of occurrence.

An anatomic classification was proposed by Longo *et al.*^[4] to classify the epidermoid cysts into three variants based on the location of the cyst in relation to the muscles of the floor of the mouth. When the cyst is present above the geniohyoid muscles, it is referred to as “median genioglossal cysts or sublingual cysts.” The cyst which is present between the geniohyoid and the mylohyoid muscles is termed as “median geniohyoid cyst” and as “the lateral cysts” when located in the submaxillary region.^[16] Cysts that develop above the mylohyoid have the potential to progressively enlarge and eventually displace the tongue toward the palate and lead to difficulty with chewing, dysphagia, dysarthria, dysphonia, and dyspnea. Cystic lesions that arise below the mylohyoid develop into submental or submandibular swelling.^[9] In three of our cases, the lesion probably originated as sublingual lesion primarily and herniated through the mylohyoid into submental and submandibular space similar to the case reported by Tandon and Gupta.^[4]

The differential diagnosis should include ranula, benign tumors, inflammatory disorders of mucosa or salivary glands, lymphatic or vascular malformations, lipoma, branchial cleft cysts, sublingual or submandibular cellulitis, sublingual or submandibular abscess, thyroglossal duct cyst, malignant tumors, and heterotopic gastrointestinal cyst.^[6,19]

It is suggested in the literature that epidermoid cysts larger than 5 cm on cheek are rare.^[20] One patient in this study presented with an epidermoid cyst measuring 5.6 cm × 5.1 cm on left cheek.

Although marsupialization of very large cysts is reported in the literature, the preferred treatment approach is complete surgical enucleation without rupturing the cyst because the intraluminal cystic contents serve as irritants for fibrovascular tissues in the vicinity leading to inflammation.^[19] The choice of the approach for the lesions in the maxillofacial region relies on the determination of exact location of the lesion through clinical diagnosis supplemented with ultrasonography, computerized tomography (CT) scan, and/or MRI. Extraoral approach is preferred for median geniohyoid or very large sublingual cysts, and the intraoral approach for medium or small size sublingual cysts.^[16] Enucleation of a very large sublingual dermoid cyst through symphyseal mandibular osteotomy has been reported by McGregor.^[21] In our study, all cases involving the floor of the mouth were enucleated through an intraoral incision.

Malignant transformation of dermoid cysts to squamous cell carcinoma and adenocarcinoma has been reported in the literature.^[22] A “5% rate of malignant transformation of oral dermoid cysts of the teratoid type” was reported by Zachariades

and Skoura-Kafoussia in his study.^[23] However, in our study, recurrence or malignant transformation was not reported in any of the cases similar to the study by Tandon and Gupta which reported that recurrence and malignant transformation are very uncommon after complete surgical enucleation.^[4]

CONCLUSION

These lesions are rare in the maxillofacial region and pose a diagnostic dilemma. In our study, the diagnosis was reached by FNAC, ultrasonography and/or computerized tomography scan and/or MRI, and subsequent histopathology of the excised specimen. Based on this study, it is recommended that imaging modalities are an important aid that provide diagnostic information about the size and location of the lesion and guide the treatment plan. Surgical enucleation is the preferred treatment modality and the choice of approach (intraoral/extraoral or combined) depends on the anatomic location of the cyst. Prognosis was good, with no incidence of relapse or malignant changes.

Ethical clearance

The study was approved by the institutional Ethics Committee of Shyam Shah Medical College, Rewa M.P (Approval No: S.no/IEC/MC/2022/4831).

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Tsirevelou P, Papamantios M, Chlopsidis P, Zourou I, Skoulakis C. Epidermoid cyst of the floor of the mouth: Two case reports. *Cases J* 2009;2:9360.
2. Passi D, Singh G, Mehta G, Singhal D. Unusually large submandibular epidermoid cyst: A case report, differential diagnosis and therapy. *Contemp Clin Dent* 2014;5:252-5.
3. Meyer I. Dermoid cysts (dermoids) of the floor of the mouth. *Oral Surg Oral Med Oral Pathol* 1955;8:1149-64.
4. Tandon PN, Gupta DS. Epidermoid cyst in the floor of mouth with sub mental component. *J Maxillofac Oral Surg* 2014;13:59-62.
5. Jham BC, Duraes GV, Jham AC, Santos CR. Epidermoid cyst of the floor of the mouth: A case report. *J Can Dent Assoc* 2007;73:525-8.
6. Walstad WR, Solomon JM, Schow SR, Ochs MW. Midline cystic lesion of the floor of the mouth. *J Oral Maxillofac Surg* 1998;56:70-4.
7. Calderon S, Kaplan I. Concomitant sublingual and submental epidermoid cysts: A case report. *J Oral Maxillofac Surg* 1993;51:790-2.
8. Vargas Fernández JL, Lorenzo Rojas J, Aneiros Fernández J, Sainz Quevedo M. Dermoid cyst of the floor of the mouth. *Acta Otorrinolaringol Esp* 2007;58:31-3.
9. Louis PJ, Hudson C, Reddi S. Lesion of floor of the mouth. *J Oral Maxillofac Surg* 2002;60:804-7.
10. Kandogan T, Koç M, Vardar E, Selek E, Sezgin O. Sublingual epidermoid cyst: A case report. *J Med Case Rep* 2007;1:87.
11. Mayer JE, Miller MD, Birlea SA. Symmetric multilocular epidermoid cysts on the face: An unusual presentation of a common lesion. *JAAD Case Rep* 2018;4:337-9. Ozan F, Polat HB, Ay S, Goze F. Epidermoid cyst of the buccal mucosa: A case report. *J Contemp Dent Pract* 2007;8:90-6.
12. Sahoo NK, Choudhary AK, Srinivas V; Kapil Tomar. Dermoid cysts of maxillofacial region. *Med J Armed Forces India* 2015;71:S389-94.

13. Ravindranath AP, Ramalingam K, Natesan A, Ramani P, Premkumar P, Thiruvengadam C. Epidermoid cysts: An exclusive palatal presentation and a case series. *Int J Dermatol* 2009;48:412-5.
14. Longo F, Maremonti P, Mangone GM, De Maria G, Califano L. Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. *Plast Reconstr Surg* 2003;112:1560-5.
15. Choi JS, Bae YC, Lee JW, Kang GB. Dermoid cysts: Epidemiology and diagnostic approach based on clinical experiences. *Arch Plast Surg* 2018;45:512-6.
16. Dutta M, Saha J, Biswas G, Chattopadhyay S, Sen I, Sinha R. Epidermoid cysts in head and neck: Our experiences, with review of literature. *Indian J Otolaryngol Head Neck Surg* 2013;65:14-21.
17. Kim CS, Na YC, Yun CS, Huh WH, Lim BR. Epidermoid cyst: A single-center review of 432 cases. *Arch Craniofac Surg* 2020;21:171-5.
18. Baek SO, Kim SW, Jung SN, Sohn WI, Kwon H. Giant epidermal inclusion facial cyst. *J Craniofac Surg* 2011;22:1149-51.
19. Hiremath R, Chandrashekarayya SH, Manswini Pol TJ, Anegundi KR. A rare case of a submental epidermoid cyst: A case report. *J Clin Diagn Res* 2011;5:1452-3.
20. de Mendonça JC, Jardim EC, Dos Santos CM, Masocatto DC, de Quadros DC, Oliveira MM, *et al.* Epidermoid cyst: Clinical and surgical case report. *Ann Maxillofac Surg* 2017;7:151-4.
21. McGregor IA. Symphyseal mandibular osteotomy in the approach to sublingual dermoid cyst. *Br J Plast Surg* 1991;44:544-5.
22. Cabibi D, Martorana A, Cappello F, Barresi E, Di Gangi C, Rodolico V. Carcinosarcoma of monoclonal origin arising in a dermoid cyst of ovary: A case report. *BMC Cancer* 2006;6:47.
23. Zachariades N, Skoura-Kafoussia C. A life-threatening epidermoid cyst of the floor of the mouth: Report of a case. *J Oral Maxillofac Surg* 1990;48:400-3.