Sublingual Dermoid Cyst: Review of 14 Cases

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Abstract

Background: Dermoid cyst is a benign congenital lesion of ectodermal origin. They are commonly found throughout the body but rare in the oral cavity. It is a developmental lesion and usually due to retention of germinal epithelium during growth of brachial arches and lower jaw. It is commonly present in the sublingual region as a swelling in the middle of the mouth. It presents as a slow growing mass, causing elevation of the tongue and interference with speech and swallowing. **Aim and Objective:** To determine the epidemiology, presentation, co-morbidities and treatment of sublingual dermoid cyst in our Centre. **Materials and Methods:** This is a retrospective review of all patients with sublingual dermoid cyst managed over a period of eight years from January 2010 to December 2017. Information was extracted from case files of patients. Data collected included: age of patient at presentation, sex, location of cyst, co-morbidities, treatment giving, findings and histological diagnosis. Only patients with a histological diagnosis of dermoid cyst were included in the study. **Results:** Fourteen cases were included in this study. Eight were males (57.1%) and six (42.9%) females. Male to female ratio is 1.3:1. Age range is from Day 1 to 25years. Five were congenital sublingual dermoid cyst in newborn, one was attached to the tongue ventral surface, two were bulging from the submandibular and submental spaces in adults. The remaining six cases were limited to the floor of the mouth. **Conclusion:** Sublingual dermoid cyst could interfere with swallowing and speech, this affects patient nutrition and breathing. Therefore, surgical excision of the cyst should be done promptly.

Keywords: Sublingual, dermoid, cyst, floor of the mouth, newborn, congenital lesion, epidermoid, submandibular, oral cavity

Introduction

Dermoid cysts are subcutaneous cysts of the ectodermal origin found along the lines of embryonic fusion, which is hamartomatous.[1] The tumor is covered by a thick dermis-like wall that contains multiple sebaceous glands, hairs, and large amounts of fatty masses and almost all skin adnexa.[2] It is a teratoma of cystic nature that contains an array of developmentally mature solid tissues. It frequently consists of skin appendages, hair follicles, and sweat glands, while other commonly found components include clumps of long hair, pockets of sebum, blood, fat, bone nails, teeth, eyes, cartilage, and thyroid tissue.[2] Dermoid cysts grow slowly and contain mature tissue; this type of teratoma is nearly always benign; in rare cases, squamous cell carcinoma usually develops from the wall of the cyst in adults.[3]

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Dermoid cyst could be congenital or acquired. Acquired type may be due to trauma. Dermoid cyst usually presents early in life as an asymptomatic mass.

However, they may reach a large size and involve more than one anatomical area and touch the hyoid bone when in the neck.[4] Most dermoid cysts on the floor of the mouth occur in individual aged 10–30 years.[4] However, there are few reports

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of oral dermoid cysts in newborns or children.[4] Dermoid cyst is treated with complete surgical excision, and the prognosis is good. Recurrences of the cyst have been recorded in literature.[4] Sometimes, they may not be noticed until a child is older or an adult. Comorbidities of the sublingual dermoid cyst include upper respiratory tract infection, anemia, respiratory obstruction, feeding difficulties, and esthetic challenges.[5] This report contains 14 cases of sublingual dermoid cyst treated in our center over 8 years.

Materials and Methods

A retrospective study of patients who were treated for sublingual dermoid cyst of the oral cavity at Barau Dikko Teaching Hospital over 8 years from January 2010 to December 2017 was done. Fourteen cases were included in this study. The patients were analyzed for age, sex, site, location of the cyst, comorbidities and approach for surgery, forms of anesthesia, treatments given, and recurrences.

All patients with suspected sublingual dermoid cysts were included in the study.

Results

Fourteen patients were treated, of which 8 (57.1%) were males and 6 (42.9%) were females. The ratio of male to female is 1.3:1.0. Age range of the patients was 1 day to 25 years [Table 1]. Five of the patients were newborn (cases 1–5) and presented with congenital sublingual dermoid cyst in the floor of the mouth [Figure 1a]. The cyst was attached to the ventral surface of the tongue in one of the patients aged 21 years (case 12); in two female adults aged 23 and 25 years (cases 13 and 14), the cysts were bulging from the submandibular and submental spaces [Figure 2a]. The remaining six cases were limited to the floor of the mouth (cases 1–12).

The lesions were no fluctuant, did not transilluminate, and were all located at the floor of the mouth.

The comorbid symptoms include: upper respiratory tract infection, which is the most common, followed by anemia in infants, feeding challenges were mostly encountered in newborns, whereas aesthetics and difficulty in swallowing were common in adults.

Two of our adult patients were restricted to fluid diet (cases 12 and 14). Periodical reduction of the cystic fluid was done by decompression, to reduce the volume of the lesion to allow for ease of breathing and feeding, especially in the newborn before the time of surgery. The newborns had surgery at the age 3 of months, hemoglobin of 10 g/dl, and body weight of 10 pounds or 5 kg, which was considered appropriate for surgery. Endotracheal intubation was difficult until cyst fluid was carefully decompressed to reduce the size. One neonate became cyanosed after intubation and the surgery was postponed. The surgery was done later after he was treated for respiratory tract infection. Axial T1- weighted Magnetic Resonance Imaging (MRI) for case 11 showed a sharply circumscribed cystic mass in the floor of the mouth

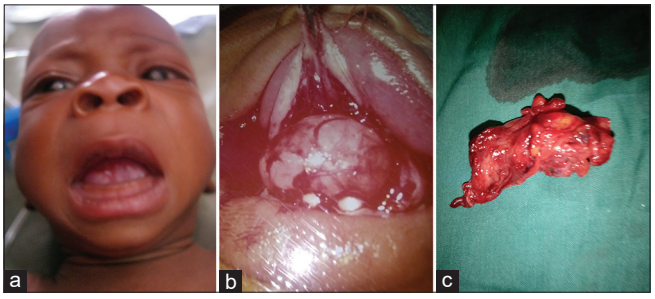

Figure 1: (a) 3-month-old baby boy with sublingual dermoid cyst. (b) 3-month-old baby boy with exposed sublingual dermoid cyst under general anesthesia. (c) Excised cyst.

Table 1: Age, sex, and other parameters

Cases age	Sex	Site	Duration	Symptoms	Comorbidity	Surgical approach	Histology
1 - Day 1	Male	Sublingual midline	Birth	SRD	URTI	Intraoral	Dermoid
2 - Day 1	Female	Sublingual midline	Birth	SRD	URTI	Intraoral	Dermoid
3 - Day 1	Male	Sublingual midline	Birth	SRD	URTI	Intraoral	Dermoid
4 - Day 1	Male	Sublingual midline	Birth	SRD	URTI	Intraoral	Dermoid
5 - Day 1	Female	Sublingual midline	Birth	SRD	URTI	Intraoral	Dermoid
6 - 2/12	Male	Sublingual midline	2 months	SRD	URTI	Intraoral	Dermoid
7 - 1 year	Female	Sublingual midline	1 year	SRD	Anemia	Intraoral	Dermoid
8 - 7	Male	Sublingual midline	7 years	RD	URTI	Intraoral	Epidermoid
9 - 9	Male	Sublingual midline	9 years	AS	None	Intraoral	Epidermoid
10 - 10	Female	Sublingual midline	10 years	AS	None	Intraoral	Epidermoid
11 - 13	Male	Sublingual midline	13	AS	None	Intraoral	Epidermoid
12 - 21	Male	Ventral tongue	21	AS	None	Intraoral	Epidermoid
13 - 23	Female	Submandibular Submental	23	SD	Esthetic	Extraoral	Dermoid
14 - 25	Female	Submandibular Submental	25	SD	Esthetic	Extraoral	Dermoid

SRD=Swallowing and respiratory difficult; RD=Respiratory difficulty; AS=A symptomatic; SD=Swallowing difficulty; URTI=Upper respiratory tract infection

[Figure 3] and a Computed Tomography Scan (CT) of the neck for case 14 showed encapsulated left sided mass with multiple cellular masses at the floor of the mouth [Figure 4].

All patients went through routine hematological and biochemical investigations. Computed Tomography Scan (CT) of the neck for case 14 showed encapsulated left sided mass with multiple cellular masses at the floor of the mouth [Figure 4] and Axial T1- weighted Magnetic Resonance Imaging (MRI) for case 11 showed a sharply circumscribed cystic mass in the floor of the mouth [Figure 3]. Eight cases were done under general anesthesia while six under local anesthesia. Two patients (cases 13 and 14) had extraoral approach for the surgical excision, while 12 patients had intraoral approach. The outcome of the treatments was satisfactory. Aspiration cytology was done which revealed cheesy material-containing nonnucleated epithelial cells. A provisional diagnosis of the sublingual dermoid cyst was made. The excised cyst was sent for histology which revealed the following: gross pathological evaluation showed a 2.6 cm × 1.5 cm × 1.0 cm thin-walled, unilocular cystic mass filled with keratin debris.

Microscopically, the cyst lining was composed of stratified squamous epithelium with keratin debris and sebaceous glands with associated hair follicles. The diagnosis of the dermoid cyst was confirmed [Figure 5].

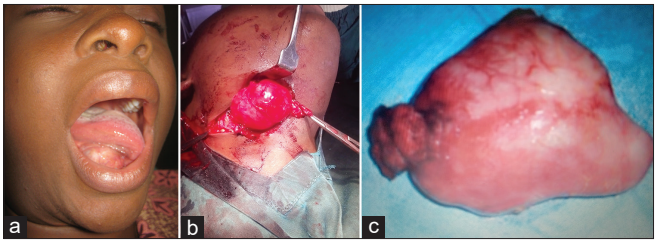

Figure 2: (a) 23-year-old female with cyst at the floor of the mouth and bulging through submandibular space. (b) The cyst being enucleated under general anesthesia. (c) Excised cyst.

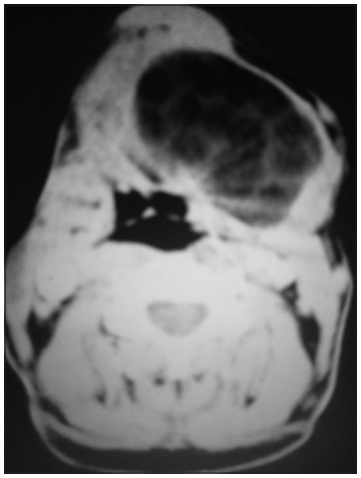

Figure 4: Computed tomography scan of the neck showing encapsulated left-sided mass with multiple cellular areas

Surgical procedure

Midline incision was made along the longitudinal ventral surface of the tongue to the floor of the mouth, and by careful dissection, the cyst was separated out of the mylohyoid muscle [Figure 1b]. This procedure was repeated for all the patients who had intraoral excision. Extraoral approach was done for cases 13 and 14 with submandibular incision 2 mm below the angle of the mandible [Figure 2b], and by careful dissection, the tumor was dissected out [Figure 2c].

Discussion

Epidermoid and dermoid cysts of the oral cavity represent <0.01% of all oral cavity cyst.[3] Dermoid cyst of the floor of the mouth is a dysembryogenetic lesion derived from the entrapment and subsequent growth of epithelial cells, during midline fusion between the first and second brachial arches.[5] They usually present early in life as a symptomatic mass.[5] Cases 1–5 in this report were newborns with a large cyst under the tongue, with marked tongue elevation which obstructs

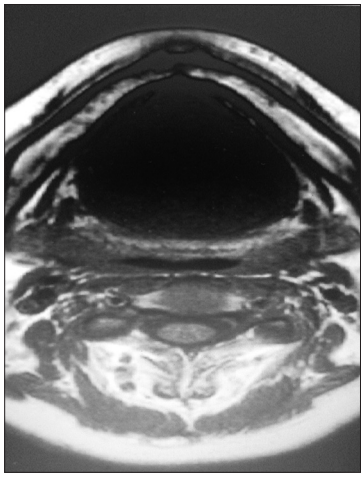

Figure 3: Axial T1-weighted magnetic resonance imaging showing sharply circumscribed cystic mass

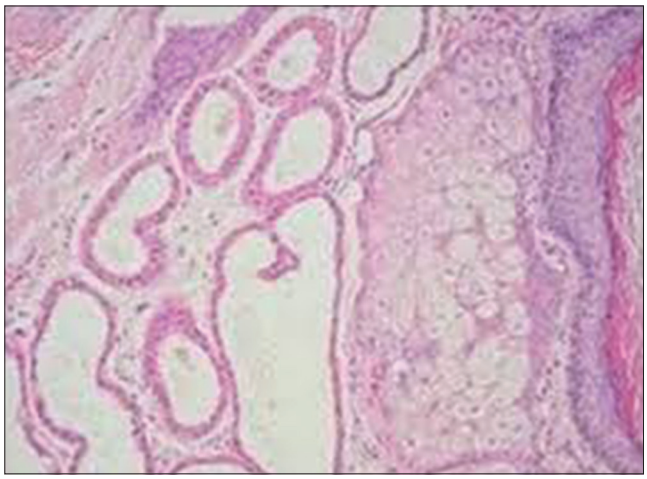

Figure 5: Photomicrograph of the dermoid cyst

feeding and breathing. Needle decompression of the fluid was commenced the same day of presentation to ease feeding and respiration and was done once a week till 3 months when the babies were operated.

Dermoid cyst can be classified into three, considering the location: sublingual, submental, and submandibular cysts.[6] Cases 1–12 were mainly sublingual, while cases 13 and 14 involved the sublingual, submental, and submandibular spaces. Histologically, midline cysts of the floor of the mouth are divided into three types: (1) epidermoid cysts consisting of an epithelial lined wall that may be partly keratinized, (2) dermoid cysts showing evidence of skin appendages such as hair follicles, hair, sweat, and sebaceous glands; and (3) teratomas containing mesodermal elements such as bone, muscle, respiratory and gastrointestinal tissues, and fibrous capsules in addition to skin appendages.[7,8] In our study, cases 8–12 were epidermoid, while cases 1–7, 12, and 13 were dermoid.

Other rare dermoid cysts in the oral cavity are those on the tongue.[9] Patients with intralingual dermoid cyst were described in the English literature.[9,10] Case 12 in this report was intralingual cyst, which protruded the dorsum of the tongue.

The cyst is a painless, slow-growing lesion with doughy consistency and is often soft and well encapsulated without associated lymphadenopathy. None of the cases in our report had any lymphadenopathy. The cyst is often located between geniohyoid and mylohyoid muscles, when it bulges out at the submental and submandibular region; careful dissection needed to be done during surgery to avoid damage to the hypoglossal nerve. Cases 13 and 14 who were adult female patients in our report had the cyst bulges out at the submental and submandibular region [Figure 2]. Complaints of swelling below the tongue, producing difficulty in feeding in neonates and swallowing of solid foods in adults, and altered speech were common to all our patients.

The sudden increase in size of the cyst is postulated to be due to the onset of puberty when there is an increase in the sebum from the sebaceous glands.[11] Cases 13 and 14 aged 23 and 25 years, respectively, gave similar history that there was fast enlargement of the tumor after attaining the age of 18 years.

Differential diagnosis of the dermoid cyst includes lymphoepithelial cyst, thyroglossal cyst, ranula, cystic hygroma, lymphangioma, and soft tissue abscess. Histologically, the contents of the cyst often contain keratin, sebaceous glands, hairs, nails, fat globules and even cartilages may be present.[10] The histology of case 13 showed a thin walled unilocular cystic mass filled with keratin, the cyst lining is composed of squamous epithelium with keratin debris (asterisk) and sebaceous glands with associated hair follicles [Figure 2]. Magnetic resonance imaging (MRI), computed tomography (CT) scan, and ultrasonography are helpful in establishing the differential diagnosis.[12] However, CT and MRI give precise localization to the geniohyoid and mylohyoid muscles and also enable a surgeon to choose the appropriate surgical approach, especially in the case of very large lesions.[9,10] Axial T1- weighted Magnetic Resonance Imaging (MRI) for case 11 showed a sharply circumscribed cystic mass in the floor of the mouth [Figure 4] and a Computed Tomography Scan (CT) of the neck for case 14 showed encapsulated left sided mass with multiple cellular masses at the floor of the mouth [Figure 3]. The treatment is surgical excision of the tumor. The gross surgical specimen of excised tumour of case 2 is shown in [Figure 2c] and Figure 1c shows excised tumour of case 14. The intraoral approach is used for small tumor, while extraoral approach is used for very large sublingual dermoid cysts affecting the submandibular and submental spaces and in cases of infection that could compromise the patient's airways. Cases 1–12 had excision through intraoral approach, while cases 13 and 14 had extraoral approach. Difficult intubation was experienced in most of the patients. Oftentimes, needle decompression of the fluid was done to reduce volume of the cystic fluids to facilitate endotracheal intubation. The major complications of the lesion were recurrent upper respiratory tract infection, anemia, respiratory obstruction, feeding difficulties, and esthetic challenges.[5] All these were managed in our cases. Case 6, male, aged 2 months was cyanosed few minutes after intubation and the surgery was aborted; he was treated for respiratory tract infection and the surgery was successful at another time. Postoperative challenge observed from many of our cases was excessive mucous secretions from the oropharyngeal region and periodic suctioning of the secretions averted respiratory embarrassment. This could be attributed to postoperative edema from the floor of the mouth that resulted in raised tongue and made swallowing difficult.

Recurrence is very rare with complete excision of the lesion; in our study, two cases of recurrence were reported (cases 11 and 14); their first surgeries were done under local anesthesia; it was likely due to incomplete excision. A 5% rate of malignant transformation of the oral dermoid cysts into the teratoid type has been reported in literature.[13,14] None of our cases showed any malignant transformation at histology. Recent advances in the management of sublingual dermoid cyst advocates the inclusion of thyroid scintigraphy in the preoperative diagnosis of the cyst of the floor of the mouth, to assess if thyroid gland is involved.[15,16] Further, surgical enucleation is the only effective treatment for this kind of lesions.[12-16] All our cases had surgical enucleation. This study has shown that sublingual dermoid cyst could be life-threatening, especially in the newborn, because of difficulty in breathing and recurrent respiratory tract infections. Therefore, special care must commence for the newborn, which should involve a pediatric physician. Prognosis of our cases was very good, with no reported case of recurrence.

Conclusion

The results obtained from this study emphasized the need for appropriate surgical treatment to reduce incidence of recurrence. In addition, there is need to treat the lesion urgently because of the difficulties the patients experience in breathing,

swallowing, and periodic respiratory tract infection.

Limitation

The diagnosis of sublingual dermoid cyst in our study was limited to clinical evaluation, fine-needle aspiration biopsy, and histopathology of the cyst wall. However, CT and MRI were not done due to limited resources.

Consent

Consent of patients included in this study was obtained from the time of presentation when the photographs were taken.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Hemaraju N, Nanda SK, Medikeri SB. Sub-lingual dermoid cyst. Indian J Otolaryngol Head Neck Surg 2004;56:218-20.
2. Howell CJ. The sublingual dermoid cyst. Report of five cases and review of the literature. Oral Surg Oral Med Oral Pathol 1985;59:578-80.
3. Etarbi MS, Aishaish H, Khalifa O. Large Sublingual cyst in the floor of the mouth and submental space. Otolaryngo 2017;7:287-91.
4. Di Francesco A, Chiapasco M, Biglioli F, Ancona D. Intraoral approach to large dermoid cysts of the floor of the mouth: A technical note. Int J Oral Maxillofac Surg 1995;24:233-5.
5. Jain H, Singh S, Singh A. Giant sublingual dermoid cyst in floor of the mouth. J Maxillofac Oral Surg 2012;11:235-7.
6. Fomete B, Saheeb BD, Onyebuchi EP, Ogbeifun JO. Dermoid cyst of the oral cavity as seen in a Nigeria tertiary institution. Niger J Surg Res 2013;157:3-6.
7. Menditti D, Laino L, Ferrara N, Baldi A. Dermoid cyst of the mandibula: A case report. Cases J 2008;1:260.
8. Park SW, Lee JJ, Chae SA, Yoo BH, Kim GJ, Lee SY. Congenital epidermoid cyst of the oral cavity: Prenatal diagnosis by sonography. Clin Exp Otorhinolaryngol 2013;6:191-3.
9. Saheeb BD, Osagonona A. Submental dermoid cyst. A case report. OJM 2005;17:24-7.
10. Chukuneke FN, Okwuowulu T. Enbloc enucleation of a large intra-oral dermoid cyst under local anaesthesia: A case report and Review of the literature. J Coll Med 2007;12:8-12.
11. 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 Suppl:S137-9.
12. Akinosi JO. Multiple sublingual dermoid cysts. Br J Oral Surg 1974;12:235-9.
13. Oginni FO, Oladejo T, Braimah OP, Adenekan AT. Sulinqual epidermoid cyst. Surg 2014;4:96-8.
14. Mohta A, Sharma M. Congenital oral cysts in neonates: Report of two cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;102:e36-8.
15. Batsaki JG. Tumours of the Head and Neck Clinical and Pathological Consideration. 2nd ed. Phikdelphia: Williams and Wilkins Company publishers; 1980. p. 226-8.
16. Gleizal A, Abouchebel N, Lebreton F, Beziat JL. Dermoid cyst of the tongue: An association of dermoid cyst with bronchogenic epithelium. J Craniomaxillofac Surg 2006;34:113-6.