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A Giant Plunging Sublingual Dermoid Cysts A Case Report and Review of Anatomy-Surgical Classification

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ABSTRACT

Oral dermoid cysts and ranulae are uncommon, frequently misdiagnosed. Dermoid cysts are subcutaneous located cystic mass that contain epithelium and adnexal structure. Dermoid cysts of the floor of the mouth (DCFOM) are rare comprise only 1.6% to 6.5% of all body dermoid cysts and account for 23% to 34% of head and neck dermoids. There are suspected theories regarding the basis of the pathology and controversy regarding the treatment. The aim of this report is to present a case of a giant dermoid cyst in the floor of mouth that initially misdiagnosed as a ranula and review of its surgical management. A 27 years old woman had a swelling in the sublingual area over 7 months. Ultrasonography (USG) examination revealed a large cystic mass which was 3.1 x 2.5 x 3.1 cm located at sublingual and extended to submental region. Intraoral approach was preferred for marsupialization procedure due to the thinness of the capsule and no recurrence or complaints were detected during follow-up period, but longterm evaluation should be conducted after the primary surgery to evaluate any form of recurrence. When a cystic mass is detected on the floor of oral cavity, we must consider dermoid cysts for differential diagnosis. Surgery is the only treatment. If possible, intraoral approach should be preferred because of its perfect cosmetic results.

1. INTRODUCTION

Dermoid cysts are uncommon, benign congenital tumors of ectodermal origin that can occur in any region of the body [1]. Oral dermoid cysts and ranulae are uncommon, frequently misdiagnosed. A dermoid cyst is defined as a closed, epithelium-lined cavity thats contains dermal adnexal structures. On the other hand, a ranula is described as a mucocele that occurs in the floor of the mouth, arising from the sublingual gland. When this swelling in the floor of the mouth also dissects through the mylohyoid muscle and produces swelling within the neck it is referred to as plunging ranula. Oral dermoid cysts and ranulae both present as painless, soft and compressible lesions, and due to their due to the common anatomical location, may be indistinguishable during clinical examination. However, it is essential to note that the therapeutic options differ considerably for each lesion. Differential diagnosis is the first step towards proper treatment, which is

crucial to prevent recurrence and sequelae. This article presents a large, lateral, dermoid cyst in the floor of mouth, initially misdiagnosed by imaging studies as a ranula. Clinical, radiological and therapeutic aspects of oral dermoid cysts and ranulae will also be discussed to aid in the correct diagnosis and management of these lesions [2].

Dermoid cysts of the floor of the mouth (DCFOM) comprise only 1.6% to 6.5% of all body dermoid cysts and account for 23% to 34% of head and neck dermoids. The floor of the mouth is the second most common site of DCFOM in the head and neck region after the lateral eyebrow [3]. The main treatment modality is surgery via extraoral or intraoral approach according to the size and location of the mass. Dermoid cysts are classified according to their location as submental, sublingual and submandibular. Intraoral approach is chosen in the literature usually for small and sublingual dermoid cysts. Extraoral approach is preferred for large and submandibular cysts [4].

We reported a patient with large dermoid cyst in the floor of the mouth at the same time discusses the pathology, clinical picture, treatment, and we also propose this paper to review comprehensive anatomic classification of the cyst with surgical correlations approach.

2. Case Report

A 27 years old woman was referred to Division of Oral and Maxillofacial Surgery Stomatology Clinic, dr. R. Soedjati Soemodiarjo Hospital, Grobogan with a chief complaint of recurrent swelling in the sublingual and submental area. Initially the swelling appeared under the submental region since 7 months ago, increasingly enlarged and extends into the oral cavity at the midline sublingual and submental. Patients complain of difficulty in swallowing. There is no history of infections, trauma to the neck and no complaints of pain, no shortness of breath, no hoarseness.

On physical examination found general condition is good, compos mentis, cooperative, adequate nutrition and normal vital signs. On inspection there was 4 x 3 x 4 cm mass located at sublingual region. The lesion was fluctuant on palpation but non tender, no lymphadenopathy, and no fistula was noted (Figure 1). Intra oral examination on the inspection found the tongue raised (Figure 2).

Figure 1. Pre-operative photograph showing a mass in submental regio

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Figure 2. Intraoral view of the sublingual bulge in the floor of the mouth

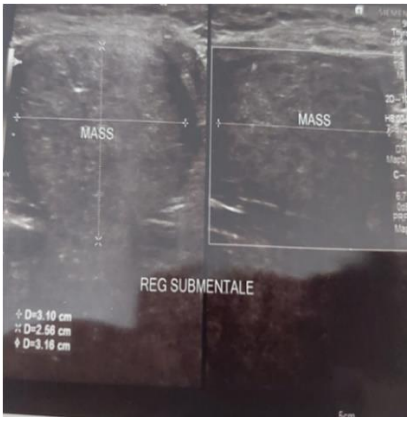

Figure 3. Ultrasonography (USG) shown 3.1 x 2.5 x 3.1 cm cystic lesion, firm boundaries, impressing thin encapsulated in the sublingual and submental region

Panoramic radiographs did not reveal any pathology. Ultrasonography (USG) shown solid lesions, 3.1 x 2.5 x 3.1 cm in size, in the sublingual and submentale region, there are no intra-lesion vascularity. No visible abnormalities of the thyroid gland, no enlarged lymph node, with conclusion cystic-solid lesions in the sublingual dan submental region (Figure 3). Then the patient is diagnosed with plunging ranula differential diagnose dermoid cyst.

After obtaining informed consent, intraoral approach was preferred for the marsupialization of cyst. Folllowing nasotracheal intubation under general anesthesia, approximately two 3 cm oral incisions were used (Figure 4). Macroscopically, the was cheesy keratinous content emerging as a paste (Figure 5). Histopathologically, cystic structure lined with squamous epithelium and its lumen containing keratinous material was noted.

Figure 4. Two sublingual incision for the marsupialization procedure

Figure 5. Cheesy keratinous content

The patient did well postoperatively and no recurrence was noticed during 3-months follow-up period (Figure 6).

Figure 6. Submental and lateral views of the patient postoperatively

3. Discussion

Several pathologic conditions can cause masses in the floor of the mouth. In this case report, dermoid cysts preoperatively diagnosed ranulae are present. This may be due to their similar clinical presentation, as both are painless, soft and compressible lesions, which can cause tongue fullness, with subsequent difficulty with swallowing, speech and breathing. This might have been a contributing factor to the initial misdiagnosis, as dermoid cysts are rare. Imaging studies are important to assist in the differential diagnosis, but may be difficult with dermoid cysts and ranulae, as both appear as thin-walled, cystic lesions [2].

DCFOM are an uncommon entity, hence rarely considered in the differential diagnosis (Table 1). In this case, patients felt swelling symptoms 7 months before. There are 3 theories with regard to the origin of cysts in the floor of the mouth. According to the 1st and most prevalent theory, these cysts originate from embryonic cells of the 1st and 2nd branchial arch entrapped in the mesenchyme of the area during the 3rd/4th week of embryonic life. With regard to the 2nd theory, it explains the pathogenic mechanism of the acquired form. The acquired cysts may be due to the implantation of epithelial cells subsequent to accidental or surgical injury (traumatic causes, iatrogenic antecedents, or an occlusion of a sebaceous gland duct). Lastly, the 3rd theory maintains that these cysts are considered a variation of the cyst of the thyroglossal pore [5].

Table 1. The Differential Diagnosis of DCFOM [6].

Criteria	Differential Diagnosis

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Mucus extravasation phenomena		Mucocele Plunging (cervical) ranula
Embryologic anomalies		Thyroglossal duct cyst Branchial cleft cyst Dermoid cyst Cystic hygroma
Infection		Acute bacterial infection/celullitis of oral floor Sialadentitis of sublingual or submandibular gland Viral lymphadentitis (Epstein-Barr virus, cytomegalovirus;)
Granulomatous disease	Infectious	Mycobacterial disease Cat-scratch disease Actinomycosis Toxoplasmosis Tularemia Histoplasmosis Blastomycosis
	Noninfectious	Sarcoidosis Wegener's granulomatosis Langerhans' cell histiocytosis Crohn's disease
Non-granulomatous inflammatory disease		Kawasaki's disease
Tumor		Benign tumors of salivary glands
	Other benign tumors	Lipoma Fibroma Hemangioma Lymphangioma Angioma Neurofibroma
	Other malignant tumors	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 Burkitt's lymphoma

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 [7]. Depending upon the anatomical location DCFOM are classified as supramylohyoid (intraoral or sublingual), inframylohyoid (cervical), peri- and trans-mylohyoid (both intraoral and cervical) [8] (Table 2) (Figure 9).

Table 2. A Comprehensive Anatomo-Surgical of Cyst of Floor of The Mouth [1].

	Supramylohyoid (Intraoral or Sublingual)	Inframylohyoid (Cervical)	Peri- and Trans-Mylohyoid (Dual Intraoral and Cervical)
Median	1. Suprageniohyoid 2. Infrageniohyoid	4. Submental	6. Submental transmylohoid
Lateral	3. Sublingual space	5. Submandibular space	7. Lateral peri- and transmylohyoid

NOTE. The anatomic location of the cyst determines the most appropriate surgical approach, which is

indicated by different shades of the background, which also corresponds with Figure 9.

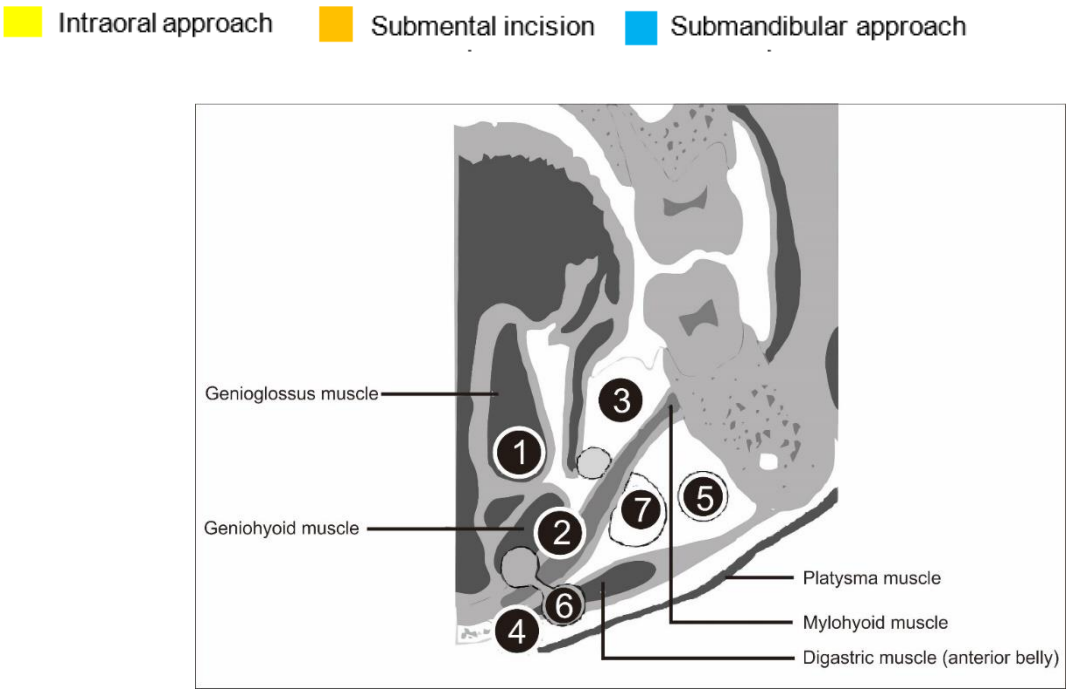

Figure 7. schematic anatomic location of the DCFOM [1].

The most common type of DCFOM is situated deep in, superior to the geniohyoid and mylohyoid muscles, perhaps developing between the fibers of the genioglossus. In the literature this type has been described as supra-geniohyoid cyst (no. 1 in Table 2 and Figure 9). The second type develops between the genial muscles and mylohyoid, for this cyst the term "infra-geniohyoid" is the most accurate description (no. 2 in Table 2 and Figure 9). The third kind of DCFOM in the original classification is an unusual entity and develops between the genioglossus and hyoglossus medially and mylohyoid laterally. It has also been designated as a true lateral cyst, sublingual space cyst is the term for this type because of the definition of the sublingual space between the hyoglossus and mylohyoid muscles (no. 3 in Table 2 and Figure 9). All 3 types of DCFOM in the original classification are located superior to the mylohyoid muscle (ie, supra-mylohyoid) and they could be regarded as intraoral lesions [1], [10].

Imaging studies are important to assist in the differential diagnosis, but may be difficult with dermoid cysts and ranulae, as both appear as thin-walled, cystic lesions, which are hypodense on CT. Nevertheless, each have certain distinguishing characteristics, such the "sack of marbles" appearance that is pathognomonic for dermoid cysts, caused by areas of fat attenuation on CT. It is important to make the correct clinical diagnosis, as therapeutic options differ considerably between ranulae and dermoid cysts. Treatment for ranulae consists of removal of the feeding sublingual gland and/or marsupialization [2]. Due to the history, clinical presentation and radiologic aspects of our case, other diagnosis were ruled out and marsupialization of a ranula was planned as treatment. Intraoperative features of a true cystic lesion with an interior of a yellowish thick creamy material ultimately led to the correct diagnosis of dermoid cyst, later confirmed with histological examination.

The diagnostic work up for suspected dermoid cyst should include ultrasonography, CT scan or MRI allows more precise localization of the lesion in relation to anatomic structures, which helps in choosing the most appropriate surgical approach [5]. The treatment employed in this case was an intraoral approach because it

was above the geniohyoid muscle and an easy access. Some authors have mentioned that an intraoral approach is ideal because it gives a good view of the cyst, an easy access as well as esthetic results. Marsupialization procedure was prefered because of the thinness of the dermoid cyst capsule, so that blunt dissection could not be done to remove the entire cyst. An intraoral approach with a mucosal incision usually through the floorof the mouth is appropriated. By the marsupialization procedure, the submandibular ducts also could be avoided. When the cyst is so large or is extending under the mylohyoid muscle, an extra-oral approach is required, but in this case intra-oral approach was prefered because it provides excellent cosmetic results. Recurrence would be rare although another cyst can arise from cyst fragments or the sinus tract left by surgery. Long-term follow-up with clinical examination and ultranosonography should be conducted after the primary surgery to evaluate any form of recurrence.

4. Conclusion

Dermoid cysts and ranulae are commonly misdiagnosed due to their scarcity and relatively similar clinical presentations. The dermoid cyst and ranulae should be considered one of the differential diagnoses in the patient presented with a cystic mass in the floor of mouth. Surgery is the only treatment. Preoperative imaging is important for localizing the lesion and determining the surgical approach. If possible, intraoral approach should be preferred because of its perfect cosmetic results. Long-term evaluation should be conducted after the primary surgery to evaluate any form of recurrence.

5. References

[1] Christian, B. T., Imad, A. E. N., Omri, E., Michal, L. and Micha, P. (2007). Dermoid Cysts of the Lateral Floor of the Mouth: A Comprehensive Anatomo-Surgical Classification of Cysts of the Oral Floor. J Oral Maxillofac Surg., 65, pp.327-332.

[2] Martins, S., Neto and Soares, Y. R. B, (2019). Large dermoid cyst of the floor of the mouth. An emphasis on differential diagnosis. Rev Esp Cir Oral Maxilofac., 41(3), pp.149-152.

[3] Sedat, A., Mehmet, G. D., Necdet, D., Sefer, S. and Sukran, S. K. (2016). A Giant Plunging Sublingual Dermoid Cyst Excised by Intraoral Approach. J. Maxillofac. Oral Surg., 15(2), pp.277–280.

[4] Mohan, B., Nandita, S., Dharnappa, P., Ram, M. and Ramdas, N. (2014). Epidermoid Cyst Of The Floor Of The Mouth. Natl J Maxillofac Surg., 5(1), pp.79-83.

[5] Paul, E. G., William, C. F., Edward, L. and Leonard, B. K. (2013). Floor-of-Mouth Dermoid Cysts: Report of 3 Variants and a Suggested Change in Terminology. J Oral Maxillofac Surg., 71, pp.1034-1041.

[6] Hanpon, K. and Chanisa, P. (2020). A large sublingual epidermoid cyst with parapharyngeal space extension: A case report. International Journal of Surgery Case Reports. 72, pp.233-236

[7] Sahoo, N. K., Choudary, A. K., Srivinas, V., and Tomar, K. (2015). Dermoid cysts of maxillofacial region. Medical Journal Armed Forces India, 71, pp.389-394.

[8] Kishore, H. R. and Reshma, P. R. (2021). Sublingual dermoid: an uncommon presentation and review of literature. Int. J. Otorhinolaryngol Head Neck Surg., 7, pp.154-8.

[9] Sahoo, N. K. and Choudhary, A. K. (2012). Dermoid Cyst of Floor of Mouth: A Case Report. National Journal of Medical and Dental Research, 1(1), pp.24-30.

[10] Kondo, S., Kamatani, T., Saito, Y., Yoshihama, Y., Shirota, T. and Shintani, S. (2015). True lateral dermoid cyst of the submandibular region: A case report. J. Oral Maxillofac Surg. Med. Pathol., 27(1), pp.74-7.

[11] Aydın, S., Demir, M. G. and Demir, N. (2016). A Giant Plunging Sublingual Dermoid Cyst Excised by Intraoral Approach. J. Maxillofac. Oral Surg., 15, pp.277–280.