



Mucocele Masquerading as Lymphangioma - A Rare Case Report

Ravina Sorout, Puneeta Vohra, Osheen Batra, Pratibha Chauhan, Archna Nagpal and Ashima Bali Behl

Oral Medicine and Radiology, SGT University, Gurugram
Corresponding author: Email id: soroutravina@gmail.com

ABSTRACT

Things are not always what they seem. They can mislead the clinician to give a different diagnosis. Tongue being a delicate, vascular and sensitive site, where mucocele is never considered, we consider the swelling as the most common diagnosis as lymphangioma. The most common cystic lesions of oral cavity are mucocele but ventral surface of tongue which is originating from anterior salivary glands are rare to find Mucocele. Only 1.9%-10.3% of the cases of mucocele have been reported involving the Blandin and Nuhn glands. No encapsulation is seen over the glands of Blandin and Nuhn and they are overlapped to the underlying muscle tissues, they are manipulated differently as compared to other oral mucoceles. Mucocele should taken into consideration if it occur on the tongue too as tongue is an high vascularity. The physical examination of the patient, evolution of the lesion and the imaging are well described and the non – invasive management, histopathological features and the follow up of the patient are also included. A minor trauma can give rise to mucocele after few days and can remain unchanged for months if treatment is not given. The size can increase in diameter from a few millimetres to few centimetres. Increase or decrease in the size of the mucocele can be seen on the basis of rupture and mucin production. A benign lesion known as a mucocele is characterised by the extravasation or retention of mucus from small salivary glands in submucosal tissue. The lower lip and the floor of the mouth are where mucoceles are most likely to develop; the buccal mucosa and tongue are the least common sites. These kinds of lesions are typically brought on by trauma and lip-biting behaviours. Although mucocele is a common oral mucosal lesion, the ventral aspect of the tongue hardly ever experiences this condition. The mucoceles most frequently occur on the lower lip (77.9%), lingual mucoceles (24.2%), and the floor of the mouth (15.6%). We present a 4-year-old child with a lesion on the ventral surface of tongue whose clinical characteristics suggested an initial diagnosis of lymphangioma but was later confirmed to be a mucocele. Mucocele pose a diagnostic and therapeutic challenge if present in the vicinity of vascular structures like tongue and floor of mouth. The purpose of this article is to aid in a step-by-step approach to clinical characteristics, differential diagnosis and therapeutic interventions for mucocele in a daily clinical practice We should always keep our mind open while giving diagnosis. And secondly we should not rely on surgical procedures. Before thinking for surgical procedures, alternative therapy should be taken into consideration. Location and age of patients should always be keep in mind as children are very elusive to cooperate with. They should be handled with proper precautions and carefulness.

Keywords- Mucocele, Follow-up, lymphangioma, tongue, aspiration

Received 09.10.2022

Revised 09.11.2022

Accepted 13.12.2022

INTRODUCTION

An accurate diagnosis in dentistry is essential for providing an effective clinical treatment, especially when it comes to oral mucosal lesions [1]. A cavity filled with mucus, which is a secretory by product of salivary glands, is known as an oral mucocele and is one of the most prevalent benign lesions of the oral mucosa. Mucus extravasation, which is typically thought to be of traumatic origin, and mucus retention, which is caused by obstruction of the duct of a minor or auxiliary salivary gland, is the two reasons underlying the formation of these lesions. A mucocele is described as a collection of mucus in a connective tissue cavity that is not epithelium-lined. Mucous escape reaction/mucus retention phenomenon, mucocele, ranula, and mucus retention cyst are some of the names given to this lesion, with mucocele and mucus retention phenomena being the most commonly used terms [3]. The lesions are seen in the upper submucosa or just under the mucous membrane (superficial mucocele) (classical mucocele). It could be found as a fluctuant nodule deep inside the connective tissue or as a fluid-filled vesicle or blister in the surface mucosa. It is possible for inspissated mucin to spontaneously drain, especially in superficial lesions, followed by recurrence. Long-lasting lesions may have fibrosis on their surface [4].

These lesions are known as ranulas [2] when they are found on the floor of the mouth. Because the lesions on the floor of the mouth mimic the protruding underbelly of a frog, the term "ranula" is derived from the

Latin word "rana," which means "Belly of frog." The excretory duct rupture, followed by saliva extravasation and deposition into the surrounding tissue, is suggested to be the cause of ranula formation [5]. These lesions grow slowly, are reddish-purple in hue, and range in diameter from one to five cm². Lower lip (77.9%) is where the mucocoeles are located. 6, as well as the floor of the mouth (15.6%). 6, anterior ventral tongue (5.0% from Blandin-Nuhn glands). 7, the palate (1.3%), the retromolar pad (0.5%), and the buccal mucosa (4.8%). [7].

The upper lip rarely develops mucocoeles [7]. A very small percentage of oral mucocoeles—2–8%—involves the ventral surface of the tongue. Sporadic occurrences of these lesions have been documented in international literature. Infants under the age of one only experience a low incidence of it (2.7%). In children, mucocoeles of the extravasation type are more common than those of the retention type. Because of their clinical features and location, mucocoeles on the ventral surface of the tongue that are produced by the Blandin Nuhn glands, mixed glands (both serous and mucous), are thought to be quite unusual because they could be mistaken for vascular lesions, squamous papillomas, or pyogenic granulomas. The lingual muscles surrounding the Blandin Nuhn glands are located close to the medial line, close to the tip of the ventral portion of the tongue. These glands form a horseshoe-shaped mass that extends laterally and behind the medial line, draining through four to six ducts that start close to the lingual frenulum⁶. It is difficult to diagnose and treat mucocoele since the lesion has no preference for one sex and affects people of all ages, with the highest frequency reported to be in the second and third decades and seldom detected in babies [2].

Oral mucocoeles rarely result in serious issues. Depending on the size and location of the mucocoeles⁸, discomfort, difficulties with speaking, mastication, swallowing, and external swelling may ensue. Due to the clinical similarities between mucocoele and many other swellings and ulcerative lesions of the oral cavity, it is important to differentiate them carefully using Fine Needle Aspiration Cytology, chemical analysis (which will reveal protein content and high amylase), Computed Tomography Scanning, and Magnetic Resonance Imaging [9]. For the evaluation of individuals with salivary gland nodules and enlargement, fine-needle aspiration is a helpful diagnostic procedure, particularly when the differential diagnosis of angiomatous lesion is involved.

Age and proximity to vital structure is an important consideration while deciding the treatment plan. Here we report an interesting unusual case whose clinical appearance suggested an initial diagnosis of lymphangioma but later was confirmed to be a mucocoele of the ventral surface of tongue and emphasis upon meticulous non- interventional treatment and follow up. .

CASE PRESENTATION

A four year-old female patient reported to the Department of Oral Medicine and Radiology at SGT Dental College , Gurugram with a chief complaint of swelling underneath the tongue with a duration of two weeks duration. Her father disclosed that the swelling was insidious in onset, gradually enlarged to attain the present size . The swelling was painless and not associated with any type of trauma from a foreign body or sharp tooth. It was not impeding speech, chewing or swallowing with no history of similar swelling in the same area in the past. There was no fluid oozing out of the swelling. Her medical and family history were non-contributory. It was patient's first exposure to dentist.

On general examination patient was normal. Extraoral examination revealed no abnormality. There was no evidence of enlarged palpable lymph nodes. The intraoral examination showed presence of a swelling in the anterior ventral aspect of the tongue. On inspection the lesion presented as a well defined, sessile, bluish pink solitary oval shaped swelling on the right side of ventro- lateral surface of tongue of approximately 1x 2 cm in size extending 2cm posterior to the tip of tongue to the 1cm anterior to root of the tongue. Surrounding area appeared normal. On palpation all the inspectory findings are confirmed with respect to size, shape and location. The swelling was fluctuant, non tender and soft in consistency with a smooth surface. There was no induration. The dorsum and lateral aspects of tongue appeared normal. There was no restriction in movements of the tongue.

Based on the clinical characteristics i.e. site of the lesion, color of the lesion and consistency ; an initial diagnostic hypothesis of oral lymphangioma was suggested for the lesion with differential diagnosis of mucocoele, minor salivary gland tumour (The glands of Blandin–Nuhn as they are localized in the ventral part of the tongue, next to the apex in the lingual median plane), irritational fibroma (excluded as the lesion was not firm), lipoma (excluded as slip sign was negative) , and neurofibroma.

A chairside investigation of FNAC (fine needle aspiration cytology) was performed after obtaining normal investigatory reports which included complete blood count, bleeding and clotting time and random blood glucose level . A written informed consent from the parents was taken prior to aspiration. A 27-gauge needle was introduced into the lesion for aspiration. A 0.8mL of clear aspirate was obtained. It was sent for cytological investigation to the Department of Oral Pathology and Microbiology for further

evaluation which confirmed the diagnosis as a mucocele . It revealed inflammatory cells and macrophages in a mucinous background suggestive of mucocele .

Surgery is the mainstay treatment for such lesions but considering the young age of the patient and location of the lesion we opted for aspiration of fluid from the lesion at every recall until the lesion completely regressed. Corticosteroids could have been injected in the lesion after eliminating the aspirate but considering the age steroid injection was advocated. Patient reported after 1 week for 1st follow up. Needle was introduced into the lesion and a 0.6mL mucous fluid was aspirated. As a result of this procedure, the lesion regressed in size. On the second follow up after a week, same treatment was repeated and the patient unveiled a marked reduction in size of the swelling. This time aspirate was 0.5 mL in quantity. On the third follow up after 7 days, similar procedure was performed and 0.4mL mucous fluid was drawn. Patient reported after 2 weeks , with complete regression of the lesion on the fourth follow up visit. Patient has been kept on a regular follow up. During this period, no reoccurrence of the lesion has been observed. Patient has been reviewed meticulously every month for more than 9 months. Routine follow up of the lesion size is crucial for managing such cases in children. During review, prognosis was excellent, and no recurrence was found. This conservative approach to the swelling presented a better option compared to surgical excision or any other therapy.

DISCUSSION

The tongue is a special essential organ that has a significant impact on oral health. It is a mucous sac-related organ that is highly vascular and muscular. Compared to the ventral surface of the tongue, this sac is smooth. With thin webbed projections (plica fimbriata) emerging from the lingual frenum, the ventral side of the body is more vascular than the dorsal one [10]. The vascular supply to the mouth floor is varied and rich. These can occasionally be linked together into one trunk. The deep lingual artery, which supplies the tongue, emerges from the lingual artery as it runs anteriorly, and the sublingual artery, which supplies the floor of the mouth [11, 12].

Hence it is very important to formulate a proper diagnosis before surgically manipulating lesions in these areas. Several cases have been reported that mimicked the clinical appearance of lymphangioma but had different definite histological diagnoses such as oral granular cell tumors , dermoid cyst [1].

To the best of our knowledge, there are no reports of lesions with clinical characteristics of lymphangioma and final diagnosis of mucocele of the ventral surface of tongue.

Lymphangiomas are rare lesions, appearing in less than one percent of biopsies performed in routine pediatric dental practice. The incidence of lymphangiomas has been reported to range from 1.2 to 2.8 per 1000 newborns. The vast majority of lymphangiomas are congenital and seen at birth or evident up to two years of age. The most common sites of occurrence are the submandibular area (close to the parotid gland) and the anterior two thirds of the tongue. They may also occur less frequently on the palate, gingiva, cheek mucosa, and alveolar ridges and are rarely seen in the lips and ventral surface of tongue¹. Localized lymphangiomas occur as nodular tumours elevated above the surface of the tongue [13]. Generally , oral lymphangiomas are vesicles resembling the appearance of tapioca pudding or frog eggs and a size varying from one to five cm in diameter. Differential diagnoses of lymphangiomas from other lesions include haemangiomas, dermoid cysts, thyroglossal duct cysts, pyogenic granulomas, and granular cell tumors¹. Surgical excision is the best alternative for most lymphangiomas presenting localized growth¹⁴. With varying degrees of effectiveness, simple drainage, aspiration, and excision have been tried. With limited success rates and a variety of side effects, radiation, laser therapy, and instillation of sclerosing drugs such hot water, quinine, sodium morrhuate, urethane, and nitromin have also been tried. Bleomycin and OK-432, two sclerosing drugs, have recently received attention for the treatment of paediatric lymphangiomas [15].

The child in the current case was 4 years old, and the lesion had only been present for a few weeks. The clinical characteristics were consistent with those of oral lymphangiomas that have been previously described. On the other hand, mucoceles are lesions resulting from the rupture of salivary gland ducts and consequent accumulation of mucus into the soft tissue around this gland. They are most frequently found in the oral cavity of children and adolescents [16].

Clinically, they are nodular, exophytic, and asymptomatic lesions with bluish color or a color similar to the surrounding mucosa and a size rarely exceeding 1.5 cm in diameter. The diagnosis of mucocele is based on the clinical appearance of the lesion . There is usually a history of trauma to the region and variations in lesion size. The Blandin-Nuhn glands are situated in the lingual median plane, ventral to the apex of the tongue. The development of a mucocele at this location is uncommon, and because the Blandin-Nuhn glands are not encapsulated and immediately overlie the muscular tissues, their manipulation is frequently unique from that of other oral mucoceles [16]. Lesions that are known to cause tongue swelling should be part of the differential diagnosis in a case like the one that was reported.

Salivary glands, adipose tissue, blood vessels, nerves, and connective tissue are all present in the tongue, making any of these tissues susceptible to pathosis.

Fine Needle aspiration might be performed to rule out a vascular cause or a cystic mucoepidermoid tumour as the aspirate of Mucoepidermoid Carcinoma is stringy mucin, as their clinical appearance may be similar [17]. No matter where they are, the pseudo cystic mucoceles appear as soft, painless swellings that range in hue from deep blue to normal pink. The colour variation is influenced by the extent of the lesion, its proximity to the surface, and the tissue's flexibility above it. When the surrounding tissue is less elastic or if it is farther away from the surface mucosa, the bluish colour may be more subdued, but it will become obvious after the initial mucosal incision [18].

There are three different strategies for managing mucoceles, according to Baumash. Before primary closure, the tiny lesion can be thoroughly excised, being sure to remove any marginal glands and tissue related with the salivary glands. Unroofing is the best way to treat large mucoceles (marsupialization). The third operation, which is typically used for moderate-sized lesions, is dissecting the mucocele and the supporting mucous glands. Additionally, cryosurgery, laser ablation, micro marsupialization, and marsupialization can also be used to treat bigger lesions [19]. An alternative to surgery is the use of intralesional corticosteroid injections since they are effective anti-inflammatory drugs and also have sclerosing properties that cause the dilated salivary ducts to constrict [20].

Marsupialization and micro marsupialization are indicated for very young children who are unable to cooperate with a surgical removal or for larger lesions. Other treatment options, such as homeopathy, injection of sclerosing agents, and watchful waiting, have also been described [19]. As seen in our case, it is important that, even for lesions with typical clinical characteristics, additional clinical investigations (e.g., palpation, needle aspiration) and complementary examinations, such as biopsy and histopathological analysis, are always recommended to confirm the diagnosis and establish the adequate treatment. So, meticulous follow up of every patient is very important to avoid recurrence.

CONCLUSION

The majority of mucocele are benign and self-limiting in nature, and are initially identified based on clinical signs before being definitively identified based on the results of a histological analysis. The majority of the reported material demonstrated how the lesion developed after trauma and repetitive lip-biting. Complete excision has historically been the simplest course of treatment, and recurrence has been linked to incomplete removal of the lesion, although it is laborious for young patients. The management/approach, anaesthesia, and sedation of paediatric patients may necessitate additional steps. Therefore, we chose the simpler, less intrusive procedure of aspirating fluid from the lesion. For more than nine months, our patient faithfully submitted monthly reports. The prognosis was excellent at the review, and the absence of a recurrence was valued.

Declaration of patient consent

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

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. de Carvalho FK, Pinheiro TN, da Silva RA, de Queiroz AM, da Silva LA, Nelson-Filho P. (2015). Lymphangioma of the lower lip mimicking a mucocele in children. *Journal of Dentistry for Children.*;82(2):116-9.
2. Bhargava N, Agarwal P, Sharma N, Agrawal M, Sidiq M, Narain P. An unusual presentation of oral mucocele in infant and its review. *Case reports in dentistry.* 2014 Jan 1;2014.
3. Urvashi S, Bhandary S, Rao K, Shetty P. Oral mucocele or mucous escape reaction an obstructive disorder: A report of 2 cases, *IJDR* 2017; 2(1): 15-17
4. More CB, Bhavsar K, Varma S, Tailor M. Oral mucocele: a clinical and histopathological study. *Journal of oral and maxillofacial pathology: JOMFP.* 2014 Sep;18(Suppl 1):S72.
5. Suresh BV, Vora SK. Huge plunging ranula. *Journal of maxillofacial and oral surgery.* 2012 Dec 1;11(4):487-90.
6. Garcia Leon N, Marrugo Pardo GE. Mucocele of the glands of blandin nuhn: a case report. *Colombia médica.* 2013 Jan;44(1):46-7.
7. Neville BW, Damm DD, Allen CM, Chi AC. *Oral and maxillofacial pathology.* Elsevier Health Sciences; 2015 May 13.

8. Rodríguez H, Parra RD, Cuestas G, Cambi J, Passali D. Congenital mucocele of the tongue: a case report and review of the literature. *Turk J Pediatr.* 2014 Mar 1;56(2):199-202.
9. Sinha R, Sarkar S, Khaitan T, Kabiraj A, Maji A. Nonsurgical management of oral mucocele by intralesional corticosteroid therapy. *International Journal of Dentistry.* 2016 Oct 16;2016.
10. Abdul Aziz MA, Yussif NM. Nonneoplastic tongue swellings of lymphatic and lymphocytic origin: Three Case Reports. *Case reports in dentistry.* 2016 Nov 20;2016.
11. Peñarrocha-Diago M, Balaguer-Martí JC, Peñarrocha-Oltra D, Bagán J, Peñarrocha-Diago M, Flanagan D. Floor of the mouth hemorrhage subsequent to dental implant placement in the anterior mandible. *Clinical, cosmetic and investigational dentistry.* 2019;11:235.
12. Ferneini E, Gady J, Lieblich SE. Floor of mouth hematoma after posterior mandibular implants placement: a case report. *Journal of oral and maxillofacial surgery.* 2009 Jul 1;67(7):1552-4.
13. Goswami M, Singh S, Gokkulakrishnan S, Singh A. Lymphangioma of the tongue. *National Journal of Maxillofacial Surgery.* 2011 Jan;2(1):86.
14. Bhayya H, Pavani D, Tejasvi MA, Geetha P. Oral lymphangioma: A rare case report. *Contemporary clinical dentistry.* 2015 Oct;6(4):584.
15. Sanlialp I, Karnak I, Tanyel FC, Senocak ME, Büyükpamukçu N. Sclerotherapy for lymphangioma in children. *International journal of pediatric otorhinolaryngology.* 2003 Jul 1;67(7):795-800.
16. Adachi P, Soubhia AM, Horikawa FK, Shinohara EH. Mucocele of the glands of Blandin-Nuhn—clinical, pathological, and therapeutical aspects. *Oral and maxillofacial surgery.* 2011 Mar 1;15(1):11-3.
17. Guimarães MS, Hebling J, Filho VA, Santos LL, Vita TM, Costa CA. Extravasation mucocele involving the ventral surface of the tongue (glands of Blandin-Nuhn). *International Journal of Paediatric Dentistry.* 2006 Nov;16(6):435-9.
18. Baurmash HD. (2003), Mucoceles and ranulas. *Journal of Oral and Maxillofacial Surgery.* ;61(3):369-78.
19. Joshi SR, Pendyala GS, Choudhari S, Kalburge J. (2012). Mucocele of the glands of Blandin-Nuhn in children: a clinical, histopathologic, and retrospective study. *North American journal of Medical Sciences;*4(9):379.

CITATION OF THIS ARTICLE

Ravina Sorout, Puneeta Vohra , Osheen Batra, Pratibha Chauhan, Archana Nagpal, Ashima Bali Behl: Mucocele Masquerading As Lymphangioma - A Rare Case Report. *Bull. Env.Pharmacol. Life Sci., Spl Issue [5]: 2022: 231-235.*