

Oral Mucocele: Presentation at a Rare Site with Review

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Abstract

Mucocele is the most common disorder of minor salivary glands, especially extravasation type. The Mucocele has a bluish translucent color and common sites of occurrence are lip followed by tongue, floor of mouth and the buccal mucosa. It is the most common minor salivary gland lesion in children and young adults. It is usually associated with history of trauma that leads to severance of salivary gland duct. The extravasation Mucocele is a pseudocyst which contains the pool of spilled mucin and surrounded by granulation tissue which further undergoes fibrosis. Diagnosis and management of Mucocele is challenging task. Various treatment options include marsupialization, surgical excision, dissection, laser ablation, cryosurgery, electrocautery, intra-lesional steroid injections and irradiation. Surgical excision is an appropriate treatment modality with least recurrence rate and good prognosis. Here, we report a case of an oral Mucocele which is present at a rare site of the oral cavity i.e. at the lower buccal vestibule, which to our knowledge is second case reported in review till date.

Keyword: Extravasation; Minor salivary gland, Mucocele, Vestibule.

Introduction:

The term Mucocele is used to define the sub-epithelial accumulation of mucous secreted from salivary glands and their ducts in the mucosa of the oral cavity.¹ Mucocele is a common lesion of the oral mucosa that results from an alteration of minor salivary glands due to mucous accumulation causing a limited swelling.² They present as fluctuant, bluish, non-tender sub mucosal swelling with a normal overlying mucosa. Mucocele can occur as extravasation and retention phenomenon. Extravasation Mucocele results from severance of salivary gland duct and the consequent spillage of mucin into the soft tissues around the gland.³ Retention Mucocele occur due to decrease or absence of glandular secretion produced by the blockage of the salivary gland ducts.⁴ Clinically, there is no difference between extravasation and retention type. When Mucocele is located in the floor of the mouth it appears as “Belly of a frog” and is called as a ‘ranula’.

These lesions are devoid of epithelial lining and are also termed as superficial or classical Mucocele. Superficial Mucocele is located under the mucous membrane and classical Mucocele is seen in the upper submucosa.^{4,5}

Case Report:

A male patient aged 21 years, reported to the Department of Oral Medicine and Radiology, Bharati Vidyapeeth Deemed Dental College and Hospital with the chief complaint of an intraoral swelling in lower left buccal vestibular region of the mouth for past 7 days. History revealed that the swelling was initially smaller in size and gradually increased to the present size. The patient did not give any relevant medical history, nor did he give any history of trauma, parafunctional habits, or history of difficulty or pain during chewing or speaking. On extraoral examination, the face of the patient was bilaterally

symmetrical with no remarkable findings.

On Intraoral inspection, a solitary well defined swelling was present on lower left buccal vestibular region in relation to tooth number 33, 34 and 35. The swelling extended antero-posteriorly from mesial surface of 33 to the distal surface of 35; medio-laterally it extended from muco-gingival junction of 33, 34 to 2 cm away towards the vestibule onto the buccal mucosa. The swelling measured approximately 3 x 2 cm in size and was oval in shape, it had smooth and shiny surface with a slight bluish hue. On palpation, all the inspectory findings were confirmed. The swelling was soft in consistency, non-tender, and fluctuant, non-reducible and compressible. It was fixed to underlying tissue (**Figure No. 1**). On correlating the clinical history with the clinical examination a provisional diagnosis of Mucocele on lower left buccal vestibule with the differential diagnosis of fibroma, lipoma and haemangioma was made. A further investigation like diascopy was found to be negative (**Figure No. 2**). Hence lesions of vascular origin were ruled out; therefore excisional biopsy was advised and performed (**Figure No. 3**). The histopathological finding revealed areas of severance of duct, eosinophilic coagulum and mucous pooling, features of which are suggestive of "Extravasation type of Mucocele" (**Figure No. 4**).

Correlating the clinical findings with the histopathological report a final diagnosis of Extravasation Mucocele in lower left buccal vestibular region was made. Patient was examined after 1 week, post-operatively (**Figure No. 5**) and was recalled after a period of 1 month wherein no recurrence was noted and now the patient is on regular periodic follow up.

Discussion:

Mucocele is a benign cystic lesion of the oral cavity that has been ranked seventeenth most common salivary gland lesion seen in the oral cavity. It is the second most common benign soft tissue tumor occurring in the oral cavity.⁵ The incidence of Mucocele is generally high, 2.5 per 1000 patients, frequently occurring in the second decade of life and rarely among children and infants under one year of age.⁶ Mucocele is a painless, slow growing and fluctuant swelling that occurs due to mucus extravasation or retention of mucous material arising

from the salivary gland. It usually occurs as an isolated lesion, although more than one may be present at a time as in case of superficial Mucocele that can present as single or multiple blister.⁷

Mucocele mostly present as a doomed shape swelling with intact epithelium over it, however in this reported case it was a single and oval shaped swelling. Mucocele occupies 70% of the salivary gland cysts (6% of all parotid lesions) and they usually arise from minor salivary glands and are non neoplastic localized lesions of the duct system.⁸ It is a rare occurrence in the major salivary glands (parotid, sublingual and submandibular). The Armed Forces Institute of Pathology collected data on 2339 cases of Mucocele and found that 33.0% occurred on the lower lip, 7.7% on the buccal mucosa, 6.3% on the floor of the mouth, 6.1% on the tongue and only 0.4% on the upper lip.⁹ Mucocele of the minor salivary glands are rarely larger than 1.5 cm in diameter. However; in our case it was measured to be 3x2 cm in size.

Various literatures on Mucocele reveal that it is equally seen in men and women, whereas other studies reported that Mucocele has a slight female prevalence of about 1.3:1¹⁰ but the reported case was a male patient and the lesion was compatible with extravasation cyst.

Mucocele can be traumatic or non-traumatic in origin. Parafunctional habits such as lip biting being the most common contributory factor for occurrence of an oral Mucocele. The literature reveals that most of the patients give a history of spontaneous development (71.4%), followed by lip biting (25.7%) and trauma (2.9%). This reported case in the lower left buccal vestibular region is a rare occurrence and was a spontaneous development.¹¹ Yamasoba et al highlighted two crucial etiological factors in Mucocele formation; traumatism and obstruction of salivary gland ducts. But in our case there was neither trauma nor any obstruction of salivary ducts. Mucous is produced exclusively by the minor salivary glands and is also the most important substance secreted by the major sublingual salivary glands.

The diagnosis of Mucocele is based principally on the clinical examination. It usually presents as a bluish, soft, transparent cystic swelling which frequently resolves spontaneously.



Figure No. 1: A Well-defined Swelling in the Left Buccal Vestibular Region



Figure No. 2: Negative Diascopy



Figure No. 3: Excisional Biopsy

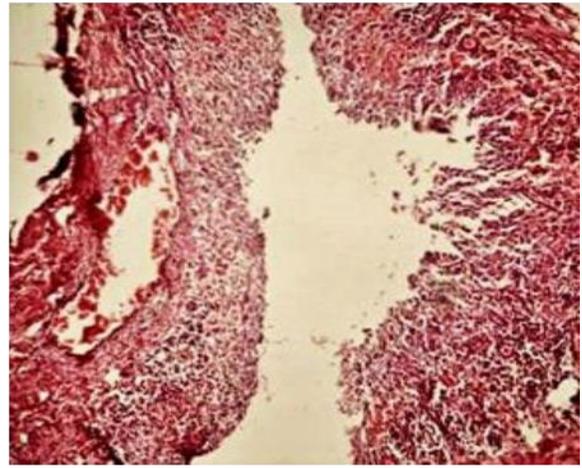


Figure No. 4: Shows Eosinophilic coagulum and Mucous Pooling



Figure No. 5: Post-Operative Shows Healing

The duration of the lesion is not constant; it varies from a few days to 3 years. The blue color is caused by vascular congestion, and cyanosis of the tissue above and accumulation of the fluid below. Coloration can also depend on the size of the lesion, proximity to the upper surface and elasticity of the superficial tissue. In our case, diascopy procedure was performed, which was found to be negative. For further investigations, Fine needle aspiration cytology could be done, that may show mucus with inflammatory cells and may aid in the diagnosis.¹²

It is important to give differential diagnosis even though Mucocele is a benign and asymptomatic lesion but clinically it resembles with many other benign or malignant swellings of oral cavity. Mucocele is always associated with history of trauma that is why it is important to elicit proper history from the patient. The clinical appearance of Mucocele is pathognomonic such as location, variation in size, bluish color and the consistency. Clinically, diascopy can be helpful for a correct diagnosis.¹³ but in our case there was no history of trauma and the site of lesion was also rare.

Mucocele is mostly a self-limiting condition and it often ruptures and leaves slightly painful erosions that usually heal within few days. There are three possible surgical approaches to management of Mucocele of the lips, cheeks and palate; complete excision, Marsupialization and Dissecting.^{14, 15}

The Lesion can be excised completely or treated with marsupialization because excision can injure the vital structures such as mental nerve. The Cryosurgery is an effective method in treatment of Mucocele. The procedure includes a gas expansion cryoprobe with a 10mm diameter round tip. The application should be for 30-seconds freeze at -81°C temperature followed by an approximately 1 minute defrost.¹⁶ Steroids play an important role in treatment of Mucocele, a single intra lesional steroid injection, preceded by aspiration of the cyst fluid can be done. It causes the pseudocyst wall to collapse and triggers a severe inflammatory reaction of the wall that results in marked fibrosis.¹⁷

In the above case the authors reported a spontaneously developed solitary Mucocele in a male patient that developed at lower vestibule which is a

very rare site of its occurrence in the oral cavity. To the best of our knowledge and with literature review this may be the second clinical report of a Mucocele that has developed in an unusual site of the oral cavity, without any history of trauma or duct obstruction.

Conclusion:

Mucocele is the most common benign self-limiting condition of the oral mucosa. One of the most common sites of its occurrence is the lower lip mostly due to trauma or lip biting habit. Our case report is till date, a second such reported case with review in literature where an extravasation type of Mucocele located at an unusual site of the oral cavity, i.e. in vestibular region of the lower jaw was reported and was not associated with history of trauma or any parafunctional habits. Majority of such cases can be diagnosed clinically; however biopsy is required sometimes to rule out any other lesions. On correlating the clinical history with the clinical examination, a provisional diagnosis of Mucocele on lower left buccal vestibule with the differential diagnosis of fibroma, lipoma and haemangioma was made. For investigation diascopy procedure was found to be negative; hence lesions of vascular origins were ruled out. Therefore excisional biopsy was advised and performed.

Management of Mucocele becomes challenging because of high possibility of recurrence, however if no spontaneous regression occurs surgical excision with dissection of surrounding and contributing minor salivary gland can lead to clinical success without recurrence and a better prognosis.

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