



Twin Mucoceles Involving the Glands of Blandin and Nuhn on the Ventral Surface of the Tongue

[PP: 05-09]

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Abstract:

A mucocele is a fluid-filled mass caused by local trauma, severing a salivary duct and causing extravasation of mucin into surrounding oral mucosa. Inflammatory response and granulation tissue formation may forge a contained pool of mucin, resulting in a typical mucocele presentation. A higher incidence of mucocele is reported in the lower lip; however, it can arise in any other region that contains minor salivary glands like the tongue, palate, and buccal mucosa. Mucoceles involving Blandin and Nuhn minor salivary glands on the ventral surface of the tongue are considered rare entities. Literature indicates Blandin and Nuhn mucoceles comprise 9.9% of the oral mucoceles. These mucoceles may present difficulty in eating, speaking, and can develop a psychological disturbance in an individual. The purpose of this paper is to present an uncommon clinical entity of twin mucoceles (superficial mucocele in combination with an almost equal size deep mucocele) involving the glands of Blandin and Nuhn on the ventral surface of the tongue in a 30-year-old individual. They were managed with excisional biopsy, and the histopathological investigation confirmed the clinically determined diagnosis of twin mucoceles.

Keywords: *Mucocele, Oral Mucosa, Granulation Tissue, Histopathological Investigation***ARTICLE INFO** The paper received on: **18/3/2020** Accepted after review on: **12/5/2020** Published on: **07/06/2020**

Cite this article as:

Alzahrani, F., Bhat, A., Zameer, M. & Syed, W. (2020). Twin Mucoceles Involving the Glands of Blandin and Nuhn on the Ventral Surface of the Tongue. *Case Reports in Odontology*. 7(1), 05-09.

1. Introduction

A mucocele is a fluid-filled mass caused by local trauma, severing a salivary duct and causing extravasation of mucin into surrounding oral mucosa. Inflammatory response and granulation tissue formation may forge a contained pool of mucin, resulting in a typical mucocele presentation. [1] Clinically, the mucocele appears as a well-defined, fluctuant, painless swelling of

the mucosa. The majority of reported mucus cysts are smaller than 1cm in diameter; however, the size can vary up to several centimeters in rare instances. There is a distinct difference in the presentation between the superficial and deep cystic lesions in terms of color. The superficial ones present a bluish to transparent hue, whereas deep lesions show normal mucosal coloration, and if they are associated with bleeding, it may impart a vascular



appearance. [2] A higher incidence of mucocele reported in the lower lip; however, it can arise in any other region that contains minor salivary glands like the tongue, palate, and buccal mucosa. [3, 4]

Mucocele involving Blandin and Nuhn minor salivary glands of the tongue has been considered uncommon. Literature indicates Blandin and Nuhn mucoceles comprise 9.9% of the oral mucoceles. [5, 6] As suggested, increased pressure caused by mucous plugs in the intraepithelial squamous cell lined portion of the duct might cause the duct to burst or rupture, which thus can create superficial mucoceles.[7]

2. Case Report:

A 30-year-old male reported to the dental department of Armed Forces Hospital, Jazan, KSA. The chief complaint was swelling in the undersurface of the tongue from the past 3months. Medical history was non-contributory with no history of trauma. The extra-oral examination showed no gross abnormality. Intraoral examination revealed a protuberant, oval-shaped, non-ulcerated, mucosal color, sessile swelling on the anterior ventral tongue measuring 1.7x1.5cms. (Figure 1&2) There was no visible pulsation. On palpation, the swelling was fluctuant, non-tender, and gave a negative diascopy test. Fine-needle-aspiration-biopsy (FNAB) revealed viscous salivary aspirate or mucus-like material, made our suspicion more towards mucocele. The provisional diagnosis was made as mucocele involving the glands of Blandin and Nuhn. The treatment plan was to excise mucocele with associated minor salivary glands.

Local anesthesia, 2% lidocaine with 1:80000 adrenaline, was administered. A longitudinal combined with the circumferential incision was made through

the mucosa, followed by blunt dissection to remove the sessile growth in toto without disrupting the integrity of the lesion wall. While dissection, another similar swelling measuring 1.7cmx1.6cm was revealed attached to the superficial one. (Figure 3) Both the swellings were cautiously excised in-toto without losing the content along with the associated minor salivary gland. (Figure 4) The biopsy sample was immediately fixed in 10% formalin and sent for histopathologic evaluation. The wound was closed with interrupted sutures using 3.0 vicryl suture material. (Figure 5) Hemostasis was achieved, and the patient was discharged on the same day. Histopathological investigation reported the tissue fragment is covered by stratified squamous epithelium with underlying cyst lined by granulation tissue. It contained inflammatory cells (macrophages and giant cells) consistent with the mucous cyst's extravasation phenomenon. Follow up revealed satisfactory healing.

3. Discussion:

Blandin and Nuhn minor salivary glands are closely-packed mixed seromucous minor salivary glands located on both sides of the midline in a horseshoe pattern. They are embedded 12 to 25mm deep within the musculature of the anterior ventral tongue. Mucoceles on the ventral surface of the tongue usually arise from the glands of Blandin and Nuhn. Mucocele can be present below the mucosal layer, in submucosal layer or in deep corium known as superficial, classical or deep mucocele respectively.[4, 8]

Two types of Blandin and Nuhn mucoceles have been described based on the clinical presentation. 1) A submucosal lesion of 2-4cm in size covered with integral mucosa, usually of a long-term development without clinical symptoms. 2) A protuberant



pedunculated swelling presents painful sensibility with a history of local traumas.[4] However, It is stated that the clinical presentations of these mucoceles are similar to squamous papilloma, vascular lesions, polyp, pyogenic granulomas based on the degree of vascularization and acinar atrophy.[9] The diagnosis of the mucocele is primarily clinical. So, the patient's anamnesis should be gathered correctly along with the clinical data of the lesion, such as site, size, shape, appearance, and history of oral trauma. FNAB can be a helpful adjunct in the diagnosis of the mucocele. In particular cases, the diagnosis may require ultrasonography, computed-tomography (CT), or magnetic-resonance-imaging (MRI) to better evaluate form, diameter, and relative position of the lesion with other anatomical landmarks. Histopathological evaluation of the excised mucocele is necessary to re-confirm the diagnosis.

Histopathologically, the mucoceles involving salivary glands have been categorized into extravasation and retention type: 1) The extravasation type results from damage to the excretory duct that leads to the pooling of mucus into the adjacent connective tissue. 2) The retention type is a true cyst lined by an epithelium. The acinus and the ducts of the salivary glands present alterations, dilatation of the interlobular and intralobular ducts with an accumulation of mucus, and decomposition of the cells of the individual acinus, results in mucus formation in the area.[4] A vesicular extravasation subtype of oral mucocele with subepithelial mucin deposition has been described and termed as a superficial mucocele. These lesions are single or multiple, often short-lived, that may burst or rupture, leaving an ulcerated surface.[10] Increased pressure caused by mucous plugs

in the duct might cause it to rupture and create superficial mucoceles. [7]

In the present case, one superficial protuberant sessile mucocele is found on the anterior-ventral surface of the tongue. FNAB was performed, and it revealed mucus-like material. In the surgical approach, we gave a combined longitudinal and circumferential incision to remove the superficial mucocele, and bluntly dissected out the deeper mucocele, which was found attached to the superficial mucocele. Simultaneously, the associated glandular tissue was also removed. The histopathologic investigation has confirmed our diagnosis of extravasation type of mucocele involving the glands of Blandin and Nuhn. The patient was followed up and had satisfactory healing with no significant complaints. Management of large lesions may require alternative approaches[11–14] that include cryosurgery, intralesional steroid, OK-432 injection, micro-marsupialization, marsupialization of the mucocele, and laser ablation.

4. Conclusion:

Mucoceleles involving Blandin-Nuh minor salivary glands of the tongue are considered to be rare entities. These mucoceles may present difficulty in eating, speaking, and can cause a psychological disturbance in some patients. A clear understanding of the mucocele regarding its clinical presentation and the use of FNAB as a diagnostic adjunct can help a clinician arrive at a correct diagnosis. The histopathologic investigation is essential to confirm the clinically determined diagnosis of mucocele. The excision of mucocele requires high levels of blunt dissection surgical skills as any puncture to the generally thin and fragile mucosal layer of the cyst may complicate its complete

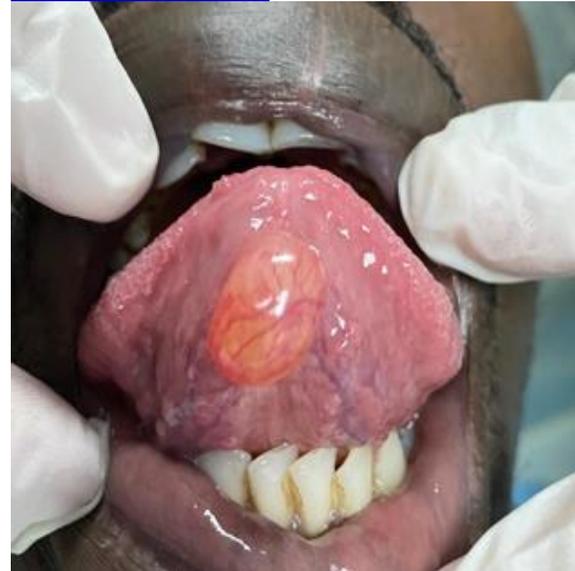


excision and would increase the chances of recurrence.

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Figures & Legends



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Figure 1&2: Mucocele on anterior ventral tongue



Figure 4: Twin Mucocele post excision along with the content



Figure 3: Twin mucocele attached to each other on anterior ventral tongue



Figure 5: Suturing done post excision

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