



Mucocele of Blandin and Nuhn: Review of Literature and A Novel Treatment Approach

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Abstract

Mucocele of Blandin and Nuhn occur on the ventral surface of the tongue. Various treatment modalities, which include surgery, laser ablation, cryosurgery, sclerotherapy, micro-marsupialization, have been used for its management. The use of intralesional sclerosing agent or corticosteroid is a good alternative non-surgical procedure that can be performed in a short span of time, economical, esthetically more beneficial than surgical methods. This article presents a case report with a review of the literature regarding Blandin and Nuhn mucocele that was treated with an unconventional mode of treatment in the present case.

Keywords: Blandin and Nuhn, intralesional steroid, mucocele, tongue

Introduction

A mucocele (muco – mucus and coele – cavity) is a cavity filled with mucus, originating from either major or minor salivary glands; is one of the most common benign soft-tissue cysts of the oral cavity.[1,2] The site more often affected by a mucocele is the lower lip, followed by the tongue.[1] Mucocele of Blandin and Nuhn occur on the ventral surface of the tongue and clinically characterized by painless, translucent, and fluctuant nodule, which is usually asymptomatic.[3] Etiologic factors include trauma to the tongue, or it may occur due to dilation of the duct secondary to its obstruction. Various treatment modalities, including surgery, laser ablation, cryosurgery, sclerotherapy, micro-marsupialization, have been used for its management[3]. However, these procedures have several disadvantages such as trauma, pain, lip disfigurement, damage to adjacent vital structures, and ducts

leading to the development of satellite lesions and can also be expensive to the patient. An intralesional sclerosing agent or corticosteroid is a good alternative non-surgical procedure that can be performed in a short span of time, economical, esthetically more beneficial than surgical methods. This article presents a case report with a review of the literature regarding Blandin and Nuhn mucocele that was treated with an unconventional mode of treatment in the present case. To the best of our knowledge, intralesional corticosteroid was never used alone in the treatment of mucocele of Blandin and Nuhn and this paper gives a new dimension for the treatment of such cases especially in pediatric population.

Case Presentation

A 5-year-old female patient was referred to the Department of Pedodontics with a chief complaint of



swelling on the undersurface of the tongue. The duration of swelling was approximately 6 months. There was no pain associated with the swelling. The parent and the patient could not recollect any history of trauma. The swelling had gradually increased in size and interfered with speech and mastication. There was no history of bleeding or any discharge from the swelling. On clinical examination, a large oval swelling soft in consistency, fluctuant and translucent, measuring about 12 mmx16 mm, was present on the left ventral aspect of the tongue (Fig. 1). The overlying mucosa was bluish in color at the central area and margins were same as that of normal mucosa. Rest of the medical and dental history was non-contributory. The differential diagnosis included mucocele, pyogenic granuloma, vascular lesions, lipoma, polyp and squamous papilloma. The clinical appearance, its location on the ventral surface, fluctuation and fluid-filled consistency were pathognomonic for the diagnosis of mucocele of glands of Blandin and Nuhn. The patient was subjected to ultrasonographic examination which revealed a solitary uniformly hypoechoic image without any calcification. On aspiration, mucous was recovered from the swelling. Blood investigation revealed the parameters within the normal limits. Considering the large size of the mucocele that could create a large defect, surgical excision was deferred. Conservative treatment with intralesional steroid injection was planned. The area was disinfected with 0.1% povidone-iodine; surface local anesthesia was applied and mucus aspirated with the help of an 18-gauge needle and a syringe. Then 1 mL of beta-methasone (4 mg/1 mL) was slowly injected by insulin syringe (0.3x8 mm size, 31 gauges) to prevent any leakage, less discomfort, and pain (Fig. 2). The solution was gradually injected into the base of the lesion and adjacent

to the periphery of the lesion. The patient was examined after a period of 7, 14, and 21 days to evaluate the response of the lesion towards treatment and consequently given the 2nd, 3rd, and 4th injections. The size of the lesion was measured by means of a dental caliper in mm in weekly evaluation. The size of the lesion was significantly reduced after 4 visits (Figs. 3, 4, and 5). The patient was evaluated after 2 months, and a complete resolution was observed. After 3 and 6 months of follow-up, there was no history or sign of recurrence or local discomfort.

Discussion

The tongue has 3 types of minor salivary glands - Blandin-Nuhn glands, Von Ebner glands, and Weber glands.[2,4,5] The Von Ebner glands are pure serous and



Figure 2. Intralesional injection using insulin syringe

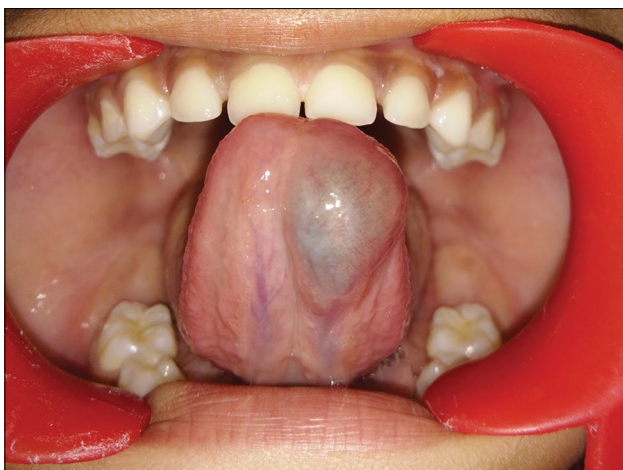


Figure 1. Blandin and Nuhn mucocele on left ventral surface of tongue

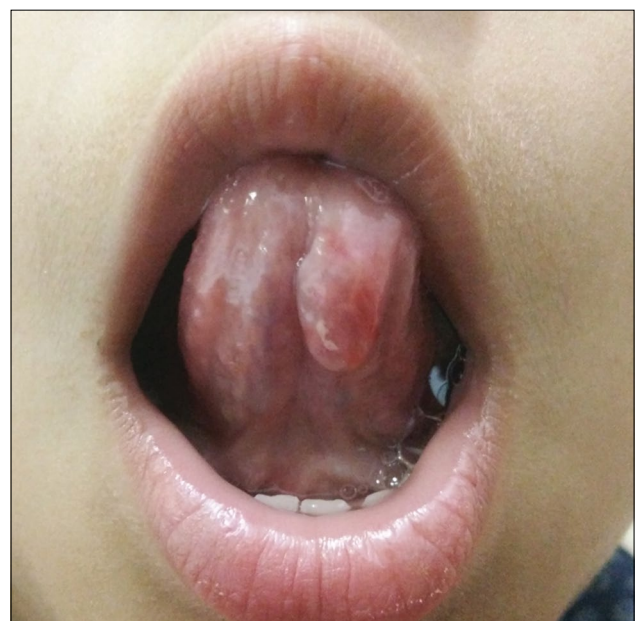


Figure 3. Regression in size of lesion on 2nd appointment



Figure 4. Regression in size of lesion on 3rd appointment



Figure 5. Almost complete regression of lesion after 4th intrale-sional injection

located near circumvallate and foliate papillae on the dorsal surface of the tongue. Weber glands open near peritonsillar space and are pure mucous glands. Blandin-Nuhn glands are a group of mixed seromucous salivary glands on ventral surface of tongue with ducts opening medial to the plica fimbriate, laterally to the lingual frenum. The gland is approximately 8 mm in width and 12–25 mm in depth.[2,6] Seromucous acini are present anteriorly and mucous acini capped seromucous demilunes are present posteriorly in the gland.[6]

Incidence

Mucocele of the oral cavity are quite common, occurring with a frequency of 2.4 cases per 1000 people, however, mucoceles of the glands of Blandin–Nuhn are considered to be rare[4]. In 1970 Heimansohn first reported a case of mucocele of Blandin and Nuhn in his 14-year old daughter.[7] The prevalence, according to different authors, varied between 2.25%-18.3%.[2,4,6,7] These mucoceles appear with higher frequency in children than in adults because younger people are more likely to experience trauma that induces mucin spillage.[4] Females are more commonly affected than males.[4]

Etiology

The most common etiological factor in these mucoceles is local traumatic injury of tongue. In some cases, mucosal inflammation of the gland duct results in blockage, dilatation, and rupture of the duct with sub-epithelial spillage of fluid. Immunological factors may be the cause in few cases.[2,5]

Rupture of the salivary gland duct results in mucus spillage into the connective tissue and such mucoceles are categorized as false cyst or pseudocyst or extravasation mucocele (92%) because there is no epithelial lining surrounding the mucin.1 On the other hand, mucus retention cyst (8%), lined by ductal epithelium, is caused by an epithelial cell proliferation of a partially obstructed salivary duct.[4,8]

Types

Two common types: a submucosal variety with no symptoms and of a long-term development covered with integral mucosa; protuberant variety, with a pedunculated base, and frequently associated local trauma history and pain.[6,7]

Clinical features

The Blandin-Nuhn mucocele occurs on the ventral surface of the tongue, in the midline, or laterally, fast-growing at the beginning and may vary in size afterward. They may be categorized as superficial mucocele (directly under the mucosa), classic mucocele (in the upper submucosa), or deep mucocele (in the lower corium).[2,5] Their clinical presentation depends upon the degree of keratinization of the overlying mucosa and their location depth.[5] Superficial lesions are more translucent and bluish in color, whereas the deeper lesions have a normal mucosal color, lack the vesicular appearance, and are more nodular.

The presence of mucinous contents gave them fluctuance. The diameter may range from a few millimeters

to a few centimeters. An episodic decrease and increase in size, corresponding to rupture and subsequent mucin production, may be observed.^[1] These lesions are often asymptomatic, however, they can cause interference with speech and mastication and sometimes suffocation or dysphagia as they grow in size.^[9]

Differential diagnosis

The differential diagnosis should include lesions known to cause swelling of the tongue. The tongue contains adipose tissue, salivary glands, connective tissue, blood vessels and nerves, so pathosis of any of these tissues is possible.^[1] Sarada et al stated that the mucoceles of the Blandin–Nuhn glands are clinically similar to pyogenic granulomas, vascular lesions, polyp, and squamous papilloma.^[2]

Palpation can be helpful tool in the diagnosis of mucocele, which are fluctuant and movable because of its mucinous contents, whereas lipomas and tumors of minor salivary glands are non- fluctuant.^[5] Needle aspiration might be performed to rule out a vascular cause or a mucoepidermoid tumor. Usually blue in color, these blanch under digital pressure, whereas other pigmented lesions such as nevi, hematomas, and melanomas lack blanching.^[1] Their clinical appearance is unique, with longitudinal swelling in the ventral surface of the tongue parallel to the lingual frenulum.^[9] Paraclinical exams (ultrasound, CT, MRI, or needle aspiration) can confirm the diagnosis but are not always necessary, as the diagnosis is clinically obvious.^[9] It is therefore important to assess the location of the lesion, the presence of trauma, any rapid changes, an increase or decrease in the lesion size, a bluish color, and fluid consistency and recovery of mucus with aspiration.^[10]

Histopathological characteristics

They exhibit mainly mucus extravasation phenomenon with no epithelium lining the mucin collection. Mucocele walls consist of granulation tissue, with a variable number of fibroblasts, leukocytes, and phagocytes.^[6] The luminal fluid from extravasation mucoceles contains many matrix metalloproteinases, especially MMP-9, MMP-2, and MMP-1, compared with saliva from Wharton's duct.^[1] These proteolytic enzymes produced by macrophages and fibroblasts are involved in the remodeling of the connective tissue and may contribute to the development of the lesion.

Treatment

When mucoceles are present on the tongue, they can be easily traumatized and become a strong source of irrita-

tion and annoyance to the patient. Therefore, early treatment with surgical excision including the associated salivary gland tissue, is usually the treatment of choice. Surgery can be performed under local anesthesia if the patient is compliant and if the lesion is small; in other cases, general anesthesia is necessary. A longitudinal incision parallel to the frenulum and 5 mm away from it is made in the ventral surface of the tongue, and the mucus is discharged.^[9] Large mucoceles are best treated with an unroofing procedure (marsupialization). In addition, larger lesion may also be managed by cryosurgery, laser ablation, micro-marsupialization, electrocautery, and steroid injections.^[1,3,4] Some authors have suggested using rubber base impression material or alginate to fill the cystic cavity, improving the visual access for surgical excision.^[10] Micro-marsupialization is an alternative technique, which involves passing a thick silk suture through the internal part of the lesion along its widest diameter.^[11] The introduction of a suture, according to the literature, causes epithelialization around the suture, establishing new excretory ducts between the surface and the underlying salivary gland tissue and leading to the disappearance of the lesion.^[11] Recurrence was seen in some cases even after the surgical excision because they cannot be removed in toto like other mucoceles; moreover the glands of Blandin and Nuhn are not encapsulated and are directly overlapped by the muscle tissues making their manipulation different from other oral mucoceles.^[5,9]

Although surgery is widely used, it may lead to disfigurement and damage to adjacent ducts and development of satellite lesions.^[8] The use of steroid injections, is an alternate to surgery. The advantages of this technique include simplicity, minimum trauma, and relative lack of pain. However, literature regarding the usage of intralesional corticosteroids is meager. Luiz et al^[12] reported a case treated with topical corticosteroid in controlling the recurrences and attributed it to the high-potency vasoconstriction and anti-inflammatory properties of the clobetasol propionate, reducing the inflammatory process involved in the pathogenesis of the mucoceles. Mortazavi et al^[13] and Javali et al^[11] had attempted combined intralesional dexamethasone and micro-marsupialization. Baharvand et al^[8] and Sinha et al^[14] had done interventional studies to evaluate the efficacy of intralesional corticosteroid injection as a nonsurgical treatment procedure in oral mucoceles. Most of the patients treated were cured completely or showed decrease in size of their lesions. No deformity or infection was observed. Corticosteroids act as the most potent anti-inflammatory agent inhibiting the

expression of multiple inflammatory genes (encoding cytokines, chemokines, adhesion molecules, inflammatory enzymes, receptors, and proteins) and may also increase the transcription of genes coding for anti-inflammatory proteins including lipocortin-1, interleukin-1, and interleukin-10 receptor antagonist. [14] They also act like a sclerosing agent causing shrinkage of the dilated salivary ducts.[11,14]. The treatment cause minimal pain and usually no local anesthesia is required. The time of procedure is short, so children can well tolerate it. In regard to cost-effectiveness, steroid injection therapy is economically and esthetically more advantageous than surgery, cryotherapy, or laser ablation.[8]. To the best of our knowledge, intralesional corticosteroid was never used alone in the treatment of mucocele of Blandin and Nuhn, and this paper gives a new dimension for the treatment of such cases, especially in the pediatric population.

Conclusion

Blandin–Nuhn mucoceles are quite uncommon, and the present paper reviewed the main features of these lesions. Intralesional corticosteroid therapy is a good alternative nonsurgical procedure which is a relatively simple, repeatable, cost effective, and potentially curative method easily acceptable by the patient. This treatment protocol can be considered the first choice or substitute for surgery to treat large oral mucoceles, especially in children.

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References

- 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). *Int J Paediatr Dent* 2006;16(6):435-439 doi:10.1111/j.1365-263X.2006.00759.x
- Sarada B, Reddy CH, A K P, Kurra S. Solitary nodular lesion of tongue- a rare entity. *J Clin Diagn Res* 2014;8(2):256-258 doi:10.7860/JCDR/2014/8073.4075
- Jose SC, Abraham KK, Khosla E. Blandin and Nuhn mucocele in a pediatric patient. *J Indian Soc Pedod Prev Dent* 2018;36(3):315-318 doi:10.4103/JISPPD.JISPPD_10_18
- Joshi SR, Pendyala GS, Choudhari S, Kalburge J. Mucocele of the glands of blandin-nuhn in children: a clinical, histopathologic, and retrospective study. *N Am J Med Sci* 2012;4(9):379-383 doi:10.4103/1947-2714.100977
- Khalekar YJ, Sande AR, Zope A, Suragimath A. Mucocele of tongue: A rare case report. *J Indian Acad Oral Med Radiol* 2016;28:462-464
- Adachi P, Soubhia AM, Horikawa FK, Shinohara EH. Mucocele of the glands of Blandin-Nuhn--clinical, pathological, and therapeutical aspects. *Oral Maxillofac Surg* 2011;15(1):11-13 doi:10.1007/s10006-010-0221-1
- Banu V, Kanneppady SK, Anusha RL. Mucocele of the glands of Blandin – Nuhn. *International Dentistry-African Edition* 2013;3(6):26-29
- Baharvand M, Sabounchi S, Mortazavi H. Treatment of Labial Mucocele by Intralesional Injection of Dexamethasone : Case Series. *JDMT* 2014;3(3):128-133
- Graillon N, Mage C, Le Roux MK, Scemama U, Chossegros C, Foletti JM. Mucoceles of the anterior ventral surface of the tongue and the glands of Blandin-Nuhn: 5 cases. *J Stomatol Oral Maxillofac Surg* 2019;120(6):509-512 doi:10.1016/j.jor- mas.2019.04.005
- Kumaresan R, Karthikeyan P, Mohammed F, Thapasum Fairozekhan A. A novel technique for the management of blandin-nuhn mucocele: a case report. *Int J Clin Pediatr Dent* 2013;6(3):201-204 doi:10.5005/jp-journals-10005-1219
- Javali R, Bhagwati B, Bhagwati S. Micromarsupialization combined with intralesional corticosteroids to treat mucocele: A case report. *J Indian Acad Oral Med Radiol* 2016;28:44-47
- Luiz AC, Hiraki KR, Lemos CA Jr, Hirota SK, Migliari DA. Treatment of painful and recurrent oral mucoceles with a high-potency topical corticosteroid: a case report. *J Oral Maxillofac Surg* 2008;66(8):1737-1739 doi:10.1016/j.joms.2008.01.050
- Mortazavi H, Baharvand M, Alirezaei S, Noor-Mohammadi R. Combination therapy in a large lower lip mucocele: A non-invasive recommended technique. *Dent Hypotheses* 2014;5:127-129
- Sinha R, Sarkar S, Khaitan T, Kabiraj A, Maji A. Nonsurgical management of oral mucocele by intralesional corticosteroid therapy. *Int J Dent* 2016;2016:2896748 doi:10.1155/2016/2896748