Extravasation Mucocele Involving The Glands Of Blandin-Nuhn Present On The Ventral Surface Of The Tongue- A Case Report.

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Mucocele are benign soft tissue masses which occurs from the retention or extravasation of mucus in the surrounding tissues of the lamina propria. They are located most frequently in the varying locations on the oral mucosal surfaces overlying accessory minor salivary glands; most frequently located on the lower lip and buccal mucosa; the occurrence in the ventral aspect of the tongue is rarely seen. This article reports a case of an uncommon presentation of mucocele with anterior lingual salivary glands (glands of Blandin–Nuhn) which are embedded within the musculature of anterior tongue ventrum.

Keywords:
Blandin-Nuhn gland, extravasation, mucocele, ventral surface of tongue

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INTRODUCTION:
Cystic swellings associated with the minor salivary glands are referred to as mucocele. Mucocele (mucocrinum & coele- cavity) by definition are the cavities filled with mucous and they are the most common soft tissue masses found in the oral cavity. They represent the 15th most common oral mucosal lesion with a prevalence of 2.4 cases per 1000 people. It is usually found as an asymptomatic, benign, mucous-containing cystic swelling caused by pooling of saliva at the site of injured minor salivary glands. Based on histology, mucoceles are classified as extravasation or retention types. The extravasation mucocele consists of extravasated mucus in connective tissue and the retention mucocele represents mucus retained by an epithelial lining. Although minor salivary glands are found in most parts of the oral cavity except the gingiva, the prevalence of mucocele varies depending on the specific location. Mucoceles involving the anterior lingual salivary glands (glands of Blandin-Nuhn), which are embedded within the musculature of anterior tongue ventrum are rarely reported. They present as soft, painless, fluctuant and movable swellings with a normal or bluish colour. The size may range from a few millimetres to a few centimetres and the colour depends on size of lesion and its proximity to mucosal surface and elasticity. On clinical presentation the mucoceles of the glands of Blandin-Nuhn are exophytic and may resemble pyogenic granuloma, polyps or squamous papillomatous growth.

CASE REPORT
A 9-year-old female patient accompanied by parents reported to the Department of Oral and Maxillofacial surgery with the chief complaint of swelling on the ventral aspect of the tongue since two month. She described the swelling as painless, had gradually increased in size associated with speech and eating difficulties. She denied any trauma to the area as well as bleeding or discharge from the swelling. There was no other relevant past medical or dental history.

On extra oral examination, there was no visible asymmetry of the face and no cervical lymphadenopathy. On intraoral examination, a protruding solitary, bluish, non-ulcerated, oval shaped swelling, measuring about approximately 2.5 X 2 cm in size was found on the left ventral surface of the tongue along the lingual frenulum, 1 cm posterior to the tip of the tongue, not crossing the midline. The mass was located superficially but firmly attached to ventral surface of tongue, freely mobile on all planes and was soft, fluctuant, smooth, non-tender, non-compressible, and the mucosa overlying the lesion was tense. Based on the clinical examination and based on peculiar location, a provisional diagnosis of mucocele involving Blandin-Nuhn glands and excisional biopsy under local anaesthesia as treatment plan was considered. Patient’s parents were explained about the condition, as well as the treatment options and its benefit, risk and complications. The routine blood investigations were done which were within the normal limits.

After informed consent taken from the patient’s relatives (parents), surgical excision of the lesion was done under local anaesthesia with 2% lignocaine with all aseptic precautions. Lingual nerve block was given on left side and local infiltration was given on the right side of the tongue. A tongue stitch was taken and the tongue was retracted. The incision was taken around the swelling ventrally and the lesion was excised by dissecting down till the muscle layer. Complete removal of the lesion along its entire periphery with associated gland was done to avoid recurrence. The wound was closed primarily with 3-0 vicryl sutures. The specimen was immediately fixed in 10% formalin and sent for histological evaluation. The procedure was uneventful.
and post procedure patient was comfortable. Patient was regularly followed up for six months and there was no recurrence. The histopathology report described the specimen as presence of connective tissue stroma with extravasation of mucin without proper demarcation and presence of few salivary glands along with fibroblasts and chronic inflammatory cell infiltration at the periphery of the cystic lining, all of which is suggestive of extravasation type mucocele.

*Fig 1.* Oral examination of mucocele of the glands of Blandin-Nuhn presenting as a solitary, bluish, non-ulcerated oval shaped swelling measuring approximately 2.5 X 2 cm, on the left ventral surface of the tongue along the lingual frenulum, 1cm posterior to the tip of the tongue, no crossing the midline.

*Fig 2.* Specimen following complete removal of the lesion along its entire periphery and associated underlying gland

*Fig 3.* Primary closure with 3-0 vicryl sutures.
DISCUSSION
There are three distinct sets of minor salivary glands located in the human tongue namely, the glands of Von-Ebner, the glands of Weber, and the glands of Blandin-Nuhn also called as anterior lingual gland.[10] The glands of von Ebner opens at the base of the grooves that surrounds the circumvallate papillae and also presents at the base of the clefts between the foliate papillae. The glands of Weber are pure mucous glands, found along the lateral border of the tongue and opens into the crypts of the lingual tonsil. The glands of Blandin-Nuhn are mixed mucus and serous glands; presents as horseshoe-shaped masses on both sides of the midline, surrounded by musculature of the ventral surface of the anterior tongue.[2,10] They are neither lobulated nor encapsulated and each gland measures about 1-8mm wide and 12-25mm deep. They consist of several small independent glands and these glands drain by means of 5-6 small ducts which opens near the lingual frenum.

Heimansohn was the first one to reported a case of mucocele of Blandin-Nuhn in a 14-year old female in 1970.[11] After 1970, more such cases were reported. Harrison in his study found that out of 400 mucocele cases, only nine had origin from the tongue salivary glands.[1] Jinbu et al who did similar studies found that mucoceles involving Blandin-Nuhn gland comprised of 9.9% of all oral mucoceles studied. Another study done by Nico et al reported 8.3% mucocele of the glands of Blandin-Nuhn among 36 pediatric patients.[12] De Camargo Moraes et al. in their series of 312 cases of oral mucoceles, 48 cases were reported as mucocele of gland of Blandin-Nuhn; stated that mucocele of the ventral surface of tongue involving Blandin-Nuhn should not be considered as rare.[13] The incidence of mucoceles in these glands were found to be high in young and female patients by a ratio of 4:1.[11-13]

Trauma to the ventral surface of the tongue is considered as the main etiological factor of mucocele of Blandin and Nuhn and are usually located on the tip of the ventral surface of the tongue, rarely these lesions are presented lateral to the midline.[2] The lesions are usually asymptomatic and relatively small in size ranging from 2mm in diameter to 20mm; sometimes they can grow relatively large enough to cause feeding difficulties in babies or difficulty in speech and mastication in adults.

In the present case report, mucocele presented as oval shaped swelling on the left ventral surface of the tongue along the lingual frenulum, 1cm posterior to the tip of the tongue, not crossing the midline, measuring about approximately 2.5 X 2 cm which is largest in size till date. The clinical presentation of these lesions depends upon their depth, adjacent soft tissues and the degree of keratinisation of the overlying mucosa. The superficial lesions of the gland presented as raised, bluish, translucent soft tissue swelling with tense overlying mucosa as it appeared in our case (Fig-1), whereas the deeper lesions are mostly nodular, lack the vesicular appearance, and have a normal mucosal colour.[7] The differential diagnosis considered in the present case were oral hemangiomia, irritation fibroma, oral lymphangioma, gingival cyst, benign or malignant salivary gland neoplasm, soft tissue abscess,[12-13] Palpation can be helpful for a clinical and differential diagnosis as fluctuation is absent in lipomas and tumors of minor salivary glands, whereas it is present in cysts, mucoceles, abscess and hemangioma. Fine needle aspiration cytology can be performed for confirmation of the diagnosis. As mentioned in our case report, the swelling was pale blue, fluctuant with a smooth surface with no ulcerations and discharge, hence was diagnosed as mucocele of Blandin-Nuhn gland. They are often asymptomatic, but as they grow in size, they may interfere with speech and mastication. There is strong possibility of trauma to the mucocele considering its location and hence also can become a site of discomfort to the patient.

Thus, excisional biopsy of the lesion is the treatment of choice and same was planned in our case. The surgical excision includes the removal of entire lesion along with evacuation of its contents and the underlying mucous glands. According to Surgerman et al. and Baurmash, the management of moderate to large Blandin-Nuhn mucoceles includes completely un-roofing of the lesion along its entire periphery to visualize and remove all of the glands.[7,14] Management also includes marsupialization,[14] cryosurgery,[15] laser ablation,[9] and micro marsupialization.[7,14] The management of small
Blandin-Nuhn mucoceles includes complete excision and primary closure, with rapid and uneventful healing.[7] Other techniques include a method where the cystic cavity is filled with rubber impression material like alginate pre-surgically which allows good visual access for surgical excision by delineating the entire lesion.[16]. The underlying glands of Blandin-Nuhn which are present deep in the musculature also needs to be removed completely; if they are left behind, they may result in recurrence.[2,7,14]. Prior clinical evaluation and preoperative awareness of the surgical anatomy of the glands of Blandin-Nuhn helps to minimize the chances of recurrences of the lesion. The above-mentioned case was managed with complete removal of the lesion along its entire periphery with associated gland under local anaesthesia and the specimen was sent for histopathological examination. Mucocele involving the anterior lingual gland are often histologically classified as extravasation or retention type. The cases of retention mucoceles are less frequent, particularly seen as a cystic cavity in elderly patients and are often long standing. This is histopathologically well defined, with the presence of an epithelial wall covered with a row of cuboidal or squamous cells produced from the excretory duct of the salivary glands.[2,7] The case of extravasation mucoceles are more frequently seen involving the anterior lingual gland.[12-14] In our case, histopathology report reveals extravasation of mucin without proper demarcation and presence of few salivary glands along with fibroblasts and chronic inflammatory cell infiltration at the periphery of the cystic lining; all of which is suggestive of extravasation type mucocele of the anterior lingual salivary gland. In our case the patient was followed up till six months and there were no signs suggestive of recurrence.

**CONCLUSION**

- Mucoceles of the glands of Blandin-Nuhn should not be considered as rare.
- Diagnosis is based mainly on clinical inspection and palpations which varies according to their location, appearance, colour, size and consistency.

- Management is completely removal of the lesion along with the underlying Blandin-Nuhn gland to avoid recurrence; however, the type of surgery depends upon the size of the mucocele.
- The surgical removal should always be followed by histopathological examination to avoid misdiagnosis.

**REFERENCE**

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