

An unusual presentation of mucocele on the tongue - a case report

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ABSTRACT

Background: Mucoceles are self-limiting mucus-containing cavities that is caused by mechanical trauma to the excretory duct of the gland, which leads to pooling of saliva into the surrounding tissues. They may vary according to its clinical presentations, depending on its depth of involvement. They are usually asymptomatic. Sometimes they may show change in size due to rupture and subsequent mucin accumulation. They more frequently occur in relation to the lower lip.

Case Presentation: We present a case report of mucocele on the ventrolateral aspect of the tongue, which is a rare occurrence. Even though it clinically resembled a mucocele, diagnostic confirmation was done after histopathological examination.

Conclusion: Although surgical excision is the treatment of choice, removal of all the adjacent minor salivary glands is required to prevent the recurrence. They pose a diagnostic and therapeutic challenge to the clinician as well as surgeon due to its clinical resemblance with other lesions of the tongue. So, a thorough clinical knowledge as well as the determination of varying aspects of the etiopathogenesis of these oral lesions is necessary for correct diagnosis and for the indication of appropriate treatment.

Keywords: Mucocele, extravasation cyst, retention cyst, case report.

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Background

Oral mucocele is one of the most common benign minor (accessory) salivary gland lesions that is caused by mechanical trauma to the excretory duct of the gland which leads to the pooling of saliva into the surrounding tissues. By definition, mucoceles (muco - mucus and coele - cavity) are cavities filled with mucus [1]. It is a self-limiting mucous-containing cyst of salivary glands commonly occurring in the oral cavity, with a relatively rapid onset and fluctuating size. Trauma and obstruction of salivary gland ducts are considered to be the most common etiological factors.

The clinical presentation of mucoceles may vary depending on the depth of the lesion. They are usually single dome-shaped soft painless fluctuant swelling with a normal or bluish color. They are usually asymptomatic because of the mucinous contents. The diameter may range from a few millimeters to centimeters. Occasionally, an episodic decrease and increase in size may occur due to rupture and subsequent mucin accumulation [2]. Oral mucoceles of minor salivary glands are usually superficial with diameters ranging between 5 and 14 mm. Mucoceles found in deeper areas are larger, but rarely larger than 1.5 cm in diameter. Even though

mucoceles occur in varying locations of oral mucosal surfaces overlying minor salivary glands, mucoceles of the tongue are rare. Such a rare occurrence of mucocele is illustrated in this case report.

Case Presentation

A 34-year-old male patient reported at OMFS OPD Govt Medical College, Kottayam, with the chief complaint of swelling over the right ventrolateral surface of the tongue. It was noted for approximately 2 weeks without any change in size. The swelling was ovoid in shape with mildly bluish discoloration, measuring approximately 2.5 × 1.5 cm in dimension with no history of bleeding or any discharge from the swelling (Figures 1 and 2). On palpation, it was non-pulsatile and nontender, but fluctuant. It was not blanched on applying finger pressure, suggestive of negative diascopy test. There was no relevant medical and dental history. Based on the clinical examination and history, a provisional diagnosis of mucocele was made. The patient was subjected to ultrasonographic examination which revealed a solitary hypoechoic image without any calcifications. Excisional biopsy along with the removal adjacent



Figure 1. Bluish coloured ovoid swelling over the right ventrolateral aspect of the tongue.



Figure 2. Solitary ovoid swelling over the right ventrolateral aspect of the tongue causing no restrictions for the tongue movements.

minor salivary glands was carried out and the surgical site was closed with sutures. The biopsy specimen was immediately fixed in 10% formalin and sent for histopathological examination. The histopathologic report suggested a confirmatory diagnosis of extravasation-type mucocele.

Discussion

The diagnostic differentiation between solitary, smooth, and nodular submucosal lesions of the tongue is a great challenge for clinicians due to close clinical resemblance of lesions like schwannoma, neurofibroma, rhabdomyoma, lymphangioma, fibroma, lipoma, leiomyomas, oral lymphoepithelial cyst, and inflammatory lesions such as fibroepithelial polyp, benign salivary gland tumors, and mucoceles. Mucocele affects at any age with equal sex predilection, with the highest incidence in the second decade of life. Marcello et al. [4], Yamasoba et al. [5], and Oliveira et al. [6] reported that more than 65% of the patients with OMs were less than 20 years of age with a male:female ratio of 1.07:1 [3]. It can be single or multiple, and are devoid of the epithelial lining or are covered by granulation tissue. Occasionally, it may rupture to leave mildly painful erosions that usually heal within few days. Depending on the depth of the lesion, it can be of three types. The lesions that are located directly under the mucous membrane (superficial mucocele), in the upper submucosa (classical mucocele), or in the lower cerium or lamina propria (deep mucocele)

[7]. The superficial lesions are usually thin-walled, bluish swellings which may rupture easily, whereas the deeper lesions are well-circumscribed nodular swellings covered by normal appearing oral mucosa. Oral mucoceles occur in varying locations on the oral mucosal surfaces overlying accessory minor salivary glands, except the gingiva, where minor salivary glands are absent. The lower lip is the most commonly affected site, while mucoceles on the tongue are rare. If it occurs on the tongue, it is almost exclusively on the ventral surface, where the glands of Blandin and Nuhn are located. The color varies with the size of the lesion, its proximity to the mucosal surface, and the elasticity of the overlying tissue. It may range from deep blue to that of normal mucosa (pink). The deep blue color results from tissue cyanosis, vascular congestion associated with the stretched overlying tissue, and the translucency of the accumulated fluid beneath. Usually, they are asymptomatic, but rarely may cause significant problems like discomfort, interference with speech, mastication, and swallowing, depending on its size and location.

Histologically, mucoceles can be classified as extravasation or retention type. The “extravasation type” is a pseudocyst without defined walls that is caused by the rupturing of the excretory duct by mechanical trauma, with subsequent extravasation of mucin into the connective tissue which may trigger some secondary inflammatory reactions. They are frequently seen in relation to lower labial mucosa, buccal mucosa, and retromolar area. On the contrary, retention type is a true cyst, lined with epithelium in which mucus get retained in the duct or acini due to the epithelial proliferation of a partially obstructed salivary duct by sialolith or strictures. The “retention type” is less common than extravasation, which usually affects the upper lip, hard palate, floor of mouth (ranula), and maxillary sinus in older individuals.

The human tongue has three sets of minor salivary glands, namely the glands of Von-Ebner, the glands of Weber, and the glands of Blandin and Nuhn. Von-Ebner glands are located in a trough of the circumvallate papillae on the dorsal surface of the tongue near the sulcus terminalis, Weber’s glands are located in the superior portion of tonsils in the peritonsillar space, and the glands of Blandin and Nuhn are mixed mucus and serous glands embedded within the musculature of the ventral surface of the anterior tongue [8,9]. Kheur et al. [10] reported a 6.26% occurrence of mucoceles of glands of Blandin-Nuhn in both adults and children. Sugerma et al. [11] have not reported any occurrence of mucoceles of the glands of Von-Ebner and Weber. The glands of Blandin and Nuhn are mixed mucus and serous glands that are approximately 1–8 mm wide and 12–25 mm deep. They consists of several small independent glands forming a horseshoe-shaped mass, extending laterally and posteriorly from the midline. They drain by means of five to six small ducts that open near the lingual frenum [1,7,11].

Mucoceles involving the glands of Blandin and Nuhn are often diagnosed as the extravasation type [7]. The presence of salivary gland tissue and sialomucin is the histological diagnostic feature. Histologically, lesion may show acute inflammation intermingling with the mucus collection or patterns of mature lesions with scarce amounts of mucus and connective tissue fibrosis. The lesion may also shows hyperplastic parakeratinized stratified squamous epithelium, small cystic spaces containing mucin and mucus-filled cells, areas of spilled mucin surrounded by a granulation tissue and sebaceous cells in the connective tissue [12].

Even though the appearance of a mucocele is pathognomonic, a thorough history and examination of the lesion play a key role in its diagnosis. The appearance of a mucocele is pathognomonic, and the following are crucial factors in its differential diagnosis as well as diagnosis: location, history of trauma, rapid appearance, variations in size, bluish color, consistency, and palpation. Signs of fluctuation are positive for cysts, mucoceles, abscess, and hemangiomas, whereas lipomas and minor salivary gland tumors present no fluctuation. But a drained mucocele would not fluctuate and a chronic mucocele has a lesser fluctuation due to fibrosis. Sometimes routine radiographs, fine needle aspiration, ultrasonography or advanced diagnostic methods like computed tomography and magnetic resonance imaging may be required to have a better diagnosis based on the assessment of its contents, form, diameter, position, and origin. Increased amylase activity, increased protein content, and reduced alkaline phosphatase activity in fibroblasts can be revealed by the chemical analysis [8,12].

Surgical excision along with servicing minor salivary glands is the treatment of choice. Usually, the surgical excision includes the servicing mucous glands with evacuation of its contents. Sugerma et al. [11] and Baumash [1] reported the technique for managing moderate-to-large Blandin and Nuhn mucoceles is to completely unroof the lesion along its entire periphery and to visualize and remove all of the glands. After the surgical excision, it may heal without complications or recurrence. Small mucoceles can be completely excised and primarily closed, with rapid and uneventful healing. Larger lesions can be managed by marsupialization, dissection, cryosurgery, laser ablation (carbon dioxide lasers), micromarsupialization, electrocautery, intralesional injection of sclerosing agent OK-432, or steroid injection. The lesion has higher chance of recurrence in marsupialized cases [1,13,14].

Conclusion

The non-neoplastic diseases of salivary gland pose a diagnostic and therapeutic challenge to clinician as well as surgeon due to its resemblance of clinical presentation and varying etiologies, such as reactionary inflammatory

processes, metabolic and immune disorders, and infections and iatrogenic responses. So, a thorough clinical knowledge as well as the determination of varying aspects of the etiopathogenesis of these oral lesions is necessary for the correct diagnosis and for the indication of appropriate treatment.

What is new?

As per the literature, mucoceles commonly occur in relation to lower lip, followed by floor of mouth. We present a case report of mucocele on tongue, which is a rare occurrence.

List of Abbreviations

cm	Centimeter
mm	Millimeter
OMFS	Oral and maxillofacial surgery
OPD	Out patient department
OM	Oral mucocele

Conflict of Interests

The authors declare that there is no conflict of interest regarding the publication of this article.

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Consent for publication

Informed consent was obtained from the patient.

Ethical approval

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Summary of the case

1	Patient (gender, age)	Male, 34 years
2	Final diagnosis	Mucocele
3	Symptoms	Nontender swelling
4	Medications	Antibiotics and analgesics after the biopsy
5	Clinical procedure	Excisional biopsy
6	Specialty	Oral and Maxillofacial Surgery