



## MYSTERY BEHIND MUCOCELES - A LITERATURE REVIEW

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### ABSTRACT

Mucocele is a clinical term used for a pseudocyst that is associated with mucus extravasation into the surrounding soft tissues. These lesions occur as the result of trauma or obstruction to the salivary gland excretory duct and spillage of mucin into the surrounding soft tissues. The present article presents a review on mucoceles.

### INTRODUCTION

Mucoceleles which are of minor salivary gland origin are also referred to as mucus retention phenomenon and mucus escape reaction. The superficial mucocele, a special variant has features that resemble a mucocutaneous disease. At times the mucus retention cyst also referred to as the sialocyst or the salivary duct cyst is included in this group of lesions but appears to represent a separate entity on the basis of its clinical and histopathologic features [1]. Although the mucus retention cyst is discussed in this article, its features are differentiated from the features of the pseudocysts. The lesions of the sinus, such as sinus mucoceles, pseudocysts, and retention cysts, are not included in this discussion.

Ranulas are mucoceles that occur in the floor of the mouth and usually involve the major salivary glands. Specifically, the ranula originates in the body of the sublingual gland, in the ducts of Rivini of the sublingual gland, in the Wharton duct of the submandibular gland, and, infrequently from the minor salivary glands at this location. These lesions are divided into 2 types: oral ranulas and cervical or plunging ranulas. Oral ranulas are superior to the mylohyoid muscle, whereas cervical

secondary to mucus extravasation that pools ranulas are associated with mucus extravasation along the fascial planes of the neck [2].

### Pathophysiology

The development of mucoceles and ranulas depend on the disruption of the flow of saliva from the secretory apparatus of the salivary glands. The lesions are most often associated with mucus extravasation into the adjacent soft tissues caused by a traumatic ductal insult; the insults include a crush-type injury and severance of the excretory duct of the minor salivary gland. The disruption of the excretory duct results in extravasation of mucus from the gland into the surrounding soft tissue. The rupture of an acinar structure caused by hypertension from the ductal obstruction is another possible mechanism for the development of such lesions. Furthermore, trauma that results in damage to the glandular parenchymal cells in the salivary gland lobules is another potential mechanism.

Regarding superficial mucoceles, trauma does not always appear to play an important role in the pathogenesis. In many cases, mucosal inflammation that involves the minor gland duct results in blockage, dilatation, and rupture of the duct with subepithelial spillage of fluid. Changes in minor salivary gland function and composition of the saliva may contribute to their development. In some cases, an immunological reaction

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may be the cause.

Studies have revealed increased levels of matrix metalloproteins, tumor necrosis factor- $\alpha$ , type IV collagenase, and plasminogen activators in mucoceles compared with that of whole saliva. These factors are further hypothesized to enhance the accumulation of proteolytic enzymes that are responsible for the invasive character of extravasated mucus [3].

Besides ductal disruption, partial or total excretory duct obstruction is involved in the pathogenesis of ranulas in some instances. The duct may become occluded by a sialolith, congenital malformation, stenosis, periductal fibrosis, periductal scarring due to prior trauma, excretory duct agenesis, or even a tumor. Although most oral ranulas originate from the secretions of the sublingual gland, they may develop from the secretions of the submandibular gland duct or the minor salivary glands on the floor of the mouth. The mucus extravasation of the sublingual gland almost exclusively causes cervical ranulas. The mucus escapes through openings or dehiscence in the underlying mylohyoid muscle.

Occasionally, ectopic sublingual glands may be responsible for the problem. When mucus secretions escape into the neck through the mylohyoid muscle, they extend into the fascial tissue planes and cause a diffuse swelling of the lateral or submental region of the neck [4].

The continuous secretions from the sublingual gland allow for relatively rapid accumulation of mucus in the neck and a constantly expanding cervical mass.

The mucus retention cyst may also develop because of ductal obstruction; however, many of these lesions actually represent a distinct cystic entity of unknown cause. When ductal occlusion is involved, it is usually caused by a sialolith or an inspissated secretion that results in ductal dilatation and focal containment of the mucoid material.

### **Epidemiology**

In the Minnesota Oral Prevalence Study [5] that included 23,616 white adults older than age 35 years, mucoceles represented the 17th most common oral mucosal lesion, with a prevalence of 2.4 cases per 1000 people. Data from the Third National Health and Nutrition Examination Survey (NHANES III) that included 17,235 adults aged 17 years or older documented an overall prevalence ranking of 44 for the mucocele and a point prevalence of 0.02%. In the same study, which consisted of 10,030 children aged 2-17 years, the mucocele had a point prevalence of 0.04%. Congenital mucoceles in newborns are rare, with sporadic case reports and small case series appearing in the literature.

Mucoceles of the anterior lingual salivary glands (glands of Blandin and Nuhn) are relatively uncommon. In the Minnesota Oral Disease Prevalence Study, Blandin and Nuhn mucoceles had a lower prevalence than mucoceles at other locations, or 0.1 cases per 1000 persons. This type of mucocele represents an estimated 2-

10% of all mucoceles [6].

Superficial mucoceles are typically located in the soft palate, the retromolar region, and the posterior buccal mucosa. They represent approximately 6% of all mucoceles. Multiple superficial mucoceles have been reported in a small number of patients. In an 11-year retrospective review of oral mucoceles and sialocysts from a university-based oral and maxillofacial pathology laboratory, most lesions were found to be mucus retention phenomenon (mucoceles, 91%). In descending order, the other diagnoses included ranulas (6%), and mucus retention cysts (5%). Mucoceles outnumbered mucus retention cysts by a ratio of 15.3:1.0. More limited histopathologic studies document that the mucus retention cyst (those lesions with an epithelial lining) accounts for 3-18% of all oral mucoceles.

Ranulas have a prevalence of 0.2 cases per 1000 persons and are ranked 41st in the Minnesota Oral Disease Prevalence Study. As noted previously, ranulas accounted for 6% of all oral sialocysts in a university-based oral and maxillofacial biopsy service. The prevalence of cervical (plunging) ranulas is not known; however, these lesions are considered uncommon. The number of ranulas that represents a true retention cyst ranges from less than 1% to 10%.

Large international population studies comparable to those undertaken in the United States are not available for oral diseases, except in Sweden. In a study of 30,000 Swedish individuals aged 15 years or older, the prevalence of mucoceles was 0.11%. In a Brazilian study of 1200 children seen at pediatric hospital clinic, the prevalence of mucoceles was 0.08%.

### **Mortality/Morbidity**

Mucoceles tend to be relatively painless or asymptomatic lesions with little or no associated morbidity or mortality. Depending on the size and location, some mucoceles may interfere with normal mastication. Oral and plunging ranulas, if large may affect swallowing, speech or mastication and may result in airway obstruction. The very rare thoracic ranula may compromise respiratory function and may be life threatening.

### **Gender**

Although no gender predilection is usually associated with mucoceles, the prevalence of the lesions in the Minnesota Oral Disease Prevalence Study was 1.9 cases per 1000 males compared with 2.6 cases per 1000 females. Other authors have shown that mucoceles are more common in males than in females, with a male-to-female ratio of 1.3:1. In the reported cases, superficial mucoceles and mucoceles of Blandin and Nuhn have a predilection for females. The sexual predilection for oral ranulas slightly favors females, with a male-to-female ratio of 1:1.4, while cervical ranulas have a predilection for males [7].



## Age

Most mucoceles occur in young individuals, with 70% of individuals being younger than 20 years. The peak prevalence occurs in persons aged 10-20 years. Although not well studied, superficial mucoceles tend to occur in individuals older than 30 years. Ranulas usually occur in children and young adults, with the peak frequency in the second decade. The cervical variant tends to occur a little later in the third decade. Mucus retention cysts occur in older individuals; the peak prevalence occurs in persons aged 50-60 years. Rarely, prenatally diagnosed and congenital mucoceles and ranulas have been reported.

## DISCUSSION

More than 70% of mucoceles arise from the minor salivary glands in the lower lip, whereas only 2.5% of mucoceles arise on the tongue. Therefore, mucoceles of the glands of Blandin and Nuhn are uncommon. They may be the result of trauma to the tongue ventrum that ruptures the draining ducts. This results in extravasation of secretions into the connective tissues, which excites a variable inflammatory and tissue repair response. The vascular and granulation tissue elements may be pronounced, as found in pyogenic granulomata associated with pregnancy.

A history of trauma and recovery of mucus with fine needle aspiration are helpful in the clinical diagnosis of mucocele of the glands of Blandin and Nuhn. The following characteristics of the mucocele are: rapid onset, increase and reduction in size, bluish color and fluid-filled consistency. During surgery, the glands that are deep in the tongue musculature are commonly left behind, resulting in persistence of the lesion. Careful clinical evaluation of these lesions and preoperative awareness of the surgical anatomy of the glands of Blandin and Nuhn

may minimize the need for repeated surgical procedures [8,9].

## CONCLUSION

Mucoceles are mucus containing cystic lesions of the minor salivary glands. The clinical appearance of a mucus cyst is a distinct, fluctuant, painless swelling of the mucosa. The patient may relate a history of recent or past trauma to the mouth or face or the patient may have a habit of biting the lip. The history and clinical findings lead to the diagnosis of a superficial mucocele. Radiographic evaluation is considered if sialoliths are considered a contributing factor in the formation of oral and cervical ranulas. Ultrasonography has been used to evaluate the lesions, and with high-frequency transducers, ultrasound demonstrates the internal structures more clearly than computed tomography (CT).

Ultrasound can rule out the type of lesion before surgical interventions can be attempted. The demonstration of mucus retention phenomenon and inflammatory cells can be done by fine needle aspiration, and high amylase and protein content can be revealed in chemical analysis. The localization and determination of the origin of the lesion can be done by CT and magnetic resonance imaging.

Surgical excision with removal of the accessory salivary glands has been suggested as the treatment. The excised tissue should be submitted to the pathological investigations to confirm the diagnosis. Laser ablation, cryosurgery, and electrocautery are approaches that have also been used for treatment of the conventional mucoceles, with variable success.

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