

Mucocele of the Ventral Surface of the Tongue: A Case Report

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ABSTRACT

Mucoceles are common cystic lesions of the oral cavity that can appear in different locations in the oral mucosa due to trauma or obstruction of salivary gland ducts with the lower lip as the predominant location. However, mucocele occurs on the ventral surface of the tongue involving the anterior lingual gland is rarely seen especially in pediatric group. In the present case, a 9 years old Saudi female was diagnosed with a mucocele on the ventral surface of the tongue. Clinical presentation, microscopical and histological features are described with the review of literature.

Key Words: Extravasation, Mucocele, Retention phenomena, Salivary gland.

INTRODUCTION

Mucocele (from the Latin terms mucus, or mucus and coele, or cavity) is one of the most common benign soft tissue lesions of the oral mucosa (Bhavna *et al.*, 2007; Jose *et al.*, 2009). They are believed to be secondary to traumatic or obstructive disorders of the minor salivary glands (Jimbu *et al.*, 2003; Marcello *et al.*, 2008; Jose *et al.*, 2009).

The incidence of mucocele is high, It accounts for 2.5 lesions per 1000 patients, frequently in the second and third decade of life (Guimaraes *et al.*, 2006; Jose *et al.*, 2009) without gender predictability (Huang *et al.*, 2007; de Camargo *et al.*, 2009). Mucoceles usually appear in the glands that secrete predominantly mucous saliva. It is therefore more frequent to find these lesions in the minor salivary glands distributed throughout the oral submucosa, except in the attached gingival regions (Jimbu *et al.*, 2003; Jose *et al.*, 2009). The lower lip is considered the most frequent site for a mucocele as it is the most probable place for such a trauma, especially at premolar level. Such trauma could be due to accidental biting, trauma by hand-held implements (e.g. toothpick, pencil, toothbrush). Other traumatic oral habits, such as lip sucking, would be among the major causes (Jing-Yi *et al.*, 2010). When the mucocele is located in the floor of the mouth, this lesion is known

as ranula which derived its name from the Latin word “rana” (meaning frog) because of its resemblance with the underbelly of a frog (Baurmash, 2003; Marcello *et al.*, 2008; Alethea *et al.*, 2010). Ranulas are related to the duct systems of the sublingual salivary glands. However, it is less frequently related to submandibular gland and minor salivary gland ducts of the floor of the mouth (Alethea *et al.*, 2010). Mucoceles that arising on the tongue occasionally involve Blandin-Nuhn glands.

Clinically, mucoceles present, as a well circumscribed, asymptomatic swelling of variable size ranging in color from deep blue to the normal pink color of the oral mucosa depending on the size of the lesion, proximity to the surface, and upper tissue elasticity (Bentley *et al.*, 2003). It is fluctuant and smooth on palpation. The blue color is caused by vascular congestion, cyanosis of the above tissue, and the accumulation of fluid below (Baurmash, 2003; Bentley *et al.*, 2003). Lesion duration is not constant. According to Harrison (Harrison, 1975), the lesions develop over a period of one week to five years, though the most common duration is three weeks to three months. As for the size, mucoceles of the minor salivary glands are rarely larger than 1.5 cm in diameter (reported lesion size varies between 0.2 mm and 2 cm in diameter as found by Baurmash,

2003; Jose *et al.*, 2009) and are always superficial. Mucocoeles found in deeper areas are usually larger. Mucocoeles rarely cause problems like difficulty in speaking, swallowing, and mastication depending on the size and location of the lesion (Baurmash, 2003; Bhavna *et al.*, 2007; Jose *et al.*, 2009). In 1965, mucocoeles were classified microscopically by Cohen (1965) into two types, i.e. Extravasation phenomena and mucus retention cyst. Extravasation mucocoele results from a broken salivary glands duct and the consequent spillage into the soft tissues around this gland. Retention mucocoele appears due to a decrease or absence of glandular secretion produced by blockage of the salivary gland ducts.

CASE REPORT:

A nine years old, Saudi girl presented to the department of oral & Maxillofacial Surgery, King Fahad Hospital-Hofuf, KSA with a complaint of swelling under her tongue for few weeks with difficulty of eating and speaking. She had unremarkable medical history. There was no history of trauma. Extra-oral examination did not show any swelling or lymphadenopathy. Intra-oral examination revealed a non-tender, dome shaped, un-ulcerated fluid filled lesion on left side of the ventral region of the tongue with intact overlying mucosa. (figure 1).

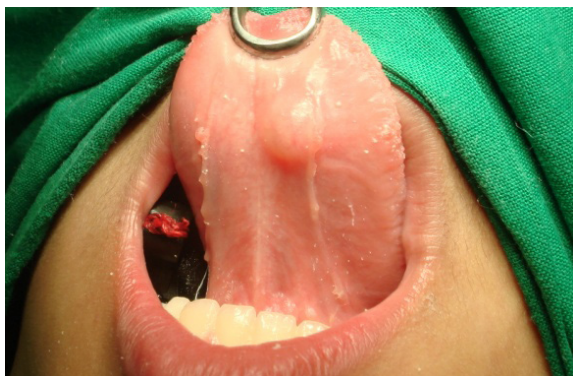


Figure 1: Intraoral view of cystic lesion of left ventral side of the tongue with normal covering mucosa (measuring about 1cm x 1cm).

Fine needle aspiration (FNA) revealed viscous salivary aspirate, which clinically

confirmed the diagnosis of mucocoele. Excision of the mucocoele was done under general anesthesia with associated normal minor salivary glands to avoid recurrence. (figure 2)



Figure 2: Intra-operative view of the tongue mucocoele.

Excised lesion was sent for histopathologic examination, which confirmed the diagnosis of mucocoele (extravasation phenomena) (figure 3). Patient was reviewed for six months without any sign of recurrence.

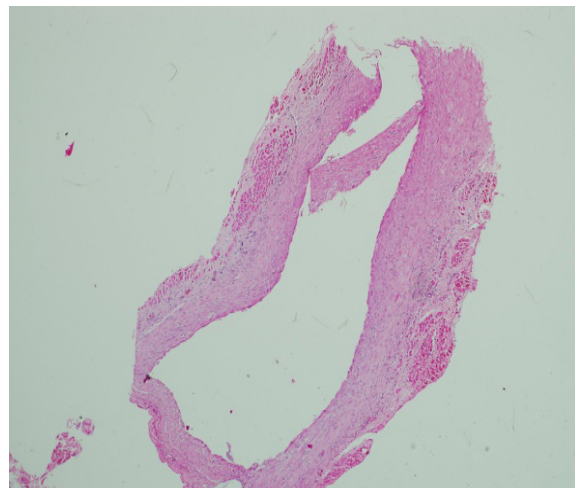


Figure 3: Histological view of pseudocyst surrounded by granulation tissue (H&E, x20).

DISCUSSION

Blandin-Nuhn glands are compact groups of small mixed salivary glands located near the ventral tip of the tongue and are arranged in a mass with a horseshoe shape. They are embedded in the muscles of the ventral aspect near the midline and covered by a thin layer

of mucosa (Baurmash, 2003; Jimbu *et al.*, 2003; Marcello *et al.*, 2008). Even though it is described as a rare and uncommon lesion as reported by Harrison (1975). In his review of 400 cases of mucoceles, he reported only 2.25% of the lesion arose from the tongue. In later studies, Jimbu *et al.*, (2003) found that Blandin–Nuhn mucoceles comprise 9.9% of the oral mucoceles cases they studied. However, de Camargo Moraes *et al.*, (2009), in their 6-years-period study, diagnosed 15.4% of the cases with Blandin–Nuhn glands mucocele.

The Blandin-Nuhn, glands' mucocele is easily diagnosed clinically. However, Sugeran *et al.*, (2000) stated that the clinical presentations of those mucoceles are similar to vascular lesions, pyogenic granulomas, polyp and squamous papiloma, depending on the vascularization degree and the atrophy of the acinus. The diagnosis of the mucocele is mainly clinical. So, the anamnesis of the patient should be collected correctly. Data such as lesion location, size, rapid appearance and history of trauma must be gathered. Lesion palpation and fine needle aspiration biopsy (FNAB) are very helpful in the diagnosis of the mucocele. In particular cases, the diagnosis may require traditional radiography, ultrasonography or advanced diagnostic methods including computed tomography (CT) or magnetic resonance imaging (MRI) to better visualize form, diameter and position of the lesion relative to adjacent organs (Zancopé *et al.*, 2009; Dario *et al.*, 2010). Performing a histopathological investigation of the excised mucocele is important too.

Histopathologically, in the case of retention mucoceles, which are less frequent and seen particularly in elderly patients, a cyst cavity can be found. This is generally well defined with an epithelial wall covered with a row of cuboidal or squamous cells produced from the excretory duct of the salivary glands. Compared to extravasation mucoceles commonly found in individuals less than 30 years old and account for over 80% of

all mucoceles, retention mucoceles show no inflammatory reaction and are true cysts with an epithelial covering. Extravasation mucoceles are pseudocysts without defined walls. The extravasated mucous is surrounded by a layer of inflammatory cells and then by a reactive granulation tissue made up of fibroblasts caused by an immune reaction. Even though there is no epithelial covering around the mucosa, this is well encapsulated by the granulation tissue (Baurmash, 2003; Guimarães *et al.*, 2006; Jose *et al.*, 2009). The histopathological examination of the mucoceles of the glands of Blandin–Nuhn reported in the literature, as well as in our case, revealed that they consist of mucus extravasation phenomenon with no epithelium lining the mucin collection.

The suggested treatment of the mucocele of Blandin–Nuhn glands was surgical excision together with the associated minor salivary glands to avoid any injury caused by the excision to any marginal glands and/or ducts that may lead to recurrence of the lesion. Alternative treatment include cryosurgery, intralesional corticosteroid, OK-432 injection, micro-marsupialization, marsupialization of the mucocele and laser ablation (Huang *et al.*, 2007; Jose *et al.*, 2009). Marsupialization of these lesions alone should be avoided since it is very likely to result in recurrence.

CONCLUSION

Mucocele of Blandin-Nuhn gland is an uncommon lesion. Clinician should know the main features of the lesion since the clinical appearance could be similar to other lesions such as vascular lesions, pyogenic granulomas, polyp, squamous papiloma or lymphangioma. Fine needle aspiration is helpful in the clinical diagnosis of mucocele. It is important that the excised lesion is followed by histopathologic examination to confirm the diagnosis. The treatment of choice of this lesion is surgical excision with associated minor salivary gland to avoid recurrence of the lesion.

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قبيلة مخاطية في السطح البطني للسان : تقرير حالة

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الملخص

القبيلة المخاطية من الأمراض الكيسية الشائعة في الفم والتي يمكن أن تظهر في أماكن مختلفة من الغشاء المخاطي للفم نتيجة لانسداد أو تمزق قنوات الغدد اللعابية، وتعتبر الشفة السفلية من أكثر الأماكن شيوعاً لظهورها. إن ظهور القبيلة المخاطية في السطح البطني للسان يعتبر من الأمور النادرة خاصة عند الأطفال.

الهدف من هذه الحالة هو استعراض قبيلة مخاطية في السطح البطني للسان طفلة سعودية تبلغ من العمر 9 سنوات من الناحية الإكلينيكية والمخبرية كما نقوم بمراجعة بعض النشرات العلمية بخصوص القبيلة المخاطية.

الكلمات المفتاحية: الظاهرة الاحتباسية، الظاهرة التسرية، غدة لعابية، قبيلة مخاطية.