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## Multiple recurrent vesicles in oral mucosa suggestive of superficial mucocele: An unusual presentation of allergic stomatitis

### *Abstract*

**Background:** Superficial mucocele presents as small, clear vesicle on noninflamed mucosa. In this study, we report several vesicles on the bucal mucosa of a woman diagnosed as superficial mucocele.

**Case Presentation:** A 48-year old woman presented with multiple vesicles on her labial mucosa, ventral surface of the tongue, floor of the mouth and palate. A mucosal biopsy was taken from the vesicle. Histopathologically, intraepithelial mucocele was diagnosed. The lesion was successfully treated with mouthwash betamethasone. There has been no recurrence for 18 months.

**Conclusion:** In the present study, several mucoceles were seen in the oral mucosa. No similar case was reported previously.

**Keywords:** Superficial mucocele, Vesicle, Mucosa, Salivary gland.

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The superficial mucocele is a small, translucent, tense and subepithelial vesicle affecting the oral mucosa. It could be either single or multiple. Occasionally, the lesions are persistently recurrent, with a pattern of rupturing, causing mild discomfort and healing within a few days. Superficial mucoceles may develop on any location where minor salivary glands exist, including the soft palate, retromolar region, and buccal mucosa. The superficial mucocele was first defined by Eveson in 1988 (1). Before his report, superficial mucoceles were often misdiagnosed as pemphigoid, bullous lichen planus or herpes virus infection (1-3). In this article, we reported a case of multiple superficial mucoceles on the labial mucosa, ventral surface of the tongue, floor of the mouth and palate.

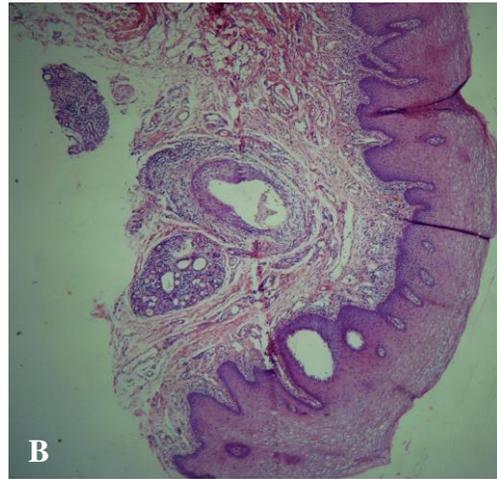
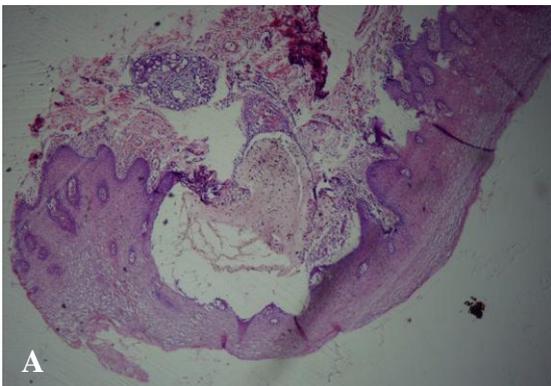
### Case presentation

A 48-year-old white woman was referred to Babol Dental Faculty in October 2010 with a chief complaint of recurrent multiple vesicles in her mouth for four months (figure1). The vesicles caused a little pain and irritation. She was visited by a physician and managed with diagnosis of aphthous ulcer with no improvement. So she was referred to the Oral and Maxillofacial Medicine Department. The patient has maxillary and mandibular removable dentures and poor oral hygiene. Both maxillary and mandibular molars had amalgam fillings. Intraoral examination showed many clear vesicles 1-3 mm in diameter with an erythematous base on the palatal mucosa, lower labial mucosa, floor of the mouth and ventral surface of the tongue. They burst spontaneously or by eating. Clinical examination revealed no submandibular, sublingual or cervical lymphadenopathy. There were not any other mucosal or cutaneous involvements. The patient had iron deficiency anemia and hypothyroidism and used levothyroxine for 6 years. She underwent lower labial biopsy.



**Figure 1. Multiple superficial mucoceles; A: Palatal surface, B: Ventral surface of the tongue**

The histopathologic examination showed that the oral mucosa consisted of stratified squamous epithelium with intra cellular edema. Intraepithelial vesicle was seen as well. Also, in subepithelial connective tissue, minor salivary glands consisted of mild chronic inflammatory cells infiltration was observed (figure 2). Considering the clinical signs and histopathologic result, final diagnosis confirmed it to be allergic stomatitis.



**Figure 2. Histopathologic features of the superficial mucocele (hematoxyllin –eosin stain: original magnificantion ×10) A: Intraepithelial mucocele were observed, B: Multiple mucoceles were observed**



**Figure 3. Palatal surface, A: 2 months after treatment, B: 3 months after treatment**

According to the diagnosis, betamethasone mouthwash 3 mg/ml was administered to the patient 4 times a day accompanied with Nystatin suspension (100,000 IU) 3 times a day, each time 40 drops were used for 3 months. Cetirizine 10 mg/day was taken for 2 months. She was advised to avoid allergenic foods and chemicals. She was followed up every two weeks for about 3 months.

During this period, signs of improvement appeared (figures 3). At the end of these 3 months, the patient reported no complaint of pain or burning. The lesions improved completely. Only white reticular striations were seen on the right and left buccal mucosa. Suspected to oral lichen planus, previous specimen of lower labial biopsy was again evaluated.

An intraepithelial mucocele and dilated ducts and inflammation in ducts of minor salivary glands were seen. Histopathologic findings were compatible with superficial mucocele. After treatment, the patient has been followed up every 2 months for 18 months without any complaint.

## Discussion

The superficial mucocele is a rare bullous oral mucosal lesion that is more frequent in women over the age of 30 (4, 5). In most researches, superficial mucocele occurs in single site with two or three blisters. The recent case was a patient with similar clinical signs and history but different multiple lesions were seen on different mouth areas such as the ventral surface of tongue, floor of the mouth, labial mucosa and palatal mucosa. The great number of vesicles and their vast area of involvement were the characters of this case which made it different from other reports.

Superficial mucoceles have not been defined as distinct entities in most pathology resources, thus these lesions were difficult to diagnose (5, 6). Superficial mucoceles were initially misdiagnosed as pemphigoid, herpes virus infection and bullous lichen planus (1, 5).

The clinical signs of pemphigoid and bullous lichen planus are absolutely different from superficial mucoceles. In these lesions, bullae are opaque, loose and bigger whereas, the vesicle of the superficial mucocele is translucent and tense, similar to a dewdrop. Also a positive Nikolsky phenomenon may show the existence of pemphigoid (7).

Superficial mucoceles represent subepithelial extravasations of sialomucin occurring at the epithelial-

connective tissue interface. Minor salivary gland ducts are often seen in the immediate vicinity of mucoceles. The histopathologic feature of current case showed the lesion was an intraepithelial mucocele.

The etiology of superficial mucocele is not specified. More often, this lesion occurs in regions which are not exposed to trauma. Hence, the traumatic etiopathologic mechanism of conventional mucocele seems unlikely (8). The obstruction or rupture of the duct might be caused by increased intra ductal pressure in the intraepithelial portion of the duct secondary to local chronic inflammation (4, 9). Navazesh suggested that the tartar that is contained in toothpaste could be a contributing factor to developing superficial mucocele (6).

Our case wears both maxillary and mandibular dentures and histopathological image showed chronic inflammation. Consequently both trauma and chronic inflammatory factors were effective on the etiology of this lesion. In some studies, superficial mucocele related to oral lichen planus was reported (5, 9, 10). Examination revealed bilateral white striations on the buccal mucosa and amalgam preparations existed in maxillary and mandibular molars. As far as we know, she was not convinced to be biopsied, the association between these lesions and oral lichen planus or lichenoid was in doubt. Most superficial mucocele is resolved spontaneously and needs no treatment unless it causes continuous irritation (1, 2). Since this disease is rare and often misdiagnosed as other diseases, careful intraoral and histopathological examination is necessary for prompt diagnosis.

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