Case Report

Multiple Mucous Retention Cysts (Mucocele) of the Oral Mucosa: A Case Report

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ABSTRACT

To our knowledge, the occurrence of multiple mucoceles is not very common. This case report presents a 62-year-old man with multiple nodules on the upper and lower labial mucosa as well as both buccal mucosae with unknown history. Histopathology evaluation showed minor salivary gland ducts dilated to the point of cyst formation. The cysts seemed to be formed either as a result of dilatation of salivary ducts due to altered secretion or because of an acquired or congenital weakness in the ductal structure. The physiopathology of these findings is discussed.

Keywords: Lip, Mucous retention cyst, Mucocele, Multiple, Nodule, Oral.

Received: *October 2007*
Accepted: *December 2007*

Dent Res J 2007; 4(2): 111-113

Introduction

Cystic lesions of the minor salivary glands are very common. These lesions clinically referred to as mucoceles may be either extravasation or retention type. Mucoceles are usually single, although more than one may be present at a time [1]. Gorlin et al [2] in 1970 illustrated a case of lower lip multiple mucous extravasation cysts, in which at least three distinct cysts were presented. Tal et al in 1984 reported two cases of multiple mucous retention cysts in which numerous minor salivary gland ducts had dilated to the point of cyst formation and the number of individual cysts exceeded 100 in each case [1]. Jinbu et al [3] in 2003 described a woman with multiple recurrent blister formation on the soft palate, proved to be superficial mucoceles. And in the 13th international congress on Oral Pathology and Medicine (Geneva, Switzerland, 2006) Kuffer et al [4] described two other cases.

Case Report

A 62-year-old man complained of multiple small nodules in the buccal and labial oral mucosa. These nodules were first noticed by his dentist about one month earlier when he extracted some remaining teeth in order to prepare a full denture. He was referred to the department of Oral Medicine in the School of Dentistry of Isfahan University of Medical Sciences in July 2007. The patient medical history was unremarkable, except for minor thalassemia (hypochromic microcytic anemia) and consumption of folic acid and mild tranquilizer (Diazepam 2 mg). He had no history of any traumatic incident or para functional habits but he was a heavy smoker (1 package per day) for more than 40 years. There was no indication of familial history of similar oral lesions. Head and neck examination disclosed nothing remarkable. In oral examination, the mucosal surface of the lips and the buccal mucosa showed numerous nodules; about 16 in the upper lip and 5 in the lower lip and 2 in both buccal mucosa (figures 1 and 2). The nodules were smooth in surface, pink color, firm on palpation, sessile and painless. Some of the nodules appeared slightly inflammed. On pressure, some

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nodules exuded a viscous saliva-like liquid, and some secreted pus, while in others a thick yellowish-white fluid was seen. The other parts of his oral mucosa were almost normal.

Two biopsy from two distinct lesions were done and histological examination revealed dilated ducts in lamina propria and submucosa. Some of the excretory ducts seemed to be dilated to the point of cyst formation (figure 3). The cysts were lined by two to three cell layers thick flattened squamous epithelium or by a pseudostratified columnar epithelium containing mucous-secreting cells (figure 4). Occasional dilated ducts were lined with oncocytes and were filled with mucous (figure 5). The alveolar elements of the minor salivary glands appeared normal. Inflammation was greatly absent except occasionally around ducts near surface (figure 6). The ducts opening onto the surface were markedly dilated and acinar cells of the adjacent minor salivary glands were unremarkable.

According to all findings above and based on the type of epithelial lining, possibility of inclusion cyst was excluded and final diagnosis rendered was multiple true retention cysts.

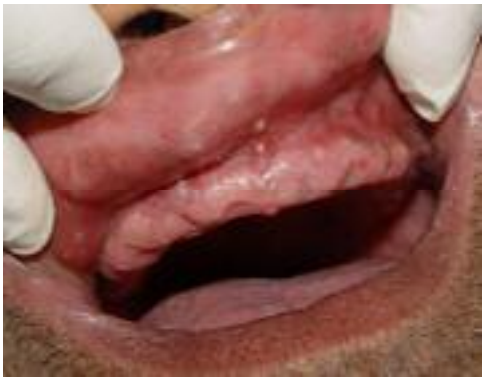

Figure 1. Numerous nodules on the upper lip.

Figure 2. Numerous nodules on the lower lip.

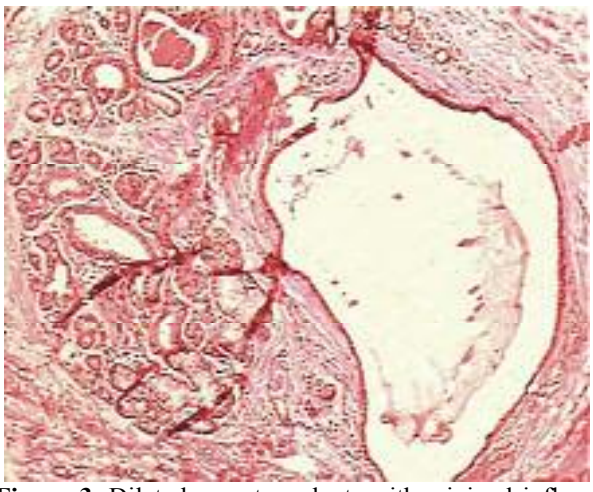

Figure 3. Dilated excretory ducts with minimal inflammatory infiltration. (H&E stain, original magnification, X10).

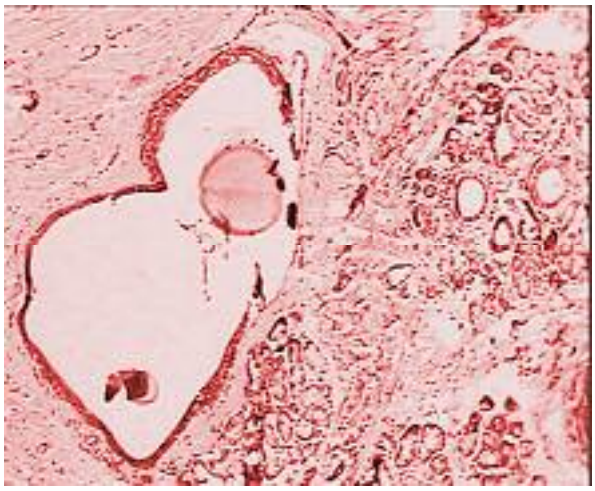

Figure 4. Dilated excretory duct with mucous plug (H&E stain, original magnification, X10).

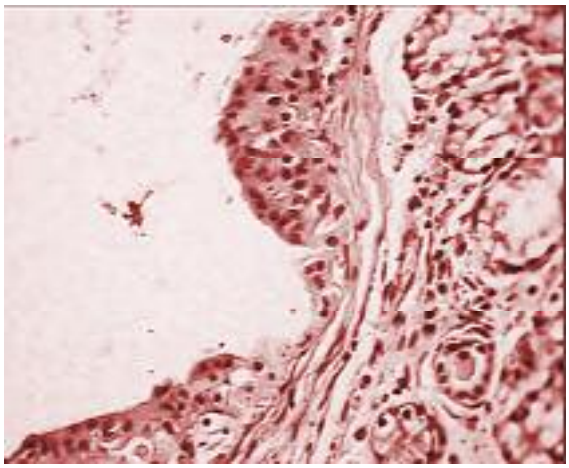

Figure 5. Dilated intralobular duct with oncocytic epithelium (H&E stain, original magnification, X40).

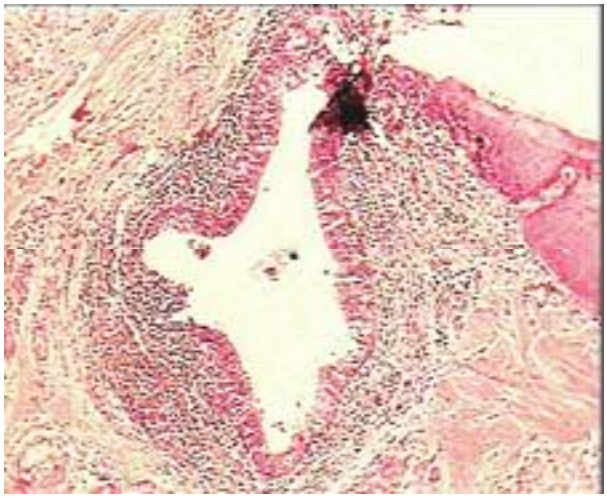

Figure 6. Dilated duct orifice with secretory pseudostratified columnar epithelium (H&E stain, original magnification, X10).

Discussion

Cheilitis glandularis was considered in the differential diagnosis of the case [1,5]. Although the dilatation of salivary ducts may occur in such a condition, cyst formation has not been described. Also in this case, other mucosal site in addition to the lip was affected and there was no histological evidence of hypertrophy of minor salivary gland tissue. Multiple mucoceles reported in this case was due to cystic dilatation of minor salivary glands excretory ducts. Meanwhile we could not find any related medical condition such as cystic fibrosis in this patient.

Multiple mucous retention cysts with dilated ductal orifices have been reported in the literature and in some cases especially those with ductal obstruction, the epithelium may undergo oncoytic metaplastia that often demonstrates papillary folds into the cystic lumen [6,7]. In this case, there was neither papillary enfolding in the cyst wall nor lymphocytic proliferation. So papillary cystadenoma and Warthin's tumor were excluded respectively.
Since there were no clinical signs of inflammation around the nodules, heat or traumatic incidence secondary to heavy smoking were excluded.

Tal and his colleagues [1] described two possible explanations for the formation of multiple mucoceles, either the duct obstruction by an altered, tenacious and viscid mucous, or congenital or acquired weakness in the structure of the ducts, which leads to stasis of secretion in dilated ducts and secondary retrograde infection. In this case, surprisingly the patient mouth was not dry and decrease of saliva flow was not remarkable. A mucous coagulum was found in the histopathological observation (figure 4). It could be due to alteration in minor salivary secretion component, or due to a combination of factors such as senile metabolic changes with poor oral hygiene and smoking. These factors may play some role in the formation of such conditions. Kuffer et al [4], also described inspissated mucous plug in one cyst with pericystic fibrosis and chronic lymphocytic infiltration. Unfortunately the patient failed to return for subsequent appointment, so no further investigation and metabolic assessment (blood and saliva biochemistry) were possible.

Nevertheless, the sequence of phenomenon is obscure. It may be due to alteration of minor salivary glands secretion and the subsequent dilatation of duct or due to dilatation of ducts, leading to salivary flow stasis and formation of cysts. This question remains a challenge for future investigations.

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