WORLD JOURNAL OF PHARMACEUTICAL RESEARCH
SJIF Impact Factor 6.805
Volume 5, Issue 9, 1450-1455. Case Report ISSN 2277– 7105

"CLEAR CELL VARIANT OF MUCOCELE – A CASE REPORT"

[1]*Dr. Sushruth Nayak, M.D.S, [2]Dr. Prachi Nayak, M.D.S, [3]Dr. Manisha Sahu

[1]Professor, Department of Oral Pathology & Microbiology, Maharishi Markandeshwar College of Dental Sciences & Research, Mullana, Ambala, Haryana.

[2]Reader, Department of Oral Pathology & Microbiology, Maharishi Markandeshwar College of Dental Sciences & Research, Mullana, Ambala, Haryana.

[3]Department of Oral Pathology & Microbiology, Chhattisgarh Dental College and Research Institute, Rajnandgaon, Chhattisgarh.

Article Received on
04 June 2016,

Revised on 25 July 2016,
Accepted on 15 August 2016

DOI: 10.20959/wjpr20169-6941

***Corresponding Author**
Dr. Sushruth Nayak
Professor, Department of Oral Pathology & Microbiology, Maharishi Markandeshwar College of Dental Sciences & Research, Mullana, Ambala, Haryana.

ABSTRACT

Mucocele is a common lesion of the oral mucosa that results from an alteration of minor salivary glands due to a mucous accumulation. Mucocele involves mucin accumulation causing limited swelling. Two histological types exist - extravasation and retention. Mucoceles can appear at any site of the oral mucosa where minor salivary glands are present. The most common location of the extravasation mucocele is the lower lip, while retention mucoceles can be found at any other site. Mucoceles can affect the general population, but most commonly young patients. We report a case of 26 -year-old man who presented an asymptomatic translucent swelling on the lower labial mucosa which was clinically diagnosed as mucocele. Histologically it showed macrophage proliferation with numerous clear cell populations. This unusual presentation expands the microscopic spectrum in diagnosis of oral mucoceles and also can lead to differential diagnosis with primary or metastatic clear cell neoplasms.

KEYWORDS: Clear cell, lip, mucocele.

INTRODUCTION

Mucocele is a common lesion of the oral mucosa that results from an alteration of minor salivary glands due to a mucous accumulation. Mucocele involves mucin accumulation causing limited swelling. Two types of mucocele can appear - extravasation and retention.[1]

Mucous is produced exclusively by the minor salivary glands and is also the most important substance secreted by the major sublingual salivary glands. Extravasation mucoceles are caused by a leaking of fluid from surrounding tissue ducts or acini. This type of mucocele is commonly found on the minor salivary glands. Physical trauma can cause a leakage of salivary secretion into surrounding submucosal tissue. Inflammation becomes obvious due to stagnant mucous resulting from extravasation. Extravasation mucoceles undergo three evolutionary phases. In the first phase, mucous spills diffusely from the excretory duct into connective tissue where some leucocytes and histiocytes are found. Granulomas appear during the resorption phase due to histiocytes, macrophages and giant multinucleated cells associated with a foreign body reaction. In the final phase connective cells form a pseudocapsule without epithelium around the mucosa. Retention mucoceles are formed by dilation of the duct secondary to its obstruction or caused by a sialolith or dense mucosa. The majority of retention cysts develop in the ducts of the major salivary glands.[2] We present an unusual case of oral mucocele being exclusively composed of macrophages showing extensive clear cytoplasmic changes. The current case can potentially lead to diagnostic difficulties with clear cell and/or lesions of the oral mucosa such as salivary gland or metastatic origin neoplasms.

CASE REPORT

A male patient aged 26 year old, reported to the department with the chief complaint of an intraoral swelling in lower left labial mucosal region of the mouth for past one year. History revealed that the swelling was initially smaller in size and gradually increased to the present size. The patient did not give any relevant medical history, with no history of trauma, parafunctional habits, or history of difficulty or pain during chewing or speaking. On extraoral examination, the face of the patient was bilaterally symmetrical with no remarkable findings (Figure-1). On Intraoral inspection, a solitary well defined swelling was present on lower left comissure region in relation to tooth number 33, 34 and 35. The swelling extended anterio-posteriorly from commisure of lip to the mesial surface of 35. The swelling measured approximately 3 x 2 cm in size and was oval in shape (Figure-2). It had smooth and shiny surface with normal colour to the adjacent muosa. On palpation, the swelling was soft in consistency, non-tender and fluctuant. On correlating the clinical history with the clinical examination a provisional diagnosis of Mucocele was made, with the differential diagnosis of fibroma, lipoma, haemangioma, and salivary gland tumour. Further hematological investigations were made which was under normal limits. Incisional biopsy was performed

and the tissue was taken up for the histopathological examination. Hematoxylin and eosin stained section revealed fibrous connective tissue wall enclosing the cystic lumen (Figure-3). The connective tissue wall showed the presence of foamy macrophages with areas of clear cytoplasmic cells. The cells were round with oval nuclei resembling the clear cells (Figure-4). Also seen was the presence of mild chronic inflammatory cell infiltrates with blood vessels. Periodic acid Schiff's (PAS) stain revealed the positivity for mucin (Figure-5). Depending on the above mentioned features, final diagnosis of Mucous Extravasation cyst with clear cell variant was made.

DISCUSSION

Mucocele is a benign cystic lesion of the oral cavity that has been ranked seventeenth most common salivary gland lesion seen in the oral cavity. It is the second most common benign soft tissue tumor occurring in the oral cavity. The incidence of Mucocele is generally high, 2.5 per 1000 patients, frequently occurring in the second decade of life and rarely among children and infants under one year of age.

Various literatures on Mucocele reveal that it is equally seen in men and women, whereas other studies reported that Mucocele has a slight female prevalence of about 1.3:1 but the reported case was a male patient and the lesion was compatible with extravasation cyst. Mucocele can be traumatic or non-traumatic in origin. Parafunctional habits such as lip biting being the most common contributory factor for occurrence of an oral Mucocele. The literature reveals that most of the patients give a history of spontaneous development (71.4%), followed by lip biting (25.7%) and trauma (2.9%). The duration of the lesion is not constant; it varies from a few days to 3 years. The blue color is caused by vascular congestion, and cyanosis of the tissue above and accumulation of the fluid below. Coloration can also depend on the size of the lesion, proximity to the upper surface and elasticity of the superficial tissue.[3] Interestingly, unusual mucocele variants have been reported. Superficial mucoceles are solitary or multiple, which microscopically show mucus extravasation at the epithelial-connective tissue interface. Myxoglobulosis-like change have been reported in some oral mucoceles, in which globular hyalinized structures interspersed with mucin and mucin-laden macrophages are present into the lumen or in the extraluminal connective tissue. Another unusual pattern characterized by a metaplastic transformation from granulation tissue that circumscribes the mucin pool to papillary folds exhibiting in their surface synovial-like changes has also been described.[4] In our case clinical and histological

findings were suggestive of an extravasation type of mucocele, which is not considered to be a challenge for a pathologist. But the presence of numerous clear cell populations within the connective tissue stroma is histologically very unusual and may present a difficulty in diagnosis.

Differential diagnosis of tumors with a predominant clear cell and/or signet ring cell (SRC) components in the head and neck region can be challenging. Usually, clear cell/SRC change results from fixation artifacts, intracellular storage of glycogen, mucin or lipid, or due to paucity of organelles. The differential diagnosis of clear cell lesions of the oral mucosa includes salivary gland, sebaceous gland, melanocytic and metastatic tumors. Although most exhibit distinctive microscopic characteristics, histochemistry and immunohistochemistry can be required as well as relevant clinical information in order to achieve the correct diagnosis.

Mucocele with clear cell features as the presented here should be distinguished from clear cell variant of salivary gland tumors such as myoepithelioma, oncocytoma, mucoepidermoid carcinoma, myoepithelial carcinoma, acinic cell carcinoma and clear cell carcinoma, not otherwise specified. The malignant salivary gland tumors exhibit the propensity for regional or distant metastasis and low recurrence. By immunohistochemistry, all these salivary gland tumors are cytokeratin positive while the mucoceles are uniformly negative.[5]

Clear cell aggregates can lead to confusion and challenge the pathologist to differentiate benign lesion like mucocele from an aggressive malignant neoplasm. In such cases, the use of CD68, CD163 (shows strong positivity) and AE1/AE3 pan-cytokeratin, S100, HMB-45 and CD1 (negative) immunomarkers are valuable tools to arrive at an accurate diagnosis.[5]

Figure-1: Extra oral view

Figure-2: Intra oral swelling

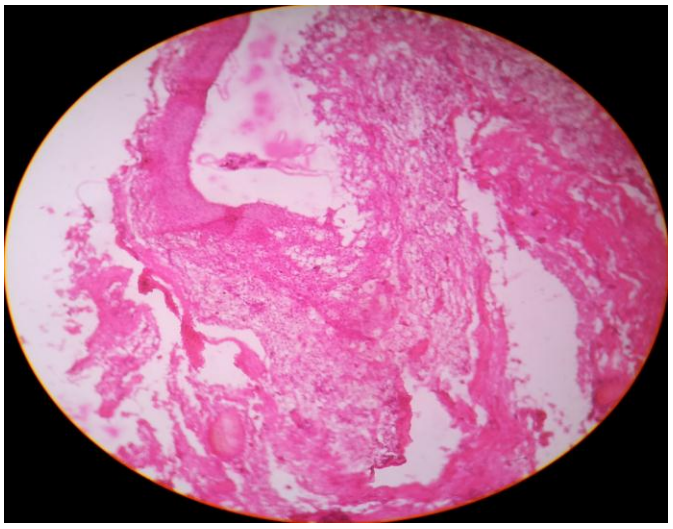

Figure-3: Cystic lumen surrounded by connective tissue stroma

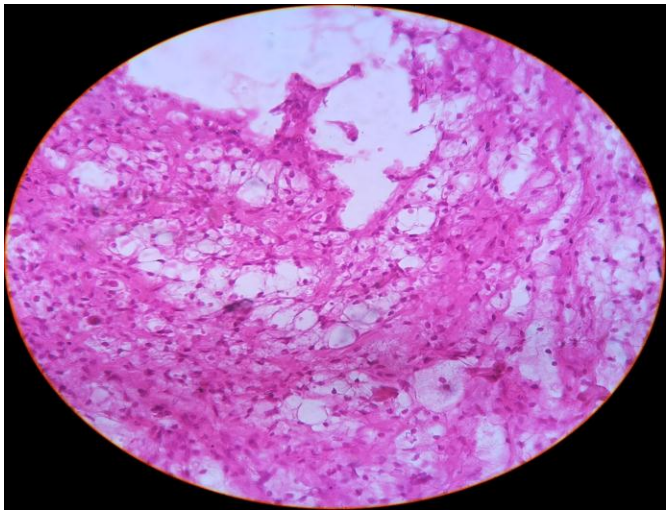

Figure-4: Clear cell population

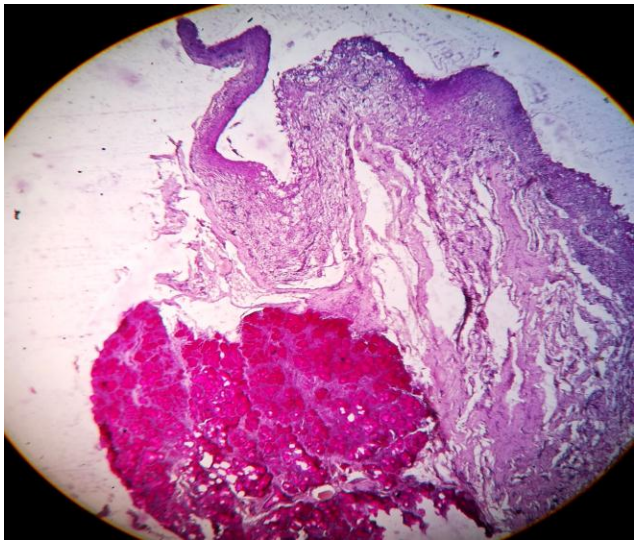

Figure-5: PAS positivity for mucin

CONCLUSION

Mucocele is the most common benign self-limiting condition of the oral mucosa. Majority of such cases can be diagnosed clinically; however biopsy is required sometimes to rule out any other lesions. Management of Mucocele becomes challenging because of high possibility of recurrence, however if no spontaneous regression occurs surgical excision with dissection of surrounding and contributing minor salivary gland can lead to clinical success without recurrence and a better prognosis.

REFERENCES

1. Singh RK, Singh A, Vivek R, Tripathi AA. Mucocele: Review and a Case Report. Healtalk 2012; 4(3): 38-39.
2. Ata-Ali J, Carrillo C, Bonet C, Balaguer J, Pefiarrocha M. Oral mucocele: review of the literature. J Clin Exp Dent 2010; 2(1): e18-21.
3. Marathe S, Hebbale M, Nisa SU, Harchandani N. Oral Mucocele: Presentation at a Rare Site with Review. International Journal of Advanced Health Sciences 2014; 1(4): 14-18.
4. Paremala K, Radhika MB, Thambaih LJ. Myxoglobulosis of lower lip: Report of two cases. J Oral Maxillofac Pathol 2011; 15(2): 232-35.
5. Pina AR, Almeida LY, Andrade BAB, Leon JE. Clear cell change in a lower lip mucocele. J Oral Maxillofac Pathol 2013; 17(2): 318-22.