INTRODUCTION

Mucoceles are one of the most common benign soft tissue lesions of the oral cavity which often results due to local trauma and consequent rupture of salivary gland ducts, especially in the lower lip.[1] They represent a localized accumulation of saliva that is classified into two types based on its pathogenesis. The term mucus extravasation phenomenon is used when there is mucin leakage into the connective tissue from a severed minor salivary duct. Whereas, mucus retention cyst is accumulation of mucin within the lumen of a distally obstructed salivary duct.[2] Apart from the conventional classification, other variants are also seen: (1) superficial mucoceles, (2) mucoceles with myxoglobulosis and (3) mucoceles exhibiting papillary synovial metaplasia like change.[3] The case reported in this article, reveals an extravasation mucocele showing unique globular organization of mucous content representing myxoglobulosis.

CASE REPORT

A 40-year-old female patient was referred for evaluation of an asymptomatic swelling on the lower lip of 6 months duration with a history of local trauma. Clinical examination revealed a well-defined, nontender, smooth-surfaced, roughly oval, fluctuant swelling [Figure 1]. No relevant medical history was elicited. Oral hygiene was fair. A provisional diagnosis of mucocele was made; the lesion was excised under local anesthesia. Histopathological examination revealed a mucus extravasation cyst having lumen filled with unique mucinous globular structures similar to a rare variant of appendiceal mucocele, myxoglobulosis.

Keywords: Mucocele, mucus extravasation cyst, myxoglobulosis
calcifications [Figure 4]. The granulation tissue forming the cystic wall was highly cellular, consisting of chronic inflammatory cells. The mucinous globules showed positive results for PAS and Alcian blue [Figures 3 and 4].

DISCUSSION

Mucus extravasation phenomenon most commonly affects children or young adults, aged between 10 and 30 years. About 80%–90% cases are observed in the lower lip as an asymptomatic swelling with translucent or bluish hue.[2] Histologically, the presence of a well-demarcated interstitial mucin surrounded by a granulation tissue containing neutrophils and multinucleated giant cells in the submucosa serves as a peculiar feature to diagnose mucus extravasation phenomenon.[2] Myxoglobulosis or caviar appendix is a variant of mucocele characterized by the presence of opaque pearl-like globules composed of mucoid material. They are generally 2–3 mm in diameter and form clusters that appear like “fish eggs” or frogspawns.” The first case of myxoglobulosis was accidentally discovered during a postmortem examination by Latham in the year 1897.[4]

In the present case, overall clinical and histopathological features were suggestive of an extravasation mucocele. Albeit, the diagnosis of extravasation mucocele is usually straightforward, the presence of globular structures within the lumen is unique and this condition is referred to as myxoglobulosis. The globules present in this case showed mildly cellular core with peripheral laminations of dystrophic calcifications. This might be a result of a long-standing lesion which has become less cellular over a period of time as stated by KA Shah.[3]

The precise etiology and pathogenesis of myxoglobulosis remain unknown. However, the etiological factors for myxoglobulosis of the intestine or appendix, as suggested by...
Probstein and Lassar include bacteria and necrotic epithelial debris which represent the focus of infection for mucin pooling. An uncommon histologic feature was observed in the lower lip mucocele which bore a resemblance to globules observed in a variant of mucocele of the vermiform appendix by Li et al. in 1997 and thus was described as “myxoglobulosis.” The globules represent an attempt to organize the mucin by the granulation tissue capsule, which is then expelled into the lumen because of persistent mechanical stresses.

Ide and Kusama proposed that the formation of these pearl-like globules of salivary mucocele develop as a result of a rich reparative interaction of the capsular granulation tissue in response to the intraluminal pooling of mucin. Despite the etiology, the significance of myxoglobulosis is exclusively academic. Mucus extravasation phenomenon associated with myxoglobulosis does not reveal distinguishing clinical features, nor is the prognosis any different from conventional types. Treatment includes excision of salivary gland and associated duct along with the pool of extravasated mucin to avoid recurrence. Regardless of not being clinically significant, myxoglobulosis continues to be a remarkable phenomenon for its relative rarity.

Acknowledgement
I am sincerely thankful to D.Y. Patil Dental College for providing me the opportunity to write a case report on the above topic.

I am also thankful to Dr. Treville Pereira for guiding me in every stage of this paper. Without his support, it would’ve been extremely difficult for me to prepare the paper so meaningful and interesting.

I am also thankful to Dr. Subraj Shetty who has helped me during this course of paper in several ways.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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