Case Report

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Myxoglobulosis: A Rare Histopathological Variant of Oral Mucocele in an Adolescent

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ABSTRACT

Background: Myxoglobulosis, is characterized as a rare variant of appendiceal mucocele. Few cases have been reported indicating the occurrence of myxoglobulosis in extravasation mucocele of oral cavity. Morphologic studies including series of case reports were carried out to identify prevalence and possible pathogenesis of this unusual variant. In present paper, a case report of extravasation mucocele showing an unusual intraluminal globular organization of lumen was described. A 16-year-old male adolescent patient with a chief complaint of painless swelling of the lower lip of approximately 2 years duration was reported. It was surgically excised and on histological examination, revealed a well demarcated cystic lumen with numerous mucinophages and globules of varying sizes. Clinicopathologically, it was diagnosed as myxoglobulosis variant of oral mucocele. Even though, these variants tend to occur infrequently, it is imperative to recognize them to avoid misdiagnosis.

Keywords: Oral Mucocele, Myxoglobulosis, Appendiceal Mucocele.

INTRODUCTION

Relatively common disorders that affects major and minor salivary glands are obstructive disorders. Mucocele is one such pathological entity, that mainly evolves due to the severance of minor salivary gland duct. Two types of pathological alterations are recognized i.e., mucous extravasation phenomenon and mucous retention phenomenon. As lesion progresses, extravasated mucin in lumen of minor salivary gland ducts gets obliterated with the progressive growth of granulation tissue, which is termed as “Organizing mucocele”.(1)

Several unusual mucocele variants were reported in literature that includes myxoglobulosis and papillary synovial metaplasia like change.(2-5) Myxoglobulosis variant is an uncommon phenomenon and was first described in the context of mucoceles of the vermiform appendix.(6) It is characterized by eosinophilic globular structures either within the lumen or budding from the wall of oral mucoceles.(7) This article presented a case of extravasation mucocele of the lower lip showing the unique pattern of myxoglobulosis.

CASE REPORT

A 17-year-old male patient was reported to a private dental clinic with a painless swelling on lower lip of 2 years duration. The lesion was asymptomatic and gradually increased to a size of 1×2 cm approximately. No relevant medical history with any specific record of systemic diseases. Patient was not aware of any antecedent local trauma that elicits such lesion. Clinical examination revealed that the lesion was dome- shaped, smooth

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surfaced and softer in consistency with no fluid discharge. The lesion was then surgically excised under local anaesthesia, cut surface showed a cavity filled with yellowish, semi-solid like material. Histopathological examination revealed a well demarcated cystic lumen surrounded by fibro-cellular connective tissue capsule with no evidence of lining epithelium. The cystic lumen showed numerous mucinophages and globules (approximately 70-80) of varying sizes (small to large), detached from each other (Figure-1). The globules are predominantly polygonal to round, with central core of eosinophilic, amorphous, acellular material surrounded by mucinous material (Figure-2). No capillaries and calcifications were evident within globules. The cystic capsule is predominantly fibrocelluar, consisting of fibroblasts and chronic inflammatory cells chiefly mucinophages. The adjacent minor salivary gland shows focal areas of degeneration with loss of architecture of the acini.

DISCUSSION

Based on the findings, the lesion was suggestive of mucous extravasation phenomenon, however the presence of unusual numerous eosinophilic globules of varying sizes posed great challenge in histopathological diagnosis. Chi et al., based on their clinicopathological review of 1824 cases concluded that when an extravasation phenomenon that constitutes more than 30 percent or more the volume of cyst with eosinophilic globules would be diagnosed histopathologically as myxoglobulosis.(7) The present case report met the above criteria and based on that diagnosis was carried out.

An unusual histological and peculiar pattern of intraluminal globular appearance was first described by Li et al., in 1997 where the author used to term "myxoglobulosis".(3) The term myxoglobulosis was first described by Latham in 1897 in the mucocele of the appendix.(6) Li et al., Ide and Kusama also reported a case of myxoglobulosis like change in lower lip.(3, 8) In 2011, Paremala and co-workers presented two case reports of myxoglobulosis with similarities in clinical presentation.(9) Li et al., reported that the extent of globules within the lumen was high and total obliteration of intraluminal cavity was observed. But the lesion in present case report showed free floating intraluminal globules, which is similar to the findings presented by Ide and Kusama.(8)

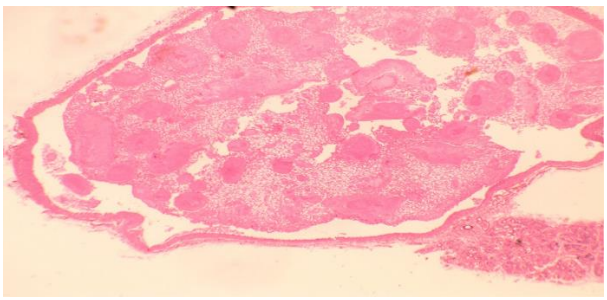

Fig 1: Photomicrograph showing cystic lumen showing mucinophages and numerous globules of various sizes that are detached from each other (16X).

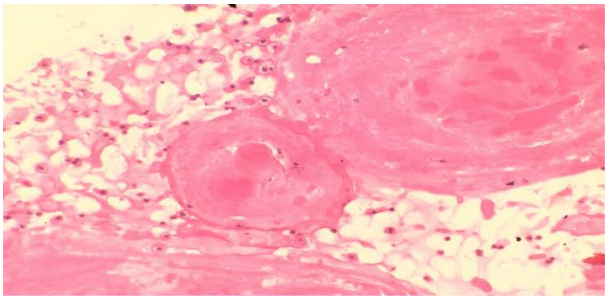

Fig 2: Photomicrograph of cystic lumen with globules having central eosinophilic, amorphous, acellular material surrounded by mucinous material (100 X)

The factors leading to transformation of mucin into globular structures were still unclear. Probstein and Lassar stated bacterial and necrotic epithelial debris may act as nidus for concentric deposition of mucin (10) whereas, Lubin and Berle proposed that the core of globules represents an organizing mass of mucin by granulation tissue originated from appendiceal wall.(6) Li et al., stated that extrusion of granulation tissue into lumen due to frictional or mechanical forces occurs as an attempt to organize mucin. Ide and Kusama believed that prolonged and extensive mucin extravasation from the salivary gland results in enlarged mucin- filled pseudocyst, and any localized changes in microenvironment might trigger the formation of these florid globules. They also stated that in terms of similarity to appendiceal lesions, myxoglobulosis of salivary mucocele seems to occur as a result of exuberant

reparative process of the capsular granulation tissue in response to mucin.(8)

Shah KA in his study to identify the possible pathogenesis of myxoglobulosis in oral mucocele, he identified 11 cases with features of globular structures out of 76 oral mucoceles.(4) He concluded that the histological changes observed in extravasation mucocele was due to disruption of salivary duct, resulting in extravasation of mucin which in turn generates host response with neutrophils initially followed by macrophages. The extrusion of acellular and cellular material in the connective tissue results in separation of the collagen fibres surrounded by macrophages giving a globular appearance. The resulted globular appearance is either mechanical due to mucin or enzymatic due to macrophage action.(4)

In present case report, we observed that globules in granulation tissue wall are cellular, but globules in lumen are acellular. This is in accordance to the findings of Shah KA and Paremala et al., for which the former stated that it was a time related phenomenon with earlier globules more cellular.(4, 9)

So far, even though large case series oral mucoceles were presented, there is no single mention of unusual mucocele variants. One important reason is the failure of the pathologists to recognize the nature of myxoglobulosis, because often these mucocele variants present classic histopathologic findings of a mucocele that could easily baffle a pathologist. Therefore, it is important to recognize these variants to avoid misdiagnosis and more case reports are required to accumulate data on its incidence and to ascertain its precise nature in oral region.

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interests regarding this article.

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