J. Oral Diag. 2019; 04:e20190012

CASE REPORT

Tongue mucocele and microlith associated with a phlebolith mimicking a hemangioma: A unique case report with its surgical management

Nyer Firdoose C.S. [1]*

Abstract:

Unique variants of oral mucocele such as the mucocele appearing on tongue are considered to be extremely rare. It is important to recognize these unique variants promptly to avoid misdiagnosis. The cases of phleboliths not associated with vascular lesions are also rarely reported in the literature, especially as solitary nodules. Conclusion: The following manuscript thereby attempts to present a unique case of a geriatric patient with a combination of Tongue mucocele with microlith mimicking a hemangioma associated with phlebolith along with its diagnosis, clinical presentation and surgical management.

Keywords: Hemangioma; Mucocele; Tongue.

[1] Riyadh Specialized Centre, Department of Oral & Maxillofacial Surgery Ishbiliyah, 13226, Riyadh, Kingdom of Saudi Arabia.

Correspondence to:
Nyer Firdoose C.S.
E-mail: drnyeromfs@gmail.com

Article received on April 18, 2019.
Article accepted on June 7, 2019.

DOI: 10.5935/2525-5711.20190012

INTRODUCTION

Mucocele by definition is cavity filled with mucus (muco - mucus and coele - cavity)[1]. It can be either retention type or extravasation type; the retention type is common in geriatric patients. They are a resultant of narrowed ductal opening caused by a sialolith or a mucus plug[2]. They are usually found in the upper lip, the palate, or the floor of the mouth[3]. Phlebolith formation is a characteristic of hemangiomas due to the result of slow circulation with subsequent thrombus formation and eventual calcification. They are caused by blood stasis[4] or trauma[5], as calcified thrombi are usually associated with vascular lesions, such as vascular malformations.

Mucoceles appearing on the dorso-lateral surface of tongue are extremely rare; there are only a few such cases in the available literature. The cases of phleboliths not associated with vascular lesions are rarely reported in the literature, especially as solitary nodules. The following manuscript thereby attempts to present a unique case of a geriatric patient with a combination of Tongue mucocele with microlith mimicking a hemangioma associated with phlebolith along with its review, diagnosis and surgical management.

CASE PRESENTATION

A 67 year old Saudi male patient presented to our department of Oral and maxillofacial surgery with chief complaint of soft swelling over his dorsal surface of tongue extending to the lateral border towards the right side. He also complained of alterations in his speech pronunciations.

Clinical presentation: A painless dome-shaped fluctuant bluish-coloured swelling sized 2.5cms x 2cms was seen on the right dorso - lateral side of the tongue with no pulsation or palpable hard nodules Figure 1- A, B. The lesion was pink with blue tinge, non pulsating and depressible. Patient gave history of similar lesion on the same part of tongue which was excised previously two decades ago after which the present lesion developed within two months after the initial excision which started as a small nodule growing up to the present size.

The duration of the present lesion was approximately 20 years thereby making it a slow growing lesion. The previous lesion was confirmed to be a hemangioma as per the patient's history. Thus the clinical picture of the lesion was suggestive of a recurred hemangioma as a provisional diagnosis.

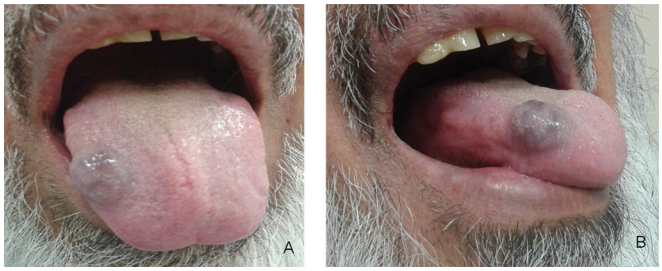

Figure 1. A: showing dorsal view of tongue at initial presentation B: showing lateral view of the lesion.

Surgical Management: Considering the previous history of recurrence of the lesion, the decision to surgically excise was made. A longitudinal elliptical incision was placed covering the entirety of the lesion Figure 2 - A, B. Surgical excision of the lesion was initiated by completely unroofing the lesion along its entire periphery, splitting the overlying mucosa and then resecting the lesion completely from the base. Adequate haemostasis was achieved and closure was done using 4.0 vicryl Figure 3.

It was worth noticing that the lesion was not attached to the underlying muscles. The lesion was then sectioned into two parts after its excision en toto which revealed clear fluid encapsulated within a clear cystic lining with presence of a sub - centimetric lith Figure 4- B.

An additional dark red coloured micro - lith was attached to the external surface of the lesion which was 3 x 3mm approximately Figure 4-A.

Histopathological picture: The excised tissue was subjected to histopathaological analysis which led to the confirmation of the lesion as being a mucocele. The H & E section shows Stratified squamous epithelium of varying thickness, underlying connective tissue stroma is fibrocellular with areas of mucous pooling with mixed inflammatory cell infiltrate Figure 5- A. The histopathologic section shows parakeratinized stratified squamous epithelium. The underlying connective tissue is loose & fibrillar with pseudocystic space containing mucinophages and few lymphocytes. The other red lith was confirmed to be a phlebolith. The H & E section shows the concentric calcification caused by repetitive mineral deposition Figure 5-B.

Follow-up: The patient was reviewed for every 3 weeks interval for up to 6 months Figure 6. During follow-up review, prognosis was excellent, and no recurrence was found.

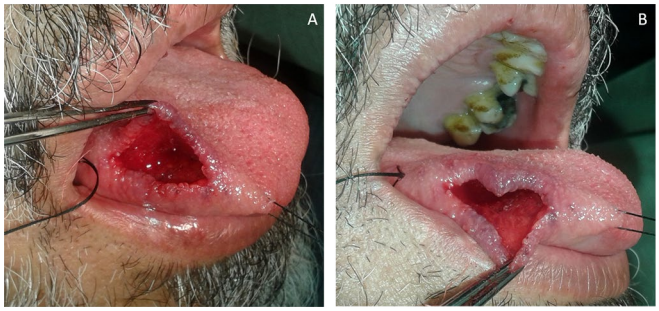

Figure 2. A -B: Intra operative view showing no muscular attachment of the lesion.

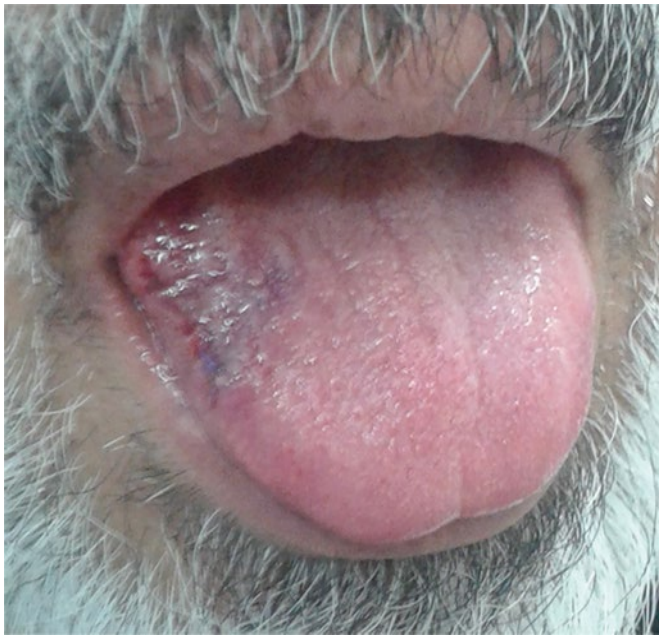

Figure 3. Immediate post operative.

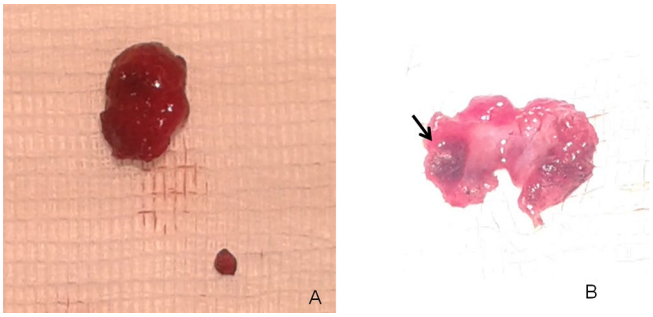

Figure 4. A: showing excised lesion with associated phlebolith B: showing lesion dissected into half with presence of another small lith inside (black arrow).

DISCUSSION

Mucoceles on the tongue are rare and occur almost exclusively on the ventral surface where the glands of Blandin and Nuhn are located. The mucocele is located directly under the mucosa (superficial mucocele), in the upper submucosa (classic mucocele), or in the lower

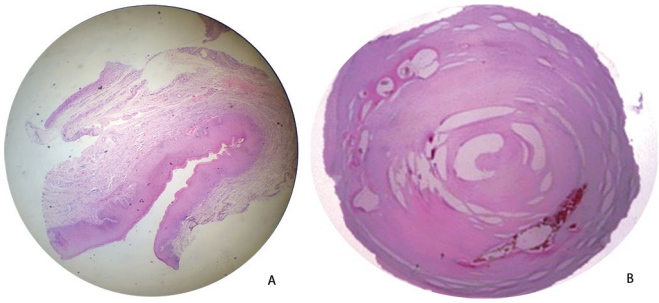

Figure 5. A. H & E section shows Stratified squamous epithelium of varying thickness, underlying connective tissue stroma is fibrocellular with areas of mucous pooling with mixed inflammatory cell infiltrate. **B.** H & E section showing concentric calcification caused by repetitive mineral deposition and lamellar fibrosis.

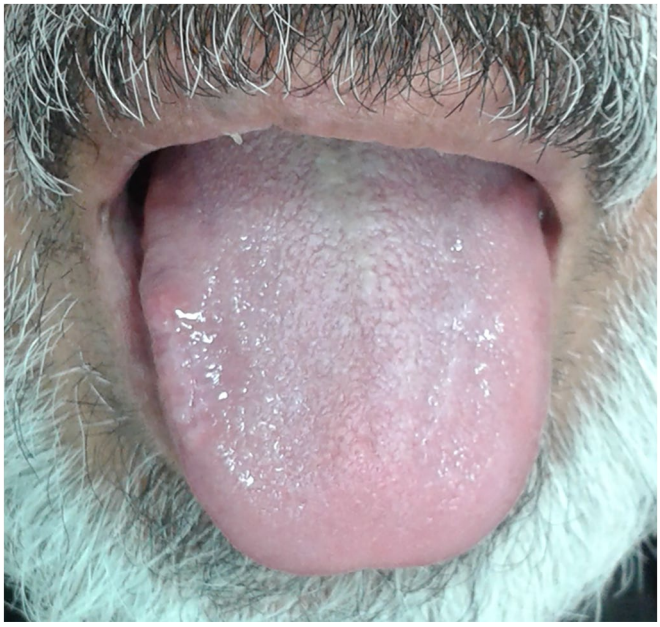

Figure 6. Six months post-operatively.

corium (deep mucocele)[1]. The clinical presentation of these lesions depends upon their depth within the soft tissue and the degree of keratinisation of the overlying mucosa superficial lesions present as raised soft tissue swelling that is translucent and having bluish colour, whereas the deeper lesions are more nodular, lack the vesicular appearance, and have a normal mucosal colour[1]. Previous available literature and earlier studies suggest that extravasation phenomenon is far more common than retention, and extravasation mucocele showed a definite male predominance which most frequently is noticed in the second and third decade of life. Mucoceles are usually asymptomatic, though in some patients they can cause discomfort by interfering with speech, chewing or swallowing[6].

Phleboliths are not unusual in the head and neck, Phleboliths of the oral region can be found in infants

to elderly individuals, but mainly between the first and third decades of life (55.2%), with no sex predilection (48. 28% female: 51.72% male), and are sometimes associated with masticatory muscles (27.6%)[7].

There are few cases in the literature of phleboliths not associated with other vascular anomalies[8]. When a phlebolith is located in salivary gland regions, it can be clinically misdiagnosed as a sialolith or salivary gland disease, especially when there is intermittent swelling, despite not being associated with food intake[9].

Cases of Tongue mucocele is very rarely reported so are the phleboliths not associated with other vascular lesions which again is an entirely unique entity in the literature, especially as solitary lesion with a micro-lith associated with a phlebolith mimicking a hemangioma. The author thereby reports such a rare entity prone to get misdiagnosed and may lead to the final diagnosis which is completely different from the provisional diagnosis reached with the deceiving clinical presentation.

CONCLUSION

Unique variants of oral mucoceles occur infrequently, and the clinician is not cognizant with the possibility of such lesions at unusual sites, it is necessary to sort the dilemma by seeking histopathological assessment and avoid misdiagnosis. A wise oral maxillofacial surgeon should always subject the lesion to histopathological confirmation owing to its close resemblance to neoplastic lesions, vesiculobullous lesions. Moreover hemangiomas closely mimic mucocele and thus requiring a meticulous histopathological examination of all the excised mucoceles. A clinician should be curious to intervene if there is more to it than that meets the eye always, before concluding the diagnosis to execute the best treatment plan for the betterment of the patient.

CONFLICT OF INTEREST

None declared.

ACKNOWLEDGEMENTS

The author thanks the patient for his kind co-operation.

REFERENCES

1. Baurmash HD. Mucoceles and Ranula. J Oral Maxillofac Surg. 2003; 61:369-78. PMID: 12618979 [DOI:10.1053/joms.2003.50074].
2. Jinbu Y, Kusama M, Itoh H, Matsumoto K, Wang J, Noguchi T. Mucocele of the glands of Blandin-Nuhn: clinical and histopathologic analysis of 26 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2003; 95(4):467-70. doi:10.1067/moe.2003.51.
3. Greebberg MS. Salivary gland disease. In: Lynch MA (ed.). Burket's Oral medicine: diagnosis and treatment. Philadelphia: Lippincott-Raven. 1997; 415-8.
4. Altuğ HA, Büyüksoy V, Okçu KM, Doğan N. Hemangiomas of the head and neck with phleboliths: clinical features, diagnostic imaging, and treatment of 3 cases. Oral Surg, Oral Med, Oral Pat, Oral Rad Endodont. 2007; 103(3):e60-e64. http://dx.doi.org/10.1155/2015/507840.
5. Mandel L, Perrino MA. Phleboliths and the vascular maxillofacial lesion. J Oral Maxillofacial Surg. 2010; 68(8):1973-6.
6. More CB, Bhavsar K, Varma S, Tailor Mansi. Oral mucocele: A clinical and histopathological study. J Oral Maxillofac Pathol. 2014 sep; 18(Suppl 1):S72-S77.
7. Kurita H, Chino M, Kurashina K, Kotani A. Phlebothrombosis with phlebolith of the tongue. Oral Surg, Oral Med, Oral Patol. 1994; 77(6):552.
8. Zachariades N, Rallis G, Papademetriou J, Konsolaki E, Markaki S, Mezitis M. Phleboliths. A report of three unusual cases. Brit J Oral Maxillofacial Surg. 1991; 29(2):117-9.
9. Lima GMG, Moraes RM, Cavalcante ASR, Carvalho YR, Anbinder AL. An Isolated Phlebolith on the Lip: An Unusual Case and Review of the Literature. Case Reports in Pathology. 2015; Article ID 507840, 5 pages.