

Oral hemangioma mimicking as a mucous retention cyst- A case report and the review of literature

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ABSTRACT

Introduction: Oral mucocele (OM) is the most common benign minor salivary gland lesion. Characterised by single or multiple, soft, fluctuant nodule, ranging from the normal color of the oral mucosa to deep blue.

Case presentation: 47 years old female reported to our hospital with complain of swelling and discomfort in right cheek since 3-4 years. She had a single swelling of 3x3cm, soft to firm in consistency, non pulsatile, non compressible and not adherent to the underlying tissue. The overlying mucosa was smooth, non pigmented with no reddish or bluish discoloration and provisional diagnosis of Oral mucocele was made. She was operated under general anesthesia where incision was made in buccal mucosa, parallel to the occlusal line and avoiding the stensen duct opening. The entire mass was removed in toto without any breach. Intraoperatively no obvious feeder vessel was identified. Histopathological examination showed numerous varying sized ectatic channels lined with flattened endothelium and these channels were filled with RBCs, diagnosis of Oral Hemangioma was made. A detailed mention of differential diagnosis and a brief literature review are also done.

Conclusion: Oral hemangioma (OH) are rare lesion in buccal mucosa, it can mimic as OM as both presents as soft fluctuant swelling with smooth overlying normal appearing mucosa. Treatment of mucocele is straightforward and complete excision of the lesion under local anesthesia is the most common practice. However, OH are highly vascular lesion with high predictability of bleeding when ruptured. Radiographic Investigations like MRI or ultrasound is necessary to rule out any vascular lesion or intra cranial involvement. The critical step is the removal of this lesion. Excision has to be done meticulously and its best excised under general anesthesia with facility of ligation and electro cautery.

INTRODUCTION

Oral hemangiomas (OHs) are benign tumors that occur in and around the oral cavity due to endothelial cell proliferation. OHs are relatively rare and most frequently involve the lips, tongue, buccal mucosa, and palate. The majority of hemangiomas will completely involute, with 10%-20% persisting into adolescence or adulthood.ⁱ While medical, interventional, and surgical regimens are available, there is no standardized treatment for OHs. Due to myriad potential complications, treatment of OHs is typically not pursued unless functional impairment exists. Treatment of choice depend on several factors including the age of the patient, size and extent of the lesions, as well as their clinical characteristics.

Oral mucous retention cyst (OMs) is the most common benign minor salivary gland lesion, usually caused due to mechanical trauma to the excretory duct of the gland. They are characterized by single or multiple, soft, fluctuant nodule, ranging from the normal color of the oral mucosa to deep blue.ⁱⁱ The superficial lesions appear as thin-walled, bluish swellings that rupture easily while the deeper lesions are well circumscribed swellings usually covered by normal appearing

oral mucosa.ⁱⁱⁱ However, a skillful removal is required as the overlying mucosa might rupture, resulting in discharge of mucous into oral cavity, and might hinder appreciation of margins of the lesion and its complete excision. Clinically, OHs in buccal mucosa can mimic a deep OMs as both presents as soft fluctuant swelling with smooth overlying normal appearing mucosa. Both OMs and OHs might have a past history of trauma. Treatment of mucocele is straightforward and complete excision of the lesion under local anesthesia is the most common practice. However to diagnose OHs, a detailed investigation like ultrasound, CT scan or MRI is necessary. The excision of OHs needs to be done carefully due to its vascular nature and high predictability of bleeding. Hence it is best excised under local or general anesthesia, depending on its size, and with the availability of option of cauterization, embolization and local measures.

CASE DESCRIPTION

A 47 years old female reported to our hospital in March 2022 with the complaint of swelling and discomfort in the right cheek since 3-4 years. She had history of trauma from cheek bite in that area 6 years ago. Patient noticed a pea sized swelling on the inner side of right cheek 2 years post trauma, which gradually increased over the years and reached to the size of a small lemon. She had no history of bleeding from the lesion. Due to the prominence of the lesion, it was causing her discomfort while talking and chewing. Past medical and family history was non-contributory. On examination, she had a single swelling of 3x3cm, soft to firm consistency, non-pulsatile, non-compressible and non-adherent to the underlying tissue. The overlying mucosa was smooth and had no reddish discoloration. There was no obvious increase in temperature of lesion. No obvious feeder vessel was identified. No obvious bleeding/ pulsations were identified on palpation of the lesion. There was no associated lymphadenopathy. OH was not suspected preoperatively. Provisional diagnosis of mucous retention cyst of right buccal mucosa was made. Other differential diagnosis was vascular malformation, pyogenic granuloma, angiomyolipoma, angiosarcoma or hemangiosarcoma. MRI was done to rule out cystercercosis or any intracranial involvement. MRI finding revealed well defined cystic lesion along the deep subcutaneous fat, superficial to buccinators and anterior to masseter muscle and located deep to the right platysma [Figure 1].

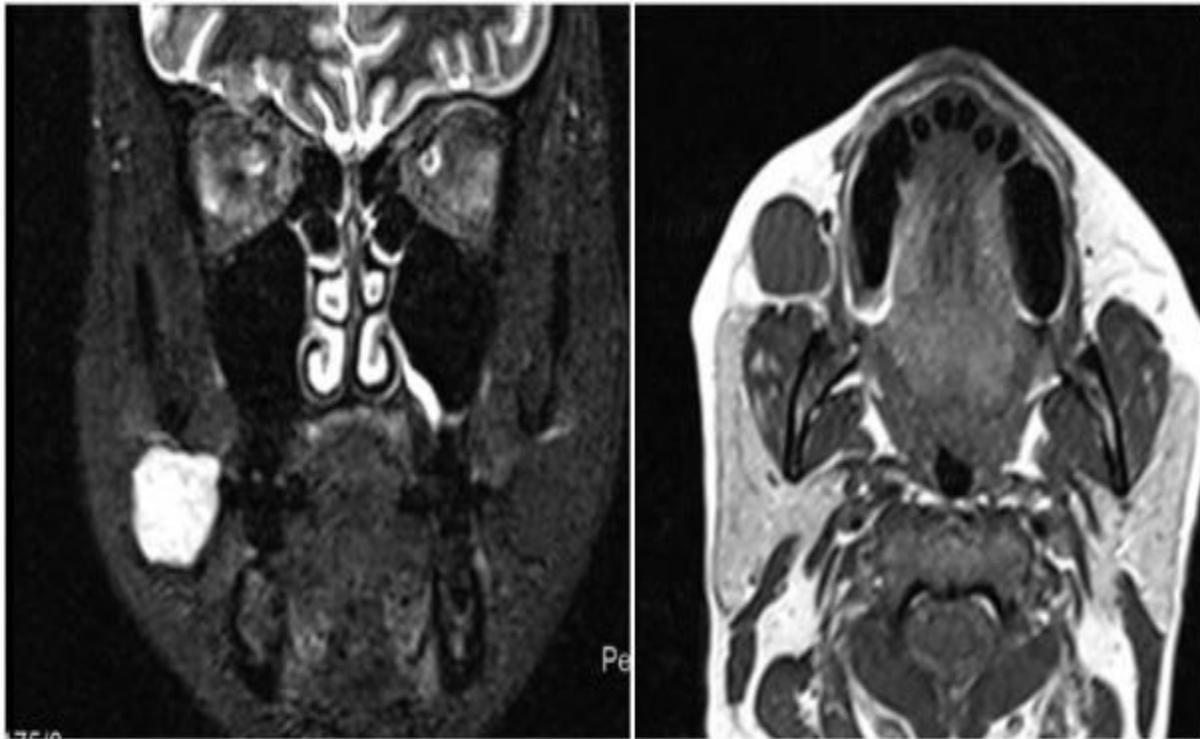


Figure 1- MRI Images

The findings suggested a benign cystic lesion, with no intra-cranial involvement. The patient was admitted in day care and operated under general anesthesia where incision was made in buccal mucosa, parallel to the occlusal line and avoiding the Stensen's duct opening. The entire mass was removed in toto, without any breach. Intraoperatively no obvious feeder vessel was identified. Mild to moderate bleeding was encountered after the excision which was managed by cauterization. Gross specimen was soft in consistency, well circumscribed, reddish-blue in color and approximately of size 2.5×3 cm [Figure 2].



Figure 2- Excised Tumour Mass

Specimen was sent for histopathological examination. The cut surface was grayish brown and congested. The tumor was composed of numerous varying sized ecstasic channels lined by flattened endothelium. These channels were filled with RBCs. Histopathologically, the diagnosis of Oral Hemangioma was made.[Figure 3]. On one year followup, patient was asymptomatic with no sign of recurrence.

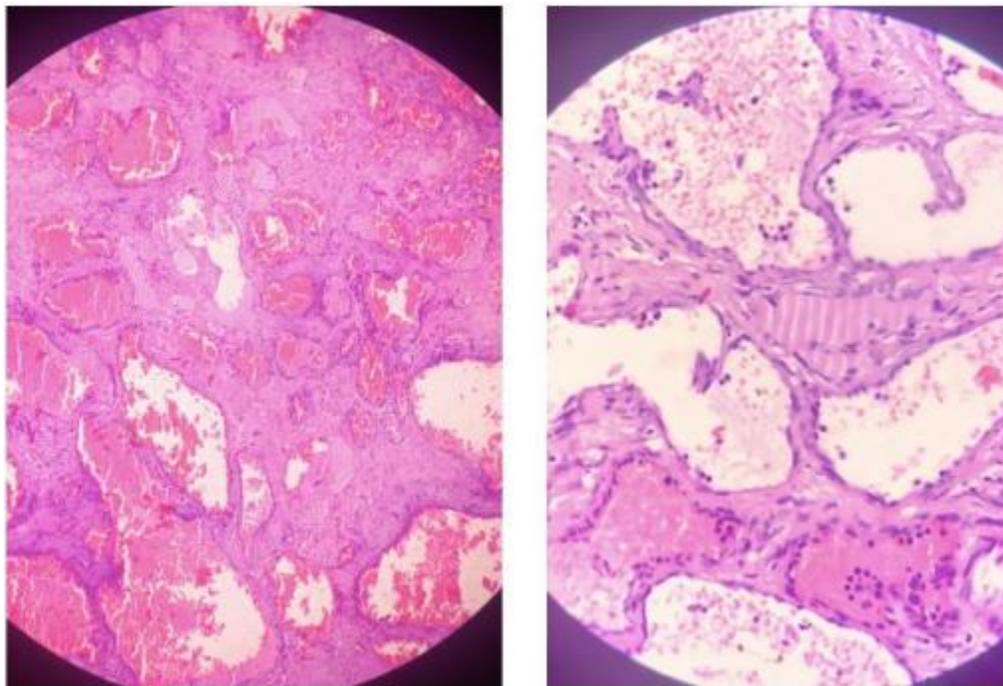


Figure 3- Histopathological Images

DISCUSSION AND REVIEW OF LITERATURE OF ORAL HEMANGIOMA

Oral Hemangioma is a rare benign tumor of head and neck area, has a prevalence of 6.4% in infants, with limited data available to estimate prevalence in older age groups^{iv}. It usually remains asymptomatic for up to two or three decades. They

become palpable after a sudden growth and the swelling is the most common initial presentation.^v In our case, OH was not suspected preoperatively. Because a vascular lesion may not be clinically suspected, there is an increased risk of bleeding complications during biopsy procedures, therefore it is advised to get ultrasound or MRI before any intervention. If the size of lesion is small, it is best to surgically remove it in-toto thus reducing the bleeding complication. The clinical differential diagnosis includes several lesions, such as vascular malformation, salivary gland and benign mesenchymal neoplasms, mainly because of its non-specific clinical features.^{vi} In 1972, Allen and Enzinger made classification of OH as three histological subtypes: small vessel (capillary), large-vessel (cavernous) and mixed subtype (cavernous and capillary).^{vii} Metaplastic calcification or Phlebolith may be present in some cases, usually cavernous hemangioma or AV malformation. The diagnosis is based on the typical histopathological features.^{viii} The lesion is composed of a well formed but often immature vascular structures lined with plump endothelial cells. The endothelial cells are amphophilic or eosinophilic, often with vacuolated cytoplasm. The large, round and vesicular nucleus with open chromatin pattern often protrudes into the lumen. The vessels are surrounded by an inflammatory infiltrate which contains lymphocytes, eosinophils, plasma cells, histiocytes and scattered mast cells. In our case, the tumour was composed of numerous varying sized ecstatic channels lined by flattened endothelium and filled with RBCs.

The histological differential diagnosis of OH includes mainly angiosarcoma, Kaposi sarcoma, angiomas and infiltrating angiolipoma.^{vii} In the differentiation of angiosarcoma and OH, some important features are present in malignancy, such as nuclear hyperchromatism, pleomorphism, the presence of multinucleated endothelial cells, as well as necrosis and solid growth of malignant cells. Due to its similarity to some cases of angiosarcoma, acantholytic squamous cell carcinoma may rarely be considered in the differential diagnosis. The initial lesions of Kaposi sarcoma are composed of the proliferation of numerous capillaries, but they differ from OH, because there is no muscular infiltration. HHV-8 immunohistochemistry is also helpful in this differentiation, associated with clinical correlations, such as endemic presentation. With a histological appearance similar to OH, angiomas is a rare condition, usually presenting during childhood, in which several segments of the body are involved by vessels that are proliferating^{ix}. The infiltrating angiolipoma is considered as an OH with abundant adipose tissue. Because oral lesions are often subjected to local trauma, worrisome inflammatory reactive changes can be present, and should be also taken into consideration in the differential diagnosis. Being a soft tissue lesion, MRI presents better detection and delimitation of lesion extension than computed tomography. OH presents as a hypo-intense lesion in T1-weighted and greater signal intensity in T2-weighted images. Also the lesion has poorly defined T1 margins and well limited T2-weighted margins. It may also present rounded hypo-intense foci that can represent a cross-section of fibrofatty septa, smooth musculature, ossification, or vascular channels with thrombus formation^x.

Several treatment options for OH have already been studied, such as cryotherapy, sclerotherapy, radiation, lasers and corticosteroids, each with their advantages and disadvantages, but the main treatment is surgical excision.^{xi} Local recurrence, ranging from 9% to 28%, has been reported and is usually associated with incomplete surgical resection.^{vii}

In conclusion, OH is an uncommon benign vascular proliferation in the oral cavity. The clinical appearance may be non-specific, leading to unexpected bleeding complications during a biopsy procedure. OH may mimic oral mucocele clinically, therefore it is very important for proper radiological investigation like ultrasound, FNAC or MRI (gold standard) for proper diagnosis.

In case a rupture in the integrity of OH occurs, it may lead to severe hemorrhage, dysphagia, thereby compromising the airway and this might cause failure to thrive. Thus, excision under general anesthesia is a safer modality, compared to that under local anesthesia as it allows securing of airway and rules out patient uncooperativeness.

OH may show microscopic features similar to those observed in infiltrative vascular malignant tumors. Oral OH should be considered in the differential diagnosis of swellings of the buccal mucosa, tongue and lips.

CONCLUSION

Oral mucous retention cyst/ mucocoele might mimic an oral hemangioma clinically. However, proper clinical examination and history, and investigations like MRI or ultrasound can help in framing a provisional diagnosis. Histopathologic examination is the gold standard for framing an accurate diagnosis; however, it does include the risk of bleeding, which might cause airway compromise. Therefore, excision in toto should be considered for such lesions, preferably under general anesthesia so as to minimize the above-said risks. Furthermore, the size of the lesion, patient cooperativeness, availability of general anesthesia, surgeon's preference and patient affordability should be considered.

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