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CASE REPORT

ORAL VASCULAR LESION: A CASE REPORT

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ABSTRACT:
Vascular lesions are tumors or malformations of the vasculature. Vascular tumors are the most common tumors in infancy; it is seen in 1.1 to 2.6% of newborn infants and 10 to 12% of children by the first year of life. There persistence in adulthood however presents as a challenge to the physician. Most of the case reports of vascular lesions in the oral cavity have been presented in relation to tongue, lips and in the palate. Report of classical lesion in the buccal mucosa has not been reported in the near past. Asymptomatic lesions tend to get neglected by patients. In this case report we present one such rare case of a vascular lesion in an adult. Classification of vascular lesions and there treatment is discussed.

Keywords: oral vascular lesion, hemangioma, buccal mucosa.
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INTRODUCTION:

Vascular lesions are tumors or malformations of the vasculature. It is the most common tumor in infancy; it is seen in 1.1 to 2.6% of newborn infants and 10 to 12% of children by the first year of life [1, 2]. There persistence in adulthood however presents as a challenge to the physician [2,3]. The hemangioma involutes with time and symptomatic lesions are effectively managed by various means [2,3]. Most of the case reports of vascular lesions has been presented in relation to, lips, tongue and some in the palate [4,5,6,7]. Asymptomatic lesions tend to get neglected by patients. In this case report we present one such rare case of a vascular lesion intraorally in an adult female patient.

CASE REPORT:

A 49 year old female patient presented to the department of oral medicine and radiology at century dental college with chief complaint of decayed teeth. There were no associated symptoms. Her history did not reveal any significant detail. She was moderately built and nourished with steady gait and normal

cognition. On General and extra oral examination no abnormality was detected. On intraoral examination there was a large purple coloured swelling on the right buccal mucosa. The swelling was sessile with broad base and was located at the line of occlusion opposing upper right teeth region. It measured about 3*2 cm, its surface was smooth and lobulated, rubbery on palpation and did not empty on application of pressure (Figures 1 &2). At the anterior margin of the swelling a pulsatile vessel could be palpated. The lady was aware of the swelling and said that it was present since 20- 25 years; it had waxed and waned in size. It never bled nor was it associated with any discomfort. In her previous visit to dentists she was advised to ignore it. She was not interested in getting treatment done for the same. We advised her to get an ultrasonography done. She however did not get any investigation done. She was reluctant in even getting a noninvasive diagnostic procedure like diascopy performed. Provisional diagnosis of low flow vascular anomaly and differential diagnosis of Non Involuting Capillary Hemangioma (NICH) and Arteriovenous malformation was made. The patient was advised to be watchful of the same and report to the nearest hospital in case of bleeding from the same.

As in this patient, the lesions are asymptomatic and tend to get neglected. They also refuse to carry out investigations for the same. This makes it difficult to diagnose the lesion accurately.

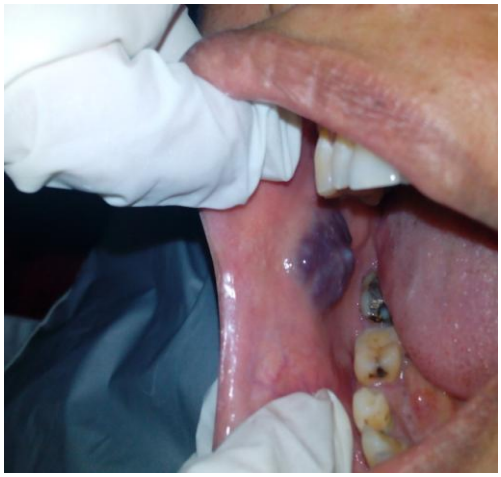

Figure 1 Clinical picture showing raised lesion at the level of occlusion on the right buccal mucosa.

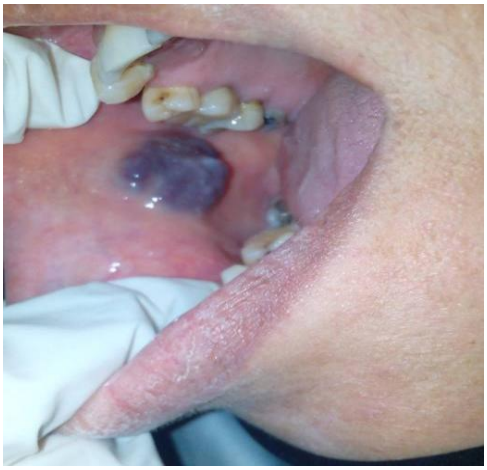

Figure 2. The swelling is dome shaped and purple in colour. The contents of the swelling did not empty on application of pressure.

DISCUSSION:

Vascular anomalies are congenital lesion of abnormal vascular development. They were first classified by Mulliken and Glowacki In 1982 [8] Recent classification of vascular lesions has clarified their diagnosis beyond doubt. Currently they have been described by International Society for the Study of Vascular Anomalies (ISSVA) classification. Revised in 2014 as given below

Vascular anomalies

Vascular tumors	**Vascular malformations**		
	Simple	**Combined**	**others**
Benign Locally aggressive or borderline Malignant	Capillary malformations Lymphatic malformations Venous malformations Arteriovenous malformations Arteriovenous fistula	capillary-venous malformation CVM capillary-lymphatic malformation CLM capillary-arteriovenous malformation CAVM Lymphatic-venous malformation LVM capillary-lymphatic-venous malformation CLVM capillary-lymphatic-arteriovenous malformation CLAVM capillary-venous-arteriovenous malformation CVAVM capillary-lymphatic-venous-arteriovenous CLVAVM	Anomalies of major named vessels (aka "channel type" or "truncal" vascular malformations) Vascular malformations associated with other anomalies Provisionally unclassified vascular anomalies

Benign vascular tumors are further classified as: Infantile hemangioma / Hemangioma of infancy, congenital hemangioma
(a) Rapidly involuting (RICH); (b) Non-involuting (NICH); (c) partially involuting (PICH)
Tufted angioma, Spindle-cell hemangioma, Epithelioid hemangioma, Pyogenic granuloma (lobular capillary hemangioma);

Locally aggressive or borderline vascular tumors: Kaposiform hemangioendothelioma, Retiform hemangioendothelioma,Papillary intralymphatic angioendothelioma (PILA), Dabska tumor, Composite hemangioendothelioma, Kaposi sarcoma

Malignant vascular tumors: Angiosarcoma, Epithelioid hemangioendothelioma

Clinical diagnosis of malignant lesions is challenging. It is usually diagnosed by histopathological examination.

Syndromes associated with vascular lesions:
Blue rubber bleb nevus (Bean) syndrome VM,
Klippel-Trenaunay syndrome: CM + VM +/- LM + limb overgrowth;
Parkes Weber syndrome: CM + AVF + limb overgrowth;
Servelle-Martorell syndrome: limb VM + bone undergrowth Sturge-Weber syndrome: facial + leptomeningeal CM + eye anomalies +/- bone and/or soft tissue overgrowth;
Limb CM + congenital non-progressive limb hypertrophy
Maffucci syndrome: VM +/- spindle-cell hemangioma + enchondroma
Macrocephaly - CM (M-CM / MCAP);
Microcephaly - CM (MICCAP);
CLOVES syndrome: LM + VM + CM +/- AVM + lipomatous overgrowth
Proteus syndrome: CM, VM and/or LM + asymmetrical somatic overgrowth
Bannayan-Riley-Ruvalcaba sd: AVM + VM +macrocephaly, lipomatous overgrowth

VASCULAR TUMORS:

Vascular tumors are common during infancy [8]. The infantile hemangioma is the commonest of the vascular lesions and is prevalent in infancy. It is rarely apparent at birth, evident in the first three months. It gradually involutes. It has been described in literature as going through three distinctive phases: proliferation 1-3 months, quiescence 9-12 months and involution by 5-9 years [1-3,8,9]. They may be superficial, deep and compound. They appear as red or nodular raised lesion. They require intervention only if problematic. Some of the complications that have been reported are: hemorrhage, ulceration, infection and disfigurement [9]. In certain cases depending on the location on compression of vital organs and may cause airway obstruction and cardiac failure [10].

As these lesions involute over time no treatment is required in majority of the cases. Only cases with severe or recurrent hemorrhage or those that interfere with vital structures require treatment. Various therapies with steroid, interferon and vincristine have been tried with variable rates of success. Surgical excision or laser therapy has been used in lesions unresponsive to medication [9]. The recent literature suggests the usage of propranalol with higher safety profile [10].

VASCULAR MALFORMATIONS:

Vascular malformations, by contrast, are present at birth and grow proportionately with the child. They are composed of dysplastic arterial, venous and/or lymphatic vessels rather than proliferating cells. They are named according to the predominant vessel type and are further classified as 'high-flow' and 'low-flow' lesions [9].

Lesions that demonstrate arteriovenous shunting such as arteriovenous malformations (AVMs) and arteriovenous fistulae (AVF) are described as high flow, whereas venous malformations (VMs), lymphatic malformations (LMs) or combined lympho-venous/veno-lymphatic malformations (VLMs), together with CMs are described as low flow. AVM the lesion enlarges due to hormonal changes, infection, trauma and surgical injury.

INVESTIGATION:

Ultrasonography or MRI can be used as the first modality of examination [11]. They help in differentiating between fast and slow flow lesions. Doppler sonography can be used to differentiate AVM from other lesions. MRI helps in defining the extent of the lesion, volume of the affected area. CT however is useful to identify osseous involvement of the lesion and to identify phleboliths. Characteristic shining pearl appearance has been reported in vascular malformations [12]. Ultrasound guided needle placement is utilized for sclerotherapy [13].

TREATMENT:

Pulsed dye laser treatment is the 'gold standard' for capillary malformations. Per cutaneous image guided sclerotherapy has become mainstay in treatment of venous malformation. Various agents have been used such as ethanol, bleomycin, sodium tetradecyl sulfate, polidoconal, OK-432. Of these absolute ethanol have been associated with most complications and least have been noted with sodium tetradeyl sulfate [12,13]. High-power lasers have an excellent therapeutic option for this type of lesion in the oral cavity. Their coagulative properties allow procedures to be done without the risk of bleeding, which promotes a better healing pattern and a differentiated postoperative appearance. Surgical excision combined with intravascular

embolization is treatment of choice for AVM however extensive AVM are still not curable [13].

Review of literature:

There have been many case reports of haemangioma in the literature, although earlier most of venous or capillary malformations were reported as hemangiomas [4,5,6,7]. The current classification of vascular anomalies which is based on clinical, radiological, and histological differences helps in differentiating accurately between vascular tumors and malformations and also helps in predicting prognosis [3].

Vascular lesions of the lip [5,6], tongue and palate[7] have been reported frequently in adult patients. Intramuscular hemangiomas involving masseter has been commonly reported and cases involving buccinators also has been found [14]. Multifocal hemangiom [15] have also found mention in recent literature.

CONCLUSION:

Vascular lesion of the buccal mucosa has not been reported in an adult individual in the recent past. However we are limited in our diagnosis because the patient was not interested in getting any further investigations done or therapy as the lesion had not caused any problem to her. Based on the clinical presentation alone the most plausible diagnosis would be a low flow venous malformation. The aim of this case report is to establish the innocuous presence of this rare benign lesion and also justifies watchful neglect for most of these.

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Conflicts of Interest: The authors have no conflicts of interest to declare.

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