Colour Doppler Imaging in Benign Malformation of Buccal Mucosa- A Case Report

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Abstract

Hamartomas, occurring in oral and maxillofacial region are basically indigenous tissue malformations that arise from epithelial derivatives, mesenchymal derivatives and odontogenic derivates. Vascular anomaly, is a mesenchymal hamartomatous condition- a broader term describing condition of blood vessels including abnormal number, structure and position imparting blue/purple colour to oral mucosa. Such anomalies are broadly classified into two basic types like vascular tumours and vascular malformations.

A clinician should possess skills and knowledge for identifying such lesions and be aware of its diagnosis to avoid any event of uncontrolled bleeding in the dental chair.

Keywords : Hemangioma, Vascular Malformation, Ultrasonography

Introduction:

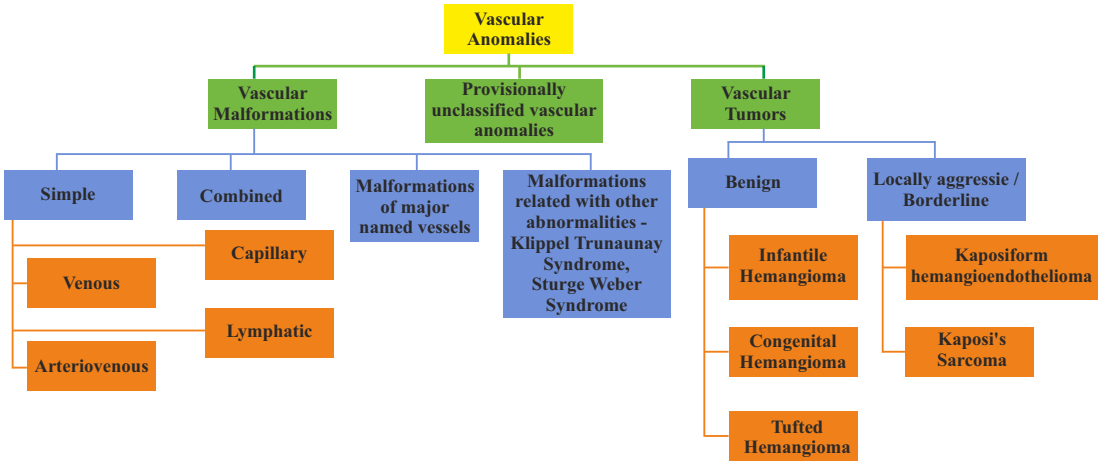
The oral cavity reflects versatility of colours ranging from pinkish colour of the oral mucosa representing a normal healthy tissue to derivatives indicating lesions (physiologic or pathologic) giving red/white appearance, grey/black appearance due to pigmented lesions, brown melanotic/heme-associated lesions and blue/purple vascular lesions.

Vascular anomaly/lesion is a broader term which consist of many conditions of blood vessels including abnormal number, structure, and position.[1] Vascular anomalies are classified into two basic types – Vascular tumours and Vascular malformations, which are often used alternatively.[2] Haemangioma refers to true neoplasm of endothelial cell origin characterized by proliferation and increased rates of endothelial cell turnover whereas vascular malformation refers to a localized defect in vascular morphogenesis with normal rates of cell turnover.[3]

Standardized diagnosis and therapeutic management have been an everlasting challenge to the clinician due to varying symptoms and morphology of the lesion. Over the past few years various terminologies and classifications have been proposed which have only added to the confusion.[4,5] First classification of vascular anomalies was presented by the Father of Cellular Pathology, Rudolf Virchow in 1863 based on microscopic channel architecture. Since then, various classifications have been proposed. The first classification of the International Society for the Study of Vascular Anomalies (ISSVA) was proposed in Hamburg in 1988 and since then it has been modified several times. A good classification that is easy to understand, should be applied by the clinicians as it is very important for classifying information, data recording, communication regarding prognostic information, and guidance of treatment.[6] Nair et al proposed Classification of Vascular Malformations based on anatomy and depth of location in the head and neck region(Table I).[6]

Type	Description
Type 1:	Mucosal/cutaneous – superficial lesions requiring excision of skin/mucosa
Type 2:	Submucosal/subcutaneous – require complete excision after skin flap elevation
Type 3:	Glandular – involves glands of head and neck and excised along with affected glands
Type 4:	Intraosseous lesions – require excision with involved bone and reconstruction when required
Type 5:	Deep Visceral - lesions involving deep visceral spaces like parapharyngeal/ infratemporal fossa

Table I: Classification of Vascular Malformations based on anatomy and depth of location in the head and neck region.



Recent classification (2018) by International Society for the Study of Vascular Anomalies (ISSVA)[2]

Knowledge about various types of vascular anomalies is vital because even though they present rarely in oral cavity, potential risk is involved and complications may arise while providing dental treatment to these patients due to uncontrolled bleeding which is iatrogenic in nature.

Patients may report to our clinic worrying about lumps/growth in the oral cavity and it is crucial that we should be capable of diagnosing and reassuring the patients appropriately. Many patients presenting with low flow lesions should be reassured and educated that the lesions are harmless and only lesions causing contraction, localized thrombosis, and those showing intermittent expansion should be treated[7] To differentiate small vascular lesions from non-blanchable red or pigmented lesion a simple chairside test is used called Diascopy.[8] In this technique pressure is applied to a suspected vascular lesion and blanching of the lesion is noted. (Figure 1) Characteristic blanching of mucosa/tissues seen in diascopy occurs due to dissipation of blood intravascularly under compression giving pale appearance by removing the camouflaging effect of congested blood to reveal a true colour of the underlying lesion.[9]

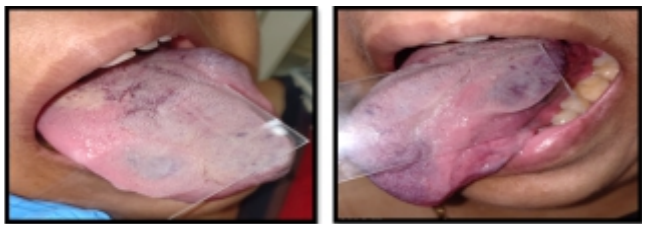


Figure 1: Diascopy- A Chair-side investigation for suspected vascular lesion.

Case report

A 47-year-old female patient was referred to the Department of Oral Medicine & Radiology at our Institute for evaluation of painless purplish intraoral swellings. These swellings appeared during adolescence and persisted since then without any change in their dimension. There were no associated symptoms like pain, change in size, fever, weight loss. No history of any secondary changes like softening, and ulceration. Past medical and family history were non-contributory. Extraoral examination did not reveal any significant findings. There was no associated lymphadenopathy and temperature on the overlying skin was normal. Intraoral picture showed discrete 3-6 lobules bluish – purplish in colour interspersed on the maxillary vestibular area and buccal mucosa, as shown in figure 2, of these 2 were solitary and rest showed coalescing (Figure 2). They were rounded, non-pulsatile and compressible in nature. Diascopy was performed which showed blanching of lesion on applying pressure with glass slab, hence it is considered as a vascular lesion. Differential diagnosis of venous malformation and arteriovenous malformation was given.

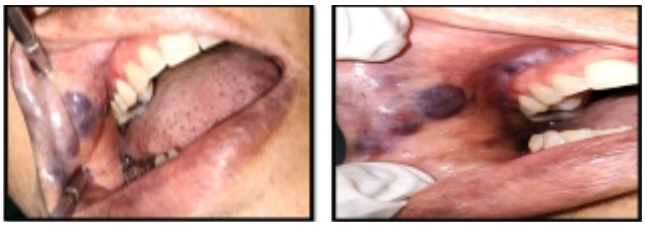


Figure 2: Purplish swelling over upper lip on right side. Multiple soft, non pulsatile,non- tendpurpalish swellings over right er Compressible purplish swellings over right buccal mucosa

Orthopantomogram was taken to rule out any underlying intraosseous involvement. There were no significant findings on OPG as shown in figure 3

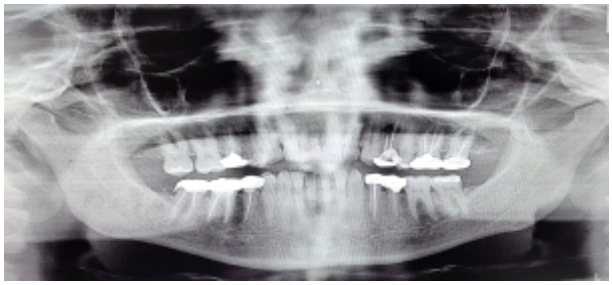


Figure 3: Orthopantomogram

Later colour doppler Ultrasound examination was performed which revealed a diffuse lesion involving entire upper labial and buccal mucosa with respect to central incisor to premolar and extending from mucosa upto subcutaneous tissue plane. It showed multiple tortuous anechoic spaces which were compressible and filled up with slow flow colour on doppler, with spectral features compatible with venous flow as shown in figure 4.

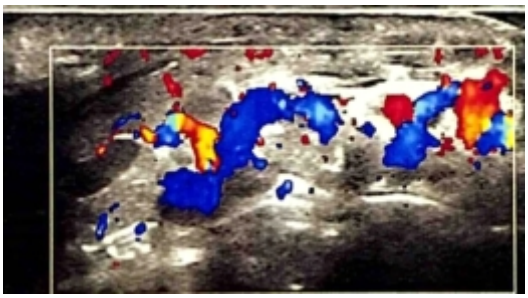


Figure 4 colour doppler ultrasound

Overall ultrasonographic features gave an impression of an unencapsulated slow flow venous malformation. Since the USG report gave the impression of slow flow venous malformation, the patient was reassured regarding the condition and was explained regarding due precautions during dental procedures. The patient refused any treatment as she was asymptomatic.

Discussion –

Vascular lesions are disorders of aberrant angiogenesis, lymphangiogenesis and vasculogenesis are relatively rare in the oral cavity as compared to head and neck[10,11] Vasculogenesis involves growth of vessels from mesoderm-derived embryonic precursors like hemangioblasts giving rise to endothelial precursors angioblasts and blood cell precursors hemocytoblasts. Fusion of angioblasts takes place in vascular islets inducing the formation of primary capillary plexus which extends and matures during angiogenesis. Haemangiomas which comprise of hamartomatous growth in capillaries are due to imbalance between intrinsic and extrinsic factors acting on endothelial cells[12] like (Table II)

Table 2: Intrinsic and extrinsic factors acting on endothelial cells.

Factors stimulating angiogenesis	Factors inhibiting angiogenesis
Local tissue environment hypoxia/acidity	Angiostatin
VEGF (vascular endothelial growth factor)	Platelet factor 4
Fibroblast growth factor	Interferon
Matrix metalloproteinases 9	Thrombospondin 1
Intercellular adhesion molecule 3	Tissue inhibitor of metalloproteinase

Sporadic mutation leading to error in morphogenesis of any combination of arterial, venous and lymphatic vascular channels during embryonic development is called as Vascular malformation.[13,14,15] Arteriovenous malformations occurring due to failure of regression of AV channels in primitive retiform plexus are abnormal connections between arteries and veins permitting blood within organ or tissue to bypass the capillary network. Diagnosis is based on clinical history, physical examination, and when unclear assisted with USG or MRI.[16]

Diagnosis of such hamartomatous lesions is based on clinical and histopathological examination. Clinical diagnosis remains a challenge because of varied clinical presentation due to difference in size, location, subtype, and tissue involvement by the lesion.[17] Pre-interventional imaging is very important for confirmation of diagnosis and assessment of flow dynamics.[18] Computed tomography, doppler ultrasonography and magnetic resonance imaging are useful modalities for confirmation of diagnosis and evaluation of the extent of lesion and flow velocity. Imaging assesses exact flow dynamics in order to differentiate various types of vascular malformations thus facilitating treatment planning.[19,20,21] Table III shows the relevance of vascular lesions to dental practitioners in terms of various procedures.

Table 3: Fate of vascular lesions

Condition	Clinical appearance	Referral to secondary care	Safety for injections	Safety for extractions	Safety for scaling
Infantile hemangioma	Most common tumours in infants manifest clinically several weeks after birth and appear as raised dark red, when located deeper produce blue hue of overlying skin.[22,23]	If a patient is already under specialist care	Not applicable	Not applicable	Not applicable
Congenital hemangioma	These are less common, seen on prenatal imaging, undergo proliferation in utero, present upto full size at birth, usually single and when subcutaneously located they are pink or purple.[22]	Yes	Yes	Yes	Yes
Pyogenic granuloma	A misnomer as this lesion is neither granulomatous nor pus-producing, so microscopically accurate more appropriate term is lobular capillary hemangioma. Clinically it appears as a small red papule either smooth or lobulated exophytic lesion with pedunculated or sessile base [24,25]	Yes	Yes	Yes	Yes
Capillary malformation	Commonly called as portwine stains. Most commonly appear as pink macular lesions in head and neck regions along distribution of trigeminal nerve dermatomes[22]	If causing cosmetic problem	Yes	Yes	Yes
Venous malformation	Most common type of vascular malformation presenting as bluish soft compressible masses on oral mucosa with no pulsation / thrill[22]	If symptoms are present or confusion regarding exact nature of lesion	Try not to inject in lesion	If lesion is not in close association with tooth	Avoid scaling near lesion
Lymphatic malformation	They appear as either microcystic or macrocystic lesion lacking typical pulsatile thrill of high flow lesions and are non compressible[22]	If lesions have not been previously seen by a specialist	Try not to inject in lesion	Not safe	Avoid scaling near lesion
Arteriovenous malformation	Usually, growth is slow but sometimes grows very rapidly in less time or growth is induced by trauma. Usually present as a pulsatile mass with associated thrill or bruit. when present in mandible, mobility of teeth, bone expansion and associated gingival bleeding is noted[22]	Yes	Not safe	Not safe	Not safe

1) Colour doppler ultrasound – Though various imaging modalities are available for the assessment of vascular anomalies but colour doppler ultrasound is a first-line approach.

Advantages – No ionizing radiation

Excellent intrinsic resolution

Cost effective, reliable and non-invasive

Identify presence, quality and type of doppler flow, mean velocity and direction Identify feeding and draining vessels

Helps in differentiating vascular and non-vascular causes of nodular lesions of oral cavity

Provide information on location, size, margins, echogenicity and tendency to invade adjacent structures.

Useful in both diagnosing and guiding treatment of the lesion.[1]

Development of new, compact, linear hockey stick shaped high frequency probe (7 – 15 MHz) allows visualization of course of vessels and anatomy of lip from external vermilion

to internal lip surface without moving patient or probe.5-7 MHz probes are needed in obese patients and in deeper intramuscular malformations. Grey scale ultrasound is performed at higher resolution frequency.[1]

Colour doppler spectral curve analysis – It assesses velocity as well as arterial / venous nature of flow

Power doppler – useful for detecting very slow flow.

Disadvantage – malformations located in deeper tissues are less precisely detected.[7]

2) Magnetic resonance imaging – It is the most important and valuable tool for assessment of vascular malformation. It includes high contrast soft tissue imaging, vascular imaging and flow measurement. MRI consist of various sequences like T1w , T2w with fat saturation , spin echo (SE), fast spin echo (FSE), short T1 inversion recovery (STIR).[7]

Disadvantage – Time consuming

Susceptibility to motion artifacts

Contrast enhanced MRI – enhanced with gadolinium

i) **Peripheral enhancement without progression throughout the mass** -lesion other than vascular entity.

ii) **Peripheral enhancement progressive to centre** – Venous malformation

iii) **Rapid enhancement throughout the mass** – Arterial malformation

iv) **Enhancement accompanied by flow voids** – Arteriovenous malformation[1]

3) Computed tomography – because of high exposure to ionizing radiation it is not recommended as a routine diagnostic modality though it allows precise evaluation of feeding and draining vessels with rapid assessment of vascular malformation.[7]

Conclusion –

It is important to differentiate the malformation from tumours, assure the patient about their nature and inform them about the precautions during routine activities and dental procedures. Diagnosis and treatment of these lesions involve a multidisciplinary approach so it is essential to refer these cases to specialists when appropriate.

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