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INTRAORAL VENOUS ANGIOMA OF BUCCAL MUCOSA – A RARE CASE REPORT

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ABSTRACT

Vascular malformations are atypical irregular anomalies of vein with a prevalence of 3% of the general population. Most of them are asymptomatic with an inconclusive etiology and are located on other routine investigations. We present a case report of intraoral manifestations of VA in a 23-year-old male patient with a complaint of a swelling on his right side of cheek for a period 8 months with gradual increase in size and associated symptoms. The lesion was diagnosed as venous angioma following an MRI scan and treated with sclerotherapy. The lesion responded to sclerotherapy and spontaneous remission was evident. No recurrence was noted during 12 months follow up. Early detection is necessary to determine the clinical behaviour and potential dentoalveolar complications of the lesion for timely diagnosis and rapid treatment and can avoid fatal events

KEY WORDS: Venous angioma, vascular malformations, developmental venous anomaly, intraoral, sclerotherapy

INTRODUCTION

Venous malformations are sporadic in nature and sometimes autosomal dominant. [1] A defect in chromosome 9 can lead to the formation of venous anomaly with slight female predilection. [2] Though congenital, they become evident during any phase of life. Relying on their hemodynamic characters they are divided as low and high flow malformations. [3] Low flow lesions include capillary, venous and lymphatic malformations whereas high flow lesions include arterial malformations and arterio-venous malformations. Treatment includes medical, conservative (sclerotherapy) or surgical (laser) management. [1] This is a report of a 23-year-old male patient presenting with unique intraoral clinical manifestation of single lesion presenting at two sites.

CASE REPORT

A 23 years old male patient came to the Department of Oral Medicine a year ago with the complaint of a swelling on his right of cheek for the past 7 months. History revealed that the growth initially started as a small one and after few days, patient had a habit of constant sucking over the growth and it reached the present size. The swelling had occurred for the first time. His past medical and dental history did not reveal anything significant.

On extraoral examination, there was facial asymmetry with a diffuse swelling on the right-side mandibular body region roughly measuring 2.5*1.5cm in dimension. The surface over the swelling was normal in colour, temperature and skin over swelling was pinchable with no secondary changes present over the swelling.

Intraoral examination revealed 2 well defined lobulated bluish red swellings were evident on the right buccal mucosa in relation to the occlusal plane measuring about 1.7*1.2cm and 1.1*0.8cm in size respectively. On palpation it was non-tender, soft and blanched on pressure. Oral examination also revealed supernumerary teeth in relation to 45 and multiple restorations with good oral hygiene. Based on the history and clinical features, diagnosis of vascular lesion on right buccal mucosa was given.

Figure 1: Bluish red swelling on right buccal mucosa a) 1.7*1.2cm b)1.1*0.8cm

The differential diagnosis of hemangioma, mucocele, neurofibroma and a minor salivary gland tumor was considered. OPG taken for diagnostic assessment showed no remarkable feature relating to the intraoral lesion. MRI (1.5 tesla) revealed hyperdense area along the right cluster of medullary veins in the midline cerebellum draining through collector vein into straight sinus suggestive of venous angioma. Later MR angiogram, confirmed the presence of venous angioma.

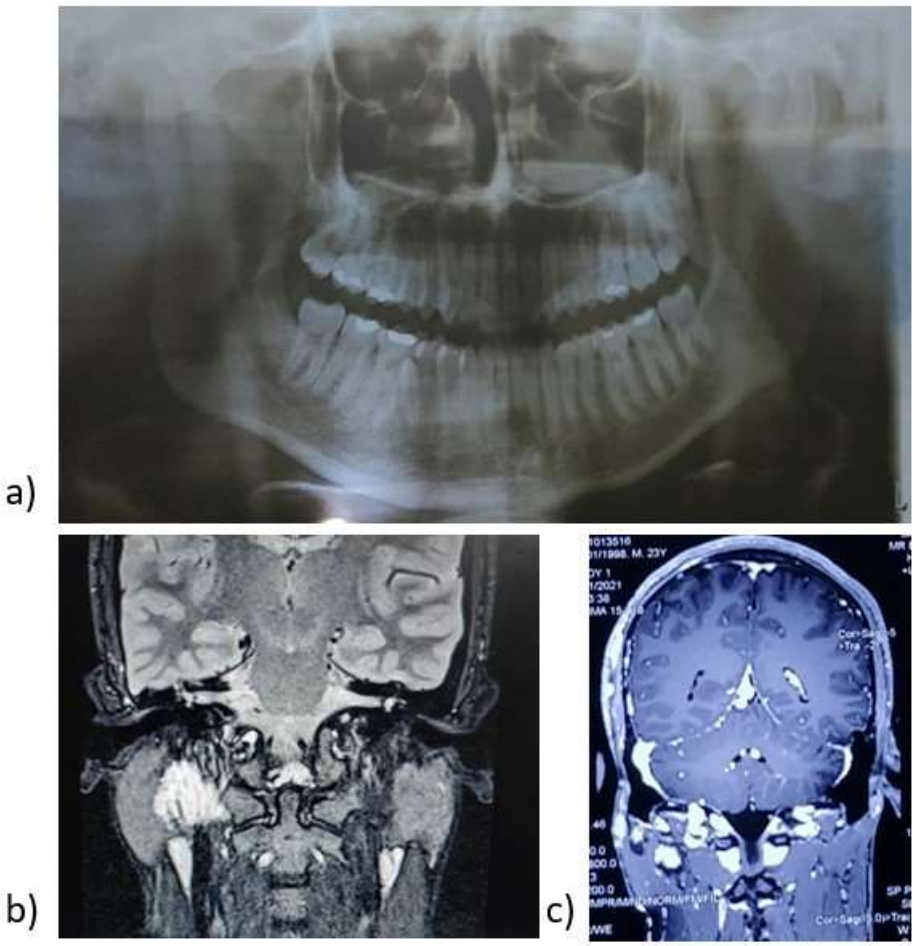

Figure 2: Investigations a) OPG with no bone involvement b) MRI c) MRI with contrast

TREATMENT AND FOLLOW UP

Conservative intervention of sclerotherapy was planned and executed with sodium tetradecyl sulphate a sclerosing agent of 2ml for 7 times over a period of 7 weeks. Reduction in size of the lesion observed within a week with good adherence and tolerability. No complications of the drug were observed. A follow up of one year was done with no recurrence or adverse and unanticipated events.

Figure 3: a)Pre Operative clinical image b)Post operative follow up after 6 months c)Post operative follow up after 1 year

DISCUSSION

Venous malformations are otherwise known as venous angioma, cavernous angioma, cavernous hemangioma and phleboangioma. They are slow flow lesions with abnormal venous network. [2] They are initially small, insidious, soft and compressible in nature. They give a bluish appearance mimicking mucocele due to ectatic venous channel within the dermis. Hormonal changes during puberty and pregnancy are the major etiology for gradual increase in its size. Acute increase in the size is due to infection or injury.

Contentious theories regarding the pathophysiology proposed by Lasjaunias at al (1986) stated that during the developmental stage "The hemodynamic need" leads to VA for evaluation by altering drainage patterns. Few literature states that they are normal variants of venous drainage and not true vascular malformations.

The common complaints are significant cosmetic defects, recurrent bleeding, obstruction of airway and interference with normal speech, dentition and venous thrombosis (rare).

The use of conventional radiographs is limited to detect the presence of dystrophic calcifications and calcified phleboliths and CT for intraosseous involvement. Doppler Ultrasound/MRI are traditionally used as the gold standard for their diagnosis. MR angiography is used to determine undermining associated vascular deformity. The differential diagnosis included were lymphangioma, mucocele, lipoma and a minor salivary gland tumor.

Haemangioma and lymphangioma are uncommon congenital malformations that are benign hyperplasia of lymphatic vessels. Common intra oral site is on the dorsum of tongue and palate. [5]

Mucocele, a salivary gland pathology occurs due to trauma leading to break in gland duct and spillage of mucin in surrounding areas. They classically present as dome shaped growth with lower lip as the commonest site. [6]

Lipoma, benign tumor of mature fats cells that frequently presents in superficial soft tissues. [7] Although they are mesenchymal in origin, they rarely occur intraorally.

Minor salivary gland tumor, are benign growth associated with minor salivary gland. Common intraoral sites include palate, lip, buccal mucosa, tongue or floor of mouth. [8]

Primary treatment includes monitoring and review, along with low dose of aspirin to reduce the risk of phlebothrombosis. [3] Superficial

dermal and oral lesions require conservative management with sclerosing agents. Sodium tetradecyl sulphate, a commonly used sclerosing agent works by increasing endothelial fibrosis and decreasing the size of large blood vessels. [9] The drug is contraindicated in conditions like bleeding disorders and severe blood infection. Complications of the drug post administration might include mild discolouration, pain and swelling at the site of injection and cutaneous necrosis. [9] Recurrence in sclerotherapy can be managed by surgery. Lasers can be used for minimum scaring in superficial skin and oral mucosa.

The strength in conservative approach of treatment includes esthetical and non-surgical management. Limitation includes its application for superficial, small and slow growing lesion with higher chances for recurrences that requires long term follow-ups. [10]

Venous malformations should be followed up regularly as they are a part and parcel of normal blood circulation, they do not require management. Any surgical excision or closure would result in complications and recurrences. Conservative management of injecting sclerosing agents on regular interval shows complete resolution of vascular lesion.

CONCLUSION

This is a unique report having a diagnostic dilemma for a venous malformation having an intraoral presentation at two sites. Definitive diagnosis of such unique intraoral presentations can be made after thorough investigative procedures, emphasizing the importance of detailed diagnostic workup.

PATIENT PERSPECTIVE

Patient's informed consent was obtained. In patient's perspective obtained through his brother, "Initially when there was a swelling, he felt very uncomfortable. His diet was slowly getting affected and he constantly developed sucking over the swelling. Though we were unaware to get medical help at that stage, when the size began to increase, we opted your institution. You gave us detailed explanation on the treatment plan and he was glad, it didn't require surgery. Though the appointments required to take off from the work, he is happy that the treatment was successful.

Table 1: Review of Literature – Intraoral Venous Angioma

S. No	Authors	Year	Intraoral Presentation	Treatment
1.	Marica et al.,	1986	Right side of the tongue from base to tip, gingiva, tonsils, hard and soft palate	Surgical removal
2.	Slaba et al.,	2010	Giant venous malformation in tongue	Embolization and sclerotherapy
3.	Ravi et al.,	2011	VA in maxillary right labial sulcus with phleboliths	Surgical excision
4.	Kim et al.,	2012	Two swellings on the lower labial vestibule and two on tongue	Sclerotherapy (ethanolamine oleate)
5.	Ujwala	2015	Lobulated swelling on maxillary left labial mucosa	Surgical excision

6	Lidiya et al.,	2019	Mandibular right buccal sulcus	Sclerotherapy (Polidocanol)

Table 2: Timeline of history

February 2020	First noticed the swelling that was initially small in size of the lesion
October 2020	Reported for medical help as there was noticeable increase in size of the lesion
November 2020	Commencement of sclerotherapy
December 2020	Completion of sclerotherapy
December 2020	Visible reduction in the size of the lesion
April 2020	6 months follow up (Figure3b)
October 2020	1 year follow up (Figure3c)

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LEGENDS OF FIGURES

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