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Unraveling Venous Malformation: A Unique Case Report from an Oral Medicine Perspective

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Abstract

Vascular anomalies of the soft tissue, encompassing malformations and tumors, arise from errors in vascular embryogenesis and represent a heterogeneous group of pathologies involving the circulatory system. The International Society for the Study of Vascular Anomalies (ISSVA) classification has standardized the terminology and approach toward these anomalies. This classification of 2014 divides the vascular anomalies between tumor lesions and vascular malformations.

Keywords

Vascular anomalies; Cavernous hemangiomas; Hemangiomas; Radioopacities; Ultrasonography; Therapeutic;

Introduction

Vascular anomalies of the soft tissue, encompassing malformations and tumors, arise from errors in vascular embryogenesis and represent a heterogeneous group of pathologies involving the circulatory system [1]. The International Society for the Study of Vascular Anomalies (ISSVA) classification has standardized the terminology and approach toward these anomalies. This classification of 2014 divides the vascular anomalies between tumor lesions and vascular malformations [2]. Historically, all vascular anomalies were broadly termed as "hemangiomas," but the work of Mulliken and Glowacki differentiated true neoplasms, such as infantile hemangiomas from vascular malformations. For instance, venous malformations (VM), previously known as "cavernous hemangiomas," are congenital anomalies typically presenting as soft, subcutaneous masses with a normal to bluish-purple hue [3].

Their classification and understanding have evolved significantly, with diagnostic imaging playing a pivotal role in their identification and management. Imaging techniques such as magnetic resonance imaging (MRI) and ultrasonography (USG) help confirm diagnoses, delineate lesion extent, monitor disease progression, and guide therapeutic interventions [1].

In the head and neck region, common sites for VM include subcutaneous tissues of the face, muscles of mastication, periorbital regions, and deep neck spaces. These anomalies vary in clinical presentation depending on their depth and location. Unlike high-flow vascular anomalies, VM lack palpable thrills or auscultatory bruits, and their low-flow characteristics are identifiable on imaging. Ultrasound serves as an ideal initial diagnostic tool, while MRI and CT provide detailed insights into lesion extent, vital organ involvement, and potential complications such as bone destruction [4].

A unique case report of venous malformation of temporal-masseter region is described here. The aim is to enhance awareness and understanding of such clinical presentations to the dentists, to facilitate accurate diagnosis and appropriate management.

Case Report

A 49-year-old male patient reported with swelling on right side of the face and in front of the ear for 2-3 months which did not increase in size. He did not experience pain but experienced mild numbness on right side in the morning. He did not give history of dental pain or trauma. He was under medication for hypertension for 4 years. There was no other relevant contributing medical history.

On examination, mild facial asymmetry was noted with oval soft swelling on the inferior part of right temporal region, which became taut on clenching. There was a soft diffuse swelling in right nasolabial fold region extending to zygoma region. No colour change was noted over the skin. On palpation, tenderness was not present, and there was no auscultatory bruits. TMJ movements were normal and mouth opening was adequate within normal limits. On intraoral examination, there was occlusal pit caries on 16, 18, 46, and 47. The right buccal mucosa appeared more bulkier with prominent blood vessels compared to left side.

A panoramic radiograph was taken to rule out any dental etiology. There was no caries or periapical lesion noted with right maxillary teeth. There were multiple small round radioopacities noted in right coronoid region suggestive of calcifications or phlebolith. Hence USG and MRI were advised to look for the soft tissue involvement and extension. USG showed bulky right temporalis muscle, whereas subcutaneous tissue and

parotid gland were normal. On MRI, a small lobulated facial soft tissue lesion was noted measuring 3.4 x 2 cm with surrounding temporal and zygomatic bone involving masseter and temporalis muscle. It was T1 hyperintense, T2 /FLAIR hyperintense with punctate areas of hypointensity within suggestive of phleboliths. Multiple flow voids were noted within this lesion. Right temporalis muscles appeared mildly bulky indicating hypertrophy. From the findings, this is a case of vascular anomaly, indicating a venous malformation, along with temporalis hypertrophy. A differential diagnosis of intramuscular hemangioma was given.

The patient was explained about the benign nature and was given reassurance. A periodic follow up was done for 3 months, and the lesion did not increase in size and patient was completely asymptomatic.

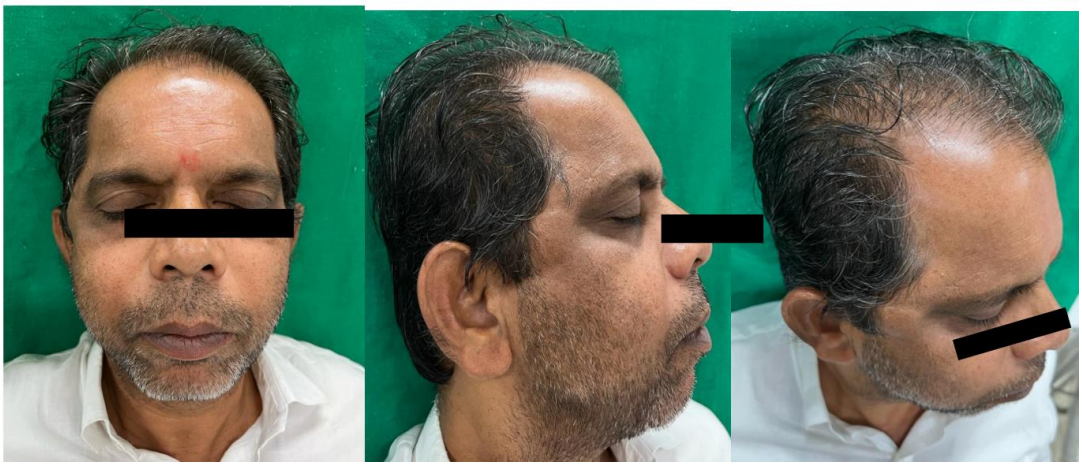

Figure 1: Extraoral pictures showing swelling in right nasolabial and temporal region.

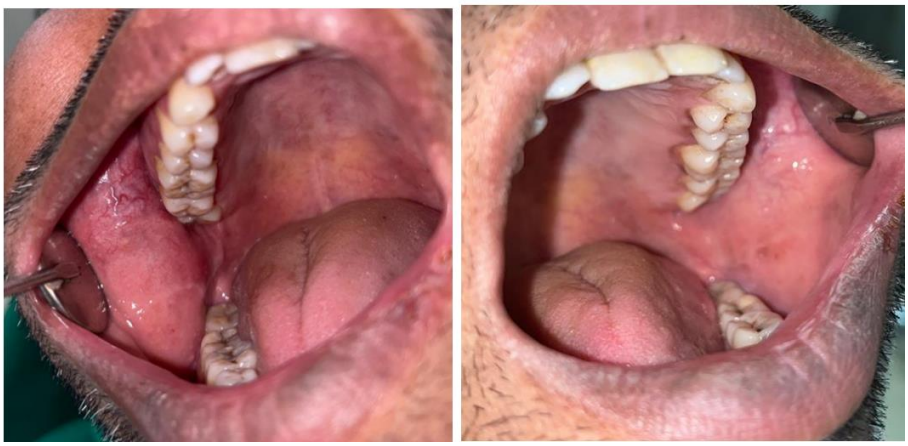

Figure 2: Intraoral pictures showing right and left buccal mucosa with prominent blood vessels on right side.

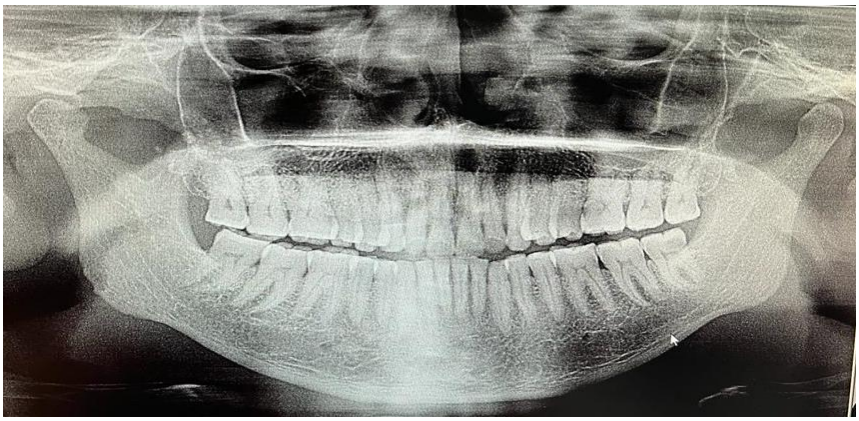

Figure 3: Panoramic radiograph showing Phleboliths.

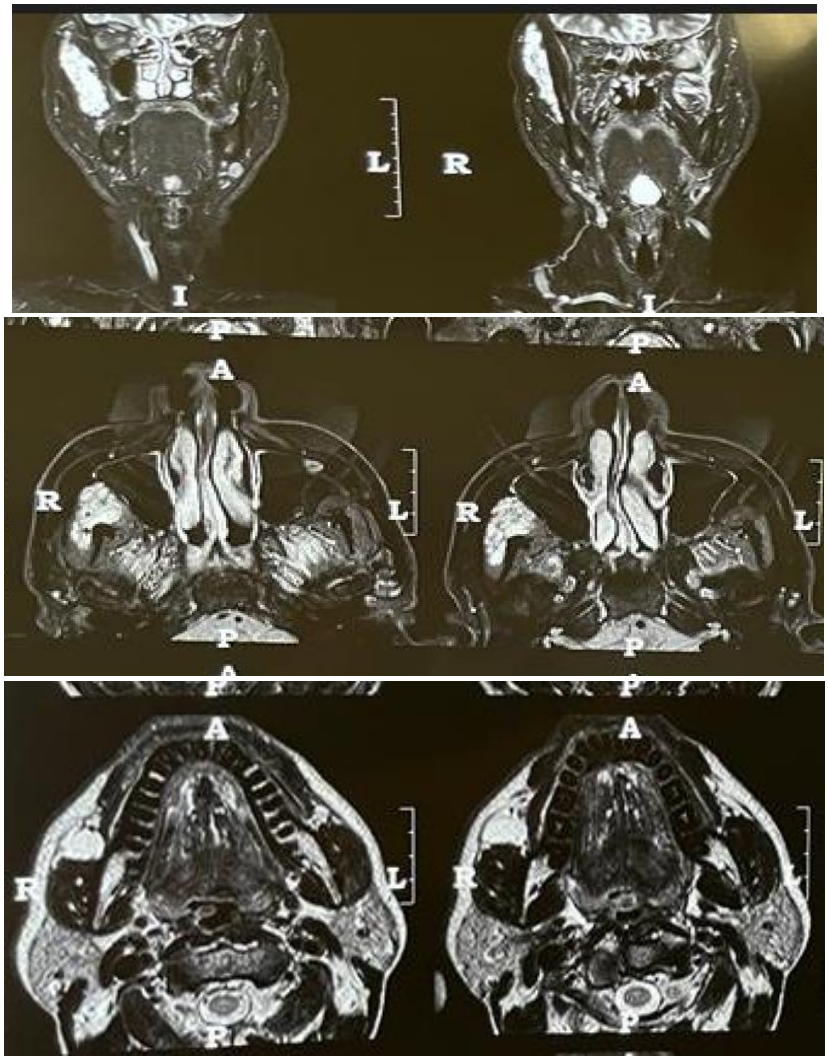

Figure 4: MRI images showing hyperintense areas in right temporalis and masseter region with areas of hypointensity, and temporalis muscle hypertrophy.

Discussion

VM, frequently not identified until later in life, are said to be present at birth, like all vascular malformations. They are seen in intradermal, subcutaneous, intramuscular, and intraosseous locations [4].

Pre-operative or clinical diagnosis is usually difficult, as pulsations and bruits are frequently absent due to the presence of surrounding muscle fibers, and absence of mucosal discoloration [5]. The present case can be easily misdiagnosed as a temporal space infection by its clinical appearance, but given the lack of pain and duration, this clinical entity can be ruled out. Secondly, the swelling appears to be temporal hypertrophy, lipoma, or a dermoid cyst, as the vascular anomalies could not be clinically detected [6]. This becomes particularly challenging to the physicians because venous malformations have wide variety of clinical appearance and severity, and some of them tend to be clinically occult as in our case, and even on angiograms they are not visible due to the slow blood flow through them. Due to their increased potential of recurrent bleeding, early detection and prompt management are demanded [7].

Some literature shows majority of hemangiomas involving the temporal muscle as cavernous hemangiomas [6, 8]. In the study by Rootman et al., they have demonstrated that cavernous hemangiomas are noninfiltrating, focal venous malformations that do not have features of hyperplasia [9]. Thus, since features of a tumour are absent, the suffix -oma should never be used to describe vascular malformations [10].

The low flow characteristic of VM is typical of their imaging characteristics. On USG, VM is depicted as poorly marginated structure which is usually hypoechoic and heterogeneous and can appear infiltrative. On doppler spectral analysis, it is identified as monophasic low-velocity internal flow with no identifiable arterial waveform [4].

Phleboliths are highly suggestive of VM, and it appears as echogenic foci with distal acoustic shadowing. It is formed due to thrombi produced by slowing of peripheral blood flow, which becomes organized and mineralized to form calcified thrombi which forms the core. The repeated process of fibrous component attaching to a developing phlebolith causes enlargement of the phlebolith [6].

Contrast based MRI is the key imaging modality when low flow lesions such as venous and lymphatic malformations are suspected. MRI plays a pivotal role in the diagnosis, treatment planning and follow-up of most vascular malformations and tumors of the head and neck. Conventional MRI provides a differential diagnosis for an atypical lesion [11].

Management strategies for vascular anomalies depend on lesion complexity and associated complications like disfigurement, pain, or venous thrombosis. While conservative measures, such as compression devices, are preferred for uncomplicated cases, interventions like sclerotherapy, laser photocoagulation, and surgical excision are employed for symptomatic or complicated lesions. Sclerotherapyis a widely accepted first-line treatment for low-flow anomalies, with agents like ethanol and sodium tetradecyl sulfate demonstrating efficacy [4]. As this patient in our case report did not have intracranial extension nor increase in size or symptoms, treatment was not necessary, but was kept on follow up.

Conclusion

Understanding the features and imaging characteristics of vascular anomalies of head and neck region is essential for oral medicine physicians to prevent misdiagnosis and complications. This case report enlightens the importance of proper clinical examination and how imaging unmasked a bigger picture of venous malformation.

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