#### **Case Report**

# Massive Arteriovenous Malformation of Tongue: A Case Report

Dr. Anila Koneru <sup>1</sup><sup>0</sup>, Dr. Vanishree M<sup>2</sup>, Dr. Deepa Mastammanavar<sup>3</sup>, Dr. Vidyachandra Katapadi <sup>1</sup><sup>6</sup><sup>4</sup>, Dr. Vardendra Manvikar<sup>1</sup>, Dr. H Aparna Latha <sup>1</sup><sup>5</sup>

 <sup>1</sup>Associate Professor, Department of Oral Pathology, Navodaya Dental College, Raichur, Karnataka, India.
<sup>2</sup>Professor and Head, Department of Oral Pathology, Navodaya Dental College, Raichur, Karnataka, India.
<sup>3</sup>Assistant Professor, Department of Dentistry, Haveri Institute of Medical Sciences, Haveri, Karnataka, India.
<sup>4</sup>Associate Professor, Department of Oral and Maxillofacial Surgery, Srinivas Institute of Dental Sciences, Mangalore, Karnataka, India.

<sup>5</sup>Assistant Professor, Department of Oral Pathology, Navodaya Dental College, Raichur, Karnataka, India.

\*Corresponding Author: Dr. H Aparna Latha; aparnalatha060593@gmail.com

Received: 16 April 2025;

Accepted: 30 May 2025;

Published: 02 June 2025

#### Abstract

"Vascular malformation" is a general term used to define a group of lesions, present at birth, shaped by an anomaly of angiovascular or lymphovascular structures. In this paper, we present a case of arteriovenous malformation involving the left dorsum of tongue of a 25-year-old female followed by a review of the literature. A 25-year-old Female patient complains of a large swelling on the left upper and lower surface of tongue since 3 years and increased in size over the last 4 months. On intraoral examination, a large reddish purple-colored lesion - the anterior portion of the tongue. The overlying skin appeared nodular. On palpation, the swelling was soft, fluctuant and compressible. Lesion blanched with pressure- Diascopy, and pulsations were felt at the posterior end. Provisional diagnosis of Vascular Malformation was given with a differential diagnosis of Hemangioma. MRI revealed Hyperintense T2-weighted images showed vessels with flow voids confirms a fast flow vessel. These findings were related to Arteriovenous Malformation. Histopathological examination revealed the presence of large and small vessels lined by endothelial cells. These vessels are filled with blood and supported by fibrous connective tissue stroma. A Final Diagnosis of AV Malformation of Tongue was given. To conclude, clinical diagnosis with proper mode of investigations such as MRI, CT, Doppler and Angiography helps in rapid diagnosis and avoiding morbidity related to the vascular lesions.

Keywords: Arteriovenous malformation, Doppler, Magnetic resonance imaging, Tongue.

## Introduction

"Vascular malformation" is a general term used to define a group of lesions, present at birth, shaped by an anomaly of angiovascular or lymphovascular structures. Vascular malformations occur in around 1% of births however majority of these patients do not present for treatment. The high-flow vascular anomalies in the head and neck are arteriovenous malformations (AVMs). These are the lesions with direct communications between an artery (or arteries) and a vein (or veins) bypassing the capillary bed <sup>[1]</sup>.

AVMs are usually present at birth but commonly evident in childhood or adolescence. These lesions can occur at any area of the body. They have gradual onset and progression. In the oral cavity, these can present at any site, but most commonly occur on anterior two-thirds of the tongue, palate, and gingival and buccal mucosa <sup>[2]</sup>.

These lesions can be diagnosed by plain radiography, computed tomography scans, magnetic resonance imaging, or angiography. Various sclerosing agents and embolization, combined

with surgical treatment, are still the most conventional modern approach to treat these lesions <sup>[3]</sup>. Here, we present a case of arteriovenous malformation involving the left dorsum of tongue of a 25-year-old female.

#### **Case Report**

A 25-year-old Female patient complains of a large swelling on the left upper and lower surface of tongue. The patient presented with a large painless swelling on the left dorso-Ventral surface of tongue since 3 years and increased in size over the last 4 months (**Figure 1**). Not associated with pain and there was no regression in its size. No significant facial asymmetry on Extraoral examination.



Figure 1: A large reddish purple-colored lesion in the left dorsal portion of the tongue.

On intraoral examination, a large reddish purple-colored lesion involving anterior portion of the tongue. The lesion measured 4 cm from tip of Tongue anteroposteriorly and mediolaterally extending from midline to whole left side and Ventral side of the tongue (**Figure 2**).



Figure 2: The lesion measuring 4 cm extending from midline to whole left side and Ventral side of the tongue.

The overlying skin appeared nodular. On palpation, the swelling was soft, fluctuant and compressible (**Figure 3**).



Figure 3: The overlying skin appeared nodular.

On diascopy the Lesion blanched with pressure, and pulsations were felt at the posterior end. Provisional diagnosis of Vascular Malformation was given with a differential diagnosis of Hemangioma.

Radiographic findings of MRI revealed Hyperintense T2weighted images showed vessels with flow voids confirms a fast flow vessel. Maximum diameter of 4 cm lesion showed loose tangle of vessels in the middle and anterior left part of the tongue (**Figure 4**). These findings were related to Arteriovenous Malformation.



Figure 4: 4 cm lesion showed loose tangle of vessels in the middle and anterior left part of the tongue.

Biopsy findings and Diagnosis: Histopathological examination revealed the presence of large and small vessels lined by endothelial cells (**Figure 5**). These vessels are filled with blood and supported by fibrous connective tissue stroma (**Figure 6**). A Final Diagnosis of AV Malformation of Tongue was given.



Figure 5: Presence of large and small vessels lined by endothelial cells.



Figure 6: Vessels are filled with blood and supported by fibrous connective tissue stroma.

#### Discussion

A vascular malformation can be slow-flow (i.e., capillary, lymphatic, or venous) or fast-flow (i.e., arterial). If there are combinations of these elements, the malformation is called an arteriovenous malformation (AVM), lymphatico-venous malformation (LVM), or capillary-lymphatico-venous malformation (CLVM). Arteriovenous malformations are high-flow lesions with direct communications between an artery (or arteries) and a vein (or veins), bypassing the capillary bed <sup>[4]</sup>.

Arteriovenous malformation of the head and neck is a rare vascular anomaly but when present is persistent and progressive in nature and can represent as a fatal disease <sup>[5]</sup>.

Regarding the origin and pathogenesis of AVM, defects in TGF-beta signaling and a genetic two-hit hypothesis are the

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prevailing theories. Trauma, ischaemic event secondary to thrombosis, ectasia, hormonal changes, and puberty can induce proliferation of the AVM and trigger the growth of the lesion and manifestation of its troublesome symptoms. These are often the cause of massive, sometimes fatal, haemorrhages <sup>[6]</sup>.

AVMs are usually present at birth but commonly manifest in childhood or adolescence. Our case was associated with a 25-yearold female. These lesions present as a pulsatile mass with a thrill, bruit, and occasionally local hyperthermia, ulceration or bleeding, functional impairment due to ischaemia <sup>[1]</sup>.

In the oral cavity, these can present at any site, but most commonly on anterior two-thirds of tongue, leading to macroglossia and difficulty in mastication, speech, and deglutition. The present case showed the involvement of massive site, that is dorsum of tongue and ventral surface of tongue <sup>[2]</sup>.

These lesions can be diagnosed by CT, MRI or angiography. Surgical resection can lead to extensive blood loss and an incomplete resection can lead to regrowth of the tumor. Multimodal treatment, including preoperative embolization and complete surgical resection are considered for the management of AVMs<sup>[7]</sup>.

Considering the size of the lesion, combined approach of Embolization and Surgical excision was chosen as a treatment option in this case. The postoperative course was uneventful without any complications. Thus, AVMs present a therapeutic challenge because of their haemodynamic characteristics and their modality of growth.

# Conclusion

A Massive case of arteriovenous malformation of dorsum and Ventral surface of tongue in a 25-year-old female patient has been presented. Clinical diagnosis with proper mode of investigations such as MRI, CT, Doppler and Angiography helps in rapid diagnosis and avoiding morbidity related to the vascular lesions.

# Declarations

# **Ethical Approval**

Ethical approval was obtained by the corresponding author.

## **Conflicts of Interest**

The authors declare that there is no conflicts of interest.

## **Data Availability**

Available on corresponding author upon responsible request.

## **Funding Statement**

None

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