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Case Report

CAVERNOUS HEMANGIOMA OF TONGUE: A NEW INSIGHT

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Abstract:

Hemangiomas are one of the most common developmental anomalies. They constitute more than 50% cases in the head and neck region. One of the most widespread and perilous site is tongue. The present case report describes such an anomaly in 47 year old Indian male on the tongue. It also emphasizes the role of ultrasonography as a sole diagnostic as an investigation in areas where biopsy can lead to loss of function. Further, it highlights various advancements in the treatment modalities of hemangiomas along with their advantages and disadvantages.

Keywords: Hemangioma, Sclerotherapy, Ultrasonography, Cavernous, Capillary

INTRODUCTION

Hemangioma is derived from greek word i.e Haima means blood, angeion means vessel and oma means tumor. It may be defined as "a benign tumor of dilated blood vessels." Hemangioma of head and neck appear a few weeks after birth and they grow rapidly. According to the classification given by Mulliken and Glovacki in 1982, vascular deformities are divided into two groups i.e hemangiomas and the vascular malformations. Further, hemangiomas can also be classified depending on the vessel type involved or flow types such as the arterial and arteriovenous (high flow) type, capillary or venous (low flow) type.

The pathophysiology of hemangiomas is attributed to genetic and cellular factors, mainly to monocytes, which are considered the potential ancestors of hemangioma endothelial cells.



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Imbalance in the angiogenesis, which causes an uncontrolled proliferation of vascular elements, associated with substances such as vascular endothelial growth factor (VEGF), basic fibroblast growth factor (BFGF) and indole-amine 2,3-dioxygenase (IDO), which are found in large amount during proliferative stages, are believed to be the cause.³

Clinically, hemangioma appears as soft and poorly defined. They readily blanch with compression. They have been likened to a "bag of worms." Histologically, hemangiomas can be classified into capillary and cavernous forms. 5,6

Capillary hemangioma is composed of many small capillaries lines by a single layer of endothelial cells supported in a connective tissue stroma of varying density, while cavernous hemangioma is formed by large, thin walled vessels, or sinusoids lined by epithelial cells separated by thin layer of connective tissue septa.⁷

This aim of this paper is to report such a case in Indian male patient who had a growth on his tip, lateral border, dorsal and lingual border of the tongue on one side.

CASE REPORT

A 47-year old male presented with a swelling in the mouth involving the left anterior 2/3rd, posterior 1/3rd and lateral border of the tongue since twenty years. According to the patient, the swelling was present from twenty years and for the past 10 years, the swelling in the tongue gradually increased to the present size. Patient was asymptomatic and there were no associated features such as fever, difficulty in the speech and swallowing. Past medical, dental and family histories were non-contributory and on physical examination, patient appeared to be healthy and, with all his vital signs being within normal limit.

Intra-oral examination, revealed multiple dome-shaped swellings in the left anterior $2/3^{rd}$ of tongue, posterior 1/3rd as well as on lateral borders of the tongue extending both dorsally and ventrally.(fig 1,2) The surface appeared to be smooth and granular with well- defined borders. The swelling was bluish purple in color. Surrounding area was normal and no ulceration was present on the surface of lesion. On palpation the swelling was soft to firm in consistency, non mobile, non tender, afebrile with no palpable thrills. Swelling blanched on compression.

Depending on the clinical features, a working diagnosis of hemangioma with differential diagnosis of a capillary hemangioma, granular cell myoblastoma, angiomyolipoma and angiosarcoma were made.

The routine blood and urine investigations were normal. Ultrasounography revealed multiple dilated cystic channels as hypoechoic lesions. In the cystic spaces multiple phleboliths were present.(fig 3) Color Doppler ultrasonography revealed low velocity venous flow. There was also presence of feeding vessel.(fig 4) Hence, ultrasonography suggested the final diagnosis of cavernous hemangioma of tongue.



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Fig 1 Multiple bluish-purple dome-shaped swellings in the left anterior $2/3^{rd}$ of tongue, posterior 1/3rd as well as on lateral borders of the tongue.



Fig 2 Multiple bluish purple dome-shaped swellings present over tip, lateral border of the tongue along extending ventrally.



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Fig 3 Grey scale US reveals multiple dilated cystic channels seen as hypoechoic lesions. There is also presence of multiple phleboliths in the cystic spaces.



Fig 4 Color Doppler US reveals low velocity venous flow along with feeding vessels.



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DISCUSSION

Haemangiomas are among the most common neoplasms encountered by the dentists. In 80% of the cases, hemangiomas occur as single lesions. Most common intraoral sites are tongue followed by buccal mucosa, lips & palate respectively. In our case, it was present on tongue.

The differential diagnosis includes capillary hemangioma, granular cell myoblastoma, angiomyolipoma and angiosarcoma. In the present case these pathologies were differentiated ultrasonographically to obtain final diagnosis.

Ultrasonographically, Cavernous hemangioma can be differented by the presence of solid echogenic mass with phleboliths on Gray scale US. Further, it is multispacial. Color Doppler US reveals monophasic/ venous or no flow pattern. These features were present in our case as well.

Capillary hemangioma can be differentiated from cavernous type by the multicystic appearance on Gray scale US. On Color Doppler US, there is presence of flow in the septa.⁸

Vasoproliferative disorders such as Angiosarcomas on Gray scale US can be seen with well-defined and homogenous pattern. Moreover, they have variable echogenecity. On Color Doppler, they are hypervascular and have arterial or venous wave form.⁸

Other pathologies such as Granular cell myoblastoma and Angiomyolipoma reveal involvement of muscle tissue whereas hemangiomas are purely vascular in origin.

Thus, Ultrasonography can be used as a sole diagnostic aid in cases of vascular tumors. Therefore, in large lesions it is preferable to undergo ultrasonography instead of histopathological sections as this will prevent functional loss.

Cavernous hemangioma of tongue has a clinical implication as slight trauma can lead to consequent bleeding or ulceration. Further, it can lead to masticatory disturbance, respiratory problems and difficulty in speech. But, in majority of the cases patient concern is focussed on the cosmetics. Therefore, management of hemangioma depends on following factors:

- 1. Site
- 2. Accessibility
- 3. Depth of invasion
- 4. Rate of growth
- 5. Age of patient
- 6. Cosmetic considerations

Various treatment modalities are present. These include Sclerotherapy, Laser photocoagulation, Surgery, Embolization, Intralesional & systemic steroids, Electrolysis & thermocautery, Immunomodulatory therapy along with interferone alpha, cryotherapy and plasma knife surgery. Advantages and disadvantages of these treatment approaches are



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described in a table (table 1). In our case, we advised patient for sclerotherapy but patient did not give any follow-up.

Table1: Treatment Modalities of Hemangiomas

Sr. No.	Treatment approach	Advantages	Disadvantages
1.	Sclerotherapy	 Simple Procedure Inexpensive No loss of blood No hospitalization is required Sclerosing agents are widely available 	1.Post-operative pain 2.Burning sensation 3. Anaphylaxis 4.Tissue Necrosis 5. Sloughing
2.	Laser photocoagulation	1. Decrease blood loss	1.Re-growth of the lesion 2.Variable response 3.Accessibility of the lesion
3.	Surgery	 Indicated in large lesions Complete resection Reduces symptoms 	1.Loss of function2. Loss of blood3. Hospitalization may be required
4.	Embolization	1.Haemorrhage control	1.Technique sensitive as there may be misplacement of embolic material
5.	Intralesional & systemic corticosteroids	1. Decreases tumor bulk	1.Disseminated Varicella 2.Herpes Infection 3.Cushinoid habitus
6.	Electrolysis & thermocautery	1.Haemorrhage control	1. No excision is required
7.	Immunomodulatory therapy with Interferone- alpha	1. Helps in treatment of large, multiple and life threatening hemangiomas	1.Irreversible spastic diplegia
8.	Cryotherapy	 Decrease blood loss Lack of discomfort Decrease risk of Infection Minimal Scarring 	 Slow procedure Lack of precision in depth Tissue necrosis Pigmentation and scarring can result
9.	Plasma knife surgery	 Method is safe No blood loss Can be used for superficial lesions 	1. Cannot be used in larger lesions

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CONCLUSION

The hemangioma is a benign proliferation of endothelial cells common in the head, neck and relatively rare in the oral cavity. The present report highlights the conservative approach in diagnosis and treatment planning in cases of cavernous hemangioma of tongue which are large in size. Ultrasonography along with Color Doppler ultrasonography can be used as a sole diagnostic tool in diagnosing cases of cavernous hemangiomas in such cases thereby, not necessitate the use of biopsy. This further emphasizes use of sclerosing solution injections as a treatment of choice in such large lesions.

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