ACTA SCIENTIFIC DENTAL SCIENCES

Volume 8 Issue 7 July 2024

Case Report

Addressing Maxillofacial Vascular Anomalies: A Case Series on Deep Hemangiomas of the Tongue Treated with Sodium Tetradecyl Sulphate

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Received: April 05, 2024

Published: June 06, 2024

Abstract

Maxillofacial vascular anomalies pose a significant challenge in diagnosis and treatment, particularly concerning hemangiomas, which are prevalent in the head and neck region, with the lips, tongue, buccal mucosa, and palate most commonly involved (more than 50% of lesions). While typically identified in infancy, adult-onset and intramuscular hemangiomas are even more rare occurrences, necessitating careful management to prevent functional or aesthetic complications. We present a case series of seven patients with deep or cavernous hemangiomas on the tongue, successfully treated with 1% sodium tetradecyl sulphate. This non-surgical approach offers a promising alternative to invasive procedures, potentially reducing the need for extensive surgical interventions and ensuring favorable outcomes for patients.

Vascular anomalies are one of the most difficult diagnostic and therapeutic enigma in the maxillofacial region. Hemangiomas are developmental vascular abnormalities and more than 50% of these lesions occur in the head and neck region, with the lips, tongue, buccal mucosa, and palate most commonly involved. Most vascular anomalies are noticed at birth or occur during infancy, and generally involve skin or subcutaneous soft tissues. Adult onset hemangiomas are rare, and intramuscular location is extremely rare. Despite its benign origin and behaviour, it is always of clinical importance to the dental professionals and requires appropriate management as it may lead to an early or continuous loss of function or lifetime esthetic impairment.

Here we present a case series of 7 patients presenting with deep or cavernous hemangioma on tongue treated with 1% sodium tetradecyl sulphate with successful remission of the lesion. Usually such patients require surgical removal of the lesion. But in consideration to the massive surgical procedure, this therapeutic approach may reduce the chances of the surgical requirement.

Keywords: Cavernous Hemangioma; Tongue; 3% Sodium Tetradecyl Sulphate; Vascular Anamolies

Abbreviations

STS: Sodium Tetradecyl Sulphate

Introduction

Hemangiomas, constituting around 7% of benign maxillofacial tumors, are vascular tumors marked by an abnormal growth of blood vessels [1,2]. Originating from mesenchymal tissue, they are regarded as benign congenital hamartomas. They are believed to stem from malformed tissue affected by repetitive trauma or hormonal influences [3]. Around half (50%) appear within the first decade of life, with the majority (94%) manifesting before the age of 30 [4]. Unlike pediatric hemangiomas, which proliferates during infancy and gradually regress over years, adult hemangiomas have distinct growth pattern [5].

Clinical manifestation: While the majority of hemangiomas affect the head and neck region, occurrences within the oral cavity are uncommon but can manifest on various oral structures includ-

ing the tongue, lips, buccal mucosa, gingiva, palatal mucosa, salivary glands, alveolar ridge, and jaw bones [6,7]. Females exhibit a slightly higher incidence compared to males [6]. The prevalence of oral hemangiomas is estimated at 8 per 1000 in males and 4 per 1000 in females [8]. The complex vascular network in the oral cavity and head and neck area may predispose to vascular lesions.

Clinical charecteristics and Classification: haemangioma presents as a soft masses, smooth or lobulated, sessile or pedunculated, varying size from a few mms to several cms. Their typical coloration ranges from deep red to bluish, and they may exhibit blanching when pressure is applied. If sizable, they can impede proper mastication [7,8].

Hemangiomas are categorized into capillary and cavernous types based on their vascular system[2]. Capillary hemangiomas primarily feature capillary structures with active proliferation, potential invasion into nearby tissues, and a brief clinical course. In contrast, cavernous hemangiomas are characterized by larger vessels, sporadic mitotic activity, and a longer clinical duration [9,10].

Incidence and resolution

The incidence of cavernous hemangioma of the head and neck is about 5% of the vascular malformations diagnosed by angiography and histology [11]. Typically observed between the third and fifth decades of life, cavernous hemangiomas are also encountered in pediatric and geriatric populations [10]. Approximately, 70% of these hemangiomas spontaneously resolve by adolescence, with half of them linked to skin hemangiomas [12]. Clinically, cavernous hemangiomas manifest more frequently in females nearly twice as in male [13], and appearing larger, less defined, and displaying no inclination towards regression.

Classification Guidelines: The International Society for the Study of Vascular Anomalies (ISSVA) has established guidelines for distinguishing between these two conditions, based on the innovative classification initially introduced by Mulliken., *et al*. in 1982. Vasoformative tumors are broadly categorized into Hemangioma and vascular malformation. Hemangioma is further histologically subdivided into capillary and cavernous forms. Capillary hemangioma consists of numerous small capillaries lined by a single layer of endothelial cells, supported within a connective tissue stroma of varying density. On the other hand, cavernous hemangioma comprises large, thin-walled vessels or sinusoids lined by epithelial cells, separated by thin layers of connective tissue septa [14].

Pathophysiology: Hemangiomas are acknowledged as benign tumors characterized by three stages: endothelial cell proliferation, rapid growth, and eventual spontaneous regression. The pathophysiology of hemangiomas is linked to genetic and cellular factors, particularly monocytes, considered potential precursors of hemangioma endothelial cells. Dysregulation in angiogenesis, leading to uncontrolled proliferation of vascular elements, is associated with substances such as vascular endothelial growth factor (VEGF), basic fibroblast growth factor (BFGF), and indole-amine 2,3-dioxygenase (IDO), which are abundant during proliferative stages [15].

Materials and Methods

Methodology

Cases were selected based on their medical history, ultrasonographic and MRI reports, clinical findings of the lesion, and a review of past published articles. It was concluded that the final diagnosis was Cavernous Hemangioma of the tongue. Among the surgical and non-surgical treatment modalities, the chosen approach was conservative non-surgical treatment.

The selected treatment option was a 3% solution of Sodium Tetradecyl Sulphate administered as an intralesional injection, with a volume of 2-4 ml depending on the size of the lesion, using an insulin syringe. For smaller lesions with a size less than 2 x 1 cm in diameter, a volume of 2 ml was injected intralesionally once a month for three months per injection.

As the lesions gradually decreased in size from 2 x 1 cm to 1.0 x 0.5 mm in diameter, the dosage was tapered to 1-1.5 ml in the third month. In the case of larger and deeper hemangiomas, a volume of 4 ml was injected in the first month, followed by a gradual decrease in dosage as observed reduction in size. Overall, six months of injections were administered for large and deeper lesions.

Adjuvant medication included Rebapemide in tablet form once a day for 3-4 weeks, adjusted according to the size of the lesion, as well as lycopene as a multivitamin, and benzedymine as an oral solution for deep lesions. Analgesics and antibiotics were administered in some cases, and in one particular case, Fluconazole in tablet form was prescribed once a day for two weeks.

Case presentation in tabulated form

S. No	1	2	3	4	5	6	7
Sex/age	33/F	27/F	32/F	19/M	42/F	51/M	48/F
Cheif complaint and clinical features	Swelling over left border of tongue since 20 yrs difficulty in swallowing and mastication	Pea size swelling over tongue since 6 yrs, difficulty in swallowing	Asymptomatic swelling over right border of tongue since 11yrs,	Aymptomatic swelling over left border of tongue since 2 yrs	Large swelling over left and dorsal border of tongue since birth Gradually increase in size slowly difficulty in swallowing and mastication	Large swelling over tongue since 25yrsdifficulty in swallowing and mastication sometimes burning sensation present and bleeding occurs	Some time painfull large bluish swelling over dorsum border of tongue since 28 yrs difficulty in swallowing and mastication, burning sensation on eating spicey and hot food
Investigation	Routine Blood investigation, USG, MRI NECK	Routine Blood investigation, USG	Routine Blood investigation, USG	Routine Blood investigation, USG	Routine Blood investigation, USG, MRI NECK	Routine Blood investigation, USG	Routine Blood investigation, USG, MRI NECK
Diagnosis	Cavernous hemangioma of tongue	Cavernous hemangioma of tongue	Cavernous hemangioma of tongue	Cavernous hemangioma of tongue	Cavernous hemangioma of tongue	Cavernous hemangioma of tongue	Cavernous hemangioma of tongue
Treatment plan	Intralesional Injection 1% Sodium Tetra Decyl Sulfate For 4 Month One In Each Month With Lycopene 5% In Tablet Form	Intralesional Injection 1% Sodium Tetra Decyl Sulfate For 3 Month Single Per Month with Tab Lycopene 5%	Intralesional Injection 1% Sodium Tetra Decyl Sulfate For 3 Month Single Per Month with Tab Lycopene 5%	Intralesional injection 1% sodium tetra decyl sulfate for 3 month single per month with tab lycopene 5%	Intralesional injection 1% sodium tetra decyl sulfate for 6 month single per month with tab lycopene 5%	Intralesional injection 1% sodium tetra decyl sulfate for 6 month ingle per month with mucoprotactive drug tab rebapemide bd for 1 month	Intralesional injection 1% sodium tetra decyl sulfate for 6 months with tab fluconazole for candidial like infections and tab rebepemide as mucoprotactive agent
Complication	Pain At Site of Injection Subside by Own	Pain At Site of Injection Subside by Own	Pain At Site of Injection Subside by Own	Pain At Site of Injection Subside by Own	Pain, Bleeding Mucosal De Epitheliaization	Pain, Bleeding, Mucosal De Epitheliaization	Pain, Bleeding, Mucosal De Epitheliaization
Prognosis	Excellent	Excellent	Excellent	Excellent	Very Good	Very Good	Very Good

Table 1

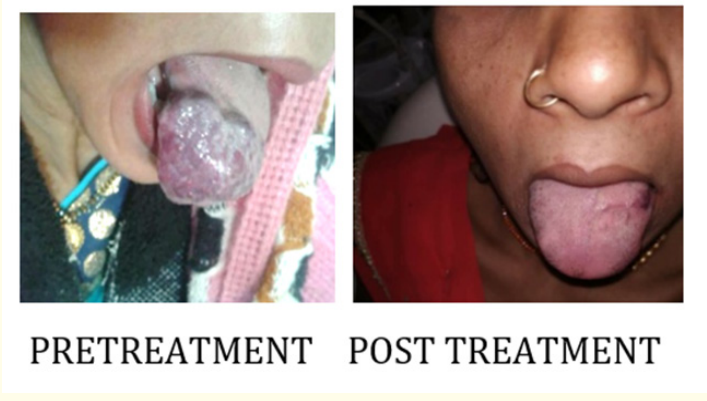

Figure a

Case of hemangioma of right lateral borger of tongue

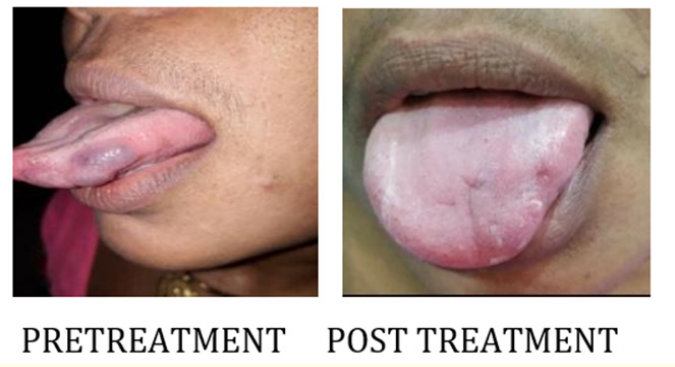

Figure b

Case of hemangioma of left lateral border of tongue

Results and Discussion

Hemangiomas exhibit a notably higher incidence among females, with the head and neck region being the most frequently affected, particularly the face, oral mucosa, lips, tongue, and trunk. The term "cavernous hemangioma" traditionally denotes lesions with significantly enlarged vascular channels. These hemangiomas consist of deep, irregular dermal channels filled with blood. They are comprised of intricate networks of thin-walled cavernous vessels or sinusoids, separated by a sparse connective tissue stroma. In terms of location and lesion number, our observations align closely with those reported in the literature, with approximately 80% of patients presenting a single lesion and the head and neck region being the most commonly affected [16].

Clinically, hemangiomas typically present as soft, smooth, or lobulated growths, either sessile or pedunculated, varying in size from a few millimeters to several centimeters. The color spectrum of these lesions ranges from pink to reddish-purple, with the tumor blanching under pressure and the potential for spontaneous or trauma-induced hemorrhage. They typically cause no pain [17].

Masses found in the submental space encompass a range of histopathological conditions. The list of potential diagnoses should encompass reactive lymphadenitis, lymphoma, metastatic lymph nodes, dermoid cysts, abscesses, as well as benign neoplasms like lipomas, hemangiomas, or neuromuscular-origin tumors, along with their malignant counterparts.

Preoperative radiographic imaging is recommended for selected cases, especially when large lesions may encroach upon critical anatomical structures like the facial nerve or orbit. Computed tomography (CT) and magnetic resonance imaging (MRI) are valuable tools for volumetric assessment of hemangiomas and vascular malformations[18]. These imaging modalities aid in diagnostic differentiation and analysis of lesion characteristics, including size, extent, and location. Additionally, they facilitate follow-up evaluations of lesions treated with systemic therapy.

In present case, Ultrasonography (USG) and magnetic resonance imaging (MRI) were employed as the preferred diagnostic imaging techniques. USG serves as an initial screening tool due to its portability, absence of ionizing radiation, and avoidance of sedation, particularly advantageous in pediatric cases. It is relatively straightforward, non-invasive, and provides reliable outcomes for assessing small, superficial, or suspected solid visceral lesions. Typically, USG can discern the fundamental nature of the lesion, guide initial management decisions, and facilitate further imaging assessments. For sonography to be effective, it should encompass grayscale imaging, color Doppler, and spectral Doppler tracings to evaluate vascularity and identify vessel types. In this specific instance, Ultrasonography revealed multiple dilated cystic channels exhibiting low-velocity venous flow on color Doppler, with the presence of phleboliths within the dilated channels, which is indicative of hemangioma [13].

MRI plays a crucial role in refining sonographic observations and assessing the full extent of larger lesions to strategize medical, interventional, or surgical treatment. The essential data required

for lesion evaluation is typically obtained from a combination of T1-weighted, fat-saturated T2-weighted, and gradient-echo (flow-weighted) MR images. In this instance, the axial plane proved most beneficial in illustrating the relationship between the lesion, neurovascular structures, and tissue planes [13].

The approach to managing hemangiomas is contingent upon numerous factors, and typically, genuine hemangiomas necessitate no intervention. Various treatment options have been documented in the literature for head and neck hemangiomas, ranging from a watchful waiting strategy for spontaneous regression to intralesional and systemic corticosteroid therapy, embolization, excision, electrolysis, thermocautery, immunomodulatory therapy with interferon alfa-2a, and laser photocoagulation [18,19]. Presently, sclerotherapy is predominantly utilized due to its effectiveness and its capacity to preserve surrounding tissues [14].

Sclerotherapy was selected as a conservative treatment approach because it leverages the physical, chemical, and biological properties of an agent to disrupt the targeted tissue [20,21]. This disruption facilitates the formation of sclerosed or "hardened" by-products, leading to significant alterations or reductions in function following therapy. Not only does it result in the occlusion of vascular structures akin to embolization, but it may also mitigate recurrence, proliferation, or collateralization by permanently disrupting the endothelium of the targeted vascular structures [22]. Moreover, the biological effects extend beyond structures with an endothelium, successfully targeting the epithelial lining of true cysts, capillary beds, lymphatic structures, and even bone cysts [14,20,21].

For a substance to be considered a potential sclerosant, it must exert a physical, chemical, and/or biological effect on the target tissue and prompt a regulated inflammatory response. This response arises from cellular damage and subsequent fibroblast proliferation, culminating in sclerosis. In addition to fibrosis, sclerosants may induce various effects such as thrombosis, extraction of proteins from lipids, protein denaturation, cellular dehydration via osmosis, and physical obstruction through polymerization. These processes collectively result in the controlled disruption of the target tissue's biological function [14]. To be effective, a sclerosant must diffuse through a fluid medium to reach its target tissue and interact with it for a sufficient duration to initiate the sclerosing process. Among the available sclerosing agents, sodium morrhuate, sodium sulfate tetradecyl, polidocanol, ethanolamine oleate, and hypertonic glucose solution have demonstrated excellent outcomes [14]. Sodium tetradecyl sulfate, an anionic surfactant approved by the FDA, is utilized as an intravascular sclerosing agent and has found extensive application in the treatment of vascular lesions. This sclerosant has been employed in various medical scenarios, including the treatment of varicoceles, vascular malformations of the extremities, upper gastrointestinal bleeding, variceal bleeding, hemorrhagic tumors, gallbladder ablation, lymphoceles, and percutaneous ablation of oral lesions such as Kaposi sarcoma and ganglion cysts [14,20,21].

The literature indicates that 3% STS is deemed safe for treating such cases, typically requiring a volume equivalent to one-fourth of the lesion size, equating to 0.5 ml of 3% STS for every 2 cm of lesion size [22]. Consequently, smaller lesion volumes necessitate less sclerosing agent injection. Moreover, Choi., *et al*. in 2016 underscored the manufacturer's recommendations for 3% STS, stating that 0.5-2.0 mL of 3% STS can be safely injected into a lesion per dose [21]. In this reported case, we adhered to the same protocol. However, due to the considerable size of the lesion, we adjusted the concentration to 1% to err on the side of caution, necessitating nearly three times the usual volume. The frequency of injections during follow-up was dictated by the texture and size of the lesion; given its substantial size, multiple injections were administered until the desired effect was achieved, at which point injections were ceased. Complications associated with STS primarily manifest at the administration site, including extravasation during percutaneous treatments.

Surgery is typically considered when systemic treatments fail to yield results or for aesthetic purposes, either as a standalone excision or in conjunction with plastic surgery. Both conservative and aggressive treatment approaches may be pursued for hemangiomas of the tongue, each with its drawbacks. Conservative treatments may result in frequent recurrences, while aggressive approaches can potentially lead to functional impairment [17,18]. Cryotherapy has been documented to yield favorable outcomes, with high success rates reported. Kutluhan [22] employed plasma knife surgery for the excision of hemangiomas of the tongue.

Conclusion

Hemangioma stands as the predominant benign vascular tumor in the maxillofacial area, necessitating careful management to avert early or persistent functional deficits or long-term aesthetic concerns. Typically, surgical excision is deemed necessary for such cases. However, given the extensive nature of surgical procedures, treatment with sodium tetradecyl sulphate presents a viable alternative, often resulting in successful remission of the lesion. This

therapeutic avenue holds promise in potentially reducing the necessity for surgical intervention.

Conflict of Interest

- **Author contributions:** All authors have sufficiently contributed to the study and agreed with the results and conclusions.
- **Funding:** No funding source is reported for this study.
- **Declaration of interest:** No conflict of interest is declared by authors.

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