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Case Report

Precision in needles: A case report of intralesional sclerotherapy for pediatric arteriovenous malformations

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ABSTRACT

This report describes the successful management of a superficial arteriovenous malformation (AVM) of the tongue in a 10-year-old female patient. The lesion was treated with weekly intralesional injections of 3% sodium tetradecyl sulfate (STS) over a period of three weeks under local anesthesia. Complete regression of the lesion was observed by the end of the third week, with no complications or recurrence noted after six months of follow-up. Intralesional sclerotherapy with STS proved to be a safe, effective, and minimally invasive treatment option for oral AVMs, providing a cost-effective alternative to surgery with excellent patient compliance and minimal adverse effects.

Keywords: Arteriovenous malformations, Pediatric, Sclerotherapy, Sodium tetradecyl sulfate, Tongue

INTRODUCTION

Vascular malformations (VMs) are a group of congenital anomalies resulting from developmental aberrations in the vascular system, characterized by abnormal proliferation of vascular (arterial, venous, and lymphatic) structures. Among them, arteriovenous malformations (AVMs), though uncommon, are characterized by their high-flow capability due to direct artery-to-vein communications that bypass the capillary network, forming a vascular tangle known as a "nidus."[1,2]

The clinical progression and proliferation of AVMs are influenced by factors such as ischemia secondary to thrombosis, hormonal changes during puberty, trauma, ectasia, and genetic predisposition.[3,4] AVMs occur at an early age in 40–60% of patients, with around 30% becoming clinically apparent during childhood or adolescence, while many manifest later in life. Traumatic AVMs typically involve a single vessel and are most commonly localized in the head-and-neck region, accounting for nearly 60–70% of cases.[5,6]

Clinically, AVMs may present as pulsatile masses accompanied by a thrill, bruit, and occasionally localized hyperthermia, bleeding, ulceration, necrosis, and impaired function due to reduced vascularity and nutrition. The overlying skin may exhibit a reddish or port-wine discoloration.[7,8]

Given their complex presentation, accurate diagnosis is crucial and typically involves a range of imaging modalities, including radiography, color Doppler ultrasound, computed tomography, magnetic resonance imaging, and angiography.[9] Subsequent management of these lesions presents considerable medical and psychosocial challenges, as they frequently result in

significant esthetic and functional impairments, necessitating a multidisciplinary approach. Treatment modalities conventionally comprise surgical resection, endovascular embolization, with recent advancements highlighting the efficacy of laser technologies and sclerotherapy.[10]

Despite advancements, surgical management remains complex due to risks such as massive intraoperative hemorrhage and the difficulty of replacing diseased vessels with healthy tissue. Hence, sclerotherapy emerges as the preferred modality for the management of AVMs, offering effective lesion control while minimizing trauma and preserving surrounding healthy tissues.

Although AVMs of the oral cavity have been reported, their occurrence on the lateral dorsum of the tongue in pediatric patients is relatively uncommon. Reports documenting successful management solely with sodium tetradecyl sulfate (STS) sclerotherapy in this location remain scarce, posing unique therapeutic challenges. This case report, documented in accordance with the CARE (CAse REport) guidelines, presents the successful management of an AVM through serial intralesional injections of the sclerosant agent, STS.

CASE REPORT

A 10-year-old girl presented to the dental outpatient department for the extraction of a root stump from a deciduous molar tooth. There was evident growth on the left lateral dorsum of the tongue with no history of bleeding, pain, ulceration, or burning sensation.

On intraoral examination, a solitary, localized, circumscribed, well-defined growth measuring approximately 1.5 × 2 cm was present on the left lateral dorsum of the tongue, in relation to 36, which was dark red to purplish in color [Figure 1]. On palpation, there was visible pulsation, and the growth was non-tender, soft in consistency.

A routine blood examination was advised and found to be within normal limits. The patient was advised to undergo an ultrasound of the tongue, which revealed a well-defined, heterogeneous hypoechoic lesion with lobulated margins measuring approximately 14 × 23 mm, located in the submucosal plane. The lesion is composed of multiple serpiginous, tubular, anechoic channels. Color Doppler imaging demonstrates high-velocity, low-resistance arterial flow with evidence of arteriovenous shunting. The lesion is non-compressible on graded pressure and shows no involvement of deeper muscular or osseous structures. These findings are suggestive of a high-flow AVM.

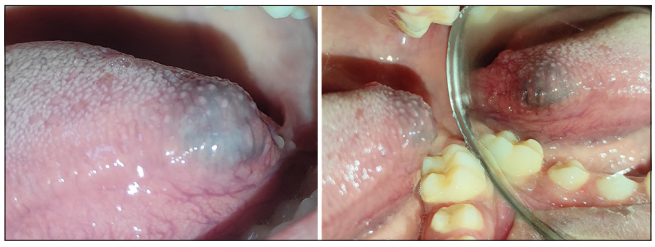

Figure 1: Arteriovenous malformation on the left lateral dorsum of the tongue.

Treatment

A comprehensive explanation of the available treatment modalities was provided to the patient's parents. Following the procurement of taking assent from the patient along with informed consent from the parents and administration of local anesthesia, an initial therapeutic intervention was made by administering an intralesional injection of 5 cc of boiling distilled water. However, this intervention proved ineffective, as the lesion exhibited no signs of improvement after a week. Subsequently, from the 2nd week onward, the therapeutic approach was revised. Under stringent aseptic conditions, 1 mL of 3% STS (Setrol, Samarth Life Sciences Pvt., Ltd., Goregaon [West], Mumbai) was meticulously administered through intralesional injections in a circumferential manner around the lesion once weekly for three consecutive weeks [Figures 2-4]. The procedure was conducted under local anesthesia achieved through a lingual nerve block, using a syringe (Dispo Van, Hindustan Syringes and Medical Devices Ltd., Faridabad, Haryana) fitted with a 26-gauge needle to ensure precise delivery of the sclerosant. Following each session, the patient was prescribed analgesics (Syrup Ibugesic Plus) to ensure adequate pain management. By the end of the 3rd week of clinical follow-up, complete lesion regression was noted, with no adverse effects reported, indicating a favorable therapeutic outcome [Figure 5].

DISCUSSION

Vascular anomalies encompass a spectrum of lesions originating from arterial, venous, and/or lymphatic channels. Mulliken and Glowacki introduced a biological classification system based on endothelial cell behavior, distinguishing lesions characterized by endothelial proliferation from those exhibiting structural anomalies, now recognized as VMs.[11,12] The International Society for the Study of Vascular Anomalies has since refined this classification, differentiating vascular tumors from malformations based on clinical presentation, imaging findings, histopathology, and biological behavior. AVMs are categorized as isolated fast-flow VMs within this framework.[13,14]

The etiology of AVMs is attributed to aberrations during embryogenesis, where improper differentiation and maturation of the primitive vascular plexus result in the persistence of arteriovenous channels. This leads to the formation of a central nidus composed of tortuous

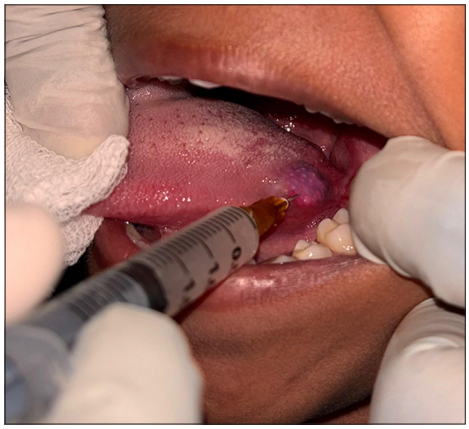

Figure 2: Intralesional injection of 3% sodium tetradecyl sulfate.

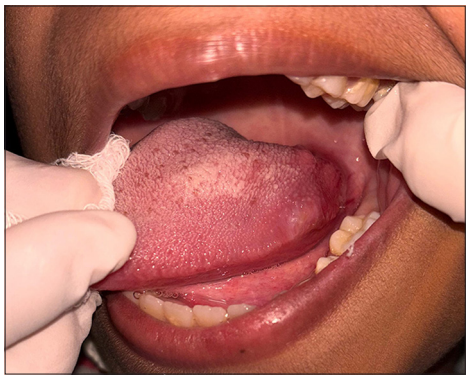

Figure 3: 1st-week follow-up.

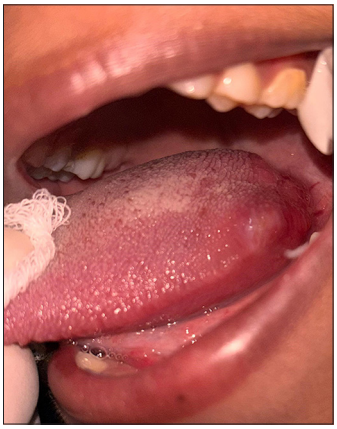

Figure 4: 2nd-week follow-up.

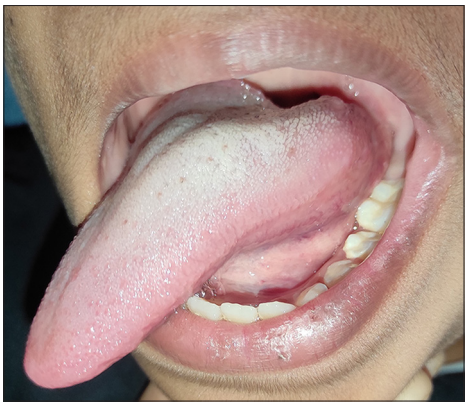

Figure 5: 3rd-week follow-up.

vascular loops.[11,15] Aberrant angiogenic signaling involving growth factors such as vascular endothelial growth factor, angiopoietin-2, and transforming growth factor beta, along with somatic mutations in genes such as mitogen-activated protein kinase kinase 1 (*MAP2K1*), kirsten rat sarcoma viral oncogene homolog (*KRAS*), and RAS p21 protein activator 1 (RASA1), contributes to the pathogenesis of AVMs by disrupting endothelial function and vascular architecture.[16-19] Persistent hemodynamic stress resulting from arteriovenous shunting contributes to vascular remodeling, collateral recruitment, and lesion expansion, thereby elevating the risk of ulceration and hemorrhage, particularly in mucosal or superficial locations.

Although AVMs within the oral cavity are rare, their clinical significance is underscored by their potential for substantial complications. AVMs of the jaw have an estimated incidence of fewer than 1/100,000 individuals annually.[20,21] Prompt identification and appropriate management are critical given their potential morbidity.

Multiple factors influence the clinical behavior and proliferation of AVMs, including ischemia from thrombosis, hormonal fluctuations during puberty, trauma, ectasia, and genetic predisposition.[7,8] Approximately 40–60% of AVMs develop early in life, with nearly 30% becoming clinically evident in childhood or adolescence; nonetheless, many cases present later in life. Traumatic AVMs generally involve a solitary vessel and predominantly affect the head-and-neck region, accounting for 60–70% of all cases.[9,10]

In a landmark study by Kohout *et al.*, the anatomical distribution of 81 head-and-neck AVMs revealed the cheek (31%) as the most frequent site, followed by the ear (16%), nose (10%), upper lip (7%), mandible (5%), neck (5%), scalp (5%), and maxilla (4%).[3] Intraorally, the tongue remains the most prevalent site of AVM involvement, as exemplified in the present case. Less common intraoral locations include the gingiva, lips, palate, and buccal mucosa. AVMs in these regions may compromise speech, mastication, and swallowing, and are vulnerable to trauma-induced ulceration and secondary infection.[22,23]

Clinically, AVMs may manifest as pulsatile masses exhibiting thrill, bruit, localized warmth, and occasional symptoms such as bleeding, ulceration, necrosis, or functional impairment due to compromised vascular integrity and nutrition. The overlying mucosa or skin may present a reddish or port-wine hue.[11,12]

Historically, a broad array of treatment modalities has been utilized for AVMs, including surgical excision, radiotherapy,

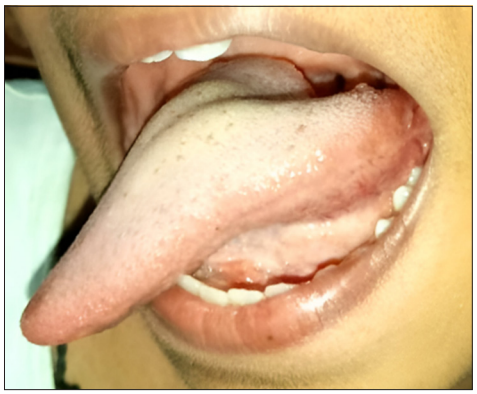

Figure 6: 6th-month follow-up.

cryosurgery, electrocoagulation, isotope therapy, systemic corticosteroid therapy, interferon-α therapy, endovascular embolization, as well as recent advancements highlighting the efficacy of laser therapy and sclerotherapy.[24-26]

Sclerotherapy, in particular, has emerged as a conservative, cost-effective strategy for managing benign vascular lesions such as AVMs, especially those located in surgically challenging regions. The technique results in minimal scarring and generally carries fewer complications than excisional procedures. A range of sclerosing agents has been employed with varied success, including STS, 5% sodium morrhuate, 5% ethanolamine oleate, sodium phyllite, 1% polidocanol, quinine urethane, chromated glycerin, polyiodinated iodine, Picibanil, sodium silicate, pingyangmycin, bleomycin, hypertonic saline, boiling distilled water, and absolute ethanol, alone or in combination.[26,27] Among these agents, STS is particularly advantageous due to its low incidence of allergic reactions, minimal local tissue necrosis compared to agents such as absolute ethanol, ease of administration, cost-effectiveness, and favorable safety profile, making it especially suitable for pediatric oral lesions.

Nonetheless, sclerotherapy is not devoid of risks; potential adverse events, including superficial ulceration, tissue necrosis, sloughing, swelling, secondary infection, transient neuropathy, hemorrhage, hyperpigmentation, and anaphylaxis are seen in some rare cases.[28] Despite these concerns, recent literature – including reports by Kwon *et al.* (2023),[29] Kaur *et al.* (2021),[30] Singal and Bhatt (2020),[31] Min *et al.* (2015),[32] and Choi *et al.* (2016)[33] – demonstrates favorable outcomes following 3% STS sclerotherapy for oral AVMs and similar vascular lesions such as mucocele or hemangioma, with most studies indicating substantial lesion regression, minimal complications, and low recurrence rates.

In the current case, intralesional sclerotherapy with 3% STS was performed circumferentially around the lesion under local anesthesia, administered weekly over three weeks. The treatment yielded complete lesion resolution by the end of the 3rd week, without any complications or scarring, and no recurrence was observed after six months of follow-up [Figure 6]. These findings reinforce the efficacy and safety of STS sclerotherapy in managing superficial high-flow AVMs of the tongue, particularly highlighting its utility in pediatric patients due to its minimally invasive nature and favorable risk profile.

Limitations

- The present report is limited by its single-case design, which restricts the generalizability of the findings.
- Histopathological confirmation was not obtained due to the non-excisional nature of treatment, potentially limiting diagnostic certainty.
- The follow-up period was confined to six months, which may be insufficient to detect late recurrences or long-term complications.

CONCLUSION

Sclerotherapy with STS has been proven to be a safe, simple, and effective treatment for superficial mucosal AVMs, resulting in complete remission without any complications or recurrence. Given its minimally invasive nature, low cost, absence of scarring, and high patient compliance, especially in pediatric patients, sclerotherapy is a valuable therapeutic approach, particularly for small and uncomplicated AVMs. Prompt diagnosis and treatment remain essential. Further comparative studies with careful case selection and a thorough understanding of potential risks are essential to establish sclerotherapy as a reliable treatment modality for such lesions.

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