CASE REPORT

SCLEROTHERAPY IN ORAL VASCULAR MALFORMATION – AN INSTITUITIONAL EXPERIENCE.

Dr. Jayanta Saha*, Dr. Abhishek Khatua**

ABSTRACT

Managing vascular anomalies of the oral and maxillofacial area is a challenging task for any surgeon. The variable nature of the vascular lesions, varying size and disfigurement adds to the difficulty in treating them. Surgical excision, embolization is a treatment of choice but has more morbidity and is infrastructure intensive.This approach has an increased burden to the patients financially as well as it will require hospital admission, multiple specialties of doctors, operating room cost etc. Thus, we treated our patients with multiple intralesional injection of sclerosing agents. The option of surgical resection was kept as an adjunct in case the injection therapy was not up to the mark. Patients were kept on weekly follow up and injectables were administered at a gap of 2 weeks depending on the size of the lesion. Sodium tetradecyl sulphate was the sclerosing agent of choice in this case series due to its reliability, low cost and easy availability. Complete resolution of the lesion was found following sclerotherapy.

KEY WORDS

Sclerotherapy, vascular malformation, sodium tetradecyl sulphate, maxillofacial, oral, sclerosing agent.

ABOUT THE AUTHORS

*Assistant Professor
Department of Oral and Maxillofacial Surgery
Burdwan Dental College and Hospital, Burdwan
**Assistant Professor
Department of Oral and Maxillofacial Surgery
Dr. R. Ahmed Dental College and Hospital, Kolkata

CORRESPONDING AUTHOR

Dr. Abhishek Khatua
Assistant Professor
Department of Oral and Maxillofacial Surgery
Dr. R. Ahmed Dental College and Hospital, Kolkata

INTRODUCTION

Vascular anomalies are a varied lesions resulting from abnormal number, structure, or position of blood vessels. They are generally a congenital or acquired lesion that occurs in the oral cavity, face, neck and may or may not be associated with syndromes. James Wardrop first differentiated between haemangioma and vascular malformation (VM) in 1818. Clinically they appear bluish, elevated, compressible. Most VMs are low flow lesions associated with small capillaries or veins and is slow filling. Lesions associated with any artery are high flow lesions and are termed as arterio-venous malformation (AVM). AVMs are excluded from this case series and vascular malformations of capillary or venous origins are considered for this series.

Various treatments are reported in literature including laser therapy, embolization, electrocauterization, steroid injections, surgical removal, or sclerotherapy. They may be used independently or in conjunction. Non-surgical therapies are generally used to reduce the size of the lesion and followed up by surgical excision. Here, we are using recurrent intralesional injection of sclerosing agent as the sole therapy for small to moderate lesions.

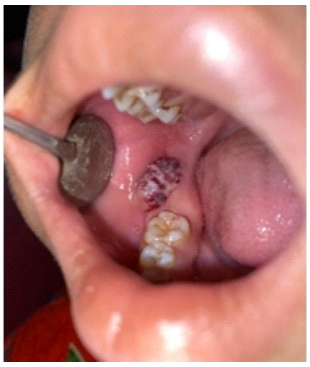
Several sclerosing agents are available for use but most widely used is sodium tetradecyl sulphate (STS) which is known to cause extra-vascular inflammation and thrombosis, causing ischemia, occlusion of abnormal vessels and subsequent necrosis of tissue. We hereby discuss few cases where sclerotherapy was used as a sole therapy for the treatment of VM. Our institutional protocol of STS injection was injecting every 2 weeks, whose frequency depended on the size and response of the lesion.

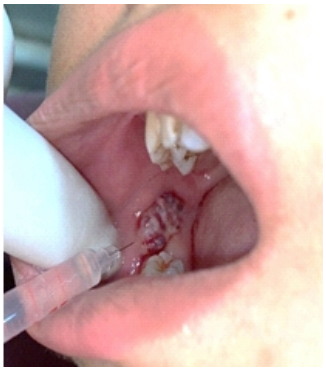
CASE REPORT

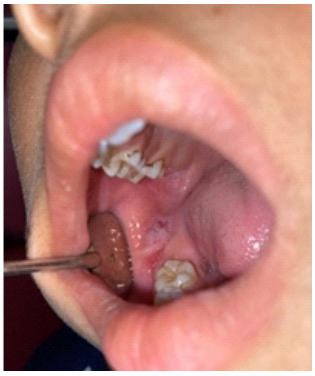
Case 1

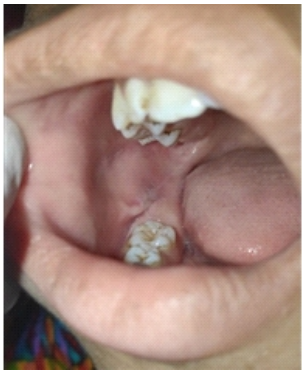
A 12 years old female patient reported to the outpatient department (OPD) of Oral and Maxillofacial Surgery (OMFS) Department of Burdwan Dental College and Hospital, Burdwan,

Case 1





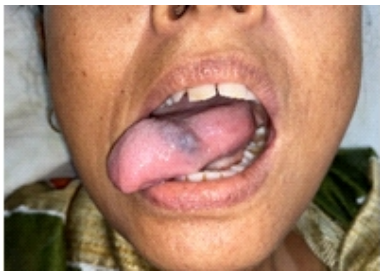


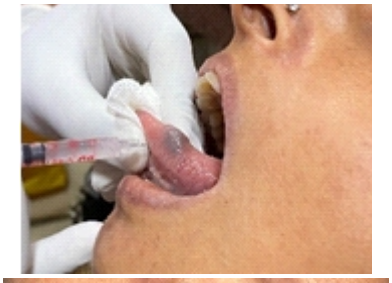


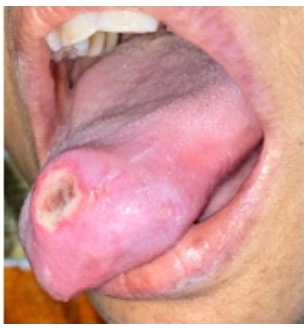
West Bengal, with chief complaint of small reddish swelling on the right retromolar region. Swelling was recurrent and occurs following cheek bite or spontaneously. The swelling was 1.5 cm by 1 cm in size, reddish, bleeds on touch. The bleeding subsides on its own. A diagnosis of low flow vascular malformation was made and a treatment plan of sclerotherapy with sodium tetradecyl sulphate (STS) was decided. 10 units of STS was injected intra lesionally and patient was followed up weekly. Patient and parents were informed about possible complications like local ulceration, swelling, infection, haemorrhage, possible recurrence. The patient was kept under antibiotics, local styptics measure if required. Second and third injections (about 1ml of STS) were given 2 weeks apart. Complete regression of the lesion was noted after 4 weeks.

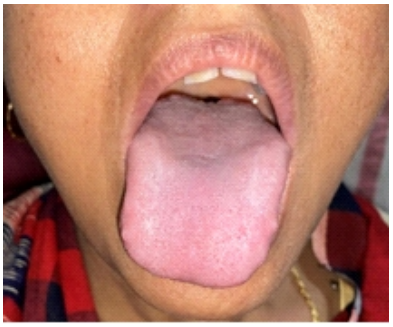
Case 2

A 52 year old female patient reported to the OPD with a congenital swelling of left lateral border of the tongue with a bluish discoloration and a dome shaped swelling of about 2.5 cm by 2.5 cm which was compressible, slow filling, painless. Diascoscopy test was positive. Clinical diagnosis of VM was made and sclerotherapy with STS (1.5 ml) was the treatment

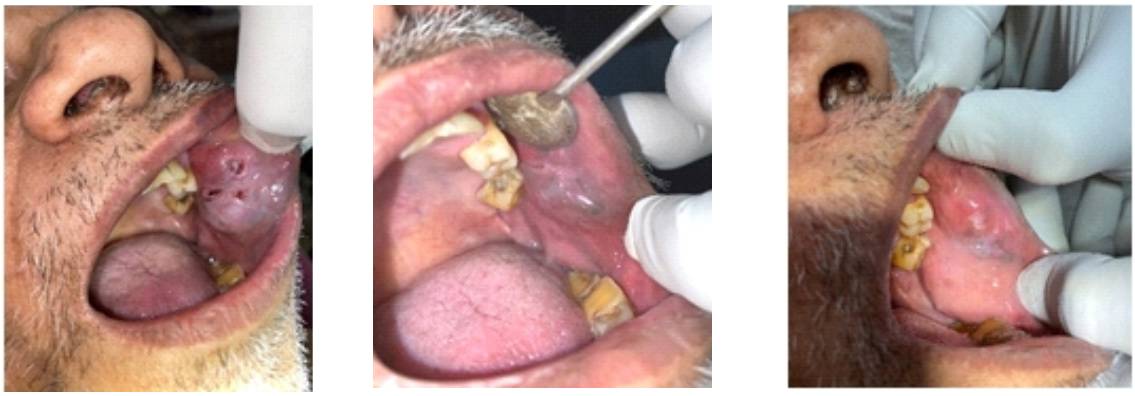




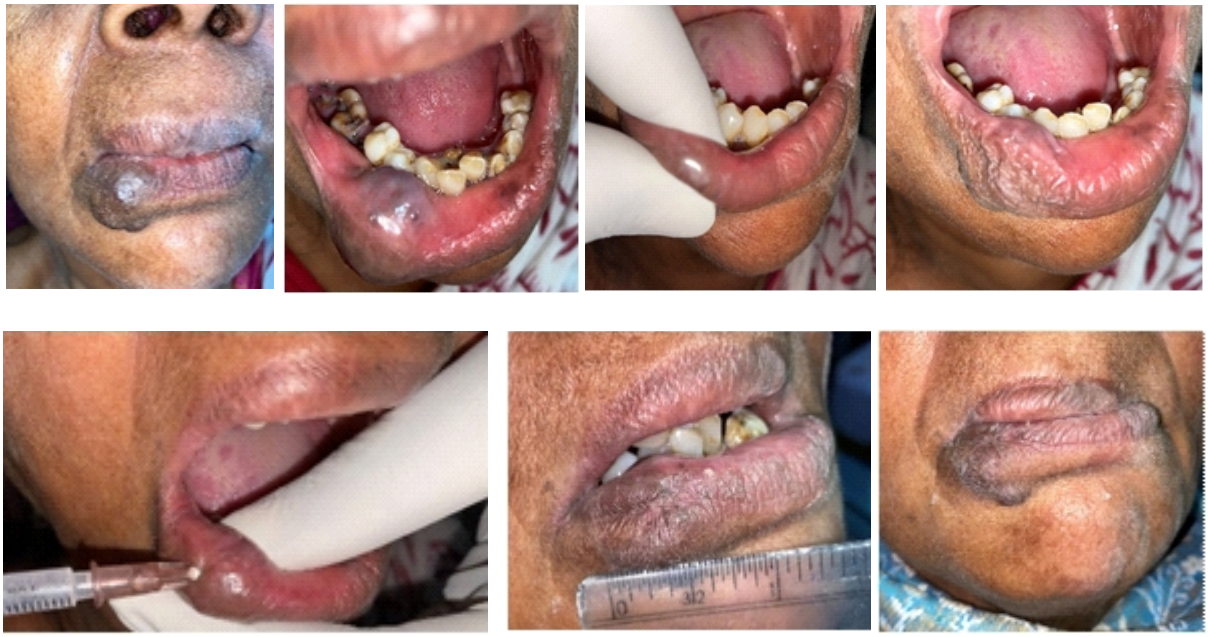




Case 2



Case 3



Case 4

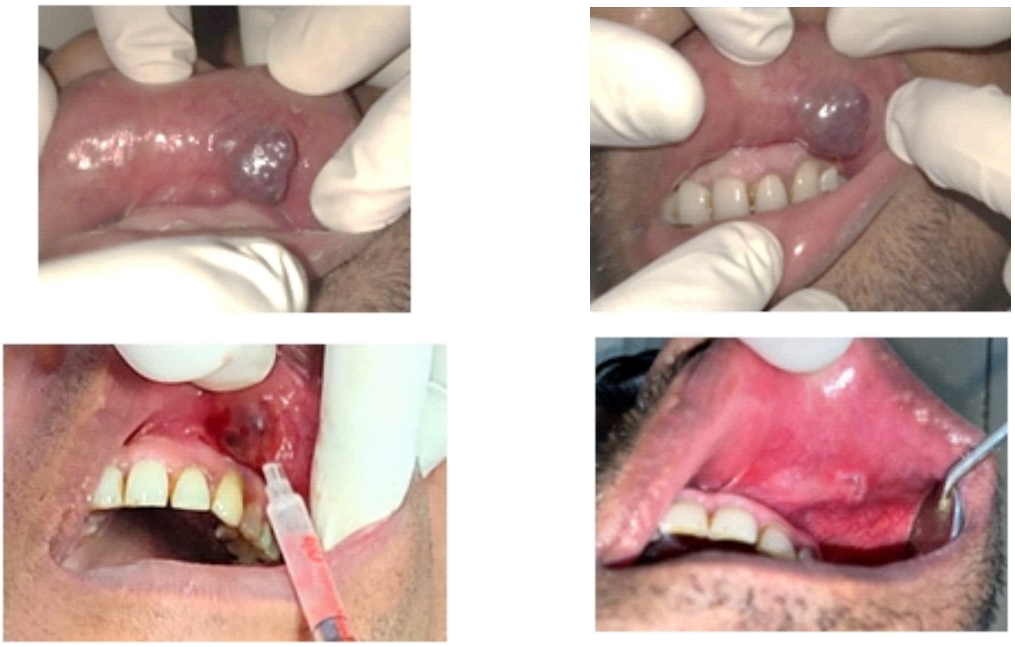
plan decided as per our institute protocol. During the first week follow up, patient gave history of swelling and pus discharge which got resolved with the antibiotics prescribed during the first injection. On examination, we found some crater like ulcerations on the surface of the tongue, but the lesion had decreased in size. 2nd Injection was given at the 2nd week period. The healing was uneventful. Final injection was administered at 4th week. Complete resolution of the lesion was seen with some residual fibrosis of the area.

Case 3

A 60 years old male patient reported with a complaint of a slow growing, large mass on the left buccal mucosa. The lesion was about 4cm by 3cm in size, elevated, erythematous, compressible, diascopy-positive. Clinically diagnosed as a VM; sclerotherapy with STS was started. More than 2 ml of STS was injected first time. 7 days follow up revealed some ulcerations and areas of necrosis as was expected. 4 sessions of injection was required for this large lesion. Complete resolution of the lesion was noted. Some residual fibrosis was observed but essentially the healing was uneventful.

Case 4

A 59 years old female patient reported with complaint of swelling in the right lower lip since birth. Lesion was dome shaped swelling with a bluish tinge, compressible, slow filling, painless. There was some hyperpigmentation on the skin covering the



Case 5

lesion. It was diagnosed as a VM and sclerotherapy was started. Mild ulceration and crustation was seen post injection. The lesion regressed in size after 3 sessions of STS intralesional injection.

Case 5

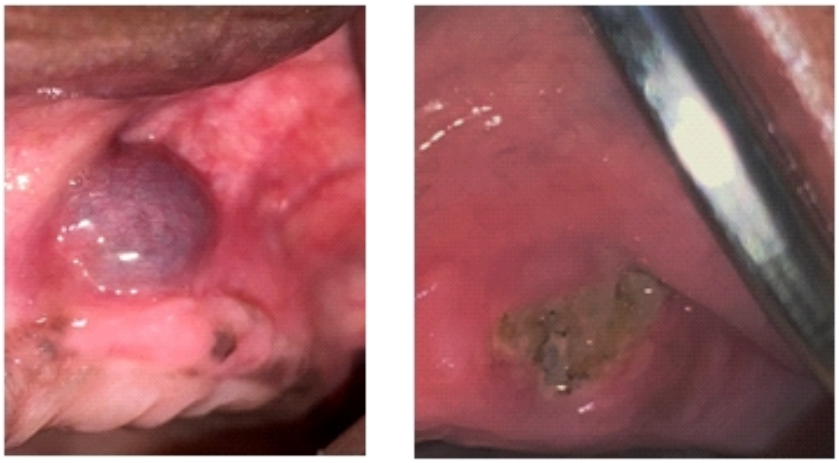
A 30 year old male patient reported with a complaint of a blood filled swelling on the inner aspect of left upper lip which bled if he accidentally bit on it. On examination, a well-defined, erythematous spherical swelling of about 2cm by 2 cm in size. Considering history and clinical presentation, it was diagnosed as VM and sclerotherapy treatment was initiated with STS. Mild ulceration, swelling, fibrosis was noted at post injectionstperiod which reduced on its own. Complete resolution of the lesion was noted.

Case 6

An 80 years old patient reported with a small ovoid bluish mass over the left alveolus of upper edentulous ridge of about 1cm by 0.5 cm in size, which was causing difficulty to fabricate a denture. The lesion was diagnosed as a VM and was treated with sclerotherapy resulting in complete resolution and healing.

DISCUSSION

Vascular Malformations are rare lesions with wide variety of presentations. They fall inside a spectrum of a small birth anomaly to a life threatening debilitating condition. They are generally caused due to embryonic remnants of a developmental stage. It is difficult for surgeons treating VM s to reach a consensus on a definite treatment plan owing to its



Case 6

erratic presentation, puzzling classification, nomenclatures and varied response to treatment.[2-5] Mulliken and Glowacki classified congenital vascular malformation of the maxillofacial region. They divided them into Hemangioma and Vascular malformation.[1] Hemangiomas are more common, superficial tumours present congenitally that generally grows rapidly in the initial phase but slowly regresses as the child grows. VM are growths that do not regress spontaneously but grows slowly with the child. VMs are be further subdivided into low flow versus high flow lesions, depending on the blood flow rate. Low-flow lesions include capillary, venous, lymphatic, or mixed malformations and High-Flowlesions include arteriole, arteriovenous, fistulae, or shunt malformations.[6]

VM appears bluish, elevated, compressible non-pulsatile with varying degree of depth from the overlying tissue. Many VMs are asymptomatic, but may cause a few episodes of bleeding that would be difficult to stop. They usually do not resolve on its own, may get secondarily infected[2]. Aesthetic concern maybe a primary intention for patients to seek treatment. Diagnosis is mainly clinical and can be supplemented with Doppler ultrasound to check the flow rate. CT angiography and MR angiography may be used to identify any feeder vessels especially in high flow lesions.[10] Diascopy test may be done at bedside to differentiate between other erythematous lesions and a VM.

Treatment of oral VM ranges from Surgical excision, embolization, steroids, lasers, cryotherapy, sclerotherapy or a combination of them. Usually, treatment plan is dictated by the type, size, location of the lesions. It also depends on other factors like availability of resources, patients' socioeconomic status, finances. Historically, most commonly used treatment was surgical excision even for smaller lesions. But owing to facial disfigurement, proximity to vital structures, unavailability of resources leads to the introduction of sclerotherapy[11]. In resource-poor settings sclerotherapy can be a very competitive treatment for oral VM especially that are smaller in size.

The sclerosants used to treat the condition include ethanolamine oleate, ethibloc, bleomycin, and sodium tetradecyl sulphate[1,12]. These agents commonly provide a stimulus to vascular endothelial intima, induce extra-vascular inflammatory reactions, and ultimately cause vascular fibrosis and occlusion. Ethanolamine oleate is known to be a sclerosant with the lowest recurrence rate, although it can affect normal structures if it flows into normal tissues[1]. Hassan et al.[11] injected a maximum of 15 mg bleomycin into intravascular lesions, three or four times on average and six times at maximum, in 69 patients and observed definite improvement in 71% of the patients. This result demonstrates that simple, non-invasive sclerotherapy, instead of surgical treatment, can generate good and aesthetically pleasing results.

Similar to the other sclerosants, sodium tetradecyl sulphate also induces extra-vascular inflammatory reactions and causes vascular fibrosis and occlusion. Baurmash and Mandel[14] reported that sodium tetradecyl sulphate injected into vascular malformation lesions could induce allergic and anaphylactic reactions. An injection of 0.5 to 2 mL per dose in lesions is possible, and the minimum dose is recommended. In addition, an injection dose greater than 2 mL is not recommended in one lesion, but interval injections over 5 to 7 days are recommended, based on the manufacturer's instructions.

In these cases, since the lesions were in the outer layer and were relatively small, sclerotherapy that applied 1% sodium tetradecyl sulphate injected directly into the lesions was used to minimize both the side effects of the agent and the surgical burden on the patient. Multiple injections were given to the patients in this case series depending on the size and response of the lesions at accurate intervals. The treatment was finished when no recurrence was found in either patient. For deeper lesions, sclerotherapy may be used as adjunct to surgical excision.[13]

CONCLUSION

Vascular malformation is a group of disease affecting various areas of the mouth and face, with significant morbidity to the patients. Surgical excision for all lesions, caries the risk of excess bleeding, facial disfigurement and scarring which may add on to the post operative morbidities. Hence sclerotherapy may be a useful alternative to surgical excision for small to moderate lesion with significantly less burden to the patients.

DECLARATION

We, the authors, declare no conflict of interest.

We, the authors. declare that no financial aid was obtained from any source.

We, the authors, declare that consent from all patients was taken regarding publication.

We declare that this article is not published or being considered for publications anywhere else as far our knowledge.

REFERENCES

1. Zheng JW, Zhou Q, Yang XJ, Wang YA, Fan XD, Zhou GY, et al. Treatment guideline for hemangiomas and vascular malformations of the head and neck. Head Neck. 2010; 32:1088–1098. PMID: 19924783.

2. Kohout MP, Hansen M, Pribaz JJ, Mulliken JB.

Arteriovenous malformations of the head and neck: natural history and management. PlastReconstr Surg. 1998; 102:643–654. PMID: 9727427.

3. Hohenleutner S, Badur-Ganter E, Landthaler M, Hohenleutner U. Long-term results in the treatment of childhood hemangioma with the flashlamp-pumped pulsed dye laser: an evaluation of 617 cases. Lasers Surg Med. 2001; 28:273–277. PMID:11295764

4. Kim JY, Kim DI, Do YS, Lee BB, Kim YW, Shin SW, et al. Surgical treatment for congenital arteriovenous malformation: 10 years' experience. Eur J VascEndovasc Surg. 2006; 32:101–106. PMID: 16478673.

5. Sadan N, Wolach B. Treatment of hemangiomas of infants with high doses of prednisone. J Pediatr. 1996; 128:141–146. PMID: 8551406.

6. Ramírez-Amador V, Esquivel-Pedraza L, Lozada-Nur F, De la Rosa-García E, Volkow-Fernández P, Súchil-Bernal L, et al. Intralesional vinblastine vs. 3% sodium tetradecyl sulfate for the treatment of oral Kaposi's sarcoma: a double blind, randomized clinical trial. Oral Oncol. 2002; 38:460–467. PMID: 12110340.

7. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. PlastReconstr Surg. 1982; 69:412–422. PMID: 7063565.

8. Waner M, Suen JY. Hemangiomas and vascular malformations of the head and neck. New York: Wiley-Liss;1999.

9. Buckmiller LM, Richter GT, Suen JY. Diagnosis and management of hemangiomas and vascular malformations of the head and neck. Oral Dis. 2010; 16:405–418. PMID: 20233314.

10. Dubois J, Garel L. Imaging and therapeutic approach of hemangiomas and vascular malformations in the pediatric age group. PediatrRadiol. 1999; 29:879–893. PMID: 10602864.

11. Hassan Y, Osman AK, Altyeb A. Noninvasive management of hemangioma and vascular malformation using intralesional bleomycin injection. Ann Plast Surg. 2013; 70:70–73. PMID:23249476.

12. da Silva WB, Ribeiro AL, de Menezes SA, de Jesus Viana Pinheiro J, de Melo Alves-Junior S. Oral capillary hemangioma: a clinical protocol of diagnosis and treatment in adults. Oral Maxillofac Surg. 2014; 18:431–437. PMID: 24263242.

13. Johann AC, Aguiar MC, do Carmo MA, Gomez RS, Castro WH, Mesquita RA. Sclerotherapy of benign oral vascular lesion with ethanolamine oleate: an open clinical trial with 30 lesions. Oral Surg Oral Med Oral Pathol Oral RadiolEndod. 2005; 100:579–584. PMID: 16243243.

14. Baurmash H, Mandel L. The nonsurgical treatment of hemangioma with Sotradecol. Oral Surg Oral Med Oral Pathol. 1963; 16:777–782. PMID: 13967026.

