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SUCCESS AND FAILURE WITH INTRALESIONAL 3% SODIUM TETRADECYL SULFATE SCLEROSING AGENT IN TWO CASES OF INTRAORAL HEMANGIOMA

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ABSTRACT: Vascular malformations are one of the most common lesions of the oral cavity. The lesion may be a congenital malformation observed in neonates or arteriovenous malformation observed in adults. The treatment of these lesions includes surgical excision, cryotherapy, selective embolization and treatment with intralesional sclerosing agents. We present two cases of benign oral vascular lesions treated with intralesional injection of 30mg/ml sodium tetradecyl sulfate. One lesion virtually disappeared after one session of sclerotherapy, leaving a fibrotic lesion. No side effects were observed. The other lesion resulted in a severe inflammatory response including pain, swelling and surface ulceration up to more than 2 weeks. Sclerotherapy with sodium tetradecyl sulfate is effective in treating smaller benign oral vascular lesions, and the use of the sodium tetradecyl sulfate provides alternative or support for surgical methods. Larger lesions in critical anatomical areas may not be the best choice for treatment with Sclerosing agents. This paper presents the success and failure of vascular lesions treated with sclerosing agent.

INTRODUCTION: Vascular malformation is a generalized term used to describe a group of lesions, formed by an anomaly of angiovascular or lympho vascular structures.

Vascular malformations are the most common tumours of the head and neck region in infants and children. They typically present at birth, enlarge during the first year of life, and then usually spontaneously involute by 5 years of age. For this reason, treatment is often unnecessary. In adults (particularly elderly persons), benign vascular proliferations are generally varicosities. Vascular malformations are structural anomalies of blood vessels without endothelial proliferation. By definition vascular malformations are present at birth and persist throughout life. They can be

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categorised according to the type of vessels involved (capillary, venous, arterial) and according to hemodynamic features (low flow or high flow). Approximately 85% of childhood onset haemangioma spontaneously regress after puberty, whereas a varix arises in older individuals, and once formed, does not regress¹.

The word “haemangioma” has been widely used in the medical and dental literature with reference to a variety of different vascular anomalies which has traditionally led to a significant amount of confusion regarding the nomenclature of these lesions. In 1982, Mulliken and Glowacki described a classification scheme which is presently accepted². These vasoformative tumours are classified under 2 broad headings of haemangioma and vascular malformation. Haemangioma is further sub classified based on their histological appearance as: (1) capillary lesions; (2) cavernous lesions; and (3) mixed lesion^{2,3}.

The classification proposed by Mulliken and Glowacki suggests that the term haemangioma can be reserved for lesions with cellular proliferation that present in infancy and usually involute by adolescence. Of the lesions restricted to the oral cavity, the tongue and floor of the mouth are the usual sites⁴. This paper highlights two cases of vascular malformation reported in the department, with the success and failure of intralesional sclerosing agents used as treatment modalities for the vascular malformation of head and neck region.

Case Report 1: A 59 year old male patient presented with a swelling in the ventral surface of the tongue at the junction of the tongue and floor of the mouth since 20 yrs.

There was no change in size of the swelling since 20 yrs. There were no associated features of pain, difficulty in the speech and swallowing. Past medical, dental and family histories were non-contributory and on physical examination no similar lesions were found, he appeared to be healthy with all vital signs within normal limits. The lesion was a solitary localized swelling seen in the ventral surface of the tongue, lateral to midline of size 1x3.5 cm surrounded by erythema (**Fig. 1**). Swelling was firm in consistency, non mobile, non tender, with no palpable thrills and blanched on

compression (diascopy). Aspiration of swelling was positive for blood. Ultrasound revealed ill-defined heterogeneous lesion noted with highly vascular and hypo echoic areas.



FIG. 1: PRE OPERATIVE PHOTOGRAPH SHOWS SWELLING IN THE VENTRAL SURFACE OF THE TONGUE SURROUNDED BY ERYTHEMA

Treatment: Based on the clinical diagnosis of the lesion, treatment with intralesional injection of 3% sodium tetradecyl sulfate (sclerosing agent) was planned.

First Visit: After anaesthetising the area, intralesional injection with 1 ml of 3% sodium tetradecyl sulfate diluted with 4ml of distilled water was given using a 25 gauge needle at multiple sites.

Recall Visit: Patient was recalled on the next day to check for development of any acute adverse reactions and the lesion appeared completely asymptomatic on examination. Patient was recalled again after a week for clinical examination, which showed complete regression and fibrosis of the lesion suggestive of good response of the lesion to the sclerosing agent (**Fig. 2**).



FIG. 2: POST OPERATIVE PHOTOGRAPH SHOWS COMPLETE REGRESSION AND FIBROSIS OF THE LESION

Case report 2: A 55-year old male presented with a swelling involving the right half of the tongue since childhood, increased in size since past 15 yrs gradually to attain the present size. There were no associated features of pain, difficulty in the speech and swallowing. Past medical, dental and family histories were non-contributory and on physical examination, he appeared to be healthy with all vital signs within normal limits.

A solitary localized swelling present in the dorsum of the tongue of size 3x7 cm extending anteriorly from the tip of the tongue, 7cm posterior, medially from the middle of the tongue, to the lateral border of the tongue. Surface appeared pebbled and erythematous in the lateral border of the tongue. Borders are ill-defined. Surrounding area appeared pale pink in colour. The swelling was soft to firm in consistency, non mobile, non tender, with no palpable thrills and blanched on compression (diascopy). Aspiration of swelling in the dorsum of tongue revealed blood (**Fig. 3**).



FIG. 3: PRE OPERATIVE PHOTOGRAPH SHOWS SWELLING PRESENT IN THE DORSUM OF THE TONGUE OF SIZE 3X7 cm

Depending on the clinical features, a working diagnosis of hemangioma with differential diagnosis of AV malformation and lymphangioma was made. The routine blood investigations were normal. Colour Doppler ultrasound revealed ill-defined heterogeneous lesion noted in the right anterior and lateral aspect of posterior part of the tongue. The lesion was highly vascular. No feeding vessel was noted. Flow inside the lesion was predominantly arterial.

MRI revealed that a large lobulated well defined homogenous soft tissue mass in the right half of the tongue measuring 40x46x60mm which appears

hyper intense on T2W and Short T1 inversion recovery (STIR) sequence and mildly hyper intense to adjacent muscles on T1W sequence suggestive of haemangioma of right side tongue with multiple phleboliths (**Fig. 4**).

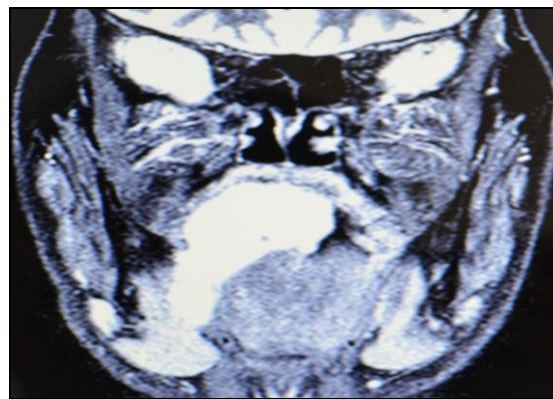


FIG. 4: MRI REVEALED A WELL DEFINED LARGE LOBULATED HOMOGENOUS MASS IN THE RIGHT HALF OF THE TONGUE WHICH APPEARS HYPERINTENSE ON T2W

Treatment: Based on the provisional diagnosis of the lesion, treatment with intralesional injection of 3% sodium tetradecyl sulfate (sclerosing agent) was planned

First Visit: After anaesthetising the area, intralesional injection with 1 ml of 3% sodium tetradecyl sulfate diluted with 4ml of distilled water was given using a 25 gauge needle at multiple sites.

Recall Visit: The patient presented with painful diffuse enlargement of the tongue which appeared erythematous with multiple ulcers in the injection sites with tender necrotic floor. It was clinically diagnosed as local inflammatory reaction to the sclerosing agent and was managed by intramuscular injections of Dexamethasone (2mg) and chlorphenaramine maleate (25 mg) followed by oral intake of chlorphenaramine maleate tablet, 25mg for 3 days.

Patient was recalled after three days for follow up. Patient reported reduction in pain and discomfort. Examination revealed reduction in the swelling, with persistent ulcers. Irrigation of the ulcer was done with hydrogen peroxide followed by prescription of chlorhexidine mouthwash. Patient was recalled after a week which revealed complete resolution of acute allergic symptoms and healing of ulcers in the injection site (**Fig. 5**).



FIG. 5: POST OPERATIVE PHOTOGRAPH SHOWS ENLARGEMENT OF THE TONGUE WITH NO IMPROVEMENT WITH SCLEROSING AGENTS

DISCUSSION: Identification and classification of vascular anomalies were hampered historically by the use of confusing nomenclature. Early classifications published by Virchow and Wagner characterized vascular lesions according to the vessel's pathologic appearance⁵. Vascular growths were divided into angiomas and lymphangiomas. The biologic behaviour and natural history of the vascular lesions were not considered. In 1982, Mulliken and Glowacki made great strides to dispel this confusion when they published a classification of vascular birthmarks, grouping them into two major categories: haemangioma and malformations. More specifically, haemangioma were differentiated from vascular malformations by their clinical appearance, histopathologic features, and biologic behaviour². In 1996, the classification was modified slightly to reflect the importance of other types of vascular tumours that exhibit different clinical and histologic characteristics than the common infantile haemangioma, including kaposiform hemangio-endotheliomas, tufted angiomas, and others.

Vascular malformation is a common benign vascular tumour and can often be found in the skin and mucosa of the head and neck region, and relatively rare in the oral cavity. Oral cavity including the lips, buccal mucosa, or tongue. Vascular malformation in the oral cavity causes discomfort and potentially serious clinical problems. This lesion in the tongue can cause particular problems consisting of recurrent hemorrhage, biting of the lesion, pain, and difficulty with speaking, mastication, or deglutition.

Clinically vascular malformations are soft, sessile or pedunculated, and painless. They may be smooth or irregularly bulbous in outline. Color varies from deep red to purple and the tumour blanches on the application of pressure.

Vascular malformation do not metastasize, but rather proliferate or involutes with time. Conventional radiography of the affected area is usually the initial diagnostic study obtained in patients suspected of having a soft tissue mass and will often reveal an ill defined soft tissue prominence or mass. In many cases, the radiographs are normal. However, the other major soft tissue finding is phleboliths, which can occur in 20% to 67% of cases. Modalities of treatment of benign vascular lesions include surgery, systemic corticosteroids, interferon - α , laser, embolization, cryotherapy, and radiation.

Systemic corticosteroids, interferon- α , and embolization are used for large lesions⁶. The most frequent options of treatment for small lesions include the surgical approach, intralesional corticoid therapy, laser, cryo-therapy, and sclerotherapy⁷. Surgical removal is possible for small vascular lesions but is considered to be more invasive than sclerotherapy and presents the risk of haemorrhaging.

Sclerotherapy is the best complement to subsequent surgery. Sclerotherapy has been used in the management of hemangioma for more than 100 years. Different sclerosing agents have been used with varying degree of success such as sodium tetradecylsulphate, sodium morrhuate, sodium citrate, invert Sugar, boiling water, sodium psylliate⁸. Most of the vascular malformation regresses spontaneously without treatment. Sodium tetradecyl sulfate, an alkyl sulfate, has been described as a powerful almost ideal sclerosing agent and its administration is associated with minimal systemic and local reactions and sclerosing technique is contraindicated in cases of superimposed local infection or uncontrolled diabetes.

The treatment employed in the present case was sclerotherapy. However, before choosing the adequate type of treatment for a vascular malformation, a number of characteristics should

be considered, such as duration, size, location and number of tumours, patient age and the haemodynamics of the tumour. Moreover, the viability of the intended technique must also be assessed.

Mechanism of Action: It causes

- Localized inflammatory reaction.
- Obliterative thrombosis of hemangiomatous space.
- Subsequent fibrosis of the endothelial spaces.
- Regression of the lesion without affecting the bone.

Advantages of Sclerosing agent is that it is simple and inexpensive, no loss of blood, no hospitalization is required, sclerosing solutions are readily available and can be kept in office. Disadvantages are post-operative pain and burning sensation, anaphylactic reaction, tissue necrosis and sloughing [4%] and airway compromise [1%] Minkow *et al.*,⁹ (1979) used a technique of intralesional injection of 0.1- 0.5 ml of 3% STS in intraoral Haemangioma at the interval of 2 - 4 weeks. Satisfactory results were reported in all patients with minimum side effects and disappearance of the lesions without scarring. The number of injections varied according to the size of lesion. The interval between the injections was usually 2 - 4 weeks. It allows the indurations and inflammatory reaction to subside.

Khandpur S and Sharma VK¹⁰ (2009) performed study on sclerotherapy with 3% STS in 13 patients with venous malformations and micro cystic lymphatic malformation. They concluded that the lesions regressed by 90 - 100% in 11 cases after a mean of four injections, with no improvement in two cases. Complications included cutaneous blister formation, erosions, and crusting at injection site in seven cases and atrophic scarring in four patients.

Hemant¹¹ (2011) used 3% sodium tetradecyl sulfate in eight patients. Effective response rate of 62.5% and a mean of five injections at an interval of 3 weeks were required to produce the response. Three patients returned with recurrence within 2 years giving an ineffective or failure rate of 37.5% which found to be quite unacceptable considering the number of times patient has to undergo the

injection procedure and overall time duration of treatment needed to treat a moderate sized low flow vascular lesion. Out of five patients complications were included cutaneous blister and ulcer formation, at injection site in two cases and scarring in one patient. Erythema appeared on skin overlying the lesion and injection site in three patients which persisted for 2 - 3 days. No other significant systemic or local complications were encountered.

Nitesh Mohan¹² (2014) conducted the study, were included 15 cases in a period of 5 years. Haemangioma regressed in all cases with relief of symptoms. All patients complaining of bleeding, inflammation and pain were given analgesic and anti-inflammatory drugs. Complete remission was seen in 86.67% of the patients. 13.33% showed partial regression with ulceration and sloughing, which subsided with subsequent treatment. Tarun¹³ (2013) suggested that local reactions consisting of pain, urticaria or ulceration may occur at the site of injection. Systemic reactions include headache, nausea and vomiting. Allergic reactions such as hives, asthma, hay fever and anaphylactic shock have been reported.

Extreme care in needle placement and using the minimal effective volume at each injection site are important. Allergic reactions have been reported. Therefore, as a precaution against anaphylactic shock, it is recommended that 0.5ml of sodium tetra decyl sulfate be injected into a varicosity, followed by observation of the patient for several hours before administration of second or larger doses.

In case 2 Patient developed inflammatory reactions like painful swollen tongue with ulcers at the injection sites covered with necrotic slough, along with headache and fever. The local inflammatory reactions subsided immediately after injections of IM Dexamethasone (2mg) and chlorphenaramine maleate (25 mg) followed by oral intake of chlorphenaramine maleate tablet, 25mg for 3 days. These reactions on follow up were similar to the reactions described by Tarun (2013).

CONCLUSION: Vascular malformation can best treated with sclerosing agent with better patient compliance and therefore sclerotherapy proved to

be an effective and conservative technique for the treatment of benign vascular lesions. However we conclude that surgery is the therapy of choice in the large vascular lesions involving the tongue and sclerosing agent for the smaller lesions.

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