CASE REPORT

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Sclerotherapy for Oral Vascular Malformation Using Sodium Tetradecyl Sulfate: Case Series

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ABSTRACT

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Vascular malformations in the oral cavity are congenital vascular anomalies with potential functional and aesthetic implication. Over the years, the demand for effective and minimally invasive treatments for oral vascular malformations has increased. This study presents a case series of six patients diagnosed with oral vascular malformations, who underwent sclerotherapy using sodium tetradecyl sulfate. Treatment showed satisfying results without any recurrence.

Key words : Vascular Malformation, Vascular anomalies, Hemangioma, Sclerotherapy

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Ⅰ. Introduction

In recent years, the classification of tumors and tumorlike proliferations of vascular origin has been made[1]. In 1982, Mulliken and Glowacki introduced a classification for vascular anomalies, grouping them into tumors and malformations according to their endothelial characteristics[2]. Vascular malformation (VM) is a common congenital anomaly in the head and neck region; the prevalence is 1.2~1.5%[3].

According to the classification proposed by Finn and Mulliken, VM results from disturbances in specific stages of vascular development, characterized histologically by the absence of endothelial cell proliferation and possessing a normal cell replication cycle. These malformations manifest due to structural abnormalities[4]. The VM shows steady growth without regression, whereas the hemangioma tends to undergo involution during early childhood, resulting in inconspicuous scarring[2].

Various treatment modalities have been used for managing VM, including observation, systemic steroids/interferon-alpha therapy, embolization, compression therapy, surgical excision, electrocauterization, cryotherapy, sclerotherapy, radiotherapy, and selective angiographic embolization depending on the location, depth, and type of the lesion and the patient's conditions[5,6]. A minor and stable malformation might not necessitate intervention, while larger and concerning lesions could be managed through a blend of sclerotherapy and surgical excision[1].

Sclerosing agents include 5% sodium morrhuate, sodium psylliate, quinine urethrone, 5% ethnolamine oleate, 1% polidocanol, sodium tetradecyl sulfate, and hypertonic saline. When these components are exposed to vascular endothelium, cells, and serum, they become inactivated and are excreted from the body within 3 days after administration. After administration, these agents destroy the intracellular matrix between vascular endothelial cells and softening the endothelium. They expose collagen fibers and cause vascular spasm, leading to platelet aggregation and the release of platelet-related factors, resulting in blood coagulation, and inducing partial vascular obstruction[4,7].

This study presents a case series of five patients with oral VM who visited the Department of Oral and Maxillofacial Surgery at Kyungpook National University Dental Hospital (Daegu, Korea). The patients underwent sclerotherapy with a 1% sodium tetradecyl sulfate (Fibrovein, STD Pharmaceutical Products Ltd, Hereford, UK) (STS) injection, with one-time or repetitive injections as a conservative therapeutic strategy. We report the outcomes of lesion regression and satisfactory aesthetic results without lesion recurrence.

Ⅱ. Case series

Case 1

A 15-year-old female visited the hospital mainly because of a bulging dark blue lesion without pain in the right labial mucosa, extending into the vestibule

(Fig. 1A). CT angiography revealed a VM measuring approximately 2cm in size(Fig. 2). It was provisionally diagnosed with a VM, sclerotherapy was chosen as the treatment approach. 1% STS solution 1ml was directly injected into the lesion. Normal saline soaking gauze was used to apply pressure about 5 minutes after the injection. Two weeks after the initial injection, a follow-up examination showed significant regression of the vascular malformation, with a substantial portion of the lesion resolving(Fig. 1B). An additional 0.3 ml of 1% STS solution was injected into the remaining lesion areas. Two weeks after the second injection, most of the lesion resolved(Fig. 1C). Two months later, there were no signs of lesion recurrence, indicating a favorable outcome(Fig. 1D). The patient remained asymptomatic. The patient experienced some bleeding and pain and had a bruise for about a week. There were no systemic side effects or allergic reactions.

Case 2

A 21-year-old female visited the hospital mainly because of a painless bulging lesion on the left dorsal surface of her tongue(Fig. 3A). Magnetic resonance imaging (MRI) found 12x13x14mm-sized severe soft tissue hypertrophy on the left posterior surface of the tongue(Fig. 4A). Patient diagnosed with VM in the left tongue. 0.5 ml of 1% STS was directly injected into the lesion. Two weeks later, the color of the lesion reduced in size as it turned red. Slight pain occurred for several days after the injection. 0.7ml of 1% STS was injected into the lesion. A month later, a follow-up examination showed that the lesion had disappeared(Fig. 3B). There have been no recurrences so far.

Case 3

A 60-year-old female visited the hospital mainly because of a blue bulging lesion on the left lower labial mucosa(Fig. 5A). The patient had a history of diabetes, hypertension, and thyroid cancer resection. The patient had no discomfort or pain but wanted to be removed. On the physical examination, a non-pulsatile dark blue bulging lesion about 3cm was observed. The lesion was clinically diagnosed as a VM in the left labial mucosa. Initial sclerotherapy was applied with 0.5 ml of 1% STS solution. After two weeks, the size of the lesion decreased significantly (Fig. 5B). Secondary sclerotherapy was applied with the same doses. Two weeks later, the lesion was completely dissolved, and it was not observed with the naked eye(Fig. 5C). There have been no recurrence findings so far.

Case 4

A 36-year-old male visited the hospital with symptoms that the dark blue lesion at the left tongue tip seemed to have grown since a few weeks ago. He had experienced the lesion for over ten years(Fig. 6A). On the physical examination, a raised blue lesion of about 1cm on the left tongue tip was observed. There was no pain on palpation, and non-pulsatile. The lesion was clinically diagnosed with as

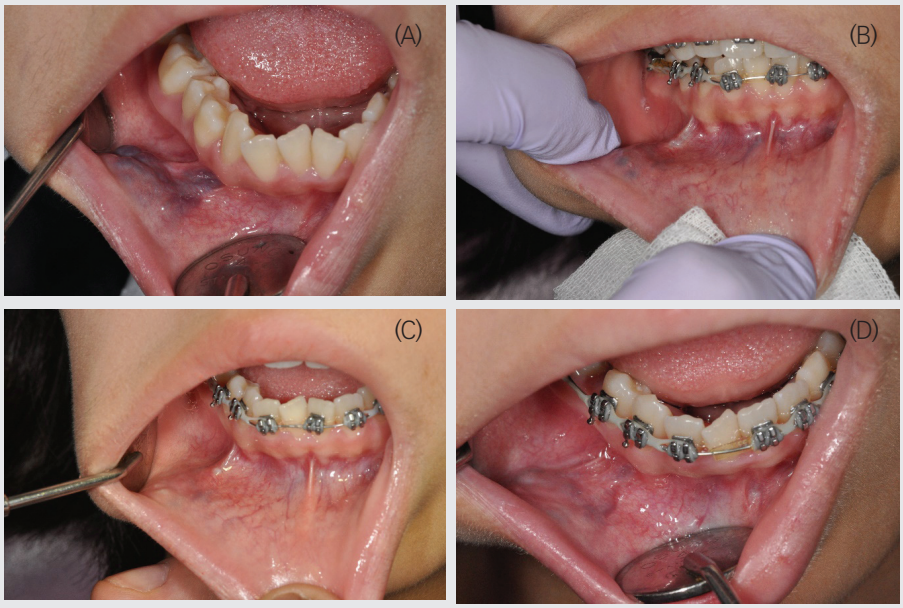

Figure. 1. (A) First visit (B) Two weeks after initial sclerotherapy (C) Two weeks after Secondary injection (D) Two months after third visit

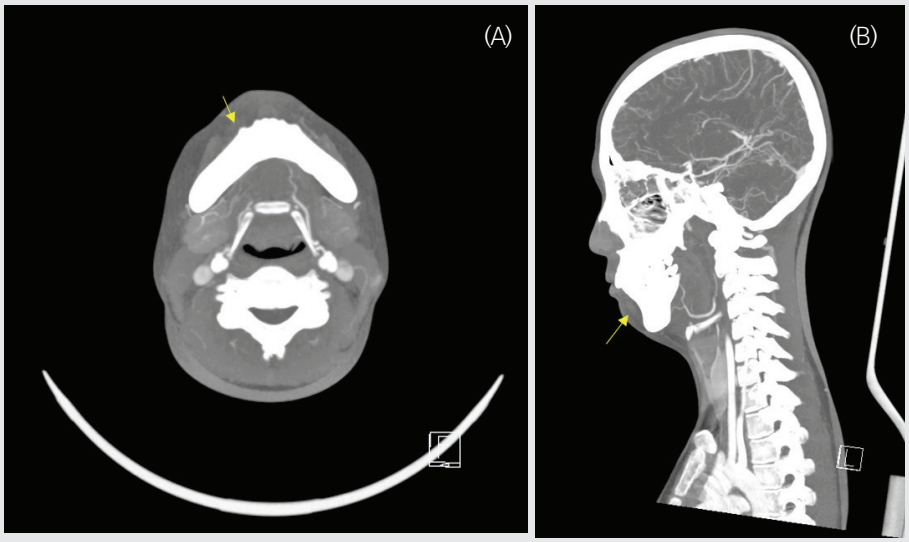

Figure. 2. CT angiograph demonstrates a vascular malformation with soft tissue swelling on right lower labial mucosa (arrow)

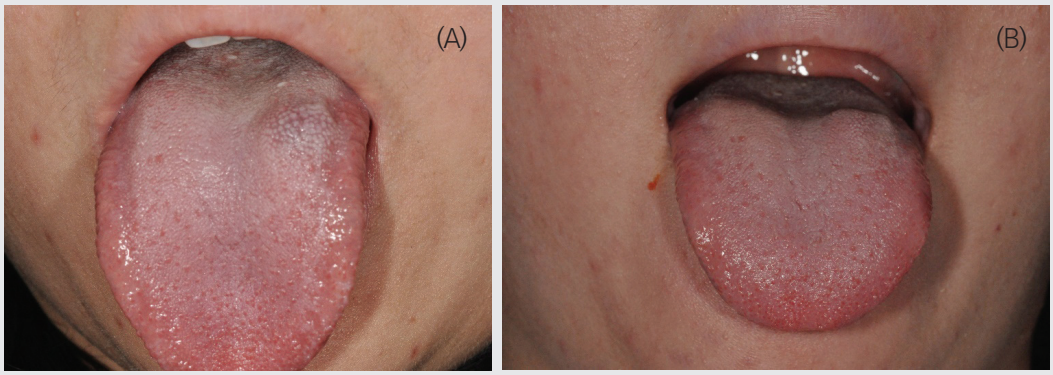

Figure. 3. (A) First visit (B) A month after initial sclerotherapy

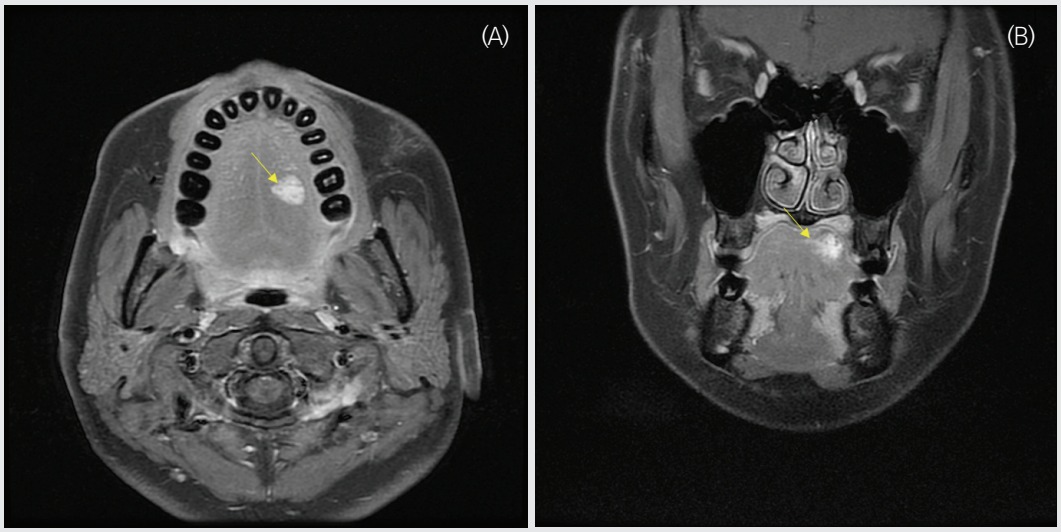

Figure. 4. Contrast-enhanced CT scan image demonstrates a vascular malformation on left tongue dorsal surface. (arrow)

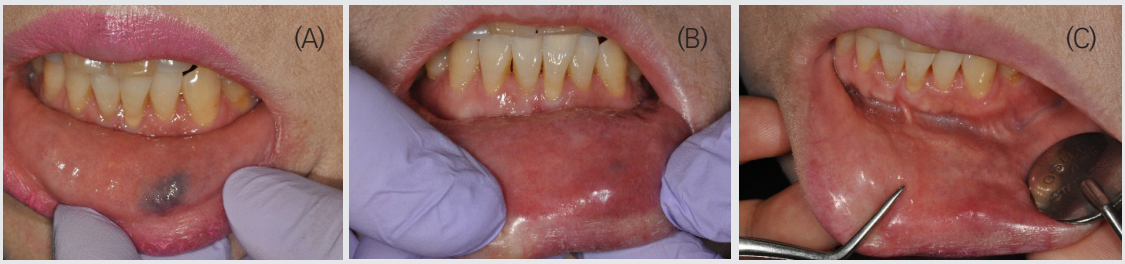

Figure. 5. (A) First visit (B) Two weeks after initial injection, residual lesion observed. (C) Two weeks after secondary injection, the lesion completely eliminated.

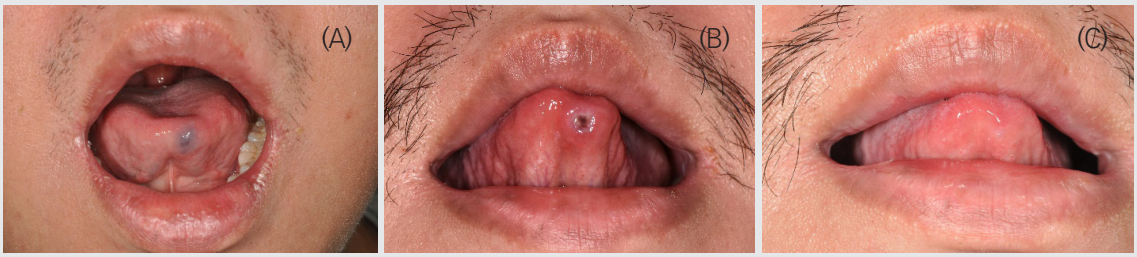

Figure. 6. (A) First visit (B) Two weeks after initial injection, showing scarring and ulcer. (C) Six weeks after initial injection, showing normal coloration of mucosa

VM. Initially, 0.5 ml of 1% STS solution was directly injected into the lesion. Two weeks later, the lesion almost dissolved, and small scar tissue remained(Fig. 6B). A month later, the lesion completely degenerated, and the site had regained normal color(Fig. 6C). The patient has maintained good conditions and has not experienced lesion recurrence.

Case 5

A 34-year-old female visited the hospital with a complaint of a bulging lesion at the posterior-lateral aspect of her tongue, which has been present for approximately 10years. Physical examination revealed an irregular, raised lesion measuring 1cm in size, showing a mixture of red and blue colors, located on the left side at the posterior aspect of the tongue(Fig. 7A). The patient has never experienced significant discomfort or pain related to the lesion. However, she expressed concern about its recent growth, which has been causing anxiety. Therefore, she sought medical attention with a desire

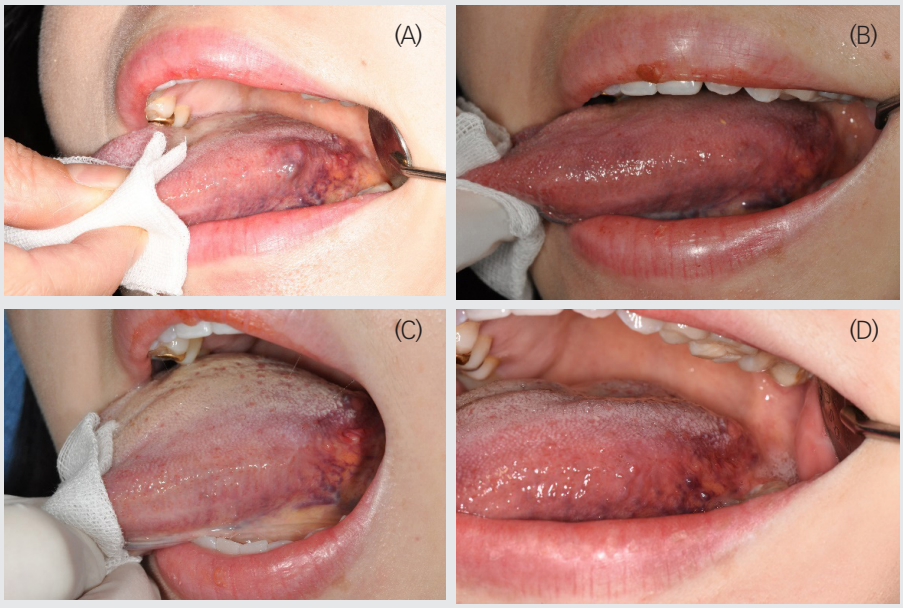

Figure. 7. (A) First visit (B) A month after first injection, small size of swelling excist (C) A month after secondary injection. (D) Threee months after sendary injection, bulging lesion completely removed.

for the removal of the lesion. Clinically diagnosed as a VM, the patient received an injection of 0.5ml of 1% STS solution directly into the lesion. After 5 weeks of follow-up observation, a reduction in the size of the lesion and normalization of its color were observed(Fig. 7B). Using the same volume and injection method, a second sclerotherapy was performed. One month later, complete disappearance of the lesion was confirmed(Fig. 7C). Subsequent follow-up at two months after complete disappearance showed no signs of lesion recurrence(Fig. 7D).

Case 6

An 81-year-old female patient presented to the hospital with a dark blue lesion in the upper lip tubercle area(Fig. 8A, B). Physical examination found a dark blue lesion of approximately 1cm in size, which showed blanching followed by recovery upon compression, while no pulse was detected. There is no pain or discomfort, but the patient described that the lesion was growing, concerned and expressed a desire for removal. The patient was diagnosed with a VM, and 0.5ml of 1% STS was directly injected into the lesion. At a follow-up observation after 2 weeks,

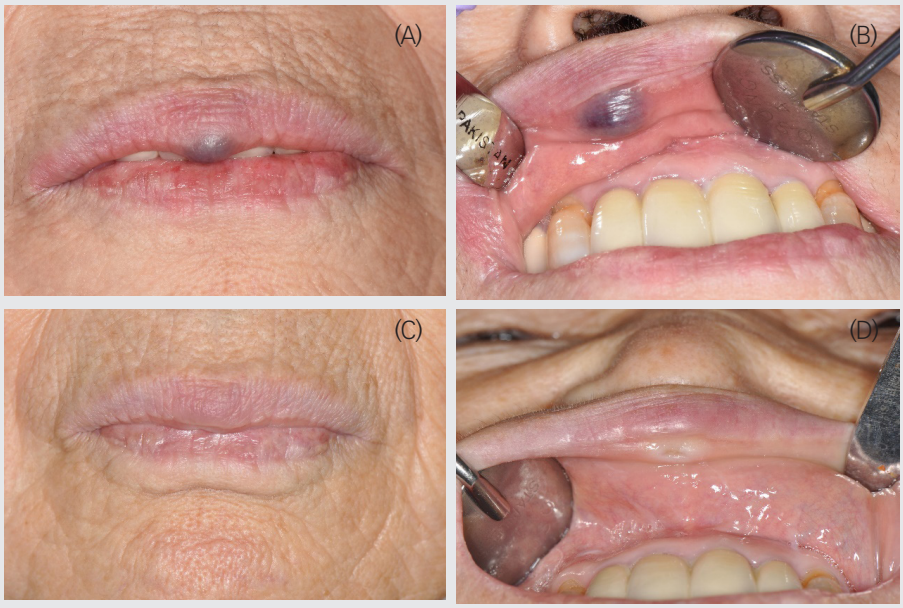

Figure. 8. (A,B) First visit (C,D) Two weeks after initial injection, the lesion completely eliminated, showing scarring and ulcer.

the lesion had disappeared, leaving a white scar with tenderness(Fig. 8C, D).

Ⅲ. Discussion

In this study, the VM were observed in individuals ranging from 15 to 81 years old, indicating a wide range of ages of occurrence. Notably, a greater occurrence of VM was found among female, which is consistent with findings from related studies[8].

Hemangiomas and VM are both benign vascular anomalies commonly found in the head and neck region. While they share similar clinical presentations when occurring in the oral cavity, there are distinct clinical and histological differences between them. Benign vascular anomalies can be classified based on the type of fluid they contain, leading to subcategories such as hemangiomas that contain blood and lymphangiomas that contain lymph fluid. Additionally, they can be differentiated based on the size of the vascular channels, with subtypes like capillary and cavernous malformations[9]. Mulliken and Glowacki introduced a biological classification that distinguishes vascular lesions with endothelial cell proliferation (e.g., hemangioma) from lesions

Table 1. Patients treated with 1% sodium tetradecyl sulfate for oral vascular malformation

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6
Sex/Age	F/15	F/21	F/60	M/36	F/34	F/81
Location	Labial mucosa	Tongue dorsal surface	Labial mucosa	Tongue tip	Tongue Lateral border	Upper lip
Dosage	1.0ml/0.3ml	0.5ml/0.7ml	0.5ml/0.5ml	0.5ml	0.5ml/0.5ml	0.5ml
Number of injection	2	2	2	1	2	1

with structural anomalies (VM) based on endothelial cell characteristics, physical findings, and natural history[2,9,10]. Hemangiomas exhibit growth through endothelial cell hyperplasia and should be distinguished from VM, which are not true neoplasms but rather localized defects of vascular morphogenesis resulting from dysfunction during embryogenesis and vasculogenesis[11].

VM can be categorized based on blood flow velocity - either slow (venous, lymphatic, and capillary) or rapid (arterial and arteriovenous)[12]. Venous VM, appearing as hypoechoic tubular structures on Doppler ultrasound, exhibit a gradual blood flow pattern. Accurate diagnosis proves crucial for the efficacy of sclerotherapy, as successful treatment relies on prolonged contact between the vascular wall and the sclerosing agent[13].

Diagnosis of these lesions is obtained through clinical and radiological evidence, as well as tissue biopsy. Particularly, for deeper-seated lesions, the initial diagnosis can be challenging. Therefore, in the case of such vascular lesions, in addition to computed tomography (CT), MRI, and Doppler imaging, the use of MR angiography, and the three-dimensional overlay of ultrasound images can be beneficial for diagnosis[14~16].

Various treatment methods have been used for benign vascular lesions manifested in the oral cavity. These methods include observation, cauterization, surgical excision, cryotherapy, systemic steroid administration, radiation therapy, laser therapy, and selective vascular embolization. In the present case, a direct sclerotherapy approach was adopted for the lesion site, which is considered a conservative and aesthetically pleasing method[17,18].

Vascular sclerotherapy is a treatment method in which a medication is injected into the blood vessels to create a thrombus, ultimately leading to fibrosis. Once the sclerosant is injected into the blood vessel, the vascular endothelium is damaged, causing the vessel to endosclerosis. The damage to the endothelium exposes collagen proteins beneath, leading to platelet aggregation and thrombus formation. If a significant amount of thrombus forms, endothelial cells may grow into the thrombus, resulting in recanalization, where blood flow is restored through the previously occluded vessel. Additionally, inflammation may occur within or around the blood ves-

sel. To prevent these occurrences, it is essential to thoroughly apply compression to the treated vascular area following vascular sclerotherapy[19].

Sclerotic agents can be broadly categorized into osmotic agents, detergents, and irritant/corrosive substances. STS belongs to the detergents among the sclerosants. Detergents act by disrupting cell membranes through a mechanism called protein theft denaturation, similar to how detergents are used in chemistry laboratories to extract proteins. Administering these agents leads to endothelial damage within few minutes of injection, and this damage can extend farther from the injection site[20].

In this cases, only 1% STS was used. Before the treatment, the oral mucosa was draped using a Chlorhexidine. An appropriate amount of STS was loaded into a 3cc syringe with a 26G needle. The location of the lesion was retracted by the assistant using a dental mirror, while the operator firmly held the oral mucosa and tongue to minimize the failure rate of vein puncture. During the injection, particular attention was paid to avoiding leakage of the medication outside the vein, and immediately after the injection, normal saline-soaked gauze was used for compression.

Ⅳ. Conclusion

STS is an effective treatment option for managing small-sized benign vascular lesions in the oral cavity. Its advantages lie in its convenience and the potential to avoid surgical risks. It should be noted, however, that STS is not a definitive treatment capable of completely eradicating the lesion. Instead, it exhibits its efficacy when dealing with smaller lesions and those with slow growth rates. Consequently, the appropriate selection and evaluation of patients are crucial prerequisites for utilizing STS effectively. Furthermore, a comprehensive understanding of its side effects is necessary to prevent potential complications.

Ⅴ. Conflicts of Interest

All authors declare that they have no conflicts of interest.

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