ORIGINAL REPORT

Alcohol sclerotherapy to treat vascular malformations in the oral cavity

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Arteriovenous malformations; Therapeutic embolization; Ethanol; Mouth

Abstract
Objective: To present our experience in treating vascular malformations in the oral cavity solely by injecting ethanol into the lesions.
Material and methods: We treated 26 patients (12 men and 14 women) with oral malformations. The diagnosis was based on clinical findings (n=26), magnetic resonance imaging studies (n=19), angiography findings (n=5), and direct puncture venography (n=2). To achieve sclerosis, we administered absolute ethanol through direct puncture. All interventions were performed under deep sedation.
Results: The vascular malformations treated ranged from 7 mm to 60 mm (median: 24.5 mm) in maximum diameter and had been present in the oral cavity for 0.2 to 54 years (mean: 13.6 years). The median age of the patients was 44.5 years (range: 12-87 years). The reason for treatment of the malformation was: an increase in size (n=8), local bleeding (n=11), risk of bleeding during dental extraction (n=5), pain (n=1), and esthetic purposes (n=3). Lesions were located in the mucosa of the cheek (n=12), in the facial gingiva (n=5), in the labial mucosa (n=6), in the tongue (n=3), in the pterygomandibular region (n=1), and in the palate (n=1). The median dose of ethanol was 3.2 mL. Twenty lesions disappeared after a single injection session, five after two sessions, two after three sessions, and one after five sessions. In 20 cases all signs of the lesions disappeared, in 6 a bluish macule persisted, and in 2 a mass effect persisted. The symptoms improved in all patients. Only transient complications of sclerotherapy were observed: local inflammation, perioral paresthesia in two patients, and necrosis of the mucosa of the cheek in one.
Conclusions: Alcohol sclerotherapy is an efficacious procedure for treating vascular malformations in the oral cavity.

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Introducción

La malformación vascular es un término que describe una variedad de condiciones que incluyen malformaciones arteriovenosas (MAV), malformaciones venosas (MV), malformaciones linfáticas (ML) y malformaciones capilares. Estas malformaciones pueden ser tanto para nutrición (venir), como para eliminación (volver). En el caso de la MAV, es una malformación vascular compuesta por vasos sanguíneos malformados que se conectan directamente entre la arteria y la vena, sin pasar por la red capilar normal. Las MAV pueden ser tanto intracranales como extracranales, y pueden presentarse de manera espontánea o como consecuencia de una lesión o accidente vascular cerebral.

Material y métodos

Se incluyeron 168 pacientes con MAV intracranales, de los cuales 120 fueron mujeres (71%) y 48 varones (29%). La edad promedio fue de 45 años, con un rango de 1 a 80 años. Todos los pacientes presentaron clínica de MAV, con la manifestación más frecuente siendo la hiperemia y la pulsación regional. Se realizó una evaluación clínica completa para determinar el compromiso neurológico y la función cerebral. Se realizaron estudios de imagen para confirmar la presencia de MAV, incluyendo resonancia magnética (RM) y angiografía por resonancia magnética (ARM).

Escleroterapia con etanol de las malformaciones vasculares de la cavidad oral

Resumen

Objetivo: Presentamos nuestra experiencia en la inyección intralésional de etanol como tratamiento único y efectivo de las malformaciones vasculares (MV) de la cavidad oral. Material y métodos: Se trataron 26 pacientes (12 varones y 14 mujeres) con malformaciones orales. El diagnóstico se estableció por los hallazgos clínicos (n = 26), los estudios de resonancia magnética (n = 19), de arteriografía (n = 5) y de flebografía percutánea por punción directa (n = 2). Para la esclerosis de las lesiones se empleó etanol absoluto mediante punción directa. Todas las intervenciones se realizaron con sedación profunda. Resultados: Se trataron 28 MV de diferentes tamaños, con una mediana de diámetro máximo de 24,5 mm (7–60), presentes en la cavidad oral durante una media de 13,6 años (0,2–54) en 26 pacientes. La mediana de edad fue de 44,5 años (12–87). Los criterios para el tratamiento de las malformaciones fueron: aumento de tamaño (n = 8), sangrado local (n = 11), riesgo de sangrado durante una extracción dental (n = 5), dolor (n = 1), trastorno estético (n = 3). Localización de las lesiones: 12 en la mucosa yugal, 5 en el cuello, 6 en la mucosa labial, 3 en la lengua, una en la región pterigomandibular y una en el paladar. La dosis mediana de etanol fue de 3,2 ml. Veinte lesiones desaparecieron tras una única inyección, 5 tras 2 sesiones, 2 tras 3 sesiones y una tras 5 sesiones. En 20 casos las lesiones desaparecieron, y persistió una mácula azulada y en 2 persistió un efecto de masa. Los síntomas mejoraron en todos los pacientes. Las complicaciones asociadas a la escleroterapia intralésional fueron pasajeras: inflamación local, parestesia perioral en 2 pacientes, y necrosis de la mucosa yugal en uno. Conclusiones: La esclerosis con etanol es un procedimiento eficaz para el tratamiento de pacientes con MV de la cavidad oral.

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Table 1  Main characteristics of patients and lesions.

<table>
<thead>
<tr>
<th>Lesions</th>
<th>Age</th>
<th>Sex</th>
<th>Location</th>
<th>Start* (age)</th>
<th>Evolution time (years)</th>
<th>Symptoms</th>
<th>Maximum diameter (mm)</th>
<th>Type of VM</th>
<th>Volume of ethanol (ml)</th>
<th>Number of sessions</th>
<th>Clinical result</th>
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<td>7</td>
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<td>30</td>
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<td>4</td>
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<tr>
<td>3</td>
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<td>Lip mucosa</td>
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<td>Cheek mucosa</td>
<td>A</td>
<td>6</td>
<td>Bleeding</td>
<td>11</td>
<td>HIVM</td>
<td>5 (2 + 3)</td>
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<td>D</td>
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<td>M</td>
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<tr>
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<td>F</td>
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<td>F</td>
<td>Vestibular gum</td>
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<td>15</td>
<td>Bleeding</td>
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<td>HIVM</td>
<td>3.2 (1.5 + 0.7 + 1)</td>
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<td>Risk of bleeding</td>
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<td>HAVM</td>
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<td>BM</td>
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<td>HIVM</td>
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<td>Enlargement</td>
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<td>HIVM</td>
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<td>M</td>
<td>Cheek mucosa</td>
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<td>Enlargement</td>
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<td>HIVM</td>
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<td>HIVM</td>
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</tbody>
</table>
Alcohol sclerotherapy to treat vascular malformations in the oral cavity

Results

A total of twenty-eight VM that had been presented for mean 13.6 years (range 0.2–54) in 26 patients, were treated (Table 1). The median age was 44.50 years (range 12–87).

Five subjects developed the malformation in childhood, 3 after puberty, and 18 in adulthood. In no case was the appearance of the lesions related to pregnancy, local injuries or operations.

Criteria for malformations treatment were: enlargement (n = 8), local bleeding (n = 11), risk of bleeding in case of tooth extraction (n = 5), pain (n = 1), esthetic problems (n = 3). The lesions presented the following locations: 12 in cheek mucosa (Figs. 1–3), 5 in vestibular gums, 6 in the lip mucosa, 3 in the tongue, 1 in the pterygomandibular region and 1 in the palate (Fig. 4). The median maximum diameter of the lesions was 24.5 mm (range: 7–60). At MRI, in three subjects the VMs had low T1 and T2 signal intensities, without hypertrophied arteries or turbulent shunts, which were classified as corresponding to HAVMs. In the remaining patients, MRI showed high signal intensity in T2 sequences, in relation to HVMs. In no case did the imaging studies reveal involvement of the facial bone structures.

The median ethanol dose used was 3.2 ml (range 0.4–11.5). In case of small lesions, a single injection proved effective for completing treatment, while larger malformations required puncture at several lesion sites. Technical success was achieved in all cases (100%). The lesions improved or disappeared after a single intervention in 20 patients (71.43%), mean size 20.7 mm; two sessions in 5 (17.86%), mean size 27.2 mm; three sessions in 2 (7.14%), mean size 27 mm; and after five sessions in 1 (3.57%). All patients improved objectively and subjectively. In 20 cases lesions disappeared completely, a bluish macula persisted in 6 and mass effect in 2. Symptoms relieved in all patients.

Complications associated with intralesional sclerotherapy were transient: local inflammation (controlled with symptomatic treatment) in 12 (42.86%), perioral paresthesia (that were decreasing spontaneously until its disappearance at the 12 months clinical control) in 2 (7.14%), and only 1 patient presented a deep ulcer (Fig. 5) that simply required

injected in each puncture. Technical success was defined as the possibility of embolizing the VM. The patients were monitored after the procedure and were then sent home after recovering from anesthesia. An outpatient prescription of steroids was made (with progressive lowered doses of prednisone [Dacortin™] over the subsequent days), together with omeprazole (20 mg/24 h), analgesics (1 g of paracetamol or 575 mg of magentic metamizol [Nolotil™]/6 h.) and local cold compression to reduce the swelling and pain. Antibiotics (500 mg of amoxicillin/clavulanate/8 h for 7 days) were prescribed in those cases in which oral mucosal necrosis was associated to overinfection.

Follow-up comprised clinical examination in all cases after 1, 2, 6 and 12 months to assess improvement. We decided to define the response to treatment according to the morphological changes of the lesion and the improvement of symptoms.

Data were analyzed using SPSS version 12.0 software (Chicago, IL, USA).
Figure 1  45-year-old female with a venous malformation of the cheek mucosa. Before treatment (A), T2W axial MRI showing a lesion with bright signal (black arrow, B), and the result after sclerotherapy (C). 1 month after the sclerotherapy session the lesion was completely missing (D).

Figure 2  61-year-old man with a small venous malformation in the left superior cheek mucosa. Before starting the treatment (A and B) and at 1 month after 1 session of ethanol sclerotherapy (D). STIR axial MRI showing a lesion with bright signal before (C) and after (E) the treatment.
antibiotic treatment for overinfection. Clinical control after 7 days confirmed complete mucosal restitution. At present, all the malformations have disappeared, and the patients are asymptomatic, with no relapses in the course of follow-up. The mean duration of follow-up was 132 days (range 30–513).

Discussion

VMs are congenital lesions that are always present from birth, and can remain stable or grow slowly in the course of life. Most are of a predominantly venous nature. The decision to treat oral VMs is taken not only when produce pain, functional problems, or esthetic disorders as dental separation, instability or early loss of some tooth, but also with the risk of recurrent or intense bleeding.

VMs treatment options include surgical resection, laser application, selective embolization of the perfusing arteries, and percutaneous sclerotherapy. However, there are few studies comparing the different techniques available today. The general trend is the use of sclerotherapy taking advantage of the existence of diverse effective material. Ethanol is probably the most widely used sclerosing agent, justifying its choice in this study. It causes protein denaturalization that induces endothelial damage— with thrombus formation, sclerosis and immediate vascular occlusion.

Ethanol sclerotherapy was used for 28 VM of different sizes with a success rate of 71.4%. We found excellent results, with symptoms relieved in all patients. This high efficacy is accord with published data of success rate of 60–95. It is difficult to accurately compare our results with alcohol with those of previous investigators because we studied an exclusively oral localization. In prior studies, lesions were diffuse, large complex, in peripheral localization or they had previously been treated. Johnson reported 11 VMs of the tongue treated with ethanol that resulted in resolution in 85.7%. Fan reported 8 cases of intraosseous mandibular VMs that were successfully embolized with absolute ethanol. No substantial difference in outcomes is evident between these investigations.

According with previous report, the cheek mucosa were the most affected site (12 lesions), followed by lip mucosa (6 lesions), vestibular gums, tongue, pterygomandibular region and palate (1 lesion). At our institution, patients with VMs undergo evaluation in a multidisciplinary team. As part of their diagnostic work-up, most patients undergo MR imaging to help determine the treatment and follow-up plans. As described, HAVMs were characterized by a lack of signal intensity in T2 sequences, while HIVMs showed high signal intensity in T2 sequences. While mandibular or upper

Figure 3  61-year-old woman with a venous malformation of the cheek, mucosa. A, B and C before treatment. Sagittal MRI STIR sequence shows a focal hyperintense (A). Direct puncture venogram after contrast medium injection into the main component of the lesion shows a trabeculated collection of venous spaces (B). Clinical lesion (C). At 17 months after 5 sclerotherapy sessions, practical disappearance of malformation (D) (this patient decided to stop the treatment).
maxillary involvement is not rare,

Different sclerosing agents have been used, including sodium tetradecyl sulfate, N-butylcyanoacrylate or poly-
docanol with or without CO2. In the mandibular region, Siu describe three cases of successfully treated VMs with cyanoacrylate. There are no known retrospective or

As previously stated, ethanol is probably the most widely

In our series, 15 patients (57.6%) experienced self-

for only a brief period. These characterizes the most com-

Severe pain associated with the

The absolute amount used per session should be <1 ml/kg. and other authors reported that the intensity and occurrence of complications does not correlate with the ethanol dose used, and the spontaneous reversion of hitches in most cases. In our study, absence of severe complications may be attributed to the extreme care with which alcohol was injected, the adequate position of the needle within the VM and the limitation to a maximum of 3 ml in each puncture. Our results show a trend to increase the number of sessions and mean volumes of ethanol injected with increasing lesion size. These data may prove useful in procedure planning.

Recurrence after treatment is common. However, although the duration of follow-up is still insufficient, we have not observed recurrence of the treated lesions, in coincidence with the observations of other authors.

Figure 4 76-year-old woman with a venous malformation at the soft palate (black arrow in A). Unenhanced T1W coronal MRI and contrast-enhanced fat-saturated T1W axial MRI showing a bright area with a phlebolith within the soft palate (white arrows in B and C). 10 days after the ethanol injection an ulceration in the mucosa appeared (D) and the patient expelled the phlebolith (E). After completing the treatment, the ulceration has been replaced by mucosal tissue (F).
Another treatment options like resection of malfor-
mation can result in deformity and it can never be viewed as 
a first treatment modality. Presurgical embolization, does 
not serve to reduce the extent of resection, and the risk 
of posterior progression cannot be predicted. Management 
should aim to reduce the size or eliminate the lesion entirely, 
taking care to preserve as much local mucosa as possible. 
Therefore, sclerosing treatment must be indicated.

Our report presents the limitations of an observational 
study and has no control group, which limits the validity 
of the results. However, our study suggests that consider-
ating their location and the potential complications associated 
with stomatological treatments, even asymptomatic VMs of 
the oral cavity should be subjected to sclerotherapy as the 
treatment of choice. The intralesional injection of ethanol 
is good thanks to its rapid action, the absence of recanal-
ization, and the tolerability profile, but although our initial 
experience points to ethanol as a great choice for the man-
gement of such malformations, larger studies are needed 
in order to draw definitive conclusions.

Authors contribution

1 Person responsible for the study’s integrity: ATF, SFM.
2 Conception of the study: ATF, SFM.
3 Design of the study: SFM, ATF.
4 Data acquisition: ATF, VMS, RPR, JMSB.
5 Data analysis and interpretation: ATF, SFM, AGCS.

6 Statistic treatment: ATF, AGCS.
7 Bibliographic search: ATF.
8 Writing of the paper: ATF.
9 Critical revision of the manuscript with intellectually relevant contributions: SFM, AGCS, VMS, RPR, JMSB, JVBS.
10 Approval of final version: ATF, SFM, AGCS, VMS, RPR, JMSB, JVBS.

Conflict of interests

The authors declare no conflict of interest.

References

1. Mulliken JB, Glowacki J. Hemangiomas and vascular malforma-
tions in infants and children: a classification based on endothe-
2. Burrows PE, Mason KP. Percutaneous treatment of low flow vas-
3. Legiehn GM, Heran MKS. Venous malformations: classification, 
development, diagnosis and interventional radiologic manage-
4. Rosenblatt M. Endovascular management of venous malforma-
5. Hyodoh H, Hori M, Akiba H, Tamakawa M, Hyodoh K, 
Hareyama M. Peripheral vascular malformations: imaging, 
treatment approaches, and therapeutic issues. Radiographics. 