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Percutaneous Ethanol Sclerotherapy of Venous Malformations of the Oral Cavity and the Oropharynx

ORIGINAL ARTICLE

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ABSTRACT

Objective: Percutaneous ethanol sclerotherapy has shown to be efficient in treating venous malformations of the head and neck. Our aim was to assess the safety and efficacy of percutaneous ethanol sclerotherapy in treating venous malformations of the oral cavity and the oropharynx.

Materials and Methods: From 2007 to 2015, 57 percutaneous procedures using ethanol were performed in 13 patients. Medical records of these patients were retrospectively analyzed. One patient was male and 12 were females. The patients' age ranged from 8 to 65 years (mean age, 30 years). The mean follow-up was 11 months. The volume of ethanol used per session ranged from 0.4 to 14 mL (mean volume, 6 mL) in 12 patients, except the syndromic patient.

Results: In 7 out of 13 patients (53.4%), the lesions were resolved completely. In 3 patients (23%), sclerotherapy alleviated the symptoms. In 2 patients (15.3%), the lesions did not sufficiently respond to the therapy. In 1 patient (7.6%), sclerotherapy failed due to misdiagnosis. No major complications were encountered. All the patients experienced pain to a tolerable degree. Swelling, induration, and darkening of the lesion occurred following injections. In 1 patient, ulceration and cleavage on the tongue was resolved within 15 days after emergence.

Conclusion: Percutaneous ethanol sclerotherapy is a reliable and efficacious method of treating venous malformations of the oral cavity and oropharynx.

Keywords: Venous malformation, oral cavity, percutaneous sclerotherapy, ethanol

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INTRODUCTION

Venous malformations (VMs) are the most common vascular anomaly (1). They are composed of dilated venous spaces with slow blood flow (2, 3). VMs are present at birth, but they usually manifest themselves by childhood or early adulthood. Their growth is proportional with the growth of the body, and they typically do not regress (4-6). Several factors, such as trauma and puberty, may enlarge them (2). While VMs may occur in any tissue or viscera, they are usually seen in the head and neck region (3, 7, 8). Particularly, involvement of the oral cavity and/or the oropharynx may cause some significant clinical problems and is challenging to treat (3). The treatment is indicated when they are symptomatic or cause unacceptable cosmetic disturbances and is aimed at eliminating or reducing the size of the lesion and relieving the symptoms (9). Various treatment modalities have been advocated, including percutaneous sclerotherapy (10-12). Percutaneous ethanol sclerotherapy has shown to be efficient for VMs of the head and neck region (3, 13). In this study, we describe our experience in treating VMs of the oral cavity and the oropharynx with percutaneous injections of absolute ethanol.

MATERIALS and METHODS

From January 2007 to December 2015, 66 percutaneous sclerotherapy procedures were performed in 17 patients with VMs of the oral cavity and/or the oropharynx. In 13 out of 17 patients, 57 procedures were performed with injections of ethanol. These 13 patients (12 females and 1 male), aged 8-65 years (mean age, 30 years), were included in this study, and their medical records were retrospectively analyzed. Six patients had a single lesion (3 patients with a single buccal lesion, 2 with a single palatine lesion, and 1 with a single lingual lesion). Four patients had 2 lesions (2 patients with 2 lingual lesions, 1 with 2 buccal lesions, and 1 with lingual and palatine lesions). One patient had 3 lesions (lingual) and 1 had multiple lesions located on the tongue, submandibular region, and vocal cords; 1 patient with Maffucci's Syndrome (MS) had several VMs originating from various parts of the oral cavity and the oropharynx (lingual, buccal, lingual base, soft palate, uvula, tonsil-

Table 1. Patient treatment summary

Patient no.	Location of the lesions	Number of the lesions	Number of the sessions	Number of procedures	Amount of injection (mL)		Outcome
					per treatment session	total	
1	Buccal	1	1	1	4	4	Failed*
2	Buccal	1	3	3	4/10/8	22	Good
3	Buccal	2	1	2	2	2	Poor
4	Buccal	1	1	1	3	3	Poor
5	Palate	1	1	1	3	3	Excellent
6	Tongue	2	1	2	7	7	Excellent
7	Multifocal#	2	2	3	9/3	12	Excellent
8	Tongue	3	2	4	14/3	17	Excellent
9	Palate	1	2	3	1/3	4	Excellent
10	Tongue	1	1	1	4	4	Excellent
11	Tongue	2	1	2	1.5	1.5	Excellent
12	Multicentric¶	Multiple	6	20	1.5/1/1.5/5/2/0.4¥	11.4	Good
13	Multicentric£	Multiple	2	14	32/20	52	Good

*Therapy failed due to misdiagnosis
#There were 2 lesions in total located on the tongue and the soft palate, respectively
¶The patient had multiple lesions located at various areas of the head and neck including the vocal cords
¥The amount of injected ethanol during the procedure performed under laryngoscopic guidance for the lesion of the vocal cords
£The patient with MS had multiple lesions located at various parts of the oral cavity and the oropharynx

lar pillars, and vallecula). Of the 2 patients who had multicentric lesions, the sclerotherapy procedures regarding the oral cavity and the oropharynx were included in this study. In addition, the procedure of the patient who had a VM on her vocal cords was also included. The most common symptoms were bleeding and difficulty with speech and chewing. The patient with MS had previously undergone a tracheatomy because of severe respiratory difficulties prior to her referral. None of the patients had received any previous therapy, except the patient with MS who had been given systemic steroid therapy. The diagnosis was based on clinical examination and confirmed by the reflux of blood in direct puncture venography. We attempted to define the initial extension of the lesion using magnetic resonance imaging (MRI) in 8 patients, and in 1 patient, contrast-enhanced computed tomography was performed for this purpose due to technical reasons. No imaging study was required in the remaining patients, because the lesions of these patients were obviously limited. At the beginning of the procedures of the isolated buccal lesions, Doppler ultrasonography was exceptionally performed so that a venous pouch convenient to puncture might be detected.

The lesions were punctured with 23-gauge butterfly needles under fluoroscopic guidance (Artis Zee, Siemens Healthcare, Nürnberg, Germany) without general anesthesia, except in case of the patient with MS and the patient with VM of the soft palate. Some of the procedures were performed with the patient under local anesthesia using midazolam and fentanyl when needed. The patient's mouth was tried to keep open using a retractor, and/or the tongue of the patient was held firmly using dry gauze and withdrawn to visualize the lesion better and to facilitate installation of the needles when necessary. A venography was then performed via the needle using water-soluble contrast media to define the morphology, volume, and draining vein of each lesion. The volume of the contrast media used to fill the lesion was noted and then the same amount of ethanol was slowly and carefully injected under fluoroscopic guidance. Required care was taken not to exceed the maximum recommended dose of ethanol (1 mL/kg of body weight) for each session of all patients (3, 11). Ethanol injection was paused when the patients reported feeling pain, and multipuncture injections were performed for larger lesions. When draining veins were detected in any venography, manual compression was performed to block the passage of ethanol into the venous system if possible. The injections continued until the lesion became thrombosed, which was determined by observing the lesion for hardening of its density and darkening of its color. Manual compression was applied on the lesions after injection to increase the contact surface of the ethanol and to stop bleeding if possible. The total volume of ethanol used per treatment session ranged from 0.4 to 14 mL (mean volume, 6 mL) in 12 patients without MS. In the patient with MS, the total injection amount was 52 mL: 32 mL in the first session and 20 mL in the second session. In the procedure of the patient who had a VM on her vocal cords, 0.4 mL of ethanol was injected under laryngoscopic guidance. All the patients received a single intravenous adminis-

tered dose of 8 mg dexamethasone phosphate at the end of the procedures.

All the patients, except the patient with MS, were discharged following the procedures. She was admitted overnight for observation. All the patients were observed on the tenth and thirtieth day following the procedures and were advised to refer whenever they encountered any problems with the illness later. The duration of referrals ranged from 1 month to 4.5 years (mean, 11 months). Follow-up observations were mainly done with physical examination. MRI studies were performed for the patients whose lesions were difficult to evaluate by sight. In case of incomplete resolution of the lesion or existence of another lesion, a new procedure was performed at the time of referral.

An assessment system was adopted to analyze outcomes of the treatments. The outcome of a treatment was defined as "Excellent" when the lesion completely resolved clinically and/or radiologically. The term "Good" was used to define alleviation of the symptoms without complete resolution of all the lesions. In the instances when an inefficient therapy resulted in relapse or a surgical intervention was required, the outcome was defined as "Poor".

Characteristics of the lesions and the procedural details are summarized in Table 1.

RESULTS

In 7 out of 13 patients (53.4%), sclerotherapy with the use of ethanol yielded an excellent result, and the lesions resolved completely. In 3 patients (23%), the therapy alleviated the symptoms. In 2 patients (15.3%), the lesions did not respond sufficiently to the therapy. Of these 2 patients, 1 underwent surgery for excision and the second referred with relapse after 4.5 years following the sclerotherapy. In 1 patient (7.6%), the lesion of the buccal mucosa was not resolved after the sclerotherapy; hence, the patient underwent surgery. She was found to have malignancy (acinar cell carcinoma) after the pathological examination following the operation (Table 1).

Most of the lesions that responded precisely to ethanol sclerotherapy were completely resolved 2 months after the procedures (Figure 1-3). The lesions decreased by more than 50% in volume in response to the 3 sclerotherapy procedures in 1 patient with buccal VM. Some of the lesions, particularly the polypoid lesions, underwent necrosis and detachment in 2 weeks, and the normal tissue was restored. The sclerotherapy in the patient with MS resulted in the resolution of symptoms, such as the breathing and swallowing difficulties, and then the tracheatomy was subsequently closed.

After the injection of ethanol, all patients experienced pain to a tolerable degree. Swelling, induration, and darkening of the lesions

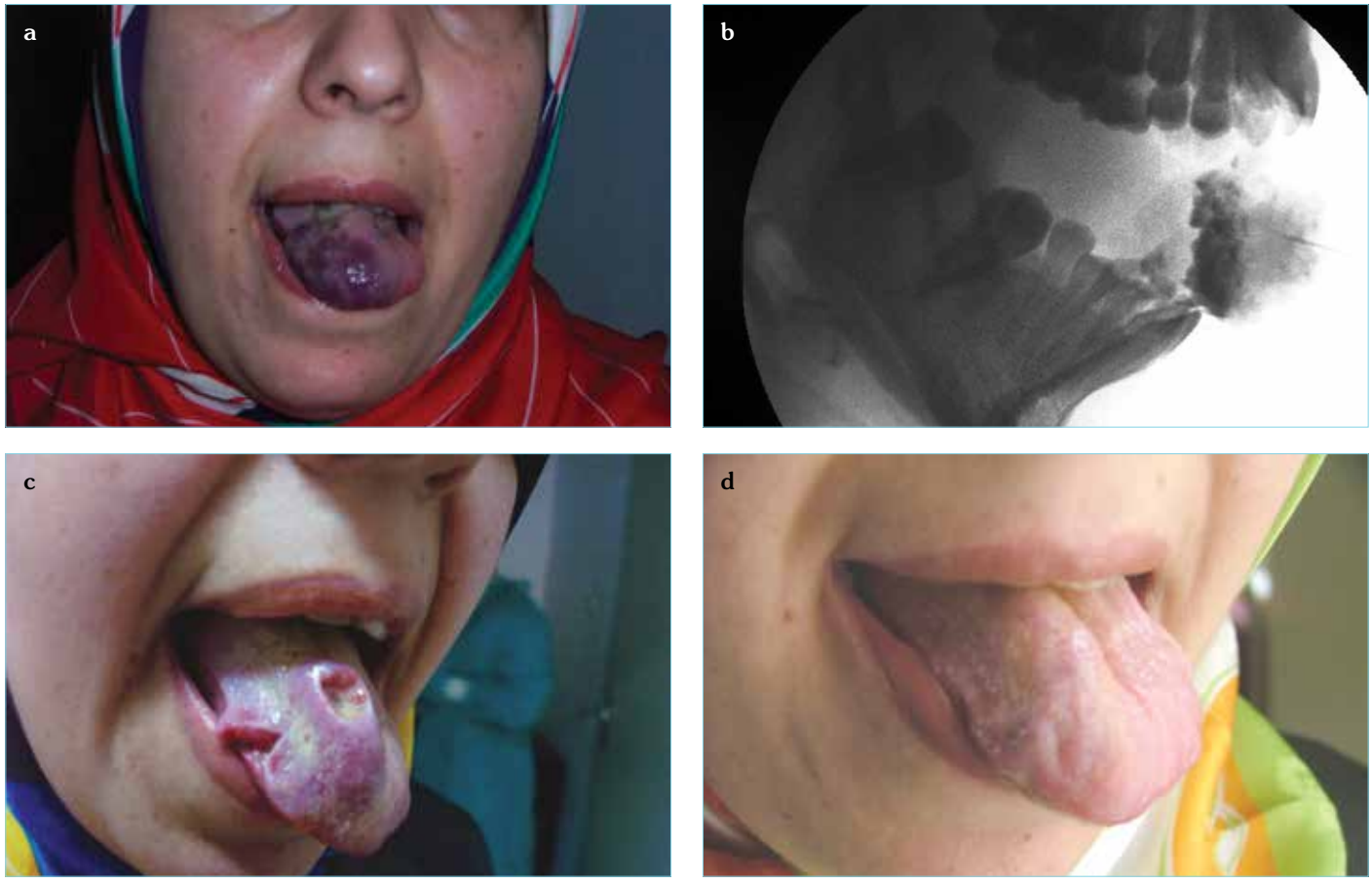

Figure 1. a-d. An image of a VM located at the right anterior part of the tongue (a), a direct puncture venogram shows opacification of the lesion (b), a view of the lesion obtained a few days after the ethanol injection shows subsequent ulceration and cleavage on the tongue (c), an image obtained 2 weeks after the injection demonstrates complete resolution of the lesion. Also, note the ulceration and the cleavage were perfectly recovered (d)

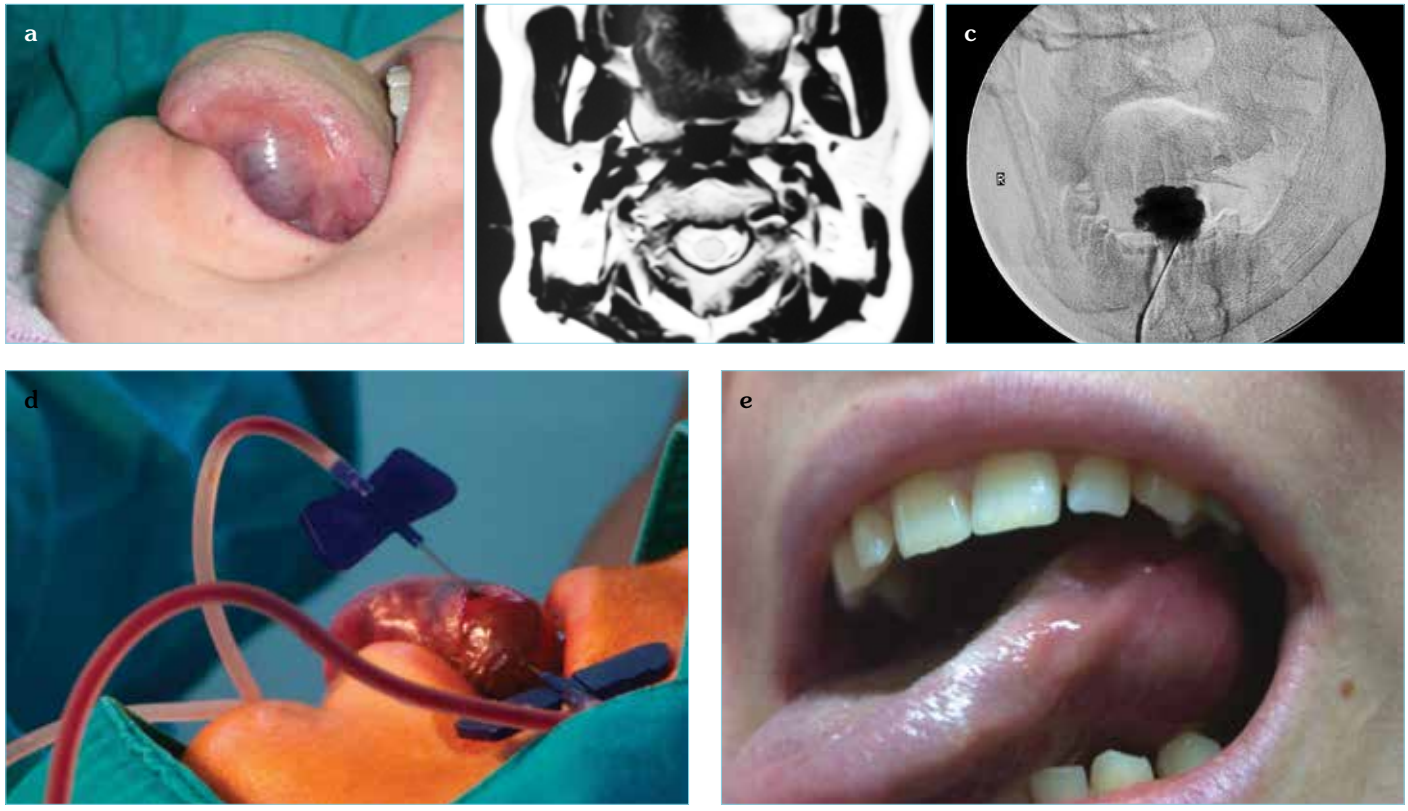

Figure 2. a-e. An image of a venous malformation located at the bottom left side of the tongue (a), an axial T2-weighted magnetic resonance image shows a hyperintense well-defined mass limited to the left side of the tongue (b), a direct puncture venography shows stagnation of contrast medium in the lesion (c), a view of the sclerotherapeutic session shows thrombosis of the lesion during double puncture injection. Note the change of the color of the lesion (d), complete resolution of the lesion is clearly seen (e)

occurred in all sessions, although no major complications were encountered (Figure 2). In a patient with lingual VM, ulceration and a cleavage developed in the area of the injection, and they were completely resolved within 15 days (Figure 1).

DISCUSSION

VMs are present at birth similar to other vascular malformations but sometimes may be latent. They grow proportionally with the growth of the body at a slow rate without regression (5). Several factors, such as trauma, failed surgical resection, alterations in the hormonal status (pregnancy, puberty, and use of steroids), thrombosis, or infection may cause expansion (2). VMs are composed of a network of thin-walled veins, deficient in smooth muscle, lined by a single endothelial layer, and dissect the host tissue (14). They are generally soft with a bluish-red discoloration and readily blanch with compression (2, 3). The diagnosis of VMs is essentially based on patient's history and clinical examination. Imaging, particularly MRI, is used to determine the initial extension of the lesions rather than differential diagnosis.

Venous malformations may arise in any part of the body, although they have a tendency to occur in the head and neck region, including the oral cavity and the oropharynx (3, 7, 8). The involvement may be localized or diffused. With regard to the location and extension, VMs of the oral cavity and the oropharynx may cause pain; bleeding; ulceration; and difficulty with swallowing, breathing, chewing, and speaking (3, 15). Indeed, VMs of these regions are a bothersome issue, which causes functional difficulties as well as cosmetic disturbances and needs to be treated carefully.

The ultimate goal of the treatment is to eliminate the anomaly and alleviate the symptoms. The main surgical challenge in treating these lesions is to excise the malformation completely without functional impairment and disfigurement due to anatomical difficulties. Percutaneous sclerotherapy has shown to be effective either alone or in conjunction with surgical interventions for treating VMs (3, 7, 11, 12). To treat VMs, various sclerosing agents including ethanol, bleomycin, polidocanol, sodium morrhuate, sodium tetradecyl sulfate, ethanolamine oleate, sodium diatrizoate tetrahydrate, hypertonic saline and OK432, alone or in combination have been used (11). Among these agents, ethanol is the most widely used, since it is cheap, easily available, and potent. Ethanol produces denudation of the endothelium, a severe inflammatory reaction, and thrombosis of the venous malformation (3, 16). Ethanol is reported to be the most reliable substance among all sclerosing agents and to have the lowest recurrence rate (11, 17). Unfortunately, percutaneous injection of ethanol is very painful and the extravasation of ethanol into the adjacent normal tissues may cause significant necrosis and nerve damage. Blistering and ulceration are not uncommon, but they resolve with conservative therapy. Besides these side effects, ethanol may induce systemic toxicity and acute pulmonary hypertension and cardiopulmonary collapse may rarely occur (2, 11).

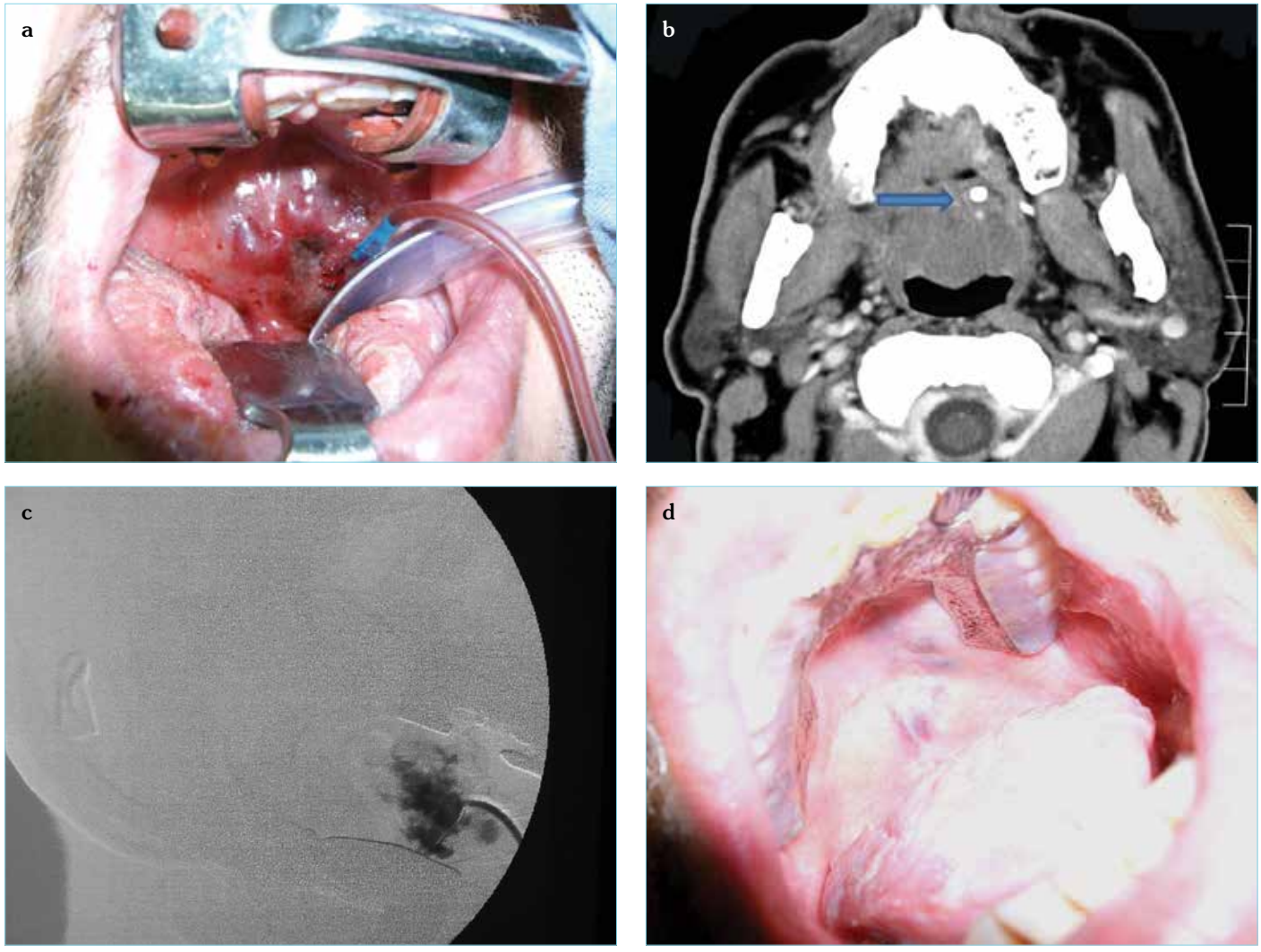

Figure 3. a-d. An image of a venous malformation located at the soft palate, obtained during the sclerotherapeutic procedure under general anesthesia shows the free return of blood through the connector attached to the needle that confirmed the diagnosis (a), a contrast-enhanced axial computerized tomography scan demonstrates subtle heterogenous enhancement of the lesion located at the soft palate with a left-sided phlebolitis typical of that of VM (arrow) (b), a direct puncture venogram shows opacification of the lesion (c), complete resolution of the lesion is clearly seen (d)

Our study shows that ethanol can be an effective and reliable sclerosing agent in treating VMs of the oral cavity and the oropharynx if used carefully. An awareness of the potential complications of the aforementioned procedure and a meticulous planning are essential for not encountering any unfavorable result. To improve the safety of a procedure, the specifications of the lesion including volume should be assessed by direct percutaneous contrast injection into the cavity before the ethanol injection and the maximum recommended aforementioned dosage should not be exceeded. In addition, ethanol must not be allowed to leak into the adjacent normal tissues and the venous system. Long-term follow-up ensures that any recurrence will be detected. Finally, in surgically compromised patients, as in the patient with MS in this study, percutaneous ethanol sclerotherapy can be chosen as a treatment modality.

This study has some limitations. The efficacy of ethanol was not compared with another sclerosant agent. In addition, the number of cases evaluated was slightly less.

CONCLUSION

Percutaneous sclerotherapy with ethanol is a reliable and efficacious method for the treatment of venous malformations of the oral cavity and the oropharynx and shows a low complication rate.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Ondokuz Mayıs University Faculty of Medicine (OMÜ KAEK 2017/381).

Informed Consent: Informed consent is not necessary due to the restorsrective nature of the study.

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Author Contributions: Conceived and designed the experiments or case: HA., FU. Performed the experiments or case: HA., FA., AİS. Analyzed the data: HA., FA., AİS. Wrote the paper: HA., FA., AİS. All authors have read and approved the final manuscript.

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REFERENCES

1. Greene AK. Vascular anomalies: current overview of the field. Clin Plast Surg 2011; 38(1): 1-5. [CrossRef]
2. Cox JA, Bartlett E, Lee EI. Vascular Malformations: A Review. Semin Plast Surg 2014; 28(2): 58-63. [CrossRef]
3. Johnson PL, Eckard DA, Brecheisen MA, Girod DA, Tsue TT. Percutaneous ethanol sclerotherapy of venous malformations of the tongue. AJNR Am J Neuroradiol 2002; 23(5): 779-82.
4. Donnelly LF, Adams DM, Bisset GS. Vascular malformations and hemangiomas: a practical approach in a multidisciplinary clinic. AJR Am J Roentgenol 2000; 174(3): 597-608. [CrossRef]
5. Hassanein AH, Mulliken JB, Fishman SJ, Alomari AI, Zurakowski D, Greene AK. Venous malformation: risk of progression during childhood and adolescence. Ann Plast Surg 2012; 68(2): 198-201. [CrossRef]
6. Greene AK, Liu AS, Mulliken JB, Chalache K, Fishman SJ. Vascular anomalies in 5,621 patients: guidelines for referral. J Pediatr Surg 2011; 46(9): 1784-9. [CrossRef]
7. Pappas DC Jr, Persky MS, Berenstein A. Evaluation and treatment of head and neck venous vascular malformations. Ear Nose Throat J 1998; 77(11): 914-22.
8. Chim H, Drolet B, Duffy K, Koshima I, Gosain AK. Vascular anomalies and lymphedema. Plast Reconstr Surg 2010; 126(2): 55e-69e. [CrossRef]
9. Enjolras O, Mulliken JB. The current management of vascular birthmarks. Pediatr Dermatol 1993; 10(4): 311-33. [CrossRef]
10. Kim KH, Sung MW, Roh JL, Han MH. Sclerotherapy for congenital lesions in the head and neck. Otolaryngol Head Neck Surg 2004; 131(3): 307-16. [CrossRef]
11. Lee CH, Chen SG. Direct percutaneous ethanol instillation for treatment of venous malformation in the face and neck. Br J Plast Surg 2005; 58(8): 1073-8. [CrossRef]
12. Lee BB, Do YS, Byun HS, Choo IW, Kim DI, Huh SH. Advanced management of venous malformation with ethanol sclerotherapy: mid-term results. J Vasc Surg 2003; 37(3): 533-8. [CrossRef]
13. Gelbert F, Enjolras O, Deffrenne, Aymard A, Mounayer C, Merland JJ. Percutaneous sclerotherapy for venous malformations of the lips: a retrospective study of 23 patients. Neuroradiology 2000; 42(9): 692-6. [CrossRef]
14. Wassef M, Blei F, Adams D, Alomari A, Baselga E, Berenstein A, et al. Vascular Anomalies Classification: Recommendations From the International Society for the Study of Vascular Anomalies. Pediatrics 2015; 136(1): e203-14. [CrossRef]
15. Lo Casto A, Salerno S, Cannizzaro F, Caronia A, Bencivinni F, Barbiera F, et al. MRI findings in lingual venous malformations. Dentomaxillofac Radiol 2003; 32(5): 333-6. [CrossRef]
16. Yakes WF, Luethke JM, Parker SH, Stavros AT, Rak KM, Hopper KD, et al. Ethanol embolization of vascular malformations. Radiographics 1990; 10(5): 787-96. [CrossRef]
17. Rautio R, Laranne J, Kahara V, Saarinen J, Keski-Nisula L. Long-term results and quality of life after endovascular treatment of venous malformations in the face and neck. Acta Radiol 2004; 45(7): 738-45. [CrossRef]