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Venous malformation at the upper lip treated with sclerotherapy – A case report

Shirou Tabe[a], Manabu Habu[a], Michinao Kawano[a,b], Osamu Sakaguchi[b], Izumi Yoshioka[b,*], Kazuhiro Tominaga[a]

[a] Division of Oral and Maxillofacial Surgery, Department of Science of Physical Functions, Kyushu Dental University, 2-6-1, Manazuru, Kokura-kita, Kitakyushu, Fukuoka, 803-8580 Japan
[b] Division of Oral Medicine, Department of Science of Physical Functions, Kyushu Dental University, 2-6-1, Manazuru, Kokura-kita, Kitakyushu, Fukuoka 803-8580, Japan

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ABSTRACT

Although vascular malformations of the head and neck present several challenges, sclerotherapy with ethanolamine oleate (EO) is effective. A 23-year-old woman presented to our institution with a mass under the mucosa and skin of the upper lip. A low-flow-type venous malformation was diagnosed based on findings from magnetic resonance imaging (MRI) and angiography. We injected 5% EO as the sclerosing agent. The superior labial artery was manually interrupted by application of pressure to prevent leakage of the 5% EO. The patient underwent a total of 3 sessions of sclerotherapy, separated by intervals of 5 months. We observed that the vascular malformations had decreased in size, with signs of scarring visible on MRI. In addition, lip asymmetry remained after sclerotherapy, and was restored with surgical recontouring. Pathological examination of the resected tissue indicated total scarring of the lesion. Postoperative cosmetic and functional outcomes were excellent, without recurrence of the venous malformation. Sclerotherapy with EO in which leakage of the agent was prevented with manual pressure is effective for treating venous malformations.

1. Introduction

Vascular malformations represent congenital angiodysplasia without cell proliferation and are classified based on the major vascular structures affected as capillary, lymphatic, venous, arterial and arteriovenous malformations, as well as various mixed types [1]. Recently, for the treatment of vascular malformations that develop in the surface layer of the face, the usefulness of conservative therapy has been reported from the perspective of esthetic preservation and recovery [2]. Sclerotherapy is a conservative management option, although a reduced effect and systemic influence may result from leakage of injected sclerosing agents such as ethanolamine oleate (EO). We report a case of venous malformation treated with sclerotherapy. Complications, which are related with EO leakage, were prevented by the application of manual pressure to interrupt blood flow through superior labial veins.

2. Case report

A 23-year-old woman presented with a dark-red-colored mass in the upper lip, which had been noted since she was 13 years old. On physical examination, the mucosa corresponding to the lesion and some portions of the vermilion border and skin above lip were colored dark red, suggesting a lesion infiltrating immediately beneath the skin and mucosa (Fig. 1A and B). Pseudofluctuation was palpable and discoloring and regression were easily visible. Furthermore, after release of applied pressure, the mass slowly returned to its original size.

Magnetic resonance imaging (MRI) showed a well-defined, multilocular, tumor-like structure measuring 20 × 23 × 10 mm in the area corresponding to the upper lip. The lesion showed intermediate to low signal intensity on T1-weighted imaging, and remarkably high signal intensity on fat-saturated T2-weighted imaging (Fig. 1C). On an angiographic image, although arteriovenous flow through the upper lip was present, no stained image corresponding to the lesion was confirmed. Feeding vessels were not specified. Based on these findings, the lesion was diagnosed as an extremely low-flow-type venous malformation.

The initial sclerotherapy was performed under sedation. Pressure was applied manually to induce regression of the lesion, to allow administration of local anesthesia in the surrounding area. Bilateral superior labial arteries were compressed manually to interrupt blood flow in order to maintain regression of the lesion. Five milliliters of 5%EO solution (Oldamine®; ASKA Pharmaceutical Co., Ltd., Tokyo, Japan)

* Corresponding author.
E-mail address: r13yoshioka@fa.kyu-dent.ac.jp (I. Yoshioka).

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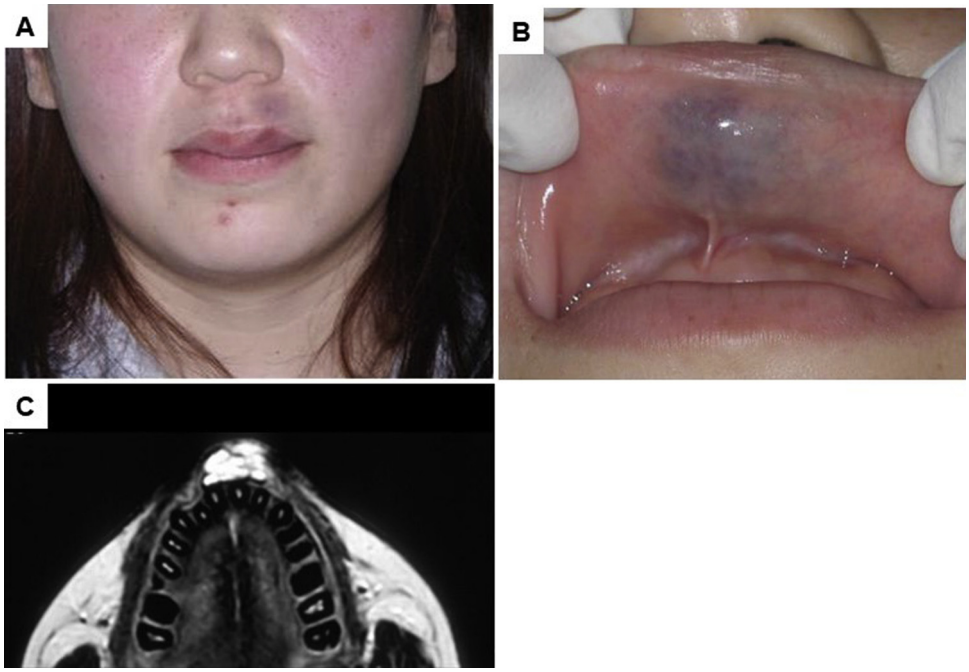

Fig. 1. Pretreatment findings.
A. Frontal view
A dark-red-colored lesion is seen on the left upper lip.
B. Intraoral view
A dark-red-colored lesion is seen paramedial on the upper lip.
C. MRI (fat-saturated T2-weighted imaging)
A well-defined multilocular structure is seen in the area corresponding to the upper lip.

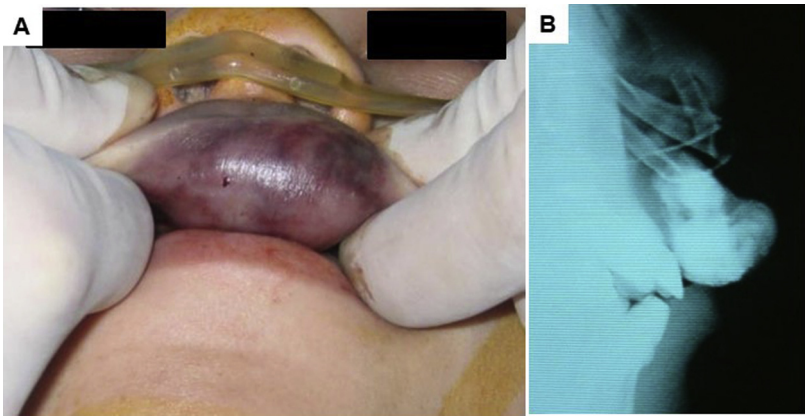

Fig. 2. Intraoral view and radiographic image during sclerotherapy.
A. Intraoral view
Upper lip is compressed manually.
B. Radiographic image immediately after 5% EO injection
EO is seen retained in the focus.

was injected into the lesion from the mucosal side of the upper lip to restore its original size (Fig. 2A). During injection, accumulation of sclerosing agent in the lesion was confirmed under radiographic guidance. We also applied manual pressure to bilateral superior labial arteries for about 5 min so that intraluminal thrombi were formed, according to the Zhi's report [3]. After radiographically confirming the absence of leakage from the vessels (Fig. 2B), the manual pressure was released. The procedure was completed by confirming the absence of bleeding from the puncture and recovery of blood flow in the skin. Immediately after the procedure, accumulation of 5% EO in the lesion was confirmed radiographically. Although there were marked swelling and ulceration in the upper lip after the injection of sclerosing agent, any systemic symptoms such as abnormal vital signs were not seen. Hematological examination also showed no abnormalities.

At 3 months after the initial sclerotherapy, scarring was observed in the vermilion border, while the lesion showed a reduction in size. However, a dark-red-colored area with fading reaction remained in the vermilion and white lip around the reduced lesion (Fig. 3A). On MRI, a signal hyperintense region remained on fat-saturated T2-weighted imaging of the lesion (Fig. 3B).

Five months after the first sclerotherapy, the second session of sclerotherapy was done by the same procedure. The volume of 5% EO was reduced to approximately 3 ml according to the size of the remaining lesion. Three months after the second sclerotherapy, efficacy

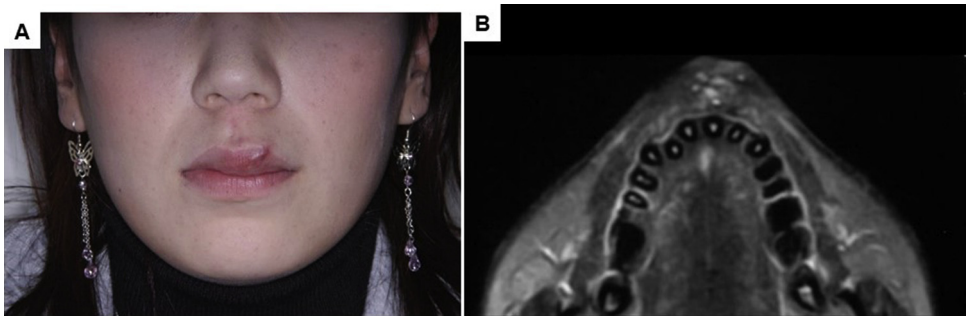

Fig. 3. Frontal view and MRI at 3 months after initial sclerotherapy.
A. Frontal view
Scarring is noted in the vermilion border.
B. MRI (fat-saturated T2-weighted imaging)
Signal hyperintense lesions with ill-defined borders are observed.

was again evaluated. Since the lesion had not completely resolved, a third session of sclerotherapy was performed 2 months later (10 months after the first session).

At 3 months after the third sclerotherapy (13 months after the initial session), the lesion was not evident on MRI and the area seemed to have become fully replaced by scar tissue. On MRI, the lesion showed a reduction of 65% in volume ratio after the first session, as compared to that before sclerotherapy, while the reduction was 85% after the second session and 97% after the third (Figs. 1C, 3 B, 4 A). Since scarring remained in the left upper lip and facial appearance was asymmetrical (Fig. 4B), labial plasty was performed under general anesthesia. Histopathological examination of the excised tissue revealed diffuse fibrosis with scattering small vessels in the subepithelial layer, resulting in disappearance of the vascular malformation (Fig. 5). The patient has since been followed, with no signs of recurrence (Fig. 4C, D).

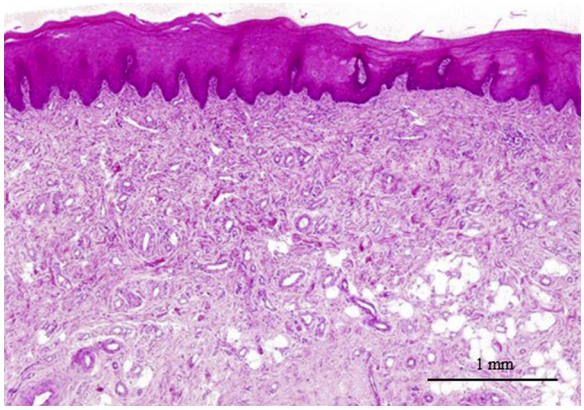

Fig. 5. Pathohistological findings of resected lesion (HE stain) (×40).

3. Discussion

Treatments for vascular malformation other than sclerotherapy include surgical resection, cryotherapy, embolization, laser vaporization, and intralesional photocoagulation [4–8]. In the present case, for postoperative esthetic and functional reasons, surgical resection and cryotherapy were considered inappropriate. Furthermore, embolization was not applicable, as the arterial feeding vessels were difficult to specify. Although laser vaporization is most useful for superficial lesions, lasers can only perforate 1–3 mm [9]. The present case is larger and thicker than 3 mm. However, Miyazaki et.al [6,5–8] reported intralesional photocoagulation technique (ILP) using a laser is effective for treatment of large submucosal vascular lesions. Furthermore, the use of ultrasound for guiding laser fiber insertion is promising and safety technique for less-invasive treatment of large vascular lesion [7]. Slow-flow vascular lesions in the oral mucosa are appropriate indications for an ultrasound-guided ILP with laser [7]. ILP is an effective

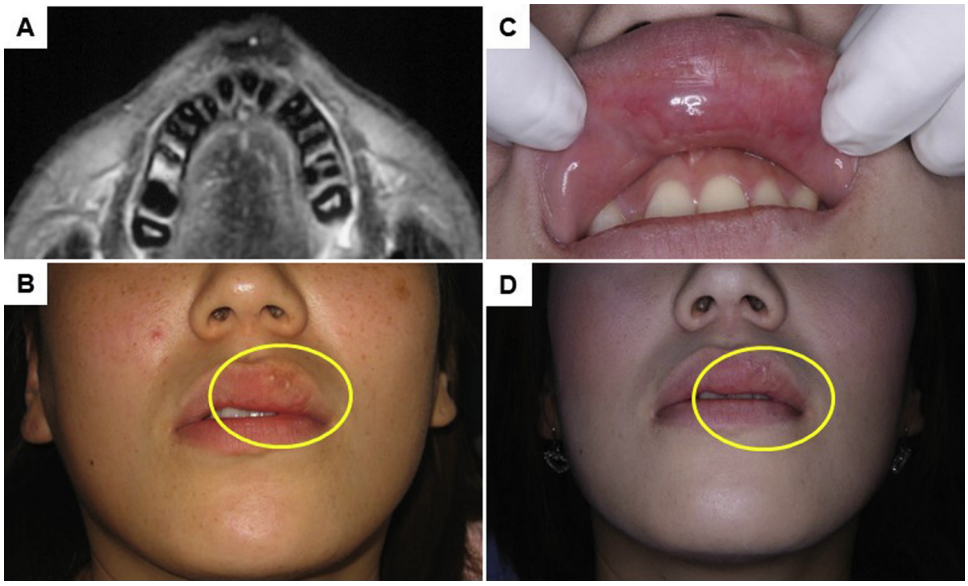

Fig. 4. MRI and Frontal view before upper lip plasty (20 months after initial sclerotherapy), Intra oral and Frontal view after upper lip plasty (10 months after third sclerotherapy).
A, B. Before surgery: MRI (fat-saturated T2-weighted imaging) and Frontal view
MRI shows more than 90% shrinkage of the lesion.
Lip asymmetry has remained in the left upper lip (inside the yellow circle) after sclerotherapy.
C, D. After surgery: Intra oral and Frontal view
Scarring has remained, but asymmetry (inside the yellow circle) has approximately been corrected.

treatment for large deep venous malformations. However, we could not detect a clear image of the lesion in the present case. Furthermore, multiple training is recommended to operate the probe and fibre simultaneously for holding the probe and fibre in each hand [7]. We have not had that experience with ultrasound-guided ILP for vascular lesion in oral cavity. Thus, we did not select ILP in the present case. Therefore, we chose sclerotherapy in the present case. However, ILP technique may have been applicable to this case.

Vascular sclerotherapy combined with embolization is recommended for high- and intermediate-flow-type lesions, while sclerotherapy alone is best for the low-flow type [5,10,11]. For determining the need for embolization, examination of blood passage through the vascular malformation over time by angiography is important to diagnose the flow type. In the present case, the patient had a low-flow-type vascular malformation, and sclerotherapy alone might have been adequate.

For sclerotherapy in the facial region, where blood flow is abundant, insufficient effect due to the leakage of sclerosing agents has been reported even in low-flow-type lesions [12,13]. In addition, renal disorder and gastric ulcer resulting from renal capillary damage are possibly caused by leakage [14]. Based on those findings, we considered that measures needed to be taken to minimize leakage of the sclerosing agent in the present case and applied manual pressure to the artery and vein of the upper lip concomitantly. Using a simple manual pressure technique, we were able to effectively interrupt blood flow and prevent leakage of the sclerosing agent.

EO causes thrombosis in the affected tissue by cellular damage of the vessel intima, followed by the deposition of fibrin and the accumulation of platelets and red blood cells on the injured endothelium, and hardens blood vessels [15]. Although use of EO results in less damage to normal tissue, multiple courses of treatment are frequently required. In the present case, we used EO because of the reduced damage to the skin, mucosa, and nerves as compared to ethanol.

When EO leakage into the systemic circulation is inevitable because of difficulty in interrupting blood flow by manual pressure, intravenous drip infusion of haptoglobin (4000 U) just before the administration of EO is necessary to prevent renal disorders [16,17]. Nishikawa et al. [18] recommended consideration of haptoglobin administration when the EO dose for a single session is $\geq$9.6 ml. In the present case, we considered that haptoglobin was not necessary because the dose of 5% EO was 5 ml and interruption of blood flow by manual pressure was effectively performed.

For quantitative evaluation of therapeutic efficacy, CT and MRI are frequently employed [19,20]. Kaji et.al [19] reported a reduction of approximately 60% over a mean of 2.6 sessions. In comparison to other reports on sclerotherapy with EO for head and neck lesions [19], the present case showed a better result in the reduction rate by fewer sessions of sclerotherapy. We considered that the application of manual pressure successfully helped EO stay in the lesion, thereby enhancing the efficacy of sclerotherapy.

Sclerotherapy with EO can necessitate multiple courses of treatment, depending on the size of the lesion. However, as lesion scarring can be reliably and safely reduced, this therapy can be used concomitantly with partial resection after lesion size reduction. In the present case, although asymmetry of the upper lip remained after sclerotherapy, esthetic improvement was obtained by surgical resection of the scar tissue.

4. Conclusion

In the present case, we obtained satisfactory outcomes from vascular sclerotherapy in which EO leakage was prevented by blood flow interruption with manual pressure.

Conflict of interest

The authors declare no conflicts of interest associated with this manuscript.

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