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MANAGEMENT OF ORAL HEMANGIOMA WITH SCLEROTIC OBLITERATION THROUGH MEDICATION THERAPY: A CASE SERIES

(Short title: Three cases of oral hemangioma sclerosis)

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Patient Consent

The case report was approved by the Ethics Committee of Paulista University (643/09), and the patients signed the consent form.

ABSTRACT

Hemangiomas or hamartomas are systemic proliferative vascular lesions that often occur in the oral cavity. The lesion usually presents a progressive growth, causing injuries and incontrollable bleeding. Its etiology is multifactorial, and it may occur at any age and there

is no gender predilection. Differential diagnosis can involve many different pathologies, including neoplasms. Patients complaints are often related to esthetics. The size, type, and degree of tissue involvement of the hemangioma dictates the need of a specific treatment. The aim of this clinical case series is to present multiple oral hemangioma scenarios managed with sclerotherapy through monoethanolamine oleate at 0.05 g/ml.The diagnosis, treatment, clinical procedures and risks of hemangiomas should be relevant to dental practitioners due to the high prevalence of this type of oral lesion.

Keywords: differential diagnosis, hamartoma, hemangioma, mouth, oleic acids, sclerosis

Background

Hemangiomas are proliferative malformations or benign vascular proliferative lesions characterized by proliferation of blood vessels (endothelial cells), which occur more often in the oral cavity, lips, tongue, buccal mucosa, and palate, however, it might develop in any part of the body. The etiology is linked to congenital anomalies, trauma, endocrine and inflammatory problems of unknown etiology.[1-4]It also can be named by hamartomas, therefore, it usually appears as developmental anomaly or congenital, appearing in most cases, at birth or early in life.[5,6]There is no predilection for gender or ethnicity. Its color ranges from deep red, blue and purple. It is usually flabby to palpation and can be circumscribed or diffuse, flat or raised, smooth or with a nodular surface. They are usually asymptomatic. It can vary in size from a few millimeters to centimeters, depending on several factors, including the patient's age, the location, and depth of the lesion, but does not appear in the form of polyp.[3,5,8,9]

The intraosseous form can often appear on both maxilla and mandible, also called central hemangioma, creating often difficulty in the differential diagnosis of other lesions.[9,10] Hemangiomas make differential diagnosis with mucous retention cysts, vascular malformations, pyogenic granuloma and malignant lesions, such as in leukemia and Kaposi's sarcoma.[11,12]To establish the correct diagnose, it is used the diascopy technic which compresses a glass slide on the lesion (glass pressing) and on the hemangioma´s cases, it will get pale, decreasing in size.[2,7,8] The association between aspiration and diascopy leads to a secure diagnose during clinical examination of the patient.[2]

The histopathology looks of the hemangioma occur by various small capillaries coated by a single layer of endothelial cells, supported by connective tissue.[5,11]It can be classified into capillary, juvenile, cavernous, and mixed arteriovenous.[1,5] The capillary

hemangioma is the most common type. The malformation venous vascular lesions correspond to the small lesions or capillary proliferation, less hemorrhagic and a little reddish.[1,6,13]The possibility of treatment depends on the size and extent of the injury and each case must be examined carefully. There are several possibilities of treatments considering each particular case, such as radiotherapy, electrocoagulation, laser application, cryotherapy, embolization, surgery, sclerotherapy added to the administration of interferon and steroids.[3,12]For instance, surgical removal by electrocautery has several advantages, such as decreased risk of bleeding, speed, professionals can easily do it and a single session is required.[2,4] Whereas other authors suggest that the use of high-power laser show satisfactory results.[14-16]However, the use of laser in the treatment of hemangiomas is controversial and it has been proven that it is more effective in superficial lesions, or in the treatment of telangiectasia or residual scarring lesions.[10]

Given the nature of the lesion, surgical excision of the hemangioma might be risky to the patient's life as may result in a hemorrhage, however, some features such as delimitation, resilience, easy access, no bone involvement and no major structures involvement, allows it to be done.[17,18]The intralesional systemic corticosteroids are intended to decrease the size of the lesion, to facilitate other adjunctive therapy approaches; however, complications associated with the use of these medications have limited their use. In the case of unsatisfactory responses or complications regarding the use of corticosteroids, recombinant interferon alpha has been used as a treatment to limit the growth of the lesion, but the neurotoxicity has reduced its application.[8,16]

Sclerotherapy often involves the intralesional injection of sclerosing agents such as sodium tetradecyl sulphate (Sotradecol®) and monoethanolamine oleate (Ethamolin®), which reduce or eliminate the lesion by sclerotic obliteration of vessels.[4,11,13,18,19]The aim of this manuscript is to report three cases of oral hemangiomas managed with medication therapy.

Clinical cases

Three clinical cases presenting oral hemangioma are reported below where therapeutic management of consisted in using Monoethanolamine Oleate 0.05 g/ml (ethamolin®; Farmoquímica, Rio de Janeiro, Brazil) as a sclerosing agent that prevents the blood flow, decreasing irrigation that feeds the vascular tumor. First, it was necessary to have the lesion area asepticby applying chlorhexidine gluconate 0.12 % with a disposable syringe and needle, as used in the application of insulin. Next, all patients were locally anesthetized with

prilocaine hydrochloride 3% (block). The needle was introduced around the lesion delicately to avoid bleeding, then the aspiration began and the drug was injected slowly aware of large liquid pressure in order to prevent an injury, embolism, ulceration, pain or severe tissue necrosis.

Each application consistedin a range from 0.5 to 1 ml of the agent because the amount is dependent on the size of the lesion. A weekly evaluation was pertinent to observe the evolution of the lesion and decide whether to repeat another dose or stop the treatment. Normally, in lesions up to 1 cm in diameter, two applications are enough for the total reversion of the hemangioma. All included patients in this case series signed the consent form following the principles of ethics in Human Research conducted inUniversity settings.

First case study

In the anamnesis, a 59 years old white skin female patientcomplained about a lesion on the tongue that feared it could bemalignant. The intraoral examination revealed a lesion on the left side of the dorsum of the tongue, with purplish color, measuring approximately 0.5 cm in diameter and lasting two years (Fig.1a). Diascopy revealed a vascular lesion method. The lesion receded afterthe eight days' evaluation *a posteriori* the single application of the sclerosing agent (Figs. 1. b,c).

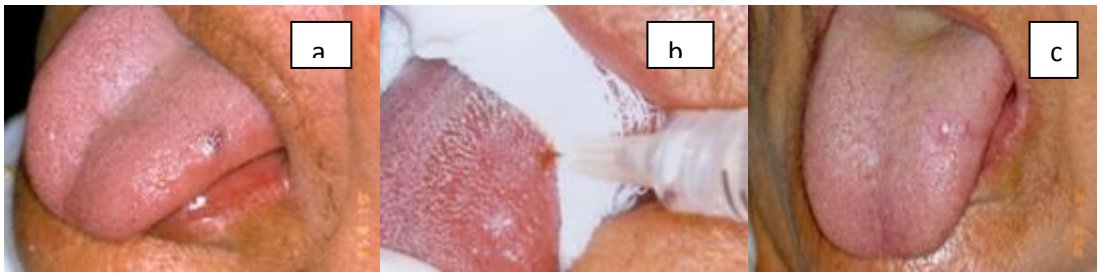

Fig. 1 - initial lesion of capillary hemangioma on the back of the tongue

Fig. 2 - application of monoethanolamine oleate - injectable

Fig. 3 - 8 days post treatment

Second case study

The chief complaint ofa 62 years old white skin female patient was: "I have a big bubble in the tongue that disturbs me to eat or speak". Later, he also complained that this bubble made his tongue look ugly and he reported being afraid of surgical treatment. The intraoral clinical examination revealed a lesion on the dorsum of the tongue measuring approximately 3 cm in diameter which has been present for approximately three years (Figs. 2a & 2b).

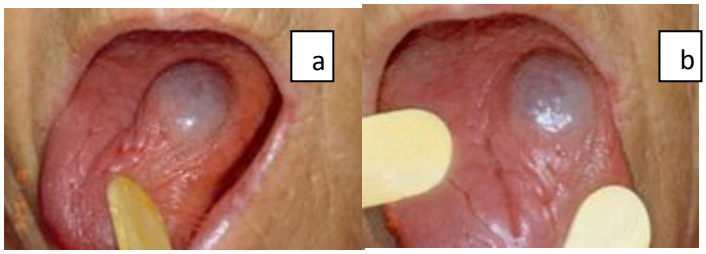

Fig.2 a A - initial lesion of capillary hemangioma on the back of the tongue

Fig. 2 B - initial lesion of capillary hemangioma on the back of the tongue

The diascopy evidenced vascular lesion because of the color change. The lesion was effectively eliminated after three applications of the sclerosing agent at a different moment each, and proper evaluation of the evolution of the treatment at each visit.Prior to the third application, it only remained a discrete erythema. Therefore, the third application was applied after two weeks from the second in order to avoid necrosis. Eight days after the third application of the medication, it was possible to notice the complete reversion of the lesion.

Third case study

During the anamnesis, a white skin 32-year-old male patient confessed beinga drug addict using needles for that purpose for several years, and in late 2006 began to present prostration, fever, cervical lymphadenopathy, night sweats, weight loss and severe headache. After medical examination and serology for HIV, syphilis, and hepatitis, positive serology for HIV was confirmed. He reported having a lip color change and volume increase for the last ten years which constantly depressed him due to the appearance. The CD4 count at time of HIV diagnosis was 146 cells/mm³ of blood and CV 73000 copies. He started antiretroviral (ARV) therapy (i.e. ARV-Combivir + efavirenz + B Complex). Three months later, the patient was diagnosed with panic disorder with severe psychotic symptoms, in addition to being diagnosed as schizophrenic. The patient evidenced a lack of commitment to ARV and later abandoned treatment. He remained over two years without using ARVs. The intraoral clinical examination detected multiple injuries in the lower lip, and two on the lower lip each measuring about 1 cm (Figs.3a & 3b). The diascopy test diagnosed a vascular lesion with color change (whitish), the injury came back to the old color when the glass was removed. After eight applications of sclerosing agent applied every two weeks, andweekly evaluations, the lesion practically disappeared.

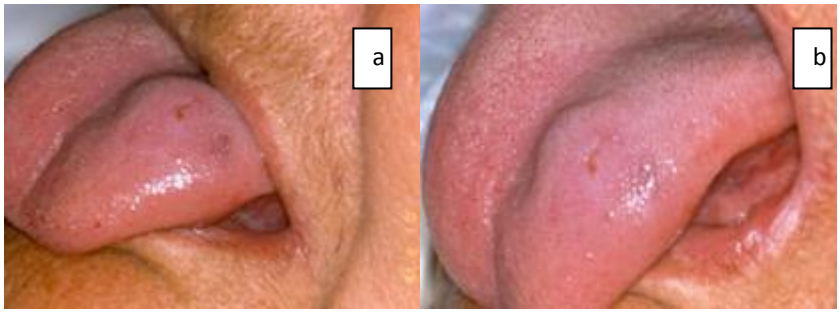

Fig. 3a and Fig 3 b. - clinical appearance of the lesion after 3 applications of monoethanolamine oleate - injectable - 1 application every 8 days

Discussion

Although hemangioma is a benign lesion, it can cause tissue compression, ulcers, bleeding, functional and aesthetic problems,[1-4,7,11] such as in those cases discussed in this case series.Esthetics are usually the main complaint and it depends on the degree of tissue involvement, the location and size of the hemangioma, it can cause facial asymmetry or interfere with the function of the involved organs. When located on the tongue or mouth floor usually interferes when talking, chewing and swallowing.[2,4,10,20]

The diagnosis is essentially clinical, and performing diascopy may cause ischemia injury, which usually occurs in vascular lesions.[3,5]The final diagnosis can only be achieved by histopathological examination.Some oral conditions do a clinical differential diagnosis of hemangiomas, including the retention cysts, mucoceles, Kaposi's sarcoma and pyogenic granuloma.[5,11,15] Due to the radiological characteristics, the intraosseous hemangioma can look similar to odontogenic cysts[5]andosteosarcoma.[15]Treatment of hemangioma depends on correct diagnosis, location, size, age of the patient, as well as the aesthetic and functional damage may be treated by several methods.[2-5,11,16,18-24]

This manuscript reported three patients with hemangiomas treated by sclerotical obliteration using monoetamolina oleate, promoting regression of the lesion by a non-surgical procedure and positive postoperative. The medication to penetration produces an extravascular inflammatory response by irritability of the venous or capillary endothelium, resulting in fibrosis and occlusion of blood vessels. The component of oleic acid can activate the release of factor Hagemman. The etamolina component can inhibit the formation of the fibrin clot by chelation of calcium. The combination of these two substances allows a hemostatic balance. Histologically, lesions treated with oleate monoetamolina have a replacement of the blood vessel by connective tissue as a result of drug-induced

inflammation. The advantages of this technique consist mainly in the elimination of surgical trauma, therefore, less risk of bleeding.[24]

The sclerosing agent should be applied carefully in the center and in the deepest portion of the lesion in order to avoidnecrosis by more superficial tissues.[2,18] The systemic dissemination may occur when the drug reaches arteries. Tissue necrosis can also be caused by the injection of a drug volume higher than recommended and can even lead to an anaphylactic reaction in sensitive patients to the drug.[24]

All patients usually report pain symptoms (burning) and there was slight edema in the perilesional region.[4,16,17] The pain symptoms are reported as transient and swelling disappears within one to three days. No tissue necrosis was observed in any of the cases presented and this is due to the following factors: early diagnose and proper indication of the technique, technical execution, application of the medication deep in the lesion, correct amount of medicine used in each session, in addition to slow and gradual drug injection.[18,24]

The use of anesthetic with vasoconstrictor agents ensures peripheral vasoconstriction, limiting the action of the drug and increasing its action time and reduces the pain symptoms. It is important to emphasize the need for anesthesia in the distance because the infiltrative local anesthetic close to the lesion could mask the injury.[24]

Conclusion

The three the clinical cases described in this article reported positive results from the medication therapy. This approach could be considered as an alternative therapy toenable sclerosis of lesions that cannot be treated by other methods due to the risk of bleeding or failures. Furthermore,the medication assessed herein is considered to be a low cost drug. The implementation of the technique was complemented with the use of local anesthetic with vasoconstriction distance which limited the action of the drug, increased the reaction time of the drug and prevented pain. Therefore, the postoperative was satisfactory and no complication was reported.

In summary, complete and satisfactory remission of lesions was achieved through medication therapy, leaving only in cases of larger volume (Figs. 2 and 3) the persistence of slight muscle laceration due to tissue proliferation of hemangiomas.

Competing interests

The authors have declared that they have no conflicts of interests.

Funding

None

Figure Legends

Figure 1: First clinical case. a) Initial appearance of the hemangioma on the dorsum of the tongue; b) Application of ethanolamine oleate 0.05 g / ml with syringe and needle for insulin around the lesion; c) Evaluation of site in 1a after eight days of treatment.

Figure 2: Second clinical case. a, b) Initial appearance of the hemangioma on the dorsum of the tongue; c, d) Clinical aspect of the lesion after three applications of the medication.

Figure 3: Third clinical case. a) Initial Clinical appearance of the lesions of the lower lip hemangioma; b) Clinical appearance of the lesions of hemangioma in track bottom and inside of the lower lip; c) Aspect of the lesion after eight applications of the medication in 15-day intervals.

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