Case report

Tackling Lip Hemangioma in Adults with Polidocanol Sclerotherapy: A Case Report and Review of Literature

ABSTRACT

Hemangiomas are the most common benign vascular tumors of the first decade of life and are rarely seen in adults. 60% to 70% of these tumors are found in the head and neck region, with oral hemangiomas being more uncommon. Hemangiomas are characterized by a proliferative growth phase that is followed by a very slow inevitable regression or involution phase. The literature reports a variety of therapeutic options for head and neck hemangiomas, such as medical management, cryotherapy, isotope radiation therapy, sclerotherapy (bleomycin, steroids), lasers, and surgery. The best course of action for every given patient is still the subject of debate. Sclerotherapy alone, or in combination has shown promising results. Polidocanol is a detergent-based solution that functions as a sclerosant by inducing localized inflammation and fibrosis. It is also an anesthetic and antipruritic agent. In this article, we describe a unique instance of a lip hemangioma in a geriatric patient and our experience with polidocanol sclerotherapy.

Keywords: sclerotherapy, hemangioma, polidocanol, intraoral, lip, geriatric.

INTRODUCTION

Hemangiomas are benign tumors that develop during infancy and childhood and are distinguished by three distinct phases: the phase of proliferation of the endothelial cells, the phase of rapid growth, and the spontaneous involution phase. Hemangioma prevalence ranges from roughly 2-3% in neonates, 10-12% in populations under 1-year-old, and 22–30% in infants weighing less than 1000g at delivery.[1] Hemangioma in adults is extremely uncommon and rarely reported. In comparison to their male counterparts, females exhibit a frequency three times higher. Hemangiomas of the oral cavity typically affect the gingiva, then the lip, tongue, and palate.[2] According to Akyol et al., the buccal mucosa (45.2%) is the most common place for intraoral

hemangiomas, followed by the tongue (35.5%), lip (9.7%), gingiva (6.5%), and palate (3.2%), in that order.[1]

The cause for the development of these tumours is unclear. The first of the two most suggested theories is that hemangiomas form from disturbed placental tissue, and the second most common explanation is that angiogenesis is improperly stimulated. The treatment options are as varied as the underlying cause and include wait-and-see measures, medication therapy, sclerotherapy (bleomycin, steroids), cryotherapy, isotope radiation therapy, laser therapy, and, last but not least, surgical therapy.[2-4]There are debates concerning the selection of therapy for each individual, as there are various options accessible, ranging from individual to combination therapy.[5]Surgical excision is the gold standard treatment technique for smaller confined lesions or peripheral hemangiomas. Hemangioma excision surgery in the lips and oral cavities can be very invasive and lead to uncontrollable bleeding wounds. As an alternative, sclerosing treatments, which attempt to destroy the blood vessels and cause fibrosis and hemangioma to vanish, may be utilized to treat these vascular tumors.[3]

We present a case of a lip hemangioma in a geriatric patient which is rare and distinctive in terms of age and location. The lesion was successfully managed by sclerotherapy with 2% polidocanol.

CASE REPORT

A 64-year-old female was referred to the Department of Oral and Maxillofacial Surgery, from the Department of Prosthodontics with a 3x1 cm, bluish-black raised lesion on the inner surface of the lower lip. The lesion was asymptomatic and posed an issue during the prossthetic rehabilitation with complete denture. A diagnosis of hemangioma was made after a thorough history, clinical examination and USG scan findings. A decision to utilize 2% Polidocanol sclerotherapy was made. Using a 30-guage needle 2% Polidocanol was injected circumferentially at the periphery of the lesion. The patient underwent 3 sessions of sclerotherapy with an interval of 2 weeks between each session. The procedure was handled well by the patient and was carried out as a daycare procedure. No complications were noted apart from mild post-operative swelling and ecchymosis of the lip and chin following the first session which resolved spontaneously. The patient was prescribed oral antibiotics and analgesics for 3 days following each session. The patient was followed up for 6 months after the last session. There was significant regression of the lesion with change in color and consistency. The patient was satisfied with the outcome and refused surgery for the

residual lesion. She was referred back to the Department of Prosthodontics where she was successfully rehabilitated with a complete denture.

DISCUSSION

Benign vascular tumors known as hemangiomas are most frequently observed in neonates and typically occur in the head and neck area of the body.[18]Hemangiomas are often categorized as mixed, cavernous, or capillary.[5]

Most hemangiomas manifest themselves in the first six weeks of life. Females are more likely than males to get these tumors, with a 3:1 ratio (F: M).[16] The head and neck area is the most frequently affected (60%) and is followed by the trunk (25%) and limbs (15%). In the head and neck they are located in the mouth, airway tract, and muscles. Intraorally they appear as deep blue lesions that are either elevated or flat.[1-4] About 60% of cases of cutaneous hemangiomas are superficial (capillary), and 15% are deep (cavernous).[5-6] Twenty percent of patients with hemangiomas have numerous tumors, but the majority of lesions are solitary. When a haemangioma first appears in a child, its future growth and involution mostly take place volumetrically.[6]

According to Harris MN et al., cutaneous capillary hemangiomas accounted for 86% of cases, whereas mucosal lesions only made up 12%. Mucosal lesions mostly affected the lips, gingiva, and tongue; the incidence was 2:1 more common in females than in males, and the lesions usually appearedin the fourth decade of life. Because mucosal lobular capillary hemangiomas were twice as common in female patients as in male patients, the study also suggested that hormones may have an impact on mucosal lesions.[6]

Hemangioma can be successfully diagnosed by computed tomography (CT), magnetic resonance imaging (MRI), and ultrasonography.[13] Among these, magnetic resonance imaging (MRI) is thought to be the most dependable imaging method for characterizing tissue and determining the extent of hemangiomas.[16]Not only does ultrasonography measure thickness, but it also measures vascularity by measuring resistance index.[14] Two-dimensional measures might not be adequate for hemangiomas since they frequently contain significant exophytic and deep components. Variations in the tumor's volume may also play a significant role in determining problems, depending on the location. With digital compression and diascopy, vascular abnormalities or suspected hemangiomas can be definitively diagnosed. The differential diagnosis of intra-osseous lesions can be more difficult because they resemble giant cell lesions or ameloblastomas on radiographs.[15]

The goals of treatment are to stop the haemangioma's proliferative growth, shrink its volume, and start the regression phase. Given the uncertainty surrounding the potential growth of a hemangioma—whether it will remain tiny and benign or develop enormously—early intervention may be the only viable solution.[13] A white or pink macule, a lesion that resembles a port-wine stain and first appears in youngsters, can be successfully treated with laser therapy, which will stop it from growing.[14]Observation should only be done for tumors that have not yet shown any signs of growth.[15]Massive hemangiomas, multiple tumors, life-threatening hemangiomas, and hemangiomas associated with comorbidities including ulceration, infection, and bleeding with dysfunction should all be treated with systemic medication therapy (steroids, interferon alpha-2a).[16]Treatment options for growing hemangiomas include laser therapy, sclerotherapy, combination therapy, and systematic medication therapy. It is possible to treat tiny, flat cutaneous hemangiomas and superficial telangiectasias with the argon laser (514 nm wavelength, 0.5 mm depth).[17] Certain patients with quickly growing hemangiomas may benefit from intratumoral injections of bleomycin or steroids in addition to topical imiquimod therapy. Surgery is reserved for lesions causing aesthetic and functional concerns and for large residuals for psycho-social reasons. These days, treating hemangiomas with cryotherapy or isotope radiation is uncommon because of the high risk of scarring, pigmentation loss, or depigmentation. Hemangiomas should be successfully treated on an individual basis, taking into account the tumor's size, location, and available therapies.[18]

In essence, sclerosing agents are irritants that harm endothelium surfaces and eventually cause the gap between them to erode. Three groups of sclerosing agents are distinguished based on the mechanism of action that results in endothelium damage.[16]

a. The *detergents* include polidocanol, also known as aethoxysklerol, sodium morrhuate, sodium psylliate, sodium tetradecyl sulfate, and ethanolamine oleate. By changing the surface tension around endothelial cells, the detergents inflict damage.[18]

b. *Osmotics*: hypertonic saline or dextrose solution. Their method of action involves dehydration-induced endothelial damage.[18]

c. *Chemical irritants*: These include substances that cause cauterization when applied and those that have a heavy metal effect on cells. These are poly-iodinated iodide and chromated glycerin.[18]

Sclerosants such as 0.05 g/mL of monoethanolamine oleate[7], 1% aethoxysklerol[8], and 5% ethanolamine oleate[9,10]have been used successfully alone or in combination with topical steroids.

Sold under the brands Asclera, Aethoxysklerol, and Varithena, polidocanol is a detergent solution that acts by causing a localized inflammatory reaction in the hemangiomatous space. This reaction starts through obliteration by thrombosis, which results in fibrosis of the endothelial cavities, leading to the regression of the lesion. Injections of polidocanol have been approved by the FDA to treat reticular veins (diameters of 1 to 3 mm) and minor varicose veins (less than 1 mm).[16] Varicose and spider veins can appear all over the lower leg when polidocanol, in the form of Varithena, is injected into the greater saphenous vein. Given that the leg's appearance may be permanently damaged, this surgery should be carried out carefully.[17-18]

Due to its anesthetic qualities, polidocanol sclerotherapy has the advantage of low or no discomfort, superior efficiency, safety when compared to other sclerosing agents, and the rare possibility of allergic reactions.[19] One thing that sets it apart from other agents is its affordability. Hyperpigmentation is less likely as the drug does not cause direct hemolysis. If complications do arise, they typically involve soft tissue loss, abscess formation, and necrosis of the surrounding tissue.[20]According to Minkow et al., a two-week gap between injections minimizes inflammatory responses and permits the surrounding tissues to heal from vascular injury.[18-20]

As the lesion did not entirely regress following the first session, we repeated the process twice with aninterval of two weeks. At each follow-up, there was a notable reduction in size, change in consistency from soft to firm and change in color from bluish-purple to pink.

CONCLUSION

It is unusual to find an intraoral lip hemangioma in the elderly population. While most hemangiomas regress on their own during infancy and childhood, therapy is necessary for a small number of cases that do not undergo involution. Sclerotherapy is a reliable, successful, affordable, and straightforward treatment for hemangiomas, making it a more prudent option than surgery which is more invasive. Polidocanol sclerotherapy is economical, has minimal or no side effects and can be performed on an out-patient basis.

Disclaimer (Artificial intelligence)

Option 1:

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc.) and text-to-image generators have been used during the writing or editing of this manuscript.

Option 2:

Author(s) hereby declare that generative AI technologies such as Large Language Models, etc. have been used during the writing or editing of manuscripts. This explanation will include the name, version, model, and source of the generative AI technology and as well as all input prompts provided to the generative AI technology

Details of the AI usage are given below:

1.

2.

3.

REFERENCES

1. Akyol MU, Yalçiner EG, Doğan AI. Pyogenic granuloma (lobular capillary hemangioma) of the tongue. International journal of pediatric otorhinolaryngology. 2001 May 11;58(3):239-41.
2. Cardoso CL, Fernandes LM, Rocha JF, Gonçales ES, Ferreira Júnior O, Taveira LA. Surgical approach of intraoral hemangioma. Odontologia Clínico-Científica (Online). 2010 Jun;9(2):177-80.
3. Gao Z, Zhang Y, Li W, Shi C. Effectiveness and safety of polidocanol for the treatment of hemangiomas and vascular malformations: A meta□analysis. Dermatologic Therapy. 2018 Jan;31(1):e12568.
4. Abidullah M, Hussain SM, Karpe T, Gaddikeri K. Cavernous hemangioma of buccal mucosa: a rare case report with a reappraisal of differential diagnosis & review of literature. British Biomedical Bulletin. 2013.
5. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. Plastic and reconstructive surgery. 1982 Mar 1;69(3):412-20.
6. Harris MN, Desai R, Chuang TY, Hood AF, Mirowski GW. Lobular capillary hemangiomas: An epidemiologic report, with emphasis on cutaneous lesions. Journal of the American Academy of Dermatology. 2000 Jun 1;42(6):1012-6.
7. Queiroz SI, Assis GM, Silvestre VD, Germano AR, Silva JS. Treatment of oral hemangioma with sclerotherapy: case report. Jornal Vascular Brasileiro. 2014 Jul;13:249-53.
8. Ademi Abdyli R, Abdyli Y, Perjuci F, Gashi A, Agani Z, Ahmedi J. Sclerotherapy of intraoral superficial hemangioma. Case Reports in Dentistry. 2016 Nov 23;2016.

9. OLIVEIRA MM, SILVA BA. Escleroterapia como tratamento de hemangioma lingual: relato de caso clínico. RGO-Revista Gaúcha de Odontologia. 2020 Jun 17;68.
10. Johann AC, Aguiar MC, do Carmo MA, Gomez RS, Castro WH, Mesquita RA. Sclerotherapy of benign oral vascular lesion with ethanolamine oleate: an open clinical trial with 30 lesions. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2005 Nov 1;100(5):579-84.
11. Agarwal S. Treatment of oral hemangioma with 3% sodium tetradecyl sulfate: study of 20 cases. Indian Journal of Otolaryngology and Head & Neck Surgery. 2012 Sep;64:205-7.
12. Minkow B, Laufer D, Gutman D. Treatment of oral hemangiomas with local sclerosing agents. International journal of oral surgery. 1979 Feb 1;8(1):18-21.
13. Bhargava DK, Dwivedi M, Acharya SK, Sundaram KR. Effect of low dosage of polidocanol in treatment of esophageal varices in cirrhotic patients. The Indian Journal of Medical Research. 1988 Dec 1;88:515-21.
14. Yonetsu K, Nakayama E, Yuasa K, Kanda S, Ozeki S, Shinohara M. Imaging findings of some buccomasseteric masses. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 1998 Dec 1;86(6):755-9.
15. Açikgözsurname A, Sakallioglu U, Özdamar S, Uysal A. Rare benign tumours of oral cavity–capillary haemangioma of palatal mucosa: a case report. International Journal of Paediatric Dentistry. 2000 Jun;10(2):161-5.
16. Witman PM, Wagner AM, Scherer K, Waner M, Frieden IJ. Complications following pulsed dye laser treatment of superficial hemangiomas. Lasers in Surgery and Medicine: The Official Journal of the American Society for Laser Medicine and Surgery. 2006 Feb;38(2):116-23.
17. Dubois J, Garel L, Abela A, Laberge L, Yazbeck S. Lymphangiomas in children: percutaneous sclerotherapy with an alcoholic solution of zein. Radiology. 1997 Sep;204(3):651-4.
18. HANGE V. Polidocanol sclerotherapy for intraoral hemangioma of buccal mucosa: a rare case report. Mucosa. 2020 Dec 12;3(4):104-9.
19. Bonet Coloma C, Mínguez Martínez I, Palma Carrió C, Galán Gil S, Peñarrocha Diago M, Mínguez Sanz JM. Clinical characteristics, treatment and outcome of 28 oral haemangiomas in paediatric patients.
20. Singh R, Agarwal SP, Sinha ON, Gaur S. A Case Report-Inj Polidocanol in Haemangioma of Tongue. Sch J Appl Med Sci. 2016;4:2433-5.

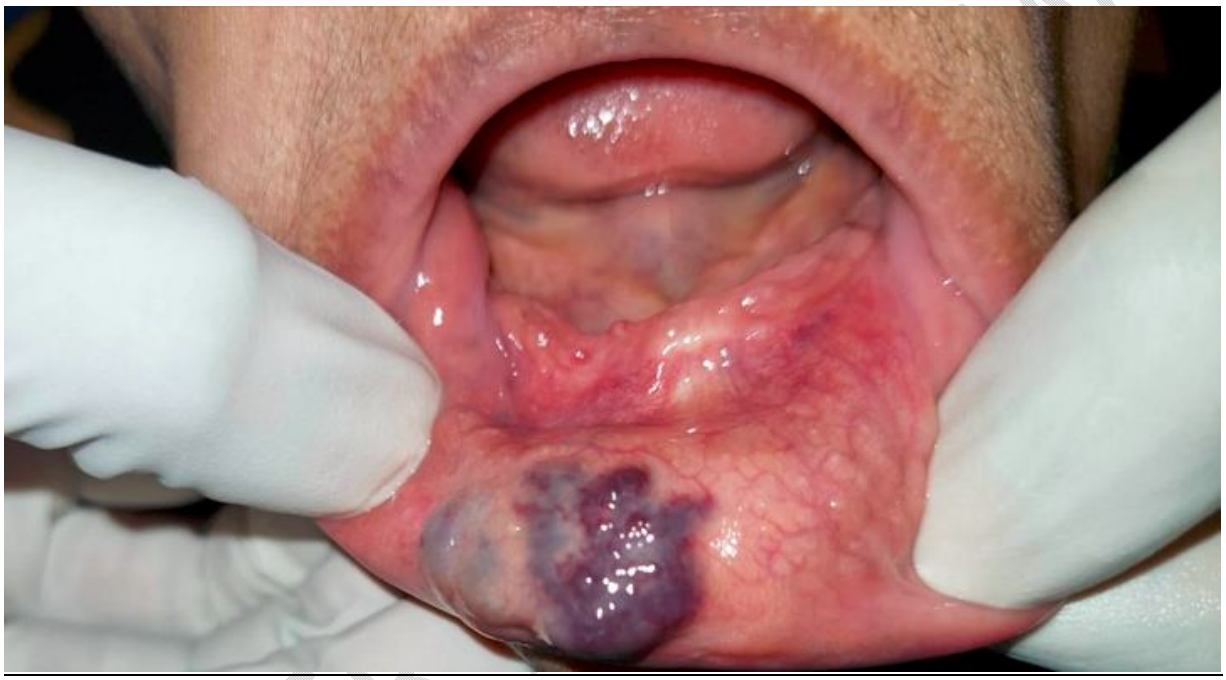

IMAGE 1: A pre-operative image describing the lesion.

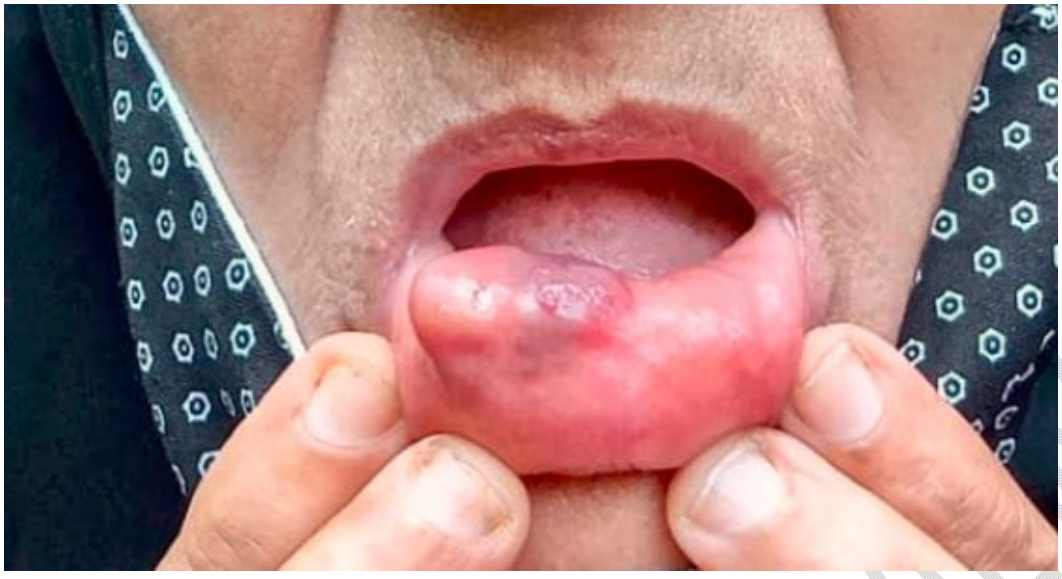

Image 2: POD-1 post the sclerotherapy treatment with 2% Polidocanol.