ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)

IJDSIR : Dental Publication Service

Available Online at: www.ijdsir.com

Volume – 4, Issue – 1, February - 2021, Page No. : 48 - 51

A Case of Lymphangioma Circumscriptum Managed With Sclerotherapy

[1]Dr. Sanjeev Kumar Singh, Senior Resident, Oral Health Sciences Center, Postgraduate Institute of Medical Education and Research, Chandigarh

[2]Dr. Manoj Jaswal , Assistant Professor, Oral Health Sciences Center, Postgraduate Institute of Medical Education and Research, Chandigarh

[3]Dr. Krishan Gauba, Professor and Head, Oral Health Sciences Center, Postgraduate Institute of Medical Education and Research, Chandigarh

[4]Dr. Ashima Goyal, Professor, Oral Health Sciences Center, Postgraduate Institute of Medical Education and Research, Chandigarh

[5]Dr. Aditi Kapur, Additional Professor, Oral Health Sciences Center, Postgraduate Institute of Medical Education and Research, Chandigarh

Corresponding Author: Dr. Manoj Jaswal , Assistant Professor, Oral Health Sciences Center, Postgraduate Institute of Medical Education and Research, Chandigarh

Citation of this Article: Dr. Sanjeev Kumar Singh, Dr. Manoj Jaiswal, Dr. Krishan Gauba, Dr. Ashima Goyal, Dr. Aditi Kapur,"A Case of Lymphangioma Circumscriptum Managed With Sclerotherapy", IJDSIR- February - 2021, Vol. – 4, Issue - 1, P. No. 48 – 51.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Lymphangioma Circumscriptum (LC) is a form of lymphatic malformation where clear or hematinic vescicles are scattered throughout the invaded cutaneous or mucosal region. Lymphangioma Circumscriptum should be considered as one of the differential diagnosis for cystic lesions of buccal cavity and sclerotherapy can be considered as a minimally invasive treatment modality for the same. A case of LC of buccal mucosa in a 13-year old female and the description investigations that led to the diagnosis of the same. Further, treatment using sclerotherapy was done and the patient was kept on follow ups for monitoring. Upon 3 doses of sclerosing agent (Sodium decamethyle sulphate) injection at 15 days intervals, the lesion was found to have considerable reduction in size without any adverse consequences. An MRI at 3 months further reported no evidence of vascular abnormalities.

Keywords: Lymphatic malformation, lymphangioma Circumscriptum, buccal mucosa, sclerotherapy.

Introduction

Lymphatic malformation (LM), formerly known as lymphangioma, is a malformation localized to the lymphatic system. [1] It has a prevalence of 1:4000 and

equally affects males and females. It is most evident at birth or within two years of life. It may be classified as being macrocystic (cystic malformation) or microcystic (Lymphangioma Circumscriptum; LC). The microcystic variety characterized by clear or hematinic vescicles scattered throughout the invaded cutaneous or mucosal region. The signs and symptoms vary depending upon the size and location of the mass. The lesion may also disfigure affected areas and disrupt the function of nearby organs or tissues. Histopathologically, LC shows plump endothelial cells lining dilated lymphatic vessels. The available treatment options for LC include medical management, laser, cryotherapy, radiation therapy, sclerotherapy, electrocautery or excision. The present case report if that of a rare case of lymphangioma circumscriptum occurring in the buccal mucosa of a 13 year old female and managed with sclerotherapy.

Case Report

Upon ultrasonography, a cystic lesion in the right buccal mucosa measuring 33*13mm could be appreciated. It had a 23*10mm superficial component with internal fine septae and debris, and a 14*14 mm deeper component, which largely appeared to have clear, anachoretic fluid. However, these findings could not make the organ of origin clear. Subsequently, an MRI was done which described the lesion as a well defined lobulated, hyperintense lesion which was of vascular origin. This narrowed down the differentials to a hemangioma and a lymphangioma. Histopathological examination following an incisional biopsy revealed tumor fragments lined by hyperplastic stratified squamous epithelium along with dilated lymphatic space in the superficial and papillary sub-epithelium. Surrounding stroma showed scattered lymphocytes. Based on these findings, a final diagnosis of Lymphangioma circumscriptum was made. Available treatment options included medical management with Imiquimod, laser, cryotherapy, radiation therapy, sclerotherapy, electrocautery or excision. Owing to the uncertainty in outcomes of medical management and the invasive nature of other therapies, sclerotherapy was chosen as the line of management. The patient was informed about a probable need for surgical excision. Treatment with sclerotherapy involved 3 ml Sodium decamethyle sulphate being injected in divided doses of 1.5 ml in lesion and 1.5 ml in surrounding tissue. After first dose sclerosing agent there was regression in size. Hence the treatment was repeated. Subsequently, 3 doses were administered at interval of 15 days in same manner. The patient was recalled at regular follow ups for monitoring. At three months, ultra-sonography was done and there were no vascular or dilated cystic lesion. No abnormal vascularity was seen. The patient did not develop recurrences for the next six months and then was lost to follow as the family shifted to a different city. However, telephonic conversation was made to enquire about any recurrence, that could be felt.

Discussion

Lymphangioma is a rare benign congenital lesion of unknown aetiology. It usually originates as a hamartomatous hyperplasia of the lymphatic vessels arising from sequestration of lymphatic tissue that do not communicate with the rest of the lymphatic channels. Lymphangioma was first described by Virchow in 1854.[1] Lymphangiomas have marked predilection for head and neck region accounting for about 75% of all cases. Approximately 50% lesions are noted at birth and 90% develop by 2 years of age.[2] These are commonly located in the head and neck region and are rarely reported in the oral cavity. Lymphangiomas are hamartomatous malformations of the lymphatic system which were first described by Redenbacher in 1828. Lymphangiomas are classified clinically into macrocystic (larger than 2 cm),

micro cystic (smaller than 2 cm) and mixed, which is a combination of the above two types. [3] Based on clinical and histological findings, lymphangiomas in the oral cavity are found to be superficial which occur as lymphangioma simplex, circumscriptum and capillary type. Deep lesions are of micro cystic type.[4] The buccal mucosa is the second most common site reported (14 cases reported) after the anterior two-thirds of tongue.[5] Oral lymphangioma presents as a superficial lesion with pebble-like surface, resembling a cluster of the translucent vesicle. This appearance has been described to be like that of tapioca pudding or frog eggs. The deep lesions clinically present as soft ill-defined masses.[6] It is most commonly found on the shoulders, neck, underarm area and limbs. The most commonly affected intraoral site is the tongue. However, it may occur on the lips, alveolar or buccal mucosa. lymphangiomas located in the mouth or neck region can cause dyspnoea, speech problems, dysphagia and feeding problems. Complications of LC include recurrent cellulitis, which may cause pain and disfigurement of the affected area, bleeding and hardening or rapid enlargement of the mass.[7] Imaging studies for LC include magnetic resonance imaging (MRI), computed tomography scanning (CT scan) and/or ultrasound to examine details of the lesion. Other examinations may be required depending upon the location and type of the lesion. [7] The best treatment options for lymphangiomas depend upon factors such as its size, location, associated symptoms and patient's tolerance towards certain procedures.[7] Involvement of vital structures, aesthetic, and functional needs may necessitate treatment modalities such as cryotherapy, electrocautery, sclerotherapy, surgical excision, radiation therapy, embolization, ligation, and laser therapy.[8]Sclerotherapy is a minimally invasive therapy that may be used to minimize the size of the lesions before surgical excision, or may be used an independent therapy.[9,10]

Conclusion

- Lymphangioma circumscriptum should considered as one of the differential diagnosis for cystic lesions of buccal cavity and a comprehensive approach has to be followed to diagnose the same
- Sclerotherapy is one of the effective means of treating the same and should be considered an effective, minimally invasive treatment option.

References

1. Kheur SM, Routray S, Ingale Y, Desai R. Lymphangioma of tongue: a rare entity. Indian J Dent Adv. 2011;3:635-37.
2. Vasconcelos MG, Santos BC, Lemos LC, Ribeiro BF, Iglesias DP, Vasconcelos RG, et al. Oral lymphangioma: Case report. Rev Sul-Bras Odontol. 2011;8:352–56.
3. Ganesh C, Sangeetha GS, Narayanan V, Umamaheswari TN. Lymphangioma Circumscriptum in an Adult: an unusual oral presentation. Journal of Clinical Imaging Science. 2013;3:44
4. Coskunses FM, Tugcu FE, Kocygit ID, Ozgul O, Karadeniz SN. Lymphangioma of buccal mucosa: an unusual case. Int J Dent Clin. 2012;4(1):50-1.
5. Kurude AA, Phiske MM, Kolekar KK, Nayak CS. Lymphangiomas: Rare presentations in oral cavity and scrotum in pediatric age group. Indian J Dermatol Venereol Leprol 2020;86:230.
6. Sunil S, Gopakumar D, Sreenivasan BS. Oral lymphangioma - Case reports and review of literature. Contemp Clin Dent 2012;3:116-8
7. Nord et al. Lymphatic Malformations. June 2011; https://www.rarediseases.org/rare-disease-information/rare-diseases/byID/646/viewAbstract

8. Balakrishnan A, Bailey CM. Lymphangioma of the tongue. A review of pathogenesis, treatment and the use of surface laser photocoagulation. J Laryngol Otol 1991;105:924-9
9. Bill AH Jr, Sumner DS. A unified concept of lymphangioma and cystic hygroma. Surg Gynecol Obstet 1965;120:79-86.
10. Suen J, Waner M. Treatment of oral cavity vascular malformations using the Meodymium: YAG laser. Arch Otolaryngol Head Neck Surg 1989;9:115-32
11. Dogan N, Durmaz CE, Sencimen M, Ucok O, Okcu KM, Gunhan O, et al. The treatment of recurrent lymphangioma in the oral mucosa by cryosurgery. Oral Health and Dent Manag Black Sea Count. 2010;9(1):7.

Legend Figures

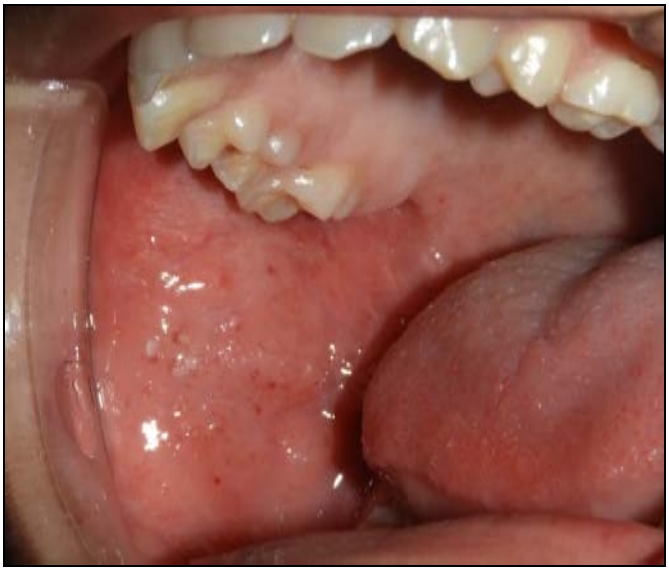

Fig. 1: Lymphangioma Circumscriptum of buccal mucosa in 13-year-old female

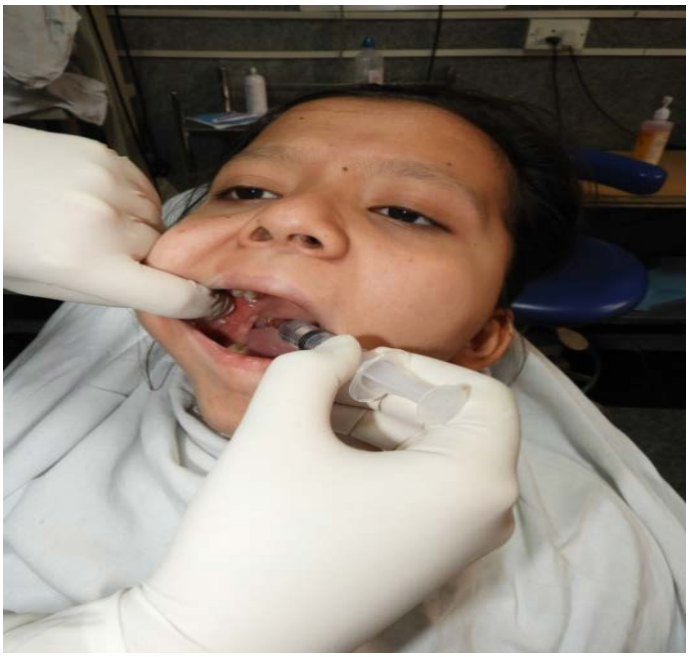

Fig. 2: Administration of 3 ml Sodium decamethyl sulphate injected- 1.5 ml in lesion and 1.5 ml in surrounding tissue.

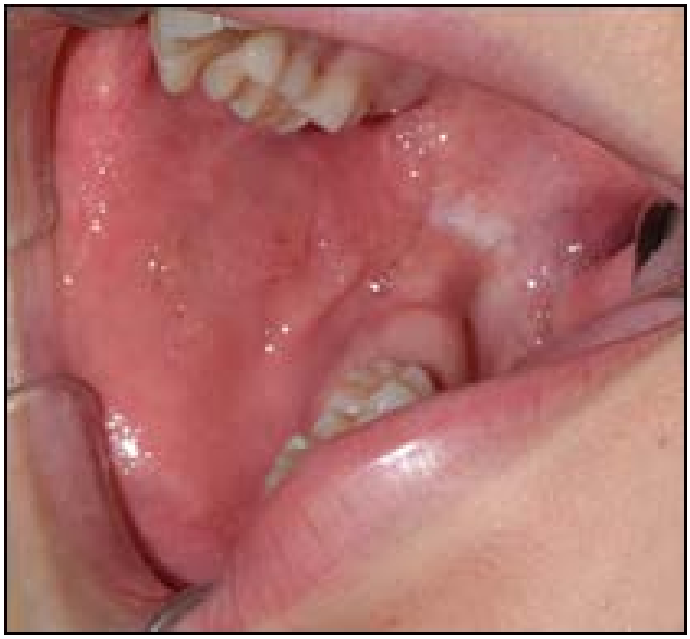

Fig 3: Intraoral site at 6 months follows up.