Case Report

JRAD Journal of Research and Advancement in Dentistry

J Res Adv Dent 2017;6:2s:93-95.

Lymphangioma Affecting Gingiva Unilaterally: A Rare Case Report

Chakravarthy YSHS[1*] Gollapalle Prabandh Reddy[2] Aileni Amarender Reddy[3] Tokala Pallavi[4] Pasupula Anoop Kumaar[5] Rampalli Viswa Chandra[6]

[1]Reader, Department of Periodontology and Oral Implantology, SVS Institute of Dental Sciences, Mahbubnagar, Telangana, India.
[2]PG Student, Department of Periodontology and Oral Implantology, SVS Institute of Dental Sciences, Mahbubnagar, Telangana, India.
[3]Professor, Department of Periodontology and Oral Implantology, SVS Institute of Dental Sciences, Mahbubnagar, Telangana, India.
[4]Senior Lecturer, Department of Periodontology and Oral Implantology, SVS Institute of Dental Sciences, Mahbubnagar, Telangana, India.
[5]Reader, Department of Periodontology and Oral Implantology, SVS Institute of Dental Sciences, Mahbubnagar, Telangana, India.
[6]Professor and Head, Department of Periodontology and Oral Implantology, SVS Institute of Dental Sciences, Mahbubnagar, Telangana, India.

ABSTRACT

Background: Lymphangiomas are benign hamartomatous tumors which are developmental malformations arising from sequestration of lymphatic tissue, which has lost its communication with rest of the lymphatic channels. More than 90% of all the cases of lymphangiomas occur in the head and neck region. In oral cavity, most frequently it involves the dorsum of the tongue followed by palate, buccal mucosa, floor of the mouth, gingiva and lips. In this report, we present a unilateral case of lymphangioma affecting gingiva of a 28 years old woman.

Keywords: Gingiva, lymphangioma, rare disease.

INTRODUCTION

Lymphangiomas are benign uncommon congenital hamartomatous lesions of lymphatic channels. They are rare benign hamartomatous lesions of lymphatic system, which can be more correctly referred to as lymphatic malformation. Although there is no clear mechanism of occurrence, it may occur from the sequestration of lymphatic tissues which have lost its communication with the lymphatic channels.[1] They have a marked predilection for head and neck of which 75% occur in the oral cavity.[2] In the oral cavity, 40-50% of the lymphangiomas involves the dorsum of the tongue with a size range from 1-2cm, other sites involvement may include palate, floor of the mouth, lips and gingiva. 50% of lymphangiomas are present at birth and 90% develop by age 2years.[1] It has no sex predilection and most commonly seen in Caucasians. Surgical excision is the treatment of choice.

This lesion on gingiva is rare and only 4 cases were reported, of which 3 were bilateral symmetrical and 1 case of unilateral lymphangioma of gingiva.[3,4,5,6] This article reports a second case of unilateral lymphangioma affecting gingiva.

CASE REPORT

A 28 years old female patient reported to Department of Periodontics, SVS Institute of dental sciences (Mahabubnagar) with a chief complaint of growth in the lower front tooth region since 3 months. On clinical examination, a sessile, erythematous, asymptomatic smooth surfaced lesion was seen on buccal aspect of right mandibular central incisor, involving the interdental papilla and extending upto attached gingiva with lingual portion intact (Figure 1). The lesion was 1 cm × 0.6 cm in size with no ulceration present on the surface and easy to bleed. The patient complained that the lesion gradually

Received: Feb. 11, 2017: **Accepted:** Mar. 26, 2017
***Correspondence Dr. Chakravarthy YSHS.**
Department of Periodontology and Oral Implantology, SVS Institute of Dental Sciences, Mahbubnagar,Telangana, India.
Email: yshschakri@gmail.com

pISSN 2278-0076
eISSN 2321-9270

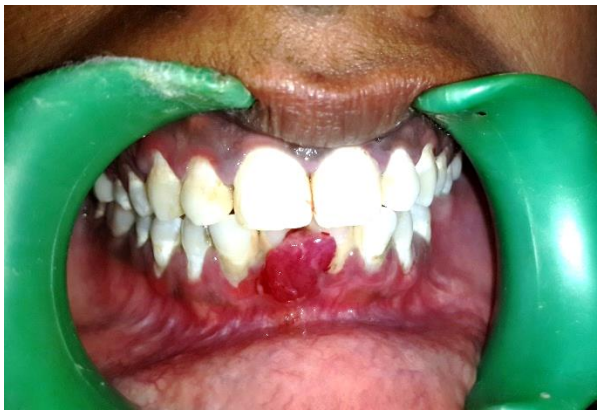

Fig 1: A sessile, erythematous, asymptomatic smooth surfaced lesion was seen on buccal aspect of right mandibular central incisor, involving the interdental papilla and extending upto attached gingiva with lingual portion intact.

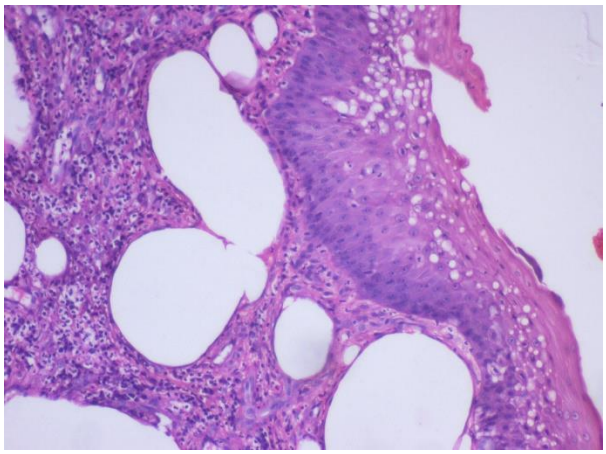

Fig 2: Histopathologically, the underlying lamina propria shows numerous thin walled lymphatic channels of varying sizes, few filled with eosinophilic coagulum in the form of lymph.

increased in size and bleeds while having solid food. Initially oral hygiene instructions and SRP was done prior to the surgery, two weeks later surgery was performed under local anesthesia. The lesion was excised by giving external bevel incision using number 15 blade and the specimen was sent for biopsy to the Department of Oral Pathology for microscopic examination with differential diagnosis as pyogenic granuloma and hemangioma.

According to histopathological features, (Figure 2) the lesion was diagnosed as lymphangioma. Histopathologically the underlying lamina propria shows numerous thin walled lymphatic channels of varying sizes, few filled with eosinophilic coagulum in the form of lymph. The lymphatic channels are seen just below the epithelium without any intervening connective tissue. The stroma also shows dense lymphocytic aggregates and endothelial proliferation in a delicate collagenous background. The epithelium in few areas is hyperplastic with marked intracellular oedema.

DISCUSSION

Lymphangioma was first discovered by Virchow in 1854 as a rare, benign, congenital malformation of unknown etiology that originates from sequestration of lymph tissue which has lost its communication with rest of the lymphatic system.[7] Embryologically it is derived from five primitive buds developing the venous system which include two paired sacs (jugular sacs, posterior sacs) and a single sac (peritoneal sac).[8] Occurrence of these lesions is rare but when affected, the most common site is the tongue and very rarely on the other sites such as the gingiva, buccal mucosa and lips. First case of lymphangioma of gingiva in 16 years old female patient was reported bilaterally, and later other cases of bilateral superficial lymphangiomas were manifested on gingiva in a 11 years old female patient and 32 years old male patient.[3,4,5] However, unilateral manifestation of lymphangiomas of gingiva were rare, only one such case was reported.[6]

In the present case, we have reported a unilateral lymphangioma manifested on the labial gingiva of 41 covering upto 3/4th of the tooth height and involving the marginal gingiva, interdental papilla and extending upto the attached gingiva. Further, the lesion was sessile, erythematous, asymptomatic, smooth surfaced and easy to bleed. Lymphangiomas are divided into 3 groups, simplex(capillary), cystic and cavernous. Some authors suggested that histological differences in various lymphangiomas are attribute to the differences in anatomical location and therefore histological classification is of little benefit.[8] According to this concept some authorities classified lymphangiomas into superficial and deep type. In our case we reported as superficial lymphangioma.

Treatment for lymphangioma varies depending on their type, size, involvement of

anatomical structures and infiltration to the surrounding tissues. Various procedures such as surgical excision, laser surgery, electrocautey, cryotherapy, embolization, radiation therapy, steroids, ligation and radio frequency tissue ablation techniques have been used.[9] Of all the above treatment options, surgical excision has been a better alternative for lesions presenting localized growth. As the lesion in our case report had superficial presentation we planned for surgical excision.

The infiltrative nature of lymphangioma is the main reason for its reccurence, and the most common places for its reccurence are tongue, larynx and hypopharynx. A recent study has concluded that the reccurence rate of lymphangioma as 39%.[10]

CONCLUSION

Lymphangiomas when occurred usually manifests in gingiva, buccal mucosa and lips. Considering that there has been only a few studies done on this lesion, it is presently impossible to specify the lesions most prevalent site. In the present study the lesion is manifested on the gingiva unilaterally, although 3 other studies report bilateral involvement. In view of the present finding of the study lymphangiomas should also be considered in the differential diagnosis of unilateral lesions of the gingiva.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Neville BW, Damn DD, Allen CM. Oral and Maxillofacial Pathology. WB Saunders: Philadelphia; 1995. p. 395.
2. Mandel L. Parotid area lymphangioma in an adult: Case report. J Oral Maxillofac Surg 2004;62:1320-3
3. Josephson P, Van Wyk CW. Bilateral symmetrical lymphangiomas of the gingiva. A case report. J Periodontol 1984;55:47-8
4. McDaniel RK, Adcock JE. Bilateral symmetrical lymphangiomas of the gingiva. Oral Surg Oral Med Oral Pathol 1987;63:224-7
5. Motaharry P, Sarrafpour B, Abdirad A. Bilateral symmetrical lymphangiomas of the gingiva: Case report. Diagn Pathol 2006;1:9.
6. Avideh Maboudi, Maryam Seyedmajidi, Unilateral gingival lymphangioma: A first case report, J Indian Soc Periodontol 2014;18:794-6.
7. 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.
8. Morley SE, Ramesar KC, Macleod DA. Cystic hygroma in an adult: a case report. J R Coll Surg Edinb 1999;44:57-8.
9. Cable BB, Mair EA. Radiofrequency ablation of lymphangiomatous macroglossia. Laryngoscope 2001;11:185961.
10. Orvidas LJ, Kasperbauer JL. Pediatric lymphangiomas of the head and neck. Ann Otol Rhinol Laryngol 2000;109:411-21.