Original Article

Lymphangioma of Gingiva: A Rare Case Report

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Abstract

One of the unconventional congenital hamartomas of the lymphatic vessels, are termed as Lymphangiomas, with a common age of diagnosis from birth to early childhood. These occurrences are most commonly seen in the region of head and neck while still being uncommon in the oral cavity. Clinically they can be superficial or deep with superficial lesions showing a pebbly surface and deeper lesions being more diffuse. Excising the lesion surgically is considered the treatment of choice. Although majority of cases have a decent rate of prognosis, the recurrence rate however is notorious for being high due to its non-encapsulated and infiltrating nature where complete removal becomes difficult. In spite of the rarity of lymphangiomas in the oral cavity, the condition should be considered by the clinicians to initiate early and proper treatment to avoid complications. The following case report establishes a case of lymphangioma present on the lingual gingiva of a male in his twenties.

Keywords: Lymphangioma, Intraoral, Gingiva.

Introduction

Lymphangiomas are considered a benign form of hamartomatous proliferation of lymphatic channels. These proliferations are not considered neoplasms but are actually classified under malformations, with a higher prediction of occurrence in the region of the head and neck.[1] Even though a rare phenomenon, the lymphangiomas can also be localized orally with tongue being a common site. The majority of cases are present at birth usually before the age of two years, with no evident gender predilection.[2] The superficial lesions usually manifesting as papillary lesions appear as frog eggs or tapioca pudding.[3]In this article, we have reported a case of lymphangioma present on the gingiva in a young individual which is extremely rare.

Case Report

A young male presented to the Department of Oral Surgery with a chief complaint of a growth on the lingual gingiva of the lateral incisor and canine of the left mandible with buccal side intact, which he noticed about a month ago. On clinical examination, a 1cm X 0.5cm nodular mass which was asymptomatic with mild bleeding was noted on lingual aspect attached to the gingiva of the left mandibular lateral incisor and canine. Lesion surface was intact without any ulceration (Figure 1). No alteration was detected in the radiograph. There were no anomalies in the medical investigations which were all within set ordinary limits. No sign of cervical lymphadenopathy was noted. A provisional diagnosis of pyogenic granuloma was made. The lesion was excised and sent for histopathological analysis. The H &Estained tissue section showed a surface lining of atrophic stratified squamous epithelium which was orthokeratinized. Numerous enlarged lymphatic vessels lined by flattened thin endothelial cells containing few lymphocytes and eosinophilic proteinaceous material were located just beneath the epithelium. Few

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Received:17-07-2019; **Accepted:**11-11-2019

Access this article online

Quick Response Code:

Website: www.nigerianmedjournal.org

How to cite this article: Shivani A, Anchal V, Ashim A, Dimple A. Lymphangioma of Gingiva: A rare case report. Niger Med J 2021; 62; (1):46-48.

channels also contained RBC's. The intervening connective tissue stroma was made up of mixed inflammatory infiltrate containing lymphocytes and PMNL (Figure 2, 3).On the basis of these features a diagnosis of lymphangioma was made. Hence after, the patient was kept under regular follow-up for 3 years and had showed no signs of recurrence.

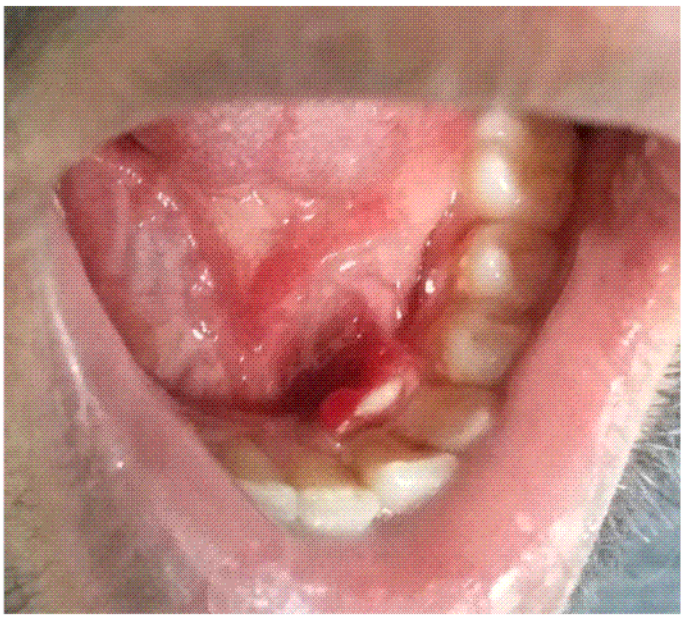

Figure 1: Clinical photograph showing a nodular lesion on the lingual gingiva of mandibular lateral incisor and canine.

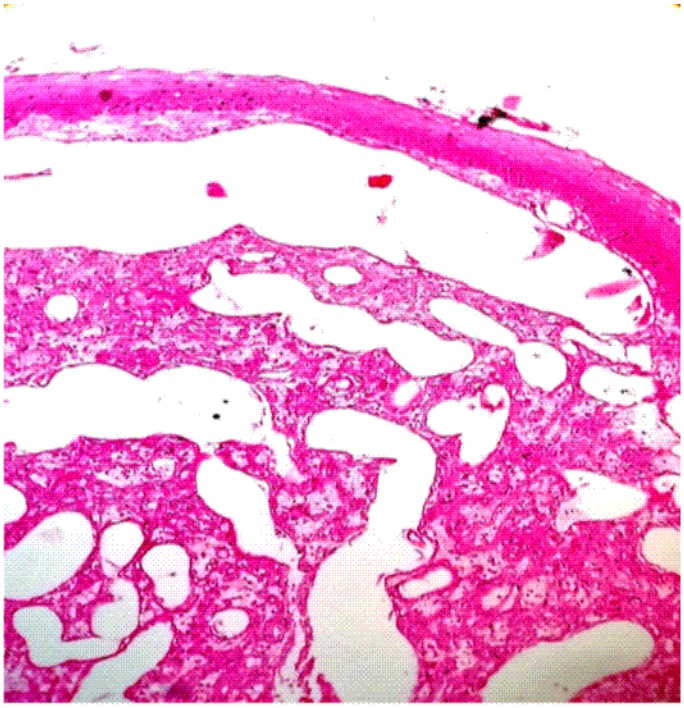

Figure 2: Photomicrograph showing atrophic orthokeratinized epithelium with enlarged lymphatic vessels (10X).

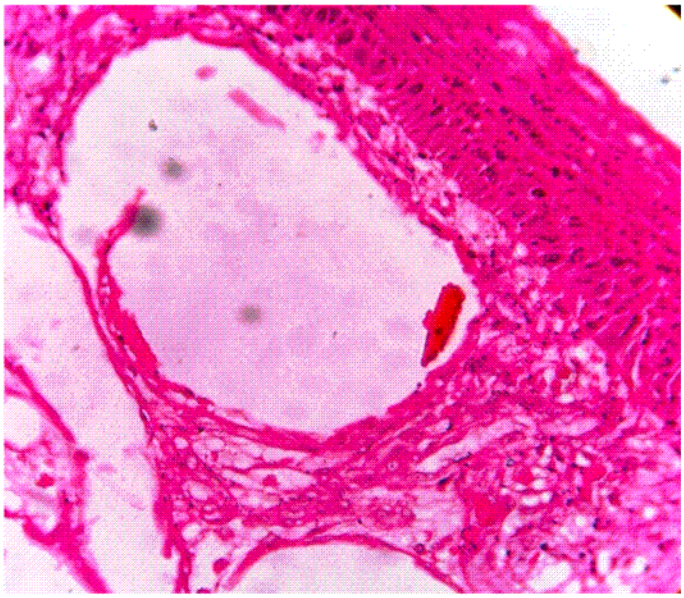

Figure 3: Photomicrograph showing enlarged lymphatic vessels lined by flattened thin endothelial cells (40X).

Discussion

Lymphangiomas represent vascular impairment affecting the lymphatic system. They are benign in nature and are developmental anomalies that arise from sequestration of the lymph sac. They are comparatively larger in size or have a tendency to enlarge due to lack of drainage and absence of normal communication with the central lymphatic system.[4]Lymphangiomas are usually present at birth and majority develop before the age of two.[2] Contrary to the above, the present case was reported in second decade of life. Literature reports that approximately 75% of cases are found in the head and neck region.[5]In this region, the most common site is the lateral neck, where a characteristic feature of large cystic spaces is observed, histopathologically, thereby being called as Cystic Hygroma.[6] Occurrence in the oral cavity is rare with the most common site being the anterior two third of the tongue followed by lips, floor of the mouth, soft palate, gingiva ,buccal mucosa, and alveolar ridge in no specific order. It is the most common cause of macroglossia, which neither becomes malignant nor has a familial tendency. There is no sex predilection and spontaneous regression of lymphangioma unlike hemangiomas, is rare.[7]

Few cases of lymphangiomas occurring bilaterally on gingiva have been reported, with dearth of evidence of its occurrence unilaterally.[3] Although lymphangiomas are benign lesions, due to its penetrating nature, compression and infiltration of vital structures, unlimited demarcation, aesthetic and

functional requirements, they should be considered in the differential diagnosis of exophytic lesions of gingivalike pyogenic granuloma, fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma and giant cell fibroma.[7]

Clinically, lymphangiomas are classified as superficial and deep, where peripheral lesions appear as pink or yellow color elevated nodules. These lesions, in due course becomes reddish or purple due to secondary hemorrhage. However, the lesions situated deeper present as soft, diffuse masses with a normal color.[8]Histologically, they have been termed as: a) Cavernous lymphangioma b)Lymphangioma simplex (capillary lymphangioma)c) Cystic Hygroma.[1]

Histopathologically, lymphangiomas consist of dilated lymphatic vessels which diffusely infiltrate the adjacent soft tissue. The lining of the vessels is done by thin endothelial cells and the spaces may contain lymphocytes and proteinaceous fluid with evidence of secondary hemorrhage sometimes. The surrounding connective tissue stroma is loose fibrillar with infiltration of inflammatory cells. Small sized capillary vessels are seen in the capillary lymphangioma while larger dilated vessels are seen in cavernous lymphangioma and large cystic spaces are exhibited in cystic lymphangioma. However, all sizes can also be seen within the same lesion.[9]According to this, our case is superficial cavernous type and is apparently confined to the gingiva. However, Bill and Sumner suggest in their study, difference in anatomic location is one of the reasons why histological differences in various lymphangiomas are present. Hence, according to them histological classification is less useful.[3]

Various treatment modalities used for the treatment of lymphangiomas include electrocautery, radiation therapy, cryotherapy, sclerotherapy, ligation, laser surgery with surgical excision being the treatment of choice. Majority of the cases have a good prognosis, but some researchers have noted high recurrence rate due to its non-encapsulated and infiltrating nature where complete removal becomes difficult.[10]

Conclusion

Lymphangiomas are benign in nature and mainly confined to the region of head and neck, but intraoral lymphangiomas are rare in occurrence with gingiva being the least involved site. Because of the rarity and the diagnostic dilemma that it poses, we report a case of unilateral gingival lymphangioma, so that the localized lesions can be treated by conservative surgical excision with less recurrence. Therefore, a precise diagnosis with proper therapeutic intervention of such lesions is crucial.

Conflict of interest: Nil
Source of funding: Nil

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