LYMPHANGIOMA OF THE BUCCAL MUCOSA

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Abstract

Lymphangiomas are uncommon congenital hamartomas of the lymphatic system, usually diagnosed in infancy and early childhood. Most lymphangiomas are present at birth (60%) and, by the age of 2 years, 80% to 90% are present. Tongue, palate, gingiva, lip, alveolar ridge and buccal mucosa are the most commonly affected sites in the oral cavity. Various methods have been tried for the treatment of lymphangioma, including surgery, radiation, laser therapy, sclerotherapy and radiofrequency ablation. This paper reports a case of lymphangioma in a 28 year-old female patient.

Keywords: lymphangioma, hamartoma, buccal mucosa

INTRODUCTION

Lymphangioma is a benign, hamartomatous tumor of the lymphatic system, widely considered as a developmental lesion rather than a true neoplasia. [1] It has been reported that approximately 75% of all cases of lymphangioma occur in the head and neck region, and that about 50% of all lesions appear at birth. About 90% of them are developed around the age of 2 years. [2] The affected sites in the oral cavity may include the tongue, palate, gingiva, buccal mucosa, lips, and the alveolar ridge of the mandible. The preferred oral involvement is the tongue. Lymphangioma of the buccal mucosa is very rare. [3] Various methods have been used for the treatment of lymphangioma, including surgery, radiation, laser therapy, and sclerotherapy. Recently, a new and more conservative surgical approach to this lesion, using a radiofrequency tissue ablation technique, has been also suggested. [4]

CASE REPORT

A 28 year-old female patient reported to the department of oral medicine and radiology with the chief complaint of swelling in the right cheek region. The medical history of the patient was common. Extraoral examination provided no new data. On clinical examination, discoloration was seen in the right buccal mucosa. The mucosal lesion was reddish purple in colour, with a smooth surface measuring approximately 2.0 X 1.0 cm (Fig. 1). The patient said that the lesion was present since birth and grew slowly with age. She had no previous dental history related to it, and no symptoms associated with the lesion. The mucosal lesion blanched on

Figure 1: Purplish lesion in the buccal mucosa
pressure. Therefore, on the basis of clinical appearance, a provisional diagnosis of hemangioma was made. Orthopantomographic examination showed no evidence of adjacent bone involvement. The lesion was surgically excised, the clean borders 2-3 mm around included, then subepithelially and histopathologically evaluated.

Histopathology showed an admixture of numerous endothelial-lined lymphatic channels and cavernous spaces (Fig 2). The lymphoid spaces, containing eosinphillic materials, suggested lymph and few red blood cells. Based on histopathological data, a final diagnosis of lymphangioma was established.

DISCUSSION

Lymphangioma is a congenital abnormality of the lymphatic system, manifested frequently as early as birth or before the age of 2 years. [1] It has been postulated that it develops from the sequestration of portions of the primitive embryonic lymphatic anlage. [5,6] These sequestered areas never achieve efficient anastomoses with the larger lymph channels; therefore, functionally they exist as localized areas of lymphatic blockage, some appearing as lymphangiomas, others as cystic hygromas. The natural history of these lesions is one of recurrent enlargement secondary to infection and trauma. [5]

Clinically, lymphangioma of the oral cavity is rather characteristic. Usually, the lesions are superficial, with a pebbly, vesicle-like feature and the so-called “frog-egg” or “tapioca-pudding” appearance. If located deeper, lymphangioma may present as a submucosal mass. [7] Although it is a benign lesion, it may lead to complications, because of its infiltrating nature, indefinite demarcation and involvement of vital structures. [4] Lymphangioma may be similar to a number of oral lesions including hemangioma, teratoma, lingual thyroid, dermoid cyst, thyroglossal duct cyst, heterotopic gastric mucosal cyst, and granular cell tumor. For inexperienced clinicians or in cases of lesions with atypical clinical features, final diagnosis should be made through biopsy and histopathological examination. [7]

Histopathogically, the proliferated vessels of the lymphatic system are lined by plump endothelial cells. The lumens of the lesion contain eosinophilic coagulum with erythrocytes and leukocytes. [3] Lymphangiomas are classified morphologically as macrocystic, microcystic, and mixed structures. Macrocystic lesions are cystic spaces of at least 2 cm, microcystic lesions are cystic spaces of less than 2 cm, and mixed lesions contain macrocystic and microcystic components. [4,8]

Besides, although lymphangioma is rare in adults, no transformation associated with age may be established. Lymphangiomas neither become malignant nor have a familial tendency. [3]

Treatment of lymphangiomas can be challenging and various methods have been attempted at, including surgery, radiation, laser therapy, and sclerotherapy. [4,9] Surgery has been the main form of treatment, but total removal is not possible as, in some cases, the lesion involves vital structures. [5] Laser photocoagulation has been reported as useful in controlling tongue size and, in some cases, removing superficial lymphangiomata. [10] Two sclerosing agents, bleomycin and OK-432 (Picibanil, a strain of A streptococcus group), have been preferred by surgeons in the treatment of lymphangioma. [4,9] Although hemangiomas respond well to the injected sclerosing agents, the experience of many oral and maxillo-facial surgeons showed that lymphangiomas are relatively unresponsive.

Figure 2: Endothelial lined large lymphoid vessels (H & E x 400)
to sclerosing agents. The reasons for this lack of response are not understood, but it is possible that differences in the endothelial lining or in the components of the vascular wall may have some part. [1]

CONCLUSIONS

Although rarely encountered in the oral cavity, lymphangiomas represent a malady that should be taken into consideration. Their early recognition allows a proper initiation of treatment and prevents possible complications.

References