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Follicular Lymphoid Hyperplasia of Palate: A Rare Case Report

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Abstract:*Lymphoid hyperplasia of the palate is a benign lympho-proliferative disorder which may be clinically and histopathologically confused with lymphomas. It is an extremely rare clinical entity with less than 25 cases reported in the English language literature.It is more common in female patients and presents as a painless, firm, well-demarcated, usually non-ulcerated, slow-growing lesion on the palate that histopathologically may resemble a lymphoma. The diagnostic challenge is to distinguish follicular lymphoid hyperplasia which is a benign disease with excellent prognosis from that of lymphomas which carries a grim prognosis. Histopathological examination will be indicative of the benign nature of FLH, however, the diagnosis will have to be substantiated with Immunohistochemical analysis and if need be, molecular studies. We report a case of a 37 year old female who reported with a slowly growing swelling in the right side of hard palate of two years duration. The lesion was treated satisfactorily by surgical excision and the diagnosis of FLH was corroborated by histopathological examination and Immunohistochemical studies.*

Key Words*:Lymphoid hyperplasia;Benigntumour of oral cavity; Lympho-proliferative lesion.*

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I. Introduction

Lymphoid lesions in the palate constitute the second largest group of neoplastic lesions in the palate after salivary gland tumours[1]. These lymphoid lesions may be benign or malignant. The malignant lesions are usually non-Hodgkin's lyrnphornas[2], which include lympho-proliferative disease ofthe hard palate (LDHP) as described by Tomich and Shafer[3]. These lesions represent an extra nodal lymphoma which mayor may not also involve widespreadsystemic disease in lymph nodes or reticulo-endothelialtissues. While lymphomas form the second most common malignancy in the oral cavity after squamous cell carcinoma involving the mucosa or the gnathic bones, [4,5]benign lymphoid lesions of the hard palate, like benign follicular lymphoid hyperplasia are exceedingly uncommon[6-10].Follicular lymphoid hyperplasia is an asymptomatic, non-ulcerated, normal-colored, firm, unilateral, sessile swelling of the hard palate[6, 11]. A case of FLH in a patient showing a unilateral soft-tissue swelling of the hard palate is described below.

II. Case Report

A 37 year old female patient reported to our clinic with complaint of a swelling in the posterior hard palate on the right side. The swelling was noticed approximately two years back and it slowly grew to the present size. Her past medical and family history as well as systems review were non-contributory.On examination, a painless, firm swelling of approximately 30mm x 20 mm in size with well-demarcated margins and an intact mucosa covering was seen on the right posterior hard palate extending across the midline. The swelling was mildly fluctuant. The patient reported numbness of the posterior hard palate for several months. The second premolar on the right side was found to be deeply carious. Occlusal radiographs and Para-nasal sinus radiographs showed no evidence of bony destruction. CT scan revealed mild cuffing of the bony bed indicating pressure effect. A provisional diagnosis of a malignant minor salivary gland neoplasm was made. An incisional biopsy was performed for histopathological examination. Microscopic examination of the specimen revealed lymphoid tissue covered with mildly keratinized, hyperplastic stratified squamous epithelium. The sub epithelial band of fibrous tissue was found to be infiltrated by lymphocytes and plasma cells. There were follicular aggregates of lymphoid cells. Many follicles showed reactive germinal centers with areas of fibrosis in between. No evidence of malignancy was found. A diagnosis of follicular lymphoid hyperplasia was made and the lesion was excised surgically. The excision biopsy also corroborated the diagnosis of FLH. To further exclude the possibility of lymphoma, Immunohistochemical tests were carried out with IHC marker CD 3 and IHC marker CD 20 which revealed normal T cell and B cell areas further confirming the diagnosis of FLH. The

surgical site healed uneventfully and the decayed premolar tooth was extracted three months after the lesion was excised. The patient is kept under follow up and there is no sign of recurrence over the last seven months.

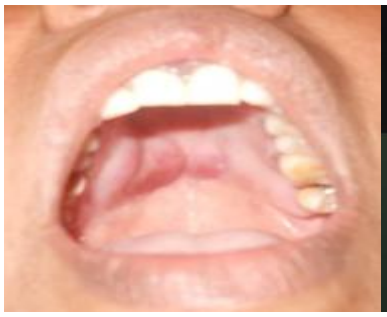

Figure 1: Pre-operative photo

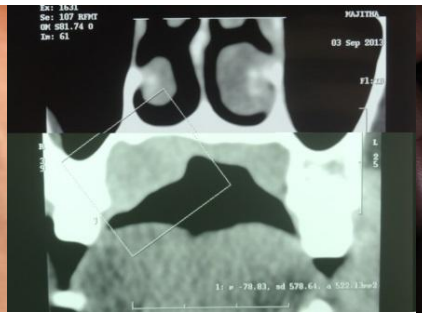

Figure 2: CT Scan view

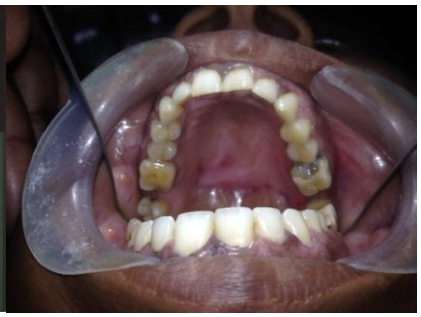

Figure 3: Post-operative photo

III. Discussion

Lymphoid aggregates are present in a variety of locations in the oral cavity, the lateral border of the tongue, the ventral surface of the tongue and floor of mouth, the buccal mucosa, the fauces, and the soft palate. Quite obviously, these lymphoid tissues are subject to most, if not all, of the various reactive and neoplastic processes occurring in the extra oral lymphoid tissues. While at least some of these processes are not commonly encountered in the oral lymphoid tissues, they are not so uncommon as to be considered rare. Such lympho-proliferative lesions encountered in the oral cavity pose the same diagnostic problems as that elsewhere, with benign proliferations being confused with malignant ones and vice versa[3].

Follicular lymphoid hyperplasia of the hard palate is a benign reactive proliferation of lymphocytes of unknown etiology[6-10] which may be confused clinically and histologically with malignant lymphoma.FLH occurs most frequently in older patients, ranging from 38 to 79 years (mean age 61 years). Women are more often affected than men, with a female: male ratio of 3.2: 1. The condition usually presents as a unilateral, painless, slow-growing, non-ulcerated mass located in the posterior hard palate. Occasionally, the soft palate is affected, or there is multicentric involvement[9]. Apart from Malignant lymphoma the differential diagnosis include salivary gland tumor's, palatal abscesses, benign lymphoepithelial lesion, adenomatoid hyperplasia, and mesenchymal tumours.[12]

Radiographically there are no intrabony abnormalities[6] and other laboratory findings are usually normal[7, 8]. However in this case there was hypoesthesia of the palate on the affected side as well as cuffing of bone on the palatal shelf which further aroused the suspicion of a malignant lesion. These features can be explained as due to pressure effect causing compression on the bone and the nerve rather than due to infiltration.

The diagnosis of follicular lymphoid hyperplasia is primarily made by histopathological examination of the excised specimen. Histologicalfeatures include well defined germinal centers surrounded by a rim of well differentiated lymphocytes together with a mixed, mainly mononuclear infiltrate with many plasmacytic lymphocytes. The histological presentation of the lesion in this case correlated well with the above picture and hence there was no difficulty in diagnosing the condition. However rarely the histological picture could be confusing with indistinct germinal centers, ill-defined mantles and a lack of tangible-body macrophages imparting an impression of monotony to the lymphoid cell population[12]. In such cases, the diagnosis is uncertain and the patient undergoes extensive clinical and laboratory investigation like immunohistochemistry and PCR as part of the staging process for lymphoma.[8, 13, 14, and 15]

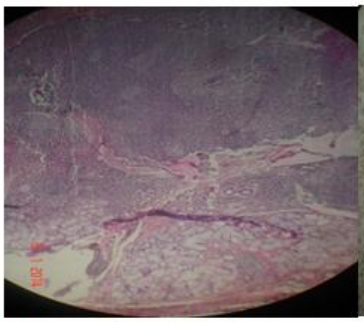

Figure 4: Histopathology

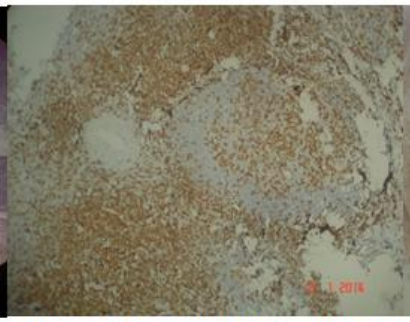

Figure 5: Immunostaining with CD3 (Positive in the follicle center and negative in the mantle zone)

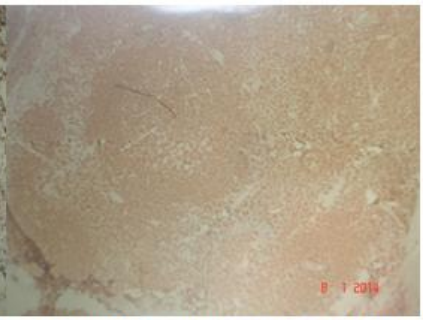

Figure 6: Immunostaining with CD20 (Uniformly positive throughout the central and mantle regions)

At present the etiology and pathogenesis of this lesion remains unclear. It has been postulated that FLH could develop in response to an antigenic stimulus or chronic irritation like that from a partial denture[6, 12]. Although our patient had an infected tooth in the immediate vicinity of the lesion it is unclear that the tooth in question initiated or contributed to the development of the lesion. According to Samoszuk, Epstein-Barr virus may be associated with a persistent and unusual form of FLH.[16] However, no correlation with the use of tobacco, alcohol, medications, or with systemic diseases has been described[6,8].

Follicular lymphoid hyperplasia of the hard palate can be successfully treated by wide excision of the lesion[6, 7, 11, 12, 15, 17]. In addition to surgical excision, other modalities like radiotherapy[11], Intralesional steroid injections[18] and no treatment[9] have been discussed as treatment options in FLH. Surgery has been found to be a very satisfactory treatment for FLH, however recurrence following surgical excision has been reported in two cases.[8,14]Intralesional injection of triamcinolone has been reported by Anjomshoaa[18] with success, however the follow up period was only seven months.

In summary FLH represents a lesion which has to be distinguished from other lymphoid swellings of the palate especially from malignant lymphomas. Histopathological examination would suggest that the lesion is FLH in cases with typical histological features but in those cases where the histological picture is not unambiguous, Immunohistochemical studies and PCR may have to be performed to eliminate the possibility of lymphoma. The most accepted treatment option is surgical excision, however radiotherapy, intralesional steroids and no treatment with close follow up, are also options that have been tested.

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