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Case Report

A Grinspan Syndrome: A Case Report

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Abstract

Oral lichen planus is common mucocutaneous condition involving skin, mucosa or both. Although exact etiology is unknown, an immune mediated pathogenesis is thought to be responsible for lichen planus. Oral lichen planus can occur with some systemic conditions also. Grinspan's syndrome shows association between diabetes mellitus, hypertension and lichen planus. The causative factors are multifactorial such as OLR (oral lichenoid reaction) caused by drugs, exacerbation during stressful conditions, autoimmune mechanism, contact hypersensitivity and association of hepatitis C. The present case discuses about middle aged female with long history of diabetes mellitus and hypertension. Initially she had poor control of blood pressure and blood sugar which led to exacerbation of oral lichen planus. Oral prophylaxis to control local factors, proper monitoring of blood pressure and blood sugar, stress management, appropriate local application with corticosteroid definitely help her to keep it to lower level.

Keywords: Grinspan Syndrome; Oral Lichenoid Reaction; Diabetes Mellitus; Hypertension

Introduction

Oral lichen planus (OLP) is a chronic inflammatory disease of an autoimmune origin. OLP is a mucocutaneous disorder of oral mucosa, skin, scalp, genital mucosa and nails. Oral lichen planus may be present alone or in association with cutaneous lichen planus [1]. The preponderance is seen between 30- 50 years and predominately female [2]. Female-to-male ratio is approximately 2:1.1 The prevalence of about 0.5% to 2%. OLP is categorized into reticular, papular, plaque like, atrophic, erosive and bullous types. The clinical presentation of reticular and plaque like forms are as papules and plaques with lace like white keratotic lines known as Wickham striae bordered by an erythematous border [3]. The buccal mucosa is the predominant site followed by tongue and gingiva. The lesions are mostly bilateral [4].

The association of OLP with diabetes mellitus and vascular hypertension was mentioned by Grinspan., *et al*. in 1966 and therefore this triad is known as Grinspan's syndrome [5].

OLP has been defined as oral potentially malignant disorder (OPMD) by the World Health Organization (WHO) Collaborating Center for Oral Cancer. Oral lichenoid lesions (OLLs) have clinical and histopathologic similarities to OLP. They are termed according to the substance causing them, if caused by dental substance it is termed as oral lichenoid contact lesions (OLCLs) and if due to systemic drug it is termed as, oral lichenoid drug reactions (OLDRs) and if graft is the cause then oral lichenoid lesions of graft-versus-host disease (OLL-GVHD) at the 2006 World Workshop of Oral Medicine IV [6].

Diagnosis of OLP is made by clinical examination with histopathologic confirmation. To eliminate any specific

autoimmune disease like pemphigus and pemphogoid, direct immunofluorescence test is mandatory. OLP and OLR share similar histopathological picture. Topical corticosteroids are the first line of treatment for OLP. Drugs of other class such as retinoids, tacrolimus, cyclosporine constitute second line of treatment. Sometimes photodynamic theory is also proven beneficial. Malignant transformation is seen in few cases but and the appropriate pathogenesis is still unclear [7].

Case Report

A 56-year female resident of Ajmer, a homemaker presented with chief complaint of white patch with discoloration on both sides of cheek, on and undersurface of tongue with difficulty in taking spicy food for the past 3-4 years. Consumption of chilies gives discomfort and burning sensation to the extent that she is taking food totally devoid of chilies. The patch is gradually increasing in size. Personal history reveals her as non-smoker, non- alcoholic and vegetarian. She has taken various treatment for it but was of no use. She was undergoing constant stress owing to her children's education. She was diagnosed as hypertensive 11 years back for which she is taking Amlodipine 5mg and Atenolol 50mg.She is diabetic for the last 6 years for which she is taking Metformin 500mg OD and Teneligliptin 20mg OD initially. Presently she is taking metformin sustained released 500mg with Glimepiride 2mg.

Clinical picture shows white lacy patch on both side buccal mucosa extending to the floor of mouth and on the dorsum and ventral surface of the tongue. Patchy areas showed period of remission and exacerbation. These patches were surrounded by erythematous zone. There was no sign of skin involvement. Complete blood count (CBC) was normal. The blood pressure 138/90mm of Hg and random blood sugar was 210mg/dl. Histopathology report showed hyper-parakeratinized stratified squamous epithelium with dense infiltration of lymphocytes in sub-epithelial zone confirming reticular lichen planus. As the patient has diabetes mellitus and hypertension along with oral lichen planus it is diagnosed as Grinspan syndrome.

Treatment was directed towards stress management, keeping blood sugar and BP within normal range. She was advised to maintain chart noting her blood sugar and BP. Oral prophylaxis was done in order to remove local aggravating factors. The patient was instructed to avoid spicy food and take a healthy diet rich in fresh fruit and vegetables. Mutlti-vitamin capsules and Vitamin C tablets were advised for 15 days and 0.1% trimiacinolone was prescribed for local application 2-3 times daily. The patient was called after 15 days. Patient was relived of burning sensation. Patient is kept on follow up after 3 months.

Figure 1: (a) Extra-oral picture of patient. (b) White lacy patch on buccal mucosa and dorsum of tongue. (c) White patch on undersurface of tongue. (d) White patch on left side of buccal mucosa with areas of hypopigmentation.

There were areas of diffuse hypopigmentation seen on both sides of buccal mucosa within remission of white patch. She is kept on follow up every 3 months.

Figure 2: Diffuse hypopigmentation patches seen on bucal mucosa bilaterally and on dorsum of tongue during follow up.

Discussion

Reticular form of oral lichen planus depicts its clinical picture as small white papules or white lines network which is known as Wickham's striae. Erosive and atrophic form of OLP are associated with high malignancy transformation. The reasoning for this increased carcinogenic potential is the atrophy or absence of the epithelium which facilitates the entry of carcinogenic agents are more [5]. Since it is considered as a potentially malignant disorder, timely diagnosis with appropriate treatment and follow-up is very vital to avoid further complications. However, the incidence of malignant transformation in OLP is quite low. Biopsy is compulsory to rule out dysplasia or malignancy. The risk can be attenuated by patient counseling, proper oral hygiene and abolition of exogenous carcinogens, effective treatment and utilization of healthy diet [1].

The rate of malignant transformation is high in smokers, alcoholics, and HCV-infected patients, compared to patients without these risk factors [8]. The malignant transformation rate of OLP and OLRs are 1.37% and 2.43%. Increase malignant transformation risk is associated with female gender, on tongue and in red clinical forms [9].

Pathogenesis

Many theories are associated with development of oral lichen planus but antigen-specific and non-specific mechanism most appropriately explains its pathogenesis. Antigen-specific mechanism in OLP include unmasking of an antigen that may peptide or heat shock protein presentation by basal keratinocytes followed by killing by $CD8^+$ cytotoxic T cells. Mast cell degranulation and matrix metalloproteinase activation explains Non-specific mechanisms of OLP. Activation of these process leads to accumulation of T cell in the superficial lamina propria, leading to basement membrane disruption, intra-epithelial T cell migration and finally keratinocyte apoptosis in OLP [10].

The principle of using corticosteroid as first line of treatment is their ability to alter the inflammation and immune response. Corticosteroid acts on lysosome by preventing the release of enzymes further preventing the release of exudate thereby reducing pain and inflammation [11]. Corticosteroids of mild nature such as triamcinolone acetonide, high-potent fluorinated corticosteroids such as fluocinonide acetonide, disodium betamethasone phosphate are applied topically, and newly developed, highly potent halogenated corticosteroids such as clobetasol are used depending on the severity of the lesion. The disadvantage with topical corticosteroids is their inability to remain in contact to the mucosa for a sufficient long time. No increased efficiency is seen in applying corticosteroid with adhesive base as compared to plain application of corticosteroid [12]. Patients with wide spread of OLP are treated with high-potent and newly developed super potent corticosteroids mouthwashes and intralesional injections. Another disadvantage for long time associated with decreased immune response of corticosteroid is development of secondary candidiasis which demand for antifungal therapy [11]. Systemic corticosteroids are indicated for resistant erosive or erythematous LP where topical approaches have failed. Low doses of systemic prednisolone is common drug used with short doses (40-80 mg for 5-7 days) [13].

The other drugs used are calcineurin inhibitors such as cyclosporin, tacrolimus, pimecrolimus, retinoid and dapsone, mycophenolate and low dose, low molecular weight heparin (enoxaparin). The non-pharmacologic modality includes PUVA therapy, photodynamic therapy and Laser therapy.

A variety of LASER are used such as 980-nm Diode laser, CO_2 laser evaporation, biostimulation with a pulsed diode laser using 904-nm pulsed infrared rays and low-dose excimer 308-nm laser with UV-B rays are studied. The laser kills the superficial epithelium containing the target keratinocytes by protein denaturation. A diode laser destroys the underlying connective tissue with the inflammatory component along the epithelium owing to its deeper penetration [14].

Mignogna., *et al*. have suggested that regular follow-up of patients with OLP should be performed up to every 4 months. OLP with dysplasia should be examined more frequently, every 2-3 months. However, patients with asymptomatic, mainly reticular type may be assessed annually [15].

Conclusion

A small subset of OLP patients (1.1%) develop OSCC; therefore, regular follow-up for these patients is recommended. A higher incidence of malignant transformation was found among smokers, alcoholics, and HCV-infected patients; however, these associations should be further investigated [8].

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