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Case Report

Discoid lupus erythematosus with epithelial dysplasia: a case report

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ABSTRACT

Lupus erythematosus is a classic example of an immunologically mediated condition and is one of the most common of the so called "collagen vascular" or connective tissue diseases. Lupus erythematosus was first described by Biett in 1828 and Kaposi in 1872. Development of squamous cell carcinoma (SCC) can occur in DLE, with about 20 cases reported in the world literature to-date, which led the World Health Organization (WHO) to classify DLE as one of the precancerous conditions. Thus malignant changes in such a condition must be given utmost importance. This article focuses on a rare case of DLE with epithelial dysplasia.

Keywords: DLE, Epithelial dysplasia, Immunofluorescence, Potentially malignant disorders

INTRODUCTION

Discoid lupus erythematosus (DLE) is a chronic, scarring, immunologic, mucocutaneous disease, which is characterized by white keratinized plaques with elevated borders, radiating white striae and telangiectasia. The prevalence of DLE is less than 5 in 10,000 individuals and is more common in women than men with female to male ratio of 1.8:1. The frequency of oral involvement in DLE is lower than skin lesions, and is found in about 20% of cases. Malignant transformation of DLE into SCC is usually observed on the lower lip, and in Caucasians.

However, there are some case reports in African Americans. Liu et al reported 6.8% of lower lip SCCs arise from lesions of DLE. Ac-cording to Liu et al epithelial dysplasia and age over 60 years are risk factors for malignant transformation in DLE, whereas sex, location of the lesion, smoking and alcohol intake were not considered to be risk factors.[1] Topical or systemic corticosteroids are usually recommended for management of DLE. Thus, this article emphasizes on a rare case of "DLE with epithelial dysplasia".

CASE REPORT

A 60-year-old female patient reported to the department of Oral Medicine and Radiology of our institution with a chief complaint of bleeding wounds on the right side of the mouth and tongue with alternating periods of remission and exacerbation. History revealed she had the lesion since two months and was not under any medication. Lesion on the right side of buccal mucosa initially started as fluid filled blisters, which later burst to form an ulcer approximately measuring 3 x 3 cm in size (Figure 1). It was typically circular and erythematous with a whitish centre, surrounded by fine white, irradiating striae. Crusting was also observed on the angle of the mouth. Lesion on the left dorsum of tongue was measuring approximately 2 x 2 cm.

No relevant medical history of any sort of systemic illness and drug allergy was reported from the patient. Patient reported with a habit history of betel nut chewing 6-7 times daily for the past 35 years. On extra oral examination, the right and left submandibular lymph nodes were tender, palpable and hard in consistency. A

complete blood cell count showed mild anaemia (haematocrit of 33.8% and haemoglobin of 11.2gm%); Erythrocyte sedimentation rate (ESR) was 64mm/hr. On her first visit patient was advised to quit the habit. The patient was also put under medication of topical corticosteroid, vitamins and folic acid supplements and analgesics. On her subsequent visit after two weeks, following poor prognosis, incisional biopsy was done on the lesion of right buccal mucosa. Gross specimen received was greyish white in color, firm in consistency measuring about 1x 0.7x 0.4 cm in size.

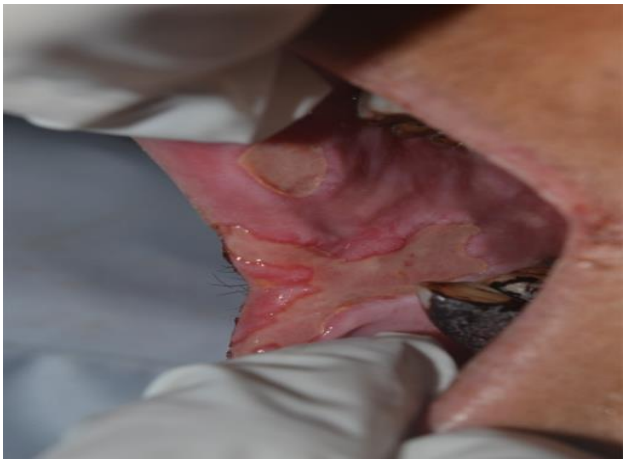

Figure 1: Photomicrograph showing lesion on the right side of buccal mucosa.

Histopathological examination revealed hyper parakeratinized stratified squamous epithelium exhibiting areas of atrophy, basal cell degeneration and acanthosis with connective tissue papillae extending upto the surface. Few areas of epithelium also showed dysplastic features and dystrophic keratinization (Figure 2).

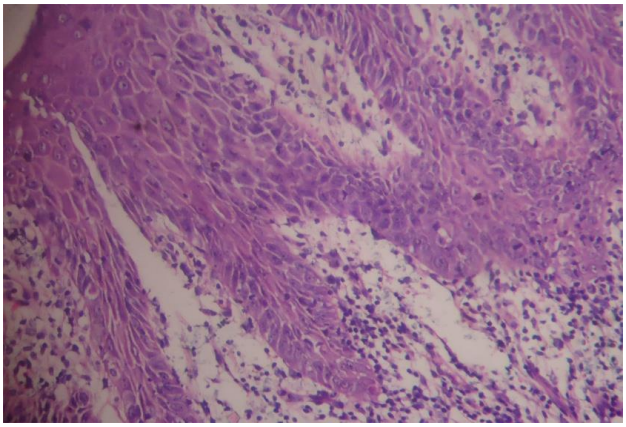

Figure 2: Photomicrograph demonstrating epithelium showing dysplastic features (H and E stain, 400 X).

Underlying connective tissue demonstrated subepithelial edema and loosely arranged collagen fibers with moderate infiltration of chronic inflammatory cells like lymphocytes and plasma cells distributed in a diffuse and perivascular pattern (Figure 3).

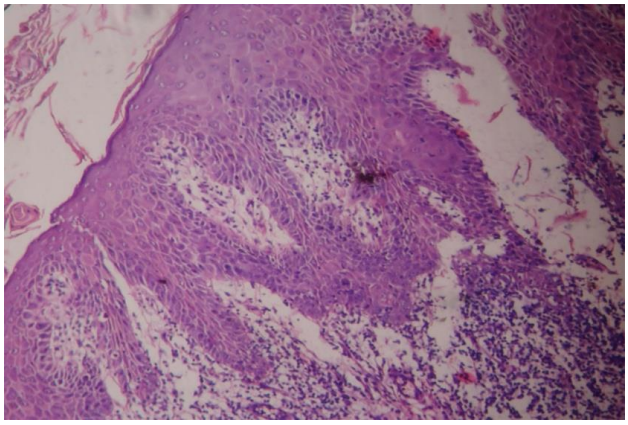

Figure 3: Photomicrograph demonstrating subepithelial edema and underlying connective tissue (H and E stain, 400X).

PAS staining revealed patchy PAS positive deposits in the epithelium-connective tissue interface (Figure 4). Based on clinical, histopathological and laboratory investigations a diagnosis of discoid lupus erythematosus with epithelial dysplasia was reported.

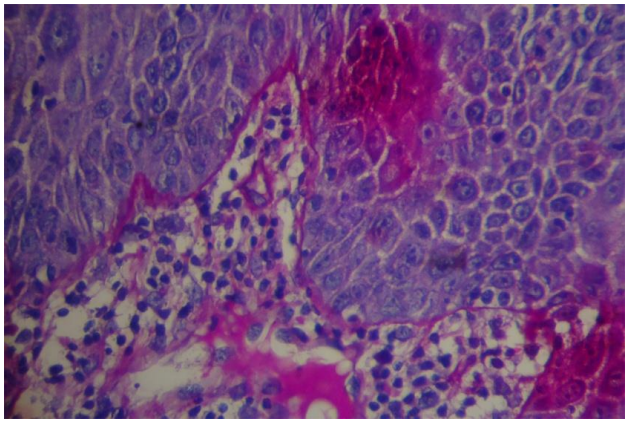

Figure 4: Photomicrograph demonstrating patchy PAS positive staining in epithelium connective tissue interface (PAS stain, 400X).

DISCUSSION

Clinically and histopathologically it is difficult to discern the discoid lesions from others. The classical discoid lupus erythematosus generally shows a central erythematous area, ulcerated or atrophic, surrounded by fine white, irradiating striae.[2] similar feature seen in case of classic Lichen planus but in lupus erythematosus, the irradiated striae are more delicate than the Wickham striae of lichen planus.

The present case showed, Discoid lupus lesions were localized to right buccal mucosa and dorsum of the tongue. Although clinical examination showed the lesions were compatible with lupus erythematosus, the diagnosis was confirmed with an incisional biopsy with histopathological examination by haematoxylin-eosin

staining (HE). In a histopathological study by Gerald Shkler and Philip L. McCarthy on 25 oral biopsy specimens of lesions of chronic discoid lupus erythematosus a characteristic and diagnostic histopathological pattern was confirmed, which consisted of parakeratosis, hydropic degeneration of stratum germinativum, collagen degeneration, and a lymphocytic infiltration in a perivascular orientation.[3]

PAS staining for the mucopolysaccharide appears to be of great value in diagnosing the oral lesions of chronic lupus erythematosus. The characteristic finding is an extremely intense reaction beneath the stratum germinativum of the epithelium and around small blood vessels. WHO (1978) reported PAS-positive deposits juxtaepithelially, which resembles a thickening of the basement membrane.[4] PAS Staining in the above reported case also demonstrated thickened basement membrane.

In a retrospective hospital based study of an ethnic Chinese population by Liu et al, documented six (6.9%) patients with DLE of lip and oral cavity related malignant transformation.[1] Out of six cases, one case with DLE being reported in a female patient of age 56 years with a high risk of epithelial dysplasia on tongue showed malignant transformation in a latent period of 6 months to stage II squamous cell carcinoma. The sites of malignant transformed lesions were located at the same sites as the previously biopsied DLE lesions. According to the proposal by the WHO, Liu et al found that high-risk dysplasia was associated with a 14.24-fold increased risk of malignant transformation after adjustment for age. These results support the view that epithelial dysplasia is a significant indicator for evaluating risk of malignant transformation in patients with DLE.[1]

The direct immunofluorescent (DIF) studies of oral DLE lesions in literature revealed three major classes of immunoglobulins IgA, IgM and IgG as well as different complement components found in the basement membrane zone in a linear and/or granular pattern. The most commonly identified are IgM and C3.[5,6] In a comparative study between DIF and histopathology by Shreekant et al on 75 cases with DLE diagnostic specificity and a positive predictive value of both methods were found to be maximal (100%).[7] No significant literature is available on the molecular events or biomarkers for the malignant transformation of DLE. However, study by Liu et al positively correlated expression of podoplanin with the progression of chronic discoid lupus erythematosus on lip to squamous cell carcinoma.[8]

CONCLUSION

DLE is a chronic scarring and potentially disfiguring disease seen in all parts of the world and among all ethnic groups. Conventional clinical and histopathological (presence or absence of epithelial dysplasia) characteristics are still the most important parameters for the prediction of malignant transformation in oral potentially malignant disorders in routine diagnostic pathology. Basal cell and more commonly squamous cell carcinoma have been reported in literature to develop in DLE lesions. Thus all the patients reported with DLE must be given utmost importance and routine follow up is mandatory.

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