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LETTER TO THE EDITOR

REPORT OF A CASE OF DISCOID LUPUS ERYTHEMATOSUS LOCALISED TO THE ORAL CAVITY: IMMUNOFLUORESCENCE FINDINGS

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Discoid Lupus Erythematosus (DLE) is a chronic disease with a typical cutaneous involvement. This pathology rarely involves mucosa: oral cavity is interested in 20% of DLE patients. We describe a case of oral DLE in a 50-year-old woman with an anamnesis for autoimmune disorders. This study shows the helpful role of immunofluorescence in the diagnosis of autoimmune diseases. The first diagnostic step was the clinical observation of the oral mucosa: the lesion area was erythematous, athrophic and hyperkeratotic. The patient then underwent laboratory examination. We utilized human epithelial cells (Hep-2010) for Indirect Immuno-Fluorescence (IIF). Moreover, the biopsy site for Direct Immuno-Fluorescence (DIF) and histopathological analysis was the untreated oral lesion. IIF detected an increase of Anti-Nuclear Antibody (ANA) and positivity for SSA-RO. By DIF, we observed IgG/IgA/fibrinogen along basal layer. Multiple biopsies reported signs of chronic basal damage. Steroid systemic therapy induced a considerable lesion regression. We suggest the use of immunofluorescence with the integration of further data to improve diagnosis of rare diseases and to establish a suitable therapy.

Discoid Lupus Erythematosus (DLE) is an autoimmune chronic cutaneous and mucosal disease, with a variety of clinical manifestations, commonly subdivided into localised and generalised forms. Only about 1% of patients with localised DLE develops Systemic Lupus Erythematosus (SLE), whereas, approximately 5% of those with generalised DLE evolves to systemic disease. Data gathered over the last ten years show a mean incidence of around 15 new cases per annum. Females are more often affected. DLE is most diffused in the third to fifth decades. Generally, lesions arise on sun cutaneous exposed sites, but DLE can also affect the mucosal membranes. An oral involvement occurs in 20-25% of patients (1).

We describe an oral case of DLE and discuss the need of comparing clinical, laboratory and histological reports to improve diagnostic and therapeutic patient management.

Our patient is a 50-year-old woman with a familiar anamnesis for autoimmune disorders. She referred a clinical history of idiosyncrasy to Non-Steroid Anti-Inflammatory Drugs (NSAID) and autoimmune hyperthyroidism treated by partial thyroidectomy. She complained of swollen gums and a moderate oral burning, stressed during mastication. The lesion area was erythematous, atrophic and hyperkeratotic. We utilized human epithelial cells (Hep-2010) for

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Indirect Immuno-Fluorescence (IIF) (2). Direct Immuno-Fluorescence (DIF) was performed on a sample of the untreated oral lesion in order to confirm the initial diagnosis of DLE.

The clinical observation of the oral mucosa showed a characteristic lesion associated to a diffuse marginal gingivitis. The plaque involved the gengival, palatal and cheek mucosa, with a symmetrical and bilateral appearance. Centrally, the lesion was speckled, erythematous and atrophic, with a scalloped white keratotic peripheral border and adjacent teleangectasia. No systemic symptoms, and only erythrosedimentation Rate (ESR) slightly above normal were present.

The active lesion was characterized by hyperkeratosis and parakeratosis; the mucosal membrane was slightly atrophic and presented, at the basal layer, a basement membrane thickening, a dermo-epidermic worm-hole, a liquefactive degeneration of basal keratinocyte (cytoid body formation) and a dense hexocitotic or perivascular lymphohistiocytic infiltrate.

The IIF showed a moderate increase of Anti-Nuclear Antibody (ANA) in two types of patterns (one dotted coarse granular, the other more homogeneous); the anti-nuclear ribonucleoprotein (SS-A-Ro 52kDa) also displayed an intense positivity. All the other typical SLE antibodies (anti-double stranded DNA; anti-histone; anti-ribonuclear protein; anti-small nuclear ribonucleoprotein or Smith's antigen) were lacking. Patient serum did not contain anti U1-RNP.

The DIF showed a weak linear and discontinuous positivity for IgG, moderate for IgA, high and more marked for fibrinogen (thick band fluorescence) along the basal layer (3).

After ten days therapy with prednisone (60 mg daily), we detected a significant phlogosis regression, an evident reduction of oral lesions, near disappearance of the erythema, atrophy and hyperkeratosis. The patient reported the clearing up of other symptoms and a less intense burning sensation.

The histopathological profile of DLE is very evocative (4), but the lesions frequently may have atypical features which can make the differentiation with lichen planus difficult.

For these reasons, there is a tremendous expansion in the use of laboratory services in rheumatology over the past 10 years (5-6). Rheumatological practice relies heavily on laboratory investigations (particularly immuno-fluorescence tests) (7). These tests cover the use and interpretation of laboratory investigations employed in DLE and highlight recent developments. Naturally laboratory personnel, hospital clinicians and primary care physicians must have a thorough understanding that a correct diagnosis must be founded on the precise comparison and integration of the all data and not on the isolated valuation of the different findings. In our case, in which even the specific autoimmune serologic pattern was lacking, immunofluorescence was necessary to establish the definitive diagnosis. It confirmed a diagnostic hypothesis, differentiated and distinguished among closely related diseases (8). An accurate and complete diagnostic protocol is the first important step in deciding a suitable and effective therapy and to set up the successful management of patients (9).

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REFERENCES

1. **Vitali C., A. Doria, A. Tincani and P. Fabbri.** 1996. International survey on the management of patients with SLE. I. General data on the participating centers and the results of a questionnaire regarding mucocutaneous involvement. *Cl. Exp. Rheumatol. 14(S):17.*
2. **Ishikawa O., K.K. Zaw, Y. Miyachi, T. Hashimoto and T. Tanaka.** 1997. The presence of anti-basement membrane zone antibodies in the sera of patient with non-bullous lupus erythematosus. *Br. J. Dermatol. 136:222.*
3. **Kulthanan K., P. Roongphiboolsopit, S. Chanjanakijskul and P. Kullavanijaya.** 1996. Chronic discoid lupus erythematosus in Thailand: direct immunofluorescence study. *Int. J. Derm. 35:*

711.

4. **Shahidullah M., Y.S. Lee and C.J. Khor.** 1995. Chronic cutaneous lupus erythematosus. An immunopathological and microscopic study. *Ann. Acad. Med. Singapore 24:789.*
5. **Togliatto M., M. Carrozzo, D. Conrotto, M. Pagano and S. Gandolfo.** 2000. Oral lupus erythematosus. Description and analysis of 11 cases. *Minerva Stomatol. 49:35.*
6. **Jonsson R., G. Heyden, N.G. Westberg and G. Nyberg.** 1984. Oral mucosal lesions in systemic lupus erythematosus - a clinical, histopathological and immunopathological study. *J. Rheumatol. 11: 38.*
7. **Schiodt M., P. Halberg and B. Hentzer.** 1978. A clinical study of 32 patients with oral discoid lupus erythematosus. *Int. J. Oral Surg. 7:85.*
8. **Dahl M.V.** 1983. Usefulness of direct immunofluorescence in patients with lupus erythematosus. *Arch. Derm. 119:1010.*
9. **Mesa M., R. Kelner, F. Pecoraro and M. Deasy.** 1979. Oral discoid lupus erythematosus. A case report and review of the literature. *J. Periodontol. 50:90.*