Coexistence of Psoriasis, Vitiligo and Oral Lichen Planus – a New Variant of Multiple Autoimmune Syndrome?

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\textbf{ABSTRACT}

Multiple autoimmune syndrome is a rare condition, defined by the association of at least three autoimmune disorders in the same patient, and first described by Humbert and Dupond in 1988. Psoriasis, vitiligo, and oral lichen planus are all common dermatological disorders in the general population, and the coexistence of these three diseases in these exact clinical variants has never been reported in the literature before. Herein, we describe an unusual case of co-occurrence of psoriasis vulgaris, vitiligo, and oral lichen planus. A 67-year-old Romanian female patient with a history of both vitiligo and psoriasis presented with white, shiny striae, organized in a reticular pattern on the mucosa of the cheek and whitish homogeneous irregularities similar to leukoplakia involving the dorsal side of the tongue. Histology examination confirmed oral lichen planus. The rarity of this case is highlighted by the coexistence of psoriasis, vitiligo, and oral lichen planus in the absence of cutaneous lichen planus lesions, an association which may be a newly described variant of multiple autoimmune syndrome.

\textbf{Keywords}: vitiligo, lichen planus, oral, psoriasis, hypopigmentation, female, autoimmunity.

\textbf{INTRODUCTION}

Psoriasis, oral lichen planus, and vitiligo are all common dermatological disorders in the general population. Coexistence of these three diseases, in these clinical variants, has never been reported in the literature, although an immunological link between them has been previously suggested (1-3). Psoriasis is a common, chronic inflammatory skin disease characterized by T-cell mediated hyperproliferation of keratinocytes. Despite the advances in the last decades, the pathogenesis of
psoriasis is still not fully understood but current data indicates that human leukocyte antigen Cw0602 is associated with an increased susceptibility for psoriasis (4). Two cytokines, interleukin (IL)-23 and IL-22, have been found to play a crucial role in psoriasis pathogenesis (5).

Vitiligo is an acquired disorder of pigmentation due to a substantial loss of functioning epidermal and/or hair follicle melanocytes. Recent research has shown that IL-17 secreting cells induced by IL-1β are present in increased numbers in the serum of vitiligo patients, thus inducing a proinflammatory state (6). In the meantime, the number of regulatory T cells is reduced and their ability to suppress CD8+ cells is diminished (7).

Oral lichen planus (OLP) is a chronic, T cell-mediated autoimmune disease, in which autocytotoxic CD8+ T cells attack the basal layer. Cytokines such as IFN-gamma, TNF-alpha, IL-1-alpha, IL-6 and IL-22 associated with T helper cell response may play a role in the pathogenesis of lichen planus (8, 9).

It has been suggested that a combination of genetic susceptibility and environmental factors, which underlie most autoimmune conditions, explains the higher possibility of multiple autoimmune diseases appearing in one patient. Lopez-Jornet et al. have shown a higher incidence of other autoimmune diseases in patients with OLP when compared with patients without OLP (7% vs 4%), thus suggesting a higher chance for these patients of associating vitiligo or psoriasis compared to the rest of the population (10).

**CASE REPORT**

A 67-year-old woman presented with an 18 year history of hypopigmented skin patches on the extremities, trunk, and face (Figure 1A), which had already been diagnosed as vitiligo after clinical examination, Wood lamp examination, and skin biopsy. About two years prior to presentation and sixteen years after the onset of vitiligo, she developed scaly erythematous plaques on the elbows, dorsum of the hands, and scalp (Figure 1A). A skin biopsy from one of these plaques revealed epidermal acanthosis with hyperkeratosis, parakeratosis, and infiltrations of neutrophils into the stratum corneum and subcorneal zone, compatible with the clinical diagnosis of psoriasis vulgaris (PV) (Figure 2A). Note that there was co-localization between psoriatic plaques and vitiliginous patches.

One year after the onset of psoriasis, she developed white, shiny striae, disposed in a reticular pattern, localised on the mucosa of the cheek (Figure 1C-D), and whitish homogeneously distributed irregularities, similar to leukoplakia, involving the dorsal side of the tongue (Figure 1B). Oral cavity examination revealed that she had dental fillings. A biopsy was taken from the lesions on the oral mucosa, which histologically showed hyperkeratosis with irregular acanthosis and focal thickening in the granular layer, Civatte bodies in the lower epidermis, a band-like inflammatory infiltrate composed of histiocytes and lymphocytes, mainly T cells, localised in the superficial dermis (Figure 2B), thus confirming the diagnosis of oral lichen planus. There was no
evidence of LP elsewhere, such as the skin, genitalia, nails, or scalp.

Routine laboratory tests were all unremarkable. No finding suggestive of any type of connective tissue or rheumatic disease was evident.

On the basis of the clinical and histological findings, the diagnoses of psoriasis vulgaris, vitiligo and oral lichen planus were established and she was started on systemic antioxidant therapy, topical steroids for psoriasis lesions and intrabuc- 
cal steroids for the OLP lesions, with a follow-up visit scheduled for a month later.

**DISCUSSION**

Coexistence of psoriasis, vitiligo, and OLP and has never been reported in the literature before. There is one report in which psoriasis, vitiligo, and cutaneous lichen planus occur together (2). However, there are more frequent reports about the association between pairs of these three immune disorders.

The association of vitiligo and lichen planus has been previously described (11, 12). Most cases associated vitiligo with the cutaneous form of lichen planus (LP), with a great majority of patients having LP lesions superimposed on vitiliginous areas (13). Lesion distribution was particular in our case due to the association of vitiligo with OLP, in the absence of cutaneous LP lesions. The common pathogenic way of LP and vitiligo could be explained by the autoimmune destruction of basal keratinocytes leading to a cross-reaction with antigens located on melanocytes.

Even though the association between vitiligo and psoriasis was first described in 1890 (14), the relationship between the two diseases is still subject to discussions. Some reported cases describe the two dermatoses occurring together with strict co-localization (15-17), while others deny strict co-localization (3). Some authors (18, 19) conclude that psoriasis lesions have the same occurring rate on both vitiliginous lesions and on normal skin. In our case there was, although not strict, a co-localization between psoriasis and vitiligo lesions. Autoimmunity, common neuropeptides, and Koebner’s phenomenon have all been called to explain the pathogenic link between the two disorders. The concurrence of vitiligo and psoriasis highlights the link between the pathogeneses of the two conditions. In vitro studies have confirmed that patients with vitiligo have enhanced neurotensin, which in turn leads to a melanocytic production of TNF-alpha (20) and Jain et al. showed that patients with vitiligo present elevated levels of TNF-alpha in peri-viti- 

**CONCLUSION**

In conclusion, we report a case of coexistence of three immune dermatoses in a single patient by the common etiological background of autoim- munity and Koebner’s phenomenon. The rarity of this case is highlighted by the coexistence of psoriasis, vitiligo, and oral lichen planus in the absence of cutaneous lichen planus lesions, an
association which, to the best of our knowledge, may be a newly described variant of multiple autoimmune syndrome. A combination of these diseases in one patient is a unique situation and, at the same time, a much greater therapeutic challenge in comparison to the each condition occurring separately or in pairs.

Conflicts of interest: none declared. 
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References