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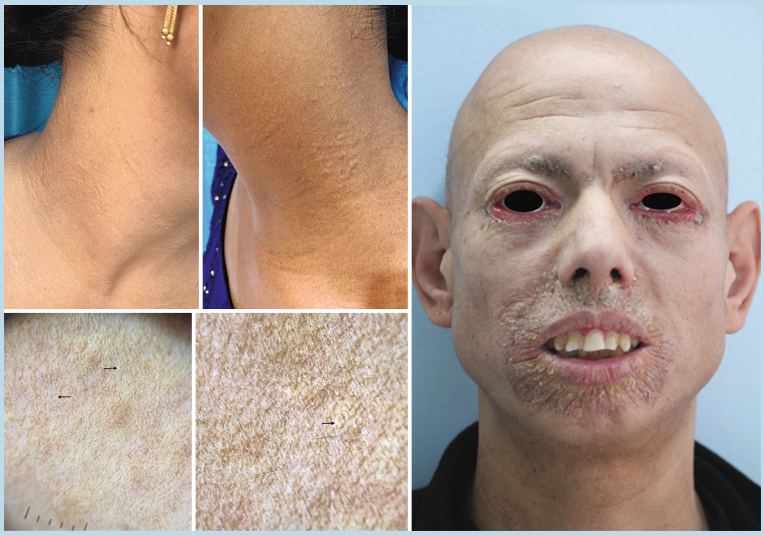

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Skin Sarcoidosis on Both Vitiligo and Tattoos in the Same Patient: Report of an Exceptional Case and Pathogenetic Hypotheses of Intriguing Associations

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Sir,

A 48-year-old man presented with an itchy, brownish-red, squamous plaque in his pubic region, surrounded by a previously appeared hypochromic halo [Figure 1a]. On dermoscopy, yellowish coalescing patches with homogenously distributed linear vessels were observed [Figure 2a]. On total skin examination, small erythematous papules on tattoos [Figure 1b and c] and hypo-pigmented macules on his fingers were found. Under Wood's light, these macules, as well as the hypochromic halo, showed a whitish fluorescence consistent with vitiligo [Figure 1d]. Histopathology of the pubic plaque and one tattoo papule demonstrated naked noncaseating granulomas composed by epithelioid histiocytes and multinucleated giant cells bordered by a moderate chronic inflammatory infiltrate in the papillary dermis [Figure 2b]. A reduction in melanin and in melanocytes number, demonstrated by immunostain for mediastinal lymphadenopathy (MLA), was seen at the dermal-epidermal junction in the pubic sample.

Suspecting skin sarcoidosis, blood tests were performed showing only an increase in angiotensin converting enzyme levels (145 U/L). Chest X-ray and quantiferon tests were negative. Chest CT showed peribronchial cuffing and millimetric consolidations at hilum and subpleural areas of the upper-medium pulmonary fields [Figure 3a] in association with hilar [Figure 3b] and MLA[Figure 3c], consistent with the hypothesis of sarcoidosis.

Diagnosis of sarcoidosis is based on clinical and radiological signs and histopathology because of the absence of definitive diagnostic tests. Its etiopathogenesis is still unclear, but a dysregulation in the immune response to undefined antigens is supposed. Tatoos may be the antigenic trigger of the cutaneous manifestations due to a chronic antigenic stimulation in predisposed subjects.[1] Other theories consider tattoos as the target of sarcoidosis because the tissue alterations, namely the scarring, create an "immunocompromised district," which is the loss of normal network of immune control. This may lead to an exaggerated local reaction to exogenous or auto-antigenic stimulation. The immune dysregulation may also be linked to the alteration of the lymphatic drainage, which compromises the transition of immune cells from the skin to the lymph nodes, and to the disruption of the neurologic innervation with subsequent alteration in the secretion of neuromodulators. The theory of the "immunocompromised district" seems to underlie or contribute to Köbner and Rebnok phenomena.[2]

In the literature, cases of association between sarcoidosis and vitiligo are described and generally explained by a common autoimmune diathesis. The two diseases appear to share the same pathogenetic background, that is the activation of interferon-γ (IFN-γ) signaling that recruits JAK1-STAT1 pathway. In sarcoidosis, IFN-γ drives macrophage activation in granulomas[3] whereas in vitiligo this leads to the transcription of chemokines as CXCL9 and CXCL10, which are involved in recruitment and homing of cytotoxic T-cells responsible for the damage of melanocytes.[4,5]

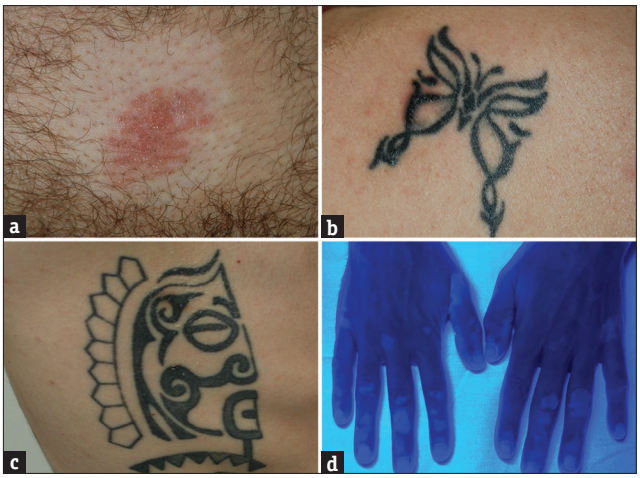

Figure 1: (a) Clinical presentation of the plaque surrounded by the hypochromic halo in the pubic region (b and c) Clinical presentation of the erythematous papules on tattoos (d) Whitish fluorescence of the hypo-pigmented macules on fingers under Wood's light

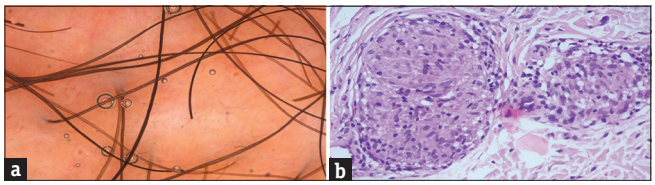

Figure 2: (a) Dermoscopy of the plaque showing yellowish coalescing patches with homogenously distributed linear vessels (b) Histopathology showing naked noncaseating granulomas composed by epithelioid histiocytes and multinucleated giant cells bordered by a moderate chronic inflammatory infiltrate in the papillary dermis

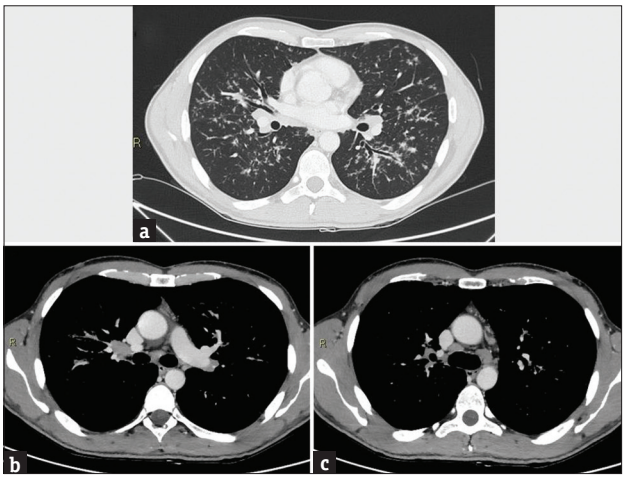

Figure 3: (a) Radiological findings of peribronchial cuffing and millimetric consolidations at hilum and subpleural areas of the upper-medium pulmonary fields (b) Radiological findings of hilar lymphadenopathy (c) Radiological findings of mediastinal lymphadenopathy

The efficacy of tofacitinib, a JAK1-JAK3 inhibitor, in both diseases, seems to support a common, pathogenetic mechanism.

In our case, skin sarcoidosis is located within the vitiliginous patch, which had been present for several years, and within the tattoos. It may be hypothesized that the hyper-expression of IFN-γ in vitiligo and the local tissue modifications due to tattooing contributed to the formation of the sarcoidotic plaque in the presence of a genetic predisposition.

Our presentation offers the opportunity to consider the possible regional immunological and tissue modifications that promote skin hyper-reactivity. The peculiarity of our case consists in the coexistence of skin sarcoidosis within both a vitiligo patch and tattoos, which is a condition never previously described, to our knowledge.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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