Strict Anatomical Colocalization of Vitiligo and Elastolytic Granulomas

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Key Words
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Abstract
Vitiligo is the most common depigmenting disorder, with a worldwide occurrence of 0.1–2% in the general population. Multiple conditions have been described colocalized in vitiligo patches, like psoriasis or lichen planus. However, actinic granuloma has not been described in association with vitiligo lesions so far.

Introduction
Vitiligo is the most common depigmenting disorder, with a worldwide occurrence of 0.1–2% in general population [1]. Multiple conditions have been described colocalized in vitiligo patches, like psoriasis or lichen planus [2–5]. However, actinic granuloma has not been described in association with vitiligo lesions so far.

Actinic granuloma was first described by O’Brien in 1975 as an uncommon disease, most frequently presented in middle-aged women [6, 7]. Its classification and pathogenesis is controversial. Some authors consider actinic granuloma a subtype of annular granuloma and others a different entity [6–9]. The pathogenesis of this condition is still uncertain [7–9].

Case Report
We report the case of a 50-year-old woman, who was referred to our Dermatology Department in 2006 with a diagnosis of lip-tip vitiligo. The milky patches were located in the face, on the dorsum of the hands, and on the forearms and legs. Her past medical history was only remarkable for hyperthyroidism
diagnosed 1 year ago. There was no other family history of dermatologic or endocrine problems. No
diagnosis of lupus, sarcoidosis or other dermatologic diseases could be elucidated in her medical or
family history. She was treated with topical steroids (Clovate® cream) and topical tacrolimus (Protopic®
0.1% ointment), with poor response in achral lesions. Oral PUVA therapy with 8-MOP was started in
2007, obtaining a moderate repigmentation in distal areas. Treatment was given 3 times weekly for a
maximum of 20 weeks. In June 2009, during a routine follow-up, the patient complained that
non-pruriginous lesions had appeared in her hands 2 weeks ago. On physical examination, she
presented with multiple asymptomatic, erythematous, annular plaques and solitary papules 5–10 mm in
diameter, located on the dorsum of both hands, strictly confined to vitiliginous lesions. No other lesions
in different areas could be observed. Full blood count, liver and renal function and urine analysis were
all in normal ranges. Based on the clinical picture, presumptive diagnosis of coincident vitiligo and
annular granuloma was made. A skin biopsy was performed.

Histological examination revealed mono- and multinucleated histiocytes and inflammatory
infiltrates in the dermis. Deformed and fragmented elastic fibers in the dermis were also revealed using
Orcein staining. Fibers phagocyted by the histiocytes were seen, too. Besides, absence of melanocytes in
Fontana-Masson staining was demonstrated. These histological findings confirmed the coexistence of
vitiligo and elastolytic or actinic cutaneous granuloma.

Conclusion

Several articles have suggested that annular and actinic granuloma are different
conditions based on histopathological characteristics [8]. Different terms have been used
to designate this disorder: annular elastolytic giant-cell granuloma (AEGCG) [9], O’Brien
granuloma [7], multiple [10, 11], actinic [7, 8] or elastolytic [12] granuloma. Some articles
suggest that actinic granuloma is a subtype of AEGCG [12]. The term ‘multiple
granuloma’ was coined by Leiker in 1964, and it is clinically similar to actinic granuloma,
being considered the same entity in some studies. Nevertheless, multiple granuloma
presents a distinctive histological finding of focal necrobiosis. A photo-mediated etiology
has been proposed in the literature for this subtype [10, 11].

The etiology of actinic granuloma remains obscure. However, several theories have
been postulated. O’Brien suggested the role of infrared and ultraviolet actinic radiation [6,
7, 13]. The importance of a granulomatous chronic inflammation in association with
another systemic illness such as sarcoidosis or diabetes mellitus was proposed by Hanke
[9].

The actinic damage hypothesis is based on the cutaneous cell-mediated immune
response. CD4 lymphocytes would act against the solar-damaged elastic fibers, which
could be recognized as antigenic products [6, 7, 13]. Histological characteristics like
sarcoidal granuloma or multinucleated giant cells suggest an inflammatory response to
small products, like altered elastic fibers. The confinement of the inflammatory reaction
to the superficial dermis, corresponding to the area of solar elastosis, supports the actinic
theory [8].

This photo-mediated damage could be considered as a trigger factor in our patient and
could explain the strict colocalization of elastolytic granuloma on vitiligo patches.
Vitiliginous skin is more photo-sensible and the actinic damage is more intense than in
the rest of the body [14]. In this case, the affected skin on the dorsum of the hands was
not only prone to photo-damage as a result of sun exposure, but it was also selectively
exposed to PUVA therapy over the last 2 years in our patient.

In this case, we present an uncommon clinical feature with strict colocalization of
elastolytic granuloma in vitiliginous areas. The location of the lesions strongly supports
the photo-damage hypothesis in our patient. Moreover, the patient received PUVA and sun therapy as an important part of her treatment.

To our knowledge, this is the first patient with coexistence and strict anatomical coincidence of actinic granuloma and vitiligo reported in the literature thus far. We conclude that the actinic radiation played an important role in this form of granuloma in our patient. However, further studies to clarify pathogenetic links between vitiligo and actinic granuloma would be necessary.

**Fig. 1.** Vitiliginous areas with coincident multiple small papules located on the dorsum and lateral aspect of both hands. The annular plaques only affect depigmented areas.
Fig. 2. Histological examination revealed mono- and multinucleated histiocytes and inflammatory infiltrates in the dermis. Deformed and fragmented elastic fibers in the dermis were also revealed (Orcein staining, 100x).
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