CASE REPORT

Antiretroviral-responsive confluent and reticulated papillomatosis: a case report of an unusual association

Rami A. Ballout¹,²,†, Gilbert Helou³ and Ismael Maatouk⁴,⁵,*,‡

¹Faculty of Medicine, American University of Beirut, Beirut, Lebanon, ²Lipoprotein Metabolism Section, Translational Vascular Medicine Branch, National Heart, Lung and Blood Institute (NHLBI), National Institutes of Health (NIH), Bethesda, MD, USA, ³Internal Medicine Department, Mount Lebanon Hospital, Beirut, Lebanon, ⁴Faculty of Health & Life Sciences, De Montfort University, Leicester LE1-9BH, UK, ⁵Dermatology Department, Clemenceau Medical Center affiliated with Johns Hopkins, Beirut, Lebanon

*Correspondence address. Tel: +9613568968; E-mail: ismael.maatouk@dmu.ac.uk

Abstract

This is the case of a 29-year-old male newly diagnosed with advanced HIV (CD4 < 35 cells/mm³), presenting to us with hyperpigmented and scaly non-pruritic macules over his chest and upper abdomen of several weeks duration. Woodlamp examination was negative, but a skin biopsy suggested confluent and reticulated papillomatosis (CRP). Given his lack of any of the condition’s identifiable triggers and the unusually rapid resolution of his lesions shortly after antiretroviral therapy initiation, an immunodeficiency-related etiology for his CRP was entertained. Autoimmune disorders and atopic conditions have been well reported previously as possible triggers of CRP. However, in this report, we raise immunodeficiency as a possible trigger of CRP as well, such that immune dysregulation overall (autoimmunity or immunodeficiency) can contribute to CRP ontogenesis. To our best knowledge, this is the first report to date suggesting a possible association between CRP, a rare dermatological condition, and acquired immunodeficiency syndrome.

INTRODUCTION

Confluent and reticulated papillomatosis (CRP) is a rare and acquired ichthyosiform skin disorder characterized by hyperpigmented, scaly and velvety reticular plaques over the neck, chest, trunk and/or upper back [1]. Its precise causes and underlying pathophysiology remain unknown and subjects of future discovery. However, multiple conditions have been documented to precede or coexist with its development, namely diabetes, obesity, ultraviolet light and/or radiation exposure, amyloidosis and fungal infections of Malassezia furfur, Dietzia spp. or actinomycetes [1, 2]. Oral minocycline remains the mainstay of treatment for CRP, with several newly emerging alternatives such as azithromycin, antifungals and/or systemic retinoids [1, 2].

CASE REPORT

A 29-year-old man presented to our clinic with new-onset, non-pruritic skin lesions that started 4 weeks ago as brown papules over his anterior neck and gradually coalesced into
larger plaques and spread caudally across his chest. Inspection revealed hyperpigmented and scaly plaques (Fig 1a).

The patient is otherwise healthy. His family history is non-revealing, and his BMI is 21 kg/m². He does not smoke and works indoors.

Before presenting to us, he saw an infectious disease specialist who initially diagnosed him with pityriasis versicolor, a great mimicker of CRP [2], and started him on oral and topical fluconazole for 3 weeks, after which he is to follow up with us. However, during that same visit, the patient was also diagnosed with acquired immunodeficiency syndrome (AIDS), given an extremely low CD4 count (35 cells/mm³) and positive HIV serology on his recently requested labs. The physician immediately started him on highly active antiretroviral therapy (HAART) and trimethoprim–sulfamethoxazole prophylaxis.

On follow-up 3 weeks later, the patient’s lesions appear to have persisted unchanged, suggesting treatment failure.

We thus opted for a skin biopsy after a wood’s lamp examination of his lesions was non-revealing. The biopsy’s histopathology report came back 2 days later indicating ‘basket-woven orthokeratosis, hypogranulosis and papillomatosis within the epidermis, and sparse perivascular mononuclear cell infiltrates in the upper dermis with negative PAS fungal stain’, findings that are consistent with CRP.

However, given his lack of any recognizable CRP-predisposing factors and non-responsiveness to antifungal therapy, we decided against prescribing any further antimicrobial therapies, but instead test whether his occult immunodeficiency status could have had any role in his CRP ontogenesis. Intriguingly, the patient returned 5 weeks after HAART initiation, exhibiting near-complete resolution of his lesions (Fig 1b). At 8 weeks post-therapy initiation, his CD4 count had reached 195 cells/mm³.

**DISCUSSION**

The finding that our patient’s lesions did not respond to antifungal therapy yet resolved completely after 5 weeks only of HAART initiation, without co-administering any antibiotics (e.g. minocycline), suggests a possible role for HAART therapy in treating his lesions. Moreover, his lack of ‘typical’ CRP-associated conditions (diabetes, obesity, family history of ichthyosis, etc.) and negative fungal stain further support a non-conventional etiology of his lesions. It is worth mentioning that few cases of spontaneous resolution of CRP have been reported to date within the literature [3, 4]. However, the time for resolution in these cases was no less than 12 months since onset, as compared to only 5 weeks in our patient. This observation in particular is what prompted us to consider a possible role of the antiretroviral therapy in the very rapid resolution of CRP in our patient [3, 4].

Based on our extensive literature search, this report appears to be the first published to date, suggesting an association between immunodeficiency and CRP development.

This is not completely surprising, however, given that immune dysregulation has been redundantly reported for over two decades as a possible trigger of CRP; Kagi et al. associated their case of CRP with atopy (5), while Tirado-Sanchez et al. treated their patient with tacrolimus, an immunosuppressive agent [6].

In fact, several subtypes of acquired ichthyosis have been previously reported to associate with an immunodeficient status [7, 8]. As such, it is not completely unusual for CRP, an acquired ichthyosiform disorder, to be associated with immunodeficiency.

Nonetheless, being the sole case reported to date suggesting a possible association of HIV-related immunodeficiency and CRP development, we are certainly unable to establish causality or conclusively confirm an HIV-induced etiology for our patient’s lesions at this time, and our statements remain mere speculations.

**ACKNOWLEDGEMENTS**

None.

**CONFLICT OF INTEREST STATEMENT**

No conflicts of interest.

**FUNDING**

This research did not receive any specific grant from funding agencies in the public, commercial or not-for-profit sectors.

**CONSENT FOR PUBLICATION**

R.B. and I.M. obtained written informed consent from the patient described in this report to have his picture taken and his case published.

**AUTHORS’ CONTRIBUTIONS**

G.H. is the infectious disease specialist who interviewed and examined the patient first and attempted to treat him for a suspected fungal infection, before referring him to us. R.B. was the 4th-year medical student shadowing I.M. in clinic when the patient presented first for thorough dermatological evaluation. R.B. and I.M. obtained full history from the patient and performed his thorough physical examination. I.M. obtained the skin biopsy and performed the wood’s lamp examination. R.B. and I.M. followed up with the patient on his skin lesions after 5 weeks and, after discussing with G.H., agreed that the patient’s CRP was likely related to his immunodeficiency. R.B. and I.M. then obtained written consent from their patient to have his case reported, and R.B. drafted the entire manuscript which was subsequently revised and approved by all authors.

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