Unilateral Nevoid Acanthosis Nigricans on the Back: A Case Report

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Abstract- Unilateral Nevoid Acanthosis Nigricans (UNAN) is a rare, benign, autosomal dominant form of Acanthosis Nigricans (AN), which is a disorder of keratinization. Unlike AN, the lesions of UNAN are distributed unilaterally while the histopathological findings are similar to the common form of AN. The first description of UNAN was given by Krishnaram in 1991, and following that report, very few cases have been reported. Herein, we present another case of UNAN localized over the back, in a young adult female.

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Keywords: Acanthosis nigricans; Unilateral nevoid acanthosis nigricans; Rare subtype; Nevoid acanthosis

Introduction

Acanthosis Nigricans (AN) typically presents as symmetric, brownish-black, velvety, hypertrophic, verrucous, and at times, papillomatous plaques most commonly involving the flexural sites (1). It is a cutaneous marker, most frequently of insulin resistance and less frequently of malignancy (2). AN has been classified into 8 types: benign, obesity-related, syndromic, malignant, unilateral, acral, drug-induced, and mixed AN which is a combination of one or more of the above (1). Nevoid AN is a variant characterized by the unilateral distribution of morphologically and histopathologically diagnosable AN without any other accompaniment of common AN. It manifests at any age at or before puberty (3). The described case is of nevoid AN, localized over the back.

Case Report

A 19-year-old female presented with asymptomatic, pigmented and hyperkeratotic plaques over her upper back. The lesions appeared since the age of 18; they were initially macular and gradually evolved to become elevated and spread to the present size over a 1-year period. There was no history of rubbing the area. The patient was the second-born among three children born to non-consanguineous parents, and she did not have any other member of her family affected with similar lesions. She was non-obese and had no past or concurrent history of diabetes mellitus, drug intake, or other associated systemic complaints.

Dermatological examination revealed 3 well defined coalescing plaques with soft ridges. The lesions were barely elevated, and light brown at the peripheries and the centers contained elevated dark brown plaques. They were distributed over a broad area of the right scapular region, respected the vertical midline, measuring approximately 10×20 cm in overall (Figure 1 and 2). The skin was otherwise normal. The nape of the neck, axillae, waistline, and buttocks were free from any skin lesions. Palms and soles, mucous membranes, genitals, hair, and nails were normal. Systemic examination was noncontributory. Complete blood cell, plasma glucose level, Fasting serum insulin, thyroid function test, liver, and renal function tests were within normal limits. Human Immunodeficiency virus (HIV) and VDRL tests were negative, and malignancies were ruled out by the systemic evaluation.

Histological examination revealed hyperkeratosis, papillomatosis, and mild Acanthosis (Figure 3). Based on clinical and histopathological findings, a diagnosis of UNAN was made.

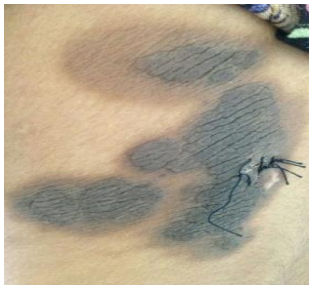

Figure 1. Coalescing pigmented plaques over the scapular region

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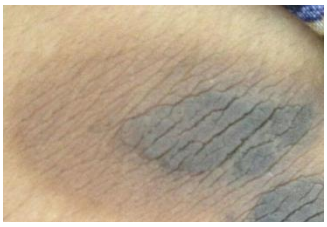

Figure 2. A close up picture of the plaque

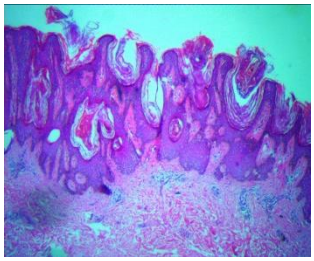

Figure 3. Histopathology of skin tissue shows the papillomatosis resulted from the upward projection of finger-like dermal papillae, which are covered by thinned epidermis. In the valleys between these papillary projections, the epithelium shows mild acanthosis with overlying hyperkeratosis. There is some hyperpigmentation of the basal layer

Discussion

AN derives from the Greek roots, acantho, meaning ”thorn,” and nigricans,”black.” It is often associated with an underlying systemic disorder, such as insulin resistance, or a malignancy. In contrast, UNAN had not been found to be associated with endocrinopathies, malignancy, drugs, or syndromes (4). The exact underlying pathologic mechanism in the development of UNAN remains speculative. It has been postulated to be inherited irregularly as an autosomal dominant trait, and the mosaic expression of activated growth factor receptors in keratinocytes has been hypothesized to play a role in its pathogenesis (5-6). The first description of UNAN was given by Krishnaram in 1991 (3) and later, Schwartz (7) was the first to include this entity in the classification of AN in 1994. According to a recent classification in a textbook, UNAN has been grouped together with “syndromic AN” and “acral acanthotic anomaly” under the heading “other” causes of AN (8). The natural history of the disease is a short period of activity for 4-5 years at the outset, following which stability without any tendency to resolution is the rule. Familial involvement in UNAN is not present, as found on an extensive literature search (9). Unlike the classical symmetrical AN that normally involves the flexural areas, UNAN does not have a predilection for intertriginous areas, as revealed from the available case reports in the literature (3). Most of the cases respected the midline with the distribution being essentially unilateral as it has been observed in our case.

Clinical differentials include ichthyosis hystrix, confluent and reticulate papillomatosis, and hyperkeratotic type of seborrheic keratosis (9). The closest differential diagnosis is epidermal nevus, but acanthosis nigricans lesions are usually more velvety and histologically show mild compact hyperkeratosis, papillomatosis, and limited acanthosis as compared to epidermal nevi (4). In our case, late age of onset and the presence of minimal acanthosis excluded epidermal nevus. Absence of horn cysts and basaloid cells ruled out seborrheic keratosis. In addition, other close mimickers were excluded due to the absence of their characteristic findings.

The affected individuals often seek treatment for cosmetic reasons. Various treatments have been described, including retinoids, calcipotriol, fish oil, ammonium lactate cream, cryotherapy, dermabrasion, an excision (if small lesion) and long-pulse alexandrite laser treatment, with variable results (3).

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