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BECKER'S NEVUS AND IPSILATERAL ACANTHOSIS NIGRICANS

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Abstract
There is paucity in world literature regarding the simultaneous occurrence of Becker's nevus and ipsilateral acanthosis nigricans in the same individual. There is only case reported previously in world literature. We speculate that our case may further strengthen the view of probable, more than a chance, association of these two entities and suggest need for further exploration of the role of androgen receptors in such cases.

Key words: Beckers nevus; ipsilateral acanthosis nigricans; hypoplasia of breast

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Introduction

The case of ipsilateral Beckers nevus and acanthosis nigricans is presented here for the extreme rarity of a distinct morphological presentation of two different pigmentary disorders in the same patient. Various theories have been proposed for the simultaneous co-existance of the same with hormone dependant theory being the most plausible explanation.

Case Report

A 15-years-old healthy boy presented with asymptomatic gradually progressing brownish-black pigmentation on left side of anterior chest since last 4 years. On examination, a well-defined, large hyperpigmented macule with irregular borders starting from the midline on the left side involving mammary area, extending superiorly upto left infra-clavicular area and anterior left shoulder and also seen involving the antero-medial side of left arm. There was slight extension even on the right parasternal area. There was coarse hypertrichosis in the sternal area as well as acneiform lesions present within the pigmented area (Fig. 1). This clinically was clearly a Becker's nevus. On more careful examination, he interestingly, had an ipsilateral smooth velvety irregular hyperkeratotic plaque with skin tags in the left axillary region. The right side of axilla was normal. The hair in left axilla were much enlarged, pigmented and coarser than the ones in the right axilla. The pigmented lesion in left axilla was clinically suggestive of acanthosis nigricans (Fig. 2). Additional findings noted on clinical examination were hypoplasia of only areola of ipsilateral breast, with scoliosis in the lumbar region and interestingly, a marginal elongation and slight overcurvature of the nail of his left index finger (Fig. 3). He was a right handed person. There was no evidence of acanthosis nigricans in other areas as right axilla, neck, groins, periumbilical areas or forehead. He was investigated by a physician for detailed endocrinal evaluation and also by an orthopedician for bony abnormalities, which were said to be insignificant from investigational and treatment point of view. His complete blood count, blood sugar levels, urinalysis, glycosylated haemoglobin level, glucose tolerance test and plasma insulin levels were all normal. His thyroid status was normal. No hormonal abnormalities were clinically suspected and investigated. His X-ray chest and abdomino-pelvic ultrasound were normal. X-ray Spine revealed slight scoliosis in the thoraco-lumbar region. There was no evidence of internal malignancy. Skin biopsy from pigmented area on chest showed mild acanthosis and hyperkeratosis with regular elongation of rete ridges in epidermis with hyperpigmentation in the basal layer confirming BN (Fig. 4).

In axillary area, hyperkeratosis, papillomatosis and acanthosis were seen with increased pigmentation in the basal layer seen, confirming AN. Thus, a clinical diagnosis of Becker's nevus with acanthosis nigricans was confirmed. We councelled the patient regarding BN and prescribed topical Tretinoin cream 0.1% for AN.

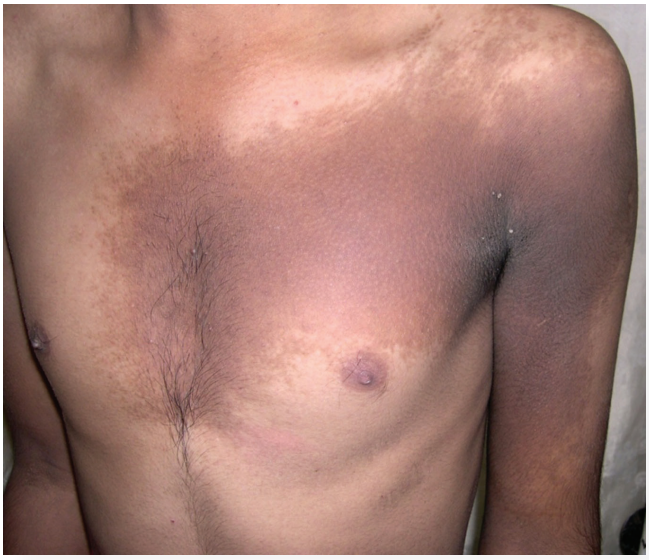

Figure 1. A well defined hyperpigmented macular lesion of Becker's nevus in left mammary area extending on to the anterior shoulder and arm with hypertrichosis and acneiform lesions.

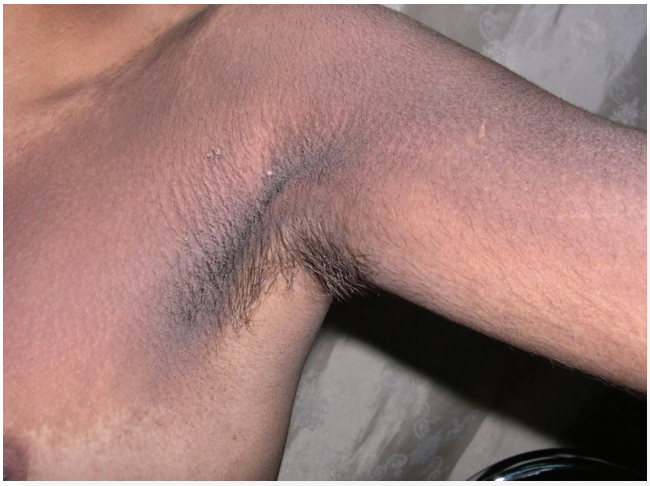

Figure 2. Ipsilateral papillomatous hyperkeratotic plaque in the left axilla with skin tags suggestive of acanthosis nignricans.

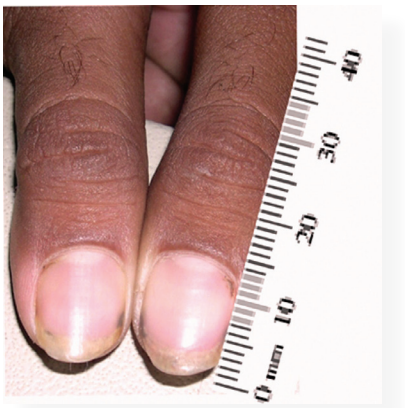

Figure 3. Increased longitudinal curvature and length of left index finger nail.

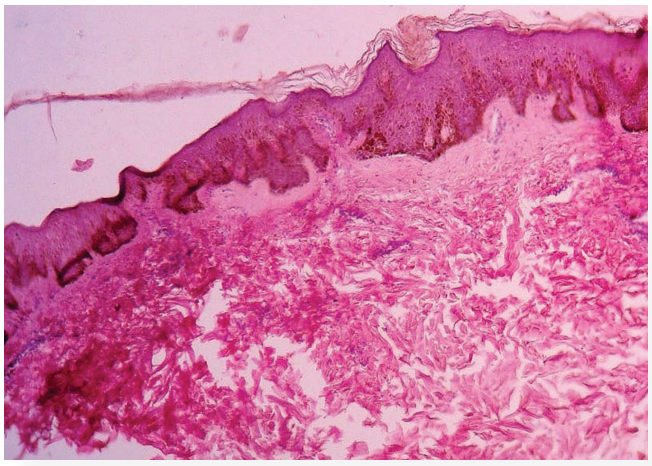

Figure 4. Histopathology showing acanthosis, elongation of rete ridges and hyperpigmentation in basal layer confirming Becker's nevus. H&E (10x10)

Discussion

Becker naevus (BN) was first described by Becker in 1949, as two cases where hypertrichosis and hyperpigmentation occurred on upper back unilaterally [1].Subsequently, term Becker Naevus syndrome [2] was proposed to encompass several developmental anomalies which were found to be associated with BN. Also they made this entity independent from the more generalised term of Hairy Epidermal nevus syndrome [3]. BN is an irregular well defined macular area, which is frequently associated with hypertrichosis and acneiform lesions. The overall incidence being 0.5% in males [4], it is usually more prominent around puberty. It has been postulated to be an ectodermal and mesodermal hamartoma, with increased epidermal (melanocyte), dermal (smooth muscle), and appendageal (hair follicle) components [5]. Some of the associated abnormalities reported are breast hypoplasia, aplasia of underlying pectoralis major, scoliosis, ipsilateral limb shortening, ipsilateral foot enlargement, spina bifida,supernummary nipples, short limbs and segmental odontomaxillary dysplasia. Congenital adrenal hyperplasia, polythelia and accessory scrotum have also been reported [2-6].

Acathosis nigricans (AN) typically presents as symmetric, brown black, velvety, hypertrophic, verrucous, papillomatous plaques most commonly involving the intertriginous sites including axillae, groins, sides of neck. Of the 8 types of AN, unilateral nevoid form is perhaps the rarest form reported [7].

Both sporadic and familial occurrences have been described for AN and it is to be demonstrating a para-dominant inheritance [7,8]. Unilateral Acanthosis nigricans is an extremely rare form of AN that manifests at any age at or before puberty and has a morphologic pattern similar to other forms of AN. It is generally not associated with syndromes, endrocrinopathies, drugs, or malignancies and is said to be inherited as an irregularly autosomal dominant trait that may first become evident at birth, in childhood, or during puberty and extends for a certain period and then either remains stationary or starts regressing [8]. It has a unilateral distribution and the histopathology is similar to the other AN. The differential diagnoses of nevoid AN include confluent and reticulate papillomatosis, Dowling Degos disease, melanocytic nevus and Becker's nevus itself. AN is more hyperpigmented and has raised velvety appaearance than a Becker's nevus.

The regional association between mammary hypoplasia and BN led to the hypothesis of a hormone-dependent disorder. Based on this, increase of the number of androgen-receptors in the affected areas was proved, which explained the appearing of lesions in puberty and alterations such as hypertrichosis and acneiform eruptions restricted to the affected regions. Androgenic sensitivity could also explain the peripubertal manifestation of classic Becker melanosis as well as the association with hypoplasia of the breast and areola in both males and females as this could antagonize estrogenic effects on breast development [9-13].

In our case, ipsilateral association of AN without any family history of Diabetes or AN in a case of BN with acneiform lesions within the nevoid zone, skin tags and hypretrichosis of terminal hairs with darkening compel us to speculate and need to explore underlying androgen sensitivity.

There is paucity in world literature regarding the simultaneous occurrence of these two disorders in the same individual. The only case reported previously being reported by Buck et al and described in detail later by Hulsmans et al [14], where both disorders, BN and AN, had been viewed to be androgen receptors mediated disorders. We were not able to perform androgen receptor studies due to constraint in resources and thus cannot comment on the relation of androgen receptors in our case. However, we speculate that our case may further strengthen the view of probable, more than a chance, association of BN with AN and suggest need for further exploration of the role of androgen receptors in such cases.

The case is presented here for the extreme rarity of a distinct morphological presentation of two different pigmentary disorders in the same patient.

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