Case Report

Idiopathic Hyperkeratosis of the Nipple and Areolar in a Nigerian Teenage Girl

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ABSTRACT

Hyperkeratosis of the nipple and areola (HNA) is a rare disorder characterised by hyperkeratotic or verrucuous plaques over the nipples and areola. Three distinct subtypes were described by Levy-Frankel in 1938. We report the first case of the idiopathic form of this dermatosis in a 14 year old Nigerian girl with remarkable improvement following the use of topical salicyclic acid ointment.

INTRODUCTION

Hyperkeratosis of the nipple and areola (HNA) is a rare disorder characterized by hyperkeratosis of the nipple with or without the areola. There have been very few case reports worldwide and the aetiology is still unknown.

REPORT

A 14 year old girl presented at the paediatric dermatology clinic with a one year history of thickened lesions over the nipples bilaterally of one year duration. It was not painful, not itchy, no nipple discharge and no associated pruritus. There was no swelling over the breasts and she had never been pregnant. There was no prior history of chronic rubbing of nipple neither was there any history of atopy. She had no similar lesions in other parts of the body. She was a sickle cell disease patient in stable state and had only been on routine haematinics given at the haematology clinic. Her mother had died of HIV-related illness about a year before but the patient was HIV-negative. She was very anxious and emotionally upset about the lesions and only confided in an older underage sibling who stumbled on it accidentally and brought her to the clinic.

Examination revealed verrucuous, hyperkeratotic plaques over the nipples bilaterally. There was a diffuse, reticulate and symmetric hyperpigmentation overlying the breasts (Fig 1). There were no other skin lesions to suggest warts, acanthosis nigricans or epidermal nevi. Patient refused biopsy and did not consent to her father being informed of the abnormality. She was subsequently placed on salicyclic acid ointment 20% for 1 month with significant improvement (Fig 2).

DISCUSSION

Hyperkeratosis of the nipple and areola (HNA) is an uncommon dermatosis with few reports if any, in a black African child. It is usually characterised by diffuse thickening, hyperpigmentation and verrucuous lesions over the nipple with or without involvement of the areola. Various classifications have been proposed but the description by Levy and Frankel in 1938 with 3 distinct subtypes is still the most popular. Type 1 is considered to be an extension of an epidermal nevus, Type 2 is in association with other skin disorders like chronic eczema, icthyosis, darier's disease, acanthosis nigricans while Type 3 was described as an idiopathic disorder called nevoid hyperkeratosis. The idiopathic form has been found to be commoner in the 2nd to 3rd decade of life presenting mostly with bilateral involvement of the breasts. It occurs in both men and women but has been found to be much commoner in women. Although aetiology is unknown, a hormonal influence has been postulated because of worsening of lesions with pregnancy and the development of hyperkeratosis following the use of oestrogen. Histopathologic findings include orthokeratotic

Figure 1: Verrucuous thickening over the nipples with diffuse reticulate hyperpigmentation over the breast and areola.

Figure 2: Nipple and areola clear of lesions following administration of salicyclic acid ointment

hyperkeratosis, papillomatosis, mild acanthosis in the epidermis and perivascular lymphocytic infiltrate in the dermis. Though we could not confirm our diagnosis, the age of the patient, the bilateral nature of the disease, the reticulate hyperpigmentation over the areola and lack of associated or similar skin lesions in other parts of the body suggests to us that this might be an idiopathic form of HNA. Various treatments like topical and oral retinoids, cryotherapy, electrocautery and surgical excision have been used in patients with no uniformity and varying success rates. Our patient made remarkable improvement within a few weeks of therapy with salicyclic acid ointment.

Awareness needs to be raised about this condition so that more reports can be made with a view to studying its pathogenesis and optimal treatment protocol.

REFERENCES

1. Burns DA. The Breast. In: Tony Burns, Breathnach S, Cox N, Griffith C, editors. Rook's Textbook of Dermatology, 7th ed. New York: Blackwell Science; 2004. p. 67.8-67.9.
2. Levy-Franckel A. Les hyperkeratoses de l'areole et du mamelon. Paris Med. 1938;28: 63- 66.
3. Revert A, Banuls J, Montesinos E, Jorda E, Ramon D, Torres V. Nevoid hyperkeratosis of areola. Int J Dermatol 1993;32:745-6.
4. Schwartz RA. Hyperkeratosis of the nipple and areola. Arch Dermatol. 1978;114(12):1844-1845.
5. Shastry V, Betkerur J, Kushalappa PA. Unilateral nevoid hyperkeratosis of the nipple: A report of two cases. Indian J Dermatol Venereol Leprol. 2006;72:303-5.