Multidermatomal nevus comedonicus: A case report

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Nevus comedonicus is an uncommon variant of adnexal hamartoma and is considered a rare subtype of epidermal nevi. It was first described in 1895 by Kofmann who used the term "comedo-nevus". It manifests as a group of closely dilated follicular openings with dark keratin plugs resembling comedones. Both unilateral and bilateral distributions are seen. The face is the most commonly affected site followed by the neck, the trunk, and the upper arms. We report a case of a 25-year-old male who presented with linear keratotic papules which on histopathology was confirmed to be nevus comedonicus. Our case was interesting because of its large size and of its multidermatomal involvement affecting the chest, back, and arms.

Keywords: nevus, nevus comedonicus, epidermal nevus

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INTRODUCTION

Nevus comedonicus (NC) manifests as groups of closely set, dilated follicular openings with dark keratin plugs resembling comedones. NC is caused by a defect in the development of the hair follicle [1]. Lesions may develop any time from birth to the middle age, but are usually present at birth or develop before the age of 10 years [2]. A few case reports have described a later onset in life including in the seventh decade. These cases usually occur after some form of trauma or a rash [2]. Men and women are equally affected. The majority of the cases are isolated. However, NC may be part of nevus comedonicus syndrome in association with skeletal or central nervous system anomalies, ocular abnormalities, and cutaneous defects [2,3].

CASE REPORT

We report a case of a 25-year-old male who presented with multidermatomal nevus comedonicus. He presented with linear, grouped, hyperpigmented, comedo like, keratotic papules on chest, left arm and back since birth. He also had multiple lesions with scarring on his back and posterior aspect of the arm since 8 years ago (Figure 1); the soles, palms, and mucosa were spared and regional lymph nodes were not palpable. The patient had no other health problem. There was no history of a similar case in the family and the patient was not the result of consanguineous marriage. He did not mention any history of preceding trauma or rash before appearing the lesions. Skin biopsy was taken from the arm lesion which showed features consistent with NC. (Figure 2)

DISCUSSION

NC manifests as single or multiple lesions. It may be linear, interrupted, unilateral, bilateral with a dermatomal distribution along the lines of Blaschko, or segmental. NC is typically found on the face, trunk, neck, and upper extremities. Rarely, it has been described on the palms and soles or the penis. When it occurs on the elbows and knees, it can appear as verrucous nodules [1,4-6]. In our case, the patient presented with multidermatomal involvement and the face, palms and soles were spared.

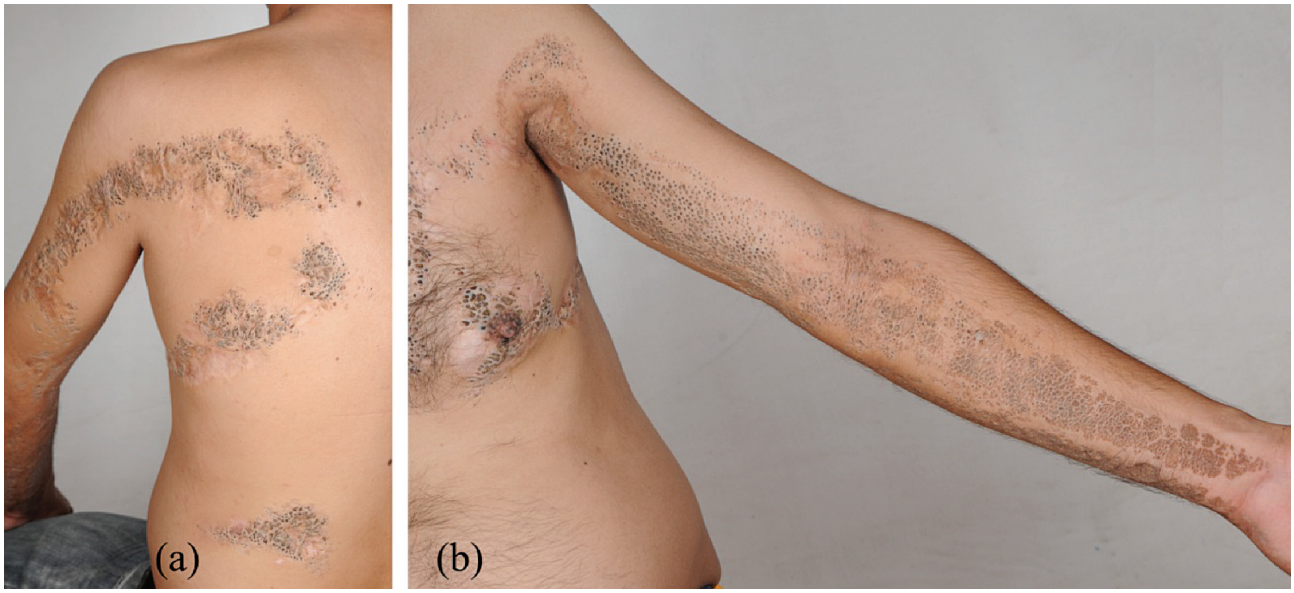

Figure 1. Linear disposition of nevus comedonicus (a) back of the patient (b) chest and arm with multidermatomal involvement.

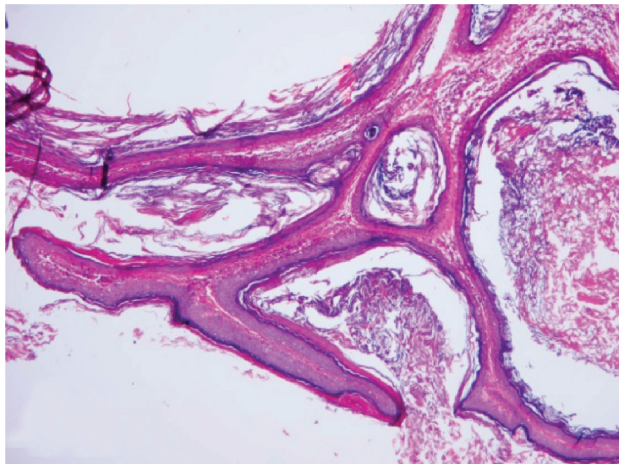

Figure 2. Large dilated pilosebaceous follicles filled with orthokeratotic keratin materials (H & E, × 400).

Histopathological examination showed large dilated pilosebaceous follicles filled with orthokeratotic keratin materials [3,6]. One to several hair shafts can be found occasionally on the base of the lesion. Similarly, small sebaceous gland lobules may be seen opening into the lower pole of the invaginations [3]. The interfollicular epidermis shows hyperkeratosis and papillomatosis resembling epidermal nevus. Occasionally, epidermolytic hyperkeratosis is seen. Inflamed and infected lesions will show the presence of dermal inflammatory cells infiltrates Histologically, differentiation from comedonal acne is important. In comedonal acne, the pilosebaceous units are complete whereas those they are poorly formed in NC. Moreover, in NC, hyperkeratosis and papillomatosis are frequently observed in the interpapillary epidermis that are absent in comedonal acne [5].

Treatment of NC is mainly for cosmetic reasons. However, for those with the inflamed entity, proper treatment is required to prevent complications of the recurrent infection and inflammation. Various types of treatments have been used in NC. Treatment outcomes are mostly unsatisfactory [4]. Topical agents like tretinoin, ammonium lactate lotion, tazarotene and calcipotriol have been reported to be cosmetically successful [6]. Our patient did not have fallow-up for treatment.

In conclusion, NC is a rare developmental anomaly mainly causing cosmetic disfigurement.

Complications due to recurrent inflammation and infection can occur.

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